

## AN EXCEPTIONAL CASE OF CERVICAL CHORIOCARCINOMA HIGHLIGHTING THE RARITY BEYOND THE UTERINE BODY: CASE REPORT AND REVIEW OF LITERATURE

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Article Received: 04 April 2026

Article Revised: 24 April 2026

Article Published: 01 May 2026



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Department of Obstetrics and Gynecology, Mohammed VI University Hospital Abdelmalek Essaâdi University, Tangier, Morocco. DOI: <https://doi.org/10.5281/zenodo.19914279>



**How to cite this Article:** W. Aarbaoui\*, C. Khalloufi, T. Mazali, F. El Hilali H. Moustaide, S. Benkirane (2026). An Exceptional Case of Cervical Choriocarcinoma Highlighting the Rarity Beyond the Uterine Body: Case Report and Review of Literature. World Journal of Advance Healthcare Research, 10(5), 220–225.  
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### ABSTRACT

Choriocarcinoma is an aggressive trophoblastic tumor<sup>[1]</sup>, the uterine cavity being its most frequent site of development.<sup>[2]</sup> Extra-uterine choriocarcinomas are extremely rare and represent a small proportion of gestational choriocarcinomas.<sup>[1]</sup> Among these rare localizations, the cervix is considered the most commonly reported site, although its overall incidence remains exceptionally low.<sup>[2]</sup> In the present case, a 29-year-old multiparous patient, was admitted to the maternal emergency department with mild pelvic pain and vaginal bleeding. Cervical pregnancy was the most likely diagnosis (considering that the patient had six weeks of amenorrhea and elevated serum  $\beta$ -hCG levels). We carried out a surgical exploration by laparotomy (laparoscopy was not available), and performed a resection of the entire mass, which included placental debris. Pathological examination returned results in favor of poorly differentiated tumor proliferation, suggesting a primary cervical choriocarcinoma. This case underscores the diagnostic challenges associated with cervical choriocarcinoma. The rarity of this entity, combined with its atypical presentation, contributes to delayed diagnosis and potential mismanagement. Early diagnosis of this rare entity is essential in order to avoid delays in treatment and improve prognosis. Clinicians must maintain heightened awareness of this rare but life-threatening condition when evaluating patients with suspected cervical pregnancy or unexplained gynecological bleeding associated with positive pregnancy markers.

**KEYWORDS:** Cervical choriocarcinoma; Ectopic pregnancy; Diagnostic challenge; Vaginal bleeding; Differential diagnosis; Cervical mass, Ultrasound diagnosis.

### INTRODUCTION

Choriocarcinoma is a highly aggressive malignant trophoblastic neoplasm, most often of gestational origin, characterized by rapid hematogenous dissemination and markedly elevated serum  $\beta$ -human chorionic gonadotropin ( $\beta$ -hCG) levels. It typically arises within the uterine cavity, whereas extrauterine presentations are rare and often associated with atypical clinical features.<sup>[1,6]</sup>

Primary cervical choriocarcinoma is an exceptionally rare entity, frequently presenting with severe vaginal bleeding and posing a significant diagnostic challenge

due to its resemblance to more common conditions such as cervical pregnancy or cervical carcinoma.<sup>[2,8]</sup>

Ectopic choriocarcinoma has been reported in various locations, including the fallopian tube, ovary, and vagina, reflecting the broad spectrum of clinical manifestations and contributing to potential diagnostic delays.<sup>[3–5,9]</sup> Diagnosis is based on a combination of clinical findings, markedly elevated serum  $\beta$ -hCG levels, and histopathological confirmation.<sup>[10]</sup>

Given its rarity and nonspecific presentation, cervical choriocarcinoma remains a diagnostic and therapeutic

challenge. We report a case of primary cervical choriocarcinoma and discuss its clinical, diagnostic, and therapeutic aspects in light of the current literature.

### CASE REPORT

A 29-year-old multiparous patient, with two live vaginal births was admitted to the maternal emergency with mild pelvic pain and vaginal bleeding. The patient had no previous history of ectopic pregnancy, molar pregnancy, or miscarriage, no concept of taking contraceptives. Her last delivery was three years ago and her last period was six weeks ago.

Upon admission, the patient was stable. Abdominal examination identified a soft and non-tender abdomen in all four quadrants. A vaginal examination revealed a closed, long, and firm cervix, minimal uterine bleeding with no vesicles or trophoblastic debris, pelvic pain with manipulation of the cervix, and a palpable, painful mass on the posterior wall of the uterus.

Transvaginal ultrasonography showed a normal-sized uterus with regular contours, a thin endometrium, and the presence of a heterogeneous posterior cervical mass measuring 34x42mm, taking the Doppler effect, associated with a thin effusion blade (figure 1). Ultrasonography does not show an intrauterine gestational sac or latero-uterine mass. beta-hCG measurement level was 160,000 IU/L and the patient's blood group was O positive.

An ectopic cervical pregnancy was the most likely diagnosis we retained. Our decision was to carry out a surgical exploration by laparotomy (laparoscopy was not available), which revealed a mass protruding from the posterior cervico-isthmic region measuring 30mm with regular boundaries and a thin wall (figure 2).

We performed a resection of the entire mass, which included placental debris, successful hemostasis was achieved (figure 3,4).

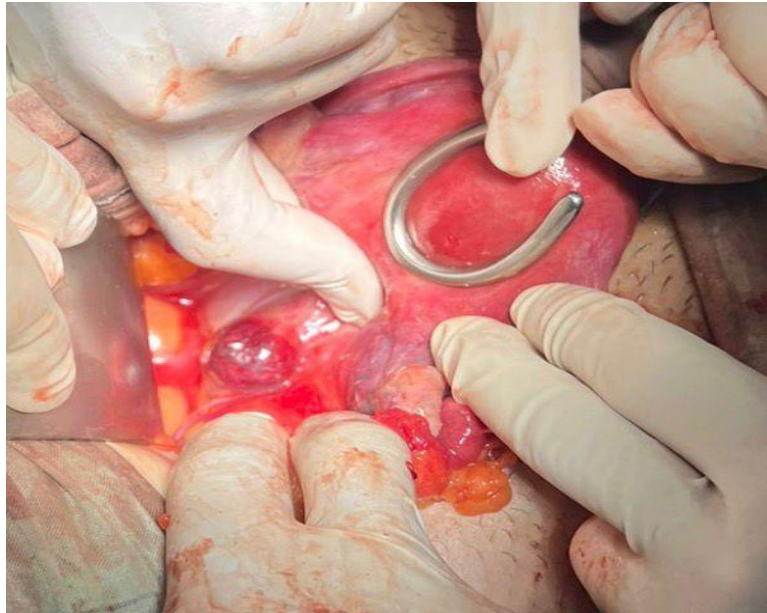
Pathological examination came back in favor of poorly differentiated tumor proliferation, suggesting a primary cervical choriocarcinoma.

We performed a complete staging workup including abdominopelvic CT scan, thoracic X-ray, and brain MRI, all of which were normal. According to the FIGO classification, the patient was classified as stage I, low-risk disease.

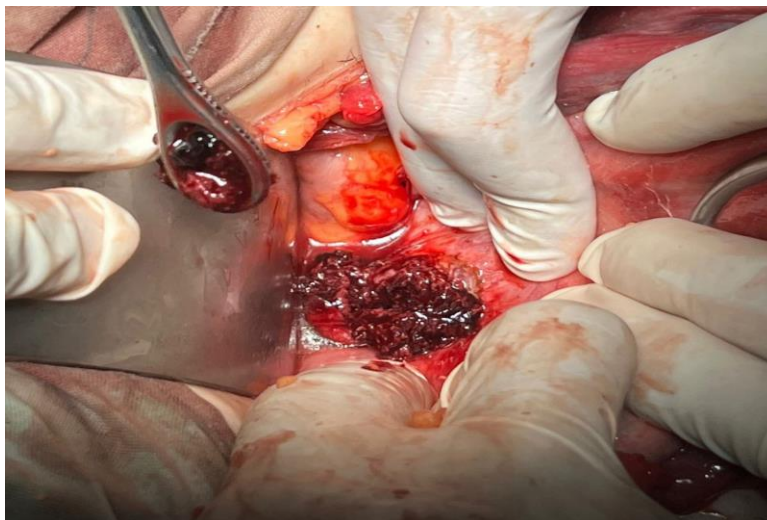
This case was presented at multidisciplinary team meetings and classified as a low-risk choriocarcinoma, then referred to the oncology department for further management, where she received a monochimiotherapy based on methotrexate and folinic acid a significant improvement was observed with weekly B-HCG levels decreasing until negativation after 4 courses of monochemotherapy.



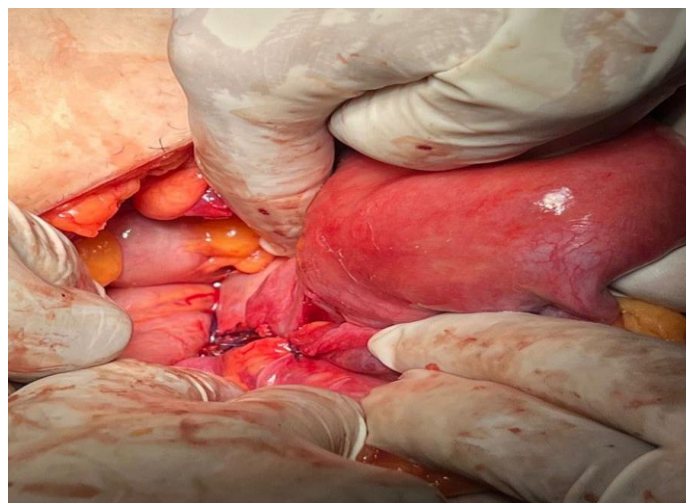
**Figure 1: Transvaginal ultrasound image demonstrating a heterogeneous mass located in the posterior cervix, measuring 34 × 42 mm.**



**Figure 2: Intraoperative view showing a mass protruding from the posterior cervico-isthmic region, measuring 30 mm, with regular boundaries and a thin wall.**



**Figure 3: Intraoperative view showing resection of the entire mass, including placental debris.**



**Figure 4: Intraoperative image demonstrating successful hemostasis following complete resection of the cervical mass.**

## DISCUSSION

Choriocarcinoma is an aggressive trophoblastic tumor.<sup>[1]</sup> the uterine cavity being its most frequent site of development.<sup>[2]</sup> Ectopic choriocarcinoma are extremely rare, while cervical choriocarcinoma are the most common site of extrauterine localization.<sup>[2]</sup> Other locations have also been reported, such as the fallopian tubes<sup>[3]</sup>, ovaries<sup>[4]</sup>, and vagina.<sup>[5]</sup> The interval between pregnancy and the appearance of the disease may vary from 5 weeks to 15 years, with occasional cases occurring in the postmenopausal period.<sup>[6][7]</sup> Due to the rarity of cervical choriocarcinoma and the nonspecific nature of its clinical presentation, establishing a diagnosis remains a real challenge. This diagnostic difficulty highlights the importance of our case, which illustrates the importance of maintaining a high degree of suspicion in the presence of any abnormal bleeding associated with elevated  $\beta$ -hCG levels. Early diagnosis of this rare entity is essential in order to avoid delays in treatment and improve prognosis.

A literature review of articles indexed in PubMed between 1998 and 2015 (Mangala and Deepak.) analyzed 121 published cases of choriocarcinoma with atypical manifestations. The geographical distribution of cases showed a predominance in Asia (47.1%) and Europe (26.44%), followed by North America (14.04%) and South America (3.3%). A few isolated observations came from Australia (2 cases), Africa (3 cases), and Eurasia (6 cases). The age of the reported patients ranged from 17 to 67 years, with 84.29% of cases concentrated in the 20-40 age group. It should also be noted that two patients were under 20 years and five were over 50 years of age.<sup>[1]</sup>

Among the 121 case reports, 50 (41%) occurred after a term pregnancy, which was also the case in our patient, 18 (14%) cases were associated with a previous abortion, 8 (6%) were molar pregnancies, 7(5%) were ectopic pregnancies, and 1 case was a partial mole.<sup>[1]</sup>

The pathogenesis of cervical choriocarcinoma is poorly understood. Several hypotheses have been put forward: it can involve from cervical metastasis of a primary uterine tumor that has regressed spontaneously, to malignancy of a product of conception implanted in the cervix, or even residual trophoblastic cells from a previous pregnancy which, after a latent phase, migrate to the cervix and undergo neoplastic transformation.<sup>[8]</sup>

Even though it's hard to make a clinical diagnosis, some clinical signs can prompt the suspicion of cervical choriocarcinoma, notably abnormal bleeding, which is the most common clinical sign<sup>[1]</sup>, despite the fact that this symptom raises the question of differential diagnosis with other more common diseases (miscarriage, polyp, ectopic pregnancy, or cervical cancer).<sup>[8]</sup> As in our observation, the patient was initially managed with an erroneous diagnosis of ectopic pregnancy. In conformity with the literature, the case reported by Kairi-Vassilatou

*et al.* (2007) describes a primary gestational choriocarcinoma of the cervix of a 43-year-old patient, initially mistaken as an ectopic pregnancy and definitively diagnosed after histopathological examination.<sup>[2]</sup> Our report also supports the findings of Wang *et al.* (2021), who, in a retrospective series of 13 patients with primary cervical gestational trophoblastic neoplasia, highlighted the rarity of this location and the diagnostic difficulty associated with its nonspecific clinical manifestations, dominated by abnormal metrorrhagia. As in our case, several patients in their cohort had initially received an erroneous diagnosis.<sup>[8]</sup> which highlights the importance of establishing the diagnosis for adequate management. Other symptoms may be observed, such as pelvic pain, and a cervical mass may be detected during clinical examination.<sup>[1]</sup>

For more than fifty years, Saito *et al* have proposed three criteria for the diagnosis of cervical choriocarcinoma: one important criterion is no primary localization of choriocarcinoma in the uterine cavity, the second criterion is the histopathological confirmation of the choriocarcinoma nature, finally the exclusion of a molar pregnancy or concomitant intrauterine pregnancy.<sup>[9]</sup>

In view of the non-specific nature of the symptoms, BHCG testing is essential, an increased level of this hormone associated with cervical mass must raise the alarm about the risk of choriocarcinoma.<sup>[1]</sup>

The presence of a pelvic mass should raise suspicion for choriocarcinoma.<sup>[1]</sup> Pelvic ultrasound plays a key role in the diagnostic process, typically revealing an echogenic or hypoechoic mass, often with a multicystic appearance, areas of necrosis and hemorrhage, or vascular cavities. Color Doppler examination commonly demonstrates marked hypervascularization resulting from arteriovenous shunts and tumor neovascularization.<sup>[10]</sup>

However, these sonographic features are not specific, and definitive diagnosis requires correlation with clinical findings and serum  $\beta$ -hCG levels.

In addition to its diagnostic value, Doppler ultrasound may also provide important prognostic information. Several studies have shown that increased tumor vascularization, a low resistance index, and a low uterine artery pulsatility index are associated with a higher risk of methotrexate resistance, even in patients classified as low-risk according to FIGO criteria. Therefore, Doppler assessment may serve as a complementary tool for early risk stratification and for optimizing therapeutic management in gestational trophoblastic neoplasia.<sup>[11]</sup> Not to mention the pathological examination, which complements the previous investigations that leads to the final diagnosis and eliminates other differential diagnoses by performing a biopsy, especially in patients with cervical lesions, as in the case reported by Wang and colleagues, in which four patients were diagnosed

with squamous cell carcinoma and received inadequate treatment.<sup>[8]</sup>

According to the joint EOTTD–ESGO–GCIG–ISSTD guidelines published in the Journal of Clinical Oncology, the management of gestational trophoblastic neoplasia is based on FIGO staging and WHO prognostic scoring. The management of gestational choriocarcinoma is based on FIGO staging and WHO prognostic scoring. Low-risk disease (FIGO score  $\leq 6$ ) is treated with single-agent chemotherapy, most commonly methotrexate, administered either as an 8-day regimen with folinic acid rescue or as weekly intramuscular injections. Actinomycin D may be used as an alternative first-line agent. Serum  $\beta$ -hCG levels are monitored weekly, and treatment is continued until normalization, followed by two to three consolidation cycles. Post-treatment surveillance includes monthly  $\beta$ -hCG monitoring for 6 to 12 months.<sup>[12]</sup>

In high-risk patients (FIGO score  $\geq 7$ ), multi-agent chemotherapy with the EMA-CO regimen is recommended. This protocol combines etoposide, methotrexate, actinomycin D, cyclophosphamide, and vincristine, administered in 14-day cycles. Weekly  $\beta$ -hCG monitoring is essential to assess response, and consolidation cycles are given after normalization. Long-term follow-up with monthly  $\beta$ -hCG measurements for 12 months is required to detect relapse early.<sup>[12]</sup>

Surgical resection of these lesions should be avoided due to their vascular structure, which can lead to severe, uncontrollable bleeding. That is why it is important to always bear in mind that, in cases of suspected cervical ectopic pregnancy, further investigations should always be carried out before deciding on surgical resection. Fortunately for our patient, hemostasis was achieved and we did not need to resort to additional surgical procedures.

In cases of uncontrollable vaginal bleeding that endangers the patient's life, a hysterectomy should be performed for patients who do not wish to preserve their fertility. Hysterectomy may also be indicated in cases of chemoresistant lesions. For young patients who wish to have children, uterine embolization may be proposed, followed by chemotherapy which constitutes the recommended treatment for choriocarcinoma.<sup>[8]</sup>

Most patients in the Chinese series required a hysterectomy in addition to chemotherapy (84.6%)<sup>[8]</sup>, whereas our management was limited to chemotherapy. These findings confirm the variability of the clinical and therapeutic profiles of this rare disease and reinforce the importance of individualized management and early diagnosis.

## CONCLUSION

Primary cervical choriocarcinoma represents a rare and diagnostically challenging entity that can mimic cervical

pregnancy due to overlapping clinical presentations of vaginal bleeding and elevated  $\beta$ -hCG levels. The rarity of this condition, combined with non-specific imaging findings, often leads to delayed diagnosis and emphasizes the critical importance of histopathological examination. Clinicians must maintain a high index of suspicion when evaluating patients with abnormal bleeding and positive pregnancy markers, as early recognition is essential to initiate timely treatment and improve prognosis. This case underscores the necessity of considering rare malignancies in the differential diagnosis of atypical gynecological presentations in reproductive-age women.

## ACKNOWLEDGEMENTS

The authors would like to thank all healthcare professionals involved in the diagnosis and management of this case.

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