High-grade glioma arising in immature ovarian teratoma

Editor,

Neuroectodermal tumors arising in ovarian teratomas include astrocytoma, oligodendroglioma, ependymoma, and glioblastoma. Herein, we present a rare case of high-grade glioma arising in immature ovarian teratoma as the histological and immunophenotypic characteristics are similar to those arising from the central nervous system. To our knowledge, it is the fourth case reported in 6 decades. The patient underwent laparoscopic fertility-sparing ovarian neoplasms surgery combined with chemotherapy. No sign of recurrence was found within 6 months of follow-up after the treatment.

A 25-year-old female was admitted in our hospital because abdominal girdle growth was noted for 1 month without abdominal pain or fever. There was no change in the menstrual cycle, color, quantity, and texture. Color ultrasound revealed a giant cystic-solid mass located in the pelvis, measured about 16.5 × 14.8 × 12.0 cm. The lining of the cyst cavity was not smooth with multiple irregular hypoechoic and mixed echo protrusions. There were high-density echo plaques in the convex surface. Laboratory tests revealed serum tumor marker: α-fetoprotein (AFP) at 25.35 ng/mL (normal <8 ng/mL). The patient underwent giant pelvic mass resection and right ovarian cyst extirpation during the surgery.

Pathological manifestations

The cut surface consisted of solid and cystic areas; the wall thickness of the cystic region was 0.1–0.8 cm, containing a small amount of hair, solid tissue region, and gray-red soft cut surface. Microscopically, the cystic-solid mass was composed predominantly of differentiated squamous epithelium, cutaneous appendages, and fibrous fat; however, a small amount of immature cartilage was seen in the cystic region. Mature ganglion cells could be seen in the background of neuroglia and one part of the area differentiated into the ependyma around the glial [Figure 1a-e]. The solid area was predominantly composed of neuroglia derived from neuroectoderm. The density of glial cells was significantly increased in the neuroglia, the ratio of nucleus to cytoplasm increased, and pathologic mitosis can be seen [Figure 2]. Increased blood vessels in glial tissue and proliferated vascular endothelial were also found in the neuroglia [Figure 3a]. Immunohistochemistry showed that the malignant cells are negative for GFAP and IDH-1 and positive for Oligo-2 and P53, and Ki-67 labeling index was about 10% [Figure 3b-f]. Hematoxylin and eosin (HE) morphology and immunohistochemical characteristics were consistent with high-grade gliomas in the central nervous system. In the background, a component of the immature neural was seen in a region less than one low-power field [Figure 1f]. Immunohistochemistry showed that the malignant cells were positive for epithelial membrane antigen (EMA) in cavity edge, were negative for GFAP, s-100, and OCT4, and Ki-67 labeling index was less than 5% [Figure 4a-f].

Based on the above findings, the patient underwent magnetic resonance imaging (MRI) plain scan, and an enhanced scan of brain parenchyma was normal.

The incidence of ovarian immature teratoma only accounts for 1% of ovarian teratomas.[1] It usually occurs on one side of the ovary and in young women under 30 years of age. Laboratory tests often reveal slightly elevated serum levels of AFP. In this tumor, somatic cells may occasionally undergo malignant transformation. Among those transformations, squamous cell carcinoma is most common, while high-grade gliomas are extremely rare.[2] Only three cases of high-grade glioma arising in immature ovarian teratoma have been reported so far.[3-5]

Immature teratomas differ from common mature teratomas in that they contain immature nerve tissue. Although reactive vascular hyperplasia can be seen around the primary neural tube, even accompanied by distinct cell-rich nuclear division activity, they are still intermixed with different maturity of ectodermal or endodermal components at intervals. This can be used in the identification of high-grade gliomas in immature teratomas. In addition, the former Ki-67 index is generally less than 4%.
The prognosis of high-grade glioma arising in mature teratoma is better than that arising in immature teratoma.\textsuperscript{[4,6,7]} Standard treatment for ovary immature teratoma accompanied with high-grade glioma is still not available. The serum level of tumor marker AFP is useful in monitoring the progression of the disease, but the effect is not that so reliable as previously reported.\textsuperscript{[4]} Even so, the serum level of AFP of this patient decreased from 25.35 ng/mL to the normal range after one course of treatment with PEB regimen (etoposide + cisplatin + bleomycin). Up to now, no abnormality was found in 4 months after surgery and combined chemotherapy; follow-up is ongoing.

In conclusion, we presented a rare high-grade glioma arising in OIT. HE morphological support as well as immunohistochemistry confirmed that this tumor was consistent with high-grade gliomas occurring in the central nervous system.

**Declaration of patient consent**
The authors would like to certify that they have obtained the patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that name and initial will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**
There are no conflicts of interest.

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