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Case Report

Fallopian Tube Hemangioma Discovered on Follow-up for Uterine Leiomyoma

Uterus Leiomiyomu Takibinde Bulunan Fallop Tüpü Hemanjiyomu

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ABSTRACT

Hemangioma in female reproductive organs, particularly in the fallopian tube (FT), is a sporadic disease. In this report, we describe a case of hidden capillary hemangioma in FT in a 39-year-old woman who suffered from uterine leiomyoma. During the preoperative stage, pelvic sonography, computed tomography, and diagnostic laparoscopy revealed a subserous leiomyomatous nodule located along the posterior wall of the uterus. Despite this, intraoperatively, a benign vascular neoplasm was diagnosed. Histologically, it is characterized by multiple thin-walled vascular spaces lined with a single layer of endothelial cells, in which single mitoses were observed. The diagnosis was then confirmed immunohistochemically by CD31 and CD34 expression in the endothelial cells lining the inner surface of the spaces and the low mitotic activity of the tumor cells. It is virtually impossible to diagnose this asymptomatic neoplasm before and during surgery, which can result in an inadequate number of surgeries. Incorrect interpretation of a benign tumor at a young age can lead to unnecessary radical surgery with a resulting loss of fertility, and an unrevealed malignant process can threaten life.

Keywords: Fallopian tube hemangioma, hemangioma, capillary hemangioma

ÖZ

Kadın üreme organlarında ve özellikle fallop tüpünde (FT) hemanjiyom sporadik bir hastalıktır. Bu yazıda, uterin leiomiyomu olan 39 yaşında bir kadın hastada FT'de gizli kapiller hemanjiyom olgusu sunulmuştur. Ameliyat öncesi pelvik sonografi, bilgisayarlı tomografi ve tanısal laparoskopi, uterusun arka duvarı boyunca yerleşmiş subseröz leiomiyomatöz bir nodülü ortaya çıkardı. Buna rağmen, intraoperatif olarak benign vasküler neoplazm tanısı konuldu. Histolojik olarak, tek mitozların gözlendiği tek bir endotelyal hücre tabakasıyla kaplı çoklu ince duvarlı vasküler boşluklarla karakterizedir. Tanı daha sonra boşlukların iç yüzeyini kaplayan endotel hücrelerindeki CD31 ve CD34 ekspresyonu ve tümör hücresinin düşük mitotik aktivitesi ile immünohistokimyasal olarak doğrulandı. Bu asemptomatik neoplazmın tanısını ameliyattan önce ve ameliyat sırasında koymak neredeyse imkansızdır ve bu da yetersiz ameliyat hacmine neden olabilir. Genç yaşta iyi huylu bir tümörün yanlış yorumlanması, doğurganlık kaybıyla sonuçlanan gereksiz radikal cerrahiye yol açabilir ve ortaya çıkarılmamış bir malign süreç hayatı tehdit edebilir.

Anahtar kelimeler: Fallop tüpü hemanjiyomu, hemanjiyom, kapiller hemanjiyom

INTRODUCTION

Fallopian tube (FT) tumors occur quite rarely compared with other female reproductive organ neoplasias. The incidence of FT cancer is no more than 1.2-1.8%, and benign neoplasms occur even less frequently[1,2]. Hemangiomas (vascular neoplasms) are benign tumors of endothelial cell origin that can be detected at any age and in many human organs. They include a heterogeneous group of malformations resulting from a congenital disorder of vascular morphogenesis as well as true neoplasms that occur due to either endothelial cell proliferation or vascular malformations without true endothelial cell proliferation[1,3]. Hemangioma in female reproductive organs, particularly in the FT, is a sporadic disease. It was first described in 1947 by Ragins and Crane[4]. Katiyar et al.[3] presented their study of capillary

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hemangioma with data up to 2016 that demonstrated ten documented cases of cavernous hemangioma in the FT of patients aged 13 to 77 years. Since 2016, four other observations of FT hemangiomas have been published, including two cases of capillary hemangiomas[3,5,6]. We present a case of FT capillary hemangioma in a 39-year-old woman.

CASE REPORT

Patient information: In 2018 (three years before the FT tumor was revealed), a 39-year-old Eastern Slavic woman was diagnosed with uterine leiomyoma, and she was regularly seen by a gynecologist. At a regular examination, rapid growth of the uterine tumor was observed. The patient's health was satisfactory and her menstrual cycle was not disturbed. The patient's past history revealed one birth (cesarean section with blood transfusion) and one medical abortion. There were no infectious or general somatic symptoms in her past history, and her heredity was not burdened by otopathology.

Diagnostic assessment: Pelvic sonography revealed a postoperative scar along the anterior wall of the lower uterine segment. A reduced echogenicity node (FIGO-7) 27 mm in diameter was visualized along the posterior wall of the uterus, which was avascular in the ROWER mode and on CDL Doppler. No structural changes were observed in the cervix or cervical canal. The endometrium was in the late proliferative phase with medium echogenicity. Both ovarian size, structure, and folliculogenesis were within the age norms. A rounded neoplasm of up to 30 mm in diameter with a well-defined contour was visualized in the right parametrium. Anechoic areas up to 2-3 mm in diameter were found in the peripheral region, which had the appearance of a neoplasm with a fluid component (a "ground-glass" appearance). Neoplasmic vascularization in the ROWER mode and on CDL Doppler corresponded to 1 point. The neoplasm's connection to the ovary and uterine body was not revealed (Figure 1). The conclusion of uterine leiomyoma and neoplasm of the right parametrium with possible endometrioma was made based on the ultrasound findings.

Computed tomography of the chest, abdomen, and small pelvis with intravenous bolus injection of the contrast agent tomohexol 350 revealed a subserous leiomyomatous node along the posterior uterine wall measuring 23x28 mm and an oval-shaped neoplasm with unclear origin in the right corner of the uterus measuring 34x30 mm, with diffuse accumulation of contrast. The other examined organs showed no pathological deviations from the age norms (Figure 2). Clinical laboratory tests did not reveal any significant deviations from the norms [Ca-125, 38 U/mL; HE4, 42.10 pmol/L; ROMA-1 (calculation before menopause), 5.4]. After a comprehensive evaluation, a diagnosis of nodular uterine leiomyoma (FIGO-7) with possible external endometrioma was made.

Surgical intervention: Considering the presence of a neoplasm of unclear origin in the small pelvis, diagnostic laparoscopy was performed, followed by decision-making regarding the appropriateness of surgery and its volume. Laparoscopy revealed a subserous leiomyomatous node with a diameter of up to 28 mm along the posterior surface of the lower uterine segment. No pathologies

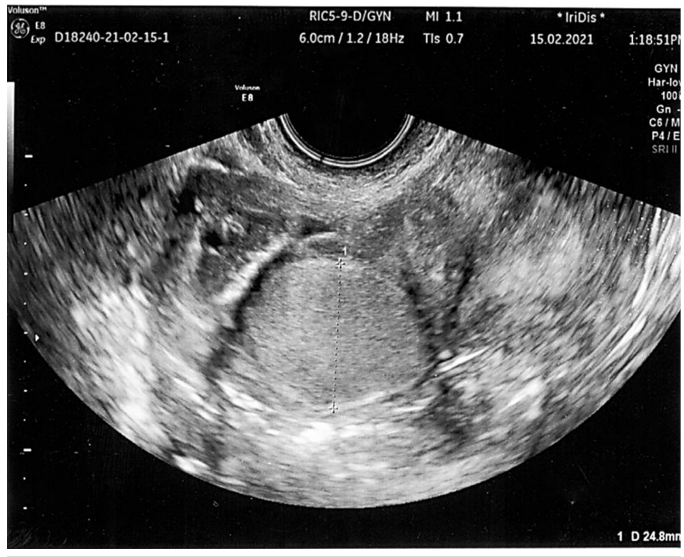

Figure 1. Parametrium neoplasm with unclear connection to the ovary and uterine body.

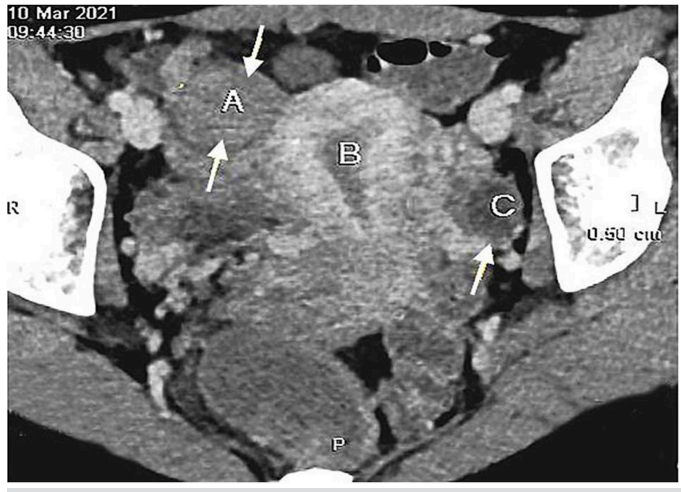

Figure 2. Computed tomography: A- small pelvis neoplasm; B- uterus; C- left ovary.

were observed in the ovaries or left FT. The right FT was pinkish-colored, deformed, and thickened up to 30 mm at the point of connection with the uterus. The decision was thus made to remove the right FT and perform a conservative myomectomy.

Pathomorphological investigation and outcomes: A frozen section examination of the intraoperative biopsy material confirmed the benignity of the neoplasms. Evaluation of the removed right FT revealed an ovoid-shaped neoplasm measuring 26x28 mm under the isthmic serous membrane. It was pale pink and had an uneven structure with a combination of dense and cystic areas. Six tumor fragments were fixed in 10% neutral formalin for 24 h for the study. On the basis of the standard procedure, paraffin-embedded tissue sections were stained with hematoxylin and eosin. Histologically, the tumor was composed of multiple thin-walled vascular spaces lined with a single layer of endothelial cells, in which single mitoses were present (Figure 3).

The pathomorphological study of the postoperative material concluded that the neoplasm was a capillary hemangioma of the right FT. Immunohistochemical analysis of the tumor sections was performed using anti-CD34, -CD31, -Ki-67, and -ER antibodies to confirm the diagnosis. The presence of strong and monotonous immunoreactivity for CD31 and CD34 in endothelial cells lining the interior surface of the spaces confirmed the diagnosis of a benign vascular tumor. In addition, low mitotic activity of the tumor cells (Ki-67 proliferation) and absence of estrogen receptor expression were revealed (Figure 4).

DISCUSSION

In addition to malignant tumors, borderline and benign neoplasms in FT can sometimes be revealed. They are worth considering, as many of them may be the first signals of malignancy or several complications[3,5,7,8].

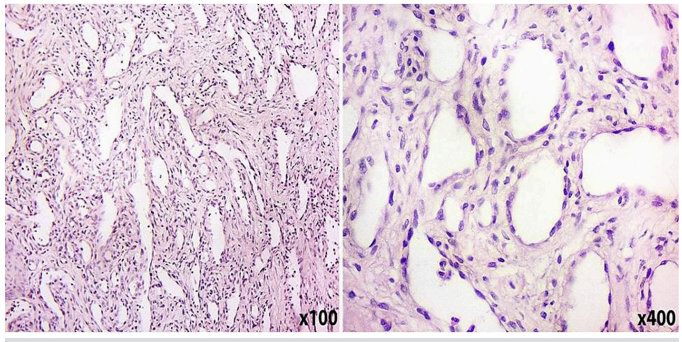

Figure 3. Right fallopian tube tumor. Hematoxylin and eosin staining. Magnification ×100 and ×400.

FT hemangioma is a sporadic benign vascular tumor, with only 16 documented cases available in the literature. Most of these neoplasms measured between 2 and 10 mm, had no clinical manifestations, and were incidentally discovered during surgeries for other diseases[1,6,9]. Thus, it should be assumed that the incidence of FT hemangioma is much higher.

The etiology of hemangiomas remains unclear. It is commonly believed that FT hemangioma is associated with the effect of estrogens, which stimulate the growth of blood vessels. This has been confirmed in some cases in which estrogen receptors were found in the endothelial cells of hemangiomas during immunohistochemical studies[1-3,6,8,10]. This case's postoperative material study demonstrated the absence of estrogen receptors in the endothelial tumor cells, which indicates that the occurrence and growth of the FT hemangioma did not depend on the effect of estrogen. In this case, the pathogenesis of the FT hemangioma remains unclear.

Differential diagnoses of FT hemangiomas include lymphangiomas, vascular leiomyomas mesothelioma and adenomatoid tumors[3,9]. Histologically, it is not difficult to distinguish hemangiomas from the above-mentioned neoplasms and FT cancer. However, for reliable confirmation of the diagnosis, an immunohistochemical study is recommended[3,8,11,12].

In our case, we found typical histological features of capillary hemangioma, characterized by multiple

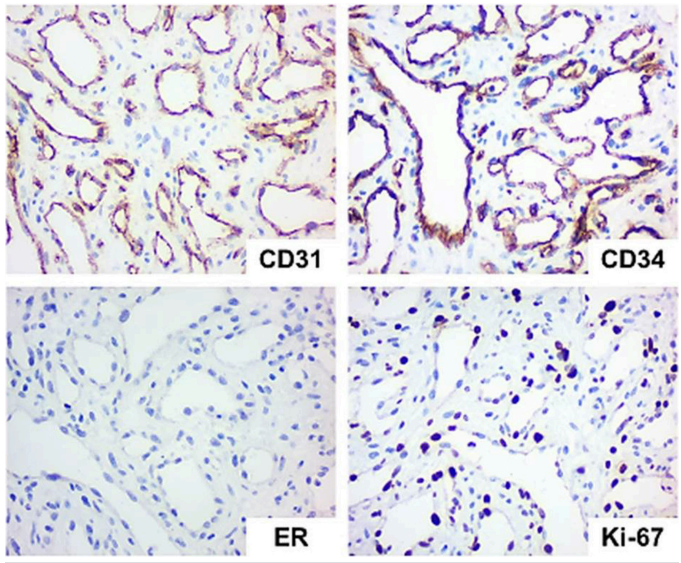

Figure 4. Tumor in the right fallopian tube. Immunohistochemical study of CD34, CD31, ER, and Ki-67 expression. Chromogen - diaminobenzidine; nuclei were counterstained with Mayer's hematoxylin. Magnification: ×200.

thin-walled vascular spaces lined with a single layer of endothelial cells, in which single mitoses were observed. The diagnosis was then confirmed immunohistochemically by CD31 and CD34 expression in the endothelial cells lining the inner surface of the spaces and the low mitotic activity of the tumor cells.

Similar to our case, hemangiomas in most patients are found incidentally during surgeries for other diseases or complications. If the tumor is small, specific clinical symptoms are virtually absent. However, in four of sixteen FT hemangiomas, the clinical picture of "acute abdomen" due to the hemoperitoneum was recorded[6,7]. In one case, the symptoms of the disease corresponded to acute appendicitis[7]. In some cases, FT hemangiomas were accompanied by uterine bleeding, although the state of the endometrium did not indicate any hormonal effect of the hemangiomas[3,11-15].

Sparing surgery should be considered an appropriate treatment for FT hemangioma because no relapses have been reported in previous cases of FT hemangioma[3].

Our case demonstrates that modern technical means make it possible to identify asymptomatic neoplasms and prevent the development of their complications. It is virtually impossible to make a diagnosis before surgery, and it is unlikely that a macroscopic diagnosis can be made during surgery. Therefore, it is necessary to conduct a frozen section examination, particularly when the size of the lesion is small. Mistakes in the interpretation of the FT tumor's origin can cause an inadequate volume of surgeries. Incorrect interpretation of a benign tumor at a young age can lead to unnecessary radical surgery with a resulting loss of fertility, and an unrevealed malignant process can threaten life.

Ethics

Informed Consent: The patient provided the written informed consent to the publication of the case report results. There are no potentially identifiable human images or data is presented.

Author Contributions

Surgical and Medical Practices: D.S., G.S., N.H., V.K., Concept: D.S., G.S., N.H., V.K., Y.R., I.G., Design: D.S., G.S., N.H., M.L., K.S., V.K., Y.R., I.G., Data Collection and/or Processing: D.S., G.S., N.H., M.L., K.S., V.K., Y.R., Analysis and/or Interpretation: D.S., G.S., N.H., M.L., K.S., V.K., Y.R., I.G., Literature Search: M.L., K.S., V.K., Writing: D.S., G.S., V.K., Y.R., I.G.

Conflict of Interest: The authors have no conflict of interest to declare.

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