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Case Report

Extra-adrenal myelolipoma presenting in the spleen: A report of two cases☆

N.S. Aguilera*, A. Auerbach

University of Virginia Health System, Charlottesville VA and +Joint Pathology Center, Silver Spring, MD

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Abstract Myelolipoma is a rare neoplasm composed of mature fat and bone marrow occurring most frequently in the adrenal gland with rare occurrences in extra adrenal locations including lung, liver, retroperitoneum, mediastinum and testes. Splenic myelolipomas are seen most commonly in non-human species including cat and dog. Only rare cases of splenic myelolipoma in humans have been reported previously. We present two cases of myelolipoma in the spleen. The first is a 62 year old female presenting with abdominal pain and a splenic mass. The second is a 44 year old male presenting with hematuria and a mass in the spleen. Both cases showed trilineage bone marrow elements with mature fat. These cases demonstrate that myelolipoma do rarely occur in human spleen and we highlight the distinction from extramedullary hematopoiesis, mature extramedullary myeloid tumor (myeloid sarcoma), lipoma and well differentiated liposarcoma.

1. Introduction

Myelolipomas (ML) most commonly present as an adrenal mass occasionally becoming hemorrhagic and rarely as an extra adrenal myelolipoma (EAML). Adrenal myelolipoma are commonly asymptomatic but may rupture and cause acute abdomen necessitating surgical intervention [1]. A wide age range at presentation is seen in ML and there is no sex predilection. They vary widely in size with most being between 5 and 10 cm in diameter. EAML are usually incidental and require no specific treatment. EAML is an extremely rare tumor presenting as a benign fatty mass or containing both soft tissue and fat in radiologic studies [2]. EAMLs have been located in the pelvis [3], chest/mediastinum [3,4], retroperitoneum [5], peri-renal [4], omentum [2], liver [3], nasal cavity [6]and rarely spleen [7–9]Splenic myelolipomas are more commonly seen in other species including cat and dog [10,11], but rarely in humans.

2. Case report

Cases were sent as consultative cases to the Armed Forces Institute of Pathology now Joint Pathology Center, Silver Spring MD. Hematoxylin and eosin stained sections of each case were examined. Paraffin embedded tissue was available for case 2 and immunohistochemistry was performed. Immunohistochemical staining for CD61 (Immunotech, Miami, FL) and MDM2 (Novus Biologials, Littleton, CO), CD8 (DAKO, Carpinteria CA), hemoglobin peroxidase (DAKO) were done with an automated immunohistochemical staining system, DAKO

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* Corresponding author at: University of Virginia Health System, P.O. Box 800214, Charlottesville, VA, 22908-0214. Tel.: +1 434 924 9492.
E-mail address: na2d@virginia.edu (N.S. Aguilera).

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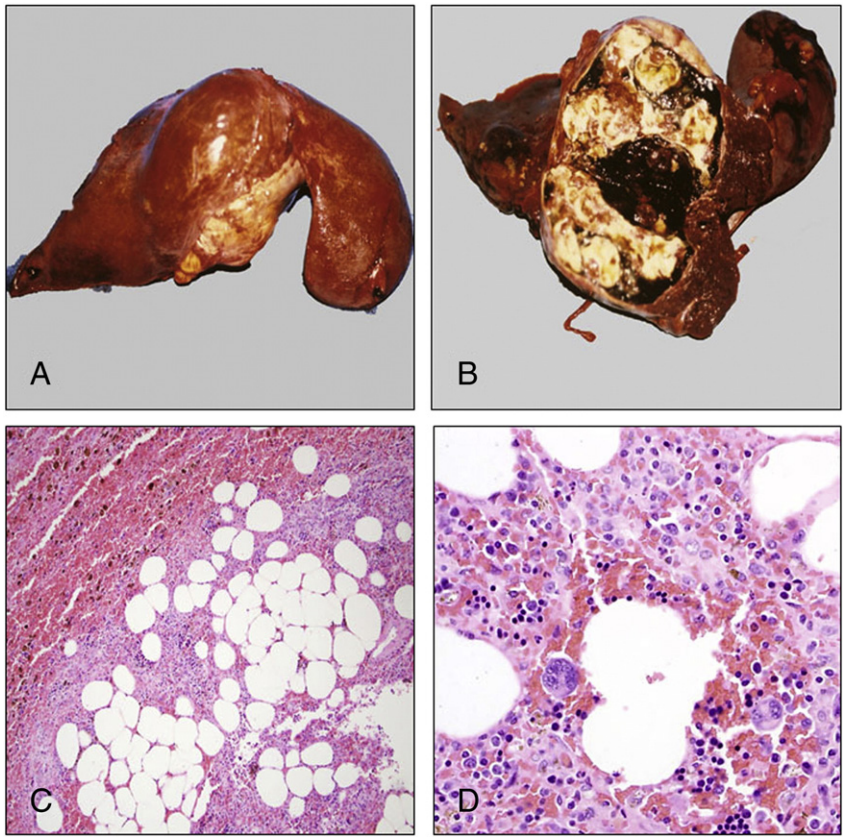

Fig. 1 A) Gross image of the uncut spleen showing a 8.5 CM mass in greatest dimension, B) Gross image of the cut surface of the mass showing a yellow-white surface and hemorrhagic areas. Normal spleen can be seen at the periphery, C) 4X H&E of case 1 showing mature fat with surrounding splenic tissue showing hemorrhage. D) 40X H&E showing hematopoiesis with all three cell lines present with normal morphology.

Autostainer, with antigen retrieval and antibody dilutions per manufacturer recommendation.

2.1. Case 1

The patient was a 62 year old African American female presenting with an abdominal pain. Imaging studies showed a 4.2 cm mass in the right ascending colon and an 8.5 cm in the spleen. To evaluate both a splenic mass and a colonic mass, the patient underwent a splenectomy and segmental resection of the right large intestine. The spleen weighed 228 g. The gross findings showed a well circumscribed mass 8.5 cm in greatest dimension. The splenic mass showed a yellow to reddish cut surface (Fig. 1). Bone marrow elements with trilineage hematopoiesis and normal fat were seen in the mass. Extensive hemorrhage was present in the mass and in the surrounding spleen. The right colon was diagnosed with colonic adenocarcinoma and the splenic mass was diagnosed as myelolipoma and considered incidental.

2.2. Case 2

The patient was a 44 year old male who presented with hematuria. CT scan detected a large splenic mass which was displacing the kidney. Grossly the spleen weighed 197 g. The mass was well defined grossly. Microscopically bone marrow elements showing trilineage hematopoiesis was present with mature fat (Fig. 2). Focal hemorrhage was present in the mass. Immunohistochemistry showed no CD8 positive lining cells in the areas of myelolipoma indicating absence of normal sinuses. Hemoglobin peroxidase marked clusters of erythroid cells. Myeloperoxidase marked the myeloid cells and CD61 highlighted scattered megakaryocytes. MDM2 staining was negative.

The splenic mass was diagnosed as a myelolipoma and the hematuria resolved after splenectomy and was felt to be unrelated to the mass.

Neither case showed boney spicules or adrenal elements. No atypia in the fat was seen in either case. Normal spleen was present at the periphery of the mass in both cases and no extramedullary hematopoiesis outside of the mass was seen.

3. Discussion

Myelolipomas (ML) are tumors composed of mature bone marrow elements with mature adipose tissue. Hematopoietic cells typically represent variable numbers of all 3 lineage of