Hydatid disease at unusual sites

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ABSTRACT

Hydatid disease is a cyclo-zoonotic parasitic infection caused by Echinococcus granulosus. 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. We present review of occurrence of hydatid disease at unusual sites and our experience with clinical presentation and management of hydatid disease.

Keywords: Hydatid disease, Usual sites, Unusual sites

INTRODUCTION

Hydatid disease (HD) has a wide geographic distribution and is considered an important public health problem of the world. This public health problem is influenced by the socioeconomic status of the population. Migration also spreads this disease [1]. The state of Kashmir, India, is endemic for hydatid disease [2]. 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 [3]. Human echinococcosis is caused by infection with the larval stage of genus Echinococcus granulosus and Echinococcus multilocularis [4]. Echinococcus cyst following a primary infection may inhabit any anatomic site from head to toe. The life cycle of Echinococcus involves two hosts, one definitive carnivore host (dogs, cats and certain wild carnivores) and the other intermediate herbivore host (sheep, goats, swine, small rodents and other wild herbivores). Humans act as an accidental intermediate host are infected after ingesting viable oncosphere-containing eggs, which have been shed in the faeces of the definitive host. The ingested ova penetrate the intestinal wall, reach the portal system and from there to the liver where most of them are lodged in the hepatic sinusoids [5]. A few ova may pass through the liver (first filter) and reach the lung (second filter) and then into the systemic circulation, causing hydatid disease in other organs. A possible dissemination through lymphatic channels accounts for cases with hydatid cysts at uncommon sites [4, 5]. 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%) [6].
Growth of hydatid cyst in different organs of the body is variable and depends both on patient factors, parasite factors, location of organ of cyst, host reaction and presence of any complications. The cyst has an inner germinal layer which is semitransparent, white-yellowish and an outer acellular or laminated membrane recognized grossly by its ivory white color. The external-most fibrous or adventitial layer forms as a reaction of the host to the cyst. Compressible organs such as the lung or brain facilitate the growth of the cyst. Free space around cyst usually makes easy growth of the cyst.

Diagnostic dilemma with hydatid cyst at unusual sites can lead to complications as sometimes these may present as acute surgical emergency or a chronic illness leading to morbidity [7]. Sometimes bacterial superinfection of hydatid cyst may occur. This results in pyogenic abscess formation within the cyst. Location of the cyst, type of organ, presence of intact pericyst and proneness to trauma determines the risk of infection. Infection usually develops only after rupture of both the pericyst and endocyst. Liver is the commonest site of involvement by hydatid disease and is very prone for getting infected due to Case of ascending infection via bile duct or portal vein. Secondary bacterial infection of hydatid cyst may lead to potential pitfalls in clinical presentation and radiological diagnosis. Histopathological diagnosis proves vital in such cases. Escherichia coli, viridans group streptococci and enterococci species in liver cysts and aspergillus fumigatus in lung cysts are most frequently involved in super infection of hydatid cysts [8]. Yersinia enterolitica and klebsella pneumoniae have been seen in some hepatic hydatid cysts [9]. Secondary infection of an intracranial hydatid cyst may occur due to clostridium ramosum, which is an extremely rare infectious pathogen in neurosurgical practice [10]. Rarely hydatid cysts can have spontaneous regression. In the natural course of healing, dense calcification of all components of the cyst can occur. If complete calcification of cyst is seen it signifies a dead cyst.

Diagnosis of hydatid disease is based on the patient’s history, clinical findings, serum biochemical profiles, serologic tests and pathologic diagnosis. Serologic tests usually allow hydatid cysts to be distinguished from nonparasitic cysts and abscesses. The sensitivity of various serological tests used for hydatid disease varies from 64 to 87 % [11]. The diagnosis is often difficult when hydatid cyst occurs at unusual locations as the imaging appearance varies at different sites [12]. Surgery for removal of the cystic lesion remains the mainstay of treatment and has a high success rate. Chemotherapy with benzimidazole compounds has also been used with some success to sterilize the cyst, decrease the chances of anaphylaxis, and reduce the complications and recurrence rate post-operatively. For patients who cannot undergo surgery PAIR (puncture, aspiration, injection, and re-aspiration) is indicated.

**Spleen**

The reported prevalence of splenic involvement by hydatid disease varies from 0.9% to 8% [13]. Hydatid cyst is the only parasitic cyst of the spleen and it is said to be twice as common as the non-parasitic variety. Any type of hydatid cyst can be seen in the spleen. Isolated splenic involvement itself is very uncommon. Splenic cysts are often unilocular and do not pose difficulty in diagnosis in endemic areas. Splenic hydatid generally develops after systemic dissemination or intraperitoneal spread from a ruptured liver cyst. Their imaging characteristics is similar to those of hepatic hydatid cysts. On ultrasound scan of abdomen, splenic hydatid cyst may present as a solitary, unilocular cyst or rarely as multiple well defined anechoic spherical cystic lesions or may present as anechoic spherical cystic lesions with hyperechoic marginal calcification. Computed tomography (CT) scan of the abdomen confirms the cystic lesions with or without daughter cysts within the spleen with attenuation value near that of water and does not enhance after intravenous contrast administration. On magnetic resonance imaging (MRI) the signal intensity of hydatid cysts is the same as any other similar cyst, except that the rim is of lower intensity on both T1 and T2 weighted images. Splenectomy should be the method of choice and is considered the gold standard for hydatid spleen [14]. Partial splenectomy with omentoplasty may be reserved for cases with unresectable cysts tightly adherent to adjacent structures.

**Pancreas**

Hydatid cyst of the pancreas is rare and very difficult to distinguish from cystic neoplasm of pancreas. It has been reported to affect the pancreas in 0.25% patients with hydatid disease [15]. Possible sources of infestation include hematogenous dissemination, local spread via pancreaticobiliary ducts, and peripancreatic lymphatic invasion. The head of the pancreas is the most frequent location (57%), followed by the corpus (24%) than the tail (19%) [16]. These hydatid cysts of pancreas often have severe adhesions to surrounding structures and do not communicate with pancreatic ducts. 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. Abdominal CT scan shows the presence of a cyst in the pancreas, with no enhancement on contrast. Magnetic resonance imaging shows the characteristic low-signal intensity rim of the hydatid cyst on T2–weighted images and is superior in demonstrating irregularities of the rim. These irregularities represent incipient detachment of the membranes [17]. Magnetic resonance cholangiopancreatography (MRCP) can show the communication of the cystic lesion with the pancreatic duct and helps in defining the type of surgical treatment. 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 [18].

**Kidney**

Primary hydatid of the kidney is rare entity and is responsible for only 2 to 3% of all hydatid disease [19].
Kidney involvement has insidious onset and usually remains asymptomatic for many years [20]. Renal involvement could be primary or secondary. Cysts are usually unilateral and located in the upper or lower pole. In primary hydatid disease cyst passes through the portal system into the liver and retroperitoneal lymphatics. The hydatid cyst of the kidney is considered closed if all three layers of the cyst are intact. When the cyst goes outside the pericyst confined by the lining of collecting system it is considered to be an exposed cyst. If all the three layers of the cyst have ruptured resulting in free communication with the calyces and pelvis, it is called an open or communicating cyst. Cystic rupture into the collecting system, causing hydatiduria is pathognomonic, though seen in only 10–20% of renal hydatidosis and is usually microscopic [21]. Sometimes when kidney is sectioned along the midcoronal plane it may demonstrate a large cyst with the typical “bunch of grapes” appearance due to the presence of daughter cysts. Ultrasonography aids in the diagnosis of hydatid cysts by demonstrating daughter cysts and hydatid sand. On changing the patient’s posture under real time, there is shifting of hydatid sand, which may give rise to the “falling snowflake pattern”. Excretory urography reveals a space-occupying lesion that displaces calices. CT scan usually demonstrates an expansile, hypo-attenuating tumor with a well-defined wall and daughter cysts within the parent cyst. Presence of dense calcification of the pericyst and cyst contents is common in end-stage hydatid disease and implies the death of the parasite. Removal of hydatid cyst with pericystectomy is possible in most cases. Nephrectomy is reserved for destroyed kidney.

Adrenal Gland
Hydatid cyst of the adrenal gland is rare, with an incidence of 0.06–0.18% at autopsy [22]. Most adrenal cysts are asymptomatic, they are usually found as incidental findings on imaging studies or incidentally during surgery. Hypertension, as the only symptom of the primary adrenal echinococcosis, has been reported in only one case [23]. To identify a hydatid cyst in the adrenal gland, ultrasound, CT scan and MRI can demonstrate cystic lesions and reveal daughter cysts. The definitive diagnosis is made by macroscopic and microscopic examination of the cyst. Treatment of hydatid disease of adrenals is mostly surgical and the operation of choice is removal of cyst preserving ipsilateral kidney and, if possible, the remaining adrenal gland to provide adequate haemostasis [23].

Omentum
Omentum is one of the rare sites of isolated hydatid disease [24]. Omental hydatid often reaches the pelvis and is erroneously diagnosed as pelvic cystic disease (Figure 1). CT scan and MRI is useful in diagnosis. Total excision of cyst without rupture is recommended in omental hydatid cyst.

Testis and Ovary
Testes are extremely rare sites for echinococcosis. The blood-testicular barrier, low temperature in the scrotum and different properties of the testicular tissue are suggested be the reasons of this defense mechanism [25]. Hydatid can be seen in both descended and the undescended testis. This can present as a swelling of the testis. A unilocular hydatid cyst of testis is often misdiagnosed as hydrocele or cyst of the testis. Histopathology of excised sac clinches the diagnosis.

Primary hydatid of ovary is extremely rare [26]. This can be isolated or secondary to liver hydatid. Preoperatively on ultrasonography it mimics polycystic disease of ovary or multilocular type on radiology. Daughter cysts may resemble septal structures and mimic complicated ovarian cysts and even ovarian malignancy. Histopathology confirms the diagnosis. Primary ovarian hydatid often requires oophorectomy.

Bone
Incidence of hydatid disease of bone from various studies is reported to be 0.5–4% [27]. Primary isolated bone hydatid is a very rare occurrence. The lesions in

Figure 1: Showing unilocular hydatid cyst of omentum.
bone may lie dormant for 10 to 20 years [28]. Perhaps no other bone infection is more difficult to eradicate as bone echinococcosis. Due to lack of connective tissue barrier in bone, progressively enlarging daughter cysts extend to fill the medullary cavity to a variable extent replacing the medulla [29]. About 60% cases of bone hydatidosis affect the spine and pelvis, 28% the long bone and 8% the ribs and scapula [30]. Spine is the common site of infection [31]. Hydatid disease of spine usually spreads over the spine by direct extension from pulmonary, abdominal or pelvic infestation and most commonly affects the thoracic (52%), followed by the lumbar (37%) and then the cervical and sacral spine [32]. Skeletal lesions in hydatidosis tend to present with pain or pathological fractures following trivial injuries.

The most common radiological manifestation of skeletal hydatid disease is a lucent expansile lesion with cortical thinning. Bone hydatid disease lacks a typical clinical appearance and image characteristics on X-ray or CT scan are similar to those of tuberculosis, metastases, giant cell tumour or bone cysts. [33] The primary role of CT is in the recognition of the extracavitary spread of the hydatid disease within the soft tissues which may be quite variable and may have the typical pattern of a cystic lesion seen as a round or oval area containing fluid, with sharp and thin margins, exhibiting no contrast enhancement.

Magnetic resonance imaging shows distinctive diagnostic features of bone hydatid disease, especially in the spine. The magnetic resonance imaging signal intensity pattern of the daughter cysts reflect their contents and may vary in cysts that are dead or alive. The only definitive treatment when bone is involved is complete resection of the involved area with a wide healthy margin. The combination of antihelmintic therapy, wide resection and the use of polymethylmethacrylate (PMMA) gives the best outcome in the treatment of bone hydatidosis [30].

Muscle

Primary muscular hydatid cysts are rare accounting for 3% of all patients with hydatidosis [34]. Muscular hydatidosis usually occur as isolated lesions without hepatic or pulmonary lesions [8]. These often mimic soft tissue tumors. Muscular hydatidosis is rare, because the cyst uses oxygen for growth while muscles contain lactic acid [35]. These are usually revealed as a painless enlarging soft tissue masses. Diaphragmatic localization is very rare, with an incidence of 1%, and most of these are generally associated with liver disease [36].

Magnetic resonance imaging is the examination of choice in case of suspicion of hydatid disease of muscle due to its ability to adequately demonstrate most features of hydatid disease, with the exception of calcifications. In diaphragmatic hydatid, the computed tomography findings consist of thickened and lobulated diaphragm with unilocular or multilocular cysts. These cysts may split the leaves of the diaphragma. The treatment of choice in musculoskeletal hydatid disease is surgical excision (pericystectomy), potentially combined with antihelmintic medication [37]. Percutaneous aspiration, infusion of scleroidal agents and reaspiration (PAIR), under imaging (ultrasound or CT) guidance can be used as alternative to surgery in inoperable cases. Aspiration of fluid is safe, simple and an effective means to reach a working diagnosis [38].

Brain

Cerebral involvement is very rare (1-3%), and more common in children [39]. Cerebral hydatid cysts are usually supratentorial, the infratentorial lesions are quite rare. Intracranial hydatid cysts are commonly solitary. Multiple intracranial cysts are rare [40]. Intracranial hydatid cysts may also be classified as primary or secondary. The primary cysts are formed as a result of direct infestation of the larvae in the brain without demonstrable involvement of other organs; secondary multiple cysts result from spontaneous, traumatic or surgical rupture of the primary intracranial hydatid cyst. They lack brood capsule and scolices [41]. Patients with intracranial hydatid cysts usually present with focal neurological deficit and features of raised intracranial pressure; the latter may be due to the large size or due to interference with pathway of CSF. The typical intracranial hydatid cysts caused by Echinococcus granulosus, present as a well defined solitary cystic lesions in the middle cerebral artery territory in the parietal lobes, although they can be seen in any location including skull vault, extradural, intraventricular, meningeal, posterior fossa and brainstem [42]. Operative diagnosis of hydatid cysts can be made by USG and confirmed by a CT scan. The magnetic resonance imaging is also of considerable value in intracranial hydatidosis. Surgically, intact cyst excision is the ideal treatment. Medical treatment with albendazole seems to be beneficial both pre- and post-operatively. Percystic hydraulic method (Dowling-Orlando technique) gives better results in removing these cysts intact [40, 43]. The definitive diagnosis can be made by histopathologic examination [44].

Heart and Vessels

Cardiac and vascular hydatid cysts are rare. Cardiac hydatid cysts are found in fewer than 2% of cases of hydatidosis and may present as complete heart block, constrictive pericarditis and congestive cardiac failure [45]. These cardiac and vascular cysts have a poor prognosis because of the risk of rupture and hematogenic dissemination. Echocardiography and MRI are of great value in diagnosing and determining the anatomic extent and relationship of the cyst in cardiac hydatidosis. The treatment of choice for cardiac hydatid cysts is surgical excision.

Intra-arterial deposits without any other organ involvement is an extremely rare manifestation of hydatid disease and only a few cases have been reported till date [46]. Arterial involvement of hydatid cysts usually develops after cardiac hydatid cyst rupture and embolization [47]. These hydatid cysts can also result in arterial occlusions in the aorta, iliac arteries, femoral
arteries, popliteal arteries and even the myocardial arteries [48].

Subcutaneous hydatid cyst
Primary subcutaneous hydatid cyst is very rare and the incidence is unknown. Subcutaneous hydatid cyst may be secondary or primary. The mechanism of the primary subcutaneous localization is unclear; direct spread from adjacent sites may be the mechanism of infection [49]. In secondary cysts, there is a primary location of hydatid disease like liver, lung, or spleen that may be operated previously or not operated. Subcutaneous hydatid cyst mimics cutaneous swellings. Magnetic resonance imaging is useful in confirming the diagnosis, size, localization, relationship to adjacent organs and type of the cyst. Final diagnosis is after excision followed by histopathology [50]. For the subcutaneous hydatid cyst, the treatment must always be total surgical excision.

CONCLUSION
The occurrence of Echinococcus granulosus in some locations of the body is very rare. These are anatomic locations may cause difficulties in making the diagnosis. Hydatid disease is a differential diagnosis of cystic lesions, especially for the cystic lesions encountered in patients who live in or have come from endemic region.

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