



Case report

Hypered laparoscopic and endoscopic management of lower gastrointestinal bleeding from metastatic Jejunal and ileal choriocarcinoma of unknown primary

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ABSTRACT

Introduction: Choriocarcinoma is form of malignant neoplasm that arise from trophoblastic cells that occurs mostly in ovaries and testes and it can metastasis to lungs, liver or to gastrointestinal tract.

Presentation of case: This is 37 years old female presented as a case of lower gi bleeding and was diagnosed to have metastatic jejunal and ilium choriocarcinoma of unknown primary and underwent ileocecal resection first then followed up with small bowel resection around 60 cm from duodenojejunal junction with primary anastomosis. The patient was found to have liver and brain metastasis and received chemotherapy with full response.

Discussion: Choriocarcinoma is the most aggressive form of gestational trophoblastic disease that metastasizes through the lymphatic and hematogenous routes, and when its metastasis to the small bowel it's considered the worst prognosis with high mortality rate. Treatment consists of surgery and combined chemotherapy which is what our patient responded with.

Conclusion: Metastatic choriocarcinoma with unknown primary can be treated surgically with chemotherapy.

1. Introduction and importance

Choriocarcinoma is form of malignant neoplasm that arise from trophoblastic cells, it's one of the subtypes of Gestational trophoblastic neoplasia which also include invasive mole, choriocarcinoma, placental site trophoblastic tumor and epithelioid trophoblastic tumor [1]. CM can be either gestational or nongestational in origin, differentiating between its subtypes has importance in prognostic and therapeutic management [2]. It is well known that CM is rapidly growing, highly invasive, widely metastasize and secretes human chorionic gonadotropin (hCG) [3]. The pathogenesis of these tumors is still uncertain, they primarily occur in the ovary or the testes. Extragonadal sites that have been reported including lung, liver, breast, prostate, urinary bladder, nose and gastrointestinal tract [4]. There's not much data about metastatic choriocarcinoma in the GI tract with unknown primary, for that we chose this case report of a young lady who presented with melena and was diagnosed with metastatic choriocarcinoma with unknown origin. With importance of how we approached, diagnosed and managed her. This case report has been reported in line with the SCARE Criteria [12].

2. Case report

37 years old female, middle eastern, housewife, married with history of hypertension on amlodipine, body mass index of 43, para 7 with complete 2 abortion 12,14 years ago and no history of allergies. Presented to the emergency department by herself Complaining of first-time history of fresh blood and melena for 2 days, large amount. At that time, her menstrual cycle was regular, and her last menstrual period was one month before admission, there was no history of hemorrhoids or anal fissure, hx of trauma, use of non-steroidal anti-inflammatory drugs, previous similar attack, loss of consciousness, history of weight loss, nausea or vomiting, family history of GI malignancy, history of decrease appetite, history of dysphagia, odynophagia, history of heart burn, reflux, history of smoking or alcohol drinking.

She was stable with heart rate of 58 beats/min, blood pressure of 122/70 mmHg, afebrile.

On examination, patient looked well, not in pain, not jaundiced. Abdominal examination: soft lax no tenderness, no organomegaly. Per rectal exam revealed melena with no hemorrhoids or anal fissure. Cardiovascular system was normal and chest examination was normal with

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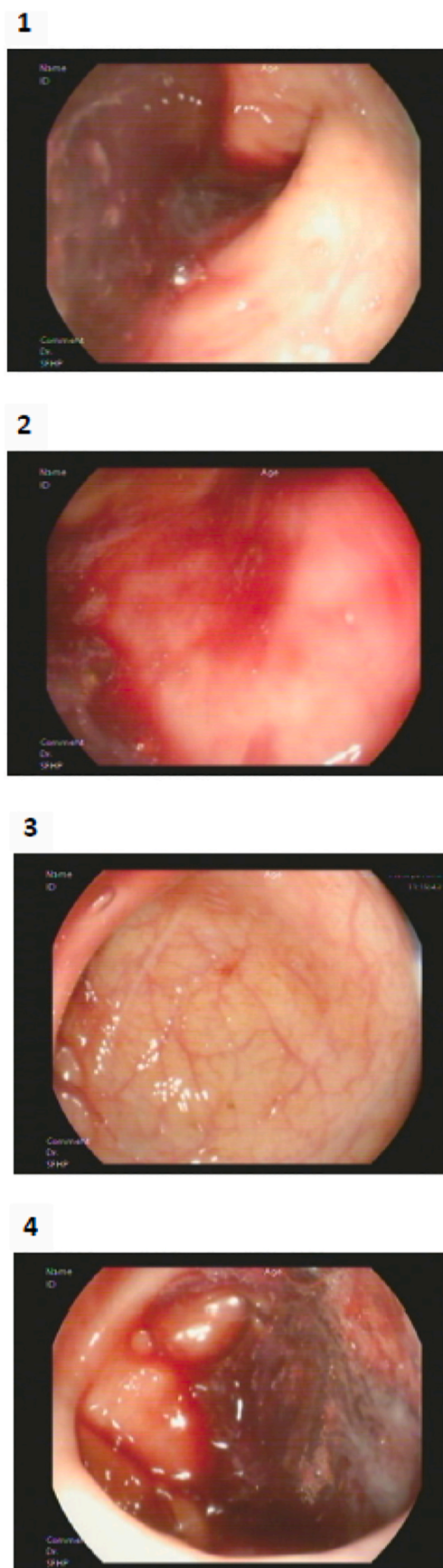


Fig. 1. Lower GI scope showing bleeding.



Fig. 2. CTA Coronal showing liner hyperintensity of TI suggestive of active bleeding.



Fig. 3. CT Axial View showing Focal liner hyperintensity at the terminal ileum suggestive of Active bleeding.

equal bilateral air entry.

Laboratory was done which showed, occult Blood was Positive. electrolytes were normal, amylase of 10 (U/I). liver function test was normal. Hemoglobin level of 11.5 (g/i), hematocrit of 0.348, MCV: 83 (fi), white blood cells of 9.8 ($10 \times 9/L$).

Patient was admitted with gastroenterology and started on conservative management with pantoprazole infusion and 1 units of packed red blood cell given. Patient underwent upper gastrointestinal endoscopy with the result of duodenitis and no active or evidence of recent bleeding, biopsy was acquired that was positive for helicobacter pylori. Colonoscopy was done that showed no signs of lower GI bleeding. Patient was offered capsule endoscopy but refused and was discharged home with H. pylori triple therapy.

One week after discharged, patient presented to the emergency department with same history associated with shortness of breath and fatigue with decrease appetite. She denied history of abdominal pain, syncope, chest pain or any genitourinary symptoms. on examination she was conscious, alert, oriented and pale, her vital signs were bp of 106/59, HR: 101b/min, afebrile, Chest; EBAE, CVS: S1 + S2 + 0, Abdomen; soft lax per rectal exam showed melena, no fresh blood, hemorrhoids or

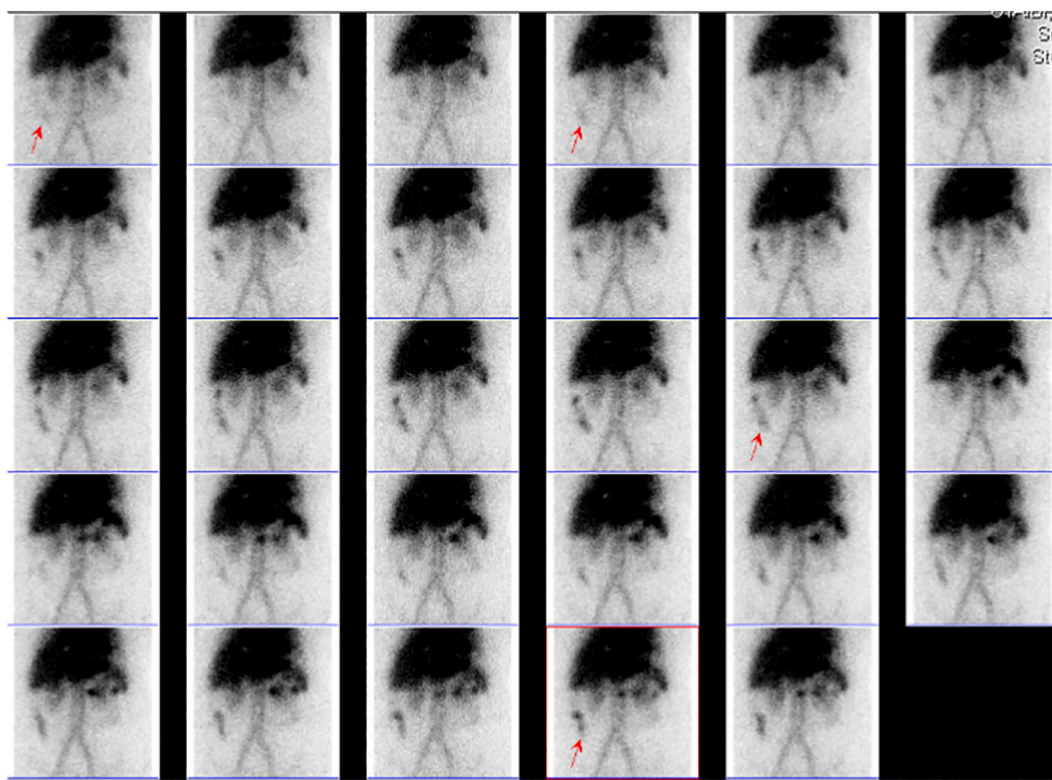


Fig. 4. Localization scan showing active bleeding from TI and proximal colon.

anal fissure.

Laboratory were requested with results of; Hgb: 6(g/i), wbc:10($10 \times 9/L$), other laboratory results were unremarkable. Patient was admitted under gastroenterology, conservative management was started and 2 units of PRBCs were given. Upper GI Endoscopy repeated, which showed mild antral gastritis with mild duodenitis. Capsule endoscopy was carried out. Colonoscopy implemented again with results of few angiodysplasia with no active bleeding with possibility of bleeding mostly from the proximal bowel (jejunum or proximal ileum) which can't be reached by scope (Fig. 1).

Computed tomography angiogram was requested, and the result was Focal liner hyperintensity at the terminal ileum seen at arterial phase, increasing in density in the Porto venous and delayed images, measuring 2.5×0.5 cm, away from ileocecal valve by 4 cm suggestive of active bleeding (Fig. 2–3).

Localization scan was done for more precise point of bleeding, which showed active GI bleeding likely originating from the terminal ileum or proximal colon (Fig. 4). At this point patient was still requiring multiple blood transfusion, and intervention radiologist was involved in the management for embolization. Angiography was performed, they reached the superior mesenteric artery through the right common femoral artery, and they didn't find the source of bleeding, so they terminated the procedure.

With these findings, and multiple requirements of PRBC, the gastroenterologist consulted the general surgery team for exploration and control of bleeding. The patient was consented for laparoscopic plus minus open exploration, plus/minus bowel resection, plus/minus stoma creation and proceed.

During operation, patient was under general anaesthesia in a supine position. The surgery was performed by consultant of upper gi and bariatric surgery with experience for more that 10 years. The procedure was done in one of tertiary hospitals in the country. 4 ports were inserted in the abdomen 5 mm supraumbilical, 5 mm epigastric, 12 mm in left upper quadrant and 15 mm in the right lower quadrant, bowel run though was done, with no significant findings. 30 cm away from the

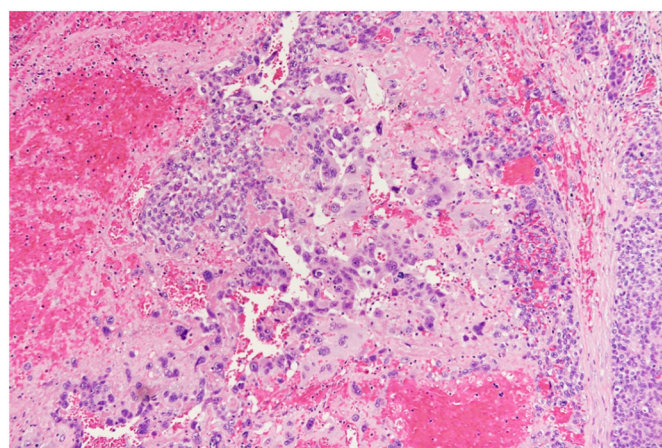


Fig. 5. Transverse and longitudinal sections of the mass showing cytotrophoblasts and syncytiotrophoblast.

ileocecal valve, a small enterotomy was done of the small bowel, push endoscopy was inserted, 4 cm from the ileocecal valve there was an area with oozing only, for that ileocecal resection was done with primary anastomosis using Endo GI Blue 60. The operation was about 2–3 h with minimal blood loss and there was no complications or adverse outcomes.

Histopathology results were metastatic choriocarcinoma, tumor located in the submucosa of terminal ileum, resection of margin was negative, Cecum and appendix were unremarkable (Fig. 5–6).

Multidisciplinary team was Formed with a gynecologist, oncologist and general surgery team. Human chorionic gonadotropin was requested with result of 50,721(iu/l). MDT decided plan was to investigate the patient and look for the primary tumor. Multiple imaging and laboratory test were requested.

We Started with ultrasonography of the pelvic to inspect for any

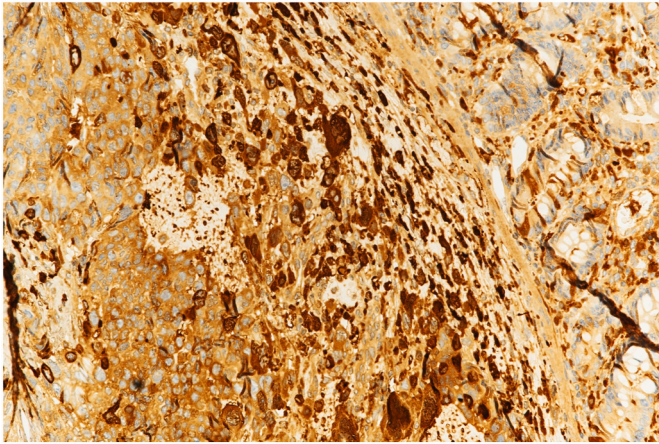


Fig. 6. Immunohistochemistry: hCG+.



Fig. 7. CT brain showing brain metastasis in the right frontal and left parietal lobes.

suspicious mass in the uterus or ovaries. Which showed, the uterus is anteverted normal in size measures $99 \times 60 \times 52$ mm, Midline endometrial thickness = 6 mm, both ovaries are normal in appearance, and there was no free fluid within the The Pouch of Douglas.

Then CT brain was requested which showed (Fig. 7). Large calvarial metastatic hemorrhagic and hypervascular mass lesion $7.5 \times 7.2 \times 6.2$ cm multiple intra-axial subcortical brain metastasis in the right frontal and left parietal lobes. Multiple acute lacunar infarcts scattered in the cerebral and cerebellar hemispheres, suggestive of micro emboli with irregular stenosis at the distal right middle cerebral artery.

After first operation by 1 week patient started to complain of melena and drop of hgb of less than 7 (g/l) that required multiple transfusion of blood. Patient passed the capsule endoscopy and was reviewed by the GI with results of the time of capsule to reach the duodenum was 1.54 min and there were multiple polyps in the terminal ileum, with different sizes. CT angiography scan was done with result of intussusception within jejunal loop that shows hyperdense foci noted within distal part that could represent hemorrhage or enhancing lesion associated with partial obstruction with multiple lung deposits and scattered liver metastasis along the right and left hepatic lobes as well as focal

hypodensity at the left (Fig. 8–9). Magnetic resonance enterography was requested with result of Partial obstruction seen in mid jejunal loops most probably due to obstruction could be secondary to bleed or mass with multiple subtle liver lesion. Tagged RBC scan was also requested with result of no scintigraphic evidence of active GI bleeding. Due to low hgb with multiple blood transfusion Patient was reoperated. She underwent laparoscopic exploration. Throughout the operation, patient was under general anaesthesia in a supine position. The surgery was performed by a consultant of upper gi and bariatric surgery with experience for more than 10 years. The procedure was done in one of tertiary hospitals in the country, the findings were, small bowel mass 60 cm from the DJ (Fig. 10). The decision was made to resect 60 cm of proximal jejunum with primary anastomosis with liver nodule resection. During the same operation gynecologic oncologist was involved for hysteroscopy dilation and curettage, with the findings of 3×4 cm lesion at the posterior uterine wall. The length of the operation was about 3–4 h with minimal blood loss with no complications or adverse outcomes.

Histopathology report was the jejunal resection showed metastatic choriocarcinoma with lymph node invasion, liver nodule resection was consistent with choriocarcinoma, the uterine wall biopsy was negative for malignancy.

Patient was kept in the ward for a week, during that time oncology team was consulted and their plan was to discharge her home if she's fit and to be readmitted for beginning of chemotherapy session after 1 month post operation. Her condition, prognosis and management were explained to the patient, and she agreed.

3. Clinical discussion

Gestational trophoblastic neoplasms (GTN) consist of multiple diseases including complete and partial molar pregnancies, invasive mole, placental site trophoblastic tumor (PSTT), and choriocarcinoma (CCA). GTN is considered to be one of the rarest malignancies that is very sensitive to chemotherapy. Mostly it's developed after molar pregnancy, but it can happen any at any gestation [5]. Choriocarcinoma is the most aggressive form of gestational trophoblastic disease, most studies have been case reports. For that reason, the clinicopathology, treatment modalities, and prognostic factors are not well established. Most CCA patient have history of hydatidiform mole, an abortion, ectopic pregnancy, or full-term pregnancy. In which Our patient had history of complete abortion 12, 14 years ago. CCA metastasizes through the lymphatic and hematogenous routes. The common sites of metastasis are the lymph node, liver, peritoneum, and lung. Generally, patients die of hepatic failure due to tumor metastasis, hemorrhage, and disseminated intravascular coagulation [6,7]. Metastasis to the small bowel is the worst prognosis [8], and it seen most commonly in the jejunum, followed by the duodenum and the ileum, respectively [9,10]. when it metastasizes, which is very rare and only 5% of patients [9], it usually presents with severe lower gastrointestinal bleeding. It may Present with intestinal intussusceptions, upper abdominal pain, vomiting or rarely perforated viscus [10].

Lower GI bleeding has multiple causes, most commonly causes of bleeding are vascular lesions, neoplasms, inflammatory lesions (like Crohn's disease), Meckel diverticulum, arteriovenous malformation, lymphangiectasia, and radiation induced. Small bowel metastasis from Breast cancer, melanoma, Kaposi's sarcoma can also manifest as LGIB [8]. Our patient initially presented with history of melena, of less than a week, but from her laboratory work (low Hgb, hypochromic microscopic) it might have been a chronic minimal bleeding that was not noticed before, on the second operation patient had an intussusception of the jejunal loops. Because the bleeding was mostly in the small intestine, Upper and lower GI scope was nonconclusive. CTA helped us in identifying the suspected area and surgery confirmed it for us. Few study reports had the same presentations that required also surgical intervention. Most patients had liver metastasis, our patient had liver, brain and lungs metastasis. The Patient was admitted in the oncology ward

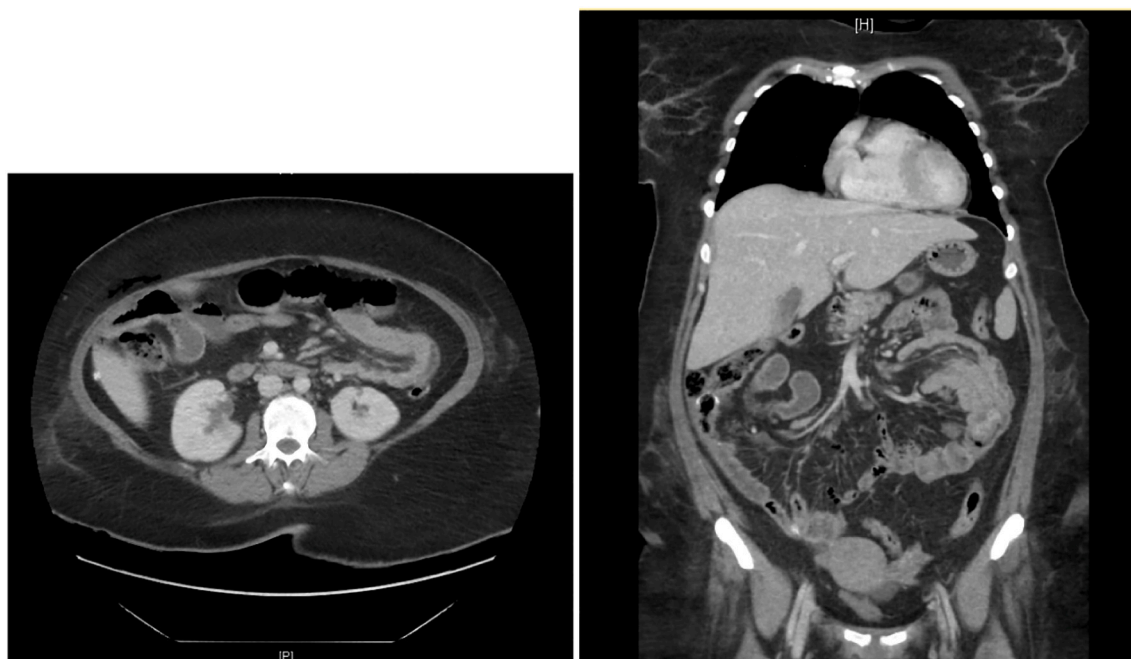


Fig. 8. CT abdomen showing intussusception of jejunal loops with signs of hemorrhage and partial obstruction with Lung and liver mets.



Fig. 9. CT Axial of lung showing multiple lung deposits.

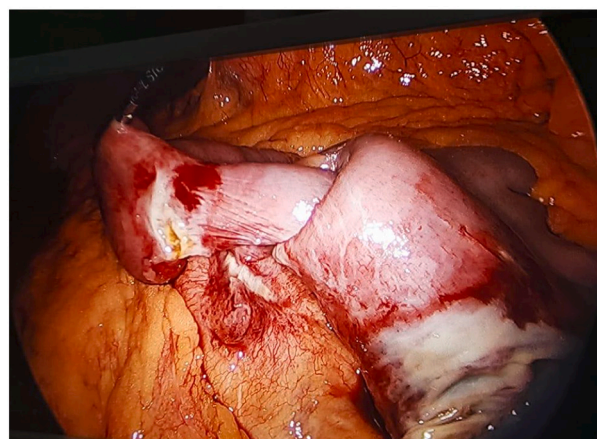
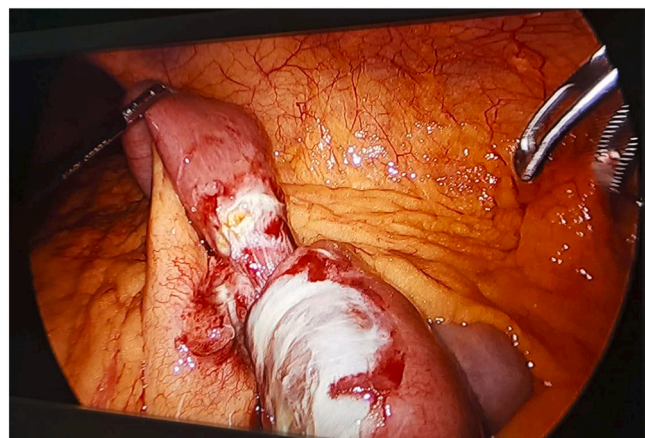


Fig. 10. Small bowel mass 60 cm from the DJ with active bleeding.

and started on chemotherapy of dactinomycin, etoposide, methotrexate and Cyclophosphamide (EMACO regimen), with repeated CT brain, CT chest and abdomen that showed regression in the metastatic lesions. And recent Bhcg showing normal result of 47(iu/l).

In conclusion, due to the uncommonness of the disease, most of the data are being extracted from case reports. For its treatments, case reports recommended to be vigilant in young patient with history of abortions or ectopic pregnancy, who present with unexplained GI bleeding. For what is known that early diagnosis is the only chance for patient's survivability rate and to help in preventing mortality and morbidity. Studies accustomed that this tumor usually responds to surgery and combined chemotherapy. [11]

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Ethical approval

In our institute, ethical approval is exempted, depend on acquired patient consent.

Consent

Written informed consent was obtained from the guardian on behalf of the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on requested.

Research registration

Not applicable.

Guarantor

DR. Amer Alshahrani.

Provenance and peer review

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CRediT authorship contribution statement

All authors contributed to manuscript preparation, manuscript editing, manuscript review.

Declaration of competing interest

The authors report no declarations of interest.

References

- [1] A. Biscaro, A. Braga, R.S. Berkowitz, Diagnosis, classification and treatment of gestational trophoblastic neoplasia, *Rev. Bras. Ginecol. Obstet.* 37 (1) (2015 Jan) 42–51.
- [2] Johanna M.D*. Savage, Emily B.S*. Adams, Emanuela M.D*. Veras, Kathleen M. PhD† Murphy, Brigitte M.M.D* Ronnett ‡., Choriocarcinoma in women, *Am. J. Surg. Pathol.* 41 (12) (December 2017) 1593–1606, <https://doi.org/10.1097/PAS.0000000000000937>.
- [3] J.H. Lee, J.K. Lee, D.B. Kang, Primary gastric choriocarcinoma coexisting with adenocarcinoma, *Korean J. Gastroenterol.* 73 (6) (2019 Jun 25) 350–354.
- [4] J. Rosai, Pregnancy, trophoblastic disease, and placenta, in: J. Rosai (Ed.), *Rosai and Ackerman's Surgical Pathology*, 9th ed., Elsevier, Philadelphia, USA, 2006, pp. 1737–1761.
- [5] Samir A. Farghaly (Ed.), *Gynecologic Cancers: Basic Sciences, Clinical and Therapeutic Perspectives*, March 2016. ISBN-10: 953-51-4278-X.
- [6] A. Kobayashi, T. Hasebe, Y. Endo, et al., Primary gastric choriocarcinoma: two case reports and a pooled analysis of 53 cases, *Gastric Cancer* 8 (2005) 178–185.
- [7] T. Noguchi, S. Takeno, T. Sato, Y. Takahashi, Y. Uchida, S. Yokoyama, A patient with primary gastric choriocarcinoma who received a correct preoperative diagnosis and achieved prolonged survival, *Gastric Cancer* 5 (2002) 112–117.
- [8] S.R. Fatemi, B. Hatami, M. Ghobakhlou, et al., Severe lower gastrointestinal bleeding due to metastatic choriocarcinoma to the jejunum: a rare presentation, *J. Gastroenterol. Dig. Dis.* 2 (1) (2017) 22–24.
- [9] J. Molina Infante, I. Beceiro Pedreno, C. Ripoll Noiseux, et al., Gastrointestinal hemorrhage due to metastatic choriocarcinoma with gastric and colonic involvement, *Rev. Esp. Enferm. Dig.* 96 (2004) 77–80.
- [10] S. Takahito, D. Tsutomu, N. Yuujirou, et al., A case of unknown primary metastatic choriocarcinoma of the small intestine in a man, *J. Jpn. Surg. Assoc.* 73 (2012) 3192–3196. Lurain JR.
- [11] Gestational trophoblastic disease I: epidemiology pathology, clinical presentation and diagnosis of gestational trophoblastic disease, and management of hydatidiform mole, *Am. J. Obstet. Gynecol.* 203 (2010) 531–539.
- [12] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 Guideline: updating consensus surgical CASE REport (SCARE) guidelines, *Int. J. Surg.* 84 (2020) 226–230.