Capillary Hemangioma of the Endometrium
A Case Report and Review of the Literature

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A 39-year-old woman with menorrhagia of 7 years’ duration was found to have a capillary hemangioma of the endometrium. Initial diagnosis by curettage was considered questionable but was later confirmed at hysterectomy. A thorough search and review of the literature was performed.

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Hemangiomas of the uterus and cervix are extremely rare. Although found at all levels of the uterine wall, including serosa, myometrium, and endometrium, most cases diffusely involve the myometrium. These lesions are associated with numerous obstetric and gynecologic complications, ranging from intermenstrual spotting, menometrorrhagia, and infertility to maternal and fetal demise from pronounced bleeding of the gravid uterus.1–3 Rare cases are associated with tuberous sclerosis and hemorrhagic telangiectasia.4,5 Only 6 cases in the literature, 2 of them in English, describe endometrial hemangioma. This is an additional report of a capillary hemangioma involving the endometrium and causing severe menorrhagia, the third such case in the English literature.

REPORT OF A CASE

The patient, a 39-year-old Hispanic woman, gravida 3, para 3, presented with menorrhagia that was refractory to conservative management. Menarche had commenced at age 13 and had been regular, typically of 7 days’ duration. The last normal menstrual when she first visited the emergency department complaining of dehydration, treated with intravenous saline, and placed on oral contraceptives. Menorrhagia was initially responsive but later became refractory, with bleeding lasting more than 10 days and almost constant spotting (1 bleeding-free week per month). Repeated dilatation and curettage during these 7 years rendered pathologic diagnoses including “endometrial hyperplasia,” “proliferative endometrium,” and “decidualized endometrial stroma with weakly proliferative glands.” Pelvic ultrasound done 7 months prior to hysterectomy showed an intraluminal endometrial nodule, believed to represent a polyp or prolapsed submucosal fibroid (Figure 1). Despite diagnostic endometrial curettage 4 months before hysterectomy, the bleeding continued, so a hysterectomy was performed.

MATERIALS AND METHODS

The tissues were fixed in 10% formalin and embedded in paraffin. Paraffin-embedded tissue sections were deparaffinized and rehydrated by passage through xylenes (5 minutes × 2), graded alcohols, and deionized water. Endogenous peroxidases were blocked by incubating the slides for 5 minutes at room temperature in 3% aqueous hydrogen peroxide, followed by rinsing with copious amounts of water. Slides for all antibodies except CD10 were then subjected to microwave antigen retrieval using 10mM citric acid, pH 6.0, followed by water rinses. All slides were soaked at room temperature for 5 minutes in Tris-buffered saline, pH 7.6, containing 0.05% Tween 20 (Dako), and placed in a Dako autostainer. The slides were incubated for 30 minutes at room temperature with the following antibodies and dilutions: monoclonal antibodies to von Willebrand factor clone F8/86 (1:50; Dako), CD34 (1:50; Dako), and monoclonal anti-CD10 (1:50; Novocasta, Vector Laboratories, Burlington, Calif). The autostainer then rinsed the slides with Tris-buffered saline with Tween, incubated them for 30 minutes at room temperature with EnVision + polymer (peroxidase) (Dako), rinsed them with Tris-buffered saline with Tween, incubated them for 10 minutes at room temperature with 3,3′-diaminobenzidine tetrahydroferrate substrate solution (Dako), and finally subjected them to a final deionized water wash. The slides were then removed from the autostainer, counterstained with hematoxylin, dehydrated by passage through graded alcohols and xylenes, and cover-slipped.

PATHOLOGIC FINDINGS

Endometrial curettage specimens of the intraluminal nodule consisted of multiple fragments of hemorrhagic tissue, measuring 8.0 × 8.0 × 4.0 cm in aggregate. Histologic examination revealed tissue fragments with numerous small, capillary-sized vascular channels, lined by monotonous endothelial cells, consistent with a capillary hemangioma. The capillaries were relatively uniform in size, with no centrally placed larger vessels, and there was no significant associated organizing hemorrhagic lesion. Separate fragments of secretory endometrium with

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Figure 1. Uterine ultrasound showing endometrial nodule.

Figure 2. a, Endometrial curettings showing capillary hemangioma (hematoxylin-eosin, original magnification ×120). b and c, Immunohistochemical stains showing positivity for factor VIII (original magnification ×120 [b]) and for CD34 (original magnification ×120 [c]).

Figure 3. Hysterectomy specimen (a) and papillary capillary hemangioma of the endometrium (b) (hematoxylin-eosin, original magnification ×40).
decidual stromal reaction and inactive glands were also seen (Figure 2, a). Immunohistochemical stains revealed that the endothelial cells were positive for von Willebrand factor F8/86 (Figure 2, b) and CD34 (Figure 2, c) and negative for CD10, thus supporting the diagnosis of a vascular tumor. On hysterectomy, a 92.0-g uterus with attached cervix was received. The uterus measured 9.0 cm from fundus to cervix, 6.5 cm from cornu to cornu, and 3.0 cm from anterior to posterior. The cervix was 3.5 cm in diameter. The external cervical os was oval and measured 0.4 cm in widest dimension. The ectocervical mucosa was tan-pink and shiny. The endocervical canal measured 3.5 cm in length. The endometrium appeared tan-pink, measured less than 0.1 cm in thickness, and contained a 0.6-cm red, polypoid mass involving its posterior wall (Figure 3, a). The myometrial wall was 1.2 cm thick and unremarkable. Histologic examination of the mass revealed a capillary hemangioma of similar histology to that of the previous endometrial biopsy. This polypoid hemangioma was well circumscribed and covered by attenuated endometrium showing multifocal superficial ulceration. The border between hemangioma and myometrium was irregular in that, although the vascular lesion remained limited to the well-circumscribed tumor, foci of smooth muscle could be seen in the base of the polyp (Figure 3, b). Uninvolved endometrium was proliferative, and the cervix showed moderate acute and chronic inflammation.

**COMMENT**

Vascular lesions of the uterus are diverse. They include capillary and cavernous hemangiomas, angiomyomas, and hemangioendotheliomas. Cavernous hemangiomas, composed of large dilated vascular channels, are the most common vascular tumors of the uterus. Lesions have been found restricted to the serosa, myometrium, and endometrium, and combinations thereof, as well as transmural. Myometrial lesions are the most common, and these are often diffuse,2,6,7 some with extension to the portio. The first case of diffuse uterine hemangiomas was described in 1897 and was an incidental discovery at autopsy after a young woman developed anemia and dyspnea and died 24 hours after delivering twins.8 The difference between the localized and diffuse forms of cavernous hemangioma is not clear, and transitional stages may exist.8

Congenital and acquired vascular tumors are found exceptionally with hereditary syndromes, including tuberous sclerosis5 and hemorrhagic telangiectasia.4 The cell of origin possibly represents pluripotential, embryogenic, mesodermal rests within the uterus.9 Typical acquired cases were associated with previous pelvic surgery, endometrial curettage, trophoblastic disease, endometrial carcinoma, and maternal ingestion of diethylstilbestrol.10

These vascular lesions may be asymptomatic, or they may cause menstrual irregularities or life-threatening bleeding. A case of rupture of hemangioma with hemoperitoneum and fetal demise has been reported in a gravid patient.3 The most common presenting sign was hemorrhage following curettage, when the thin endometrial tissue covering the hemangioma was removed and the blood vessels were exposed.6 Bleeding tendencies are believed to occur from erosion of thin endometrium overlying lesions, trophoblastic growth into these vascular malformations with their fistulous connections to systemic circulation, mechanical forces, and platelet trapping.8 The epithelium covering capillary hemangiomas is usually intact, but in exposed areas, such as the cervix and vagina, traumatic ulceration of the overlying epithelium may create a lesion that bleeds with only slight contact or trauma.11 The trophoblastic tissue of the placenta may erode into the hemangioma, causing severe bleeding or placental separation.12 The vascular lumina within the hemangiomas are connected to the surrounding blood vessels.11

Endometrial hemangiomas are extremely rare, possibly because of the periodic cyclical endometrial shedding. Though extension from the minimal myometrium involvement seen on the hysterectomy specimen is probable, this vascular lesion may also theoretically originate from angiomatous proliferation in a polypoid endometrial lesion that persists for a prolonged period.10 It has been proposed that the latter may occur because increased blood flow provided by the endometrial hemangioma may prohibit normal cyclic shedding associated with the usual hormonal flux.10

The differential diagnosis included hemangiopericytoma and angiosarcoma; however, a regular 1-layer architecture, bland benign-appearing endothelial cells, and virtually no nuclear atypia or mitotic activity excluded these entities.

Of the endometrial hemangiomas described in the English literature to date,4,10 including our own, all cases studied have shown progressive symptoms of uterine bleeding, refractory to conservative therapy, including dilation and curettage. Investigative modalities including vaginal examination, endometrial curettage, ultrasound, and hysteroscopy were noninformative, although the uterus may on rare occasions feel or appear pulsatile.7 Pelvic angiography and computed tomography may confirm the vascular nature of the lesion if there is a clinical suspicion of this abnormality in cases refractory to hormonal therapy.1 Treatment of uterine vascular anomalies that occur during pregnancy includes conservative therapy such as close follow-up during the second and third trimesters, with close observation during delivery. Numerous patients have had successful vaginal and cesarean deliveries despite the presence of extensive myometrial hemangiomas. The treatment of endometrial hemangiomas remains unclear, however, because this is an extremely rare entity, with few reports in the literature. Examination of the few cases in the literature suggests that conservative therapy typically fails, and the treatment of choice is hysterectomy. Nonsurgical modalities such as radiation therapy would probably cure the lesions but in the process would destroy ovarian function.10

**References**