Author's response to reviews

Title: A solitary primary subcutaneous hydatid cyst: a case report

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Author's response to reviews: see over
JMCR Editorial Team

Object: Manuscript ID 6125006183597470 - A solitary primary subcutaneous hydatid cyst: a case report. Dr Abdelmalek Ousadden et al.

Thank you for consideration of our manuscript for publication in your journal. We have reviewed the above manuscript according to your reviewer’s comments.

Reviewer # 1 : Abuzer Dirican

Comments to authors:
1. Explanation of Figure 2 can be changed as “The image of totally excised hydatid cyst”.
   - Done
2. In referance section “;” should be used instead of “,” after year of publication.
   - in JMCR reference style “,” comes after year of publication.
Quality of written English: Needs some language corrections before being published
   - Done

Reviewer # 2 : Levent CANKORKMAZ

Comments to authors:
Accept after minor revisions and needs some language corrections
Quality of written English: Needs some language corrections before being published.
   - Done

Additional material submitted by the reviewer # 2

Introduction
Solitary primary hydatid cyst in the subcutaneous abdominal wall is an exceptional entity even in countries where echinococcus infestation is endemic.

Case presentation
We report a new case involving in a 70-year-old woman presenting a subcutaneous mass in the paraumbilical area with non-specific clinical presentation. The diagnosis of subcutaneous hydatid cyst was suspected by radiological findings. A complete surgical resection of the mass was performed with uneventful postoperative recovery. The histopathology confirmed the suspected diagnosis.

Conclusion
Hydatid cyst should be considered in the differential diagnosis of every subcutaneous cystic mass, especially in regions where the disease is endemic. The
best treatment is the total excision of the cyst with an intact wall.

Introduction

Hydatid disease is a parasitic infestation caused by the *Echinococcus granulosis* which life cycle is well described [1]. Endemic areas are still countries of the temperate zones, where the common intermediate hosts, sheep goats and cattle, are raised (such as North Africa, Middle East, Central Europe, Australia and South America) [1, 2]. The liver is the most frequently involved organ (75%), followed by the lung (15%) [2, 3]. The solitary primary subcutaneous localisation is extremely rare, and its incidence is unknown [2]. That makes this case located in the abdomen anterior wall, without any other involvement, interesting to be reported.

Case presentation

A 70-year-old moroccan Caucasian woman presented with a subcutaneous cystic mass in the right para-umbilical abdominal wall, evolving for 6 months. On physical examination, a 6 cm diameter abdominal parietal mass was palpated 5 cm right of the umbilicus. It was cystic, fluctuant, mobile, and painless. The overlying skin was normal. Abdominal ultrasound showed a rounded cystic mass well limited in the right para-umbilical abdominal wall, measuring 60 mm. No other abdominal cystic mass has been found (which were diagnosed by examination). The preoperative examinations (chest radiograph, complete blood count, urine analysis, and blood biochemistry) revealed no abnormality. Hydatic serology was negative.

Upon surgical exploration, the mass was attached to the subcutaneous adipous tissue but not associated to any muscular or cutaneous structure (Fig 1). The macroscopic appearance suggested a hydatid cyst (Fig 2). Perforation was avoided by means of meticulous dissection. Histopathologic examination of the specimen revealed a hydatid cyst. The patient has been followed for 2 years and no recurrence of hydatidosis has been detected.

Discussion

The mechanism of primary subcutaneous localisation is unclear [2, 4]. The ingested parasite’s ova penetrates the intestinal wall, joins the portal system and reach the liver where most of them are caught in the hepatic sinusoids [2]. A few ova may pass through the liver filter to reach the lung (second filter), than the systemic circulation to cause hydatid disease in other organs [1, 2]. A possible dissemination through lymphatic channels has also been reported and accounts for cases with solitary cysts in uncommon sites [3-5]. Direct spread from adjacent sites may be another mechanism of infection [6].

In our case, the hydatid cyst was located subcutaneously, the patient had not undergone previous surgery for hydatid cysts, and no hydatid cysts were found in other organs. Therefore, our patient was diagnosed as having primary subcutaneous hydatid cyst.

In a large series from Greece, the frequency of extrahepatic and extrapulmonary hydatidosis was 9% [5]. In different series, the frequency of subcutaneous tissue involvement, which is usually associated with involvement of other solid organs, was approximative 2% [1, 7, 8]. But, primary isolated hydatid cysts located in the abdominal wall remains extremely rare, even in geographic areas in which echinococcal infestation is frequent [3, 4].

The clinical course is non-specific and depends on the site of involvement, the size, and pressure caused by the enlarged cysts [1]. Usually, it present as an inert painless noninflammatory mass, without deterioration of general condition [4, 9]. If superinfected or cracked, the cyst can simulate an abscess or a cancer [8, 9]. The radiological imaging (Ultrasoundography, Computed Tomography scan, MR Imaging) is useful in diagnosis, showing the size, localisation, relationship to adjacent organs and type of the cyst. It can also search for another hydatid
The radiological findings of a thick cyst wall, calcification, daughter cysts, and a germinative membrane separate from the cyst wall are findings specific to hydatid cysts [1-4]. Enhancement of the peri-cystic soft tissues can be considered as a suggestive MR Imaging feature of soft tissue hydatid disease [9]. Serology is a useful tool to confirm the diagnosis, although it is rarely positive for extrahepatic and extrapulmonary locations (25%) [1, 4, 8]. It is furthermore associated with false-negative and false-positive results [4].

The best treatment option is complete surgical excision of the intact cyst, to avoid leakage of cyst content that can cause anaphlaxis and local recurrence [1, 2, 8]. If ideal surgery (surgery) is impossible, the cyst content (fluid, membrane and daughter cysts) has to be removed intraoperatively, and the cyst pouch has to be irrigated with scolecidal—(scolicidal) solutions [1, 2]. Other options include percutaneous treatment under ultrasound guidance, with needle aspiration and irrigation of scolecidal—scolicidal solutions, and medical treatment with the use of Albendazole (albendazole) [2, 8].

Conclusion
Hydatid cyst should be considered in the differential diagnosis of every subcutaneous cystic mass, especially in regions where the disease is endemic. The best treatment is the total excision of the cyst with an intact wall.

Changes have been made as indicated by the reviewer.

(has patient history bites or pinprick??)

• no

Reviewer # 3 : Bulent Unal

Comments to authors:
This presentation is extremely rare for hydatid disease. I think the manuscript is suitable for publish in the JMCR. But it needs some language corrections.

• Done

Here is the new manuscript with all the changes done:

Abstract

Introduction

Solitary—The solitary primary hydatid cyst in the subcutaneous abdominal wall is an exceptional entity even in countries where the echinococcus infestation is endemic.

Case presentation

We report a new case involving—in of a 70-year-old woman presenting a subcutaneous mass in the paraumbilical area with non-specific clinical
presentation. The diagnosis of subcutaneous hydatid cyst was suspected by radiological findings. A complete surgical resection of the mass was performed with uneventful postoperative recovery. The histopathology confirmed the suspected diagnosis.

Conclusion

Hydatid cyst should be considered in the differential diagnosis of every subcutaneous cystic mass, especially in regions where the disease is endemic. The best treatment is the total excision of the cyst with an intact wall.

Introduction

Hydatid disease is a parasitic infestation that is caused by the Echinococcus granulosis which life cycle is well described [1]. Endemic areas are still countries of the temperate zones, where the common intermediate hosts, sheep, goats and cattle, are raised; such as in North Africa, Middle East, Central Europe, Australia and South America [1, 2]. Thus the liver is the most frequently involved organ (75%), followed by the lung (15%) [2, 3]. The solitary primary subcutaneous localisation is extremely rare and its incidence is unknown [2]. That makes this case located in the abdomen anterior wall, without any other involvement, interesting to be reported. In our case, the hydatid cyst is located in the abdomen anterior wall, without any other involvement. That makes this case interesting to be reported.

Case presentation

A 70-year-old Moroccan Caucasian woman presented with a subcutaneous cystic mass in the right para-umbilical abdominal wall, which has been evolving for 6 months. On physical examination, a 6 cm diameter abdominal parietal mass was palpated 5 cm on the right of the umbilicus. It was cystic, fluctuant, mobile, and painless. The overlying skin was normal. The abdominal ultrasound showed a rounded cystic mass well limited in the right para-umbilical abdominal wall, measuring 60 mm. No other abdominal cystic mass has been found. The preoperative examinations (chest radiograph, complete blood count, urine analysis, and blood biochemistry) revealed no abnormality. The hydatic serology was negative. Upon surgical exploration, the mass was attached to the subcutaneous adipose tissue but was not associated to any muscular or cutaneous structure (Fig 1). The macroscopic appearance suggested a hydatid cyst (Fig 2). Perforation was avoided by means of meticulous dissection. The histopathologic examination of the specimen revealed a hydatid cyst. The patient has been followed for 2 years and no recurrence of hydatidosis has been detected.

Discussion

The mechanism of the primary subcutaneous localisation is unclear [2, 4]. The ingested parasite’s ova penetrates the intestinal wall, joins the portal system and reaches the liver where most of them are caught in the hepatic sinusoids [2].
A few ova may pass through the liver filter (first filter) reaching the lung (second filter) and the systemic circulation, to cause causing hydatid disease in other organs [1, 2]. A possible dissemination through lymphatic channels has also been reported and accounts for cases with solitary cysts in uncommon sites [3-5]. It accounts for cases with adjacent sites may be another mechanism of infection [6].

In our case, the hydatid cyst was located subcutaneously. The patient had not undergone previous surgery for any hydatid cysts, and no hydatid cysts which were never found in other organs. Therefore, our patient was diagnosed as having a primary subcutaneous hydatid cyst.

In large series from Greece, the frequency of extrahepatic and extrapulmonary hydatidosis was 9% [5]. However, in different series, the frequency of subcutaneous tissue involvement, which is usually associated with involvement of other solid organs, was approximately 2% [1, 7, 8]. But, the primary isolated hydatid cysts located in the abdominal wall remains extremely rare, even in geographic areas in which the echinococcal infestation is frequent [3, 4].

The clinical course is non-specific and depends on the site of involvement, the size, and the pressure caused by the enlarged cysts [1]. Usually, it presents as an inert painless non-inflammatory mass, without any deterioration of the general condition [4, 9]. However, if superinfected or cracked, the cyst can simulate an abscess or a cancer [8, 9].

The radiological imaging (Ultrasonography, Computed Tomography scan, MR Imaging) is useful in diagnosis, showing the size, localisation, relationship to adjacent organs and type of the cyst. It can also search for another hydatic location [1, 4]. The radiological findings of a thick cyst wall, calcification, daughter cysts, and a germinative membrane separated from the cyst wall, are all findings that are specific to hydatid cysts [1-4]. Enhancement of the peri-cystic soft tissues can be considered as a suggestive MR Imaging feature of soft tissue hydatid disease [9]. Serology is a useful tool to that confirms the diagnosis although it is rarely positive for extrahepatic and extrapulmonary locations (25%) [1, 4, 8]. It is furthermore associated with false-negative and false-positive results [4].

The best treatment option is a complete surgical excision of the intact cyst, to avoid which avoids leakage of cyst content that can cause anaphylaxis and local recurrence [1, 2, 8]. If the ideal surgery is impossible, the cyst content (fluid, membrane and daughter cysts) has to be removed intraoperatively and the cyst pouch has to be irrigated with scolicidal solutions [1, 2]. Other options include percutaneous treatment under ultrasound guidance, with needle aspiration-irrigation of scolicidal solutions, and medical treatment with the use of Albendazole [2, 8].

**Conclusion**

The hydatid cyst should be considered in the differential diagnosis of every subcutaneous cystic mass, especially in regions where the disease is endemic. The best treatment after all is the total excision of the cyst with an intact wall.
References


Figure legends

Figure 1: Per-operative view of the subcutaneous hydatid cyst.

Figure 2: Hydatid cyst after its surgical debulking. Image of the totally excised hydatid cyst.