DOI: 10.7860/JCDR/2015/15828.6534

Case Report

Obstetrics and Gynaecology Section

Primary Tubal Choriocarcinoma Presented as Ruptured Ectopic Pregnancy

ERBIL KARAMAN[1], ORKUN ÇETIN[2], ALI KOLUSARI[3], IRFAN BAYRAM[4]

ABSTRACT

The primary tubal choriocarcinoma associated with ectopic pregnancy is very rare. A 31-year-old woman was admitted to the emergency room due to amenorrhea, left lower abdominal pain and fatigue. Her β-Hcg level was 29251.4 mIU/ml and transvaginal ultrasund revealed a 24x21 mm of left tubal ectopic pregancy mass with large amount of free fluid in pelvic cavity. The patient was diagnosed with ruptured tubal ectopic pregnancy and thus, she underwent laparotomy with left total salpingectomy. The pathological assessment was reported as primary tubal choriocarcinoma with the involvement of whole tubal layer. The patient was defined to have stage I choriocarcinoma with good prognostic factors and methotrexate monotherapy was administered. Serum β- Hcg levels of the patient gradually declined and eventually became negative at the first month of the treatment. With this case report we aimed to implicate that when diagnosing the ectopic pregnancy, even very rare, the tubal choriocarcinoma should be kept in mind.

Keywords: Human chorionic gonadotropin, Tubal rupture, Tubal pregnancy

CASE REPORT

A 31-year-old white-turkish woman (gravida 2 para 2) was admitted to the emergency room due to amenorrhea of 7 weeks, left lower abdominal pain and fatigue. It was learnt that she had regular menstrual cycles, two vaginal deliveries and no previous gynecologic operations. Her physical examination revealed a sharp left adnexal pain with rebound tenderness. She had hypotension (systolic/diastolic blood pressure: 80/50 mmHg) and tachycardia (pulse: 105 beats/min). On laboratory examination, haemoglobin: 7.4 g/dl, haematocrit: 24.1%, leukocyte count: 15100/mm^3 and β-Hcg: 29251.4 mIU/ml were detected. The patient underwent transvaginal ultrasonography which showed a normal uterine cavity with endometrial thickness of 8 mm and no signs of intrauterine gestational sac or embryo. On the other hand, an ectopic mass of 24x21 mm was visualized in the left 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.

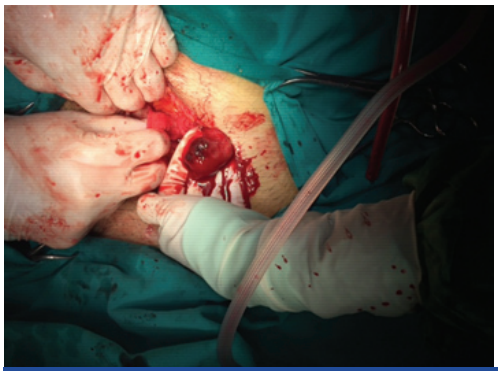
On exploration, a ruptured and actively bleeding ectopic mass with size of 4x3 cm was noted on the ampulla of left fallopian tube as well as abundant haemoperitoneum with 600 cc of blood. A complete left salpingectomy was performed [Table/Fig-1,2]. Histopathological examination revealed the final diagnosis as primary tubal choriocarcinoma of 2 cm in size which involved the whole layer of tubal wall and ruptured into pelvic cavity at the ampulla region. The tumour displayed the typical biphasic feature of choriocarcinoma in which atypical cytotrophoblasts and syncytiotrophoblasts are mixed [Table/Fig-3]. Extensive vascular invasion within the layer of tubal wall was observed.

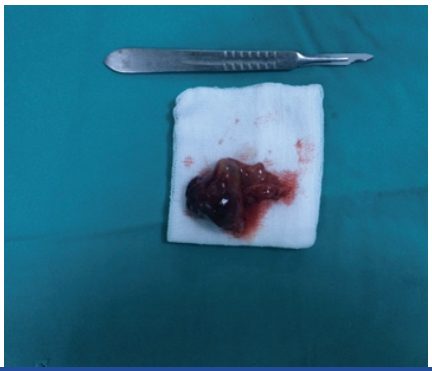
Immunohistochemical analysis was positive for β- Hcg and negative for human placental lactogen and P 63 whereas, Ki 67 proliferation index was 50%. Based on these findings, the patient was transferred to the Department of Medical Oncology unit so that comprehensive radiologic and laboratory examinations could be done. Computed tomography scans of the abdomen and thorax were demonstrated no metastases. This workup indicated that the patient had FIGO stage I disease with good prognostic factors. In order to prevent any recurrences, adjuvant monochemotherapy with three courses of methotrexate was administered to the patient.

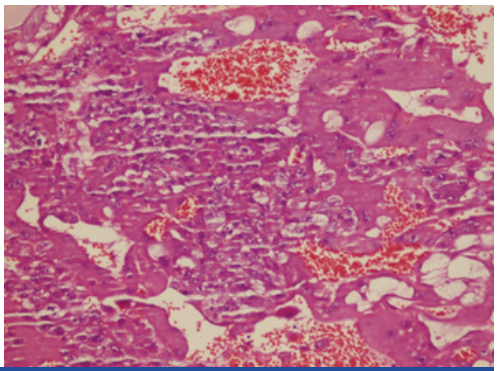
The patient was put on a follow up of weekly β- Hcg measurements during the first month and then monthly measurements. The β- Hcg level declined to 1488.75 mIU/ml at the end of the first week after surgery and just before starting the chemotherapy. The negative β- Hcg level was achieved at the end of the first month after the operation. The patient responded well to the chemotherapy and no side effects were observed. The patient was followed up for six months and was disease free till that period. The patient was counselled to avoid pregnancy for two years.

DISCUSSION

Gestational choriocarcinoma is a malignant type of the gestational trophoblastic disease which usually originates from uterine cavity.







[Table/Fig-1]: Intraoperative view of left tubal mass measuring 4x3 cm between two fingers of the operator **[Table/Fig-2]:** Postoperative macroscopic view of left salpingectomy material **[Table/Fig-3]:** Histopathologic view of tubal choriocarcinoma presenting the biphasic feature of mixed atypical cytotrophoblasts and syncytiotrophoblasts

This neoplasia also may rarely affect fallopian tubes, ovaries, cervix, vagina or other pelvic organs [1,2]. Gestational trophoblastic diseases comprise a variety of disorders in which choriocarcinoma is the most malignant form and usually develops after a molar pregnancy and relatively to a lesser extent after a term pregnancy, abortion or ectopic pregnancy [3]. The choriocarcinoma associated with ectopic pregnancy, fortunately, is extremely rare with an incidence of 0.76-0.4% of all ectopic pregnancies but it is generally very aggressive [4]. It is reported in a study that its incidence is one in 5333 tubal pregnancies and one in 1.6 million normal intrauterine pregnancies [5]. Gestational choriocarcinoma is a highly responsive tumour to chemotherapy and has a good prognosis even in the advanced stage. However, tubal choriocarcinoma associated with ectopic pregnancy has a very aggressive course. This discrepancy may be attributed to the relatively thinner muscular layer of fallopian tubes and early rupture of tubal wall into the pelvic cavity. Therefore, tubal choriocarcinoma is mostly metastatic when diagnosed. A study by Horn et al., reported that 75% of cases have metastasis during the initial diagnosis [6]. The clinical findings are quite similar to those of an ectopic pregnancy including amenorrhea, increased serum β- Hcg levels, vaginal bleeding, and pelvic pain [7]. Therefore, the histopathological verification of an ectopic pregnancy is essential for the confirmation of the diagnosis and the exclusion of any other tubal pathology [6]. In this case, an initial diagnosis of ectopic pregnancy was made on admission and the final diagnosis of choriocarcinoma was affirmed by postoperative histologic 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 gynecologic diseases such as ovarian cyst, tuba-ovarian abscess and ectopic pregnancy [3]. For instance, Munkdur et al., reported a case of tubal choriocarcinoma presenting as an ovarian tumour which was treated by adenexectomy and partial infracolic omentectomy followed by multiple agent chemotherapy [8]. In this case, tubal choriocarcinoma was incidentally diagnosed after the postoperative pathological examination of the salpingectomy specimen which was obtained due to a suspicion of ruptured tubal pregnancy. 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. It has been reported that trophoblastic tumours, particularly choriocarcinoma, produce up to hundred times the amount of β- Hcg which is secreted by a normal pregnancy. This is also valid for our case as the initial β-Hcg level measured up to 29251.4. Therefore, the measurement and monitorization of β- Hcg concentrations are the most useful markers for the detection of any recurrences after the treatment.

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 [3]. Therefore, due to the great tendency for vascular invasion, choriocarcinoma can metastasize into the lungs, brain, liver, and even very rarely into the fetus [4]. Because of this highly metastatic potential, chemotherapy is essential and proves to be very effective in trophoblastic tumours. However, adjuvant chemotherapy is recommended to the patients with extrauterine choriocarcinoma and Mitrovic et al., reported a case of cervical gestational choriocarcinoma in which hysterectomy was the only option due to the severe uterine bleeding [1,9]. There is a challenging issue regarding the women who are incidentally diagnosed with tubal choriocarcinoma or persistent trophoblastic disease after undergoing salpingectomy for ectopic pregnancy. To the best of our knowledge, the literature shows that a woman who has incidentally diagnosed tubal choriocarcinoma achieves complete remission with chemotherapy even if there were metastases [10].

The women who are treated for extra-uterine choriocarcinoma should receive effective contraception for at least one year after the completion of their treatment. After complete remission, the patient should be followed for the rest of her life because re-occurrence may occur several years after the initial treatment. The follow up marker is β- Hcg and the monitorization of β- Hcg is the most useful diagnostic tool in case of tubal choriocarcinoma. Thanks to the efficieny of adjuvant chemotherapy, it is well known that fertility can be preserved successfully in women with extra-uterine choriocarcinoma. However, an increase in serum β-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

Primary tubal choriocarcinoma can be seen rarely with ectopic pregnancy. The counseling about possibility of tubal choriocarcinoma should be given to the patient when managed for ectopic pregnancy. When treating the ectopic pregnancy, the important role of pathologic examination associated with monitoring serial β-Hcg concentration will be crucial not only to diagnose persistant trophoblastic disease but also to avoid a surprise like choriocarcinoma.

Patient consent: The patient gave consent for the case and images

REFERENCES

[1] Chen MJ, Yang JH, Lin MC, Ho HN, Yang YS. An unusual gestational choriocarcinoma occurring primarily on the surface of a subserous leiomyoma. *BJOG*. 2004;111:188-90.
[2] Mitrovic SLj, Arsenijevic PS, Kljakic D, Djuric JM, Milosavljevic MZ, Protrka ZM, et al. Gestational choriocarcinoma of the cervix. *Arch Iran Med*. 2014;17(11):783-85.
[3] Muto MG, Lage JM, Berkowitz RS, Goldstein DP, Bernstein MR. Gestational trophoblastic disease of the fallopian tube. *J Reprod Med*. 1991;36(1):57-60.
[4] Nayama M, Lucot JP, Boukerrou M, Collinet P, Cosson M, Vinatier D. [Tubal choriocarcinoma: a case report and review of the literature]. *J Gynecol Obstet Biol Reprod (Paris)*. 2007;36(1):83-86.
[5] Rettenmaier MA, Khan HJ, Epstein HD, Nguyen D, Abaid LN, Goldstein BH. Gestational choriocarcinoma in the fallopian tube. *J Obstet Gynaecol*. 2013;33(8):912-14.
[6] Horn LC, Bilek K, Pretzsch G, Bajer D. Choriocarcinoma in extrauterine tubal pregnancy. *Geburtshilfe Frauenheilkd*. 1994;54:375-77.
[7] Lurain JR, Sand PK, Brewer JI. Choriocarcinoma associated with ectopic pregnancy. *Obstet Gynecol*. 1986;68(2):286-87.
[8] Mundkur A, Rai L, Hebbar S, Guruvare S, Adiga P. Fallopian tube choriocarcinoma presenting as ovarian tumour: a case report. *J Clin Diagn Res*. 2015;9(1):QD01-02.
[9] Mitrovic SLj, Arsenijevic PS, Kljakic D, Djuric JM, Milosavljevic MZ, Protrka ZM, et al. Gestational choriocarcinoma of the cervix. *Arch Iran Med*. 2014;17(11):783-85.
[10] Salleh S, Arthrur I. Persistent peritoneal trophoblastic implantation followingsalpingotomy, salpingectomy and methotrexate for ectopic pregnancy. *GynecolSurg*. 2005;2(3):195–96.

PARTICULARS OF CONTRIBUTORS:
1. Assistant Professor, Department of Obstetric and Gynecology, School of Medicine, Yuzuncu Yil University, Van, Turkey.
2. Medical Doctor, Department of Obstetric and Gynecology, School of Medicine, Yuzuncu Yil University, Van, Turkey.
3. Professor, Department of Obstetric and Gynecology, School of Medicine, Yuzuncu Yil University, Van, Turkey.
4. Professor, Department of Pathology, School of Medicine, Yuzuncu Yil University, Van, Turkey.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Erbil Karaman,
Assistant Professor, Hafiziye Mahallesi Iki Nisan Caddesi, Efes Sitesi A Blok No:9 Ipekyolu, Van, Turkey.
E-mail: erbil84@gmail.com

FINANCIAL OR OTHER COMPETING INTERESTS: None.

Date of Submission: **Jul 22, 2015**
Date of Peer Review: **Aug 04, 2015**
Date of Acceptance: **Aug 05, 2015**
Date of Publishing: **Sep 01, 2015**