Hepatic artery aneurysm secondary to epithelioid angiosarcoma

Vanessa W. Davis, MD;[*] Karim Somani, MD;[*] Todd P. Chaba, MD;[†] Jennifer Andruchow, MD;[‡] A.M. James Shapiro, MD, PhD[*]

Angiosarcomas, which are rare, aggressive mesenchymal neoplasms, are responsible for less than 1% of all soft-tissue sarcomas. The incidence of these tumours is 2–3 cases per 1 000 000 annually.[1] A small subset of these tumours may arise within a large vessel wall, and of the few reported cases, there seems to be a predilection for the pulmonary vasculature or the aortic wall.[2] To our knowledge, angiosarcomas have not been described within a visceral artery aneurysm. We report a case of epithelioid angiosarcoma arising from the common hepatic artery with aneurysm formation and metastatic involvement of the liver and perigastric soft tissue.

Case report

A 58-year-old man presented with a history of epigastric pain and melena stool for 1 week. Prostate cancer had been diagnosed 7 years previously, and this was treated with pelvic irradiation (35 fractionated treatments) with no recurrence and a normal prostate-specific antigen level. There was no known exposure to vinyl chloride, arsenic compounds or thorium dioxide. The finding of microcytic anemia prompted a gastroscopy, which revealed an ill-defined submucosal mass along the greater curvature of the stomach, biopsies of which were nondiagnostic. Computed tomography (CT) revealed a 6.5-cm aneurysm of the common hepatic artery extending to the base of the gastroduodenal artery and an exophytic mass, 2.7 cm in diameter, arising from the stomach. No metastatic disease was found (Fig. 1).

Laparotomy demonstrated an exophytic mass arising from the gastric body, with acute bleeding into the surrounding omentum, a nodule measuring 0.5 cm in diameter in segment 2 of the liver and a large aneurysm of the common hepatic artery. The patient underwent a Bilroth II gastrectomy with Roux-en-Y reconstruction. Proximal and distal aortic control was obtained, and the celiac artery was completely exposed. The aneurysm was intimately connected to the superior border of the pancreas. A segment of the aneurysm wall lining was sent for pathological examination and culture. The splenic artery was transposed onto the proper hepatic artery with interrupted 7-0 Prolene sutures, and the spleen was preserved on the short gastric vessels (Fig. 2). A low-grade leak from the duodenal stump, likely induced by ischemia after ligation of the gastroduodenal artery, necessitated a second laparotomy and wash-out on day 5, with repair over a duodenal tube. Subsequently, the

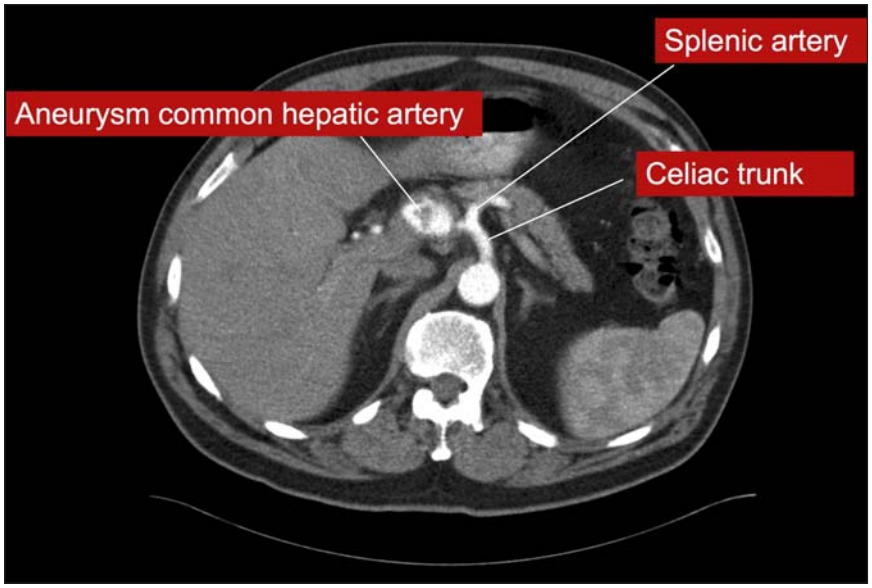

FIG. 1. Computed tomography scan showing a 6.5-cm aneurysmal dilatation of the common hepatic artery extending to the origin of the gastroduodenal artery, intimately associated with the head of the pancreas.

*From the Departments of *Surgery and †Pathology, University of Alberta Hospitals, and the ‡Sturgeon Hospital, Edmonton, Alta.*

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Correspondence to: *Dr. A.M.J. Shapiro, 2000 College Plaza, 8215–112th St., Edmonton AB T6G 2C8; fax 780 407-6933; shapiro@islet.ca*

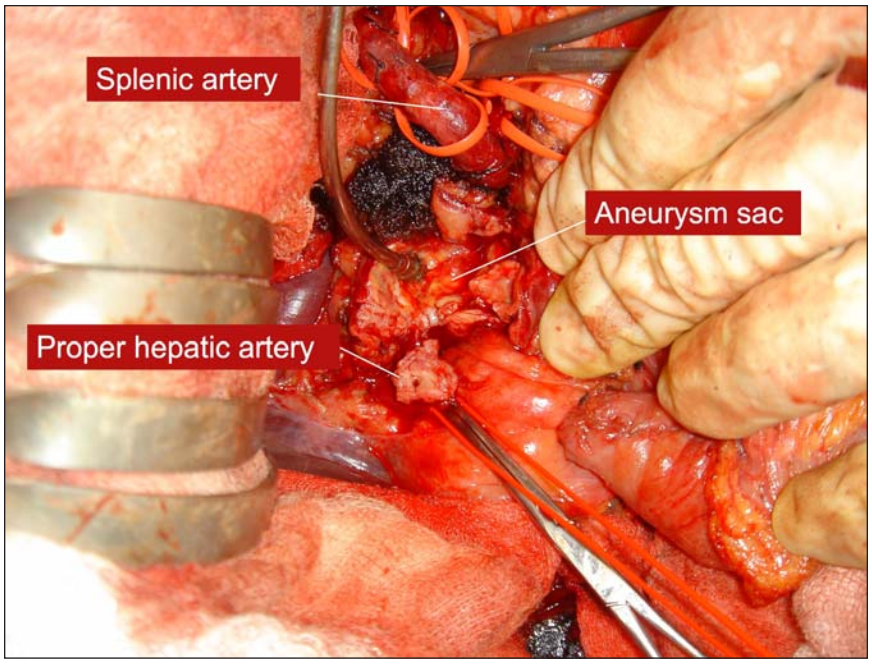

FIG. 2. Operative view showing the aneurysmal sac and splenic artery dissected in preparation for transposition onto the proper hepatic artery.

patient was discharged home on a normal diet.

Histologic sections from the hepatic artery aneurysm revealed pleomorphic epithelioid malignant tumour cells with a fibrous wall. Sections from the stomach and omentum showed similar cytologic features with lymphovascular invasion consistent with metastatic disease. Twenty-four sampled lymph nodes were negative for malignancy. Biopsy of the liver nodule also revealed metastatic infiltrating malignant pleomorphic epithelioid cells with solid nests and sheets. Immunohistochemical studies demonstrated positive staining for vimentin, CD31 and CD34 and negative staining for keratin, CD117 (c-kit), Mart-1 and LCA. S-100 showed weak focal positivity. Overall, the morphologic and immunohistochemical features were diagnostic of epithelioid angiosarcoma arising from the common hepatic artery, with metastases to the liver, perigastric soft tissue and omentum. The tumour was designated as pT1b pNo pM1 according to the TNM staging system.

Discussion

Although angiosarcomas may occur in various anatomic locations, this vascular malignant tumour has a predilection for cutaneous involvement of the head and neck and is commonly seen in chronically lymphadematous limbs or after radiotherapy.[1,3] Although angiosarcomas characteristically display vascular differentiation, primary intravascular involvement is relatively rare.[1] Epithelioid angiosarcoma causing a visceral aneurysm in the hepatic artery is an unusual and previously unreported manifestation of this mesenchymal malignant tumour. Only 6 cases of angiosarcoma with associated aneurysm formation have been reported in the literature, and visceral artery aneurysms secondary to any cause represent only 0.1%–0.2% of all vascular aneurysms.[4]

Several known predisposing factors for angiosarcomas exist, including exposure to vinyl chloride, arsenic compounds and thorium dioxide. Areas of chronic lymphadema are prone to the development of angiosarcomas, as are previously irradiated areas. In the current case, previous exposure to high-dose fractionated irradiation for prostate cancer may be the underlying etiologic factor.

Surgical resection currently offers the only possible curative therapy for epithelioid angiosarcoma, but the prognosis remains dismal, even with aggressive multimodal therapies, since metastatic disease is frequent and this tumour is resistant to currently available chemotherapeutic agents and radiotherapy. One surgical series of 44 patients with angiosarcoma at various sites reported a median patient survival of 20 months, and visceral involvement was associated with a far inferior prognosis when compared with other sites.[5] At 4 months' follow-up, our patient was alive with stable metastatic disease and had not received adjuvant chemotherapy.

Competing interests: None declared.

References

1. Hottenrott G, Mentzel T, Peters A, et al. Intravascular ("intimal") epithelioid angiosarcoma: clinicopathological and immunohistochemical analysis of three cases. *Virchows Arch* 1999;435:473-8.
2. Scholsem M, Raket D, Flandroy P, et al. Primary temporal bone angiosarcoma: a case report. *J Neurooncol* 2005;75:121-5.
3. Allison KH, Yoder BJ, Bronner MP, et al. Angiosarcoma involving the gastrointestinal tract: a series of primary and metastatic cases. *Am J Surg Pathol* 2004;28:298-307.
4. Overhaus M, Lauschke H, Schafer N, et al. The coeliac aneurysm: a rare cause of abdominal pain. *Vasa* 2006;35:201-5.
5. Fury MG, Antonescu CR, Van Zee KJ, et al. A 14-year retrospective review of angiosarcoma: clinical characteristics, prognostic factors, and treatment outcomes with surgery and chemotherapy. *Cancer J* 2005;11:241-7.