EJGO European Journal of Gynaecological Oncology

Case Report

Primary fallopian tube carcinoma with isolated appendiceal metastasis: A case report and review of literature

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Summary

We report the first documented case of primary fallopian tube carcinoma with isolated appendiceal metastasis. A 67-year-old postmenopausal woman was referred for an asymptomatic adnexal mass that was discovered incidentally by transvaginal ultrasound during a routine cancer screening. Abdominal-pelvic CT and pelvic MRI showed a 6 cm right tubal mass suggesting malignancy. A second suspicious 2 cm solid appendiceal mass was found to be metastatic. Debulking surgery diagnosed a right tubal high-grade serous carcinoma. No gross or microscopic metastasis was found in the abdominal cavity, apart from an appendiceal metastasis without sites of serosal involvement.

Key words: Appendiceal metastasis; Metastasis; Fallopian tube carcinoma.

Introduction

Primary fallopian tube carcinoma (PFTC) is a rare tumor of the female tract with an incidence 0.3 to 1% in all gynecologic malignancies [1, 2]. At the time of diagnosis, 70% of cases are late stage, and previous studies reported the 5-year PFTC patient survival rate is 44.5% [3]. PFTC and ovarian carcinoma use similar mechanisms - cells exfoliate to the peritoneal cavity, and there's sustained invasion characterized by transluminal migration, hematogenous dissemination, or by lymphovascular invasion as cancer cells break into blood vessels or lymphatic channels [4-6]. Distant metastasis is extremely rare, and only a few cases have been reported previously [7-11]. This is the first documented case of PFTC associated with appendiceal metastasis.

Case report

Transvaginal ultrasound on a 67-year-old woman (G5P4) found a left ovarian cyst measuring 5.7 × 3.0 cm. Initial outpatient screening showed cancer symptoms, but the patient had a 7-year history of hypertension. Patient G5P4 was referred to our clinic and no remarkable abnormalities were found during physical and pelvic examinations. CA125 testing showed the protein was elevated (444.6 U/mL) and this triggered further imaging studies to look for gynecologic malignancies. Abdominal and pelvic computed tomography (CT) revealed a 5.0 cm cystic fluid-attenuated right adnexal mass containing a 3.5 cm enhancing mural nodule. There was also focal distension with a 2.0 cm enhancing solid mass at the appendiceal tip. Lymph nodes were not enlarged nor were other suspicious lesions found on images of her abdominal and chest CT scan. Pelvic magnetic resonance imaging (MRI) showed a 6.0 × 3.5 cm mass in the right fallopian tube with a prominent solid portion and lobulated contour with accompanying hydrosalpinx (Fig. 1A). The mass was heterogeneously isointense on T1- and T2-weighted images, with restricted diffusion. In addition, a mural nodule measuring approximately 1.9 × 1.4 cm with intermediate signal intensity on T2-weighted images was seen at the tip of the appendix (Fig. 1B), without evidence of peritoneal or lymph node metastasis. At exploratory laparotomy, the right fallopian tube was enlarged, with the tumor occupying the lumen, but the tubal surface was intact and absent of tumor invasion. Histopathology diagnosis was serous carcinoma. The right ovary was grossly normal and didn't show any abnormal pathology except for senile atrophy, and the left ovary and tube were also normal. Pelvic cavity ascites fluid measured less than 10cc. Fluid was aspirated and pelvic washing was performed for cytology. An abdominal hysterectomy was performed, followed by bilateral salpingo-oophorectomy with total omentectomy, pelvic and para-aortic lymph node dissection, and appendectomy. The patient's appendix was enlarged and the appendiceal mass measured approximately 3 cm. Serous membrane tissue adhered to the right paracolic gutter, but the mass was confined to the appendix and did not breach the serosa. There was no visible tumor seeding elsewhere in the abdominal cavity. The final pathology diagnosis was high-grade serous carcinoma of the right fallopian tube. The tumor measured 4.5 × 3.5 × 3.0 cm and was confined to the lumen (Fig. 2). There was no lymphatic, venous, or perineural invasion. The uterus, both ovaries, and the left fallopian tube were also free from tumor, as were the omentum, pelvis, and para-aortic lymph nodes. There was no malignancy on cytology of the pelvic

Published: 15 April 2020

Eur. J. Gynaecol. Oncol. - ISSN: 0392-2936
XLI, n. 2, 2020
doi: 10.31083/j.ejgo.2020.02.5242

ascites. A second tumor was found on the appendix. This solid tumor measured 3.2 × 1.8 × 1.5 cm, with growth into the lumen and extension to the subserosa layer. However, the serosal surface was not involved. The histopathologic diagnosis was metastatic high-grade serous carcinoma of the appendix (Fig. 3). The patient received six cycles of adjuvant carboplatin (carboplatin: AUC 5) and paclitaxel (paclitaxel: 175 mg/m^2) chemotherapy. Patient monitoring at 36-months showed no sign of cancer recurrence.

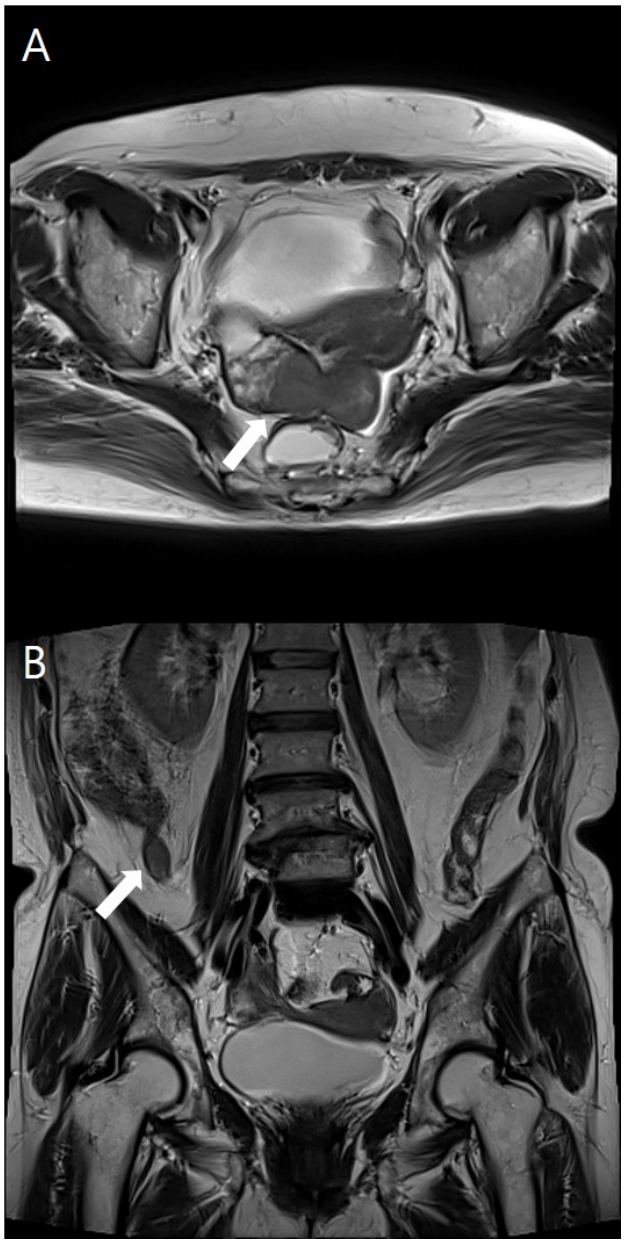

Figure 1. — (A) Axial contrast-enhanced T2-weighted pelvic MRI shows a 6.0 × 3.5 cm sized lobulated right fallopian tubal mass with heterogenous enhancement (arrow). (B) Coronal contrast-enhanced T2-weighted pelvic MRI shows a 1.9 × 1.4 cm sized mass in the tip of appendix with heterogenous enhancement (arrow).

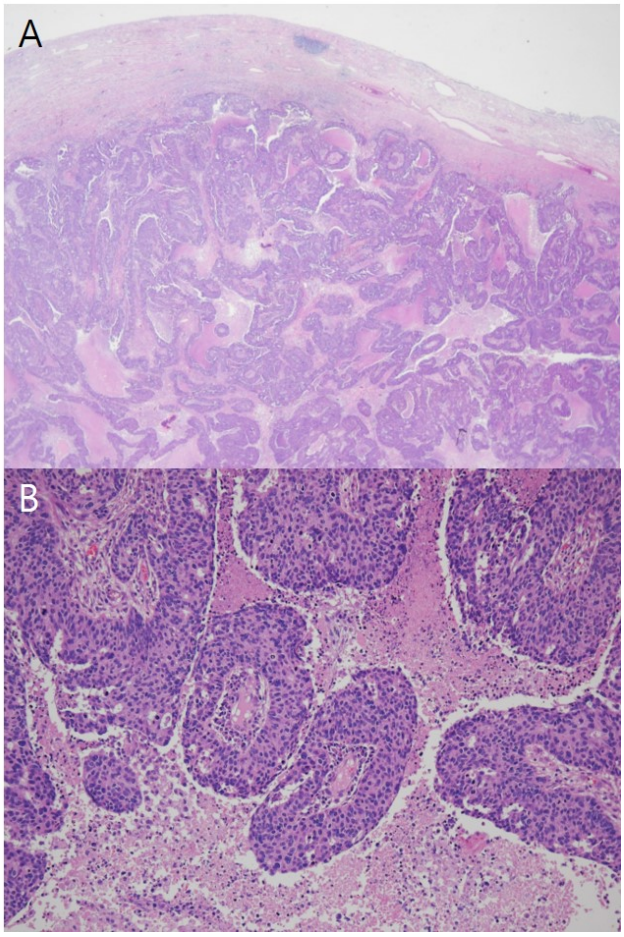

Figure 2. — Hematoxylin and eosin (H&E) stained cross-section of the right fallopian tube. The tumor is confined to the fallopian tube. Histology demonstrates high-grade serous carcinoma. (A) × 10 and (B) × 100.

Discussion

Primary fallopian tube carcinoma (PFTC) is a rare gynecological tumor. PTFC incidence in the United States was 0.63 per 100,000 between 2011 to 2014. When comparing to incidence rates between 2001 to 2005, this represents a 4.19-fold increase [12]. The highest incidence was seen in women aged 70 to 74 years old [12].

The etiology of PFTC is unknown, but fallopian tube mucosal chronic inflammation has been suggested as a cause of both PFTC and ovarian cancer, whereas oral contraceptives, high parity, and infertility are associated with reduced PFTC risk [13]. *BRCA1* and *BRCA2* gene mutations might also have a causal relationship with PFTC [14].

Patients with PFTC may present vaginal bleeding or spotting (50 to 60%), colicky abdominal pain (30 to 49%), or abdominal mass (12 to 61%) [5]. PFTC tends to be diagnosed at an earlier stage than epithelial ovarian cancer (EOC) because the latter is often asymptomatic in early stage disease [5, 15, 16]. However, preoperative PFTC di-

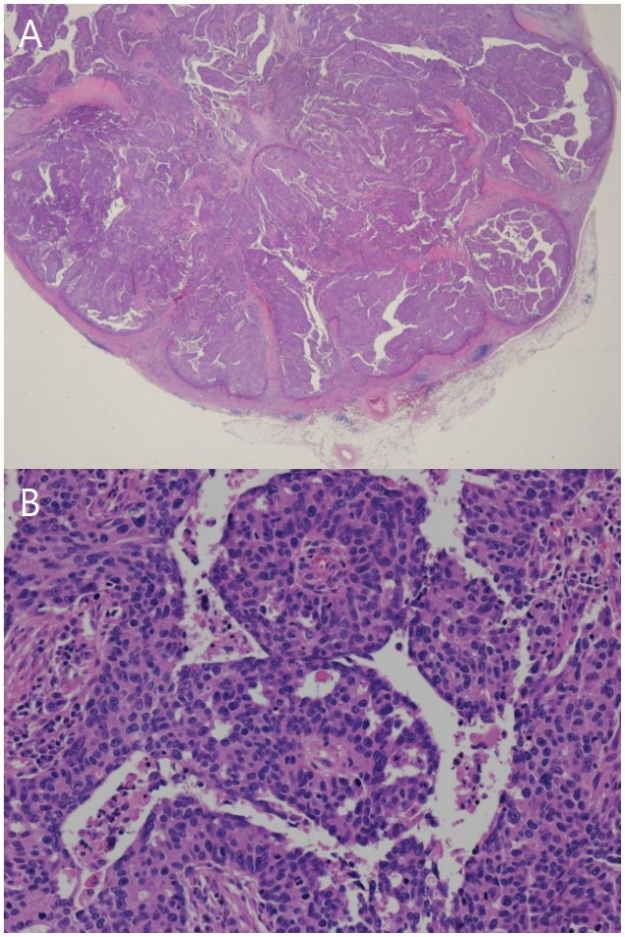

Figure 3. — Hematoxylin and eosin (H & E) stained cross-section of the appendix. Histology demonstrates metastatic high-grade serous carcinoma from the right fallopian tube. Note that appendiceal serosa was not penetrated by the tumor. (C) ×10 and (D) ×200.

agnosis is difficult due to its rarity and nonspecific clinical manifestations. It is often misdiagnosed as an ovarian tumor or pelvic inflammatory disease. Imaging tools such as ultrasonography, CT, and MRI are helpful to make an exact preoperative diagnosis and for preoperative staging [15, 17].

Fewer than 5% of PFTC patients are accurately diagnosed before surgical exploration. The ideal outcome of surgical PFTC treatment is to achieve tumor debulking and optimal cytoreduction resulting in no gross residual disease. Total abdominal hysterectomy, bilateral salpingo-oophorectomy, omentectomy, peritoneal washings, peritoneal biopsy, pelvic and para-aortic lymph node dissection, are procedures that are used to reduce tumor burden. Adjuvant combination chemotherapy with paclitaxel and carboplatin is considered. Tumor stage, maximum diameter of residual tumor after cytoreductive surgery, and tumor histology and grade are all associated with patient prognosis [16, 18].

As with ovarian carcinoma, PFTC can spread by cell exfoliation to the peritoneum, contiguous invasion, transluminal migration, and hematogenous or lymphatic spread [4-6]. In particular, the abundant supply of lymphatics to the fallopian tube contributes to more frequent lymph node metastasis, even when the tumor is apparently confined to the fallopian tube [10, 11, 19].

The appendix is a common site for EOC because of its proximity to the right ovary and adjacent paracolic space, where ascites drains [20]. However, isolated appendiceal metastasis in association with EOC is rare - especially, in the absence of pelvic disease at secondary sites. Bese *et al*. reported 10 cases of appendiceal metastasis among 90 patients with EOC, and all of them had macroscopic metastasis elsewhere in the abdomen [21]. Fontanelli *et al*. had 37 patients with appendiceal involvement among 160 patients with EOC, but none had isolated appendiceal metastases [22], and Ayhan *et al*. published patients with stage I EOC had isolated microscopic appendiceal metastasis of 2.8% [20]. To date, there has been only one reported case of a metastatic serous papillary adenocarcinoma infiltrating through to the serosal surface [23].

Our case differs from the above-mentioned reports in that the primary tumor was confined to the fallopian tube, the amount of ascites was small, and no metastatic tumor was observed in the pelvic washings. Furthermore, there was no tumor infiltration to the appendiceal serosa and surrounding peritoneum, which implies a lower possibility of metastasis by cell exfoliation. Although it is difficult to determine the exact route of appendiceal metastasis in our patient, we can hypothesize that hematogenous spread is the most likely route. Transluminal migration of a small number of exfoliated cells from the primary tumor is another distinct possibility.

Conclusion

Preoperative diagnosis of fallopian tube carcinoma is difficult due to the silent nature of PFTC malignancy. A better understanding of metastatic lesions lead to complete resection and improve PFTC patient outcome.

Conflict of Interest

The authors declare no competing interests.

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