Article published online: 2020-01-29

Case Report

Uncommon Manifestations of Endocervical Malignant Mixed Mullerian Tumor with Incidental Bilateral Fallopian Tube Carcinoma

Kishan Prasad H L, J H Makannavar, Jayaprakash Shetty, Vinutha[1], Channappa Patil[2], Harish S Permi

Departments of Pathology and [2]Oncology, K S Hegde Medical Academy of Nitte University, Mangalore, Karnataka, [1]Department of OBG, Bombay Hospital, Mumbai, India

Address for correspondence: Dr. Kishan Prasad H L, E-mail: dr_kishanpath@yahoo.com

ABSTRACT

A 43-year-old perimenopausal lady presented with bleeding per vagina and lower abdominal pain. On evaluation, she had cervical polyp, which expelled spontaneously during the per speculum examination. Histopathology revealed malignant mixed Mullerian tumor. Extended hysterectomy with salphingo oophorectomy was carried out, which showed bilateral fallopian tube carcinoma and leiomyoma uterus. The patient was treated with carboplatin regime and found to be disease-free for 1 year. This case presented because of a rare combination of the lesions.

Keywords: Fallopian tube, malignant Mullerian tumor, primary adenocarcinoma

INTRODUCTION

Malignant mixed Mullerian tumors (MMMTs) are metaplastic carcinomas including both sarcomatous and carcinomatous elements commonly involving the uterus and ovary.[1,2] MMMTs constitute less than 5% of the uterine malignancies, rarely involving the endocervical region. Recent studies have provided evidence to classify them as variants of carcinoma.[2] Primary adenocarcinoma of the fallopian tube is a rare tumor, accounting for only 0.3–1% of all gynecological malignancies.[3] Bilateral involvements are still rare. The diagnosis is seldom made pre-operatively because the signs and symptoms are not specific and are often mistaken for ovarian tumor or tubo ovarian mass.[3,4] Some tubal primaries with disseminated disease involving the ovaries are misclassified as ovarian in origin.[3,5] We are reporting an extremely rare case of endocervical MMMT, leiomyoma uterus and bilateral fallopian tube carcinoma in a perimenopausal woman.

Access this article online

Quick Response Code:

Website: www.jlponline.org

DOI: 10.4103/0974-2727.78569

CASE REPORT

A 43-year-old gravida 2, para 2, non-tubectomised perimenopausal female presented with bleeding per vagina (PV) and lower abdominal pain since 1 month. Bleeding PV was continuous, with no clots. Lower abdominal pain was of the spasmodic type, mainly in the supra pubic region. She had intermenstrual bleeding since 6 months. The patient had last child birth 23 years ago by caesarian section. General and systemic examinations were unremarkable. Per-speculum examination revealed a polyp protruding through the external os measuring 2cm × 1cm × 1cm, which was expelled spontaneously. Her hemogram, biochemical parameters, X-ray chest, ultrasound of abdomen and pelvis were within normal limits. Human immunodeficiency virus, hepatitis B surface antigen and hepatitis C virus tests were negative. The computed tomography abdomen and pelvis revealed a small soft tissue density lesion in the anterior uterocervical junction measuring

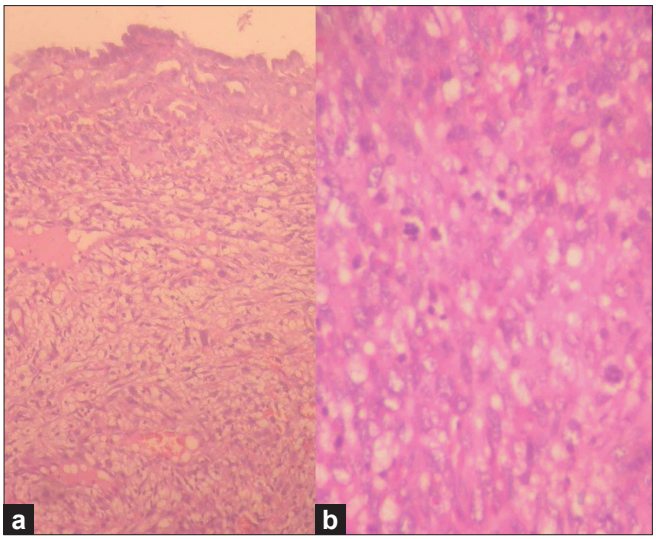

Figure 1: (a) The endocervical epithelium and stroma showing malignant features (hematoxylin and eosin, ×100), (b) Deeper stroma showing endometrial stromal sarcoma features (hematoxylin and eosin, ×400)

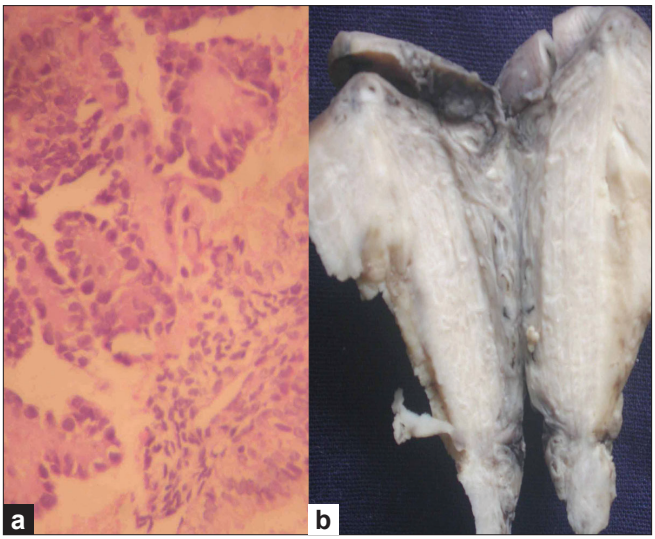

Figure 2: (a) Curettage material with papillary carcinoma (hematoxylin and eosin, ×100), (b) Cut surface of the hysterectomy specimen with friable tissue in the endocervical region

2cm × 1cm. The endometrial cavity was normal and no ascites or lymph nodes were detected. Hysteroscopy revealed a growth within the lateral wall of the uterus. Curettings from endometrial and endocervical region were collected for examination. Based on histopathology report of expelled polyp and curettings, extended hysterectomy with bilateral salphingo oophorectomy was performed. Intraoperatively, the uterus was 8 weeks in size. Both the fallopian tubes showed hydrosalphynx. Both the ovaries were normal. No free fluid was seen in the peritoneal cavity.

PATHOLOGICAL FINDINGS

The spontaneously expelled grey-brown polyp [Figure 1a] on microscopy showed biphasic tumor cells with the epithelial component arranged in papillary structures and tubules. The stromal component [Figure 1b] showed oval to spindle tumor cells within a myxoid background and numerous proliferating vascular channels. Also, areas of hemorrhage and necrosis were seen. Features were suggestive of MMMT. Curettings from the endometrium and endocervix tissue [Figure 2a] showed papillary carcinoma-endocervix.

The extended hysterectomy specimen showed tiny friable material in the endocervical region [Figure 2b] and intramural leiomyoma, 1cm × 0.5cm in the posterior wall of the uterus. The left fallopian tube was tortuous and dilated, 4cm × 2cm × 1cm. The right fallopian tube was enlarged and tortuous, 5cm × 2cm × 2cm, with a solid grey white to yellowish appearance on the cut surface. Microscopy showed a proliferative endometrium. The endocervix showed minute papillary carcinomatous tissue tethered to the surface. The myometrium showed tiny intramural leiomyoma. The left fallopian tube showed *in situ* papillary carcinoma [Figure 3a].The right fallopian tube showed invasive serous papillary carcinoma with *in situ* changes in the adjoining tubal epithelium [Figure 3b]. Both ovaries were normal. In view of the histological findings in the expelled polyp, curettage material and with the hysterectomy findings, a final diagnosis of MMMT-endocervix, intramural leiomyoma and bilateral primary fallopian tube carcinoma was made.

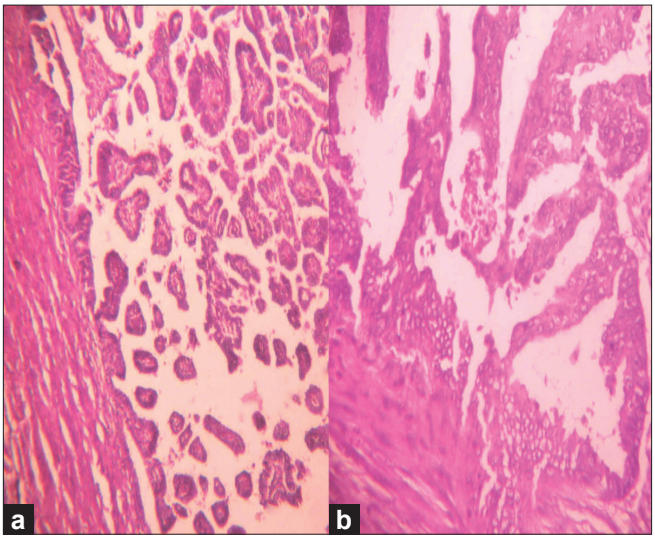

Figure 3: (a) Left fallopian tube carcinoma with in situ carcinoma (hematoxylin and eosin, ×100) (b) Right fallopian tube carcinoma with invasive carcinoma (hematoxylin and eosin, ×400)

DISCUSSION

This collision tumor of MMMT-endocervix, leiomyoma uterus and bilateral fallopian tube carcinoma is an extremely rare entity. Such a combination of manifestations could not be found on extensive review of the literature. The tubal carcinoma may occur as a part of multiple upper genital tract malignancies.[6]

The MMMT accounts for <5% of the uterine corpus neoplasms and, rarely, involves the endocervical region.[1,2,7,8] They are similar to uterine lesions, commonly presenting as post-menopausal bleeding. They present with enlarged irregular uterus and tumor protruding through the cervical os as a polypoidal mass.[2] Our patient was a perimenopausal lady with pervaginal bleeding, lower abdominal pain and with friable polyp protruding through the external os.

MMMTs on histology show an admixture of malignant epithelial and mesenchymal elements. The epithelial element may be endometrioid, serous, clear cell, mucinous, squamous and undifferentiated carcinoma. The stroma may be homologous in the form of endometrial stromal sarcoma, fibrosarcoma and leiomyosarcoma or heterologous like rhabdomyosarcoma and chondrosarcoma.[1,2,7,8] In our case, the epithelial component was of a serous type and homologous stroma showed endometrial stromal sarcoma. The MMMTs metastasize to the pelvic and para aortic lymph nodes, pelvic soft tissues, vagina and peritoneal surfaces of the upper abdomen. Surgical stage is the most important prognostic factor in MMMT. Small MMMTs confined to the tip of a polyp can have a favorable prognosis; such tumors can metastasize and be fatal.[2] In our case, pelvic organs, peritoneal surfaces and the pelvic and para aortic lymph nodes were free. Primary adenocarcinoma of the fallopian tube was first described by Renand in 1897, and is the rarest malignancy of the female genital tract.[3] These tumors occur between 14 and 88 years of age, with the majority in the 6^{th}–7^{th} decades.[3,5] Vaideeshwar *et al*.[9] described tubal carcinoma in a 40-year-old female, which was comparable to our case. The duration of symptoms in such patients ranges from 3 days to a few years.

Patients with abdominal pain seek medical attention earlier than those with vaginal discharge or menstrual problems.[6] However, in our case, the patient had abdominal pain and menstrual problems.

Primary tubal carcinoma is rarely found in the *in situ* stage.[6] In our case, left fallopian tube carcinoma was in an *in situ* stage while right tube carcinoma was invasive. Sarangetham *et al*. reported bilateral tube carcinoma associated with leiomyoma.[3] Hydrops tubae profluens is the typical finding of primary fallopian tube carcinoma in magnetic resonance imaging.[4,6] Pre-operative and imaging studies show findings similar to fallopian tube carcinoma in hydrosalphynx, tubo-ovarian abscess and ovarian neoplasm, making its diagnosis difficult.[4] This could explain why its diagnosis was missed in our case on imaging and pre-operatively.

Diagnosis of fallopian tube carcinoma is based on the following two criteria:[10] (a) main tumor mass to be confined to the tube with demonstrable carcinoma *in situ* changes in the adjacent mucosa and (b) uterus and ovaries should be normal. In our case, bilateral fallopian tubal carcinoma was confined to the lumen of both the tubes, with *in situ* changes in the adjacent tubal mucosa. The ovary and endometrial tissue were normal. However, MMMT was seen within the endocervix.

The tumor stage has a better prognostic value than the histological grade in fallopian tube malignancies. Serous papillary tubal carcinomas may be associated with marked chronic inflammation. Various forms of salphingitis, including tuberculosis, may produce severe hyperplasia of the tubal epithelium with sufficient reactive atypia and mitotic activity to mimic *in situ* or invasive adenocarcinoma. Close attention to severe nuclear atypia and abnormal mitosis and evidence of invasion is always necessary to prevent misdiagnosis.[3]

Serum CA 125 is reported to be elevated in fallopian tube carcinoma, and its pre-treatment level is considered to be an independent prognostic factor.[3] In our case, bilateral fallopian tube carcinoma was an incidental finding and, hence, the serum CA 125 level was not assessed. However, its post-operative level was within normal limits.

Post-operatively, the patient received five cycles of chemotherapy (carboplatin 600 mg). After 1 year of therapy, she remained asymptomatic and is still under regular follow-up.

ACKNOWLEDGMENT

We are thankful to Dr Supriya Rai, Professor of OBG, K S Hegde Medical Academy of Nitte University for her valuable suggestions and guidance.

REFERENCES

1. Rosai J. Female reproductive system. In Rosai and Ackerman's surgical pathology. 9th ed. New Delhi: Elsevier publication; 2004. p. 1483-761.

2. Ronnett BM, Zaino RJ, Ellenson LH, Kumaran RJ. Endometrial carcinoma. In: Kumaran RJ, editor. Blausteins pathology of the female genital tract. 5th ed. New Delhi: Springer Publication; 2002. p. 501-59.
3. Sarangthem B, Laishram S, Sharma B, Konjegbam R, Debnath K. Primary bilateral tubal adenocarcinoma associated with uterine leiomyomas. Indian J Pathol Microbiol 2008;51:32-3.
4. Hosokowa C, Tsubakimoto M, Inoue Y, Nakamura T. Bilateral primary fallopian tube carcinoma:findings on sequential MRI. AJR Am J Roentology 2006;186:1046-50.
5. Sonmezer M, Ustun Y, Gungor M, Ensari A, Ortac F. Primary carcinoma of the fallopian tube in a 88years old woman:review of literature. Ind J Cancer 2001;38:61-4.
6. Wheeler JE. Diseases of the fallopian tube. In: Kumaran RJ, editor. Blausteins pathology of the female genital tract. 5th ed. New Delhi: Springer Publication; 2002. p. 617-22.
7. Ho SP, Ho TH. Malignant mullerian tumors of the uterus-A ten year experience. Singapore Med J 2002;43:452-6.
8. Fujii H, Yoshida M, Gong ZX, Matsumoto T, Hamano Y, Fukunaga M, *et al*. Frequent genetic heterogenecity in the clonal evolution of gynecological carcinosarcoma and its influence on phenotypic diversity. Cancer Res 2000;60:114-20.
9. Vaideeswar P, Desai MS, Prabhat DP, Desai DP. Fallopian tube carcinoma: A report of two cases. Indian J Pathol Microbiol 1997;40:397-9.
10. Yogiraj SS, Vasantrao KP, Ranganathrao SB. Fallopian tube carcinoma. Indian J Pathol Microbiol 2008;51:261-2.

How to cite this article: Prasad KH, Makannavar JH, Shetty J, V, Patil C, Permi HS. Uncommon manifestations of endocervical malignant mixed mullerian tumor with incidental bilateral *Fallopian tube* carcinoma. J Lab Physicians 2011;3:52-5.

Source of Support: Nil. **Conflict of Interest:** None declared.