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Case Report

Splenic hamartoma associated with abdominal discomfort and pain: Case report

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ABSTRACT

Hamartomas are benign splenic neoplasms asymptomatic in most of the cases. Symptoms, when present, may either be related to the growth of the mass with abdominal discomfort and pain or be related to a hypersplenism syndrome. Certain preoperative diagnosis cannot be made with current diagnostic imaging. Splenectomy is therefore indicated in order to obtain histological diagnosis, rule out malignancy or achieve regression of symptoms. We report the case of a 39-year-old woman referred for a splenic hamartoma causing pain located on the upper abdominal quadrant. She underwent splenectomy through a left subcostal access followed by complete resolution of symptoms.

Resection of splenic masses is indicated to complete diagnosis, achieve cure and, when present, relieve symptoms.

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1. Introduction

Hamartomas are rare, benign tumors of the spleen [1,2] and their exact, preoperative diagnosis is difficult [3]. They are asymptomatic in most of the cases, nonetheless, as they may hide a malignant lesion or progress towards symptoms of compression on other structures, pain, hypersplenism or rupture, their resection is usually warranted [4].

In line with the SCARE criteria [5], we now report the case of a splenic hamartoma associated with left upper quadrant abdominal pain, treated by splenectomy followed by resolution of symptoms.

2. Presentation of case

A 39-year-old woman presenting a non-specific left upper quadrant pain lasting since a month underwent an abdominal ultrasound examination (US) which disclosed a 4 cm diameter, hypoechoic mass of the upper pole of the spleen with inner blood flow at Power Doppler. She was, then, addressed to our Department for treatment. At physical examination the lower pole of the spleen was palpable and non-tender. Laboratory examinations were all

within normal limits. CT-scan revealed a 4.5 cm diameter, round mass of the upper pole of the spleen, iso-dense at non-contrast CT (Fig. 1), with homogeneous contrast enhancement on the arterial phase and mild, late enhancement on the portal phase (Fig. 2). The spleen with the mass appeared to compress the posterior wall of the stomach and the anterior surface of the kidney without infiltration (Fig. 3). On 3-D reconstructions, the mass appeared well round and fed by an upper hilar branch of the splenic artery, almost mimicking an aneurysm (Fig. 4). A selective, splenic artery angiogram showed the main trunk of the splenic artery dividing at the hilum with an upper and an inferior branch. The mass was fed by the upper branch but also by multiple, collateral feeding vessels thus hindering a satisfactory and complete embolization of the mass without massive necrosis of the spleen (Fig. 5). The decision was to proceed with a splenectomy, after obtaining patient's informed consent for open surgery. The operation was performed by the senior author (GI), through a left subcostal incision and was carried on in a standard fashion, with separate ligation of the splenic artery and vein (Fig. 6). The mass was well capsulated and arose in a context of splenomegaly, with a major axis of the spleen of 15 cm (Fig. 7).

Postoperative course was uneventful and the patient was discharged home on postoperative day 5. At one-month follow-up, left upper quadrant abdominal pain completely regressed.

Histology on the resected specimen was consistent with the diagnosis of splenic hamartoma (Fig. 8).

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Fig. 1. CT-scan of the abdomen showing an iso-dense mass of the upper pole of the spleen.

3. Discussion

Hamartomas are rare benign tumors of the spleen consisting of aberrant splenic tissue [4] and were first reported by Rokitansky in 1861 [6]. They may arise from the splenic pulp, lymphoid splenic tissue or may have a mixed pattern of origin [4]. Their frequency in

splenectomy specimens ranges from 0.015 to 2.7% [4] and 0.024-0.13% in autopsy specimens [7]. Overall, slightly less than 200 cases should have been reported so far [2,8] They may affect any age group, with equal incidence in men and women [1], although women may tend to have larger lesions than men, probably due to hormonal factors [1,9]. When affecting children they usually become symptomatic when associated with multiple hamartomatosis or isolated and still of a smaller diameter compared to adults [10-14]. In most of the cases they are asymptomatic and may be detected at abdominal US examinations performed for search of other conditions. However, sometimes and usually when reaching large diameters of around 8 cm and more in adults or when multiple, they may exhibit symptoms of hypersplenism with thrombocytopenia, anemia and pancytopenia [1,8]. They may also be associated with multiple hamartomatosis, tuberous sclerosis and with Wiskott-Aldrich-like syndromes and Kasbach-Merrit syndrome [8,15]. They may also cause symptoms of abdominal discomfort and pain due to compression on adjacent organs or simply to mass effect as in the reported case or cause rupture of the splenic capsule and bleeding [8].

As an exact preoperative diagnosis can seldom be established [2], their resection is usually always indicated, as they may turn out to be malignant lymphomas or Hodgkin's disease [4]. Further differential diagnosis with other benign and malignant conditions affecting the spleen include vascular tumors such as hemangioma, hemangiopericytoma, lymphangioma, angiosarcoma and thrombosed hilar aneurysm of the splenic artery [1,16,17]. Hamartomas should also be

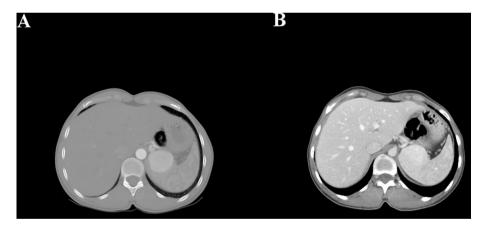


Fig. 2. CT-scan of the abdomen: after contrast injection the splenic mass presents a homogeneous enhancement on the arterial phase (A) and a light enhancement on the portal phase (B).

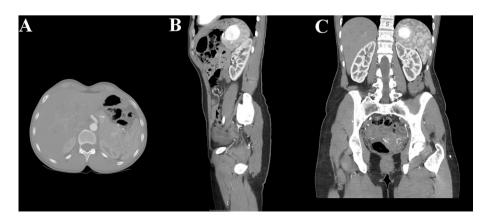


Fig. 3. CT-scan of the abdomen. On both transverse (A), lateral sagittal (B) and anterior sagittal (C) reconstruction splenomegaly and mass compress the anterior face of the upper pole of right kidney and posterior wall of the stomach without infiltration.

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