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Atypically Located Hydatid Cysts in Human Morphology

Quistes Hidatídicos de Localización Atípica en la Morfología Humana

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SUMMARY: Hydatid cysts (HC) caused by Echinococcus granulosus (EG) larvae are commonly observed in the liver and lungs; however, atypical localizations also occur. This study investigates the frequency of atypically located HC and their relationship with surgical history. Cysts can be found in regions such as the diaphragm, mediastinum, and myocardium within the thoracic cavity, and in the spleen, kidneys, brain, and bone tissue outside the thoracic cavity. Patients with a surgical history exhibit an increased risk of complications and longer hospital stays, which affect treatment processes. The findings suggest that atypical cysts may be related to surgical incision lines. As a result, systematic screening and the development of existing surgical techniques are recommended for patients at risk of Echinococcus granulosus infection. This study provides important insights into the diagnosis and treatment of atypical hydatid cysts and lays the groundwork for future research.

KEY WORDS: Atypical hydatid cysts; Echinococcus granulosus; Hospitalization duration.

INTRODUCTION

Hydatid disease is globally prevalent but more commonly seen in endemic regions such as the Mediterranean, the Middle East, South America, and parts of Asia and Africa. It imposes a significant public health burden in these areas, affecting both rural and urban populations. The prevalence in endemic areas can vary, but in some regions, up to 5-10 % of the population may be affected. The disease incurs a high economic cost due to healthcare requirements, surgical interventions, and long-term follow-up. The clinical manifestations of hydatid cysts depend largely on their location, size, and the involvement of surrounding structures. In atypical locations, cysts may present with nonspecific symptoms that mimic other conditions, leading to diagnostic delays. For instance: neurological symptoms like seizures or focal deficits, arrhythmias, pericarditis, or even life-threatening cardiac tamponade and pathological fractures or osteolytic lesions. Imaging techniques such as ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) are critical for diagnosing hydatid cysts, particularly in atypical sites. However, misdiagnosis is common due to the unusual presentation, and thus, serological tests (e.g., ELISA, indirect hemagglutination) are often employed to confirm echinococcosis. Hydatid disease, or echinococcosis, is a parasitic infection caused by the larval stage of Echinococcus species, primarily *E. granulosus* and *E. multilocularis*. The disease is typically acquired through the ingestion of parasite eggs, which are transmitted by definitive hosts, often canines. Humans are accidental hosts, and the larval cysts primarily develop in the liver (approximately 70 %) and lungs (10-20 %) due to the portal vein system and blood flow dynamics, respectively. These cysts form slowly over time, creating space-occupying lesions that can lead to organ dysfunction and other systemic effects (Fig. 1) (Çörtelekoglu *et al.*, 2003; Karakol & Tatar, 2021). Cysts located outside the liver and lung parenchyma are called atypically located HS and can be seen in the diaphragm, bone, and muscle in the chest wall, mediastinum, myocardium, and pulmonary artery in the thorax. Outside the thorax; spleen, lobar fissure, pleural cavity, kidneys, bone tissue, brain tissue, muscle tissue and rarely arterial and venous structures. The management of hydatid cysts in atypical locations often requires tailored approaches due to their anatomical proximity to critical structures. Surgical intervention remains the primary treatment modality, especially for accessible cysts in organs where surgical excision is feasible. However, complete resection can be challenging in atypical sites, and the risk of cyst rupture or spillage during surgery poses additional risks of anaphylaxis or secondary echinococcosis. Adjunctive antiparasitic therapy with albendazole or mebendazole is often used postoperatively to prevent recurrence (Isitmangil *et al*. 2003; Craig *et al*., 2007; Karakol & Tatar, 2021).

Despite the clinical significance, the literature on atypically located hydatid cysts remains limited, with most cases being isolated reports. This study aims to consolidate knowledge on the clinical presentations, diagnostic challenges,

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and management strategies associated with atypically located hydatid cysts. By analyzing a series of cases and reviewing current diagnostic and therapeutic options, we hope to provide insights that enhance clinical decision-making and patient outcomes in hydatid disease involving atypical sites.

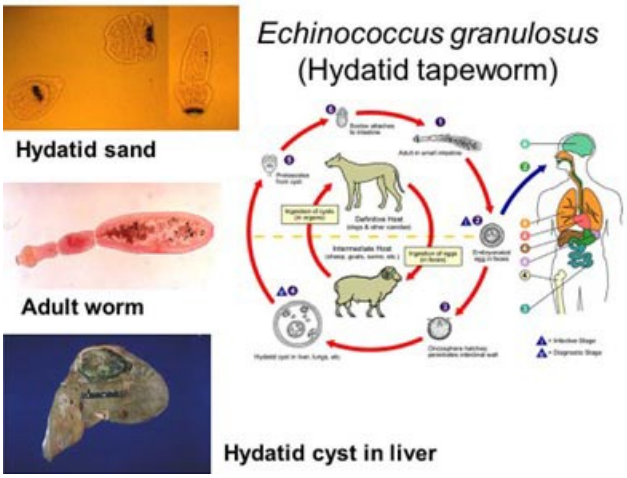

Fig. 1. Gross morphology of hydatid cysts.

MATERIAL AND METHOD

In this study, 23 HC patients with atypical locations treated in our hospital between January 2010 and January 2024 were retrospectively analyzed. Our diagnostic methods include chest X-ray (X-ray), computed tomography (CT), magnetic resonance imaging (MRI), and ultrasonography (USG). Demographic characteristics, localization of cysts (muscle-bone and only soft tissue), cyst size, cyst status (ruptured and intact), serological test results (I˙mmun Floresan Antikor test results were recorded to detect echinococcosis antibodies), surgical histories of patients due to HC, and length of hospital stay were examined in detail.

Cyst size was determined by intraoperative measurements together with imaging methods. Cyst sizes were divided into two groups: the first group consisted of cysts < 4

Table I. Characteristics of atypical cysts.

	Age	Sex	Surgery history	Cyst location	Clinical presentation	Investigations	Management	Duration of stay in the hospital
1	36	F	Liver	Parasternal	Bloating, pain	Chest radiography and usg	Enucleation	3
2	24	F	No	Thoracic cavity	Back pain	IFA, Chest radiography and thorax tomography	Removal by exploration	11
3	44	F	Lung	Mediastinum	Back pain, Stinging pain in the chest	Thorax tomography	Removal by exploration	8
4	65	M	No	Diaphragm	Stinging pain in the chest	IFA, Thorax tomography	Removal by exploration	6
5	17	F	No	Diaphragm	No symptoms	Thorax tomography	Removal by exploration	7
6	36	F	Lung	4.Rib	Bloating, pain	Chest radiography, USG, MR	Removal with ribs and surrounding tissue	4
7	56	M	Lung	6.Rib	Bloating, pain	IFA, USG, MR	Removal with ribs and surrounding tissue	4
8	54	M	Heart tissue	Vertebra	Back pain	Thorax tomography, IFA	Removal of cyst contents and laminektomi	8
9	45	F	Lung	6.Rib	Bloating, pain	USG, Thorax tomography,	Removal with ribs and surrounding tissue	6
10	19	M	Lung	7.Rib	No symptoms	USG, Thorax tomography, MR	Removal with ribs and surrounding tissue	5
11	56	M	No	Mediastinum	Shortness of breath and burning	Thorax tomography, IFA	Removal of cyst contents	8
12	44	M	No	Parasternal	No symptoms	Chest radiography, IFA, USG	Enucleation	3
13	35	M	Liver	Diaphragm	No symptoms	Thorax tomography, IFA, MR	Removal of cyst contents	5
14	23	M	No	Mediastinum	No symptoms	Thorax tomography, IFA, MR	Removal of cyst contents	5
15	56	M	Liver	Diaphragm	Sometimes shortness of breath	Thorax tomography, IFA	Removal of cyst contents	13
16	17	M	No	Diaphragm	Sometimes pain	Thorax Tomography, IFA	Removal of cyst contents and the diaphragm was cut and repaired again.	8
17	45	M	Lung	7.Rib	Pain	USG	Enucleation	3
18	29	F	No	Mediastinum	No symptoms	Tomography, IFA	Enucleation	6
19	41	F	No	1.Rib	Bloating, pain	IFA, USG,	Enucleation	3
20	24	M	No	Muscles	Pain	Tomography, IFA	Removal of cyst contents	4
21	58	F	Soft tissue	Gluteus muscle	Pain	Tomography, IFA	Removal of cyst contents	2
22	64	F	No	Brain tissue	Throbbing headache	Tomography, IFA	Removal of cyst contents	10
23	3	M	No	Parasternal	No symptoms	USG,IFA	Enucleation	2

cm (2-3.5 cm), and the second group consisted of cysts ≥ 4 cm (4-9 cm). In addition, cysts are divided into two separate groups according to their location: soft tissue and bone tissue cysts.

RESULTS

Of the 23 patients examined in our study, 56.5 % were female (13) and 43.5 % were male (10). The mean age of the patients was 38.74 ± 17.033 (3-65) years. The most preferred diagnostic methods are X-ray, USG, and CT. Two patients were diagnosed using MRI because of mass imaging of the chest wall (Table I).

Atypical cysts were present in many places such as between the rib and the intercostal muscle, in the soft tissue on the sternum, in the mediastinum, on the diaphragm, in the thoracic vertebrae, in the brain parenchyma, and on the gluteus maximus muscle (Figs. 2, 3, 4).

Fig. 4. Germinative membrane image with atypical HK enucleation method.

Fig. 2. Intraoperative view of HC in the mediastinum.

Fig. 3. Cranial tomography image of atypical hydatid cyst in the brain parenchyma.

When the history of these atypical HC was questioned, it was determined that 11 (48 %) of these patients had undergone surgery for HC. Of these surgical interventions, 27.3 % were from the liver, 54.6 % from the lung, 9.1 % from the heart, and 9.1 % from soft tissue.

52 % of atypical HC were observed in the soft tissue and 48 % in the bone tissue. Bone involvement was usually observed in areas close to the incision lines, tubes, or drain areas of previous surgical operations for HS (Fig. 5).

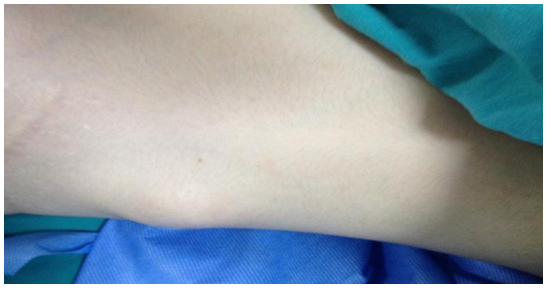

Fig. 5. Image of the cyst close to the previous surgical incision line in patients who have previously had surgery due to hydatid cyst.

In 60 % of the patients, the size of the HC was <4 cm, while in 40 %, it was ≤4 cm. It was detected in 26 % (n = 6) of perforated cysts and 74 % (n = 17) of non-perforated cysts. Approximately 70 % of patients underwent (Immunofluorescent Antibody) IFA testing. Of the patients who underwent IFA, 64 % were positive and 36 % were negative (Table II).

The mean lenght of hospital stay was calculated as 5.82 ± 2.93 (2-13) days. Bone involvement was found to be statistically significant in surgeries performed because of a history of HC. In addition, soft tissue-derived HC were found to increase the hospitalization duration (Table III).

Table II. Sizes of cysts and tissues in which they are located.

	Size under 4 cm	Size 4 cm and above	p
Cyst-related surgical history	6 (55 %)	5 (45 %)	
Cyst-related no surgery history	8 (67 %)	4 (33 %)	0.68
Bone tissue location	8 (73 %)	3 (27 %)	0,021*
Soft tissue location	6 (50 %)	4 (50 %)	0.4
Perforated cyst	3 (50 %)	3 (50 %)	
Intact cyst	11 (65 %)	6 (35 %)	0.64
IFA test positivity	7 (58 %)	5 (42 %)	
IFA test negativity	7 (63 %)	4 (37 %)	1
Female sex	7 (54 %)	6 (46 %)	
Male sex	7 (70 %)	3 (30 %)	0.66

Table III. Analysis of cases who previously had surgery due to hydatid cyst.

	Previus surgery due to cyst	Medical treatment after surgery	Surgery performed	Perforation of the cyst	Location of recurrence	Recurrence time (year)	Treatment duration (months)
1	Lung hydatid cyst	albendazol	Cystectomy & capitonage	Perforated	Mediastinum	3	3
2	Lung hydatid cyst	albendazol	Cystectomy & capitonage	Perforated	4. Rib	4	3
3	Muscle hydatid cyst	albendazol	Total cystectomy		Gluteus muscle	6	4
4	Lung hydatid cyst	albendazol	Cystotomy &capitonage	Perforated	7. Rib	5	4
5	Liver hydatid cyst	albendazol	Interventional procedure guided by USG		Parasternal	6	3
6	Lung hydatid cyst	albendazol	Cystotomy & capitonage		6. Rib	4	6
7	Liver hydatid cyst	albendazol	Interventional procedure guided by USG		Diaphragm	3	3
8	Liver hydatid cyst	albendazol	-		Diaphragm	4	3
9	Lung hydatid cyst	albendazol	Cystotomy & capitonage	Perforated	7. Rib	3	4
10	Lung hydatid cyst	albendazol	Cystotomy & capitonage		4. Rib	5	3
11	Heart hydatid cyst	albendazol	Cyst removal		Vertebra	5	3

DISCUSSION

Hydatid cyst is frequently associated with factors such as low socio-economic status, regional climate impacts and uncontrolled slaughter in regions associated with agriculture and animal husbandry. In addition, the growing population of stray dogs may increase their incidence (Çörtelekoglu *et al*., 2003; Isitmangil *et al*., 2003; Alho *et al*., 2023). Hydatid cysts occurring outside the liver and lungs are atypical. Fourteen % of atypical cysts are detected in endemic regions (Gougoulias *et al*., 2010). Hydatid cyst can be considered as an endemic region due to many factors such as agriculture and animal husbandry being the main source of livelihood in the region, inadequacies in preventing the transmission of infection, increase in stray dogs, inadequate public health measures, uncontrolled animal slaughter. The combination of these factors facilitates the spread of the disease and threatens the health of people in the region.

Imaging techniques such as radiography, US, CT, and MRI are used to diagnose hydatid cyst disease. Serological tests, such as immunofluorescence tests, indirect hemagglutination, immunoelectrophoresis, and enzyme-linked immunosorbent assays (ELISA) can be used for diagnosis. These tests help determine the presence of the parasite and severity of the disease (Akyildiz *et al*., 2009).

Hydatid cysts of the skeletal system can be difficult to diagnose and additional evaluation may be required. Serological examination may not always be helpful, but magnetic resonance imaging devices can indicate the presence of cysts, especially multivesicular cysts (Garcia-Diez *et al*., 2000). In our study, radiography, CT, and US were the most commonly used diagnostic tools. MRI was used for the diagnosis of cysts in lesions with bone involvement, since tomography was seen as a mass. MRI was used to diagnose two patients with thoracic vertebral involvement and rib involvement. Serological tests were performed when patients were available in our hospital. Serological tests were used instead of CT for some bone pathologies.

Following the oral ingestion of EG eggs, primary spread can be seen in many organs. These eggs are primarily filtered through the liver, lungs, and spleen (Craig *et al*., 2007; Mandal *et al*., 2022). If they settle in filtered tissues or in a tissue by escaping filtration, they form cysts with a diameter of 1–30 mm and cause a reaction in the host tissue, which in turn forms a pericyst or ectocyst, which is a fibrous layer of tissue. An endocyst usually consists of one or two layers, and cysts with only laminated layers are called universal or sterile cysts. Protoscolexes are released into the cyst fluid and daughter cysts develop. This condition may be responsible for perforated hydatid cysts, recurrence after surgery, and inflammatory and parasitic complications. Although secondary spread is not clearly understood, EG larvae may spread to their environment due to accidental spread of cysts or spontaneous perforation during surgery. There are opinions that argue that if HC are poured into the pleural cavity intraoperatively, recurrence may occur between 5-22 %. In other studies, the recurrence rate after surgery was 17 % (Mottaghian & Saidi, 1978; Sokouti *et al*., 2013). The vast majority of recurrences were found at or near the surgical site (Aghajanzadeh *et al*., 2022). Contrary to these two views, a study stated that although 30 % of cases were perforated, no recurrence was detected (Mahmodlou *et al*., 2013).

The spread in areas far from the intraoperative site probably supports a mechanism of spread related to the lymphatic system. In case of rupture of the cyst during surgery and the necessary precautions are not taken, the juvenile cysts may spread around and reappear through lymphatics or contact (Ammann & Eckert, 1996; Iuliano *et al*., 2000; Cirenei & Bertoldi, 2001; Sokouti *et al*., 2013; Mandal *et al*., 2022; Aghajanzadeh *et al*., 2022). In a study on mediastinal HC, it was stated that the presence of microruptures and the transmission route may be possible. In the same study, atypical recurrence of HC in the mediastinum was observed in 2 of 24 patients who had previously undergone surgery for lung and liver HC (Erog˘lu *et al*., 2016). In our study, 11 patients who had previously undergone surgery for HC reappeared atypically after an average of 4.5 years. More than 80 % of HC surgeries recurred after lung and liver surgeries. After lung cyst surgery, 54 % of recurrences were atypical in or on the rib cage. The peculiarity of these recurrences is the region that fits the surgical site. These were observed around the tube thoracostomy incision, under or above the thoracotomy incision or ribs, diaphragm, and mediastinum. Recurrences after liver cyst surgery were examined in two diaphragms and one thoracic vertebra. In one case of cardiac cause, recurrence of HC was observed in the thoracic vertebrae. 82 % of these relapses were recurrences in nearby areas

In one study, 282 hydatid cyst-related articles published between 1991 and 2014 were examined. In this review, atypical hydatid cysts were found in endemic areas in 14 % of articles. In this study, the most commonly affected areas were the thighs (78 % thighs, 17 % head and neck and 10 % upper limbs. These findings provide important information regarding the prevalence and localization of hydatid cysts (Gougoulias *et al*., 2010; Salamone *et al*., 2016). In another study, muscle involvement was examined in 5 % of patients, and bone involvement in 2-4 % of patients. On average, muscle and bone involvement was observed in 3 % of patients. The majority of bone involvement is vertebral involvement, the most common of which is thoracic vertebral involvement. Among bone involvements, rib involvement is observed in 6 % of all bone involvements and is rare (Grove *et al*., 1976; Faber *et al*., 2010; Sioutis *et al*., 2021; Aleissa *et al*., 2023). As observed in many studies, bone and muscle involvement associated with HC is rare. In another atypical study, cysts were localized in soft tissues (spleen, skin, muscle) (Khanna *et al*., 2005). Some atypical localizations can be seen in regions such as the thyroid, kidney, spleen, and peritoneum (Versaci *et al*., 2005).

It was observed that the recurrences of patients who had previously undergone surgery for HS after an average of 5 years were atypical cysts with bone location. These cyst lesions were mostly located in the ribs, sternum, and vertebrae, close to the surgical site.

CONCLUSIONS

In conclusion, while hydatid cysts commonly present in the liver and lungs, atypical locations can pose significant diagnostic and therapeutic challenges. Recognizing the possibility of hydatid disease in uncommon sites is essential, especially in endemic regions or in patients presenting with nonspecific symptoms that do not clearly indicate typical infection sites. Early and accurate identification of these

atypically located cysts, aided by imaging and serological testing, enables timely intervention and minimizes complications. Our study underscores the importance of considering hydatid disease in differential diagnoses across a range of anatomical sites, advocating for increased clinical awareness to improve patient outcomes.

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RESUMEN: Los quistes hidatídicos (HC) causados por larvas de *Echinococcus granulosus* (EG) se observan comúnmente en el hígado y los pulmones; sin embargo, también se producen localizaciones atípicas. Este estudio investiga la frecuencia de los HC de localización atípica y su relación con el historial quirúrgico. Los quistes se pueden encontrar en regiones como el diafragma, el mediastino y el miocardio dentro de la cavidad torácica, y en el bazo, los riñones, el cerebro y el tejido óseo fuera de la cavidad torácica. Los pacientes con antecedentes quirúrgicos presentan un mayor riesgo de complicaciones y estancias hospitalarias más prolongadas, lo que afecta los procesos de tratamiento. Los hallazgos sugieren que los quistes atípicos pueden estar relacionados con las líneas de incisión quirúrgica. Como resultado, se recomienda la detección sistemática y el desarrollo de las técnicas quirúrgicas existentes para los pacientes con riesgo de infección por *Echinococcus granulosus*. Este estudio proporciona información importante sobre el diagnóstico y el tratamiento de los quistes hidatídicos atípicos y sienta las bases para futuras investigaciones.

PALABRAS CLAVE: Quistes hidatídicos atípicos; *Echinococcus granulosus;* Duración de la hospitalización.

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