Splenosis of the peritoneal cavity resembling an adnexal tumor: case report

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Summary

Introduction: Splenosis is the autoimplantation of ectopic spleen tissue in various anatomic cavities of the body resulting after trauma or rupture of the splenic parenchyma. The major localization sites of this phenomenon are mainly intraperitoneal, the gastroenteric tract, genitalia, intrahepatically and the kidneys. Extraperitoneal locations occur less frequently and include the thorax and brain. Also localization in the subcutaneous fat has been described. Case report: We present the case of a 32-year-old woman with symptomatic peritoneal cavity splenosis occurring ten years after traumatic splenectomy. The patient was admitted to our department with the clinical presentation of an adnexal tumor. US and CT confirmed an adnexal mass. Exploratory laparotomy was performed and multiple focal lesions were noticed on the uterus, ovaries and intestinal tract. Biopsies were taken and sent for histological analysis. The pathology specimen revealed ectopic splenic tissue. After surgical intervention the patient remained asymptomatic. Conclusion: Splenosis is a rare phenomenon which clinicians should be aware of in order to spare patients from pointless surgical interventions. Patients with abdominal masses and post-traumatic splenectomy should be checked for splenosis.

Key words: Splenosis; Autoimplantation; Splenectomy; Adnexal tumor.

Introduction

The first description of the splenosis phenomenon in humans was made by Albrecht [1] and later by Schilling [2] in 1907. The mechanism of autoimplantation was first analyzed by von Kuttner [3] in 1910. The term splenosis was first coined in 1939 by Buchbinder and Lipkoff during exploratory laparotomy for an abdominal tumor [5]. Splenosis may present in 16-67% of patients with traumatic rupture of the spleen [6]. Approximately 100 cases have been reported universally [7]. Splenosis is a rare condition which can progress asymptomatically. Its clinical performance is not characteristic and varies depending on whether the site is intraperitoneal or extraperitoneal. It may be characterized by mild pelvic pain or may demonstrate acute abdominal manifestations in patients with abdominal masses of ectopic splenic tissue [8]. Cases involving the ileum have been described [9], as well as gastrointestinal hemorrhage following rupture of the capsule of the ectopic splenic tissue, [10] and hydrenephrosis because of amenable compaction of the ureter in the small pelvis [11].

The diagnosis of splenosis is difficult to make with the usual imaging media, such as ultrasound (US) and computed tomography (CT), giving false imaging findings which refer to neoplastic procedures [5]. Sometimes an erroneous clinical estimate leads to pointless surgical management [5]. The use of radioactive technetium-99 intravenously provides important information on the existence of ectopic splenic tissue [12].

Peripheral blood examination may show the existence of Howell-Jolly bodies which are frequently recognized in splenosis cases. These bodies are intracellular deposits in the red blood cells, nigrescent after Wright staining, and are constituted by condensate DNA of fragmented splenic cell nuclei.

Case Report

A 32-year-old woman came to the Outpatient Gynecology Department with atypical abdominal pain and accompanying symptoms of dysmenorrhea, dyspareunia and a feeling of metrorrhagia. The most remarkable fact in the patient’s history was a splenectomy, ten years before, because of traumatic rupture of the organ in a car accident. She had no gynecological or pathological history. Her menses were normal with a 28-day-cycle and duration of five days.

The patient brought the results of the US of her genitalia and CT of the upper and lower abdominal area. US revealed that the uterus was of normal size, with 9 mm thick endometrium. In the left adnexa a firm tumor measuring 35 x 40 mm was found but there was no fluid in the Douglas space. Upper and lower abdominal CT proved the absence of the spleen and confirmed the existence of a firm left adnexal tumor measuring 35 x 50 mm, with no other complementary pathological findings. The patient was hospitalized in the clinic for further investigation and management.

No pathological findings were discovered during physical examination and inspection of the external genitalia. The vagina and cervix were normal. The uterus was mobile, normal in size but mild sensitivity was found in the anatomic region of the left adnexa during the gynecological examination with both hands. The abdomen was smooth, easily compressed, with moderate pain.

The laboratory blood tests of the patient showed hematocrit 37%, Hgb 12.2 g/dl, platelets 200,000, WBC 12.5, slightly

Revised manuscript accepted for publication February 1, 2007
increased (INR 1.07). The remaining laboratory biochemical
tests were absolutely normal. Tumor markers CA 125 and CA
19.9 were within normal limits. The presurgical diagnosis was
left adnexal tumor. The patient was subjected to exploratory
laparotomy. Exposure of the abdominal cavity revealed a
normal uterus with multiple angiomatosus, blue-red lesions,
measuring up to 1 cm in the anterior surface. The adnexa pre-
sented multiple varices on both sides and were normal in
texture. On the surface of the large intestine serosa, as well as
in the greater omentum, the same lesions as those in the ante-
rior surface of the uterus were observed. Biopsy samples of
various abdominal sites were taken and sent for rapid biopsy. It
should be noted that the biopsy sampling was an extremely
hemorrhagic procedure and that hemostasis was difficult. The
frozen section results were negative for malignancy and indi-
cated the existence of ectopic splenic tissue in the sampled
lesions. Cytology lavage of the peritoneal fluid was also car-
died out and the cytological examination was negative. The
patient was not subjected to total surgical dejection of the foci and only
selective biopsy sampling was performed.

Results

The final histological diagnosis verified the results of the
rapid biopsy and determined the existence of ectopic splenic
tissue with a surrounding capsule. The postsurgical
course was normal with no symptomatology. The patient
remained in the clinic for three days and was in good con-
dition when released. Six months after the exploratory
laparotomy the patient remains asymptomatic.

Discussion

Post-traumatic splenosis of the genitalia and peritoneal
cavity is a rare clinical entity which presents characteris-
tics common to different diseases such as endometriosis,
benign and malignant adnexal neoplasms, hemangiomas
of the gastrointestinal tract, lymphomas etc. [8]. The
patient’s history should be taken under serious considera-
tion when traumatic spleen rupture or splenectomy is
noted[6]. Apart from the history, the diagnosis of spleno-
sis can be determined with the help of laboratory find-
ings, such as the presence of Howell-Jolly bodies in
peripheral blood, absence of spherocytosis and with
imaging techniques – by means of scintigraphy and using
Tc-99m tagged red blood cells [13]. Usually splenosis is
found in multiple sites, is asymptomatic and most times
is discovered accidentally [14]. Experimental studies
have shown that subcutaneous fat, the greater omentum
and peritoneum are the ideal tissues for autoimplantation
of ectopic spleens [15].

As has already been mentioned, splenosis should be
distinguished from the congenital abnormality of sec-
ondary spleens that originate from the posterior mesog-
gastrium and are found the entire length of the gastro-
plenic ligament [16], and rarely in the pancreas or in the
pelvis [17]. Splenosis of the female genital tract should
take under consideration in cases of non-diagnosed
chronic pelvic pain, as well as in those with the presence
of a pelvic mass of unknown origin in patients with
splenectomy history [6]. The main difference between
splenosis and endometriosis is that splenosis does not
create adhesions [18].

If we consult the international literature we can see that
intrapelvic splenosis may give symptomatology in the
majority of cases compared to intraperitoneal splenosis
where patients have a higher probability of not develop-
ing clinical symptoms [19].

It is impossible to predict which patients will develop
splenosis post-traumatically [8].

The immunological significance of post-traumatic
splenosis is still under dispute and study. It has been
shown that patients who have been subjected to post-trau-
tmatic splenectomy have less risk of developing infection
compared to patients undergoing selective splenectomy
due to myelodyplastic syndrome [20]. A dangerous
complication which is observed is the so-called OPSI
(overwhelming postsplenectomy infection) is rapid post-
splenectomy sepsis, with 60-80% morbidity in the first
48 hours [21]. This is due to post-traumatic autoimplan-
tation of splenic tissue resulting in the maintenance of
basic immunological functions such as phagocytosis,
antibody production against pneumococcus and
opsonophagocytosis. Studies report that roughly 20-30%
of splenic tissue is required to have an immune response
against bacteremia [20].

Complete surgical excision in patients with diagnosed
disease is contraindicated laparoscopically. In recent
years the destruction of splenosis foci has been per-
fomed with an argon laser [22]. When splenosis is diag-
nosed casually in symptomatic patients complete surgical
excision should not be performed [8].

Splenosis represents a problem of differential diagno-
sis that may often lead to confusion and cause the patient
to be subjected to pointless surgical operations [5].

Conclusions

Splenosis is a rare pathology which, in most cases, pro-
gresses asymptptomatically. Its clinical performance has
not been clarified and depends on the location. The diag-
agnosis is difficult and misleading. Imaging findings many
times lead the physician to an erroneous estimation and
treatment. The history of post-traumatic splenectomy and
accompanying pelvic pain should always be taken into
consideration. In case of pelvic pain with vague etiology,
surgical investigation should always be taken into
account. In asymptomatic patients with a diagnosis of
splenosis made casually surgical excision is contraindi-
cated [23].

In general, this is a benign disease of immunological
importance concerning the body’s defenses.

References

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toneum”. Beier z Pathol Anat u z Allg Path., 1896, 20, 513.
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