Echinococcosis: Unusual localizations and different imaging techniques for diagnostic evaluation.

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Learning objectives

Echinococcosis is the widest spread human parasitic infection in the world caused by a cestode: Echinococcus granulosus and it may develop in any part of the body. Rare sites of hydatitosis lead to atypical presentation causing difficulties in establishing the diagnosis.
Imaging techniques are the main methods of definite diagnosis and consist in ultrasound, computed tomography, and magnetic resonance imaging.
Echinococcosis (or hydatid disease) is the widest spread serious human parasitic infection in the world caused by a cestode: Echinococcus granulosus. It is endemic in areas with tropical or subtropical climates particularly in the mediterranean region, South America, Africa and Australia due to the close association between humans and domestic animals (1). Hydatid disease, common in Mediterranean countries, still remains endemic in Tunisia. Man is an accidental, intermediate host, and infection of humans represents a terminal event for the parasite. In fact, humans become infected through contact with definite host or consumption of contaminated food. After it enters the body, larval stage of Echinococcus parasite forms cysts. Since the cysts are slowly growing, the symptoms are seen several years after primary infection.

According to the literature, hydatid disease may develop in almost any part of the body. The liver is the most frequently involved organ (75%), followed by the lung (15%) and the remainder of the body (2). Primary rare localizations of parasitosis range approximately from 5% to 30% but the exact incidence of unusual locations is difficult to ascertain as they are only reported as few sporadic cases (3).
Findings and procedure details

Atypical locations of the disease included: brain, orbit, neck, mediastinum, intraperitoneal cavity, pelvis, adrenal gland, psoas, quadriceps muscles, and breast. Diagnosis was confirmed by hydatid serology, US, CT, MRI and histopathological examination of the specimen for operated patients.

Cysts may be single or multiple, uni- or multiloculated and thin- or thick walled. More specific signs include visualization or calcification of the cyst wall, the presence of daughter cysts and membrane detachment (2). CT and MR imaging, alone or in combination, are helpful in the diagnosis of hydatid disease. Although CT is superior in detecting calcification of the cyst wall or septa, MRI is better in detecting multiplicity and defining the anatomic relationship of the lesion with the adjacent structures and helps in surgical planning (5, 6).

**Brain:**

Hydatid disease affects rarely the central nervous system (1% of cases) and is more common in children (4, 5). Intracranial hydatid cysts are commonly solitary (Figure 1) and multiple cysts are rare. Intracranial hydatid cysts may also be classified as primary or secondary depending on whether other organs haven't / have been involved (4). The secondary multiple cysts result from spontaneous, traumatic or surgical rupture of the primary intracranial cyst (Figure 2). Patients with intracranial hydatid cysts usually present with focal neurological deficit and features of raised intracranial pressure.

The typical intracranial hydatid cysts present as a well defined solitary oval or cystic mass in the middle cerebral artery territory in parietal lobes, although they can be seen in any location including skull vault, extradural, intraventricular, meningeal, posterior fossa and brainstem (4).

CT and MR imaging demonstrate attenuation or signal intensity similar to that of cerebrospinal fluid (Figure 1, 3). Although the lesion may cause extrinsic compression of the ventricular system with subsequent hydrocephalus, there is no associated edema as is typically seen in abscesses and cystic tumors (5). The lesion does not enhance after intravenous administration of contrast material, and calcification is extremely rare. When present, edema and post contrast enhancement indicate ongoing inflammation. Presence of significant edema may indicate the rupture of the cyst and may be present in postoperative cases (Figure 2). Such cases are difficult to differentiate from other cystic lesions with enhancement and peripheral edema such as abscesses, large granulomas or cystic gliomas (7).

Kohli et al. (8) performed in vivo MR spectroscopy (MRS) studies in a patient with intracranial hydatid cyst and found, besides lactate, alanine and acetate, a large resonance for pyruvate. The MRS pattern appeared different from the other cystic lesions
of the brain and they have suggested MRS as an adjunct to imaging in the differential diagnosis of intracranial hydatids.

**Orbit:**

Orbital involvement occurs for 0.3% of all hydatid cysts and it is more prevalent among young patients (5, 9). Typically, an orbital hydatid cyst is unilateral and occurs with or without hydatid cysts located elsewhere in the body. It tends to involve the retrobulbar tissues either within the muscle cone or outside in the superolateral or superomedial angle. The most frequent clinical findings are proptosis, chemosis, lid edema, visual impairment, and restriction of extraocular motility (10).

Radiological findings play an important role in preoperative differential diagnosis. Orbital ultrasonography is a useful test when a "double wall sign" has been observed (10). CT of the orbital hydatid cyst demonstrates a hypodense, unilocular, well-defined homogenous mass with a hyperdense rim (Figure 4). MRI demonstrates a cystic lesion with a low signal intensity on T1-weighted images and high signal intensity on T2-weighted images, without rim enhancement after contrast medium administration (Figure 5) (11).

**Neck:**

Hydatid disease in the neck is quite rare, even in areas where the disease is endemic, with only a few cases reported in the literature (12). The diagnosis of cervical Echinococcus infection mainly depends on the clinical history of the patient, diagnostic radiological findings and serologic tests. Cervical hydatid cysts are usually slow-growing, fluctuant, painless masses.

Histopathological evaluation of the excised specimen and fine needle aspiration cytology usually leads to the diagnosis. Since puncture of the cyst may lead to an anaphylactic reaction due to spillage of hydatid fluid, the use of fine needle aspiration is controversial at present (12). Imaging modalities like Ultrasound, CT and MRI help to determine the cystic avascular nature of the lesion (Figure 6, 7). Daughter cysts, vesicles and internal septa can also be demonstrated (13).

**Mediastinum:**

Primary mediastinal hydatid cysts are quite rare. Symptoms of mediastinal hydatid cysts depend on their size and location and whether or not they exert any compression on the neighboring structures. The most serious complications include leakage of the hydatid material to the neighboring vessel, causing embolization and/or anaphylaxis with possibly fatal consequences (14).

Chest radiograph, Ultrasound, CT and MRI facilitate diagnosis. Chest radiograph oriented the diagnosis by showing a mediastinal water tone, often rounded or oval (16). Thoracic ultrasound allows confirmation of the diagnosis when the lesion is accessible and also reveals the fluid character of the opacity and, in many cases, the proliferous membrane,
pathognomonic of hydatid cysts (15). CT often accurately defines the relationship of the lesion to the adjacent structures. The most common CT finding of a mediastinal hydatid cyst is a homogenous mass with fluid (Figure 8). The presence of peripheral calcifications supports diagnosis (14). MR imaging, with its superior soft tissue resolution, can also provide us with a clear delineation of the cyst.

**Intraperitoneal and pelvis:**

Peritoneal hydatidosis is almost always secondary to hepatic disease, although some unusual cases of primary peritoneal hydatidosis have been described (2) with an overall frequency of 13% (17). Peritoneal involvement is usually undetected unless cysts are large enough to cause symptoms. Most of the cases of peritoneal hydatid disease are secondary to previous surgery for liver hydatidosis.

Accurate and rapid diagnosis of peritoneal hydatid disease is possible because of the availability of modern imaging techniques, and the surgical procedures are decided on radiological findings (Figure 9, 10). Ultrasound and CT scan are the radiological methods of choice for assessing the number of hydatid cysts in the abdomen and assessing the changes in size, number and density of lesions in response to drug therapy (18). Ultrasound is particularly useful for detection of cystic membrane, septa, and to look for hydatid sand. CT scan best demonstrates cyst wall calcification and cyst infection and enables better delineation of extent of disease (Figure 9). Thickening, calcification of wall, reduction in size and number of cysts are taken as therapeutic response on follow up (17).

**Adrenal Gland:**

Hydatid cyst of the adrenal gland is rare as this entity has been reported in only 7% of all adrenal cysts and constitutes less than 1% of all cases (19). Adrenal hydatid cysts usually form in association with generalized echinococcosis. Most adrenal cysts are asymptomatic, they are usually found as incidental findings on imaging studies or incidentally during surgery. When symptoms are present, most are related to local visceral compression. Rarely, endocrine abnormalities like arterial hypertension are seen in adrenal hydatidosis (4).

Hydatid cyst identification in the adrenal gland is based mainly on ultrasound and CT scan (Figure 11, 12). MRI shows the characteristic low signal intensity rim of the hydatid cyst on T2-weighted images. Both MR and CT images are able to show the exact anatomic extent, size, volume and position of the mass, the number of cysts, the relationship to other organs and possible complications (9). The presence of calcifications in the adrenal mass very much supports a diagnosis of hydatidosis. However, the definitive diagnosis is made by macroscopic and microscopic examination of the cyst.

**Muscle:**
Primary muscular hydatid cysts are rare, accounting for 3% of all patients with hydatidosis (4, 9).

Primary hydatid cysts of the lower extremity muscles are usually solitary and unilocular or multilocular (Figure 13). Psoas muscle is an unusual location for hydatid cyst accounting for only 1 to 3% of cases and can be unilateral or bilateral (21). Hydatid of psoas muscle can be isolated (Figure 14, 15) or associated with hydatid disease elsewhere in body.

Ultrasound is a useful tool for orienting the diagnosis of any tumefaction of the soft tissues, showing the size, localization and type of the cyst (22). CT is especially valuable for surgical planning. On CT scan they appear as a well-defined cystic lesion with daughter cysts that may contain wall calcification, septae or debris without the enhancement on intravenous contrast (Figure 14) (20). MRI 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 (9). The classic MRI findings include a multivesicular cyst, a low-intensity rim ("rim sign") on T2- weighted images or a detached membrane (Figure 13, 15). The most pathognomonic sign is that of daughter cysts within larger cysts (22).

**Breast:**

Hydatid cyst of the breast is very rare even for patients living in endemic areas and accounts for only 0.27% of all cases. It is very challenging to differentiate it from other tumoral lesions of the breast. However, it should be included in differential diagnosis of breast lumps.

Typically, the patient presents with painless breast lump, which increases slowly in size without regional lymph node involvement. It might mimic fibroadenoma, phyllodes tumors, chronic abscesses, or even carcinoma. Preoperative diagnosis can be made by fine needle aspiration cytology where scoleces, hooklets or laminated membrane can be identified. It is a safe procedure, as no complications were mentioned in the literature. The disease can be diagnosed by radiologic or serologic means, both of which are not definitive. Mammogram may show a circumscribed mass, the characteristic ring-shaped structures inside the mass in over penetrated view strongly suggests breast hydatid cyst. The ultrasound and Magnetic Resonance Imaging (Figure 16, 17) are helpful diagnostic tools. Hemagglutination tests may be helpful in diagnosis. The treatment of a hydatid cyst of the breast is complete excision. Albendazole may decrease the recurrence rate of hydatid cyst disease (23).
Fig. 1: Cerebral CT: axial (a), sagittal (b) and coronal (c) images after injection of contrast: Left temporo-parietal lobulated mass, unilocular, presenting a thin and regular rim with homogeneous density close to that of CSF, unmodified after injection of contrast, compressing the left lateral ventricle and driving back the midline structures to the right.

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Fig. 2: Cerebral CT : axial images (a, b) after injection of contrast: Postoperative oedemato-hemorrhagic alterations associated with daughter cysts in the left temporal lobe.

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Fig. 3: Cerebral MRI: (a) axial unenhanced T1-weighted image, (b) axial T2 FLAIR - weighted image, (c) axial and (d) coronal contrast-enhanced T1-weighted images: pure fluid mass with low-signal-intensity on T1 and high signal-intensity on T2, canceling completely on FLAIR, surrounded by a thin rim discreetly hyperintense on T1 and hypointense on T2, unmodified after injection of Gadolinium and associated with perilesional edema. This lesion excerts a mass effect on the median structures. Furthermore, extra-dural left frontal abscess with fluid collections of the scalp.

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Fig. 4: Orbito-cerebral CT : axial images before (a) and after (b) injection of contrast: left intra-orbital hypodense, unilocular and homogenous mass with a hyperdense rim, unenhanced after injection of contrast.

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Fig. 5: Orbito-cerebral MRI: (a) axial and (b) coronal fat-saturated T2-weighted images, axial (c) unenhanced and (d) contrast-enhanced T1-weighted images: left intra-orbital cystic lesion with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images, without rim enhancement after injection of Gadolinium.

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Fig. 6: Ultrasound axial image: left basi-cervical cystic mass, rounded, heterogeneous and avascular on Doppler, measuring 10 cm, with posterior enhancement of echos.

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Fig. 7: Cervical CT: axial images before (a) and after (b) injection of contrast: subcutaneous cystic mass, thin-waled, unenhanced after injection of contrast and containing a serpiginous structure realizing the aspect of a detached membrane.

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**Fig. 8:** Thoracic unenhanced CT: axial images (a, b): homogenous thin-walled mediastinal mass with fluid density and thin parietal calcifications, coming into contact with the right edge of the heart.

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Fig. 9: Contrast-enhanced abdominal CT: axial images (a, b, c, d): multiple cystic masses, multi-compartmentalised, unmodified after injection of PDC and localized in the mesentery (a, b, c) and the pelvis (d). Note the presence of a thin parietal calcification (a).

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**Fig. 10:** Pelvic MRI: (a) axial, (b) coronal and (c) sagittal T2-weighted images: Cystic pelvic mass, multi-compartmentalised with high signal intensity on T2-weighted images, and presenting a thin hypointense rim.

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**Fig. 11:** Ultrasound axial image: multi-compartimentalised anechoic mass with posterior enhancement of echos.

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**Fig. 12:** Contrast-enhanced abdominal CT: axial images (a, b): large thin-walled cystic mass, with homogeneous content, driving back the right hepatic lobe.

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Fig. 13: MRI of the thigh: coronal unenhanced T1-weighted image (a) and fat-saturated T2-weighted image (b), axial T2-weighted (c) and contrast-enhanced T1-weighted (d) images: a solitary muscular cystic mass, hypointense on T1-weighted images and hyperintense on T2-weighted images with a low-intensity rim on T2-weighted images and a detached membrane, without enhancement after injection of Gadolinium.

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**Fig. 14:** Contrast-enhanced abdominal CT: axial images (a, b): oval and thin-walled cystic lesions, multi-vesicular, with heterogeneous density, located in the right psoas muscle.

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**Fig. 15:** MRI of the psoas muscles: coronal (a) unenhanced and (b) contrast-enhanced T1-weighted images, (c) coronal and (d) sagittal fat-saturated T2-weighted images: multiple multi-vesicular cystic lesions, hypointense on T1-weighted images and hyperintense on T2-weighted images, without enhancement after injection of Gadolinium, located in the right psoas muscle. Note the hypointense signal on T1- and T2-weighted images of the internal partitions and the rim.

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**Fig. 16:** Axial T2-weighted MR images show a hyperintense cystic lesion in the left breast with a well-defined borders in low signal intensity "rim sign". Inside there is a serpiginous structure which represents the collapsed parasitic membranes "serpent sign".

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**Fig. 17:** Axial T1 weighted MR images showing a hydatic cyst in the left breast with low signal intensity.

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Conclusion

Hydatid disease should be considered in the differential diagnosis of the all cystic masses in all anatomic locations, especially when they occur in areas where the disease is endemic. The combination of clinical history, imaging finding, and serologic test results usually help the diagnosis.
References


