Torsion Ovary, an Unusual Presentation of Carcinoma Appendix Detected in Whole-Body F-18 2-Fluoro 2-deoxy Glucose Positron Emission Tomography/Computed Tomography Scan

Abstract

Torsion ovary is one of the common emergencies in gynecology requiring surgery. Torsion ovary is generally caused by cystic lesions of ovary and benign tumors. Malignant tumors rarely present as torsion ovary. Krukenberg tumor presenting as torsion ovary is very rare with only a few case reports described in literature. Stomach is the most common primary site (70%) followed by colorectal, breast, lung, contralateral ovary, pancreatic, cholangiocarcinoma, and gallbladder carcinomas. Krukenberg tumor with primary in appendix is relatively rare. Here, we are presenting an unusual case of mucinous carcinoma appendix with Krukenberg tumor presenting as unilateral torsion ovary, demonstrating the role of whole-body F¹⁸ 2-Fluoro 2-deoxyglucose positron emission tomography/ computed tomography scan in identifying the primary.

Keywords: Appendix mucinous adenocarcinoma, Krukenberg tumor, positron emission tomography/ computed tomography scan, torsion ovary

Introduction

Carcinomas of appendix can be divided mucinous nonmucinous into and adenocarcinomas. Mucinous carcinoma usually causes peritoneal carcinomatosis and ovarian deposits. Stomach is the most common site for primary of Krukenberg followed by colorectal, breast, tumor lung. contralateral ovary, pancreatic. cholangiocarcinoma, and gallbladder carcinomas,^[1] Krukenberg tumor with primary in appendix is relatively rare. Mucinous adenocarcinoma of appendix with Krukenberg tumor presenting torsion ovary is extremely rare. as Whole-body F¹⁸ 2-Fluoro 2-deoxyglucose positron emission tomography/computed tomography (WB FDG PET/CT scan) is the most sensitive imaging modality to detect unknown primary. Here, we are presenting an unusual case of Krukenberg tumor with primary in the appendix manifesting as unilateral torsion ovary where PET/CT scan is very helpful in diagnosing the disease and guiding the clinician for further management.

Case Report

A 44-year-old female with a history of hysterectomy 6 months ago for fibroids

acute abdominal pain in the right iliac fossa. Ultrasonography of the abdomen showed bilateral ovarian cysts with torsion of the right ovary. Under emergency laparoscopy which was done elsewhere, she was diagnosed to have a large solid-cystic right ovarian mass measuring $10 \text{ cm} \times 8 \text{ cm}$ with one turn of torsion and signs of gangrene. She was also found to have a small hemorrhagic cyst measuring 5 cm \times 4 cm in the left ovary. In view of the difficulty in retrieval of the large ovarian mass, laparoscopy was converted to laparotomy suprapubic transverse incision. with Histopathology of the surgical specimen was suggestive of primary mucinous adenocarcinoma of bilateral ovary: Krukenberg tumor was given as second possibility. The patient was referred to our hospital for further management. Slide review and immunohistochemistry (IHC) examination revealed high-grade mucinous carcinoma [Figures 1 and 2]. In view of normal upper gastrointestinal (GI) endoscopy and colonoscopy and IHC for cytokeratin 7 and 20 being positive and CDX2 being negative, the possibility of primary mucinous carcinoma was favored. However, a guarded report was

presented to the emergency department with

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Figure 1: Whole body F-18 2-fluoro 2-deoxy glucose positron emission tomography/computed tomography scan. Left is the maximum intensity projection image showing 2-fluoro 2-deoxy glucose avid lesion in right iliac fossa (yellow arrow). Right is the axial and coronal positron emission tomography/computed tomography fused images showing diffusely enlarged appendix with increased 2-fluoro 2-deoxy glucose uptake (White arrows).

given as IHC does not conclusively differentiate primary mucinous carcinoma of ovary from secondaries. Magnetic resonance imaging (MRI) and ultrasonography (USG) abdomen did not reveal significant abnormality except for postoperative findings in the pelvis. Tumor markers: Serum carcinoembryonic antigen (CEA) - 4.89 ng/mL (0-3), serum CA 125-12.2 IU/mL (0-35.0), CA - 19.9-161.3 IU/mL (0-37). The patient underwent whole-body FDG PET/CT scan [Figure 1]. Scan showed diffusely enlarged (20 mm diameter) appendix with asymmetrically thickened wall and periappendiceal mild fat stranding (SUV_{max} - 13.5). Few subcentimetric mesenteric lymph nodes (short-axis diameter 9 mm) were also noted with faint FDG uptake. In view of histopathological diagnosis in favor of primary ovarian carcinoma and imaging morphology, subacute to chronic appendicitis was given as the first diagnosis with close differential diagnosis of neoplastic etiology as there were no other associated clinical or imaging features to suggest acute appendicitis. Metastases to appendix were less likely as entire length of appendix was involved. The patient underwent laparoscopic appendicectomy, and the frozen sections were suggestive of mucinous adenocarcinoma following which right hemicolectomy, lymph nodal dissection, and omentectomy were performed. Postoperative final histopathology diagnosis was Grade II Mucinous adenocarcinoma of appendix with positive two lymph nodes along caecum [Figure 3]. On IHC, microsatellite markers (MLH1, MSH2, MSH6, and PMS2) for MSI status revealed intact expression.



Figure 2: ×100 – H and E stain of ovarian tumor shows neoplastic glands and floating in pools of mucin

Discussion

Secondary ovarian tumor constitutes around 10% of ovarian tumors. Krukenberg tumors constitute up to 40% of secondary ovarian tumors.^[2] Route of spread is not clearly understood, but transcoelomic spread is considered the major route of spread. Differentiating primary mucinous ovarian carcinoma from Krukenberg tumor is crucial because management differs for both the conditions. Krukenberg tumors with primary in appendix are less common and vary from very rare to up to 3% of cases in literature.^[3] There are very few case reports of Krukenberg tumor presenting as unilateral torsion ovary.^[4] To the best of our knowledge, there are no case reports of carcinoma appendix with metastases to ovary presenting as torsion. Mucinous appendicular carcinoma with Krukenberg usually has associated peritoneal carcinomatosis which is absent in the present case. In the absence of peritoneal carcinomatosis, Krukenberg tumor poses a major challenge in pathological and radiological diagnosis.

Whole-body FDG PET/CT is the best modality to detect unknown primary tumor.^[5] Primary detection rate in PET/CT scan varies from 24% to 44% in various meta-analysis.^[6,7] In the present case, clinical presentation, histopathology, and IHC (Diffuse CK7 and C20 positive, CDX2 negative) could not differentiate primary versus secondary mucinous adenocarcinoma in ovary. Characteristics in favor of Krukenberg tumor were bilaterality, small size, surface deposits, normal CA-125, and mildly elevated CEA levels. Diagnosing carcinoma appendix in this case was challenging because (1) histopathology was more in favor of bilateral primary ovarian carcinoma even though Krukenberg tumor was not ruled out completely, (2) postoperative (total abdominal hysterectomy + bilateral salpingo-oophorectomy) USG abdomen and MRI did not pick any other lesion in the abdomen and pelvis, and (3) there was no peritoneal carcinomatosis. PET/CT scan was helpful in identifying the primary tumor and thus in



Figure 3: $\times 100$ – H and E stain of appendix tumor shows mucinous adenocarcinoma

guiding the clinician for further management of the patient with unusual presentation.

Conclusion

WB FDG PET/CT scan may be indicated in cases with clinical/pathological dilemma between bilateral primary ovarian carcinoma and Krukenberg tumor in identifying the unknown primary tumor.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understand that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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