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CASE REPORT

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Ovarian hemangioma: A rare encounter

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Key Clinical Message

Ovarian hemangioma, though rare and asymptomatic, can mimic malignant ovarian tumors, thus it is necessary for comprehensive histopathological and immunohistochemical evaluation for accurate diagnosis and appropriate management.

KEYWORDS

benign ovarian tumor, capillary hemangioma, diagnostic challenge, ovarian hemangioma, vascular lesion, vascular tumor

1 | INTRODUCTION

Hemangiomas are benign vascular tumors and hemangioma of the female genital tract, particularly those developing in the ovaries, are extremely rare. Ovarian hemangioma (OH) are typically accidental findings following surgery or discovered by chance during autopsy.[1] Although they are asymptomatic, few of them can sometimes be symptomatic when large enough to cause abdominal pain, adnexal torsion or resemble an ovarian neoplasm.

The article's focus is on the rare diagnosis, along with its clinicopathological characteristics and alternative diagnosis in light of contemporary literature.

2 | CASE REPORT

A 63-year-old postmenopausal woman presented with 2 months of lower abdominal pain to our hospital for evaluation. Her past medical and family history were insignificant.

3 | METHODS

Ultrasonography (USG) and non-contrast and contrast-enhanced computed tomography (NCCT and CECT) revealed a solid, enlarged left ovary measuring 38 × 40 × 46 mm with significant enhancement, and a right ovary measuring 14 × 16 mm. Small lymph nodes were noted in the mesentery, bilateral external and internal iliac regions. Cancer antigen 125 (CA-125) and carcinoembryogenic antigen (CEA) were elevated to 327 IU/mL and 0.85 ng/mL respectively. Physical examination showed a non-tender abdomen.

Staging laparotomy and frozen section was planned. Frozen sections from the left ovarian mass showed atypical cells exhibiting moderate pleomorphism and anisonucleosis without mitosis or necrotic features, suggesting stromal tumor. Considering the age of the patient and suspected stromal tumor on frozen section examination, total abdominal hysterectomy with bilateral salpingo-oophorectomy (Figure 1B), total omentectomy, and the removal of para-aortic and pelvic lymph nodes was

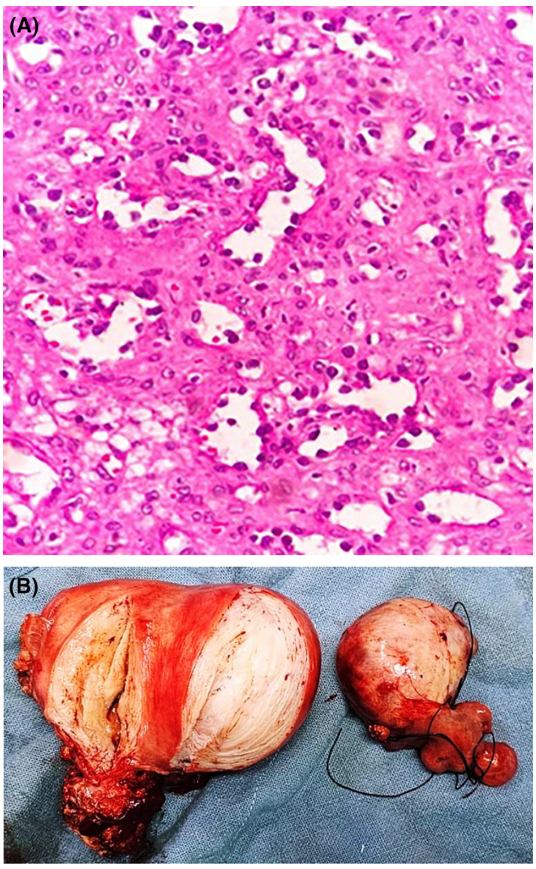

FIGURE 1 (A) H&E stained sections revealing variable calibered blood vessels of cavernous and capillary type lined by single layer of endothelial cells revealing hobnailed morphology. No necrosis, marked nuclear atypia and increased mitosis noted. (B) Surgical image showing left ovarian mass and hysterectomy specimen.

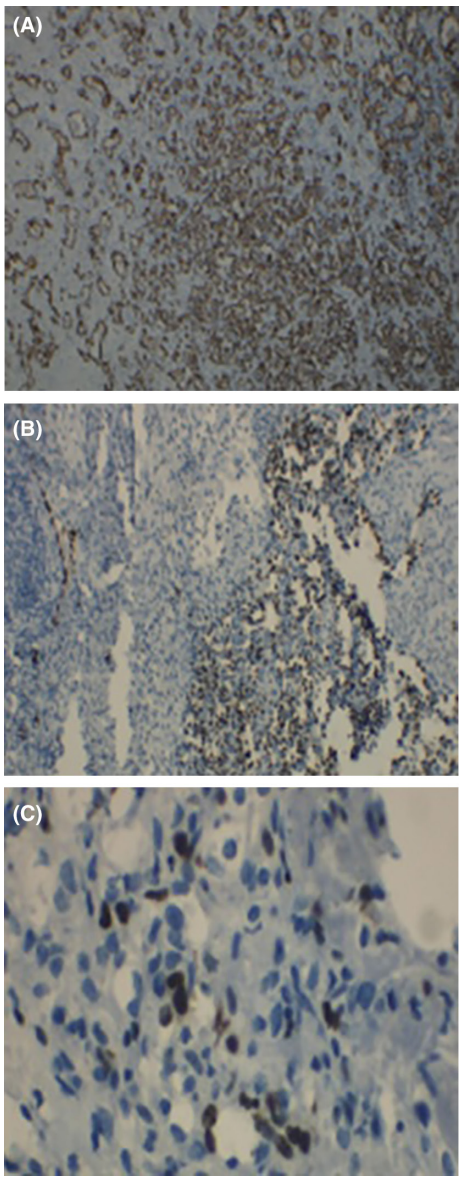

FIGURE 2 (A) IHC image shows tumor cell positive for CD34. (B) IHC image shows tumor cell positive for ERG. (C) IHC image shows nuclear positive for KI 67.

performed. On gross examination of the specimen, the left ovary was enlarged measuring 5 × 4 × 2 cm with a gray-brown glistening outer surface. Cut section was solid. Endometrial polyp of 3.5 × 2.5 × 1 cm attached to the right lateral wall of the uterus was also identified. The right ovary, bilateral fallopian tubes and parametrium appeared grossly normal and no tumor deposits were observed in the omentum.

On microscopy, sections from left ovarian mass showed variable-sized blood vessels lined by endothelial cells with hobnail features, surrounded by an edematous stroma with areas of hemorrhages. (Figure 1A) No evident features of marked atypia, necrosis and increased mitosis noted in sections studied. Hence, immunohistochemical analysis was performed to further evaluate.

Immunohistochemistry (IHC) demonstrated positivity for endothelial cell markers CD34, CD 31 and ERG, with a low proliferation index (KI 67 of <5%). (Figure 2A–C) These overall histomorphological features along with immunohistochemical findings are those of mixed cavernous and capillary hemangioma, characterized by hobnailed features.

4 | CONCLUSION

Her post-operative period was uneventful and she was consequently discharged without any fresh complaints.

5 | DISCUSSION

Hemangiomas are benign tumors of blood vessels occurring due to malformation of blood vessels during

canalization, leading to abnormal channels. Despite the ovary being a highly vascular organ, ovarian hemangiomas are rare non-cancerous growths with fewer than 60 cases documented in medical literature.[2] OH can develop at various ages spanning from toddlers at 2 years[3] to elderly at 81 years.[1] While some individuals may experience no symptoms, others with larger tumors may present with abdominal distention, vomiting, or abdominal pain, as observed in our case.[3]

Histologically, OH are three types depending on the size of the blood vessels: cavernous, capillary, and a mixed type.[4] Cavernous are the most common type while other two occur less frequently. A true hemangioma has proliferating vascular channels with minimal stroma, forming a distinct and separate mass within the ovary.[5] In this instance, the presence of multiple thin-walled vascular channels of variable size surrounded by an edematous stroma primarily supported the diagnosis of hemangioma.

The various differential diagnoses for ovarian hemangiomas include sclerosing stromal tumor and microcystic stromal tumor. Sclerosing sex cord-stromal tumor exhibits a pseudolobular architecture characterized by alternating hypocellular and hypercellular regions. The tumor primarily consists of round-to-oval luteinized cells and spindle cells, with hemangiopericytomatous vessels interspersed throughout,[6] which mimics our case due to the presence of vascular proliferation. Clinically, OH can mimic ovarian carcinoma when patients present with elevated CA-125 and ascites and pseudo-meigs.[7,8] OH can be differentiated from angiosarcoma based on the absence of marked cytologicatypia, papillary endothelial tufting, necrosis, and increased mitotic activity.[9] Other differentials of OH include tubo-ovarian mass, twisted ovarian cyst, chocolate cyst and pathologically they are distinguished from vascular growths, lymphangiomas, and monodermal teratomas with significant vascular component, while vascular elements aren't typically found in ovarian teratomas, there have been reports of bilateral ovarian teratomas containing hemangiomatosis component which can be differentiated from pure hemangioma by the presence of respiratory epithelium.[10] Ovarian hemangioma when associated with decreased platelet count are considered as Kasabach–Merritt syndrome, particularly in bilateral cases associated with widespread abdominopelvic hemangiomatosis.[5]

In middle-aged women, ovarian hemangioma may present as pseudo-Meigs syndrome. Notably, in the oldest documented case of an 81-year-old woman with ovarian hemangioma, symptoms included hypertension and hyponatremia.[1] These diverse age-related presentations underscore the variability in symptomatology associated with ovarian hemangioma.

In one such case, a 35-year-old woman presented with a two-year history of abdominal pain and heavy menstrual bleeding. Imaging revealed a 10 cm × 10 cm mass in the right iliac fossa, prompting surgical intervention. Initial frozen section analysis during surgery suggested a sex-cord-stromal tumor, leading to a hysterectomy and salpingo-oophorectomy due to suspected malignancy. However, post-operative immunohistochemistry unexpectedly revealed anastomosing ovarian hemangioma, challenging the initial diagnosis.[6] Conversely, in our case, for a 63-year-old, it was also initially suspected as a stromal tumor based on frozen section analysis, subsequent immunohistchemistry examination revealed mixed cavernous and capillary hemangioma through immunohistochemistry. The age difference between the cases and the differing initial diagnoses underscore the variability in symptomatology and diagnostic challenges associated with ovarian hemangiomas, emphasizing the importance of comprehensive histological evaluation for accurate diagnosis and management.

In another situation, a 38-year-old woman presented with symptoms indicative of Meigs' Syndrome, including shortness of breath, chest tightness, and bilateral lower leg edema. Imaging revealed a cystic tumor in the right adrenal area and the left ovary, suggesting ovarian cancer with nodal involvement, alongside elevated CA125 levels. The initial diagnosis impression was Meigs' Syndrome. However, upon histological examination, well-circumscribed hemangiomas of mixed type were identified within the ovarian parenchyma.[10] In contrast, in our case the diagnosis was, initially suspected as a stromal tumor. Surgical intervention and subsequent examination revealed mixed cavernous and capillary hemangioma, differing from the initial suspicion. While both situations involved ovarian hemangiomas, they occurred within distinct clinical contexts and diagnostic pathways, highlighting the variability in presentation and diagnostic challenges associated with these lesions.

Ovarian hemangiomas can be effectively diagnosed using color-doppler ultrasound and magnetic resonance imaging (MRI). Due to the variability in presentation it is essential to consider ruling out the diagnosis of ovarian hemangioma in patients who present with ovarian or abdominal masses to prevent unnecessary investigative procedures and surgical interventions.

AUTHOR CONTRIBUTIONS

M. Singh: Conceptualization; supervision. **P. Subedi:** Methodology; resources; writing – original draft. **B. Adhikari:** Writing – original draft. **S. Mohanty:**

Investigation. **A. Sherchan, A. Bajrachrya, and H. P. Dhakal:** Manuscript reviewing.

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CONFLICT OF INTEREST STATEMENT

The authors declare that there is no conflict of interest.

DATA AVAILABILITY STATEMENT

The data supporting the findings in this case report is available within the article and its supplementary materials.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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