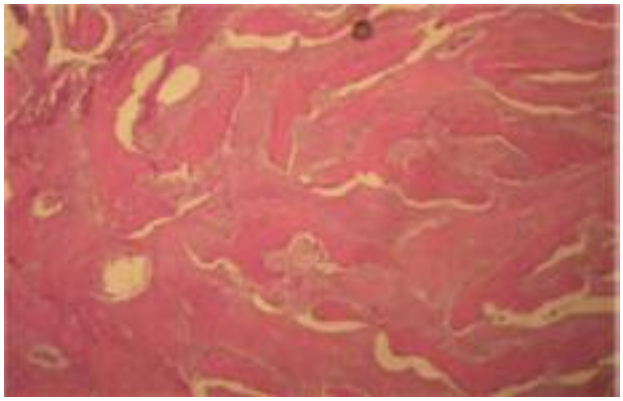
Figure 4 : tissu fibroblastique dans lequel on observe des travées osseuses épaisses lamellaires.



Trois ans après, réapparition de la déformation faciale. Celle-ci est toujours isolée : pas de signes associés en particulier neurologiques ou ophtalmologiques. La TDM a mis en évidence la même lésion diagnostiquée en 2002 mais avec atteinte du sphénoïde. La décision était l'expectative et la surveillance clinico- radiologique.
Actuellement, on est à trois ans de recul ; l'asymétrie faciale ne s'est pas accentuée. Il n'y a pas de signes de compression notamment ophtalmologique.

Conclusion

Le diagnostic de fibrome ossifiant est posé sur un faisceau d'arguments cliniques, radiologiques, histologiques et évolutifs. Il est aisé lorsqu'il s'agit d'une forme typique. Mais, des formes atypiques existent. La distinction avec une dysplasie fibreuse peut ainsi être difficile. Le diagnostic dans ce cas repose essentiellement sur l'examen histologique ainsi que l'évolution clinique.

Références

1) Tuméfaction maxillaire révélatrice d'une dysplasie fibreuse à propos d'un cas/ Pierre Gangloff, Anne Polo, Herve Moizan, Nicolas Froment, Eric Gerard 2004.

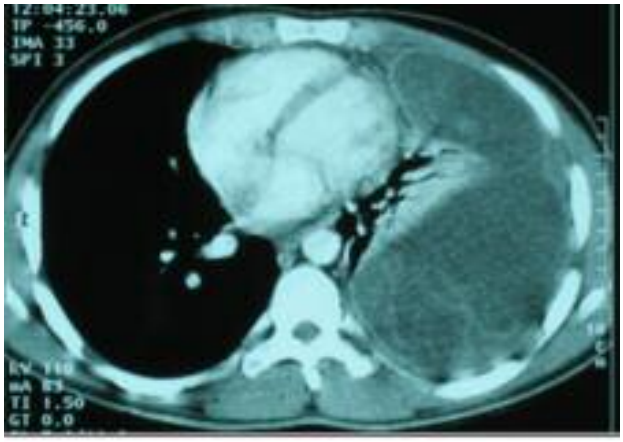
Landolsi Amany, Jbali Souheil, Ben Rejeb Maha, Ben Slimen Najoua, Zitouni Karima, Zairi Issam, Adouani Ali.
Service de chirurgie maxillo-faciale et esthétique, l'hôpital Charles-Nicolle, Tunis, Tunisie.
Faculté de médecine de Tunis, Tunis El Manar, Tunisie.

Left hepato-pleural fistulae complicating primary hepatic hydatidosis

Hydatid disease, a worldwide zoonosis which is caused by infestation with the *Echinococcus granulosus,* may occur in almost any organ of the host. The evolution of hepatic hydatid cysts is unpredictable; indeed the complications are numerous like the infection, the compression and the rupture in adjacent organs: bile ducts, peritoneal cavity, digestive tract and the thorax [1].

A 22-year-old man presented with cough, high fever and pain in the left side of his chest of 1 week's duration. The patient reported a history of contact with dogs. At admission, results of physical examination were normal except for signs of left pleural effusion. The leukocyte count showed 12,400 cells/mm with 4% eosinophils.
A Chest Roentgenogram disclosed a huge left pleural effusion, consolidation and atelectasis at the lung base. Ultrasonographic examination of the abdomen revealed an enlarged liver with a large multilocular cyst in the segment II which is open in the left pleural cavity and a unilocular cyst in the segment I.
A computed tomographic scan confirmed the ultrasonographic findings by showing a hepatic hydatid cyst invading the left thoracic cavity throughout the diaphragm concomitant with parasitic membrane floating on the pleural effusion [Fig1].

Figure 1 : Computed tomographic scan of lungs shows a characteristic image of a parasitic membrane floating on the pleural fluid.



The *Echinococcus* indirect hemaglutination titer was 1/2560.
A left sixth interspace posterolateral thoracotomy was made, allowing evacuating 750ml of purulent and bilious liquid. After aspiration, we found to contain a hydatid cyst with daughter cysts. Visceral and parietal pleura were very thick. Dissection of the adherences to the lung has been the first step.
The left hemi diaphragm was elevated, and adhesions between the left lower lobe and the diaphragm were noted. At the level of the diaphragmatic dome, there was a hepatic-pleural fistula of 3cm. We proceed to a wide pleural toilet with H2O2 followed by a decortication. The phrenotomy was sutured by points separated in the vicryl. The thoracotomy was sutured in layers after placing two left-sided chest drainage tubes on the pleura.
The second step was a laparotomy allowing a pericystectomy with suture of the biliary fistulas. Thereafter the cavities were washed with hypertonic saline solution. Epiplooplastie and External marsupialization over one transabdominal tubes was performed.
Postoperative recovery was uneventful, the tubes were removed after 6 days, when they no longer drained.

Albendazole (800 mg /day) was administered for 3 months. There has been no recurrence for more than three years.

Hydatid disease, which is caused by the *Echinococcus granulosus* tapeworm, has been acknowledged as a clinical entity since ancient times. Organs of scarified animals were described in the Talmud as "bladders full of water".

Transdiaphragmatic migration of hepatic hydatid disease in the posterior segments of the right hepatic lobe attributable to their proximate location in regard to the diaphragm occurs in 0.6–2.5% of cases [1].

Many factors may promote the intrathoracic occurrence of a hepatic hydatid cyst [2]: - The pressure gradient between the pleural and abdominal cavities favors a thoracic direction;

Compression and ischemia of the diaphragm secondary to inflammation of the cyst;

And the chemical action by bile on the diaphragm, lung, and pleura.

If enough adhesions precede the erosion, the cyst will rupture into the pulmonary parenchyma causing pneumonitis and broncho-biliary fistula. When the cyst crosses the diaphragm, the possibility of rupture into the pleural cavity, or even into great thoracic vessels [3] or pericardium increases.

In 1987 Mestiri and colleagues [4] enhanced Deve's classification into four types:

•Type I: Direct fistulisation of the cyst into the bronchi with small, I-A, or large fistula, I-B.

•Type II: Intrapulmonary parenchymal collection without fistula, II-A, or with large fistula, II-B.

•Type III: Intrapleural encysted collection without, III-A, or with bronchitic fistula, III-B, or parietal fistula, III-C;

•Type IV: Rupture into the pleural cavity with pleural effusion (hydatidothorax), IV-A, or a secondary pleural hydatidosis, IV-B.

This last type, like in our case, raises problems difficult to resolve so much on the diagnostic plan that therapeutics.

Because the hydatid fluid is highly antigenic, rupture into the pleural cavity results in chest pain and sometimes anaphylactic shock.

Diagnostic imaging studies have been very helpful in identifying the communication and in delineating its location.

The relevant chest radiography findings are elevation of the diaphragm, pleural effusion, consolidation and atelectasis at the lung base, and an hourglass-shaped lesion or a loculated pleural effusion in the posterior thorax [5].

Ultrasonography (US) is helpful for evaluating the hepatic hydatid disease and the pleural effusion, but CT can clearly demonstrate the transdiaphragmatic migration and the thoracic component of the disease [6].

CT scan is a very important diagnostic tool as the information they provide about the local extension of the disease, the evaluation of adjacent organs, or the existence of other cysts are essential for surgical management.

Bronchoscopy had a limited role because it was impossible to visualize the fistula due to the small size of the tract. It was mainly used for bronchial hygiene.

Surgery remains the treatment of choice and the acquired experience in hepatic echinococcosis and pulmonary brings to light four fundamental points in the therapeutics namely [7,10]

The necessity of reconstituting the diaphragmatic septum:

•The necessity of an excision lungwort in case of advanced lesion

•To realize the hepato-diaphragmatic disconnection

•The necessity of assuring the free court of the bile

•The necessity of treating the original cyst

Extended pulmonary resections are not justified taking into account that echinococcosis is a benign disease and has always the risk of recurrence so there is a vital need for sparing as much viable lung tissue as possible [8].

Medical treatment with mebendazole or albendazole has been reported [9]. It is mainly used perioperatively when dissemination is confirmed and total resection of the cysts is not possible.

Athough the incidence of hydatidosis has been really decreased today, rupture of hepatic hydatid cysts into the thorax remains a dangerous entity with a high morbidity and mortality. Careful assessment and early treatment of septic complications are essential in successfully treating this rare condition.

References

[1] Kabiri H, El Maslout A, Benosman A. Thoracic Rupture of Hepatic Hydatidosis (123 Cases). Ann Thorac Surg 2001; 72: 1883– 6.

[2] Kabiri H, Chafik A, Al Aziz A, El Maslout A, Benosman A. Traitement des fistules bilio-bronchiques et bilio-pleuro-bronchiques d'origine hydatique par thoracotomie. Ann Chir 2000 ; 125 : 654-9.

[3] Turgut A T, Altınok T, Topc¸u S, Kos U. Local complications of hydatid disease involving thoracic cavity: Imaging findings. European Journal of Radiology 2009; 70 : 49–56.

[4] Mestiri S, Kilani T, Thameur H, Sassi S. Les migrations thoraciques des kystes hydatiques du foie: proposition d'une classification. Lyon Chir 1987; 83:12–6.

[5] Turgut AT, Altın L, Topcu S. Unusual imaging characteristics of complicated hydatid disease. Eur J Radiol 2007; 63: 84–93.

[6] Pedrosa I, Saiz A, Arrazola J. Hydatid disease: radiologic and pathologic features and complications. Radiographics 2000; 20:795–817.

[7] Sakhri J, Ben Ali A, Letaif R, Derbel F, Dahman Y, Ben Hadj Hmida R. Les kystes hydatiques du foie rompus dans le thorax. J Chir 1996 ; 133 : 437-41.

[8] Gomez R, Moreno E, Loinaz C. Diaphragmatic or transdiaphragmatic thoracic involvement in hepatic hydatid disease: surgical trends and classification. World J Surg 1995; 19:714–9.

[9] Abi F, El Fares F. Fistules bilio-thoraciques : diagnostic aisé mais problèmes thérapeutiques et pronostiques. Maghreb Méd 2004 ; 204 : 11-6.

[10] Kilani T, Daoues A, Horchani H, Sellami M. Place de la thoracotomie dans les complications thoraciques des Kystes hydatiques du foie. Ann Chir Thorac Cardiovasc 1991; 45: 705–10.

Sonia Ezzine Baccari, Mehdi Bouassida , Selim Sassi , Hassen Touinsi, Fethi Chebbi, Mongi Mighri, Sadok Sassi

Department of thoracic and general surgery, Tahar Maamouri Hospital, Nabeul, Tunisia, North Africa.
