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UNCOMMON SITES OF HYDATID DISEASE

Pathology

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ABSTRACT

BACKGROUND: Diagnostic dilemma with hydatid cysts at unusual sites can lead to complications as sometimes these may present as acute surgical emergency or chronic illness leading to morbidity. The aim of the study is to highlight the fact that this disease should be suspected in cystic lesions involving any organ of the body, especially in endemic areas like India.

MATERIAL & METHODS: It is a descriptive study of duration of one year in which all cases of hydatid cysts signed out in the Department of Pathology from June 2017 to May 2018 were included. 25 cases were identified in a period of one year. Haematoxylin and eosin stains sections of 5 micrometres thickness were re-examined in all cases to confirm the diagnosis.

RESULTS: Out of 25 patients, 23 (92%) had single organ involvement and 2 (8%) had more than one organ involved. Liver was involved in 14 patients (56%), either solitary or in association with other organs. Lung involvement was seen in 7 patients (28%). 4 patients (16%) had cyst at extrahepatic site (ovary, pelvis, kidney, pancreas).

CONCLUSION: A high index of suspicion is required for pre-operative diagnosis of hydatid disease at rare and unusal sites in the body. A possibility of hydatid disease should be considered in the differential diagnosis of cystic swellings present anywhere in the body especially in patients from endemic areas.

KEYWORDS

Hydatid disease, Liver, Lungs, Extrahepatic site.

INTRODUCTION

Hydatid disease or Echinococcosis is a zoonotic disease caused by the larvae (metacestode) of the cestode species of the genus Echinococcus like *E. granulosus, E. multilocularis, E. vogeli or E. oligarthus*. It is estimated that the worldwide incidence of cystic echinococcosis is about 100,000-300,000 cases annually [1]. Increased prevalence of the parasite is found in parts of Europe, around Mediterranean region, the Russian Federation, China, Africa, Australia, and South America [2]. Hydatid disease is a significant health problem in India and has been reported in many states of which the highest prevalence is reported in Andhra Pradesh, Tamil Nadu and Jammu and Kashmir [3].

Humans acquire primary Cystic Echinococcosis by ingestion of *E. granulosus* eggs excreted by infected carnivores [4]. The cysts most frequently occur in liver (63%) followed by the lungs (25%), muscles (5%) and bones (5%). They can also be found uncommonly in the kidney, brain and spleen [5]. It is rare to diagnose hydatid cyst in the pelvis especially as a primary localization [6]. The incidence reported in the literature of pelvic hydatid cyst is 0.2-2.25% [7]. Cystic echinococcosis has a multitude of clinical presentations ranging from asymptomatic disease to acute emergencies. Intact and small-sized cysts have no specific characteristic symptoms. Their clinical manifestations depend on the organ involved; site and size of the cyst; interaction between the expanding cysts and the adjacent organ structures and complications related to cyst rupture; spread of protoscoleces; and bacterial infection [8, 9, 10, 11]. Diagnostic dilemma with hydatid cysts at unusual sites can lead to complications as sometimes these may present as acute surgical emergency or chronic illness leading to morbidity [12]. The aim of the study is to highlight the fact that this disease should be suspected in cystic lesions involving any organ of the body, especially in endemic areas like India.

MATERIAL & METHODS

It is a descriptive study of duration of one year in which all cases of hydatid cysts signed out in the Department of Pathology from June 2017 to May 2018 were included. All cases of hydatid cysts signed out in the above mentioned period were retrieved from surgical pathology files of Govt Medical College Jammu. 25 cases were identified in a period of one year. Haematoxylin and eosin stains sections of 5 micrometres thickness were re-examined in all cases to confirm the diagnosis. Criteria for diagnosis of cystic hydatid disease are visualisation of a lamellated structure of cyst wall, germinal layers, scolices and protoscolices on Haematoxylin and Eosin (H&E) stained paraffin embedded sections. Clinical features and follow up data was obtained from consult files and referring surgeons.

RESULTS

25 cases which were diagnosed as hydatid cyst were included in the present study. Majority of the patients were in the age group of 20-40 years. The mean age of patients was 37.3 years. The sex incidence showed female predominance (M: F- 1:1.3). Majority of the patients were farmers (48%), followed by housewives (36%) and students (6%). The main presenting symptom was pain in the abdomen (88%) followed by lump abdomen (68%). Leucocytosis, eosinophilia and elevated liver enzymes were common haematological abnormalities observed in the study. Out of 25 patients, 23 (92%) had single organ involvement and 2 (8%) had more than one organ involved. Liver was involved in 14 patients (56%), either solitary or in association with other organs. Lung involvement was seen in 7 patients (28%). 4 patients (16%) had cyst at extrahepatic site (ovary, pelvis, kidney, pancreas). Right lobe of the liver was involved in 10 patients (71.4%). The left lobe of the liver was found to be involved in 4 patients (28.6%). All four cases at uncommon locations (ovary, pelvis, kidney and pancreas) were of primary hydatid disease with no past history of liver involvement.

Figure 1: Picture showing gross of ovarian tissue with cyst wall.

Figure 2: Picture showing pearly white cyst wall in patient with ovarian hydatid cyst.

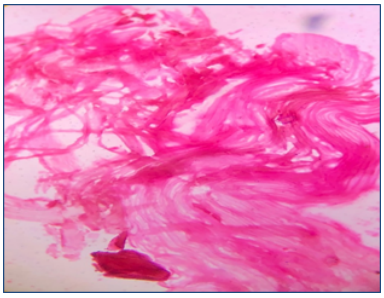

Figure 3: Photomicrograph showing lamellated membranes in hydatid cyst of ovary.

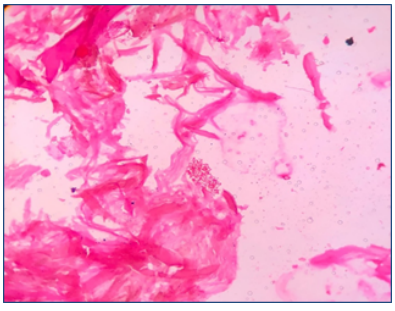

Figure 4: Photomicrograph showing cyst wall in hydatid cyst of pancreas.

DISCUSSION

Cystic echinococcosis has a wide geographical distribution and is a cause of serious concern in the endemic region due to severe disease, increase in morbidity and considerable economic loss [13, 14]. Our study revealed that of all patients of Cystic echinococcosis presenting to our hospital, this disease commonly affects patients in their second and fourth decade, is seen frequently in farmers and has a female predominance. In our study, patients with cystic echinococcosis presented commonly with pain in abdomen followed by lump in abdomen. Liver was the most commonly affected organ followed by lung. Irshadullah *et al* also reported similar distribution with the highest percentage of patients in the age group of 21-30 years (25.83%) and 20% in the fourth decade. It has been consistently seen in various studies through the years and confirmed by our study that this disease in seen in young population who are in their active years. In our study females were affected more than the male which is identical to findings of previous studies [15].

Our study established that pain and lump in the abdomen were the common presenting features of Cystic Echinococcosis. According to Balik *et al* [16] and Jacob *et al* [17] pain in abdomen has been the most common presenting symptom (74.01% and 85%, respectively). This disease is usually found in liver and lungs but no organ of body is immune. Location at unusual sites in the body can have atypical presentations and can pose a diagnostic challenge. A high index of suspicion, radiological investigations as well as histopathological examination is necessary in establishing the diagnosis of hydatid disease at unusual sites in the body. In endemic areas any patient presenting with a cystic mass, in any tissue or organ, should be considered a potential case of the hydatid disease [18]. The exact percentage of site involvement varies and the exact incidence of unusual locations is difficult to ascertain as they are only reported as case reports. In 10% cases, hydatid disease arises in the viscera; mainly in the spleen (0.9–8%) and also in kidney, bone, heart muscles and peritoneal cavity (0.5–5%) [19]. In our study four patients presented with hydatid cysts at uncommon location ie in ovary, pelvis, kidney and pancreas.

Single case of hydatid disease of the ovary was seen in our study in a multiparous woman of 40 years age with symptoms of pain abdomen and menstrual irregularities. A primary hydatid cyst of the ovary is extremely rare. Most cases occur as a result of the rupture of hepatic cysts. In the present case, a hydatid cyst was not found in the liver and there was no history of previous echinococcosis, indicating a primary ovarian cyst. Pelvic Echinococcosis symptoms are not specific and can present with abdominal pain, menstruation irregularities, infertility and urinary disturbances. Ovarian echinococcosis can mimick either polycystic disease or malignancy. The diagnostic challenge is due to the nonspecific symptomatology, along with atypical ultrasonographic finding of a solid ovarian mass [20]. The ultrasonography is an important imaging examination that allows knowing the cystic aspect of the lesion, revealing the characteristic fluctuating membranes of the multilocular cyst. CT scan confirms the diagnosis showing daughter cysts and calcifications of the cyst's wall. Serological test like indirect haemagglutination tests and ELISAs to differentiate hydatid cyst from nonparasitic cysts or abscess has sensitivity varying from 64% to 87%. Histopathological examination is required for final confirmatory diagnosis.

Our study showed a single case of hydatid cyst of kidney. Primary hydatid of the kidney is rare entity and is responsible for only 2 to 3% of all hydatid disease [21].Renal involvement could be primary or secondary. Cysts are usually unilateral and located in the upper or lower pole [22]. Ultrasonography aids in the diagnosis of hydatid cysts by demonstrating daughter cysts and hydatid sand. Removal of hydatid cyst with pericystectomy is possible in most cases. Nephrectomy is reserved for destroyed kidney.

In our study there was a case of hydatid cyst involving the pancreas. Hydatid cyst of the pancreas is rare and very difficult to distinguish from cystic neoplasm of pancreas. The clinical presentation is variable and insidious, depending on the location and the size of the hydatid cyst. On abdominal ultrasonography a cystic mass in the pancreas is seen [23]. Cysts in body and tail are best treated by resection methods whereas, for those in the head region, a cystectomy with simple drainage is a simple, quick and effective solution [24]. One patient 55 years of age in our study had hydatid cyst in the pelvis. Omental hydatid cysts often reach the pelvis and is erroneously diagnosed as pelvic cystic disease. CT and MRI are useful in the diagnosis. In our study, USG was proved diagnostic in all cases. In the study conducted by Balik *et al* (1999), ultrasonography showed diagnostic accuracy of 97.7% [15].

Diagnosis of hydatid disease is based on the patient's history, clinical findings, serum biochemical profiles, serologic tests and pathologic diagnosis [25]. The diagnosis is often difficult when hydatid cyst occurs at unusual locations as the imaging appearance varies at different sites [26].

CONCLUSION

Incidence of hydatid disease at unusual sites in India is higher than in other parts of the world. A high index of suspicion is required for pre-operative diagnosis of hydatid disease at rare and unusual sites in the body. A possibility of hydatid disease should be considered in the differential diagnosis of cystic swellings present anywhere in the body especially in patients from endemic areas. Early treatment is mandatory to avoid local and general complications which are directly related to duration of cyst [27].

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