Utility of radioguided surgery in splenosis

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A B S T R A C T

Splenosis is a common finding after traumatic rupture of the spleen or therapeutic splenectomy, defined as a heterotopic autotransplantation of the spleen in peritoneal cavity and surface. In splenectomized patients due to hematologic disease, splenosis can lead to disease recurrence. We present a case of splenosis in a patient with idiopathic thrombocytopenic purpura who relapsed after splenectomy. For its localization, conventional imaging and scintigraphy with 99mTc-denatured red cells were performed, and at least five splenic foci were observed. Given the difficult intraoperative localization of these nodules, radioguided surgery was performed, with excellent localization and removal of all known nodules and multiple peritoneal implants of millimeter size that were not previously observed. We conclude that radioguided surgery is an excellent tool for locating foci of peritoneal splenosis, which have difficult access, thus avoiding early recurrence of the disease.

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Utilidad de la cirugía radioguiada en la esplenosis

R E S U M E N

La esplenosis es un hallazgo frecuente tras la rotura traumática del bazo o esplenectomía terapéutica, definiéndose como un autotrasplante heterotópico del bazo por la superficie y cavidad peritoneal. En pacientes esplenectomizados por enfermedad hematológica, la esplenosis puede llevar a recidiva de la enfermedad. Presentamos un caso de esplenosis en un paciente con púrpura trombocitopénica idiopática que presentó una recidiva tras esplenectomía. Para su localización se realizaron pruebas de imagen convencional y gammagrafía con hematíes desnaturalizados marcados con 99mTc, donde se apreciaron al menos 5 focos esplénicos. Dada la difícil localización intraoperatoria de dichos nódulos, se practicó cirugía radioguiada, con excelente localización y extirpación de los nódulos conocidos y de múltiples implantes peritoneales de tamaño milimétrico no visualizados previamente. Concluimos que la cirugía radioguiada es una excelente herramienta para la localización de focos de esplenosis peritoneal de difícil acceso y visualización, evitando así una recurrencia precoz de la enfermedad.

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Introduction

Splenosis is a frequent finding in 26–67% of the patients with traumatic rupture of the spleen or following therapeutic splenectomy. It is defined as a heterotopic autotransplantation of the spleen on the surface of the peritoneum and peritoneal cavity. Unlike accessory spleens, splenosis is an acquired condition and may be distributed anywhere in the abdominal cavity. Extra- as well as retroperitoneal localizations and those of the lung or abdominal wall are rare and are usually caused by intraoperative or traumatic implantation of splenic tissue.1,3

The number of splenic nodules varies from a few to hundreds and their size ranges from a few millimeters to several centimeters.

In patients undergoing elective splenectomy due to hematologic disorders such as idiopathic thrombocytopenic purpura (ITP), splenosis may lead to disease recurrence. In these cases the localization of the active splenic tissue by scintigraphy is essential to allow surgical treatment.4 Differentiation between tumor masses and splenosis is equally necessary since it may simulate lymphomas or peritoneal metastasis which cannot be exclusively differentiated with the conventional imaging techniques (CT, MR, ultrasonography).

Identification of the splenic tissue is performed by scintigraphy with heat-denatured red cells which are phagocyted by Kupffer cells of the reticuloendothelial system.

Radioguided surgery has been used in numerous contexts and is based on the use of an intraoperative probe for the detection of tissue marked with the radiotracer. The main and most extensive application of this approach is in the detection of the sentinel lymph node in oncologic disease. The idea of using radioguided surgery in this case arose from the difficulty in intraoperatively detecting the implants, considering the uncertainty regarding their exact number, localization and small size. Its application in splenosis is related to the detection of gamma radiation in the splenic implants by
an intraoperative probe after the injection of denatured red cells labeled with $^{99m}$Tc. To our knowledge there are no studies on the use of radioguided surgery for this application.

We present a clinical case of splenosis of difficult localization in which radioguided surgery was successfully performed.

**Clinical case**

A 15-year-old male was diagnosed with ITP in 2000 at the age of 3 years, being initially treated with prednisone and cyclosporine. The patient presented exacerbations of the disease in 2003 and 2004 and was admitted to the emergency department for hematomas, epistaxis and gingivohemorrhage with brushing with no other clinical manifestations of active bleeding. Complementary tests always showed frank thrombocytopenia (4000–15,000/mm$^3$).

In 2005, the patient presented an exacerbation resistant to medical treatment (corticoids, intravenous immunoglobulin and cyclosporine) and was admitted for severe thrombocytopenia (12,000/mm$^3$) and the appearance of spontaneous hematomas. Pharmacologic treatment was administered (intravenous antiD immunoglobulin) with no response, leading to the consideration of splenectomy.

Due to the torpid evolution of the disease uneventful splenectomy was performed by laparoscopic surgery, maintaining adequate hematologic levels following surgery. The patient remained in complete remission since then.

In 2012 the patient was attended for severe thrombocytopenia (10,000/mm$^3$) and initiated new treatment with unspecific immunoglobulin, achieving good response with an increase in the platelet count of greater than 50,000/mm$^3$ at 24 h.

An abdominal ultrasonography was performed showing a single nodule in the left hypochondrium possibly related to a small accessory spleen in a splenectomized patient. Given the important symptomatology presented which could not be explained by the presence of a single small size spleen, the study was complemented with splenic scintigraphy.

Scintigraphy was performed with denatured red cells labeled in vitro with $^{99m}$Tc by intravenous administration of tin pyrophosphate and posterior extraction of 8 ml of blood to which 10 mCi of $^{99m}$Tc was added for the labeling of the red cells which were thereafter heated in a water bath at 49°C during 15 min for denaturation.

One hour after the reinjection of the denatured red cells, anterior and posterior planar images were obtained as well as left lateral planar images of the thorax and abdomen with a ADAC Vertex (Philips) gamma camera, showing the presence of at least 5 pathologic foci of rounded appearance distributed along the left abdominal flank from the lateral region to the iliac fossa along the paracolic gutter (Fig. 1).

The MR confirmed the presence of several small sized pseudo-nodular lesions (largest being of 3 cm at its greatest axis) in the left paracolic gutter-left iliac fossa in the localization described in the previous scintigraphy.

The scintigraphy with denatured red cells was repeated 25 days after the diagnosis in the same conditions previously described after which an exploratory laparotomy was performed with resection of splenic nodules guided by a gamma probe.

Surgery was carried out 2 h after the reinjection of the denatured autologous red cells labeled with $^{99m}$Tc, finding a total of 33 splenic nodules (6–7 nodules of a size between 1 and 4 cm in diameter and multiple subcentimetric sized implants) (Fig. 2).

The implants were localized on the posterior parietal peritoneum and above the intestinal loops. On measurement with the probe, very much elevated values of counts/second (c/s) were obtained in the splenic nodules (2000/2500 c/s in the largest fragments and of 300 c/s in the subcentimetric sized implants). On finalization, the abdominal cavity was revised by quadrants, only finding background activity in the hepatic and renal territory.

The histological results of the samples confirmed the diagnosis, and splenic remnants were not observed in the postoperative CT scan.

**Discussion**

The treatment of ITP is essentially medical. Lack of response to this treatment constitutes the principal indication of splenectomy in these patients, with good results in up to 85% of the patients.\(^4\)
Recurrence following splenectomy may be due to splenosis and thus, specific imaging techniques should be performed to localize the new foci.

Scintigraphy with denatured $^{99m}$Tc-red cells is a reliable and easily applicable, non invasive technique with a high positive predictive value and is highly specific and profitable for the localization of splenic tissue. Nonetheless, this technique has difficulty in determining the precise anatomical localization of the foci of splenosis and since this site usually involves peritoneal seeding of numerous small implants which are difficult to localize in the surgical field, it is necessary to use a technique which helps to achieve their detection during surgery.

Radioguided surgery is routinely used to locate a tissue which selectively accumulates a radiotracer administered by a gamma detector and to thereafter perform precise resection of this tissue.

The use of radioguided surgery has recently been of great utility in the definitive identification of parathyroid adenomas which are difficult to localize, achieving clear advantages compared to conventional surgery, allowing reduction of the incision size, surgical time, postoperative pain and the global costs. One study carried out by McGreal et al. reported a success rate of 97% in the localization of parathyroid adenomas by radioguided surgery after the administration of $^{99m}$Tc-MIBI.

Radioguided surgery seems to be a technique with great utility in the localization of foci of splenosis, with no studies comparing recurrence in conventional surgery with radioguided surgery. This procedure is quick and easy to perform and allows a considerable increase in surgical precision. Similar to other radioguided surgical procedures (biopsy of sentinel lymph node, parathyroidectomy), success in the global result requires a close relationship and coordination between surgeons and nuclear physicians.

Conclusion

The performance of splenic scintigraphy together with the use of an intraoperative detection probe is a very useful technique for the localization of peritoneal splenosis in splenectomized patients, allowing intraoperative detection and probably avoiding recurrences.

Conflict of interest

The authors declare no conflict of interest.

References