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Case report

18F FDG positron-emission tomography findings of gliomatosis peritonei: A case report and review of the literature

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1. Case

A 31 year-old nulliparous woman presented with abdominal discomfort. Ultrasound and computed tomography (CT) revealed a right adnexal mass. She underwent diagnostic and therapeutic laparotomy with right salpingo-oophorectomy, omental biopsy and pelvic washings. Intra-operatively, she was noted to have a moderate amount of intra-abdominal serous fluid. At the completion of surgery, examination of the pelvis and abdominal organs did not identify visible residual disease. Pathology revealed an intact 1.6 kg, 18 cm high-grade immature teratoma with neural/neuroepithelial differentiation (Fig. 1A), with areas of necrosis on the serosal surface. Washings were positive for immature teratoma with neuroepithelial elements; there were no omental implants but surface exudate contained immature teratoma (again, with neuroepithelial components). The post-operative recovery was unremarkable. Post-operative radiologic staging by CT revealed no evidence of residual intra- or extra-abdominal disease; tumour markers (LDH, HCG, and CEA) were normal except for AFP 73 mcg/L, CA19-9 73 kU/L, and CA-125 380 kU/L. The patient's final FIGO stage was 1C3 and she went on to receive 3 cycles of BEP chemotherapy (30 units of bleomycin on days 2, 9 and 16, etoposide 100 mg/sq. m/day on days 1–5 and cisplatin 20 mg/sq. m/day on days 1–5 of 21-day cycles); treatment was well-tolerated with no major side-effects.

After completion of chemotherapy, all tumour markers normalized. A post treatment CT of the abdomen and pelvis showed multiple new soft tissue nodules over the right diaphragmatic dome, suspicious pericardial and cardiophrenic adenopathy. To investigate the possible etiologies of the new masses (e.g. growing teratoma syndrome versus active malignant germ cell elements), a positron-emission tomography (PET)/CT scan was ordered. This revealed intense fluorodeoxyglucose (FDG) accumulation in periphrenic and aorto-caval lymph nodes and in soft-tissue deposits over the dome of the liver and in the right para-rectal area (SUV max of 22.1) (Fig. 2A). The AFP remained normal at the time of the PET/CT scan.

A diagnostic laparoscopy was undertaken, and revealed multiple peritoneal implants including a 2 × 3 cm right pararectal mass; biopsies were taken from multiple lesions. All samples revealed mature well-differentiated glial tissue (Fig. 1B) immunohistochemically positive for NeuN, synaptophysin and GFAP (Fig. 1C), with some atypical cells but no necrosis or increased mitotic activity, consistent with a diagnosis of gliomatosis peritonei (GP).

Follow-up CT four months after completion of treatment showed no evidence of progression. Repeat PET nine months after treatment completion revealed partial metabolic response compared with the initial post-treatment PET. At the time of her last follow-up (12 months after completion of all treatment), the patient remains well and continues to have normal tumour markers.

2. Discussion

To our knowledge, we present the first case report of FDG-PET/CT imaging of gliomatosis peritonei (GP), and demonstrate that this entity can be strongly FDG-avid. GP is a rare histological entity. It has been described in relationship to immature teratomas and growing teratoma syndrome

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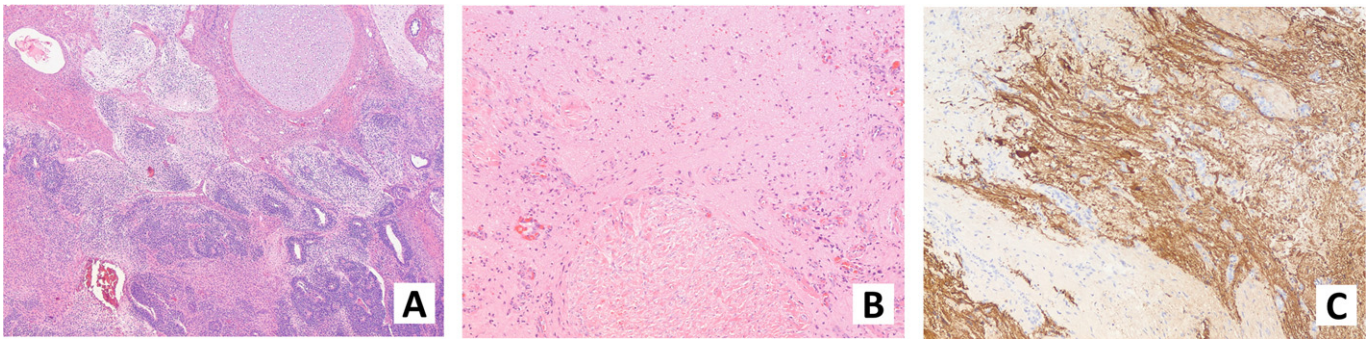

Fig. 1. Representative histology from the initially resected immature teratoma (A) and GP (B) and immunohistochemistry showing GFAP positivity confirming the diagnosis of GP (C).

(GTS) (Merard et al., 2015). GTS is defined as the presence of growing mature teratoma during or after completion of chemotherapy for a malignant germ cell tumour. In GP, the mature teratoma is exclusively composed of well-differentiated glial tissue (Li et al., 2016; Nogales et al., 2012). It has generally been described as an intra-abdominal phenomenon, although there have been reports of intrathoracic (Wu and Lai, 2015; Webman et al., 2015; Lipskar et al., 2009) and nodal (Wang et al., 2016) GP. Some cases of GP have shown spontaneous regression after resection of the

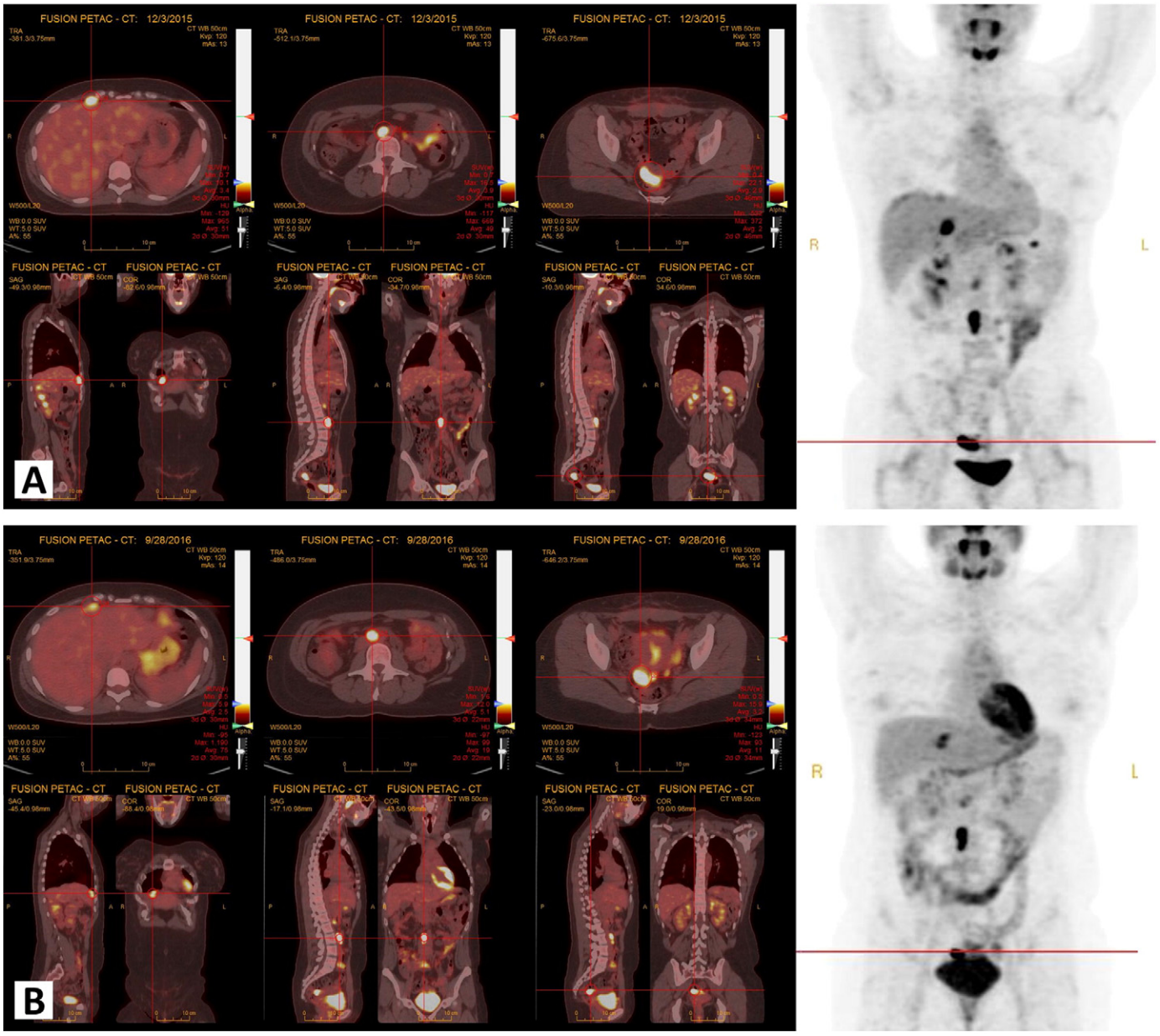

Fig. 2. FDG-PET/CT images at the end of treatment (A) and at 9 month follow-up (B).

primary tumour (Webman et al., 2015). In three case series the presence of GP was not associated with decreased survival (Wang et al., 2016; Liang et al., 2015; Bentivegna et al., 2015). In one case series of 16 patients, the presence of GP in association with malignant ovarian germ cell tumours decreased recurrence-free, but not overall survival (Yoon et al., 2012).

In our case, post-treatment functional imaging was requested to differentiate between resistant/progressive malignant GCT and mature teratoma (Yokoyama et al., 2015; Balink et al., 2012). Persistent mature teratoma should present with low FDG uptake (Kikawa et al., 2011), while immature teratoma and active malignant germ cell carcinoma usually show high uptake. However, the presence of glial tissue, with its baseline reliance on glucose metabolism, could logically also be FDG avid, as this case illustrates. GP should be added to the differential diagnosis of FDG-avid residual mass(es) following germ cell therapy, especially if the initial diagnosis was of immature teratoma containing neural/neuroepithelial differentiation. Supportive of the benign nature of the masses were the persistently normal tumour markers. However, any FDG avid mass remaining after treatment for germ cell tumour should be further evaluated either by complete resection or biopsy for pathologic assessment. Malignant germ cell tumours growing during or immediately after completion of platinum-based chemotherapy have a poor prognosis and accurate diagnosis and treatment is paramount (Broun et al., 1992). Observation of remaining GP lesions may be reasonable, as stability and sometimes regression are to be expected. However, if residual lesions are resectable then the safest option is removal as until they have been histopathologically examined, it is not possible to be sure all of them will also be GP. Furthermore, masses that are non FDG-avid on PET/CT may be compatible with either necrotic tumour or persistent mature teratoma since PET/CT is unable to reliably discriminate these two entities. These lesions must be followed closely and resected if growing in order to prevent GTS or de-differentiation to/development of active cancer. Generally, a multidisciplinary approach that includes surgical and medical experts is recommended.

In this case, the uniform gradual decrease in FDG uptake with follow up imaging, the stability of the size of all these lesions, as well as the persistently normal tumour markers, are reassuring. The evolution of FDG-PET findings for GP is not known, but given that GP is known to be stable and occasionally to spontaneously regress, a slow reduction in FDG uptake over time could be expected. While GTS commonly shows low-grade FDG uptake, it typically increases in size over time and residual immature teratoma/malignant germ cell carcinomas should, in many cases, produce germ cell serum markers along with rapidly progressing FDG-avid lesions. As FDG PET/CT is now widely available and becomes an important diagnostic modality used for assessing residual masses after treatment for germ cell carcinomas, the current case report suggests it could be useful for the follow-up of GP.

Disclosure

The authors have declared no conflicts of interest.

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