Gliomatosis peritonei with a mature cystic teratoma: a rarity

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Abstract

Gliomatosis peritonei (GP), a rare condition related to ovarian teratomas, is characterized by miliary implants of mature glial tissues on the peritoneum or omentum. Although grossly mimicking intra-abdominal tuberculosis or carcinomatosis, a mature gliomatosis peritonei is a benign condition and therein lies the importance of recognition of this condition. Conservative surgery without adjuvant therapy is sufficient for this condition. We present here a very rare case of a unilateral mature ovarian teratoma with gliomatosis peritonei where fertility sparing surgery was done.

Keywords: Gliomatosis peritonei, Teratoma, Mature glial cells

1. Introduction

Gliomatosis peritonei (GP) (coined by Neuhauser in 1906) is defined as miliary implantation of glial tissues on the surface of the visceral or parietal peritoneum, omentum and bowel wall with secondary maturation into glial nodules of 1 to 10 mm. This condition is usually seen in patients with immature ovarian teratomas. Until now, reports of only 90 cases of gliomatosis peritonei have been reported which are mostly associated with immature teratomas. Gliomatosis peritonei constitutes a harmless situation with good prognosis even when associated with an immature teratoma of the ovary [1].

Injury to the ovarian teratoma capsule may have a role in peritoneal implantation [2]. When the teratoma is associated with GP, the prognosis is usually better, irrespective of original tumor grade [2].

There are no established protocols for monitoring these patients. Computed tomography (CT), magnetic resonance imaging (MRI) and tumor markers have all been proposed [3, 4].

However, malignant transformation of GP has been reported [5]. Gliomatosis Peritonei can be identified during first surgery or a second look laparotomy [6].

2. Case history

A 24 years old P1L1 was admitted in our gynecology Outpatient Department with complain of pain abdomen since 1 year and mass abdomen since 3 months which was consistently growing in size. She also complained of nausea and loss of appetite.

Her menstrual cycles were regular and she had delivered a term female baby by normal vaginal delivery 16 months back. She had breastfed the baby for 1 year.

Her general examination was normal. Per abdomen, there was a 32 weeks size pelvic lump with solid and cystic component, nontender, mobile, with regular margin and variegated surface. Per vaginum examination revealed, normal sized retroverted uterus. There was a distinct groove between the uterus and the ovarian tumor which was felt through the anterior and right lateral fornix predominantly.

Ultrasonography (USG) examination was done which revealed that the tumor appeared to be ovarian teratoma of size 18 × 20 cm with solid and cystic component and calcified areas. There was ascitis and right sided hydronephrosis. Cancer antigen (CA-125) was done which was 58 U/ml (normal less than 35 U/ml).

The patient was put up for laparotomy. Intraoperative findings were as follows:

1. Right sided ovarian tumor with irregular surface about 18 × 20 cm.
2. Moderate degree of ascitic fluid.
3. Five millimeter size greyish white nodules densely scattered equidistantly on whole peritoneal surface and omentum (Fig. 1).

Fig. 1. Glial nodules on omentum.
The first impression was that of intraperitoneal carcinomatosis. Ascitic fluid was sent for histopathological examination. Liver, undersurface of diaphragm and both kidneys were palpated. Since the patient had only one child, and was only 24 years old, a fertility sparing surgery with right sided salpingoopherectomy was done. Partial omentectomy was done. Multiple peritoneal punch biopsies were taken including the peritoneal nodules.

Cut section of the tumor showed a tuft of hair, a tooth, sebum, a piece of bone and was sent for histopathological examination (Fig. 2). The cytoanalysis of the ascitic fluid was unremarkable. The histopathology report of the ovarian tumor confirmed it to be a mature teratoma and the peritoneal nodules were mature glial elements confirming the diagnosis to be gliomatosis peritonei with mature teratoma (Fig. 3).

No adjuvant therapy was given but the patient was asked to come for regular follow-up.

3. Discussion

There are three possible sources of GP as follows:

1. Deposition of immature neural tissue with consequent maturation,
2. Lymphogenous metastasis,
3. Mature glial cells extruded through a defect in the capsule of the primary tumor [7, 8].

The occurrence of GP and its accompanying ovarian teratoma is in the early decades of life. It has been found to occur almost exclusively in females with ovarian teratomas. The size of the ovarian tumor varies from 12 to 33 cm in diameter and the size of the nodules of glial implants are 1 to 10 mm in size. Macroscopically, they are indistinguishable from intra-abdominal tuberculosis and carcinomatosis. Microscopically, GPS may consist of mature or immature glial tissue, the former implies a favorable prognosis.

Immunohistochemistry of the glial tissue is positive for vimentin and neural markers, such as GFAP, S100 and NSE. Negative MIBI indicates the nonproliferative nature of the tissue and negative AFP rules out metastasis of an immature germinal cell tumor [1, 10].

The management of GP is directed by the nature of the glial implants, which should be extensively sampled and thoroughly examined. For a mature implant, conservative surgery is indicated. There is a high incidence of adhesion sequelae in GP.

Eighty-five percent of the cases of ovarian teratomas initially present with abdominal pain and mass abdomen. Imaging studies and tumor markers guide the initial diagnosis as well as monitoring of the disease [9].

Surgery is the basic treatment for both mature and immature teratomas as well as for peritoneal gliomatosis, which is carried out to confirm the diagnosis in order to exclude malignancy and to prevent the malignant transformation of the peritoneal implants.

A favorable prognosis is determined by the following:

1. Histological nature of glial tissue implants that are completely mature regardless of the nature of the ovarian teratoma.
2. Loss of proliferative activity of the peritoneal implants. Therefore, in addition to the primary tumor grade, peritoneal gliomatosis grade is also important.

4. Conclusion

The low frequency of this condition indicates that there is no consensus on the monitoring of peritoneal gliomatosis. Further studies should be carried out to determine the protocols for the management and follow-up of this disease. This case report is directed toward making the gynecologists and surgeons aware about this seemingly harmless condition and to refrain from doing aggressive surgery especially in young patients desirous of further child bearing as in our case.

5. Abbreviations

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<tr>
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<tr>
<td>GP</td>
<td>Gliomatosis peritonei</td>
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<td>CA-125</td>
<td>Cancer antigen 125</td>
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<td>GFAP</td>
<td>Glial fibrillary acidic protein</td>
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<td>NSE</td>
<td>Neuron specific enolase</td>
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<td>MIBI</td>
<td>Methoxy iso butyl isonitrite</td>
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<td>AFP</td>
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Fig. 2. Cut section of ovarian teratoma showing tuft of hair, tooth and bone.

Fig. 3. HPE photograph showing mature teratoma with mature glial elements.
Conflict of interest

The author has no conflicts of interest.

References