



Littoral Cell Angioma of the Spleen in a Patient with Metastatic Abdominal Leiomyosarcoma. Case Report of Rare Tumor and Review of the Literature

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Abstract

Primary splenic neoplasms are rare and among these neoplasms are littoral cell angiomas (LCA). LCAs are benign vascular tumors arising from the littoral cells lining the splenic red pulp sinuses. LCAs are most often identified incidentally, although patients may present with hypersplenism such as anemia, thrombocytopenia, and splenomegaly. The incidence is found to be equal in both males and females and most reported cases are in middle aged adults. Most cases of LCA described in the literature have been composed of multiple lesions of varying size in the spleen. The pathogenesis of LCA is unknown; however the neoplasm is associated with immunological disorders and malignancies. We report a case of a patient diagnosed with a LCA of the spleen in association with metastatic abdominal leiomyosarcoma, along with a literature review.

KEYWORDS: Littoral cell, Endothelial, Red pulp, Angiosarcoma, Benign

ABBREVIATION

LCA: littoral cell angiomas, **AS:** Angiosarcoma, **IHC:** Immunohistochemistry

INTRODUCTION

Primary tumors arising from the spleen are uncommon. They are classified as lymphoid, non-lymphoid, and tumors like lesions. Within the non-lymphoid tumors, vascular neoplasms are the most common. LCA arises from vascular elements that form the splenic red pulp, while lymphoid neoplasms arise from the splenic white pulp. In regard to vascular tumors of the spleen, the biologic behavior can be both benign and malignant [1]. Littoral cell angioma (LCA) of the spleen is a particularly rare vascular tumor that was first described by Falk et al. in 1991. LCA arises from the littoral cells lining the splenic red pulp sinuses. LCA affects both men and women equally with no specific age correlation [2]. Commonly, it is asymptomatic and is discovered incidentally. LCA may also present with a myriad of possible signs and symptoms, such as: splenomegaly with or without abdominal pain, hypersplenism with anemia or thrombocytopenia. More dramatically, LCA has been reported to present as splenic rupture and hemoperitoneum [3].

The origin of LCA is generally unknown; however there are suggestions of immune dysregulation as a hypothesis due to the reported association with Crohn's disease [4]. In addition, several reported cases of LCA have been associated with inflammation and other tumors [5]. LCA was first described as benign but further investigations indicate there is commonly association with other visceral malignancies and even malignant forms of LCA [6]. Diagnosis of LCA can be challenging as it mimics other neoplasms of the spleen clinically and on imaging [2]. The current management of LCA is long term follow-up due to the malignant potential of the neoplasm and the frequent association with other malignancies as in our case [7]. Splenectomy is commonly the preferred treatment of the neoplasm. We present a case of a patient with LCA in the spleen associated with the recurrence of metastatic abdominal leiomyosarcoma.

CASE PRESENTATION

A 57-year-old woman presented with abdominal pain and swelling of her lower abdomen. She also complained of weakness, epigastric distress, weight loss, nausea, vomiting, and occasional upper gastrointestinal tract bleeding. Patient reported history of high grade localized uterine leiomyosarcoma two years prior to current presentation. FNA cytology studies and biopsy sampling confirmed the diagnosis of abdominal wide-spread metastatic leiomyosarcoma. A multidisciplinary tumor board meeting recommended extensive debulking surgical removal of all abdominal tumor masses to be followed by Adriamycin-based chemotherapy. Surgical debulking specimens included; segment of jejunum, segments of left lobe of the liver, spleen, distal pancreas and segment of the diaphragm, in addition to segments of ileum. All organs were involved by metastatic leiomyosarcoma except for the pancreas and the spleen.

An incidental two small splenic lesions measuring 1.3 cm and 0.6 cm were grossly identified displaying features different from other abdominal tumor masses. The cut surface of these lesions showed relatively circumscribed, spongy, blood filled cystic

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spaces separated by fibrous septae (**Figure 1A**). Microscopic examination displayed proliferation of anastomosing, tortuous, blood filled vascular channels with irregular lumina forming papillary projections and cystic spaces. The vascular spaces were lined by non-atypical tall bland endothelial cells with sloughing of endothelial cells into the vascular spaces. With absence of nuclear atypia or necrosis (**Figure 1 B-C**). Angiosarcoma was ruled out and a benign vascular lesion was favored. Immunohistochemistry (IHC) studies were utilized for definitive diagnosis. The endothelial lining cells were positive for CD31 (**Figure 1D**), CD68, Factor VIII, and vimentin. The cells were negative for Pan Cytokeratin (**Figure 1E**), CD34 (**Figure 1F**), and HHV8. The cells showed low Ki67 proliferation index. The histomorphology, together with the IHC studies were diagnostic of Littoral cell angioma.

Patient received Adriamycin-based chemotherapy as a post-operative treatment and was free of recurrence or metastasis for 18 months after which she expired due to extensive metastatic leiomyosarcoma to the bone, liver, and lung unrelated to littoral cell angiomata.

DISCUSSION

LCA, a benign vasoformative tumor, originates from the sinus lining endothelial cells, known as littoral cells, of the splenic red pulp. The littoral cells are considered to be unique lining cells possessing both vascular endothelial and macrophage-like properties [8]. However, littoral cells display higher lysosomal activity than vascular endothelial cells [9]. These cells can

undergo neoplastic proliferation resulting in LCA of the splenic red pulp. The pathologic diagnosis of LCA is based on neoplastic proliferation of anastomosing vascular channels rimmed by cells reacting immunohistochemically to both endothelial cell and macrophage markers [10]. LCA can present clinically with a broad array of symptoms ranging from asymptomatic to findings of abdominal pain, splenomegaly, and hypersplenism [11]. Patients with LCA less commonly present with clinical features of anemia, thrombocytopenia, hepatitis, cirrhosis, portal hypertension, and extramedullary hematopoiesis [12]. Although first described as benign, LCA has recently been shown to exhibit malignant potential and is increasingly reported in association with other visceral malignancies as in the current case [5]. Grossly, the cut surface of the spleen shows dark red, brown, or black nodules of blood or blood products [13]. Histopathologically, LCA is characterized by the presence of anastomosing vascular channels, which are lined by elongated endothelial cells and papillary fronds extending into the vascular channels.

The etiology of LCA remains mostly unknown, however it is postulated that immune dysregulation may play a role as it is commonly reported in patients having history of other tumors or inflammation. LCA is distinct from other splenic vascular lesions such as angiosarcoma, due to presence of both endothelial and histiocytic pattern of differentiation [7].

There are several differential diagnoses of lesions that mimic LCA clinically and in imaging. These lesions include angiosarcoma, lymphangioma, hamartoma, lymphoma, Kaposi's sarcoma, hemangioma, and littoral cell angiosarcoma [14].

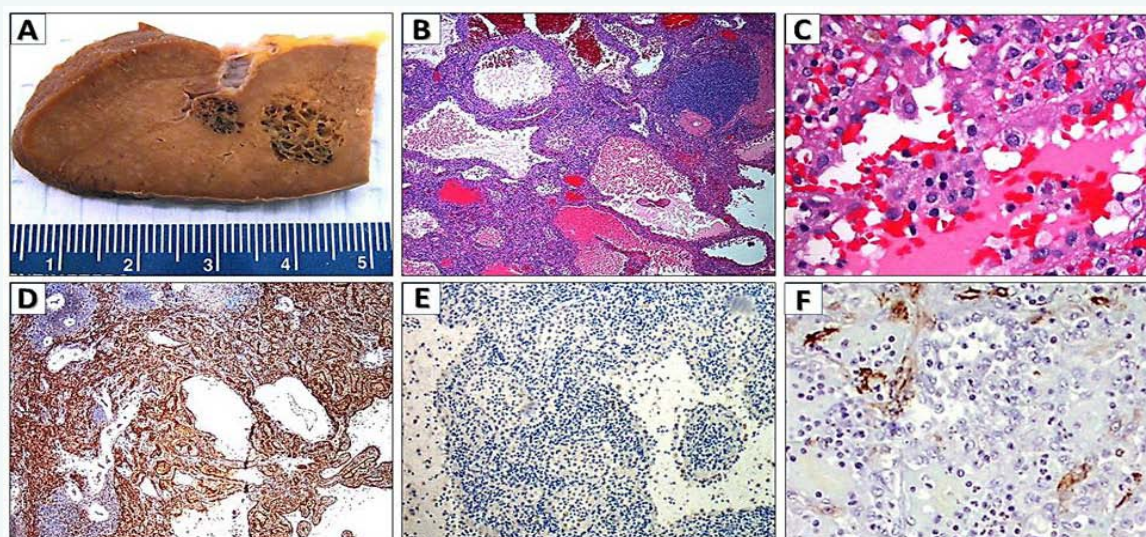


Figure 1 Pathological examination of the spleen Littoral cell tumor

1A: The cut surface showing relatively circumscribed, spongy, blood filled cystic spaces separated by fibrous septae

1B: Proliferation of anastomosing, tortuous, blood filled vascular channels with irregular lumina forming papillary projections and cystic spaces. (H&E stain x20)

1C: The vascular spaces were lined by non-atypical tall bland endothelial cells with sloughing of endothelial cells into the vascular spaces (H&E stain x40)

1D: Tumor cells positive for CD31

1E: Tumor cells negative for pan cytokeratin

1F: Tumor cells negative for CD34



Angiosarcoma (AS) of the spleen is the most common primary non-hematopoietic malignant tumor of the spleen [16]. Like other sarcomas, splenic angiosarcoma is a highly aggressive tumor [17]. Patients diagnosed with splenic angiosarcoma have poor prognosis with only 20% surviving for more than 6 months [16]. Angiosarcoma frequently metastasizes to the liver, followed by lung and bone [18].

Imaging studies such as magnetic resonance imaging (MRI), computed tomography (CT) scan, and ultrasound are generally not useful in differentiating LCA from other splenic lesions. However, CT of LCA shows solitary or multiple hypoattenuating nodules [14]. In one study, T1 and T2 weighted MRI revealed hypodense lesions due to hemosiderin deposits [2]. Radiological appearance of angiosarcoma often shows a large splenic mass or multiple masses with associated splenomegaly [19]. On contrast-enhanced CT, angiosarcoma is commonly seen as hypervascular masses in the spleen. These lesions have a heterogeneous appearance due to internal areas of hemorrhage and necrosis. Calcifications in the lesions are rarely reported [20]. Splenic lymphangioma is a rare, benign splenic tumor that often presents during childhood [21]. They may be isolated or may be part of rare lymphangiomatous syndrome involving multiple organs. They often present as an abdominal mass in children, and as an incidental finding in adults [22.] In imaging studies, they appear as multiloculated cysts of varying size that are predominantly subcapsular in location. They are hypoechoic on ultrasound and hypodense on CT, with few enhancing septa and occasionally with peripheral rim of calcification [20]. Hamartoma of the spleen is a rare benign lesion composed of disrupted splenic red pulp elements with unorganized lymphoid follicles. On ultrasound, hamartomas are usually seen as well-circumscribed homogeneous solid masses [23]. Typical hamartomas appear isodense on non-contrast CT and are usually isointense on T1-weighted images and heterogeneously hyperintense on T2-weighted images. More often, splenectomy is needed for a definitive diagnosis [24]. Lymphoma of the spleen is mostly due to secondary involvement, whereas primary lymphoma of the spleen accounts for less than 1% of all lymphomas [25]. If the splenic lymphoma is secondary, abdominal lymphadenopathy is often present [26]. Splenic lymphomas are characterized by diffuse infiltration which can be manifested as splenomegaly; focal, small or miliary nodules; or as multiple large nodular lesions; and bulky solid masses [27].

Kaposi sarcoma is a low-grade vascular tumor associated with Kaposi sarcoma Human Herpesvirus 8 (HHV-8) infection. Kaposi sarcoma is one of the most common AIDS-defining malignancies where it is usually high grade with aggressive course. It is often seen in mucocutaneous sites, but also may involve other organs and anatomic locations, including the spleen. HHV8 is the most specific immunohistochemical marker available to help distinguish Kaposi sarcoma from other neoplasms [28]. Hemangioma is the most common benign neoplasm of the spleen and is often found incidentally on imaging [2]. Splenic hemangiomatosis consists of vascular endothelial cells forming cavernous and capillary vessels but lacks macrophages [29]. They have varied radiological appearances depending on the

capillary or cavernous components of the hemangioma. On ultrasound, smaller hemangiomas often are discrete echogenic lesions while larger lesions may have a more complex appearance [30]. Punctate peripheral calcifications may be noted on CT. On MRI, splenic hemangiomas are typically isointense to hypointense on T1-weighted images and hyperintense on T2-weighted images [20]. Littoral cell angiosarcoma is cytologically and immunophenotypically similar to a LCA but has lining cells that are more atypical than seen in a littoral cell angiomas [31]. Histological examination of littoral cell angiosarcoma shows a well-differentiated neoplasm forming ballooning blood channels as well as intraluminal papillary fronds. Tumor cells display malignant nuclear features and hemophagocytosis. Solid neoplastic areas with mitotic figures are present. The tumor cells in littoral cell angiosarcoma show the concomitant presence of lysosomes and Weibel-Palade bodies. Immunohistochemically, tumor cells are positive for both endothelial Factor VIII-AG, and CD34 along with histiocytic markers cathepsin D, lysozyme, and alpha-1-antichymotrypsin. One study indicated that angiosarcoma may originate from all the vascular compartments of the spleen, including red-pulp sinuses similar to LCA [32].

LCA can be differentiated from these other splenic lesions based on histopathological findings. Gross appearance of the spleen shows slight-to-moderate splenomegaly with enlarged size and weight. Cross-section shows widened splenic trabeculae and often multifocal nodules rather than a solitary nodule. In our case, two separate lesions were identified. Histological examination shows sinus-like anastomosing channels with an irregular lumen that resemble splenic sinusoidal architecture. These sinus-like channels may have a papillary pattern or form a cyst-like space that is lined by tall, plump endothelial cells. These endothelial cells can function in hemophagocytosis and lack features of nuclear atypia or mitotic activity [7].

Immunohistochemical findings can help differentiate LCA from other splenic lesions. LCA shares some morphologic and IHC features with hemangiomas of other sites, such as immunoreactivity for vascular endothelial markers CD31 and factor VIII. However, IHC staining for littoral cells usually reveals a dual differentiation pattern. The cells stain positive for endothelial markers factor VIII, CD31, as well as histiocytic marker CD 68. In almost all cases LCA is CD68 positive. In addition, LCA is characteristically CD 21 positive and CD 8 negative [33]. The epithelial cells in LCA occasionally express S-100 protein and have a low Ki67 proliferative index [13]. In addition, high expression of formin homology domain protein 1 (FHOD1) distinguishes non neoplastic littoral cells from the neoplastic LCA. FHOD1 protein is expressed by normal littoral cells, but not by LCA [1].

LCA has been linked to Crohn's disease in some reports. Inborn metabolic diseases such as Gaucher's disease and immune system dysfunction have been postulated as a possible important pathogenic mechanism [4]. There have been reports describing an association of LCA with immunological disorders, including; ankylosing spondylitis, myelodysplastic syndrome, non-Hodgkin lymphoma, Wiskott Aldrich syndrome, chronic glomerulonephritis, and aplastic anemia [32]. Supporting



this hypothesis, other reports have suggested that chronic infection and systemic immunosuppression may contribute to LCA development [34]. There have been previous reports describing the association of cancers of the lung, colon, pancreas, kidney, and ovary with LCA [1]. Our case was associated with widely spreading metastatic leiomyosarcoma initially uterine in origin [39]. A 2016 study done by Pekova et al. studied 25 cases of LCA. Approximately one third of cases were accompanied by other existing malignancies, including; lung cancer, gastrointestinal cancer, genitourinary cancer, melanoma, lymphoma and sarcoma. Immune disease was found in another third, including; ankylosing spondylitis, psoriasis, Castleman's disease, lymphocytic colitis, Systemic Lupus Erythematosus and Crohn's disease. These findings suggest a possible relationship between LCA and malignancy or chronic inflammation. This provides evidence that LCA may have a TNF-alpha related pathogenesis [6].

Current treatment of LCA is long term follow up due to a potential to become malignant. Splenomegaly is commonly associated with LCA and so a laparoscopic splenectomy can be challenging and should be performed by an experienced surgeon with a particular focus to prevent splenic capsule rupture and tumor cell dissemination. Conversion to hand-assisted laparoscopic splenectomy (HALS) or even open surgery may be necessary when dealing with cases having extensive adhesions or massive splenomegaly, especially in patients with malignant tumors [7]. One case of successful partial splenectomy reported with a localized LCA that could be removed completely for histological analysis [35]. A 2018 case report by Takayoshi et al. has shown promise in cytotoxic agents such as etoposide, paclitaxel, and vincristine, while also prescribing immunosuppressants prednisolone and cyclosporine to treat metastatic LCA [36].

One of the largest LCAs on record, an abdominal CT scan showed a spleen measuring 35×18 cm and containing multiple lesions, the largest nodule being 11.3×9.2 cm [37]. Bisceglia et al suggested the possible existence of a clinical syndrome. Further clinical observations along with more in-depth genetic and molecular studies are needed before any conclusions can be drawn [38].

We present this report to add another case of Littoral cell angioma to the medical literature, a rare splenic lesion for which pathogenesis is mostly unknown. As large series studies reported a strong association of LCA with co-existing malignancies or chronic inflammatory conditions, a search for these conditions should be seriously investigated whenever there is a diagnosis of LCA. We hope this report will raise the awareness of clinicians and pathologists to include this rare tumor in the differential diagnosis of splenic lesions.

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