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Case Report

Gliomatosis peritonei associated with mature cystic teratoma – A case report

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ABSTRACT

The case report aims to highlight a rare entity called gliomatosis peritonei. We report a case of a 23-year-old female presenting with an ovarian mass along with multiple omental nodules. On histopathological examination, the ovarian mass was diagnosed as mature cystic teratoma and the omental nodules showed features of gliomatosis peritonei. Gliomatosis peritonei is a rare entity characterized by peritoneal or omental implants of mature glial tissue. GFAP (Glial Fibrillary Acidic Protein) was positive in the omental nodules. Gliomatosis peritonei is an exceptional finding and requires long term follow up of the patient.

1. Introduction

Gliomatosis peritonei (GP) is a rare entity characterized by peritoneal and omental implants of mature glial tissue which is associated with Immature Teratomas, less commonly Mature Teratomas and rarely, Ventriculo-Peritoneal shunts. Approximately 100 cases have been reported in literature.[1] It is considered as Grade 0 by WHO, has a favorable prognosis and the treatment of the patient depends only on the stage of the associated Immature Teratoma.

2. Case Report

We present a case of a 23 year old female with complaints of lump in abdomen since 3 months. On ultrasonography of abdomen, a large multiloculated solid cystic mass was seen in midline of pelvis arising from the right adnexa suggestive of right ovarian neoplasm. Patient also had moderate ascites. The specimen of Ovarian mass along with biopsy from the omental nodules was received for histopathological examination. The Ovarian mass was 14 x 18 x 10 cm in size. Externally capsule was disrupted by a nodule measuring 1.5 x 1.5 cm. On cut surface, tumor showed a variegated, solid-cystic appearance and hair follicles, bony gritty tissue, calcification, mucinous material, yellowish areas etc. were identified. (Figure 1) The ovarian mass was extensively sectioned and showed mature glial tissue, nerve bundles, ganglion cells, mature adipose tissue, skin, salivary gland, glandular epithelium, cartilage, lymphoid aggregates and skeletal muscle. (Figure 2) No evidence of immature or malignant elements was seen. The diagnosis on histopathology was Mature Cystic Teratoma. The omental nodule biopsy showed mature adipose tissue studded with multiple nodules composed of mature glial tissue with fibrillary matrix and benign glial cells. A small focus of keratin scales was also seen. (Figure 3) Immunohistochemistry was performed and GFAP showed strong diffuse cytoplasmic positivity within the gliomatous nodules, thus confirming the diagnosis of Gliomatosis Peritonei. (Figure 3)

3. Discussion

GP is rare entity associated with ovarian teratomas with only approximately 100 cases reported in literature.[1] It is associated with immature teratomas, less commonly mature

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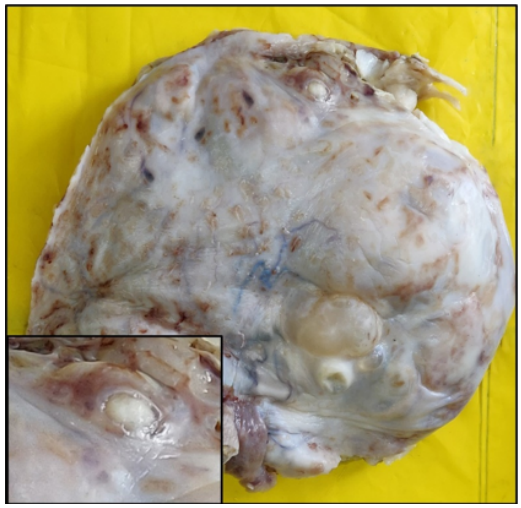

Fig. 1: External surface of solid-cystic ovarian mass is bosselated and shows a nodular area of capsular disruption.

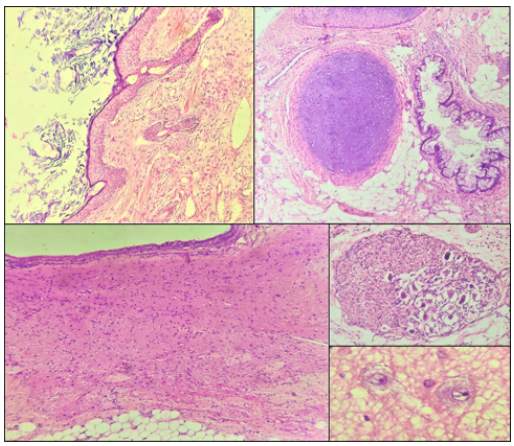

Fig. 2: H & E stained sections from ovarian mass show mature cystic teratoma composed of mature glial tissue, ganglion cells, skin with adnexal structures, glandular epithelium, cartilage etc.

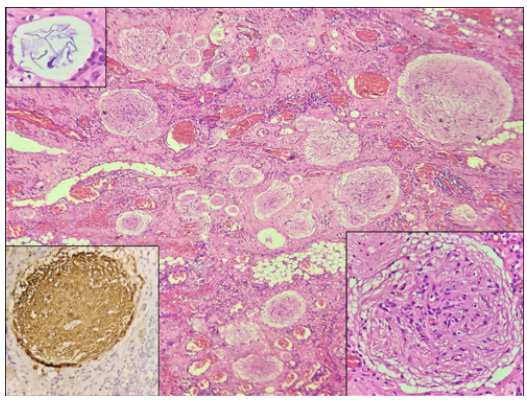

Fig. 3: H &E stained sections from omental nodule biopsy show mature adipose tissue studded with multiple nodules of mature glial tissue which was positive for GFAP. A small focus of keratin scales was also seen.

teratomas and rarely, ventriculo-peritoneal shunts.[1] In our case, the patient had GP associated with mature cystic teratoma. Patients are generally young women in the first two decades of life with abdominal distension or lump in abdomen,[2] similar to our case. The diagnosis of the ovarian neoplasm can be made by a combination of clinical findings and radiological studies. However, smaller omental nodules may be overlooked causing a delay in accurate diagnosis due to the rarity of this disease.[1] In some cases, GP can show extensive involvement of omentum and peritoneal surfaces resulting in clinical misdiagnosis as peritoneal tuberculosis or peritoneal carcinomatosis.[3] Histopathology is a gold standard investigation in such cases for reaching accurate diagnosis.

The exact mechanism of implantation in GP is not known and there are different hypothesis. The first hypothesis suggests that GP is genetically identical to the associated teratoma and occurs due to peritoneal seeding of the mature glial tissue via capsular rupture.[4,5] The presence of other mature tissues like shed keratin scales, hair etc. in GP support this hypothesis,[5] as seen in our case. In some cases of GP, mature glial tissue has been found within lymph nodes suggesting an angiolymphatic mode of spread.[4] The second hypothesis states that the ovarian teratoma is genetically different from GP. Some studies show that the teratoma shows homozygosity at certain microsatellite loci, whereas the gliomatous nodules show heterozygosity at the same loci.[4,6] It has also been suggested that growth factors secreted by the teratoma cause the peritoneal stem cells or peritoneal mesenchymal cells to undergo differentiation into mature glial tissue.[6] GP is classified as Grade 0 by WHO. It has a favourable prognosis. GP may remain stable for long periods of time[1] or it may undergo fibroblastic transformation and disappear gradually.[7] Patients with GP have higher chances of recurrence,[8] and very rarely, GP may also turn malignant. Liang L et al reported a case of immature ovarian teratoma which showed glioma arising from gliomatosis peritonei after 6 months.[9] Dadmanesh F reported a case of with GP who developed glioblastoma multiforme 7 $\frac{1}{2}$ years later.[10] Hence, long term follow-up and close monitoring is required in GP. In our study, as GP was associated with mature cystic teratoma, no further treatment was needed post-operatively and 3 months later, the patient is fine and has no recurrence.

4. Conclusion

In patients with ovarian teratoma with omental involvement, a possible diagnosis of GP should be considered. As the radiological diagnosis of GP is challenging due to the rareness of the disease, histopathology should be performed for accurate diagnosis in all such cases. The follow-up of patients with GP should be long term and should not be discontinued even if symptoms are alleviated.

5. Conflict of Interest

None.

6. Source of Funding

None.

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