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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 Gobindsingh government hospital, Jamnagar for hydatid disease located outside liver and lungs.

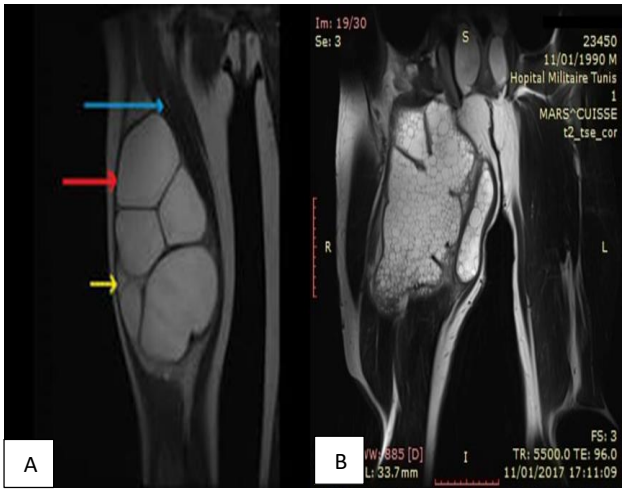

Figure 1 (A and B): MRI thigh (Hydatid cyst with daughter cyst) and MRI local region (Hydatid cyst).

METHODS

A retrospective study was done for all patients operated for hydatid disease between September 2022 to August 2023 in Guru Gobindsingh 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 Gobindsingh 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).

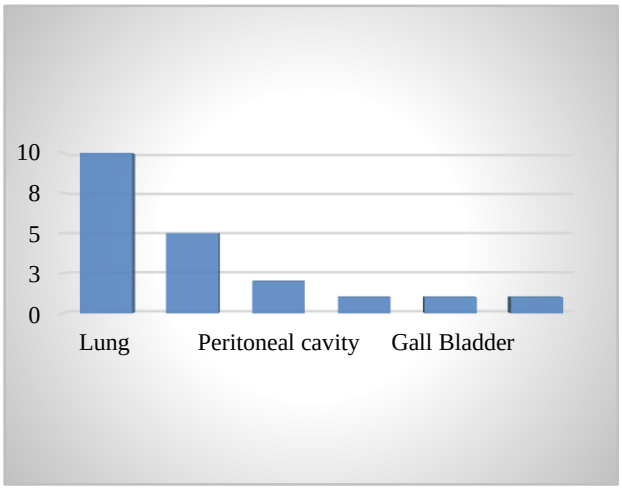

Figure 2: Sites where hydatid cysts found worldwide percentage-wise.

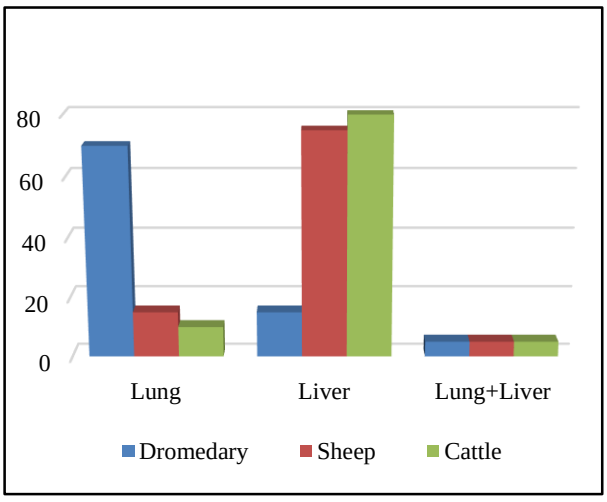

Figure 3: Comparison between percentages of organ involved in hydrated cyst between dromedary, sheep and cattle.

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.

Infected organs	Frequency of infected people to cyst	Percentage (%)
Liver	36	69
Lung	7	13
Liver+ lung	3	6
Peritoneum	2	4
Liver+ kidney	1	2
Liver+ lung+ bone	1	2
Liver+ peritoneum	1	2
Spleen	1	2
Total	52	100

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

Symptoms and signs	Serological tests	Radiologic imaging	Localization
Abdominal pain			Spleen
	IHA ±	CT /MRI / US	Peritoneal cavity
Palpable mass, lump ±		US abdomen, MRI and CECT	Right thigh
			Head of pancreas Scapular region

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.

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

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.[1] Treatment involves a multidisciplinary approach, with surgery being the cornerstone for cyst removal, supplemented by medical therapy to prevent recurrence and complications.[2] 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.[1,2,8,12]

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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Conflict of interest: None declared
Ethical approval: The study was approved by the Institutional Ethics Committee

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