

Case Report

Ovarian torsion caused by hyperreactio luteinalis in the third trimester of pregnancy: a case report

Qin Li, Xiaotian Li, Pengnan Zhang

Department of Gynecology, Obstetrics and Gynecology Hospital of Fudan University, Shanghai 200011, China

Received August 20, 2015; Accepted October 10, 2015; Epub October 15, 2015; Published October 30, 2015

Abstract: In this case report, a rare case of an adnexal torsion caused by hyperreactio luteinalis (HL) in the third trimester is described, since adnexal torsions are mainly restricted to the first trimester of pregnancy. In an emergency Cesarean section, the patient gave birth to a healthy female baby weighing 3,300 g and we found an enlarged benign multiple luteinized follicular cyst mass in the right adnexum, which led to an adnexal torsion. After detorsion, both ovaries recovered to their normal sizes two months after the intervention.

Keywords: Hyperreactio luteinalis, third trimester pregnancy, adnexal torsion

Introduction

Adnexal torsion is the main complication for adnexal masses during pregnancy and occurs mainly during the first trimester [1-3]. It is usually caused by ovarian weight increases due to cystic or solid masses, hemorrhages, or edemas and defined as a partial or complete rotation of the adnexa on its axis together with the vascular pedicle. The incidence of adnexal torsion during pregnancy is low and reported to be 0.03% to 0.045% in all pregnancies [1, 4]. It is extremely rare in the third trimester and up to now, a case of adnexal torsion caused by HL in the third trimester of pregnancy has not yet been reported to our knowledge. Despite the association with elevated β hCG, 60% of reported HL cases are found in normal singleton pregnancies [5] and tend to occur later in pregnancy, typically in the third trimester or less commonly in the puerperium. Patients are typically asymptomatic or have relatively mild symptoms, but in as many as 28% of women, maternal virilization may occur [6, 7]. Herein we report the case of an acute ovarian HL caused ovarian torsion in the third trimester of pregnancy. The study protocol was approved by the ethics review boards of the Obstetrics and Gynecology Hospital of Fudan University and written informed consent was obtained from the patient.

Case report

A 27-year-old Chinese woman had her first pregnancy 2 years after her marriage. Her menstruation was not regular and the menstrual cycle duration was between 37 and 90 days. When she came to the hospital, we detected acne in her face during a physical examination. The serum β hCG concentration was 4,3187 mIU/ml in a Down's syndrome screening test during the 13th gestational week. The values of oral glucose tolerance tests (OGTT) were 4.8, 10.6, 9.8 and 4.2 mmol/L in the 0, 1st, 2nd and 3rd hours during the 24th week of gestation. During week 38, day 3 of gestation, the patient appeared in the hospital complaining of severe right abdominal pain without interval and was admitted the same evening. The white blood cell count was 15,500/ μ L with 85% neutrophils. Ultrasound examination revealed that her liver, cholecyst, pancreas, spleen and kidneys were normal, but an ultrasound image revealed that the size of the right ovary was 8.8 \times 6.3 \times 4.4 cm, and that of the left ovary was 5.7 \times 4.4 \times 4.3 cm (**Figure 1**). There was no free peritoneal fluid in the peritoneal cavity. The tumor markers CA125 (39.2 U/mL (normal value < 35 U/mL) and AFP (119.55 ng/mL (normal value < 25 ng/mL) were enhanced, whereas CA153 (10 U/mL (normal value < 28 U/mL), CA199 (10.17 U/mL (normal value < 37 U/mL) and CEA (1.86 ng/mL (normal

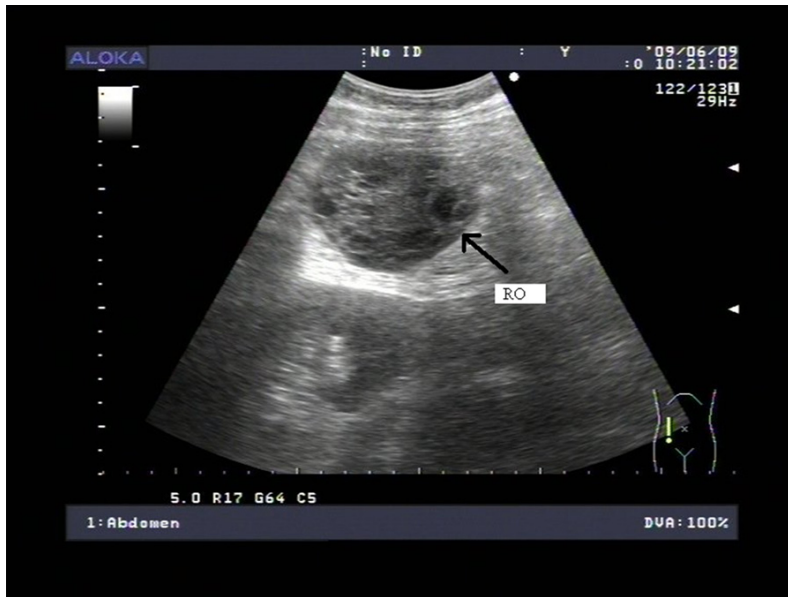


Figure 1. Ultrasound image of the ovaries. The size of the right ovary was 8.8 × 6.3 × 4.4 cm and appeared with multi-follicular cysts. (RO: right ovary).

value < 5 ng/mL) were in the normal range. Antibiotic therapy the next morning produced no relief and 7 hours later a female baby (3300 g) was born by Cesarean section. During the delivery, the two ovaries presented with enlarged multi-follicular cysts. The right ovary was about twofold bigger than the left one and was twisted twice through 360°, with involvement of the right fallopian tube, but was viable. During the operation, the right adnexum was untwisted without biopsy. The patient did not develop fever after the operation and the abdominal pain completely disappeared. Two months after the intervention, an ultrasound examination showed that both ovaries had returned to normal size.

Discussion

This is the first Chinese case report about an ovarian torsion caused by HL in the third trimester, which is extreme rare since HL induced adnexal torsion occurs mainly during the first trimester [1]. Because HL triggered follicular cysts are often misdiagnosed as malignancy [8, 9], unnecessary oophorectomies are frequently performed [7, 10]. For masses suspicious for malignancy or clinically symptomatic masses, evaluation by laparoscopy is recommended [11]. In our case, the serum α -fetoprotein concentration was more than 2.5 times the multiple of the median, but elevated α -fetoprotein

levels are not considered to be conclusive in the 3rd trimester [12]. Also, the enhanced CA125 serum concentration of 39.2 U/mL was in the upper normal range during the 3rd trimester [13]. The serum β hCG concentration of 43,187 mIU/mL in the 13th gestational week was not enhanced (normal values are 13,300-254,000 mIU/ml in the 13-16 gestational weeks), indicating that in our patient an abnormal sensitivity of the β hCG receptor was the cause of the follicular cysts.

Patients with ovarian torsion typically present after the acute onset of unilateral pain accompanied by nausea and vomiting, but the condition must be distinguished from appendicitis and other diseases associated with abdominal pain, and from placental abruption in pregnant women. A prolonged period of ovarian tension can lead initially to intraovarian congestion and ultimately to hemorrhage, necrosis and infarction. Massive ovarian edema may occur when there is impeded venous and lymphatic drainage resulting in marked ovarian enlargement [10]. Due to its ability to assess vascular flow and not cause harm to the fetus, ultrasound is the preferred imaging modality. Sonographic features of ovarian torsions are enlargements with prominent, heterogeneous central stroma and multiple, small, peripheral follicles. The follicles in twisted ovaries are smaller, usually measuring 5 mm or less in diameter. The classic progression of Doppler findings during ovarian torsion is loss of venous flow, decreased diastolic flow, and finally loss of systolic flow [14, 15]. The MRI appearance of adnexal torsions are reported as follows: fallopian tube thickening (protrusion), wall thickening of the twisted adnexal cystic mass, ascites, uterine deviation to the twisted side, adnexal hemorrhage, and lack of contrast enhancement of the adnexal mass [16].

Historically, oophorectomy has been the procedure of choice when an ovarian torsion is encountered. This practice stemmed from the

fear of thromboembolic complications, secondary to untwisting of the ischaemic adnexa. Venous thromboembolism was thought to occur after torsion reduction, which allowed coagulated blood to escape into the vascular tree, resulting in emboli. In addition, adnexectomy was also performed when the adnexa appeared necrotic or did not return to a viable-looking appearance after untwisting the ischaemic adnexa. However, recent literature mainly recommends a conservative treatment of ovarian torsions by simply untwisting the adnexa [17-19] during laparoscopic surgery, which is a safe procedure for pregnant women [20]. In our opinion, if a patient presents with acute abdominal pain in the late third trimester, Cesarean section should be performed, since for a late third trimester pregnancy with acute abdominal pain any clinical investigations carry more risk than premature delivery of the infant.

In our case, the twisted right ovary with HL was untwisted following the Cesarean section. Historically, biopsy was necessary when an enlarged ovary was encountered, but since HL recovers with decreased β hCG levels during puerperium, a biopsy of the edematous ovary could have increased the risk of complications, such as bleeding, ovary necrosis and loss of ovary functions.

Summary

In our case report, we introduce a rare case of an ovarian torsion caused by HL in the 3rd trimester of pregnancy. Acute abdominal pain was the sole clinical symptom and occurred in the 38th gestational week, which allowed us to perform a Cesarean section without a laparoscopic intervention, after which we diagnosed HL and untwisted the ovaries without complications.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Qin Li, Department of Gynecology, Obstetrics and Gynecology Hospital of Fudan University, 128 Shenyang Road, Shanghai 200011, China. Tel: +86-21-33189900; Fax: +86-21-55122025; E-mail: liqinbm@163.com

References

[1] Kumari I, Kaur S, Mohan H and Huria A. Adnexal masses in pregnancy: a 5-year review. *Aust N Z J Obstet Gynaecol* 2006; 46: 52-54.

[2] Terzic M, Aksam S, Maricic S and Arsenovic N. Acute abdomen caused by adnexal torsion in the first trimester of pregnancy: a case report. *Srp Arh Celok Lek* 2011; 139: 239-241.

[3] Hasiakos D, Papakonstantinou K, Kontoravdis A, Gogas L, Aravantinos L and Vitoratos N. Adnexal torsion during pregnancy: report of four cases and review of the literature. *J Obstet Gynaecol Res* 2008; 34: 683-687.

[4] Zanetta G, Mariani E, Lissoni A, Ceruti P, Trio D, Strobelt N and Mariani S. A prospective study of the role of ultrasound in the management of adnexal masses in pregnancy. *BJOG* 2003; 110: 578-583.

[5] Bidus MA, Ries A, Magann EF and Martin JN. Markedly elevated beta-hCG levels in a normal singleton gestation with hyperreactio luteinalis. *Obstet Gynecol* 2002; 99: 958-961.

[6] Tanaka Y, Yanagihara T, Ueta M, Hanaoka U, Kuno A, Kanenishi K, Yamashiro C, Ohnishi Y, Tanaka H, Hara K, Kushida Y, Kobayashi S and Hata T. Naturally conceived twin pregnancy with hyperreactio luteinalis, causing hyperandrogenism and maternal virilization. *Acta Obstet Gynecol Scand* 2001; 80: 277-278.

[7] Cavoretto P, Giorgione V, Sigismondi C, Mangili G, Serafini A, Dallagiovanna C and Candiani M. Hyperreactio luteinalis: timely diagnosis minimizes the risk of oophorectomy and alerts clinicians to the associated risk of placental insufficiency. *Eur J Obstet Gynecol Reprod Biol* 2014; 176: 10-16.

[8] Wajda KJ, Lucas JG and Marsh WL Jr. Hyperreactio luteinalis. Benign disorder masquerading as an ovarian neoplasm. *Arch Pathol Lab Med* 1989; 113: 921-925.

[9] Yapar EG, Vural T, Ekici E, Kuscu E and Gokmen O. Hyperreactio luteinalis masquerading as an ovarian neoplasm in a triplet pregnancy. *Eur J Obstet Gynecol Reprod Biol* 1996; 65: 177-180.

[10] Glanc P, Salem S and Farine D. Adnexal masses in the pregnant patient: a diagnostic and management challenge. *Ultrasound Q* 2008; 24: 225-240.

[11] Hoover K and Jenkins TR. Evaluation and management of adnexal mass in pregnancy. *Am J Obstet Gynecol* 2011; 205: 97-102.

[12] Simpson JL, Palomaki GE, Mercer B, Haddow JE, Andersen R, Sibai B and Elias S. Associations between adverse perinatal outcome and serially obtained second- and third-trimester maternal serum alpha-fetoprotein measurements. *Am J Obstet Gynecol* 1995; 173: 1742-1748.

[13] TOUTOU Y, DARBOIS Y, BOGDAN A, AUZEY A and KEUSSEOGLU S. Tumour marker antigens during menses and pregnancy. *Br J Cancer* 1989; 60: 419-420.

Ovarian torsion caused by hyperreactio luteinalis in the third trimester of pregnancy

- [14] Kupesic S and Plavsic BM. Adnexal torsion: color Doppler and three-dimensional ultrasound. *Abdom Imaging* 2010; 35: 602-606.
- [15] Nizar K, Deutsch M, Filmer S, Weizman B, Beloosesky R and Weiner Z. Doppler studies of the ovarian venous blood flow in the diagnosis of adnexal torsion. *J Clin Ultrasound* 2009; 37: 436-439.
- [16] Birchard KR, Brown MA, Hyslop WB, Firat Z and Semelka RC. MRI of acute abdominal and pelvic pain in pregnant patients. *AJR Am J Roentgenol* 2005; 184: 452-458.
- [17] Huchon C and Fauconnier A. Adnexal torsion: a literature review. *Eur J Obstet Gynecol Reprod Biol* 2010; 150: 8-12.
- [18] Brun JL, Fritel X, Aubard Y, Borghese B, Bourdel N, Chabbert-Buffet N, Collinet P, Deffieux X, Dubernard G, Huchon C, Kalfa N, Lahlou N, Marret H, Pienkowski C, Sevestre H, Thomas-sin-Naggara I, Levêque J; Collège National des Gynécologues Obstétriciens Français. Management of presumed benign ovarian tumors: updated French guidelines. *Eur J Obstet Gynecol Reprod Biol* 2014; 183: 52-58.
- [19] Sasaki KJ and Miller CE. Adnexal torsion: review of the literature. *J Minim Invasive Gynecol* 2014; 21: 196-202.
- [20] Palanivelu C, Rangarajan M, Senthilkumaran S and Parthasarathi R. Safety and efficacy of laparoscopic surgery in pregnancy: experience of a single institution. *J Laparoendosc Adv Surg Tech A* 2007; 17: 186-190.