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Ovarian Hemangioma: Report of Two Cases and Review of the Literature

Ovaryan Hemanjiyom: İki Olgu Sunumu ve Literatürün Gözden Geçirilmesi

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ABSTRACT

Ovarian hemangiomas are rare tumors. Most of them are asymptomatic and of the cavernous type. The ovaries have a rich vascular supply and the rarity of vascular tumors in the ovary is therefore surprising. Although often found incidentally during the surgery, these lesions may rarely be associated with systemic manifestations. Here, we report two cases of cavernous ovarian hemangiomas. One was accompanied by with an endometrial carcinoma in a patient who presented with postmenopausal bleeding, and the other was found incidentally during the histopathological examination of the hysterectomy and bilateral salphingo-oophorectomy specimen that was performed for leiomyoma uteri.

Key Words: Ovary, Hemangioma

ÖZ

Ovaryan hemanjiomlar nadir tümörlerdir. Çoğunluğu asemptomatik ve kavernöz tiptedir. Overlerin zengin bir damar ağına sahip olmalarına karşın vasküler tümörlerin bu kadar az görülmesi şaşırtıcıdır. Bu lezyonlar çoğunlukla insidental olarak saptanmakla birlikte nadir de olsa sistemik bulgular da verebilirler. Biz, burada iki kavernöz hemanjiyom olgusu sunmaktayız. Bu olgulardan biri postmenopozal kanaması olan bir hastada endometriyal karsinom ile birlikte, diğeri ise leiomyoma uteri nedeniyle yapılmış bir total abdominal histerektomi ve bilateral salpingo-ooferektomi materyalinin histopatolojik incelenmesi sırasında tesadüfen saptanmıştır.

Anahtar Sözcükler: Over, Hemanjiyom

INTRODUCTION

Vascular tumors of the female genital tract, especially those of the ovary, are very rare. Most ovarian hemangiomas are of the cavernous type and they may present either as an isolated ovarian mass, usually discovered incidentally, or in conjunction with diffuse abdominopelvic hemangiomatosis (1-3). Ovarian hemangiomas are "nonfunctional" neoplasms. However, it is well known that luteinization of ovarian stromal cells commonly occurs as a reactive phenomenon, and may be associated with androgenic, estrogenic or progestagenic effects (2,4). This tumor is due to the cyclic changes that the ovaries undergo during the reproductive years. The histogenesis of hemangioma of the ovary is however debatable.

Here, we report two cases of cavernous ovarian hemangiomas. One was accompanied an endometrial carcinoma in a patient who presented with postmenopausal bleeding, and the other one was found incidentally in the histopathological examination of a hysterectomy and bilateral salphingo-oophorectomy (TAH+BSO) specimen that was performed for leiomyoma uteri.

CASE REPORT

Case 1: A 68-year-old woman underwent TAH+BSO and pelvic paraaortic lymph node dissection for endometrial carcinoma. The endometrial tumor was diagnosed as a moderately differentiated endometrial carcinoma (Figure 1) with less than 50% myometrial invasion. There was no lymphovascular space invasion. The uterine serosa and adnexa were free of the tumor. Pelvic (n=33) and paraaortic (n=5) lymph nodes, omentum, and peritoneal washings were negative. The right ovary was 2x1.5x1 cm and contained a 0.5x0.5x0.2 cm, well-circumscribed hemorrhagic small nodule on cut surface. The histopathologic examination revealed a cavernous hemangioma, consisting of multiple thin-walled blood vessels filled with red blood cells (Figure 2). A single layer of flattened endothelium lined all the vessels. Mitotic activity was not noted and no atypical cells were seen, and the final diagnosis of the ovarian nodule

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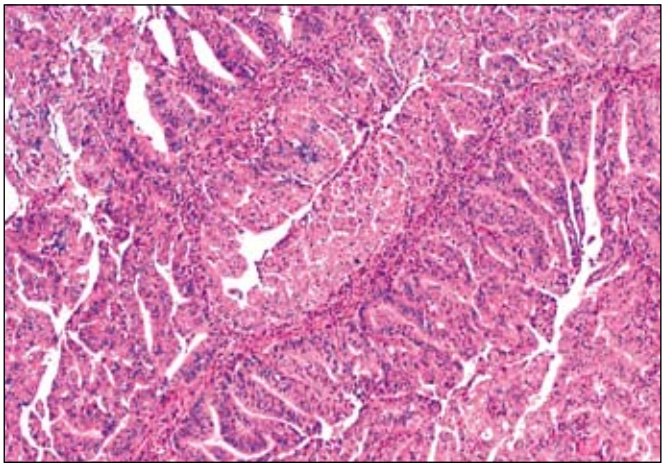

Figure 1: Endometrial adenonocarcinoma, moderately differentiated (H&E, x100).

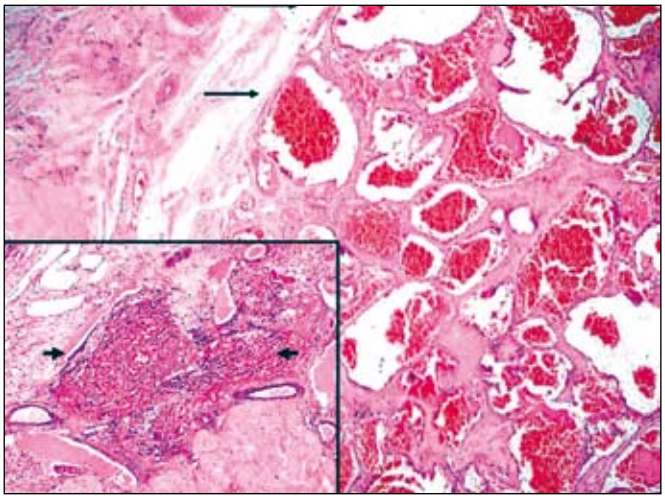

Figure 2: Multiple thin walled vessels of ovarian hemangioma replacing the ovarian stroma in Case 1 (H&E, x40). Inset: A cluster of luteinized cells in the ovarian stroma (arrows) (H&E, x100).

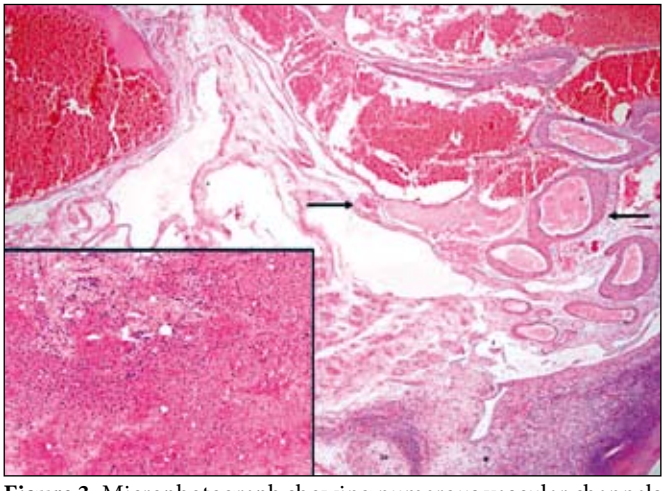

Figure 3: Microphotograph showing numerous vascular channels in ovarian hemangioma in Case 2 (arrows) (H&E, x40). Inset: Stromal luteinization in the peripheral ovarian stroma (H&E, x100).

was cavernous hemangioma. The adjacent cortical stroma contained small clusters of luteinized cells.

Case 2: A 44-year-old woman underwent TAH+BSO for symptomatic uterine leiomyoma. The left ovary measured 3.5x2.5x2 cm and the stroma was edematous on gross inspection. Histologically, an incidental cavernous hemangioma was noted in the left ovary (Figure 3). The peripheral ovarian stromal cells had undergone extensive luteinization. The uterus showed multiple leiomyomata and secretory endometrium.

DISCUSSION

Vascular tumors are rare in the female genital tract, particularly in the ovary (1-7). Hemangioma of the ovary was first described by Payne in 1869 (cited by Talerman) (8). Ovarian hemangiomas have been reported both in adults and children with an age range from infancy to 81 years (9,10). Histologically hemangiomas are benign lesions arising from a failure in the vascular malformation, particulary in the canalizing process, forming abnormal vascular channels. There are two types, cavernous and capillary, with the cavernous type being the most common (6,7). Although they have been found in different parts of the ovaries, the medulla and hilar region are the most common locations. This is consistent with the presence of larger vascular channels in these regions. The tumor was located in the ovarian medulla in case 1 and the ovarian hilus in case 2. Ovarian hemangiomas range from few millimeters to 20 cm in size and generally possess a smooth glistening outer surface on gross examination (7).

Conditions associated with ovarian hemangiomas include trombocytopenia (11), ascites (2,3), stromal luteinization with or without ascites (3,11), and endometrial hyperplasia or carcinoma. Stromal luteinization and stromal hyperplasia in relation to an ovarian hemangioma have been reported in three patients with endometrial hyperplasia, and one patient with endometrial adenocarcinoma (1,4,12,13).

The etiology of ovarian hemangiomas is unknown. Some state that it is a true tumor or hamartoma, or stimulated vessels. However, some autors have proposed that they are congenital malformations, or neoplasms, which may be stimulated by pregnancy, other hormonal influences, or infection (14). One hypotesis has been that the inciting event in the development of ovarian hemangiomas is hyperestrogenism resulting from stromal hyperplasia or stromal hyperthecosis, which may also result in endometrial stimulation (15,16). This hypothesis is based on the fact that estrogens have known growth stimulatory effects on

the vasculature and that most hemangiomas carry estrogen receptors.

An alternative hypothesis is that the presence of an ovarian hemangioma is the primary event in the pathway leading to hyperandrogenism or hyperestrogenism. In this way, stromal luteinization that occurs in the presence of expansile ovarian lesions is usually restricted to the stroma of the neoplasm or to the ovarian stroma adjacent to the neoplasm. In our case 1, stromal luteinization was limited to one ovary while stromal luteinization was markedly prominent in the peripheral ovarian stroma in case 2, and there was no stromal hyperthecosis in either cases. Furthermore, immunohistochemical staining for estrogen and progesteron receptors was negative in the endothelial cells of the hemangioma in both patients but positive in the co-existing endometrial carcinoma in case 1. These findings suggested that the ovarian hemangioma may initiated by a reactive phenomenon of stromal luteinization culminating in hyperandrogenism and hyperestrogenism. Differential diagnosis of luteinized stromal cells neighbouring hemagioma from steroid cell tumors was based on the absence of a dense reticulin fiber pattern, intracellular lipid and lipochrome pigment. The hemangioma had a hilar localization in one of our cases. Luteinized cells did not lie between the vascular channels, but were located at the periphery of the hemangioma as a small solid group. The location and the pattern of the Leydig cells were not compatible with the typical findings of Leydig cell tumors and Reinke crystals were not present in our cases. The differential diagnosis from the steroid cell tumor was made with the above-mentioned microscopic appearance, and the lack of an observable mass on macroscopic evaluation.

The differential diagnosis also includes angiosarcoma and ovarian teratoma with a large hemangiomatous component. Grossly, angiosarcomas are usually unilateral, cystic, soft, friable and spongy with hemorrhage and necrosis. Histologically, angiosarcomas show marked cytologic atypia, pleomorphism, papillary endothelial tufting, necrosis, hemorrhage, and increased mitotic activity. Ovarian teratoma with a large hemangiomatous component was described by Feuerstein et al. (17). The lesions were distinguished from a pure hemangioma by the presence of skin adnexa or other ectodermal, endodermal, or mesodermal tissue components.

Hemangiomas of the ovary are usually discovered incidentally as in our cases. They may occasionally be large and symptomatic. There have been three previous reports of ovarian-hemangioma–induced stromal luteinization resulting in endometrial hyperplasia. To our knowledge, the ovarian hemangioma that synchronously existed with a well-differentiated endometrial carcinoma in case 1 is the second case that represents simultaneous occurrence of an endometrial carcinoma in conjuction with hemangioma of the ovary in the literature, and the absence of estrogen and progesterone receptors in the endothelial cells of the hemangioma suggests that ovarian hemangiomas may occur independent from estrogen and progesterone stimulation.

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