Evaluation of imaging features of pelvic echinococcosis based on multimodal images

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Abstract

Background: Hydatid disease (HD) is a zoonotic parasitic disease caused by the larvae of Echinococcus, It is mainly prevalent in pastoral areas. Bone echinococcosis is rare, accounting for 0.5 to 4.0% of all echinococcosis. It is likely to miss the diagnosis and misdiagnose due to non-specific early symptoms and the clinical manifestations and imaging features. The clinical data of 15 patients with pelvic cystic echinococcosis were analyzed retrospectively, and the X-ray, CT, and MRI imaging features of the disease were discussed, which are reported below.

Methods: All 15 patients underwent CT scan evaluation. A total of 8 patients underwent coronal, sagittal, and three-dimensional reconstruction with 3-mm-slice thickness, and 4 patients underwent X-ray plain film examination. Five cases underwent MRI scan. Eight cases underwent MRI or CT enhanced scan.

Results: X-ray plain film is characterized by continuous cystic bone destruction, irregular low-density shadow when invading soft tissue, and sometimes calcification which can be seen on the wall or inside the cyst. The involved sacroiliac joint or hip joint may narrow or disappear. The involvement of pelvic cystic echinococcosis is relatively wide, and 80% of patients with pelvic cysts in this group had multiple lesions in the same period. Cystic expansive bone destruction was the most common. Pelvic CT revealed a lobulated hypodense lesion of varying size with internal septae, causing cortical thinning and destruction. Most of them had no periosteal reaction. The iliopsoas muscle is most easily invaded. Single cystic echinococcosis of pelvis showed intermediate or low signal intensity on T1-weighted images and hyperintensity on T2-weighted images in the involved bone and surrounding soft tissue on MRI, and the cyst wall showed linear low signal in T1WI, T2WI, and STIR sequences. The polycystic type is characterized by multiple cysts of varying signal intensity (daughter cysts) within a larger cyst is the typical MRI finding, forming “small vesicles” high signal daughter cysts. Osteosclerosis or calcification showed low signal in T1WI and T2WI.

Conclusions: The results of this study suggest that the lesions of pelvic cystic echinococcosis are mostly cystic expansive and osteolytic bone destruction, which is easy to invade the surrounding soft tissue, often accompanied with calcification; among them, multiple cystic lesions are characteristic.

Keywords: Pelvis, Echinococcosis, Computed tomography, Magnetic resonance imaging, X-ray computed, Management

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Background
Hydatid disease (HD) is a zoonotic parasitic disease caused by the larvae of Echinococcus, also known as echinococcosis. It is mainly prevalent in pastoral areas, especially in Xinjiang, Qinghai, Ningxia, Gansu, Inner Mongolia, and Tibet. Bone echinococcosis is rare, accounting for 0.5 to 4.0% of all echinococcosis [1, 2]. It is likely to miss the diagnosis and misdiagnose due to nonspecific early symptoms, and the clinical manifestations and imaging features are similar to those of bone tumor, bone cyst, and other diseases. The clinical data of 15 patients with pelvic cystic echinococcosis were analyzed retrospectively, and the X-ray, CT, and MRI imaging features of the disease were discussed, which are reported below.

Materials and methods
General information
Altogether, 15 patients with pelvic cystic echinococcosis were confirmed by histopathological examination or clinical and imaging examination in the first affiliated hospital of Xinjiang Medical University from February 2011 to June 2019, including 10 males and 5 females, aged 19–58 (37.7 ± 9.8) years. There were 4 cases of Kazak and 4 cases of Han nationality, 2 cases of Uygur, Hui and Mongolian each, and 1 case of Kirgiz. Fourteen cases had lived in Xinjiang for a long time, and one case had lived in Qinghai for a long time, with a course of 1–20 years. Clinical manifestations are the following: pain in the hip, lower limbs, and lumbosacral region in 5 cases and limitation of movement in 5 cases (including 1 case with nerve compression symptoms such as incontinence). Local infection occurred in 7 cases (1 case of subcutaneous mass of buttock, 4 cases of sinus formation after operation, 2 cases of non-healing incision). There were 1 case of chronic osteomyelitis, 3 cases of pathological fracture, and no symptoms in 2 cases. There were 7 cases of echinococcosis with other systems, 4 cases of pulmonary echinococcosis, 4 cases of hepatic echinococcosis, and 1 case of prostate echinococcosis. Among them, there were 7 cases confirmed according to the clinical history of echinococcosis combined with laboratory examination and imaging findings, and 8 cases were confirmed by histopathological examination.

Methods
To record the imaging data of the patients according to the medical records, all 15 patients underwent CT scan evaluation (GE 16- or 64-slice CT machine). A total of 8 patients underwent coronal, sagittal, and three-dimensional reconstruction with 3-mm-slice thickness, and 4 patients underwent X-ray plain film (Philips DR system) examination. Five cases underwent MRI scan (Siemens 1.5T Avanto MR scanner). The scanning sequence included T1WI, T2WI, and short time inversion recovery (STIR); 8 cases underwent MRI or CT enhanced scan (MRI enhanced in 3 cases, CT enhanced in 5 cases), with iohexol (300 mg I/ml), 1.5 ml/kg, and injection flow rate of 3.0 ml/s; MRI enhanced contrast agent was GD DTPA, 0.2 ml/kg, injection flow rate 2.0 ml/s.

Results
Location and number of lesions
In 15 cases, sacrum, ilium, pubis, ischia, acetabulum, sacroiliac joint, hip joint, and soft tissue of surrounding muscles were the main affected parts. There were 4 cases of echinococcosis in the same side of femur and 3 cases of echinococcosis in the lumbar and sacral vertebra. The lesions occurred in 7 cases of the right pelvis, 6 cases of the left pelvis, and 2 cases of the bilateral pelvis. There were 38 lesions involving 3 or more parts in 7 cases, and 13 lesions invaded the surrounding soft tissue.

Imaging findings
X-ray findings
All the 4 patients underwent X-ray examination showed multiple bone destruction, including 2 cases of single cystic expansive bone destruction, 1 case of irregular bone destruction with small bone shadow on the edge, and 1 case of cystic expansive bone destruction with patchy bone destruction; 4 cases of marginal osteosclerosis without periosteal reaction (Fig. 1). There were 4 cases of irregular low-density shadow in the surrounding soft tissue, 2 cases of irregular calcification shadow in
the soft tissue, and sacroiliac joint narrowing in 3 cases, showing hyperostosis in the lower edge of sacroiliac joint and blurred articular surface.

**CT findings**

CT scan was performed in all 15 cases, and different degrees of bone morphological changes after the removal of bone echinococcosis were found in 14 cases. Among them, there were 9 cases of cystic expansive bone destruction and thinning of bone cortex, and no periosteal reaction can be seen in the expansion area (Fig. 2), osteolytic bone destruction in 3 cases, erosive bone destruction in 3 cases (Figs. 3 and 4), and cystic expansive bone destruction and osteolytic bone destruction occurred in 1 case simultaneously. The lesions invaded the surrounding soft tissues (iliac muscle, iliopsoas, gluteal muscles, erector spinae muscle, internal obturator muscle, etc.) in 13 cases. There were 10 cases of single cystic type and 4 cases of multicystic type. On plain CT scan, the boundary of the cyst showed round water-like, well-defined shadow, and the daughter cyst in the multicystic mother cyst showed septal round-like, low-density shadow, calcification inside the cyst or cyst wall in 9 cases, gravel-sand-like calcification in the cyst wall in 3 cases, shell-like calcification in 2 cases, irregular calcification in soft tissue in 4 cases. There were 7 cases of sacroiliac joint narrowing, 2 cases of sacroiliac joint disappearance, and 2 cases of multiple cystic lesions of sacroiliac joint.

**MRI findings**

A total of 5 patients underwent MRI examination. Among them, there were 2 cases of cystic expansive bone destruction, 2 cases of osteolytic bone destruction, and 1 case of erosive bone destruction. The lesions involved multiple parts. There were 3 patients of single cyst type, which showed intermediate or low signal intensity on T1-weighted images and hyperintensity on T2-weighted images, while in 2 cases of multiple cyst
type, the mother capsule was filled with septate daughter cysts, and the signal intensity of daughter cysts was higher (Figs. 5 and 6). There was one case of arc calcification of cyst wall with hyperintensity on T1-weighted and T2-weighted. In one case of postoperative recurrent infection, MRI showed long T1 and mixed long T2 signal in the sacrum subcutaneous tissue, and the subcutaneous fat showed obvious heterogeneous enhancement. CT or MRI enhancement features
In CT enhanced scan, there were no enhancement in 3 cases, a slight enhancement in 2 cases, and a linear enhancement of the cyst wall in 2 cases. In contrast-enhanced MRI scan, 3 cases showed slight linear enhancement at the edge of the lesion and partial septal enhancement and 1 case showed obvious heterogeneous enhancement of subcutaneous fat in the sacrum (indicating the presence of secondary infection).

Discussion
Clinical features and epidemic
Echinococcus granulosus is the main pathogen leading to hydatid disease in the pastoral areas of China, which seriously endangers human health and the development of animal husbandry. Granulosus has a carnivorous animal as a definitive host and an intermediate host such as sheep, cattle, and other herbivores. Humans are incidental hosts due to ingestion of the parasitic eggs from contaminated food or water. The larvae released from the eggs penetrate the intestinal wall and migrate to various organs. In addition to having a clear history of pastoral contact, a relatively higher prevalence has been noted in the rural populations that is mainly attributable to the slaughtering of animals in these areas [1]. In this study, we found that three patients were shepherds with a definite epidemiological history, and nine patients were all farmers. Pelvic bone disease is particularly difficult to diagnose, given the vague symptomatology and typically delayed clinical presentation. Hydatid lesions grow slowly; when located in the bone marrow cavity, they can spread along the cancellous bone to form bone destruction and cause pathological fracture. The lesions can penetrate through the bone cortex and invade the surrounding soft tissue to form a mass. When they break to the skin, chronic suppurative osteomyelitis can be developed by the formation of long-term unhealed fistulas and the outflow of hydatid detritus and pus. If the joint is involved, it can cause pathological dislocation. However, by the time clinical symptoms appear, such as pain, swelling, abnormal walking, incontinence due to nerve compression, paraplegia, and other related symptoms, the lesion is already large. Therefore, this disease warrants awareness and updated knowledge on part of clinicians, especially in endemic areas when patients present with generalized musculoskeletal complaints. Early radiologic investigations, laboratory evaluation, such as serological detection, and histopathologic analysis of the biopsy specimen are of paramount importance [3]. Serological tests are important in differential diagnosis. The immunoreaction of the human body is related to the hydatid cyst’s integrity, growth vigour, and location. Immunoreaction is heavier in ruptured hydatid cysts and lower when intact. Serological tests are frequently negative when the hydatid cyst is ageing, calcified, or dead. At present, the serologic examinations used in diagnosing hydatid cyst disease are classified into two categories. One is the detection of antigen from the hydatid fluid and protoscoleces, and the main antigenic components are Ag 5
EcgF antibody, and anti-Em2 antibody were all positive. Routin
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Imaging features

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intensity (daughter cysts) within a larger cyst is the typi-
cal MRI finding, forming “small vesicles” high signal
daughter cysts [6]. Daughter cysts can be seen as cystic
structures attached to the germinal layer that are
hypointense relative to the intracystic fluid on T1-
weighted images and hyperintense on T2-weighted im-
ages. After enhanced scanning, it could be marginal lin-
ear enhanced or septal partial enhanced, but there was
no obvious enhancement inside the lesions. Osteosclero-
sis or calcification showed low signal in T1WI and
T2WI. In addition, MRI has an important value for the
ruptured infection of echinococcosis due to its good soft
tissue resolution. When hydatid cysts are ruptured or in-
fected, the signals of T1WI and T2WI are significantly
enhanced due to the increase of protein content, espe-
cially T2WI, and the boundary of the cyst changed from
sharp to blurred. It has been reported in the literature
[5] that MRH (magnetic resonance hydrography) can
clearly show the structure of the sinus when the lesion
ruptures outside the skin to form a non-healing fistula
and help to accurately assess the number of daughter
cysts and degree of the disease.

Management
In terms of management, hydatid disease of the pelvic
bone is particularly a serious clinopathologic entity as
the cyst in this location may invade pelvic joints, which
can potentially make complete recovery difficult. Al-
though the definitive treatment of bony hydatidosis is
surgery, a number of studies have highlighted the com-
bination of antihelminthic chemotherapy and surgery as
a feasible choice [2]. In the published medical literature,
several surgical methods, including simple drainage or
debridement, complete excision, total hip arthroplasty,
bone grafting, and hemipelvectomy have been reported
thus far [8]. Furthermore, bone cement filling is a reli-
able option to avoid the relapse of the cystic lesions due
to its ability to kill the daughter cysts due to necrotizing
effects of increased temperature in the polymerizing ce-
ment [9]. Among the collected cases, most of the pa-
tients were removed the lesion thoroughly and filled it
with bone cement and took albendazole before and after
operation. Hemipelvectomy was performed in patients
with extensive pelvic bone invasion on one side. Fortu-
nately, no recurrence has been observed since the oper-
ation in 2015. A patient who had a proximal fracture of
the femur was performed bone cyst removal and tumor-
type prosthesis replacement. Three patients received
radiotherapy after surgery. Albendazole was used in the
postoperative management of all patients. Chemotherapy
using mebendazole or albendazole as only treatment is
not adequate in most patients. However, it can be em-
ployed as neoadjuvant therapy to shrink the cyst load
before surgery and/or as adjuvant therapy to decrease
the recurrence risk [10]. However, the major goal of
these procedures is to restore the limb function rather
than complete eradication of the infectious etiology due
to echinococcosis. In this study, most of them had a his-
tory of bone hydatid disease and had undergone at least
one surgical treatment. Only 4 cases were secondary to
liver hydatid or lung hydatid. Therefore, clinicians
should perform a lifelong follow-up for early detection
of potential recurrence and sequels.

Differential diagnosis
Aneurysmal bone cyst, giant cell tumor, and bone tuber-
culosis can be considered as potential differential diag-
noses in this case [3, 11]. The most common of these
underlying lesions is the giant cell tumor. On conven-
tional radiographs, giant cell tumors appear as eccentric
epiphysal lytic lesions with nonsclerotic but well-
defined borders. MRI features are not specific, inter-
mediate, or low signal intensity on T1-weighted images,
and hyperintensity on T2-weighted images are common.
Enhancement after intravenous gadolinium chelate chelate
administration may be seen. The aneurysmal bone cyst can
be seen as an expanding lytic mass is seen on radio-
graphs and CT images. It can appear a stepped liquid-
liquid plane and diverticulum processes of different
sizes. Bone tuberculosis generally invades the vertebral
body and intervertebral disc at the same time, while
bone echinococcosis often involves the paraspinal soft
tissue on one side, and the intervertebral disc is rarely.

Conclusion
The results of this study suggest that the lesions of pel-
vic cystic echinococcosis are mostly cystic expansive
and osteolytic bone destruction, which is easy to invade
the surrounding soft tissue, often accompanied with calcifi-
cation. Among them, multiple cystic lesions are charac-
teristic, which can improve the early diagnosis rate and
achieve the purpose of early treatment.

Abbreviations
MRI: Magnetic resonance imaging; CT: Computed tomography;
MHR: Magnetic resonance hydrography; STIR: Short time inversion recovery

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Authors’ contributions
Chen Yuwei was responsible for the data analysis and manuscript writing. AI
ERKEN alkebaier contributed to the data collection and language editing. All
authors read and approved the final manuscript.

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Consent for publication
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All authors have seen the manuscript and approved it to submit to your journal.

Competing interests
The authors declare that they have no competing interests.

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