CASE REPORT

Serous surface papillary cystadenoma of fallopian tube clinically presenting with ectopic pregnancy

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ABSTRACT

Herein reported is the rare case of serous surface papillary cystadenoma (SSPC) of fallopian tube who presented with signs and symptoms of ectopic pregnancy. A 31-year-old woman presented with abdominal pain and elevation of serum human chorionic gonadotropin (hCG) (2,530 IU/L). Pelvic ultrasound examination suggested a rupture of right tubal pregnancy. Emergency laparotomy was performed, and right fallopian tube was resected. Pathologically, the fallopian tube was totally replaced by SSPC, but no evidence for pregnancy (chorionic villi, fetus, decidua, amnion, and cord) was seen. The SSPC was unilocular, and was surrounded by multiple papillary proliferations of serous cells with fibro-vascular cores, which were recognized both in the surface areas and in the intracystic inner aspects. The tumor cells of the SSPC were negative for hCG. After operation hCG returned to the normal range, and the patient was recovered with no complications. A rare case of SSPC of fallopian tube with pseudo-manifestations of ectopic pregnancy is reported. The raised serum hCG and its return to the normal ranges after the tube resection are completely unclear and enigma.

Key Words: Fallopian tube, Serous surface papillary neoplasm, Human chorionic gonadotropin, Ectopic pregnancy

1. INTRODUCTION

Neoplasms of the fallopian tube are rare. Serous tumors of the fallopian tube are exceedingly rare. Only a few cases of serous cystadenoma or cystadenofibroma of the fallopian tube have seen reported.[1–4] Serous surface papillary neoplasm (SSPN) of the ovary is rare. SSPN of the ovary shows papillary epithelial proliferations on the surface of ovary with or without cyst formations. In this report, benign SSPN with cystadenoma features with intracystic papillary epithelial proliferations are termed as "serous surface papillary cystadenoma" (SSPC). The author herein reports a very rare case of SSPC of the fallopian tube which presented with signs and symptoms of tubal pregnancy, but operative and pathological studies revealed an SSPC of fallopian tube but no evidence for pregnancy.

2. CASE REPORT

A 31-year-old woman presented with right abdominal pain. A prompt pelvic ultrasound study showed possible right tubal pregnancy, and laboratory data showed an increased serum human chorionic gonadotropin (hCG) (2,530 IU/L). The patient denied previous coitus during recent one year. Right tubal pregnancy was suspected and emergency laparotomy was carried out. The gynecologists found enlarged right fallopian tube, and resection of the tube was carried out. Grossly, the fallopian tube showed a mono-locular cystic tumor (5 cm $\times$ 4 cm $\times$ 4 cm) with many papillary structures. The tumor was located in fallopian tube, and fimbriae and

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meso-salpinx were not involved. The papillary structures were present apparently both in the surface areas and in the inner cystic spaces (see Figure 1). The cyst contained bloody fluid.

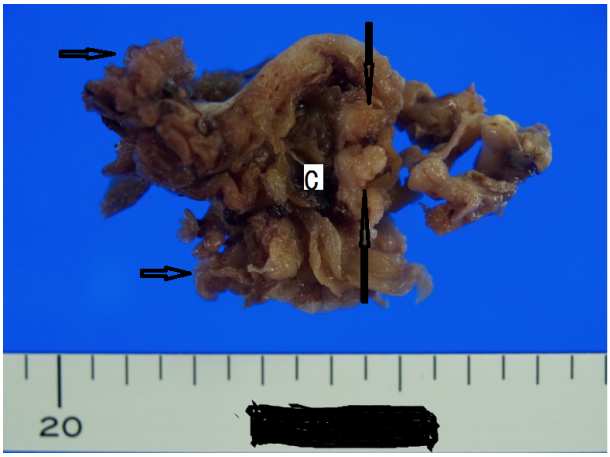

Figure 1. Gross features of the right fallopian tube
The tumor is monolocular, cystic (the cystic cavity is indicated by "C"), and measures 5 cm × 5 cm × 4 cm. Papillary proliferations is seen both on the outer surface (small arrows) and in the cystic inner spaces (large arrows).

Microscopically, the tumor showed cystic and papillary proliferations composed of one layer of columnar epithelium with luminal cilia that was arranged in a papillary pattern with fibrovascular cores (see Figure 2A). The epithelium showed mild atypia, but it was not enough to be diagnosed as borderline (see Figure 2B-C) because there were neither obvious features of nuclear atypia such as hyperchromasia, increased N/C ratio, prominent nucleoli, and irregular nuclear membranes nor overt features of structural atypia including nuclear stratification, loss of nuclear polarity, and nuclear protrusion into outer papilla. No invasive features were seen, nor lympho-vascular permeation. No fetal elements (chorionic villi, trophoblasts, amnion, and fetus) and decidual cells were recognized in multiple sectioning.

An Immunohistochemical analysis was carried out with the use of Dako Envision method.[5,6] The immunoreactivities of tumor cells were as follows: cytokeratin (CK) 7 +++ (see Figure 2D), CK20-, estrogen receptor (ER) +++ (see Figure 2E), progesterone receptor (PgR) +++, WT-1 +++ (see Figure 2F), Ki-67 ++ (labeling index = 30%) (see Figure 2G), p53 ++ (relatively diffuse) (see Figure 2H), CDX-2 -, TTF-1 -, and hCG -. SSPC of the fallopian tube was the final pathological diagnosis. The patient quickly recovered after the operation and is now free of disease. The hCG returned to normal range after the operation.

3. Discussion

Tumors of the fallopian tube is very rare. According to 2003 WHO blue book,[7] tumors of the fallopian tube and related ligaments are classified into the following categories: malignant epithelial tumors, borderline epithelial tumors, carcinoma in situ, benign epithelial tumors, tumor-like epithelial lesions, mixed epithelial-mesenchymal tumors, soft tissue tumors, mesothelial tumors, germ cell tumors, trophoblastic diseases, lymphoid/hematopoietic tumors, and secondary tumors. Each of these categories are sub-classified into several subtypes. Among these, tumors associated with the present SSPC are serous adenocarcinoma, borderline serous tumor, and serous cystadenoma. Since no comprehensive studies of these tumors are unavailable, the author herein describes relatively common tumors, i.e. primary fallopian tube carcinomas, because there are no significant data on benign and other tumors of the fallopian tube. According to Clayton et al.,[8] patients with fallopian tube carcinoma had a mean age of 70 years, and most presented with postmenopausal bleeding with a rare case having a second carcinoma. Some were nulliparous. None were diagnosed pre-operatively. All were treated surgically and received platinum-based chemotherapy. Although patients with tubal carcinoma had better outcomes than those with advanced ovarian carcinomas, the difference was not statistically significant. Accurate diagnosis and differentiation of tubal carcinomas from advanced ovarian carcinoma are important for monitoring trends in incidence, for better characterization of prognostic features and improved management. Kalampokas et al.[9] also reported similar but slightly different findings of fallopian tube carcinomas. As is well known, epithelium of fallopian tube and mesothelium particularly around fimbria play an important role in the development of ovarian serous cystadenoma and cystadenocarcinoma.[10,11] In this respect, it is very interesting that primary carcinoma indistinguishable from serous tumors of ovary can arise from mesothelium.[12]

The present case clinically presented with signs and symptom of right tubal pregnancy. The data of elevated serum hCG supported the diagnosis of tubal pregnancy. In the current case, immunoreactivity of hCG was negative in the tumor of the fallopian tube. However, no pathological findings of pregnancy were noted; neither trophoblasts, chorionic villi, decidua, fetus, amnion, nor cord were noted. Instead, the fallopian tube showed an SSPC. The reason and mechanism of the raised serum hCG is only speculative or entirely enigma. hCG is produced mainly in trophoblasts during pregnancy or rarely in tissues of aberrant production such as ectopic-hCG-producing tumors. The explanation of elevated serum hCG and its decline into normal level after the resection of the tubal SSPC in the present case is totally enigmatic; how-

ever several unlikely explanations are present. First, the first hCG value is artificial error. Second, the tubal SSPC actually produced hCG but it was not detected by the immunohistochemistry, Thirdly, complete rupture of left tubal pregnancy was present without leaving any fetal and mother elements in the tube, and they subside and become necrotic, thus causing decreased hCG.

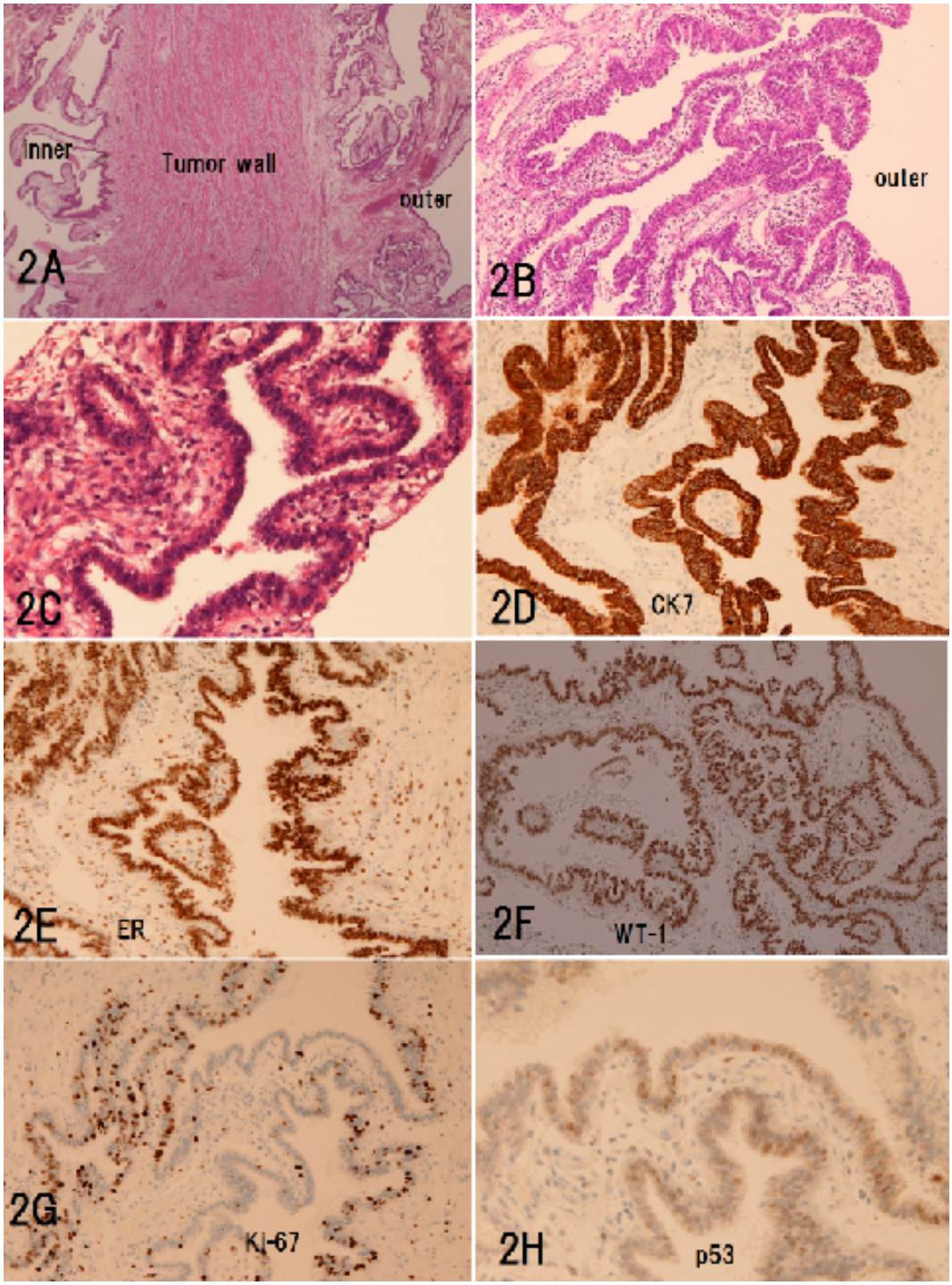

Figure 2. Histological (A-C) and Immunohistochemical (D-H) features of the fallopian tube tumor

A: Low power view of the tumor shows papillary proliferation of one-layered columnar epithelium with fibrovascular cores both in the outer and inner surface. The central fibrous tissue is a tumor walls. HE, ×20. B,C: Moderate (B) and high power (C) views show atypia. C: HE, ×150. D: HE, ×280. C-F: The tumor cells are positive for cytokeratin 7 (D), estrogen receptor (E), MT-1 (F), Ki-67 antigen (labeling index = 30%) (G), and p53 protein (H). ×200.

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The present SSPC was also characterized by mono-locular cystic tumor with intracystic papillary proliferation of tumor cells in addition the outer surface papillary proliferation; thus the author has termed SSPC. The present case is the first case of SSPC of fallopian tube. The present tumor of right fallopian tube is not typical for serous tumors; the present tumor showed marked papillary proliferations in both inner cystic areas and outer surfaces; these inward and outward papillary proliferations with cyst formations are extremely rare even in ovarian tumors. Although serous tumor as in the present case is extremely rare in fallopian tube, serous surface papillary neoplasm, i.e. SSPC of fallopian tube as in the present case, has not been reported. Therefore, the present tumor in fallopian tube location seems valuable.

The Immunohistochemical study is consistent with primary fallopian tube tumor. Positive WT-1 suggest tumor of serous papillary group. Although the present tumor seemed benign, the positive p53 can detect wild-type and mutant p53; the former reflect rapid fixation and the latter p53 mutations.[13] It is because half time period of protein degradation is far quicker in wild-type p53 than is mutant p53. However, p53 expression of relatively diffuse pattern, as seen in the present case) in pathologic tissue sections can be evaluated to be mutant p53, suggesting that the present tumor might show p53 gene mutation. The high Ki-67 labeling index (LI) was 30% in the present study. In general, Ki-67 LI cut off value is different among tumors, the cut-off value is 10% in many tumors. The high Ki-67 LI in the current tumor show rapid turnover of tumor cells and high proliferative fractions. It also suspects that the tumor is true neoplasm. However, malignant potential cannot be evaluated only in Ki-67 LI. Other factors should be considered for the biologic behaviors.

Neoplasms of fallopian tube have been shown to play a pivotal role in the carcinogenesis of ovarian surface epithelial tumors. Serous tubal intraepithelial carcinoma (STIC) is suspected to become serous neoplasms of ovary and peritoneum.[10, 11] Although it is not obvious intraepithelial carcinoma, it is conceivable that the present tumor may give rise to serous tumors of ovary or peritoneum if the tumor was not resected.

CONFLICTS OF INTEREST DISCLOSURE

The author declares no conflict of interest.

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