CASE REPORT

Intestinal choriocarcinoma without primary source: A diagnostic enigma

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ABSTRACT

Intestinal choriocarcinoma is a very rare phenomenon. This is the first reported case of intestinal choriocarcinoma following an ectopic pregnancy. We report a 24 year-old woman who presented with severe abdominal pain, distension, and vomiting. She had a history of an ectopic pregnancy nine months prior. Emergent laparotomy exploration revealed abscess formation and obstructions at two sites in the small intestine, with unremarkable gynecological organs. The obstructed sections of the intestinal were excised and subsequently diagnosed as intestinal choriocarcinoma in histopathological evaluation. Postoperative positron emission tomography (PET) revealed a non-contrast-enhancing lesion on the small intestinal wall with increased metabolic activity consistent with healing tissue at the anastomosis site. No other primary focus and/or metastatic lesions were detected. Multi-agent chemotherapy was planned for the patient. No residual and/or recurrent tumoral lesions were detected on the PET scan at the one-year follow-up.

Keywords: Choriocarcinoma; gestational trophoblastic neoplasia; intestinal choriocarcinoma; multi-agent chemotherapy; positron emission tomography.

INTRODUCTION

Gestational choriocarcinoma is a rare, aggressive neoplastic subtype of gestational trophoblastic disease (GTN), reported in approximately one in 20,000 to 40,000 pregnancies.[1] An even rarer form of gastrointestinal tract metastatic choriocarcinoma with small bowel involvement have been reported in a total of 15 cases worldwide, with 5% of these cases occurring in women.[2] Here, we report an extremely rare case of intestinal GTN presented with atypical clinical findings without any primary source.

CASE REPORT

A 24-year-old woman (G3P1) presented with complaints of severe abdominal pain associated with the absence of flatulence and defecation for the last ten days. She had vomiting without any gynecological complaints. Her past medical history was notable for a full-term pregnancy three years prior and an ectopic pregnancy treated with methotrexate nine months before the current presentation. At the time of the ectopic pregnancy, the endometrial thickness was 8 mm on ultrasound, and serum beta-human chorionic gonadotropin (b-hCG) and progesterone levels were 607.19 mIU/mL and 2.09 ng/mL, respectively.

The gynecological examination on current admission was unremarkable. The abdominal examination revealed distension and tenderness with guarding and rebound tenderness. She had high serum levels of b-hCG (25,160 mIU/mL), platelet count (521×103/mL (159-388)), and C-reactive protein (CRP) (29.84 mg/dL (0-0.5)) and severe combined serum electrolyte imbalances (sodium of 119 mEq/L (136-146); potassium of

Cite this article as: Tutcu Şahin S, Solmaz Hasdemir P, Atmış Ö, Aliyeva A. Intestinal choriocarcinoma without primary source: A diagnostic enigma. Ulus Travma Acil Cerrahi Derg 2024;30:771-773.

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Ulus Travma Acil Cerrahi Derg 2024;30(10):771-773 DOI: 10.14744/tjtes.2024.57242
Submitted: 17.03.2024 Revised: 27.07.2024 Accepted: 04.08.2024 Published: 07.10.2024

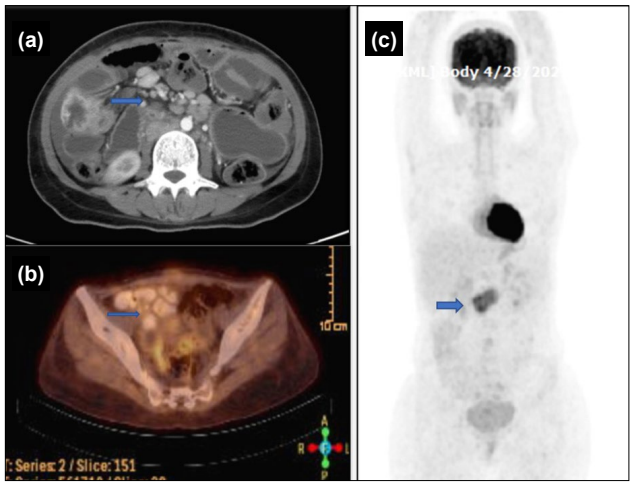

Figure 1. Preoperative abdominal computed tomography (CT) imaging **(a)** showing signs of ileus and intestinal implants (arrow). Postoperative positron emission tomography (PET) scan displaying high metabolic activity in the small intestines, viewed in transverse **(b)** and coronal **(c)** planes (arrows).

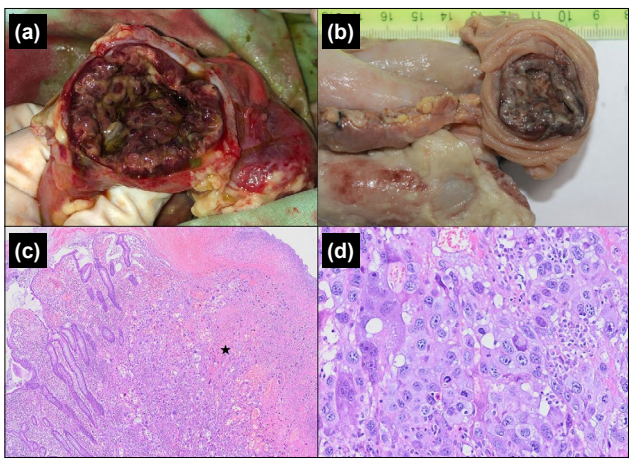

Figure 2. Macroscopic (a and b) and microscopic (c and d) appearances of the choriocarcinoma. **(a)** The obstructed section of the intestine as seen during the operation. **(b)** Hemorrhagic, brown, ulcerated, polypoid tumoral lesion, sharply demarcated from the non-neoplastic intestinal mucosa. **(c)** Transition from non-neoplastic intestinal mucosa to ulcerated, necrotic, and hemorrhagic tumoral lesion (*), stained with hematoxylin and eosin, magnification x40. **(d)** Tumor cells consisting of atypical cytotrophoblasts and syncytiotrophoblasts with frequent mitotic figures, stained with hematoxylin and eosin, magnification x200.

2.8 mEq/L (3.5-5.1) and calcium of 8 mg/dL (8.8-10.6)). The serum progesterone level was 2.09 ng/mL. Abdominopelvic computed tomography (CT) imaging revealed normal genital organs, an abscess formation (10 x 5 cm in diameter), and free fluid between the intestines along with enlarged (>1 cm) para-aortic and mesenteric lymph nodes (Fig. 1).

She underwent emergent exploratory laparotomy due to clinical findings consistent with acute abdomen. The genital organs appeared normal, and the intestines were found to be dilated after the abscess fluid was aspirated. Two intestinal obstructions, nearly 5 cm in diameter, were observed at 240 and 320 cm in proximity to the ileocecal valve. The enlarged lymph nodes were found in the intestinal mesenteric tissue. The obstructed intestinal tissues were excised, and side-to-side anastomoses were performed. Histopathological examination of the biopsy specimens was consistent with GTN (Fig. 2). The serum b-hCG on the second day post-operation regressed to 5,865 mIU/mL. The serum b-hCG level at the one-month follow-up was 502 mIU/mL. Postoperative positron emission tomography (PET) scan performed to detect the primary source and/or metastases revealed a non-contrast lesion on the small intestinal wall with a diameter of 50x31 mm with increased fludeoxyglucose (^{18}F-FDG) uptake (SUV-max: 10.9) without any other metabolic activity in her body parts (Fig. 1). The metabolically active area on the intestinal side was consulted by the Departments of Radiology, Nuclear Medicine, and Surgery and was considered to be healing tissue at the anastomosis site. Multi-agent chemotherapy was planned to be warranted in accordance with the recommendations of the Society of Gynecologic Oncology and the International Federation of Gynecology and Obstetrics (FIGO).[3,4] No residual and/or recurrent tumoral lesion was detected on the PET scan at the one-year follow-up. Written consent was obtained from the patient for this report.

DISCUSSION

Choriocarcinomas may present following term pregnancies (~50%), molar pregnancies (~25%), and other gestational events.[1] To the best of our knowledge, there is no reported case of choriocarcinoma of the small intestines following an ectopic pregnancy and without any primary source.

It has been reported that patients are at higher risk for post-molar gestational trophoblastic neoplasia if they have any of the following: age older than 40 years, pre-evacuation b-hCG greater than 100,000 milli-international units/mL, excessive uterine enlargement, or theca lutein cysts greater than 6 cm.[1] Interestingly, our case did not have any of these predictors.

Ectopic molar pregnancy is a rare gynecological occurrence. GTN following an ectopic molar pregnancy is extremely rare. Only six cases with GTN originating from interstitial ectopic pregnancy have been reported in the literature.[5] All these GTN cases were reported in genital organs. Our case is the first intestinal GTN following an ectopic pregnancy. The combination of histopathology and serum b-hCG is the gold standard for diagnosing molar pregnancy and GTN.[6]

Disseminated metastasis usually occurs only after pulmonary metastases are established. Another unique feature of our case was the absence of any characteristic findings in physical examinations and imaging modalities that would indicate any other metastatic and/or primary GTN source. Several

cases of GTN have been reported after testicular GTN presented with gastrointestinal bleeding and/or perforation.[7-9] Our case presented with only abdominal pain and distension, without any bleeding and/or perforation.

Our case was considered stage 4 GTN according to the FIGO staging system.[4] Currently, there are no standard management and follow-up guidelines at this stage of the disease in this particular situation. Aggressive multi-agent therapy is recommended for patients with high-risk diseases.[3,4] Currently, the EMA-CO (etoposide, methotrexate, and dactinomycin alternating with cyclophosphamide and vincristine) chemotherapy protocol is used most frequently. The increased risk of early menopause following multi-agent chemotherapy should be taken into consideration in such cases.[1] Adjuvant surgical procedures are especially recommended for controlling symptoms related to metastasis, like hemorrhage, and managing high-risk GTN.[10,11]

CONCLUSION

Intestinal choriocarcinoma should be included in the differential diagnosis of acute abdomen in the presence of high b-hCG levels among women of reproductive age. This is a curable type of malignancy with correct diagnosis and appropriate treatment.

Peer-review: Externally peer-reviewed.

Authorship Contributions: Concept: S.T.Ş., P.S.H.; Design: S.T.Ş.; Supervision: P.S.H.; Resource: Ö.A.; Materials: Ö.A.; Data collection and/or processing: A.A., Ö.A.; Analysis and/or interpretation: S.T.Ş.; Literature search: P.S.H.; Writing: P.S.H., S.T.Ş.; Critical reviews: P.S.H., A.A.

Conflict of Interest: None declared.

Financial Disclosure: The author declared that this study has received no financial support.

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OLGU SUNUMU - *ÖZ*

Primeri bilinmeyen intestinal koryokarsionom: tanısal bir bilmece

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İntestinal koryokarsinom oldukça enderdir. Ektopik gebelik sonrasında gelişen intestinal koryokarsinom olgusu ise daha önce bildirilmemiştir. Olgumuz şiddetli abdominal ağrı, distansiyon ve kusma yakınmaları ile merkezimize başvurmuş 24 yaşında kadın olup, anamnezinde 9 ay önce ektopik gebelik geçirmiş olması dışında hastalık öyküsü yoktu. Akut batın tablosu ile acil laparotomik eksplorasyona alınan olgunun batın gözleminde ince barsak ansları arasında abse formasyonu ve iki alanda obstruktif kitle tespit edilmiş, jinekolojik organlar olağan olarak izlenmiştir. Obstrukte intestinal alanlar eksize edilmiş, histopatolojik değerlendirme sonucu intestinal koryokarsinom olarak saptanmıştır. Postoperatif pozitron emisyon tomografi değerlendirmesinde ince barsak duvarında anostomoz hattı ile uyumlu alanda kontrast tutmayan bir lezyonda artmış metabolik aktivite dışında patoloji saptanmamıştır. Olgu multi-ajan kemoterapiye yönlendirilmiş, birinci yıl kontrol PET taramasında rezidü ve/veya rekürren lezyona rastlanmamıştır.

Anahtar sözcükler: Koryokarsinom; gestasyonel trofoblastik neoplazi; intestinal koryokarsinom; multi-ajan kemoterapi; pozitron emisyon tomografi.

Ulus Travma Acil Cerrahi Derg 2024;30(10):771-773 DOI: 10.14744/tjtes.2024.57242