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Case Report

An unusual case report of metastatic periampullary carcinoma

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ABSTRACT

Pancreatic cancers usually metastasize through the lymphoid system to organs such as the lung, liver, bone and spleen. Ovarian metastasis in pancreatic cancers is extremely rare, hence, it is difficult to distinguish between primary and metastatic ovarian tumors, especially in tumors with a primary source from the GIT & Hepatobiliary system. We present the case of a periampullary carcinoma with ovarian metastasis in a middle-aged female who presented with complaints of abdominal pain, constipation, yellowish discoloration of eyes and dark colored urine along with loss of appetite and weight loss for a duration of 6 to 8 weeks. Radiological examination revealed right adnexal lesion and nodular thickening along periampullary region. ERCP guided biopsy of the growth in periampullary region revealed moderately differentiated adenocarcinoma. She underwent pancreatico-duodenectomy with bilateral salpingo-oophorectomy. The histopathological examination revealed invasive carcinoma in both the ovaries, and moderately differentiated adenocarcinoma in periampullary and intra-ampullary region. As per the findings in previous studies, bilateral ovarian tumors of any size, or a unilateral tumor less than 10 cm likely represents metastatic disease rather than primary ovarian tumor. The rarity of co-presentation of pancreatic and adnexal mass makes the diagnosis tough however it is important to differentiate between primary ovarian mucinous cancers and ovarian metastasis from primaries in GIT for further treatment and follow up.

Keywords: Periampullary cancer, Adnexal lesion, Ovarian metastasis, Pancreaticoduodenectomy, Salphingo-ophorectomy

INTRODUCTION

Pancreatic cancer usually metastasizes through the lymphoid system to organs such as the lung, liver, bone and spleen. Ovarian metastasis in pancreatic cancers is rare, hence, it is difficult to distinguish between primary and metastatic ovarian tumors, especially in tumors with a primary source from the GIT and hepatobiliary system.[1,2] The pathological features of ovarian metastasis which may simulate primary ovarian tumor is another reason for misdiagnosis between the two. Additionally, the incidence of mucinous ovarian cancer (mEOC) as per latest studies is around 3% only and generally occur in young women and are diagnosed at an early stage, with 83% being diagnosed at stage I and only 17% at stage II or higher.[3] A significant decrease in the cases of Mucinous neoplasms of ovaries has been seen in the last two decades because a significant proportion of Mucinous carcinoma or mucinous borderline tumors (MBT) of the ovary, which were originally classified as primarily ovarian tumor, were actually of metastatic origin with the primary source located most commonly in the gastrointestinal tract.[4] We present a case of a 48 year lady who underwent surgery for periampullary carcinoma with right ovarian mass.

CASE REPORT

A 48-year-old woman, known case of hypertension and a renal donor, presented in our hospital with complaints of

abdominal pain, nausea/vomiting, constipation, yellowish discoloration of eyes and dark colored urine and loss of appetite and weight loss for a duration of 6 to 8 weeks. MRI of abdomen revealed mild nodular thickening along periampullary region of D3 segment of duodenum and diffusely dilated CBD (13.7 mm) with bilateral IHBRD (Figure 1). ERCP guided biopsy of the growth in periampullary region revealed moderately differentiated adenocarcinoma.

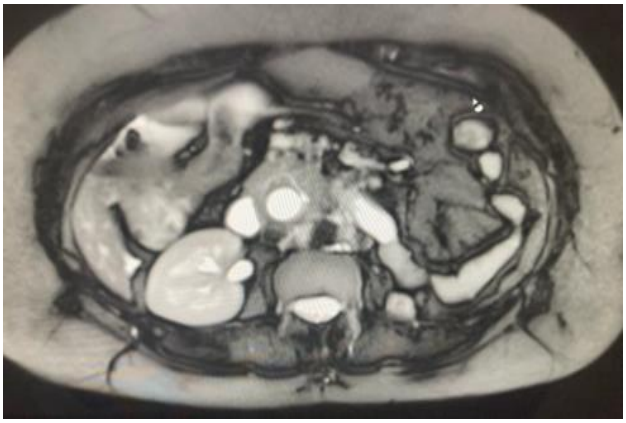

Figure 1: Dilated CBD.

FDG PET/CT scan was done which showed FDG avid hypo-attenuating soft tissue lesion showing variable enhancement in periampullary region involving D3 segment of duodenum measuring approximately 2.7×1.6 cm (SUV Max 9.70). Lesion abutting distal CBD with features suggestive of mitotic etiology. It also showed right tubo-ovarian complex cystic lesion measuring approximately 7.1×7.5×5.7 cm showing multi-loculation and mild FDG uptake (SUV Max 2.2) in enhancing internal septations suggestive of complex cystic lesion. There was no evidence of metabolically active lesion elsewhere in visualized region of body. Serum CA-19-9 was 2126.50 U/ml and CA-125 was 8.94U/m.

She underwent exploratory laparotomy. Intraoperatively there was 2×2 cm growth in periampullary region with indentation of transverse mesocolon to the pancreatic head region with dilated CBD and a solid cystic lesion in right ovary. There was no significant lymph nodes and no omental, peritoneal or mesenteric deposits and no ascites. Pancreaticoduodenectomy with reconstruction and bilateral salpingo-oophorectomy and feeding jejunostomy was done. Intraoperative frozen section of ovarian lesion was suggestive of serous borderline cystadenofibroma. Post operative period remained uneventful and patient was discharged in satisfactory condition.

The histopathological examination revealed invasive carcinoma in both the ovaries, and moderately differentiated adenocarcinoma in periampullary and intra-ampullary region. Stroma showed desmoplasia with capsular infiltration with few goblet cells. On immunohistochemistry, tumor cells in all the specimens from ovaries and pancreaticoduodenectomy specimen showed expression of CK19, EMA (MUC-1), CK20 and were negative for WT-1, ER and PAX8. Based on histopathology, diagnostic possibilities were: Metastatic adenocarcinoma to both ovaries, in known case of periampullary adenocarcinoma and primary bilateral ovarian adenocarcinoma (likely sero-mucinous type).

The patient received adjuvant chemotherapy with FOLFIRINOX. She is currently in regular follow up.

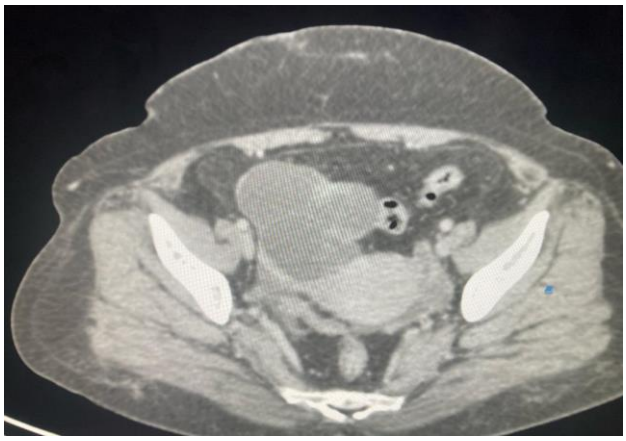

Figure 2: Complex right adnexal cystic lesion.

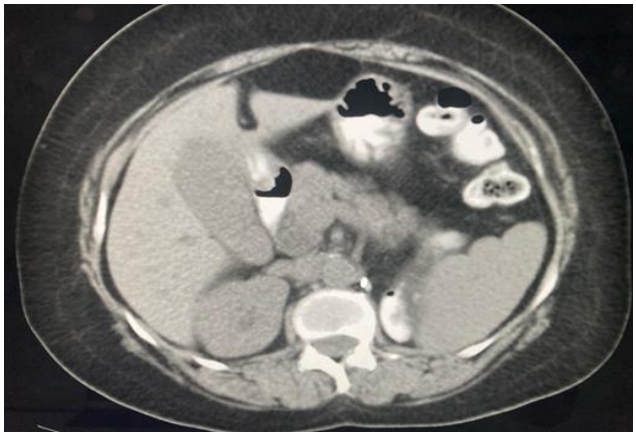

Figure 3: Tapering of distal CBD.

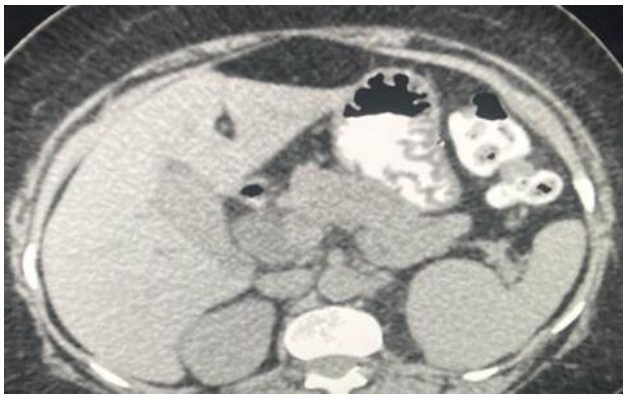

Figure 4: Periampullary lesion.

DISCUSSION

It is important to differentiate between primary ovarian mucinous cancers and ovarian metastasis from primaries in GIT for further treatment and follow up. Ovarian metastasis from the gastrointestinal tract including periampullary cancers show considerable morphological similarity that makes differential diagnosis difficult, especially in advanced disease.[3] The fact that both the ovaries were involved with cancer on histopathology, and knowing that a significant proportion of cases of metastatic pancreatic adenocarcinoma to the ovary tends to present bilaterally, the possibility of a periampullary cancer with ovarian metastasis cannot be ruled out.[2,4] The pancreas is an important source of metastatic tumors that simulate primary ovarian mucinous cystadenocarcinomas and borderline tumors.[1] Furthermore, it has been found in previous studies that bilateral ovarian tumors of any size, or a unilateral tumor less than 10 cm likely represents metastatic disease, while a unilateral tumor of size more than 10 cm likely represents primary disease.[2-4] In our case both the ovaries of the patient were involved with tumor, which further points towards ovarian metastasis rather than a primary ovarian neoplasm. Additionally, ovarian tumors with multinodular growth pattern, or desmoplasia around neoplastic glands as seen in our case, are more often seen in metastatic ovarian masses as opposed to primary ovarian carcinomas.[2,4] Raised CA19-9 and normal CA-125 values also favor the diagnosis of periampullary malignancy rather than a primary ovarian cancer. Dundr et al in their study showed expression of CK20, MUC-1 (EMA) and CK19 in both primary ovarian mucinous cancers and periampullary cancers, whereas markers like ER and PAX8 were shown to express only in ovarian cancers and not in pancreatic cancers, which further supports the diagnosis of ovarian metastasis from periampullary cancer in our case.[4] Recently, Liliac et al found that the co-expression of WT1 and PAX8 demonstrated a valuable association in confirming the ovarian origin of malignant effusions. They recommended WT1 in the differential diagnosis of primary ovarian tumors or in exclusion of uterine, breast, pancreatobiliary or gastrointestinal metastases, showing similar morphologic features, however the expression of WT-1 and PAX8 being strongly positive for serous ovarian cancers and extremely low for mucinous ovarian cancers. WT-1 positivity has also been proven to have a significant effect on the OS and DFS in cases of pancreatic ductal carcinoma. Kanai T et al, found in their study that the median DFS time for patients with PDA with weak and moderate to strong cytoplasmic WT1 expression was 543 and 196 days, respectively.[6] The differential diagnosis between primary mucinous epithelial ovarian cancers and mucinous metastasis from other organs also relies on other clinical characteristics including bilaterality, surface involvement, signet ring cell presence, and lympho vascular invasion, which are more common in metastases and quite rare in primary mucinous ovarian cancers.[3,4] There is still a certain proportion of tumors for which, based on the morphological and immunohistochemical (IHC) features alone, the distinction between a primary and a metastatic tumor is not possible. The reason is that the tumor morphology, IHC features, and even the molecular changes of primary and metastatic tumors may overlap. These tumors require a close clinical-pathological cooperation to decide for further plan.

CONCLUSION

This was an interesting case with periampullary tumor with isolated metastasis to bilateral ovaries. Although it is difficult to conclusively prove that whether this was a periampullary tumor metastatic to bilateral ovaries or primary ovarian malignancy with metastasis to GIT. Most reliable approach to the diagnosis of these tumors combines macroscopic, microscopic and IHC assessment combined with a close clinic-pathological correlation to decide appropriate plan of management.

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