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Bilateral lung and liver hydatid cysts. Case report

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SUMMARY: **Bilateral lung and liver hydatid cysts. Case report.**

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Introduction. *Synchronous occurrence of pulmonary and hepatic hydatid cysts is an uncommon manifestation of hydatid disease that is observed in less than 10% of cases. We report a rare case of bilateral lung (with bronchial fistula) and liver cyst, surgically treated after medical therapy.*

Case report. *A 44-year-old housewife reporting fever, anorexia and fatigue that had been present for the previous 20 days received diagnosis of bilateral lung and liver hydatid cyst. Because of the dimensions of right lung cyst and the successive bronchial fistolization, we proceeded to three-stage operation of two thoracotomies and a laparotomy to control the risk of further rupture. After surgery, all post-operatives were uneventful. Complete resolution of the therapy with no evidence of recurrence at 2 years follow-up.*

Conclusion. *We emphasize the need to search for additional hydatids in patients who present with either pulmonary or liver hydatids. The simultaneous treatment of liver and lung should be reserved to patients in good conditions; in all other cases, especially when one cyst is more symptomatic than the others or has more risk of rupture, we prefer to treat single cyst.*

RIASSUNTO: **Idatidosi epatica e polmonare bilaterale.** ***Case report.***

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Introduzione. *L'idatidosi epatica e polmonare sincrona è una rara manifestazione della infestazione da echinococco, osservata in meno del 10% dei casi che giungono in ospedale. Riportiamo un raro caso di localizzazione polmonare bilaterale (con fistola bronchiale) ed epatica, trattata chirurgicamente dopo terapia medica.*

Caso clinico. *Una casalinga di 44 anni si presenta alla nostra osservazione con febbre, anoressia e facile stancabilità da circa 20 giorni. Con l'ausilio dell'imaging toraco-addominale, è stata posta diagnosi di idatidosi epatica e polmonare bilaterale. A causa delle dimensioni della cisti polmonare destra e per la successiva fistolizzazione nel bronco, siamo stati costretti ad eseguire la procedura in 3 tempi, con due toracotomie e una successiva laparotomia sottocostale destra, per controllare l'ulteriore rischio di rottura. Dopo ogni intervento chirurgico, il decorso post-operatorio è stato regolare. Il follow-up clinico e strumentale a 2 anni non ha mostrato alcun segno di recidiva.*

Conclusioni. *Enfatizziamo la necessità di ricercare ulteriori localizzazioni, anche rare, in soggetti con idatidosi epatica e polmonare sincrona. Il trattamento simultaneo delle localizzazioni multiple va riservato a pazienti in buone condizioni cliniche generali; in tutti gli altri casi, e specialmente quando una cisti è maggiormente sintomatica rispetto alle altre (rischio di rottura), è preferibile trattare prima questa cisti.*

KEY WORDS: Hydatid - Cist - Lung - Liver - Bilaterality - Synchronous occurrence - Surgery.
Idatidosi - Polmone - Fegato - Bilateralità - Sincronicità - Chirurgia.

Introduction

Hydatid disease is an antropozoonosis caused by tapeworm *Echinococcus granulosis* and is prevalent in sheep-and cattle-raising areas in the Mediterranean region, South America, Australia, and New Zealand.

The liver is the most commonly affected organ (> 65%) followed by the lung (> 25%) (1).

We report a case with concomitant hepatic and bilateral pulmonary hydatid disease in a 44-years old girl.

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Case report

A 44-year-old caucasian housewife of rural province of Agrigento (Sicily) was admitted to our University Hospital reporting fever, anorexia and fatigue that had been present for the previous 20 days. Her vital signs were as follows: body temperature, 36.8°C; blood pressure, 130/70 mm Hg; heart rate, 85 beats/min; respiratory rate, 14 breaths/min. The oxygen saturation was 96% while she was breathing ambient air. Pulmonary examination revealed bilateral decreased breath sounds and dullness to percussion over the right and the left costophrenic angle. The rest of the physical examination were normal. Laboratory investigations included a complete blood count, liver function tests and urine analysis; all results were within normal ranges except for eosinophils 2.83 10^3/µl (NV < 0.8) and fibrinogen 533 mg/dl (NV 150-450).

On chest radiography (Figs. 1A,B) circular cystic-like lesions were visualized in the anterior-inferior segment of inferior right (9 x 6.8 cm) and left lobe (8.3 x 6.6 cm). CT abdominal scan examination revealed the presence of a hypodense cystic lesion of 11 × 8.2 cm in the right lobe of the liver (S5-S7-S8) with detachment of the inner membrane and a mark on porta vein (Fig. 1C) and confirmed bilateral cystic fluid-filled mass in lung (Fig. 1D). Serologic test results for echinococcus by means of an enzyme-linked immunosorbent assay and an indirect haemagglutination test were negative. To sterilize the hepatic cyst, chemotherapy with albendazole (400 mg twice daily) was started for 8 weeks.

Surgical excision of the pulmonary cyst was performed through a right anterolateral minithoracotomy (Fig. 2A). After identification of the cyst, the viscerolisis was performed (Fig. 2B), the most superficial part of the cyst was opened (cystotomy) under positive pression ventilation by the anesthetist, and the laminated membrane was full removed with ring forceps (Fig. 2C). The remaining pulmonary cavity was irrigated with saline hypertonic solution and cleaned with sterile gauze sponges. Then, the residual cavity was obliterated by absorbable pursestring sutures of polyglactine starting from the deepest level to the surface (Fig. 2D). Figure 2E shows the specimen. The postoperative period was uneventful. The chest drain was removed on the fourth postoperative day and the woman was discharged on the sixth day after surgery with oral therapy with albendazole (400 mg twice daily).

One month after, the patients had a ruptured cyst in the bronchus (Fig. 3A) and presented with cough and expectoration, so the patient received surgical excision of left pulmonary cyst via anterolateral mini-thoracotomy; the cysts was identified and the pericardium and pleura protected with wet sponges soaked in hypertonic saline solution; cruciate incision was made over the cyst (Fig. 2B), which was attempted to enucleate under positive-pressure ventilation by the anesthetist using Barrett's technique (Fig. 2C). The bronchial communications were sutured and the cyst cavity (Fig. 2D) closed with multiple purse-string sutures. The postoperative period was uneventful and chest drains was removed on the 3rd postoperative day. The patient was discharged on the 7th day after surgery with oral therapy with albendazole (400 mg twice daily) to prevent recurrence.

One month after, the hepatic cyst was removed through a subcostal incision and near-total pericistectomy with opened cyst (Figs. 4A,B,C,D). The subdiaphragmatic drainage was removed in 3rd post-operative day and the patient was discharged on 6th post-operative day; postoperative course was uneventful. All cysts were subjected to histopathologic examination, which confirmed the diagnosis.

Adjunctive chemotherapy with oral albendazole (400 mg twice daily) was administrated for two months to prevent recurrence. During the 24-month follow-up with four-month intervals of ultrasonography and upright chest radiography, there was no evidence of recurrence.

Discussion

Bilateral lung hydatid cysts with liver hydatid cysts are an uncommon manifestation of hydatidosis but many reports do not mention such a coincidence. Tomalino de-

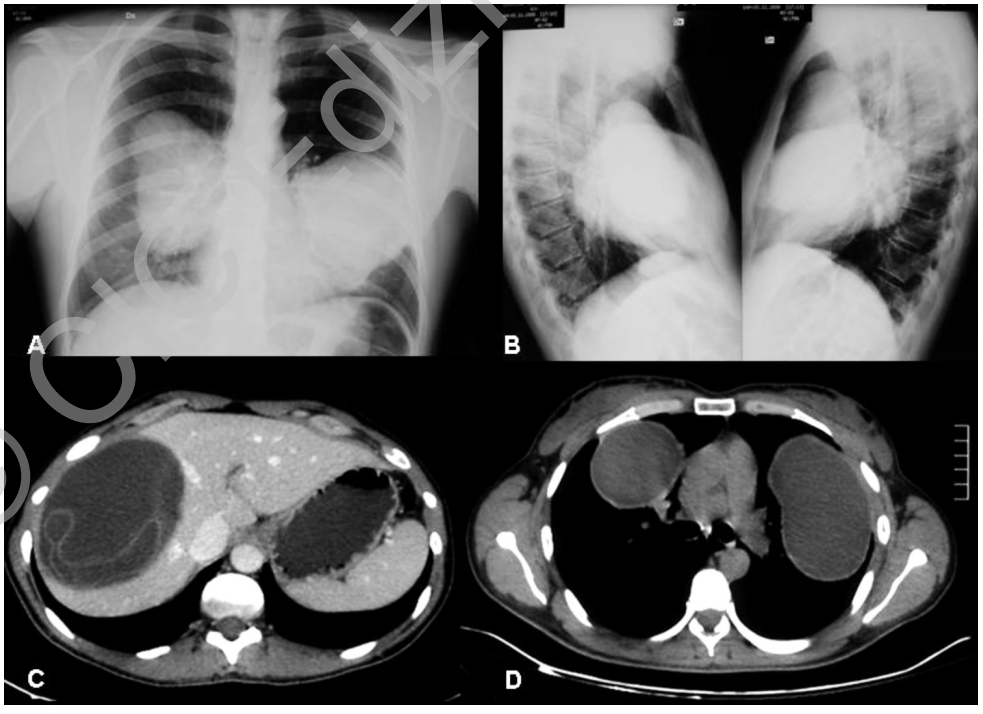

Fig. 1. - Chest radiography (Figs. 1A,B) and CT (Figs. 1C,D) scan that confirmed bilateral cystic fluid-filled mass in lungs.

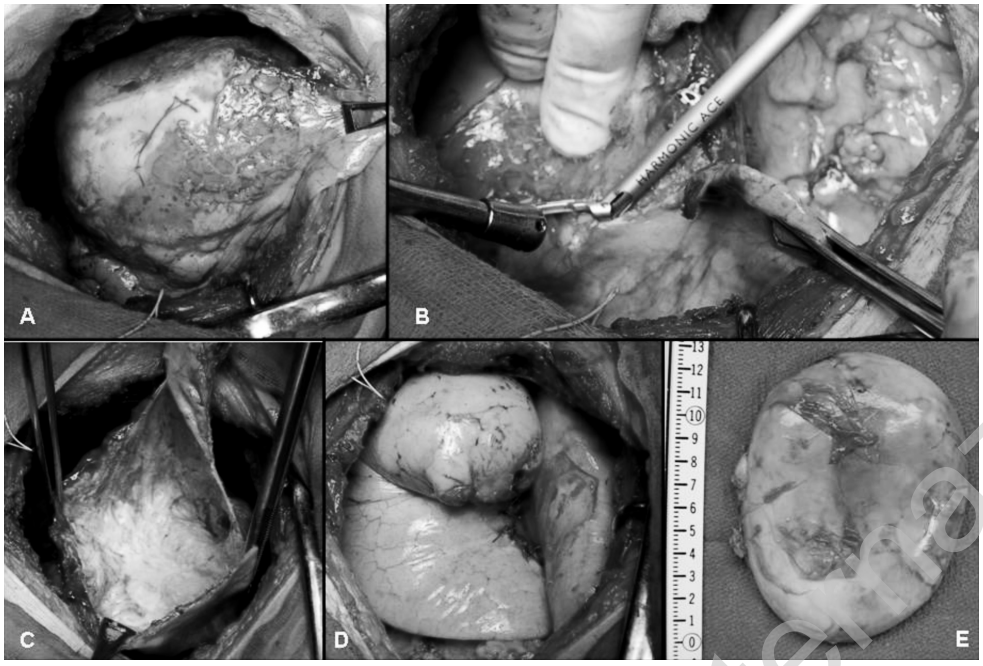

Fig. 2. - Right anterolateral minithoracotomy (Fig. 2A); viscerolisis (Fig. 2B), removing of laminated membrane (Fig. 2C); obliteration of residual cavity (Fig. 2D); specimen (Fig. 2E).

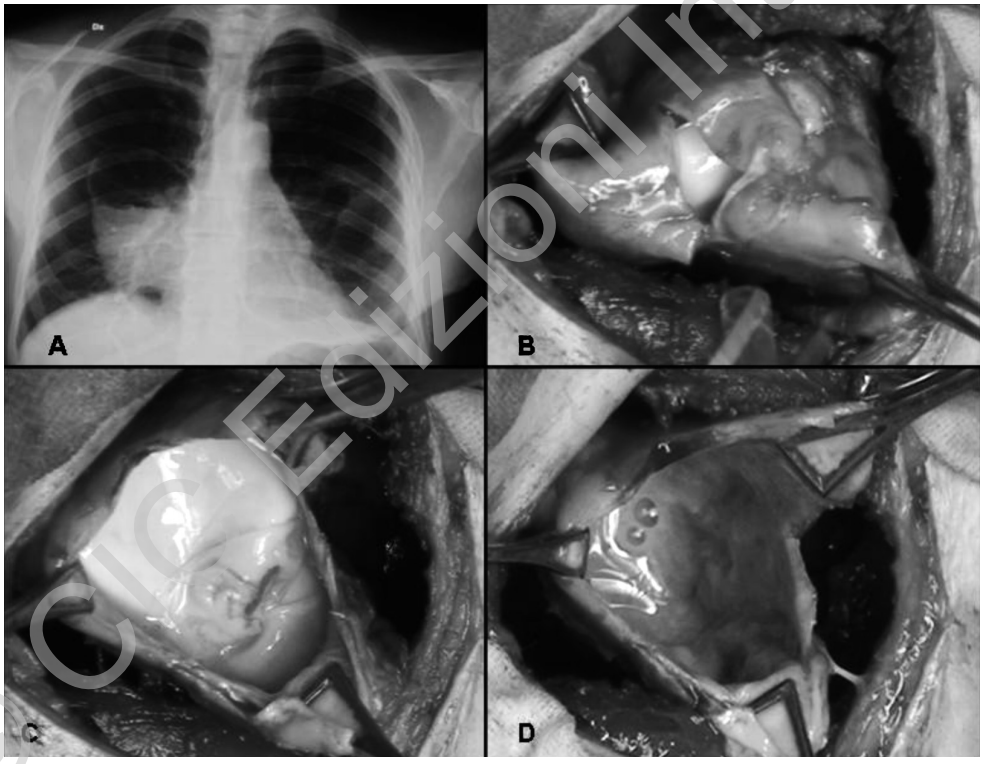

Fig. 3. - Ruptured cyst in the bronchus (Fig. 3A); cruciate incision over the cyst (Figs. 3B,C); the bronchial communications is closed (Fig. 3D).

scribed this manifestation of hydatidosis in 1961. Crauzaz and Saidi discuss the intrathoracic evolution of liver hydatids and their approach through right thoracotomy but the simultaneous problem of liver hydatid cysts was not dealt with. Peleg and coworkers reported 10% of patients with pulmonary hydatid cysts on the right side had their liver hydatid cysts removed in the same operation. Burgos and associates also removed hepatic cysts transdiaphragmatically in 7 of 331 patients with pulmonary hydatids (2). Synchronous occurrence of pulmonary and

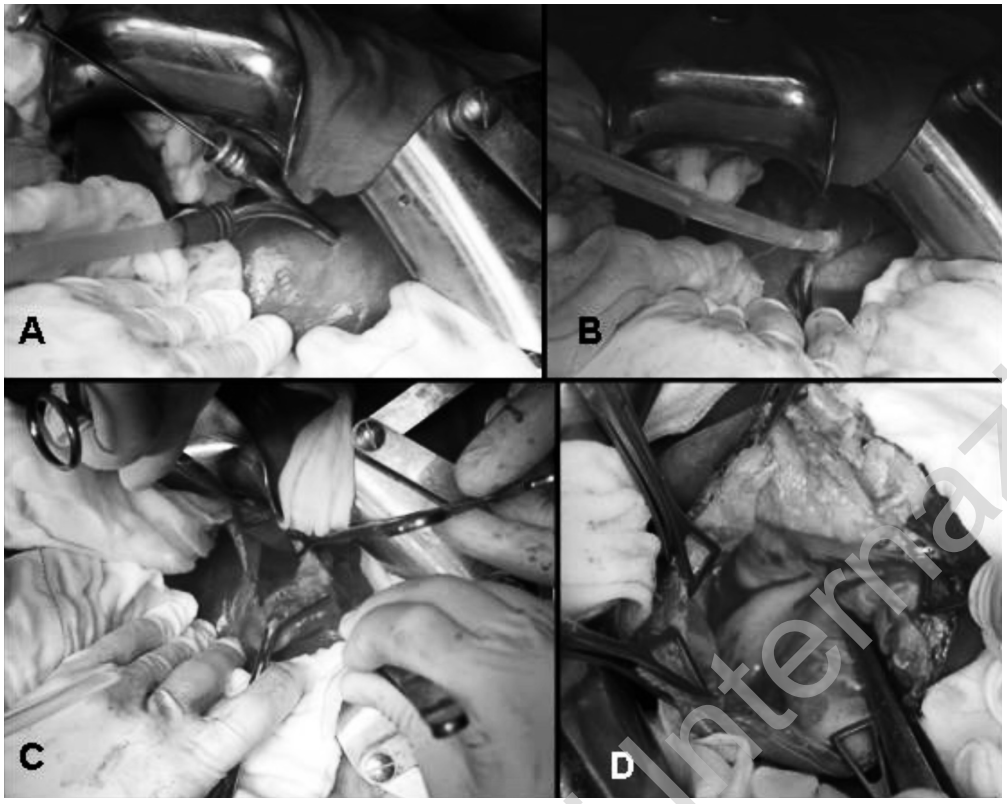

Fig. 4. - Near-total pericistectomy with opened cyst (Figs. 4A,B,C,D).

hepatic hydatid cysts is an uncommon manifestation of hydatid disease that is observed in less than 10% of cases (2).

Plain chest films can usually establish the diagnosis; round homogenous opacities in the lung parenchyma are characteristic of simple uncomplicated cysts (unruptured cysts present as radiodense shadows on chest roentgenograms), whereas radiologic signs caused by the entrance of air into the cyst or floating hydatid membranes in the remaining liquid after vomique are pathognomonic for ruptured cysts (3). The image of pneumopericyst and waterlily sign are characteristic features of complicated cysts. Hepatic cysts present as round shadows which may be calcified. Rarely secondary infections with gas-producing organisms may produce daughter cysts. With intrabiliary rupture, gas is noted in the remaining cavity. Ultrasound scan helps in substantiating the diagnosis. Computer tomographic scan furnishes useful information regarding both pulmonary and hydatid cysts in liver and correlates well with the operative findings.

The cysts may remain asymptomatic for a long time. As they enlarge, patients complain of cough, expectoration of membranes, hemoptysis, and thoracic pain in cases of pulmonary cysts. Patients with liver hydatids may present with abdominal pain, and a palpable mass in the right hypochondrium and epigastrium. Occasionally patients may have sputum stained with bile in the case of liver cysts rupturing into the lungs, or jaundice hydatidenteria or hydatidemesis if they rupture into the bile ducts. The surgical goals are total eradication of parasite, prevention of cyst rupturing at operative field, and care of the residual cavity (3).

In case of multiple localization, hepatic cyst is usually treated first, the lung cyst few weeks later. Some locations (right hepatic lobe cysts and cysts of the right lower lung lobe) favourite a single intervention by frenothoracotomy. Above all, surgery should start from the symptomatic location, or from the more massive, or one in which any lesion can be more dangerous or life-threatening (4).

Otherwise, combined resection for bilateral pulmonary and hepatic hydatid cysts is superior to three-stage approach as it decreases morbidity, mortality and the stay in the hospital (5). Cetin and colleagues reported removal of bilateral hydatid cysts of the lungs through midsternotomy. However, simultaneous removal of bilateral lung and hepatic hydatid cysts has only been referred as a single case reported by Jacob and coworkers. One-stage procedure through a thoracoabdominal incision or a thoracic or transpleural approach is preferable if the patient is a good surgical risk (6). Most authors advocate conservation of lung parenchyma, reserving resections for ruptured cysts that have caused destruction or infection of the adjacent tissue (7). Various surgical

procedures have been described in the literature, namely, excision of entire cyst by enucleation (Barrett's technique), wedge resection, segmentectomy, lobectomy, and needle aspiration of the cyst in situ. Enucleation of lung hydatid cyst was first described by C.V. Armand Ugon in Uruguay in 1947 under the name of "hydatic delivery". Barrett and Thomas described a similar technique in 1952 (enucleation of the cyst followed by obliteration of the residual cavity with purse-string sutures) (7). Cysts larger than 10 cm in diameter can be better managed by needle aspiration followed by enucleation to prevent tracheobronchial flooding with hydatid fluid. Before opening the cyst, the lung and especially the cyst-containing lobe should be freed from any adhesions to the chest wall. Spillage of the cyst contents into the thoracic cavity after needle aspiration is common (3).

In 1948, Perez-Fontana introduced cystopericystectomy for management of lung hydatid cysts followed by obliteration of the remaining cavity to achieve complete eradication of the parasite; the cyst is fully extracted and subtotal removal of the adventitia follows. However, this procedure carries the risk of hemorrhage and air leak during dissection of the pericystic space (3).

The hydatid cyst in the liver can be excised by using the natural plane of cleavage that exists between the germinating layer and adventia. Primary closure of the residual cavity with drainage was accomplished by us without any complications (2).

Chemotherapy alone is not reliable in controlling this disease: even if the parasite in the lung and liver dies, the membranes retained are the source of recurrent infections or bacterial superinfection, with also the high risk of damage or partial rupture of the germinal layer of the lung hydatid cysts between 3rd and 14th days of the treatment (8). We routinely prescribe mebendazole starting 4 weeks before surgery and continuing postoperatively for about 6 to 8 weeks (20-40 mg/kg each day), monitoring red blood cells count and hepatic function.

Conclusions

This case report highlights the necessity of maintaining a high level of vigilance because hydatid disease is still an existent public health problem of worldwide significance that may remain asymptomatic and undiagnosed for a long period.

We are of the view that surgical treatment of the lung cyst should be preferred firstly in cases of lung hydatid cyst disease with the involvement of multiple organs because rupture of cyst should be considered as inevitable during the medical therapy and the patient should be hospitalized.

The choice of which cyst (liver or lung or combined) must be assessed on the basis of symptoms, spillage or rupture risk and performance status of the patient.

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Written consent for publication data and images was obtained from the patient during the compilation of informed consent for surgery.

Declaration of no conflict of interest

The Authors declare that they have no competing interests

Authors contribution

GG and CLN are the major contributors in writing the manuscript; MG, CS, GG, CLN and AS performed surgical interventions; GG, FLV, FC and EC performed photos and graphical integration of histological specimen and examination; GG, CLN and FC performed literature review and final revision of the manuscript. All Authors read and approved the final manuscript.

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