

Growing teratoma syndrome: A case report and review of the literature

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A rare case of growing teratoma syndrome (GTS) of the ovary is presented, with a review of the literature. A 36-year-old woman had 6 months' duration of back pain and abdominal fullness. She had undergone surgery elsewhere (right salpingo-oophorectomy) followed by chemotherapy for a histopathologically diagnosed immature teratoma of the right ovary 6 years previously. Imaging showed evidence of extensive peritoneal cystic metastases indenting the liver and in the pelvis and right suprarenal region. As tumour markers were within the normal range, a diagnosis of GTS was made. The patient underwent exploratory laparotomy with total hysterectomy and left salpingo-oophorectomy, and debulking of peritoneal deposits. Histopathological examination identified mature cystic teratoma. The available literature on this condition, with definitions, understanding of the pathophysiology and prognosis, is reviewed.

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Case report

A 36-year-old woman presented 6 years after oophorectomy for a right adnexal mass reported on histopathology as an immature teratoma. She underwent four cycles of chemotherapy (bleomycin, etoposide, cisplatin), and remained asymptomatic until 6 months before consulting us. In the intervening period (3 years previously), she had had a child, delivered by caesarean section with apparently normal intraoperative findings. She presented to us with backache and a sensation of abdominal fullness of 6 months' duration. She had mild hepatomegaly, but findings on abdominal, speculum and vaginal examination were otherwise normal. Magnetic resonance imaging of the abdomen showed well-defined multiloculated cystic lesions along the liver capsule indenting liver parenchyma. These were initially considered suggestive of peritoneal metastases. Peritoneal metastases were also noted in the pelvis and adjacent to the left kidney and right suprarenal region. As the tumour markers (CA-125, beta-human chorionic gonadotrophin and alpha-fetoprotein) were within the normal range, a working diagnosis of growing teratoma syndrome (GTS) of the ovary was made. She underwent exploratory laparotomy with total abdominal hysterectomy and left salpingo-oophorectomy, and debulking of tumour tissue from the pelvis, Morison's pouch, the diaphragm and the sub-hepatic region. Intraoperatively, deposits were noted on the left ovary and both uterosacral ligaments, and nodules on the undersurface of the diaphragm, in Morison's pouch, on the right side of the paracolic gutter, and between the liver and the kidney. There was no ascites, and the uterus and tubes were normal. She had blood loss of 3 L intraoperatively with subsequent disseminated intravascular coagulation that required correction and monitoring in the intensive care unit for 48 hours. She was discharged on the 8th postoperative day after an otherwise uneventful recovery. Histopathology was reported as mature cystic teratoma in specimens from the posterior surface of the uterus and nodules in the diaphragm, Morison's pouch and the infrahepatic region. There was an extensively hyalinised

nodule and no tumour in the mesothelium. Other findings were mild chronic cervicitis, secretory endometrium, myometrium with no specific lesion, and bilateral fallopian tubes with no specific lesion. The pathologists could find no immature teratomatous component in multiple sections examined. This was consistent with GTS.

Review of the literature

The diagnosis of GTS, first described in 1982, is based on three criteria:^[1] (i) an increase in tumour size or detection of metastases during or after chemotherapy for malignant germ cell tumour; (ii) normal tumour markers (which were high initially); and (iii) mature teratoma without evidence of malignancy on histopathology of the post-chemotherapy surgical specimen.

One of the initial suggested names for the diagnosis was 'chemotherapeutic retroconversion'; however, the pathogenesis remains uncertain.^[2] There is either selective elimination of the immature components of the teratoma or differentiation of malignant cells into mature teratoma cells. Clinically both these processes can mimic malignant metastases. In our patient radiographs suggested the possibility of malignant metastases, but clinically she was only minimally symptomatic for the extensive cystic peritoneal deposits. She had had initial surgery 6 years previously and had undergone a caesarean section 3 years previously.

At a multidisciplinary meeting, it was concluded that a diagnosis of GTS would need histopathological confirmation. A biopsy would serve no purpose, as both diagnosis and treatment depend on complete surgical removal and histopathological confirmation of the same. Although this involved fairly extensive surgery for this patient, with significant blood loss, it was hoped that the long-term outcome would be good.

There have been reports of carcinoids and sarcomatous conversion mimicking GTS;^[3-5] however, the pathologists were unable to find immature components in any of the specimens from our patient. Peritoneal gliomatosis has also been associated with GTS,

with some suggestion that initial association of immature teratoma with peritoneal gliomatosis may predict future development of GTS.^[1,2]

The outcome of GTS has uniformly been described as good; however, the key is early recognition so that resection can be as complete as possible, as chemotherapy is ineffective.^[6]

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