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CASE REPORT

Growing Teratoma Syndrome : A Woman with Mixed Germ Cell Ovarian Tumor

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ABSTRACT

We reported a 24 year-old woman presented with pelvic mass who underwent right salpingo-oophorectomy. Histopathological examination revealed mixed endodermal sinus tumor and dysgerminoma. During chemotherapy with PEB regimen, although the serum alpha-feto protein level decreased, pelvic examination with confirmation of ultrasonography revealed further enlarged pelvic mass. Exploratory laparotomy and excision of tumor mass was performed and microscopic examination of specimen revealed mature teratoma. This patient was free of disease 3 months after diagnosis.

The growing teratoma syndrome is an infrequent entity of nonseminomatous germ cell tumors of ovaries. In this condition, masses that enlarge during or after chemotherapy are found to contain mature teratoma without malignant elements.

A 24 year – old, nulliparous, presented with pelvic mass gradually increased in size within three months. She was operated in October 1998 with right salpingo-oophorectomy and excision adherent omentum at a general hospital. The operative findings showed minimal peritoneal fluid, solid enlargement of right ovary about 15 cm. in diameter with ruptured tumor capsule, small tumor seeding on pelvic peritoneum and cul de sac. The histopathological slides were reviewed and presented with endodermal sinus tumor and dysgerminoma of right ovary with omental metastasis.

She was referred to Siriraj Hospital about one month after surgery with good performance (ECOG=0), normal general physical examination and normal profile of hematological, liver function and renal

function. After patient counselling, we started chemotherapy with PEB regimen (Cisplatinum 20 mg/m^2 D1-5; Etoposide 100 mg/m^2 D1-D5; Bleomycin 15 mg/m^2 D1, 5). The pretreatment tumor markers were alpha-fetoprotein 10,053 U/ml.; lactic dehydrogenase 465 U/ml.; and beta hCG 18.7 mIU/ml. During treatment, tumor markers decreased as shown in table 1 with some degree of hematologic toxicity. After four courses of chemotherapy, the pelvic examination revealed nodular mass about 3 cm. in diameter in cul de sac. Because of the declination of the tumor marker levels, we continued the 5th course of PEB regimen. In the next visit the cul-de-sac-mass had increased by 6 cm. in diameter, tense cystic to solid consistency, and fixed which corresponded with mixed echogenic mass behind the uterus depicfed by ultrasonography. After counselling the patient and her husband, reexploration to cytoreduce the growing tumor mass was performed. The operative findings showed a solid-cystic tumor packed in cul-de-sac, 7 cm. in diameter, smooth surface, cut-surface showed dark-brown and some grayish areas with stony hard consistency; normal uterus with left adexa; no ascites and normal intraabdominal organs. The tumor mass was totally removed after peritoneal washing including partial omentectomy.

The cytology was negative. The microscopic pathology revealed stratified squamous epithelium with keratinization and skin appendage and some areas of cartilage, fat cells and upper respiratory tract structure and unremarkable omentum. This pathology was compatible with mature teratoma. She was free of disease for 3 months after second surgery.

Table 1. Tumor markers profiles

Chemotherapy	AFP(U/ml.)	LDH(U/ml.)	Beta hCG (mIU/ml)
C1 PEB	10,053	465	18.7
C2 PEB	4,121	618	-
C3 PEB	87.8	382	-
C4 PEB	6.9	377	-
C5 PEB	10.0	498	-

Note : AFP, alpha – fetoprotein; LDH, lactic dehydrogenase

Discussion

The growing teratoma syndrome has been first described by Logothetis et al[1] with regard to gonadal and extragonadal germ cell neoplasms in males, but few cases have been reported in females. These tumors enlarged after chemotherapy and were first described to be refractory and chemotherapy was the treatment of choice rather than surgery. Tonkin et al[2] described the case of a 15-year-old girl who had a nondysgerminomatous germ cell tumor of the ovary that was completely resected. Within two months of beginning chemotherapy, an enlarged abdominal mass was found; on resection it contained only mature germ cell elements. Kattan et al[3] reported a 38-year-old woman who developed the growing teratoma syndrome after initial adnexectomy, composed of a mixture of mature teratoma, immature teratoma and endodermal sinus tumor. Several laparotomies were performed but only mature teratoma was identified. As reported by Maroto et al,[4] that the involved sites (e.g. retroperitoneal, lung, supraclavicular lymph node, inguinal lymph node) which previously affected by the disease were common recurrence and time to relapse ranged from 37-118 months with efforts to complete resection of the masses.

The patient in our report fits the description of

the growing teratoma syndrome. She had small residual tumor seeding in cul de sac before chemotherapy, and growth of mass occurred during chemotherapy despite decreasing AFP levels within three months. In three other cases reported by Geisler et al,[5] all their patients with immature teratoma had residual diseases before chemotherapy. Patients with nondysgerminomatous germ cell tumor of the ovary who have masses that increase in size during or after chemotherapy with elevated tumor markers must be evaluated for drug resistance. In the case of decreasing or normal tumor markers, a growing or persistent mass may be due to mature teratoma, fibrosis, or necrosis. Sugawara et al[6] suggested that a FDG PET scan with kinetic analysis could visually differentiate a viable tumor from mature teratoma, necrosis, or scar. However, after diagnosis of growing teratoma syndrome, these masses should be resected because they may cause obstruction, compression, or undergo sarcomatous degeneration.[7]

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