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Case Report

Littoral Cell Angioma of Spleen: A rare case report

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ABSTRACT

Introduction: Spleen's Littoral Cell Angioma is a rare benign vascular tumor. The main clinical presentation is isolated splenomegaly.
Case presentation: we present the case of a 37 years old male patient who suffers from chronic pain in the left hypochondrium.The abdominal examination finds a painful splenomegaly related to a biological bicytopenia. The CT Scan shows a 32 cm splenomegaly.An exploratory laparotomy with splenectomy was performed. The histological and immunohistochemical study confirmed The final diagnosis of Littoral Cell Angioma.
Clinical discussion: Isolated splenomegaly of unknown etiology is the main clinical sign. Splenectomy is required for a diagnostic purpose and substratum for histological study.
Conclusion: Littoral Cell Angioma of the Spleen is a rare benign vascular tumor, however it should be highlighted by clinical and radiological features, the definitive diagnosis is made upon histological study.

1. Introduction

Splenic Littoral Cell Angioma is a rare benign vascular tumor that grows at the expense of the littoral cell of the sinuses of the red pulp of the spleen [1]. The clinical presentation is non specific.Isolated splenomegaly of unknown etiology is the main clinical presentation.the discovery of hypersplenism is possible.the splenectomy is almost required for the diagnosis.

The diagnosis of certainty is based on histological study [2]. Through this case report we highlight the main clinical, histological, and therapeutic characteristics of this rare clinical entity. This case has been reported following the SCARE guidelines [3].

2. Case presentation

A 37 years old male patient, consulting in the internal medicine department for the management of a splenomegaly, he had no significant pathological history and he suffers from chronic pain in the left hypochondium.without any other digestive symptoms. The clinical examination showed an isolated splenomegaly, without associated hepatomegaly related to the collateral veinous circulation, or lymph nodes or ascitis. The blood counts showed a thrombopenia of 32000 g/l (normal range 150000–500000g/l). Normocytic normochromic anemia at 6.6 g/dl (normal range 10–16 G/L), mean corpuscular volume of 74 fl (normal range 82–98 fl),a mean corpuscular concentration of hemoglobin of 26 (normal range 28–32). The C-protein reactive was 11mg/l (normal range is less than 5mg/l). The liver function tests was normal and the serology of hepatitis was negative.An abdominal ultrasound shows a splenomegaly with multiple hyperechogenic lesions.A computed tomography (CT) shows a spleen increased in size by 32 cm with multiple nodular lesions containing calcifications measuring 37 a 38 mm in lower polar and 50*31 mm in medio-splenic with a permeable portal vein (Fig. 1). The upper fibroscopy did not show any signs of portal hypertension.The case was discussed in a multidisciplinary reunion and a splenectomy with a liver biopsy was performed (Fig. 2) with an overall stay of four days.

The histological examination was in favor of a littoral cell angioma (Figs. 3 and 4).

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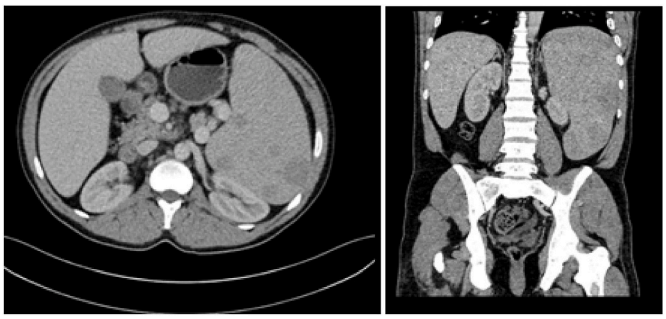

Fig. 1. CT images show multiple splenic lesions.

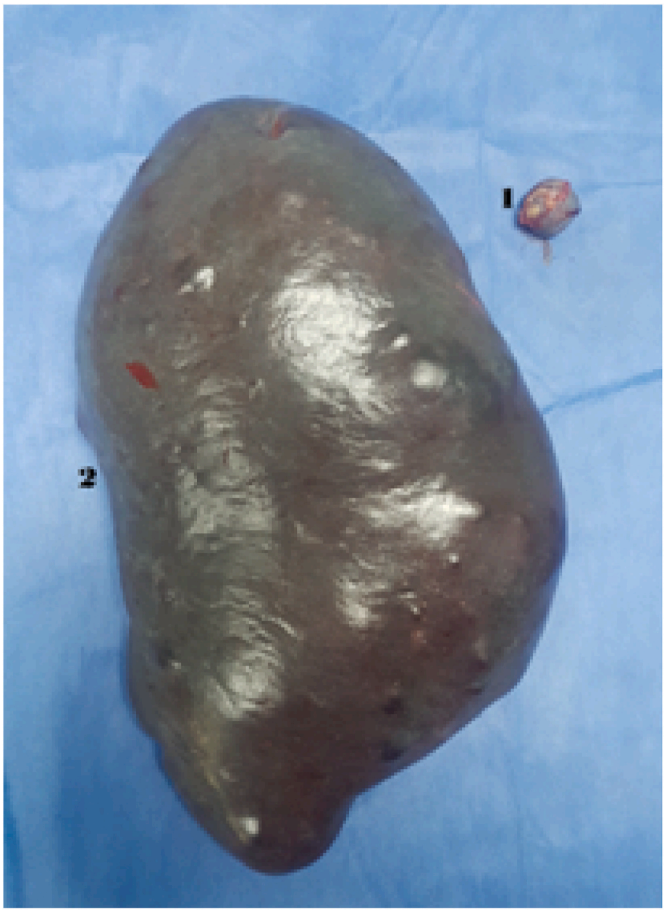

Fig. 2. Image showing 1: liver biopsy 2: splenectomy.

The patient was afterward transferred to the internal medicine department for follow-up.

3. Discussion

Littoral Cell Angioma of the Spleen is a rare vascular tumor, it can be a single lesion or multiple lesions of small size [4].

This disease was described for the first time in 1991 by FALK and al [5].it affects men and women regardless of de age of the patient [6].

Usually asymptomatic, sometimes the revelation can be fortuitous or linked to an abdominal distension, pain, or splenomegaly [10].

The biological diagnosis is poor. It shows sometimes a hypersplenism [6]. A Littoral Cell reported in the literature describes a bulky limited and heterogeneous masses [7–9], sometimes it can be presented with complications due to the voluminous and compressive character of

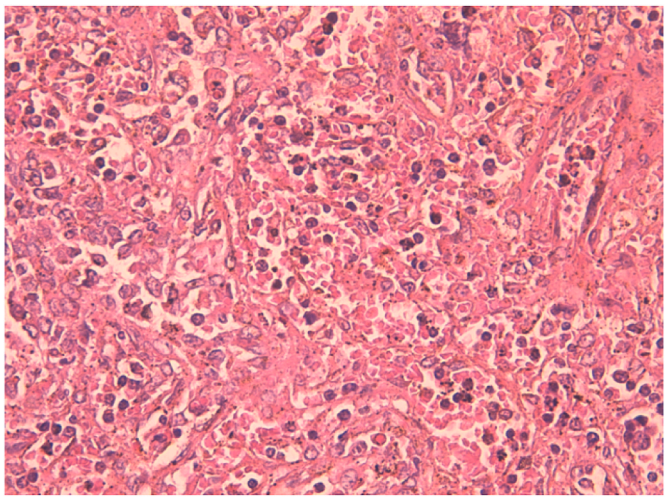

Fig. 3. The vascular spaces are lined by plump cells with the appearance of sinus lining ("littoral")cells. They show nuclear enlargement with no atypia or necrosis.

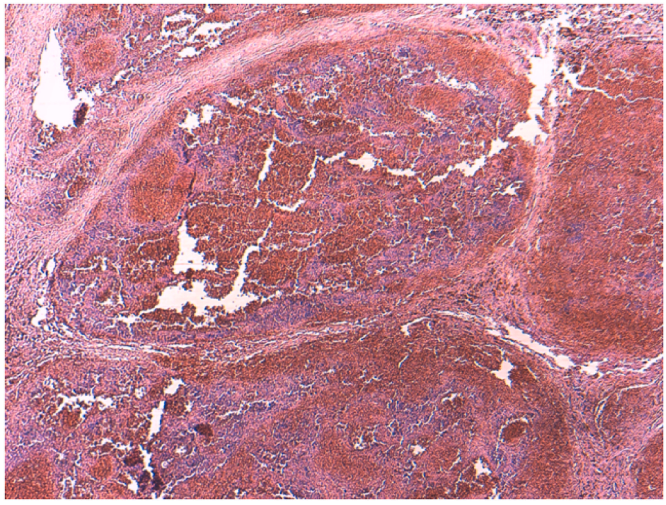

Fig. 4. Multiple splenic nodules made of anastomosing, tortuous and blood-filled vascular channels.

Angioma such as a portal hypertension by the compression of the splenic vein [8].

The unusual aspect of imaging mimicks other splenic pathologies and don't allow a precise diagnosis.however, it helps reduce assumptions of tumoral or infectious diseases.

The splenectomy is almost required for the diagnosis in the case of clinical impact or acute complications of splenic angioma.

The proved diagnosis is histological (2). and characterized by a proliferation composed by vascular space of varied size, interconnected, containing long papillary projections. The immuno-histochimistry of these tumors is very particular combining a double vascular and histiocytic phenotype (CD 31 and CD 68, KP1) and the basal component express CD 34 while the big cells are negative the CD 21 is sometimes positive the CD 8 is negative [2].

4. Conclusion

Littoral Cell Angioma of Spleen is a rare benign vascular tumor; however it should be highlighted with clinical and radiological features. The definitive diagnosis is based on the histological study.

Ethical approval

No ethical approval necessary.

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Author contribution

Dr Bouzayan Laila: Have written the article, have consulted the patient, and participated in the surgery.

Pr Jabi Rachid: supervised the writing of manuscript.

Dr Mabrouk Mohamed Yassine: have helped writing the article, data collection.

Dr Madani Ayoub: supervised the writing of the manuscript.

Dr GUELIL Abdelali: supervised the writing of the manuscript.

Dr Haloui Anas: Interpretation of histological data.

Dr ABOU WIDAD: Interpretation of radiology.

Pr SKIKER Imane:(radiology professor): confirm the radiology Interpretation.

Pr BenaniAmal (anatomopathology professor): confirm the histological diagnosis.

Pr Bouziane Mohammed (oncology surgery professor): have supervised the writing of the paper, and has been the leader surgeon of the case.

Trial register number

Not available.

Guarantor

Dr Bouzayan Laila,

Consent of the patient

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

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Declaration of competing interest

The authors declared no potential conflicts of interest concerning research, authorship, and/or publication of the article.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103904.

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