

Primary Soft Tissue Hydatid Cysts

Primer Yumuşak Doku Yerleşimli Kist Hidatik

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ABSTRACT

Objective: Hydatid cyst disease is a helminthic infection caused by *Echinococcus granulosus*, which is encountered with cysts in many organs, especially the liver and lungs. Soft tissue and intramuscular hydatid cyst are rare even in endemic countries. It is challenging to distinguish subcutaneous and intramuscular hydatid cysts from soft tissue tumors. This study aimed to present the clinical features of primary soft tissue hydatid cyst cases without liver and lung hydatid cyst in the Southeast Anatolian region, where hydatid cyst disease is endemic.

Methods: Patients admitted to the Şanlıurfa Training and Research Hospital General Surgery and Orthopedics and Traumatology Outpatient Clinic between September 2018 and December 2019 with complaints of pain and/or swelling under the skin and soft tissue were evaluated. After the examinations, the records of the patients who were operated on with a pre-diagnosis of hydatid cyst and whose histopathologic evaluation was reported as a hydatid cyst were reviewed retrospectively.

Results: Eight patients were included in the study. The mean age of the patients was 39.75±14.80 years. Lesions were located in neck (12.5%), left thoracic posterior area (25%), gluteus (25%), thigh (12.5%), right upper quadrant of abdominal wall (12.5%), and under the right clavicle (12.5%). When imaging methods were examined, ultrasonography was performed in 7 patients (87.5%), chest computed tomography was performed in 1 patient (12.5%), and magnetic resonance imaging was performed in 2 patients (25%).

Conclusion: Diagnosis of hydatid cyst should be considered in the differential diagnosis of soft tissue tumors in countries of endemic regions for hydatid cyst disease such as Southeastern Anatolia Region, Turkey.

Keywords: *Echinococcus granulosus*, hydatid cyst, soft tissue mass

ÖZ

Amaç: Kist hidatik hastalığı, başta karaciğer ve akciğer olmak üzere birçok organda kistlerle karşımıza çıkan, *Echinococcus granulosus* kaynaklı helmintik bir enfeksiyondur. Yumuşak doku ve kas içi kist hidatik, endemik ülkelerde bile nadirdir. Deri altı ve kas içi kist hidatikleri yumuşak doku tümörlerinden ayırt etmek zordur. Bu çalışmada, kist hidatik hastalığının endemik olduğu bölgemizde karaciğer ve akciğer kist hidatik öyküsü olmayan primer yumuşak doku yerleşimli kist hidatik olgularının klinik özelliklerinin sunulması amaçlanmıştır.

Yöntemler: Eylül 2018 ile Aralık 2019 tarihleri arasında Şanlıurfa Eğitim ve Araştırma Hastanesi Genel Cerrahi ve Ortopedi ve Travmatoloji Poliklinikleri'ne deri altında ve yumuşak dokuda ele gelen kitle, ağrı ve şişlik şikayeti ile başvuran olgular değerlendirilmiştir. Yapılan tetkikler sonrası kist hidatik ön tanısıyla ameliyata alınan ve patoloji sonucu kist hidatik olarak raporlanan hastaların kayıtları retrospektif olarak incelenmiştir.

Bulgular: Sekiz hasta çalışmaya dahil edilmiştir. Hastaların yaş ortalaması 39,75±14,80 idi. Lezyon yerleşim yerlerinin 1 (%12,5) hastada boyunda, 1 hastada sağ klavikula altında, 1 hastada karın duvarı sağ üst kadranda, 2 hastada sırtta vertebra lateralinde, 2 hastada gluteusda ve 1 hastada uylukta olduğu görülmüştür. Görüntüleme yöntemi olarak ise, 7 hastaya (%87,5) ultrasonografi yapıldığı, 1 hastaya (%12,5) toraks bilgisayarlı tomografi çekildiği ve 2 hastaya da (%25) manyetik rezonans görüntüleme yapıldığı saptanmıştır.

Sonuç: Kist hidatik, ülkemiz gibi endemik olan bölgelerde yumuşak doku kitlesi olan hastalarda akılda tutulmalıdır. Tedavide kist total olarak eksize edilmelidir.

Anahtar Kelimeler: *Echinococcus granulosus*, kist hidatik, yumuşak doku kitlesi



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INTRODUCTION

Hydatid cyst disease is a parasitic disease caused by *Echinococcus granulosus*, occurring most commonly in the liver and lungs. *Echinococcus granulosus* is transmitted to a human, an intermediate host, through infected food. Following the removal of the larvae in the infected food, it is absorbed from the duodenum and comes to the liver via the venous route and clings to the sinusoids, forming the hydatid cyst. Larvae that exceed the sinusoids cause disease in peripheral organs through systemic circulation (1,2). Hydatid cyst disease is most common in the liver (70%) and lungs (12%). Liver and lung act as a filter for interference. Rarely, parasites involved in the systemic circulation may also cause lesions in organs and soft tissues (3). Due to the presence of lactic acid, muscle tissue forms an unfavorable environment for the parasite, so hydatid cysts in the muscles are extremely rare (4). Soft tissue and intramuscular hydatid cysts are rare even in endemic countries for hydatid cyst disease (0.7-0.9%). It is extremely difficult to differentiate subcutaneous and intramuscular hydatid cysts from soft tissue tumors (5). Hydatid cyst disease is a pathology that should be immediately removed when determined, and its differentiation from other benign events of soft tissue should be made strictly.

This study aimed to contribute to the literature by evaluating the clinical features of our primary soft tissue hydatid cyst cases with no history of liver and lung hydatid cyst disease in the Southeast Anatolian region, where cyst hydatid disease is endemic.

METHODS

Patients admitted to the Şanlıurfa Training and Research Hospital General Surgery and Orthopedics and Traumatology outpatient clinic between September 2018 and December 2019 with complaints of pain and/or swelling under the skin and soft tissue were evaluated. The records of the patients who were operated on with a pre-diagnosis of hydatid cyst and whose postoperative pathology report was determined as a hydatid cyst were analyzed retrospectively. Patients' age, gender, liver and lung cyst hydatid history, serology test results, preoperative imaging [abdominal and/or superficial ultrasonography (USG), computed tomography (CT), magnetic resonance imaging (MRI)], and pathology reports were obtained from the hospital registry system and patient files. Only isolated soft tissue hydatid cyst cases were included in the study. Patients with hydatid cysts in the liver and/or lungs along with soft tissue, patients whose preoperative imaging and biochemical data were not available, patients who did not have proper imaging examinations, and refugees living in the camps were not included in the study. The study was performed according to Helsinki Declaration. Informed consent was obtained from the patients and/or their relatives.

Statistical Analysis

Statistical Package for the Social Sciences (SPSS 21 Inc., Chicago, IL, USA) computer software was used for bio-statistical analyses. The data were presented either as mean and standard deviation values or median with minimum-maximum values.

RESULTS

Between September 2018 and December 2019, 98 patient was operated at our hospital due to hydatid cyst. Eight patients with

primary soft tissue hydatid cysts were included in the study. Patients whose pathology results were reported as hydatid cysts did not have a history of hydatid cysts of the lungs or liver. Two patients were referred to the orthopedic outpatient clinic, and six were referred to the general surgery outpatient clinic. Three patients (37.5%) were male, and five patients (62.5%) were female. The mean age of the patients was 39.75 ± 14.80 years. All of the patients had lived in the Southeastern Anatolia region, an endemic region for hydatid cyst disease. Patients' complaints for the referral to the outpatient clinic were pain and/or swelling at the lesion site. The serology test (IHA) results examined in the preoperative and/or postoperative period were negative in 1 patient (12.5%) and positive in 7 patients (87.5%). Lesions were located in neck (12.5%), left thoracic posterior area (25%), gluteus (25%), thigh (12.5%), right upper quadrant of abdominal wall (12.5%), and under the right clavicle (12.5%). When imaging methods were examined, it was determined that USG was performed in 7 patients (87.5%), chest CT in 1 patient (12.5%), and MRI in 2 patients (25%) (Table 1). Lesions were reported as anechoic cystic lesions or heterogeneous hyperechoic lesions (hydatid cyst?) in USG. The CT of only patient was reported as "approximately 6x4 cm cystic lesion under the right clavicle" (Figure 1). MRI determined 45x23x30 mm cystic mass lesion in the left vastus lateralis muscle" in one patient and "cystic mass lesion in the right gluteal region, measuring 151x133x91 mm" in another (Figure 2, 3). In all patients, the lesions were completely excised without deteriorating the tissue integrity (Figure 1). Two patients were operated on under spinal anesthesia, and six patients were operated on under local anesthesia. Perioperative anaphylaxis and postoperative complications were not observed in patients. All patients were discharged on the first postoperative day without any complications. Albendazole 10-15 mg/kg was administered to patients for an average of 3 months in the postoperative period. Pathology results of all our patients were

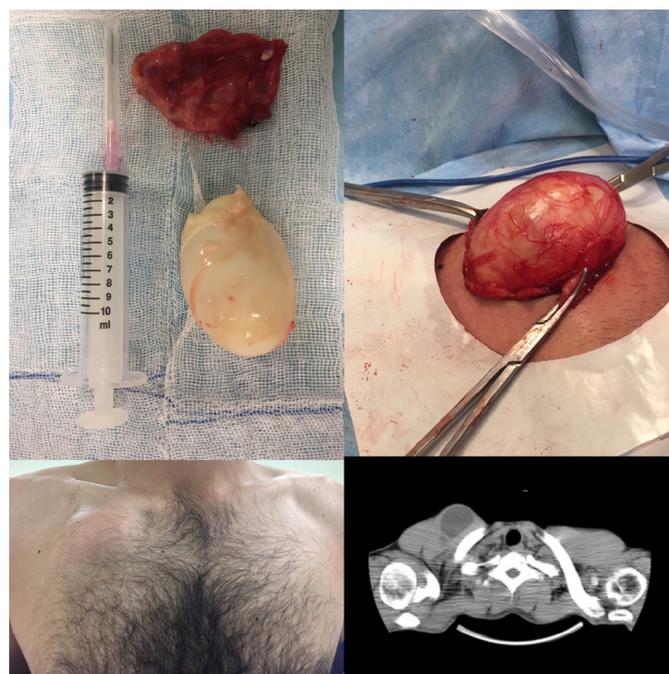


Figure 1. Images of the patient with a diagnosis of hydatid cyst on the anterior chest wall

compatible with hydatid cyst. Recurrence was not observed in any patients at 6 and 12 months of follow-up.

DISCUSSION

Hydatid cyst disease is widespread in most developing livestock raising countries such as Turkey, and hydatid cyst disease remains an important health problem. The organs where the disease is most common are the liver and lungs (6,7). Infrequent settlements besides the liver and lungs are spleen, soft tissue, abdomen, kidney, brain, bone, pancreas, breast, pelvis, joint, bladder, heart, ovary, thyroid, retroperitoneum, incision scar, and choledochal cord, in order of frequency (7). However, parasitic cysts tend to grow in the muscles of the trunk, neck, and legs, where blood flow is high, and muscle contraction is relatively low. Intramuscular hydatid cyst has been reported in pectoral major, sartorius, quadriceps and gluteus muscles (8,9). Gougoulias et al. (10) described cyst hydatid cases in the thigh, popliteal fossa, posterior humerus, and scapular region. Demirel et al. (11) presented retroperitoneal cases in the muscle behind the right hemithorax, under the left clavicle, and in the right iliac fossa. A case of primary subcutaneous lumbo-vertebral hydatid cyst was presented in a study (12). In Tekin et al.'s (13) study, a subcutaneous hydatid cyst in the neck was presented. In the study of Akbaş et al. (14), a primary hydatid cyst on the iliac muscle was presented during pregnancy. The initial symptoms are oftenly swelling and pain. In our cases, the complaints of the patients were pain and swelling as well.

It is vital to distinguish soft tissue and intramuscular hydatid cysts from soft tissue tumors. In the diagnosis of hydatid cyst, history, radiological imaging methods, and serological tests are used. Serological tests are diagnostic tests, but they are also used in post-treatment follow-up (15). Serological and immunological tests may be negative in the early period; therefore, radiological imaging methods are more reliable (16,17). Intravesicular female

cysts, commonly observed in liver hydatid cyst disease imaging, are generally not seen on USG or CT imaging of skeletal muscle cysts. Classic MRI findings are a multivesicular cyst, a low-density peripheral ring ("rim sign"), or a detached membrane on T2-weighted images (18,19). In only one of our patients, serological tests were negative, and in all remaining patients, serological tests were positive. Preoperative imaging methods have guided physicians at the point of diagnosis.

In patients presenting with pain and swelling in soft tissue, benign and malignant tumors should be considered (20). Diagnosis of hydatid cyst can be made by determining the layered membranes or hooks in fine-needle aspiration biopsy (FNAB). However, it should be remembered that during this procedure, scolexes may pass into the systemic circulation and cause an anaphylactic reaction or cause the disease to spread (15). The exact diagnosis is made by the histopathological methods (17,21).

The recommended treatment for soft tissue hydatid cysts is surgical excision (22). Physicians should be careful during the operation and should avoid contact with the tissue around the cyst. If there is contact, the recurrence rate increases and complications such as fever, eosinophilia, and anaphylactic shock may also occur. Albendazole treatment can be administered before and after surgery. The recommended dose of albendazole is 10-15 mg/kg/day for an average of 3 months (23,24). To ensure sterilization during surgery, it is recommended to wash the area with alcohol, formol (1%), and hypertonic sodium chloride (20%), where the cyst is removed (25). Total cystectomy was performed surgically in our cases. None of the patients had postoperative complications. Albendazole treatment was administered to all patients for three months.

Study Limitations

Our study has some limitations. First, its design was retrospective. Therefore, determining the clinical features of the patients

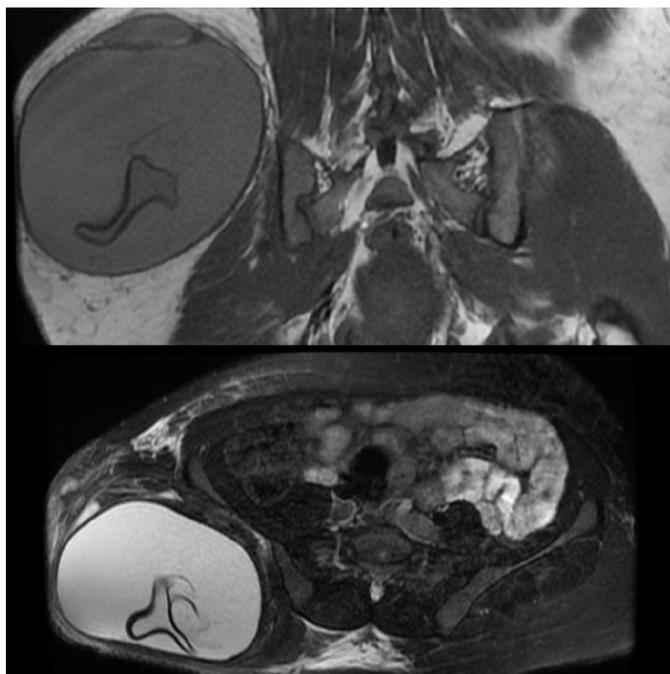


Figure 2. Pelvic MRI axial and coronal image

MRI: Magnetic resonance imaging

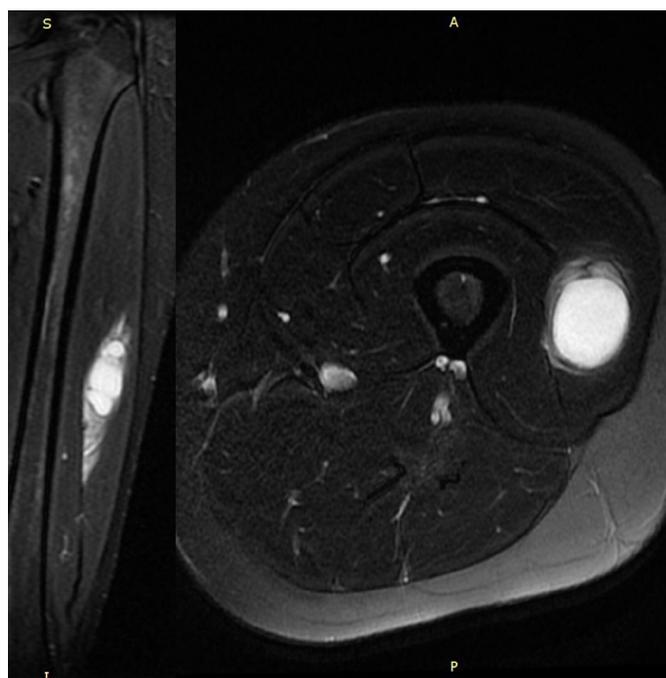


Figure 3. Left lower extremity contrasted MRI axial and coronal image

MRI: Magnetic resonance imaging

Table 1. The demographic, clinical, and radiological characteristics of the patients

Patient no	Age	Gender	Lesion location	Liver and lung hydatid cyst	Serology	Preoperative USG findings	Preoperative CT/MRI findings	Pathology
1	24	M	Under right clavícula	None	1/640	6x3.5 cm anechoic cystic lesion (hydatid cyst?)	6x4 cm cystic lesion under the right clavícula	Hydatid cyst
2	35	F	Neck	None	1/80	3x2 cm anechoic cystic lesion	None	Hydatid cyst
3	45	F	Torasic posterior area	None	1/320	11x6 cm cystic lesion about 2 cm from the skin	None	Hydatid cyst
4	67	M	Right gluteus	None	1/1.280	Heterogeneous hyperechoic 10x5 cm lesion containing cystic areas under the skin (hydatid cyst?)	None	Hydatid cyst
5	48	F	Left thoracic posterior area	None	1/640	5x4 cm anechoic cystic lesion under the skin (hydatid cyst?)	None	Hydatid cyst
6	33	F	Right gluteus	None	1/640	Revealed a thick-walled heterogeneous lesion of 106x70 mm in soft tissue in the right gluteal region	Right hip revealed cystic mass lesion sized as 151x133x91 mm with uniformly limited, localized lobulation including dissociated membranes subcutaneously located in the postero-superior of the right gluteal region and in the posterior of the iliac crest	Hydatid cyst
7	21	F	Left thigh	None	1/640	None	Athick-walled cystic mass, including a small amount of thin septa and microcysts, sized as 45x23x30 mm in the left vastus lateralis muscle	Hydatid cyst
8	45	M	Abdominal wall	None	1/320	7x4 cm heterogeneous hypoechoic lesion under the skin (hydatid cyst?)	None	Hydatid cyst

USG: Ultrasonography, CT: Computed tomography, MRI: Magnetic resonance imaging

was limited. Second, long-term results of patients could not be achieved.

CONCLUSION

In conclusion, hydatid cyst disease remains a serious public health problem in developing countries and in countries where livestock raising is widespread, such as Turkey. Hydatid cyst disease should be kept in mind in the differential diagnosis of soft tissue tumors, especially in patients presenting with pain and swelling in the endemic regions for hydatid cyst disease.

*Ethics

Ethics Committee Approval: Since it is a retrospective study, we did not apply for ethical committee approval.

Informed Consent: Informed consent was obtained from the patients and/or their relatives.

Peer-review: Internally peer-reviewed.

*Authorship Contributions

Surgical and Medical Practices: M.P., T.G., A.E.Y., Concept: M.P., D.A.Ç., G.P., A.E.Y., Design: T.G., G.P., Data Collection or Processing: D.A.Ç., T.G., A.E.Y., Analysis or Interpretation: M.P., D.A.Ç., Literature Search: D.A.Ç., G.P., Writing: M.P.

Conflict of Interest: No conflict of interest was declared by the authors.

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