

Muscular hydatid cyst in the calf – a clinical case

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Abstract— Muscular echinococcosis is a rare parasitic disease caused by the tapeworms *Echinococcus granulosus* and *Echinococcus multilocularis*. Clinical symptoms are non-specific and appear late, usually when the hydatid cyst is of significant size. The combination of diagnostic imaging studies and serological tests is the key to making a correct diagnosis. The treatment of muscular echinococcosis is mainly surgical, in the form of complete resection of the hydatid cyst. Adjuvant anthelmintic therapy is also administered to reduce the risk of hydatid cyst recurrence.

We present a clinical case of a 17-year-old girl with muscular echinococcosis, diagnosed and treated in our clinic.

Keywords—hydatid cyst, echinococcosis

1. Introduction:

Echinococcosis is a zoonosis caused by the parasites *Echinococcus granulosus* and *Echinococcus multilocularis* from the *Taenia* family¹. Infection in humans occurs most often when consuming contaminated food or water, as well as through close contact with some of the final hosts - dogs or wolves. The most commonly affected organs in humans are the liver, lungs and brain. The muscular form of echinococcosis is rare, accounting for between 0.5 and 0.9% of all cases².

The clinical presentation of muscular echinococcosis is non-specific, and often the first complaints come from the compression of adjacent vascular and nerve structures by the growing cyst^{1,2,3,4}. In some cases, the only sign of the disease is a slowly growing formation, which requires a differential diagnosis with a primary soft tissue tumor. The results of diagnostic imaging studies and especially conventional radiography are also non-specific and may lead to wrong diagnosis and inadequate therapeutic behavior⁵. Serological diagnosis of echinococcosis is possible by a hemagglutination test and can be of great benefit as an additional method to establish the correct diagnosis⁶.

The treatment of muscular echinococcosis is mostly operative, in the form of en bloc excision of the entire cyst^{2,3,4}. Care must be taken to preserve the integrity of the cyst due to the risk of anaphylactic shock and disease dissemination in the event of intraoperative rupture. An alternative method is percutaneous aspiration of the cyst con-

tents with subsequent infusion of scoliced drugs under ultrasound or CT control¹. Adjuvant anthelmintic therapy is needed in most patients due to the high frequency of disease recurrence. We present a rare case of an echinococcal cyst located in the musculature of the lower leg of a 17-year-old female patient.

2. Clinical case:

In 2020 a 17-year-old girl with complaints of a slow growing soft tissue formation in the right calf was admitted in the bony pathology ward of the University Orthopedic Hospital Boycho Boychev. The patient did not report any other complaints or a traumatic event. No other previous illnesses or surgeries were noted. Clinical examination revealed an oval soft-tissue formation with a dense-elastic consistency located on the dorso-medial surface of the right calf. The described finding was about 10 cm in diameter, painless and movable. The range of motion in the right knee and ankle joint was without limitation. No abnormalities were found in the vascular-nervous status of the limb. Initially, a conventional radiograph was performed, which revealed an enlarged soft-tissue shadow corresponding to the triceps surae muscle. An MRI followed, which revealed a lobulated heterogeneous formation with isointense signal on T1 and hyperintense on T2 sequence, filled with multiple oval cystic formations (Fig.1). The described finding was located entirely in the posterior muscle group of the lower leg and more precisely in m. gastrocnemius. After clinical discussion and in view of the results of the imaging studies, it was decided that the most likely diagnosis is muscular echinococcosis, necessitating surgical treatment. Through a longitudinal skin incision on the dorsal surface of the right calf, the formation localized intramuscularly in m. gastrocnemius was reached. After incision, a cavity filled with pus-like fluid and numerous small whitish structures resembling pieces of a surgical glove were noted. Material was taken for histological examination. Extensive drainage and lavage of the cavity was performed. This was followed by placement of an aspiration drain and closure of the operative wound. After a smooth postoperative period, the patient was discharged without any walking aids. Histological examination confirmed the diagnosis of hydatid cyst. The patient was referred for subsequent adjuvant anthelmintic treatment. At the follow-up examination after 6 months and 1 year postoperatively, no recurrence of the cyst was detected.

3. Discussion:

Echinococcosis is most commonly caused by the tapeworm *Echinococcus granulosus*^{1,2,3,4}. The disease has an endemic distribution, being most often found in the Mediterranean, the Middle East, South America and Africa⁷. After the parasite enters the bloodstream, it is most often filtered through the liver or lungs, which are the target organs of the parasite. Isolated localization in the skeletal muscles without the presence of cysts in other organs is rare. The presence of lactic acid and the contractile ability of

the muscles make it an unfavorable environment for the development of the parasite. The muscles of the thigh, lower leg and shoulder girdle are most often affected ^{2,3}.

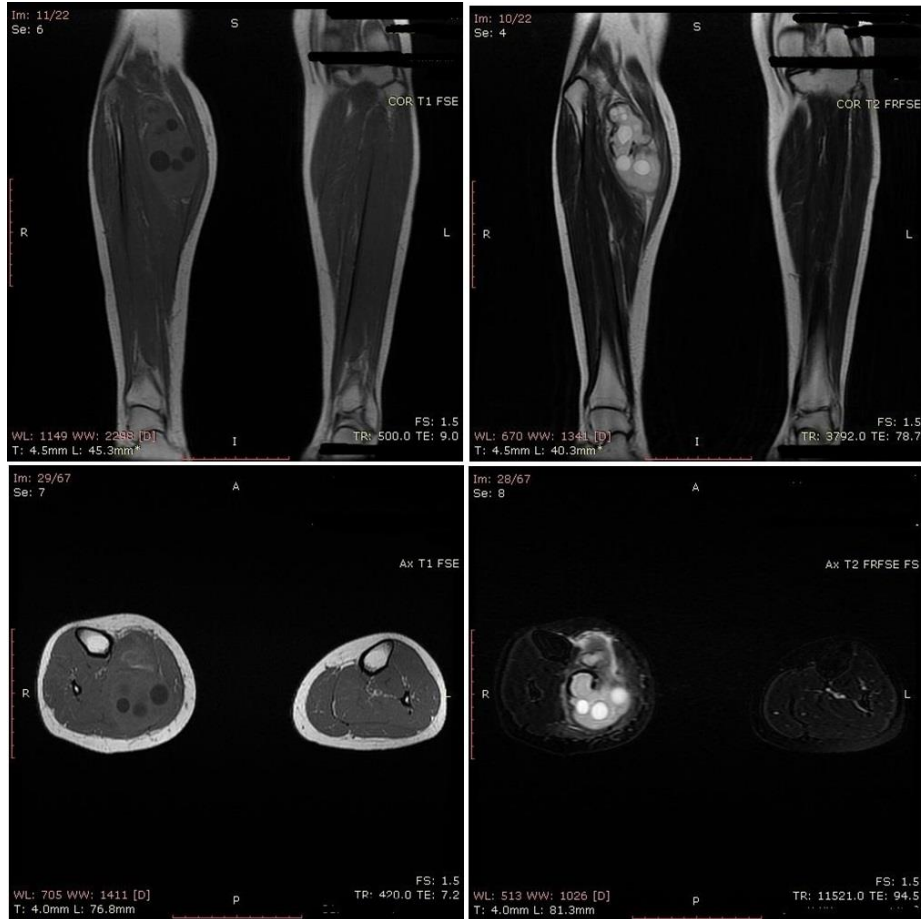


Figure.1 MRI showing a cystic formation with low signal on T1 and high signal on T2 sequence. Multiple secondary cystic formations are found inside the lesion.

When the parasite infiltrates the skeletal muscles, an echinococcal cyst is formed, which has 3 layers⁷. The outer layer is made of a dense fibrous membrane and is called the pericyst. The middle layer does not contain cells and serves for the filtration of nutrients. The inner layer is the place of production of scolices, which are the larvae of the parasite. With the growth of the cyst numerous daughter vesicles are formed, the fusion of which results in the formation of a daughter cyst. The content of the vital echinococcal cyst is clear to pale yellow. The fluid is highly antigenic due to its high protein content.

In the initial phase of the disease, there are usually no clinical symptoms. Very often, complaints appear when the echinococcus cyst is of significant size. Cases have been described in which the first manifestation of the disease is a loss of sensation or paresthesia coming from compression of peripheral nerves². Of the diagnostic imaging studies, the most valuable is MRI, where the detection of a cystic formation filled with numerous small daughter cysts is pathognomonic for echinococcosis. Usually, the cyst has a hypointense signal on T1 and a hyperintense signal on T2 sequence, and in most cases the peripheral part of the cyst has a low signal more commonly known as low-intensity rim sign⁵. Ultrasound diagnostics is an easy and fast method for differentiating muscular echinococcosis with a sensitivity of almost 100%². Computer tomography can also be used to diagnose the disease. A uni- or multilocular cyst filled with fluid and soft tissue expansion is well appreciated. Conventional radiography does not provide reliable information and should be used as a method to exclude other pathology². Serological diagnosis has a rather complementary role in muscular echinococcosis, due to its relatively low sensitivity and specificity. The number of false positive samples reaches 25% and the number of negative samples 30%. The most commonly used tests are ELISA and indirect hemagglutination⁶.

In terms of differential diagnosis, muscular echinococcosis must be distinguished from a primary soft tissue tumor^{1,2,3,4,5}. Usually, the distinction between the two pathologies is made by performing an MRI or CT scan, which reveals the previously described finding in muscular echinococcosis. Soft tissue abscess is another pathology that should be included in the differential diagnosis. Unlike muscular echinococcosis, changes in the patient's laboratory parameters will be present, as well as possible febrile episodes.

Due to the risk of anaphylactic shock and local dissemination of echinococcosis, open biopsy and marginal excision are contraindicated^{1,2}. The method of choice is the complete excision of the cyst without disturbing its integrity. In inoperable cases, the so-called PAIR method is applied, characterized by percutaneous puncture of the cyst, aspiration, irrigation with scolicidal agents and subsequent respiration². This type of procedure is usually performed under ultrasound or CT guidance. Adjuvant anthelmintic therapy is mainly carried out with Albendazole and Praziquantel for 6 weeks post-operatively in order to reduce the risk of disease recurrence, especially in cases where there is intraoperative rupture of the cyst².

4. Conclusion

Muscular echinococcosis is a rare parasitic disease, but nevertheless it should be present in the differential diagnosis of soft tissue formations. Inadequate treatment of the disease can lead to local dissemination, which will ultimately worsen the function of the affected limb and the patient's quality of life.

5. References

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