Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at https://doi.org/10.1016/j.ejogrb.2017.11.009.

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Increased human Chorionic Gonadotropin levels five years before diagnosis of an ovarian dysgerminoma

A 35 year-old nulliparous white woman was referred to the Department of Obstetrics and Gynecology at the University of Bari in July 2007 with a 6 weeks' history of amenorrhea and increased level of serum human Chorionic Gonadotropin 125 IU/ml. Transvaginal ultrasound revealed the presence of a 2 × 2 cm right adnexal mass and diagnosis of ectopic pregnancy was placed. HCG level dropped in the next days to 38 and remained stable for the next two months. Diagnosis of persistent trophoblastic disease was made and the patient received 3 cycles of metothrexate (1 mg/sqm on day 1-3-5-7 with folinic acid rescue) but h-CG levels remained still stable (31 UI/ml). Pelvic ultrasound, total body CT scan, MRI of the pelvis, D&C were all negative. Diagnostic laparoscopy was performed but no pathologic finding was found. Blood samples revealed nohyperglycosilated hCG which is specific to trophoblastic disease.

The patient was then referred to the Charing Cross Hospital in London (UK) in May 2009, were diagnosis of persistently elevated h-CG levels was confirmed and strict follow-up was advised. Follow-up consisted of pelvic examination, transvaginal ultrasound every three months, MRI of the pelvis and chest X ray every six months. During follow up her serum beta-hCG levels remained stable in the range between 19 and 35 UI/ml.

In January 2013 on a routine follow-up examination a 6 cm solid right adnexal mass was found at transvaginal ultrasound. Serum beta-hHCG level was 23, all other tumor markers were within normal range. Total body CT scan was negative and the patient was submitted to laparotomic right adnexaectomy with infracolic omentectomy and multiple peritoneal biopsies. Gross examination revealed a greysh mass with tumor reaching the surface of the capsule. Pathologic examination revealed a pure ovarian dysgerminoma, and immunohistochemistry staining was positive for CD117 and PLAP but negative for beta-hCG, CK pool and inhibin. All other specimens (omentum, tube, biopsy and peritoneal cytology) were negative. Serum beta-hCG levels fall immediately after surgery to normal values. The patient received three cycles of adjuvant chemotherapy with the BEP regimen. At 10 months the patient is doing well, her hCG values are normal and she has no sign of recurrent disease.

Positive serum beta-human chorionic gonadotropin (beta-hCG) in reproductive-age women generally indicates a pregnancy, and to a lesser extent, gestational trophoblastic disease or ovarian germ cell tumors [1]. Besides these gynecologic neoplasms, benign gynecologic condition such as ovarian teratoma or non gynecologic cancers such as ovarian small cell carcinoma [2] and spindle cell carcinoma of the shoulders [3] can be associated with beta-hCG positivity as well.

This is the first reported case of persistently (five years) elevated serum beta-hCG levels in a patients in whom an ovarian disgerminoma was diagnosed five years later. The complete disappearance of beta-hCG following surgery was a confirmation of the ectopic production by the tumor despite the negativity of immunohystochemisry staining for beta-hCG.

A number of papers have documented increased serum beta-hCG levels in patients with diagnosis of ovarian dysgerminoma and elevation of serum markers beta-human chorionic gonadotropin and alpha-fetoprotein were also found to be significant predictors of overall survival in malignant ovarian germ cell tumors [4].

Persistently elevated serum and urinary beta-hCG levels was reported in 14 healthy patients who were not pregnant and had no previous diagnosis of GTN at the Charing Cross Hospital, London. False-positive beta-hCG was excluded in all. Three patients developed gestational choriocarcinoma after 9–29 months. However, in 11 women no cause for the persistently elevated beta-hCG was found. One of these achieved chemotherapy-induced normalisation of serum beta-hCG, but the remaining 10 underwent surgery and/or chemotherapy without benefit. Thus, 71% (10/14) of patients remain well with unexplained elevated beta-hCG levels [5].

Persistently elevated serum beta-hCG levels can be detected many years before diagnosis of an ovarian dysgerminoma. Strict follow-up in these patients is mandatory.

Conflict of interest

None.

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Epiploic appendagitis at term pregnancy. The importance of expert imaging: A case report

Dear editor,

A 39 years old GP0AB2 at term gestation presented to our emergency department with complaints of sharp right lower quadrant abdominal pain. She denied fever or other symptoms. current pregnancy was normal. Fetal weight estimation matched the 97% percentile – hence she was scheduled for an elective cesarean section in the following week.

The Patient suffered severe pain without contractions. The uterus was soft and tenderness was noticed in her right lower quadrant. Fetal heart rate monitoring was normal. Laboratory examinations revealed an elevated liver enzymes without leukocytosis, C-reactive protein was slightly elevated. Abdominal ultrasound scan was performed – the Appendix was not visualized, and a suspicious mass which could represent epiploic appendigitis (EA) was found located to the maximal point of tenderness (Fig. 1A).

No further diagnostic tools were performed and the patient was delivered by an emergency cesarean section. An examination of the bowl was undertaken by summoned general surgeons which revealed a necrotic epiploic appendage (Fig. 1B) on the rightly displaced Sigmoid, which was removed. Post operative course was normal and laboratory results normalized.

EA are peritoneum covered fatty masses found mostly on the left colon which supplied by blood vessels running at their bases [1]. Ischemia of EA is commonly due to torsion or a spontaneous thrombosis [1,2] and causes severe pain mimicking appendicitis or diverticulitis. Patients' present usually with localized sharp left lower abdominal pain and normal temperature and blood count [2]. Torsion of EA during pregnancy is extremely rare event and was only reported once in the literature [3], thus it is uncommon for a physician to contemplate on this etiology as a cause of an acute abdominal pain during pregnancy. The diagnosis is made by ultrasound that shows a hyperechoic non-compressible ovoid structure adjacent to the colonic wall. Doppler study can demonstrate absence of blood flow to the structure [4]. In the present case, the patient was at term pregnancy, thus our team didn't hesitate to deliver her by emergency cesarean section and no MRI was performed. Yet, In cases of preterm pregnancy, MRI should definitely be considered in the setting of acute abdomen in pregnancy.

AEA during pregnancy is extremely rare and causes a true diagnostic dilemma, since clinicians and surgeons often do not consider lesions of these structures in the differential diagnosis of intra-abdominal disease. Thus, – the physician should keep in mind this entity and a matching directed imaging study should be done.

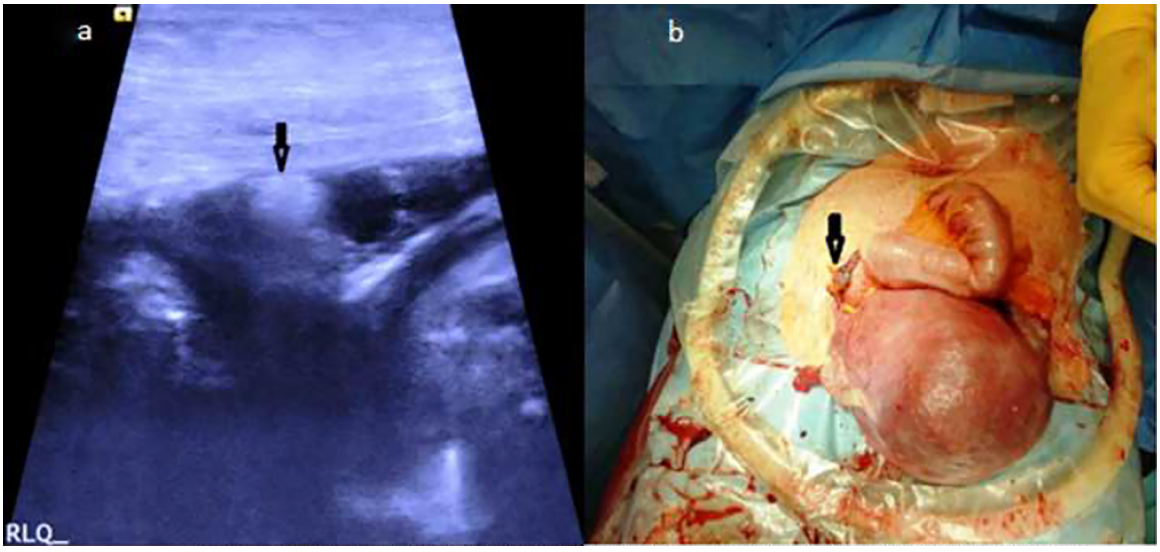

Fig. 1. a: Hyperechoic thickened epiploic appendagitis on ultrasonography, b: epiploic appendagitis attached to the rightly displaced sigmoid.