Case report

Littoral cell angioma of the spleen: CT and MR imaging appearance

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Abstract. We report a case of littoral cell angioma (LCA) of the spleen, a recently described splenic pathology, which imaging characteristics and pathologic morphology have been discussed only by a few authors. The imaging findings in unenhanced and contrast-enhanced MRI and CT as well as histologic specimen are presented. Diagnosis was made after elective splenectomy. Differential diagnosis of splenic tumors as well as the imaging findings in this particular case are discussed.

Key words: Spleen – Tumor – CT – MRI

Introduction

The most common primary neoplasms of the spleen are vascular tumors [2, 6, 18, 27] which occur mostly as benign hemangiomas [16, 19], followed by malignant vascular tumors such as hemangioendotheliomas whose incidence seems to be much lower [5, 18, 20, 27]. Other rare primary neoplasms of the spleen include splenic lymphangiomas, lymphangiosarcomas, or vascular hamartomas of the spleen [3, 4, 6, 9, 18, 21, 23, 24, 26, 27]. In 1991 a novel splenic vascular lesion, littoral cell angioma (LCA), was described [4] as a benign splenic vascular tumor derived from splenic sinus lining cells. Their clinical and morphologic features have already been reported, but according to our knowledge there are only few published data about radiologic imaging of these rare splenic lesions.

In this case report we describe the CT and MR imaging appearance of an LCA of the spleen, incidentally found in a patient with malignant melanoma.

Case report

A 66-year-old man suffering from malignant melanoma of the skin (Clark level IV) was transferred for surgical treatment. Primary location of the tumor was the back.

During clinical staging CT examinations of the thorax and abdomen were performed. As the CT scans of the abdomen revealed multiple hypodense splenic masses (Fig. 1), the patient was referred to an MRI study of the upper abdomen for further evaluation of this pathology.

Methods

Routine CT was performed as conventional incremental CT on a Somatom Plus scanner (Siemens, Erlangen, Germany) in 8-mm sections (120 kV, 210 mA) before and during contrast media administration (120 ml Ultravist, flow 3 ml/s, Schering, Berlin, Germany).

Magnetic resonance imaging was performed twice, both examinations on a 1.5-T superconducting clinical imager (Magneton Vision, Siemens, Erlangen, Germany) using a semiflexible cp-array coil. T1- and T2-weighted breathhold sequences were basically performed in the transverse plane (6-mm sections, 1.25-mm interslice gap). For T2-weighted imaging we used a turbo spin-echo sequence (TSE) with TR/TE of 3200/138 ms, a flip angle of 180°, and an echo train length of 29. Eleven slices can be performed in one breathhold of 17 s; thus, two separate sets have to be obtained to cover the entire upper abdomen. T1-weighted imaging was done with a 2D fast low-angle-shot (FLASH) sequence (TR/TE = 174.9/4.1 ms, flip angle 80°, 23 slices) with an acquisition time of 19 s. Additionally, GRE T1-weighted fat-suppressed images (TR/TE = 155/4.8 ms, flip angle 75°, five slices) were used.

In a first examination we applied gadopentatate dimeglumin (Magnevist, Schering, Berlin, Germany) to perform dynamic studies. The contrast medium was administered intravenously as a bolus (0.1 mmol Gd-
DTPA/kg bodyweight, flow 2 ml/s) directly followed by a flush of 20 ml sodium saline using the same flow rate. Bolus injection was performed with an automatic MR power injector (Injektron 82 MR, Medtron, Saarbrücken, Germany). The dynamic MR protocol includes an early phase T1-weighted scan with a delay of 20–25 s after start of the contrast medium administration followed by a second scan with a delay of 50–55 s. Additionally, delayed phase scans were obtained 2 and up to 15 min later.

In a second examination we used SPIOs as contrast medium (15 mmol/kg bodyweight, Endorem, Guerbet, Aulnay-sous-Bois, France) and obtained T2-weighted TSE scans before and 60 min after contrast medium administration.

**Imaging findings**

On unenhanced CT scans multiple well-circumscribed hypodense lesions were found. After contrast medium administration, there was no significant increase in density (Fig.1).

In unenhanced T2-weighted MRI (Fig.2a), the splenic lesions showed a high signal intensity (SI), comparable to what is found in hemangioma of the spleen. Unenhanced T1-weighted images depicted the lesions as slightly hypointense masses (Fig.3a), and on early contrast-enhanced images the lesions were clearly hypointense with inhomogeneous contrast media uptake (Fig.3b,c). On delayed T1-weighted images, applied 15 min after contrast media administration, the masses showed homogeneous contrast media uptake, now appearing hyperintense in comparison with surrounding splenic tissue (Fig.3d), indicating a vascular lesion with contrast media pooling. Contrast media retention in splenic lesions was even better visualized on T1-weighted fat-suppressed images (Fig.3e) 15 min after injection of Gd-DTPA.

Sixty minutes after administration of SPIO, T2-weighted images showed a decrease of signal intensity in parts of the lesions (Fig.2b) most likely due to contrast media uptake in macrophages, although most of the lesions did not show a significant change of SI. No focal liver lesions and no lymphoma could be detected in the upper abdomen. Differential diagnosis of splenic lesions included multiple atypical hemangiomas, diffuse low-grade hemangiosarcoma, or hemangiendothelioma as well as metastases of malignant melanoma with atypical appearance.

Due to the generally good conditions of the patient and the uncertain nature of splenic lesions, elective splenectomy was performed. The pathologic, especially immunohistochemical features revealed an LCA. Up to date, 18 months after treatment, the patient is in good clinical condition, without complications concerning splenectomy and follow-up investigations, performed in a 6-month interval, did not reveal any recently developed metastases of malignant melanoma.

**Pathologic characterization of the splenic lesions**

Macroscopic examination of the surgical specimen revealed multiple, well-circumscribed blood-filled spongy nodules between 2 and 30 mm replacing the normal splenic tissue. These nodular lesions were composed of markedly dilated, cavernous blood vessels and a complex network of narrow vascular channels (Fig.4) forming altogether a mesh of vascular and solid tissue, reflecting well the imaging findings. At high magnification, the channels were lined by a simple layer of endothelial cells, which attained a pseudopapillary pattern in some areas (Fig.5). The luminal spaces contained exfoliated cells. The lining cells expressed endothelial markers such as factor-VIII-related antigen, CD 31 and CD 34 (Fig.6). Nuclear abnormalities and atypical mitosis were absent. Infrequent mitosis were observed. These features correlate to those previously described [1, 4] for LCA.