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RESEARCH ARTICLE

OVARIAN FIBROTHERCOMA: A CASE REPORT

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ABSTRACT

Fibrothecoma is a rare type of ovarian sex cord stromal tumor with only a few case reports available in the literature. Histologically, they resemble both fibroma and thecoma. Thecomas are composed of lipid-containing cells that resemble theca interna cells. Fibromas show spindle, oval, or round cells forming variable amounts of collagen. Here we report a case of 52 years old postmenopausal lady who presented with pain abdomen. Clinically, she had a firm lump predominantly in the right flank and her sonographic reports suggested a solid cum cystic mass in right ovary. She was operated with a clinical suspicion of ovarian malignancy but the histopathological examination confirmed it to be a benign fibrothecoma.

INTRODUCTION

Ovarian tumors represent tumors of epithelial, germ cell, and sex cord-stromal origin. Common epithelial tumors are the commonest histological subtype seen accounting for approximately 60% of all ovarian tumors, followed by germ cell tumors. (Kurman *et al.*, 2014) Ovarian sex cord-stromal tumors are uncommon neoplasms that represent approximately 5% of all ovarian tumors. These tumors comprise a heterogeneous group and are formed by diverse cell types that arise from the primitive sex cords or stromal cells. (Haroon *et al.*, 2013) The stromal cells include theca cells, fibroblasts, and Leydig cells whereas the gonadal primitive sex cords include granulosa cells and Sertoli cells (Wilkinson *et al.*, 2008). These cell types may be present separately or admixed and display different degrees of differentiation. Fibrothecoma of ovary is a tumor arising in stroma and containing a mixture of cells comprising of fibroblasts, producing collagen like a fibroma and spindle cells with lipid droplets characterizing a thecoma. (Pratt, 2004) Fibrothecoma of the ovary is very rare as it only accounts for 1.2% of all ovarian cancers. (Quirk *et al.*, 2005). In the present study, we report a case of ovarian fibrothecoma in a postmenopausal lady with a clinical and radiological suspicion of malignancy.

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CASE REPORT

A 52yrs old postmenopausal, female presented with pain lower abdomen and gaseous distension for the last 1 month. She had a history of two spontaneous abortion and no live birth. There was a history of irregular treatment for infertility. She was hypertensive and diabetic on oral medication for the last 3 years. On examination, a midline lump about 12 weeks size was found in pelvis which was more towards the right side. Per vaginum examination showed that both the vaginal fornices were full and there was a lump, which was hard, relatively fixed and more towards right adnexa. Uterus could not be palpated separately from the lump. The cervix was normal and there was no vaginal bleeding or discharge.

Complete blood count and routine blood biochemistry was normal. The level of CA-125 was 28.40U/ml. Abdominal and pelvic ultrasound showed a right adnexal solid and cystic lesion of 68mm x 40mm. A cyst of size 42mm x 30mm was seen in left ovary. The uterus was normal in shape and size. There was no ascites or lymphadenopathy. The patient underwent an exploratory laparotomy. A right sided solid mass, with stretched but intact capsule and left ovarian chocolate cyst was found intraoperatively. Bilateral salphingoopherectomy was done as the uterus could not be removed because of the presence of adhesions. The left ovary with the cyst was easily removed, however there was presence of adhesions around the right ovary and it had to be

meticulously dissected before removal. The removed left ovarian cyst and right ovarian mass were then sent for histopathological examination. The macroscopic examination showed a partly cut open cystic 4.5cm x 3cm x 0.6cm left ovary with a soft grey brown to light brown cut surface. The right ovary was seen as one grey white soft tissue piece measuring 8cm x 5cm x 4cm with a grey white cut surface. The microscopic examination showed a chronic hemorrhagic left ovarian cyst with no viable lining epithelium. Multiple sections taken from the right ovarian mass showed nests of oval to polyhedral cells with uniform round nuclei and clear to eosinophilic cytoplasm. These nests were surrounded by fibroblastic proliferation without any atypia or mitotic activity. The final histopathological diagnosis was confirmed as benign fibrothecoma. The post-operative period was uneventful and the patient recovered completely.

DISCUSSION

Fibromas are the most common sex cord-stromal tumors comprising of spindle stromal cells that produce a collagenous stroma. (Chen *et al.*, 2003) These fibromas range in size from small to large lesions. Small lesions are frequently asymptomatic, but the patient can present with pelvic discomfort or acute abdominal pain due to ovarian torsion as the size increases. (Montoriol *et al.*, 2013) Though microscopically they often show no atypia or mitotic figures, 10% of all ovarian fibromas may demonstrate low malignant potential, exhibiting mild nuclear atypia, and more than four mitotic figures per 10 high-powered fields. These generally exhibit the same clinical manifestations but tend to be larger than fibromas, thus potentially leading to necrosis and hemorrhage. (Kurman *et al.*, 2014) Ovarian fibrosarcomas are rare entities that follow a malignant clinical course and tend to exhibit moderate-to-severe nuclear atypia and mitotic figures. These tumors usually present in postmenopausal women as large unilateral masses. (Ray *et al.*, 2012) Thecomas are composed of lipid-laden stromal cells that resemble theca cells, which usually encircle the ovarian follicles, and exhibit estrogenic activity in most cases. They account for 0.5%–1% of all primary ovarian tumors and are more likely to occur in postmenopausal women. They are usually benign but the patient may experience estrogen-related symptoms such as uterine bleeding and endometrial hyperplasia. (Tanaka *et al.*, 2004) Thecomas that manifest in combination with fibrous tissue may be classified as fibrothecomas. They show significant cellularity, a relatively large amount of collagen and, in up to 50% of cases, pronounced edema. The collagenous component is usually fibrillary in appearance but hyalinization is sometimes seen. The typical thecal cells may present in small clusters, often being observed only on close examination of several sections, or as larger and isolated fields. The presence of luteinized thecal cells would change the diagnosis to luteinized thecoma. (Scully *et al.*, 1999). These tumors are usually seen in the postmenopausal age group similar to that seen in our report. The clinical presentation of ovarian fibrothecoma is usually nonspecific such as pelvic and abdominal pain or distension. However, some ovarian thecomas may be associated with estrogenic manifestations such as irregular bleeding, menorrhagia and endometrial hyperplasia. Similarly, ovarian fibromas may be associated with Meig's syndrome, which is defined as a triad of ovarian fibroma, unilateral hydrothorax and ascites. (Choi *et al.*, 2006)

It can also be associated with basal cell nevus syndrome, comprising of bilateral ovarian fibromas, multiple basal cell carcinoma of skin and odontogenic keratocysts. (Howell *et al.*, 1990) However, in our case none of the above abnormalities were reported. Grossly, ovarian fibrothecomas are unilateral in about 90% of all cases and are usually solid, spherical or slightly lobulated, encapsulated hard gray white masses covered by an intact ovarian serosa. However, ovarian fibrothecomas of size larger than 10 cm tend to be associated with varying degree of edema, myxoid change and cystic degeneration. Their characteristic sonographic appearance is of a round or oval solid tumor, with regular margins. They may have stripy acoustic shadows, but these are present in just a small percentage of cases. Fibrothecomas can also show cystic areas, due to hemorrhage, edema or necrosis within the stromal tissue. (Paladini *et al.*, 2009).

CA-125 is a tumor marker used for evaluating ovarian tumors. Our patient had a normal value of CA-125. Generally, fibrothecomas are not associated with an increased level of CA125. However, there are reports of thecoma and fibrothecoma associated with Meig's syndrome and elevated CA-125 values upto 600 IU/ml. Choi K *et al* in 2006 reported a case of granulosa cell tumor associated with Meig's syndrome and elevated CA-125 levels of 82.49 IU/ml. (Choi *et al.*, 2006) Macci *et al* in 2014 reported an ovarian fibroma in association with Meig's syndrome, hemolytic anemia and raised CA-125. (Macci *et al.*, 2014). The presence of hemorrhagic cyst in the left ovary along with adhesions around the uterus suggests endometriosis as the most probable cause of infertility seen in this patient. Incidentally, a case of secondary amenorrhea and infertility due to Inhibin B producing ovarian fibrothecoma has also been reported by Meyer *et al* in 2000 and this could be a contributory factor. (Cho *et al.*, 2013) However ashormonal estimation was not done in this case no further comments could be made in this regard. Fibrothecomas have a very good prognosis and the treatment depends primarily on the age of the patient. In case of young patients, laparoscopic tumorectomy can be done, whereas in postmenopausal women radical surgery in terms of bilateral salpingoophorectomy is indicated. (Cho *et al.*, 2013)

Conclusion

Ovarian sex cord-stromal tumors are infrequent tumors, which develop from cells arising from the primitive sex cords or the stroma. They are usually benign and can be treated with simple surgical excision. However, the rarity of sex cord-stromal tumors contributes to a low index of suspicion; and therefore, a thorough knowledge of the clinicopathologic and radiologic findings of these tumors is important to facilitate surgical planning and avoid extensive morbid surgical procedures.

REFERENCES

- Angela, C. Meyer, John C., Papadimitriou, Steven G. Silverberg, *et al.* 2000. Secondary amenorrhea and infertility caused by an inhibin-B-producing ovarian fibrothecoma. *Fertility and sterility* vol. 73, no. 2, February.
- Chen, V.W., Ruiz, B., Killeen, J.L., Coté, T.R., Wu, X.C., Correa, C.N. 2003. Pathology and classification of ovarian tumors. *Cancer* 97:2631–2642.

- Cho, V.J., Lee, H.S., Kem, J.N. 2013. Clinical characteristic and surgical management options for ovarian fibrothecoma/fibroma: Gynecologic and obstetrics investigation. vol 76 :182-187.
- Choi, K., Lee, H.J., Pae, J.C., Oh, S.J., Lim, S.Y., Cho, E.Y. and Lee, S.S. 2006. Ovarian granulosa cell tumor presenting as Meigs' syndrome with elevated CA125. *Korean J Intern Med.*, 20:105-109.
- Haroon, S., Zia, A., Idrees, R., Memon, A., Fatima, S., Kayani N. 2013. Clinicopathological spectrum of ovarian sex cord-stromal tumors; 20 years' retrospective study in a developing country. *J Ovarian Res.*, 6:87.
- Howell, C.G., Jr, Rogers, D.A., Gable, D.S., Falls, G.D. 1990. Bilateral ovarian fibromas in children. *J PediatrSurg.*, 25: 690–691.
- Kurman, R.J., Carcangiu, M.L., Herrington, C.S., Young, R.H. 2014. Classification of tumors of the ovary. In: WHO Classification of Tumors, Volume 6. 4th ed. Lyon: IARC, 44–56.
- Macci, A., Madeddu, C., Kotsonis, P., Pietrangeli, M. and Paoletti, A.M. 2014. Large twisted ovarian fibroma associated with Meigs' syndrome, abdominal pain and severe anemia treated by laparoscopic surgery. *BMC Surgery*, 14:38.
- Montoriol, P.F., Mons, A., Da Ines, D., Bourdel, N., Tixier, L., Garcier, J.M. 2013. Fibrous tumours of the ovary: aetiologies and MRI features. *ClinRadiol* 68:1276– 1283.
- Paladini, D., Testa, A., Van Holsbeke, C., Mancari, R., Timmerman, D., Valentin, L. 2009. Imaging in gynecological disease (5): clinical and ultrasound characteristics in fibroma and fibrothecoma of the ovary. *Ultrasound Obstet Gynecol.* 34:188–195. doi: 10.1002/uog.6394.
- Pratt, J. 2004. Pathology of the ovary. 1st ed. Philadelphia: Saunders, 197–226.
- Quirk, J.T., Natarajan, N. 2005. Ovarian cancer incidence in the United States, 1992-1999. *GynecolOncol.* 97(2): 519-23.
- Ray, S., Biswas, B.K., Mukhopadhyay, S. 2012. Giant primary ovarian fibrosarcoma: Case report and review of pitfalls. *J Cytol* 29:255–257.
- Scully, R., Sobin, L. 1999. Histological Typing of Ovarian Tumours, vol. 9. Springer: New York, Berlin,.
- Tanaka, Y.O., Tsunoda, H., Kitagawa, Y., Ueno, T., Yoshikawa, H., Saida, Y. 2004. Functioning ovarian tumors: direct and indirect findings at MR imaging. *Radiographics* 24:S147–166.
- Wilkinson, N., Osborn, S., Young, R.H. 2008. Sex cordstromal tumours of the ovary: A review highlighting recent advances. *DiagnHistopathol.* 14(8): 388-400.
