An unusual presentation of secondary pleural hydatidosis


ABSTRACT: Although pleural involvement is relatively common in cystic hydatid disease, one of the rarest and least known complications is secondary pleural hydatidosis.

We report the case of a patient who presented with polycystic secondary pleural hydatidosis 4 yrs after treatment for a pyopneumothorax caused by rupture of a pulmonary cyst near the pleural space. Due to the coexistence of severe chronic obstructive pulmonary disease, surgery was ruled out.

The patient was treated with albendazole, with a favourable outcome.


Cystic hydatid disease (CHD) is an infection produced by larvae of the parasite platyhelminth Echinococcus granulosus [1]. It has a wide geographic distribution and humans may be infected incidentally as intermediate hosts in the parasite’s life cycle. The organs most commonly affected are the liver and the lungs [1, 2]. Pleural involvement is rare, and usually follows the rupture of a pulmonary or hepatic cyst inside the pleural space [3]. Since in about 90% of episodes the cyst is no longer fertile after rupture, secondary pleural hydatidosis (SPH) is a rare event occurring in less than 10% of such cases [3]. Surgery is the treatment of choice for CHD, but antihelmintic therapy has been advocated in patients with recurrent hydatidosis or in those in whom surgical intervention involves a high risk of morbidity or mortality [4]. This report describes an unusual case of a SPH treated successfully with albendazole.

Case report

A 66 yr old male resident of Tarragona (Catalonia, Spain) was admitted to hospital with left pleuritic pain and increasing dyspnoea of 2 months duration. He was a tobacco smoker (45 pack-years). His past history included a surgical excision of a hepatic hydatid cyst at the age of 40 yrs, and the diagnosis of severe chronic obstructive pulmonary disease (COPD) (forced expiratory volume in one second (FEV1) 1.05 L, 35% of predicted value) with bullous emphysema at 50 yrs of age. He was receiving treatment with bronchodilator drugs.

Four years prior to the current admission, he had had left pyopneumothesis (fig. 1a), which was diagnosed as a complication of the rupture of an infected emphysematous bulla and was successfully treated with closed thoracic drainage and antimicrobial drugs. Physical...
examination was normal except for bilateral hypopho-
ness on chest auscultation. Chest radiographs were con-
sistent with bullous emphysema and there were diverse
nodular images in the left lower hemithorax (fig. 1b).
Main biochemical and electrocardiographic (ECG) 
data were normal. Arterial blood gas values, on room
air, revealed mild hypoxaemia (arterial oxygen tension
\( P_{\text{a},\text{O}_2} \)) 10 kPa (76 mmHg). Blood eosinophil
count was 514 eosinophils·mm\(^{-3}\) (table 1). Total immunoglobulin
E (IgE) was 2,850 U·mL\(^{-1}\) with a positive radioaller-
gosorbent test (RAST) specific IgE for \( E. \) granulosus
(2.10 Phadebas RAST units (PRU)·mL\(^{-1}\)) (table 1).

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Determinations of antinuclear antibodies, anti-deoxyri-
bonucleic acid (DNA), rheumatoid factor, carcinoembr-
yonic antigen (CEA) and alpha\(_1\)-antitrypsin were either
normal or negative. The tuberculin skin test with two
tuberculin units (TU) purified protein derivative (PPD),
RT-23, was negative. Examination of three samples of
spum showed no acid-fast bacilli, fungi or atypical
cells. Hydatidosis was diagnosed and, as surgery was contra-
indicated, treatment was initiated with albendazole, 10
mg·kg\(^{-1}\) daily, in cycles of 28 days followed by 14 days
with no treatment. Clinical status as well as analytical
data of the patient improved progressively, as reflected
in table 1. After the sixth cycle, thoracic CT scan sho-
ved a marked reduction in the number and size of the
cysts, which subsequently became completely opacified
(fig. 2b).

**Table 1.** Total IgE, specific RAST IgE (Echinococcus
granulosus) and eosinophilia on admission and after
therapy

| IgE: immunoglobulin E; RAST: radioallergosorbent test; PRU: Phadebas RAST units. |

<table>
<thead>
<tr>
<th></th>
<th>On admission</th>
<th>After therapy</th>
<th>After therapy</th>
</tr>
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<tbody>
<tr>
<td>Eosinophils n·mm(^{-3})</td>
<td>514</td>
<td>215</td>
<td>80</td>
</tr>
<tr>
<td>Total IgE U·mL(^{-1})</td>
<td>2,850</td>
<td>911</td>
<td>825</td>
</tr>
<tr>
<td>RAST IgE PRU·mL(^{-1})</td>
<td>2.10</td>
<td>1.31</td>
<td>1.07</td>
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**Fig. 2.** a) Chest computed tomography (CT) scan indicates mul-
tiple cysts located in the left lower hemithorax and implanted in both
diaphragmatic and mediastinic pleura. b) Chest CT scan (after ther-
apy with albendazole) shows a reduction in the number and size of the
cysts, which became completely opacified.

**Discussion**

Human echinococcosis is caused by three species of
Echinococcus: \( E. \) granulosus, \( E. \) Multilocularis and \( E.
\) vogeli [1, 2]. \( E. \) granulosus produces CHD, which is
the commonest form in humans, representing some 90%
of cases [5]. CHD is found worldwide. Spain is an
endemic zone, and its annual incidence is estimated at
5.21–7.55 cases per 100,000 inhabitants [6]. CHD of
the lung is asymptomatic in 30% of patients [4], and,
when symptoms occur, they are usually due to the
compression of the underlying pulmonary tissue by the
cyst and/or to the presence of complications, such as
rupture or infection [2, 5]. A hydatid cyst may be con-
fused with a bulla if it cracks and becomes empty [7].
This may have been the case in our patient when he
presented pyopneumothorax 4 yrs before developing
SPH.

Although, in rare instances, SPH has been associat-
ed with a haematogenous dissemination of the larvae,
usually it is caused by a rupture of a neighbouring cyst
with dissemination of the contents of the cyst (multiple
daughter cysts and scolices) along the pleura. Less than
10% of these patients develop SPH [5]. This low per-
centage has been attributed to bacterial superinfection
of the pleural space [3]. Three different forms of SPH
have been described: pleural granulomatosis, hydatido-
thorax, and pleural hydatid graft [3]. The case reported
here is a peculiar example of this last form. In endemic
zones, hydatid cysts are among the major causes of
well-defined nodular pulmonary lesions [5].

Medical therapy is useful when surgery is techni-
cally difficult or contraindicated [1–5]. Albendazole is the
drug of choice, and the usual dose is 10–15 mg·kg\(^{-1}\)
daily for 4 weeks in repeated cycles (usually more
than three), separated by 2 weeks without treatment [1,
8].

This case is an example of secondary pleural hydati-
dosis which emphasizes that when pleural effusion and/
or pyopneumothorax are detected in a patient with previous hepatic cystic hydatid disease, the possibility of secondary pleural dissemination should be considered.

References