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IMAGING DIAGNOSIS OF MUSCULOSKELETAL HYDATID DISEASE

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Abstract: Hydatid disease has a worldwide distribution and causes health problems in endemic countries. Musculoskeletal hydatid disease may be a difficult differential diagnosis. The aim of this retrospective study is to present some different imaging features of musculoskeletal hydatid disease.

We evaluated imaging findings of 15 patients with musculoskeletal hydatid disease over a 15-year period. Ten (67%) were men and 5 (33%) women, ages ranging between 12 and 69 years. All underwent radiographic evaluation. CT was done in 9 patients and MRI in 3 patients with soft tissue involvement. All underwent surgery, followed by histological examination.

Bone involvement occurred in 11 patients and soft-tissue involvement in 4 patients respectively. Bone lesions in the spine were found in 4 (36%), in the pelvis 2 (18%), in the proximal femur 2 (18%), in the tibia 2 (18%) and 1 (9%) in the first metatarsal. On plain film we identified three different patterns of presentation of bone HD, defined as "typical" osteolytic lesion (33%), "tumour-like" features (41.6%) and lesions resembling infection (25%). Soft-tissue hydatidosis was localized on the distal part of the thigh, paravertebral and deltoid muscle. MR imaging showed a cystic mass, containing multiple vesicles, and in one of them the pathognomonic for hydatid disease, the

"water lily" sign, was present. We present three cases of primary musculoskeletal hydatid disease localized on metatarsal bone, proximal femur and deltoid muscle.

Musculoskeletal hydatid disease, presenting with a variety of patterns, may resemble a tumour or an inflammatory process. Because of the rare presentation of the disease it should be kept in mind in the diagnostic work-up of musculoskeletal tumours.

Key words: hydatid cyst, soft tissue, bone, water-lily sign, magnetic resonance imaging, primary musculoskeletal.

Introduction

Hydatid disease (HD) has a worldwide distribution and causes health problems in endemic countries such as the Mediterranean region, Central Asia, East Africa, Australia and New Zealand [1]. This is a zoonotic infection involving larval forms of small tapeworms of *Echinococcus granulosus*. This parasite inhabits the small intestine of carnivores, such as dogs and wolves, the definitive hosts in the cycle [2]. Humans are known to be accidental intermediate hosts for *Echinococcus* organisms.

Hydatid disease mostly affects the hepatic (50–77%) and pulmonary (8.5–43%) regions, and occurs in only 10–15% in other regions of the body [3, 4, 5, 6]. Skeletal involvement is seen in only 1–4% of cases [2]. Muscular hydatid cysts have a prevalence of primary presentation of only 0.5–4% [7, 8, 9].

The musculoskeletal hydatid disease may mimic benign or malignant cystic tumours, metastases, congenital cysts, abscesses, pseudocysts, empyemas, infarcts, haematomas and other lesions [5, 10]. Preoperative diagnosis of hydatid disease is essential to avoid rupture and dissemination of the cyst which may lead to recurrence, and intraoparative spillage of the antigenic cyst fluid may lead to a severe anaphylactic response.

The aim of this retrospective study is to present some different imaging features of musculoskeletal hydatid disease.

Materials and methods

Retrospective imaging findings of 15 patients with histologically proved bone and soft-tissue involvement of hydatid disease were evaluated. The patients were treated at the Clinic for Orthopedic Surgery, primarily understood as a bone or soft tissue tumour, over a 15-year period. The cases with soft tissue localization of hydatid disease were detected in the last 1.5 year period.

Ten (67%) were men and 5 (33%) women, of ages ranging between 12 and 69 years.

In all cases plain films of the affected region (bone and soft-tissue structures) were available. Computed tomography (CT) was done in 9 patients and magnetic resonance imaging (MRI) in three. One case with hydatid disease of the *m. deltoideus* was diagnosed at the operation. Localization and characteristics of the lesion were analysed on CT and MRI.

Involvement of other organs, such as the liver and lung, was ruled out with sonography and chest radiography, respectively. All underwent surgery, followed by histological examination.

Results

Of the 15 patients, bone involvement occurred in 11 patients and four had soft-tissue involvement. One of the cases with soft tissue localization of hydatid disease, in paravertebral muscles, was secondary to the vertebral affection.

The clinical presentation consisted of slow-growing masses; pain (n = 10/15); fever (n = 1) or weakness (n = 3). In 3/15 cases the disease had primary musculoskeletal localization: the metatarsal bone, proximal femur and deltoid muscle. Serology for hydatidosis was positive in all cases.

Bone lesions in the spine were found in 4 patients (36.4%), in the pelvis 2 (18.2%), in the proximal femur 2 (18.2%) patients, in the tibia 2 (18.2%) patients and in 1 (9%) patient in the first metatarsal bone. Soft tissue lesions were localized intramuscular, two on the distal part of the thigh, one within paravertebral muscles and one in deltoid muscle.

Findings on plain film of bone HD were categorized into three groups: the first was "typical" – a rounded osteolytic lesion in four patients (33%); the second was "tumour-like" features, presented in 5 (41,6%) patients, and the third was lesions resembling infection in 3 (25%) patients (Fig.1a). The cases with soft-tissue localization of the thigh presented like the shadow of a soft tissue tumor, without calcification of the cyst wall.

CT contributed to a better evaluation of the extension within the bone with a clear demarcation of the lesion (Fig. 1b). CT of the cases with soft-tissue involvement of hydatid cyst was presented as a hypodense oval lesion within the muscles, without clear demarcation of the daughter cysts (Fig. 1c, 2a), without any calcification. Destruction of the vertebra was seen in the patient with involvement of paravertebral muscle. The bone was normal in the cases with soft tissue involvement of the thigh.

MR imaging was performed in both cases of soft-tissue lesions in the thigh and one in the paravertebral muscle. On MRI at the level of the third lumbar vertebra, on the T1-weighted image a low signal intensity on the ver-

tebral body and pedicle can be seen, with cysts within the paravertebral muscle. On sagittal and coronal T2-weighted images of the lumbar spine and the paravertebral muscle using high signal intensity the lesion of the vertebra can be seen, and a lot of cysts within the paravertebral soft tissue (Fig. 1f, g). On T1- and T2-weighted images, a well-defined oval cystic mass in the soft tissue can be seen. One of the cases localized on the thigh contained round-shaped daughter cysts (Fig. 2) which were well delineated on MRI (2b, c, g). A continuous low intensity rim could be seen in T2-weighted images (Fig. 2c). The fluid in the daughter cysts showed slight hypointensity compared to the main cyst in our case on T1 sequence. There was no sign of wall thickening, but detached germinative membranes were well presented as a hypointense band within the cysts in T2-weighted images (Fig. 2c) and SPIR images (Fig. 2d, f), which is pathognomonic for hydatid disease, the so-called "water-lily" sign. T1-weighted images, after contrast, depict the wall enhancement (Fig. 2g).

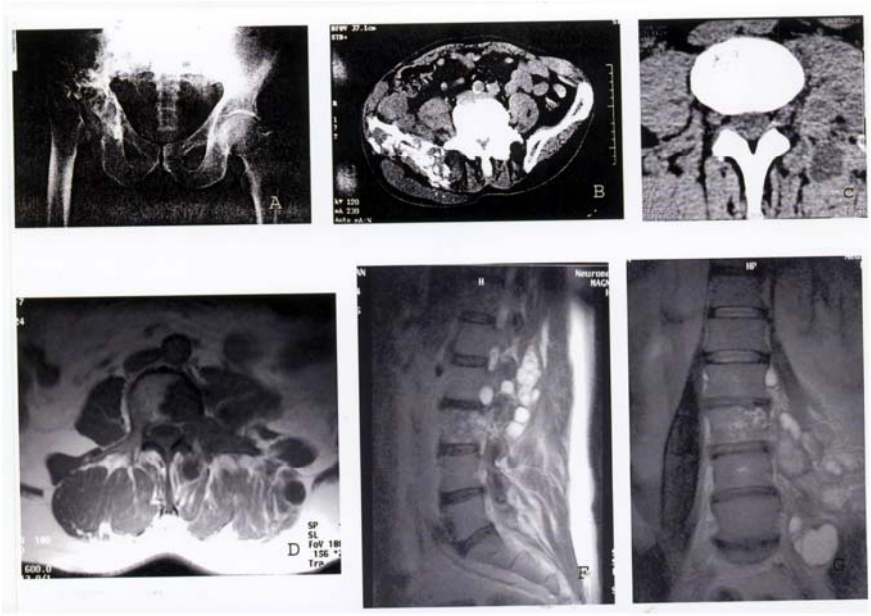

Figure 1 – A*) plain film of the pelvis showed a "tumour-like" lesion of the right iliac bone; B) CT obtained at the iliac crest demonstrated destruction of the right iliac bone with a multiloculated small cystic mass partly surrounded by sclerotic changes of the bone; C) CT at lumbar vertebra, hypodense oval soft – tissue lesions within left paravertebral muscles; D) T1 weighted image: hyposignal of the L3 vertebra with affection of the body and pedicle of the vertebra. Round lesion in muscle-hydatid cyst; F) and G) T2 weighted images, sagittal and coronal, of the lumbar spine with paravertebral muscle. High signal intensity can be seen in the lesion of the vertebra, and a lot of cysts within the paravertebral soft tissue.*

Слика 1 –А) Рентгенографија на карлица прикажува лезија која личи на тумор на десната илијачна коска; В) Компјутеризирана томографија (КТ) направена на ниво на илијачното крило прикажува деструкција на десната илијачна коска со мултилокуларни мали цистични лезии опкружени од склеротични промени на коската; С) КТ на лумбален пршлен – хиподензна овална мекоткивна промена во паравертебралната мускулатура лево; D) Магнетна резонанца (МР)-Т1 пулс секвенца: хипосигнал на Л3 пршлен со зафаќање на телото и педиклот. Округла лезија во мускулатурата – хидатидна циста; F) и G) МР-Т2 пулс секвенца, сагитална и коронална рамнина, на лумбален 'рбет со паравертебралната мускулатура. Зголемен интензитет на сигналот се гледа на лезијата на пршленот и многу цисти во паравертебралното меко ткиво

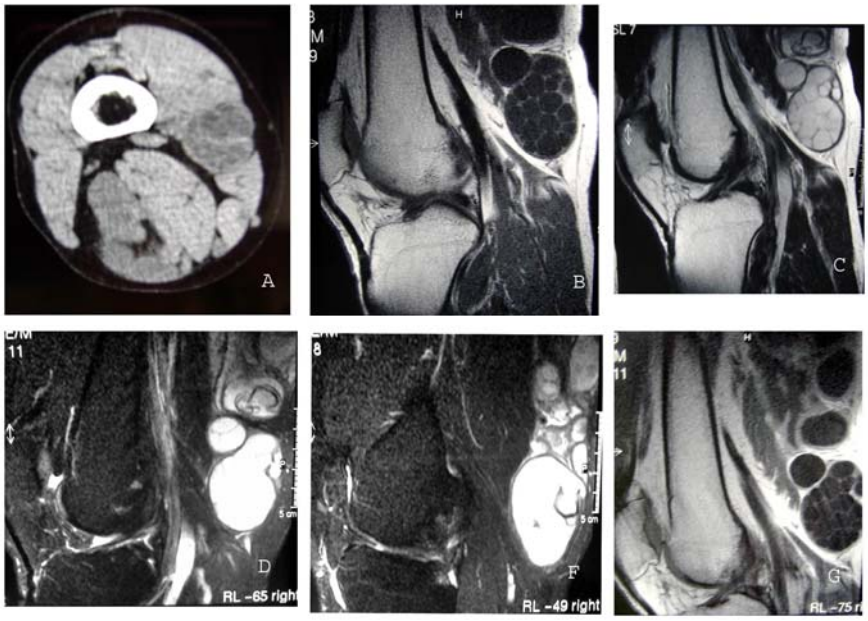

Figure 2 – A) CT of thigh: a lobulated hypodense lesion within the muscle; B) T1-weighted image, sagittal plane: hypodense oval soft-tissue lesions with daughter cyst and one round antero-superior; C) T2-weighted image, sagittal plane: the same lesion with a continuous low intensity rim. Within the cyst localized above, the pathognomonic "water-lily" sign is present; D) and F) SPIR image: clearly show the detached germinative membranes of the "water-lily" sign; G) T1-weighted images after contrast, depicts the wall enhancement.

Слика 2 –А) КТ на надколеница: лобулирани хиподензни лезии во мускулот; В) МР-Т1 пулс секвенца, сагитална рамнина: хиподензна овална мекоткивна лезија со цисти ќерки и една округла циста енгеро-супериорно; С) МР-Т2 пулс секвенца, сагитална рамнина: истата лезија со континуиран раб со намален сигнал. Во цистата локализирана

над неа присутен е патогномоничен "water lily" знак; D) и F) МР-СПИР секвенца: јасно прикажува одлепената германативна мембрана со "water lily" знак; G) МР-Т1 пулс секвенца по контраст, прикажува пребојување на ѕидот

Three cases showed primary manifestation of musculoskeletal hydatid disease. One case was localized on the first metatarsal bone, one on the proximal femur and one within the deltoid muscle.

Concomitant hepatic hydatid disease was seen in 7 patients, pulmonary hydatid disease in one patient and both hepatic and pulmonary hydatid disease in 4 patients.

The treatment of choice in our cases was surgical removal of the intact cyst and curettage if that was not possible.

Discussion

Primary localization of hydatid disease on the musculoskeletal system is very rare. We present three cases of hydatid disease primary localized on the first metatarsal bone, on the proximal femur and in the deltoid muscle. We describe a case with soft tissue hydatid disease with the pathognomonic "water-lily" sign.

Musculoskeletal infection by *Echinococcus multilocularis* is rare, but has a more invasive nature, sometimes mimicking a malignant lesion [3]. A preoperative radiological work-up may reveal the characteristic features of a hydatid cyst, but radiological findings can range from purely cystic lesion to a completely solid appearance [3].

Cystic HD affecting the bones is a rare condition, with the location distributed as follows: 35% in the spine, 21% in the pelvis, 16% in the femur, 10% in the tibia, 6% in the ribs, 4% in the skull, 4% in the scapula, 2% in the humerus, and 2% in the fibula [2]. Of the cases that we present 36 per cent of bone lesions had a vertebral localization, and the pelvis, proximal femur and tibia 18% each. We have one case with primary localization on the first metatarsal bone which, to our knowledge, has not previously been reported.

There is a minimal host reaction in the bone, and as a result the cyst has a much thinner wall. This fact, together with the rigid nature of the bone, prevents the cyst from forming its typical spherical shape [11]. The cyst shows slow growth in a direction of low resistance and, if destroys the cortex, causing the spread of HD into the surrounding soft tissue and adjacent part of the skeleton [12]. We had one case of soft tissue spread in the paravertebral muscle from the vertebral hydatid disease.

Muscular hydatid cysts are rare [5], with the prevalence of primary presentation being only 0.5–4% [7, 8,] because muscle is an unfavourable site for infestation as a result of its high levels of lactic acid. The most frequent localizations were paravertebral, pelvic-gluteus and the lower extremity [7]. In our study we present two cases of soft tissue hydatid disease with muscular localization on the thigh, one in the paravertebral muscles and one case within deltoid muscle. This last was misunderstood as being a benign soft tissue tumour, and was diagnosed at the operation.

Preoperative diagnosis of echinococcosis is necessary to prevent rupture of the cyst and to avoid the resulting anaphylactic shock and local recurrence [13, 14, 15, 16]. Serology alone is insufficient to diagnose echinococcosis [7]. The diagnosis is based on the identification of a hydatid cyst in tissue. Imaging modalities such as ultrasound and CT may reveal a calcified cyst wall and microcalcification in daugher cysts and varying fluid densities between cysts and surrounding organs [1]. The CT appearance of bone lesions is similar to those demonstrated on plain film. A well-defined, typically multiloculated, osteolytic lesion sometimes with coarse trabeculae within it is usually seen, giving a honeycomb appearance, which is accompanied by expansion of the bone and thinning of its cortex [17]. The primary role of CT is in the recognition of the extraosseous spread of HD within soft tissue. The association of bone lesions with soft tissue calcification allows a reliable diagnosis of HD [17]. In spinal HD the most common differential diagnosis problem is tuberculous spondilitis. The absence of damage to the disc surface of the vertebral bodies and the spread of the disease through a subperiosteal and subligamentous path are typical of vertebral HD [10].

A palpable mass is the most constant clinical finding of hydatid disease affecting soft tissue. The clinical manifestations are caused by compression of the organ involved. The CT findings are variable, including a unilocular cyst, a multivesicular lesion, or an atypical complex or solid lesion. The multivesicular lesion is characteristic of HD and presents with multiple daughter cysts within the parent cyst, whereas complex or solid lesion is the result of inflammatory changes mimicking a tumour [5]. In this case MR and ultrasonography could be useful in the diagnosis [8]. Magnetic resonance imaging findings of hydatid cysts in the liver are well described, but the diagnosis is more difficult to make in the soft tissue of the musculoskeletal system because the MRI findings are not described in a lot of cases [1, 7, 13, 18, 19]. A low-intensity rim can be seen on T1-weighted and T2-weighted images, but is more prominent on T2-weighted images. This rim is less developed in muscles [1]. It was seen in our cases with soft tissue localization. The detachment of the germinative membrane from the pericyst (the "water-lily" sign) is considered to be pathogno-

monic [20,21]. To the best of our knowledge, only two cases of the "water-lily" sign has been reported in intramuscular hydatid disease [20]. In our cases with soft tissue localization, on MRI the pathognomonic "water-lily" sign was present in one case on the posterior aspect of the thigh.

Although hydatid disease is not a common disorder and rarely involves soft tissue, it should be taken into consideration in the differential diagnosis of a soft tissue mass on the musculoskeletal system [18, 19, 22]. It should be kept in mind in the differential diagnosis of a slow-growing soft tissue mass of the musculoskeletal system, especially in regions where this zoonosis is endemic.

Using all available imaging methods contributes a lot in preoperative diagnosis. Different imaging modalities are complementary and give, in most of the cases, a definitive preoperative diagnosis.

In conclusion we can say that if hydatid disease is in the group of the benign lesions of the musculoskeletal system, it is necessary that this is well established preoperatively. If not, there is a high risk of dissemination and recurrence, which can be avoided with adequate preoperative and postoperative medical therapy, with complete surgical resection as the preferred treatment for isolated echinococcosis.

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R e z i m e

„ИМИЦИНГ“ ДИЈАГНОЗА НА МУСКУЛОСКЕЛЕТНА ХИДАТИДНА БОЛЕСТ

Василевска В.[1], Зафировски Ѓ.[2], Кирјас Н.[3], Јаневска В.[4], Самарџиски М.[2], Костадинова-Куновска С.[4], Лозанче К.[2], Антевски Б.[2], Серафимоски В.[5]

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Хидатидната болест е широко распространета во светот и предизвикува здравствен проблем во ендемските земји. Мускулоскелетната хидатидна болест може да има тешкотии во диференцијалната дијагноза. Целта на оваа ретроспективна студија беше да презентира некои различни „имицинг“ презентации на мускулоскелетната хидатидна болест.

Евалуиравме „имицинг“ наоди кај 15 пациенти со мускулоскелетна хидатидна болест во 15-годишен период. Десет пациенти (67%) беа мажи и пет (33%) жени, на возраст од 12 до 69 години. Кај сите беше направена радиографска евалуација. Компјутеризирана томографија (КТ) беше направена кај девет пациенти и магнетна резонанца (МР) кај три пациенти со мекоткивно зафаќање. Сите се оперирани, проследено со хистолошки преглед.

Инволвираност на коска имаше кај 11 пациенти и мекоткивна инволвираност кај четири пациенти. Коскени лезии на ’рбетот беа најдени кај 4 (36%), карлица 2 (18%), проксимален фемур 2 (18%), тибија 2 (18%) и 1 (9%) кај прва метатарзална коска. На рендгенографија беа идентификувани три различни изгледи на промената кај коскена хидатидна болест: дефинирани како типична остеолитична лезија (33%), со изглед како тумор (42%) и лезии кои личат на инфекција (25%). Мекоткивната хидатидоза беше локализирана на дистален дел на надколеница, паравертебрална мускулатура и делтоиден мускул. Магнетната резонанца покажува цистична маса, која содржи мултипни везикули и кај еден од нив беше присутен "water lily" знак патогномоничен за мускулоскелетна хидатидна болест. Презентираме три случаи на примарна мускулоскелетна хидатидна болест локализирана на метатарзална коска, проксимален фемур и на делтоиден мускул.

Мускулоскелетната хидатидна болест, презентирајќи се со различен изглед на промената, може да биде слична со тумор или со воспалителен

процес. Поради ретката презентација на болеста треба да се има на ум дијагностичката обработка на мускулоскелетните тумори.

Клучни зборови: хидатидна циста, мекоткивен тумор, коска, "*water-lily"* знак, магнетна резонанца, примарна мускулоскелетна.

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