

RESEARCH ARTICLE

BILIO-PLEURO-BRONCHIAL FISTULA COMPLICATING A HYDATID CYST OF THE LIVER: ABOUT A RARE CASE

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Abstract

Introduction: The hydatid cyst is a parasitic disease still endemic in Morocco as well as in several countries, it affects with predilection the liver, biliopleurobronchial fistula is a rare (2.5 to 16%) and threatening complication of hepatic hydatidosis with a poor prognosis.

Observation: We report the case of a 40-year-old female from a rural area, suffering for 2 months from pain in the right hypochondrium, with notion of biliptysia and who was presented to the emergency room for respiratory distress, clinical examination finds a right fluid effusion syndrome. The posteroanterior chest roentgenogram showed a homogeneous opacity occupying the totality of right thoracic field. The thoracic CT showed a right pleural effusion of high abundance associated with a hydatid cyst of the liver, the pleural puncture brought back a frankly purulent liquid, the patient thus benefited from a thoracic drainage in emergency with resulting from an immediately purulent then saffron-like liquid whose chemical study objectified the presence of bilirubin, because of the presence of bile in pleural fluid and the notion of biliptysia a bronchial fibroscopy was made and showed the presence of bile in the bronchial suction fluid. The diagnosis of a biliopleurobronchial fistula was retained, and the patient benefited an endoscopic sphincterotomy with a good evolution then a pleural decortication and closing of the fistula.

Conclusion: The incidence of bilio-pleurobronchial fistula complicating a hydatid cyst of the liver is rare which can be life-threatening, we report a new observation and we insist through this work on a rapid initial management of the hydatid cyst of the liver, in order to avoid progression to serious complications such as biliopleurobronchial fistula as well as rapid and adequate management of this medical, endoscopic and surgical fistula.

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Introduction:-

Bilio-pleurobronchial fistula results from abnormal communication between the bile ducts, the bronchi and / or the pleura. It is a rare complication often of hydatid origin in our context [1]. Hydatid cyst is a parasitic infection still endemic in Morocco as well as in several countries

The passage of hydatic cysts of the liver is done through a breach located at the diaphragmatic barrier, this complication is rare: 2.5 to 16% of cases, especially very serious, due to the polymorphism of the lesions, knowing that the bile is caustic at the bronchial level.

Case Report:

Mrs . R. E .a 40 years old women , never treated for tuberculosis and without any recent tuberculosis contagion, without notable pathological history, living in a rural area and havingcontact with dogs, reported since 2 months a pain in the right hypochondrium, complicated 1 week before hospitalization by right thoracic pain and stage IV of mMRC dyspnea , a productive coughbringing back mucous expectoration as well as biliptysia, evolving in a context of apyrexia and deterioration of the general state.

The clinical examination found a patient who was conscious, pale, polypneic 35 cycles per minute, with signs of respiratory struggle, tachycardia at 100 beats per minute, 85% ambient oxygen saturation with theright fluid effusion syndrome and pain on palpation of the right hypochondrium.

The posteroanterior chest roentgenogram showed a homogeneous opacity occupying the totality of right thoracic field (figure 1)

The thoracic CT showed a right pleural effusion of high abundance and total pulmonary atelectasia (figure 2) with a hydatid cyst of the liver (figure 3)

the pleural puncture brought back a frankly purulent liquid, the patient thus benefited from a thoracic drainage in emergency, with resulting from an immediately purulent liquid then saffron yellow whose chemical study objectified the presence of bilirubin, and scolex.

Fibroscopy was made and showed a stenosis of the apical sub-segment of the right upper lobar and confirmed the presence of bile in the bronchial suction fluid.

Hydatid serology was positive at 43 DU on the ELISA test, and> 1/2560 on the indirect hemagglutination test.

During her hospitalization, the patient presented biliptysies of medium abundance, and the thoracic drain brought back daily saffron yellow liquid corresponding to the bile.

The diagnosis of a biliopleuro bronchial fistula complicating a hydatid cyst of the liver was retained.

The patient benefited from thoracic drainage, antibiotic treatment, as well as respiratory physiotherapy with good clinical and radiological evolution (figure 4) and benefited from an endoscopic sphincterotomy before thoracotomy with decortication and closure of the fistula.

Discussion:-

A biliobronchial fistula (FBB) results from an abnormal communication between the bile ducts and the bronchi, to which can be added a prior rupture in the pleura thus constituting a bilio-pleurobronchial fistula (FBPB) [3], The frequency of FBB and FBPB is variable according to the series: from 2.5% [4] to 16% [5] of hydatid cysts of the liver.

Intimate contact of the hepatic dome with the diaphragm promotes the crossing of the diaphragmatic barrier by hydatid cysts of the liver, sometimes causing rupture of these cysts in the pleura or in a pulmonary lobe or in the bronchi.

Thus, the intrathoracic evolution of hydatid cyst of the live depends on several factors [5,6]: intimate contact of the hepatic dome with the diaphragmatic dome; the thoraco-abdominal pressure gradient which tends to draw the contents of the hydatid cyst from the liver towards the thoracic cavity; erosion of the diaphragm by ischemianecrosis caused by possible superinfection of the cyst and inflammatory phenomena and corrosion of all tissues in contact with bile [7].

It is a serious complication of hydatid cysts of the liver due to the multiplicity of lesions which concern

simultaneously the abdominal and thoracic stage passing through the diaphragm [6,8]. Ruptured hepatic hydatid cysts in the thorax are rare and occur more frequently in the bronchi than in the pleural cavity [9]. The pleural rupture is made either in a symphysical pleura with the constitution of an encysted pleural pocket communicating with the hydatid cyst, or in a free pleura with the appearance of purulent pleurisy.

These two lesioned aspects are the basis of the classification of Dévé [10] improved by Mésteri [6,11] and his colleagues in 1987, that they took into consideration four types of intrathoracic rupture of the hydatid cyst of the liver ,based on the importance of bronchial fistulas; and pleura parietal fistulization.

The various clinical pictures produced when the thoracic fistula is installed are variable and depend on the degree of importance of the transdiaphragmatic fistula and the organ where the bile pours and the suppurative content [12].

In case of biliopleural fistula, the symptomatology can be insidious, especially when the fistulous paths (s) are minimal. The table produced is that of the bilithorax: pleural fluid effusion of biliary nature,

The respiratory signs are reduced to an irritative cough and right basithoracic pain. Examination will reveal minimal right effusion syndrome. The biliopleural fistula can manifest itself in a more noisy way and in general realize the picture of a pyothorax, it then settles a brutal side point, a fever, coughs that are productive and bring back purulent sputum and dyspnea more or less important depending on the intensity of the effusion.

In the case of biliopleurobronchial fistula, a complication which follows the neglected biliopleuralfistula, the symptomatology will produce a pyothorax table associated with purulent bronchorrhea or biliptysia as in the case of our patient, indeed biliptysia is the main symptom, it is pathognomonic but inconstant [13], present in 77.8% for Chehab et al. [14] But it is only present in 29.5% and 12.5% of cases for Sakhri et al. [8] and Kilani et al. [15].

Radiological investigations must include a front and side chest roentgenogram and an abdominal ultrasound. On the chest roentgenogram, there is generally an elevation of the diaphragmatic dome (right), hydro-aeric thoracic levels or pleural effusion as the case of our patient.

The abdominal ultrasound allows to find the hydatid cysts of the liver, to specify its seat, its size, its vascular reports and to sometimes objectify the transdiaphragmatic fistula but also to examine the vesicle and the main bile duct.

The thoracic CT would bring a more precise study of the hepatic, pleuropulmonary lesions and the diaphragmatic breach. Bronchoscopy helps clarify the severity of lesions in the bronchial tree. [16]

The first step in the treatment of FBB and FBPB is a general, nutritional and respiratory preparation: respiratory physiotherapy, antibiotic therapy, blood transfusion in the event of anemia and hydro-electrolytic and caloric rebalancing.

Some [17,18] have proposed before the intervention an endoscopic sphincterotomy to reduce the flow of very productive bile fistulas and avoid corrosive contact of bile with the bronchi, in order to reduce the rate of postoperative complications which is consistent with taking charge of our patient.

Surgery generally has five objectives: the treatment of endothoracic lesions, which often requires significant parenchymal excision; treatment of hydadid cysts of the liver after hepatodiaphragmatic disconnection; research and treatment of intracystic bile fistulas; repair of the diaphragm; adequate drainage of the pleural cavity and the intrahepatic (cystic) cavity and / or the interhepato-diaphragmatic space. [8.15, 19.20]

The progression after biliobronchial fistula surgery can be marked by complications in 16% of cases, dominated by pleuropulmonary suppurations. [5, 21]

Conclusion:-

The incidence of bilio-pleurobronchial fistula complicating a hydatid liver cyst is rare. It is a severe complication of hepatic hydatidosis. Despite the improvement of resuscitation and operating techniques, it has a severe prognosis. Thoracotomy remains the best way, preoperative endoscopic sphincterotomy is a crucial step in preparing for this

surgery, it can even be offered as an exclusive treatment in non-operable patients with minimal hepatopulmonary lesions.

Through this work, we insist on rapid initial management of the hydatid cyst of the liver, in order to avoid progression to serious complications such as biliopleurobronchial fistula which can be life-threatening.



Figure 1:- The posteroanterior chest roentgenogram showed a homogeneous opacity occupying the totality of right thoracic field.



Figure 2:- The thoracic CT showed a right pleural effusion of high abundance and total pulmonary atelectasia.



Figure 3:- Abdominal CT scan showed a hydatid cyst of the liver.



Figure 4 (A): The posteroanterior chest roentgenogram showed a good radiological evolution .



Figure 4 (B): CT scan showing an encysted pleural effusion containing hydroaeric levels with a normal pulmonary parenchyma.

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