

**Case Report** 

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# A Complex Case of Multi-organ Hydatid Cystic Echinococcosis: Atypical Presentation and Multidisciplinary Surgical Management

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# **ABSTRACT**

This case report discusses a rare case of multi-organ hydatid cystic echinococcosis involving the liver, bilateral breasts, peritoneal and retroperitoneal cavities, mesentery, and omentum. This complexity requires a multidisciplinary surgical approach to manage the atypical spread of the disease. The challenges in diagnosis, specific surgical techniques, and postoperative management strategies were explored. The patient's successful recovery during the two-year follow-up highlights the importance of thorough diagnosis, surgical planning, and collaboration among specialists, offering valuable insights into managing extensive hydatid disease.

#### **INTRODUCTION**

Hydatid disease or cystic echinococcosis is caused by *Echinococcus granulosus*. It typically affects the liver and lungs but can also involve multiple organs. Humans usually contract the disease through contact with infected animals or ingestion of contaminated food. The liver is often the first organ affected as it serves as a major site where the parasite initially establishes itself; however, other organs, such as the spleen, kidneys, and muscles, can also be involved <sup>[1,2]</sup>.

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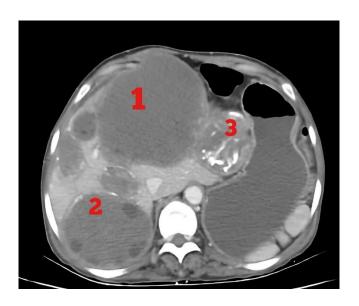
The disease is more common in Central Asia, the Middle East, and parts of South America<sup>[1]</sup>. Multiorgan involvement, such as in this case, is rare and presents challenges in diagnosis and treatment. This report highlights these challenges and the need for a coordinated, multidisciplinary approach.

#### **CASE PRESENTATION**

A 50-year-old Afghan woman presented with vague abdominal pain, palpable masses in the abdomen and breasts, and chronic fatigue. Examination revealed tenderness in the right upper and lower abdomen, with hepatomegaly and masses in both breasts. Laboratory tests, including liver function and serological tests for hydatid disease, were performed. Imaging, including ultrasound and CT scans, showed multiple cystic lesions in the liver, breasts, peritoneal and retroperitoneal cavities, omentum, mesentery, and soft tissue of the buttocks, consistent with hydatid disease [3]. The imaging also revealed cysts corresponding to different CE classes, ranging from simple cysts (Class 1) to calcified and complex cysts (Class 3 and 4) [3].

# **Diagnostic Approach**

The diagnosis was based on imaging and serology. Ultrasound was initially used because of its accessibility and sensitivity, especially for liver and peritoneal cysts. CT imaging provides a comprehensive view of cyst size, number, and anatomical details, which are essential for planning surgery [4]. In this case, CT was preferred over MRI because of its better availability and suitability for surgical planning. Serological tests, such as ELISA, have also been performed, although their sensitivity is limited, especially in cases with extrahepatic cysts [1,4].



This axial CT scan image demonstrates multiple well-defined cystic lesions involving the liver and retroperitoneal space, consistent with hydatid disease. The cysts showed a multilocular appearance with both daughter cysts and a laminated membrane, which are typical features of Echinococcus granulosus infection. The presence of daughter cysts provides a characteristic "wheel spoke" pattern within the main cyst,

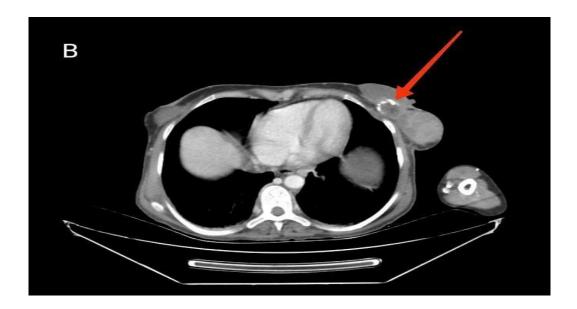
aiding the confirmation of the diagnosis. No significant calcification or rupture was observed in the cyst walls. Imaging findings are critical for the assessment of surgical planning and the extent of organ involvement.

Cyst 1 is classified as Class 1 due to its simple appearance without internal structures.

Cyst 2 is classified as Class 2, characterized by the presence of daughter cysts.

Cyst 3 is classified as Class 3 and 4 because of the presence of calcifications and its location anterior to the stomach.

Figure 1: CT scan showing Multiorgan Hydatid Cystic Lesions.



This Axial CT scan image reveals the presence of a cystic lesion in the left breast (indicated by the red arrow), consistent with hydatid disease. The cyst exhibited well-defined borders and internal septations, characteristic of daughter cyst formation. Breast involvement is an uncommon manifestation of hydatid disease, making this an unusual presentation. Identifying such cystic lesions is crucial for surgical planning and management to prevent complications. Clear delineation of the cyst from the surrounding breast tissue helps in determining the surgical approach for complete and intact removal.

Figure 2: CT scan Demonstrating Bilateral Breast Hydatid Cysts.



This Axial CT scan demonstrates a well-defined cystic lesion in the retroperitoneal space. The lesion was consistent with hydatid disease, displaying typical features of a hydatid cyst, including a smooth outer wall and internal fluid density similar to that of cerebrospinal fluid. No calcification or rupture was observed. Retroperitoneal involvement is uncommon in hydatid disease, making it an atypical manifestation that requires careful surgical planning to avoid complications. Imaging findings are crucial for determining the extent of involvement and appropriate surgical approach.

Figure 3: CT scan showing Retroperitoneal Hydatid Cystic Lesion.

# **Management and Treatment**

Surgical removal is the cornerstone of treatment. The main goal is to drain the cysts with sufficient care to prevent the possibility of spillage, thereby minimizing the risk of recurrence. A multidisciplinary team performed the staged surgeries, including hepatotomy and cyst removal from various affected organs. Hypertonic saline-saturated gauze was applied around the cysts to reduce the chance of spillage and infection during the procedure [4]. Intraoperative ultrasound was used to localize deeper cysts, which was particularly important given the involvement of breast cysts.

#### **Surgical Intervention**

A midline laparotomy was performed to access the abdominal cavity for cyst removal from multiple organs. The procedure involved draining the cysts, ensuring no residual daughter cysts or hydatid sand remained, and resecting the cyst walls that did not interfere with the main viscera. The laminar layer of the hydatid cysts was removed completely, while the fibrotic capsule was resected unless it was attached to a specific organ. Hypertonic saline-saturated gauze was applied around the cysts to reduce the chance of spillage and infection during the procedure. Intraoperative ultrasound was used to help localize deeper cysts and ensure complete removal <sup>[5]</sup>. The open surgical approach was chosen over laparoscopic surgery because of the complexity and extensive involvement of multiple organs <sup>[5]</sup>.

#### **Post-operative Care**

Albendazole therapy was initiated preoperatively and continued postoperatively at a dose of 12–15 mg/kg/day for three months to reduce the risk of recurrence <sup>[5]</sup>. Liver function was monitored regularly to detect signs of hepatotoxicity. Collaboration among specialists in radiology, infectious diseases, and surgery is critical for comprehensive treatment.

# Follow-up and Long-Term Management

Follow-up included regular imaging and monthly liver function tests to monitor recurrence or new cysts. Albendazole was continued for three months, with careful monitoring for hepatotoxicity. The patient was also educated on recognizing the symptoms of recurrence and on preventive measures, such as maintaining hygiene and

avoiding contact with infected sources. During the two-year follow-up, the patient remained symptom-free.

#### LIMITATIONS IN DIAGNOSIS AND TREATMENT

# **Diagnostic Limitations**

**Serological Tests:** Serological tests such as ELISA and indirect hemagglutination have limited sensitivity and specificity. False positive or false negative results can occur, especially in cases with extrahepatic involvement. Cross-reactivity with other parasitic infections can also affect accuracy and limit reliability [1].

**Imaging Challenges:** Imaging techniques such as ultrasound and CT scans can have difficulty distinguishing hydatid cysts from other cystic lesions, especially in unusual locations, such as the breasts or gluteal regions. Interpretation of imaging results can vary depending on the expertise [3].

#### **Treatment Limitations**

**Surgical Complications:** Surgical treatment carries risks including cyst rupture and potential spillage of hydatid fluid, which can lead to secondary echinococcosis or anaphylactic shock. Complete removal is challenging when cysts are in inaccessible areas or near critical structures, increasing the risk of complications <sup>[4,5]</sup>.

**Recurrence risk:** Recurrence is a common limitation in treating HD, even with surgical removal and anthelmintic therapy. Factors, such as incomplete removal, cyst rupture during surgery, and microscopic residual cysts, contribute to recurrence. Albendazole is used to mitigate this risk but is not always effective, and prolonged use raises concerns about hepatotoxicity [4].

**Medical Treatment Challenges:** Anthelmintic drugs, such as albendazole, have limited effectiveness when used alone, particularly for larger or calcified cysts. Prolonged use can also cause side effects such as gastrointestinal issues and liver toxicity, which limit their use, particularly in patients with pre-existing liver conditions <sup>[4,5]</sup>.

## **DISCUSSION**

Managing multiorgan hydatid disease is challenging because of the risk of rupture, systemic dissemination, and recurrence. Combining surgery with albendazole is effective in reducing these risks <sup>[5]</sup>. A multidisciplinary approach involving surgeons, radiologists, and infectious disease experts was crucial for treating the extensive involvement observed in this case.

The involvement of unusual sites, such as the bilateral breasts and retroperitoneal cavity, adds complexity to surgical planning, particularly regarding cosmetic outcomes. Imaging is critical for determining the extent of the disease and

surgical planning. Intraoperative ultrasound helped to ensure that all cysts were located and removed without complications.

The use of hypertonic saline as a scolicidal agent minimizes the risk of recurrence. Although laparoscopic techniques are useful for other hydatid surgeries, they are not suitable owing to their complexity and multi-organ involvement [5]

Recurrence is a significant concern and is often caused by residual microscopic cysts or intraoperative spillage. In this case, albendazole therapy effectively reduced the viability of protoscolices and the risk of recurrence, although prolonged use carries risks <sup>[4]</sup>. Therefore, close monitoring is required to avoid adverse effects, particularly hepatotoxicity.

The patient's favorable outcome, with no recurrence after two years of follow-up, emphasizes the importance of individualized treatment and multidisciplinary management strategies. A comprehensive follow-up plan, including imaging and patient education, played a key role in ensuring long-term success.

This case highlights the importance of early diagnosis, thorough imaging, careful surgical planning, and multidisciplinary collaboration for managing complex multiorgan hydatid disease. Future research should explore novel imaging modalities, such as MRI or PET, for better detection and improved outcomes, as well as minimally invasive techniques for complex cases.

# **CONCLUSION**

This case emphasizes the importance of a multidisciplinary approach to managing complex cases of multiorgan hydatid disease. Early diagnosis, precise imaging, and comprehensive treatment planning, including surgical removal and adjuvant albendazole therapy, are crucial for achieving favorable outcomes. Close collaboration among specialists is essential for the successful recovery of the patient. Future research should investigate novel imaging and surgical techniques for improved management of complex hydatid cases.

# **REFERENCES**

- Wen H, Vuitton L, Tuxun T, et al. Echinococcosis: Advances in the 21st Century. Clin Microbiol Rev. 2019;32(2). doi:10.1128/CMR.00075-18.
- 2. Clinical Features and Imaging of Cystic Hepatic Lesions: An updated Review of cystic hepatic lesions. Available from: https://www.ncbi.nlm.nih.gov/pmc/articles/PMC6997642/.
- 3. Zalaquett E, Menias C, Garrido F, et al. Imaging of Hydatid Disease with a Focus on Extrahepatic Involvement. Radiographics. 2017;37(3):901-923. doi:10.1148/rg.2017160172.

4.	Safioleas M, Misiakos EP, Kakisis J, et al. Surgical Treatment of Human Echinococcosis. Int Surg. 2000;85(4):358-
	365. Available from: https://pubmed.ncbi.nlm.nih.gov/11589608/.
5.	Vuruskan E, Ercil H, Anil H, et al. Comparison of Laparoscopic and Open Surgery in the Treatment of Renal Hydatid
	Cysts. J Laparoendosc Adv Surg Tech A. 2022;32(4):427-431. doi:10.1089/lap.2021.0031.