REVIEW
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Rare anatomical localizations of hydatid cysts (2020-2025): Narrative review of presentation, imaging, and management

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ABSTRACT

Hydatid disease, caused by *Echinococcus granulosus*, typically affects the liver and lungs. However, cyst development in rare anatomical sites presents unique diagnostic and therapeutic challenges.

We conducted a narrative review using a structured search of PubMed, Scopus, and Google Scholar (first 200 records), with predefined eligibility criteria and dual-reviewer screening where applicable. Findings were synthesized narratively; numeric summaries reflect reporting frequency among included case-level reports rather than prevalence or comparative effectiveness.

Seventeen studies (case reports/small series) were included. Among the included reports, the most commonly reported rare sites were the brain, bone, and heart. Imaging narratives most frequently supported magnetic resonance imaging triage for central nervous system/spine/cardiac disease and computed tomography for osseous/retroperitoneal involvement, while surgery was most often reported as the index treatment with context-dependent use of albendazole. Percentages in this review reflect the share of reports among included studies (i.e., reporting frequency), not population prevalence, incidence, or comparative effectiveness.

Rare-site hydatid cysts require a high index of suspicion in endemic areas. Optimal outcomes are achieved through multidisciplinary evaluation, site-specific surgical planning, and tailored pharmacotherapy. Further research into minimally invasive techniques, molecular diagnostics, and vaccine development is warranted to improve diagnosis and long-term disease control.

Keywords: Albendazole therapy, diagnostic imaging, echinococcosis, extrahepatic involvement, general surgery, hydatid cyst, rare anatomical localization, narrative review

INTRODUCTION

Hydatid disease caused by *Echinococcus granulosus* predominantly involves the liver and lungs, but clinically meaningful presentations also occur at uncommon sites that can mimic neoplastic or inflammatory conditions (1,2). In this review, we systematically map reports published between January 2020 and March 2025, with the practical aim of summarizing site-specific presentations, imaging pitfalls, and management decision points that are most relevant to real-world multidisciplinary care (3). Rather than claiming an exhaustive synthesis, we focus on transparent methods and verifiable case-level details to support safer diagnostic and peri-operative choices across specialties.

Unusual localizations of hydatid cysts—such as the brain, spine, heart, bone, retroperitoneum, spleen, and musculature—are relatively rare but clinically important due to their non-specific presentations, diagnostic ambiguity, and increased risk of surgical complications (4,5). These atypical manifestations often mimic malignancies, abscesses, or congenital cysts, leading to delayed or inappropriate interventions (6). From a general surgery standpoint, such cases demand heightened suspicion and careful planning to minimize perioperative morbidity and recurrence risk. Recent literature has reported an increase in the detection of extrahepatic and extrapulmonary hydatid cysts, likely owing to advances in imaging technologies and improved clinical awareness (7). However, there remains a lack of consensus on the diagnostic and therapeutic approach to these rare presentations. While surgical excision remains the cornerstone of treatment, particularly in complicated or

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inaccessible locations, the roles of minimally invasive procedures and adjunctive antiparasitic therapy continue to evolve (8,9). The 2020–2025 window was selected a priori to reflect contemporary practice—including wider magnetic resonance imaging (MRI) availability, improved cardiac/central nervous system (CNS) imaging, and evolving minimally invasive and peri-operative strategies—while limiting historical heterogeneity. Because rare-site hydatid cysts are commonly co-managed, we explicitly adopt a multidisciplinary team frame throughout (radiology, general and specialty surgery, infectious diseases, neurosurgery/cardiothoracic as applicable). Our objective is therefore not to estimate prevalence or comparative effectiveness, but to provide verifiable, site-aware guidance—with study identifiers and DOIs—to help teams avoid hazardous pathways (e.g., inadvertent biopsy) and align around shared, pragmatic decisions.

MATERIAL and METHODS

Information Sources and Search Strategy

We performed a narrative review with a structured search (PubMed, Scopus, Google Scholar-first 200 records) covering January 1, 2020 to March 31, 2025, applying predefined inclusion/exclusion criteria and dual-reviewer screening with consensus resolution. Where helpful for transparency, we retained PRISMA-style reporting elements (search strings, selection flow), but we do not claim a protocol-registered systematic review. The full electronic strategies are provided below.

- PubMed (last searched March 31, 2025):

("hydatid cyst"[Title/Abstract] OR "cystic echinococcosis"[Title/Abstract] OR "Echinococcus granulosus"[Title/Abstract]) AND (brain OR cerebral OR spine OR spinal OR vertebral OR cardiac OR heart OR myocard* OR bone OR osseous OR pancreas OR pancreatic OR spleen OR splenic OR retroperitone* OR mediastin* OR breast OR muscle OR musculoskeletal OR renal OR kidney) AND (case OR "case report" OR series OR management OR surgery)

Filters: Humans, English, 2020/01/01–2025/03/31.

- Scopus (last searched March 31, 2025):

TITLE-ABS-KEY ("hydatid cyst" OR "cystic echinococcosis" OR "Echinococcus granulosus") AND TITLE-ABS-KEY (brain OR spine OR cardiac OR heart OR bone OR pancreas OR spleen OR retroperitone* OR mediastin* OR breast OR muscle OR musculoskeletal OR renal OR kidney) AND TITLE-ABS-KEY (case OR "case report" OR series OR management OR surgery) AND (LIMIT-TO (PUBYEAR, 2020–2025) AND LIMIT-TO (LANGUAGE, "English"))

- Google Scholar (last searched March 31, 2025):

"hydatid cyst" AND (brain OR spine OR cardiac OR bone OR pancreas OR spleen OR retroperitoneum OR mediastinum OR breast OR muscle OR renal) AND (case OR "case report" OR series OR management OR surgery).

First 200 records screened; backward citation tracking was performed from included articles.

We additionally hand-searched reference lists of eligible studies to identify missed reports (10). The study selection process is summarized in the PRISMA 2020 flow diagram (Figure 1).

Eligibility Criteria

We included peer-reviewed human reports published 2020-2025 that described primary hydatid cysts at uncommon, non-hepatic/non-pulmonary sites and provided clinical and/or imaging and/or management details. We excluded articles focused solely on hepatic or pulmonary disease, non-English or non-human studies, reviews without case-level data, and veterinary reports.

Study Selection

Two reviewers independently screened titles/abstracts, then assessed full texts against eligibility criteria. Disagreements were resolved by consensus. Reasons for exclusion at full-text stage were recorded (e.g., hepatic/pulmonary only; review without case-level data; non-English; non-human). We included 17 reports (case reports/small series). Within the included literature, the most frequently reported rare sites were brain, bone, and heart. Imaging descriptions most commonly supported MRI for CNS/spine/cardiac involvement and computed tomography (CT) for osseous/retroperitoneal disease, while initial management most often involved surgery with context-dependent albendazole. All quantitative summaries herein reflect reporting frequency within the included case-level literature and should not be interpreted as population prevalence, incidence, or comparative effectiveness (Table 1).

The search identified 537 records-523 through database searching and 14 from other sources. After removing 56 duplicates, 481 titles/abstracts were screened. 89 full texts were assessed for eligibility; 72 were excluded for the following reasons: not rare location (n=45), insufficient data (n=19), non-English (n=8). Seventeen studies were included in the narrative synthesis (with no quantitative synthesis/meta-analysis).

Data Collection and Items

Using a piloted extraction form, we captured: first author/year, patient demographics, anatomical site, presentation, imaging modalities/findings, management (surgery/percutaneous/medical), outcomes (recurrence/re-intervention/complications), and follow-up. Where available, DOI/PMID identifiers were recorded for verification.

Risk of Bias and Quality Appraisal

Given the case-report/series nature, methodological quality was appraised using the CARE checklist (case reports) and JBI

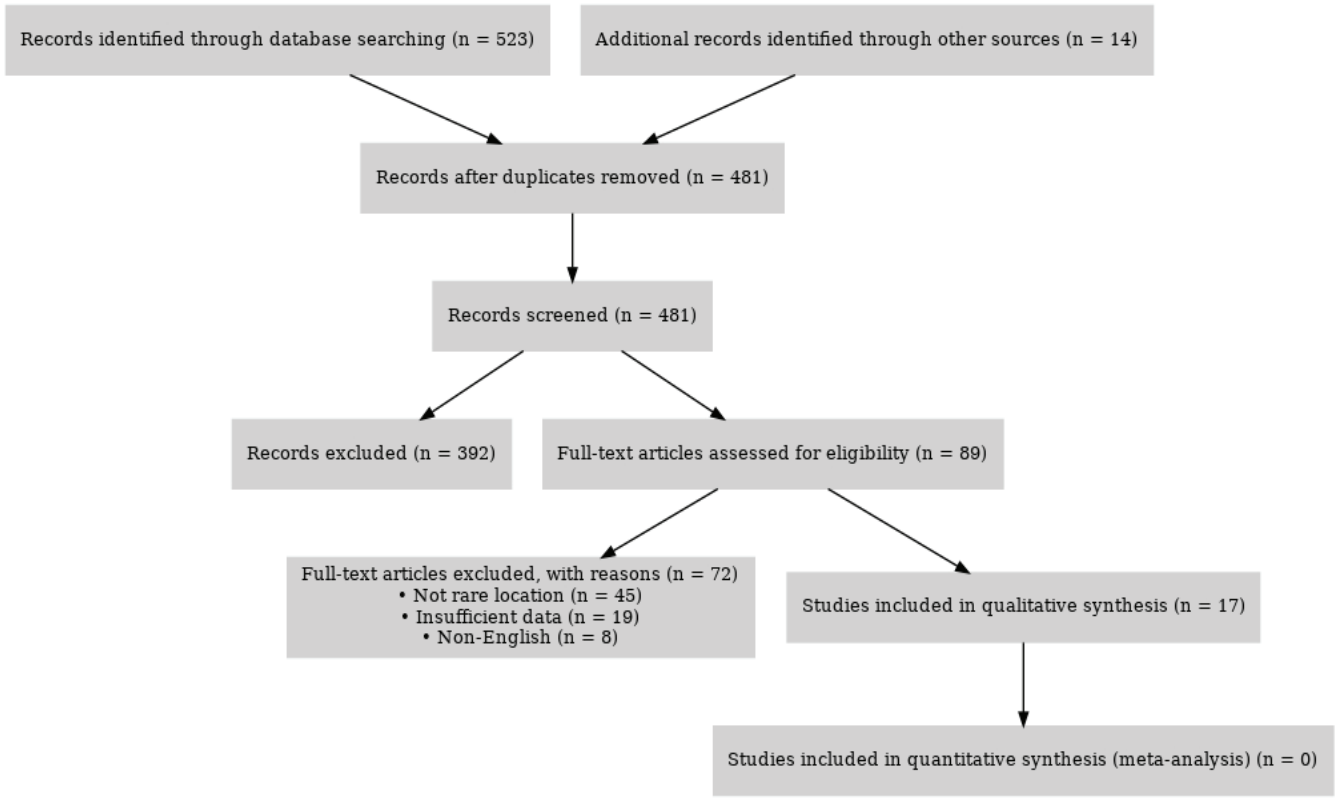

Figure 1. PRISMA flow.

Critical Appraisal Tool (case series) (28). We summarize study-level results in a dedicated table and contextualize limitations in the discussion.

Protocol and Registration

No review protocol was registered (e.g., PROSPERO); this is acknowledged as a limitation and mitigated through transparent reporting of search strategies, selection procedures, and identifiers.

Case reports were appraised against the CARE checklist and case series against the JBI critical appraisal tool. For each report, we assessed reporting domains including clinical findings, diagnostic assessment, therapeutic intervention, and follow-up/outcomes; when items were not explicitly stated, they were coded as "unclear." Study-level results are provided in Supplementary Table S2 (CARE and JBI worksheets).

Pathophysiology and Mechanisms of Dissemination to Uncommon Sites

Hydatid disease originates when the larval stage of *E. granulosus*—released from ingested eggs—penetrates the intestinal mucosa and gains access to the bloodstream or lymphatic system. The liver and lungs typically function as the primary and secondary filters, respectively, trapping the majority of oncospheres. However, when these barriers are bypassed, the larvae may disseminate to virtually any anatomical location (29). The hematogenous route is the most widely accepted mechanism for the spread of hydatid cysts to distant or atypical sites. Once in systemic circulation, oncospheres can seed tissues with rich vascular supply, such as the brain, heart, spleen, and musculoskeletal system (30). In certain locations—particularly the spine or pelvis—retrograde venous flow or lymphatic spread has also been proposed, especially in the absence of pulmonary or hepatic involvement (31). Osseous hydatidosis is uncommon in the included case literature and is typically reported to exhibit an infiltrative, trabecula-penetrating, multicystic growth pattern with limited pericyst formation. Unlike soft tissue cysts that develop a pericyst—a fibrous host reaction wall—bone lesions grow infiltratively, expanding through trabecular bone and mimicking malignancy or osteomyelitis (32). The absence of a containing capsule leads to diffuse extension and significant structural destruction. In cerebral hydatid disease—often described in children within the included case literature—parasites are thought to reach the CNS via arterial embolization, most commonly along the middle cerebral artery. The cysts grow slowly, with minimal inflammatory response, allowing for significant expansion before clinical symptoms arise (6). Moreover, immunological factors may influence the predilection of cysts for certain unusual locations. Some

studies suggest that immune evasion by the parasite or local immunosuppression may facilitate survival in otherwise reactive tissues, such as myocardium or splenic parenchyma. Genetic variations among *E. granulosus* strains (e.g., G1, G6) have also been linked to differential organ tropism, potentially impacting tissue specificity and dissemination patterns (33,34). A thorough understanding of these dissemination pathways is critical not only for diagnosis but also for planning site-specific surgical interventions. Misinterpretation of rare localizations can lead to diagnostic delays, inappropriate treatment, or inadvertent cyst rupture with potential anaphylaxis, underscoring the importance of pathophysiologic insight in clinical decision-making.

Diagnostic Challenges and Radiological Mimics of Uncommon Hydatid Cysts

The diagnosis of hydatid disease in atypical anatomical sites poses considerable clinical and radiological challenges. Unlike hepatic and pulmonary hydatidosis, which often display pathognomonic imaging features, cysts in rare locations frequently mimic neoplastic, congenital, or inflammatory lesions, leading to diagnostic delays or mismanagement (35). Differential radiological features of hydatid cysts in uncommon anatomical localizations, their frequent imaging mimics, and key differentiating points to aid diagnosis in Table 2.

Table 1. This table summarizes 17 case reports and series describing hydatid cysts outside the liver and lungs published between 2020 and 2025

Reference (first author, year)	Anatomical site(s)	Presentation	Imaging	Treatment	Outcome
Sebai et al. (11)	Retroperitoneum	Abdominal mass/ pain	CT	Total excision + albendazole	No recurrence at 12 mo
Mohamed Babiker et al. (12)	Multiple rare sites	Site-specific symptoms	US/CT	Site-specific surgery ± albendazole	Good overall; site-dependent
Khan et al. (13)	Cardiac (left ventricle)	Dyspnea, chest pain	Echocardiography + MRI	Open surgery + albendazole	Recovered
Wu et al. (14)	Pancreatic head	Epigastric pain; fever	CT/MRI ± s	Surgical excision + albendazole	Improved
Sqalli Houssaini et al. (15)	Orbit	Proptosis; visual disturbance	MRI	Orbitotomy + cyst excision	Vision preserved
Hakimi et al. (16)	Spleen	LUQ pain; palpable mass	CT/US	Splenectomy ± albendazole	Recovered
Getie et al. (17)	Musculoskeletal	Painful swelling	MRI	Excision + albendazole	No recurrence at 6-12 mo
Aryal et al. (18)	Cerebral	Headache; seizures	MRI	Craniotomy (en bloc) + albendazole	Seizure-free
Dhadve et al. (19)	Spinal (thoracolumbar)	Back pain; paraparesis	MRI/CT	Two-stage decompression + albendazole	Residual deficit
Ines et al. (20)	Breast	Painless mobile mass	US ± MRI	Lumpectomy + albendazole	Recovered
Bouchaala et al. (21)	Adrenal	Flank pain; incidental mass	CT/MRI	Adrenalectomy + albendazole	Recovered
Fourati et al. (22)	Retroperitoneum	Deep pelvic pain	CT/US	Excision + albendazole	Improved
Gadre et al. (23)	Mediastinum	Chest discomfort; cough	CT/MRI	Resection + albendazole	Recovered
Maamri et al. (24)	Intraventricular brain cyst	Headache; vomiting	CT/MRI	Craniotomy + albendazole	Recovered
Ganjipour Sales et al. (25)	Pelvic cavity	Pelvic pain; mass	MRI/CT	excision + Albendazole	Recovered
Ayub et al. (26)	Pericardium	Dyspnea; chest pain	Echocardiography + CT	Pericardial cyst excision + albendazole	Recovered
Maghbool et al. (27)	Kidney	Flank pain; hematuria	CT/US	Partial nephrectomy + albendazole	Recovered

CT: Computed tomograhpy, US: Ultrasound, MRI: Magnetic resonance imaging.

Imaging Modalities and Pitfalls

Ultrasound (US) remains a useful first-line tool, particularly for superficial or abdominal cysts, but its sensitivity decreases significantly for deep or bony structures. CT is valuable for identifying calcifications, osseous destruction, and cyst wall integrity, particularly in bone, retroperitoneum, or spine involvement (36). MRI, owing to superior soft-tissue resolution, is the modality of choice in CNS, musculoskeletal, and cardiac localizations, allowing for better delineation of cystic morphology and its relationship to neurovascular structures (37). However, in rare sites, hydatid cysts may lack classic radiological features—such as the presence of daughter cysts or the "water lily sign"—leading to frequent misinterpretation. For instance: Spinal cysts can resemble vertebral tuberculosis, metastases, or aneurysmal bone cysts.

Cardiac hydatidosis may appear similar to myxomas or fibromas on echocardiography or MRI. Intracranial cysts may be confused with arachnoid cysts, neurocysticercosis, or low-grade gliomas when daughter vesicles are not evident (38). Pancreatic or splenic cysts can radiologically overlap with pseudocysts, serous cystadenomas, or cystic neoplasms. In osseous echinococcosis, the lack of a pericyst results in a multilocular, expansile, and infiltrative lesion, often mistaken for aggressive bone tumors. The absence of significant periosteal reaction or sclerosis on CT may further obscure the diagnosis (32). Given the diagnostic ambiguity in rare localizations, a pragmatic stepwise pathway is summarized in Table 3 to guide imaging selection, ancillary testing, and safe tissue diagnosis.

Serologic and Molecular Tools

While serological tests—such as ELISA, indirect hemagglutination, or immunoblotting—can support diagnosis, their sensitivity is highly dependent on cyst location. Cysts in the brain or bones often provoke a muted immune response, resulting in false-negative serologies (39). Polymerase chain reaction (PCR) and next-generation sequencing (NGS) techniques are increasingly used to detect *E. granulosus* DNA in biopsy or aspirate specimens, especially when imaging and serology are inconclusive. However, their clinical utility remains limited due to accessibility and cost (40).

Histopathology: Final Confirmation

Definitive diagnosis often depends on histopathological analysis, typically obtained intraoperatively or post-excision. Presence of laminated membrane, germinal layer, and scolices confirms the diagnosis. However, this retrospective confirmation highlights the persistent difficulty of non-invasive preoperative diagnosis, particularly in rare sites. Given these challenges, clinicians must maintain a high index of suspicion when encountering cystic lesions in unusual locations,

Table 2. Radiologic differentials and site-specific pitfalls at uncommon locations of hydatid disease (2020-2025)

Anatomical site (reference #)	Typical hydatid cyst features	Radiological mimics	Key differentiating points
Brain (cerebral/ intraventricular) (8,14)	Well-defined, spherical cyst; CSF-isodense; no enhancement; minimal perilesional edema on MRI	Arachnoid cyst, cystic glioma, colloid cyst	Absence of mural nodule; smooth cyst wall; no solid component; lack of diffusion restriction
Cardiac (3,13,16)	Cystic intramyocardial lesion; hypointense rim; possible daughter cysts	Myxoma, fibroma, thrombus	No vascularity on Doppler; calcified rim possible; relation to myocardium or pericardium
Musculoskeletal/bone (7,9)	Multiloculated, expansile lesion; cortical thinning; minimal periosteal reaction	Aneurysmal bone cyst, giant cell tumor, metastasis	No periosteal aggressive reaction; presence of daughter cysts; lack of matrix mineralization
Spleen (6)	Cyst with internal septations or daughter cysts; calcified wall possible	Epidermoid cyst, pseudocyst, lymphangioma	Water attenuation; presence of daughter cysts; peripheral calcification
Breast (10)	Well-circumscribed cystic lesion; internal septations	Fibroadenoma, cystic carcinoma	No vascularity; laminated membranes may be visible on US
Pancreas (4)	Cystic lesion without communication to pancreatic duct; possible daughter cysts	Mucinous cystic neoplasm, serous cystadenoma, pseudocyst	No enhancement; absence of solid component; hydatid serology positive
Retroperitoneum (1,11,12,17)	Well-defined cystic lesion with or without calcification	Lymphangioma, mesenteric cyst, cystic sarcoma	Daughter cysts; laminated membranes on CT/MRI
Orbit (5)	Cystic mass causing proptosis; well-circumscribed	Dermoid cyst, hemangioma	No internal vascularity; fluid-fluid levels rare in hydatid

Each row cites supporting included studies using manuscript Ref# (11-27).
Note: Imaging patterns summarize reported features in the included case literature and may not generalize to population-level performance.
CT: Computed tomography, MRI: Magnetic resonance imaging, CSF: Cerebrospinal fluid.

Table 3. Site-aware diagnostic and management considerations synthesized from the included reports (2020-2025)

Clinical presentation (reference #)	First-line imaging	Supportive diagnostics	Differential diagnosis	Definitive diagnosis	Suggested next step
Non-specific symptoms (pain, mass, neuro signs) or incidental cystic lesion in endemic patient	US for superficial/ abdominal; CT for retroperitoneum/ bone; MRI for CNS/ spine/cardiac	Serology (ELISA/IHA) if safe to obtain; baseline labs (CBC, LFTs)	Abscess, cystic neoplasm, congenital cyst, TB/fungal infection	Not yet applicable	If hydatid suspected, proceed to targeted MRI/CT; avoid FNA in pancreas/CNS/ cardiac/bone
Cyst shows features suggestive of hydatid (daughter cysts, laminated membranes) (1,4-7,9,10,12,14,17)	MRI (soft-tissue detail) or CT (calcification/ bone destruction) depending on site	Repeat/confirm serology; consider PCR on surgical specimen when available	Arachnoid cyst, aneurysmal bone cyst, pseudocyst, myxoma, metastasis	Preoperative working diagnosis of hydatid	Plan site-specific management conference (surgery vs. PAIR vs. medical)
Equivocal imaging in deep or critical location (brain/heart/spine) (8,14)	High-resolution MRI ± contrast; gated cardiac MRI/echo if cardiac	Serology may be negative; consider advanced molecular assay (PCR/ NGS) if accessible	Low-grade glioma, thrombus, cysticercosis, sarcoma	Provisional diagnosis remains uncertain	Avoid percutaneous biopsy; prioritize surgical exploration only if benefits outweigh risks
Soft-tissue or muscular lesion with safe percutaneous window (7)	US/CT guidance to define safe tract	Pre-procedure anaphylaxis precautions; limited aspiration ONLY if diagnostic uncertainty persists	Pyomyositis, lymphangioma, softtissue sarcoma	Cyst fluid positive for hooks/ membranes or PCR (if performed)	If confirmed/ suspected hydatid: schedule total excision or carefully selected PAIR; start albendazole
Osseous/spinal destructive lesion without pericyst (9)	CT (cortical integrity) + MRI (neural elements)	Serology often negative; multidisciplinary review	TB spondylitis, metastasis, primary bone tumor	Intraoperative pathology (frozen) favors hydatid	Plan staged debridement/ reconstruction + prolonged albendazole; long-term imaging follow-up
Pancreatic or cardiac cystic mass (3,13,16)	Pancreas: contrast CT/MRI (no FNA); cardiac: echo + cardiac MRI	Serology as adjunct only	MCN/SCN/ pseudocyst (pancreas); myxoma/fibroma/ thrombus (heart)	Surgical specimen confirms hydatid	Proceed to definitive surgery in experienced center; postoperative albendazole

Each decision node is annotated with supporting manuscript Ref# (11-27).
Note: This site-aware pathway synthesizes reported practice patterns and should be applied within an MDT, tailored to local expertise and patient context.
IHA: Indirect hemagglutination, CT: Computed tomography, MRI: Magnetic resonance imaging, PCR: Polymerase chain reaction, CNS: Central nervous system, FNA: Fine-needle aspiration, LFT: Liver function test, TB: Tuberculosis, NGS: Next-generation sequencing, PAIR: Puncture, aspiration, injection, re-aspiration, CBC: Complete blood counts, MDT: Multidisciplinary team, MCN: Mucinous cystic neoplasm, SCN: Serous cystic neoplasm.

especially in patients from endemic areas. Multimodal imaging supported by serological and, when available, molecular techniques can guide accurate preoperative planning.

Clinical Presentation and Site-specific Manifestations

The clinical presentation of hydatid cysts in rare anatomical sites is highly variable and largely dictated by cyst size, rate of growth, and proximity to adjacent critical structures. Unlike hepatic or pulmonary echinococcosis, which often remains asymptomatic until large sizes are reached, cysts in atypical locations frequently produce early and misleading symptoms, leading to misdiagnosis (41).

Splenic Involvement

Isolated splenic hydatid cysts are uncommon in the included case literature and are only infrequently reported as solitary lesions. Patients often present with left upper quadrant pain, splenomegaly, or a palpable mass. Rupture into the peritoneal cavity may lead to acute abdomen or anaphylactic shock, especially in endemic regions. The condition can be confused with splenic pseudocysts, abscesses, or cystic neoplasms (42).

Osseous and Musculoskeletal Hydatidosis

Skeletal hydatid disease, particularly in the spine, pelvis, femur, and humerus, may mimic malignancies or chronic infections. Symptoms typically include chronic pain, pathological fractures, or neurological deficits in vertebral involvement. The cysts exhibit an infiltrative, trabecular pattern without encapsulation, leading to aggressive local destruction and high recurrence rates (43).

CNS

Intracranial hydatid cysts are more commonly seen in pediatric patients and often involve the parietal or occipital lobes. Symptoms include increased intracranial pressure (headache,

vomiting, papilledema), seizures, or focal neurological deficits. Cysts grow slowly and are frequently misdiagnosed as arachnoid cysts or brain tumors unless daughter cysts are clearly visualized (44).

Cardiac Hydatid Disease

Cardiac involvement is uncommon in the included case literature but is associated with substantial morbidity, particularly when intracavitary extension or conduction pathways are involved. The left ventricle is most commonly affected, followed by the interventricular septum and right atrium. Clinical presentation ranges from asymptomatic murmurs to arrhythmias, embolic events, or sudden cardiac death. Echocardiography and cardiac MRI are essential for diagnosis and surgical planning (45).

Breast, Thyroid, and Other Soft Tissues

Hydatid disease of the breast may resemble fibroadenoma or cystic carcinoma, while thyroid involvement can mimic multinodular goiter or thyroid cancer. In both cases, symptoms are often limited to a slow-growing, painless mass, and diagnosis is typically made postoperatively (46).

Other rare sites include:

- Adrenal glands (mimicking pheochromocytoma)

- Retroperitoneum (mimicking sarcoma or lymphoma)

- Scrotum and epididymis (suggesting hydrocele or neoplasm)

- Orbit (causing proptosis or visual disturbances)

Multiorgan and Disseminated Hydatidosis

In immunocompromised individuals or following surgical rupture of a primary cyst, multiple secondary cysts may develop. Disseminated disease may manifest with multifocal pain, systemic symptoms, or acute abdomen, necessitating comprehensive imaging and multidisciplinary management (47).

Management Strategies: Surgical, Percutaneous, and Medical Approaches in Rare Sites

The therapeutic management of hydatid disease in atypical anatomical locations is complex, requiring individualized strategies based on cyst location, complications, and the patient's overall health status. Unlike hepatic hydatidosis, where standardized protocols exist, rare site involvement often demands a multidisciplinary, case-based approach (2).

Surgical Management

Surgery remains the cornerstone of treatment for hydatid cysts in most uncommon locations, especially when vital organs are involved or complications such as rupture, compression, or infection occur. Recommendations are MDT-conditioned and context-dependent; peri-operative albendazole is considered as an adjunct per site and spillage risk.

Osseous involvement often necessitates wide excision or curettage, sometimes with reconstructive procedures like bone grafting or spinal stabilization. Complete resection is often challenging due to the infiltrative nature of cysts and the absence of a true capsule (32). Cerebral hydatid cysts are typically removed via en bloc excision using techniques like the Dowling-Orlando method, which allows gentle extraction under irrigation, minimizing rupture risk (6). Cardiac hydatidosis requires open-heart surgery with cardiopulmonary bypass. The cysts must be removed without intraoperative rupture to avoid fatal embolism or anaphylaxis (45). Breast, thyroid, and retroperitoneal cysts may be approached via total excision, but adhesions and anatomical distortion may complicate dissection. The main surgical goal is complete cyst removal without spillage, as rupture increases recurrence and systemic allergic reactions.

Percutaneous Techniques Puncture, Aspiration, Injection, Re-aspiration (PAIR)

The PAIR technique, widely accepted for liver cysts, has limited but growing application in select extrahepatic cases. Suitable for superficial or isolated soft tissue cysts (e.g., muscle or subcutaneous tissue) where adjacent vital structures can be avoided under imaging guidance (48). Contraindicated in CNS, osseous, cardiac, and retroperitoneal locations due to rupture risk. Can be used as a bridge to surgery or for palliative management in inoperable cases, though long-term recurrence rates remain uncertain. For non-hepatic sites, indications are highly selective and evidence is limited; decisions should be MDT-driven and center-experienced.

Pharmacologic Therapy

Albendazole remains the mainstay medical therapy, administered at 10-15 mg/kg/day in divided doses over several weeks to months. Mebendazole, though less effective, may be used in certain settings. Pharmacologic treatment is indicated in three scenarios:

- Preoperative sterilization to minimize intraoperative rupture risk.

- Postoperative prophylaxis, particularly after partial excision or spillage.

- Primary treatment in inoperable or disseminated cases, including CNS or bone involvement (49).

Therapeutic monitoring is essential due to potential hepatotoxicity and myelosuppression. Some studies support combination therapy (e.g., albendazole + praziquantel), which may enhance scolicidal efficacy, but evidence remains limited.

Peri-operative albendazole: Conditional, MDT-driven use (dose/monitoring by site and context) We suggest peri-operative albendazole within an MDT when timing and safety

permit—especially after spillage, partial resection, multiple cysts, or uncertain margins. Typical regimens in guidance and practice are 10-15 mg/kg/day in two divided doses (or 400 mg BID in adults; max 800 mg/day), started pre-operatively when feasible and continued post-operatively with duration tailored to site, spillage, and residual disease (49). Pre-op lead-in: Short courses (~2 weeks) can reduce protoscolex viability; longer pre-op courses are used when logistics allow, recognizing mixed evidence (50). Post-op continuation: Common practice ranges 1-3 months after uncomplicated resection and longer when spillage/ residual disease is suspected; pulmonary reviews suggest ≥3-6 months in selected cases. Longer courses (e.g., in bone/ CNS) are reported but rest on low-certainty evidence (50). Site-aware nuances:

• CNS/cardiac: Surgery is primary; adjunct albendazole is often used pre/post but data are case-level. Decisions should be individualized (51).

• Bone: Prolonged courses are described due to infiltrative growth and difficult clearance; evidence remains heterogeneous (49).

Monitoring & safety: Check LFTs at baseline and periodically; consider biliary/hematologic labs if prolonged therapy. Avoid in 1st trimester pregnancy; review drug interactions and counsel on taking with fat-containing meals to increase bioavailability (51).

Framing: These are practice-supporting suggestions derived from guidance and case-level literature, not prescriptive standards or comparative-effect estimates. Local expertise and patient factors should drive the final plan (52).

Avoiding Biopsy/FNA When Hydatid Disease is Suspected (MDT-conditioned)

Because needle biopsy/FNA can precipitate cyst rupture, dissemination, or anaphylaxis, routine tissue sampling should be avoided when the clinical-imaging context suggests hydatid disease, particularly in pancreas, spleen, bone, brain, and heart. In rare, inconclusive scenarios, selective sampling may be considered within an MDT at centers with immediate capability to manage anaphylaxis, using protective measures and only after a risk–benefit discussion that documents why imaging and serology were insufficient. This approach is aligned with major guidance emphasizing non-invasive diagnosis and risk-avoidance where hydatid disease is on the differential (48). Practical note (reporting level): If biopsy is undertaken despite caution, teams should report pre-procedure risk stratification, intra-procedure protections, and outcomes to strengthen the case-level evidence base.

Emerging Therapies

Recent years have witnessed exploration of alternative interventions:

- Microwave ablation, radiofrequency ablation, and stereotactic radiosurgery for inoperable CNS lesions.

- Robotic-assisted surgeries in complex anatomical regions (e.g., mediastinum).

- Novel anthelmintics (e.g., oxfendazole, nitazoxanide) are under investigation.

- Fluorescence-guided surgery has shown promise in achieving complete cyst removal in experimental models.

- Although these modalities are not yet standard practice, they reflect an evolving approach toward less invasive and more targeted therapy.

Prognosis, Recurrence, and Follow-up Considerations in Atypical Hydatid Disease

The prognosis of hydatid cysts in unusual anatomical sites is highly variable and generally less favorable compared to hepatic or pulmonary involvement. Factors influencing prognosis include anatomical location, cyst size, surgical accessibility, presence of complications, and the completeness of cyst removal (53).

Recurrence Rates and Contributing Factors

Recurrence remains a major concern in atypical-site hydatidosis and was frequently reported across included cases, with signals varying by anatomical site and management approach:
- Osseous hydatidosis exhibits the most frequently reported recurrence due to its infiltrative growth, absence of pericyst formation, and challenges in achieving radical resection. In spinal hydatidosis, recurrence rates can exceed 40%, even with aggressive surgical techniques (32).
- In CNS involvement, recurrence is low if en bloc removal is achieved. However, intraoperative rupture can lead to intracranial dissemination or ventricular seeding, complicating future management (54).
- Cardiac and muscular cysts may recur if daughter cysts remain or if surgical margins are insufficient due to anatomical constraints (55).
Inadequate preoperative sterilization, incomplete excision, and lack of postoperative albendazole therapy are widely recognized as modifiable risk factors for recurrence.

Postoperative Monitoring

A structured follow-up protocol is essential, particularly during the first two to three years, when recurrence is most likely to occur.

- MRI is preferred in CNS and spinal cases, given its ability to detect early recurrences and evaluate soft tissue detail.

- CT scans are valuable for follow-up in osseous, retroperitoneal, and thoracic cysts.

- US remains useful for superficial cysts or abdominal surveillance.

- Serologic monitoring can be adjunctive, although anti-echinococcal antibody levels may remain elevated for prolonged periods and are not always reliable indicators of recurrence (56).

Long-term Outcomes

Long-term prognosis varies according to site and extent of disease:

- Brain hydatidosis has good outcomes if complete removal is achieved early, but neurological deficits, seizures, or hydrocephalus may persist.

- Spinal cysts often result in permanent disability, including paraplegia or chronic pain, despite adequate decompression and adjuvant therapy.

- Cardiac hydatidosis, while rare, carries the risk of sudden death, but surgical outcomes are favorable if diagnosis is timely and excision is complete (57).

Rehabilitation and Supportive Measures

Patients with significant neurological impairment or extensive skeletal involvement may benefit from rehabilitation programs, including:

- Physical therapy

- Orthotic support or prosthetic fitting

- Anticonvulsant therapy in CNS cases

- Psychosocial support, especially in younger or disabled individuals.

In endemic areas, community-based follow-up and patient education are crucial to minimize reinfection and promote long-term surveillance.

Future Directions and Research Perspectives in Atypical Hydatid Disease

Despite notable progress in the diagnosis and treatment of cystic echinococcosis, hydatid disease in rare anatomical sites continues to present diagnostic, therapeutic, and prognostic challenges. Future advancements across molecular biology, immunology, pharmacology, and surgical technologies may offer innovative solutions to these complexities.

1. Advances in Diagnostic Imaging

Emerging radiologic techniques such as diffusion-weighted MRI, positron emission tomography-CT, and radiomics-based artificial intelligence algorithms are gaining attention for their potential to enhance the differentiation of hydatid cysts from malignant or inflammatory lesions, especially in deep or unusual locations (58). AI-assisted interpretation of imaging data may support earlier diagnosis by recognizing subtle and atypical imaging patterns in locations like the spine, retroperitoneum, or brain, where hydatidosis often mimics neoplasms.

2. Molecular and Serological Innovations

While serological assays remain suboptimal in specificity and sensitivity for rare-site disease, recent developments in PCR-based methods and NGS of cyst aspirates or surgical samples offer improved diagnostic accuracy (59). Additionally, point-of-care molecular diagnostics using loop-mediated isothermal amplification are under development and may enhance field-level diagnostics, particularly in endemic but resource-limited areas.

3. Immunotherapeutic Strategies and Vaccines

The modulation of host immune responses to *E. granulosus* is an emerging focus area. Novel therapies targeting Th1/Th2 immune polarization, dendritic cell activation, and regulatory T-cell suppression are under experimental investigation. Promising vaccine candidates such as EG95 recombinant antigen and EgAgB-based multiepitope formulations have shown protective efficacy in animal models. Their translation to human use, especially for high-risk occupations or endemic populations, is under active exploration (60).

4. Novel Antiparasitic Agents and Delivery Systems

Standard agents like albendazole and mebendazole exhibit limited efficacy in poorly vascularized sites such as bone or brain. Recent research has focused on agents like oxfendazole, tribendimidine, and nitazoxanide, which have shown superior protoscolicidal activity in experimental models (50). Moreover, liposomal formulations and nanoparticle-conjugated antiparasitic drugs are being tested to improve tissue penetration, reduce systemic toxicity, and enable targeted drug delivery.

5. Surgical and Technological Innovation

Minimally invasive techniques, including robot-assisted surgery and endoscopic approaches, are under investigation for anatomically challenging cysts in the mediastinum, orbit, or retroperitoneum. These methods may reduce morbidity and improve postoperative recovery. Intraoperative technologies such as fluorescence-guided resection and real-time 3D navigation may also enhance cyst delineation and reduce the risk of rupture, particularly in CNS and musculoskeletal locations.

6. One Health Surveillance and Genotype Mapping

Hydatid disease control hinges on integrated One Health strategies, including veterinary, environmental, and public health interventions. Genotyping of *E. granulosus* strains using cox1 and nad1 markers has revealed significant strain-related differences in tissue tropism and pathogenicity (61). Real-time surveillance of parasite circulation in livestock and canine populations, coupled with education and sanitation measures, remains fundamental in reducing human infection—especially with cysts arising in unusual anatomical sites.

Controversies and Consensus in Management of Atypical Hydatid Cysts

The management of hydatid disease in rare anatomical sites is fraught with challenges, and in many areas, consensus is lacking. Despite general agreement on the importance of surgical resection and antiparasitic therapy, several controversies remain regarding timing, technique, and adjunctive measures.

PAIR in Uncommon Sites: A Matter of Debate

While the PAIR technique has gained widespread acceptance in hepatic hydatid cysts, its application in atypical localizations remains contentious. Proponents argue that with appropriate precautions and imaging guidance, PAIR may offer a minimally invasive alternative in selected superficial or muscular cysts. However, critics highlight the elevated risk of cyst rupture, dissemination, and anaphylaxis, particularly in CNS, osseous, or cardiac locations where containment is anatomically limited (62). There is currently no standardized guideline governing PAIR in non-hepatic hydatidosis, and most published cases are isolated reports or small series, limiting the generalizability of outcomes.

Duration of Antiparasitic Therapy: Is More Always Better?

The optimal duration of albendazole therapy is also debated, especially when used as adjuvant or neoadjuvant treatment. Standard regimens of 3 months may be insufficient for osseous, cerebral, or disseminated cysts, leading some experts to advocate for 6-month or even longer treatment courses. However, prolonged therapy raises concerns regarding hepatotoxicity, bone marrow suppression, and patient adherence (51). Emerging data suggest that combination therapy with praziquantel may enhance efficacy, yet consensus is lacking on its routine use in rare-site disease (52).

Role of Corticosteroids: Adjunct or Risk?

The role of perioperative corticosteroids in preventing anaphylaxis remains uncertain in the case-level literature. Several reports describe the use of a single preoperative dose—typically considered in contexts of high cyst burden or suspected leakage—as a potential adjunct within an MDT. At the same time, others caution against routine use in CNS disease because of concerns about immunosuppression and potential parasitic persistence (63). Taken together, decisions should be individualized (site, spill risk, and timing) and made within an MDT, rather than viewed as a standard recommendation.

Watchful Waiting in Asymptomatic Cases: A Reasonable Option?

In certain scenarios, especially with incidentally discovered, asymptomatic cysts in surgically challenging sites (e.g., retroperitoneum, deep musculature), some clinicians opt for a watch-and-wait strategy with close radiological surveillance. While this approach reduces surgical morbidity, it is not without risk—cyst rupture or secondary infection may occur unpredictably (1). These ongoing debates reflect the complexity of treating hydatid cysts outside of classical hepatic and pulmonary sites. There is a growing need for multicenter data, standardized treatment algorithms, and controlled studies that evaluate outcomes of conservative vs. interventional approaches in rare anatomical presentations.

Overall Certainty/Limitations

Given that the vast majority of included records are single-patient case reports, the level of evidence is extremely low; therefore, statements herein should be read as practice-supporting, MDT-conditioned suggestions, not as prevalence or comparative-effect claims.

This is a narrative review based largely on single-patient case reports, so findings are fragmentary and should not be over-interpreted. Although we describe the use of CARE and JBI tools, the risk-of-bias assessment is limited by the underlying reports and is not comprehensive; item-level results are summarized but remain insufficiently detailed for formal grading. We restricted inclusion to English-language publications, which introduces language bias. The Google Scholar component was limited to the first 200 records, further adding non-systematic elements. Accordingly, all numeric statements reflect reporting frequency within the included case-level literature (not prevalence/effectiveness), and all clinical suggestions are MDT-conditioned and context-dependent.

Overall, the evidence base consists predominantly of single-patient case reports, which limits internal validity and precision. In our CARE-based appraisal (n=17), reporting was consistently adequate for clinical findings (17/17), diagnostic assessment (imaging/labs, 17/17), therapeutic intervention (17/17), and follow-up/outcomes (17/17). In contrast, several key domains were frequently not explicitly reported, including timelines (Unclear in 17/17), adverse events (Unclear in 17/17), patient perspective (Unclear in 17/17), and informed consent statements (Unclear in 17/17). Titles explicitly identifying a case were present in 13/17, and a structured abstract appeared likely in 11/17 (journal formatting dependent). When small case series are present, JBI domains such as consecutive inclusion

and complete inclusion are seldom verifiable from the text. Accordingly, our conclusions are positioned as practice-supporting, MDT-conditioned suggestions, not estimates of prevalence or comparative effectiveness, and should be interpreted with these reporting limitations in mind.

Future Directions and Unresolved Questions

Over the 2020-2025 period, case-level evidence at uncommon sites highlights practical opportunities for MDT-driven standardization while underscoring persisting gaps. Prospective, site-specific registries with harmonized reporting (presentation, imaging sequence, peri-operative details, cyst stage, spillage, and follow-up) are needed to move beyond narrative signals. Candidate endpoints include peri-operative complications, recurrence at site, need for re-intervention, and functional recovery (e.g., neurologic or cardiac performance where applicable). Imaging research should compare MRI vs. CT triage by anatomical site, and clarify when US suffices for surveillance. Pharmacologic studies could define timing and duration of peri-operative albendazole, especially after spillage or uncertain margins. Finally, consensus criteria for when to avoid percutaneous biopsy/FNA at pancreas, spleen, bone, brain, and heart should be formalized within MDT pathways.

Unresolved questions:

- Which site-specific imaging algorithms reduce misdiagnosis and biopsy-related risk most effectively?

- What minimal effective duration of peri-operative albendazole balances safety with recurrence prevention?

- How should recurrence be defined and monitored across sites (imaging schedule, markers)?

- Can MDT checklists/decision tools improve time-to-treatment and outcomes in low-volume centers?

CONCLUSION

Rare-site hydatid disease is best approached through MDT coordination with site-specific imaging triage and risk-aware operative planning. Based on 2020-2025 publications, three practical actions are feasible now: (i) prioritize MRI for CNS/spine/cardiac involvement and CT for osseous/retroperitoneal disease, reserving US for screening or superficial sites; (ii) use peri-operative albendazole when timing and safety permit, with postoperative continuation after spillage, partial resection, or uncertain margins; and (iii) avoid percutaneous biopsy/FNA when imaging and clinical context suggest hydatid disease in the pancreas, spleen, bone, brain, or heart, unless a specialized MDT determines that benefits outweigh risks. These are practice-supporting suggestions derived from recent case-level evidence rather than claims of prevalence or efficacy. Standardized reporting, prospective registries, and consensus criteria for non-hepatic PAIR would further improve consistency and outcomes across centers.

Footnotes

Author Contributions

Concept – B.Z.; Design - B.Z., Ö.A., T.D.; Data Collection or Processing - Ö.A., T.D.; Analysis or Interpretation - B.Z., Ö.A., T.D.; Literature Search - B.Z., Ö.A., T.D.; Writing - B.Z., Ö.A., T.D.

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