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Case Report

A Hepatic hydatid cyst: four case reports

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ABSTRACT

We report four cases who were referred to Dr. D. Y. Patil Medical Hospital, Pune suffering from abdominal pain and gastrointestinal manifestations. The patients' history was unremarkable, except that they had contact with dogs and live in rural communities. Laboratory findings showed peripheral blood eosinophilia, leukocytosis, and elevated liver enzymes. Ultrasonography showed well-circumscribed cystic masses in the liver. Diagnosis of hydatid cysts was confirmed by computed tomography (CT). Surgical treatment was performed and all patients recovered well. The results of these cases support the nation that CT scan can led to increased clarity, regarding surgical management, because of discordance between radiographic and laboratory findings.

Keywords: Abdominal pain, CT, Hydatid cysts

INTRODUCTION

Human cystic echinococcosis (CE)/hydatidosis is a dog-borne zoonoses caused by infection with the larval stage of the dwarf tapeworm of the genus *Echinococcus*. *E. gran-ulosus* and *E. multilocularis* are the most important members of the genus in respect of their public health importance and their geographical distribution.[1] The life cycle of *Echinococcus* is indirect and involves two hosts, one definitive carnivore host and the other intermediate herbivore host.[2] The problem arises when humans act as an accidental intermediate host and ingest viable onco-sphere containing eggs, which have been shed in the faeces of the definitive host. The oncospheres invade the intestines, enter the vasculature and develop into hydatid cysts in any organ or tissue, where a variety of symptoms can be produced. However, the liver acts as the first filter for hydatid larvae, making it the most commonly affected organ followed by lung.[2]

The diagnosis of CE is based on the patient's history, clinical findings, haematological and serum biochemical profiles, and serological testing, which may be negative in 10% to 20% of cases.[3] Efforts to improve diagnostic accuracy have led to integration of a range of imaging techniques into the diagnostic armamentarium.[1,4] The radical surgical removal of the cystic lesion remains the mainstay of treatment with a high success rate.[1,4] Chemotherapy, with benzimidazole compounds has also been used with some success to sterilize the cyst, decrease the chance of anaphylaxis, and reduce the complications and recurrence rate post-operatively. In recent years, a third treatment option was introduced (PAIR, puncture, aspiration, injection, and re-aspiration) and is indicated for patients who cannot undergo surgery.[5,6] CE is re-emerging as a major public health issue, with potentially life-threatening complications.[7]

CASE REPORT

The four patients seen and evaluated in the Department of Surgery, Dr. D. Y. Patil Medical Hospital, Pune between April 2015 and September 2017 for complaints of abdominal pain and gastrointestinal (GI) manifestations. Patients were interviewed and examined by a Surgeon. Blood samples were assessed for some haematological parameters, including red blood cell count (RBC), white blood cell count (WBC), eosinophil (%) and hemoglobin

(Hb) content. Total serum bilirubin using commercial kit (Quimica Clinica Aplicada), and (2) activity levels of alanine aminotransferase/aspartate aminotransferase (ALT/AST) and alkaline phosphatase (ALKP), markers of hepatocellular damage according to Reitman-Frankel colorimetric transaminase procedure. The serum samples were tested by the echinococcosis hemagglutination test (IHA) Fumouze® (Laboratories Fumouze, Levallois-Perret,) and ELISA (F-Form, Maxisorp, Nunc, Roskilde, Denemark). Pre-operative diagnosis of hepatic cystic lesions was made ultrasonically and confirmed by a computer tomography (CT) scan. All four patients were successfully managed surgically and all were asymptomatic at the two years follow-up (Table 1).

Table 1: Pertinent details of hepatic hydatid cases.

	Case 1	Case 2	Case 3	Case 4
Gender	**Female**	**Male**	**Male**	**Male**
Age (years)	22	11	37	43
Occupation	Student	Farmer	Farmer	Shepherd
Residence	Suburban	Rural	Rural	Rural
Dog contact	No	Yes	Yes	Yes
Symptoms	Abdominal pain, nausea	Indigestion, easy fatigability	Generalized abdominal pain, nausea, vomiting, diarrhoea	Pain in right, hypochondrium, nausea, vomiting, weight loss
Physical examination	Hepatomegaly	Normal	Normal	Hepatomegaly
Hemoglobin (g/dl)	12	13.2	14	14.1
RBCs (106/ml)	3.9	4.4	4.8	4.5
WBCs (103/ml)	8.4	12.7	6.6	19.2
Eosinophils (%)	8	18	4	17
Bilirubin (mg/dl)	1.7	2.2	0.9	1.2
ALT (U/ml)	58	34	22	41
AST (U/ml)	74	61	34	36
ALKP (U/L)	18	22	16	11
Serology (test)				
IHA	Positive	Negative	Positive	Positive
Maximal diameter (cm)	6	14	5	6
Cyst type	III	I	I	I
Concurrent locations	Spleen	--	--	--
Surgical Treatment	Complete pericystectomy	Complete pericystectomy	Complete pericystectomy	Complete pericystectomy
Postoperative complications	--	--	--	--

A provisional diagnosis of hydatid disease was made based on the clinical manifestations, haematological and biochemical parameters, and serological tests. The diagnosis was confirmed by radiological imaging. The important demographics and clinical features, laboratory findings and therapeutic intervention in these patients are summarized in Table 1. The mean age of the patients was 28.3 (range 11-43) years. Of the four patients, three were male and one was female. Three of the four cases had frequent contact with dogs and farm animals and live in agri-cultural sheep-grazing area which is in line with the general epidemiology of hydatidosis. All patients were non-alcoholic and non-smoker. Presenting signs and symptoms for the 4 patients are listed in Table 1. All of them had intermittent colicky pain associated with nausea and vomiting. Except one patient (case one) who has a history of acute symptoms lasting 1-2 months, their mild symptoms began more than one year ago. Haematological studies revealed leukocytosis in all cases and eosinophilia in three cases. RBC count and Hb content were within normal range. Total serum bilirubin was elevated in three cases and was within normal range in one case (case three).

The biochemical serum level of ALT was significantly raised in one patient (case one) and the AST level was increased in cases one and two. Analysis of the liver enzyme ALKP revealed abnormal increase in three cases and normal level in case four. Serologic echinococcal tests are valuable when they are positive, but one of the four hepatic hydatidosis cases gives a false negative. Although serology tests like ELISA and IHA can help

make the diagnosis, complete reliance on them is not recommended.

Radiographic evaluation confirmed the variable nature of the clinical hydatid disease that has been previously reported.8 Hepatic radiograph appearance was variable and revealed abnormal findings (Figure 1). In case one (Figure 1a) liver had a relatively large size, normal echopattern but, with two well defined sonolucent cysts with thin internal septae. Liver in case two (Figure 1b) showed average size with a well-defined hypodense non-enhancing hydatid cystic lesion and is seen affecting segment V and VI. It also compressed the gall bladder, displaced the patent nondilated portal vein (PV). No other focal lesions and no dilatation of the intra or extra-hepatic bile ducts were noticed. Liver in case three (Figure 1c) had a large well-defined hypodense hydatid cyst involving the postro-superior segment of the right lobe (segment VII). The liver of case four (Figure 1d) was enlarged with smooth contour and uniform enhancing parenchyma with normal patent PV. Multiple variable-sized cystic lesions are seen scattered in both hepatic lobes. Further investigations ruled out coexistent hydatid disease in other organs except spleen in case one.

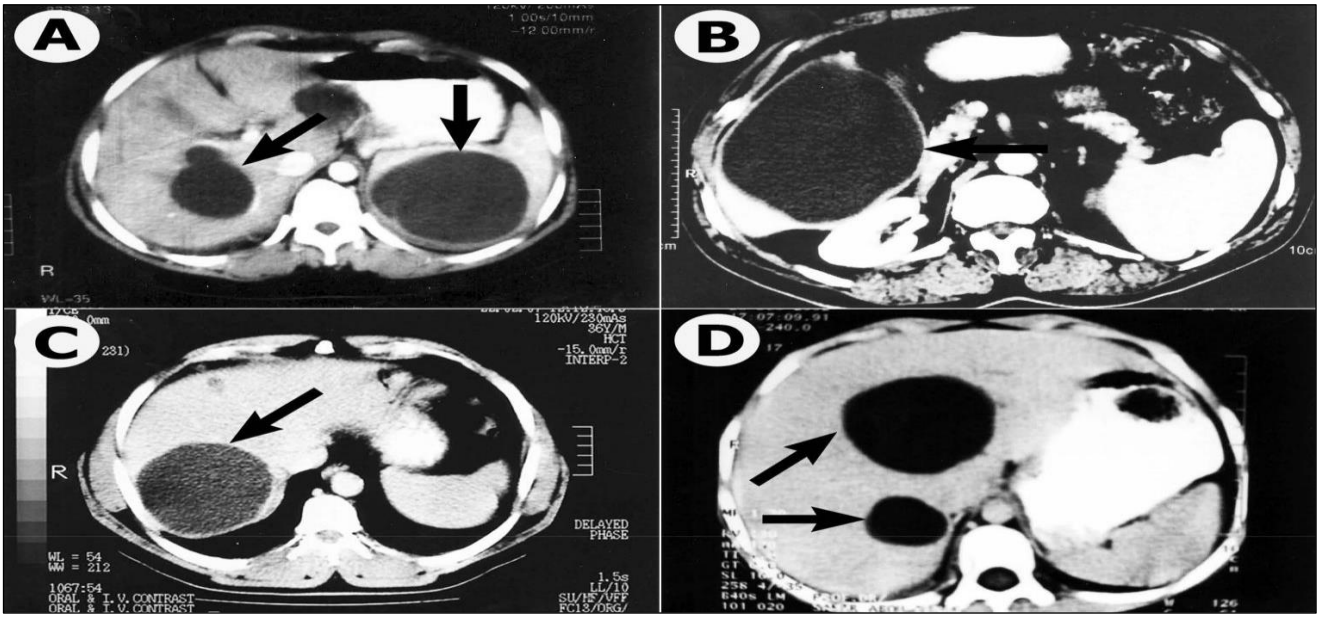

Figure 1a, 1b, 1c and 1d: Cystic hydatid lesions. Figures a, b, c, and d refer to case one to four, respectively; Arrows indicate the hydatid cyst.

DISCUSSION

From a clinical point of view, clinical manifestations of hydatidosis in humans are variable, most patients seem to tolerate the infection for extended periods without any symptomatology, or they may suddenly show dramatic and acute symptoms.[8] All four patients made an excellent recovery following surgical excision of the entire cystic masses. Adjunctive Albendazole chemotherapy 10 mg/kg/day was prescribed for 4-6 weeks before surgery to sterilize the cyst and for 2 months post-operatively to reduce the recurrence rate. At the 24-month follow-up after surgery, the patient remained free of symptoms and continued to do well. This result is consistent with other studies that found that CT scan has become an extremely useful and valuable diagnostic tool in the management of patients with hydatidosis.[1,9] Accuracy would have been enhanced by concomitant magnetic resonance imaging (MRI). But, MRI was not performed in this study, though we recognize that these might have been helpful.[10]

As frequently as it occurs, hydatidosis can sometimes be a diagnostic quandary. With the concern that, left untreated, even solitary hydatid cyst in critical organ(s) will likely progress to grave and life-threatening illness, surgeons have historically coped with diagnostic uncertainty by accepting high negative surgical intervention rates as an alternative to risking disease progression. Taken haematological, biochemical, serological and imaging findings into account, some recommendations for more accurate and sensitive detection of clinical cases, regarding surgical management, can be made:

- Diagnosis is best accomplished by a combination of imaging, serologic, and laboratory investigations

- Radiographic examination must be considered for patients in whom hydatidosis is a potential diagnosis
- Patients with equivocal history, physical findings, or laboratory results should not be considered negative unless there are no obvious positive radiographic findings.

Despite major advances in our understanding and treatment of hydatidosis over the past two decades, control of this zoonotic disease remains a challenging endeavor. Fortunately, because of the high risk of infection, the high morbidity rate and the unpredictable outcome of hydatid infection, there is an increasing realization in international health agencies that hydatidosis is an important disease that causes life-threatening morbidity. Like many diseases, prevention is the key to control, and we know that simple changes in social habits and hygiene can prevent infection and disease in humans. Also, chemoprophylaxis is an effective measure to protect humans living in endemic or hyperendemic areas of hydatid infection.

CONCLUSION

In conclusion, we stress the importance of maintaining a high level of clinical awareness that hepatic hydatidosis continues to be a clinical problem especially in endemic regions of the world and travelers to these areas should be made aware of the need to take on extra-measures to avoid hydatid disease. In endemic areas like Egypt any cystic enlargement of soft tissue should raise the suspicion of clinicians of hydatid disease. Serologic tests, ultrasonography, and CT should be performed before any invasive procedure.

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