A rare case of ovarian splenosis

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Abstract

Splenosis is a very rare entity that often appears following a traumatic rupture of the spleen or after splenectomy and represents heterotopic transplantation of splenic tissue. The ovary is reported as an atypical and rare localization. We report a case of a middle-aged woman, which presented with a left adnexal mass. Transvaginal ultrasonography, computed tomography (CT) and high-field 3T magnetic resonance imaging (MRI) revealed the left adnexal mass. Laparoscopy was performed, and histological and immunohistochemical examination revealed that resected mass was splenic tissue.

Keywords: ovarian splenosis, MRI, immunohistochemistry, lymphoid follicles, white pulp, macrophages.

Introduction

Splenosis was first reported in 1939 [1]. It represents heterotopic auto-transplantation of splenic tissue in case of traumatic or non-traumatic splenectomy. Its incidence is unknown, being a rare diagnosis, like in our case, and an incidental finding during surgery or autopsy. The pathogenesis of splenosis is unclear, so the development of splenic implants cannot be predicted.

The gynecological cases are even more rare and the differential diagnosis includes endometriosis, mesothelioma, abdominal lymphomas, hepatic adenomas and peritoneal metastases depending on the location [2, 3]. An important differentiation between endometriosis and splenosis is the absence of intraperitoneal adhesions in cases of splenosis. Splenosis has also a different appearance in terms of color and consistency when compared to malignant tumors.

Imaging techniques such as US (ultrasonography), CT (computed tomography), MRI (magnetic resonance imaging) play an important role in characterizing lesions and evaluate the relations with adjacent organs of the mass, but they are not specific for diagnosis of splenosis, even with the introduction of high-resolution imaging techniques [4–6]. Usually, the final diagnosis of splenosis is made by pathology and with the development of new techniques by immunohistochemistry [7].

Case presentation

A 38-year-old woman presented in the Department of Gastroenterology, Emergency County Hospital, Craiova, Romania, in July 2015, for left lower quadrant abdominal pain with recent onset (three weeks) and partial relief after non-steroid anti-inflammatory drugs administration. No association with menstrual cycle, defecation or urination was noted. The clinical history included chronic viral B and D hepatitis treated with hepatoprotective drugs. No fibrosis of the liver was revealed. She was also treated one year ago for Burkitt’s lymphoma with splenectomy. The blood tests after hospital admission were normal, including the carbohydrate antigen-125 (CA-125) value and inflammatory tests. We performed an ultrasound which revealed an isoechoic lesion in the left ovary with Doppler signal (Figure 1, a and b). The patient was referred to the imaging department where she performed 16-slices computer tomography (CT). The CT reported a left adnexal mass, isodense, with moderate contrast-uptake (Figure 2, a and b). High-field 3T MRI was also performed showing a well-lined mass in the left ovary with isosignal in T1, iso/hypersignal T2 weighted and fat suppressed sequences and moderate enhancement (Figure 3, a–c). The imaging diagnosis was benign lesion, possible adnexal fibroma. In these conditions, the patient signed an Informed Consent (including the using of all the data for scientific research and publishing) and the laparoscopic ovariectomy was proposed. The uterus and right ovary were normal on gross examination with the presence of a well-defined mass on the left ovary.
After performing laparoscopic ovariectomy, the biological material was fixed in 10% neutral formalin and sent to pathological anatomy for the microscopic examination. The material was included in paraffin, sectioned in a microtome and stained with Hematoxylin–Eosin (HE) and green light Goldner–Szekely (GS) trichrome. For the immunohistochemical examination, there were performed sections with a 3 μm thickness that were collected on poly-L-lysine covered blades, after which they were held in a thermostat at 37°C for 24 hours in order to increase the biological adherence material.

Subsequently, there was performed the deparaffinization and hydration of the histological sections, after which the biological material was passed into a 1% hydrogen peroxide solution for 30 minutes for the blocking of the endogenous peroxidase. After the section washing in tap water for 5 minutes, there followed the antigen demasking through boiling in sodium citrate pH 6 for 20 minutes, in the microwave oven. After that, there was performed the blocking of non-specific sites in 2% skimmed milk, for 30 minutes. Prepared like this, the sections were incubated with the primary antibodies for 14 hours (overnight), in the refrigerator at 4°C, and the next day, there was applied the secondary biotinylated antibody for 30 minutes, at room temperature. Subsequently, there was performed a washing of the samples in 1% phosphate-buffered saline (PBS), three baths of 5 minutes each, followed by the application of Streptavidin–HRP (Horseradish peroxidase) for 30 minutes at room temperature, followed by another washing in 1% PBS (three baths ×5
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The signal was detected with 3,3’-Diaminobenzidine (DAB) under a microscopic control, the reaction being stopped by a PBS washing when the interest structures had a maximum of staining. The contrasting was performed with Mayer’s Hematoxylin for 1–2 minutes. Then, there was performed the dehydration in ethyl alcohol in increasing concentrations up to an absolute ethyl alcohol, xylene clarification and blade fixing using the DPX (Fluka). For the immunohistochemical study, we used the following antibodies: CD20 (clone L26, sodium citrate buffer, pH 6, Dako, 1:100 dilution) for highlighting B-lymphocytes; CD3 (clone F7.2.38, sodium citrate buffer, pH 6, Dako, 1:100 dilution) for highlighting T-lymphocytes; CD68 (clone KP1, sodium citrate buffer, pH 6, Dako, 1:200 dilution) for highlighting macrophages.

The tissue histopathology exam revealed splenic tissue with white and red pulp (Figures 4 and 5) partially delineated by a fibroconjunctive septa. The splenic tissue was lined up by a fibrous capsule made up of collagen fibers orderly aligned and rare fibroblasts, similar to a splenic capsule. In some areas, and subcapsularly, there were highlighted areas of edema and microhemorrhages, a proof of an immature splenic conjunctive structure (Figure 6). In the red pulp, both in the cords of Billroth, as well as in the sinusoid capillaries, there was identified a great number of macrophage cells, with hemosiderin granules (Figure 7). We believe that this aspect may be due to a more intense process of hemodialysis that took place in the splenic ectopic tissue.

Figure 4 – Microscopic image of splenic parenchyma with white pulp (splenic Malpighi follicle) and well-represented red pulp. HE staining, ×100.

Figure 5 – Conjunctive septa made up of collagen fibers starting from the capsule that crosses an area of red pulp. HE staining, ×100.

Figure 6 – Subcapsular area with edema and microhemorrhages. GS trichrome staining, ×200.

Figure 7 – Red pulp with numerous macrophages full of hemosiderin present in the sinusoid capillaries and the cords of Billroth. HE staining, ×400.

The immunohistochemical examination was performed in order to highlight the possible architectural abnormalities of the splenic tissue. The immunomarking with the anti-CD20 antibody highlighted the presence of a very large number of B-lymphocytes in the germinative centers of the lymphoid Malpighi follicles and a smaller number of diffusely disseminated B-lymphocytes in the splenic red pulp (Figures 8 and 9).

T-lymphocytes, specifically highlighted by using the anti-CD3 antibody allowed us to highlight a larger number of T-lymphocytes placed in the perivascular lymphoid casings or diffusely disseminated in the red pulp (Figures 10 and 11).

Macrophages were found in a larger number in the clear germinative center of the splenic follicle, in the lymphoid infiltrate around the Malpighi follicles, in the cords of Billroth and in the sinusoid capillaries of the red pulp (Figures 12 and 13).
By evaluating the arrangement of lymphocytes and macrophages in the ectopic splenic tissue, we may state that we did not find an arrangement of this type of cells, in comparison to the normal splenic tissue.

The ovarian fibroconnective tissue found at the periphery of the tumor formation had a normal aspect, but the left uterine tube presented atrophic aspects and intramural fibrosis.

The final pathology diagnosis was splenic heterotopic tissue, which led to final diagnosis of ovarian splenosis.

Figure 8 – B-lymphocytes present in a very large number in the clear germinative center of the follicle. Anti-CD20 immunomarking, ×200.

Figure 9 – B-lymphocytes diffusely disseminated in the cords of Billroth of the red pulp. Anti-CD20 immunomarking, ×200.

Figure 10 – Perivascular lymphoid infiltrate (white pulp) rich in T-lymphocytes. Anti-CD3 immunomarking, ×200.

Figure 11 – Red pulp with numerous T-lymphocytes diffusely disseminated in the cords. Anti-CD3 immunomarking, ×200.

Figure 12 – Lymphoid Malpighi follicle with numerous macrophages in the germinative center. Anti-CD68 immunomarking, ×40.

Figure 13 – Heterogeneously disseminated macrophages in the cords of Billroth from the red pulp. Anti-CD68 immunomarking, ×200.
Discussion

Since the first report in 1939 [1], fewer cases of splenosis were reported in the medical specialized literature. The pathogenesis of splenosis depends on the dispersion of the splenic pulp into the peritoneal cavity, and the number of ectopic spleen nodules that develop correlates with the severity of splenic injury [8]. Another possible mechanism of splenosis suggested by the localization of splenic tissue in the brain is a hematogenous spread of the splenic pulp [9].

The incidence of splenosis in post-traumatic splenectomy is considered to be between 26% and 67%, although the true incidence is unknown, this condition being an incidental finding during surgery or autopsy [10]. Splenosis can appear after a spleen traumatic rupture and also in non-traumatic cases of splenectomy if the splenic tissue is spread on the peritoneal surface at the time of the surgical intervention or in case of an incomplete excision of the splenic tissue [11]. Similar to our case, the patient was treated of Burkitt’s lymphoma with splenectomy and presented after one year with a left adnexal mass. In some studies, from spleen trauma and the diagnosis of splenosis there can pass an average time of 19 to 25 years [2, 12]. The particularity of our case is a time of one year from splenectomy to ovarian splenosis.

The patients are usually asymptomatic but they can present with pain, like in our case. The complications are rare and include bowel obstruction, hemorrhage or torsion of the splenic implants [13].

The most frequent localization of splenosis is the abdominal cavity, the uncommon location being the genital organs as in our case of ovarian mass, the thorax and brain [14, 15].

Usually, the imaging diagnosis of splenosis is made incidentally during the ultrasonography (US), computed tomography (CT), or magnetic resonance imaging (MRI) examinations [16].

The ultrasound in our case revealed a mass in the left ovary with benign characteristics. Although multislice CT scan or high field MRI, as in our case a 3T MRI, are very useful for the diagnosis of ovarian masses, as a differential diagnosis between metastases, lymphoma or other adnexal tumors, like fibroma, could not be made. The MRI splenic implants have the same signal as normal splenic tissue with low signal on T1-weighted images and high signal in T2-weighted images. In our case, the 3T MRI showed a well-delineated ovarian mass iso/hypointense on T1-weighted images, iso/hyperintense on T2-weighted image and moderate gadolinium enhancement [17, 18]. The imaging studies that use 99mTc sulphur colloid, 99mTc-labeled heat-autologous red blood cells or 111In-labelled platelet are more specific for the diagnosis of splenosis [17, 18]. The use of super paramagnetic iron oxide (SPIO) on a MRI examination is another new specific method for the diagnosis of splenosis with ectopic splenic tissue demonstrating low signal intensity as the normal spleen after intravenous administration [19–21].

The diagnosis on a peripheral blood smear can be made by the absence of Howell–Jolly bodies and siderocytes and by the relapse of a hematological condition in the case of hematological patients with splenectomy [4].

Histologically, the splenic tissue can be undistinguishable from the parent organ because of the poorly developed architecture [7]. Several reports suggested that the tissue in splenosis is often with disturbed architecture, with no hilum or capsule. In one study, two cases were reported in which histology and immunohistochemistry was not helpful for distinguishing between splenosis and normal splenic tissue [22]. Lymph nodes and low-grade lymphoma should be taken into consideration as pathological differential diagnosis in any reactive condition [23].

The treatment of splenosis depends on the patient’s symptoms. The patients without symptoms should not be treated because of the immunological function of splenic tissue and complication after surgery, as serious bleeding and damage to the surrounding organs. In case of symptomatic patients, the treatment is resection of splenic tissue [24, 25]. The development of laparoscopic surgical techniques may increase the frequency of the splenosis diagnosis as being a minimally invasive surgical procedure, which can visualize suspicious masses, and also allows access for potential biopsy or resection [26]. At gross pathology, the splenosis differs from malignant tumors having various colors from pink to dark red/black and being bluish, with no hilum and artery supply by local arteries that penetrate their fibrotic capsule. Lack of adhesions within the abdominal cavity and their consistency can differentiate the splenosis tissue from fibroids [27].

The limit of our case is the lack of experience in diagnosis and management of patients with splenosis, especially in the case of clinical history of malignancy. As in our case, the suspected diagnosis was adnexal mass, without differential diagnosis between metastases, lymphoma or other adnexal tumor. Taking into consideration our patient clinical history of non-Hodgkin’s lymphoma and that a benign condition may mimic metastases, we performed surgery which revealed splenic tissue.

Conclusions

Our case emphasizes the rare diagnosis of pelvic splenosis, which must be kept in mind as a differential diagnosis for splenectomy patients presenting with uncertain masses, especially in malignant cases in which therapeutic management and follow-up are essential. Although advanced imaging techniques (contrast enhanced ultrasound, CT, high-resolution 3T MRI) are today available, the role of histology and immunohistochemistry in order to establish a certitude diagnosis is unquestionable.

Conflict of interests

The authors declare that they have no conflict of interests.

References


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