The Treatment of Soft Tissue Hydatidosis through an Oncological Approach: A Review of 13 Cases with an Average Follow-Up of 43 Months

Original Research Article

ABSTRACT

Aim: The primary aim of this retrospective study was to assess the clinical results, instances of local recurrence, and the occurrence of complications related to the utilization of antihelminthic chemotherapy and comprehensive resection in patients diagnosed with muscle or bone hydatidosis.
Methods: A retrospective analysis was conducted on 13 patients diagnosed with muscular hydatid disease. The study included 13 patients (8 males and 5 females) who were treated for muscle hydatidosis between 2010 and 2020. The average age at the time of surgery was 38 years, ranging from 23 to 52 years. All patients underwent comprehensive resection and received preoperative and postoperative chemotherapy using albendazole. The follow-up period averaged 43 months, ranging from 32 to 60 months.
Results: All patients exhibited positive clinical outcomes, and no instances of local recurrence were observed. Surgical complications were encountered in 3 patients (23%): one case of superficial infection and two cases of hematoma. An additional surgical procedure (local debridement and vacuum-assisted drainage system) was required for one patient (8%). Employing an aggressive approach involving antihelminthic chemotherapy and extensive resection resulted in favorable clinical outcomes and reduced the risk of local recurrence in patients with musculoskeletal hydatidosis. While potential complications from aggressive surgery should be weighed, they are still preferable over the potential morbidity associated with local and systemic dissemination.
Conclusion: Muscularhydatidcystsarerare,andwide resection together albendazoltreatmentareeffectiveinpatientswithsoft-tissuehydatidcysts.

Keywords: muscular, hydatid, cyst, albendazole, wide resection

1. INTRODUCTION

Hydatid disease, resulting from the larval phase of the tapeworm Echinococcusgranulosus, is a parasitic infection. This situation presents a notable health issue in areas where the parasite is widespread, encompassing South America, the Middle and Far East, as well as the Mediterranean region.The liver and lungs are commonly afflicted by this disease, serving as frequent sites of infection. In contrast, muscle infection is a rarity, accounting for approximately 0.5-4.7% of cases[1].

Muscle hydatidosis is an infrequent occurrence, constituting only 1% to 5% of all cases of hydatidosis, even within regions where the condition is prevalent[2]. The infrequency of this occurrence can be credited to the existence of lactic acid within muscles, which generates an unfavorable setting for the parasite's development. [3]. A comprehensive examination

of existing literature highlights that the available information mostly consists of isolated case reports, encompassing a diverse array of affected muscle groups.[4-6].

In areas where hydatid disease is prevalent, it's important to include the possibility of a hydatid cyst in the list of potential causes when encountering atypical soft-tissue masses. Clinically and on radiological assessments, it closely mimics a soft-tissue tumor. Prior to surgery, a thorough radiological assessment, with a focus on techniques like magnetic resonance (MR) imaging, becomes crucial. This step is essential to circumvent unnecessary biopsies and ensure proper surgical management of the cyst[7]. The presently advised approach for treating musculoskeletal hydatidosis involves a combination of antihelminthic chemotherapy and the surgical removal of affected areas with extensive margins[8-11].Antihelminthic chemotherapy is believed to lower the probability of recurrence rates[11, 12].
The surgical management of muscular hydatid cysts necessitates extensive resection to avoid the rupture or leakage of cyst contents, a situation that could pose a life-threatening risk. This assertive oncological strategy aims to eliminate the lesion and halt the systemic dissemination of the disease.

The objective of this retrospective investigation was to assess clinical results, rates of local recurrence and complications related to antihelminthic chemotherapy and extensive resection in individuals with muscle hydatidosis.

2. MATERIAL AND METHODS

A retrospective investigation involved 13 patients diagnosed with muscular hydatid diseaseat the Orthopedic clinic from 2010 to 2020. The diagnosis was confirmed via serological validation utilizing an enzyme-linked immunosorbent assay, accompanied by radiographic evaluations. None of the patients underwent biopsy before diagnosis. After assessment and confirmation by the Tumor Council, patients were further evaluated at the Infectious Diseases Department. They were administered preoperative albendazole (15 mg/kg). Post-operative treatment plans were also determined by the Infectious Diseases Department. Comprehensive resection was performed on all lesions.

Following the surgery, the Infectious Disease clinic administered albendazole treatment for up to 6 months during the post-operative period, with the duration contingent upon the extent of extraskeletal involvement of the disease.

During the surgical procedure, hypertonic and povidone iodine solutions were employed in the operative area. No instances of anaphylactic reactions were noted among our patients during surgery. Among the cases managed by the orthopedics department, two patients exhibited liver involvement, and one patient had lung involvement. Subsequent treatments and follow-up care were overseen by the respective medical clinics.
The retrospective data collection encompassed various aspects: age and gender, lesion location, symptom duration and type, radiographic and MRI findings, histological results, prior treatment approaches, extent of dissemination, specifics of surgical procedures and supplementary treatments, clinical and radiological outcomes at the latest follow-up, and any complications, including instances of local recurrence. The mean follow-up period was 43 months, ranging from 32 to 60 months.

The study comprised 8 men and 5 women, with a mean age of 38 years at the time of surgery (ranging from 23 to 52 years). The anatomical distribution of the muscular lesions is outlined in Table I. None of the patients had previously undergone surgical intervention for muscular hydatid disease, except for one who had received chemotherapy and surgical treatment for liver involvement approximately two years prior.

Clinical and radiological follow-up, which encompassed MR imaging of the affected area, occurred every 6 months during the initial 2 years and subsequently transitioned to annual intervals. A systemic assessment was reiterated on an annual basis. Instances of complications, encompassing local recurrence and the necessity for supplementary surgery, were meticulously documented. Patients who underwent MRI imaging for early diagnosis of cystic hydatid were also subjected to ultrasound (USG) imaging. Radiologists reported identifying the characteristic features of cystic hydatid lesions, such as a multi-septated, hypoechoic cystic mass, during the USG evaluations.

3. RESULTS AND DISCUSSION

A painless, gradually enlarging mass emerged as the primary symptom in the 12 patients affected by muscle hydatidosis. These masses exhibited a moderate to large size, were soft to the touch, slightly tender upon palpation, and positioned deep within the tissue. Local warmth or redness was absent.

All patients underwent both ultrasound (USG) and MRI imaging. The MR imaging revealed soft-tissue masses within the involved muscles, ranging in size from 39 × 16 × 20 mm to 102 × 42 × 31 mm. The lesions typically displayed distinct and well-defined borders(Figure 1). MR imaging findings were subject to variation based on the developmental stage of the cysts. During the phase of a simple viable hydatid cyst (observed in 11 patients), the lesions demonstrated noticeable hyperintensity on fat-saturated T2-weighted images in comparison to the surrounding muscle tissue. These lesions displayed a uniform and cystic composition, with the cyst wall itself appearing hypointense. The contents of the cysts exhibited hypointensity on T1-weighted images and were either hypointense or hyperintense on T2-weighted images, when contrasted with the primary mother cyst (as depicted in Figure 1). In both stages, extensive edema was evident in the surrounding muscles and adjacent subcutaneous tissues, characterized by significant changes in high-signal intensity.

All the pathological investigastions reported with wide surgical margins. Post operative all the patients had albendazole . None of the patients had local or systemic recurrence.

In one patient, there was postoperative drainage which was monitored and managed through superficial debridement and the use of a negative drainage system. In two patients, discharge resulted from hematoma, and no further intervention was required for resolution. None of the patients experienced systemic anaphylaxis.

CT scans revealed the engagement of additional organs, such as the cranium, lung, and liver. Concurrent hepatic anomalies were observed in one patient, while a lung abnormality was detected in another patient with muscle hydatidosis. Surgical intervention and antihelminthic chemotherapy were employed to address these soft tissue anomalies prior to the excision of the muscular lesion. Among the patient cohort, the remaining 13 individuals exhibited solitary muscular lesions.

In 11 cases, the comprehensive blood cell count, electrolyte levels, erythrocyte sedimentation rate (ESR), and C-reactive protein values all fell within the standard range. However, elevated ESR levels were observed in 1 case involving muscle and another involving the liver. The Casoni test, also known as the indirect hemagglutination test, yielded positive results in 8 cases.

CT scans revealed the implication of other organs, including the lungs, liver and skull. Concomitant hepatic lesions were seen inone patient, lung lesions were seen in one patient with muscle hydatidosis. Surgical intervention and antihelminthic chemotherapy were employed to address these extramuscular lesions prior to the excision of the muscular lesion. The remaining 11 patients exhibited solitary muscular lesions.

In 6 cases, the full blood cell count, electrolyte levels, erythrocyte sedimentation rate (ESR), and C-reactive protein values were all within the normal range. However, 2 cases showed an elevated ESR level. The Casoni test, also referred to as the indirect hemagglutination test, yielded positive results in 5 cases.

Hydatid disease, also referred to as cystic Echinococcosis, is a parasitic condition originating from the larval stage of Echinococcus.[13]EchinococcusGranulosus and Echinococcus. Multilocularis, are responsible for this disease which is prevalent mainly in Australia, Asia, South America, Middle East and North-East Africa. While echinococcal cysts most frequently occur in the liver (62%) and lungs (20%), they have the potential to affect any region of the body[14].

The possibility of hydatid disease should be included in the list of potential diagnoses for any soft cystic mass in various anatomical locations, particularly in regions where the disease is prevalent.[15]

Musculoskeletal Echinococcosis is an uncommon scenario, accounting for only 3-5% of all cases. Several factors contribute to the infrequency of muscular involvement. Firstly, muscles are regarded as unsuitable habitats for Echinococcus due to their elevated lactic acid levels, which render them inhospitable for parasite growth. Secondly, the intramuscular growth of cysts is hindered by muscle contraction. Thirdly, the role of the hepatic barrier plays a part, as implantations in this area necessitate passing through the filtration mechanisms of the liver and lungs[16][10].

Diagnosing musculoskeletal hydatid disease is challenging both in clinical and radiographic terms. The cysts often remain asymptomatic for an extended period, and the radiographic alterations are typically lacking specificity. In cases of muscular lesions, the sole radiographic observation is usually the outline of a soft-tissue swelling.

Conventional X-rays displayed an increase in soft tissue density without any indications of calcifications or bone irregularities. Ultrasound examination of the right arm revealed intramuscular, hypoechogenic, and diverse formations within the soft tissue. Magnetic resonance imaging exhibited rounded, multivesicular muscular hydatid cysts.

Magnetic resonance imaging effectively illustrates the majority of characteristics associated with muscle hydatid disease. The morphological attributes and signal intensity characteristics vary based on the particular stages of cyst development[17-19].During the phase of a simple viable hydatid cyst, the cyst wall exhibits an isointense appearance relative to the fluid inside the cyst on T1-weighted images. On T2-weighted images, it presents as a low-signal intensity rim encircling the high-signal intensity contents.

During the phase when a hydatid cyst contains daughter cysts, the contents of both the mother and daughter cysts generally display similar intensity on both T1- and T2-weighted images. In the scenario where a hydatid cyst contains detached parasitic membranes, these membranes can be seen floating within the cyst, appearing as areas of darkness on both T1- and T2-weighted images. In the later stages of development, the cyst undergoes spontaneous collapse and calcification, leading to the formation of a calcified area within the surrounding tissue.

To verify the diagnosis, various common tests are typically employed, including serologic tests such as the indirect immunofluorescence antibody test, immunoelectrophoresis , ELISA and immunoblot test[13].

The Weinberg test, also known as the Complement Fixation test (CF), is a frequently utilized serological approach for diagnosing hydatid disease. The sensitivity of the CF test varies between 36% and 93%. Additionally, this method plays a significant role in postoperative assessment.[3]

Overall, MRI exhibits greater specificity and sensitivity compared to ultrasound for the detection of hepatic hydatidcysts[20].

Moreover, it provides enhanced insights into anatomical relationships and facilitates the surgical handling of cysts. For skeletal muscle hydatid cysts, confirming the definitive preoperative diagnosis is crucial. This process discourages treatment approaches such as marginal excision or incisional biopsy due to the risk of dissemination and the potential for anaphylactic shock upon cyst rupture[21]. As a result of this approach, we refrained from conducting any biopsies before the surgical procedures. In light of the radiological diagnoses, all patients were initiated on antihelminthic therapy. Additionally, the confirmation of diagnoses for all patients was achieved through serological tests.

Subsequent to the commencement of preoperative antihelminthic therapy, we conducted MRI and USG imaging to strategically plan the resection. Our stance is that resection of muscular hydatid cysts aligns with oncological principles. The significance of wide resection lies in its ability to prevent local recurrence and potential cyst rupture, which can trigger anaphylaxis. Hence, before proceeding with resection, having a clear understanding of the margins of the cystic mass is essential for achieving an extensive resection. In our series, we were able to successfully carry out wide resection in all 13 patients.

We firmly believe that wide resection plays a pivotal role in preventing both local and systemic recurrence, similar to its significance in sarcoma surgery. Notably, we haven't observed any instances of local recurrence or anaphylaxis, which we attribute to our consistent practice of wide resection across all patients. Furthermore, to attain the appropriate thickness for adequate wide resection, it's imperative to ensure that the cyst borders are sufficiently thick. During the surgical procedure, this border thickness safeguards against cyst damage and the potential dispersion of cystic materials. The achievement of this border thickness is aided by the administration of preoperative albendazole.

Histopathological analysis of the excised tissue specimens confirmed the presence of hydatidosis in every case. Soft tissue samples exhibited cysts characterized by an outer chitinous (fibrous laminar) layer and an inner germinal layer. This chitinous layer was enveloped by granulation tissue. The surrounding muscular tissues displayed an inflammatory infiltrate, with eosinophils being the predominant cell type.

The lesions were excised en-bloc, involving a wide margin of surrounding tissue (as detailed in Table I)Throughout all cases, wide surgical margins were successfully attained, and there were no instances of cyst material rupture or spillage.

All patients underwent a standard regimen of preoperative and postoperative adjuvant antihelminthic chemotherapy, involving albendazole. Albendazole was administered at a dosage of 15 mg/kg/day for patients weighing less than 60 kg, and at a dosage of 400 mg twice daily for patients weighing more than 60 kg. This treatment was continued for a duration ranging from 3 to 6 months.

Ten patients who presented with intramuscular hydatid cysts in their lower extremities achieved full clinical recovery within 2 to 4 months after surgery. They regained complete function and strength in their affected limbs. The sole patient with involvement in the lower extremity (gluteus medius muscle) experienced limited hip motion postoperatively. However, she remained pain-free and capable of performing daily activities throughout the follow-up period.

No cases of recurrence were observed in this series. At the latest follow-up, after an average of 43 months, all patients remained free of the disease. In the early postoperative phase, three patients (23%) encountered surgical complications, including one case of superficial infection and two cases of hematoma (as detailed in Table I). The superficial infection was successfully managed through local debridement and the use of a vacuum-assisted drainage system, coupled with antibiotic treatment.

This study is subject to several limitations. Firstly, its retrospective nature poses a constraint. Given the rarity of muscular hydatidosis, conducting a prospective study is a challenging endeavor. Secondly, the study's limited number of enrolled patients represents another limitation. Finally, the location of the lesions not being specific to either upper or lower extremities could also be considered a limitation.

Table 1. Demographics, age, gender, location,treatment, morphology,complications,follow-up

Casenumber	Age,gender	Location	Albendazole with wide resection	Cystmorphology (uniloculated-multiloculated)	Complications	Follow-up time
1	38 M	Left gastrocnem.	+	multiloculated	–	43months
2	42 M	Right vastuslateralis m.	+	multiloculated	–	38months
3	44F	Right gluteus medius m.	+	multiloculated	hematoma	34months
4	23 F	Rightbrachialis m.	+	uniloculated	–	60months
5	46 M	Righttibialis anterior m.	+	multiloculated	–	38months
6	34 F	Left gluteus maximus m.	+	multiloculated	superficialinfection	46months
7	41 M	Leftsoleus m.	+	multiloculated	–	32months
8	43 F	Rightinfraspinatus m.	+	multiloculated	–	44months
9	33 M	Rightgluteus maximus m.	+	multiloculated	–	34months
10	38F	Left adductor magnus m.	+	multiloculated	–	56months
11	15 M	Rightvastusmedialis m.	+	uniloculated	hematoma	35 months

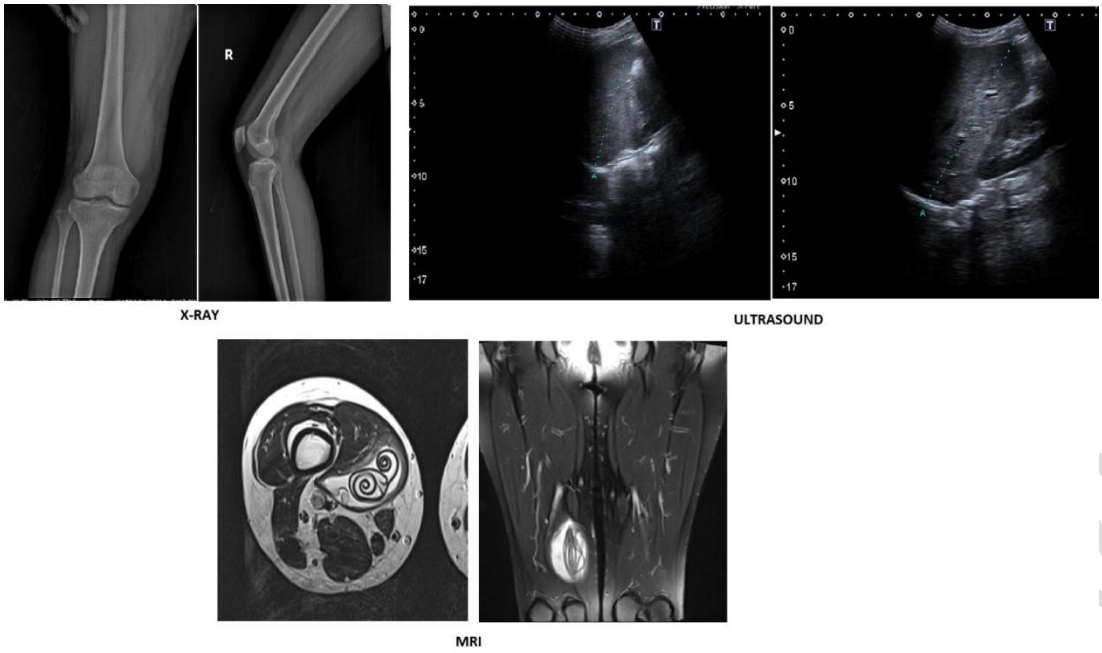

Fig. 1.Imaging the location of the muscular hydatid cyst in the vastusmedialis (x-ray, ultrasound and MRI)

4. CONCLUSION

Muscular hydatidosis should be taken into consideration when differentiating. Soft tissue tumors located in areas affected by the condition is prevalent. Achieving an accurate diagnosis through preoperative ultrasonography and MRI is vital, as avoiding biopsy is crucial to prevent potential intraoperative and postoperative complications. The same surgical principles utilized for resecting soft tissue sarcomas should be applied in the first management of muscular hydatid disease, with a strong emphasis on achieving wide surgical margins.
The combination of wide resection and albendazole treatment proves to be an effective approach for eliminating muscular hydatidosis.

CONSENT (WHEREEVER APPLICABLE)

"All authors declare that 'written informed consent was obtained from the patient (or other approved parties) for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editorial office/Chief Editor/Editorial Board members of this journal."

ETHICAL APPROVAL (WHEREEVER APPLICABLE)

"All authors hereby declare that all experiments have been examined and approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki."

DEFINITIONS, ACRONYMS, ABBREVIATIONS

HERE IS THE DEFINITIONS SECTION. THIS IS AN OPTIONAL SECTION.

TERM: DEFINITION FOR THE TERM

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