

# Hydatid Cyst With Various Organ and Tissue Involvement: A Single-Center Data

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## ABSTRACT

Hydatid cyst (CH) is a zoonotic infection caused by the parasite *Echinococcus Granulosus*. This study aims to determine the prevalence of hydatid disease presenting to our center and to present and discuss rare localized CH samples.

A total of 1247 patients were evaluated in the study. All patients were noted as Type 1 through Type 5. In addition, cysts not included in the Gharbi classification and diagnosed with *Echinococcus alveolaris* were defined as Type 6, infected CH as Type 7, post-operative (PAIR and/or surgery) CH as Type 8, and ruptured CH as Type 9, for ease of lesion identification and classification.

A total of 936 patients were included in the study. The average hydatid cyst lesion size is  $6.6 \pm 3.7$  (min=0.6-max=47) cm. When the hydatid cyst lesions of the patients were examined, it was seen that 90.9% had one stage (type), 7.8% had two stages, and 1.3% had three stages. 28.5% of the patients had Type 1, 12.7% had Type 2, 12% had Type 3, 16.9% had Type 4, 22.8% had Type 5, 2.5% had Type 5. It was observed that 0.5% was Type 6, 0.5% was Type 7, 4.8% was Type 8, and 9.5% was Type 9.

Since the condition or stage of the lesion is important in the treatment and follow-up of hydatid cysts, it would be logical and guiding to classify it as in our study.

**Keywords:** Hydatid cyst, Gharbi classification, Radiological imaging, Imaging features

## Introduction

Hydatid cyst (CH) is a zoonotic infection caused by the parasite *Echinococcus Granulosus* (EG). CH disease is a preventable public health problem and can spread to all body organs and systems locally, infiltratively, hematologically or lymphatically. Although the most common form is the disease caused by EG, the invasive and metastasizing malignant form caused by *Echinococcus alveolaris* can also be seen very frequently. Our region is among the geographies where this parasitic infection is highly endemic, such as South America, China, Africa, the Middle East, and Mediterranean countries (1). Its prevalence is quite high in regions of the world where there is animal husbandry, especially in our country, where small livestock such as sheep and goats are raised, and specifically in the eastern and southeastern Anatolian regions of our country. For this reason, our hospital, which is a tertiary health institution, has become a major center in terms of diagnosis, treatment and follow-up of hydatid disease, and our experience in this regard has increased considerably.

Most hydatid cyst patients are generally asymptomatic. However, it may present with a complex clinical picture as a result of local compression or complications in the location of the cyst. The most common complication is the local compression effect or rupture of the cyst (2). Another important complication is the hypersensitivity reaction or anaphylactoid reaction caused by immune complexes entering the tissue stroma and circulation as a result of the rupture of the cyst (3). This study aims to determine the prevalence of hydatid disease presenting to our center and to present and discuss rare localized CH samples. In addition, case examples will be presented, taking into account the radiological features in ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI).

## Materials and Methods

All radiological images of patients who were admitted to our hospital with various complaints and underwent surgical operations or

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interventional procedures between 2010 and 2022 were retrospectively examined or recorded. Ethics committee approval was received from Van Yuzuncu Yıl University Institutional Ethics Committee.

A total of 1247 patients were evaluated in the study. 311 patients with missing data or no clinical, radiological, or serological data or diagnosed as simple cysts were excluded from the study because they were unsuitable. A total of 936 patients were included in the study.

All laboratory and radiological findings of the 936 patients included in the study were accessed and evaluated individually via the hospital information management system (HIMS) and picture archiving and communication systems (PACS). Examinations were performed on all patients was noted. To avoid duplication of patients with multiple examinations, each patient was confirmed with their file number. The diagnosis of CH was made based on serological, radiological or histopathological-parasitological results of patients who underwent interventional procedures or surgery.

There are currently two different classifications in the diagnosis and follow-up of hydatid disease: the World Health Organization (WHO) classification and the Gharbi classification. In our study, CH lesions were classified according to the Gharbi Classification, and lesions that were treated or complicated were also classified separately. Gharbi classification consists of five stages (4). Type 1: Pure homogeneous anechoic thin-walled cystic lesion, type 2: Cystic lesion with detached membranes, type 3: Cystic lesion with daughter vesicles, type 4: Solid lesion causing pseudotumor, and type 5: partially or completely calcified lesion (inactive).

All patients were noted as Type 1 through Type 5. In addition, cysts not included in the Gharbi classification and diagnosed with *Echinococcus alveolaris* were defined as Type 6, infected CH as Type 7, post-operative (Puncture, Aspiration, Injection, Re-Aspiration (PAIR) and/or surgery) CH as Type 8, and ruptured CH as Type 9, for ease of lesion identification and classification.

**Statistical Analysis:** The analyses were evaluated using the SPSS (Statistical Package for Social Sciences; SPSS Inc, Chicago, IL) 22 software package. In this study, descriptive data were presented as n and % values for categorical data and as mean  $\pm$  standard deviation (Mean  $\pm$  SD) values for continuous data. Chi-square analysis (Pearson Chi-square) was carried out to compare

categorical variables among groups. The normality of continuous variables has been assessed using the Kolmogorov-Smirnov test. The Mann-Whitney U test was utilized to compare paired groups. In the analyses, the statistical significance level was accepted as  $p < 0.05$ .

## Results

A total of 936 patients, 566 (60.5%) women and 370 (39.5%) men, were included in the study. The average age of the patients is  $36.1 \pm 21.5$  (min=1-max=90) years and the average hydatid cyst lesion size is  $6.6 \pm 3.7$  (min=0.6-max=47) cm. When the examination modalities of the patients were examined, 45.7% were US, 48.4% were CT, and 5.9% were MRI (Table 1).

The average age of women was found to be significantly higher than the average age of men ( $p < 0.001$ ). There were no significant differences between genders in terms of size ( $p = 0.869$ ), modality ( $p = 0.795$ ) and number of stages ( $p = 0.431$ ) (Table 2).

When cyst localization was analyzed, it was observed that the most common cyst localization was in the liver (80.6%), and the second most common cyst localization was in the lung (11.5%). The frequency of liver and lung coexistence was 1.7% (Table 3).

When the hydatid cyst lesions of the patients were examined, it was seen that 90.9% had one stage (type), 7.8% had two stages, and 1.3% had three stages. 28.5% of the patients had Type 1, 12.7% had Type 2, 12% had Type 3, 16.9% had Type 4, 22.8% had Type 5, 2.5% had Type 6. It was observed that 0.5% was Type 7, 4.8% was Type 8 and 9.5% was Type 9 (Table 4).

## Discussion

In hydatid cyst disease, humans serve as the intermediate host and become infected after ingesting the parasite's eggs through the fecal-oral route. The first place it reaches is the hepatobiliary system because it is taken through the gastrointestinal tract and absorbed through the intestinal mucosa and then transported to the portal system. Therefore, in our study, the most common localization was determined to be the liver. According to literature information, the liver is the most frequently affected organ, with a rate of 70%, and the right lobe of the liver is affected more frequently than the left lobe. In our study, this rate was 80.6% and was found to be higher

**Table 1:** Demographic Characteristics, size and Modalities of the Patients

		N	%
Gender	Female	566	60,5
	Male	370	39,5
Age, Mean±SD		36,1±21,5	
Size, Mean±SD		6,6±3,7	
Modality	US	428	45,7
	CT	453	48,4
	MRI	55	5,9

**Table 2:** Comparison of Various Parameters By Gender

		Female		Male		P
		Mean±SD		Mean±SD		
Age		38,7±20,9		32,3±21,8		<0,001*
Dimension		6,5±3,2		6,7±4,4		0,869*
		Number	%	Number	%	
Modality	US	263	46,5	165	44,6	
	CT	268	47,3	185	50,0	0,695**
	MRI	35	6,2	20	5,4	
Number of stages	1 type	520	91,9	331	89,5	
	2 type	39	6,9	34	9,2	0,431**
	3 type	7	1,2	5	1,4	

\*Mann-Whitney U test, \*\*Chi-square analysis applied

than the rate reported in the literature. The lung is the next most commonly affected organ and is involved in approximately 20% of cases (5). In our study, lung involvement was less common compared to the literature information.

#### Common and Uncommon Locations of Hydatid Cyst

**Liver and Lung:** Liver and lung hydatid cyst localizations constitute approximately 90% of the cases. The remaining 10% covers other organs and systems. In our study, the most common localization was the liver - biliary system in 80.6% (n=754), and the second most common localization was the lung in 11.5% (n=108). Hydatid cyst lesions can be seen in five different stages and can be observed in a spectrum from pure cystic lesions to solid calcified lesions, depending on the stage (Figures 1 and 2).

**Gastrohepatic ligament:** At the level of the gastrohepatic ligament, hydatid cyst can spread from the liver or through the neighbouring structures as a result of rupture of hydatid cyst lesions in other abdominal organs, or, which is much rarer, by hematogenous spread (Figure 3).

**Hydatid Cyst Opening Into The Bronchus Or Causing Embolism:** Hydatid disease is

complicated and can spread anywhere hematogenously. One of the common complications when it occurs in the lung is bronchial opening or rupture (6). Similarly, when it reaches the heart hematogenously, it can go to the lungs, that is, the pulmonary artery, and cause pulmonary embolism (Figure 4).

**Spleen:** Spleen involvement of hydatid disease is between 0.9% and 8% in the literature, and spleen involvement has been stated as the third most common location. In the differential diagnosis, epidermoid, lymphangioma, pseudocyst, splenic abscess, and cystic tumors of the spleen should be considered (7). Radiological findings of hydatid cysts in the spleen are generally similar to those in the liver. For differential diagnosis, radiological, clinical and laboratory findings, along with simultaneous hydatid cysts in another organ, are helpful in diagnosis (Figure 5).

**Mesentery and Appendix:** Hydatid cyst cases in the mesentery and omentum are very rare, and there is limited information about this in the literature. However, we know that hydatid cysts can mimic many lesions and are usually achieved by transplantation into the abdomen secondary to the operation in patients who have had intra-

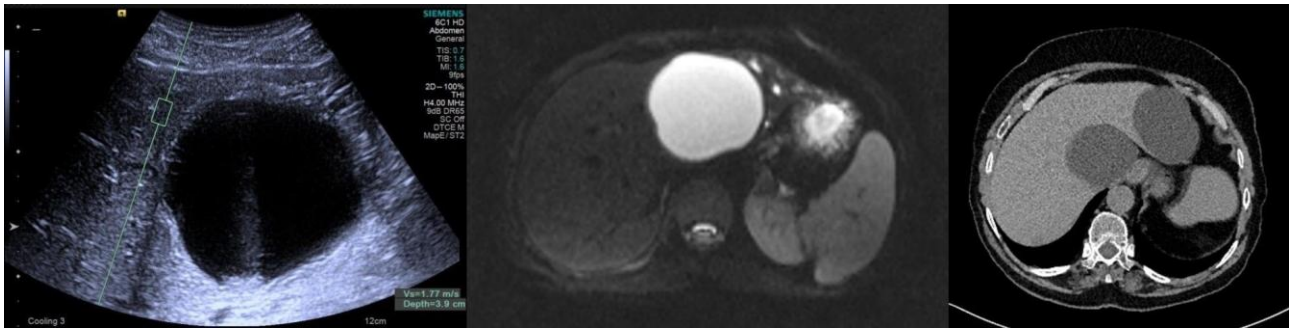
**Table 3:** Localization of Hydatid Cyst Lesions According To Organ Involvement

	Number (n)	%
Liver	754	80,6
Lung	108	11,5
Spleen	19	2,0
Mesentery	3	0,3
Pericardium	4	0,4
Pelvis	1	0,1
Axilla	1	0,1
Kidney	2	0,2
Surrenal	1	0,1
Brain	3	0,3
Ovary	1	0,1
Subcutaneous	1	0,1
Gastrohepatic ligament	3	0,3
Vertebra	1	0,1
Intramuscular	1	0,1
Hilum of the lung	1	0,1
Retroperitoneum	1	0,1
Heart	1	0,1
Paraesophageal	1	0,1
Liver + Lung	16	1,7
Liver + Spleen	4	0,4
Liver + Mesentery	2	0,2
Liver + Pelvis	2	0,2
Liver + Gastrohepatic ligament	1	0,1
Liver + Lung + Kidney	1	0,1
Liver + Lung + Brain	1	0,1
Lung + Pericardium	1	0,1
Lung + Hilum	1	0,1

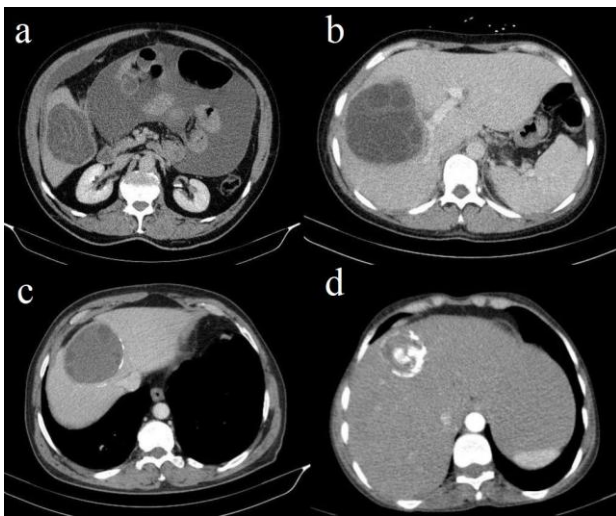
**Table 4:** Cyst types and number of related patients

	Number of patients (n)	%	
Number of types	1 type	851	90,9
	2 types	73	7,8
	3 types	12	1,3
Type*	Type 1	267	28,5
	Type 2	119	12,7
	Type 3	112	12,0
	Type 4	158	16,9
	Type 5	213	22,8
	Type 6 (Alveolar)	23	2,5
	Type 7 (Infected)	5	0,5
	Type 8 (post-op, PAIR or surgery)	45	4,8
	Type 9 (Ruptured)	89	9,5

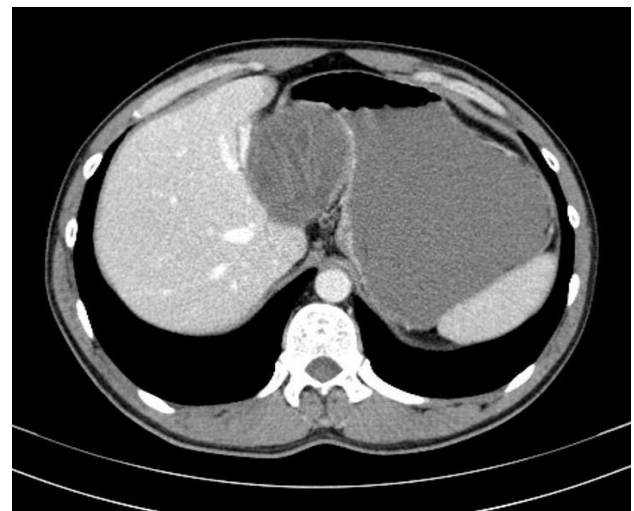
\* There is more than one type



**Fig. 1.** Ultrasound, MRI, and CT images of Type 1 hydatid cysts in the left lobe of the liver in three different patients



**Fig. 2. (a-b-c-d)** Type 2, Type 3, Type 4, and Type 5 hydatid cyst cases are observed on CT scans in different parts of the liver in four separate patients, respectively



**Fig. 3.** A type 2 hydatid cyst located in the gastrohepatic ligament and compressing the liver and stomach is seen on the CT image

abdominal surgery. It is also possible to see cystic lesions in the abdomen by hematogenous means (8). When hydatid cyst lesions are seen in the abdomen, they are seen as multiloculated daughter vesicles or multiple cystic foci, but they can also be seen as single or unilocular cysts and can be confused with mesenteric cysts and duplication cysts (Figure 6).

**Pelvis and Pouch of Douglas:** It is very rare to see hydatid disease in the pelvic area or the pouch of Douglas. However, there may also be hydatid cysts that rupture from other organs and systems in the abdomen or spread through the neighborhood structures. The most common cystic lesions in the pelvic area are ovarian lesions; hydatid cysts should be considered in the differential diagnosis in endemic regions (9). If any invasive procedure is to be performed, it is crucial to make preparations accordingly (Figure 7).

**Ovary:** The majority of cysts in the ovary are benign cysts originating from the ovary. To a lesser extent, it can be observed in malignant cystic lesions in the ovary. However, a hydatid cyst in the ovary is very rare and can generally occur as a result of hematogenous spread (10). In our study, a patient diagnosed with a hydatid cyst in the right ovary is presented (Figure 8).

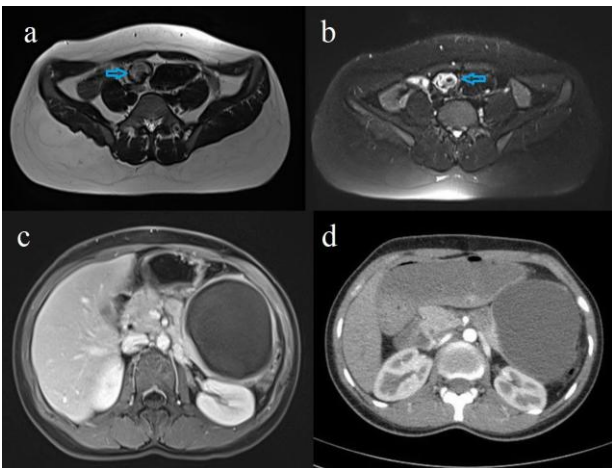
**Pancreas:** Pancreatic involvement constitutes approximately 2% of all hydatid cyst lesions, and primary involvement is extremely rare. It usually appears as a single lesion and is observed in the head of the pancreas (10). Symptoms generally develop due to local compression on the common bile duct or duodenum, depending on its location. The differential diagnosis includes serous and mucinous cystadenomas, cystic tumors of the pancreas such as intraductal papillary mucinous neoplasm (IPMN), solid pseudopapillary epithelial neoplasm (SPEN), benign simple cysts and pseudocysts (Figure 9).



**Fig. 4.** This case reveals the computed tomography findings of a hydatid cyst that spread to the lung parenchyma via the bloodstream and caused pulmonary embolism



**Fig. 5.** There are different types of hydatid cyst lesions in the spleen, some of which contain calcification and can mimic malignant lesions, and contrast-enhanced CT images of cases whose postoperative diagnosis has been confirmed histopathologically are observed

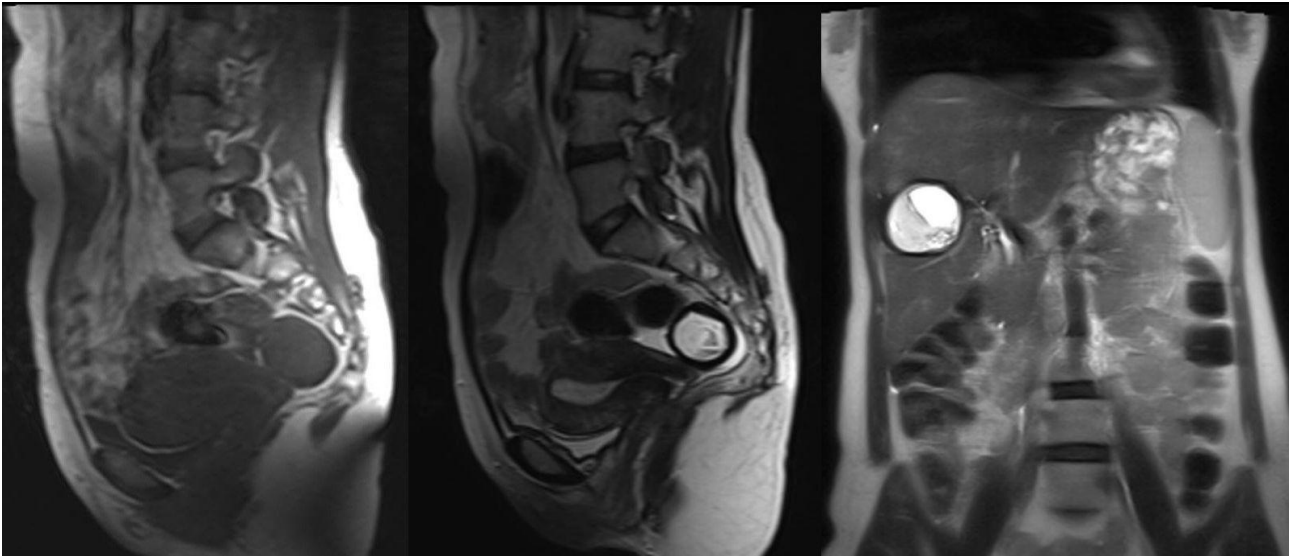


**Fig. 6. (a-b)** Hydatid cyst lesion located in the pelvic mesenteric region (arrows) is seen on T1 and T2 weighted MRI images, respectively. **(c-d)** Contrast-enhanced T1-weighted MRI and CT images of a type 1 hydatid cyst in the mesenteric region of the upper left quadrant of the abdomen are presented, respectively

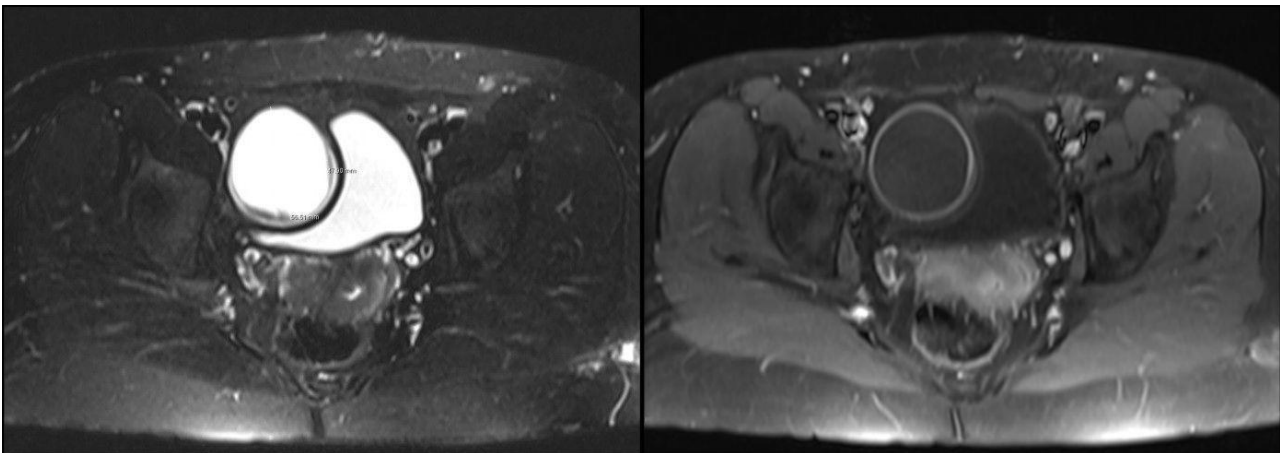
**Adrenal Gland:** Adrenal cysts are rare lesions and are often detected incidentally. The incidence of adrenal cysts in autopsy series studies was reported as .073% (11). Rarely, malignant neoplasms can mimic benign cysts. Therefore,

distinguishing adrenal hydatid cysts from other adrenal cysts and cystic changes of adrenal solid tumors by imaging findings alone is a diagnostic challenge. This is especially difficult with large-sized cysts. A study showed that 1.2% of the detected lesions were malignant and all of them exceeded 5 cm (12). Hydatid cysts constitute approximately 6-7% of all adrenal cysts. Adrenal hydatid cyst is usually seen together with liver hydatid cyst. Primary or isolated adrenal hydatid cyst constitutes less than 1% of hydatid cyst cases (13) (Figure 10).

**Kidney:** Renal hydatid cyst involvement is rare, accounts for approximately 3% of cases, and is usually solitary and located in the renal cortex (10). Renal hydatid cyst disease usually has no noticeable signs or symptoms, often remaining asymptomatic for years, and serological tests only play a confirmatory role in a small number of cases due to their high rate of false-negative results (14-15). The occurrence of cystic hydatid symptoms is quite variable, and the most common symptoms are flank pain, palpable mass, fatigue, hematuria, fever and hydraturia (16). Ultrasound is the most important diagnostic tool for hydatid disease, capable of clearly showing detached



**Fig. 7.** In this case, the MRI findings on T1 and T2 weighted images of a patient with hydatid cyst lesions in the Douglas pouch and simultaneously in the liver are observed, respectively



**Fig. 8.** A cystic lesion is seen in the right ovary, which is hyperintense on T2-weighted imaging, shows increased wall contrast on contrast-enhanced T1-weighted imaging, and pushes the ovarian parenchyma posteriorly. The diagnosis of the lesion was confirmed as a hydatid cyst

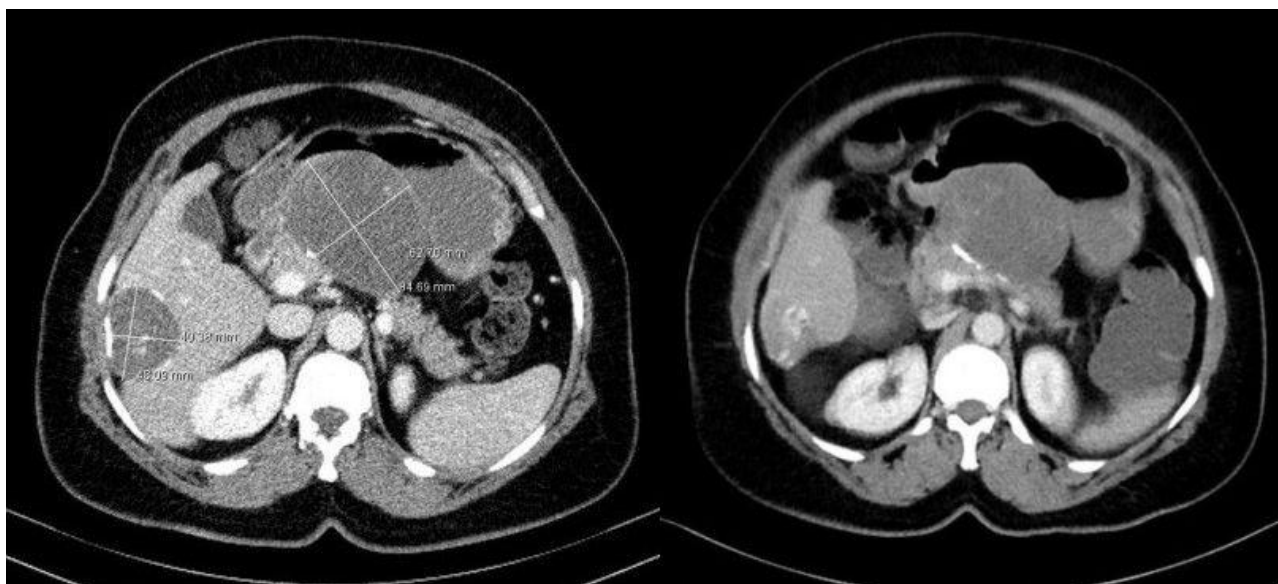
membranes, daughter vesicles, and hydatid sand that are characteristically seen in cystic lesions. Routine imaging with CT or US is considered the primary method for diagnosis, with serology and other tests serving as complementary tools (17) (Figure 11).

The preferred procedure in the treatment of renal hydatid cyst lesions is simple excision of the cyst and, if possible, kidney-sparing surgery is performed. A nephrectomy becomes necessary when the kidney is damaged. Due to the absence of a potent systemic scolicidal agent, the only definitive treatment seems to be surgery. However, PAIR also provides benefits for selected patients. Surgery may completely cure the patient, but it does not completely prevent recurrence. Generally, albendazole is used to prevent

recurrence for up to six months after capsule excision (17).

**Brain:** Only 1-2% of hydatid cysts can reach the brain after passing through the liver and lungs. It constitutes only 1-2% of the lesions located in the entire intracranial space (18). Most cases occur in the pediatric population (19).

Primary cerebral hydatid disease usually occurs as a single lesion. Sometimes, multiple cerebral cysts can form as a result of the rupture of a primary cerebral cyst or embolization of a ruptured cyst (20). Hydatid cyst symptoms depend on the location and size of the cyst. These occur either due to the local compression effect or as a result of increased intracranial pressure. Headache is the most common complaint; other symptoms consist of nausea, vomiting, drowsiness, dizziness,



**Fig. 9.** A mass lesion is observed covering the neck and body of the pancreas, containing a daughter vesicle in the center and wall calcification in the peripheral area. A similar calcified hydatid cyst is also present in the right lobe of the liver



**Fig. 10.** Contrast-enhanced CT images of a type 4 hydatid cyst with calcified walls compressing the kidney in the left adrenal gland are examined, and the PAIR procedure, which provides diagnosis and treatment, has been performed

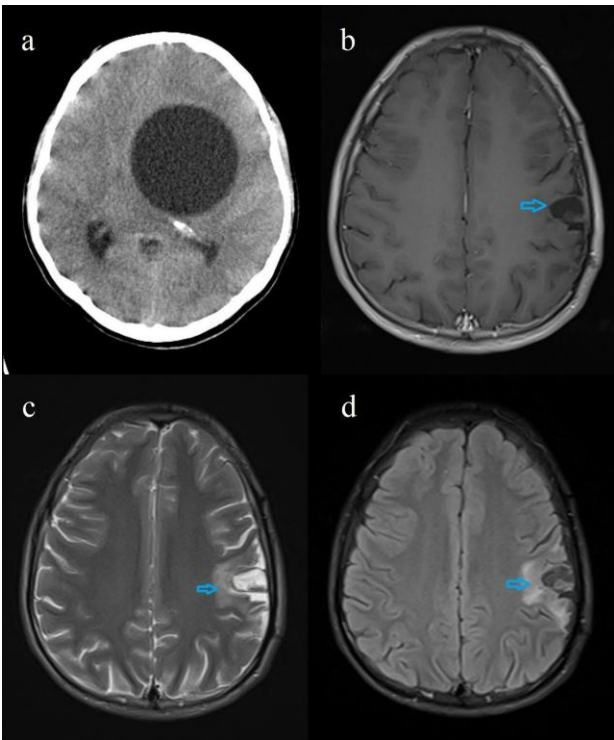
seizures, ataxia, visual disturbances, and cranial nerve palsy with focal neurological deficits that may develop depending on the location and size of the cyst (21). When the cyst is located in the posterior fossa, cerebellar symptoms may occur (21-22). MRI and CT scans are examination methods used for radiological diagnosis, and MRI is preferable for surgical planning (23-24) (Figure 12).

**Musculoskeletal and Soft Tissues:** We know that the majority of hydatid cysts occur in the liver, lungs, or both, but primary soft tissue involvement in the musculoskeletal and

subcutaneous tissues, although very rare, can be seen and cause a diagnostic difficulty (25). The most common areas of musculoskeletal involvement include the pelvic, thigh and paravertebral levels (26). US is an ideal modality to demonstrate hydatid cyst sensitivity in superficially located lesions such as muscle, soft tissue and subcutaneous tissue, floating membranes, daughter vesicle cysts and pure cystic lesions. MRI can also be a suitable examination method as it provides a better definition of anatomical relationships and is useful in pre-surgical planning.

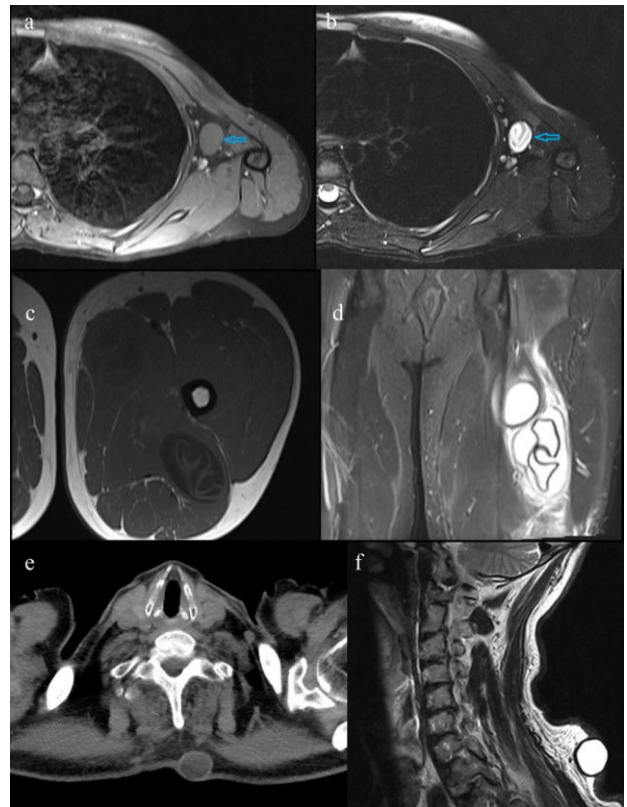


**Fig. 11.** Contrast-enhanced dynamic CT images exhibit a hydatid cyst with calcified walls and daughter vesicles causing enlargement in the upper pole of the right kidney

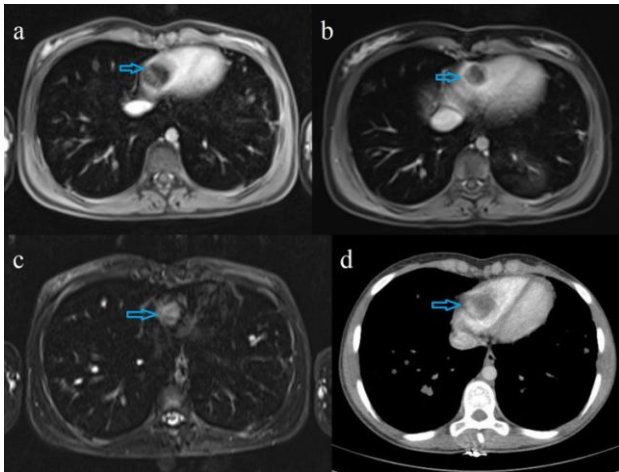


**Fig. 12.** (a) Non-contrast brain CT shows a type 1 hydatid cyst compressing the lateral ventricle in the left hemisphere. (b-c-d) In another patient, T1, T2, and FLAIR-weighted imaging findings of a hydatid cyst in the posterior left frontal lobe, causing adjacent vasogenic edema, are observed, respectively

It is essential to make a definitive preoperative diagnosis of skeletal muscle hydatid cysts. It is important in terms of complications such as possible anaphylactic shock, and some diagnostic approaches such as excision or incisional biopsy. In a study, they reported cyst rupture after a



**Fig. 13.** (a-b) Findings of a hydatid cyst with detached membranes are observed on contrast-enhanced T1-weighted and T2-weighted fat-saturated images in the left axilla, respectively. (c-d) MRI findings of a type 2 hydatid cyst with detached membranes on the left thigh are observed on T1 and T2-weighted images. On the T2-weighted image, the lesion shows an increased edematous signal in the adjacent soft tissues. (e-f) A well-circumscribed cystic lesion with CT and MRI findings in the posterior subcutaneous tissue in the inferior cervical region was excised, and its pathology was reported as a hydatid cyst



**Fig. 14.** In this case, MRI (a-b-c) and CT findings of a hydatid cyst (arrows) located in the right ventricle of the heart are observed (d)

previous fine needle aspiration, which complicated the surgical drainage of an infected primary musculoskeletal hydatid cyst (27). Pericystectomy is the preferred treatment option for musculoskeletal hydatid cysts (Figure 13).

Cardiac hydatid cysts are rare and are reported to be around 0.02-2% in the literature. Hydatid cyst larvae spread through pulmonary veins, lymphatic vessels or coronary arteries. It is a rare entity that can spread from the heart to all organs and systems (28) (Figure 14).

Hydatid cyst cases in vertebrae and bone structures are very rare. Especially, the hydatid cyst located in the vertebra can cause expansion or pathological fracture in the bone structure and cause spinal cord compression symptoms (10) (Figure 15).

Limitations: Our study has some limitations because it is retrospective. Firstly, since our study was in the form of scanning data from the PACS system and was a retrospective study, not all patients could be included. Patients with missing data were excluded from the study.

Our study is single-center and may not represent the entire endemic region. In our study, histopathological verification, which is the gold standard method for diagnosis, could not be made in all patients. In fact, the diagnosis of hydatid cyst is mostly made based on clinical-radiological findings, but in a small number of cases, the diagnosis is supported by the specimen sent to pathology after surgery. Typical radiological findings of hydatid cyst lesions, serological findings and patients diagnosed as a result of the procedure or operation history of the patients were evaluated in our study. However, since



**Fig. 15.** (a-b) The hydatid cyst located in the T8 vertebral body, causing the pathological fracture, appears as a hypodense and lytic lesion on CT. (c-d) The lesion is observed as hypointense on T1-weighted images and heterogeneously hyperintense on T2-weighted images, respectively

clinical follow-up of all patients could not be performed, there is a possibility that an atypical diagnosis may have been made in subsequent surgical interventions.

Hydatid cyst is a parasitic disease that can spread systemically to all organs and systems, firstly via the fecal-oral route and then locally invasively or through the neighbourhood or hematogenously, and multimodal systems are used in diagnosis. However, in description and classification, Gharbi and WHO classifications evaluate hydatid cyst lesions in five stages, and no classification or definition has been made in the re-imaging of patients with complicated hydatid cysts or those who have undergone an interventional procedure. Since the condition or stage of the lesion is important in the treatment and follow-up of hydatid cysts, it would be logical and guiding to classify it as in our study. In addition, hydatid disease should be kept in mind when making a differential diagnosis of cystic lesions in endemic areas.

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