# Ovarian Cavernous Hemangioma Presenting as an Left Adnexal Mass with Elevated Ca-125: A Case Report

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Hemangiomas are benign vascular tumors that are rarely found in the ovaries. Ovarian hemangiomas are usually discovered incidentally at operation or autopsy. Sometimes, they present with abdominal mass and/or pain, acute abdomen, ascites or elevated CA-125 levels, simulating ovarian neoplasm. Hemangioma should be considered in the differential diagnosis of hemorrhagic ovarian lesions grossly, and proliferations of dilated blood vessels frequently seen in the hilar region of the ovary, lymphangioma, teratoma with a prominent vascular component and angiosarcoma microscopically. The treatment of choice is oophorectomy or adnexectomy, which results in a complete cure. We report a case of very rare tumor of the ovary with an unusual presentation; an ovarian cavernous hemangioma with elevated serum CA-125 levels and uterine leiomyoma.

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**Key Words:** Ovarian hemangioma, CA-125, Cavernous hemangioma, Ovarian neoplasm

## Introduction

Hemangioma of the ovary was first described by Payne in 1869 (cited by Talerman).[1] There are fewer than 50 documented cases in the literature. The majorities of ovarian hemangiomas are of the cavernous type and may present either as isolated ovarian masses or in conjunction with diffuse abdominopelvic hemangiomatosis. Most cases of ovarian hemangiomas are small and are incidental findings at operation or autopsy. Large lesions tend to present clinically with symptoms typical of an adnexal mass such as abdominal pain, acute abdomen, ascites or elevated CA-125 level.[1-5]

We report a case of a cavernous ovarian hemangioma in a 46-year-old woman presenting with adnexal mass, elevated serum CA-125 levels and uterine leiomyoma.

## Case Report

A 46 year old multiparous postmenopausal woman underwent total abdominal hysterectomy and bilateral salpingo-oophorectomy for uterine leiomyoma and left ovarian mass. Serum CA-125 levels was 190 IU/ mL (35< normal). Serum CA19-9 and carcinoembryonic antigens were normal. Findings were negative for Papanicolaou cytology at the uterine cervix and peritoneal fluid. The left ovary measured 6x3x2 cm. A hemorrhagic mass occupied most of the ovary. The cut surface of the ovary was spongy and had a hemorrhagic appearance. Microscopic examination revealed a cavernous hemangioma, consisting of multiple thin walled blood vessels, filled with red blood cells. All of the vessels lined with a single layer of flattened endothelium. There was no necrosis, mitotic activity and atypical cells. The lesion replaced the majority of the ovary (Figure 1a-1b). Immunostains confirmed a vascular origin for the tumor. The tumor cells stained positive with CD31, CD34, and factor VIII "related antigen" (Figure 2-3). The right adnexa was normal. The uterus revealed an intramural leiomyoma of 16 cm in diameter and secretory endometrium. After operation, patient's serum CA-125 levels returned to the normal range.

*Figure 1a*

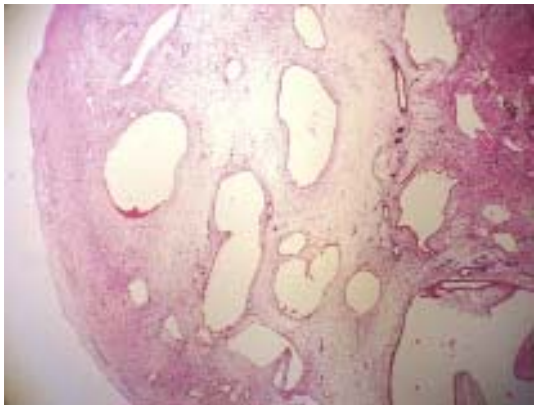

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*Figure 1b*

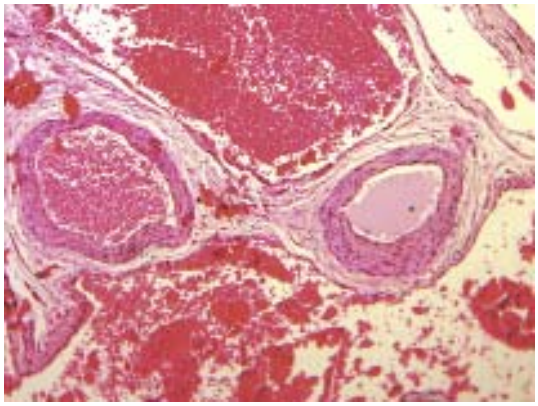

*Figure 1a - 1b : Microscopic apperance of the cavernous hemangioma (H&E, 1a; x10, 1b; x60)*

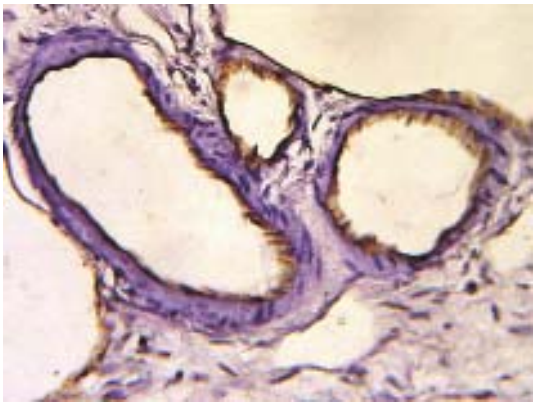

*Figure 2: Cavernous hemangioma immunohistochemically stained for Factor VIII RA (B-SA, DAB, X 200)*

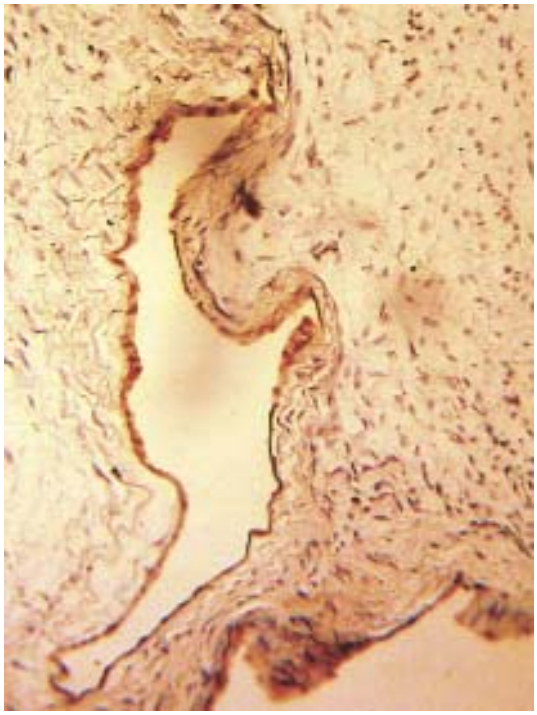

*Figure 3: CD34 expression in tumor (B-SA, DAB, X 200)*

## Discussion

Hemangioma is found only occasionally in the ovary; the number of well- documented cases does not exceed 50. This is somewhat surprising, as the ovary has a very rich and the complex vasculature. Some authors have explained this on the basis of cyclical changes during reproductive period.[1-4]

The origin of ovarian hemangioma in common with hemangioma in general is a matter of controversy; it is considered either a hamartomatous malformation or a true neoplasm. It is likely that both modes of origin are responsible for their formation.[2,3]

The reported age of patients with ovarian hemangioma ranges from 4 months to 63 years and does not show predominance in any decade.[2-4] Our case is a 46 year old female and is within the limits reported in the literature.

Ovarian hemangiomas are usually discovered incidentally at operation or autopsy. Sometimes, they present with abdominal mass and/ or pain, acute abdomen, ascites or elevated serum CA-125 levels, simulating ovarian neoplasm.[5,6] Timmerman[7] and Lin[8] have suggested that the changes occurring in the peritoneal mesothelium lead to ascites and CA-125 elevation. Kaneta et al. have reported that mesothelial cells on the surface of hemangiomatous ovarian tissue express CA-125 immunohistochemically.[6] Ovarian hemangiomas have been noted in patients with generalized hemangiomatosis and in patients with hemangiomas in other parts of the genital tract. Ovarian hemangiomas are usually unilateral, though bilateral cases have been reported.[1-3, 9-13] The lesion was unilateral in our case, too and serum CA-125 levels were higher than normal, and upon radiological examination there was a mass appearance in the left ovary. No systemic hemangiomatosis findings were detected in clinical and pathological examinations.

Macroscopically, the lesions are small, red or purple, round or oval nodules, measuring from a few millimeters to 11.5 cm in diameter. On cut section, they usually are spongy and show a “honeycomb” appearance. Although they have been found in different parts of the ovary, the medulla and the hilar region appear to be the most common sites.[1-4] In our case the lesion had a diameter of 6 cm and was occupying almost the whole ovarian tissue and contained hemorrhagic cystic structures.

Microscopically, ovarian hemangioma is of the cavernous, capillary or mixed capillary- cavernous type. In contrast to vascular tumors in other parts of the body, the most common histological type in ovary is cavernous or mixed cavernous-capillary type.[1-4] Our case had the cavernous type of hemangioma that is noted in the literature as the most frequently encountered type of hemangioma of the ovary.

Ovarian hemangioma must be differentiated from prolifer-

ations of dilated blood vessels, frequently seen in the hilar region of the ovary. In order to be regarded as a true hemangioma, a mass of vascular channels, large as well as small, and with minimal amounts of stroma should form a reasonably circumscribed lesion distinct from the remainder of the ovary. The presence of numerous blood cells within the vascular spaces and the absence of pale eosinophilic homogenous material usually distinguish hemangioma from the less common lymphangioma. Hemangioma also must be distinguished from teratoma with a prominent vascular component. In such cases, careful sampling will detect other teratomatous elements, the presence of which distinguishes the lesion from a hemangioma.[12] Other differential diagnostic entities included an angiosarcoma of the ovary. The absence of characteristic features such as marked cytological atypia, papillary endothelial tufting, necrosis, hemorrhage, and increased mitotic activity distinguish hemangioma from the less common angiosarcoma. In our case, grossly there was a distinct hemorrhagic mass involving almost the entire ovary. Upon microscopic examination the lesion was evaluated as a cavernous hemangioma since it was comprised of enlarged vascular structures lined with benign, single-layered endothelium with erythrocytes in their lumens and since other teratomatous elements were not found.

Some of the ovarian hemangiomas are difficult to differentiate clinicoradiologically from other neoplastic conditions. Pathologic examination is necesssary in all such cases to exclude a possibility of malignant tumors. Associated conditions like generalized hemangiomatosis and stromal luteinization should be excluded.[5,9,11-13]

In summary, we report a case of a very rare tumor of the ovary with an unusual presentation; an ovarian cavernous hemangioma with elevated serum CA-125 levels and uterine leiomyoma. Although it is very unusual, an ovarian hemangioma may present with elevated CA-125 and the differential diagnosis from ovarian cancer should be considered.

## Ca-125 Yüksekliği Ile Birlikte Sol Adneksial Kitle Bulgusu Veren Overin Kavernöz Hemanjiomu

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Benign vasküler tümörler olan hemanjiomlar overde nadiren görülürler. Ovarian hemanjiomlar genellikle operasyon veya otopsi sırasında rastlantısal olarak saptanırlar. Bazı olgularda ise abdominal kitle/ ağrı, akut batın, asit veya yükselmiş CA–125 seviyeleri gibi over neoplazilerini taklit eden bulgular görülebilir. Hemanjiomlar makroskobik olarak hemorajik over lezyonlarından, mikroskobik olarak ise overin hiler bölgesinde sık görülen genişlemiş vasküler proliferasyonlardan, lenfanjiomdan, vasküler komponenti belirgin teratomdan ve anjiosarkomdan ayrılmalıdır. Tedavide etkilenmiş overin veya adneksin cerrahi çıkarımı yeterlidir. Bu çalışmada oldukça nadir görülen, yüksek serum CA–125 düzeyi ve uterin leiomyoma ile birliktelik gösteren ovarian kavernöz hemanjiom olgusu sunulmuştur.

**Anahtar Kelimeler:** Over hemanjiomu, CA-125, Kavernöz hemanjiom, Over tümörü

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