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Primary splenic ectopic pregnancy in a 35-year-old woman: a case report and review of literature

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Summary

Primary splenic ectopic pregnancy is quite a rare condition. It is predominantly composed of chorionic tissues. Ectopic pregnancy in the abdomen is seldom seen, accounting for fewer than two percent of all ectopic pregnancies, but for it to be in the spleen is exceptionally rare. Its etiology is very complicated. In this report the authors describe an extremely rare case of advanced splenic ectopic pregnancy in a 35-year-old woman. The clinical presentation, radiological features, and histopathologic findings are described, and the relevant literature is reviewed.

Key words: Pregnancy; Spleen; Female; Tomography; Computed X-ray; Differential diagnosis.

Introduction

Ectopic pregnancy in the abdomen is seldom seen, accounting for fewer than two percent of all ectopic pregnancies [1], but for it to be in the spleen is exceptionally rare and often misdiagnosed. It is composed of chorionic tissues predominantly. In the literature, ectopic pregnancy that occurs in fallopian tubes, cervix, ovaries, broad ligaments, peritoneum, and liver are reported [2-5]. In this case report the authors describe an ectopic pregnancy in the spleen in a 35-year-old patient.

Case Report

A 35-year-old woman presented in the present Department of Hepatobiliary Surgery with intermittent pain and distension in the left upper abdomen for half a month. She had been treated in local hospital for three days and there was no improvement. There was no history of nausea, vomiting, constipation or fever. The history also did not reveal any trauma or other cause. The woman admitted a 20-day history of a small amount of vaginal bleeding with dark red blood, with no evident outflow of granulation tissue after the last menstruation on November 1, 2015. A T-type contraceptive ring was well detected in her uterine cavity by ultrasonography, and the uterine, fallopian tubes, and ovaries were intact. There was no significant family, and past medical and surgical history. The abdomen was flat but a slightly stiff. Upper left abdominal tenderness was remarkable and rebound tenderness was obvious. The spleen was a slightly large, and there was no percussion tenderness over the kidney region. Gynecological examination revealed no evident abnormality.

CT of the upper abdomen showed an irregular, heterogeneous mass measuring 7.0×5.0×4.0 cm under the splenic capsule, containing patchy slight hyper-attenuation (Figure 1A). The lesion did not infiltrate adjacent diaphragm, intercostal muscles, and ribs, with adherent and compressed splenic parenchyma. On lobitridol-enhanced CT, the lesion demonstrated heterogeneous significant enhancement with dilated intratumoral vessels, which originated from the splenic artery during the arterial phase. A progressive contrast pattern was present in the venous and delayed phase, while the patchy slight hyper-attenuation areas still had no enhancement (Figures 1B-D). No enlarged lymph nodes could be detected in the retroperitoneum. The radiological differential diagnosis therefore included subcapsular hematoma, rupture, and hemorrhage of hemangioma, hamartoma, and arteriovenous malformation. The lesion's location, smooth outline, and defined CT characteristics excluded an abscess and malignant tumors, such as lymphoma and metastasis.

A splenectomy was performed. There was a ruptured mass with clotted blood and a tissue mixture under the splenic capsule, with adherent and compressed splenic parenchyma. The clotted blood was firstly cleared and chorionic villi were separated. Then, the mass was excised from the spleen. Finally, bleeding was arrested with gauze and the abdominal incision was closed layer by layer. The operative process was uneventful. During this surgical procedure, the spleen weighting 194.0 grams was excised. Histological examination of the specimen confirmed it to be placental villi. Chorionic tissues, hyperplastic granulation, and splenic tissues were seen under a microscope (Figure 1E). The result of pathological diagnosis was splenic ectopic pregnancy.

Five days after the surgery, no bloody drainage outflow was observed and the drainage tubes were removed. Ten days later, the abdominal CT scan suggested that no abnormal signs were present. At two-month follow-up, the patient did not experience any complaints.

Discussion

Ectopic pregnancy in the fallopian tube is very common form of extrauterine pregnancy, then in ovaries, broad ligaments, and so on, but in the abdominal cavity it is relatively rare, accounting for only 0.9% to 1.4% of the total

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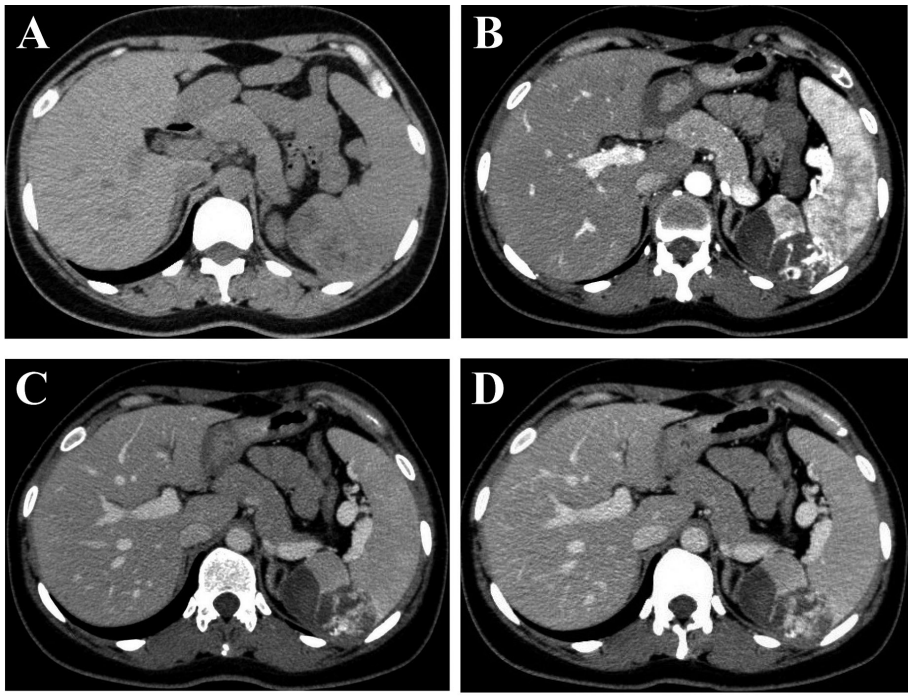

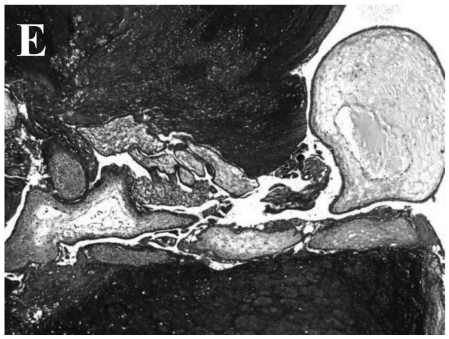

Figure 1. — A-E) CT of the upper abdomen. A) Plain-CT shows an irregular, heterogeneous mass under splenic capsule, containing patchy slight hyper-attenuation (white arrow), which is suggestive of hemorrhage. B-D) On lobitridol-enhanced CT the lesion demonstrates a heterogeneous significant enhancement with dilated intratumoral vessels in it (red arrow), which originate from the splenic artery during the arterial phase. A progressive contrast pattern is present in the venous and delayed phase, while the patchy slight hyper-attenuation areas still have no enhancement. E) Chorionic tissues, hyperplastic granulation, and splenic tissues are seen under a microscope (H&E, ×40).

number of ectopic pregnancy [2], and the maternal mortality rate can be as high as 20% [6]. This is primarily because of the risk of massive hemorrhage from partial or total placental separation due to the shortage of a blood supply [7]. In the literature, abdominal pregnancy that occurs in the liver and peritoneum are reported. In the present case, primary pregnancy in the spleen is described, which was never reported before.

Abdominal pregnancy is classified as primary and secondary; Studdiford suggested the diagnosis of primary abdominal pregnancy should be based on the following standards: 1) normal tubes and ovaries, 2) absence of an uteroplacental fistula, and 3) attachment exclusively to a peritoneal surface early enough in gestation to eliminate the likelihood of secondary implantation [8]. In the present case, the uterus, fallopian tubes, and ovaries of the patient were intact, which was considered as primary.

The presentation of patients varies and depends on the gestational age, and unexplained abdominal pain is the most common symptom. In advanced pregnancy, variable complaints can include a history of abdominal pain, persistent nausea and vomiting, and vaginal bleeding [9]. Once the gestational sac ruptured, clinical manifestation included acute abdomen, such as rebound tenderness, left upper abdominal pain, hemorrhage in abdominal cavity, and so on [4]. The present patient in this case was also marked with abdominal pain and vaginal bleeding for days. As reported, the laboratory examination showed a rise in partial hCG, but in this case, the serum of hCG was available because the ectopic pregnancy was unsuspected due to the poor cognition.

Splenic ectopic pregnancy is easily missed and mostly diagnosed after substantial emergency bleeding, which is caused by an unsecure splenic pregnancy placenta, a weak gestational sac, and the lack of protection of the myometrium [10]. The pathogenesis is unclear, but contraception, inflammation of fallopian tubes and pelvis, and profuse blood supply in the spleen may play a significant role in the development of splenic ectopic pregnancy [11, 12]. It often leads to early spontaneous abortion, causing

abdominal bleeding, and it is rare for it to develop to a late stages, as the implantation site does not involve a vascular area [13]. The present patient had been using the T-type contraceptive ring, which could be an explanation for its occurrence, and the gestational sac was ruptured for less than four weeks, which was consistent with the document reporting.

Because of the high risk of massive hemorrhage and maternal mortalities, splenic ectopic pregnancy should be terminated as soon as it is diagnosed, and laparoscopic treatment should be recommended [13, 14]. In this case, laparoscopic splenectomy was performed.

There is no clear description of the CT appearances of splenic pregnancy documented in the literature. It is easily misdiagnosed pre-operatively, excluding subcapsular hematoma, rupture and hemorrhage of hemangioma, hamartoma, and arteriovenous malformation. In this case, the authors misdiagnosed it as splenic hemangioma, due to poor awareness of the disease. The splenic ectopic pregnancy of this report showed an irregular complex-density lesion attaching to the splenic capsule on a CT plain scan, with a patchy hyper-attenuation, suggestive of hemorrhage. Heterogenous significant enhancement can be observed after intravenous lobitridol administration, whose feeding arteries originating from the splenic artery can be seen during the arterial phase.

Subcapzular hematoma is commonly encountered in patients of all age groups with a history of trauma. The lesion shows hyper-attenuation to the spleen on CT plain scan with no signs of intensification.

Hemangioma is most common tumor in the spleen with rich vascularity. It can also present hypo-attenuation on CT plain scan with peripheral nodular enhancement during the arterial phase, with persistent enhancement until the center fills with contrast, which is a distinct radiological feature that helps differentiate them from splenic ectopic pregnancy.

Hemartoma of the spleen is a rare benign tumor and occurs most commonly in adults. Spontaneously ruptured splenic hemartoma has also been reported [15, 16]. Most patients are asymptomatic and incidentally diagnosed. A plain CT could reveal a homogeneous or heterogeneous mass with calcification or fatty components, which are the typical CT features, and dense spreading enhancement and obviously prolonged enhancement on post-contrast CT [17].

Arteriovenous malformation has a low incidence in the spleen and has feeding arteries and gross draining veins, which is quite characteristic.

In summary, the authors have presented the case of a 35-year old woman with intermittent pain in the upper left abdomen. Excision biopsy of the mass-like revealed the splenic ectopic pregnancy. The differential diagnosis should include hematoma, hemangioma, hemartoma, and arteriovenous malformation.

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