

PRIMARY OVARIAN HEMANGIOMA: CASE REPORT AND REVIEW OF LITERATURE

HATİCE BAYRAMOĞLU*
TAYFUN GÜNGÖR**
MURAT ÖZ**
NASUH U. DOĞAN**
HÜSEYİN REYHAN**
MUSTAFA BEŞLİ**

SUMMARY: Hemangiomas are benign and rare tumors of female genital tract and most of which are asymptomatic. A 55-year-old woman was referred to our department due to postmenopausal bleeding with 3 cm diameter adnexial mass. Transvaginal sonography revealed a 3-cm right ovarian semicystic mass with a low-resistant vascular flow pattern in the Doppler examination, supporting malignant neoplasia of the ovary. Serum Ca-125 levels of the patient were inconclusive. The pathology result of the mass was primary ovarian hemangioma, a benign vascular tumor after the surgery. Primary ovarian hemangiomas are extremely rare conditions. Some of the ovarian hemangiomas are presented as an ovarian mass with ascites and serum Ca-125 elevation, mimicking advanced stage ovarian cancer. Frozen sections may not always give a definitive histologic behavior of the mass. Thus, unnecessary radical surgery may be performed for benign ovarian vascular neoplasm.

Key words: Hemangioma, ovary, cystic mass.

INTRODUCTION

Hemangiomas are benign (1) and rare tumors of female genital tract (2) and most of which are asymptomatic and of the cavernous type (3). The age range of cases in the literature was 12-76 (2,4). Rarely an ovarian hemangioma may be one manifestation of Kasabach-Merritt Syndrome (systemic hemangiomata) (5). In some cases ovarian hemangiomas may be presented as an ovarian mass with ascites and Ca-125 elevation, mimicking advanced stage ovarian carcinoma (6-10). Thus,

ovarian hemangiomas can present with symptoms similar to epithelial malignancies and may lead to unnecessary radical surgery(2). In most patients, ovarian hemangiomas are discovered incidentally, and sizes range from 0.3 to 24 cm (6,11). Hemangioma is a benign tumor with proliferative vessels with unclear borders. Many such tumors occur in the skin, head, and neck (7,12-14). Hemangiomas are rarely located in the genital tract. Approximately 50 documented cases about hemangiomas are available in the English literature. A review of the literature revealed that some ovarian hemangiomas are associated with endometrial hyperplasia and malignancies including endometrial cancer and germ cell tumor (11,15-18).

*From Pathology Department, Dr. Zekai Tahir Burak Women's Health Educational and Research Hospital, Ankara, Turkey.

**From Gynecologic Oncology Department, Dr. Zekai Tahir Burak Women's Health Educational and Research Hospital, Ankara, Turkey.

CASE REPORT

A 55-year-old woman was referred to our department due to postmenopausal bleeding with a 3-cm diameter adnexial mass. In a bimanual pelvic examination, the painless right adnexial mass was palpated. Transvaginal sonography revealed a 3-cm right ovarian semicystic mass with a low-resistant vascular flow pattern in the Doppler examination. There was no sign of ascites in the ultrasonographic examination. The serum Ca-125 level of the patient was 32 IU/ml which was inconclusive. Pap smear revealed normal cytologic findings and pipelle endometrial sampling showed atrophic endometrium. A laparotomy and a frozen section procedure were planned and performed on a patient with the early diagnosis of adnexial mass. The surgical exploration was nonremarkable except for the mass in the right ovary. The mass was resected with ipsilateral adnexa and the frozen section was performed. No malignancy was noted in the frozen section. Total abdominal hysterectomy and left salpingo-oophorectomy were performed. The pathology result of the mass was primary ovarian hemangioma, a benign vascular tumor.

DISCUSSION

Hemangiomas are benign lesions arising from a failure in vascular formation, particularly in the canalizing process, forming abnormal vascular channels. These are of two types: cavernous and capillary (1). The difference between these two types relates to the size of the blood vessels formed. Hemangioma in the ovary must be differentiated from proliferations of dilated blood vessels of the ovarian hilar region (1). To define the lesion as a true hemangioma, a mass of vascular channels with minimal

amounts of stroma should form a reasonably circumscribed lesion distinct from the remainder of the ovary.

We have been able to cite 45 hemangioma cases from the literature with appropriate definition (Table 1). Most of the cases were admitted to hospital with abdominal pain or vaginal bleeding. Some of the cases were defined during other undergoing surgical process such as hysterectomy or appendectomy. Ovarian hemangiomas are rare and nonfunctional vascular neoplasm of the ovary. However some of ovarian hemangioma cases are related with thrombocytopenia (19), ovarian stromal luteinization, postmenopausal bleeding, endometrial hyperplasia, or even endometrial carcinoma (12, 15, 16, 18, 20-23). Our case had postmenopausal bleeding due to atrophic endometrium. Only a few of the ovarian hemangiomas have been associated with ascites and serum Ca-125 elevation. In these circumstances ovarian hemangiomas can mimic advanced stage ovarian cancer. Preoperative Doppler ultrasonographic examination of the lesion may show a low-resistant vascular flow pattern, supporting malignant neoplasia of the ovary. The frozen section may not always give a definitive histologic behavior of the mass. Ovarian hemangiomas are rare and benign conditions. Sometimes both preoperative findings and intra-operative structures can be confusing. It can be misdiagnosed as malignancy with preoperative imaging studies. The surgical approach differs in the diagnosis of hemangioma. Thus, unnecessary radical surgery may be performed for benign ovarian vascular neoplasm. To avoid this unnecessary radical surgery hemangioma should be kept in mind in differential diagnosis before surgery and during the frozen section investigation.

Table 1: Reported ovarian hemangioma cases in the literature.

Author	Age	Symptom	Size	Location	Type	Coexisting Lesion
McBurney <i>et al.</i> 1955 (24)	57	Ascites	5 cm	Unilateral	NA	NA
Mann <i>et al.</i> 1961 (25)	19	Acute abdominal pain	11 cm	Unilateral	NA	Periappendicitis
Talerman <i>et al.</i> 1967 (26)	41	NA	5 mm	Unilateral	Cavernous	NA
Gay <i>et al.</i> 1969 (27)	4	Abdominal enlargement	NA	Unilateral	Cavernous	Benign cystic teratoma
Fundaro <i>et al.</i> 1969 (28)	NA	NA	NA	Bilateral	Cavernous	NA
Ebrahimi <i>et al.</i> 1971(29)	41	Asymptomatic	NA	Unilateral	NA	NA
Brunner <i>et al.</i> 1972 (30)	37	NA	NA	Unilateral	Cavernous	NA
Rodriquez <i>et al.</i> 1979 (31)	81	Uterin prolapse	5 cm	Unilateral	Cavernous	No
DiOrio <i>et al.</i> 1980 (32)	21	Acute abdominal pain	20 cm	Unilateral	NA	Pregnancy

Table 1: Continue

Author	Age	Symptom	Size	Location	Type	Coexisting Lesion
Lawhead <i>et al.</i> 1985 (19)	NA	Abdomino pelvic mass	NA	Bilateral	NA	Thrombocytopenia
Alvarez <i>et al.</i> 1986 (11)	68	Ovarian mass	NA	Unilateral	NA	NA
Grant <i>et al.</i> 1986 (20)	59	Postmenopausal bleeding	1.5 cm	Unilateral	NA	Endometrial hyperplasia
Miyauchi <i>et al.</i> 1987 (5)	NA	Disseminated pelvic and abdominal mass	NA	Bilateral	Capillary Cavernous	Kasabach-Meritt Syndrome
Betta <i>et al.</i> 1988 (33)	52	Lower abdominal discomfort	7 cm	Unilateral	NA	NA
Gunes <i>et al.</i> 1990 (34)	11	Acute abdomen	NA	Unilateral	NA	NA
Pethe <i>et al.</i> 1991 (35)	NA	NA	NA	NA	NA	NA
Ozana <i>et al.</i> 1994 (36)	NA	Ovarian torsion	NA	Unilateral	NA	NA
Savargaonkar <i>et al.</i> 1994 (21)	69	Postmenopausal bleeding	NA	Unilateral	NA	Stromal luteinization Tubal carcinoma
Carder <i>et al.</i> 1995 (18)	62	Postmenopausal bleeding	1.5 cm	Unilateral	NA	Stromal luteinization
Talerman <i>et al.</i> 1995 (17)	NA	NA	5 mm	Unilateral	NA	Gonadal germ cell tumor
Yamawaki <i>et al.</i> 1996 (22)	62	Pelvic mass and ascites	NA	Unilateral	NA	Stromal luteinization
Rivasi <i>et al.</i> 1996 (16)	46	Asymptomatic	3 mm	NA	NA	Endometrioid ca
	50		5 mm			Leiomyoma
	74		30 mm			ascites
Cormio <i>et al.</i> 1998 (37)	32	Adnexial mass Severe vaginal bleeding	10 cm	Bilateral	NA	Multiple other hemangiomas
Mirilas <i>et al.</i> 1999 (38)	8	Ovarian torsion Acute abdomen	NA	Unilateral	Cavernous	NA
Jurkovic <i>et al.</i> 1999(39)	32	Asymptomatic	NA	Unilateral	NA	Mucinous cystadenoma
Gehriq <i>et al.</i> 2000 (6)	39	Ascites	8 cm	Unilateral	Capillary	Ca-125 elevation Stromal luteinization
Miliaras <i>et al.</i> 2001 (23)	71	Asymptomatic	NA	Unilateral	NA	Stromal luteinization
Kaneta <i>et al.</i> 2003 (7)	NA	Ascites Pleural effusion	NA	Unilateral	NA	Ca-125 elevation
M'pempa <i>et al.</i> 2003 (40)	13	Ovarian torsion	NA	Unilateral	Cavernous	NA
Itoh <i>et al.</i> 2004 (12)	63	Adnexial mass	NA	Unilateral	NA	Mature teratoma Stromal luteinization
Correra <i>et al.</i> 2003 (41)	11	Adnexial mass	NA	Unilateral	Cavernous	NA
Uppal <i>et al.</i> 2004 (1)	32	Asymptomatic	3 cm	Unilateral	Cavernous	NA
	48	Asymptomatic	4 cm	Unilateral	Cavernous	Stromal luteinization
	48	Asymptomatic	3 mm	Unilateral	Cavernous	NA
Gucer <i>et al.</i> 2004 (15)	70	Postmenopausal bleeding	1,5 cm	Unilateral	NA	Endometrium ca Stromal luteinization
Ortiz <i>et al.</i> 2005 (42)	36	Pelvic pain	NA	Unilateral	Hobnail	Endometriosis
Abu <i>et al.</i> 2006 (8)	48	Pelvic mass Ascites	NA	Unilateral	NA	Ca-125 elevation
Erdemoglu <i>et al.</i> 2006 (9)	57	Pelvic mass Ascites	6 cm	Unilateral	AN	Ca-125 elevation
Gupta <i>et al.</i> 2006 (2)	5	Ovarian hemangioma cases				
Koh <i>et al.</i> 2007 (10)	NA	Ascites Pleural effusion	NA	NA	NA	Ca-125 elevation

Table 1: Continue.

Author	Age	Symptom	Size	Location	Type	Coexisting Lesion
Akbulut <i>et al.</i> 2008 (3)	65	Postmenopausal bleeding	6 cm	Unilateral	NA	Endometrial polyp
Kim <i>et al.</i> 2008 (43)	69	Ovarian torsion	NA	Unilateral	NA	Serous papillary ca Calcified ovarian mass
Liapis <i>et al.</i> 2009 (4)	2	Ovarian hemangioma cases				
Comunoglu <i>et al.</i> 2010 (44)	81	Pelvic mass	3.5 cm	Unilateral	Cavernous	Contralateral mature cystic teratoma
Present Case	55	Postmenopausal bleeding	3 cm	Unilateral	Cavernous Capillary	NA

REFERENCES

- Uppal S, Heller DS, Majmudar B. Ovarian hemangioma-report of three cases and review of the literature. *Arch Gynecol Obstet* 2004; 270:1-5.
- Gupta R, Singh S, Nigam S, Khurana N. Benign vascular tumors of female genital tract. *Int J Gynecol Cancer* 2006; 16:1195-200.
- Akbulut M, Bir F, Colakoglu N, Soysal ME, Duzcan SE. Ovarian hemangioma occurring synchronously with serous papillary carcinoma of the ovary and benign endometrial polyp. *Ann Saudi Med* 2008; 28:128-31.
- Liapis A, Bakalianou K, Salakos N, Iavazzo C, Dalainas I, Kondi-Pafiti A. Vascular tumors: rare neoplasms of the female genital tract. *Eur J Gynaecol Oncol* 2009; 30:220-2.
- Miyauchi J, Mukai M, Yamazaki K, Kiso I, Higashi S, Hori S. Bilateral ovarian hemangiomas associated with diffuse hemangioendotheliomatosis: a case report. *Acta Pathol Jpn* 1987; 37:1347-55.
- Gehrig PA, Fowler WC, Jr, Lininger RA. Ovarian capillary hemangioma presenting as an adnexal mass with massive ascites and elevated CA-125. *Gynecol Oncol* 2000; 76:130-2.
- Kaneta Y, Nishino R, Asaoka K, Toyoshima K, Ito K, Kitai H. Ovarian hemangioma presenting as pseudo-Meigs' syndrome with elevated CA125. *J Obstet Gynaecol Res* 2003; 29:132-5.
- Abu J, Brown L, Ireland D, Sizeland E. Mesovarian hemangioma presenting as massive ascites, pelvic mass, and elevated CA125. *Int J Gynecol Cancer* 2006; 1:412-4.
- Erdemoglu E, Kamaci M, Ozen S, Sahin HG, Kulusari A. Ovarian hemangioma with elevated CA125 and ascites mimicking ovarian cancer. *Eur J Gynaecol Oncol* 2006; 27:195-6.
- Koh LW, Sun YL, Koh PH, Chiu HY, Chen SY, Huang MH. Ovarian capillary hemangioma presenting as pseudo-Meigs' syndrome: a case report. *J Minim Invasive Gynecol* 2007; 14:367-9.
- Alvarez M, Cerezo L. Ovarian cavernous hemangioma. *Arch Pathol Lab Med* 1986; 110:77-8.
- Itoh H, Wada T, Michikata K, Sato Y, Seguchi T, Akiyama Y, *et al.* Ovarian teratoma showing a predominant hemangiomatous element with stromal luteinization: report of a case and review of the literature. *Pathol Int* 2004; 54:279-83.
- Prus D, Rosenberg AE, Blumenfeld A, Udassin R, Ne'eman Z, Young RH *et al.* Infantile hemangioendothelioma of the ovary: a monodermal teratoma or a neoplasm of ovarian somatic cells? *Am J Surg Pathol*, 1997; 21:1231-5.
- Talerman. Nonspecific tumors of the ovary. Kurman RJ, ed *Blaustein's Pathology of the Female Genital Tract*, 5th edn New York: Springer-Verlag, 5th edn, pp 1014-44, 2002.
- Gucer F, Ozyilmaz F, Balkanlı-Kaplan P, Mulayim N, Aydin O. Ovarian hemangioma presenting with hyperandrogenism and endometrial cancer: a case report. *Gynecol Oncol* 2004; 94:821-4.
- Rivasi F, Philippe E, Walter P, de Marco L, Ludwig L. [Ovarian angioma. Report of 3 asymptomatic cases]. *Ann Pathol* 1996; 16:439-41.
- Talerman A. Gonadal mixed germ cell tumor combined with a large hemangiomatous lesion. *Arch Pathol Lab Med* 1995; 119:992-3.
- Carder PJ, Goulesbrough DR. Ovarian haemangiomas and stromal luteinization. *Histopathology*, 1995; 26:585-6.
- Lawhead RA, Copeland LJ, Edwards CL. Bilateral ovarian hemangiomas associated with diffuse abdominopelvic hemangiomatosis. *Obstet Gynecol*, 1985; 65:597-9.
- Grant JW, Millward-Sadler GH. Haemangioma of the ovary with associated endometrial hyperplasia. Case report. *Br J Obstet Gynaecol* 1986; 93:1166-8.
- Savargaonkar PR, Wells S, Graham I, Buckley CH. Ovarian haemangiomas and stromal luteinization. *Histopathology*, 1994; 25:185-8.
- Yamawaki T, Hirai Y, Takeshima N, Hasumi K. Ovarian hemangioma associated with concomitant stromal luteinization and ascites. *Gynecol Oncol* 1996; 61:438-41.
- Miliaras D, Papaemmanouil S, Blatzas G. Ovarian capillary hemangioma and stromal luteinization: a case study with hormonal receptor evaluation. *Eur J Gynaecol Oncol* 2001; 22:369-71.

24. Mc BR, Trumbull M. Hemangioma of the ovary with ascites. *Miss Doct* 1955; 32:271-4.
25. Mann LS, Metrick S. Hemangioma of the ovary. Report of a case. *J Int Coll Surg* 1961; 36:500-2.
26. Talerman A. Hemangiomas of the ovary and the uterine cervix. *Obstet Gynecol* 1967; 30:108-13.
27. Gay RM, Janovski NA. Cavernous hemangioma of the ovary. *Gynaecologia* 1969; 168:248-57.
28. Fundaro P. [Bilateral cavernous hemangioma of the ovary: report of a case and review of the literature]. *Folia Hered Pathol (Milano)* 1969; 18:45-50.
29. Ebrahimi T, Goldsmith JW, Okagaki T. Hemangioma of the ovary. A case report. *Obstet Gynecol* 1971; 38:677-9.
30. Brunner P. [Cavernous ovarian hemangioma in a 37-year-old woman (case report)]. *Zentralbl Gynakol* 1972;94:343-4.
31. Rodriguez MA. Hemangioma of the ovary in an 81-year-old woman. *South Med J* 1979; 72:503-4.
32. DiOrio J Jr, Lowe LC. Hemangioma of the ovary in pregnancy: a case report. *J Reprod Med* 1980; 24:232-4.
33. Betta PG, Robutti F, Spinoglio G. Hemangioma of the ovary. *Eur J Gynaecol Oncol* 1988; 9:184-5.
34. Gunes HA, Egilmez R, Dulger M. Ovarian haemangioma. *Br J Clin Pract* 1990; 44:734-5.
35. Pethe VV, Chitale SV, Godbole RN, Bidaye SV. Hemangioma of the ovary--a case report and review of literature. *Indian J Pathol Microbiol* 1991; 34:290-2.
36. Ozana M, Formanek P, Sula F, Silhan J. [Ovarian hemangioma with torsion]. *Ceska Gynekol* 1994; 59:18-20.
37. Cormio G, Loverro G, Iacobellis M, Mei L, Selvaggi L. Hemangioma of the ovary. A case report. *J Reprod Med* 1998; 43:459-61.
38. Mirilas P, Georgiou G, Zevgolis G. Ovarian cavernous hemangioma in an 8-year-old girl. *Eur J Pediatr Surg* 1999; 9:116-8.
39. Jurkovic I, Dudrikova K, Boor A. Ovarian hemangioma. *Cesk Patol* 1999; 35:133-5.
40. M'Pemba Loufoua-Lemay AB, Peko JF, Mbongo JA, Mokoko JC, Nzingoula S. [Ovarian torsion revealing an ovarian cavernous hemangioma in a child]. *Arch Pediatr* 2003; 10:986-8.
41. Correa-Rivas MS, Colon-Gonzalez G, Lugo-Vicente H. Cavernous hemangioma presenting as a right adnexal mass in a child. *PR Health Sci J* 2003; 22:311-3.
42. Ortiz-Rey JA, Gonzalez-Ruiz A, San Miguel P, Alvarez C, Iglesias B, Anton I. Hobnail haemangioma associated with the menstrual cycle. *J Eur Acad Dermatol Venereol* 2005; 19:367-9.
43. Kim MY, Rha SE, Oh SN, Lee YJ, Jung ES, Byun JY. Case report: Ovarian cavernous haemangioma presenting as a heavily calcified adnexal mass. *Br J Radiol* 2008; 81:e269-71.
44. Comunoglu C, Atasoy L, Baykal C. Ovarian hemangioma occurring synchronously with contralateral mature cystic teratoma in an 81-year-old patient. *Ups J Med Sci* 2010;115:297-9.

Correspondence:

Tayfun Güngör
 Dr. Zekai Tahir Burak
 Kadın Sağlığı Eğitim ve Araştırma Hastanesi,
 Jinekolojik Onkoloji Bölümü,
 Hamamönü, Samanpazarı, 06230
 Ankara, TÜRKİYE.
 e-mail: gungortayfun@yahoo.com