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Tubal Choriocarcinoma Cured by Surgery Alone: A Case Report

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Case report

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ABSTRACT

Background: Fallopian choriocarcinoma is extremely rare and is still treated with a combination of chemotherapy and surgery. We report a case of gestational fallopian tubal choriocarcinoma treated solely with surgery and describe its clinical characteristics, treatment, and follow-up prognosis.

Case presentation: We report a patient who developed fallopian tube choriocarcinoma during the perimenopausal period. After surgical treatment, the patient was diagnosed with histopathology using immunohistochemistry staining combined with short tandem repeat detection, and the diagnosis was confirmed as "gestational fallopian tubal choriocarcinoma". Following the surgery, serum Beta-Human Chorionic Gonadotropin (β-HCG) levels were closely monitored. The patient's β-HCG values demonstrated an exponential decrease, returning to the normal range within six weeks. This trend was maintained for a period of three years. Consequently, the patient did not undergo chemotherapy.

Conclusion: This report offers insights into a simplified surgical approach for treating gestational fallopian tubal choriocarcinoma, designed to meet specific criteria that aim to minimize unnecessary side effects of chemotherapy for these patients.

Keywords: Fallopian tubal choriocarcinoma; Ectopic pregnancy; Surgery; β-HCG; Histopathology

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Abbreviations: CT: Computed Tomography; STR: Short Tandem Repeat

INTRODUCTION

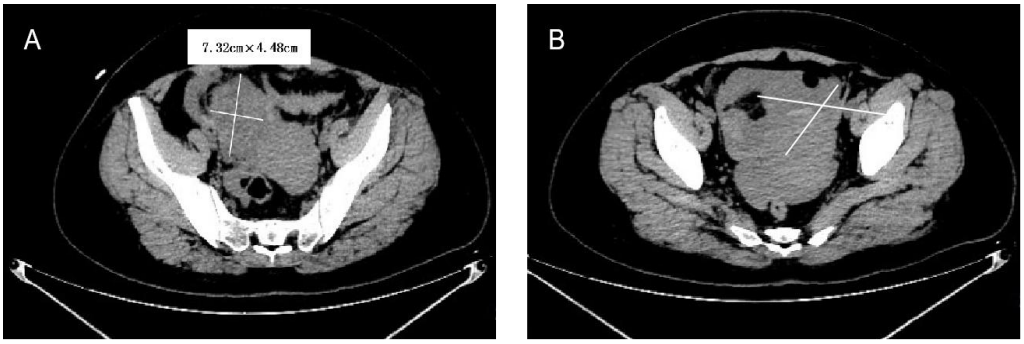
Choriocarcinoma, a rare yet highly malignant trophoblastic tumor, can originate from either gestational or non-gestational sources. It represents a malignant form of gestational trophoblastic tumor, comprising both mononuclear trophoblastic cells and syncytiotrophoblastic cells [1]. Using Short Tandem Repeat (STR) analysis combined with immunohistochemical techniques, we can distinguish between gestational choriocarcinoma and non-gestational choriocarcinoma, thus providing useful information on the conceptual type of etiology [2,3]. In the treatment of tubal choriocarcinoma, chemotherapy serves as the primary method, with surgery being a supplementary comprehensive treatment approach. We report the case of a patient with tubal choriocarcinoma with no metastasis who was successfully cured by surgery and showed no recurrence at her three-year follow-up.

CASE PRESENTATION

A 45-year-old woman, who had previously delivered a full-term fetus 24 years ago, was admitted to our hospital after experiencing fatigue for one week. Her physical examination results revealed stable vital signs, blood pressure of 100/60 mmHg, and pulse rate of 92 beats/min. The gynecological examination revealed a slightly enlarged uterus and a palpable mass approximately 5 cm in diameter in the right adnexal area with mild tenderness. The left adnexal area was normal. A posterior fornix puncture yielded 5 ml of non-clotting blood.

Auxiliary examinations included an abdominal CT, which showed fluid accumulation in the abdomen, blood Beta-Human Chorionic Gonadotropin (β-HCG) levels at 235303 mIU/ml, and blood routine results indicating hemoglobin at 56 g/L. A preliminary diagnosis of "ruptured ectopic pregnancy" was made on 2020.07.27. The patient underwent right tubal resection and left tubal ligation. During surgery, the peritoneum appeared purple-blue and approximately 2,500 ml of blood was found in the abdomen. The ampulla of the right fallopian tube was enlarged and thickened, measuring about 5 cm × 4 cm, with a completely ruptured cavity containing placenta-like tissue. Postoperatively, the patient received 5 units of suspended red blood cells and 400 ml of plasma (Figure 1).

Figure 1. Preoperative abdominal CT scan; (A) Adnexal mass (B) Pelvic effusion.



The pathological findings showed extensive bleeding and necrosis of the fallopian tubes, active proliferation of trophoblast cells with severe atypia, and no villi detected, which is consistent with the pathological features of chorionic epithelial carcinoma. Head magnetic resonance imaging, chest Computed Tomography (CT), and whole abdominal color ultrasound were performed to rule out the possibility of systemic metastasis. Ultimately, the patient was classified as FIGO stage I, with a World Health Organization prognostic score of 12, suggesting a higher prognostic risk. The components of the prognosis score are detailed as follows: Age-score 1 (45 years), previous pregnancy–score 2 (one full-term birth), interval from first pregnancy-score 4 (24 years since full-term birth), pre-treatment beta-HCG level-score 4 (>105 mIU/ml, no specific value noted), maximum tumor diameter-score 1 (approximately 5 cm × 4 cm), metastatic site-score 0 (none present), number of metastatic lesions-score 0 (none), previous chemotherapy failure-score 0 (none).

Postoperative monitoring β-HCG gradually decreased and fell to the normal range 41 days after the operation (Table 1). Considering that the patient's indicators have been normal, and there is no metastasis, there is no chemotherapy, but regular monitoring β-HCG.

Table 1. Perioperative serum β-HCG level monitoring results.

Date	β-HCG (mIU/mL)
Pre-operation	235303
6th day after operation	5855
9th day after operation	2485
13th day after operation	748
18th day after operation	372
22nd day after operation	252.35
30th day after operation	92.1
34th day after operation	60.62
41st day after operation	2.55

Follow-up

During the follow-up period, the patient was asymptomatic and had normal menstruation. The follow-up included serum β-HCG, gynecological color ultrasound, and pulmonary CT examination. After 3 years of follow-up, no signs of recurrence were found. At present, the patient is still being regularly followed up with serum β-HCG testing and imaging tests.

RESULTS AND DISCUSSION

Choriocarcinoma of the tubal tube is a rare and highly malignant ectopic trophoblastic tumor, predominantly reported in the literature. Among these cases, the majority are classified as gestational choriocarcinoma (commonly referred to as secondary choriocarcinoma) rather than non-gestational choriocarcinoma (often known as primary choriocarcinoma). Differentiating between gestational and non-gestational choriocarcinoma is important because

non-gestational choriocarcinoma has a poorer prognosis and requires more aggressive treatment. STR genotyping can be used for differential diagnosis of trisomy/monosomy syndrome and dimale triploid. By DNA polymorphism analysis at several STR sites, pregnant and non-pregnant tumors can be distinguished. In addition, the p57 [KIP2] gene is patrilineally imprinted and maternally expressed, and its protein product can serve as a surrogate marker for the maternal nuclear genome. Combining these two methods allows for an accurate diagnosis [4-6].

In this case, the clinical profile of the patient was as follows: she was 45 years old, had a previous pregnancy 24 years ago, and had a short interval between her last menstrual period and the identification of the tubal mass that confirmed choriocarcinoma. Thus, the choriocarcinoma may have developed as a primary or secondary ectopic pregnancy rather than from a previous mole lesion. These histories are important in determining the origin of the disease. For a clear diagnosis, immunohistochemical staining was performed on the tissue, and the result was P57 (-) (Figure 2), indicating that the tumor lacked maternal contribution. Consequently, the STR test was additionally conducted, revealing that the tumor was a female single male triploid (Figure 3). This indicates the presence of maternal alleles in the tumor. If a complete hydatidiform mole is accompanied by trisomy 11 (that is, one more maternal chromosome 11) or partial hydatidiform mole is accompanied by deletion of maternal chromosome 11, the p57 immunohistochemical staining results are contrary to the foregoing. This is because the imprinted gene CDKN1C is located on chromosome 11. However, this contrary conclusion can be corrected by ploidy analysis and STR genotyping. Therefore, we still consider this case to be tubal pregnancy choriocarcinoma.

Figure 2. Immunohistochemistry findings; (A) Proliferative syncytiotrophoblast leaf cells and intermediate trophoblast cells and are arranged alternately; (B) tumor cells are surrounded by massive necrosis; C-F show immunohistochemical CK, HCG, HPL, and Ki67 staining, respectively, magnification x400.

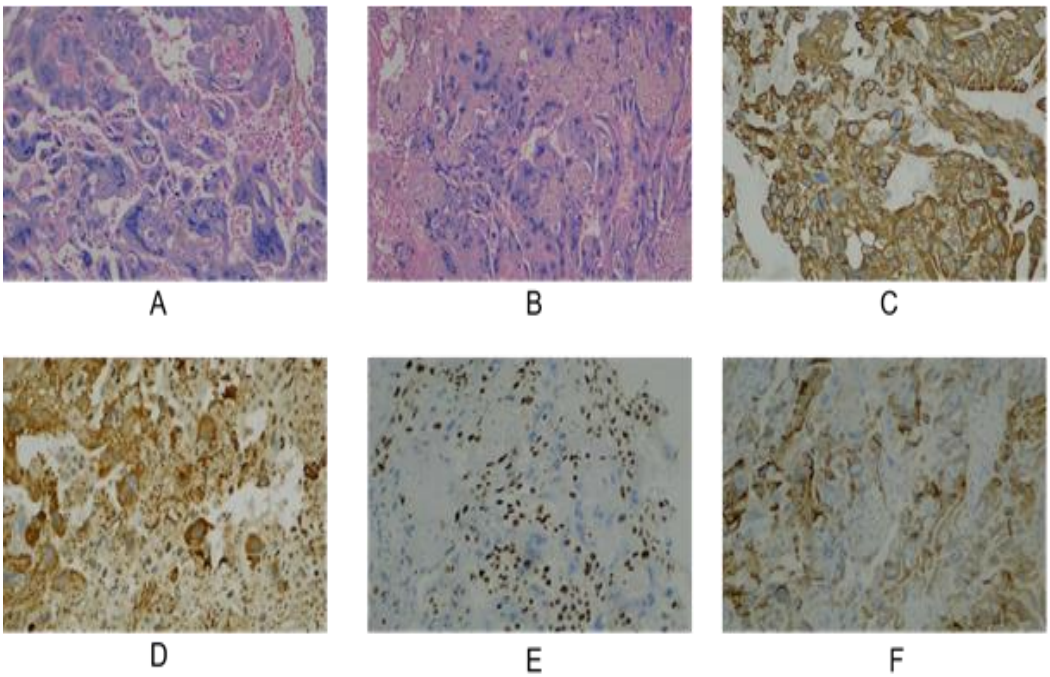
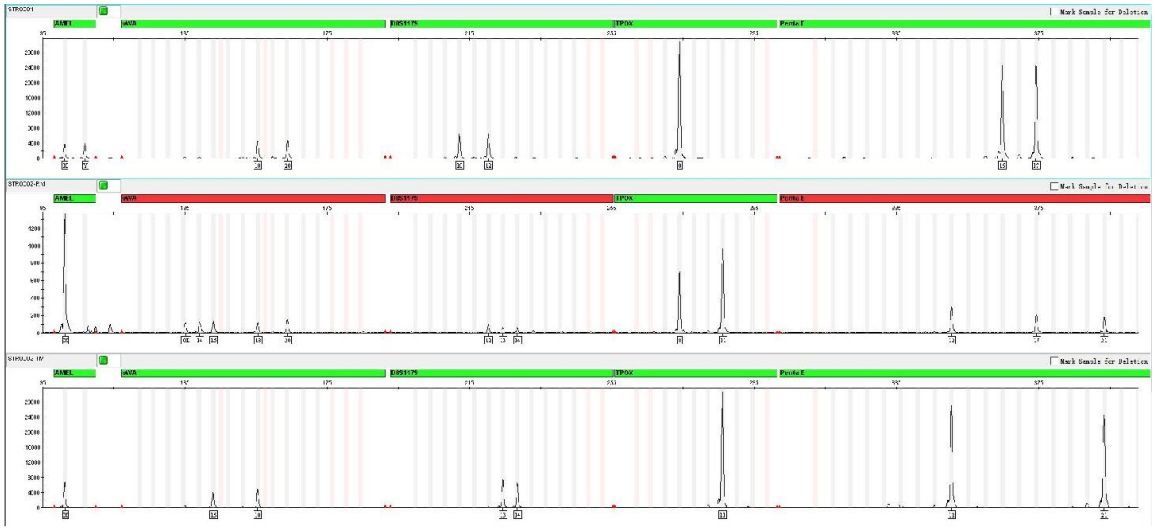


Figure 3. Short tandem repeat analysis; The tumor is a difemale monomale triploid; (A) Parent (father) (B) choriocarcinoma; (C) Parent (mother).



The clinical symptoms of tubal choriocarcinoma are similar to those of ectopic pregnancy and often manifest as abdominal pain, vaginal bleeding, and appendicular mass, and elevated serum β-HCG levels. According to relevant literature reports at home and abroad, tubal choriocarcinoma mostly manifests as an ectopic pregnancy and is treated by surgery and confirmed by histopathology after the operation. There is no unified standard for the clinical treatment of tubal pregnancy choriocarcinoma at this stage, and comprehensive treatment based on chemotherapy supplemented with surgery is advocated. Staging and prognostic scoring were performed according to tumor staging and prognostic scoring criteria of the FIGO2000 trophoblastic tumor. The chemotherapy regimen may refer to the treatment regimen for trophoblastic tumors and adopt a single drug or a combination of chemotherapy to achieve good results [7].

A review of the literature revealed that some patients with tubal choriocarcinoma had no previous pregnancy history [8-11], which raised specific questions about the origin of the disease. Tubal choriocarcinoma may originate in the fallopian tube. Some researchers have suggested that this may be caused by the invasion of the fallopian tube wall and the nourishing of vessels by the abnormal proliferation of trophoblast cells caused by the rich local blood flow, the thin tubal cavity, and a thin muscle layer during pregnancy; on this basis, cell mutations occur [12]. For the first pregnancy, the prognostic factors of the previous pregnancy in the FIGO scoring system could not be scored according to the time interval of the previous pregnancy; therefore, the diagnosis and treatment of tubal choriocarcinoma is worth further discussion. In this case, 24 years before the previous pregnancy, STR genotyping revealed primary gestational choriocarcinoma of the tubes; however, if metastatic choriocarcinoma was considered, the previous pregnancy was considered high-risk. The FIGO scoring system is suitable for uterine lesions; however, there are some difficulties in applying this prognostic scoring system to choriocarcinoma. In contrast, patients with tubal choriocarcinoma who do not have a prior pregnancy present conditions that differ from gestational trophoblastic tumors of uterine origin. However, this patient did not have a metastatic disease. Most tubal choriocarcinomas are misdiagnosed as ectopic pregnancies, leading to their surgical resection. In early and non-metastatic cases, supplemental chemotherapy is necessary after surgical resection. In 1981, Ober and Maier described eight patients who were cured by surgical resection alone [11]; however, the full text of this article and the details of these eight patients are not available. A 2011 case report described a diagnosis of androgenic primary

choriocarcinoma of the Salpinx tubes that was evaluated using a combination of p57 [KIP2] immunostaining and STR analysis, without metastasis, cured by surgery without adjuvant chemotherapy and followed for 15 months without recurrence [13]. The patient underwent fallopian tube resection due to the consideration of ectopic pregnancy rupture, and her blood β-HCG level decreased from 235,303 mIU/mL before surgery to 5855 mIU/mL six days after surgery. Due to the patient's referral to two hospitals, her blood β-HCG levels dropped to the normal range when she was admitted to our hospital six weeks after surgery. The patient did not receive chemotherapy, and there were no signs of recurrence during the three-year follow-up.

CONCLUSION

This case report describes, for the first time, a rare difemale monomale triploid tubal choriocarcinoma that was identified by surgical treatment without chemotherapy and evaluated using a combination of p57 [KIP2] immunostaining and STR analysis. Therefore, we believe that for this type of tubal pregnancy choriocarcinoma, which is challenging to differentiate from an ectopic pregnancy and requires surgical exploration, following the initial tumor resection, it may be possible to consider not administering chemotherapy and following the patient with imaging and rigorous β-HCG monitoring to avoid any risk of metastasis and recurrence. The following conditions must be met: No metastasis in the early stage, an exponential decline in β-HCG, and a drop to the normal range at 6-8 weeks. At present, the number of patients cured by surgery without adjuvant chemotherapy is very small, and the accumulation of similar cases is needed to provide further evidence for our hypothesis. Notably, this case report provides a reference for the clinical diagnosis and treatment of tubal choriocarcinomas.

AUTHOR CONTRIBUTIONS

LS and YCX participated in conception of the idea and writing of the manuscript. JL was responsible for collecting intraoperative and pathological images. ZXY and YBQ performed the clinical investigation of this case. All authors contributed to the article and approved the submitted version.

CONFLICTS OF INTEREST

The authors declare no conflict of interest.

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