Primary fallopian tube carcinoma co-existing with pregnancy: A case report

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Primary fallopian tube carcinoma is the rarest of all gynecologic cancers and usually occurs in postmenopausal women. Thus, its presence in pregnancy is extremely remote. A 31-year-old term primigravida with a history of right lower quadrant pain, diagnosed with dermoid focus on imaging was admitted due to ruptured membranes and delivered a live and healthy infant. Intraoperatively, the right fimbria was dilated and fimbriectomy was performed which revealed papillary serous cyst adenocarcinoma. There was no residual disease on re-exploration. She was classified as International Federation of Gynecologists and Obstetricians stage IA. No adjuvant chemotherapy was given. The incidence of primary fallopian tube carcinoma co-existing with pregnancy is extremely low. This article review is possibly the first reported case of primary fallopian tube carcinoma co-existing with pregnancy, locally.

Key words: Term pregnancy; Papillary serous cyst adenocarcinoma

Introduction

In 1847, Renaud first described fallopian tube malignancy. However, Orthmann submitted the first genuine case report in 1888.

Primary fallopian tube carcinoma (PFTC) is the rarest of all female genital tract malignancies accounting for 0.1-1.8% of all gynecologic cancers.1 In the United States, the average annual incidence is 3.6/1,000,000 women per year.2 At the Philippine General Hospital, from 1972 to 1986, the 8 reported cases comprised a very small percentage of the total gynecologic malignancies seen during that period.

More than 60% of cases occur in postmenopausal women, at a mean age of 55 years.1 Thus, the possibility of its presence in pregnancy is quite remote.

To date, there are only two reported case reports of fallopian tube carcinoma co-existing with pregnancy. A search of local literature yielded no such case. This is possibly the first reported case locally.

Case Report

A 31-year-old, primigravida, on her 39 2/7 weeks gestational age was admitted due to ruptured membranes. She had been experiencing an intermittent, colicky, non-radiating right lower quadrant pain for three years and upon previous consultation a transvaginal ultrasound revealed a 4 cm dermoid focus in the right ovary. No other imaging studies were requested. She was advised close surveillance with serial ultrasound scans but without compliance due to financial constraints. With the persistence of the symptoms, she was referred to the department of obstetrics and gynecology for evaluation. A repeat transvaginal ultrasound still showed the previously seen dermoid focus in the right ovary of almost the same dimensions.

One year prior to admission, the patient got pregnant. During a first trimester ultrasound “a single live intrauterine...
pregnancy a thin-walled unilocular cystic structure within the right ovary containing linear densities measuring 2.5×2.4 cm, consider a dermoid focus" (Fig. 1) were noted. Eight weeks later, surveillance of the mass showed a slight increase in the size (Fig. 2). Congenital anomalies scan at 20 weeks gestational age showed a normal fetus in breech presentation. The right ovarian cyst was noted to still have the same dimensions and characteristics.

There was a recurrence of right lower quadrant pain at 24 weeks gestational age prompting admission, however the right ovarian mass was not visualized on pelvic ultrasound. Her complete blood count and urinalysis were normal. She was given analgesics and was discharged improved on the 2nd hospital day.

Succeeding prenatal consultations were unremarkable. Her medical, personal, social, sexual and menstrual histories were unremarkable. Family history is positive for breast cancer in a cousin and an aunt on her paternal side.

Vital signs were stable on physical examination. The fundic height was 35 cm with an estimated fetal weight of 3,000 grams. Fetal heart tone of 157 beats per minute was best heard at the left lower quadrant. Speculum examination revealed minimal pooling of clear amniotic fluid. On internal examination, the cervix was 1 cm dilated with beginning effacement, ruptured membranes, fetus in cephalic presentation, station -3.

Our admitting diagnosis was “Pregnancy uterine, 39 2/7 weeks gestational age, cephalic, in early labor, G1P0; premature rupture of membranes (PROM) for 30 minutes; ovarian new growth, right probably benign.”

After 14 hours of augmentation of labor, cervix remained 1 cm dilated. She thus underwent a primary cesarean section for PROM of 18 hours and delivered a live and healthy infant.
Intraoperatively, the fimbrial end of the right fallopian tube was bulbously dilated to 4×4×3 cm (Fig. 3). The left fallopian tube and bilateral ovaries were grossly normal. During this time, malignancy was not suspected, so Frozen section was not contemplated. A right fimbriectomy was performed. Cut section of the mass revealed creamy fluid and a tan-yellow soft friable nodule measuring 2×2×1.5 cm (Fig. 4). The capsule was thin and smooth with no excrescences noted.

The final histopathology report was “Primary papillary serous cystadenocarcinoma of the fimbria, right”. Microscopically, the tubal mucosa at the margins of resection had normal papillary configuration (Fig. 5). Going towards the tumor, the lumen of the tube becomes dilated resulting in the flattening of the papillary folds (Fig. 6). The area of transition between the non-neoplastic tubal mucosa and the tumor is shown with the benign mucosa lined by a single layer of flattened epithelium, which starts to form complex branching papillary structures in the area where it becomes malignant (Fig. 7). The tumor is confined to the mucosal surface with no evidence of invasion into the muscular wall.

Due to inadequate staging on her previous surgery, a re-exploration postpartum was contemplated and proposed to the patient. Serum CA-125 showed a normal result. To avoid technical difficulties, which may be encountered in an immediate postpartum uterus, the patient underwent a staging laparotomy seven weeks following delivery.

Intraoperatively, there was no ascitic fluid. The uterus, left ovary and its corresponding fallopian tube were grossly normal. The remaining right fallopian tube segment and right ovary were removed. Frozen section revealed benign ovarian tissue and right fallopian tube segment. Other procedures performed were peritoneal fluid cytology, infracolic omentectomy, bilateral lymph node dissection, and biopsy of right and left paracolic gutters, left paracolic gutter adhesion, paravesical adhesion and anterior peritoneal wall.

There was no malignancy observed on final histopathology report.

Our final diagnosis was primary papillary serous cystadenocarcinoma of the right fallopian tube, stage IA with plan of close surveillance using CA-125 and regular gynecologic examinations every three months.
Discussion

PFTC is the rarest of all female genital tract malignancies, accounting to 0.1-1.8% of all gynecologic cancers.\(^1\) It has an average annual incidence rate of 3.6 per million women per year in the United States.\(^2\) From 1972 to 1986, 8 reported cases comprised a very small percentage of this gynecologic malignancy at the Philippine General Hospital. Since its first recorded case in 1847, fewer than 2500 cases have been reported in different journals worldwide, most of them being case reports.

More than 60% of PFTC cases occur in postmenopausal women, at a mean age of 55 years.\(^1\) Thus; the possibility of its presence in pregnancy is quite remote.

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Adolph et al.\(^3\) in Canada reported the first case in 2001. The patient was diagnosed to have fallopian tube carcinoma Stage IIB. She underwent surgery and was given adjuvant chemotherapy. A year after chemotherapy, she got pregnant and was diagnosed to have recurrent fallopian tube carcinoma. She received chemotherapy and delivered by cesarean section to a healthy baby.

Batra et al.\(^4\) in the United Kingdom reported the second case in 2006. The patient had an adnexal mass, which grew rapidly at 9 weeks gestational age. She underwent salpingo-oophorectomy, which revealed serous papillary carcinoma of the fallopian tube. No adjuvant chemotherapy was administered. She delivered spontaneously to a healthy term infant.

Our case had intermittent right lower quadrant pain and a non-palpable adnexal mass, which on ultrasound did not increase in size prior to and during her entire pregnancy. Due to an obstetric indication, she underwent cesarean section and was later diagnosed with PFTC.

There is no available theory on how PFTC develops during pregnancy. In our case, an adnexal mass was seen on ultrasound even before she got pregnant, thus, we can only surmise that the malignancy could have been developed prior to or during her pregnancy.

Differential diagnoses must include benign fallopian tube pathologies such as tuberculous salpingitis or hydrosalpinx, ovarian tumor of low malignant potential, and malignant ovarian epithelial tumors. We initially did not suspect a malignant process occurring in the fallopian tube during the cesarean section. The young age of the patient, the rarity of occurrence of fallopian tube carcinoma in pregnancy, and the benign features of the mass after dissection swayed us to believe that we were dealing with a benign or infectious condition. Though it is easy to say that in retrospect, a high index of suspicion should be observed, given the same situation, a diagnosis of fallopian tube carcinoma will still be one of the least considerations.

PFTC co-existence with pregnancy poses a challenge in diagnosing Fallopian Tube Carcinoma. In the presence of an adnexal mass by palpation or sonography during pregnancy, an ovarian pathology will almost always be the primary consideration. Adnexal masses less than 5 cm seen on routine scans with benign features may only warrant close observation. Surgery is indicated in the following situations: if the mass is greater than 6-8 cm, it is enlarging during gestation, it has features suggestive of malignancy, or it is producing acute symptoms akin to an acute abdomen. This is what happened to our patient. An adnexal mass less than 5 cm was only monitored since we believed that it was a benign ovarian tumor.

The use of tumor markers has definite limitations in pregnancy. Alterations associated with the gravid state make the interpretation of tumor markers difficult. The clinical significance of an abnormally high CA-125 is, therefore, reduced since this marker is normally elevated in pregnant patients.\(^5\) Our patient had a normal level of CA-125 after her delivery and removal of tumor.

Pregnancy affords an opportunity for the detection of a tumor, since the patient usually consults for prenatal check-up. Our patient had regular prenatal check-ups and the adnexal mass was monitored using sonography.

Ultimately, a benign etiology cannot be completely diagnosed with certainty by imaging. About 10% to 20% of adnexal masses will remain indeterminate after ultrasound evaluation.\(^6\) Computed tomography (CT) and magnetic resonance imaging (MRI) may provide additional information on tissue characterization and origin of the mass. However, a CT scan is contraindicated in pregnancy due to the effects of ionizing radiation on the developing fetus. Other ancillary procedures were not done such as color Doppler sonography because malignancy was not suspected.

To rectify the inadequacy of the initial surgical procedure, a second laparotomy with appropriate staging procedures were performed. Confirmation of early disease by clinical evaluation and frozen section analysis thus warranted conservative surgery.
The close proximity of the fallopian tubes to the ovaries and to the uterus makes it difficult to identify the true primary. Hu et al. developed a pathologic criteria that was later modified by Sedlis that is currently being used in the diagnosis of PFTC:

1. The tumor clearly arises from the endosalpinx,
2. The histology represents the epithelium of tubal mucosa,
3. Transition from benign to malignant epithelium is evident (Fig. 4),
4. The ovary and endometrium are either normal or there is a tumor smaller than the one in the tube.

Our patient fulfilled all 4 criteria, thereby making the tumor within the fallopian tube a true primary.

The management of fallopian tube carcinoma is principally the same, as that for ovarian carcinoma. Patients with apparently early stage fallopian tube carcinoma should undergo total abdominal hysterectomy with bilateral salpingo-oophorectomy, peritoneal fluid cytology, infracolic omentectomy, multiple peritoneal biopsies, and pelvic and para-aortic lymph node dissection. Surgical conservation to maintain reproductive function in young women follows the same criteria as in ovarian carcinoma. In young women desirous of future pregnancies like in our case, with unilateral tumors confined to the tube, a unilateral salpingectomy or salpingooophorectomy, with omentectomy, and lymphadenectomy may be performed.

Our case caused a diagnostic and management dilemma. Extensive counseling was done with the patient and her husband whereby the surgical staging was explained with the probability of a total hysterectomy and contralateral salpingooophorectomy should frozen section of the ipsilateral ovary and tubal segment reveal metastases.

However, it was also emphasized that a more conservative surgical procedure is the priority, considering her age and desire to maintain her reproductive potential. It was also explained to her that if there were an evidence of metastasis, adjuvant chemotherapy would be indicated.

Close surveillance for this patient is important, as she has had conservative surgery. Physical and pelvic examinations, CA-125 determination every three months, and pelvic ultrasound every 6-12 months should suffice as monitoring tools to detect possible disease recurrence.

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References