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Ovarian fibroma: A diagnostic dilemma

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Abstract
Introduction: Ovarian fibroma is a benign stromal tumor composed of spindle fibroblasts like cells that produce collagen. It is seen in perimenopausal and postmenopausal women and is seldom seen in women with the age of less than 30 years.
Case description: A young lady presented with irregular menses and secondary amenorrhoea of 8 months with gradual distension of abdomen. Raised Ca-125 levels with CECT impression raised the suspicion of malignant right ovarian tumor. Staging laparotomy was done followed by right salpingo-ovariotomy. Histopathology with immunohistochemistry confirmed the diagnosis of ovarian fibroma.
Conclusion: There are no specific markers for an accurate pre-operative diagnosis of an ovarian fibroma. Improved understanding of this entity is essential to prevent misdiagnosis and missing the diagnosis.

Keywords: Ovarian fibroma, staging laparotomy, histopathology, immunohistochemistry

Introduction
Ovarian fibromas are very rare tumors which arise from the gonadal-stromal cell origin and grow from the connective tissue of the ovarian cortex. Such benign tumors account for 1-4% of all ovarian neoplasms and are generally seen in perimenopausal and postmenopausal women [1]. In most cases, these tumors are not hormonally active but few patients may present with endocrine manifestations such as metrorrhagia due to estrogenic or androgenic stimulation [2]. They are unilateral in majority of the cases but few patients may have bilateral lesions as seen in Gorlin syndrome. Gorlin-Goltz syndrome is a rarer entity also known as nevoid basal cell carcinoma syndrome occurring in younger age groups with recurrent multiple fibromas including ovarian fibromas. Ovarian fibroma in less than 1% of cases are associated with Meig's syndrome, a triad of ovarian fibroma, ascites and plural effusion. These tumors may be also form a part of Peutz- Jeghers syndrome which includes rectal, gastrointestinal polyposis and periorificial lentiginosis [3].

Case Description
Twenty-two-year-old patient reported to the out-patient clinic with complaints of irregular menses since 2 years followed by amenorrhoea since 8 months. On further questioning, she also felt a gradual increase in the size of her abdomen since almost a year.
She was married 3 years back but was separated since more than a year and had no history of sexual contact after separation. She was averagely built with no significant findings on general examination. On per abdomen examination, 24-26 weeks' size hard mass was felt with smooth surface, regular margins and solid consistency. It was mobile side-to-side but mobility was restricted in vertical direction. On per vaginal examination, uterus was 24-26 weeks' size with right forniceal fullness but no tenderness was elicited. Her Ca-125 level was 218.38 units/mL. Ultrasound was suggestive of 14.3 x 7 cms heterogeneous mass with poor acoustic window extending from periumbilical region to pelvic region with a probability of dermoid cyst. CECT demonstrated 18.3 x 15.4 cms solid enhancing mass in right abdomino-pelvic region including right adnexa with extension to midline suggesting possibility of right ovarian neoplasm. The clinical findings and reports along with the high index of suspicion for malignancy were explained to the patient and her mother who were willing to go ahead with the required plan of management. Decision for staging laparotomy was taken and patient was posted for the same. Intraoperatively, ascites was noted and fluid was sent for cytology. Right sided 18 x 20 cms solid adnexal mass was noted which was not separable from the right sided tube and ovary. Uterus and left sided tube and ovary were normal.

No deposition was felt on the surface of the diaphragm and liver. Right sided salpingo-ovariotomy with partial omentectomy was performed and specimen sent for histopathology. The post-operative period was uneventful and patient was discharged on day 10 of surgery after suture removal.

Cytology was suspicious of malignant cells. Histopathological analysis revealed multiple sections showing spindle shaped cells arranged in interlacing fascicles and whorls with mild nuclear atypia but scanty mitotic activity with no areas of necrosis. On Immunohistochemistry (IHC), the tumor cells were positive for Vimentin, SMA, WT1, CD56, CD10 and negative for Desmin, Inhibin, Calretinin and S100. ER and PgR were positive with moderate intensity staining and Ki-67 labelling index was approximately 4-5%. Histomorphology and Immunohistochemistry features favored the diagnosis of Ovarian Fibroma. This reduced the patient's anxiety as there is a negligible risk of recurrence with good prognosis.

Discussion

Ovarian fibromas are solid benign tumors classified under sex cord stromal tumors that comprise of granulosa cells, theca cells, sertoli cells, leydig cells and spindle shaped fibroblasts in various combinations along with abundant collagen [4]. This entity is commonly seen in women in their fifth decade of life during their perimenopause or postmenopausal period [5]. Rarely, they occur in younger age groups as seen in our patient at 22 years of age. Most of the patients are asymptomatic or complain of vague abdominal pain, distension, urinary symptoms or torsion [6]. Patients with torsion present with severe acute abdominal pain, seen in 8% of the cases [5]. Our patient presented with irregular cycles and eight months of amenorrhoea along with feeling of mass in abdomen which was gradually increasing in size.

Ovarian fibromas are difficult to diagnose pre-operatively and are usually diagnosed as other ovarian cysts, uterine leiomyoma and ovarian malignancy [7]. Serum Ca-125 levels and ultrasound of the whole abdomen are the initial set of investigations in patients presenting with above complaints. Normal level of serum Ca-125 is seen in majority of the cases but few patients may have elevated levels due to inflammation or necrosis caused by torsion [8]. The raised serum Ca-125 levels can raise the suspicion of malignant ovarian tumor as in our case.

On ultrasound, typical appearance is seen in almost half of the cases while others have a variable appearance. These fibromas are generally well defined solid with hypoechoic appearance in majority of the patients, homogenous in few while others have a posterior acoustic shadowing. Some patients may even have cystic components due to degeneration [2]. CECT helps in delineating the tumor from its origin, preservation of fat planes and describes fibroma as diffuse hypoattenuating mass with poor post contrast enhancement [4]. Ultrasound was suggestive of dermoid cyst while CECT diagnosed a probability of right ovarian neoplasm in our patient. Surgery is the mainstay of treatment which can be performed by laparotomy or laparoscopically. Salpingo-ovariotomy should be performed in perimenopausal or postmenopausal women while cystectomy should be considered in younger patients [9]. In our case, conservation of the ovary was not possible as no ovarian tissue could be separated from the large solid mass and there was high index of suspicion for malignancy. Diagnosis is confirmed only on histopathology.

Macroscopically, ovarian fibromas are firm, white masses with smooth lobulated appearance, sometimes cystic degeneration or stromal edema is also noted. This generally resembles a uterine leiomyoma [9]. Microscopically, intersecting bundles of spindle cells producing collagen are seen. The cellular variety has hypercellularity, mild to moderate nuclear atypia and increased mitotic activity, the degree of which differentiates it from fibrosarcoma. Minor sex cord elements are seen which are defined as sex cord elements occupying no more than 10% of the area of the tumor on any slide. These are seen as small nests of cells resembling cells of sex cord type, granulosa cells or sertoli cells [9]. These minor sex cord elements are positive for inhibin, calretinin, CD99, CD56, antikeratin antibody KL1 and MIC on immunohistochemical analysis of ovarian fibroma [9].

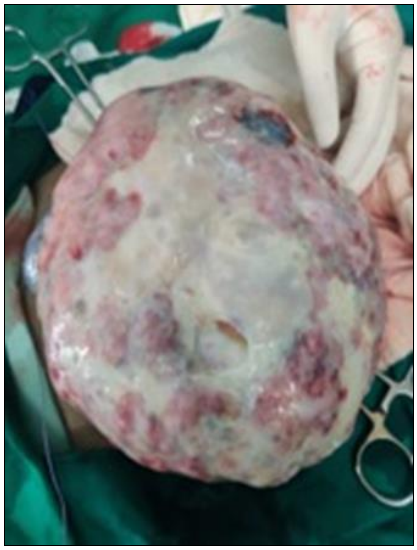

Fig 1: Intraoperative findings of a large mass

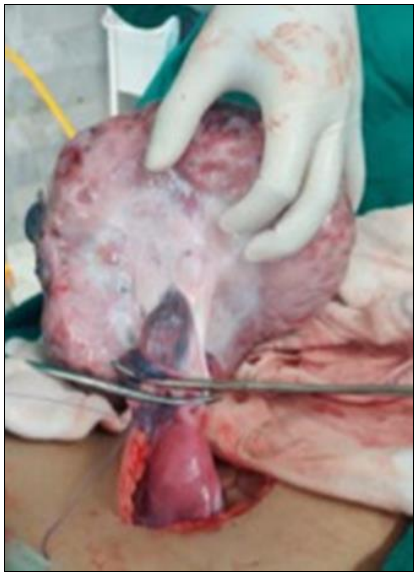

Fig 2: Excision of the large adnexal mass (salpingo-ovariotomy)

Conclusion

Ovarian fibroma though rare is the most common solid benign sex cord tumor of the ovary. It may have varied set of clinical presentations and also poses an imaging challenge in diagnosis. Most of the cases are misdiagnosed pre-operatively as they can mimic uterine leiomyoma or malignant ovarian tumors. Surgical excision followed by histopathological analysis is the preferred mode of treatment as these tumors generally have a good prognosis. This case thus highlights the diagnostic and operative dilemma which is solved only after histopathology and immunohistochemistry.

Conflict of Interest
Not available

Financial Support
Not available

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