

Primary Muscular Cystic Echinococcosis Disease: A Case Report and Review of the Literature

Primer Musküler Kistik Ekinokokkoz Hastalığı: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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Abstract

Cystic echinococcosis is a zoonotic disease that arises from the parasite called *Echinococcus granulosus*, which has an isolated primary muscle involvement. The clinical presentation of the disease is very similar to that of lipomas. In endemic regions, such as Türkiye, even if the physical examination findings are consistent with lipoma, primary muscular cystic echinococcosis disease should be considered in the differential diagnosis of soft tissue masses. In this case report, we present a rare case of primary muscular cystic echinococcosis that was diagnosed by histopathological examination who was admitted to our clinic with a mass in the left arm which was diagnosed as lipoma preoperatively, and which caused strong suspicion of cytic echinococcosis during the operation under general anesthesia.

Keywords: Echinococcosis, lipoma, parasite, soft tissue mass

Öz

Kistik ekinokokkoz, *Echinococcus granulosus* adlı parazitin neden olduğu zoonotik bir hastalık olup, bu hastalığın izole primer kas tutulumu ile giden bir formu da mevcuttur. Bu hastalığın kliniği yumuşak doku kitlelerinden özellikle lipomlar ile oldukça benzerdir. Fizik muayene bulguları lipomla uyumlu olsa bile Türkiye gibi ekinokokkoz hastalığının endemik olduğu bölgelerde yumuşak doku kitlelerinin ayırıcı tanısında mutlaka primer musküler kistik ekinokokkoz akla gelmelidir. Bu olgu sunumunda kliniğimize sol kolda kitle ile başvuran ve lipom ön tanısı ile genel anestezi ile eksizyon yapılırken, kistik ekinokokkozdan şüphelenilmesi üzerine yapılan histopatolojik incelemede primer musküler kistik ekinokokkoz tanısı konulmuş nadir görülen bir olgu sunulmaktadır.

Anahtar Kelimeler: Ekinokokkoz, lipom, parazit, yumuşak doku kitlesi

Introduction

Cystic echinococcosis is a parasitic disease arising from *Echinococcus* tapeworms (1). *Echinococcus granulosus* is responsible for approximately 99% and *Echinococcus multilocularis* is responsible for approximately 1% of the total number of echinococcosis (2). Adult parasites are mainly located in the small intestines of carnivorous animals. Parasitic eggs from the feces of these animals are transmitted to humans by

the fecal-oral route (2). Cysts can localize in virtually any organ and structure, such as abdominal or pleural cavities, kidney, spleen, bone, brain, eye, ovary, testis, and pancreas (3).

Case Presentation

A 32-year-old female patient was admitted to our clinic with a complaint of a mass in her left arm that was noticed eight months before. The patient's medical history revealed that she was living in Ankara, Türkiye. Physical examination revealed

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a 3x2 cm mass in the posterior aspect of the left arm between the musculus deltoideus and musculus triceps brachii. There was no erythema or change in skin secondary to inflammation. The palpation revealed a soft, painless, non-fluctuating mass that was fixed to subcutaneous tissues. The preoperative appearance of the patient is shown in Figure 1.

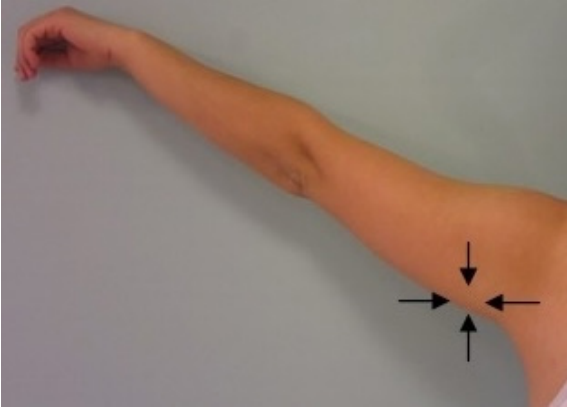


Figure 1: Image of the mass lesion at the time of application

Lipoma was considered as a preoperative diagnosis due to the ovoid shape of the mass, localization at the extremity, more common incidence of lipomas in women and absence of signs, such as aggressive growth and homogeneity loss, which suggests liposarcoma. Since most of the lipomas are located subcutaneously and do not require diagnostic imaging, the patient was prepared for excision under general anesthesia with the preliminary diagnosis of lipoma.

A 3 cm incision on the posterior aspect of the left arm was made to reach the mass. The mass was dissected from the surrounding tissues. When the mass was reached, it was seen that it was not a subcutaneous lipoma, rather a lesion located between the muscles and separated from the surrounding tissues by a fibrous capsule. The capsule was opened, and a white membranous structure was encountered. The lesion was thought to be germinative membrane because of the white and firm appearance, and the lesion was approached as a hydatid cyst. The intramembranous transparent colored viscous fluid was aspirated. The capsule structure and the white-colored germinative membrane were excised without contact with surrounding tissues free of complications. The pouch was washed with a protoscolocidal hypertonic saline solution. Albendazole 400 mg 2x1, per oral was added empirically to the patient's postoperative medication. The structures encountered during excision of the patient's mass are shown in Figure 2. Radiological imaging was performed on the patient to investigate the primary focus, and no other organs were affected.

The pathological examination was reported as hydatid cuticle and cyst wall. No complication was observed during follow-up. The pathological material is shown in Figure 3.

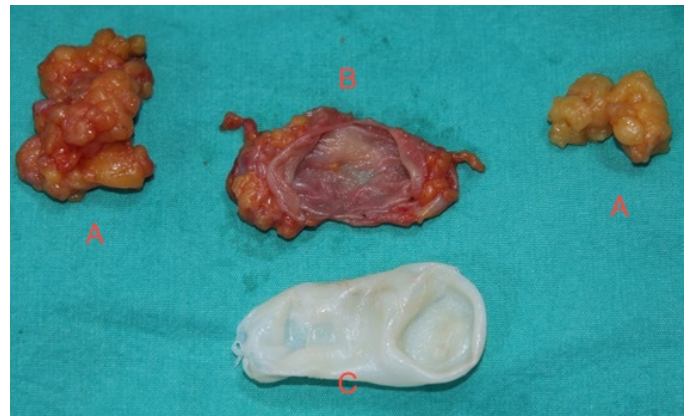


Figure 2: Intraoperative view of pericapsular adipose tissue, fibrous capsule and white germinative membrane. A) Pericapsular adipose tissue, B) Capsule structure, C) Germinative membrane

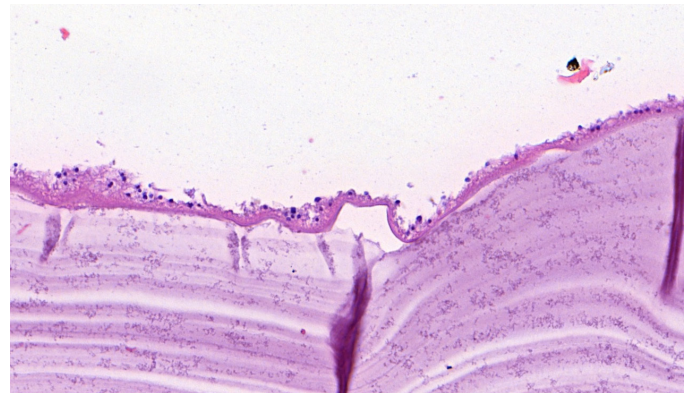


Figure 3: The eosinophilic lamellar hydatid cyst cuticle and superficially located germinative membrane, H-Ex30

Discussion

To perform a review of the literature about the intramuscular hydatid cysts, we used the keywords "hydatid cyst" and "intramuscular" on PubMed, without a time limit. Fifty-six articles were composed of 36 case reports, four case reports and the review of the literature, 12 experimental studies, four clinical trials in a period from 1970 to 2018. Forty-one articles mention about the localization of the hydatid cysts. More details are given in Figure 4.

There are fifteen cases from Türkiye, nine from India, five from Russia, three from the USA and Italy, two from Lebanon and Iraq, and one from Israel, Austria, England, China, Saudi Arabia, Albania, Brazil, Tunisia, Greece, Spain and Nigeria. The geographical distribution of the intramuscular hydatid cysts is given in Figure 5. The sex distribution of 39 intramuscular hydatid cysts cases were 21 (53.8%) female and 18 (46.2%) male. Of nine cases in the literature, four patients were farmers, two were housewives, two were construction workers, and one was a medical student.

ARTICLES

TOTAL:56

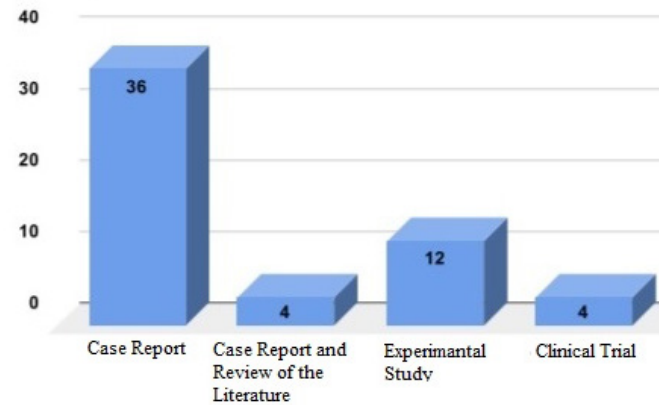


Figure 4: Forty-one articles mentioned about the localization of the hydatid cysts

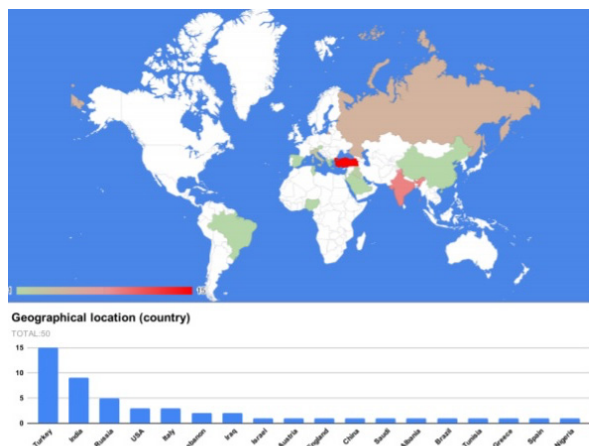


Figure 5: The geographical distribution of the intramuscular hydatid cyst cases

When the preoperative imaging was inspected, the most frequent used radiological imaging was magnetic resonance imaging (MRI) with 26 cases which are followed by detailed ultrasound (USG) in 23 cases and X-ray imaging in 22 cases. Computed tomography was used less frequently in 12 cases and fine needle aspiration biopsy was used only in three cases.

The symptoms of the patients seem to be in a wide range of the spectrum. While the majority of the patients with 18 cases had only pain, there was no pain in 13 patients. Five patients complained with limitation of movement, four patients with erythema, two patients with pruritus, two patients with pyrexia, and one patient with paresthesia. Present symptoms are given in Figure 6.

Humans are the intermediate host of the parasitic life cycle of the disease (1). The most common organs that the disease is seen in human are liver with a ratio of 50-70%, lung with a ratio of

Symptoms

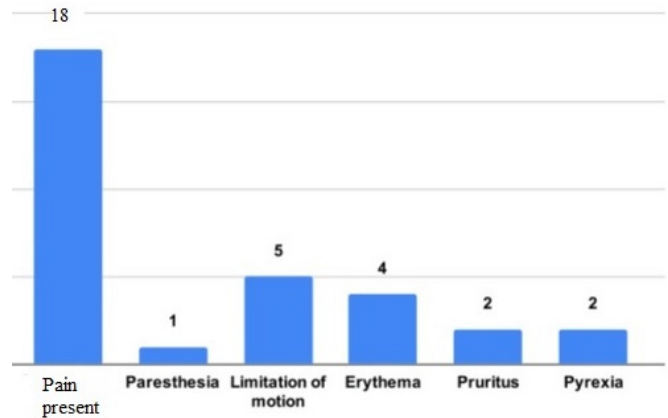


Figure 6: The most common symptoms of the patients

11-17%, soft tissues with a ratio of 2.4-5.3%, and subcutaneous tissues with a ratio of 0.5-4.7% (2). When the literature is reviewed, it is seen that, between 1991 and 2014, 242 of the 282 articles mentioning soft tissue involvement of hydatid cysts have muscle or bone involvement. In the remaining 40 articles, there are cases of hydatid disease of the skin and subcutaneous cyst without muscle or bone involvement (4). Rarely, cysts are in striated muscles, resulting in a primary muscular hydatid cyst. The reason why the primary muscular disease is as rare as 3% is thought to be the lactic acid in striated muscles, which makes the environment inappropriate to strive for larvae (1,2). The most commonly involved muscles are supraspinatus, biceps brachii, gracilis, and quadriceps femoris (5). Hydatid cysts mostly present in the proximal area of the extremities due to the higher blood supply of the proximal muscles (6). Figure 7 reveals the localization of the disease. Although the hydatid cyst can be seen at any age, the disease peaks in the 30-50 age (1). The most common endemic areas of hydatid disease are the Mediterranean, Middle East, Central Asian countries, New Zealand and Australia (7,8). Especially in the eastern parts of Türkiye, echinococcosis is still commonly encountered, and the disease is regarded as a public health problem (6). The study conducted by Gun et al. (6) with 329 echinococcosis cases, revealed that the disease presents in females more than males. The hydatid cyst may be

Localization

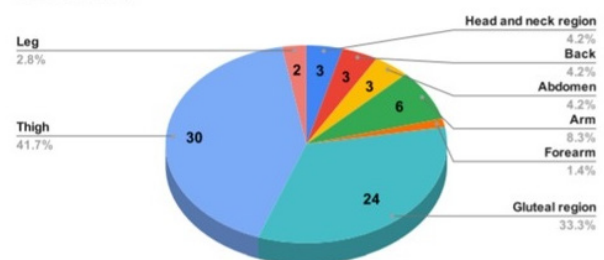


Figure 7: The number and the percentage of the localizations of the intramuscular hydatid cysts

associated with the patient's occupation and history of contact with animals in endemic areas (5). In the patient's history, which was reevaluated postoperatively, it was found out that the patient was housewife and had cattle and had been feeding a dog at home. Although the primary muscular hydatid cyst is usually asymptomatic, it reveals itself as a painless mass in most patients. Therefore, radiological imaging should be used for diagnosis. Because calcification can be seen in only 38% of plain radiographs, utilizing USG and MRI is more efficient in diagnosis (1). The daughter cysts that appear on imaging and constitute the living form of cysts are pathognomic for the disease and show the viability of cystic structures (1). Lipoma, sebaceous cyst, calcified hematoma, abscess, and soft tissue tumors must be thought in the differential diagnosis of hydatid cyst (9). Lipomas are usually round or ovoid, usually located subcutaneously and do not require imaging (10). Considering that lipomas are the most common benign tumor of soft tissues and have the most important place in the differential diagnosis of hydatid cysts, the incidence of lipomas is higher among female patients and may increase up to 16% (10), whereas the incidence of echinococcosis varies between 1-220 in 100,000 patients even in the endemic areas (11,12).

Indirect hemagglutination test, which is one of the serological diagnostic methods, has a 90% sensitivity for hydatid cyst in the liver (13). However, and it can give false-negative results up to 50% in patients with solitary lung cyst (14). In this manner, the importance of utility of radiological and serological tests together in the diagnosis of echinococcosis should be kept in mind (13). The sensitivity and specificity of serological tests in with soft-tissue involvement reduce to 25% (15). The postoperative indirect hemagglutination test was negative for the patient mentioned in this case report. In 18 cases in the literature, indirect hemagglutination tests were used to diagnose intramuscular hydatid cysts. Eleven positive and seven negative results were obtained.

Treatment of hydatid cyst disease is the total excision of cysts without allowing the rupture of cysts and dissolution of cyst contents (1). The rupture of cysts during surgery causes the discharge of antigenic and toxic cyst contents and may cause local or systemic spread and anaphylactic reaction (5,14). The patient's mass was dissected completely from the surrounding tissues and excised without any tearing or shedding. It is recommended that medical antihelminthic treatment should be added to surgical treatment for 1-2 weeks preoperatively and 3-6 weeks postoperatively (2). The patient in the case report was not suspected to have echinococcosis preoperatively, hence preoperative treatment was not started, but in the postoperative period, 2 weeks of medical treatment was arranged.

Conclusion

In this case report, a patient with isolated muscle involvement without systemic hydatid cysts findings is

presented. Given the rarity of intramuscular cyst hydatid disease, it is crucial for clinicians to consider it in differential diagnoses, as early detection and appropriate treatment are essential for preventing complications.

Ethics

Informed Consent: Oral and written informed consent were obtained.

Authorship Contributions

Concept: M.E., Design: M.E., Data Collection or Processing: A.Ö., Analysis and/or Interpretation: S.S., Literature Search: A.Ö., Writing: A.Ö.

Conflict of Interest: The authors declared that there was no conflict of interest during the preparation and publication of this article.

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