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Tubal Cancer in a Young Woman Camouflaged as Bilateral Salpingitis

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Abstract

Introduction: Primary fallopian tube carcinoma (PFTC) is extremely uncommon, accounting for between 0.1–1.8% of all gynecological cancers diagnosed. Due to nonspecific symptoms, the diagnosis is often mistaken for an ovarian carcinoma or a tube-ovarian mass.

Objective: To report an interesting case of tubal cancer diagnosed in à nullipare patient.

Materials and Methods: We report about a 26-year old premenopausal patient presented with a 15-day history of mesocycle vaginal bleeding and lower abdominal pain, with multiple abdominopelvic masses, detected through pelvic ultrasound.

Results: Exploratory laparotomy was performed, and one of the tumors was diagnosed as a primary fallopian tube carcinoma (PFTC), as well as bilateral salpingitis' formations. The patient underwent complete cytoreduction. Pathology confirmed a high-grade serous carcinoma of the left salpinx with a FIGO stage considered as 1A2, and the patient received adjuvant chemotherapy. Ten months after initial surgery, the patient is alive and in good condition.

Conclusion: Tubal cancer is extremely rare in young patients, however it should be considered in the differential diagnosis of adnexal tumors.

Keywords: Fallopian tube; PFTC; Adenocarcinoma premenopausal female; Intraoperative assessment; Histopathological diagnosis; Chemotherapy

Introduction

Primary fallopian tube carcinoma (PFTC) is a very rare gynecologic malignant tumor accounting for approximately 0.14-1.8% of female genital malignancies [1,2]. More than 60% of cases occur in postmenopausal women, with a mean age of 55 years. Since 1888, when it was first described in the literature, approximately 2000 cases have been reported. It is the rarest of all the female malignancies and it histologically and clinically resembles epithelial ovarian cancer (EOC). Moreover, many support the theory that high-grade serous ovarian carcinoma, as well as peritoneal carcinoma, may in fact originate from occult high-grade serous carcinoma in the fallopian tubes [3]. However in the last few years, the reported cases of PFTC seem to rise in number. For this reason, the hypothesis that it was often misdiagnosed and underestimated in the past has been formulated. This is supported by the fact that PFTC may have been mistakenly identified as ovarian tumors during initial surgery and/or during microscopic examination by a pathologist, as the histological appearance of these tumors is identical [4].

Preoperatively, a correct diagnosis is rarely achieved and in many cases, the diagnosis is made after incidental surgery for unrelated conditions as it is often mistaken for benign pelvic disease or ovarian cancer. Frequently, PFTC present symptoms that include abdominal pelvic pain or symptoms of pressure and vaginal bleeding. This bleeding is frequently associated with a watery vaginal discharge. The upper symptoms are known as the Latzko's triad, but they are only present in less than 15% [1].

Primary adenocarcinoma of the fallopian tube with papillary features is the most common histological type of primary tubal cancer (>90%). Especially, serous carcinoma appears to be the most common histologic type. Compared with ovarian carcinoma, PFTC more often presents at early stages, but it has a worse prognosis. PFTC is usually managed in the same manner as ovarian cancer [5].

Our aim is to present the clinical and imaging characteristics of one case of PFTC, its differential diagnosis and management in our clinic and in the long term to raise awareness that it seems to be more frequent than it was previously thought.

Case Report

A 26-year-old premenopausal nulliparous woman, with no significant personal or family history presented with a 15-day history of mesocycle vaginal bleeding accompanied by lower abdominal pain, defecation disorders and increased vaginal discharge. The pain was described as intermittent, sudden and sharp, lasting about 2 hours, then turned to continuously dull pain, surrendered by NSAIDS but intensifying during sexual intercourse. The defecation disorders included episodes of diarrhea escorted by pain and occasional constipation. The vaginal discharge was noticed to be increased and thicker for almost 30 days.

Her medical history included a loop cone biopsy of the cervix that showed CINIII and contraceptive use. She was a "social" smoker with almost 1,5 PY and she denied alcohol consumption. Her BMI was 19,1.

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The patient was admitted in our clinic for further investigation. On the admission day, her blood pressure was 105/60 mmHg, the pulse rate was 68 beats per minute, and her temperature was 37.5°C. During the gynecological examination, a pelvic mass was felt and a bimanual examination showed a normal sized anteverted uterus with cervical motion and adnexal tenderness. Speculum examination showed a normal cervix and vagina.

The white cell count on admission was 7.74×109/L and the C-reactive protein level was 43.3 mg/L. The CA 125 antigen level was 871.10 U/mL, much elevated in comparison with a level of 146.8 U/mL, which was the count 2 weeks earlier. A second CA 125 count was performed 2 days later and the level had been thrown up to 1275.70 U/ml. Other tumor markers including serum CA19–9, alpha fetoprotein (AFP) and carcinoembryonic antigen (CEA) were within normal limits. B-HCG was negative and Pap smear showed a normal cytology. A chest radiograph was also normal. Her hemogram, hepatic and renal functions were normal. Blood sugar and urine examination were normal.

Pelvic ultrasonography detected a 16.5 mm cystic mass in the right side of the pelvic cavity, a uterus of normal appearance, and a left adnexa without pathological findings. Transvaginal ultrasonography displayed a cyst on the right ovary a cyst of maximum diameter 14mm. Its walls were clear and smooth, without any echogenic elements inside. Another large cystic formation of a maximum diameter of 63mm was located in the right ovary, with echogenic elements within it (hemorrhagic cells - chocolate cyst?) with an overwhelmed wall. A similar pattern was observed in the left ovary, approximately 70mm (36X70mm). No fluid collection was observed in the pouch of Douglas. A pelvic CT scan was performed to evaluate the pelvic masses further which showed bilateral adnexal cystic masses with fusiform or sausage-like shapes. The right adnexal mass appeared as a complex cystic mass without any papillary projections and the left adnexal mass had just a regular thick wall. The right ovary was normal, but the left ovary was not visualized by CT.

The preoperative differential diagnosis included bilateral endometrioma, chronic pelvic inflammatory disease, tubo-ovarian abscess and tubo-ovarian cancer.

A diagnostic laparoscopy was performed. During the operation, free peritoneal fluid was found and was sent for cytology examination. The fallopian tubes were both enlarged, fixed to the pouch of Douglas. Deposits from the sigmoid, the left adnexa, the (infiltrated) ileum and the right fallopian tube were sent for a frozen section, with the second one being malignant and described as a high grade serous adenocarcinoma infiltrating the tubal wall. The right fallopian tube was also sent for a frozen section, when during its incision, a process of maximum diameter 5 cm and macroscopic characteristics similar to those of the left fallopian tube, was identified. However, this frozen section was negative.

Based on these findings, the surgery was converted to laparotomy and the patient underwent a total abdominal hysterectomy, bilateral salpingo-oophorectomy, adhesiolysis, supracolic omentectomy, bilateral pelvic and paraaortic lymph node dissection up to the level of the left renal vein. Peritoneal washing was also performed.

The section of the left tube in the operation room revealed a dilated tube d.9,5cm containing mostly pus-like fluid without a solid component, measuring 6cm, with a white homogeneous appearance, muco-elastic composition and a region measuring 1,2cm with unclear margin and friable composition.

The histopathologic examination revealed a left tubal solid high grade serous adenocarcinoma which was found to infiltrate the tubal wall. Neoplastic infiltrations are recognized in the wall without, however any extension to the outer surface / serosa. The described findings of the fallopian tubes (maximum diameter: 6cm left and 5cm right respectively) were related to severe, chronic, active inflammation that leaded to mucosal ulceration of several areas with a complete loss of the mucus. The inflammation at some points infiltrates the wall through its entire thickness and extends to the outer surface / serosa. There are also a few sites of reactive hyperplasia of the mucosal epithelium, the type of pseudo carcinomatous hyperplasia. The uterus, omentum and the lymph nodes were free of disease. The sigmoid deposits were all free of disease, with inflammatory lesions similar to those of the fallopian tubes. The peritoneal washings showed no malignant cells.

Immunohistochemistry findings included positive ER and P53 in all the neoplasmatic cells), PR focally positive almost at 40% of the neoplasm and positive WT1 and CK7 when CK20 was found negative.

Furthermore, a second pathology opinion was requested, which confirmed the diagnosis.

On the basis of these findings, a stage IA2 primary fallopian tube cancer was confirmed. She received an adjuvant chemotherapy with carbotaxol, based on the MDT's decision. She was also recommended to undergo a gene control scanning, which was negative. One year later, the patient is alive and in good condition.

Discussion

Primary cancer of the fallopian tube is a very rare gynecological cancer, with an average annual incidence of 3.6 per million women per year and a peak incidence between the ages of 60 and 64 years [5]. Bilateral disease is even more uncommon and represents fewer than 25% of cases. However, although more than 60% of cases occur in women with a mean age of 55 years, it can rarely occur in premenopausal women as well [6]. Reviewing the literature in the light of our case we found very few similar cases, with Boufettal and Samouh reporting a primary tubal adenocarcinoma in a woman aged 42 discovered as a result of an abdominopelvic mass [7] while You and Wang describe an even more uncommon type of a primary leiomyosarcoma (LMS) of the fallopian tube [8].

The average annual incidence of PFTC is estimated at around 3.6 per million women per year [5]. The true incidence of PFTC may, however, have been underestimated. It is common that during initial surgery and/or microscopic examination PFTC are mistaken for ovarian tumors, due to the similar histological appearance of these neoplasms [9]. However, the consultation period for patients with cancer of the fallopian tube is shorter than that of patients with ovarian cancer, because of abdominal pain secondary to distention of the fallopian tubes. Besides ovarian cancer, a peritoneum neoplasia can be first noted as a pelvic mass with elevated tumor markers while benign masses can also be disguised as malignancy. Lee et al report a case of pseudocarcinomatous hyperplasia of the fallopian tube

mimicking tubal cancer in a 22 years old woman [10] and Khatib et al describe a case of cystadenofibroma [11]. Pelvic inflammatory disease should be always taken under consideration, especially in young females. Wang et al conducted a review of patients (both pre-and postmenopausal) with tubo-ovarian abscess disguised as pelvic mass, putting this diagnosis among the others suspected when dealing with a fallopian malignancy [12]. Cases of tubal choriocarcinoma have also been reported [13,14].

Rarely, PFTC may affect young women who have not yet completed childbearing. The age of our patient is 30 years younger than the average age reported in the literature. Unluckily, the literature regarding the management of the surgical procedure and the remaining fertility of the patient in such cases is poor. Any data we found concerned the management of ovarian cancer in young women and in the end, it could only play the role of recommendation, as no specific guidelines are formed yet. These techniques are mainly compromised by the surgical procedure, and chemotherapy can pose additional risks for fertility, but fertility sparing strategies exist. These include preservation of the uterus only (FIGO IC G1/G2) or the healthy ovary as well, only in well-selected patients (FIGO IA G1/G2) after adequate staging and informed consent about associated risks. Exact staging, risk assessment and oncological monitoring until birth are required. Completion of surgery is recommended, after family planning is complete [15]. As a conclusion, the patients' selection is crucially dependent on the disease stage and the histologic data (tumor type and grade) which makes the complete surgical staging surgery and the careful pathological analysis (or review) of the tumur mandatory for this treatment.

Assisted reproductive technology (ART) provides alternatives for fertility preservation such as oocyte, embryo or ovarian tissue cryopreservation. This option has been reported in several case reports. Ito et al describe an ultrasound-guided laparotomic oocyte retrieval during surgery for fertility preservation in a case of tumor recurrence after a unilateral salpingo-oophorectomy [16] while Gallot et al describe a similar technique of frozen-thawed embryos obtained immediately before radical surgery for stage IIIa serous borderline ovarian tumor [17]. Ovum donation worked in Navot's [18] and Pouly's [19] cases, where ovarian-cancer patients whose uteruses were preserved, succeeded in conceiving and delivering healthy infants. Unfortunately, the stage and the grade of our patient's tumor could not allow us to take under consideration any of these methods.

However, our patient is now in excellent condition and has already completed a full circle of chemotherapy, 10 months after the initial intraoperative diagnosis. Being so uncommon, PFTC prognosis and follow up have never been fully estimated. Similarly to the ovarian cancer, no screening method is available to detect it and the lack of symptoms in the majority of the patients contributes to a belated diagnosis and treatment which usually has to deal with more advanced stages of malignancy. With the adoption of FIGO staging classifications and clinical practice guidelines in the management of gynecologic cancers the postoperative procedures of PFTC are basically of the same principle with those of the ovarian cancer. That implies that adjuvant chemotherapy is an option but not a mandatory one for early stage patients and postoperative CA125 in patients with preoperatively high levels, during the follow-up period, plays a leading role in the prediction of relapse and metastasis [20].

Few retrospective studies analyzing the follow up of PFTC patients have been conducted, and most of them mainly report that the overall survival rates of PFTC patients for 3-year and 5-year length lie between almost 80-87% and 55-65% accordingly, depending on the stage of the disease [21-23]. Advanced tumor stage, in particular the presence of lymph node metastases, is considered to be worsening the prognosis of patients with PFTC and a careful paraaortic and pelvic lymphadenectomy, is suggested to avoid underestimating the clinical stage [24,25].

We report this case for its rarity along with the typical clinical features and the young age of the patient. The aim is to raise suspicion of PFTC even in young women and point out the significance of a preoperative diagnosis, so that the right treatment can be succeeded. Multicenter experience should be used in order to clarify the approach in such uncommon cases that affect young, possibly nullipara women.

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