International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Pawar AP et al. Int J Reprod Contracept Obstet Gynecol. 2025 Mar;14(3):913-917
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20250529

Case Report

Ovarian hemangioma: rare case report and review of literature

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Received: 25 December 2024
Revised: 21 January 2025
Accepted: 22 January 2025

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ABSTRACT

Ovarian haemangiomas are vascular tumours arising from ovary of female genital tract, are very rare. Not more than 60 well-documented cases of ovarian haemangioma are noted. Authors reports 77-year-old woman presented with pain in lower abdomen and discomfort along with bloating sensation since 1 month in gynaecology outpatient department (OPD) at Nowrosjee Wadia Maternity Hospital on august 2023. Ultrasonography suggestive of large cystic mass is seen the left hemi pelvis measuring 7.0×5.4 cm with multiple calcifications. Multidetector computed tomography (MDCT) suggestive of about 8×7.5×6 cm well-defined cystic lesion with multiple thick septae with calcification within it seen in left adnexa likely to be teratoma. Patient was posted for exploratory laparotomy. Total abdominal hysterectomy with bilateral salphingoopherectomy with partial dissection of retroperitoneal mass was done. Histopathology report suggested microscopic examination as ovarian tissue is replaced by variably sized thick and thin vascular channels and blood vessels. These vascular channels have partially denuded endothelial lining. The vascular channels are filled with frank haemorrhage suggestive of ovarian haemangioma. Ovarian haemangiomas sometimes coexist with genital tract diseases or even malignancies and thus ovarian haemangioma can be clinically significant.

Keywords: Ovary, Haemangioma, Vascular tumours, Adnexal Mass, Cystic mass

INTRODUCTION

Ovarian haemangiomas are benign and rare vascular tumours of female genital tract with less than 50 reported cases in the literature.[1-3] These neoplasms occur in both adults and children with the age ranging from infancy to postmenopausal women. Most ovarian haemangiomas are incidental finding in surgery for other causes or autopsy.[4-6] Larger or multiple lesions produce symptoms of which abdominal pain is the most common complaint. Ovarian haemangiomas are “non-functional" neoplasms. However, stromal luteinization has been observed some cases but exact pathogenesis is not known.[1]

Histologically, ovarian haemangiomas are of the cavernous, capillary or mixed types. Most ovarian haemangiomas are of the cavernous type.[7] Microscopic examination reveals numerous thin-walled dilated blood channels lined by single layer of endothelial cells arranged haphazardly in the ovarian parenchyma or forming nodule which can contain thrombin. There can be small capillary sized blood vessels with a larger feeding vessel.

Occasionally there can be presence of variable amount of ovarian tissue stroma showing inflammation, haemorrhage, hemosiderin deposits or calcification. Ovarian haemangiomas sometimes coexist with genital tract diseases or even malignancies and thus ovarian haemangioma can be clinically significant.[8]

Here, we report a case of ovarian cavernous haemangioma which manifested as a large growing mass in a postmenopausal 77-year-old female, and diagnosed on histopathological examination.

CASE REPORT

A 77-year-old woman presented with pain in lower abdomen and discomfort along with bloating sensation since 1 month. Pain was dull in nature, non-radiating more towards left side. No history of weight loss, vaginal discharge or bleeding per vaginum. No other significant medical or surgical illness in past. Her menstrual history and obstetric history was uneventful with menopause attained 15 years back. On physical examination, per abdomen was palpable mass in lower abdomen of approximately 20 weeks size and immobile. Tenderness present on left pelvic region on deep palpation. On per speculum examination, cervical erosions present, no other significant changes. On bimanual pelvic examination, uterus small in size. Firm mass palpable through left fornix upto 20 weeks, immobile, with tenderness present.

Ultrasonography showed uterus is normal in size and echotexture measuring 4.4×2.9×2.8 cm (post-menopausal). Retroverted and retroflexed in position. Normal central endometrial echo measuring 3.7 mm. Both ovaries are not visualised. A large mass is seen the left hemi pelvis measuring 7.0×5.4 cm. Mass is heterogeneous n echotexture and shows e/o multiple cystic areas and multiple calcific foci. No neovascularisation seen within mass. No free fluid notes.

MDCT suggestive of about 8×7.5×6 cm well-defined cystic lesion with multiple thick septae with calcification within it seen in left adnexa. Left ovary is not separately visualised. The left lower ureter is displaced medically by the lesion. No hydronephrosis seen. The fat plane with the rectum and sigmoid colon is maintained. It abuts the left internal and external iliac vessels. No significant lymphadenopathy seen with Possibility of neoplastic aetiology like teratoma is likely (Figures 1-4).

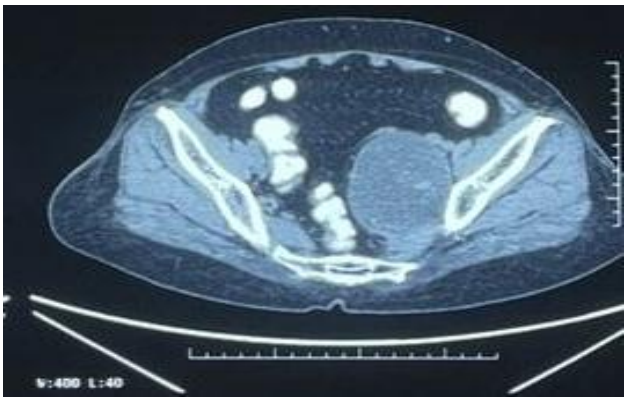


Figure 1: Computed tomography plane images revealed 8×7.5×6 cm sized, well-circumscribed hypodense mass lesion in left adnexa, left ovary not separately visualised. Surrounding fat plane appears normal. No evidence of surrounding soft tissue/bone invasion.

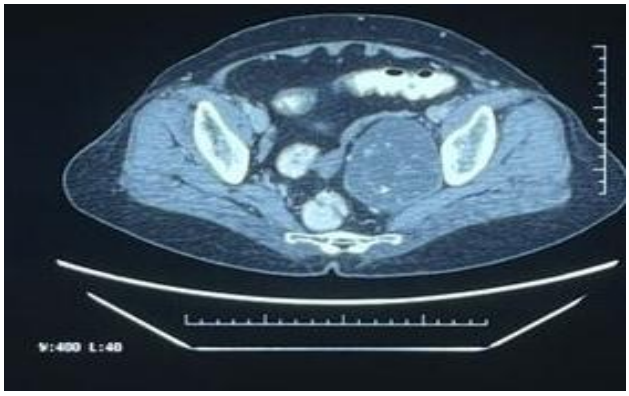
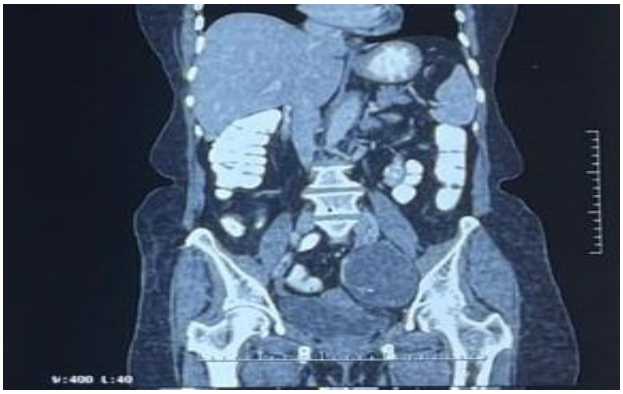


Figure 2: On contrast images, arterial phase Few thin enhancing septations are seen. Wall of mass lesion also shows enhancement. No solid enhancing component is seen.



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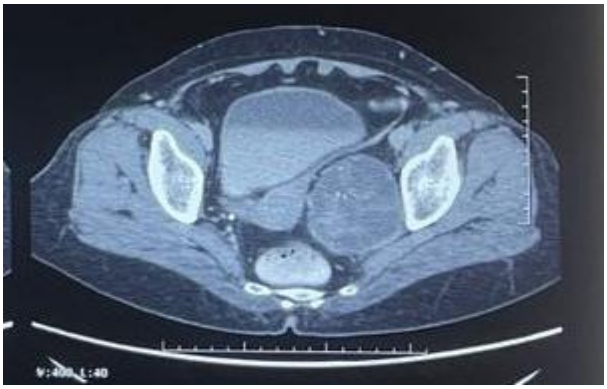

Figure 4: On contrast images, venous phase delayed enhancement is seen.

Routine haematological and biochemical parameters were within normal limits. Tumour markers which included CA 125, CA 19-9, CEA, AFP, b-HCG were within normal limits. Lactate dehydrogenase (LDH) was 224 U/l which is mildly raised. PAP smear report suggested of ascus.

Decision for exploratory laparotomy taken and patient was posted. On exploration, retroperitoneal bulge noted on left pelvic side. Retroperitoneal dissection done and mass exposed which was located just below common iliac vessels. Mass was of size approximately 8×6×5 cm, smooth, grey white with cystic consistency. Uterus with bilateral ovaries were atrophic. Peritoneal wash given and sent for cytology. Total abdominal hysterectomy with bilateral salphingoopherectomy done and sample sent for HPR. Cyst fluid aspirated and sent for cytology. On mobilising the left ureter and prophylactically ligating left internal iliac vessels, attempt to remove whole mass was made. Exact extent of mass was not delineated. It was found to be adherent to sacrum, internal iliac vessels and lateral pelvic wall. Cyst was removed partially in fragments and sent for histopathology. With attempts to remove more mass, excessive bleeding was present from retroperitoneal vessels hence decision to stop further dissection was made. 3 PCV transfusion was given intraoperatively.

On gross examination irregular fragments, part of ovary measures 4×2×0.5 cm. It is dark red to blackish in colour. Other ovary measures 3×1.5×0.5 cm. The uterus with cervix measures 6×2.5×2 cm and weighs 30 grams. The cervical length is 3.5 cm. On cut section, the endometrium is 0.1 cm and myometrium is 1.3 cm. One fallopian tube measures 3 cm and other fallopian tube measures 4 cm.

On microscopic examination, Sections through uterus show atrophic endometrium. There is no cytoarchitectural atypia. Sections through myometrium show foci of atrophic endometrial stroma; suggesting possibility of old adenomyosis. Sections through cervix show features of chronic polypoidal cervicitis with extensive squamous metaplasia. There are proliferated, hyperplastic endocervical glands seen in the cervical stroma. These glands are filled with secretions. Sections through both the tubes show presence of normal tubal lumina. Tubes show para tubal cyst formation.

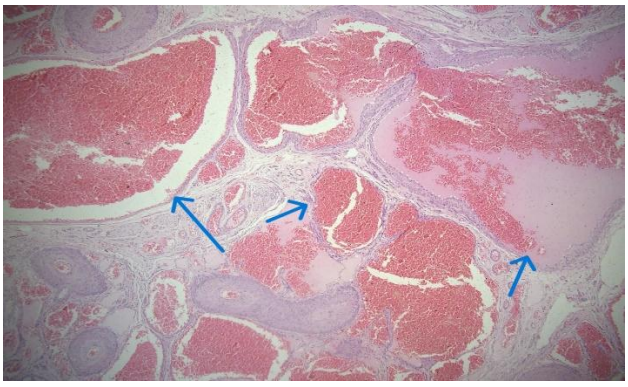

Figure 5: Variably sized dilated vascular channel filled with blood.

Sections through dark red to brown ovary (on gross examination) show only part of ovary. There is a compressed ovarian stroma at periphery with few corpus alibicans. Rest of ovarian tissue is replaced by variably sized thick and thin vascular channels and blood vessels. These vascular channels have partially denuded endothelial lining. The vascular channels are filled with frank haemorrhage. There are few foci of aggregation of hemosiderin laden macrophages; indicating old haemorrhages, these features are suggestive of benign ovarian haemangioma. There is no atypia. Sections through the other ovary show corpus albicans and compressed ovarian stroma. There are congested vessels and extravasation of RBCs seen in the ovarian parenchyma. Sections through the left pelvic node and omental tissue show areas of haemorrhages, few dilated and congested vessels in the adipose tissue.

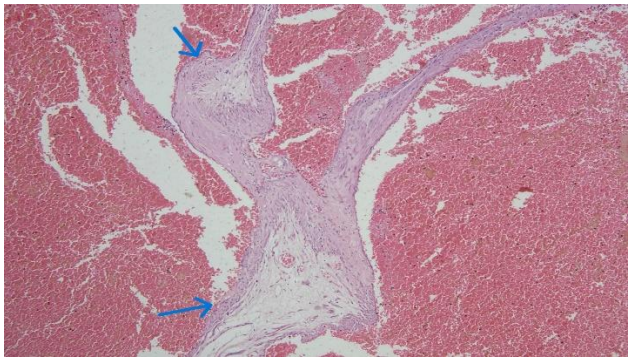

Figure 6: Dilated vascular channel lined by endothelial cells.

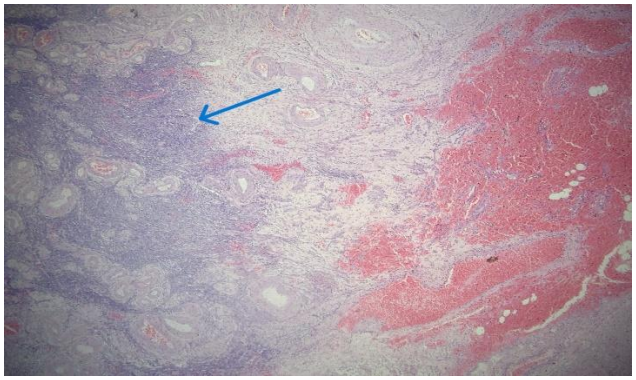

Figure 7: Compressed ovarian stroma.

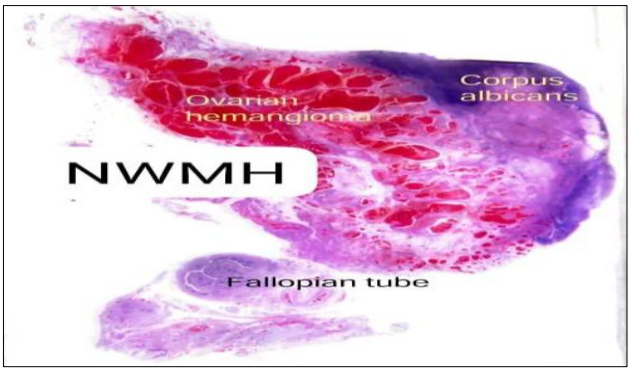

Figure 8: Microscopic view of ovarian haemangiomas.

Three small lymph nodes identified in the left pelvic lymph node and omental tissue, show features of reactive lymphoid hyperplasia. Few areas of haemorrhages are also seen in the lymph nodes. No granulomas or atypia seen in the lymph node.

DISCUSSION

Haemangiomas are highly vascular tumours and in ovary finding is very rare. Payne et al in 1869 first described bilateral ovarian haemangioma in a 25-year-old female co-existing with abdominopelvic hemangiomatosis.[9] Ovarian haemangiomas can occur at any age with wide range of 4 months to 81 years. In this case report, age was female was 77 years old. Grossly the size of ovarian haemangioma has wide range measuring 0.3 cm to 24 cm and is usually unilateral with occasional bilateral cases.[1,10] Most ovarian haemangiomas are asymptomatic and small. In present case, the tumour was unilateral presented with pain in abdomen with USG suggestive of a large mass is seen the left hemi pelvis measuring 7.0×5.4 cm. Larger ovarian haemangiomas can present with abdominal distension due to mass or abdominal pain and vomiting due to torsion.[11] Cases of ovarian haemangioma presenting with ascites and pleural effusion (pseudo-Meigs' syndrome), thrombocytopenia (Kasabach Merritt syndrome) and elevated CA 125 mimicking ovarian surface epithelial tumours have also been reported.[6,12,13] In present report, none of the above findings was seen in investigations, including USG, CT scan and lab data.

Al-Shaikh et al described that ovarian haemangioma may synchronously occur in patients with ovarian neoplasm such as mature cystic teratoma of contralateral ovary, papillary serous carcinoma and mucinous cystadenoma as well as non- ovarian neoplasms such as hyperplasia, polyp or carcinoma of endometrium, cervical carcinoma, tubal carcinoma or rectosigmoid carcinoma.[13] In present report, our case mimics ovarian teratoma as suggested in MDCT.

The exact pathogenesis and aetiology of ovarian haemangiomas is not known and different theories have been proposed to explain its etiopathogenesis. Some states that ovarian haemangioma as true tumour, hamartoma, or congenital malformation. Some authors described hormonal factors, pregnancy and infection play important role in growth of these tumours.[14] Estrogen has a growth stimulatory effect on vasculature. Due to pre-existing stromal luteinisation from stromal hyperplasia or hyperthecosis, there is hyperestrogenism which has been implicated for pathogenesis of ovarian haemangioma by some authors.[15,16] Study by Miliars et al have found endothelial cells in ovarian haemangiomas to be immunopositive for estrogen receptor (ER) and progesterone receptor (PR). The neoplastic endothelial cells can secrete stroma stimulating substance leading to stromal luteinisation. The luteinized stromal cells produce androgen and subsequently estrogen in adipose tissue leading to hyperandrogenism and hyperestrogenism features in few reported cases.[13,17,18] However, those were not seen in our case report.

Radiological features of ovarian haemangioma by Yamawaki et al have been proposed like complex mass with solid and cystic component on computed tomography (CT) scan and marked enhancement pattern with contrast enhanced T1 weighted images on magnetic resonance imaging (MRI) scan, however these findings are nonspecific.[5] The present case MDCT suggestive 8×7.6×6 cm well defined cystic lesion with multiple thick septae with calcification within it seen in the left adnexa likely to be teratoma of neoplastic aetiology. MRI was not done in our case which would have been more appropriate to differentiate haemangiomas. Our present case, USG suggestive of multiple calcifications.

The differential diagnoses of ovarian haemangiomas are angiosarcomas, lymphangiomas, proliferating hilar blood vessels, anastomosing haemangioma, degenerated uterine or intraligamentous myoma, or an ovarian tumour like fibrothecoma, Brenner tumour and mature or immature teratoma. The main differential diagnoses of ovarian haemangioma pathologically are closely packed ovarian medullary blood vessels and proliferating hilar blood vessels. Careful sampling is important to exclude the presence of teratomatous elements before diagnosing the tumour as a pure haemangioma as teratomas have prominent vascular components.[12,13] Our case presented with vascular channels with partially denuded endothelial lining. The vascular channels are filled with frank haemorrhage with few foci of aggregation of hemosiderin laden macrophages; indicating old haemorrhages. No presence of pale eosinophilic homogeneous material within the vascular channels ruled out lymphangioma. Features of angiosarcoma like increased mitosis, cytological atypia and necrosis was not seen. Haemangioma should be considered in the differential diagnosis when a growing adnexal mass or a haemorrhagic ovarian lesion is encountered in postmenopausal women.[7]

CONCLUSION

Ovarian haemangiomas are rare benign vascular tumour incidentally discovered during surgery. It should be considered for differential diagnosis with haemorrhagic lesions. Although surgical removal of the involved areas is treatment of choice, a correct preoperative and postoperative diagnosis is necessary to avoid unnecessary radical surgery and radiotherapy and chemotherapy. As ovarian haemangioma can be associated with other non-ovarian neoplasm and hemangiomatosis, a thorough evaluation of contralateral ovary, endometrium for malignancy and abdominopelvic areas for hemangiomatosis are essential.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Pawar AP, Garude SG, Patwardhan LM, Solanki VP. Ovarian hemangioma: rare case report and review of literature. Int J Reprod Contracept Obstet Gynecol 2025;14:913-7.