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CASE REPORT

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Cavernous hemangioma of corpus imitating endometrial polyp in a young non-pregnant woman: A case report study

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Key Clinical Message

Cavernous hemangiomas are rare and have various non-specific clinical presentations, such as menorrhagia. It can mimic different diseases such as endometrial polyps. Pathologists and gynecologists should be aware of performing histopathological examinations of this neoplasm for accurate diagnosis and to avoid unwarranted therapeutic interventions.

Abstract

Internal genital tract hemangiomas are rare and can be divided into capillary and cavernous. We present a rare case of cavernous hemangioma (CH) of the corpus in a young, non-pregnant woman. The patient was a 28-year-old woman who had complained of menorrhagia for 2.5 years. Sonography showed a hypoechoic intramural area measuring 35 × 23 mm. Histomorphology revealed neoplastic proliferation of dilated thin-walled arteries of various sizes within the myometrium. Due to the variable clinical presentations of CH, histopathological examination should be performed for an accurate diagnosis. It is a rare entity and we recommend training pathologists and gynecologists on this neoplasm for accurate diagnosis and to avoid unwarranted therapeutic interventions.

KEYWORDS

cavernous hemangioma, myometrium, uterus, vascular lesion

1 | INTRODUCTION

Vascular tumors in the female genital tract are rare and only about 140 cases have been reported in the literature with an age distribution of 5–81 years.[1] They can be divided into arteriovenous malformations, cavernous and capillary hemangiomas, lymphangiomas, hemangioendotheliomas, and angiosarcoma. These tumors can be either single or multifocal.[2] Vascular tumors of the female genital tract are located in the cervix, vagina, vulva, fallopian tubes, ovaries, and placenta and rarely in the uterus.[3]

Hemangiomas of the female genital tract have several clinical presentations. If small, they can be asymptomatic and discovered by chance in cases of overt abnormal uterine bleeding, postpartum hemorrhage, dyspareunia, or postcoital bleeding.[4] When they become larger, they can mimic gynecological complaints such as AUB.[5] Genital hemangiomas are located in the female genital tract or pelvic organs. Pelvic hemangiomas are associated with hereditary diseases.[3,5]

The first uterine hemangioma was reported in a woman during a postpartum examination in 1897.[6]

Hemangiomas are benign neoplasms derived from uterine mesenchymal cells.[7] These lesions can be classified as congenital or acquired. Congenital forms are associated with diseases such as hereditary hemorrhagic telangiectasia, tuberous sclerosis, Klippel–Trenaunay syndrome, Maffucci syndrome, Kasaback–Merritt syndrome, and blue rubber bleb nevus syndrome.[8] The acquired type is more common and is caused by physical injuries, including pregnancy and curettage, hormonal therapy, carcinomas, and consumption of oral contraceptive pills.[3,9,10]

The most common symptoms of hemangiomas are dysmenorrhea, menorrhagia, infertility, and pelvic pain.[5] They can appear and be incidentally detected during pregnancy. This can result in obstetric difficulties during pregnancy, including preterm rupture of the membrane, postpartum hemorrhage, disseminated intravascular coagulation (DIC), and even death.[11]

Ultrasound and color Doppler imaging followed by MRI can confirm the diagnosis of a hemangioma as a vascular lesion. On ultrasound, a thickened wall of the uterus with a hypoechoic space can help in diagnosis.[12] In color Doppler imaging, a low velocity or no flow may be reported. Hemangiomas are hypointense or isointense on T1 cuts and isointense on T2 cuts on MRI, depending on the number of venouses.[13] Ultrasound-guided biopsy can be used for the histological diagnosis of soft tissue hemangiomas if typical clinical and the US signs are inadequate. Specifically for women of reproductive age, as this may help avoid unnecessary hysterectomies.[8]

The underlying pathogenesis of CH is not yet fully understood and requires further research. Here, we present a rare case of uterine CH in a 28-year-old Iranian nonpregnant woman and review the associated literature. The study was approved by the institutional review board and the ethics committee. Informed consent was obtained from the patient. All applicable international, national, and institutional guidelines were followed in this study.

2 | CASE REPORT

The patient was a 28-year-old non-pregnant woman with a history of three pregnancies with the outcome of three live children complaining of abnormal uterine bleeding in the form of menorrhagia for 2.5 years. She was admitted to the emergency room of the Kowsar Teaching Hospital (Qazvin, Iran) on December 2018. The patient did not have any underlying disorders. The patient had no history of surgery in her life. The patient's drug and family disease history were negative. Physical and speculum examinations revealed no abnormalities. Laboratory results revealed mild anemia (Hb: 11.6 mg/dL), exclusively. The beta-human chorionic gonadotropin (hCG) level was negative (<2 mLU/mL).

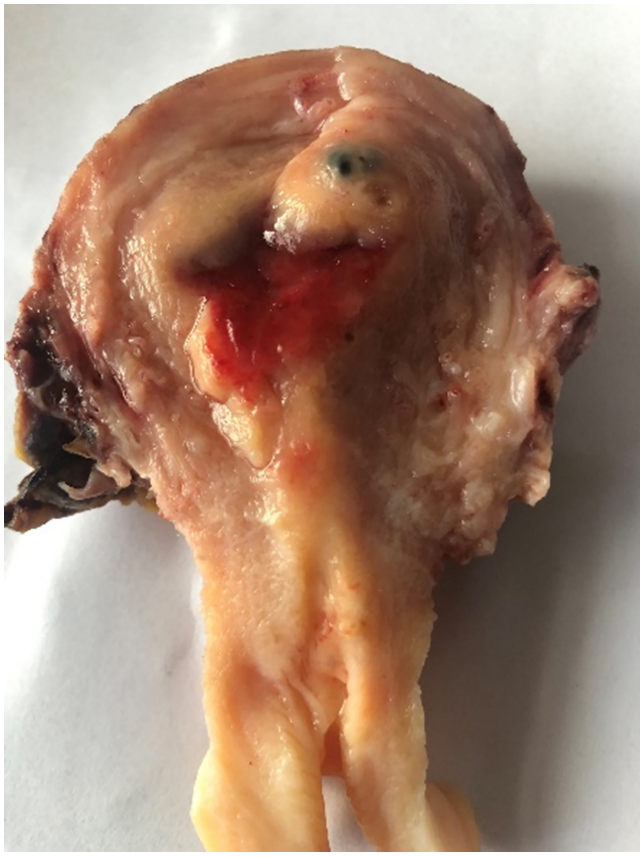
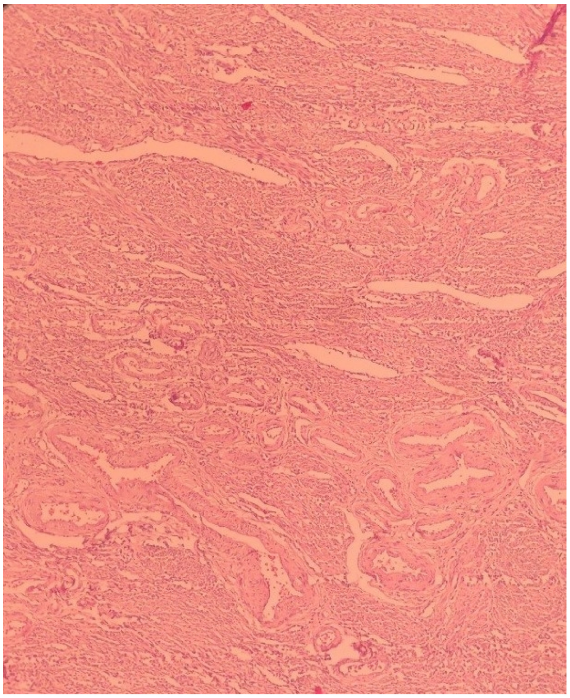


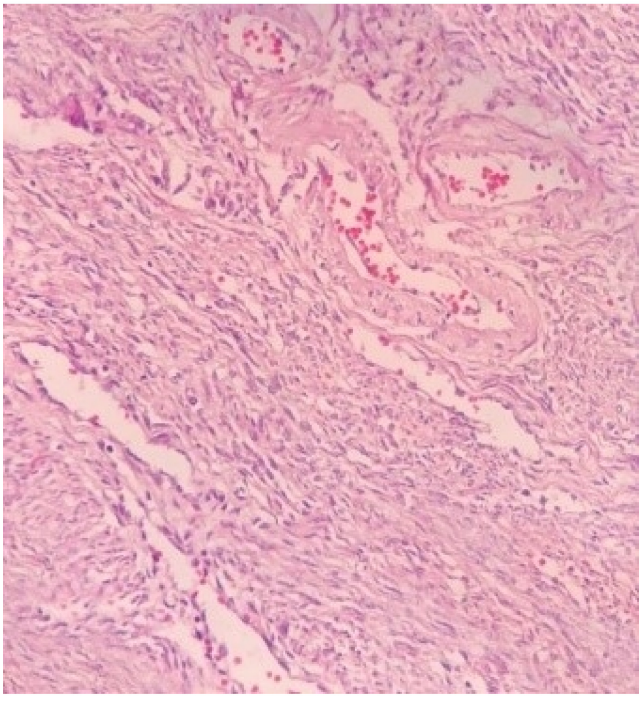
FIGURE 1 Macroscopic view of total hysterectomy specimen.

Abdominal ultrasonography revealed a hypoechoic area (M: 34 × 27 mm) with definite boundaries in the endometrial cavity. In transvaginal sonography, a hypoechoic area, measuring 35 × 23 mm was observed which was attached to the anterior wall of the endometrium. The primary differential diagnoses were endometrial polyps, submucosal leiomyomas, and blood clots. As an endometrial polyp was the highest probability, the patient underwent dilation and curettage. The biopsy was performed through dilation and curettage. However, her menorrhagia did not stop, and as she had three children she did not want any more children, so she underwent a supracervical hysterectomy (Figure 1).

Macroscopic examination revealed a localized, enlarged area of the corpus protruding into the endometrial cavity. The cut sections showed an irregular intramural lesion measuring 3 × 3 × 1.8 cm protruding into the endometrial cavity. Histomorphological examination revealed mid-proliferative endometrium. The underlying myometrium is occupied by neoplastic proliferation of variously sized dilated thin-walled arteries lined by bland-looking flat endothelial cells and containing erythrocytes (Figure 2A,B). The final diagnosis of CH was made. There



(A)



(B)

FIGURE 2 (A) Histomorphology results neoplastic proliferation of various sized dilated thin wall arteries. (B) Vascular channels lined by flat endothelial cells and containing erythrocytes. 100× (A) 400× (B), Hematoxylin & Eosin stain.

was no evidence of malignancy based on the histomorphological criteria of the examined slides. The patient has been free of any disease for the following 25 months.

3 | DISCUSSION

The histopathological findings in our case were consistent with those of CH. Histomorphological examination revealed a mid-proliferative endometrium and myometrium with neoplastic proliferation of various-sized dilated thin-walled arteries lined by bland-looking flat endothelial cells containing erythrocytes.

Uterine hemangiomas are rare tumors that occur in the fourth and fifth decade of life.[1] Numerous syndromes are associated with congenital hemangiomas, and their pathogenesis is characterized by embryonic sequestration. However, hormonal changes play an important role in the pathogenesis of acquired hemangiomas.[3] Estrogen increases endothelial progenitor cells and angiogenic factors such as matrix metalloproteinase, vascular endothelial growth factor, and nitric oxide.[14] Other angiogenic factors that increase with estrogen include basic fibroblast growth factor, insulin-like growth factor, and transforming growth factor-β.[15] Our patient was 28 years old and had no family history, which indicated that her CH had been acquired, and her past three pregnancies could have caused this lesion in the uterus. Other case reports of these lesions in the uterine cavity have also demonstrated an acquired pathogenesis.[3,8,16]

The most reported symptoms of hemangioma are vaginal bleeding, infertility, dyspareunia, postcoital bleeding, and even clinically mimicking endometriosis.[4] According to the literature, approximately half of the patients had abnormal uterine bleeding.[1] Our patient was also symptomatic and had abnormal uterine bleeding (menorrhagia) for 2.5 years.

Ultrasonography is the first step in the evaluation of gynecological problems. Our patient's ultrasonography findings showed a thickened uterus with a hypoechoic area in the endometrium. These findings have been described in other case reports of CH during pregnancy.[12,17]

Hemangiomas are mainly divided into two types: capillary and cavernous. The capillary polyps are detected as nodular or localized endometrial polyps. The cavernous types diffuse with dilated blood spaces. Diffusely dilated blood vessels filled with blood are the most common diagnostic characteristics of CH.[18] The clinical findings in our case demonstrated endometrial polyps and histomorphological examination revealed cavernous vessels with thin walls that were far away from the myometer vessels.

Differential diagnoses have various possibilities. Angiomatosis manifests as a proliferation of vessels of diverse calibers that displace the adjacent muscle and fat. Lymphangioma, on the other hand, reveals itself as large lymphatic channels in loose connective tissue stroma. These channels contain mature lymphocytes. Adenomatoid tumors with numerous irregularly shaped

anastomosing vascular channels present a highly infiltrative architecture and poor demarcation. The tumor cells are typically plump, pleomorphic, and mitotically active. Lastly, angiosarcomas are characterized by tubules, cords, or small nests that are lined with bland cuboidal cells.[19] In complicated cases besides morphology, we can use immunohistochemistry of CD34, CD31, and von Willebrand factor to rule out adenomatoid tumors.[3]

Hysterectomy is the best treatment choice in most cases, but conservative treatment may also be considered, such as cryotherapy, laser ablation, radiotherapy, uterine artery embolization, knife excision, and local excision.[20,21]

4 | CONCLUSION

Uterine hemangiomas are rare and have various symptoms ranging from asymptomatic to gynecological and obstristic complications. For a definitive diagnosis, histopathological examination should be performed, and we recommend training pathologists and gynecologists on this neoplasm for accurate diagnosis and to avoid unwarranted therapeutic interventions.

AUTHOR CONTRIBUTIONS

Ali Emami: Investigation; writing – original draft; writing – review and editing. **Ensiyeh Bahadoran:** Investigation; writing – original draft; writing – review and editing. **Fatemeh SamieeRad:** Conceptualization; methodology; project administration; visualization; writing – review and editing.

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CONFLICT OF INTEREST STATEMENT

The authors declare that they have no conflicts of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no new data were created or analyzed in this study.

ETHICS STATEMENT

The study was approved by the institutional review board and ethics committee. All applicable international, national, and/or institutional guidelines were followed and participant consent was obtained.

CONSENT

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

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