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CASE STUDY

SPONTANEOUS RUPTURE OF HYDATID CYST OF THIGH-A VERY RARE PRESENTATION

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ABSTRACT

Hydatid disease is quite common in countries like India, Egypt, Australia, New zealand, Mediterranean countries, Middle East, Africa and sheep rearing countries (Kayaalp *et al*., 2007). This common disease is known to affect many uncommon sites in the human body as face, mediastinum, retroperitoneum, breast, thyroid and intermuscular planes. Other unusual sites include heart (2%), kidney (1.5%-3%), brain (3%), and spleen (3%). Incidence of musculoskeletal disease including subcutaneous tissue has been reported to be 0.5 to 5 % (Gole *et al*., 2013). Hydatid cysts at multiple unusual sites have also been reported (Abbey *et al*., 2002). These unusual sites, where suspicion of hydatid disease is unlikely, present as diagnostic dilemma. Incidence of hydatid disease of thigh has been reported to be 0.37% In a series of 272 cases of hydatid disease and thigh is the commonly involved unusual site (Gole *et al*., 2013). This could be explained due to high vascularity and less muscular activity at this site (Gracia-Diez *et al*., 2000) These unusual sites may present as diagnostic dilemma as suspicion of hydatid disease at these unusual sites is unlikely. Spontaneous rupture of hydatid cyst of these externally visible sites need no specific investigations when the diagnosis is more than evident on clinical examination as in the case being presented here.

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INTRODUCTION

Hydatid disease is quite common in countries like India, Egypt, Australia, New zealand, Mediterranean countries, Middle East, Africa and sheep rearing countries (Kayaalp C. Hydatid, 2007). This common disease is known to affect many uncommon sites in the human body as face, mediastinum, retroperitoneum, breast, thyroid and intermuscular planes. Other unusual sites include heart (2%), kidney (1.5%-3%), brain (3%), and spleen (3%). Incidence of musculoskeletal disease including subcutaneous tissue has been reported to be 0.5 to 5 % (Gole *et al*., 2013). Hydatid cysts at multiple unusual sites have also been reported (Abbey *et al*., 2002). These unusual sites, where suspicion of hydatid disease is unlikely, present as diagnostic dilemma. Incidence of hydatid disease of thigh has been reported to be 0.37%. In a series of 272 cases of hydatid disease and thigh is the commonly involved unusual site (Gole *et al*., 2013). This could be explained due to high vascularity and less muscular activity at this site (Gracia-DiezAL *et al*., 2000). These unusual sites may present as diagnostic dilemma as suspicion of hydatid disease at these unusual sites is unlikely. Spontaneous rupture of hydatid cyst of these externally visible sites need no specific investigations when the diagnosis is more than evident on clinical examination as in the case being presented here.

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Case report

A Thirty six year female presented with a thirteen years long history of a lesion of the size of 5x 4 cm on medial aspect of her left mid thigh (Fig. 1). The swelling was small in size to start with, and had been gradually increasing in size. The swelling finally ruptured on its own, four days back with discharge of whitish grape like contents (Fig.1). There was no known history of trauma, abdominal pain, cough, fever and urticaria. There was multiple (12 in number), whitish grape like hydatid daughter cysts along with portions of the cyst wall protruding from the lesion (Fig.1). No other lesion or swelling in any other part of the patient was present. On applying pressure around the lesion more hydatid cysts along with pieces of cyst membrane came out leaving behind the open empty wound (Fig. 2). Routine investigations were within the normal limits, USG abdomen and x- ray chest did not reveal hydatid disease at any other site in the patient. Specific diagnostic tests like CT scan, ELISA, IHA, were not performed in this patient. The patient was put on albendazole therapy and complete surgical excision was done. Copious use of scolicidal agent,

betadine 10%, was done. The diagnosis was confirmed by histopathological examination. The post operative period remained uneventful.

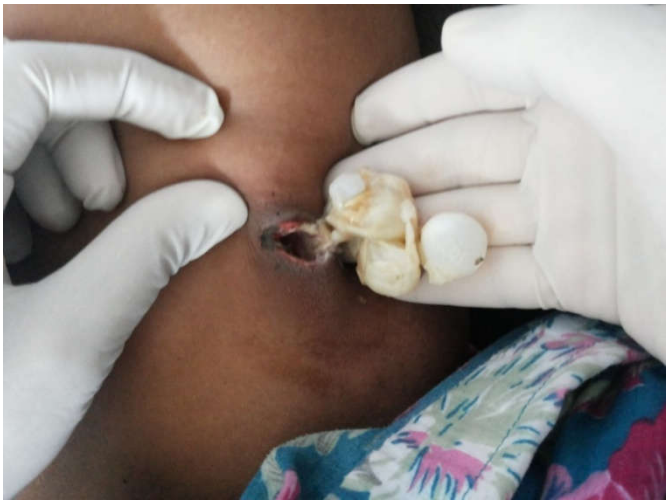

Figure 1. Spontaneous rupture of hydatid cyst of left thigh extruding hydatid cysts and portions of hydatid membrane

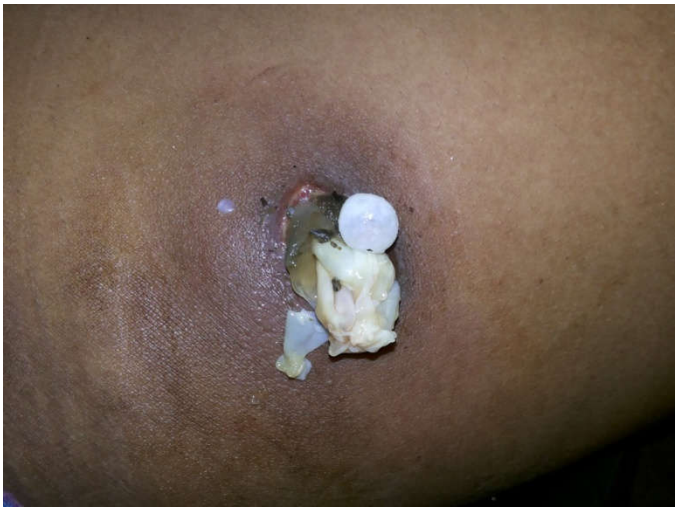

Figure 2. Open and empty thigh lesion after extrusion of the hydatid contents on applying manual pressure around the lesion

DISCUSSION

Hydatid disease is quite common in some countries and India (Gole *et al*., 2013; Abbey *et al*., 2002). The most affected organ is the liver (55%-70%) and lungs (18%-35%). Hydatid cyst has been described at many uncommon sites as heart (2%), kidney (1.5-4%), brain (3%), spleen (3%), musculoskeletal and subcutaneous planes (.5-5%). The incidence of hydatid disease of thigh, has been reported to be 0.37% in a series of 272 cases of hydatid disease (Gole *et al*., 2013). These uncommon sites may present as diagnostic dilemma as suspicion of hydatid disease at these unusual sites is unlikely. Muscular and subcutaneous hydatidosis is usually secondary in nature, resulting from the spread to normal tissue from a primary site, after spontaneous/ iatrogenic rupture or after release of viable parasites. The diagnosis becomes further complicated where primary involvement of lung and liver is not present. Sometimes subcutaneous hydatid cyst can be part of disseminated disease. Rarely, spontaneous rupture of visceral hydatid cyst into subcutaneous tissue can cause hydatidcyst (Kismet *et al*., 2006). Mechanism of subcutaneous hydatid cyst is not clear. It could also be the result of direct contamination to subcutaneous tissue through an injury (Kayaalp C. Hydatid, 2007). Implantation of ingested eggs escaping from primary filters seems to be another possibility. Under reporting of subcutaneous hydatid may be there as the lesion can be easily misdiagnosed (Kayaalp C. Hydatid, 2007). In the present case history of 13 years seems to be long but the duration of 30 years has been reported in the literature (Bansiwal *et al*., 2011). Late presentation as in this case may be mainly due to two reasons. This patient belongs to a very remote rural area with minimal medical facilities. She visited local medical practitioner many times, but the disease could not be diagnosed. Second possibility of late presentation is that the patient did not have any aesthetic reason. Subcutaneous hydatid disease could be a sign of disseminated hydatid disease in the body and clinician should be aware of this. The entity in the present case may probably be labeled as primary hydatid of the thigh as no other associated hydatid cyst was detected on clinical examintion and USG abdomen. In some of the highly endemic areas of Greece a tumour at any sight is considered as hydatid cyst until proven otherwise (Kazakos *et al*., 2005). R. Though the present case is very rare yet the diagnosis was very clear and apparent and did not require any specific investigation like USG, CT SCAN, ELISA, IHA and were therefore not done. Complete surgical excision was done and the patient put on albendazole therapy. Some times hydatid cyst can form communicating fistula with the surrounding muscle group therefore complete excision must be done otherwise recurrence is the possibility (Thekdi *et al*., 2014). Scolicidal agent, 10% povidone iodine was used locally in this case as it prevents recurrence (Thekdi *et al*., 2014). The prognosis is generally good. There was no recurrence seen in 22 cases out of 26 cases (Kayaalp C. Hydatid, 2007). The diagnosis was made merely on clinical inspection of lesion only. The presence of germinating membrane separate from daughter cysts are findings specific to hydatidcyst (Firky *et al*., 1997). The case is presented for its straightforward clinical diagnosis in spite of its being a very rare presentation.

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