

Case Report

Solitary subcutaneous gluteal hydatid cyst: a case report

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Abstract. A 50-year-old female presented with a swelling in the left gluteal region (upper outer region) since two years which was gradually increasing in size. Fine needle aspiration cytology of the swelling was done which was inconclusive. Incisional biopsy of the swelling done at some peripheral hospital revealed hydatid cyst. Based on histopathological examination, ultrasonography and magnetic resonance imaging features along with positive serology diagnosis of solitary gluteal subcutaneous hydatid cyst was made and total cystectomy (cystopericystectomy) was done. Post-operatively patient was put on albendazole. Larval stage of Echinococcus is the cause of hydatid cyst. Four species of Echinococcus tapeworm can produce disease in humans viz E.granulosus and E.multilocularis cause cystic and alveolar hydatid disease respectively, while E.vogeli and E.oligarthus rarely infect humans. Hydatid cyst of subcutaneous tissue is extremely rare, and this case report highlights the rarest location of the hydatid cyst.

Key words: Hydatid cyst; subcutaneous tissue

1. Introduction

Hydatid cyst has a worldwide distribution and has been recognised since ancient times. Human hydatidosis is a parasitic infection of the liver and other organs caused by the flatworm Echinococcus, most commonly E.granulosus which is a 5 mm long hermaphroditic tapeworm that has dog, foxes or coyotes as the definitive host and sheep, swine, cattle and zebra as the intermediate host. Man is an accidental, intermediate host and infection of humans represents a terminal event (dead end) for the parasite. Once within the man or other intermediate host the ingested eggs hatch in the duodenum to release the true larvae (oncospheres) that penetrate the mucosa of small intestine and enter the portal circulation. Liver acts as the first effective filter for most of the larvae and therefore being the commonest site of involvement (65-75%). If the larvae pass through the first filter, they reach the lungs which are the second most frequently involved site (10-25%).

If the larvae are not trapped in either liver or lungs, or if they by-pass the liver by travelling via lymphatics, it may lodge itself in any part of the body including the peritoneal cavity (8-18%), spleen (2-3%), kidneys (1-4%), uterus and adnexa (0.5-1%), retroperitoneum (0.5-1%), pancreas (0.5-0.8%), others (0.1-3%)(1). Different studies reveal the incidence/frequency of subcutaneous involvement in approximately 1-2% (2,3,4,5,6).

2. Case report

A 50-year-old female presented in the surgical outpatient department at S.K.I.M.S, Srinagar, with chief complaints of swelling in the upper outer area of left gluteal region for two years. Fine needle aspiration cytology of the swelling was done which was inconclusive, which was followed by incisional biopsy revealing hydatid cyst. Swelling was progressively increasing in size. There were no associated symptoms or any similar swelling in any other part of the body. On examination, a small 2 cm scar was present over the swelling. Swelling was about 7x5 cm in size, globular in shape, non-tender, firm in consistency, fixed with ill-defined margins. Haematological investigations were normal. ELISA (enzyme linked immunosorbent assay) for hydatid disease was positive. USG (ultrasonography) abdomen and chest x-ray were also normal. USG of left gluteal region (Fig. 1) revealed thick walled well defined

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cystic lesion 5.1x3.4x2.6 cm in size in the subcutaneous tissue overlying the gluteus muscle.



Fig. 1. Ultrasound of left gluteal region showing thick walled, well defined, cystic lesion with septa in the subcutaneous region overlying gluteus maximus muscle.

MRI (magnetic resonance imaging) documented the swelling in the subcutaneous tissue overlying gluteus maximus muscle, which was a thick walled multicystic lesion, hyperintense on T2WI/STIR images and hypointense on T1WI. Thick septa were seen within surrounding fat and muscle showing evidence of oedema (Fig. 2).

With the diagnosis of solitary gluteal subcutaneous hydatid cyst, the patient was subjected to total cystectomy under spinal anaesthesia. Dense pericystic adhesions were present (Fig. 3) and on cut-section small amount of clear fluid with laminated membranes and daughter cysts were seen. Principles of surgery for hydatid disease were strictly followed including using 1% citrimide to disinfect the wound. Histopathology of the specimen was consistent with the diagnosis of hydatid cyst. Post-operative period was uneventful. Patient was discharged on the third day and put on albendazole (10 mg/kg/day) for three cycles of 21 days each with a gap of one week between each cycle.

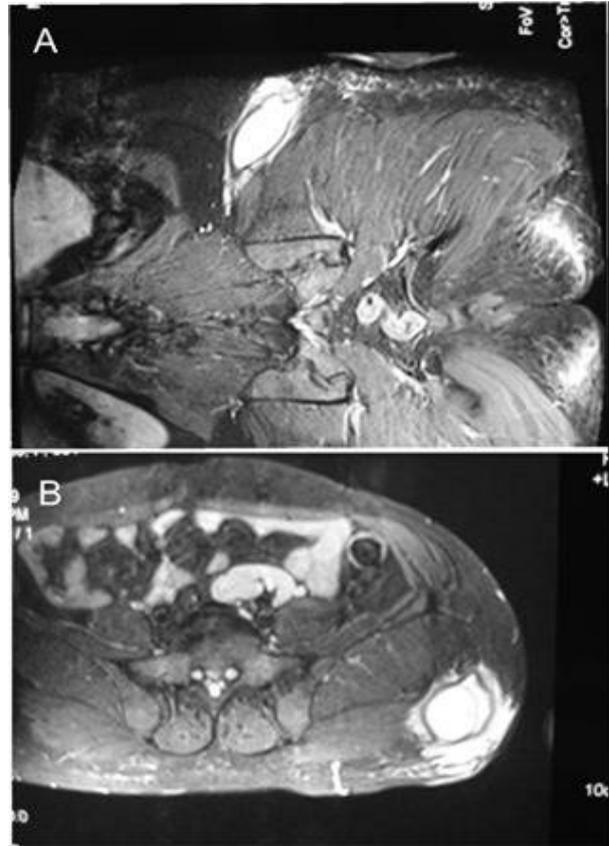


Fig. 2. Coronal (A) and axial (B) MRI showing multicystic swelling overlying gluteus maximus which is hyperintense on T2WI/STIR and hypointense on T1WI images. Thick septa seen within and surrounding tissue showing edema.

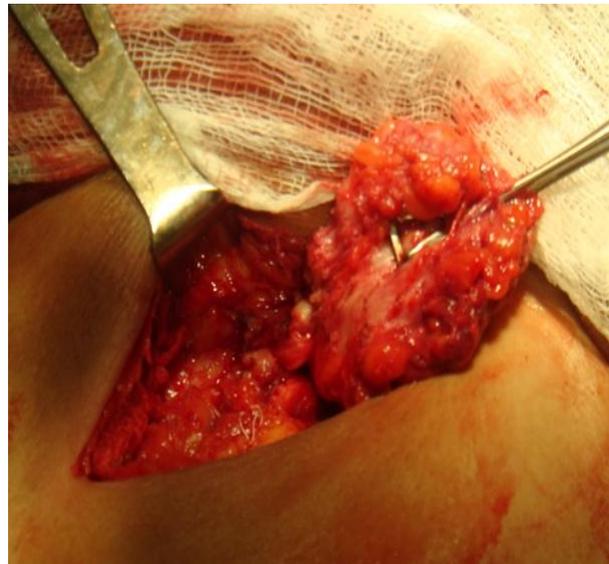


Fig. 3. Subcutaneous gluteal hydatid cyst being excised in toto.

3. Discussion

Hydatid disease is seen endemically among sheep raising communities. The disease still continues to be a serious problem in countries like Australia, New Zealand, Middle East, Africa, India, South America, Turkey and Southern Europe (7). Various soft tissue sites involved by hydatid cysts and reported in literature include those of muscles and subcutaneous tissue (neck, chest, axilla, abdomen, thigh, and palm) (8-10). To our knowledge, there has been only one case of gluteal subcutaneous hydatid cyst reported in literature until today (11).

Hydatid cyst of the subcutaneous tissue is usually associated with the disease of other structures of the body (12). The exact mechanism how the larvae pass through the liver and lungs and form solitary cyst elsewhere is not well understood. It is suggested that lymphatics are responsible for systemic dissemination of the larvae and formation of solitary cysts at uncommon sites (4). Hydatid cyst is usually asymptomatic and grows very slowly at the rate of 0.5-3 cm/year in their diameter. Symptoms and their degree depend on the site of location and size of the cyst. Our patient presented with a painless swelling in the gluteal region, which was gradually increasing in size and was associated with minimal discomfort in the sitting position.

Routine laboratory tests can only reveal eosinophilia. A number of serological tests can be done for screening, diagnosis and follow up for recurrence of hydatid disease. Highly sensitive tests include indirect haemagglutination and Latex agglutination test. Confirmation of diagnosis can be done by highly specific tests including immunoelectrophoresis, double diffusion test, and ELISA and radioallergosorbent test (1). Radiological imaging including USG, CT and MRI are excellent imaging modalities for hydatid cysts, which can delineate exact site as well as identify the daughter cysts and hydatid sand which are specific to echinococcal infestation. MRI can also show a typical distinctive feature of cyst within cyst in case of the multicystic hydatid cyst (2). We performed ELISA test on our patient who was positive (titre 1:160). USG of the gluteal region revealed a thick walled, well defined multicystic lesion measuring 51x34x26 mm in the subcutaneous tissue overlying the gluteal muscle. MRI was also

done for documentation of the rare site of hydatid disease.

Surgery remains the treatment of choice for hydatid cyst (2-5-9). Antihelminthic chemotherapy alone may be effective in 30-40% of patients. It is most effective in alveolar hydatid, less so for liver infections and essentially ineffective for the diseases of the bone, brain, eye and other sites (1). Hydatid cyst is best treated by complete excision of the cyst. We treated our patient with total cystectomy (cystopericystectomy). We used scolicidal agent for disinfecting the wound and prevent possible recurrence as there was previous history of incisional biopsy of the lesion. Patient was discharged on the third day and put on albendazole postoperatively.

Late complications which should be kept in mind are the local recurrence of the disease and development of hydatidosis at the primary sites. The patient has to be kept on regular follow up paying attention to these possibilities (9-13).

In conclusion, subcutaneous hydatid cyst is rare and should be kept in differential diagnosis of a cystic lesion especially in regions where hydatid disease is endemic. It is better not to biopsy the lesion if one suspects that it could be a hydatid cyst and should be excised totally to avoid recurrence (13).

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