

Unusual Presentations of Hydatid Cyst - A Case Study

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Abstract

Introduction: Hydatid disease is commonly found in the endemic countries within sheep rearing population of the temperate zones such as Middle East, India, Africa, South America, New Zealand, and China. Apart from liver and lung which are most often affected organs, hydatid cysts in any other location are considered uncommon.

Materials and Methods: We report 4 cases of hydatid disease presenting with cysts at unusual locations, namely, anterior abdominal wall, gluteal region, interscapular region, and right kidney. 3 of the cases, we report in the subcutaneous tissue which is very rarely seen and one in the kidney.

Results: Cystic echinococcosis remains a health issue in developing countries because of the lack of strict control programs to prevent the transmission of this infection and problems such as high populations of stray dogs, illegal butchering of animals, and poor public education about the disease. The gluteal hydatid cyst posed a diagnostic dilemma with a gluteal abscess and similarly the interscapular hydatid cyst with a tubercular abscess as both these were unusual sites.

Conclusion: Occurrence of hydatid cysts in the gluteal region, anterior abdominal wall, and interscapular region has been reported very rarely in literature. Whenever a cystic lesion is encountered in any region of the body, the possibility of hydatid cyst should be borne in mind especially in the endemic areas.

Key words: Cystic echinococcosis, Hydatid cyst, Renal hydatid, Subcutaneous hydatid, Unusual hydatid

INTRODUCTION

Hydatid cyst disease is a zoonotic infection where the causative organism is *Echinococcus granulosus*. It is commonly found in the temperate zones of the world in endemic countries India, Africa, Middle East countries, South America, New Zealand, and China where sheep rearing is commonly practiced. Lack of access to clean potable water supplies and close association of people with domestic animals like sheep and dogs makes the disease endemic in India.¹ The liver is the most frequently involved organ

(75%), followed by the lung (10%).² Primary hydatid cysts of the skeletal muscles and subcutaneous tissue are rarely encountered even in areas where echinococcal infestation is common, and some isolated case reports of such presentation have been reported.³ Apart from liver and lung which are most often affected organs, hydatid cysts in any other location are considered uncommon. In this paper, we present a series of 4 cases of hydatid cyst disease found in the anterior abdominal wall, gluteal region, interscapular region, and kidney.

MATERIALS AND METHODS

We report 4 cases of hydatid disease (Table 1) presenting with cysts at unusual locations, namely, anterior abdominal wall, gluteal region, interscapular region, and right kidney. 3 of the cases we report were in the subcutaneous tissue which is very rarely seen and one in the kidney.

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Table 1: Location, presentation, and management of four cases of hydatid cyst

S.No.	Location	Age/sex	Eosinophil (%)	Investigation	USG/CT	Management
1.	Subcutaneous hydatid cyst in anterior abdominal wall	62 year/male	8	CT-	large cystic mass with multiple daughter cysts in extraperitoneal plane	Total excision of the cyst followed by medical therapy (Albendazole)
2.	Subcutaneous hydatid cyst in left gluteal region	45 year/female	9	USG-	rounded cystic mass limited to the subcutaneous plane, superficial to gluteal muscles	Total excision of cyst followed by medical therapy (Albendazole)
3.	Subcutaneous hydatid cyst in interscapular region	36 year/male	7	Not done		Total excision of cyst followed by medical therapy (Albendazole)
4.	Renal hydatid cyst	75 year/female	6	CT and USG-	well-defined 7*6 cm cystic lesion occupying mid and lower region of right kidney	Total cystectomy leaving the pericyst <i>in situ</i> through right lumbar incision and extraperitoneal approach

USG: Ultrasonography, CT: Computed tomography

RESULTS

Case Studies

Case 1

A 62-year-old male patient presented to our hospital with a history of progressive increase in the size of a painless swelling over the abdomen which he had noticed one and a half month back (Figure 1). The patient had undergone laparotomy with a right paramedian incision 3 years back. There was no history of trauma and no history of reducibility of the swelling. Rest of the general physical examination was unremarkable. Ultrasonography revealed a cystic mass which was later confirmed by computed tomography (CT) findings which revealed a large extraperitoneal cystic mass with multiple daughter cysts in the extraperitoneal plane in the anterior abdominal wall. The cyst was herniating into the left inguinal region and reaching the left scrotal sac. On surgical exploration, there was a white cystic mass in the subcutaneous plane with macroscopic appearance of hydatid cyst (Figure 2). The sac was excised into and sent for HPR which revealed dense inflammatory infiltrate comprised predominantly of neutrophils, few lymphocytes, histiocytes in a necrotic background which was consistent with hydatid disease. The post-operative course was uneventful.

Case 2

A 45-year-old lady presented to our hospital with a swelling in the left gluteal region since 1 month. There was no history of any previous intramuscular injections. The patient had received over the counter drugs for the past 1 week; however, the swelling had not decreased in size. Her general physical examination was unremarkable. On local examination, a mildly tender solitary spherical swelling situated in the upper and outer quadrant of the left gluteal region measuring 5 cm in diameter, with a smooth surface and cystic consistency was noted. Skin over the swelling was normal. A soft tissue ultrasonography (USG) was performed which revealed a rounded cystic mass that was limited to the subcutaneous plane, superficial to gluteal muscles on the left side. The cystic mass contained fluid and showed no flow on Doppler. A provisional diagnosis



Figure 1: Subcutaneous hydatid cyst in anterior abdominal wall

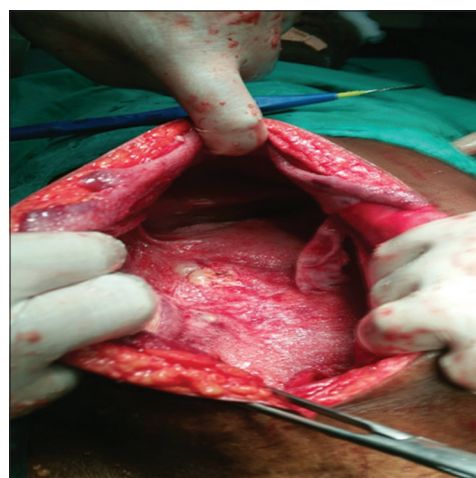


Figure 2: Intraoperative image of anterior abdominal wall hydatid cyst

of resolving gluteal abscess secondary to folliculitis or anti-bioma was made and patient posted for surgery. A cruciate incision was taken keeping in mind the possibility of a gluteal abscess (Figure 3).

However, on surgical exploration, there was a white cystic mass attached to the subcutaneous adipose tissue but free

from underlying muscles. The macroscopic appearance appeared to be that of a hydatid cyst. It contained nonpurulent thin serous fluid which was suctioned out. The cyst wall was excised into and sent for histopathology. The resultant cavity was washed with hypertonic saline and left open to heal by secondary intention. The post-operative period was uneventful, and the patient recovered well. The histopathological examination showed fibrosis, necrosis, mixed type of inflammatory cells, fibroblastic activity and vascular proliferations in adipose and connective tissues.

Case 3

A 36-year-old male patient presented to our surgical outpatient department with a slowly growing painless swelling over his back in the interscapular region for the past 4 months. Physical examination revealed a diffuse nontender swelling of size 8*6 cm. There was no history of trauma or weight loss, and the chest X-ray was normal. The patient had a history of contact with farm animals. The only significant hematological examination was raised eosinophil count. At operation, the cyst was found to be surrounded by a thick fibrous capsule and was not adherent to the underlying muscles. A total pericystectomy was done without any spillage. The cavity was closed keeping a suction drain. On microscopic examination, it showed three layers: Adventitia, laminated, and germinal layers. The germinal layers had multiple brooding capsules with many scolices in it. The post-operative course was uneventful.

Case 4

A 75-year-old female patient presented to our hospital with pain and gradually increasing lump in the right flank for 6 months. The pain was mild, dull aching, and non-radiating. A nontender lump occupying the right lumbar region was palpable measuring about 10*8 cm with a smooth surface, firm consistency and moved cephalon-caudally with respiration. Rest of the abdomen and general physical examination was normal. The USG of the abdomen revealed a well-defined 7*6 cm cystic lesion occupying mid and lower region of right kidney, with multiple cysts of varying sizes inside and hyperechoic stroma. Other organs were normal. A subsequent CT scan confirmed the possibility of a renal hydatid cyst (Figure 4). The patient was operated with a right lumbar incision and extraperitoneal approach. A total cystectomy was performed leaving the pericyst *in situ*. No bleeding or urine leak was observed from the pericystic wall. The cavity was irrigated with scolicedal agent, and the cavity was closed keeping a drain.

DISCUSSION

Cystic echinococcosis remains an important health issue in developing countries because of the lack of strict control



Figure 3: Hydatid cyst in gluteal region

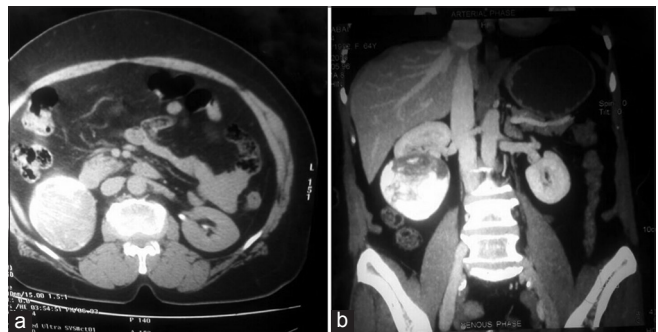


Figure 4: (a and b) Computed tomography film of renal hydatid cyst

programs to prevent the transmission of this infection plagued by other problems such as high populations of stray dogs, illegal slaughtering of animals, and poor public education, and awareness about the disease.⁴ The liver (50-70%) followed by the lung (20-30%) continue to be the commonly affected organs encountered in surgical practice. Finding hydatid cyst in a striated muscle is rare, and this has been attributed to two factors - the presence of lactic acid and contraction of the muscles. However, parasitic cysts are inclined to grow in the trunk, neck, and legs because of relatively less muscle contraction and rich blood supply to these areas. Intramuscular hydatid cysts have been reported in the muscles of the chest wall and in the pectoralis major, gluteal muscles, sartorius, and quadriceps.⁵

Typically, cystic hydatidosis consists of a single unilocular cyst. However, in as much as 30% of cases, there may be synchronous multiple cysts located in the same or multiple organs⁶ Musculoskeletal hydatid cyst is usually associated with concomitant involvement of other solid organs.⁷ The frequency of subcutaneous tissue involvement in conjunction with involvement of other solid organs has been reported to be approximately 2%.⁸ Pre-operative diagnosis of a hydatid cyst poses diagnostic challenges, and moreover the clinically slow growing nature of the

cyst resembles soft tissue tumors.⁹ Ultrasound, CT, and magnetic resonance imaging have a valuable role in pre-operative diagnosis as well as follow-up of cases of hydatid disease. The role of fine-needle aspiration cytology (FNAC) has often been controversial as there are concerns over the microscopic spillage along the needle tract. Concerns over microscopic spillage along the needle tract at the time of needle biopsy do not appear warranted, especially when patients receive subsequent medical treatment and biopsy tracts are resected at the time of definitive surgery.¹⁰ In our case series, we did not subject the patient for FNAC as the diagnosis was fairly confirmed on radiological imaging and hydatid cyst disease is common in our region.

Case 1 and 2 highlights the unique presentation of isolated subcutaneous hydatid cysts. Infected subcutaneous hydatid cysts can mimic abscess there by creating a diagnostic dilemma and may easily mislead the surgeon to perform incision and drainage with the potential to cause systemic dissemination and anaphylaxis. A very high index of suspicion is required in endemic areas to consider the possibility of hydatid cysts in a subcutaneous plane which is still quite rare.

The case 3 described in our study presented a diagnostic dilemma as the swelling in the interscapular region was suspected to be a tubercular abscess which is far more common rather than a hydatid cyst. Paraspinal and spinal hydatid cysts were first described by Chaussier in 1807. The pathogenesis of muscular invasion still remains to be understood clearly. Most authors believe that the embryo can reach the muscles via the systemic circulation after leaving the intestine and passing through two important filters: The liver and the lungs.²

Isolated primary hydatidosis of the urinary tract is a rare entity, and hence it poses several challenges incorrect pre-operative diagnosis. Ultrasound is considered to be the most accepted method for the differential diagnosis of a renal cystic tumor and highly sensitive (95%) for diagnosis. It is very safe and inexpensive. CT should be reserved for equivocal cases where a conclusion cannot be drawn only by USG.¹¹

There are various treatment options for the uncomplicated hydatid cyst and include needle aspiration under ultrasound guidance, laparoscopic approach, direct surgical intervention, or medical treatment with the use of albendazole. *En-bloc* resection without inducing rupture and spreading of the daughter cyst is a recommended treatment strategy and accepted to be curative even for intramuscular hydatid cyst.¹² In our series, all the cases were treated by surgical resection followed by medical treatment. Vaidyanathan *et al.*¹³ reported 2 cases of pelvic

Table 2: Incidence of hydatid cysts at various anatomical sites

Site	Incidence (%)
Breast	1 ²³
Thyroid	1 ²³
Heart	1 ²³
Pancreas	0.5-0.8 ^{21,22}
Spleen	2-2.5 ¹⁵
Gallbladder	0.4 ²⁰
Kidney	1-3 ¹⁸
Pelvis	2.25 ¹⁶
Soft tissue	0.5-4.7 ¹⁹
Bones	1-2.4 ¹⁹

hydatid cysts which were communicating with the bladder and they used this communication advantageously for intravesical instillation of a scolicedal agent so as to destroy the germinal layer, with a good result.

In majority of the cases in clinical practice surgery is the mainstay of treatment for hydatid cysts and offers a definite cure. The principle of surgical therapy is to excise the cyst or cysts totally whenever possible. The surgeon must be careful to remove the cyst totally avoiding spilling its contents.¹⁴

Incidence at Unusual Sites

Incidence of hydatid disease (Table 2) involving the spleen is about 2-2.5%. It can occur as a primary lesion or in association with hepatic, pulmonary, or multi-organ hydatidosis.¹⁵

Pelvic hydatid disease is uncommon, and the reported incidence is about 2.25%.¹⁶ Due to its location in a fixed cavity, it manifests with pressure effects on adjacent organs such as the urinary bladder (most common) or rectum.

Hydatid cyst of the ovary is quite unusual, and the incidence lies in the range 0.2-2.25%.¹⁷

Renal hydatid disease is also rare entity (1-3%) and located in the upper or lower pole, and these cysts are usually unilateral. 18% of renal hydatid cysts can rupture into the collecting system, leading to acute colicky pain and hydatiduria. However, primary hydatid disease of these structures is very rare.¹⁸

Primary involvement of soft tissue by hydatid cyst is unusual even in endemic areas, and the incidence is about 0.5-4.7%. The frequency of bone involvement in hydatid cyst is 1-2.4%. It is most commonly encountered in the spine and pelvis.¹⁹

According to Krasniqi *et al.*, out of 241 patients of liver hydatidosis treated surgically, only one patient (0.4%) was found to have primary hydatid cyst in gallbladder.²⁰

The incidence of hydatid disease in pancreas is reported to be 0.5-0.8%.^{21,22}

The incidence of hydatid cyst in breast, thyroid, and heart is reported to be 1% each by McManus *et al.*²³

CONCLUSION

In this paper, we described 4 cases of hydatid cyst occurring at unusual sites and their surgical management. Occurrence of hydatid cysts in the subcutaneous tissue in gluteal region, anterior abdominal wall, interscapular region, and renal hydatid cysts has been reported very rarely in literature. Whenever a cystic lesion is encountered in any region of the body, the possibility of hydatid cyst though very rare should be borne in mind especially in the endemic areas. Surgical treatment remains the mainstay wherever possible and should be supplemented by medical therapy.

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