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Abstract

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Reference


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Management of a ruptured hydatid cyst involving the ribs: Dealing with a challenging case and review of the literature

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ABSTRACT

INTRODUCTION: Hydatid liver cysts can rupture into neighboring structures in 15–60% of patients, and most often involves the bile duct, the bronchi, and the peritoneal/pleural cavities. Rarely, chest or abdominal wall involvement occurs that are challenging to manage. This case report and literature review describes the management of patients with chest wall and rib invasion.

PRESENTATION OF CASE: A 74-year-old woman, of Spanish origin, presented with right upper quadrant abdominal pain and tender localized swelling. On computer tomography (CT) assessment, the rupture of a hydatid cyst into the right anterior chest wall was identified. Partial involvement of the 10th and 11th rib were noted. The diagnosis was confirmed by a serological test. Surgical treatment involved a radical en bloc right hepatic resection together with resection of the involved ribs, diaphragm and subcutaneous tissue. Primary diaphragm and wall closures were performed. The postoperative course was uneventful with three weeks of albendazole treatment. CT follow-up at six months demonstrated the absence of recurrence.

DISCUSSION: Complete resection is the gold standard treatment of patients with hydatid cysts with the aim to remove all parasitic and pericystic tissues.

CONCLUSION: The present report illustrates that an aggressive surgical en bloc resection is feasible and should be preferred for the treatment of hydatid cysts with rupture into the chest wall, even when the ribs are involved.

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1. Introduction

Echinococcal disease is most often caused by the dog tapeworm Echinococcus granulosus and occasionally by the fox tapeworm Echinococcus multilocularis. The other two species, E. vogelli and E. oligarthrus cause polycystic echinococcosis and are much less frequently associated with human infection. The hydatid disease is caused by E. granulosus and mainly occurs in sheep grazing areas, including South and Central America, the Middle East, China and countries of the former Soviet Union with prevalence rates of 2–6 percent. The disease predominantly affects the liver (60–70%) and lungs (30%), and the surgical management, despite its difficulties, is considered as the cornerstone of treatment.

Beside anaphylactic reactions, the most frequent complication of the hydatid liver disease is the rupture into neighboring structures (15–60% of patients with hydatid cysts), often affecting the bronchus, gastrointestinal tract, and peritoneal/pleural cavities. We report and discuss the possible management strategies of a patient with a rarely observed rupture into the thoracic wall, involving the ribs and subcutaneous tissue.

2. Presentation of case

A 75-year-old female patient, only known for high blood pressure, was admitted to our department with a one-month history of right upper quadrant abdominal pain, associated to a 48-h episode of fever up to 39 °C (without jaundice, chill or weight loss). Leucocytes, C-reactive protein and liver function tests were within normal range. Physical examination revealed a tender swelling in the right upper quadrant. A computer tomography (CT) revealed a partially calcified right hepatic cyst (8.3 cm × 7 cm) with images of rupture into the lower chest wall (Fig. 1A and B). The rupture had created a 5 cm fluid collection, without evidence of dissemination into the peritoneum or into pleural cavity. The diagnosis of hydatid cyst was confirmed by a positive E. granulosus serological ELISA test (512, normal superior value: up to 128). After a four-week prophylactic treatment with albendazole 400 mg bid, a resection was performed.

The lesion was approached through a right sub-costal incision. There were many adhesions between the hydatid liver cyst, the
gallbladder, the diaphragm and the abdominal wall, preventing any communication between the peritoneal cavity and the lesion. Due to adhesions, the diaphragm was removed locally and primarily sutured (without pleural drainage). Part of ribs 10 and 11 were removed en bloc with the lesion and the involved subcutaneous tissue (Fig. 2A and B), and the resection was completed by a right hepatic lobectomy. The five centimeter abdominal wall defect was closed primarily. In order to treat any potential contamination, the abdomen was washed locally with hypertonic saline solution (NaCl 20%). The diagnosis of hydatid cyst was confirmed histopathologically.

The postoperative course was uneventful with three additional weeks of albendazole treatment. A follow-up CT did not show any evidence of recurrence at six months.

3. Management of previously published hydatid liver cyst rupture involving the ribs

In an effort to identify the possible management strategies of hydatid liver cysts with rupture into the chest wall and involving the ribs, an electronic search of the relevant literature was performed using the Medline Library. The search looked for all published articles from 1950 until November 2011 in any language, and was performed with the use of the following keywords: “hydatid*”; “liver” and “rib”.

The search only identified one previously published report of a patient with hydatid liver cyst (liver dome) involving the rib and inducing a left pleural empyema treated by rib resection with pleural drainage, and an uneventful recovery.16

A number of reported patients demonstrated primary rib hydatidosis, which were most often treated by aggressive surgical resection.17-20 Authors have reported that bone hydatid involvement of vertebrae, long bones, pelvis and skull usually result from vascular or lymphatic migration of viable parasites from other locations.14 A systematic review from Tunisia, published in 2009, showed that the gold standard for an osseous cystic echinococcosis remains a wide surgical excision which can lead to excellent outcomes.13

4. Discussion

The present report illustrates that an aggressive en bloc surgical resection is feasible and should be the preferred treatment for hydatid cysts with rupture into the chest or abdominal wall, even when the ribs are involved.

Fig. 1. Contrast-enhanced CT scan of the abdomen showing a large hydatid cyst involving the right lobe of the liver and ruptured into the subcutaneous tissue of the right anterior wall (A). Another fluid collection was present in the subcutaneous tissue (B).

Fig. 2. (A, B): In toto excision of the right liver lobe including the hydatid cyst involving the ribs and the subcutaneous tissue [1 square = 1 cm].
Table 1

<table>
<thead>
<tr>
<th>Type</th>
<th>Incidence</th>
</tr>
</thead>
<tbody>
<tr>
<td>Non complicated</td>
<td>79</td>
</tr>
<tr>
<td>Rupture in the biliary tracts</td>
<td>13</td>
</tr>
<tr>
<td>Rupture in the peritoneal cavity</td>
<td>1</td>
</tr>
<tr>
<td>Rupture in the pleural space and bronchi</td>
<td>5</td>
</tr>
<tr>
<td>Rupture in the diaphragm</td>
<td>2</td>
</tr>
<tr>
<td>Rupture in the abdominal wall</td>
<td>1</td>
</tr>
<tr>
<td>Total</td>
<td>101</td>
</tr>
</tbody>
</table>

Echinococcal disease is caused by the *E. granulosus* larva. Its initial phase is mostly asymptomatic and latent periods of several decades can be observed.\(^{11,12}\) This explains the late presentation of symptoms in our patient, approximately 60 years after she left her native endemic region in Spain. Of note, this type of presentation is similar to the one of most patients treated in Switzerland, where the prevalence of hydatid liver disease (estimated mean prevalence: 0.2%)\(^{13}\) is proportional of the number of migrants from endemic areas (mainly from Mediterranean countries, North Africa and South America). In Geneva, where a large proportion of migrants live, the incidence tends to be higher with 101 hydatid liver cysts resected over 20 years in our unit, with only one subcutaneous rupture (our current case) (Table 1).\(^7\)

The rupture into the chest wall with involvement of the ribs is a rare complication and our observation represents, to our knowledge, the second reported case of such a spontaneous rupture over the last 60 years. The rupture of a cyst results from its growth in the direction of the least resistance. The continuous friction of its thick, partially calcified and inflamed walls induces pressure erosions and adhesions to adjacent structures, and, as a result, spillage of the cyst’s content. Fortunately, the adhesions prevent the dissemination of the scolices into the peritoneal cavity and a possible anaphylactic shock.

Complete resection is the main curative treatment for all hydatid cysts with the aim to remove all parasitic and pericystic tissues. Along this line, the surgical management of a cyst ruptured into the chest/abdominal wall should include an in toto resection of all the involved structures, including the ribs (as needed in our case). Such a strategy should be selected regardless of the size of the abdominal wall involvement and leads to good outcomes as shown in the previously published report.\(^16\) This said, when the cyst is close to major liver vessels (which have to be preserved for the normal hepatic function), an incomplete resection (subtotal pericystectomy) or a percutaneous puncture-aspiration-injection-reaspiration (PAIR) procedure can be considered. Whatever the extent of the surgery, any intra-operative cyst rupture should be avoided and any potential dissemination should be prevented with the use of drapes soaked with hypertonic sodium chloride.\(^8,10\)

The length of the post-operative anti-parasitic treatment is linked to the presence of a complication, the intra-operative findings, the type of the operation (complete resection or not) and the presence of viable *E. granulosus* in the cyst.\(^9,13\) A treatment by albendazole of three to six months is usually recommended when viable cysts have been removed. Until recently, albendazole was administered intermittently in four week courses, followed by an interval of one drug-free week because of safety concerns. However, current evidence suggests that continuous therapy does not increase the risk of side effects, and efficacy may be improved.\(^15\) In our case, only a 3-week post-operative albendazole treatment was given, due to the curative nature of the resection.

5. Conclusion
Spontaneous chest wall rupture of a hydatid cyst is a rare condition. Although the management must be individualized for each patient, a surgical resection is the best curative procedure, even if the ribs are involved.

Conflict of interest statement
The authors declare no financial or any other conflict of interest.

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Ethical approval
Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contributions
All authors contributed in data collection, writing and design of the study.

References

