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Case Report

A rare location of primary extrahepatic hydatid cyst in the soft tissue

S. Uma Shanker[1]*, U. V. Sai Sreenivas[1], N. Swathanthra[2]

[1]Department of General Surgery, [2]Department of Pathology, Lalitha Super Specialties Hospital, Guntur, Andhra Pradesh, India

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***Correspondence:**
Dr. S. Uma Shanker,
E-mail: drsurgeon77@yahoo.co.in

ABSTRACT

Hydatid cyst or cystic echinococcosis in human is rare disease caused by tapeworm *Echinococcus* granulosa. Hydatid cyst of soft tissue is a rare condition and we had a case of large hydatid cyst in lumbar region. This is a case of 48 years old female presented to us during October 2018 with a large diffuse swelling over the left lumbar region which was gradually grown over the last 2 years without pain. The diagnosis of hydatid cyst was not sure even in CT scan but it was in our differential diagnosis. It is confirmed during surgery after seen the multiple cyst removed from the large cystic lesion. No recurrence even after 15 months of follow up. Presence of hydatid cyst in soft tissue is usually rare and its diagnosis is not expected clinically when it presents as a soft tissue swelling. We need to confirm it during surgery or by presence of typical germinal membrane under microscope.

Keywords: Hydatid cyst, Extra hepatic hydatid cyst, Soft tissue hydatid cyst

INTRODUCTION

Hydatid cyst or *echinococcus* is a zoonotic disease caused by *Echinococcus granulosus* a larva of Cestode species.[2] The definitive host are dogs and other canids but they differ in their choice of intermediate host. Even though it is prevalent globally, in India, these diseases are commonly seen in rural areas of Andhra Pradesh and Tamil Nadu.[3] In humans, it commonly affects the visceral organs like liver (75%), lungs (15%), spleen (5%) and other organs (5%).[1] Diagnosis of hydatid disease in humans are usually based on epidemiological background of the patients, clinical examination, radiological investigations like ultrasound of suspected part of the body, X-ray and computerized tomography etc. The diagnosis can be confirmed further by detecting specific antibodies (immunomodulation test). Other tests like, intradermal Casoni's test, human basophil degradation test and complement fixation test have only historical relevance. Incidence of extrahepatic that too soft tissue hydatid cyst is extremely rare and difficult to diagnose. This case of extrahepatic primary soft tissue large hydatid cyst is diagnosed per operatively.

CASE REPORT

A 67 years old female from the rural part of our place came with a swelling in her left loin. According to her the swelling was noticed two years ago from the time of presentation and it was gradually increasing in size with vague pain over the swelling. she didn't give any other relevant present or past history. On examination, she had a non tender smooth diffuse swelling of size 11 × 15 cm occupying the left loin and lumbar region. It was not warm, arises from the subcutaneous plain without any cough impulse. Our clinical diagnosis was large subcutaneous mulilocculated lipoma. Her hemoglobin level was 10.2 gm/dl, total count was 7,300 cells/cumm and other blood parameters are with in normal limits. Chest X-ray was normal, plain CT scan abdomen showed an encapsulated lesion with loculation suggestive of cystic lesion with differential diagnosis of hydatid cyst and lipoma. FNAC was not done for this patient due to

the fear of anaphylactic reaction. Casoni's test was not done due to unavailability of new antigen in our location. After assessment of her physical status, she was posted for surgery under general anaesthesia with informed consent. She was kept in right lateral position and draping was done. Through transverse incision over the swelling, skin and subcutaneous tissues are incised. Grey white cystic wall identified and it is dissected all around without breaking it to avoid contamination. Once adequately dissected, wound margin is protected by covering it with 0.5% chlorhexidine-soaked gauze.

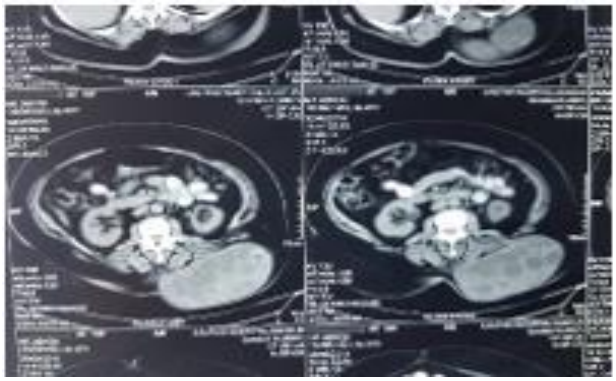

Figure 1: Plain CT abdomen showing swelling.

Figure 2: Swelling over left loin.

Figure 3: Cyst wall exposed preoperatively.

Contents of the cyst is aspirated and injected with chlorhexidine solution and re-aspirated after 10 minutes of contact with inner cyst wall and daughter cyst. Then the cyst wall was opened and all the daughter cyst, fluids are removed with large suction tube. The cyst wall was completely dissected from the surrounding tissues and send for histopathological examination along with daughter cyst. Post operatively she was advised to take albendazole 400 mg for 2 weeks. We followed her with ultrasound for 1 year and there was no recurrence and new lesion.

Figure 4: Daughter cyst removed from main cyst.

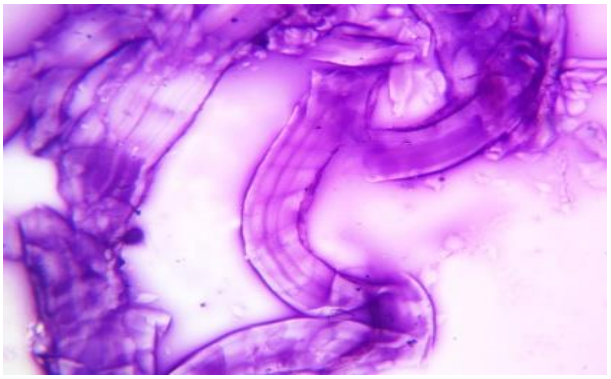

Figure 5: Cyst wall under microscope.

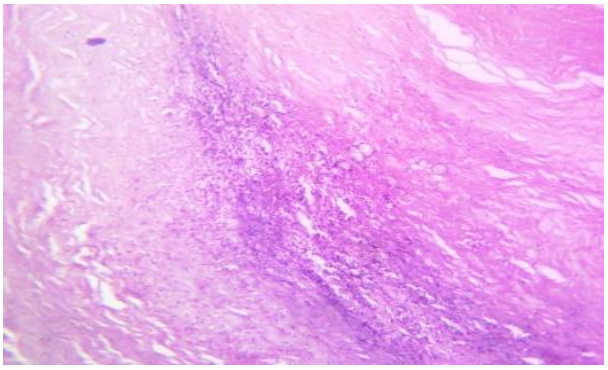

Figure 6: Pericyst with inflammation.

DISCUSSION

The hydatid cyst is a zoonosis caused by adult or larval stages of tapeworms belonging to the genus *Echinococcus granulosus*.[4] The tapeworm stage is harbored in the intestine of carnivores such as dogs, which constitute the definitive host, and the eggs are passed in the feces of the infected carnivores and ingested

by herbivores such as sheep, which comprise the intermediate host.[4] Humans are the incidental intermediate host. Larvae emerge from the eggs in the intestine penetrate it and enters the portal venules and reaches the portal vein.[6] Liver acts as the first filter and lungs act as second filter. A few embryos escape and reaches the systemic circulation.[6] Human infection with *Echinococcus granulosus* leads to the development of one or more hydatid cysts located most often in the liver and lungs, and less frequently in the bones, kidneys, spleen, muscles and central nervous system.[5] The disease is prevalent in most part of the world, though it is most expensive in sheep and cattle raising regions of Australasia part of Africa and South America.[6] It is also common in Europe, Middle East and the China.[6] It occurs in many parts of India particularly in South India.[6] The asymptomatic incubation period of the disease can last many years until hydatid cysts grow to an extent that triggers clinical signs.[5]

Table 1: The world health organization classification of hydatid cysts.

WHO stage	Characteristics	Activity
CE1	Unilocular, anechoic cyst with double line sign	Active
CE2	Multiseptated 'rosette-like' 'honeycomb pattern' cyst	Active
CE3a	Cyst with detached membrane (water-lily sign)	Transitional
CE3b	Daughter cysts in solid matrix	Transitional
CE4	Heterogeneous cyst, no daughter vesicles	Inactive
CE5	Solid matrix with calcified wall	Inactive

Even though cystic echinococcosis affects the liver and spleen commonly, it also involves brain, bone, muscles, kidney, ovary, uterus, fallopian tube, breast, peritoneum, bladder wall, omentum.[4] Ours is also a rare case of cystic echinococcosis that involved the subcutaneous plain in the loin region. We do not have the reason how the larva reaches the subcutaneous plain that too in loin and lumbar region. The diagnosis of the cystic echinococcosis was usually by ultrasound and CT scan if necessary, MRI can be useful in some cases.[7] There are no workups amongst the routine blood workups that may be used specifically for cystic echinococcosis.[7] Serologic diagnostic methods are used to support the radiological diagnosis and for follow-up assessment[7]. The indirect hemagglutination (IHA) is usually non-specific and is of value in tandem with other investigations such as enzyme-linked immunosorbent assay (ELISA) and immunoblotting.[8] Concomitant use of IHA and ELISA is associated with diagnostic sensitivity rates up to 85-96%.[7] Immunoblotting is generally used to confirm the diagnosis in cases where IHA and ELISA findings are not definitive.[9] After diagnosis, the treatment for cystic echinococcosis is based on WHO classification of hydatid cyst (Table 1).

In case CE1 and CE3a cysts are <5 cm in diameter, albendazole alone may suffice, while for cysts exceeding 5 cm in size, the puncture, aspiration, injection of a scolecidal agent, and re-aspiration (PAIR) treatment in tandem with albendazole is preferred. Types CE2 and CE3b cysts are treated by catheterization or surgery. For types CE4 and CE5 inactive cysts, monitoring is often sufficient.[10] These treatments are basically poised for liver hydatid disease. We think this can be applicable to all extrahepatic hydatid cyst including our rare case.

CONCLUSION

Occurrence of cystic echinococcosus in subcutaneous plain over the loin and lumbar region is rare. Diagnosing hydatid cyst may be confusing and difficult if arise in unusual locations and needs high level of suspicion. But CT scan may give us clue regarding preoperative decision making like avoiding FNAC and further planning. Surgery is the main stay of treatment along with frequent postoperative follow up.

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