American Journal of Obstetrics and Gynecology (2004) **191**, 1733–5

ELSEVIER

AMERICAN JOURNAL of OBSTETRICS and GYNECOLOGY

www.ajog.org

Leiomyosarcoma in a premenopausal patient after uterine artery embolization

Jay Goldberg, MD,[a,*] Irina Burd, MD, PhD,[a] Fredric V. Price, MD,[b] Robert Worthington-Kirsch, MD[c]

Department of Obstetrics and Gynecology, Jefferson Medical College, Philadelphia, Pa,[a] Pittsburgh Gynecologic Oncology, Pittsburgh, Pa,[b] and Department of Radiology, Roxborough Hospital, Philadelphia, Pa[c]

Received for publication March 2, 2004; revised April 22, 2004; accepted May 19, 2004

KEY WORDS
Leiomyosarcoma
Leiomyoma
Uterine artery embolization

A premenopausal 45-year-old woman underwent uterine artery embolization for suspected symptomatic leiomyomata. Fourteen months later, with renewed symptoms and a new pelvic mass, metastatic leiomyosarcoma was diagnosed. A lack of clinical response to a technically successful embolization should alert care providers that further evaluation and/or therapy is needed.

Uterine artery embolization is an increasingly popular treatment for symptomatic leiomyomata. Although it was only recently reported as a primary treatment for leiomyomas in 1995, more than 40,000 uterine artery embolization procedures have been performed globally. The MeSH terms "uterine artery embolization," "embolization," "leiomyomsarcoma," and "sarcoma" were used to search MEDLINE and PubMed for published reports of leiomyosarcoma after uterine artery embolization, identifying only 3 cases. Two of these cases occurred in women in their 50s, with the third in a 49-year-old woman, who became menopausal at age 43 years.[1-3] Our report depicts a case of leiomyosarcoma in a 45-year-old premenopausal woman who underwent uterine artery embolization for suspected symptomatic uterine fibroids.

* Reprint requests: Jay Goldberg, MD, Department of Obstetrics and Gynecology, Jefferson Medical College, 834 Chestnut St, Suite 400, Philadelphia, PA 19107.
E-mail: Jay.Goldberg@jefferson.com

0002-9378/$ - see front matter © 2004 Elsevier Inc. All rights reserved.
doi:10.1016/j.ajog.2004.05.028

Case report

A healthy 45-year-old multiparous woman with a 4-year history of leiomyomata elected to undergo uterine artery embolization for worsening pelvic pain, dysmenorrhea, and menorrhagia. A pre-embolization ultrasound revealed an enlarged (15.5 × 8.7 × 9.1 cm) uterus, with 2 prominent myomas, 5.4 × 4.7 × 9.1 cm and 4.9 × 2.7 × 4.0 cm. Five weeks after embolization, the patient underwent a hysteroscopic resection of a necrotic degenerating leiomyoma, because of continued pain, discharge, and fevers. The pathologic diagnosis was severe and acute purulent endometritis, granulation tissue, and a fragmented submucosal leiomyoma. Her symptoms completely resolved shortly thereafter.

Thirteen months later, with renewed pelvic pain and bleeding, examination revealed a tender mass filling the posterior pelvis. Imaging showed a solid 13.2 × 9.3 × 10.2-cm cul-de-sac mass. At laparotomy, a soft friable mass was noted posterior and separate from the uterus, with a soft necrotic center and parasitic blood supply

from the anterior rectum and cul-de-sac. There were multiple adjacent pelvic and omental adhesions with fine nodularity. A total hysterectomy and unilateral oophorectomy were performed. The intraoperative pathologic diagnosis was benign leiomyoma, with disseminated benign leiomyomatosis of the peritoneum. The final pathologic diagnosis, however, was reported as a leiomyosarcoma with low-grade endometrial stromal sarcoma. The prior embolization did not appear to be the cause of the difficulty in making the pathologic diagnosis on a frozen section. A subsequent laparotomy removed the left ovary, appendix, and debulked peritoneal implants. Leiomyosarcoma was seen throughout the specimens.

Although chemotherapy and radiation were recommended, she opted for nutritional and other alternative therapies. After becoming symptomatic with recurrent disease, she underwent chemotherapy, radiation, and multiple surgeries before dying of metastatic disease 44 months after the uterine artery embolization.

Comment

Leiomyosarcoma of the uterus occurs in approximately 1.3% of patients with uterine cancer and is the most common form of uterine sarcoma. It is one of the most aggressive cancers of uterine tract, with an early hematogenous spread to lung, bone, and liver.[4] Five-year survival rate for stage I tumor is 50%, decreasing to 20% with extrauterine spread. The signs and symptoms associated with leiomyosarcoma are usually indistinguishable from those caused by fibroids alone. However, in comparison to leiomyoma, the mean reported age of patients with leiomyosarcoma is 52 years, nearly a decade older.[3,5]

It is very difficult to make the diagnosis of a leiomyosarcoma. A rapid increase in the size of the uterus, especially after menopause raises suspicion. Parker et al,[6] however, found only a 0.27% incidence in 371 women who had undergone hysterectomy for suspected leiomyosarcoma. Endometrial biopsy, performed as part of the routine pre-embolization workup, largely excludes endometrial cancer; however, its sensitivity in detecting leiomyosarcoma is less than 25%.[6]

There is no current imaging standard used to identify sarcomas before uterine artery embolization. In one study of color Doppler ultrasonography as a screening technique for detecting leiomyosarcomas before embolization, all 10 cases of leiomyosarcoma, all having lower Doppler indices and abnormal vascularization when compared with normal or fibroid uteri, were detected in approximately 2000 women screened before hysterectomy. The sensitivity and specificity were 90.9% and 99.8%, respectively.[7] In real practice, however, this may not be practical because of the operator dependency of the examination and continuum of the findings. Magnetic resonance imaging (MRI) has also been described as a diagnostic tool for leiomyosarcoma, often showing atypical degeneration with an irregular contour.[8] Positron emission tomography may also help in identifying uterine sarcomas. A small Japanese series found this technology to have greater sensitivity in identifying uterine sarcomas than MRI or ultrasound.[9] Another recent Japanese prospective study that evaluated the combined use of dynamic MRI and serum measurement of lactate dehydrogenase (LDH) levels in 10 patients with leiomyosarcoma and 130 patients with degenerating leiomyomas had 100% sensitivity and specificity in differentiating leiomyosarcomas from degenerating leiomyomas of the uterus.[10] Given their rare occurrence and usually unsuspected identification, no large prospective imaging series containing significant numbers of leiomyosarcomas exists.

Given the difficulty in diagnosing leiomyosarcoma and the increasing popularity of uterine artery embolization, in lieu of myomectomy or hysterectomy, which provide a pathologic specimen, an increasing number of leiomyosarcomas will be unavoidably delayed in diagnosis. In this case, although we now suspect that sarcoma was present at the time of the embolization, we do not believe that the tumor was related to the embolization, but its diagnosis may have been delayed. As other gynecologic problems, including cancer, may coexist or develop later in the presence of uterine fibroids, a lack of clinical response to a technically successful embolization should alert care providers that further evaluation and/or therapy is needed.

References

1. Al-Badr A, Faught W. Uterine artery embolization in an undiagnosed uterine sarcoma. Obstet Gynecol 2001;97:836-7.
2. Joyce A, Hessami S, Heller D. Leiomyosarcoma after uterine artery embolization: a case report. J Reprod Med 2001; 46:278-80.
3. Common AA, Mocarski EJM, Kolin A, Pron G, Soucie J. Therapeutic failure of uterine fibroid embolization caused by underlying leiomyosarcoma. J Vasc Interv Radial 2001;12:1449-52.
4. Christopherson WM, Williamson EO, Gray LA. Leiomyosarcoma of the uterus. Cancer 1972;29:1512-7.
5. Marchese MJ, Lishow AS, Crum CP, McCaffrey RM, Frick HC 2nd. Uterine sarcomas: a clinicopathologic study. Gynecol Oncol 1984;18:299-312.
6. Parker WH, Fu YS, Berek JS. Uterine sarcoma in patients operated on for presumed leiomyoma and rapidly growing leiomyoma. Obstet Gynecol 1994;83:414-8.
7. Kurjak A, Kupesic S, Shalan H, Jukic S, Kosuta D, Ilijas M. Uterine sarcoma: a report of 10 cases studied by transvaginal color and pulsed Doppler sonography. Gynecol Oncol 1995;59:342-6.
8. Pattani SJ, Kier R, Deal R, Luchansky E. MRI of uterine leiomyosarcoma. Magn Reson Imaging 1995;13:331-3.

9. Umesaki N, Tanaka T, Miyama M, Kawamura N, Ogita S, Kawabe J, et al. Positron emission tomography with (IS)F-fluorodeoxyglucose of uterine sarcoma: a comparison with magnetic resonance imaging and power Doppler imaging. Gynecol Oncol 2001;80:372-7.

10. Goto A, Takeuchi S, Sugimura K, Maruo T. Usefulness of Gd-DTPA contrast-enhanced dynamic MRI and serum determination of LDH and its isoenzymes in the differential diagnosis of leiomyosarcoma from degenerating leiomyoma of the uterus. Int J Gynecol Cancer 2002;12:354-61.