

## Case report

## Anastomosing hemangioma of the ovary mimics metastatic ovarian cancer

Andrew Rezk<sup>a</sup>, Stephanie Richards<sup>b</sup>, R. Patricia Castillo<sup>c</sup>, Matthew Schlumbrecht<sup>d,e,\*</sup><sup>a</sup> University of Miami Miller School of Medicine, Miami, FL, USA<sup>b</sup> Department of Pathology, University of Miami Miller School of Medicine, Miami, FL, USA<sup>c</sup> Department of Radiology, University of Miami Miller School of Medicine, Miami, FL, USA<sup>d</sup> Sylvester Comprehensive Cancer Center, Miami, FL, USA<sup>e</sup> Department of Obstetrics, Gynecology, and Reproductive Sciences, University of Miami Miller School of Medicine, Miami, FL, USA

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## ABSTRACT

Anastomosing hemangioma (AH) is an uncommon benign vascular tumor reported to occur in the kidney and, in rare instances, the ovary. While most cases of AH in the ovary are incidental findings, we report a case of ovarian AH presenting with abdominal ascites and elevated CA-125 suggestive of metastatic ovarian cancer. Post-operative histopathologic examination demonstrated a tumor consisting of numerous vascular spaces lined by benign-appearing endothelial cells with exuberant hilus cell hyperplasia. These characteristics led to the diagnosis of anastomosing hemangioma of the ovary. A summary of the characteristics of AH, along with a review of all previously reported cases and possible theories for its presentation, are discussed.

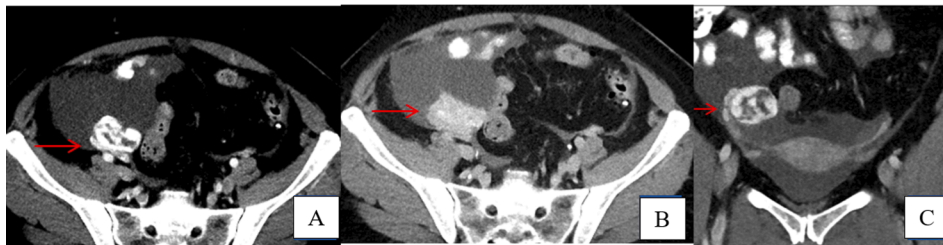
## 1. Case report

A 60-yr-old G4 P4004 with an incidental finding of a right-sided ovarian cyst on pelvic ultrasound performed by her PCP was referred to gynecology but ultimately lost to follow-up. A year later, the patient presented with abdominal bloating and persistent discomfort. She reported a 12-pound weight-loss over the previous few months. Family history was negative for malignancy. Past surgical history was significant for Cesarean section and laparoscopic salpingo-oophorectomy secondary to an ovarian cyst.

She was referred to gastroenterology, where an abdominal

ultrasound revealed diffuse ascites and a fatty liver. CA-125 was 5221 U/mL. Paracentesis yielded more than 1L of ascites fluid; however, cytologic examination showed no malignant cells. The right ovarian lesion was better defined on CT Abdomen/Pelvis with avid enhancement after contrast administration in the arterial phase and retention of the contrast in the delay images as well as association with large amount of ascites (Fig. 1A-C).

She underwent a diagnostic laparoscopy with aspiration of 1L of ascites. No peritoneal carcinomatosis was identified (Fagotti score = 0). The procedure was converted to exploratory laparotomy. Frozen section of the right ovarian mass noted 'stromal tumor, unable to characterize',



**Fig. 1.** Axial contrast-enhanced CT image in arterial phase (A) delay phase (B) and coronal arterial phase (C) shows a lobulated avid enhancing mass in the right ovary (red arrow) which pools the contrast in the delay image. Noted associated large volume ascites.

\* Corresponding author at: Division of Gynecologic Oncology, Sylvester Comprehensive Cancer Center, University of Miami Miller School of Medicine, 1121 NW 14th St, Suite 345C, Miami, FL 33136, USA.

E-mail address: [mschlumbrecht@miami.edu](mailto:mschlumbrecht@miami.edu) (M. Schlumbrecht).

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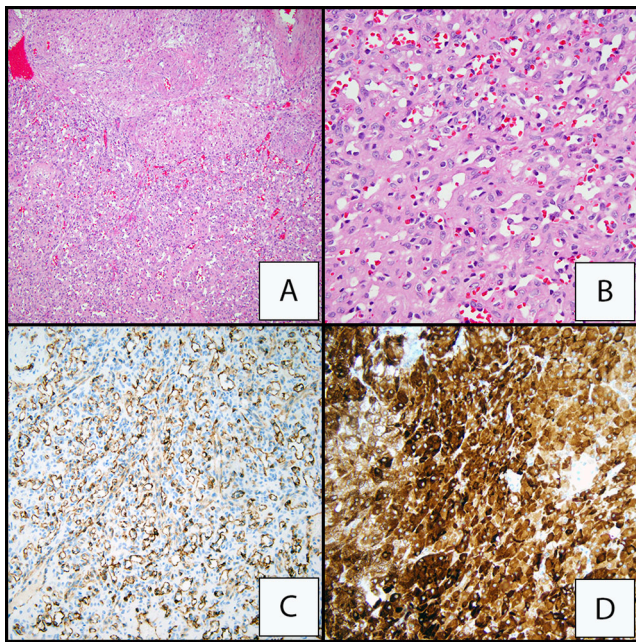
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**Fig. 2.** A) Anastomosing hemangioma (bottom) with adjacent hilus cell hyperplasia (top) (H&E stain, 100X). B) Higher power image showing a neoplastic proliferation of small, anastomosing vascular spaces containing red blood cells and lined by bland endothelial cells (H&E stain, 400X). C) Vascular spaces (CD31 immunostain, 200X). D) Hilus cell hyperplasia (Inhibin immunostain, 200X).

so total abdominal hysterectomy with bilateral salpingo-oophorectomy (TAH/BSO), infracolic omentectomy, and peritoneal biopsies was performed. Postoperatively the patient had resolution of her ascites and normalization of the CA125, and at her final postoperative appointment was experiencing no symptoms.

Pathologic examination of the right ovary revealed a 6.5 cm neoplastic vascular proliferation consisting of numerous capillary sized

vessels with anastomosing architecture and a benign-appearing endothelial lining (Fig. 2A and B). The tumor cells lacked nuclear atypia and had no appreciable mitotic activity. The endothelial origin of the tumor was demonstrated with a CD31 immunostain (Fig. 2C), and an inhibin immunostain (Fig. 2D) highlighted exuberant hilus cell hyperplasia surrounding the tumor. After careful consideration of clinical presentation, radiographic findings, and morphologic features, the diagnosis of anastomosing hemangioma was rendered. Consensus of the multidisciplinary team was routine gynecologic surveillance for this patient with a benign lesion.

**2. Discussion**

Anastomosing Hemangioma (AH) was first described in 2009 by Montgomery and Epstein (2009). Yet, out of the 6 cases the authors investigated, none were within the ovary. The morphological appearance is consistently described as non-lobular proliferation of anastomosing capillary sized vessels with sinusoidal-like arrangements resembling the red pulp of the spleen, with vessels lined by bland endothelial cells (Dundr et al., 2017). Importantly, the lack of atypical endothelial cells and mitotic figures helps to differentiate AH from its malignant counterpart, angiosarcoma (Bösmüller et al., 2011). On imaging, the differential diagnosis of AH includes ovarian teratoma with a large hemangiomatous component and ovarian angiosarcoma.

Since the definition of AH was established in 2009, 14 identifiable cases in the ovary have been described (Table 1) (Dundr et al., 2017; Kryvenko et al., 2011; Metodiev et al., 2015; John and Folpe, 2016; Gunduz et al., 2019; Subbarayan et al., 2019; Stewart and Salfinger, 2020). In these cases identified from the literature, ages at diagnosis ranged from 43 to 81 and size of tumors ranged from 1 mm to 11.2 cm. Almost all tumors were positive for CD31 and CD34 immunostains. Another common feature described is the association with prominent stromal luteinization, stromal hyperthecosis, and/or hilus cell hyperplasia (Montgomery and Epstein, 2009; Dundr et al., 2017; Kryvenko et al., 2011; Metodiev et al., 2015; Gunduz et al., 2019; Subbarayan et al., 2019; Stewart and Salfinger, 2020). The presence of these histologic and immunohistochemical findings in our case confirmed the

**Table 1**  
Previously reported cases of anastomosing hemangiomas of the ovary.

References	Age	Tumor Location	Size	Clinical Presentation	Histological Findings	IHC Staining
Kryvenko et al.	70	R. Ovary	2 mm	Endometrial Ca.	No Stromal Luteinization	CD31, CD34
	49	R. Ovary	1 mm	Benign Serous cyst	No Stromal Luteinization	CD31, CD34
	77	L. Ovary	1.1 cm	Serous cystadenoma	Stromal Luteinization	CD31, CD34
Metodiev et al.	70	R. Ovary	7 mm	TAH/BSO* Incidental Ultrasound Finding of ovarian cyst	Stromal Luteinization	CD34
Folpe et al.	74	NA	6.7 cm	NA		CD31, CD34
Dundr et al.	66	Ovary	0.5 cm	AUB	Stromal Luteinization	CD31, CD34
	43	L. Ovary	1.3 cm	Leiomyoma	Stromal Luteinization	CD31, CD34
	69	R. Ovary	1.5 cm	Leiomyoma	Stromal Luteinization	CD31, CD34
	81	R. Ovary	3.5 cm	Adenomyosis	Stromal Luteinization	CD31, CD34
	68	L. Ovary	3.5 cm	Ovarian tumor, ascites, increased CA125 (470 U/ml)	Stromal Luteinization, Hilus cell hyperplasia	CD31, CD34
	69	R. Ovary	1.2 cm	Suspected ovarian tumor (patient under surveillance because of lymphoma)	Stromal Luteinization	CD31, CD34
	62	R. Ovary	11.2 cm	Elevated CA-125 (114 U/ml)	Stromal Luteinization	CD31, CD34
Gunduz et al.	62	R. Ovary	11.2 cm	Elevated CA-125 (114 U/ml)	Stromal Luteinization	CD31, CD34
Subbarayan et al.	50	R. Ovary	3 cm	Mild ascites, Normal CA-125. CT defined heterogeneous poorly enhancing mass.	Stromal Luteinization	CD31, CD34
Stewart et al.	48	L. Ovary	8 mm	AUB	Stromal Luteinization, Hilus Cell hyperplasia	CD31, CD34

AUB Abnormal Uterine Bleeding. NA Not Available.

presence of hilus cell hyperplasia.

Out of these 14 cases, only two cases presented similarly to the case discussed herein with an elevated CA-125 and ascites. One other case presented with an elevated CA-125 alone (Gunduz et al., 2019). One of the cases featuring elevated CA-125 and ascites also notably demonstrated hilus cell hyperplasia (Dundr et al., 2017).

Few hypotheses exist about the pathogenesis of these benign lesions, their characteristic vascular nature, and the association with stromal luteinization. The most likely hypothesis characterizes these masses as behaving like enlarging follicles that cause pressure on the neighboring tissue and lead to the development of theca-like luteinized stromal cells (Dundr et al., 2017). To our knowledge, no hypothesis exists as to why these benign tumors specifically present similarly to ovarian carcinoma with elevated CA-125 and ascites. The pathogenesis of malignant ascites is mediated by vascular endothelial growth factor (VEGF) and interleukin 6 and 8, which activate native mesothelial cells and increase vascular permeability, leading to effusion accumulation (Smolle et al., 2014). This may serve as a possible explanation to why these heavy vascularized tumors produce ascitic fluid despite lacking malignant characteristics.

In summary, AH of the ovary with hilus cell hyperplasia presenting with abdominal ascites and elevated CA-125 is an extraordinary presentation of this rare benign vascular tumor. It is important that clinicians are aware of the similarity of some AH presentations to malignant ovarian carcinoma to reduce misdiagnosis, unnecessary treatment, and appropriate follow-up.

#### Author Contribution

AR wrote the manuscript. SR provided pathologic figures and contributed to writing the manuscript. RPC provided radiology figures and contributed to writing the manuscript. MS treated the patient and

edited the manuscript.

#### Declaration of Competing Interest

The patient has provided consent for this manuscript. The authors have no affiliations with or involvement in any organization or entity with any financial interest or non-financial interest in the subject matter or materials discussed in this manuscript.

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