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RUPTURED TUBAL ECTOPIC CHORIOCARCINOMA – A RARE CASE

KEY WORDS: Gestational Choriocarcinoma, Ectopic Pregnancy, Beta-hcg.

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ABSTRACT

Gestational choriocarcinoma presenting as ectopic pregnancy is a very rare event with incidence of 1.5 in every 10,00,000 pregnancies. Here, we are presenting a case of 24yr old woman with obstetric formula G2P1L1 presented with acute abdominal pain and bleeding per vaginum. On TVS, she was diagnosed as ruptured right tubal ectopic pregnancy and she had undergone emergency laparotomy and right salpingectomy. Histopathological assessment of the specimen revealed choriocarcinoma stage 1 with low risk disease. Her initial beta-hcg levels were 2,60,000mIU/ml. Single agent chemotherapy with Methotrexate was administered. Follow up beta hcg gradually declined over a period of 8 weeks. With this case report we aimed to implicate that choriocarcinoma, although a rare entity should be kept in mind and the as it has high risk of metastasis, all ectopic pregnancies must undergo histopathological examination.

INTRODUCTION

Gestational trophoblastic diseases were first described in 400 BC by Hippocrates(Seckl et al., 2010). GTN primarily encompasses pathologic entities characterized by aggressive invasion of the endometrium and myometrium by trophoblastic tissues. Histological categories include tumors such as invasive mole, gestational choriocarcinoma, placental site trophoblastic tumor.

Choriocarcinoma, a subtype of gestational trophoblastic disease, (Bruce & Sorosky, 2019) is one of the most malignant tumour of genital tract. Choriocarcinoma usually follows a pregnancy, 50% of the cases follow evacuation of hydatidiform mole, 25% follow an abortion, 20% follow a full term pregnancy and 5% follow an extrauterine pregnancy. Choriocarcinoma develops from an abnormal trophoblastic population undergoing hyperplasia and anaplasia, most frequently following a molar pregnancy(John R. Lurain, 2010). Non-gestational choriocarcinoma arises from pluripotent germ cells(Stockton et al., 2018) and is seen both in males and females, in the gonads, or midline structures with pluripotent germ cells(Stockton et al., 2018).

CASE REPORT

A 24-year-old woman (gravida 2 para 1 and live 1) was admitted to the emergency room due to amenorrhea of 8 weeks with severe abdominal pain and fatigue. she had regular menstrual cycles, previously delivered by elective lower segment caesarean section in view of cephalo pelvic disproportion. Physical examination revealed a right adnexal tenderness. She had hypotension (blood pressure: 80/50 mmHg) and tachycardia (pulse: 105 beats/min). On laboratory examination, haemoglobin: 7.0 g/dl, haematocrit: 25%, leukocyte count: 16100/mm³ and β-Hcg: 2,60,000 mIU/ml were detected. The patient underwent transvaginal ultrasonography which showed a normal uterine cavity with no signs of intrauterine gestational sac or embryo. On the other hand, an ectopic mass of 2*3cm was visualized in the right adnexal region and there was a large amount of free fluid in the pelvis. Due to the haemodynamic instability of the patient and clinical suspicion of ruptured tubal pregnancy, an emergency laparotomy was made.

On exploration, a ruptured and actively bleeding ectopic mass with size of 5*4 cm was noted in the ampullary region of right fallopian tube extending on to cornual end of uterus[Figure 1,2] as well as abundant hemoperitoneum with 500 ml of blood and right salpingectomy was performed.

5*4cms cyst was found in left ovary. Histopathological examination revealed the final diagnosis as primary tubal choriocarcinoma of 4 cm in size which involved the whole layer of tubal wall and ruptured into pelvic cavity at the ampullary region. The tumour displayed the typical biphasic feature of choriocarcinoma in which atypical cytotrophoblasts and syncytiotrophoblasts are mixed with degenerated red blood cells[Figure3]. Extensive vascular invasion within the layer of tubal wall was observed. No chorionic villi are identified. Beta- HCG levels were 2,60,000mIU/ml. Chest X-Ray demonstrated no metastases. Thyroid profile was within normal limits. MRI Brain was normal. WHO Prognostic score was 5 demonstrating that the patient has FIGO Stage1 disease with low risk and good prognostic factors. Medical oncologist opinion was taken and started single agent chemotherapy with Methotrexate and folic acid regime in order to prevent any recurrences. Three courses of methotrexate was administered to the patient alternating with folic acid.

The patient was put on a follow up of weekly β- Hcg measurements until 8 weeks when beta-Hcg came negative. The β- Hcg level declined to 89,040mIU/ml at the end of the first week of chemotherapy. The negative β- Hcg level was achieved at the end of the 8 weeks after the surgery. The patient responded well to the chemotherapy and no side effects were observed. The patient was followed up with every 2 weekly beta-Hcg for 3 months and monthly Beta-Hcg for six months and was disease free till that period. The patient was counselled to avoid pregnancy for two years.



Figure 1

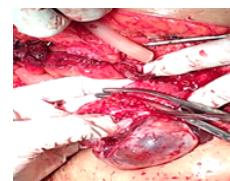


Figure 2

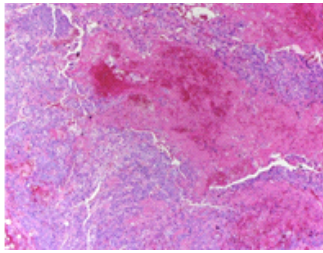


Figure 3

DISCUSSION

Gestational choriocarcinoma is a malignant type of the gestational trophoblastic disease which usually originates from uterine cavity. It rarely affects fallopian tubes, cervix, vagina, ovaries or other pelvic organs(Chen et al., 2004; Mitrovic et al., 2014). The extra uterine choriocarcinoma fortunately, is extremely rare with an incidence of 0.76-0.4% of all ectopic pregnancies(Nayama et al., 2007) . It is reported in a study that its incidence is one in 5333 tubal pregnancies and one in 1.6 million normal intrauterine pregnancies(Rettenmaier et al., 2013). However, tubal choriocarcinoma associated with ectopic pregnancy has a very aggressive course, attributed to the relatively thinner muscular layer of fallopian tubes and early rupture of tubal wall into the pelvic cavity. Horn et al., conducted a study and reported that 75% of cases have metastasis during the initial diagnosis(Horn et al., 1994).The clinical findings are quite similar to those of an ectopic pregnancy including amenorrhea, increased serum β - Hcg levels, vaginal bleeding, and pelvic pain(J. R. Lurain et al., 1986). Therefore, the histopathological verification of an ectopic pregnancy is essential for the confirmation of the diagnosis and the exclusion of any other tubal pathology(Horn et al., 1994). In this case, an initial diagnosis of ruptured ectopic pregnancy was made and the final diagnosis of choriocarcinoma was affirmed by postoperative histopathological examination.

The diagnosis of primary tubal choriocarcinoma is challenging because the related clinical symptoms and findings are often non-specific and can simulate the other gynaecologic diseases such as ectopic pregnancy, ovarian cyst and tubo-ovarian abscess(Muto et al., 1991). In this case, tubal choriocarcinoma was incidentally diagnosed after the postoperative. The most typical sonographic feature of an intrauterine choriocarcinoma is a large echogenic irregular mass with hyper vascularization which occupies the uterine cavity. However, no specific imaging findings have been defined for extra-uterine choriocarcinoma. Overexpression of p53 and MDM2 have been demonstrated in choriocarcinoma, with no evidence of somatic mutation. Other genes implicated with either overexpression or down-regulation via hyper-methylation include NECC1, epidermal growth factor receptor, DOC-2/hDab2, Ras GTPase-activating protein, E-cadherin, HIC-1, p16, TIMP3. HLA-G is demonstrated at very high levels in choriocarcinoma(Shih, 2007).

The typical histologic features of choriocarcinoma consist of the columns of trophoblastic cells without any villous structures and the invasion of vessels and muscular tissue with extensive necrosis and haemorrhage(Muto et al., 1991). Choriocarcinoma has high risk of metastasis and metastasize into the lungs, brain, liver, and even very rarely into the fetus(Nayama et al., 2007). The management in women who are incidentally diagnosed with tubal choriocarcinoma after undergoing salpingectomy for ectopic pregnancy is challenging. Gestational choriocarcinoma is a highly responsive tumour to chemotherapy. It has a good prognosis even in the advanced stage. Low risk gestational trophoblastic diseases have 100% cure rate with chemotherapy and high risk gestational trophoblastic diseases have 90% cure rate.

The women who are treated for extra-uterine choriocarcinoma should receive effective contraception for at least two years after the completion of their treatment. Monitoring β -Hcg is the most useful diagnostic tool in case of tubal choriocarcinoma .After complete remission, the patient should be followed up for the rest of her life because risk of recurrence is there several years after the initial treatment.. With the advent of efficient adjuvant chemotherapy, the fertility can be preserved successfully in women with extra-uterine choriocarcinoma. However, an increase in serum -Hcg concentration should be interpreted carefully during the long-term follow up as a new pregnancy can be easily confused with a probable recurrence.

CONCLUSION

In the summary, extra uterine choriocarcinoma is a rare entity with aggressive course. Choriocarcinoma is highly sensitive to chemotherapy. Depending on the FIGO stage and WHO prognostic scoring, management with chemotherapy is recommended. Every ectopic pregnancy specimen should be examined histologically to rule out other pathologies. Monitoring serial β -Hcg concentration will be crucial to diagnose persistent trophoblastic disease and recurrence of the disease.

CONFLICTS OF INTEREST

There are no conflicts of interest.

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