Giant Uterine Haemangioma similar to Myoma Nascendi Clinic: a case report

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Abstract

Haemangiomas of the genital tract are extremely rare. Most genital tract haemangiomas are smaller than 2 cm, asymptomatic and detected incidentally. The majority of symptomatic haemangiomas are detected as a result of severe obstetric bleeding during or after pregnancy. In this case report, a case of giant uterine haemangioma in the vagina accompanying acute vaginal bleeding and severe pelvic pain in a non-pregnant woman, which is rarely observed in the literature, is discussed.

Introduction

Haemangiomas are tumours originating from vascular endothelial cells and are frequently seen in the oral cavity, kidneys, skin and liver. They are rarely observed in the female genital tract (1). In the female genital system, haemangiomas originate in the cervix, vagina, vulva, fallopian tubes, ovaries, placenta and rarely in the uterus (2). It can be seen as single or multifocal (3). Haemangiomas of the genital tract occur in women of all ages between 5 and 80 years, including pregnant women (4). In the literature, the first uterine haemangioma was described in 1897 in the autopsy of a young mother who died 24 hours after giving birth to twins due to post partum haemorrhage (5). The incidence of uterine haemangioma is not known with certainty (6). Haemangiomas of the female genital tract may present with different clinical pictures due to differences in size, localisation and growth pattern. Although most of them are asymptomatic, in some cases they may cause a life-threatening clinical picture such as dyspareunia, post-coital haemorrhage, post partum haemorrhage and even massive abnormal uterine bleeding and should be kept in mind in the differential diagnosis (7). In the literature, cases of genital haemangioma causing severe haemorrhage are usually diagnosed during pregnancy or post partum period bleeding (8,9). İncreased hormones during pregnancy have been blamed for the excessive bleeding and large-sized haemangiomas seen in these cases (10).

In our case report, we discuss a giant uterine haemangioma in the clinic of myoma arising in the vagina causing massive abnormal uterine bleeding in a 39-year-old non-pregnant woman, which is very rarely observed in the literature.

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Case Presentation

A 39-year-old woman with a history of gravida 4, parity 3, 1 D/C. The patient was admitted to the gynaecology clinic because of long-standing abnormal uterine bleeding and pelvic pain. She stated that her bleeding and pelvic pain had been increasing for a week and both the bleeding and pain had intensified for the last 2 days. On physical examination, the uterus was approximately 12 pregnancy weeks in size and there was active vaginal bleeding and severe pelvic pain. There were no abnormal laboratory findings except Hb: 8.2 and Hct: 27.7%. General condition was moderate, hypotensive and slightly tachycardic. Vaginal examination revealed active vaginal bleeding and a limited mobile, semisolid mass originating from the lower segment of the uterus and filling the vagina completely, consistent with the clinic of myoma arising into the vagina. Pelvic USG showed a mass filling the vagina and multiple fibroid nodules in the uterus, the largest of which was 8x6 cm. The patient was hospitalised due to active bleeding and severe pelvic pain. Because of her clinical findings due to acute blood loss, 2 units of ERT transfusion was performed. After blood replacement, she was taken to emergency operation with a prediagnosis of myoma uteri nassendi without waiting for other imaging methods. Under general anaesthesia, we completely excised the vaginal mass extending into the uterine cavity through the vaginal route (figure 1). After the excision of the mass, the bleeding stopped and the pain disappeared. The operation for the other fibroid nodules covering the uterus was planned to be performed under elective conditions according to the pathological results of the mass taken from the vagina. Post operative active haemorrhage was not observed and the mass was sent to the pathology department. In the examination performed in the pathology clinic, the entire mass was reported as giant cavernous haemangioma (Figure 2).

After stabilisation of the patient, a contrast-enhanced MRI was performed to detect possible haemangiomas in the uterus or other organs. MRI revealed no pathology except for fibroids observed in the uterus similar to pelvic USG. After the patient was discharged, she continued to have dysfunctional uterine bleeding although she did not have active vaginal bleeding. Upon the patient's request for surgery and hysterectomy, hysterectomy was performed with the prediagnoses of myoma uteri, dysfunctional uterine bleeding and other undetected haemangioma nodules in the uterus. There were no intraoperative or postoperative complications. The pathology clinic was informed about the patient's past medical and pathological history. While adenomyomas were detected in the hysterectomy material, no other haemangioma focus was found.
No pathology was observed at the 1st and 6th month controls.

Conflict of interest

None declared

Informed consent of the patient

In this case report, the patient was informed in detail that his/her case could be published in a scientific study and gave written consent for the use of his/her images. The study was conducted in accordance with the Declaration of Helsinki.

Discussion

Haemangiomas are belign tumours generally characterised by increased number and size of vessels (9). Haemangiomas may be observed congenitally as part of a syndrome (11) or may be acquired due to a history of dilatation curettage, pelvic surgery, pregnancy, endometrial carcinoma, trophoblastic disease or maternal diethylstilbesterol (DES) intake (12). Although the etiology of haemangiomas is not clearly known, there are studies suggesting that estrogen causes haemangiomas by stimulating angiogenesis through angiogenic factors including vascular endothelial growth factor and nitric oxide (2). In another study, estrogen receptors detected in endothelial cells were held responsible for haemangiomas (13). While congenital genital haemangiomas are observed at younger ages, acquired uterine haemangiomas are frequently observed in women aged 40-50 years (4). Our case was 39 years old with a history of 4 pregnancies, 3 labour and 1 dilatation curettage. She had no history of abnormal bleeding in any pregnancy, during pregnancy or after delivery. There was also no family history. This suggests that our case was an acquired haemangioma.

Half of uterine haemangiomas are asymptomatic (14). In the other half, when pregnant women and post partum patients were excluded, the most common symptoms were observed as non-excessive abnormal uterine bleeding, dyspareunia, abdominal pain and chronic anaemia (15). Our patient presented with severe bleeding and severe pelvic pain. In the vaginal examination of our patient, approximately 10-12 cm, stemmed myoma originating from the uterine endometrial cavity and arising into the vagina was macroscopically surrounded by a capsule-like structure on the outer surface, filling the entire vagina. Unlike other cases in the literature, our case had anaemia findings due to active vaginal bleeding and severe pelvic pain due to torsion of the mass in the vagina.

As in other pelvic masses, the primary method for the diagnosis of uterine haemangiomas is abdominal or transvaginal ultrasonography (16). Doppler USG is also important in the diagnosis, especially in haemangiomas showing large arterivenous malformations. Contrast-enhanced MRI may be preferred in stable cases which cannot be diagnosed by USG because of its sensitivity in the detection of haemangioma (17). It should also be kept in mind that 3D CT angiography, if available, is a diagnostic method that can be used in unstable cases in which time loss cannot be tolerated (18).

In our case, pelvic USG showed a mass filling the entire vagina and multiple fibroid nodules in the uterus, the largest of which was 8 cm in size. Elective conditions were waited for other imaging modalities. After excision of the mass, contrast-enhanced MRI of our patient did not show any structure compatible with haemangioma in another focus similar to pelvic USG. Similar to the imaging methods, no other haemangioma focus was found in the pathological examination of the uterus after hysterectomy performed under elective conditions.

The clinical significance of our case is that a haemangioma of this size filling the entire vagina in the clinic of myoma uteri nascendi has not been described in the literature. Although it is rarely observed in patients presenting in this clinic, we think that it is important to consider giant haemangiomas in the differential diagnosis. In our case, we excised the mass completely, but it should be kept in mind that emergency operations performed without knowing the exact diagnosis may lead to severe unstoppable bleeding, emergency hysterectomy or mortality in cases where the entire mass cannot be removed or the mass ruptures during vaginal operation. Therefore, we believe that preoperative blood preparation, warning of the anaesthesia team and

informing the patient about possible complications and obtaining her consent are important in similar vaginal masses.

Conclusion

Genital system haemangiomas are rare pathologies. They are often small in size and asymptomatic. The majority of symptomatic cases present with bleeding during or after pregnancy. Rare genital tract haemangiomas observed without pregnancy may present with clinical findings such as dysfunctional uterine bleeding, dyspareunia, pelvic pain and post-coital bleeding. Although giant uterine haemangiomas filling the vaginal cavity completely as in our case are observed very rarely, they should be considered in the differential diagnosis. In these cases, preoperative medical preparations should be planned multidisciplinarily. Especially in young patients, all complications, including the possibility of hysterectomy, should be explained preoperatively and consent should be obtained.

Author Contributions

Asst. Prof Alihan Tığlı, Assoc.Prof.Dr Yakup Baykuş Performed the diagnosis and treatment of the case, and drafted the initial manuscript. Prof.Dr Rulin Deniz, Asst. Prof Nazlı Şener Conducted the literature review and contributed to the writing process. Dr. Erdem Gürkan, Dr. Güzide Ece Akıncı. Prepared the visual materials and managed the ethics committee approval process. Assoc.Prof.Dr Yakup Baykuş, Assoc.Prof.Dr Rulin Deniz, Asst. Prof Alihan Tığlı Reviewed and edited the final version of the manuscript for scientific accuracy.
All authors have read and approved the final version of the manuscript and agree with its submission for publication.

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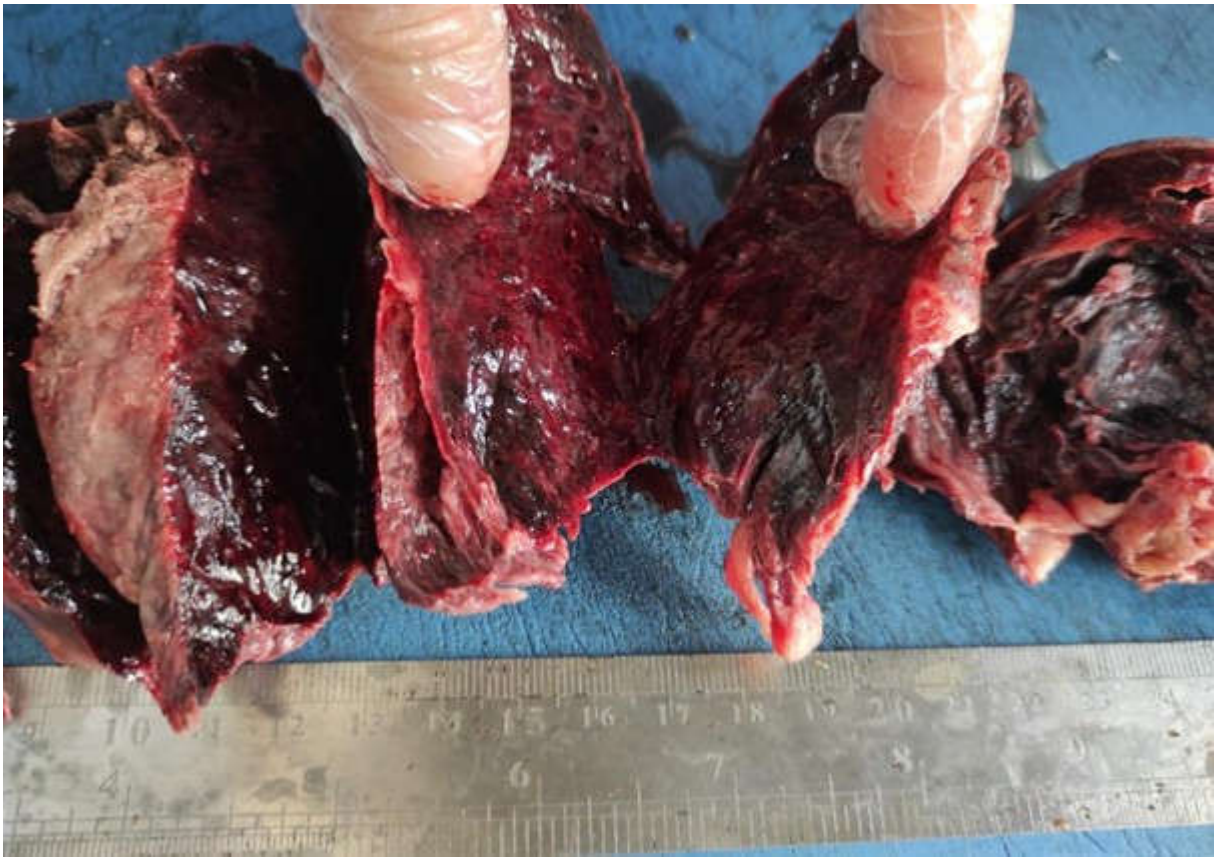

Figure 1: Macroscopic view of hemangioma

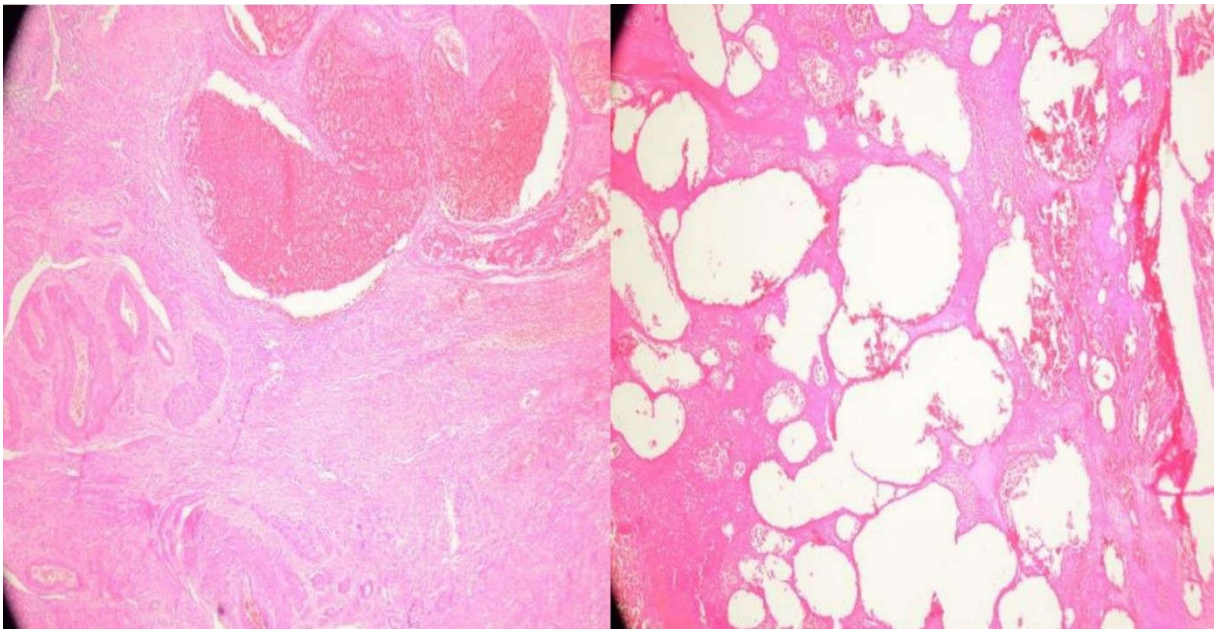

Figure 2: Microscopic image of haemangioma