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Case Report

Ovarian Haemangioma with Synchronous Xanthogranulomatous Inflammation - A Rare Pathological Finding

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ABSTRACT

Ovarian hemangiomas are extremely rare tumors of the female genital tract. 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 a case of cavernous ovarian hemangioma with xanthogranulomatous salpingooophoritis in the contralateral adnexa in a 61 year old patient who presented with postmenopausal bleeding and pain abdomen.

Keywords: ovarian haemangioma, vascular tumor, xanthogranulomatous salpingo-oophoritis, female genital tract.

INTRODUCTION

Vascular tumors of the Female Genital Tract (FGT), especially of the ovary, constitute a very small percentage of all the tumors of FGT. There are only a few case reports and short series of these tumors reported in the literature. [1, 2] Ovarian haemangiomas are an extremely rare benign vascular tumors,most commonly of cavernous type. [1] They are nonfunctional vascular neoplasm, usually discovered incidentally or in conjunction with diffuse abdominopelvic haemangiomatosis, endometrial hyperplasia and rarely associated with gynecological cancers.

The pre-operative diagnosis of ovarian hemangiomas may be facilitated by radiological methods, thus making it possible to avoid radical surgery. [3] Simple oophorectomy is curative for ovarian hemangioma. So, a clinicopathologic correlation is usually essential. [3, 5] Here we report a case of a post-menopausal woman who presented with pain abdomen and vague abdominal mass which showed features of ovarian hemangioma on one side of ovary and xanthogranulomatous oophoritis on contralateral side of other ovary.

CASE REPORT

A 61 year old woman was admitted to the Department of Obstetrics and Gynecology for pain in the hypogastrium and vague abdominal mass. The medical history of the patient was significant with

diabetes mellitus and hypertension. . Pelvic examination and ultrasonography revealed normal sized uterus with left tender adnexal mass(Tubo-ovarian mass). CAT scan confirmed a thick walled multiloculated left ovarian cyst with a solid component .Total abdominal hysterectomy with bilateral salphingo-oophorectomy was performed and the patient was discharged after an uneventful post-operative course. Specimen fixed in 10% formalin was sent for histopathological examination.

Grossly, the uterus, cervix and right tube appeared unremarkable. The right ovary measured 3.5x2.0x1.0cm with sectioned surface showing brownish blue ill-defined cystic area filled with blood clot (Fig.1). The left tubo-ovarian mass measured 6x5x1 cm. Cut surface was grey brown .Tubal lumen was obliterated .The left ovary showed a foci of yellowish white solid area.

Fig 1- Sectioned surface showing cystic spaces filled with blood.

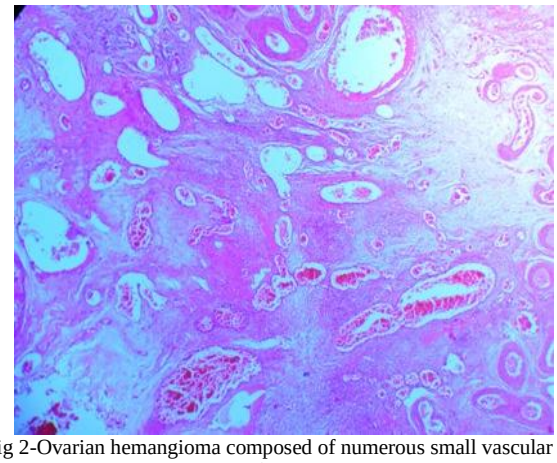
Fig 2-Ovarian hemangioma composed of numerous small vascular spaces, lined by a single layer of endothelial cells. (H&E X10).

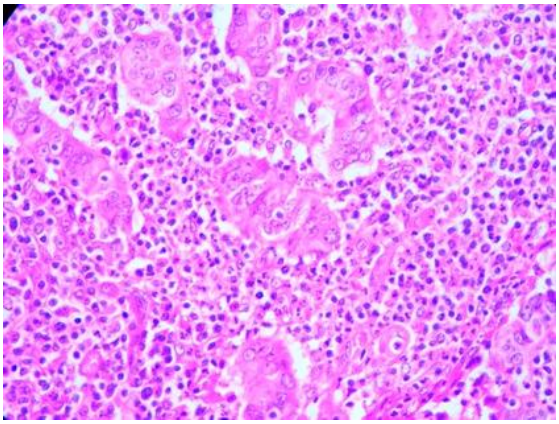
Fig 3-Ovarian tissue with foam cells and multinucleated giant cells. (H&E X40)

Histopathologically, most of the right ovary was replaced by numerous dilated thin walled vascular channels separated by connective tissue septa. The vessels were lined by single layer of endothelium without atypical features, consistent with features of cavernous hemangioma (Fig 2). Sections from left tubo-ovarian mass showed ovarian stroma with areas of diffuse and dense mixed inflammatory cell infiltrate of foamy macrophages, neutrophils, lymphocytes, plasma cells and congested blood vessels. The above findings were consistent with left xanthogranulomatous salphingo-oophoritis (Fig 3).

DISCUSSION

Hemangioma of the ovary was first described by Payne in 1869. [3] Ovarian hemangiomas are commonly discovered incidentally at autopsy or surgery. Ovarian

hemangiomas are benign lesions arising from failure in vascular formation, particularly in the canalizing process, forming abnormal vascular channels. [2, 4] They are of two types, capillary and cavernous type. There are about 50 documented cases in the literature. These tumors are usually unilateral, though bilateral cases have been reported. [4] They have a wide age group which ranged from 4 months to 81 years and size varied from 0.3 to 24 centimeter. [3] Ovarian hemangiomas are usually situated in the medulla and the hilus. Sometimes they present with an abdominal mass and /or pain and acute abdomen or ascites, simulating the more common ovarian neoplasms.

The etiology of ovarian hemangiomas is unknown and controversial. It may be a true tumor or hamartoma or stimulated vessels by hormonal influences, congenital malformations, pregnancy or infection. Though ovarian hemangiomas are non-functional, however it is well known that leutinization of the ovarian stromal cells commonly occurs as a reactive phenomenon, and that it may be associated with androgenic, estrogenic or progestrogenic effects. [1, 3] Other hypothesis is 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. This hypothesis is based on the fact that estrogens have known growth stimulatory effects on the vasculature and that most hemangiomas carry estrogen receptors. The conditions commonly associated with the tumor are thrombocytopenia, ascites, and stromal luteinization with or without ascites and endometrial hyperplasia or carcinoma. [3]

The differential diagnosis includes steroid cell tumor, angiosarcoma and ovarian teratoma with a large hemangiomatous component. [1, 3, 5] The lesion has a smooth outer surface and is red or purplish on the cut surface. In contrast to the vascular tumors in other parts of the body, the most common histologic type found in the ovary is the cavernous or mixed cavernous-capillary type. [3, 4]

Ovarian teratoma with a large hemangiomatous component was described by Feuerstein et al. The lesions were distinguished from a pure hemangioma by the presence of skin adnexa or other ectodermal, endodermal or mesodermal tissue components. [4] The pre-operative diagnosis of ovarian hemangiomas may be facilitated by radiological methods, thus making it possible to avoid radical surgery. On MRI hemangiomas should be considered as it is a richly vascularised tumor and its prominent blood flow is detected on color Doppler sonography. [3, 4]

Xanthogranulomatous salphingo-oophoritis was present in the contralateral adnexa.This is a rare form of chronic granulomatous inflammation. Bacterial infections, immunosuppression, chronic inflammatory conditions, luminal obstruction, endometriosis, leiomyoma, abnormal lipid metabolism, ineffective antibiotic therapy, ineffective clearance of bacteria by phagocytes and chronic irritation of the urachal remnant have been implicated in the pathogenesis. [6] There are very few reported cases of xanthogranulomatous salpingitis and oophoritis in the literature. Ultrasound and CAT scans demonstrate a well defined solid mass. The mass on gross examination reveals yellowish necrotic material. The affected organs suffer disorganization and infiltration with focal or sheets of foam cells admixed with chronic inflammatory cells such as lymphocytes, plasma cells, occasional neutrophils with or without multinucleated or Touton giant cells. [3, 6]

Treatment of choice for both the lesions synchronously is surgical

intervention. To the best of our knowledge, this is the first reported case that demonstrates a vascular and chronic granulomatous inflammation that demonstrates different stages in the pathogenesis of the lesions and provides an insight into the histiogenesis of this entity.

REFERENCES

1. Filiz Bolat, Serkan Erkanli, Nazım Emrah Kocer. Ovarian Hemangioma: Report of Two Cases and Review of the Literature. Turkish Journal of Pathology. 2010 Cilt 26, Sayı 3, Sayfa (lar) 264-266.
2. Uma S. Andola, Sainath K. Andola. Vascular Tumours of the Female Genital Tract: A Clinicopathologic Study of 11 Cases.Journal of Clinical and Diagnostic Research.2011, vol 5 issue 6, Pg 1241-1246.
3. Metin Akbulut., Ferda Bir,Nagihan Colakoglu et al.Ovarian hemangioma occurring Synchronously with serous papillary carcinoma oo the ovary and benign endometrial polyp.Ann Saudi Med 2008;28(2):128-131.
4. Hatci Bayramoglu,Tayfun Gungor, Murat oz, et al. Primary ovarian haemangioma:case report and review of literature .Medical Journal of Islamic World of Sciences;2012 :20:3,106-110.
5. S.Shariat Torbaghan, R. Nazari,S. Jolodari et al. Ovarian hemangioma: A rare Pathologic Finding.Acta Medica Iranica,Vol 36 No 1:Pg 44-45.
6. Nese Yener, Erdin IIter, Ahmet Midi. Xanthogranulomatous salpingitis as a rare pathologic aspect of chronic active pelvic inflammatory disease. Indian Journal of Pathology and Microbiology: January-March 2011:54(1).

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