



Evaluation of Musculoskeletal Hydatid Cyst Cases in Terms of Clinical Manifestations, Method of Dealing, Treatment, and Recurrence

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Abstract

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Hydatid disease is one of the most common parasitic diseases caused by *Echinococcus granulosus* worm. In this disease, involvement of all organs is likely. However, primary hydatid cysts of muscle tissue are so rare that occasionally in differential diagnosis, cystic masses are not placed in endemic areas. Timely and accurate diagnosis is crucial to prevent unnecessary biopsy. In this case-series study conducted in hospitals of Semnan and Tehran, Iran, from 2009 to 2014, 15 patients with musculoskeletal problems due to hydatid cyst were evaluated. In each case, information about the patient's clinical manifestations, including initial complaint, cyst location, treatment method, location of involvement organ, duration, dose of drug used and diagnostic method was evaluated. According to results of the present study, the prevalence of musculoskeletal system involvement was 7.21%. The most common site of involvement was spine and paravertebral muscles (7 cases) and the most common symptom was compressive symptoms on the spinal canal. Magnetic resonance imaging was the most commonly used diagnostic method and only in 1 case a wrong pre-operative diagnosis was made. Eleven patients reported a history of recurrence at the site of previous surgery (3 cases of hip involvement and 6 cases of spine involvement). There were 5 cases of recurrence following recent treatment, 2 of which were hip involvement and 2 cases of spine involvement. Furthermore, most patients (12 cases) underwent cyst resection surgery. The results of this study showed that although hydatid cystic skeletal muscle is rare, due to the possibility of repeated recurrence, especially in cases of spine and bone involvement, endemic areas should be considered in the differential diagnosis of each cystic mass of muscle and bone.

Introduction

Hydatid disease is one of the most common diseases in the Middle East, Australia, and South America and is one of the most common health problems in developing countries. The disease is transmitted to humans through the eggs of the *Echinococcus granulosus* parasite [1]. This parasite is one of the types of hermaphrodite tapeworms up to 5 mm long and human is an intermediate and random host of it [2]. The disease is more prevalent in areas with livestock occupation, especially in areas where dogs are used to protect animals. The economic damage caused by parasitic infections in developed and developing countries is reported to be 16% and 30% of total livestock production, respectively, and in countries where there is no serious fight against parasitic infections, infection rates are much higher [3], [4].

Liver and lung are the body's first capillary barriers passing high blood volume and are therefore the most common sites of involvement (70–85%). If not removed during these two stages of filtering, it migrates

to any part of the body. Symptoms of the disease range from asymptomatic to biliary obstruction, frequent cholecystitis mimicking and depending on the site involved and its growth rate, they can vary [5]. Symptoms of non-visceral type of disease depend on the rate of growth and the site of involvement. For example, bones 2% (invasion of bone marrow and slow bone erosion and subsequent pathological fractures), brain 0.5% (symptoms of space-occupying mass), heart 2.5% (conduction defects and pericarditis), and pelvis (pelvic mass symptoms), they can be different [6], [7]. Involvement of the musculoskeletal system is rare, accounting for about 1–5.4% of musculoskeletal hydatid cyst cases. Due to the low prevalence and similarity of these lesions to soft tissue masses, pre-operative diagnosis is difficult in this disease and in many cases, diagnosis is made intraoperatively or after histological examination of the sample. The presence of calcification in the computed tomography (CT) scan as well as the presence of several primary cysts can help to differentiate these cysts from carcinomas, amoebic, and bacterial abscesses as well as hemangiomas [8], [9].

Given the complications mentioned above, especially the problem of how to deal with it, definitive diagnosis before treatment, recurrence of the disease at the previous site and spread of the disease to other tissues, we decided to investigate the cases of musculoskeletal hydatid cysts.

Methods

In this case series study, after referring to Amir al-momenin and Kowsar Hospital in Semnan, Iran, as well as Imam Khomeini, Shariati, Sina, Amir Alam, Baharloo, Valiasr, and Ziaian Hospitals, Tehran, Iran during 2009–2014, all hydatid cyst cases were extracted from the patients' records and their musculoskeletal cases were evaluated. In the present study, of the 208 patients with hydatid cysts, 15 had musculoskeletal type. In each case, information about the patient's clinical manifestations, including initial complaint, cyst location, treatment method, location of involvement organ, duration, dose of drug used and diagnostic method was evaluated. Then, the recurrence cases were assessed by examining the record number and demographic information including age, sex, and occupation of the patient. In the case of report failure, the patient was contacted and was asked about follow-up

after the intervention, referral to another hospital, and the presence of manifestations and recurrence and their information recorded. In addition, the type of the parasite carrying the disease was evaluated by pathological evaluation. All patient information and records were kept confidential and no intervention was performed due to the retrospective study.

Data analysis

After sample collection, data were entered into the SPSS database (IBM SPSS Statistics, Version 22) and the results were reported in tables using descriptive statistics, frequency, and percentage.

Results

After reviewing patient records, a total of 15 patients with hydatid cysts with musculoskeletal involvement were included in this study. Demographic information and other factors are listed in Table 1. Furthermore, the study of 15 patients showed that only 5 patients experienced recurrence after treatment and the rest were without recurrence. In addition, regarding concurrent involvement of other organs, patients with number 5, 11, and 15 had

Table 1: Demographic characteristics and record data of the patients

N	Age/Sex	Job	Clinical signs	Initial complaint	Cyst location	Treatment method	History other organs	History same location
*P1	78/Male	Butcher	Shortness and claudication of left lower extremity	Recurrent pathologic fracture	Left hip joint	Due to the high risk of surgery, needle drainage and medical treatment with albendazole continuously	Left femoral head	Yes
P2	30/Male	Driver	Left armpit mass and discharge from previous operation site	Inflation in left axilla	Left armpit muscle	Surgery and oral albendazole 5 days before surgery up to 3 months after surgery	Brain cyst at age 9	Yes
P3	22/Female	Student	Pain in pelvis and right hip joint	Claudication and hip joint pain	Right hip joint	Surgery and oral treatment with albendazole	Right femoral head	Yes
P4	27/Male	workless	Pelvic pain along with discharges	Mass inflation	Left hip joint	Medical treatment with albendazole, the patient did not consent to surgery due to complications of previous operations.	Left hip joint	Yes
P5	51/Male	Self-employed	Low back pain and inability to walk	Low back pain	Lumbar muscles with extension to the spinal canal	Surgical and medical treatment with albendazole	No	Yes and involvement of different parts of the lumbar muscles
P6	30/Male	Self-employed	Low back pain and paresthesia in the anterior thigh	Low back pain	Upper lumbar muscles and psoas muscle	Surgery and oral treatment with albendazole from 5 days before up to 5 months after surgery	No	Yes
P7	45/Male	Self-employed	Right lower extremity pain associated with urinary and fecal incontinence	Local pain	L5, S1 lumbar vertebrae	Surgery and oral treatment with albendazole before and after operation	Yes in the gluteal area	No
P8	60/Male	Self-employed	Urinary retention and lower extremity paresthesia with swelling and tenderness at the site of previous operation	Symptoms of pressure on the spinal canal	Spinal canal in T11 abutments	Surgery and oral treatment with albendazole	No	Yes
P9	61/Male	workless	Lower extremity weakness and paraplegia	Symptoms of pressure on the spinal canal	Lumbosacral vertebrae	Drug treatment with albendazole	Femoral head and pelvis from about 40 years ago	Yes
P10	26/Male	workless	Inability to walk with urinary retention	Symptoms of pressure on the spinal canal	Thoracolumbar spinal cord	Surgery and oral treatment with albendazole 5 from years ago	No	Yes

(Contd...)

Table 1: (Continued)

N	Age/Sex	Job	Clinical signs	Initial complaint	Cyst location	Treatment method	History other organs	History same location
P11	59/Male	workless	Movement disorder and paresthesia up to the lower half of the trunk	Mass inflation	Thoracic spinal canal	Surgery and medical treatment with albendazole	No	Yes
P12	50/Female	housewife	Turgid growing mass in the left axillary	Mass inflation	left axillary	Surgery and medical treatment with albendazole	No	Yes
P13	51/Female	housewife	Pain and tightness in right anterior thigh	Local pain	Quadriceps femoris muscle	Surgery and medical treatment with albendazole	No	No
P14	54/ Female	housewife	A growing mass in the right lateral thigh that gradually causes pain during weighting	Mass inflation	Right lateral thigh	Surgery and medical treatment with albendazole	No	No
P15	33/ Female	housewife	A Turgid mass in the left arm	Mass inflation	Left arm	Surgery and medical treatment with albendazole	No	No

liver, pulmonary, and spleen and liver involvement, respectively. Furthermore, 5 patients had a history of previous involvement in other organs, including patient 1 left femoral head involvement, patient 2 brain injury involvement since the age of 9 years old, patient 3 right femoral head involvement, patient 4 left hip joint involvement, and patient 9 femoral head and pelvis involvement from 40 years ago. Diagnosis was reported in 14 patients just before surgery and only 1 patient (patient 15) underwent surgery by misdiagnosis (lipoma).

Results showed that of 15 patients, 10 (66.66%) were male (patients 1, 2, 4, 5, 6, 7, 8, 9, 10, and 11) and the rest were female. Among the patients, the most common age group involved was 50–60 years (5 patients including 5, 8, 11, 13, and 14) and 20–30 years (5 patients including 2, 3, 4, 6, and 10), then patients over 60 years old (patients 1 and 9) and between 40 and 50 years (patients 7 and 12) and age group of 30 and 40 years (patient 15). Furthermore, the overall mean age of patients was 45.13 years and the age range of patients ranged from 22 to 78 years.

According to Table 2, clinical manifestations and symptoms, involved tissue, diagnostic method, treatment, and involvement of other organs were evaluated. Our results showed that the most common clinical symptoms were symptoms of pressure on the spinal canal (including lower extremity weakness, urinary retention, paresthesia, etc.), including

7 (46.66%) patients (patients 5, 6, 7, 8, 9, 10, and 11) had come over with this complaint. It is noteworthy that, according to Table 1, some patients had two simultaneous symptoms. It was also found that the most common site of involvement was the spine and muscles surrounding it extending to the spinal canal which was 46.66% in 7 patients (5, 6, 7, 8, 9, 10, and 11). The most common diagnostic method was magnetic resonance imaging (MRI) that 6 patients (5, 6, 10, 11, 13, and 14), equivalent to 40% of the total, were diagnosed by this method. According to the treatment method, it was shown that 12 patients (80%) of 15 patients underwent open surgery and concurrent medical treatment (patients 2, 3, 5, 6, 7, 8, 10, 11, 12, 13, 14, and 15), 3 patients (20%) were treated medically and by drugs due to high risk, lack of consent for surgery, etc. (patients 1, 4, and 9). In addition to multiple surgeries, 1 patient (6.66%) underwent closed drainage and drainage with a needle at 1 time (patient 1). Drug treatment was used in all patients for different periods before and after surgery; 1 patient from 3 days before surgery to 3 months thereafter (patient 2), in one case 5 years before surgery (patient 3). Moreover, even in 1 case, the patient was on continuous drug treatment with albendazole for 30 years (patient 9). In 3 cases (patients 6, 7, and 11), it is directly stated in the patient's recent operation report that the cyst got opened during surgery and its contents were removed and immediately washed with hypertonic saline. Furthermore, in previous operation reports of eight patients (3, 5, 6, 7, 8, 9, 10, and 11), there were reports about opening of the cysts during surgery. In patient 12, the cyst was accidentally ruptured when removing and its contents were shed into the surgical area. In the operation report of patient 2, it is mentioned that as the skin got opened, the pus immediately discharged, indicating high adhesion of the cyst to the skin or incomplete removal of the cyst in previous surgeries. Furthermore, 11 cases (73.33%) had a history of involvement in the same site. The follow-up interval of recurrence was approximately 1–10 years.

The parasitic species in the patients were also examined and it was found that according to the pathological results, all patients had *E. granulosus* and no case of *Echinococcus multilocularis* was found.

Table 2: Number and percentage of the patients under study regarding the studied cases

Variable	Types	n (%)
Clinical manifestations and symptoms	Inflation and turgid mass	5 (33.33)
	Pain and feeling heavy at the site involved	5 (33.33)
	Symptoms of pressure on the spinal canal (including lower extremity weakness, urinary retention, paresthesia, etc.)	7 (46.66)
Tissue involved	Discharges	2 (13.33)
	Claudication	3 (20)
	Spine and related muscles	7 (46.66)
Diagnostic method	Muscles (excluding muscles of spine)	5 (33.33)
	Joints	3 (20)
	MRI	6 (40)
Treatment method	CT-scan	0
	Ultrasound	1 (6.66)
	Clinical examination	1 (6.66)
Involvement of other organs	Open surgery plus albendazole	12 (80)
	Medical treatment with albendazole	3 (20)
	Concurrent involvement of other organs	3 (20)
	History of previous involvement in other organs	5 (33.33)

MRI: Magnetic resonance imaging, CT: Computed tomography.

Discussion

Hydatidosis is one of the most important diseases common between animals and humans not only in terms of the life cycle of the parasite and having the different strains but also it has many complexities in terms of timely diagnosis in human and animal populations, as well as therapeutic aspects [8,9]. Diagnosis of patients with hydatid cysts is based on epidemiological findings, disease records, clinical signs, laboratory findings, and immunological tests [10]. The patient usually feels uncomfortable with a mass in the abdominal cavity, one of the diagnostic measures used is imaging techniques that include plain radiography, ultrasound, CT scans, and MRIs, though none of the imaging methods is capable of 100% and specific diagnosis of the hydatid cyst and only provides the presence of a mass in the organ under examination with little information on the physicochemical properties of the tissue [11], [12].

Although hydatid cysts are the most common disease in humans caused by intestinal worms, musculoskeletal involvement rarely occurs [13]. In the present study, of 208 patients with hydatid cyst, 15 patients (7.21%) had skeletal muscle type. Similar studies have reported the prevalence of musculoskeletal involvement caused by hydatid cysts of 4%, 1–4%, or 0.7–3% [13], [14], [15]. According to the findings of the present study, the prevalence of musculoskeletal involvement in our study is about twice that of these studies, which is justified by the fact that Iran is endemic to hydatid cysts [16]. In this regard, a study by Bulut *et al.* reported the musculoskeletal cysts of prevalence of 0.5–5%, and it was reported that in endemic areas after liver and lung, musculoskeletal hydatid cyst is the second most common site [17]. In general, musculoskeletal involvement is less prevalent in this complication and one of the reasons may be that the liver sinusoids and the capillary bed of the lung act as a barrier to the spread of cyst hematogenesis and possibly high concentration of lactic acid in the muscle and mechanical factors such as muscle contraction and prevent cyst placement [18], [19].

In our study, the majority of patients were male (66.6%) and the mean age of patients was 45.13 years. Most studies in this area have been case reports. However, in some case series studies, similar to the present study, they had similar findings. For example, in the study of García-Alvarez *et al.* on 13 patients with musculoskeletal hydatid cysts, most of the patients were male with a mean age of 53 years [13], or in a study on 9 patients with the same cysts, most patients were male [20].

From the clinical point of view, the disease encompasses a wide range of painless masses, swelling, pain, venous artery insufficiency, mimicking symptoms of intervertebral disc herniation,

etc., mainly depending on where the cyst is located [17], [21], [22], [23]. In studies that the cyst involved only the muscle, the most common symptom was a painless mass that has grown over time [20], [24]. However, sometimes more than one muscle and surrounding tissues, such as bones, are involved. The most common symptom in our study was compressive symptoms on the spinal canal, which is justified as the most common site of involvement is spine and its muscle (7 cases). In various studies, as in the present study, the most common site of involvement has been spine and paravertebral muscles. In the study on 13 cases, 35% were in the paravertebral area, 21% in the pelvic area, and the rest in the lower extremities [13], [25]. Another study reported that vertebral hydatid cysts account for 50% of musculoskeletal hydatid cysts [26].

In the present study, 3 patients had concurrent involvement of other organs; liver (1 case), lung (1 case), and liver and spleen (1 case), although in most studies musculoskeletal hydatid cysts are usually isolated and are not associated with involvement of other organs [14], [24], [25]. However, in the study of García-Alvarez *et al.*, similar to the present study, 1 patient had concurrent involvement of liver hydatid cyst [13].

MRI is usually the preferred diagnostic method for musculoskeletal hydatid cysts and is even mentioned as the first diagnostic method [27], [28]. In our study, 6 patients were diagnosed by MRI. In many cases, musculoskeletal hydatid cysts are not diagnosed or diagnosed intraoperatively or postoperatively by pathological examination. In the Ammari study, only 3 of 6 patients were diagnosed preoperatively [24]. In the García-Alvarez *et al.* study, only 1 patient with a misdiagnosis of lipoma underwent surgery, and the remaining patients were diagnosed preoperatively [13]. In our study, only 1 patient was misdiagnosed with open lipoma and underwent surgery and the rest was diagnosed correctly. Of course, this study may not be compared with other studies because most patients in similar studies had primary musculoskeletal hydatid cysts, but in the present study, 11 patients had a history of previous surgery and recurrence at the same site and therefore during the time of study their diagnosis was specified according to the previous pathology.

According to our results, recurrence at the site of previous surgery was 73.33%. In the study by García-Alvarez *et al.*, 2 of 13 patients had recurrence at the surgical site [13]. In another study on 9 patients with muscle cyst without bone involvement, no recurrence was seen in the follow-up period of 1–8 years [20]. One of the reasons for the differences between these studies and the present study and the cause of high recurrence seems to be bone involvement and spine involvement. In addition to the high involvement rate of spine and bone cases in the present study, another cause of high

recurrence may be due to cyst rupture during surgery and cyst fluid leakage. In the operation report of the 4 patients under study, a report of cyst rupture during surgery was given, which was immediately rinsed with hypertonic saline.

In this study, 12 patients underwent surgery and drug therapy. Three patients received drug treatment only. One of the 3 patients did not undergo surgery due to the high risk and underwent closed drainage. The drug treatment consisted of imidazole derivatives and mainly albendazole which was used in all of our patients. It was used in 12 patients along with surgical treatment and was used alone in 3 patients with high risk of surgery. There is little difference in the studies regarding the need for antiparasitic drugs in musculoskeletal hydatid cysts. One study reported that the usefulness of the drug depends on the dose and duration of use [20], [26]. In the study of Madhar *et al.*, it was stated that there were no cases of using imidazole derivatives except in cases of bone hydatid cysts and the use of these drugs in muscular hydatid cysts has no place [28]. In another study of 6 patients, 25 mg/kg/day albendazole was administered to 4 patients for 6 months after surgery. In the study of Garcia, 9 of the 13 patients who underwent radical surgery were given no drug treatment, and albendazole was administered to the other 4 cases where complete resection was not possible. In the follow-up in 3 of them that sacrum was involved, the addition of drug therapy failed to eliminate the infection and eventually they had to undergo surgery again [13]. On the other hand, it has been shown that administration of the drug 3 weeks before surgery significantly reduces cyst pressure. Administration of 1 and 3 months of drug in liver hydatid cysts resulted in the death of 72% and 94% of cysts, respectively [27], [29]. Therefore, one of the factors contributing to the high recurrence rate in this study may be non-administration of the drug before surgery or continuing it properly in the post-operative period.

Finally, given that the study was retrospective and that many information was not complete in the patients' records, it is recommended to conduct a prospective study for a long time to confirm the findings with greater certainty.

Conclusion

Although hydatid disease in muscular tissue is rare even in endemic areas, it should be considered as one of the differential diagnoses in all cases of soft tissue masses. Furthermore, given the likelihood of frequent recurrence, especially in cases of vertebral or bone involvement, it should be considered in the differential diagnosis of any cystic mass of muscle and bone in endemic areas.

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