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Annals of Clinical Pathology

Case Report

Multiple Hydatid Cyst In Gluteal Region: A Case Report

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Submitted: 07 December 2015

Accepted: 19 January 2016

Published: 20 January 2016

ISSN: 2373-9282

Keywords

- Hydatid cyst
- Echinococcus granulosus
- Soft tissue mass
- Double layered wall

Abstract

Introduction: Human hydatid disease is a parasitic infestation caused by Echinococcus granulosus . The disease is prevalent in most parts of the world, especially in sheep farming and cattle farming areas . Primary subcutaneous hydatid disease is a rare pathology even in endemic area of Echinococcosis. Preoperative diagnosis is essential to prevent intraoperative dissemination and rupture of the cyst.

Case presentation: We present a 54-year-old caucasian female referred with left gluteal mass. Ultrasound examination showed multiple cystic lesion in the subcutaneous tissue localized on gluteus maximus Magnetic resonance images revealed cystic mass approximately 64x58x51 mm in adipose tissue at lateral gluteal region and 4 further cysts that the major one approximately 22x15x8 mm at posterolateral region of the major cyst. Cyst had a double layered wall appearence. Patient was treated with surgical excision followed by medical therapy. Small cysts treated by long term albendasole therapy. Clinical, radiological and serologic tests showed no recurrence after treatment.

Conclusion: We suggest direct spreading of E. granulosus to adjecent sites in our case. Surgical excision and postoperative albendasole therapy used for treatment of hydatid cyst. Subcutaneous hydatid cyst should be remembered in differental diagnosis of slow growing soft tissue mass especially in endemic regions.

INTRODUCTION

Human hydatid disease is a parasitic infestation caused by Echinococcus granulosus [1]. Humans are infected by consuming food and water that are contaminated with the eggs of the Echinococcus granulosus [2]. The disease is prevalent in most parts of the world, especially in sheep farming and cattle farming areas of Asia, North and East Africa, South America, Australasia and the Middle East [2,3]. In humans, the infestation is usually located in the liver (65%) or lungs (25%), and rarely involves the brain, heart, bone, or other organs [4,5]. Musculoskeletal and soft tissue cysts account for 0.7–3% of total cases of hydatidosis [1]. These cysts appear as slow-growing masses of soft tissue, sometimes with inflammatory signs and fistulization [6-8].

In this report, we described a rare case of subcutaneous hydatic disease with multiple cysts formation in gluteal region. Preoperative diagnosis of hydatid disease is essential because rupture and dissemination of the cyst may result in recurrence, and intraoperative spillage of the antigenic cyst fluid may lead to severe anaphylactic response [1,3,7]. It is critical to suspicious clinical diagnosis in cystic swelling of muscle in rural endemic areas [6].

CASE PRESENTATION

A 54 years old caucasian female presented to the orthopaedic surgery outpatient clinic with a mass in left gluteal region. She had a 3 months history of slowly growing mass and pain on the mass region. She had not trauma history. There was not history of contact with animals. Physical examination revealed a localized, tender, cystic swelling of approximately 5x5 Cm fixed to tissues in left gluteal region. There was an extensive region of tenderness around the cystic mass. There were not ecchymosis or warmth on the gluteal region. Laboratory tests showed a showed a total leukocyte count (WBC) of 8500/mm^3, erythrocyte sedimentation rate (ESR)of 22 mm/h (Westergen) and C-reactive protein (CRP) of 2.

Ultrasound examination was performed and it showed multiple cystic lesion in the subcutaneous tissue localized on gluteus maximus. Magnetic resonance (MR) images revealed cystic mass approximately 64x58x51 mm in adipose tissue at lateral gluteal region (Figure 1) and 4 further cysts that the major one approximately 22x15x8 mm at posterolateral region of the major cyst (Figure 2). Double layered wall of the cyst was easily seen in MRI (Figure 1, Figure 2)

Indirect hemagglutination test was performed for suggestive diagnosis of Echinoccus granulosus and it was positive . Thorax and abdomen computed tomography (CT) was performed to exclude another localisations of hydatid cyst. There were not any localisation except left gluteal region.

The mass was operated under spinal anesthesia. Cystic mass was determined in subcutaneous tissue behind gluteus maximus muscle. The cyst had a 7x5x4 cm diameter in size. This cystic mass in gluteal region was removed with surrounding fibrous thick capsule and fasia. Cyst area was irrigated with hypertonic 20% saline and povidone iodine after removal of mass to reduce risk of recurrence. We decide not to remove multiple cysts that were seen on MRI in medial site of huge mass. They were very small to extripted without spillage. Incision was closed primarily after inserting suction drain and patient was discharged after removal of drain on day two. Histological examination of the specimen revealed a mass of 7x5x4 cm diameter cyst. It was contained daughter cysts and fragments of lamellar membrane of the hydatid cyst. No bacterial pathogen was cultivated in cyst fluid.

Albendazole therapy, 200 mg twice daily, was given for six months after operation. Long term albendazole therapy was selected to inactivate other small hydatid cysts. MRI and serological tests were performed to determine recurrance. Clinical, radiological and serologic tests show no recurrence after sixth month of surgical treatment.

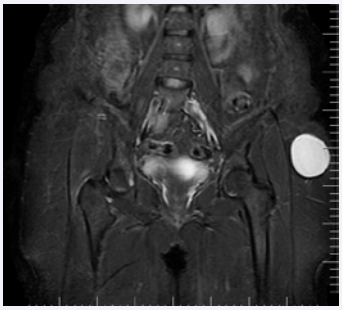

Figure 1 Coronal view of MRI with T2 weighted image demonstrate double-layered cystic mass (black arrrow) approximately 64x58x51 mm in subcutaneous adipose tissue of left gluteal region.

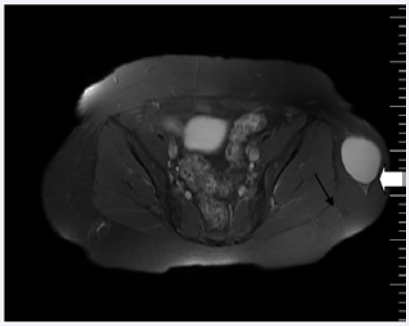

Figure 2 Axial view of MRI with T2 weighted image demonstrates double-layered cystic mass (white arrow) and small cysts (black arrow) in posterolateral gluteal region.

DISCUSSION

Musculoskeletal hydatid cyst is usually associated with involvement of other solid organs [3]. The frequency of subcutaneous tissue involvement associated with involvement of other solid organs, has been reported to be approximately 2% [9]. A solitary primary subcutaneous localization is an extremely rare entity, even in countries where the echinococcus infestation is endemic and real incidence is not known [1,3,10]. We present an extremely rare case of multiple hydatid cysts in gluteal region.

The mechanism of primary subcutaneous localization of hydatid cyst is still not clear. Bagga et al. [11] suggested three mechanisms for this localisation; ingested parasite larva enter to the circulation by penetration of intestinal wall, dissemination through lymphatic channels, direct spread from adjacent sites. We present a case of multiple adjacent localizations. We suggest direct spreading of E. granulosus to adjecent sites in our case. But primary cyst formation in this localisation is still in doubt.

Preoperative diagnosis of subcutaneous hydatid cyst is difficult. Clinically slow growing nature of this cyst resembles soft tissue tumors [1,3,7,12]. Ultrasonography (US), computed tomography (CT), and MR imaging have a valuable role in the radiologic diagnosis and follow-up of hydatid disease [13,14]. We presented US and MRI before diagnosis and for follow-up after surgical excision. Fine needle aspiration cytology (FNAC) was recommended by several authors for preoperative diagnosis [1,11,12]. We did not prefer this method because of potential spoilage. We could easily diagnosed hydatid disease in our case because were familiar with this disease [7]. E. granulosus infestations in not very rare in our area [12].

Surgery is the most effective way to treat hydatid cysts [1,5,9,15]. Complete surgical resection and medical therapy is the preferred treatment for isolated subcutaneous echinococcosis [1,15]. Rupture or spoilage of cysts should be avoided to prevent local or distant dissemination and immediate anaphylaxis [9,15]. Irrigation should be made in operation with hypertonic saline or povidone iodine in an attempt to kill scoleces [1,15]. We performed wide resection with capsule combined with hypertonic saline and povidone iodine irrigation. Small cysts were prefered to follow-up with chemotherapy. Mebendazol and albendazol are used for hydatid disease, but albendazol has better intestinal absorption and higher concentration within cystic material, making it a more effective treatment [16,17]. Albendazole preferred in 400 mgr daily dose for this case in curative dose.

CONCLUSIONS

Hydatid cyst of multiple form in gluteal region is very rare disease. Differential diagnosis should be made by using MRI and serologic tests. FNAC have a potential riscs of spoilage. We suggest direct spreading of E. granulosus to adjecent sites in our case. Atypical locations of hydatid cyst should be kept in mind especially for slow growing mass in endemic regions.

AUTHORS CONTRIBUTIONS

Özgür Çiçekli participated in data collection and drafted the manuscript. Felat Öncel participated in data collection. Alauddin Kochai participated in the design of the study.

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Cite this article

Çiçekli Ö, Öncel F, Kochai A (2016) Multıple Hydatıd Cyst In Gluteal Regıon: A Case Report. Ann Clin Pathol 4(1): 1060.