

Hepatic teratoma: Case Report

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Abstract

Hepatic teratoma is an extremely rare tumour. Since 1898 only 28 cases of hepatic teratoma have been reported in the literature, including 21 in pediatrics. The diagnosis is often confirmed by the anatomopathological study and the therapeutic approach is multidisciplinary based essentially on surgery. We report the case of a 45-year-old woman with a history of ductal carcinoma of the right breast for which she underwent surgery. The patient presented with pain in the right hypochondrium, a diagnosis of hydatid cyst of the liver was made based on a cystic image of the right liver on ultrasound. It showed a heterogeneous thin-walled cystic image in the right liver suggesting a type IV hydatid cyst. The MRI was in favor of a fluid lesion of the hepatic dome evoking a remodeled hydatid cyst. During surgery, the liver was found of normal size and segment VIII is the seat of an oval formation 70/60 mm in diameter. A resection of the cystic formation with a closed cyst was performed. Histological analysis concluded with the diagnosis of mature teratoma with no signs of malignancy. After a follow-up of 12 years, the patient is in good health and without recurrence.

Keywords: Hepatic teratoma, rare, differential diagnosis, histology.

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INTRODUCTION

Teratomas (monster tumors) are rare tumors characterized by abnormal growth of embryonic multipotential cells from the three germ layers: of ectodermal (skin, nervous tissue), mesodermal (muscle, adipose tissue) or endodermal (tube digestive system, bronchi), they are differentiated by the nature and maturity of the tissues grouped together within the tumor [1].

Hepatic teratoma is an even rarer tumour. Few cases have been published in the medical literature. Since 1898 only 28 cases of hepatic teratomas have been reported in the literature, including 21 in pediatrics.

We report in this work an observation of hepatic teratoma in a 45-year-old patient collected in the surgery department B of the University Hospital of Rabat and we discuss the clinical, radiological, histological, evolutionary and therapeutic characteristics of this pathology, in the light of the literature.

CASE REPORT

This is a 45-year-old patient, married, mother of 4 children, from and living in Sidi Slimane, mutualist. The patient was hospitalized for pain in the right hypochondrium.

She had no medical history, hydatid contagion or toxic habits, and she was operated on for intraductal carcinoma of the right breast in 2010. The patient does not report a similar case in the family.

The story dates back to January 2011 with the onset of pain in the right hypochondrium, which prompted a general medical consultation. An abdominal ultrasound was performed showing a cystic image of the right liver evoking a hydatid cyst of the liver.

The clinical examination finds a patient in good general condition on admission. WHO: 0 Weight: 63kg Height: 1.58m BMI 25.2.

- The abdominal examination is unremarkable, in particular there is neither hepatomegaly nor splenomegaly.
- The pleuro-pulmonary examination is normal, as is the cardiovascular examination.
- The patient showed no neurological signs and the lymph nodes were free.

- Ultrasound showed in the right liver (anterior sector) a heterogeneous cystic image with a thin wall evoking a type IV hydatid cyst.

Abdominal MRI showed a liver of normal size, with regular contours, site of a well-defined segment VIII lesion, surrounded by a hypo signal border on all sequences.

This lesion is roughly oval measuring 57*48 mm. It is the seat of a liquid level with a supernatant in T1 hypersignal, T2 hypo signal and a sloping part in T1

and T2 isosignal without enhancement after injection of gadolinium.

This sloping part is made of a serpiginous structure evoking a hydatid cyst. Also, absence of suspicious focal lesion in the rest of the liver and absence of deep ADP. There is no intraperitoneal fluid effusion.

The MRI aspect is therefore in favor of a fluid lesion of the hepatic dome evoking a remodeled hydatid cyst

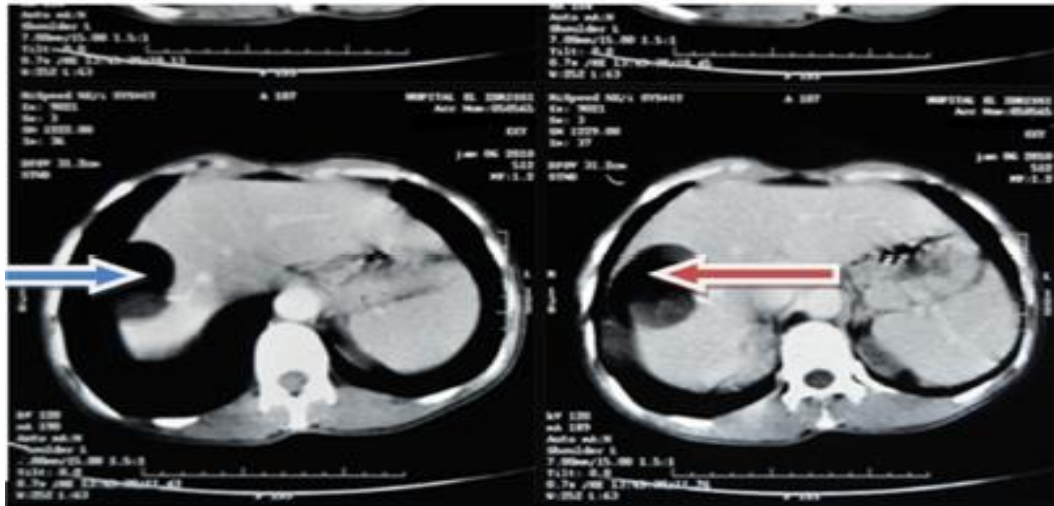


Figure 1: Abdominal MRI axial C+ section

Blue arrow: well-circumscribed lesion of the right liver with cystic-looking liquid level not taking contrast; Red arrow: right suprahepatic vein



Figure 2: Red arrow: frontal section C+ abdominal MRI showing a rounded lesion of the right liver with a watery tone without contrast uptake in the periphery

The preoperative biological assessment did not reveal any anomaly.

The patient was operated on. On exploration, segment VIII is the seat of an oval formation 70/60 mm in diameter on intraoperative ultrasound, the protruding dome of which is whitish in appearance. The content seems thick.

The diagnosis retained at this stage is that of a remodeled type IV hydatid cyst. The cyst wall appears thick. A resection of the cystic formation with a closed cyst is decided. The resection is performed under vascular clamping of the hepatic pedicle for 25 min, the cutting plane passes through healthy tissue carrying the entire wall of the cystic mass.



Figure 3: Operative resection piece

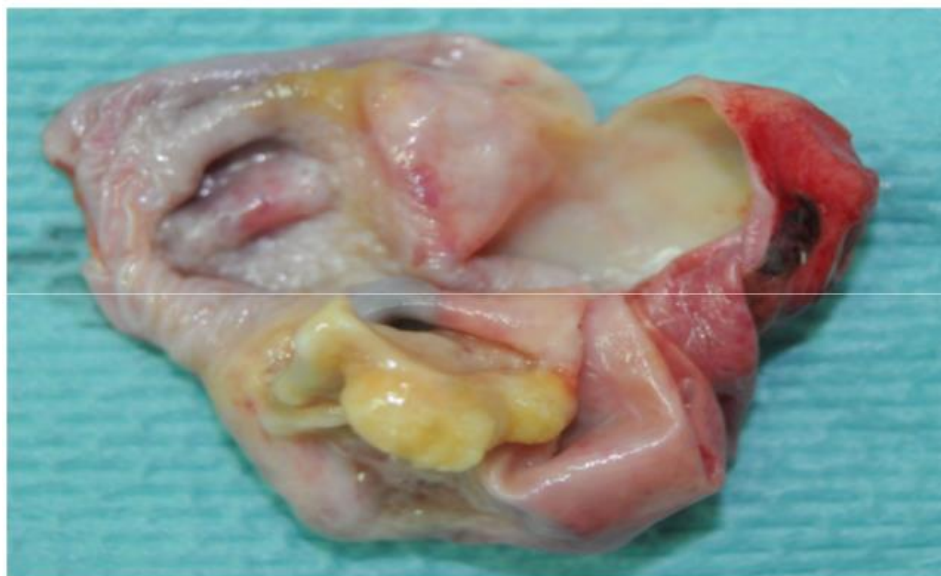


Figure 4: Operative specimen after opening showing a smooth-walled cystic appearance Slightly thick seat of some endocystic vegetations

The anatomopathological study of the surgical specimen showed a hepatic parenchyma seat of a cyst bordered by a regular flattened epithelium, containing a connective tissue surmounted by a respiratory type epithelium, made of ciliated columnar cells and a cartilaginous tissue, made of chondrocytes arranged in clusters separated by an amorphous matrix, evoking a mature teratoma of the liver.

The postoperative follow-up was simple, the patient left the hospital on the fifth postoperative day, with a follow-up for 6 years without abnormality.

DISCUSSION

Liver teratomas are exceptional neoplasms representing less than 1% of all teratomas. Even in pediatrics, where they are most commonly seen, they represent only 1% of all liver tumors [2].

Since 1898, only 28 cases of hepatic teratomas have been reported in the literature, 21 of which were observed in pediatrics [2].

The majority of cases were in female children under three years of age, with most occurring in the right lobe of the liver [1].

Less common hepatic locations include: the left lobe of the liver [2], the posterior part [3] and the falciform ligament of the liver [4].

According to our case and in accordance with data from the literature, hepatic teratoma is usually discovered by abdominal pain, but sometimes by an abdominal mass or jaundice [1].

Symptoms are usually specific to the location of the tumor, related to the mass effect on adjacent organs [10].

In adults, benign teratomas do not directly affect the general condition of the patient. Compression of adjacent structures is the cause of other symptoms that may be the reason for consultation, such as abdominal distension, nausea and vomiting [5].

Occasionally, acute abdominal pain may develop in the case of malignant hepatic teratomas most often associated with rapid degeneration [10].

Moreover, a fortuitous discovery during surgery or during an abdominal ultrasound requested for another reason is not uncommon.

In our case, abdominal pain in the right hypochondrium motivated the patient to consult a general practitioner where an ultrasound was performed showing a cystic image in the liver.

According to data from the literature and in our patient, in the majority of cases the clinical examination is strictly normal.

Imaging plays a key role in the diagnosis of these tumors, thanks to the possibility of identifying the different components of the teratoma, thus making it possible to evoke the diagnosis before surgery [11].

Currently the abdominal ultrasound-computed tomography pair or abdomino-pelvic is at the origin of the discovery of the majority of these tumors.

It makes it possible to specify the seat, the size and the contours of the tumor [12]. When the size of the tumor is less than 2 cm, it may go unnoticed.

Conversely, when the tumor is large, it poses the problem of its starting point. On average, the size of a teratoma at the time of diagnosis is 10 cm [12].

This ultrasound makes it possible to evaluate the locoregional extension, to check the contralateral structures and to carry out an assessment of extension to the vessels, to the local lymph nodes and to the adjacent organs. In addition, it plays a major role in medium- and long-term postoperative monitoring [13].

It also makes it possible to specify the structure of the mass (solid, cystic or mixed), its vascularization using Doppler [14] and the differentiation between mature and immature teratomas [15].

The sonographic appearance of mature teratomas is well known but is a frequent source of pitfalls, responsible for false positives and false negatives [16].

Classically, three forms are described:

- Cystic formation with an echogenic nodule hanging from its wall (Rokitansky's nodule);
- Attenuating echogenic mass. Posterior attenuation is secondary to the presence of hair, thick sebum within the lumen of the lesion, fat, or ossified structures.
- Cysts containing multiple linear echoes corresponding to hair floating in the lumen of the cyst

After ultrasound, CT or MRI is only done if a malignancy is suspected. For mature teratoma, CT scan or MRI are of no interest except in the case of an atypical form. CT greatly contributes to the exploration of retroperitoneal tumors and the assessment of their relationship with neighboring organs with a reliability of 95% [17].

It provides interesting information for the diagnosis of teratoma by identifying its three tissue

components, fatty and calcium which can exist in variable proportions.

Cystic teratomas result in a mass of fluid density, well circumscribed, encapsulated with areas of fatty density and calcifications. These cystic teratomas often present with a Rokitansky protuberance, which may be a simple localized thickening or septum within the mass. On injection of contrast product, an enhancement of the wall and of the Rokitansky prominence is noted.

The fleshy component of teratomas gives an intermediate and heterogeneous signal in T1 and a hyper signal lower than that of fat in T2. The cystic component gives an intense hypo signal in T1. The fatty component is in hyper signal T1 and in less intense signal in T2. The calcifications appear in hypo signal on all the sequences, but they are less well identified than in CT.

After injection of gadolinium, the signal of the fatty component remains unchanged [18].

The half-life of α FP is 5-7 days, a high level of α FP indicates the presence of malignant components. When the ablation is complete, the level of α FP decreases rapidly in the serum to normalize in a few weeks. The re ascent of the rates indicates recurrences or metastatic dissemination. The unexpected increase in α FP after chemotherapy is due to cell lysis [19].

Free β -HCG can be increased in malignant germ cell tumors but also in other malignancies such as bladder cancers.

The increase in these tumor markers affirms with certainty the presence of the secreting component in the tumor, even if it is not found on histology, despite a very careful analysis. It is essential to carry out these assays before surgery [20].

As part of the follow-up, the assay of these markers precisely identifies the presence or absence of the secreting tumour, since the secreting nature constitutes a pejorative factor as regards the evolution and the risk of recurrence [21]. It will be repeated regularly in all patients with a germ cell tumor regardless of the histological type. Indeed, secreting tumors sometimes recur in non-secreting tumors and vice versa. A secondary and confirmed rise in the level of α FP or β HCG testifies to a recurrence or a metastasis, sometimes several months before any clinical sign.

There are four histological variants of teratomas described: mature teratoma, immature teratoma, teratoma with malignant transformation and mesodermal teratoma [22].

The differential diagnosis of hepatic teratoma arises essentially with all cystic lesions of the liver.

The hydatid cyst of the liver represents the main differential diagnosis because of our epidemiological context. This is the most common cystic lesion of the liver after the biliary cyst.

In our patient, in view of the initial symptomatology, the notion of contact with dogs as well as the cystic image on ultrasound, the diagnosis that was first evoked was the hydatid cyst of the liver.

Surgical resection of hepatic teratoma is the treatment of choice. The approach used is a laparotomy. It makes it possible to meet oncological requirements [23].

Anatomopathological examination is the only means that allows a diagnosis of certainty and surgical resection is the only curative treatment. The presence of immature tissue within the tumor drastically alters the patient's prognosis [22, 10, 24, 25].

Surgery is a safe and effective treatment for patients with a resectable teratoma. Adhesions to neighboring structures, particularly vascular ones, can make complete resection difficult and constitute the main risk factor for recurrence if tumor resection is incomplete [26, 27].

In our patient, a right subcostal laparotomy was performed and a resection of the cystic formation with a closed cyst was performed.

Postoperative follow-up is essential, whether or not adjuvant treatment is started, which can be readjusted or started, if necessary, in the event of progression or recurrence of the disease.

Maximum monitoring efforts should be made within the first three to five years after treatment. Monitoring after surgery is carried out at regular time intervals, close together at the beginning, and can be more and more spaced out in the event of favorable evolution. Thus every 3 months the first year, every 6 months for 5 years then every year. This monitoring is based on:

- The clinic: to assess the evolution of a residual mass or a serous effusion.
- Biology: to detect a possible sub-clinical recurrence or relapse by measuring tumor markers. Monitoring of the markers must be carried out every 2 months during the first two years following the end of treatment and any re-ascent of the markers must be considered a priori as a relapse.
- Radiology: to monitor the appearance of possible recurrences or the evolution of a residual mass.

Our case benefited from regular clinical and paraclinical monitoring with a 6-year follow-up without recurrence.

The evolution of teratomas is unpredictable. However, some complications may be observed, namely cyst rupture and malignant degeneration which may cause liquefied sebaceous content to leak into the peritoneum and further complicate the patient's course and prognosis [6, 26, 22, 29]. The prognosis of hepatic teratomas depends on several parameters, in particular, the malignant or benign nature of the tumour, the patient's age and the AFP level. Malignant teratoma has a poor prognosis; however, complete resection of a benign tumor is curative [30]. The prognosis of teratomas containing AFP-producing cells appears to be poor. Carter et al [26] observed that all patients over the age of 15 died within a year.

CONCLUSION

Teratomas are rare malformative tumors derived from embryonic multipotential cells from at least two of the three germ cell layers and must show evidence of organ formation. The hepatic localization is exceptional and raises the problem of diagnosis.

Abdominal pain is the main revealing sign of these tumors. Ultrasound is the first-line examination. The diagnosis is histopathological and the therapeutic approach is multidisciplinary based essentially on surgery.

The anatomopathological examination is essential in the search for malignant elements which will guide the prognosis and possible future treatment. Regular postoperative clinical, biological and radiological monitoring in the medium and long term is necessary because of the risk of recurrence or the occurrence of metastasis. Although hepatic teratoma has almost always been reported in children, a diagnosis of hepatic teratoma should also be kept in mind in adult patients.

Data Availability Statement

The data that support the findings of this article are available from the corresponding author upon reasonable request.

Conflicts of interest: Drs Moustapha Traore, Jihane Sabar and Abdellatif Settaf declare no conflict of interest.

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