CASE REPORT

Gynandroblastoma: A Rare Presentation

Arzoo Alagh[1], Samarth Shukla[2], Neema Acharya[3], Sunita Vagha[4]

Abstract

Gynandroblastoma is the sex cord-stromal tumor composed of clearly identifiable granulosa–theca cell and Sertoli–Leydig cell elements in variable amounts which comprise less than 1% of the sex cord-stromal tumors. Very few cases of gynandroblastoma are present in the literature to our knowledge. We discuss a case of gynandroblastoma of the ovary in a 62-year-old menopausal female. The microscopic features of the tumor showed both Sertoli cell and granulosa cell component. These tumors are associated with hormonal imbalance as the cells involved participate in the ovarian hormonal function. Thus, extensive sectioning of the tumor must be done by the surgical pathologist to rule out the possibility of other tumors and provide with a correct diagnosis of the tumor which can help the clinicians to plan an adequate management protocol for this rare tumor which will further elucidate into a properly regulated hormonal status and a better quality of life for the patients.

Keywords: Granulosa–theca cell, Gynandroblastoma, Sertoli–Leydig cell, Sex cord-stromal tumor.

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Background

In 1930, Meyer first coined the term "Gynandroblastoma" to describe a tumor having features of both granulosa cell tumor and androblastoma.[1] These are extremely rare tumors which are probably derived from undifferentiated mesenchyme. Gynandroblastoma is the term used for the sex cord-stromal tumors composed of a mixture in similar amounts of clearly identifiable granulosa–theca cell and Sertoli–Leydig cell elements.[2] These comprise <1% of sex cord-stromal tumors. Reported cases of this entity have been accompanied by androgenic, estrogenic, or no hormonal effects.[3] Very few cases of gynandroblastoma are present in the literature to our knowledge. We report a case of gynandroblastoma of the ovary in a menopausal woman.

[1,4]Department of Pathology, Jawaharlal Nehru Medical College, Wardha, Maharashtra, India

[2]Department of Pathology, DMIMS University, Wardha, Maharashtra, India

[3]Department of Obstetrics and Gynaecology, Jawaharlal Nehru Medical College, Wardha, Maharashtra, India

Corresponding Author: Arzoo Alagh, Department of Pathology, Jawaharlal Nehru Medical College, Wardha, Maharashtra, India, Phone: +91 7020011534, e-mail: drarzooalagh@gmail.com

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Case Description

A 62-year-old menopausal female presented in gynecological outpatient department of our hospital with complaints of pain and lump in abdomen since 8 months. Also, patient complains of constipation and increase in frequency of micturition since 6 months. No virilization symptoms such as hirsutism were seen at the time of presentation. The CA-125 levels were 75 U/mL. The CT scan was suggestive of multiloculated, heterogeneous, solid cystic mass with intensely enhancing walls and solid component filling the entire pelvis reaching up to the umbilicus with loss of fat planes most likely to be malignant, mucinous cystadenocarcinoma. Patient underwent hysterectomy with bilateral salpingo-oophorectomy amidst which a frozen section was done. The frozen section of the tumor showed microfollicular pattern and hallow tubules which were suggestive of gynandroblastoma.

The resected specimen was sent to histopathology section in department of pathology in 10% buffered formalin as uterus with cervix with bilateral adnexa along with posterior and anterior visceral peritoneum and infracolic omentum. Uterus was measuring 9 × 6.5 × 4 cm. Cervix was measuring 4 cm in length. Right ovary was measuring 2.5 × 0.5 × 0.5 cm, while the right sided fallopian tube was measuring 5.5 cm in length. Uterocervix along with both fallopian tubes and right-sided ovary were grossly unremarkable. Left side ovary along with irregular mass attached was measuring 11 × 9 × 6.5 cm. Cut surface of ovary showed yellowish blackish, solid cystic areas (Fig. 1). Sections were taken from the representative areas and were processed routinely. The slides were stained with hematoxylin and eosin stain.

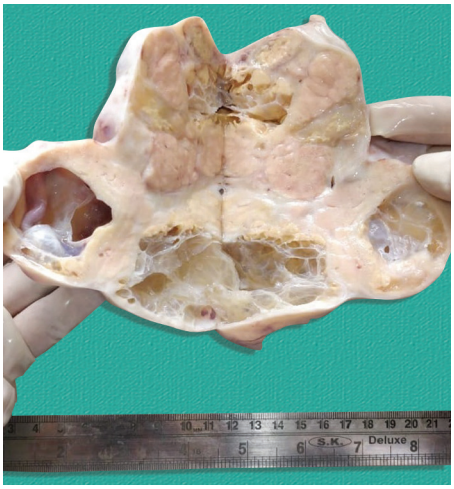

Fig. 1: Cut section of the ovarian mass showing solid and cystic areas

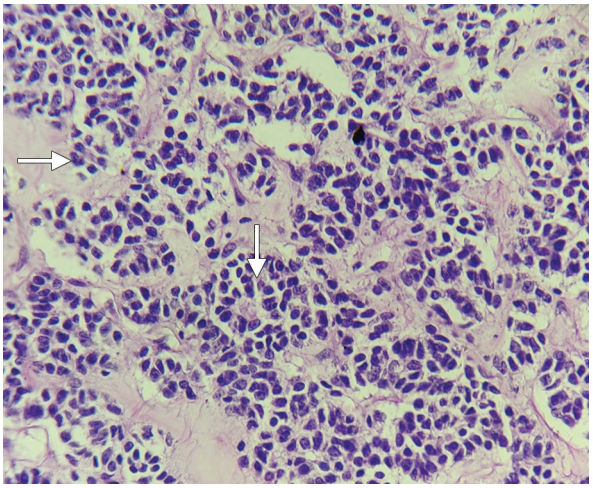

Fig. 2: Granulosa component with diffusely placed granulosa cells along with Call-Exner bodies (white arrow) (40×)

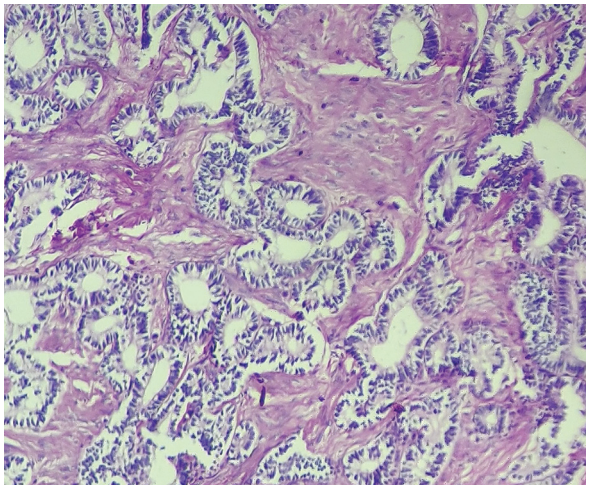

Fig. 3: Sertoli component showing hollow tubules (40×)

The microscopic feature depicted granulosa cells having diffuse growth of cells in a uniform arrangement along with angulated nuclei, scant cytoplasm. It presented mainly as microfollicular pattern, and occasionally at places, insular pattern was noticed. The distinctive microfollicular appearance of the tumor was due to the presence of many scattered Call-Exner bodies (Fig. 2). The areas of granulosa cells which comprised 70% were intermixed with cord-like structures and poorly differentiated tubules. In some areas, well-differentiated tubules were appreciated that were surrounded by abundant stroma. The stromal cells morphology varied from spindle fibroblast like cells to plump cells. The Sertoli cell component of the tumor was presented by hollow tubules that constituted columnar cells and a true lumen comprised 30% of the tumor (Fig. 3). Thus, the microfollicular pattern of granulosa cells along with presence of Sertoli cell component was suggestive of gynandroblastoma.

Also, the section from the omentum was free from invasion of metastasis by the tumor cells. The cytological study of the peritoneal fluid showed occasional polymorphs, lymphocytes, and degenerated macrophages. The smear was negative for malignant cells.

Discussion

Gynandroblastomas are rare ovarian tumors. These tumors contain variable but high proportions of granulosa cells and Sertoli–Leydig cells. In majority of cases, tumors are benign while few malignant tumors have also been reported.[3] These tumors are also associated with endometrial hyperplasia.[4] In our case, endometrial hyperplasia and adenomyosis were noted. Usually, it presents as a pelvic mass in second and sixth decades.[5] These tumors are associated with hormonal imbalance as the cells involved participate in the ovarian hormonal function. The symptoms such as amenorrhea or polymenorrhea, dysfunctional uterine bleeding, and/or hirsutism (due to Sertoli cell component) maybe present.[5]

Novak et al. has described eight gynandroblastoma cases from John Hopkins Hospital, Novak, while the WHO define gynandroblastoma as a tumor that requires more than 10% admixture of both testicular (Sertoli) and ovarian (granulosa cell) elements and that the minor component which is composed of at least 10% of the tumor.[4,6] In our case, the granulosa cell component depicted distinctive histological features, including diffuse, solid, and microfollicular patterns. In the Sertoli cell component, an hollow tubular arrangement was appreciated along with areas of poorly differentiated tubules. Microscopically, gynandroblastomas are reported to constitute the cells with similar features but with a different architectural pattern.

Neubecker and colleagues reported five cases of gynandroblastoma, whereas Gomes-Macias et al. have reported a case of gynandroblastoma in a 28-year-old female which they accidentally found during a C-section.[7,8] Also, Fukunaga et al. in 1996 reported an ovarian gynandroblastoma in a 60-year-old female and conducted immunohistochemistry and ultrasound study. In their study, Vimentin was observed in both Sertoli cell element and granulosa cells and was also positive for cytokeratin CAM 5.2. The tumor cells were reported negative for epithelial membrane antigen, CD31, CD34, and membrane-specific antigen in their study.[1]

Clinically, most patients of gynandroblastoma present with stage I disease; generally, the tumor is unilateral with no neighborhood or lymphatic invasion. Few cases of recurrence have been described in the literature.[4] This neoplasm is considered to be well differentiated and have low malignant potential. Surgery is an important therapeutic modality and can be effectively combined with chemotherapy.[3] In this case, the patient too presented with disease in stage I but did not manifest symptoms indicative of combined estrogenic and androgenic stimulation and had undergone surgical management.

Conclusion

Gynandroblastoma, being a rare tumor, becomes very challenging for pathologist to diagnose. Thus, extensive sectioning of the tumor must be done by the surgical pathologist to rule out the possibility of other tumors, as a differential diagnosis, which may change the management of the tumor. Therefore, a correct diagnosis of the tumor can help the clinicians to plan an adequate management protocol for this rare tumor with now known potential for being a malignant tumor. These management protocols will further elucidate into a properly regulated hormonal status and a better quality of life for the patients.

Orcid

Samarth Shukla https://orcid.org/0000-0002-3289-7006

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