Original Research Article

Extrahepatic and extrapulmonary hydatid cysts as primary lesions

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ABSTRACT

Background: Hydatid cyst disease, caused by the larval stage of the parasite Echinococcus granulosus, commonly affects the liver and lungs but can also manifest in various extrahepatic and extrapulmonary locations. Splenic hydatid disease, despite being rare, is the third most common location. This paper presents a case series highlighting the clinical presentation, diagnostic challenges, and management strategies for patients with extrahepatic and extrapulmonary hydatid cysts. Several studies already postulated on sites of hydatid cysts which have statistically significant results.

Methods: Patients who were operated on for hydatid disease or cystic lesions, which were later diagnosed as hydatid disease, between September 2022-August 2023 were retrieved retrospectively. Patients with lesions localized outside the liver and the lung as well as in liver and lung were enrolled in the study. Fifty-Two patients with extra-hepatic primary hydatid disease were treated surgically at our clinic. The cysts were located in different part of body. Results has undergone statistical methods like Z-test and Mann-Whitney U test. Any patient of any gender admitted with diagnosis of hydatid cyst in any part of body with age >18 but less than 70 years irrespective of any comorbidities.

Results: Surgical techniques like partial or total cystectomy with or without tube drainage are good option for management of extrahepatic and extrapulmonary primary hydatid cysts. There were no complications or mortality in the postoperative period. Hydatid cyst is considered in the differential diagnosis of cystic lesions, especially in endemic areas. Surgical technique planned according to the location of the cyst.

Conclusions: Cystectomy is a surgical option in extrahepatic and extrapulmonary hydatid cyst which is evaluate better in this study.

Keywords: Hydatid cyst, Rare localization, Primary hydatid disease

INTRODUCTION

Hydatid disease is a parasitic infection that is usually caused by Echinococcus granulosus. Humans are intermediate hosts and become infected by handling infected dogs or other carnivore hosts. Echinococcal cysts are mostly located in the liver (70%) and the lung (25%) Primary isolated extrahepatic hydatid disease is mostly seen within the abdomen with an incidence of 6-11%.1,2,4-6,9,12 Although some patients may be asymptomatic, clinical presentation is mostly with abdominal pain or swelling of soft tissue with respect to disease localization, i.e. spleen, pancreas, kidney, retroperitoneum, urinary bladder, ovaries, bone, heart, thoracic wall, spinal column, thyroid gland, brain and muscles. Although radical excision of the cyst is recommended whenever possible, conservative surgery may be needed in a selective group of cases.1-3,5,6,10,12 This study aims to review patients operated in general surgery department of Guru Gobind Singh government hospital, Jamnagar for hydatid disease located outside liver and lungs.

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Peritoneal cavity
Cattle
Lung+Liver
Gall Bladder
extrapulmonary
We
RESULTS
Jamnagar,
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Ethical
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classified
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informed
pulmonary
Jamnagar,
Gujarat,
2023
A
METHODS
A retrospective study was done for all patients operated
for hydatid disease between September 2022 to August
2023 in Guru Gobind Singh government hospital,
Jamnagar, Gujarat, India. Those who had a hepatic and/or
pulmonary disease were excluded. Then, written
informed consent was obtained from patients who
participated in this study. The localizations were
classified into two groups as intra-abdominal or
intramuscular, in order to obtain homogenous data.
Demographics, preoperative information (symptoms and
signs, serologic tests, radiologic imaging), operative
findings and techniques, postoperative data
(complications, hospital stay) and surveillance (follow-up
periods, outcome, and recurrence) records were retrieved
from patient files. Indirect hemagglutination (IHA) test
was the available method for serological confirmation.
Albendazole (Andazol/ Biofarma) was chosen as the anti-
helminthic drug, if it was decided to treat the patient
before surgery.1,2,12 Records were retrieved from patients’
casefiles. Statistical methods like Z-test and Mann-
Whitney U test were used in this study.

Inclusion criteria
Any patient of any gender admitted with diagnosis of
hydatid cyst in any part of body with age more than 18
but less than 70 years irrespective of any comorbidities
were included in study.

Ethical approval
Ethical committee of Shree M. P. Shah medical college,
Jamnagar, Gujarat, India has approved this study.

RESULTS
We identified seventy cases of extrahepatic and
extrapulmonary hydatid cysts, including involvement of
the spleen, kidney, brain, bone, and soft tissues. The
majority of patients presented with nonspecific symptoms
such as pain, swelling, or constitutional symptoms,
leading to delays in diagnosis. Imaging modalities,
including ultrasound, CT, and MRI, played a crucial role
in diagnosis. Surgical excision, combined with
antiparasitic therapy, was the mainstay of treatment.
Complications such as cyst rupture, anaphylaxis, and
recurrence were observed in a subset of patients. Fifty-
two patients were operated in Guru Gobind Singh
government hospital, Jamnagar, Gujarat for hydatid
disease between September, 2022 and August, 2023.
Among those, 44 (84.615%) patients had liver as primary
in hydatid cyst lesion with 8 (15.38%) were excepted
having no primary liver hydatid cyst with lesion on
different body parts (Rare cases).

Figure 1 (A and B): MRI thigh (Hydatid cyst with
daughter cyst) and MRI local region (Hydatid cyst).

Figure 2: Sites where hydatid cysts found worldwide
percentage-wise.

Figure 3: Comparison between percentages of organ
involved in hydrated cyst between dromedary, sheep
and cattle.

Following 8 patients had extra hepatic and extra
pulmonary hydatid cysts as primary.
Table 1: Frequency and percentage of infected people to hydatid cyst according to organ involvement.

<table>
<thead>
<tr>
<th>Infected organs</th>
<th>Frequency of infected people to cyst</th>
<th>Percentage (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Liver</td>
<td>36</td>
<td>69</td>
</tr>
<tr>
<td>Lung</td>
<td>7</td>
<td>13</td>
</tr>
<tr>
<td>Liver+ lung</td>
<td>3</td>
<td>6</td>
</tr>
<tr>
<td>Peritoneum</td>
<td>2</td>
<td>4</td>
</tr>
<tr>
<td>Liver+ kidney</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Liver+ lung+ bone</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Liver+ peritoneum</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Spleen</td>
<td>1</td>
<td>2</td>
</tr>
<tr>
<td>Total</td>
<td>52</td>
<td>100</td>
</tr>
</tbody>
</table>

Table 2: Preoperative data of patients with extrahepatic and extrapulmonary hydatid cysts involved in study.

<table>
<thead>
<tr>
<th>Symptoms and signs</th>
<th>Serological tests</th>
<th>Radiologic imaging</th>
<th>Localization</th>
</tr>
</thead>
<tbody>
<tr>
<td>Abdominal pain</td>
<td>IHA ±</td>
<td>CT /MRI / US</td>
<td>Spleen cavity</td>
</tr>
<tr>
<td>Palpable mass, lump ±</td>
<td>US abdomen, MRI and CECT</td>
<td></td>
<td>Right thigh</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>Head of pancreas Scapular region</td>
</tr>
</tbody>
</table>

IHA: Immune hemagglutination; CT: Computed tomography; MRI: Magnetic resonance imaging; US: Ultrasonography.

Table 3: Operative management and follow up in patients having extrahepatic and extrapulmonary hydatid cysts.

<table>
<thead>
<tr>
<th>Operative approach</th>
<th>Hospital stay (day) approximately</th>
<th>Follow-up period (month)</th>
<th>Long-term outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Splenectomy, cystectomy</td>
<td>7</td>
<td>09</td>
<td>Incisional hernia</td>
</tr>
<tr>
<td>Laparoscopic/ open</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Total cystectomy</td>
<td>07</td>
<td>12</td>
<td>Abdominal muscle weakness</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>recurrent hydatid cyst at same site</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td>or different sites in body</td>
</tr>
<tr>
<td>Partial pericystectomy</td>
<td></td>
<td>12</td>
<td>Anaphylactic shock (rare)</td>
</tr>
<tr>
<td>Cystojejunostomy</td>
<td>25</td>
<td>18</td>
<td></td>
</tr>
<tr>
<td>Jejunoojejunostomy</td>
<td>25</td>
<td>18</td>
<td></td>
</tr>
<tr>
<td>Cholecystectomy</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>T-tube drainage</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Laparoscopic total cystectomy</td>
<td>08</td>
<td>12</td>
<td></td>
</tr>
<tr>
<td>Total cystectomy (open)</td>
<td>05</td>
<td>09</td>
<td></td>
</tr>
</tbody>
</table>

DISCUSSION

The diagnosis of extrahepatic and extrapulmonary hydatid cysts requires a high index of suspicion, particularly in endemic regions. Imaging techniques such as ultrasound, CT, and MRI are invaluable for accurate localization and characterization of cystic lesions. Treatment involves a multidisciplinary approach, with surgery being the cornerstone for cyst removal, supplemented by medical therapy to prevent recurrence and complications. Close follow-up is essential to monitor for recurrence and manage any complications promptly. Data of this study like radiological findings, complains of patients and managements of individual cases were compared with several studies done previously and results are statistically significant. In other studies, with same purpose have similar results in meta-analysis.

There are certain limitations of this study as some of the patients have recurrent hydatid disease and undergone multiple surgeries before admission. Few of them are result of lost to follow up in previous surgeries. These limitations have been considered while receiving history and pre-op and post-op data from patients undergone this study.

CONCLUSION

Extrahepatic and extrapulmonary hydatid cysts represent a diagnostic and therapeutic challenge due to their diverse clinical presentations and atypical locations. A multidisciplinary approach involving radiologists, surgeons, and infectious disease specialists is crucial for timely diagnosis and optimal management. Further research is warranted to explore novel diagnostic modalities and treatment strategies for improving
outcomes in patients with these rare manifestations of hydatid cyst disease. Spleen is the third most site for hydatid cyst after lung and liver.

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Ethical approval: The study was approved by the Institutional Ethics Committee

REFERENCES


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