Dear Editor,

The natural history of Granulosa cell tumor (GCT) is characterized by long disease free interval with multiple episodes of recurrences. Hepatic metastasis occurs in 4% of cases and the treatment in this scenario is usually palliative. We report a case of a stage IIIb GCT undergoing HR twice with good outcome and hope this encourages gynecologic oncologists to consider HR in selected cases.

A 35-year-old lady underwent total abdominal hysterectomy on 11/11/1997 for a stage IIIb (omental metastasis) grade 3 GCT. She refused adjuvant chemotherapy and was lost to follow-up.

She was evaluated for abdominal discomfort and detected to have ascites with pelvic and hepatic recurrence in 04/02/2003 and was managed with four cycles of bleomycin, etoposide and cisplatin, followed by debulking of pelvic disease with removal of remnant omentum and pelvic node sampling. She had residual disease in omentum and received a postoperative abdominopelvic radiation.

She was referred to our institute for pain in the right hypochondrium with nausea, loss of weight and appetite and computed tomography (CT) abdomen showing a large hypodense lesion in segment VII of the liver. Tumor markers alfa fetoprotein, carcinoembryonic antigen and CA 125 were normal. Biopsy was consistent with metastatic GCT. In view of good performance status, long disease free interval and solitary nature of the lesion a decision for surgical resection of the lesion was taken. Intraoperatively, there was a mass of 5 cm × 5 cm arising from the diaphragm and partially infiltrating segment VII of the liver while the rest of the abdomen was free of disease. She underwent excision of diaphragmatic deposit with nonanatomical liver resection on 27/04/2007. During her postoperative stay, she developed right-sided pneumothorax which was managed conservatively. No adjuvant treatment was offered, this being a solitary lesion which was completely excised.

She developed a second hepatic recurrence in 21/01/2010 with CT abdomen showing a 27 mm × 22 mm lesion in segment VIII of liver. In view of the previous hepatic resection, it was decided in the multidisciplinary tumor board to offer a right hepatectomy. Again, no adjuvant treatment was given.

Long-term survival following repeat liver resections in metastatic ovarian granulosa cell tumor: Case report with review of the literature

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Letters to the Editor

Table 1: Patients with metastatic GCT who underwent liver resection

| Authors          | Timea (months) | Timest (months) | Symptoms          | Surgery                                                                 | PHRS (months) | OS (months) | Present status  |
|------------------|----------------|-----------------|-------------------|---------------------------------------------------------------------------|---------------|-------------|----------------|
| Crew et al[3]    | 144            | 120             | Dyspnoea          | Partial HR, wedge excision right lower lobe lung                          | 5             | 276         | Alive with disease |
| Madhuri et al[3] | 204            | 32              | Dyspnoea          | Partial HR, omentectomy, excision of abdominal disease                    | 40            | 276         | NED            |
| Madhuri et al[3] | 108            | 12              | Abdominal pain   | Partial HR, stripping right subdiaphragm peritoneum, cholecystectomy, right hemihepatectomy, anterior resection | 34            | 156         | NED            |
| Madhuri et al[3] | 72             | 0               | Abdominal pain   | Partial HR, stripping bilateral subdiaphragm peritoneum, omentectomy, excision pelvic disease | 24            | 96          | NED            |
| Chua et al[4]    | 144            | 0               | NA                | Partial HR, right lower lobectomy of the lung, stripping right subdiaphragm peritoneum, right adrenalectomy | 95            | 239         | NED            |
| Chua et al[4]    | 72             | 172             | NA                | Right hepatectomy, stripping right subdiaphragm, total anterior parietal and pelvic peritoneum, stripping of the omental bursa, cholecystectomy, small bowel resection, anterior resection, right adrenalectomy | 10            | 254         | Alive with disease |
| Rodriguez Garcia et al[5] | 72     | 60              | Abdominal mass   | Right hepatectomy                                                        | 9             | 141         | Died of other causes |
| Present study    | 63             | 50              | Abdominal pain   | Asymptomatic                                                              | 86            | 200         | NED            |

GCT=Granulosa cell tumor, NA=Not available, PHRS=Posthepatic resection survival, OS=Overall survival, HR=Hepatic resection, NED=No evidence of disease. *Time from primary diagnosis to hepatic recurrence, †Time from hepatic recurrence to hepatic resection

In 10/01/2012, CT abdomen showed a 2.4 cm × 1.8 cm lesion in left pelvic wall. Biopsy was consistent with recurrent GCT. Initially, patient was not willing for surgery or intravenous chemotherapy and was managed with oral etoposide and later with letrozole. But due to progressive disease, excision of the lesion was done on 04/09/13. She is clinically and radiologically free of disease till date (31/07/2014).

This is probably the first reported case in the English literature of a patient with metastatic GCT undergoing liver resection twice in a span of 3 years and still doing well 4½ years later.

Our decision for surgical management of the recurrences has been based on the available literature which has shown the benefits of debulking surgeries both in the primary and recurrent scenario for GCT.[1]

Table 1 lists the patients with metastatic GCT, who underwent liver resection.

Unlike epithelial ovarian tumors, GCT are indolent tumors and slowly grow to occupy a large volume of the liver causing compression of neighboring structures. Liver resection was successfully done in two cases presenting with dyspnea.[2,3] Both cases had good quality of life (QoL) post HR.

The solitary nature of the liver metastasis strengthens the argument for HR. However, the presence of extrahepatic disease need not be a contraindication for surgery if complete debulking can be achieved as shown by Madhuri et al. and Chua et al.[3,4]

In view of the hypervascularity noted in these tumors,[5] there exists a grave danger of tumor rupture causing intraabdominal bleed and even death. Hence, it can be justified to perform HR even if asymptomatic which prompted us to offer surgery the second time without any delay.

We hope the above justifications for HR in metastatic GCT will encourage more gynecologic oncologist to consider HR and work in tandem with hepatobiliary surgeons to achieve optimal cytoreduction. The selection criteria for justifying hepatic metastectomy should be formulated by a consensus from the available literature. Repeated liver metastasis can also be managed surgically leading to an improved QoL as seen in our case. Whether this translates to an improved recurrence-free survival and OS cannot be commented at this stage due to the limited data on the subject. However, we can probably say that an aggressive surgical approach may be justified in metastatic GCT where there is limited disease with a long recurrence-free survival and good performance status as in our case.

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