Spontaneous rupture of splenic hemangioma in puerperium

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Summary

Atraumatic splenic rupture is a rare clinical entity and in the absence of trauma, the diagnosis and treatment are often delayed. In this article the authors discuss a case of a 45-year-old woman, gravida 5, para 4, with spontaneous splenic rupture on her second postpartum day. The rupture was related to a splenic hemangioma that is a vascular malformation and the most common neoplasm of the spleen. Despite the fact that hemangiomas are the most common primary neoplasms of the spleen, only few cases of splenic rupture have been described in pregnancy or puerperium. However, spontaneous splenic rupture is a rare event and the rupture should be suspected in woman with unexplained abdominal pain or with clear signs of haemorrhage.

Key words: Spontaneous splenic rupture; Splenic hemangiomas; Puerperal disorder; Postpartum splenic rupture; Splenectomy in postpartum.

Introduction

The first reported atraumatic spleen ruptures were described in 1891, by Rokitansky, in a patient with leukemia [1] and in 1874 by Atkinson who reported the rupture of an apparently normal spleen [2]. Since then numerous atraumatic splenic ruptures have been described in literature and often a distinction has been made between the rupture of a normal spleen defined as a "true spontaneous" rupture and the rupture of a diseased spleen described as a "pathologic" or "occult" rupture.

True spontaneous splenic rupture has been reported in literature, but its validity has often been challenged. Wright and Prigot stated "There is no such clinical entity as spontaneous rupture of the normal spleen" implying that thorough questioning and investigation will reveal a history of trauma or splenic pathology [3]. After reviewing reports of spontaneous splenic ruptures through 1958, Orloff and Peskin found that most cases had an identifiable pathologic or traumatic source [4].

Splenic rupture in pregnancy has been attributed to the patient's hypervolemic state, splenic enlargement, and diminished peritoneal cavity volume due to the enlarged uterus and muscular contractions during pregnancy [5]. Several authors have suggested that contributing factors to spleen rupture may include congenital malpositioning of the spleen or anatomical characteristics such as a short splenic pedicle or deeply recessed location [6]. This would predispose the spleen to trauma from a compressing diaphragm during coughing, sneezing or contractions [7].

Case Report

A 45-year-old woman was admitted to the San Salvatore Hospital of Aquila at 39 weeks of gestation, gravida 5, para 4. She had previously been successfully treated for a superficial phlebitis in her left leg and during late pregnancy had developed a mild hypertension that was treated with Methyldopa.

Two days later she gave birth to a healthy child weighing 3,800 grams. The morning after, the patient was anxious but well-orientated as to time, place, and person. She was febrile, pale, and had a mild circulatory decompensation (blood pressure of 90/60 mmHg, pulse rate 110 hr/min). There was no previous history of direct trauma and the labor was uneventful.

Physical examination revealed a distended abdomen. Tenderness was elicited in the left and right hypochondrium and in the left subcostal region on deep palpitation with no guarding or rebound tenderness. No mass could be palpated. The liver and the spleen were not palpable. Kehrs sign was positive. Bowel sounds were present with no signs of peritonitis. Blood counts were as follows: Hb 9.6 g/dl, Hct 32.8%, Plt 296 mmc, WBC 25,000 mmc, Pmn 84%, lymphocytes 7.6%, monocytes 6.4%.

X-rays showed a marked distension of the central abdominal jejunum-ileum loops and a relaxed cecum with plenty of fecal material.

An abdominal ultrasound scan of the spleen showed large, round low-density multiple areas similar to cysts, the largest of which was located in the lower pole of the spleen.

Blood tests were repeated two hours later and a fall in the hematocrit was noted: Hb 8.5 g/dl, Htc 28.8, Plt 299 mmc, WBC 17,000 mmc, Pmn 69%.

Urgent computed tomography (CT) with contrast was performed showing significant haematic effusion, probably in the organizational phase, which extended along the omentum as far as the back of the epiploon space. A significant hematic effusion seemed to affect the peri-hepatic, peri-splenic, and long parieto-colic space. A midline laparotomy was performed urgently for hemoperitoneum.

Marked bleeding in the left hypochondrium and the presence of a lesion was noted on the hilar surface of the spleen. Approximately 2,000 cc of free blood were found in the peritoneal cavity.

Splenectomy was performed. The patient was transfused with five units of blood and 600 ml of fresh frozen plasma during the operation. She had an uneventful recovery and was discharged from the hospital on the tenth post-operative day. At one month follow-up, the patient was healthy.

The histopathological examination of the spleen revealed cystic cavities of 2, 1.5, and 0.4 cm. The biggest cavity measured 17 x 10 x 4 mm. A laceration of 3 cm in length was noted on the outer surface of the spleen. Histopathologic examination confirmed capillary hemangioma as the source of hemorrhage.

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After careful questioning, trauma and delivery complications were ruled out as possible causes of the rupture, leading the authors to conclude that the rupture was spontaneous.

Discussion

Hemangiomas are vascular neoformations composed of rapidly proliferating endothelial vascular channels filled with blood cells which are usually found incidentally. Indeed most cases are of a sporadic nature although they can be inherited in a autosomal dominant pattern [8]. Splenic hemangiomas are thought to be congenital in origin [9]. They can be solitary or multiple and are frequently detected by chance during CT or ultrasounds in patients in their fourth or fifth decade of life or at autopsy. Serious complications of hemangiomas include rupture or malignant transformation [10]. Risk of rupture is thought to be increased in late pregnancy or in puerperium, especially in multiparous women, secondary to the effects of estrogens on hemangiomas. In fact, hemangiomas show immunopositivity to estrogen receptors [11]. Other factors that increase the risk of splenic rupture in late pregnancy or in puerperium are spleen enlargement, diminished peritoneal cavity, a hypervolemic state, uterus enlargement, and contractions [12]. All these factors increase the overall risk of rupture of an hemangioma which in this case occurred after labor.

Conclusion

Spontaneous spleen rupture is an uncommon event and delay in its diagnosis can lead to an increase in morbidity and mortality. The diagnosis of splenic rupture should be taken into consideration when a patient presents with sudden onset of upper abdominal pain. The clinical diagnosis can be backed up by radiology and ultrasound with power Doppler.

However magnetic resonance imaging is the most sensitive and specific means of diagnosing of splenic rupture. Without immediate splenectomy the prognosis is always fatal.

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