Section: Pathology

Original Research Article

# HYDATID DISEASE AT UNUSUAL ANATOMICAL SITES: HIGHLIGHTING DIAGNOSTIC CHALLENGES AND LIMITATIONS OF SEROLOGY

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*Received* : 10/02/2026
*Received in revised form* : 24/03/2026
*Accepted* : 13/04/2026

*Keywords:*
*Hydatid Disease, Unusual Location, Kidney, Muscle, Brain.*

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DOI: 10.47009/jamp.2026.8.2.205

Source of Support: Nil,
Conflict of Interest: None declared

*Int J Acad Med Pharm*
2026; 8 (2); 1125-1127

## ABSTRACT

**Background:** Hydatid disease commonly involves the liver and lungs; however, involvement of unusual anatomical sites is rare and poses significant diagnostic challenges. **Materials and Methods:** We conducted a retrospective observational study over 6 years at a tertiary care centre, including patients with histopathologically confirmed hydatid disease at atypical sites. **Results:** Ten patients (age range 7–75 years) were identified, with involvement of skeletal muscle (n=4), kidney (n=3), pericardium (n=1), adrenal gland (n=1), and brain (n=1). Radiologically, all lesions were cystic, with occasional multiloculated morphology. Only 30% of cases showed positive hydatid serology. Three cases were due to Echinococcus multilocularis. All patients were treated with surgery and albendazole with good outcomes. **Conclusion:** Hydatid disease should be considered in the differential diagnosis of cystic lesions at unusual sites, especially in endemic regions. Serology has limited sensitivity in extrahepatic disease, necessitating reliance on imaging and histopathology.

## INTRODUCTION

Echinococcal species, mostly E. granulosis and E. multilocularis, are mainly responsible for causing a parasitic infection known as hydatid disease. They are commonly found in endemic regions of the world like arts of South America, the Middle East, and Eastern Europe, where transmission is facilitated by livestock farming and close contact with domestic dogs. The definitive host of these organisms are mostly animals such as dogs and humans serve only as intermediate hosts.[1]

Lungs and liver are the most commonly affected organs accounting for approximately 75% and 15% of these cases respectively. In contrast involvement of sites like primary musculoskeletal system and some organs kidney and adrenal are extremely uncommon.[2] Although theoretically hydatid disease can involve any site, the exact reason why some sites are preferred over others is largely unknown.

Their occurrence at these unusual sites poses a great diagnostic challenge and are often misdiagnosed as more common lesions like abscess, hematoma or soft tissue tumors.[3,4] We aimed to study the clincopathological and radiological features of hydatid disease at these unusual sites.

## MATERIALS AND METHODS

**Study Design:** This was a retrospective, cross-sectional, observational study conducted at Sher I Kashmir institute of medical sciences, Soura, Srinagar for a period of 6 years.

**Patient Selection**

A thorough search for patients with hydatid disease in the database of department of pathology was conducted. Patients with hydatid disease at unusual sites with complete clinicopathological data were collected. Haematoxylin and Eosin-stained slides of all the patients were retrieved for evaluation.

**Data Variables**

Patient demographics, clinical presentation, site involved, radiology, serology and follow-up outcomes were collected.

**Ethical approval and consent**

Informed consent was not sought, as it was a retrospective study and did not involve any active participation of the patients. The patients whose data were analysed could also not be contacted further. Many patients were deidentified, and the waiver of their consent did not adversely affect the rights and welfare of the participants.


ISSN (O): 2687-5365; ISSN (P): 2753-6556

## RESULTS

The cohort included patients aged 13–75 years, with a slight female predominance (5 females, 4 males). The sites of involvement included skeletal muscle (axillary region, gluteal region, posterior thigh, and deltoid muscle; n=4), kidney (n=3), pericardium (n=1), brain (n=1) and adrenal gland (n=1).

Radiologically, all lesions were described as cystic masses, with sizes ranging from 3.2 cm to 24 cm. Among these, multiloculated morphology was observed in two cases (one renal and one intramuscular thigh lesion), while the remaining lesions were predominantly unilocular cystic masses. The largest lesion was identified in the posterior thigh (24 × 7.6 × 7.7 cm), extending along myofascial planes up to the popliteal fossa. The pericardial cyst measured 6×6 cm and was associated with compression of the tricuspid valve, indicating a clinically significant mass effect.

Species differentiation revealed that 7 cases were due to Echinococcus granulosus while 3 cases were due to Echinococcus multilocularis, involving the posterior thigh, gluteal region, and adrenal gland.

Hydatid serology was positive in only 3 cases. All of these cases were managed with a combination of surgery and albendazole. On follow-up all these patients are doing fine.

The clinical and radiological details of these patients are given in Table 1.

**Table 1: Clinicopathological and radiological features of patients with hydatid disease at unusual sites**

| CASE NO | AGE | GENDER | SITE | TYPE | RADIOLOGY |
|---|---|---|---|---|---|
| 1 | 75 | F | AXILLARY MASS | E. Granulosus | 10x4 cm cystic lesion in the muscular planes. |
| 2 | 23 | F | GLUTEAL REGION | E. Multilocularis | 9x8cm cystic lesion with a thin peripheral enhancement extending from paravertebral region to gluteal region. |
| 3 | 56 | M | KIDNEY | E. Granulosus | Cystic mass measuring about 5x4cm in diameter. |
| 4 | 13 | M | KIDNEY | E. Granulosus | Multi loculated cystic lesion measuring 5.4 × 3.6 × 2.7 cm |
| 5 | 32 | M | KIDNEY | E. Granulosus | Cystic mass measuring about 3.2x2.8 cm in diameter. |
| 6 | 38 | F | POSTERIOR THIGH | E. Multilocularis | multi loculated cystic lesion measuring 24 × 7.6 × 7.7 cm in myofascial planes of posterior compartment of left thigh extending till popliteal fossa. |
| 7 | 42 | M | DELTOID MUSCLE | E. Granulosus | 5x4 cm cystic lesion in the muscular planes. |
| 8 | 14 | F | PERICARDIUM | E. Granulosus | 6x6cm cystic lesion in the pericardium compresing the tricuspid valve. |
| 9 | 34 | F | ADRENAL GLAND | E. Multilocularis | 6x5cm cystic mass in the right adrenal gland. |
| 10 | 7 | M | BRAIN | E. Granulosus | 6x5cm cystic swelling in right parietal parafalcine region. |

## DISCUSSION

This series highlights the atypical distribution of hydatid disease, extending beyond its classical hepatic and pulmonary predilection to involve musculoskeletal tissue, kidney, pericardium, adrenal gland, and central nervous system. Such unusual localizations are collectively reported in less than 10% of all echinococcosis cases and frequently pose a diagnostic challenge due to their ability to mimic neoplastic or other cystic lesions.[5,6]

A common unusual site noted in this series is musculoskeletal involvement, which is considered rare, accounting for only 1–5% of hydatid cases.[7] Skeletal muscle is generally an unfavourable environment for parasite growth due to continuous contractility and high lactic acid levels, which inhibit larval development. Despite this, the cases demonstrate that when involvement occurs, cysts tend to be deep-seated, large, and occasionally multiloculated, often extending along fascial planes, as seen in the posterior thigh lesion.

Renal involvement, observed in three cases, is also uncommon, representing 2–4% of hydatid disease. The radiologic spectrum ranges from simple cystic lesions to multiloculated cysts with daughter cysts, which may mimic cystic renal neoplasms or abscesses. The variability in morphology—from simple cysts to multiloculated lesions—highlights the potential for misdiagnosis as cystic renal neoplasms, particularly in the absence of classical imaging features such as daughter cysts or calcification.

Cardiac hydatid disease is rare (0.5–2% of cases), with pericardial involvement being even less common7. The case in this series demonstrated hemodynamically relevant compression of the tricuspid valve, emphasizing that hydatid cysts at unusual sites may lead to significant organ-specific complications, beyond being incidental findings.

Similarly, adrenal hydatid cysts are extremely rare, comprising less than 1% of cases, and are often misdiagnosed as other cystic adrenal lesions such as pseudocysts or cystic neoplasms8. Our case was misdiagnosed radiologically as adrenal cortical carcinoma. The adrenal case in this series reinforces the need for considering hydatid disease in the differential diagnosis of any cystic adrenal mass in endemic regions.

Brain involvement in hydatid disease is reported in only about 1%–2% of cases and occurs disproportionately in children, usually as a solitary supratentorial cystic lesion.[9]


ISSN (O): 2687-5365; ISSN (P): 2753-6556

Across all cases, the lesions were consistently described as well-defined cystic masses, either unilocular or multiloculated. In the absence of classical features such as detached membranes, hydatid sand, or calcification, definitive staging based solely on imaging may be limited.

An important finding is the low sensitivity of hydatid serology, which was positive in only 30% of cases. This aligns with existing literature demonstrating that serologic tests for echinococcosis have variable and often reduced sensitivity in extrahepatic disease, particularly in musculoskeletal, renal, and other unusual locations. The reduced sensitivity is attributed to limited antigen leakage from intact cysts, resulting in a diminished host immune response. Furthermore, serologic performance is known to depend on cyst stage, viability, and burden, with higher sensitivity observed in active, multivesicular lesions and lower sensitivity in inactive or isolated cysts.[10,11]

## CONCLUSION

Hydatid disease should be considered in the differential diagnosis of any well-defined cystic or multiloculated lesion at unusual anatomical sites, especially in endemic areas, as failure to recognize it preoperatively may lead to misdiagnosis and potential intraoperative complications. Serology has limited sensitivity in extrahepatic disease, necessitating reliance on imaging and pathologic confirmation for accurate diagnosis.

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ISSN (O): 2687-5365; ISSN (P): 2753-6556