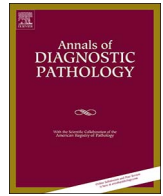




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Unusual locations of hydatid disease: A 10-year experience from a tertiary reference center in Western Turkey



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ABSTRACT

Introduction: Hydatid disease is an endemic parasitic infection caused by *Echinococcus granulosus* mostly seen in the Mediterranean countries. The most affected organ is the liver, however hydatidosis can be found anywhere in the human body.

Methods: The records of patients who were diagnosed with hydatid disease in our hospital from December 2005 to February 2016 were analyzed retrospectively. The cases were evaluated and recorded depending on their gender, age and the localization of the cysts.

Results: A total of 329 patients diagnosed over a 10-year period were included in our study. There were 202 females (61.4%) and 127 males (38.6%). The hydatid cysts were located in the liver in 257 (78.1%) patients and in unusual locations in 72 (21.9%) patients. The most common unusual site for hydatid cysts was the spleen followed by bones, central nervous system, soft tissue, the kidney and the gall bladder. Amongst these 72 patients who had hydatid cysts in unusual locations; 33 patients had concomitant liver hydatidosis, whereas 39 patients had primary involvement of unusual sites. Two patients with malignancies also had hydatid cysts in different locations.

Conclusion: Hydatid disease affects many organs in the body and therefore it can pose a major diagnostic dilemma and it may mimic other entities. In endemic areas, a differential diagnosis of hydatid disease should be considered for cystic masses in any anatomical location.

1. Introduction

Hydatid disease, also known as echinococcosis, is a parasitic infection caused by the larval forms of *Echinococcus granulosus* tapeworms. It is seen throughout the world, however it is endemic in South America, Middle East and the Mediterranean countries [1]. Turkey is amongst the countries where hydatid disease is frequently seen [2].

The parasite is found in the small intestines of the definitive hosts (most commonly dogs and foxes) and transmitted to the intermediate hosts such as sheep and humans by the ingestion of the eggs. After ingestion, the larval forms of the parasite pass to the liver via the portal vein rendering the liver the most common location for cyst formation (75%). The second most common site for the disease is the lungs (5–15%). If the larvae pass through the first filters that are the liver and the lungs or by-pass these organs using lymphatics, they can cause hematogenous and lymphatic dissemination of the disease. Therefore hydatid disease can develop in any part of the human body [3].

The hydatid cyst has a spherical shape and contains clear fluid. The cyst wall consists of three layers: (1) the outer pericyst which is a dense,

fibrous protective zone, (2) the middle acellular laminated membrane, (3) the inner germinal layer which is the only living part of the cyst. The protoscolices and the laminated membrane are produced by the germinal layer [3–4].

Hydatid disease remains as a serious health problem in Turkey. The aim of this study is to report the unusual locations of the hydatid disease on a series of patients with hydatid disease operated and diagnosed in our hospital which is located on the western part of Turkey over a period of 10 years.

2. Materials and methods

Three hundred and twenty-nine cases of hydatid disease which were diagnosed from December 2005 to February 2016 at Izmir Katip Celebi University Ataturk Training and Research Hospital's Pathology Department were analyzed retrospectively. The cases were operated after having been suspected of having hydatid disease with the imaging procedures and serology profiles. The histopathological diagnosis was made by the evaluation of surgically resected specimens both grossly

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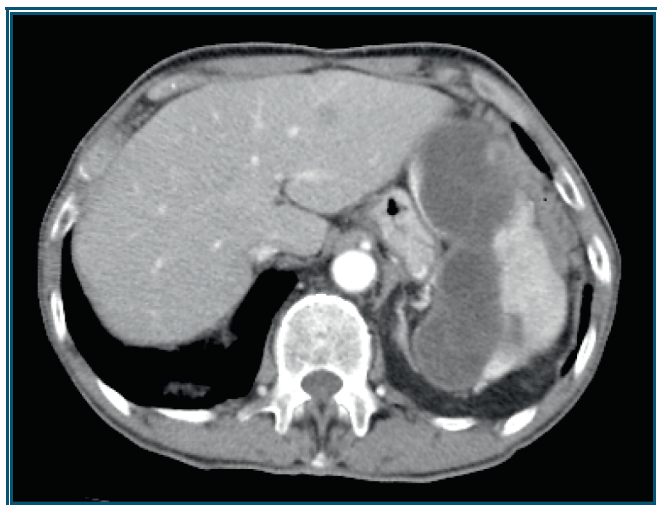


Fig. 1. Magnetic resonance imaging of a splenic hydatidosis.

and microscopically on routine hematoxylin-eosine stained sections. The age and gender of the cases and the localization of the cysts were analyzed and specified. The numbers of hepatic and extrahepatic involvement of the cases were counted and listed.

3. Results

3.1. Clinical findings

A total of 329 cases diagnosed with hydatid disease over a 10 year period were included in our study. There were 202 females (61.4%) and 127 males (38.6%). Patient age ranged from 4 to 82 years with a median of 42 years. Most of the cases presented with nonspecific symptoms such as fatigue, pain, nausea, itchiness or secondary symptoms due to the growth of the cysts including symptoms of local pressure, rupture and secondary infection. On radiological examinations, the majority of the cases raised suspicions of a cystic disease or a cystic malignancy. The cysts in the liver were easily identified by radiologists with their multicystic components, however in unusual locations other cystic diseases and malignancies with cystic structures were also included in the differential diagnosis (Fig. 1, Fig. 2).

The hydatid cysts were located in the liver in 257 (78.1%) patients and in unusual locations in 72 (21.9%) patients. Amongst these 72 patients who had hydatid cysts in unusual locations; 33 patients had concomitant liver hydatidosis, whereas 39 patients had primary involvement of unusual sites. The distribution of the unusual locations are shown in Table 1.

The most common unusual site for hydatid cysts was the spleen. There were 16 cases (4.8%) of splenic hydatidosis. Four of these patients had isolated splenic involvement. The second most common sites were the bones (11 patients, 3.3%) followed by the central nervous system (10 patients, 3%). In 7 patients bones were primarily involved. The most common osseous site was the lumbar vertebrae with 3 cases. Table 2 shows the distribution of different localizations of hydatid cysts in the musculoskeletal system. In the central nervous system 4 of the patients had cysts in the spinal medulla, five of them in intraparenchymal areas and one in the cerebellum. Other unusual locations included the kidney (5 cases, 1.5%), the gall bladder (5 cases, 1.5%), omentum (3 cases, 0.9%), peritoneum (2 cases, 0.6%), retroperitoneum (2 cases, 0.6%), common bile duct (1 case, 0.3%), pancreas (1 case, 0.3%), small bowel (1 case, 0.3%), intraorbital (1 case, 0.3%), breast (1 case, 0.3%), female adnexa (1 case, 0.3%), uterus (1 case, 0.3%), vagina (1 case, 0.3%), pericardial area (1 case, 0.3%), aortic surface (1 case, 0.3%). Two patients with malignancies (1 gastric neuroendocrine tumor and 1 endometrial adenocarcinoma) also had hydatid cysts in different

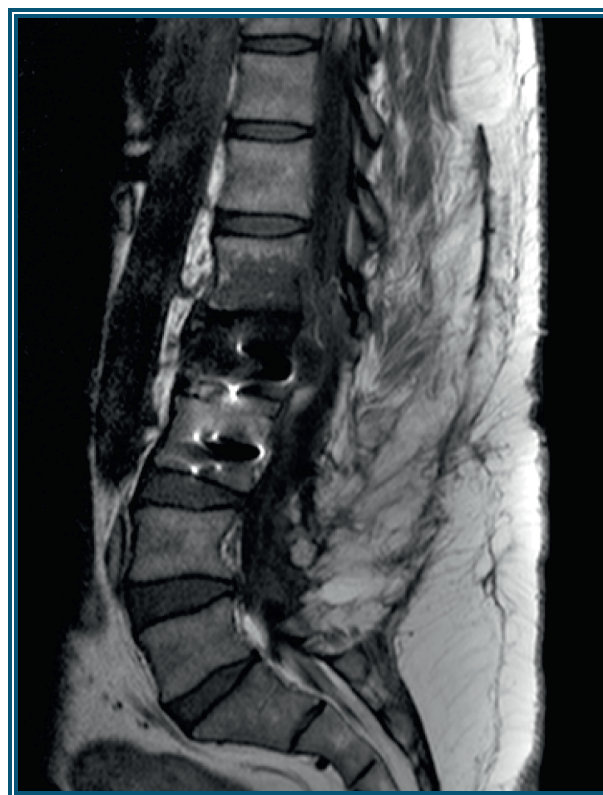


Fig. 2. Magnetic resonance imaging of hydatid disease in L4 and L5 vertebrae.

Table 1
The distribution of unusual locations of hydatid disease.

Localization	Number of cases	Concomitant liver hydatidosis	Overall percentage (%)
Spleen	16	12	4.8%
Bone	11	3	3.3%
Central nervous system	10	2	3%
Soft tissue	7	2	2.1%
Gall bladder	5	5	1.5%
Kidney	5	2	1.5%
Omentum	3	3	0.9%
Peritoneum	2	2	0.6%
Retroperitoneum	2	–	0.6%
Common bile duct	1	–	0.3%
Intraorbital	1	–	0.3%
Breast	1	–	0.3%
Pancreas	1	–	0.3%
Pericardial area	1	–	0.3%
Bowel	1	–	0.3%
Female adnexa	1	–	0.3%
Uterus	1	–	0.3%
Vagina	1	–	0.3%
Aortic surface	1	–	0.3%
Stomach + pelvis	1	–	0.3%
Total	72	33	21.9%

locations (omentum, pelvic region).

3.2. Pathological findings

The pathological specimens consisted of cystectomies and partial or radical organ resections. The gross examination of the cases revealed whitish, thin-walled, intact cystic structures also known as daughter vesicles. The cysts ranged in size and were filled with colorless fluid. Also, white gelatinous membranous material with the collapsed walls of the previously ruptured cysts were identified (Fig. 3). In splenectomy

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