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Spontaneous rupture of a giant hepatic hemangioma. Sequential treatment with preoperative transcatheter arterial embolization and conservative hepatectomy

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SUMMARY: **Spontaneous rupture of a giant hepatic hemangioma. Sequential treatment with preoperative transcatheter arterial embolization and conservative hepatectomy.**

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Hemangioma is the most common benign tumor of the liver and it is often asymptomatic. Spontaneous rupture of liver hemangiomas is a rare but potentially lethal complication. Emergent hepatic resection has been the treatment of choice but carries high operative morbidity and mortality. Recently, preoperative transcatheter arterial embolization (TAE) has been used successfully for the management of bleeding ruptured liver tumors and non-operative treatment of symptomatic giant liver hemangiomas.

We report a case of spontaneous rupture of a giant hepatic hemangioma that presented with thoracic and abdominal pain and shock due to hemoperitoneum. Once proper diagnosis was made the patient was successfully managed by TAE, followed by conservative hepatic resection.

RIASSUNTO: **Rottura spontanea di emangioma epatico gigante. Trattamento sequenziale con embolizzazione arteriosa transcatetere preoperatoria ed epatectomia conservativa.**

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L'emangioma è il più comune tumore benigno del fegato ed è spesso asintomatico. La rottura spontanea di un emangioma del fegato è una complicanza rara ma potenzialmente letale. La resezione epatica è stata il trattamento di scelta, ma ha un'alta morbilità e mortalità operatoria. Recentemente, l'embolizzazione arteriosa transcatetere (TAE) preoperatoria è stata utilizzata con successo per la gestione del sanguinamento da rottura di tumore epatico e per il trattamento conservativo degli emangiomi epatici giganti.

Riportiamo un caso di rottura spontanea di emangioma epatico gigante sintomatici manifestatasi con dolore toracico e addominale e shock da emoperitoneo. Posta la diagnosi corretta la paziente è stata trattata con successo con TAE, seguita da resezione epatica.

KEY WORDS: Giant hepatic hemangioma - Hemoperitoneum - Transcatheter arterial embolization - Liver resection.
Emangioma epatico gigante - Emoperitoneo - Embolizzazione arteriosa transcatetere - Resezione epatica.

Introduction

The first reported use of transcatheter arterial embolization (TAE) prior to surgical resection of ruptured hepatic hemangioma was by Yamamoto et al. in 1991 (1). Since that time only 6 more cases have been reported in the literature, with no patient mortality (2-5). We present a case of hemorrhagic shock after severe bleeding of a ruptured hepatic hemangioma which was percutaneously embolized in an emergency setting and surgically ressected after hemodynamic stabilization.

Case report

A 50 year-old woman was admitted in our emergency room with sudden onset of upper abdominal and right thoracic pain, and nausea. There was no history of trauma in the last 24 hours neither significant medical conditions. She was submitted to the emergency room protocol for acute thoracic pain and addressed to surgery once cardiac conditions were excluded. At this moment the patient was conscious, the blood pressure was 100/70 mmHg, pulse was 130 per min, and the initial blood test showed hemoglobin levels of 12,1 g/dL and high alanine amino-transferase levels (996 U/mL). After good response to volume resuscitation, she had an abdominal ultrasound which revealed a heterogeneous 13x10cm mass in the right liver lobe.

Six hours after admission and initial adequate response to conservative mesures the patient turned pale, the blood pressure dropped to 90/60 mmHg and pulse increased to 140 heart beats/minute. After initial resuscitation with 2000ml of saline and 2 packed red blood cells (RBC), patient underwent a contrast enhanced computed tomography (CT) (Fig.1) that demonstrated a vascular lesion in right lobe of the liver with signs of rupture and acute bleeding. There were associated hemoperitoneum and mild right pleu-

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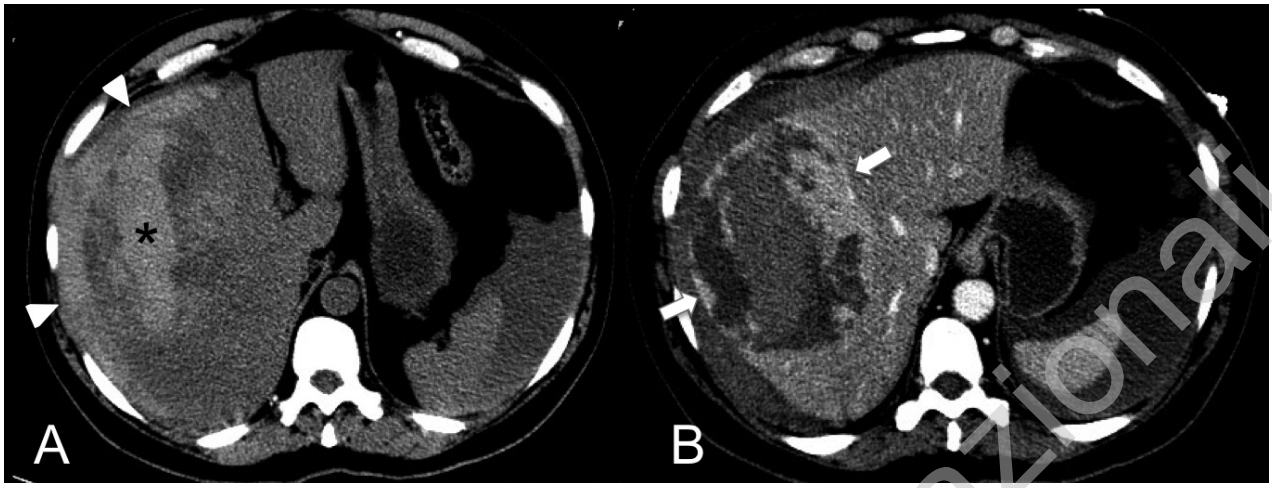

Fig. 1 - Unenhanced CT (A) showing large hepatic mass on the right lobe with heterogeneous hyperdense (blood) within the lesion (*) and extending to the abdominal cavity (hemoperitoneum; arrowhead). Enhanced CT (B) depicting centripetal peripheral nodular enhancement (arrows), typical pattern of hepatic hemangioma.

ral effusion. Remained liver parenchyma was normal with no other focal lesion. Digital Subtraction Angiography (DSA) revealed angiographic findings of a liver hemangioma and arterial contrast extravasation was noted on the celiac angiogram, after which a selective right hepatic arteriogram confirmed active contrast extravasation from the lesion.

The hemangioma was embolized in a single session, via selective right hepatic artery embolisation using PVA (polyvinyl alcohol) particles 500-750 μ (Cook Bloomington Inc., USA) delivered using a Cobra catheter. At the end of the procedure the patient was hemodynamic stable, and a check angiogram showed successful embolisation with no contrast leak.

The patient received a total of 2 packed RBCs and was submitted 6 days after angiography to MRI (Fig. 2) that showed partial reabsorption of the hemoperitoneum, a large ovoidal hypervascular lesion with centripetal peripheral nodular enhancement, and signs of recent bleeding. After 9 days from the initial episode she underwent laparotomy that revealed an steatotic liver, small amount of residual hemoperitoneum and a large hepatic mass with no active bleeding at all. A segment VIII anatomical resection was performed (Figs. 3 and 4) with no blood transfusion.

The postoperative course was uneventful, the patient fully recovered and discharged 6 days following surgery. Histologic examination revealed a hepatic hemangioma. Patient remained asymptomatic on follow-up at 10 months.

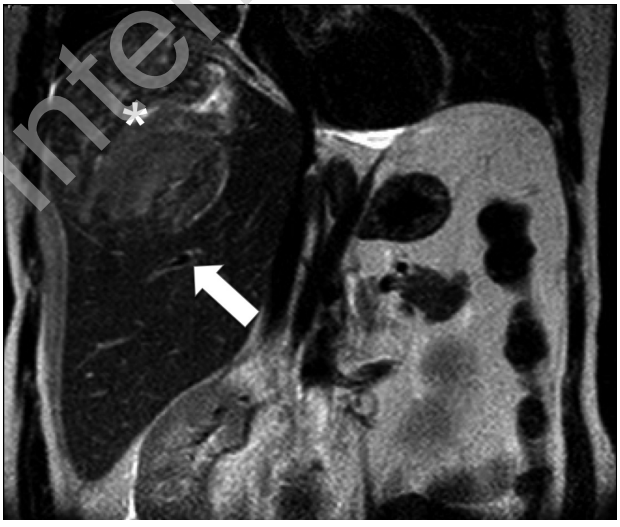

Fig. 2 - Coronal MR image after embolisation showing that the lesion (*) was confined to segment VIII, superior to the right hepatic pedicle (arrow).

Discussion

Hemangiomas are the most common primary liver tumor, with a prevalence in the general population estimated to range between 0.4% and 7.3% (5,6). They are usually diagnosed unexpectedly during routine abdominal ultrasound and generally present as small-sized, asymptomatic nodules. Those whose diameter exceeds 4 cm are called "giant hemangiomas" but there is no proven relationship between this classification and risk of complication and spontaneous ruptured were reported in bigger ones (4, 7-9).

Some authors showed a great enthusiasm with surgical therapy with excellent results (10,11). However, based on their benign clinical course conservative management is usually the best option (8, 9). Absolute surgical indications for hepatic hemangioma are spontaneous or traumatic rupture with hemoperitoneum, intratumoral bleeding, and consumptive coagulopathy (Kasabach-Merrit syndrome). Persistent abdominal pain, obstructive jaundice, portal hypertension, superficial location of tumors larger than 5 cm with a risk of trauma, and an uncertain diagnosis are relative surgical indications (9,12).

Intra-abdominal hemorrhage has been rarely described as a complication due to rupture of hepatic hemangioma after biopsy, after trauma, or spontaneously. Corigliano

Fig. 3 - Intraoperative picture showing liver after resection of segment VIII. Such a conservative approach was possible in a stable and elective condition.

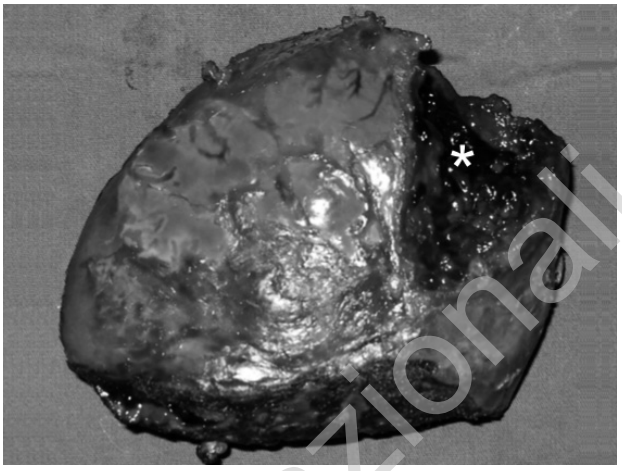

Fig. 4 - Resected segment VIII with ruptured giant hemangioma (*).

et al., reviewed 32 cases of spontaneous rupture of hepatic hemangioma in adults, 22 (95.7%) patients underwent surgery and the mortality rate of all surgery patients was 36.4% (8/22) (4). Treatment for symptomatic hemangiomas includes steroids, radiation therapy, surgical resection, hepatic arterial ligation, and transcatheter arterial embolization (13). Embolization should be as selective as possible, in branches feeding the lesion. Recent studies have emphasized the role of TAE in the effective treatment of symptomatic hemangiomas, progressively growing hemangiomas and those at risk of bleeding (13,14). The most common complications of embolization are pain, pyrexia, leukocytosis, and nausea, which last for a few days (13). Postembolization pain is due to thrombosis and necrosis. Severe complications are rare and include infection, hepatic abscess and sepsis, and migration of the embolization agent (13,15).

In our case, the lesion was subcapsular and located in segment VIII of the liver. Abdominal CT not only established the diagnosis of ruptured hemangioma but also demonstrated the site of rupture and suggested the possibility of active intra-abdominal bleeding. The successful preoperative embolization of the lesion in our patient proved to be very useful in reducing the intra-operative blood loss. The liver resection was perfomed with alternating partial inflow vascular occlusion, called "hemi-Pringle", allowing to minimal hemorrhage from the hepatic raw surface. No blood transfusion was required. Furthermore, there was no bleeding from the mobilization of the right liver lobe from the retroperitoneal space, and the shrinkage of the hemangioma following embolization resulted in an easier and quicker dissection of its margins what made possible a parenchyma preserving segment VIII resection (Figs. 3 and 4), otherwise our patient would probably have been submitted to a standard right hepatectomy during an emergency laparotomy.

Conclusion

In conclusion, we showed a case where TAE was helpful as an emergency option to quickly stop and control hemorrhage of a ruptured hepatic hemangioma and to postpone surgery to a stabilized and safer condition, enabling a parenchyma preserving surgical approach. The question if embolization alone should be the optimal treatment stands still, however the large size of lesions and their frequently subcapsular situation makes difficult to complete rule out the risk of future complications, and may cause important side effects such as pain and fever. Moreover, only surgery provides tissue confirmation.

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