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Spontaneous rupture of giant hepatic hemangioma: a rare source of hemoperitoneum. Case report

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SUMMARY: **Spontaneous rupture of giant hepatic hemangioma: a rare source of hemoperitoneum. Case report.**

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Background. *Hemoperitoneum due to spontaneous rupture of a hepatic hemangioma is a rare and serious clinical event with a high mortality rate.*

Case report. *25-year-old woman under hormonal treatment for pregnancy with abdominal pain with distension followed by vomits, palpable epigastric mass and paleness of the skin and mucosas. Computed tomography of the abdomen without oral and venous contrast showed a heterogeneous and capsulated tumor of the liver. Exploratory laparotomy was carried out that revealed a large tumor occupying the gastro-hepatic site with partial rupture of the tumor's capsule with bleeding. Because of the close relationship between the tumor and the retro-hepatic inferior vena cava, the partial resection of the tumor was realized. The patient had a good post-operative evolution. The study of the tumor revealed hepatic hemangioma.*

Conclusion. *Hepatic Hemangiomas may evolve to spontaneous rupture leading to hemorrhagic acute abdomen. Surgery is mandatory and the resection of the Hemangioma will depend on the clinical condition of the patient and on the relationship of the tumor with the vascular structures of the liver. Partial resection is a safe choice that saves lives in urgency situations, as the one described herein.*

RIASSUNTO: **Rottura spontanea di emangioma epatico gigante: causa rara di emoperitoneo. Case report.**

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Background. *L'emoperitoneo da rottura spontanea di un emengioma epatico è un evento clinico raro e grave con alta mortalità.*

Caso clinico. *Una paziente di 25 anni di età, in terapia ormonale per avere una gravidanza, ha presentato un improvviso dolore addominale con distensione e vomito, massa epigastrica palpabile e pallore cutaneomucoso. La tomografia computerizzata dell'addome senza contrasto ha evidenziato una formazione capsulata nel fegato.*

L'esplorazione chirurgica ha rilevato un grosso tumore localizzato nella zona epato-gastrica con rottura della capsula e sanguinamento. A causa dello stretto rapporto tra massa tumorale e vena cava inferiore retro-epatica è stata realizzata la rimozione parziale del tumore.

La paziente ha avuto un'evoluzione post-operatoria soddisfacente. L'esame istopatologico ha rivelato un emangioma epatico.

Conclusione. *L'emangioma epatico può evolvere in una rottura spontanea e causare un quadro di addome acuto emorragico. La chirurgia è necessaria e l'asportazione dell'emangioma dipenderà dalle condizioni cliniche del paziente e dal rapporto tra il tumore e le strutture vascolari epatiche. La rimozione parziale è un'alternativa sicura che può salvare la vita in caso di urgenza, come è stato descritto nel presente caso.*

KEY WORDS: Giant hepatic hemangioma - Liver - Hemoperitoneum - Surgery.
Emangioma epatico gigante - Fegato - Emoperitoneum - Chirurgia.

Introduction

Hemangiomas are benign vascular tumors constituted by a web of blood vessels, covered with endothelial cells and, histologically, are always followed by thrombosis, necrosis, hemorrhagic areas, fibrosis and calcifications (1). The liver is a common site for hemangiomas. It's more frequent in women and its incidence may have some relationship with hormonal patterns (1).

The aim of this report is to describe a case of a young woman, near the end of her pregnancy, under hormonal treatment, that evolved with hemoperitoneum due to rupture of a giant Hepatic Hemangioma (HH). She was operated in a state of urgency. The diagnostic aspects and surgical strategy employed are controversial.

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Case report

A 25-year-old woman was admitted with abdominal pain followed by vomits, palpable mass in the epigastrium and cutaneous-mucous paleness for 3 days. She was under hormonal treatment in order to become pregnant. She had a slightly distended abdomen, diffuse pain on palpation with a painless mass in epigastrium. Abdominal ultrasound revealed a lesion with a heterogeneous aspect in the liver's left lobe and a moderate amount of free liquid in the cavity. Computed tomography of the abdomen without oral and venous contrast showed a heterogeneous and capsulated tumor, pushing the stomach to the left and having a close relationship with the visceral face of the liver, occupying all lesser omentum's space (Fig. 1). It was opted for the performance of a culdocentesis with the aspiration of live blood.

The patient underwent a median exploratory laparotomy and a great quantity of live blood and clots were found in the abdominal cavity, the presence of a large capsulated tumor occupying the gastro-hepatic site, moving the stomach to the left and the liver's hilum to the right. Such a tumor spread to the visceral face of the liver, penetrated the 4th segment and had a close relationship with retro-hepatic inferior vena cava. There was also a rupture of the tumor's capsule located inferiorly to segment III of the liver, where the bleeding was originated. With these findings, a partial resection of the tumor was performed, starting from the place of the rupture of the tumor's capsule, taking away all of the tumor which occupied the gastro-hepatic site, leaving the posterior part of the tumor located at the visceral face of the liver. Hemostasias of the cruent area of the tumor was carefully performed with electric-clotting, stitching was carried out with Prolene 3-0 and application of Gelfoam®. Later, an exhaustive lavage of the abdominal cavity and introduction of a tubular drain positioned in the gastro-hepatic space exteriorized by a counter-opening of the left flank were performed. The patient, during surgery, received a total volume of 1,500 ml of erythrocyte concentrate.

She evolved quite well after surgery being discharged from the hospital in good clinical conditions 10 days after surgery. The study of the resected tumor confirmed the diagnosis of HH. At the moment, 10 months after the surgery, the patient is performing her usual activities without symptoms.

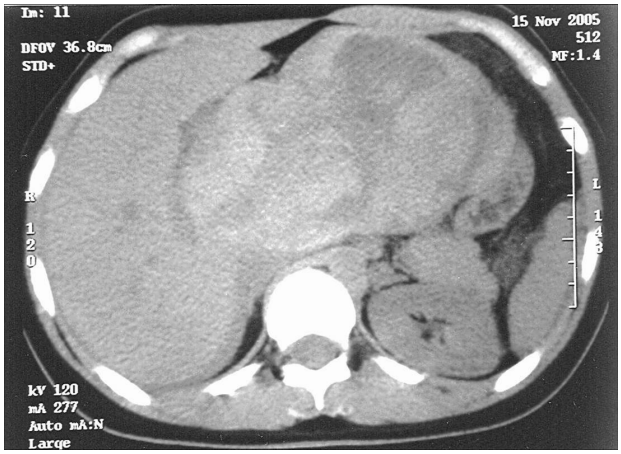

Fig. 1 - CT scan showing a large, heterogeneous and capsulated tumor.

Discussion

The hemangioma is the most common solid benign tumor of the liver (1-4). In most cases, HH have little dimensions and are asymptomatic, randomly discovered in most cases when complementary image exams are requested in order to diagnose a different disease, like cholelithiasis. Its prevalence is estimated to be between 0.4% and 7.3% of the general population (3). It is interesting, according to Genzini et al. (1), the fact that some authors report the association between the usage of stimulating hormones for ovulation with the growing incidence of HH. Facts reported by us in this case.

Ultrasound, computed tomography and magnetic resonance are useful exams for the evaluation of the HH (2). However, biopsy of the HH with a fine needle is not recommended due to the risk of complications related to bleeding (2).

HH that are bigger than 4 cm of diameter are considered giant HH; several therapies are assigned for these lesions, like radiation, ligature of the hepatic artery, trans-arterial embolization, resection and liver transplantation (3). Chui et al. (5) reported two cases of liver transplantation in patients carrying giant HHs. The surgical treatment of the HHs is indicated whenever it ruptures, evolving with hemoperitoneum, as the case described here, when there is a bleeding inside the tumor, rapid growth of the HH and when there is a consumption coagulopathy, a phenomenon known as Kasabach-Merritt syndrome (2, 6).

Spontaneous rupture of HH is rare, according to Capellani et al. (7), it is around 1 to 4%. Although rare it is associated with a high mortality rate, estimated between 60-75% of cases (6). Pietrabissa et al. (8) state that the spontaneous rupture of HH is an exceptional phenomenon.

In 2003, Corigliano et al. (2), while reviewing the medical literature, found 33 cases of hemoperitoneum by spontaneous rupture of HH in adults. The case reported herein, besides being rare, must be the first case described of a patient from the Amazon region, as no similar case in the medical literature was found after thorough bibliographical research.

As for surgical strategies, there is not, in the literature, a case of partial resection of ruptured HH as described here. Such strategy was performed taking into consideration the close relationship of the tumor with the retro-hepatic vena cava, the urgent character of the surgery and the clinical condition of the patient. Gedaly et al. (4), in a series of 28 patients with HH who underwent surgery, has performed enucleation of the HH in 23, anatomic hepatic resection in 5 patients and concluded that either enucleation or resection are safe procedures to be carried out for the surgical treatment of HH.

Conclusion

With the experience acquired in this case, we conclude that HHs may evolve to spontaneous rupture leading to hemorrhagic acute abdomen; in these cases, the association between the use of ovulation stimulating hormones and HH must be considered. Surgery is mandatory and the resection of the tumor will depend on the clinical condition of the patient and on the relationship of the HH with the vascular structures of the liver. Partial resection is a safe choice that saves lives in urgency situations, as the one described herein.

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