Chirurgia (2025) 120: 714-722
No. 6, November - December

http://dx.doi.org/10.21614/chirurgia.3238

Primary Musculoskeletal and Retroperitoneal Hydatid Disease: A Case Report and Literature Review

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Abbreviations:
ER: emergency department;
CT: computed tomography;
SBO: small bowel obstruction;
COPD: chronic obstructive pulmonary disease;
FNAC: fine needle aspiration cytology;
MRI: magnetic resonance imaging;
CE: cystic echinococcosis.

Rezumat

Boala hidatică primară musculoscheletală și retroperitoneală: prezentare de caz și review al literaturii

Boala hidatică cauzată de Echinococcus granulosus afectează rar sistemul musculoscheletal, reprezentând doar 1-4% dintre cazuri. Prezentăm cazul unui bărbat de 65 de ani care s-a prezentat cu detresă respiratorie acută şi o hernie inghinală stângă ireductibilă. Tomografia computerizată a evidențiat leziuni chistice multiloculare extinse la nivelul muşchilor iliopsoas şi coapsei, asociate cu distrucție osoasă pelvină şi un chist secundar la nivelul testiculului drept, fără implicare hepatică sau pulmonară. Exeresisul chirurgical şi examenul histopatologic au confirmat infecția cu Echinococcus granulosus. A fost inițiat tratament postoperator cu albendazol. Evoluția postoperatorie a fost dificilă din cauza bolii pulmonare obstructive cronice, dar pacientul s-a recuperat. Analiza datelor instituționale şi a literaturii confirmă raritatea bolii hidatice musculoscheletale primare şi dificultățile sale de diagnostic. Conştientizarea localizărilor atipice şi integrarea datelor imagistice, chirurgicale şi epidemiologice sunt esențiale pentru un diagnostic corect şi o conduită terapeutică optimă. Boala hidatică musculoscheletală primară poate apărea în absența unei localizări hepatice sau pulmonare şi trebuie luată în considerare în diagnosticul diferențial al leziunilor chistice ale părților moi la pacienții din zone endemice.

Cuvinte cheie: boala hidatică musculoscheletală, hidatoza retroperitoneală, Echinococcus granulosus

Abstract

Hydatid disease is a parasitic infection caused by Echinococcus granulosus, most commonly affecting the liver and lungs. Musculoskeletal localization accounts for only 1-4% of cases, and primary muscular hydatidosis without

Received: 15.10.2025
Accepted: 10.12.2025

thoracic or abdominal organ involvement is extremely rare. We report the case of a 65-year-old man who presented in an emergency setting with acute respiratory distress and an irreducible left inguinal hernia. Computed tomography revealed an extensive multiloculated cystic lesion involving the left iliopsoas and proximal thigh muscles, associated with pelvic bone destruction and a secondary right testicular cyst, in the absence of hepatic or pulmonary hydatid disease. Emergency surgery included hernia repair and excisional biopsy of the thigh masses, which intraoperatively showed the typical appearance of hydatid cysts. Histopathology confirmed Echinococcus granulosus infection, and postoperative albendazole therapy was initiated. The postoperative course was complicated by severe chronic obstructive pulmonary disease, and, despite initial recovery, the patient later developed a hip abscess and repeatedly refused further surgery, ultimately being lost to follow-up. An institutional 10-year review and a focused literature review underline the rarity and diagnostic difficulty of primary musculoskeletal hydatid disease. Primary musculoskeletal and retroperitoneal hydatid disease should be considered in the differential diagnosis of cystic soft-tissue lesions, even in the absence of hepatic or pulmonary involvement, particularly in patients from endemic areas.

Keywords: musculoskeletal hydatic disease, retroperitoneal hydatic disease, Echinococcus granulosus

Introduction

Cystic echinococcosis (hydatid disease) is one of the most common helminthic infections in humans and remains an important public health problem in many parts of the world. It is endemic in large areas of southern Europe, Asia, Africa, the Middle East, and South America, where close contact between livestock and dogs facilitates transmission. Humans become accidental intermediate hosts after ingestion of Echinococcus eggs, most often Echinococcus granulosus (1,2). The oncospheres pass throw the intestinal wall and reach the portal circulation. They are usually trapped in the liver and, less frequently, in the lungs, which act as filters for the systemic circulation. Consequently, the liver and lungs are the most frequent sites of hydatid cysts, while involvement of other organs is much less common (3,4).

Musculoskeletal hydatid disease is rare, representing only a small proportion of all cases, and muscle involvement is considered particularly uncommon (5). Several mechanisms, including high lactic acid content and continuous muscle contraction, are thought to make skeletal muscle a relatively unfavorable environment for cyst implantation and growth (6). Primary musculoskeletal and retroperitoneal hydatid disease without hepatic or pulmonary involvement is therefore an exceptional finding.

Romania is recognized as an endemic country for cystic echinococcosis, with persistent hyperendemicity documented both in humans and in livestock, despite control efforts. This epidemiological background is essential for interpreting atypical presentations and underscores the clinical relevance of hydatid disease in our region.

We describe an unusual case of primary musculoskeletal and retroperitoneal hydatid disease involving the left iliopsoas and thigh muscles, with associated pelvic bone destruction and a secondary testicular cyst, discovered incidentally during emergency surgery for an incarcerated inguinal hernia. We also present our institutional experience with hydatid disease and review the published literature on musculoskeletal involvement. This structure allows us to highlight both the particularities of the index case and the broader context of hydatid disease in endemic settings.

Case Presentation

A 65-year-old man with unknown medical history was brought to the emergency department intubated and mechanically ventilated after his family called an ambulance for acute shortness of breath. Initial clinical examination revealed an irreducible left inguinal hernia surrounded by multiple diffuse, tumor-like masses in the groin and proximal thigh. A keloid postoperative scar was present on the lateral side of the left thigh, without local signs of inflammation, suggesting prior undocumented surgery.

Imaging Findings

Emergency thoraco-abdominal computed tomography (CT) showed a left inguinal hernia containing ileal loops and omental fat, exiting medial to the inferior epigastric vessels and consistent with a direct hernia. There were no signs of small-bowel obstruction, no dilated loops, fluid levels or free intra-abdominal air. CT scan also demonstrated a large, thick-walled, multiloculated cystic mass

extending from the left retroperitoneal space into the iliopsoas muscle and the proximal anterior thigh muscles (rectus femoris, vastus lateralis, and vastus medialis). The lesion was associated with osteolytic destruction of the ilio- and ischiopubic rami, sacrum, left sacral ala and proximal femur. Contrast-enhanced images showed well-defined, non-enhancing cystic components with internal septa and no calcifications or solid tissue. A small round cystic lesion with similar characteristics was noted in the right testis. The liver and lungs were free of cystic lesions on CT (*Figs. 1-7*).

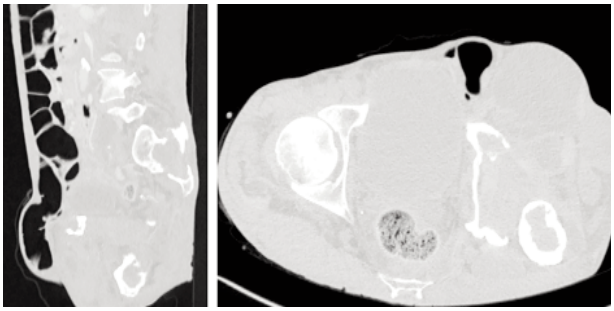

Figure 1. Sagital and axial CT scan - lung view – inguinal hernia. No evidence of SBO

Surgical Findings and Histopathology

Given the irreducible hernia, emergency surgery was performed. Herniotomy confirmed a direct inguinal hernia without bowel or mesenteric ischemia, and the hernia was repaired uneventfully. An excisional biopsy of the left thigh masses was then carried out. On incision, the lesions displayed the classical appearance of hydatid disease, with multiple daughter cysts surrounded by a thin fibrous capsule adherent to the surrounding musculature (*Fig. 8*). Because of the extent of involvement, complete pericystectomy without spillage was not feasible. Partial excision and curettage of the cystic masses were performed, followed by copious irrigation with hypertonic saline and placement of multiple drains.

The postoperative course in the intensive care unit was prolonged and challenging due to severe chronic obstructive pulmonary disease (COPD) with long-term home oxygen therapy, which was only disclosed after admission. The patient required nine days of mechanical ventilation before successful extubation. Once stabilized and transferred to the surgical ward, a more detailed history revealed an undocumented left thigh operation under local anesthesia approximately 20 years earlier, reportedly after a fall, but with no available records. The patient had a low socio-economic status, no current livestock or pets, and no recent contact with wildlife; however, his family reported that he had lived for several years on a landfill together with stray dogs and wild animals such as foxes.

Histopathological examination confirmed cystic echinococcosis due to Echinococcus granulosus. Postoperative albendazole therapy was initiated. The patient was discharged at his request after 19 days and remained compliant with albendazole treatment, but repeatedly refused additional imaging and further surgical procedures during three outpatient visits.

Follow-up

Approximately six months after the initial surgery, he was admitted to another hospital for COPD exacerbation and developed an abscess around the left hip joint. Despite local wound care being initiated, the patient again refused any surgical intervention. No further medical information is available, and the patient was ultimately lost to follow-up.

Institutional Experience

We retrospectively reviewed the records of patients

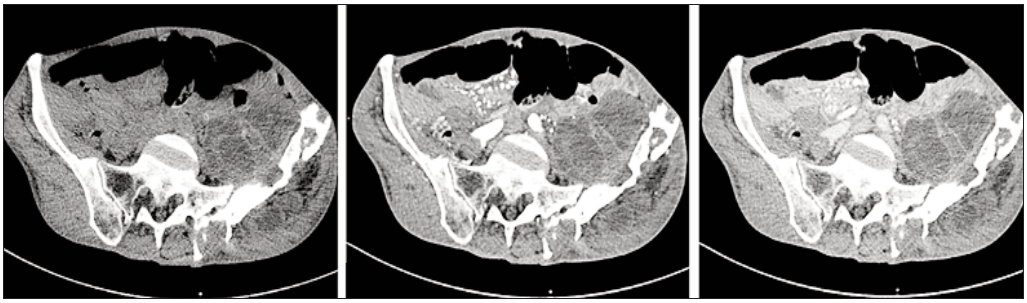

Figure 2. Axial CT scan native – arterial – venous phase

treated for cystic echinococcosis in Târgu Mureş Emergency County Hospital over a 10-year period (2013-2023). Only surgically managed and histologically confirmed cases of Echinococcus granulosus or Echinococcus multilocularis infection were included. A total of 82 patients were identified. The liver was involved in 53 patients (64.6%), the lungs in 27 patients (32.9%), and other localizations (including spleen and kidney) in 2 patients (2.4%). Among hepatic cases, 32 involved the right lobe, 19 the left lobe and 2 both lobes. For pulmonary cases, 17 involved the right lung, 8 the left lung and 2 were bilateral.

In all instances, open surgery was performed. Total cystectomy was achieved whenever feasible; in cases where the cyst wall was firmly adherent to critical structures, a more conservative approach was used, and residual cavities were managed by marsupialization with omentoplasty when local conditions allowed. All patients received preoperative albendazole for 14 days and continued treatment for 40 days postoperatively.

Information on presentation and case management was reviewed and the results presented as summary statistics (*Table 1*). Other localizations included spleen and kidney.

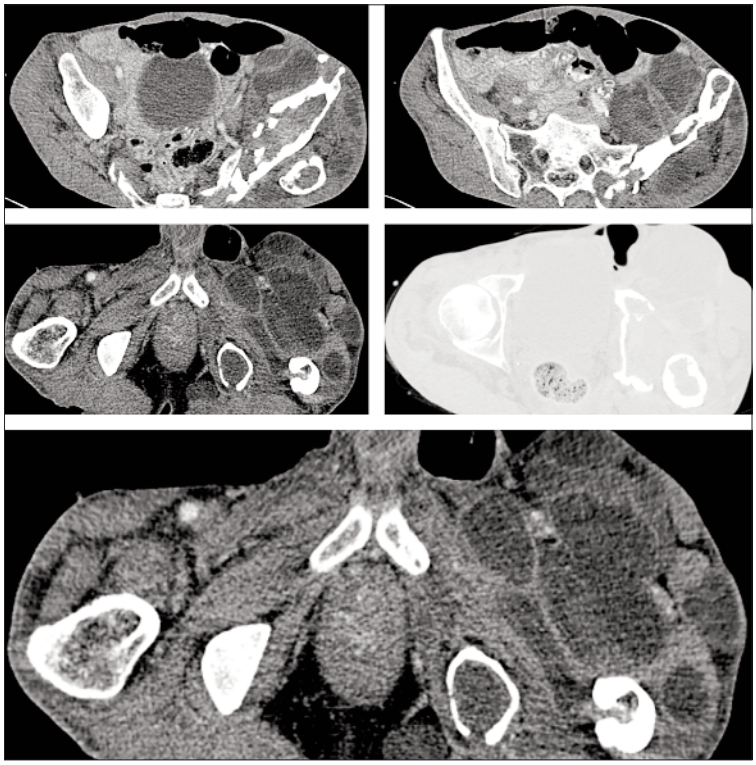

Figure 3. Axial CT scan

Literature Review

A literature search was performed in the PubMed database using "musculoskeletal hydatid" as the main keyword. We limited the search to the last 15 years and included older articles if they reported larger case series. Only original studies or case series with at least three patients and a clear description of musculoskeletal and/or bone involvement were selected. Single case reports were excluded to focus on series that allowed some assessment of frequency and patterns of involvement, although we acknowledge that case reports can provide valuable insights into very rare presentations. The main characteristics of the selected series, including the number of patients and the distribution of muscle, bone, liver and lung involvement, are summarized in *Table 2*.

Because the included series originate from different institutions and geographical regions, patient overlap is unlikely, and the 64 cases presented in *Table 2* can reasonably be considered as distinct individuals. However, the possibility of duplication cannot be entirely excluded.

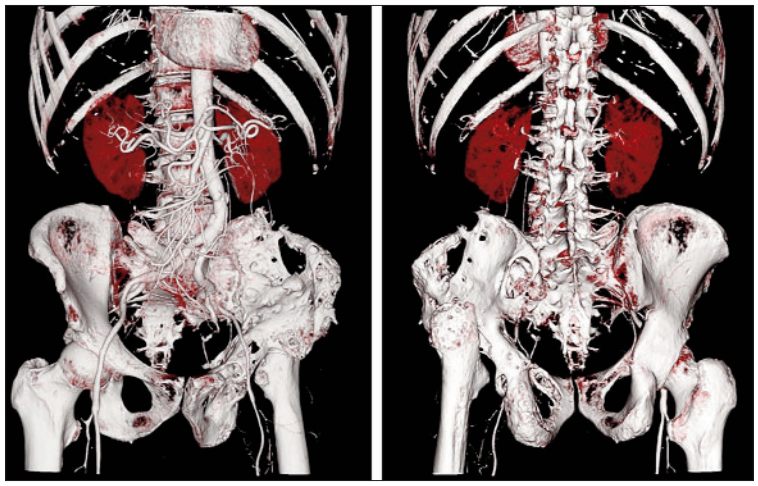

Figure 4. Anterior and posterior view VRT reconstruction

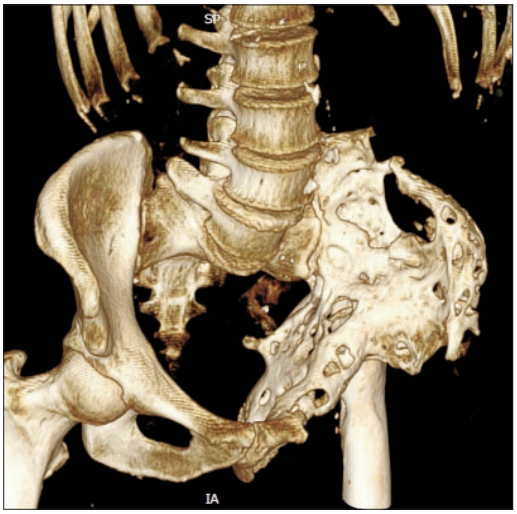

Figure 5. Oblique VRT reconstruction

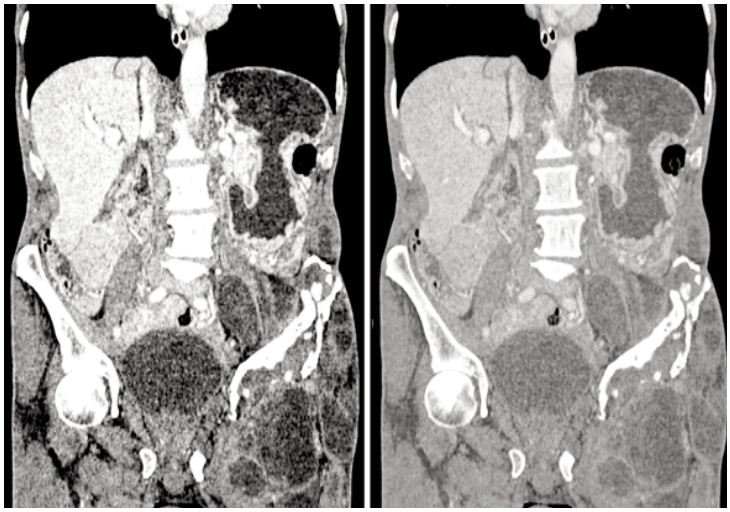

Figure 6. Core CT - venous phase

Table 1. Distribution of hydatid disease localizations in our institutional experience.

Total nr. of cases over 10 years:	82	
Liver localization	53	64.63%
Right lobe	32	60.3%
Left lobe	19	35.84%
Both lobes	2	3.77%
Pulmonary localization	27	32.9%
Right lung	17	62.96%
Left lung	8	29.62%
Bilateral	2	7.4%
Other localizations	2	2.43%

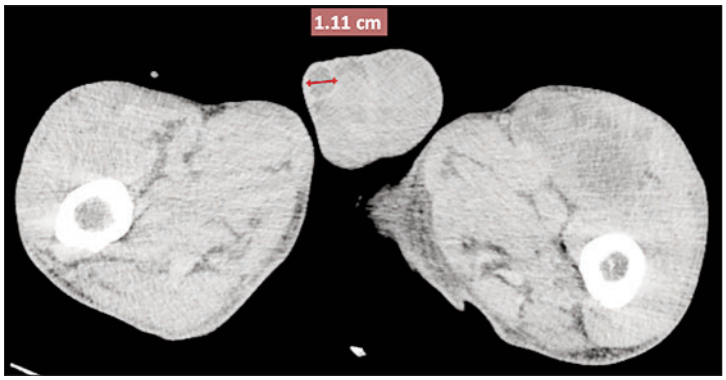

Figure 7. Axial CT – venous phase

Geramizadeh et al. published in 2013 a review from Iran totaling a staggering 463 cases over the past 20 years that has not been included in the above table but rather explained (12). The authors report that only 55 patients out of 463 cases presented with musculoskeletal hydatic cysts comprising 11.87% of cases. These findings belonging to a known endemic area showcase the uncommon character of this localization, even in such an area.

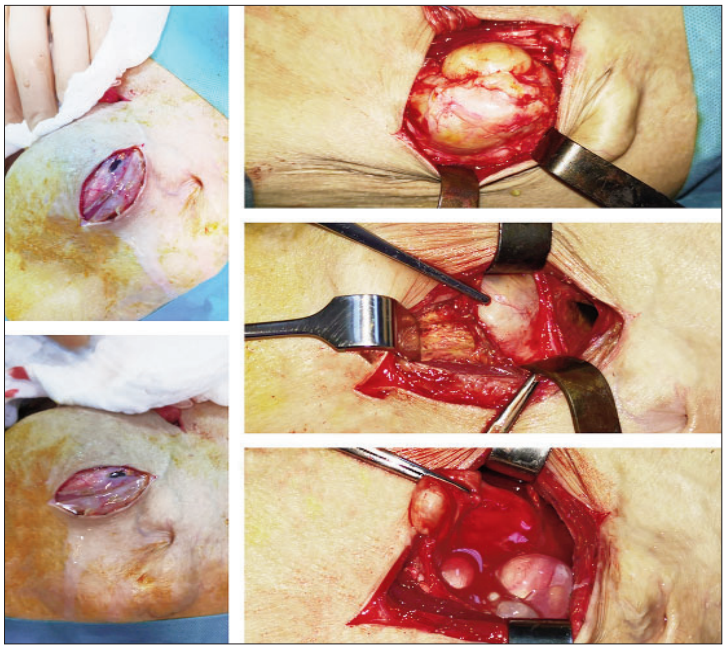

Figure 8. Intraoperative appearance

Table 2. Sites of musculoskeletal infection and associated hepatic or pulmonary involvement in published series of hydatid disease

Author	No	Site of infection	Liver	Lung	Muscle	Bone
Torricelli et al. (7)	26	Bone infection with adjacent soft tissue involvement in 12 cases	-	-	12	26
Arazi et al. (8)	15	2 cases of subcutaneous hydatic cysts	0	0	6	7
Merkele et al. (3)	8	Iliopsoas, left adductor musculature, left femur, left medial gluteal muscle, musculature of right upper leg	6	-	8	6
Dahniya et al. (9)	7	5 bone infections with no soft issue involvement, 2 primary intramuscular infections	0	0	2	5
Toğral et al. (10)	5	3 patients with left ilium and acetabulum involvement, 1 left femur, 1 left thigh muscle compartments.	1	-	1	4
Natarajan et al. (11)	3	3 cases all with pathological bone fracture	-	-	-	3
Total no. cases	64		7	-	29	51

Despite the comprehensive analysis, it is essential to acknowledge the potential limitations of the literature review for the following: the fact that case reports were not considered, even if case reports have their limitations, they can provide in-depth understanding of rare diseases; the literature review is limited to a relatively small number of studies; no statistical analysis of the literature review was performed as, in the authors' view, it would minimize the impact of the case presentation.

Discussions

Summary of the Present Case

We report a rare case of primary musculoskeletal and retroperitoneal hydatid disease in a 65-year-old man who presented in the emergency setting with acute respiratory distress and an incarcerated left inguinal hernia. CT revealed a large multiloculated cystic lesion involving the iliopsoas and proximal thigh muscles, with associated pelvic bone destruction and a secondary cyst in the contralateral testis, in the absence of hepatic or pulmonary hydatid disease. Intraoperative findings and histopathology confirmed cystic echinococcosis. The postoperative course was significantly influenced by severe COPD, and the patient repeatedly refused definitive surgery for the extensive musculoskeletal involvement, ultimately being lost to follow-up.

Comparison with Previously Reported Cases

Musculoskeletal involvement in cystic echinococcosis is reported in only 1-4% of cases, even in endemic regions. Most series describe either isolated bone disease or muscle involvement associated with hepatic or pulmonary cysts. Iliopsoas and pelvic bone involvement have been reported by several authors, but simultaneous involvement of iliopsoas, multiple thigh muscle compartments, pelvic bones and a secondary testicular cyst, without liver or lung disease, appears exceptional. Our institutional review of 82 surgically treated patients over 10 years identified only two cases with "other" localizations, underscoring the rarity of primary musculoskeletal hydatid disease even in a hyperendemic country. Hydatid cyst disease is relatively widespread throughout regions of southern Europe, Asia, Australia, Africa, and the Middle East (13-15). It is a known fact that in most cases it takes years for the patient to develop symptoms (5,10). In humans, the liver is the most common site of cyst development (60%), followed by the lungs at (20%), and seldom meet, the spleen, kidney, brain, and soft tissue (3,4). Other unusual location include the heart, mediastinum, intramedullary or various other organs (16-19). Given the rarity of primary musculoskeletal hydatid disease, it is helpful to include this information on preventive strategies and public health interventions to raise awareness and reduce the incidence of the disease in the future.

Diagnostic Considerations: Imaging, Serology, and FNAC

Clinical presentation of musculoskeletal hydatid disease is often nonspecific, with slowly progressive pain, swelling or a "tumor-like" mass. In our patient, the masses were overshadowed by acute respiratory failure and the incarcerated hernia, making preoperative suspicion of hydatid disease unlikely. In an ambulatory setting, preoperative imaging studies are mandatory for diagnostic purposes. Serological tests or enzyme-linked

immunosorbent assay may aid in the diagnosis. However, serology alone is insufficient to diagnose muscular hydatid cyst disease, often yielding false negative results (20,21) although other authors note that serology is commonly used for diagnostic and follow-up of extrahepatic hydatic disease (20). Fine needle aspiration cytology (FNAC) is another option to be take into account, but it seems one of the best methods to use is MRI (22-24). Concerns over spillage during FNAC do not appear warranted, especially if patients receive proper medical treatment and biopsy tracts are resected at the time of surgery although the procedure is seldom described for muscular cyst (25). Other authors recommend using chitosan/carboxymethylcellulose/ß-glycerophosphate hydrogel for effective control of spillage during aspiration of hydatid cysts (26). This is a new technique that still requires the proof of time, but it shows potential.

In an emergency setting as it is in our case, it is difficult to consider this diagnosis, especially in the absence of patient history; the patient had an unknown medical history, making it difficult to assess the complete background of the case, although similar localizations have been reported (3,9,22,27). Romania was listed in 1995 among the countries with the highest prevalence of cystic echinococcosis (CE) worldwide (28). Nevertheless, CE is not a notifiable disease in this country. Other authors examined cattle and sheep in endemic areas of north-eastern and southern Romania. The study finds that, in a endemic area, roughly 32% of cattle and 50% of sheep are infected emphasizing the hyperendemic presence of E. granulosus in Romania and pleading for urgent development of sustainable surveillance and control strategies both in animals and humans (29).

Clinical presentation of musculoskeletal hydatid disease depends on the location and size of the cysts. Patients may experience pain, swelling, and restricted movement in the affected area. In some cases, the cysts may rupture, leading to an acute inflammatory reaction and release of the cyst contents, which can cause anaphylaxis or systemic dissemination.

Diagnosis of musculoskeletal hydatid disease is challenging and often requires a combination of clinical, radiological, and serological investigations. Imaging techniques such as X-ray, ultrasound, computed tomography (CT), and magnetic resonance imaging (MRI) play a central role in detecting the cysts and assessing their characteristics. Serological tests, including enzyme-linked immunosorbent assay (ELISA) and immunoblotting, can aid in confirming the diagnosis (30).

Imaging represents a key aspect in diagnosis. Radiographs and CT may show expansile osteolytic lesions and cystic soft-tissue masses, whereas MRI provides better delineation of cysts, daughter cysts, and surrounding soft tissues. Features such as well-defined multiloculated cysts, the presence of daughter cysts, internal septa, and absence of significant enhancement raise the suspicion of hydatid disease. According to the WHO Informal Working Group on Echinococcosis (WHO-IWGE) classification, cystic echinococcosis is staged from CE1 to CE5 based on imaging morphology: CE1 and CE2 are active, CE3 transitional, and CE4-CE5 inactive. In our case, the non-calcified, purely cystic, multiloculated lesions without solid components are most compatible with an active-stage cyst (CE1–CE2) in this system (31).

Even in an emergency setting, imaging studies can raise the suspicion that hydatic disease may be present based on "typical" osteolytic lesions, "tumor-like" features and lesions resembling infection, but, in low volume centers, it is easy to miss this diagnosis (32). The disease can also appear as well-defined cystic lesion with daughter cysts, may contain septa or debris in it with no enhancement on intravenous contrast (23). It is even more difficult to consider hydatic disease in the absence of somewhat pathognomonic liver or lung cysts.

Differential Diagnosis

The differential diagnosis of a multiloculated cystic lesion involving muscle and bone includes chronic abscess, tuberculous osteomyelitis, soft-tissue sarcoma with cystic degeneration, benign cystic tumors (such as synovial or ganglion cysts), and other parasitic infections. In patients from endemic regions, hydatid disease should be systematically considered when imaging reveals "tumor-like" cystic masses with osteolysis, even when hepatic and pulmonary imaging is normal. Lack of awareness, especially in low-volume centers and emergency settings, may contribute to missed or delayed diagnoses.

Treatment Options and Risk of Recurrence

Treatment of muscular hydatic disease is a combination of albendazole and surgery. Although a more comprehensive discussion on different

treatment modalities and their pros and cons would provide a broader perspective it is beyond the scope of this paper.

Wide excisions and complete pericystetcomy following cyst inactivation with alcohol solutions is routinely practiced. Treatment options and chronology vary, many surgeons using short term albendazole therapy followed by surgery and long term albendazole treatment following surgery (20,24,33). Cystic rupture can lead to anaphylactic shock or can release viable scoleces that implant elsewhere and disseminate the disease (33). Considering our patient history this may have been the case. Osseous hydatidosis can be dormant for decades with nothing other than indirect signs like limb deformation or abscesses (34). In our case, complete surgical excision was not an option considering the acute setting. We opted for two stage surgical approach to treat the cystic lesions found in the abdomen and left hip, but the patient refused further surgery.

Public health considerations in endemic regions

Our case highlights several public health issues relevant to endemic areas. First, socio-economic vulnerability and close contact with stray dogs or inadequately controlled livestock remain important risk factors for transmission. Second, hydatid disease is frequently under-reported, particularly when extrahepatic or asymptomatic, and is not a notifiable disease in many countries, including Romania. Finally, atypical localizations are easily overlooked in emergency settings, which emphasizes the need for continued clinician education and integration of epidemiological data into routine decision-making.

Limitations

The literature review presented here is descriptive rather than systematic and is limited to a relatively small number of retrospective series. Case reports were intentionally excluded to focus on larger cohorts, but this approach may omit some unusual presentations. No formal statistical analysis was performed, as the primary aim of the review was to contextualize our case rather than to provide pooled estimates. Nevertheless, the available data consistently confirm that primary musculoskeletal hydatid disease is rare, even in hyperendemic regions.

Conclusions

Primary musculoskeletal and retroperitoneal hydatid disease is a rare but a clinically important entity, even in highly endemic regions. Diagnosis is particularly challenging when liver and lung imaging are normal and when the initial presentation is an emergency condition unrelated to the cysts, as in our patient.

Radiological evaluation with CT and/or MRI, integrated with the epidemiological background and intraoperative findings, is essential for recognizing atypical localizations. In doubtful cases, serology and carefully planned tissue sampling may contribute to diagnosis, but negative serology does not exclude musculoskeletal hydatid disease. Close collaboration between surgeons, radiologists, pathologists, and infectious disease specialists is crucial to optimize management and minimize the risk of intraoperative spillage and recurrence.

From a public health perspective, our findings emphasize the need to maintain a high index of suspicion for hydatid disease in endemic settings, particularly in patients with low socio-economic status and a history of contact with stray or rural dogs. Surveillance and control programs in livestock and dogs remain key components of prevention.

Key learning point: primary musculoskeletal hydatid disease can occur in the absence of hepatic or pulmonary involvement and should be considered in the differential diagnosis of cystic soft-tissue lesions in patients from endemic areas or with relevant exposure history.

Author's Contributions

BVO, SBA, BUK – surgical intervention, follow-up, MA, ZRA – image acquisition, image processing, BVO – original manuscript draft, BM, SBA, MCD – literature review, PubMed search, literature review draft, – manuscript revision, project administration, supervision. All authors have read and agreed to the published version of the manuscript.

Conflicts of Interest

The authors report no conflicts of interest related to this work.

Funding

This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Ethical Statement

The study was conducted in accordance with the ethical standards of the institutional research committee and the Declaration of Helsinki. Written informed consent was obtained from the patient(s) for publication of this case report and any accompanying images. All identifying information has been anonymized to ensure patient confidentiality.

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