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Female Adnexal Tumor of Wolffian Origin (FATWO): A Rare Ovarian Tumor

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ABSTRACT

FATWOs are rare tumors arising from the remnants of the mesonephric duct with a low malignant potential. Here we report a case of 55 year old post menopausal female with abdominopelvic mass. The diagnosis of FATWO becomes challenging because of the rarity and heterogeneity of its histologic appearances. Therefore, a careful histo-pathological and immunohistochemical evaluation should always be performed.

Keywords: Adnexal mass, Wolffian origin, benign.

INTRODUCTION

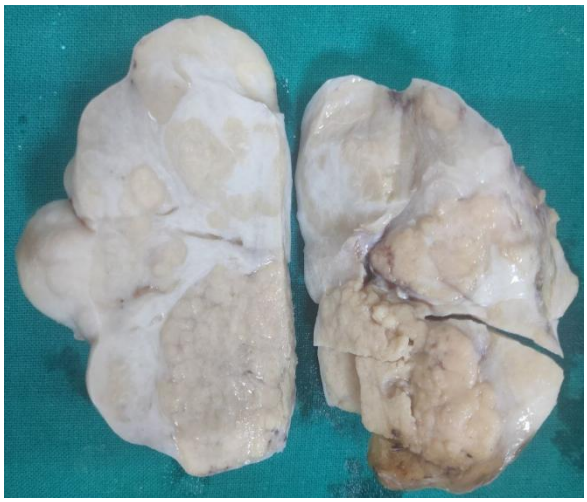
Adnexal tumors of Wolffian duct origin (FATWO) in the female population belong to the group of epithelial tumors that are rare. They were first described in 1973 by Kariminejad and Scully[1]. Reports of similar tumours occurring in the ovary[2,3] and in the male genital tract[4] were reported subsequently but they are uncommon. The ovarian and extraovarian tumours show the same morphology and are considered to have the same origin. The belief that they are of mesonephric origin is based on: (a) the finding that they are found in the same sites as mesonephric remnants; (b) their morphological dissimilarity to other ovarian tumours of either epithelial-stromal or sex cord-stromal types; and (c) some ultrastructural[5,6] and immunohistochemical[7-9] homology with the mesonephric duct. Their recognition and diagnosis depend largely on anatomical site and a morphology which is typically varied.

CASE REPORT

A 55 year old post menopausal women presented with pain abdomen and abdominal distension. On CT abdomen a solid cystic heterogenous enhancing abdominopelvic masses were seen. Serum CA 125 levels were mildly raised (62.2 units /ml) while serum CEA and CA19.9 were normal. Patient had undergone hysterectomy 7 years back for leiomyoma. A clinical diagnosis of carcinoma ovary was kept and the abdominopelvic masses were surgically removed and sent for histopathology examination. We received bilateral ovarian masses with attached fallopian tubes. The ovarian masses measured 17x16x14 cm and 16x10x6 cm. Outer surfaces of both were well encapsulated with nodular and lobulated appearance with no breach in the capsule.

On cut surface the ovarian masses were solid with pale yellow to grey white/ tan homogenous appearance (Figure 1). We also received a part of omentum measuring 30x5 cm which showed no lymph node or tumor deposits. On microscopy, the sections from bilateral ovarian masses revealed nodular

growth pattern with nests, cords, trabeculae, tubules/ glands of tumor cells with intraluminal bright eosinophilic secretions (Figure 2). The tumor cells were polygonal with mildly pleomorphic central round nuclei with fine chromatin, variably conspicuous small nucleolus, focal intranuclear grooving and moderate amount of eosinophilic cytoplasm. Occasional mitotic figures were seen. The stroma was fibrous with foci of hyalinization and myxoid change (Figure 3,4). A diagnosis of female adnexal tumor of probable Wolffian origin (FATWO), bilateral ovaries was given and IHC was advised to the patient. On immunohistochemistry pancytokeratin, CK 7 and CD10 was immunoreactive while PAX 8, CK20 and CEA were non immunoreactive which confirmed the diagnosis of FATWO.



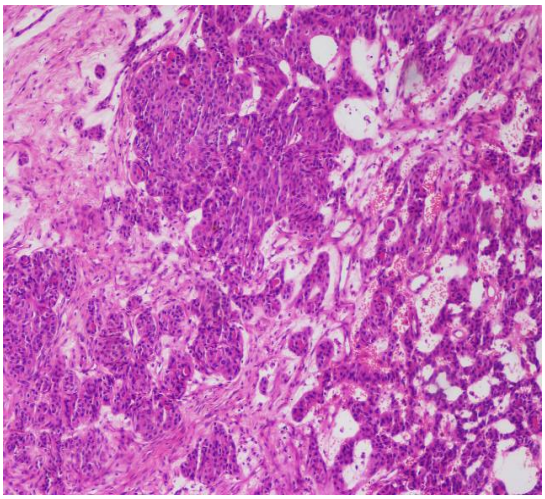
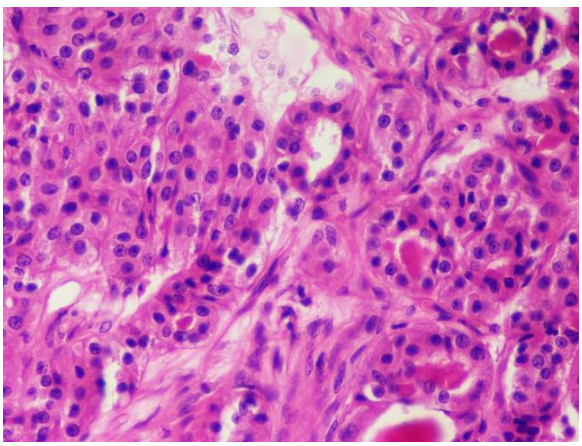


Figure 1: gross image of ovarian mass revealing pale yellow to grey white homogenous appearance on cut surface.
Figure 2: Ovarian tumor showing nests, trabeculae and tubules of tumor cell in a fibrous stroma with myxoid change. (H&E, 100X)



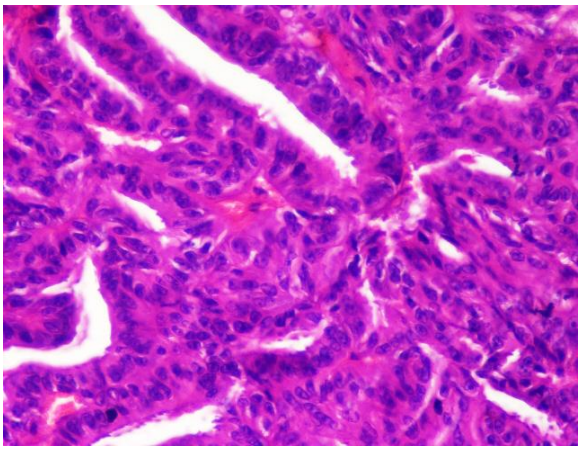


Figure 3: High power view showing poygonal tumor cells with central round nucleus, fine chromatin and eosinophilic cytoplasm with bright eosinophilic secretions. (H&E, 400X)
Figure 4: High power view revealing tumor cells arranged in glandular pattern resembling endometriod adenocarcinoma (H&E, 400X)

DISCUSSION

The diagnosis of FATWO becomes challenging because of the rarity and heterogeneity of its histologic appearances. FATWOs are quite rare tumors with approximately 100 cases documented in the literature. The patients range in age from 15 to 87 yr. Common clinical presentations are lower abdominal pain/distention, pelvic mass, or abnormal genital bleeding, but some patients often have an incidental finding. The symptoms are usually similar to ovarian tumors or uterine and intraligamentous leiomyoma. FATWO is reported primarily to originate within the

broad ligament which is coincident with Wolffian duct remnants.

On MRI, FATWOs are described as slightly hyperintense adnexal masses with cystic degeneration which is similar to subserosal leiomyomas and other ovarian tumors such as thecomas [10]. On CT the lesions can be cystic or solid with heterogeneous enhancement and ultrasound reveals similar findings with typically well-vascularized flow. FATWOs commonly have some calcified component which is visualized on both radiologic and gross examination[10,11].

Most FATWO tumors are unilateral and are well-encapsulated with a solid, rubbery, partially cystic, and often, a hemorrhagic appearance on the cut surface. They may vary from 0.8 to 20 cm in diameter.

Kariminejad and Scully[1] have highlighted varied morphologies of FATWO including tubular, trabecular, solid, and microcystic (sieve-like) growth patterns. The tumor cells comprise a small cytoplasm and show the form of epithelial cells; in some instances, the tumor cells resemble a spindle. In addition, tubular structures without mucus secretion and Müllerian tubes on the origin of tumor histological features can support the diagnosis. Gartner et al[12] suggested three major differences between Wolffian and Müllerian structures in microcystical features: (a) Wolffian epithelial cells and nuclei are significantly smaller than those of Müllerian cysts; (b) Wolffian ducts and tubules have a better-defined basement membrane than Müllerian ducts and tubules; and (c) Müllerian epithelium responds to cyclical hormonal stimulation, but Wolffian epithelium does not.

Features of aggressiveness of tumors are cellular atypia, necrosis, capsular invasion and an increased number of mitosis but malignant cases with minimal nuclear atypia and a very low mitotic figure may also occur. Metastasis may bwe seen at the time of presentation or few years after initial presentation. Most common site of metastases are liver and lung[5,13].

Based on clinical presentation, intra-operative examination, and pathological analysis, the differential diagnosis for a FATWO includes endometrioid adenocarcinomas and sex cord stromal tumors such as Sertoli-Leydig cell tumor and granulosa cell tumor. The location of the tumor is an important factor in supporting the diagnosis of a FATWO. FATWOs typically do not involve the fallopian tube or ovary and most commonly arise in the broad ligament, whereas Sertoli-Leydig tumors have not been identified in the paratubal area or broad ligament. Endometrioid adenocarcinomas that mimic FATWOs typically involve the fallopian tube or ovarian parenchyma [14].

Although there is no single specific immunohistochemical stain for FATWO, immunohistochemistry can help in narrowing the differential diagnosis. Endometrioid carcinoma typically has diffuse staining of EMA, PAX8, CK7, ER and vimentin while in Wolffian tumors, EMA and PAX8 are typically negative[15], Distinguishing FATWO from sex cord stromal tumor is a challenge for pathologists. The overlapping stains are calretinin, inhibin, and CD10[16]. However, sex-cord stromal tumors are typically diffusely positive for inhibin, whereas FATWO may have focal staining. Stains including CK7 and pancytokeratin (AE1/3), for which FATWO is immunoreactive. CK7 is not seen and AE1/3 is rarely seen (33-37%) in granulosa cell tumors. Variable expression of ER, PR, and c-Kit is reported in FATWO [13].

The most common treatment is total abdominal hysterectomy with bilateral salpingo-oophorectomy since multiple recurrences have been reported. The role of chemotherapy/radiotherapy is not clear in the management[13]. As these tumors are rare therefore the prognosis of this tumor cannot be accurately assessed.

CONCLUSION

FATWOs are rare tumors arising from the remnants of the mesonephric duct with a low malignant potential. Due to its rarity, the diagnosis can be difficult and

challenging and is one of exclusion, Therefore, a careful histopathological and immunohistochemical evaluation should always be performed.

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