J Cases Obstet Gynecol, 2016;3(3):100-103

Case Report

Choriocarcinoma in association with gonadoblastoma of the ovary with coexisting dysgerminoma of the contralateral ovary

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Abstract

Concomitant ovarian tumors are rare neoplasms that usually contain germ cell tumors of origin. Their prognosis worsens when it is associated with more malignant germ cell neoplasm. A 15-year-old virgin girl was consulted to our clinic with the diagnosis of abdominal mass. CA125, LDH and B-HCG levels are elevated. On ultrasonography bilateral pelvic mass was observed. Pathological examination revealed choriocarcinoma and gonadoblastoma in the left ovary and dysgerminoma in the right ovary. According to our investigation this is the first case that these three malignancies concomitantly present in one patient in literature. Coexistence of gonadoblastoma and a choriocarcinoma with the same gonad suggests that gonadoblastomas are genetically unstable and can give rise to other germ cell tumors.

Key Words:

Choriocarcinoma, gonadoblastoma, dysgerminoma, concomitant tumors

Introduction

Non-gestational choriocarcinoma of the ovary is an extremely rare and highly malignant tumor. They account approximately ≤0.6% of all ovarian neoplasms [1]. Histologically, it has exactly the same appearance as gestational choriocarcinoma which metastasize to the ovary [2]. The positivity of serum Human Chorionic Gonadotropin (hCG) can be used for monitoring of response in the treatment. Secretion of HCG can be noticed in pre-pubertal children with the evidence of isosexual precocious puberty as growth of pubic and axillary hair, menarche and development of mammary glands. On the other hand gonadoblastoma is also a rare germ cell tumor that consists of both germ cells and gonadal stromal cells [3]. They arise almost exclusively in patients with gonadal dysgenesis or abdominal male gonadal tissue. The association of gonadoblastoma with dysgerminoma is seen approximately in %50 of cases. Embryonal carcinomas, yolk sac tumors or choriocarcinomas can also arise from gonadoblastomas in up to 8% percent of cases. Herein we present a case of choriocarcinoma in association with gonadoblastoma in one ovary and dysgerminoma in the contralateral ovary which is the first case that these three malignancies concomitantly presented in one patient in literature according to our search in English database.

Article History:
Received: 06/04/2016
Accepted: 13/06/2016

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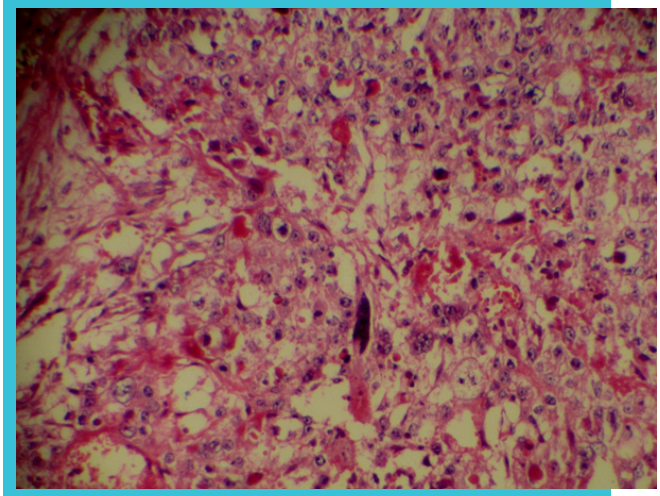
Case Presentation

A 15-year-old virgin girl was referred to Zekai Tahir Burak Women Health Education and Research Hospital Youth Center Outpatient Clinic with the diagnosis of pelvic mass. Her medical and family history were unremarkable. The physical examination revealed abdominal tenderness and mass arising from pelvis which extended to 2 cm below the umbilicus. She had Stage 4 breast and pubic hair development according to Tanner Scale. The laboratory findings were inconclusive except elevated tumor markers. The CA-125 level was 167,9 U/ml (<35.0 U/ml), lactate

dehydrogenase (LDH) was 1153 U/ml (<30.0 U/ml) and β-human chorionic gonadotropin (β hCG) was 10.902 U/ml (<5.0 U/ml) respectively. Abdominal ultrasound report showed a heterogeneous mass, 15 cm in diameter with solid and cystic component that covers entire pelvic region.

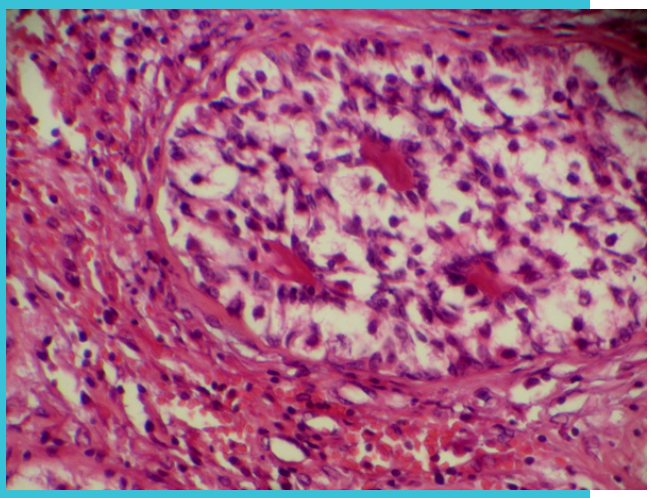
Figure 1.



Atypical trophoblastic cells with vesicular nucleusand large clear cytoplasm (10X, H.E.)

Uterus and ovaries could not be distinguished clearly. She was consulted to Oncology Department with the pre-diagnosis of ovarian malignancy. After written informed consent was obtained from the patient's parents, an exploratory laparotomy was undertaken for a suspected ovarian tumor. Intraoperatively, a solid, dark brown in color 12x15x13 cm mass originating from left ovary and 5 cm solid mass originating from right ovary was observed. Both masses were densely adhering to the colon, rectum, appendix, and posterior surface of the uterus. Moreover dense adhesions were observed between two separate adnexial mass. Frozen investigation revealed malign features intraoperatively. Due to the abdominal spread of the disease and inability to distinguish healthy ovarian tissue, total abdominal hysterectomy, bilateral salpingo-ooforectomy, omentectomy, appendectomy and pelvic paraaortic lymphadenectomy and peritoneal biopsies were performed instead of fertility sparing surgery. Final pathological examination revealed choriocarcinoma in association with gonadoblastoma in the left ovary (Figure 1, 2) and dysgerminoma in the right ovary (Figure 3). Moreover, malignant implantations were determined in omentum, sigmoid colon mesothelium. There were no signs of malignancy in 105 lymph nodes that were collected during surgery. Postoperative second day of surgery colonic material was determined in the drainage tubes. Patient underwent re-operation with the participation of gastroenterological surgery unit. Intraoperatively an aperture in sigmoid colon which is 2 cm in diameter was observed. Excision of the primary defected area in sigmoid colon and colostomy was performed during the operation. After 15 days of recovery period after second operation a combined chemotherapy was planned for the patient. The general condition of the patient as determined as unsatisfactory by medical oncologists and the chemotherapy was postponed. During follow-up general condition deteriorate and the patient was lost in postoperative 28th day.

Figure 2.

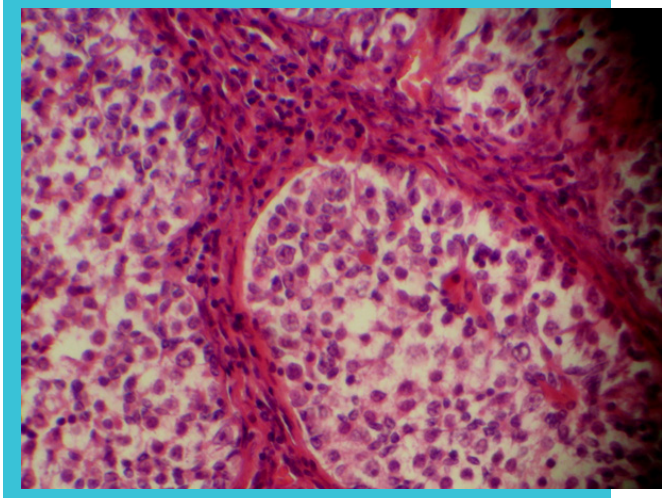


Germ cells and Sex cord stromal cells with Call-Exner Bodies (10X,H.E.)

Discussion

Gonadoblastomas are unusual mixed germ cell sex cord tumors occurring mostly in dysgenetic streak ovaries or abdominal testes that are generally incapable of female sex hormone production but may produce androgenic hormones [4]. They are composed of a mixture of supportive pre-sertoli/granulosa cells and germ cells at different stages of maturation [5]. Germ cell originated malignancies of the ovary especially dysgerminomas can be associated with gonadoblastomas [6]. Other germ cell elements such as yolk sac tumor, immature teratoma, choriocarcinoma and mixed germ cell neoplasms occur in less than 10 % cases [7]. The prognosis of gonadoblastoma and dysgerminoma association is generally excellent. Conversely, when it is associated with other germ cell tumors such as yolk sac tumor or choriocarcinoma, the prognosis may be unfavorable [8,9].

Figure 3.



Dysgerminoma cells and lymphocytes on fibrous band. (20X,H.E.)

On the other hand, non gestational ovarian choriocarcinomas account <0,6% of all ovarian neoplasms [1]. It is a highly aggressive tumor that Beta-Human Chorionic Gonadotropin (Beta-hCG) secretion is the key point in the diagnosis and follow-up. To date, no definitive and specific treatment modality has been established for pure ovarian choriocarcinomas due to the low incidence [10]. Because non-gestational choriocarcinoma is considered as a germ cell tumor differentiating to trophoblastic components, a germ cell tumor treatment protocol may be also effective [11]. In our case, we determined choriocarcinoma and gonadoblastoma in the right ovary and dysgerminoma in the left ovary of the patient. According to our literature search Zhao et al. presents a case that gonadoblastoma associated with a mixed germ cell tumor composed of five histological cell types: choriocarcinoma; embryonal carcinoma; yolk sac tumor; immature teratoma and dysgerminoma [12]. Furthermore Obata et al. presents a case of gonadoblastoma with dysgerminoma and yolk sac tumor association [13]. So far these two reports are the only cases in literature that coexistence of more than two different ovarian malignancies in one patient. The prognosis when gonadoblastoma is associated with more malignant germ cell neoplasms such as embryonal carcinoma, yolk sac tumor, choriocarcinoma is generally poor. None of these patients would have survived longer than 18 months before cisplatin was developed [14]. Although combined chemotherapy regimens and successful surgical interventions increase the survival of these patients, aggressiveness of the tumor components makes the surgery challenging and avoiding from surgical complications can be in evitable just as in our case. As a result, gonadoblastomas may have poor prognosis contrary to expectations when it is associated with other malignancies. Two or maybe more malignancy possibility should always be kept in mind before surgery. Also delay of chemotherapy due to surgical complications is the key point in those situations.

Acknowledgement
None

Declaration of Interest
None

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