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CASE STUDY

A CASE OF CYSTIC DEGENERATION OF OVARIAN FIBROTHECOMA

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ABSTRACT

Ovarian malignancies are on the rise worldwide. Important strategy in management is to differentiate benign from malignant lesions since for benign lesions ovariectomy is sufficient but malignant tumours require cytoreductive surgery with IV or intraperitoneal chemotherapy. Though imaging can help in diagnosis, histopathology is confirmatory. This report highlights the role of intraoperative frozen section in diagnosis of such malignancies.

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INTRODUCTION

The term fibrothecoma of the ovary is a term for a tumour of stromal cell origin. These tumours are rare as they only accounts for 1.2% of all ovarian cancers. (Quirk and, Natarajan, 2005) Histologically, fibromas are composed of variable amounts of collagen from entirely spindle, oval or round cells. The comas resemble theca interna cells of the ovary and are composed of lipids. There is a large histological and immunohistochemical overlap between the two that resulted in the term fibrothecoma. (Scully *et al*., 1998) The management of this type of tumour remains surgical and has good prognosis as they usually present as stage 1 lesions. (Jung *et al*., 2005)

Case presentation

A 45 year old post menopausal female came to us with complaints of pain in the abdomen for 6 months. On examination there was a abdominopelvic mass in the left iliac and supra public region. Mass was firm, mobile and non tender. Contrast CT scan of the abdomen was suggestive of solid to cystic lesion in the left adnexal region. Left ovary was not seen separately. Her CA 125 level was 102kU/ litre. Patient was planned for laparoscopy and proceed for cytoreduction surgery if required. On laparoscopy there was 24 x 17 x 9 cm predominantly cystic mass in the left adnexal region. Midline laparotomy was done and left adnexal mass was sent for frozen. (Figure 1) Frozen section was suggestive of benign tumour. Total abdominal hysterectomy with bilateral salpingo-opherectomy was done. On gross examination (Figure 2), surface was smooth. Cut section showed multiple large cysts largest of size 250 and 300 ml. Cysts contained clear fluid. No papillary projections, calcification or haemorrhage was seen. On microscopy (Figure 3) closely packed spindle cells seen at places in storiform pattern and at others showing lipid droplets and central nuclei. Evidence of myxoid degeneration and cystic changes was present. No evidence of epithelial lining, mitosis or atypia. Postoperative period was uneventful.Patient is now on follow up without any adjuvant treatment.

DISCUSSION

Ovarian fibrothecomas are benign tumours that come under sex cord- stromal tumour group of ovarian cancers. They composed of an admixture of fibrous and the comatous elements. (Salemis *et al*., 2011)

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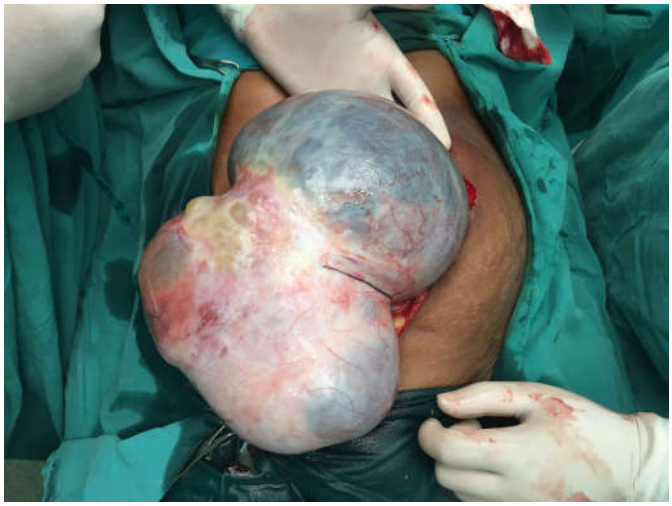

Figure 1. A 24 x 17 x 9 cm predominantly cystic mass arising from the left adnexal region

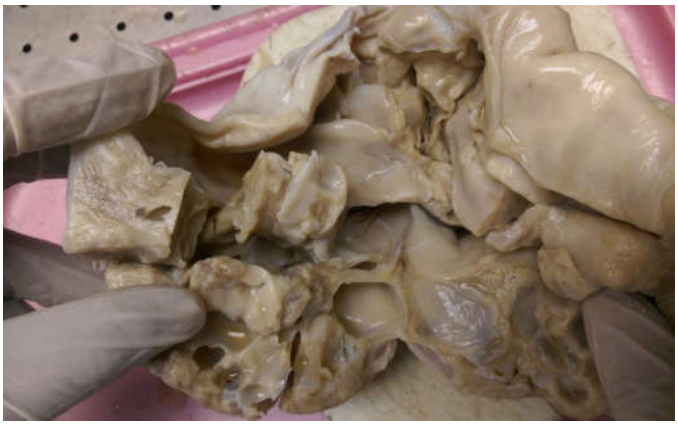

Figure 2. Cut section showing multiple cysts that largest of size 250 and 300 ml containing clear fluid. No papillary projections seen. No calcification or haemorrhage seen

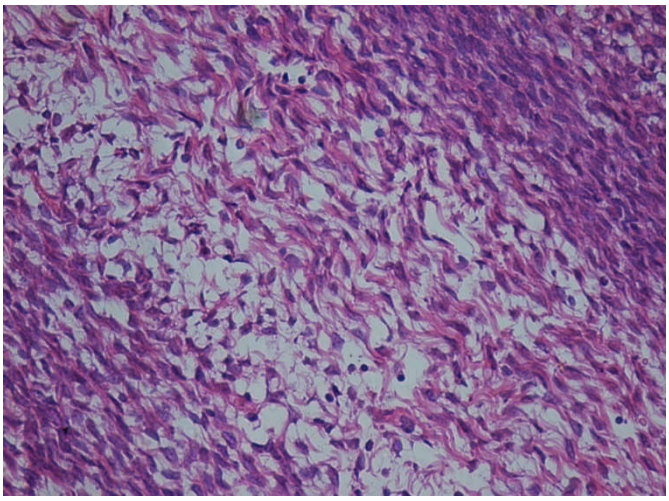

Figure 3. Closely packed spindle cells seen at places in storiform pattern and at others showing lipid droplets and central nuclei. Evidence of myxoid degeneration and cystic changes was present. No evidence of epithelial lining, mitosis or atypia

The reported incidence is between 0.4% and 8.0% of all ovarian tumors, and this wide variability can be attributed to the dif culty in differentiating fibrothecoma from ovarian fibromas or hyper- plastic stromal nodules (Stage and Grafton, 1977). They are rarely malignant and in 90% of the cases are unilateral (Salemis *et al*., 2011). These tumors occur generally in older menopausal females. However, some authors report 2 peaks of frequency: the first peak of onset is after menopause and the second is between 20 and 40 years. The occurrence of these tumors before the age of 20 is extremely rare (Laufer *et al*., 1996). The tumor is unilateral in 90 per cent of cases and with an average diameter of 6 cm. In 4.5 per cent of cases it may be more than 20 cm (Mawad and Hassanein, 1994). Fibrothecomas are usually asymptomatic and are typically found incidentally. (Sivanesaratnam *et al*., 1990) Patients present with nonspecific complaints like pelvic pain and metrorrhagia. The clinical examination generally finds a solid, mobile tumor with a regular surface and a variable size (Sivanesaratnam *et al*., 1990) Rarely, fibrothecomas present with endocrine manifestations related to hormonally active tumors. In these cases, estrogenic or even some rare androgenic manifestations are reported (Siekierska-Hellmann *et al*., 2006). Ovarian fibrothecomas can be associated with Meigs' syndrome, which is defined as the triad of ascites, unilateral hydrothorax, and benign ovarian tumor (Abad *et al*., 1999). Meigs' syndrome resolves with the removal of the benign ovarian mass. The presence of ascites, such as in Meigs' syndrome, can cause inflammation of the peritoneum and subsequently elevate the serum CA-125. However, CA-125 being a nonspecific test, can also be elevated in a number of nonmalignant conditions, including liver disease, pelvic inflammatory disease, benign fibroids, or pregnancy. CA-125 is expressed by mesothelial cells of the serosal membrane in pleura, pericardium, and peritoneum, and its production is increased with involvement of these structures (Takemori *et al*., 2000; Morán-Mendoza *et al*., 2006). In a series of 16 patients, 5 (28%) showed elevated levels of CA-125. (Yen *et al*., 2013) The vast majority of fibrothecomas behave in a benign fashion and malignant variants are exceedingly rare. Occasional cases of malignant fibrothecoma have been reported, but there is doubt as to whether a true malignant variant exists. (Waxman *et al*., 1979) On gross examination they are usually hard masses with a boss elated external surface. Edematous tumors may be soft in consistency and cyst formation is common. The cut surface is grey-white and homogeneous with a whorled pattern and occasional areas of calcification (Arya *et al*., 2008). Edema and cystic degeneration is common. Calcification and haemorrhage is rare. (Nikolaos *et al*., 2011) The histopathological varieties are the following:

- Fibromas, composed of spindly fibroblastic cells, with abundant cytoplasm, which produce collagen. Occasionally lipid laden thecal cells may be seen. The neoplasm is then called fibroth-ecoma.
- Thecomas, composed only of theca cells. These lipid-containing cells may produce estrogens or androgens. The neoplasm can be associated with en- dometrial carcinoma in 21% of the cases.
- Cellular fibromas, composed of more cellular fibroblasts containing cytonuclear atypia and high

number of mitosis. The tumor is benign if there are less than 3 mitosis per field. (Chechia *et al*., 2008)

Even though the definitive diagnosis of fibrothecoma is histologic, preoperative imaging can help in assessment. On sonography, presence of a homogenous echogenic pattern, with marked posterior acoustic absorption, is highly suggestive of a predominantly fibrous ovarian fibrothecoma. An echogenic pattern, with no posterior acoustic shadowing appears to be correlated to the presence of a mixed fibrothecoma, whereas, the finding of a diffusely hypoechoic mass, with no posterior echo enhancement, is characteristic of pure ovarian the comas. Sonographic findings, even though nonspecific, can provide the clinician with useful information which permits to detect these rare neoplasms preoperatively.

On computed tomography, fibrothecomas usually appear as a homogeneous solid tumor with varying degrees of enhancement. Calcification may be present and, as these tumors enlarge, myxoid or cystic degeneration may occur, resulting in a heterogeneous pattern. (Su Kon and Jin Wan Park, 2012) Differential diagnosis of fibrothecomas includes pedunculated and intraligamentous leiomyomas and other solid ovarianmasses such as Brenner tumors, granulosa cell tumors and dysgerminomas. In the presence of extensive cystic degeneration, the fibrothecoma can be easily mistaken for a malignant ovarian tumor. MRI can help differentiating. Fibrothecomas characteristically have low signal intensity on T1 and T2 images with post contrast enhancement. (Nikolaos *et al*., 2011) Treatment of these ovarian tumors is based on surgery. The surgical intention is always that of a complete resection of the mass, or even en bloc resection with adjacent or infiltrated organs and structures. As it is benign surgery is curative and adjuvant treatment is not required. (Su Kon and Jin Wan Park, 2012) The present case report emphasizes the varied presentation and unique gross morphology of ovarian fibroma. It also reinforces the non specificity of CA-125 as a marker of ovarian malignancy. It beckons us to re evaluate the presumption that thorough clinical examinations supported by laboratory investigations and imaging modalities are fool proof in themselves. The role of a histopathological diagnosis should not be underestimated even in cases with a strong suspicion of malignancy.

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