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Review of Hydatid Cyst with Focus on Cases with Unusual Locations

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ABSTRACT

Objective: This study has been conducted so as to contribute to health statistics of hydatid cyst by the data obtained from our clinic, and to discuss hydatid cysts in unusual locations.

Material and Method: Cases diagnosed as hydatid cyst at Dr. Lutfi Kirdar Kartal Research and Education Hospital Pathology Clinic between 2007 and 2015 have been evaluated based on criteria such as age, sex and location.

Results: A total of 364 cases, 209 females and 155 males, have been included in the study. The subjects in the cases are aged between 4 and 81 (mean: 38.84). Regarding the sites, 254 (69.8%) of the cases are located in liver, and 53 (14.6%) in the lung. Fifty-seven cases (15.6%) have been detected in unusual sites other than the lung or liver. The rate of isolated organ involvement, other than the lung and liver, has been found to be 10.3%.

Conclusion: Since hydatid cyst can be found in all the body sites, it should be taken into account in the differential diagnosis of all cystic lesions.

Key Words: Hydatid cyst, Echinococcosis granulosis, Unusual locations, Fine needle aspiration

INTRODUCTION

Hydatid cyst (HC), cystic echinococcosis (CE), hydatid disease (HD), is a zoonotic parasitic disease caused by *Echinococcus granulosus* (EG). It threatens human health and leads to huge economical loss both in our country and around the world (1). The prevalence of HC shows regional differences and ranges between 0-79 per 100.000 population (2). Its prevalence amounts to 50-400/100,000, and its incidence is 3.4/100,000 in Turkey (3).

EG is a 5 mm long hermaphroditic tapeworm (4,5). Dogs or other carnivores are considered as definitive hosts, while sheep or other ruminants, as intermediate hosts. After ingestion of food or water that has been contaminated by dog feces containing parasite eggs, most of embryos die in the hepatic capilleries but some of them turn into cysts and even move to lungs and/or other organs. (6-8). While parasitic larvae develop, cysts are seen in organs, giving the disease its name, CE or HC (8,9). Parasites may settle in almost every site of the body forming primary HC (10). Liver and lung are reported as the most frequent locations, yet unusual sites such as peritoneum, retroperitoneum, spleen, kidney, heart, pelvic, urinary tract, bladder, bone, soft tissue, pancreas, gallbladder, inguinal, supraclavicular, cerebral, spinal cord, and abdominal wall have also been described (4,6,7,11-29). Secondary HC mostly occurs after spontaneous or travmatic rupture of a cyst in abdominal cavity when free protoscolices and/or tiny cysts develop to become larger cysts (10).

During the diagnosis, lung X-ray, ultrasound (USG), computerized tomography (CT), and magnetic resonance (MR) were used to locate lesions (30,31). Histopathological examination and detection of three cyst layers that are the outer layer (pericyst), the middle layer and the inner germinal (or germinative) layer play a significant role in HC diagnosis (6). Fine neddle aspiration cytology (FNAC) has lately been introduced for the diagnosis of HC. Laminated membrane, scolex and hooks can be found in cyst fluid (32,33).

HC cases may be seen in many locations of the body; and when it is localized in unusual regions, this could lead to certain difficulties with regard to diagnosis. We have aimed to contribute to health statistics by presenting HC data of our clinic while discussing HCs with unusual locations.

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MATERIAL and METHODS

In the study, 364 cases of HC, diagnosed between June 2007 and June 2015 in our clinic, were included. Based on the archival records, cases were reviewed for age, sex, and locations. Histology slides were re-evaluated by two pathologists (SHK, AS), and patients were grouped based on their sex, age and the location of HCs. Five groups were formed based on age. Patients, 0-14 yrs of age was considered Group 1; those aged 15-29 were in Group 2; those aged 30-44 yrs in Group 3; 45-59 yrs in Group 4, and those above the age of 60 were included in Group 5. All the cases were classified based on the locations, number of lesions or other organ involvements.

RESULTS

Total biopsies (n=250041) were investigated for an 8-year period. The annual incidence rate of HC was 5.4%. As for the cases, 209 individuals were female and 155 were male. The ages of the individuals varied between 4-81 years (mean:38.84). The distribution of the cases based on the age is presented in Figure 1. When the cases were classified by their sites, 254 cases (69.8%) were located in the liver, and 53 cases (14.6%) in the lung. Besides, 57 cases (15.6%) were detected in unusual sites, other than the lung or liver. Clinical features of 57 unusual cases were investigated, and 27 cases revealed a previous diagnosis of liver HC. Distribution of cases based on involvement sites is presented in Table I. To reach a diagnosis, lung X-ray, USG, CT and MR examinations have been used based on the features of the cases (Figure 2A,B). In the histopathological examination of all cases, we have searched structures in conjunction with HC to be able to come up with a definite diagnosis (Figure 3A,B). In three of our cases, HC was diagnosed solely with FNAC. In one case, structures resembling HC were seen in bronchial lavage. In 40 of the cases, both biopsy and cytology materials revealed findings in conjunction with HC. No reaction or complication was recorded after FNAC (Figure 4A-D).

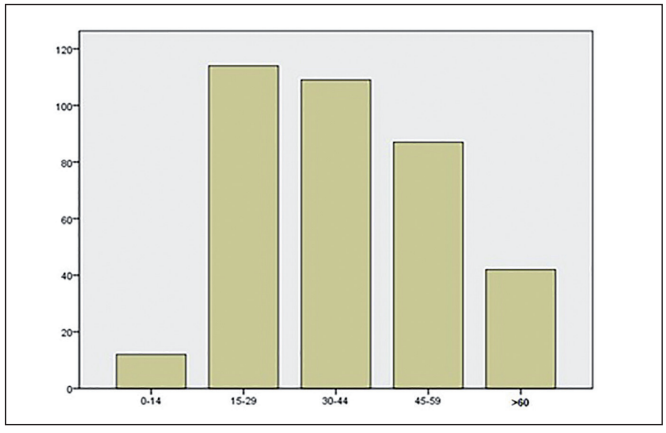

Figure 1: Distribution of the cases based on age groups.

DISCUSSION

Incidence of HC, which is one of the most important zoonotic disases worldwide and in our country, is approximately 5.7/100.000 per year (9), and it has a high prevalance in Central Anatolia, Eastern Anatolia and SoutheasternAnatolia (7,10). In our study of 8-year period, 364 cases of HC were included. Our clinic has a large number of HC cases that might be due to the high immigration rate in our region, and also to ovine-cattle breeding still being practiced in places close to urban areas.

Although found in all ages, HC is frequently observed in adulthood (10). In the national studies, cases were detected between the ages of 4 and 79 years; mean age ranges between

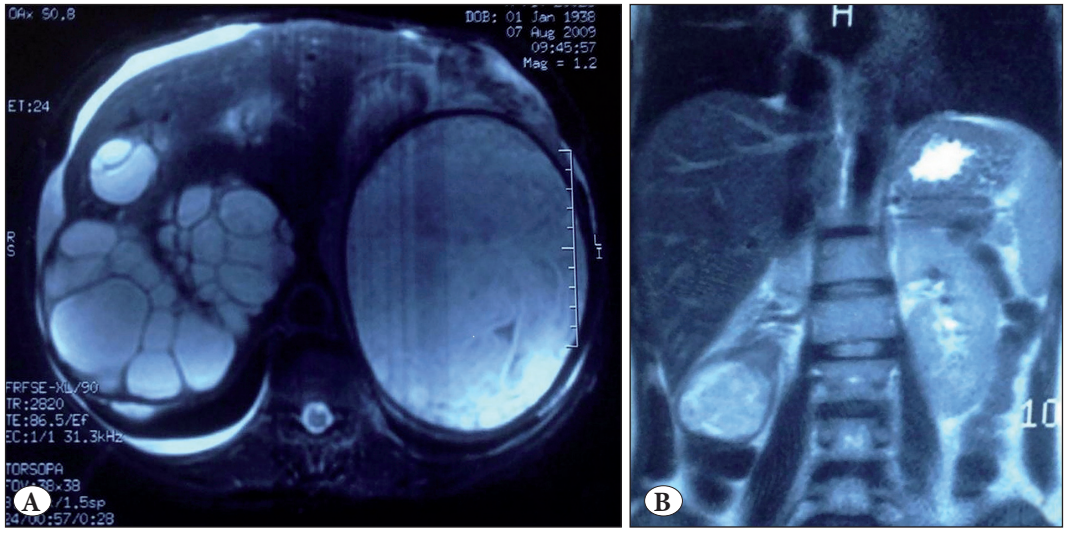

Figure 2: A) MR image of hydatid cysts that include multiple daughter cysts and semisolid areas in liver and spleen. **B)** MR image of hydatid cyst in the lower pole of the right kidney.

Table I: Distribution of cases based on involvement sites

Location (single organ)	Number	%	Location (multiple organ)	Number	%
Liver	254	69.8	Liver+Peritoneum	11	3
Lung	45	12.4	Liver+Lung	8	2.2
Kidney	6	1.6	Liver+Spleen	5	1.4
Heart	6	1.6	Liver+Pelvic region	1	0.3
Pelvic region	5	1.4	Liver+Retroperitoneum	1	0.3
Bone	3	0.8	Liver+Gallbladder	1	0.3
Bladder	3	0.8			
Pancreas	2	0.5			
Spleen	2	0.5			
Supraclavicular	2	0.5			
Spinal cord	2	0.5			
Abdominal	1	0.3			
Inguinal	1	0.3			
Muscle	1	0.3			
Paravertebral	1	0.3			
Cerebral	1	0.3			
Adjacent to right ventricle	1	0.3			
Subdiaphragmatic	1	0.3			

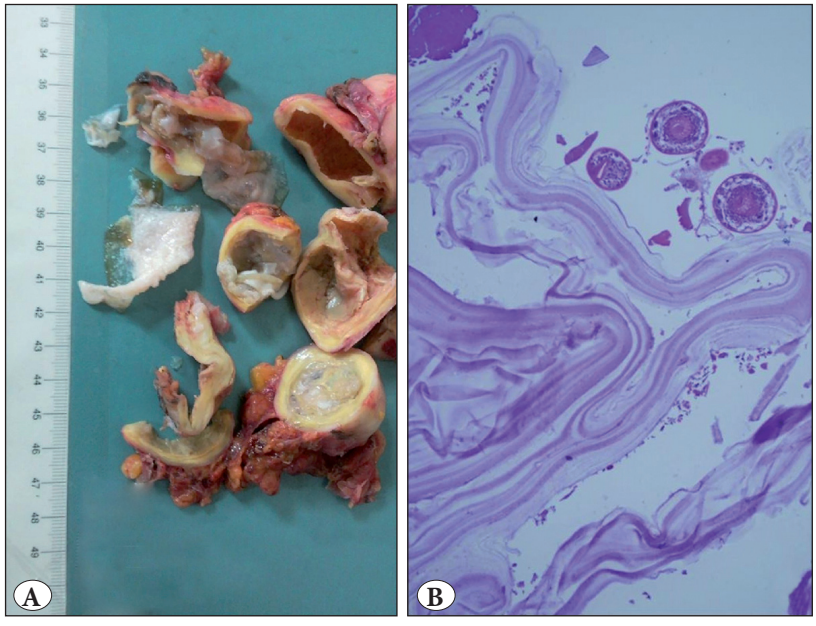

Figure 3: A) Pericyst and membrane structures in hydatid cyst localized in the bladder. **B)** Acellular eosinophilic laminated membranous structures and scoleces (H&E; x20).

28.2 and 35.9 years for females, and 25.3 and 41.6 years for males (10). HC was mostly seen among women both in the world and in our country (1), and it is considered to be due to domestic pet care and food preparation in houses by women (34). Ages of our cases, similar to the literature, were between 4 and 81years while the mean age was 40.55±1.066 (5-80) for women and 36.55±1.325 (4-81) for men. Our patients were mostly females (57.4% female, and 42.6% male) as well.

In the previous national studies, liver (66.4-89.3%), and lung (7.1-21.6%) were the mostly reported locations of HCs (9). Likewise, 69.8% of our cases were in liver, and 14.6% were in the lung.

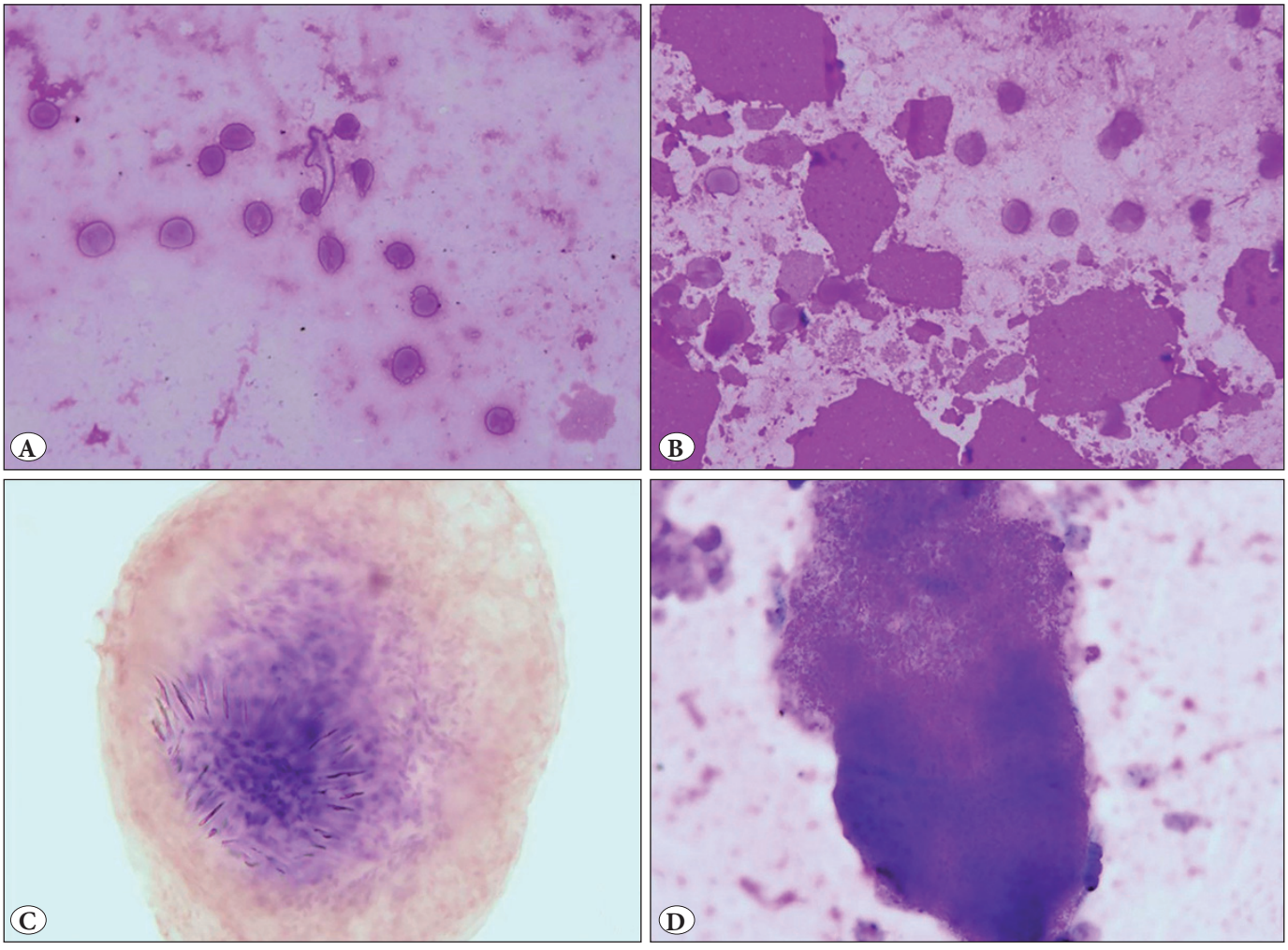

Figure 4: A) Hook structure in cyst fluid (May Grunwald Giemsa, x10). **B)** Degenerated membrane structures in cyst fluid (May Grunwald Giemsa, x20). **C-D)** Degenerated scolex in cyst fluid **C)** PAP; x100, **D)** May Grunwald Giemsa, x100).

In one study, the rate of involvement of more than one organ was reported as 6.7% (7) and in the same study, liver and lung were both involved in 2.2% of the cases (7). Our results were similar to that study: the rate of cases with a history of liver HC in the clinical history was found to be 7.5 % and 2.2 % of these cases had involvement of liver and lung.

The rate of the cases located in unusual sites in our study amounts to 15.6%. 5.3 % of these cases had a liver HC history as well. In the literature, the rate of organ involvement other than lung or liver was reported between 5.7-13.9% (4,7), and our result was similar (10.3%). In several studies, HCs of unusual locations were seen mostly in adults (7-13%) and in children the rate amounts to 7% (7). In our study, cases with unusual locations were all seen in adults but not in children. In the group with individuals aged 0-14, the most frequent involvement was seen in the lung (n:8) and then in the liver (n:4).

Although a few unusual cases of primary peritoneal involvement have been described, peritoneal HCs are almost always secondary to hepatic involvement (6). The overall prevalence of peritoneal involvement in cases of abdominal HD is approximately 13% (6). Isolated retroperitoneal HCs are also rare, and secondary to the involvement of other organs, especially liver, or in cases where there is a history of surgery (6,11). It has to be differentiated from mesenteric cyst and intestinal duplication cyst (6). Our cases with peritoneal involvement ranked in 3rd place regarding frequency (3%), and all of our cases had a history of liver HC. However, our retroperitoneal case was a secondary HC located on the left adrenal lodge.

The involvement of spleen in HC is rare. It is the 3rd most common site and the prevalence of splenic involvement ranges between 0.9% and 8% (1,7,9). Splenic HC generally develops by means of systemic dissemination

or intraperitoneal spread from a ruptured liver cyst. Isolated splenic involvement is very infrequent (6) and splenic HCs are generally solitary (6). Splenic hydatidosis should be differentiated from other splenic cystic lesions, such as epidermoid cyst, abcess, hematoma, post-traumatic pseudocyst, neoplasms like lymphangioma and haemangioma (12,13). We had 7 cases with splenic involvement (on 4th place), two cases were primary HC, and five had a previous HC.

Renal involvement is rare (1-4%) (6,7), and the frequency was found to be only 0.4% in one study (4). It is reported as the common site following liver and lung in several articles (14). They are mostly solitary and located at upper pole or cortex (6). Multilocular HCs can be misdiagnosed as simple renal cysts, cystic nephroma, and cystic variants of renal cell carcinoma (6) and infected HCs can be misdiagnosed as renal abcess (6). In our study, the renal involvement rate was 1.6%. One of our six cases was located at the right lower pole; two at the left lower pole; and two at the right upper pole. Location of one case was reported as the 'left kidney'.

Heart HC is very rare (0.02-2% of cases) and may occur owing to hematogenous spread or rupture of a lung HC (6). The cyst grows very slowly and the disease is diagnosed long after the infection (15). Diagnosis is made through echocardiography (14). The most commonly affected cardiac chambers (in decreasing order of frequency) are the left ventricle (50-60% of cases), interventricular septum (10-20%), the right ventricle (5-15%), pericardium (10-15%), and the right or left atrium (5-8%) (6,11). Nevertheless, CT scan and MR are also helpful in other parts of the body (14). We have observed six cases. All of these cases had isolated heart involvement. One case was located in the right atrium, one case in the right ventricle, and one in the myocardium. Three cases were reported as 'located in the heart'. One case (0.3%) was reported to be adjacent to the right ventricle.

Primary involvement of the pelvic cavity is very rare and patients usually manifest pressure symptoms that affect the adjacent organs (16). Commonly, cysts are detected in other parts of the body, mostly in the liver (16). Only a few cases of primary pelvic HC have been reported and primary pelvic involvement is very unusual (16). We have diagnosed 6 cases; one located at the stump line of a patient with a history of total abdominal hysterectomy with bilateral salpingo-oophorectomy. In the history of another case, the lesion was located adjacent to the urinary bladder, and the patient had operations in liver, kidney and urinary bladder with HC described. One case was at perirectal area and one was placed in the left iliac wing deviating surrounding tissues to the right. The other three cases were reported as 'located in pelvic region'.

Urinary tract and bladder involvement can occur secondary to kidney HCs (6). Primary HCs of these structures are extremely rare (6). It is observed approximately in 0.2-0.5% of the cases (17). There have been certain reports in the literature of retrovesical HC causing urinary retention (6). The rate of our urinary bladder cases was 0.8%. All of the three cases were primary ones that were located at posterior wall of urinary bladder.

Bone involvement is also rare (0.5-2.4% of cases) (6,18), and diagnosing osseous hydatidosis takes more time compared to that of other sites (19). The most commonly involved bone structures are spine (35% of cases), pelvis (21%), femur (16%), tibia (10%), ribs (6%), skull (4%), scapula (4%), humerus (2%) and fibula (2%) (6). Pericyst formation does not occur in bone, and the cyst has a much thinner wall (6). In the spine, HC bears a resemblance to tuberculosis spondylitis or chronic osteomyelitis (6), fibrous dysplasia of bone, osteosarcoma, benign cystic lesion of bone, Brown tumor (hyperparathyroidism), and various other neoplastic lesions (20). We found three cases, one with bone involvement at the right humeral shaft, and another at the right femur. The case with sacral bone involvement had a history of three operations due to spinal bone HC.

Soft-tissue HC occurs in 0.5-4.7% of patients living in endemic areas (6,18). The growth of the cyst within a muscle is difficult due to the contractility of muscles and presence of lactic acid (6,7). HC has an affinity for muscles of the neck, trunk and limbs because of increased vascularity and decreased activity of these muscle groups (6,7). Soft tissue HCs are often confused with benign soft tissue tumors (7). We had a case located in the left psoas muscle (0.3%). Our right paravertebral HC was at the T12 level, not related with muscles but in the neighborhood of the ribs.

Pancreas HCs cause diagnostic and therapeutic problems (7). Primary HC of the pancreas is uncommon, with a reported prevalence of 0.25-2% (4,6,7). Among the pancreas HCs, the head of the pancreas is more frequently involved (57%), followed by the body (24%) and the tail (19%) (21,22). It has also been reported that 9.3% of these lesions perforate in a spontaneous way or rupture into the peritoneal space (23). In the differential diagnosis, it is important that pancreatic pseudocysts and cystic malignancies of the pancreas are kept in mind (23). Both of our cases were primary HCs located in the distal pancreas.

Primary HC of the gallbladder is highly uncommon, and the incidence is 0.4% (4,24). Cysts can be located either in the lumen of the gallbladder or on its external surface (24). It is important that the gallbladder HCs are differentiated from the liver HCs and other extrahepatic cystic lesions (25). Liver hydatidosis shows a long asymptomatic period of cystic growth, whereas in primary gallbladder HC, symptoms begin at an earlier phrase, and the diagnostic imaging shows smaller cysts with deformation of the gallbladder (25). We diagnosed only one gallbladder HC case (0.3%) which coexisted with a hepatic HC.

Diagraphmatic and subdiaphragmatic locations are seldom (1%) involved, and they are mostly associated with liver disease (18,26). The diagnosis of the diaphragmatic location is frequently incidental, particularly when the cyst is small and isolated (26). Differential diagnoses include adrenal tumor, cystic lymphangioma and splenic exophytic HC (26). Our singular case (0.3%) was a primary HC located at subdiaphragmatic area, between the liver, spleen and pancreas.

A literature review has revealed that less than five cases of HC were in the inguinal canal in adults and only one case in a child (27). These cases manifest themselves mostly with swelling and the differential diagnosis of swelling includes inguinal hernia, encysted hydrocele of the cord and lipoma of the cord (27). Our case was a 30-year-old primary HC, operated due to a mass lesion at the left inguinal area.

Only a few case reports are found in the literature (28), including secondary cases as well. The most common differential diagnosis in neck swelling includes masses of thyroid, salivary gland and lymph node, all of which may be inflammatory, noninflammatory and malignant. Soft tissue sarcomas, especially the ones with rhabdomyomatous differentiation, may manifest themselves as neck swelling (28). We recognized two cases, one at the left supraclavicular area and the other at the incisura jugularis.

Incidence of cerebral HC is 0.8-4% (1,10) and 50-75% of them are at the pediatric age (7,11). Cysts are often solitary, supratentorial and intraparenchymal (7). The parietal lobe is the most frequently involved location (6,11). Our case was an adult with a HC located at occipital lobe.

Spinal HCs account for less than 1% of all HCs (6). The thoracic spine is the most frequently involved (50% of cases), followed by the lumbar (20%), sacral (20%), and cervical (10%) spine (6). Spinal HC is clustered into five groups, which are intramedullary, intradural extramedullary, extradural intraspinal, vertebral, and paravertebral (6). The first three groups of HC are uncommon (6). HCs are typically multiple when they are located in the spinal cord (6). In our study, the spinal HC rate was 0.5% (2 cases), one of which was located in thoracic intramedullary extradural, and the other in the thoracic intradural extramedullary areas.

Solitary abdominal parietal wall HC is a finding that is rare, with only 5 cases reported. All of the cases in literature had HC located either in the right iliac region or right paraumblical region (29). We also observed a case with a primary HC found at the abdominal left lateral wall.

In conclusion, the distribution of our HCs as well as our unusually located cases has been presented in this study. The results establish that HC that can be found in all parts of the body and this should always be taken into account in the differential diagnosis of cystic lesions.

CONFLICT OF INTEREST

The authors have no conflict of interest to declare.

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