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Rare Location of Hydatid Cyst in the Thigh

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Abstract: Primary muscular hydatidosis is very rare. 52 year old male presented to surgical OPD with the complaint of right thigh swelling for 2 years. Initially swelling was small and painless, then gradually it was increased in size and became painful. MRI was advised for the characterization of the swelling. MRI showed the presence of voluminous cystic mass in the adductor compartment of thigh having peculiar imaging features of hydatid cyst. Correlative ultrasound also supported the MRI findings. Furthermore serology also came out to be positive for Echinococcus infection. The patient underwent surgical excision of the cyst followed by histopathological analysis which confirmed the diagnosis of primary muscular hydatid disease.

Key words: Hydatid cyst · Thigh · Rare location

INTRODUCTION

Hydatid disease is a systemic parasitic disease that is endemic in many countries of the world and is caused by the larva of Echinococcus tapeworm. The word 'hydatid' has Greek origin which means 'watery vesicle' [1]. There are two types of Echinococcus infections. E granulosus and E multilocularis, the former one is the more common type and classically leads to unilocular cystic form. Hydatid disease is an endemic disease in the underdeveloped areas where animal husbandry is common but there is no efficient veterinary control (e.g.), parts of South America, the Mediterranean region, the Middle East, Africa and Australia) [2]. The life cycle of Echinococcus consists of two hosts. Dogs are definitive hosts while sheep are intermediate hosts. Humans can become intermediate hosts by the ingestion of food or water that has been contaminated by dog feces containing the eggs of the parasite [2]. Hydatid cyst can occur almost in any location in human body from head to toe, most commonly involving the liver in 75% of cases, the lungs in 15% and other anatomic locations in 10% [3].

It is essential that before proceed for any intervention keeping in view the differential diagnosis of hydatid cyst, firstly diagnosis of hydatid disease should be ruled out, so as to avoid intra operative rupture and leakage of cyst contents leading to anaphylaxis and secondary hydatidosis [4].

Differential diagnosis of hydatid disease should be considered for every soft cystic mass in any anatomical location, especially in areas where the disease is endemic [4].

Case Report: 52 year aged male presented with the complaints of right thigh diffuse swelling for 2 years. He was a farmer by profession and had no significant past medical history. At first swelling was small and painless. Gradually it was increased in size and became painful for last 2 months. There was no history of fever or trauma. On physical examination, there was 10 x 10 cm size swelling involving the posteromedial aspect of right thigh. The swelling was tender and firm in consistency. No overlying skin changes. Rest of general physical examination was unremarkable.

On the basis of clinical assessment, MRI was suggested to rule out possibility of sarcomatous lesion and further characterization. Surprisingly the MRI depicted a voluminous intramuscular cystic mass in the adductor compartment of right thigh measuring 21 x 9 x 10 cm in CC, AP and transverse dimensions. The mass showed peripheral post contrast enhancement along with intra substance crumpled floating membranes forming serpent sign, characteristic of hydatid cyst.

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Fig. 1: On physical examination, there was large swelling on the posterior aspect of thigh

Fig. 2: Coronal T2WI FAT SAT image showing a large hyper intense lesion in the adductor compartment of thigh having low signal internal floating membranes and peripheral low signal intensity rim forming serpent and rim signs respectively. No Surrounding edema is seen

Correlative ultrasound also showed the similar findings having a large cystic lesion containing fine debris and floating membranes forming a waterlily sign. Furthermore X ray chest and abdominal USG were done to look for any visceral location of hydatid cyst.

So keeping in view radiological features, rare diagnosis of primary muscular hydatid cyst was made and referred to surgery department for further management.

Echinococcal antibodies came out to be positive on serum serology. Complete excision of the cystic mass was done under general anesthesia and histopathology likewise validated the diagnosis of muscular hydatidosis (Histopathological images are not available).

Fig. 3: Axial T1WI FAT SAT Post Gadolinium image showing peripheral enhancement of intramuscular lesion having iso intense signals and containing low signal intensity floating membranes

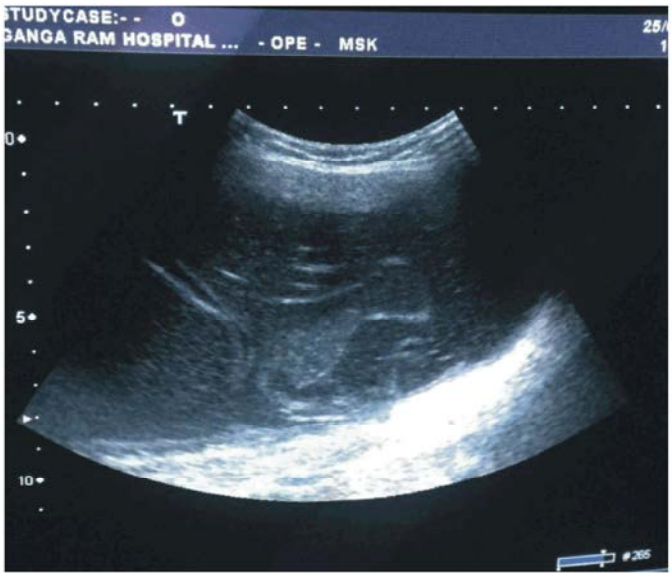

Fig. 4: USG image showing a large cystic lesion in the thigh having internal debris along with multiple crumpled echogenic floating membranes forming a characteristic waterlily sign

Fig. 5: Intra operative image showing the excision of hydatid cyst

DISCUSSION

The musculoskeletal form of hydatid disease can present both as primary and secondary disease. In secondary form, mostly the primary disease involves liver, lung or spleen and the hydatid cysts may spread from these sites to involve the musculoskeletal system secondarily, either spontaneously or as result of previous surgery of the primary diseased sites. The primary involvement of musculoskeletal system by the hydatid cysts without any other visceral cyst is extremely rare even in endemic countries, where its frequency varies from 1 to 5 % [5].

The continual muscle contraction and the production of lactic acid makes the hostile environment for echinococcal infection. That's why the muscular hydatid cysts develop rarely. However among the muscles, neck, trunk and limb root muscles are more commonly infested by Echinococcus due to decreased activity and profuse vascularization of these areas [5].

The clinical symptoms were mostly non-specific and included painless slowly growing swelling over the course of years while unaffecting the patient's general health. However infrequently hydatid cysts present due to their complications, such as neural compression or infection raising the concern for abscess or some neoplastic tumor [5].

Serological tests and ultrasonography should be performed initially for the diagnosis of hydatid disease. ELISA is 80-100% sensitive and 88-96% specific for hydatid liver disease but less sensitive for lung (50-56%) or other organ involvement (25-26%). Hydatid serology is only valuable when it is positive, negative serology does not exclude the diagnosis [4].

Ultrasonography is very helpful noninvasive and readily available diagnostic tool for detection of hydatid disease of soft tissue. The sensitivity of US is 95% and if vesicular fibrils are present the sensitivity of US increases to 100% [4].

Recently, WHO proposed an international consensus classification of US features of cystic echinococcosis (CE) [6]. According to this classification, hydatid cysts are categorized in five categories. In category 1, there are unilocular simple cysts with anechoic internal contents. Moreover snow flake sign relating to hydatid sand; pathognomonic sign can also be seen in category 1. Category 2 cysts are multiseptated and multivesicular. Wheel-like and honeycomb-like structures are formed by the septae and daughter cysts respectively. Category 3 cysts have predominantly anechoic contents with internal floating membrane (waterlily sign); as also seen in our case. Category 4 cysts have characteristic ball of wool sign due to heterogeneous degenerating membranes and contain no daughter cysts. Category 5 cysts are characterized by a thick calcified wall.

MRI is the first choice diagnostic method for the evaluation of hydatid disease in the soft tissues. It is the best modality to detect the locoregional extent of the lesion along with involvement of neighboring structures including neurovascular bundle and to plan the surgical excision of the lesion [2, 4, 5].

MRI is capable of adequately demonstrating most features of hydatid disease, with the exception of calcification [4]. On MRI, hydatid cysts commonly display iso to hypo intense signals on T1WI and hyper intense signals on T2WI [1]. Typically, the cyst wall and septae will demonstrate post contrast enhancement and pericystic hypointense rim seen on T2WI forming a "rim sign" that is reminiscent of hydatid or endocyst membrane [2, 5].

Another important sign "snake sign" or "serpent sign" is seen on MRI in the form of collapsed and detached parasitic membranes, displaying low intensity signals on all sequences and represents the degenerating hydatid cyst [2]. Both rim sign and serpent sign are illustrated in our case report leading us to the radiological diagnosis of hydatid cyst.

Surgery is the treatment of choice of muscular hydatid cyst. Pericystectomy is performed in which entire cyst is excised without breaking the wall. However to prevent local spread of cyst contents, the remaining cavity is washed with hypertonic saline [5]. There is another method of avoiding anaphylactic reaction i.e. injecting intraoperatively mixture of 0.5% cetrimide, 15% hypertonic saline and 0.5% silver nitrate solution, before the cyst opening. That will effectively kill the daughter cysts and avoid the risk of dissemination [7].

CONCLUSIONS

The purpose of this case report is to always consider hydatid cyst as a differential of any cystic soft tissue swelling, especially in a patient living in the endemic areas for hydatidosis. Prompt radiological diagnosis of hydatid cyst before any intervention is vital in preventing the possible anaphylactic shock due to disease dissemination. Moreover MRI has been the radiological study of choice in detecting the hydatid cyst in soft tissue, its locoregional extension and to look for possible complications.

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