Clinical Case Report

Medicine®

OPEN

Pregnancy with giant ovarian dysgerminoma

A case report and literature review

Xi-Wen Zhang, MM[a], Li-Rong Zhai, MM[b], Dong-Wei Huang, MM[c], Zhen-De Jiang, MD[d], Tong Yu, MD[d], Shu-Yan Liu, MD[e,*], Man-Hua Cui, MD[a,*]

Abstract

Rationale: Dysgerminoma is an extraordinarily rare neoplasm arising from the malignant germ cells of the ovary. Early antenatal diagnosis and proper management of the neoplasm to improve maternal-neonatal results are the considerable challenges facing the gyne-oncologist. We summarize the clinical features and discuss treatment strategies of the ovary dysgerminoma (OD). Besides, we also review the literature on OD in PubMed, Web of Science Core Collection, Library of Congress, and LISTA from 1939 to 2019 to evaluate its clinical characteristics, feto-maternal compromise, management, and fertility outcome.

Patient concerns: A 25-year-old pregnant woman reported lower abdominal pain and vomiting.

Diagnosis: The patient was diagnosed as right OD.

Interventions: She received a cesarean section due to severe abdominal pain, delivered a healthy girl at 38 C 4 weeks of gestation, and accepted fertility-preserving surgery. However, the patient refused chemotherapy postoperatively.

Outcomes: The patient was followed up 42 days, 3 months, and 6 months after surgery, and no tumor recurrence was observed.

Lessons: OD has non-specificity characteristics, including age, symptoms, image date, and tumor marks. However, these abnormal indicators may provide some evidence for accurate antenatal diagnosis. The management strategies should be considered comprehensively on an individual basis, and fertility-preserving surgery should be carried out in the second trimester if further pregnancy is desired. Adjuvant chemotherapy needs to be applied to the treatment of OD patients with The International Federation of Gynecology and Obstetrics (FIGO) stages II, III, and IV and timely chemotherapy is suggested if there are several weeks before the expected date of delivery. The overall prognosis of OD patients is excellent.

Abbreviations: AC = abdominal circumference, AFI = amniotic fluid index, AFP = α-fetoprotein, BEP = bleomycin-etoposide-cisplatin, BPD = biparietal diameter, CA = cancer antigen, CEA = carcinoembryonic antigen, CS = cesarean section, FIGO = The International Federation of Gynecology and Obstetrics, FL = femur length, HC = head circumference, HCG = human chorionic gonadotropin, IHC = immunohistochemical, LDH = lactic dehydrogenase, MGCT = malignant germ cell tumor, NSE = neuron-specific enolase, OD = ovarian dysgerminoma, SCC = squamous cell carcinoma antigen, TC = paclitaxel-carboplatin.

Keywords: chemotherapy, dysgerminoma, malignant germ cell tumor, pregnancy, pregnancy outcome, surgery, treatment

Editor: Maya Saranathan.

X-WZ and L-RZ have contributed equally to this work.

This research received no external funding.

Disclosure statement: None.

The authors have no conflicts of interest to disclose.

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

[a] Department of Gynecology, [b] Department of Gynecology and Obstetrics, Peking University People Hospital, Beijing, [c] Department of Pathology, [d] Department of Orthopaedics, [e] Department of Ophthalmology, The Second Hospital of Jilin University, Changchun, Jilin Province, China.

[*] Correspondence: Man-Hua Cui, Department of Gynecology and Obstetrics, The Second Hospital of Jilin University, 218 Ziqiang Road, Changchun, Jilin Province, China (e-mail: cuimanhuajlu@163.com); Shu-Yan Liu, Department of Ophthalmology, The Second Hospital of Jilin University, Changchun, Jilin Province, China (e-mail: 125787061@qq.com).

How to cite this article: Zhang XW, Zhai LR, Huang DW, Jiang ZD, Yu T, Liu SY, Cui MH. Pregnancy with giant ovarian dysgerminoma: A case report and literature review. Medicine 2020;99:41(e21214).

Received: 1 October 2019 / Received in final form: 26 May 2020 / Accepted: 9 June 2020

http://dx.doi.org/10.1097/MD.0000000000021214

1. Introduction

Malignant germ cell tumor (MGCT) is an extraordinary rare ovarian cancer, which occupies no >5% of all ovarian cancers[1–4] and 18% to 26% of all ovarian cancer with pregnancy.[5,6] MGCT mainly includes the following subtypes: ovary dysgerminoma (OD) (38.2%), yolk sac tumor (30.4%), and immature teratoma (15.7%).[2] OD is the most common subtype of MGCT and often occurs in adolescence and early adulthood.[1,7–10] In pregnant women, OD patients only account for about 0.0002% to 0.001%,[11] and OD usually has a unilateral onset and is diagnosed at an early stage. It is difficult to achieve a large sample of OD due to its relatively low incidence. Thus, more studies are needed to summarize the clinical features and determine the optimal management strategies of OD. Furthermore, OD associated with mental retardation in pregnant women is even rarer. Therefore, the purpose of this study is to report our seldom case, as well as to review the literature on OD features, differential diagnosis, management strategies, and prognosis of pregnant patient with OD.

2. Ethic

This case report was approved by the institutional review board of the second hospital of Jilin University. Informed written consent was obtained from the patient for publication of this case report and accompanying images.

3. Methods

We report a case of OD with mental retardation and review relevant literature in PubMed, Web of Science Core Collection, Library of Congress, and LISTA from 1939 to 2019 (Table 1).

4. Case report

A 25-year-old pregnant woman with mental retardation who had abdominal pain and vomiting for 7 hours was transferred to our department. The previous history was gravida 1, para 0, without surgery history. Her initial prenatal examination was performed at 12 weeks of gestation. The ultrasound indicated pregnancy status and revealed a large mass in the pelvic cavity. The regular ultrasound examination during pregnancy revealed that the volume of the mass increased gradually. At 30 C 2 weeks of gestation, the ultrasound revealed cephalic presentation. The biparietal diameter (BPD) was 6.9 cm, the head circumference (HC) was 26.6 cm, the abdominal circumference (AC) was 24.7 cm, and the femur length (FL) was 5.6 cm. The posterior wall of the placenta was grade I and the lower margin was 1.6 cm from the inner cervix. The amniotic fluid index (AFI) was 16.1. The ultrasound also revealed a hypoechoic mass in the lower part of the posterior wall of the uterus with a size of 14.8 cm × 8.5 cm. At 38 C 4 weeks of gestation, the ultrasound before admission of the patient revealed cephalic presentation. The BPD was 7.8 cm, HC was 31.5 cm, AC was 32.9 cm, and FL was 6.8 cm. The right wall of the placenta was late grade II and AFI was 12.2 cm. A U-shaped impression was found on the neck of the fetus. The ultrasound also revealed a hypoechoic mass located at the right rear of the uterus with a size of 23.0 cm × 12.5 cm (Fig. 1). Some of her tumor markers were positive. Human chorionic gonadotropin (HCG) was 14,333.94 mIU/mL (0–5 mIU/mL), the α-fetoprotein (AFP) was 142.59 ng/mL (0–8.78 ng/mL), Cancer antigen (CA)-125 was 148.10 U/mL (0–35 U/mL), CA-199 was 610.46 U/mL (0–37 U/mL), CA-50 was 59.10 U/mL (0–20 U/mL), Cytokeratin 19 fragment was 4.86 ng/mL (0–2.08 ng/mL), and neuron-specific enolase (NSE) was 76.04 ng/mL (0–15 ng/mL). Conversely, some of her tumor markers were negative, such as carcinoembryonic antigen (CEA), CA-153, and squamous cell carcinoma antigen (SCC).

On abdominal examination, the uterine fundal height was 33 cm and the abdominal circumference was 98 cm. The abdominal tenderness was positive, especially in the right lower abdomen, and the rebound tenderness was also positive. The patient could not cooperate in the other examination.

Termination of pregnancy was performed due to severe abdominal pain. She delivered a 2540 g healthy girl with a 1-minute Apgar score of 9 by cesarean section (CS) and a 10-minute Apgar score of 10 by CS.

Intraoperatively, we found a large solid mass of 25 cm × 19 cm × 24 cm, which originated from the right ovary, with a moderate amount of pale-yellow ascites. The tumor was substantially lobulated, the texture was soft, the surface was intact, and the tissue was crunchy. Large blood vessels were visible, and the boundary between the tumor and adjacent organs (the right lining of the uterus, the rectal serosa) was not clear. No abnormalities in the appearance of the ovaries and fallopian tubes were found. At sectioning (Fig. 2), the mass was grayish-white, grayish-yellow, grayish red, and homogeneous. The tumor was almost solid, while some areas were soft, of which the density was similar to brain medulla. No enlarged lymph nodes were found in the pelvis and abdominal cavity. The right fallopian tube was 7 cm long and 0.3 to 0.7 cm in diameter. Tumor biopsy and contralateral ovarian biopsy were conducted primarily.

The pathological results of the intraoperative frozen section showed right adnexa dysgerminoma; left ovarian biopsy showed no tumor, but localized old bleeding and interstitial fibrosis. Fertility-preserving surgery, including giant tumor and right adnexa resection, omentectomy, appendectomy, pelvic lymphadenectomy, abdominal aortic lymph node biopsy, was performed.

The final pathological results showed dysgerminoma (Fig. 3A–D) of the right adnexa, but no tumor metastasis was found in the right fallopian tube, left ovary, appendix, omentum, and pelvic lymph nodes. Immunohistochemical (IHC) results were as follows: D2–40, CD117, PLAP, and SALL-4 were positive (Figs. 4 and 5); CK (AE1/AE3), Vimentin, epithelial membrane antigen (EMA), estrogen receptor (ER), progesterone receptor (PR), Alpha fetoprotein (AFP), and Glypican-3 were negative, and the positive index of Ki67 was 70% (Fig. 6). The final clinical diagnosis of the patient was OD, stage IIB (according to the 2014 FIGO staging system).

The patient's postoperative vital signs were stable, and the incision healed well. However, the patient refused chemotherapy postoperatively. The follow-up results of the patient 42 days, 3 months, and 6 months after the surgery showed no tumor recurrence.

5. Discussion

OD is the most common subtype of MGCT, which originates from ovarian primordial germ cells. It often occurs in adolescence and early adulthood, but has been found only about 0.0002% to 0.001% of pregnant women.[11] It is hard to achieve a large sample of OD due to its extraordinary low incidence. Therefore, this paper is aimed to report our rare case, as well as to review the relevant literature summarizing the features, differential diagnosis, management strategies, and prognosis of pregnant patients with OD.

5.1. Features of ovarian dysgerminoma

OD can occur in women aged from 7 months to 70 years,[31] but predominantly in young pregnant women.[1,7,8,30,32] The majority of OD pregnant women usually have non-specific symptoms,[33] including the most common abdominal pain (35.3%), followed by abdominal distention (19.6%), a growing mass (19.6%), multiple symptoms (18.6%), and non-symptoms (21.6%).[2] In our study, abdominal pain was the main complaint of the patient and led to a cesarean section.

Considering the gross pathologic features of OD, it usually presents well encapsulated and characteristically solid, with a diameter range from 8 to 15 cm.[25,31] At sectioning, the tissue is lobulated, soft, fleshy, and gray-white or light tan. Occasionally, areas of hemorrhage and coagulative necrosis, which are typically related to cystic changes, can be observed. OD is most commonly unilateral in pregnancy, accounting for approximately 95%,[1] while only 5% to 20% is bilateral.[1,34–36] In our study, the tumor was unilateral and showed substantially lobulated soft texture and the entire surface. This finding was consistent with the previous literature.[1,25,31–33]

Regarding the microscopic pathologic feature of OD, it is like that of testicular seminomas. OD is composed of round cells with

Table 1

Clinical features in 34 cases of ovarian dysgerminoma in pregnancy.

Year	Author	Age, y	Gravida, parity	Size, cm, side	Delivery GA weeks	Symptoms	Delivery mode	Treatment	FIGO stage	Fetal outcome	Maternal prognosis
1939	Dockerty and MacCarty,[12]	29	–	12 × 10 × 10, Right	Term	AM by palpation	–	Ovarian tumor resection	IA (unstaged)	Terminated normally	A/W 8 months
1947	Schneider, H[13]	23	G2, P1	13 × 13 × 6, Left	39+2	AP	CS	LSO, at CS	IA (unstaged)	2892g, healthy	–
1956	Watson, SL[14]	25	G2, P1	14 × 12 × 11, Right	Term		CS	Oophorectomy, at CS	IA (unstaged)	healthy	A/W 32 months
1962	Kawahara, H[15]	27	G1, P1	17 × 16 × 8, Left	None	AM by palpation	None	Ovarian tumor resection + RTY	IA (unstaged)	None	A/W 12 months
1964	Pece, G[16]	26	P2	26 × 23 × 19, Right	40	None	VD	RSO, 2 months after VD + CTY	IIIC (unstaged)	3000g, Hare lip	NA
1966	Smith, AH[17]	18	P0	15 × 14 × 7, Left	42	Obstructed labor	CS	BSO, at CS + RTY	IIIB (unstaged)	3544 g	Dead, 10 weeks
1979	Karlen, JR[18]	20	P0	28, Left	36	AP	CS	TAH+BSO+AE+OE at CS	IB	2730g, healthy	A/W 3 years
1985	Lelle, RJ[19]	36	P1	Left	None	AM by ultrosound	None	Hysterectomy+bilateral adnectomy + OE + RTY	IA (unstaged)	none	A/W
1985	Rabinowitz, R[20]	22	P0	5, Right	33	None	CS	RSO, at CS	IA (unstaged)	2900g, dead of Potter's Syndrome	A/W 2 years
1985	Rabinowitz, R[20]	26	P2	6, Right	39	None	CS	RSO, at CS	IA (unstaged)	2750g, healthy	A/W 3 years
1986	Kulenthran, A[21]	17	P0	5 × 8 × 6, Right	40+5	None	VD	RSO, 24+4 weeks	IA (unstaged)	2950g, healthy	A/W 15 years
1986	Kulenthran, A[21]	21	P1	20 × 18 × 15, Left	34	None	VD	LSO, 3rd day after VD TAH+BSO+AE+OE, after 5 cycles of CTY	IA	1900 g	A/W 1 year
1992	Buller RE[22]	26	P0	16; –	Term	None	VD	Oophorectomy, 25 weeks	IA (unstaged)	–	A/W 2 years
1992	Buller RE[22]	28	P0	13 × 9 × 7, Right	38	None	VD	Stage surgery, 10 weeks	IA	healthy	A/W 7 months
1992	Buller RE[22]	21	P2	–, Right	38	None	VD	RSO C OE C left ovary biopsy 26 weeks + CTY	IV	2,320g, IUGR	A/W 9 months
1996	Ueda, M[23]	30	P1	Left	None	–	CS	LSO, 13 weeks TAH+RSO, 15 weeks + RTY	IB	None	A/W 10 years
2002	Sayedur Rahman, M[24]	32	P5	–	–	AP		RSO	IA	–	A/W 5 years
2002	Sayedur Rahman, M[24]	44	P10	–	–	Retained second twin		TAH + BSO + OE	IIA	–	A/W 6 years
2002	Sayedur Rahman, M[24]	21	P0	–	–	AP, vomiting	–	LSO at 13 weeks	IA	–	A/W 14 years
2002	Sayedur Rahman, M[24]	30	4	–	–	AP	–	Ovarian cystectomy, TAH + BSO + OE, + CTY	IC	–	A/W 16 years
2002	Sayedur Rahman, M[24]	42	7	–	–	AP	CS	Debulking + OE + CTY	III	–	Dead, 13 months
2002	Sayedur Rahman, M[24]	34	6	–	–	AP, vomiting	–	LSO at 21 weeks	IA	–	A/W 16 years
2002	Sayedur Rahman, M[24]	30	3	–	–	None	CS	RSO	IA	–	A/W 18 years
2002	Sayedur Rahman, M[24]	36	7	–	–	Asymptomatic	–	TAH + BSO + CTY	IC	–	A/W 15 months
2002	Sayedur Rahman, M[24]	18	0	–	–	AP, vomiting	–	RSO at 24 weeks + RTY	IA	–	A/W 5 years
2007	Hubalek, M[25]	33	P0	18 × 12 × 8, Right	35+3	None	CS	TAH+BSO+OE+SLA, at CS	IC	2450g, healthy	A/W 22 months
2010	Gauza, JE[26]	25	P0	23 × 18 × 11, Left	39	AP	CS	LSO, at CS OE + peritoneal biopsy + lavage, 45 days after CS	IA	2700g, healthy	–
2011	Akhtar, K[27]	22	P0	35 × 25, Right	32	Abdominal distention	CS	RSO, at CS + CTY	IA (unstaged)	healthy	A/W 2 years
2011	Gezginç, K[28]	32	P1	30	36	Abdominal distention	CS	Bilateral ovarian biopsy, at CS + CTY	IV	1700g, IUGR	Dead 10 months
2012	Montesinos, L[29]	24	P0	14, Right	–	AP	Ectopin pregnancy	RSO + left salpingostomy, 1st trimester	IA (unstaged)	None	A/W 7 years

(*continued*)

Table 1
(continued).

Year	Author	Age, y	Gravida, parity	Size, cm, side	Delivery GA weeks	Symptoms	Delivery mode	Treatment	FIGO stage	Fetal outcome	Maternal prognosis
2016	Gupta, M[9]	28	G2, P1	15 × 9 (Left) 3 × 5 (Right)	35+3	None	CS	Bilateral salpingostomy + tumors resection, at CS, +,CTY	IB (unstaged)	2220 g, healthy	A/W
2018	Chen, Y[1]	23	P1	18 × 13 × 10, Right	34+6	None	CS	RSO + retroperitoneal mass rection, at CS	IIB	2200 g, healthy	A/W 6 months
2019	Ni Luh, LCP[30]	24	G3	19 × 16 × 13, Left	35	Dyspena with a swollen abdomen	VD	LSO + OE + CTY + Staging	IC	2100 g, healthy	A/W 3 months
–	Our case	25	G1, P0	29 × 19 × 24, Right	38+4	AP, vomiting	CS	RSO + OE + AE + pelvic lymphadenectomy + abdominal aortic lymph node biopsy	IIB	2540 g, healthy	A/W 3 months

The International Federation of Gynecology and Obstetrics (FIGO) staging system is used for staging.
"–" = not available, A/W = alive and well, AE = appendectomy, AM = abdominal mass, AP = abdominal pain, BSO = bilateral salpingo-oophorectomy, CS = cesarean section, CTY = chemotherapy, EP = ectopic pregnancy, GA = gestational age, IUGR = intrauterine growth restriction, LSO = left salpingo-oophorectomy, OE = omentectomy, RSO = right salpingo-oophorectomy, RTY = radiation therapy, SLA = systematic pelvic and paraaortic lymphadenectomy, TAH = total abdominal hysterectomy, VD = vaginal delivery.

Figure 1. Ultrasound showed a hypoechoic mass behind the uterus, about 23.0 × 12.5 cm in size.

a uniform population, which is usually infiltrated by T lymphocytes and separated by fibrous strands. A large round or flattened nucleus that contains one or a few prominent nucleoli and clear eosinophilic cytoplasm can be observed in the center of cells. In addition, mitoses are always in large quantities.[31]

Regarding the imaging features of OD, it is characterized by pure solids. In ultrasonography, they show well-defined borders, smooth lobulated contours, and component lobules, with heterogeneous echogenicity. At Doppler ultrasonography, they are abundantly vascularized at power and color.[37–39] In our study, ultrasound results show unclear boundaries, component lobules, with heterogeneous echogenicity. This feature suggests that the mass may be malignant. At CT, the lobular pattern may also be observed with a predominantly solid tumor accompanied by enhancing septa and areas of cystic change.[38,40] Kim and Kang[38] claimed that calcification might be shown as a speckle. In magnetic resonance imaging (MRI), the most characteristic appearance is a solid mass, which is divided into lobules by fibrovascular septa. On T2-weighted images, the signal intensity is isointense or slightly hyperintense. On T1-weighted images, the signal intensity of OD is lower than that of muscles. Kitajima et al[41] described that the MRI features of epithelial ovarian neoplasms were similar to those of multilocular cystic masses with irregular septations. Unfortunately, this patient did not undergo CT and MRI examinations during the hospitalization.

Figure 2. (A and B) Gross appearance of tumors resected. (C) Tumor biopsy. (D) Contralateral ovarian biopsy.

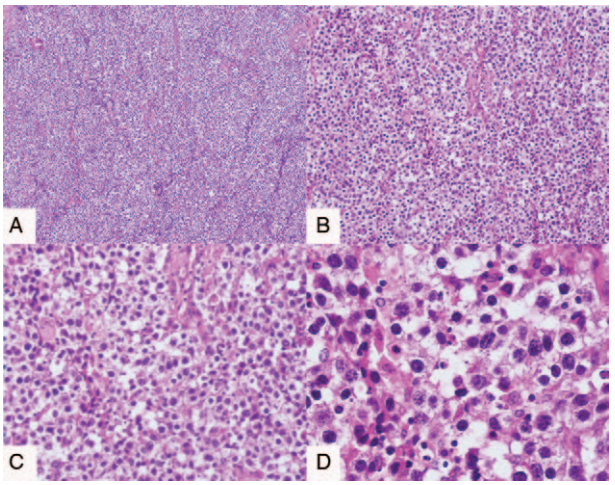

Figure 3. Hematoxylin-eosin (HE) staining results of the adnexa dysgerminoma, consist of round to atypical oval cell separated with complex thin fibrous tissue septal network which has rich lymphocyte infiltrate. (A ×40, B ×100, C ×200, D ×400).

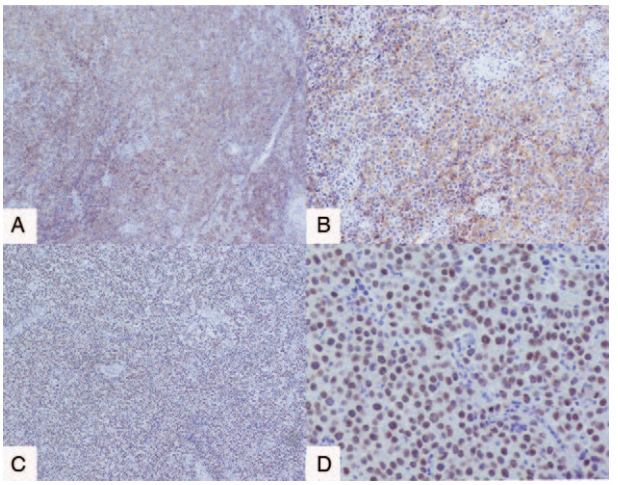

Figure 5. Immunohistochemical staining results of the adnexa dysgerminoma. (A and B) PLAP staining was positive (A, PLAP, ×40, B, PLAP, ×100). (C and D) SALL4 staining was positive (C, SALL4, ×40, D, SALL4, ×100).

Considering the tumor marks of OD, CA125, and NSE may provide reliable evidence in OD.[42] Literature reported that high levels of serum CA125 rapidly fell after chemotherapy.[25] Previous studies described that partly OD patients exhibited increased NSE content and positive NSE of IHC[43,44] The serum levels and IHC expression of NSE in pediatric patients with OD may be of value in patient monitoring.[42] In this study, CA125 and NSE increased significantly preoperatively. Some other indicators are abnormal, including HCG, AFP, CA-199, and CA-50. We hope that these positive indicators can provide some help for other scholars to diagnose OD accurately. Besides, LDH is another reliable indicator for predicting the effect of chemotherapeutic intervention.[25,45]

5.2. Differential diagnosis of ovarian dysgerminoma

OD has nonspecific features, which lead to the difficulty in making an accurate diagnosis. However, the age of patients, the imaging features of the neoplasm, and the abnormal tumor markers may help to determine a correct differential diagnosis. In general, OD should be distinguished from other purely solid masses of ovarian, including fibrosarcomas, granulosa cell tumors, Brenner tumors, epithelial ovarian, and metastatic carcinomas.[46]

5.3. Treatment strategies of ovarian dysgerminoma

With regard to surgery treatment of OD, accurate surgical staging is relatively critical to determination of the reasonable and accurate risk-based management. Currently, the FIGO classification is the most accepted method.[47] OD staged IA-C could achieve acceptable surveillance by fertility-sparing unilateral salpingo-oophorectom.[31] Bilateral salpingo-oophorectomy and hysterectomy are recommended for stage II and III diseases. In addition, if tumors do not invade the contralateral reproduction organs, unilateral salpingo-oophorectomy can be considered. The management strategies of stage IV patients mainly include fertility-sparing surgery, cytoreduction, and adjuvant chemotherapy.[48,49] Regarding second-look surgery of OD, if the tumor contains teratomatous elements or has residual disease, patients may benefit from second-look surgery after initial cytoreductive surgery and chemotherapy.[48,50] However, if the tumor does not have a teratomatous element, <5 cm of residual disease, or normal tumor marker levels after chemotherapy, second-look surgery is not recommended.[50]

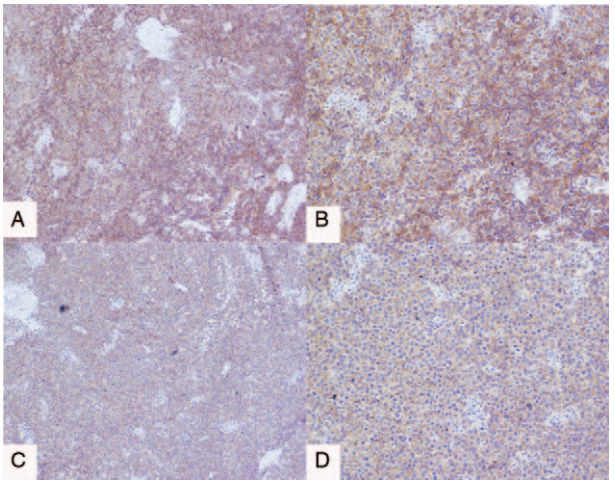

Figure 4. Immunohistochemical staining results of the adnexa dysgerminoma. (A and B) D2–40 staining was positive (A, D2–40, ×40, B, D2–40, ×100). (C and D) CD177 staining was positive (C, CD177, ×40, D, CD177, ×100).

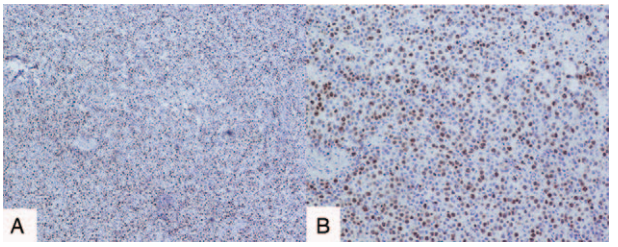

Figure 6. Immunohistochemical staining results showed that the positive index of Ki67 was 70%.

In this study, the patient's mental retardation and lack of awareness of contraception may lead to repregnancy and increase the family burden. Thus, the patient's guardian strongly requests hysterectomy and bilateral appendectomy for the patient. However, the patient does not meet the indications for hysterectomy and bilateral appendectomy according to the FIGO stage, age, and grade of malignancy. Also, in China, especially in rural areas, women who have lost fertility function are not competitive in the remarriage population. What is worse, she has mental retardation, which means that it is difficult for the patient to reconstitute a family once she divorces after hysterectomy and appendectomy. Therefore, after careful consideration, we performed fertility-preserving surgery for the patient. The patient showed a satisfactory treatment effect during the follow-up visit.

With regard to chemotherapy of OD, OD with FIGO stages II, III, and IV are indicated for chemotherapy.[47] Chemotherapy is recommended based on pathological evidence,[1] especially in cases with advanced-stage tumors, mixed epithelial and germ cell tumors, large tumor size, and rapidly increasing ascites. To date, platinum-based chemotherapy is the main strategy, including paclitaxel-carboplatin (TC) and bleomycin-etoposide-cisplatin (BEP).[25,51–56] In 2004, Hubalek et al[25] claimed that TC could elicit an excellent response and posed no adverse impacts on the fetus. BEP is usually applied to the treatment of nonepithelial ovarian tumors of nonpregnant patients. However, the incidence of adverse advents (plagiocephaly, fetal ventriculomegaly with cerebral atrophy, hearing loss, and syndactyly) of etoposide is high.[57–60] Therefore, in pregnancy, paclitaxel-carboplatin chemotherapy instead of BEP is an optimized scheme for the treatment of nonepithelial ovarian cancer.[61] The influence of chemotherapy during pregnancy on maternal survival must be considered. Literature[62,63] reported that chemotherapy during the first trimester could increase the incidence of fetal death, abortion, and malformations. Furthermore, the study also showed that the central nervous system, hemopoietic system, the eyes, and genitals were still vulnerable to sustained exposure to antineoplastic agents after organogenesis.[64] However, increasing evidence suggests that chemotherapy for the second and third trimesters is relatively safe.[65]

In our study, the patient was diagnosed as OD staged II B, and chemotherapy was recommended by gynecologists postoperatively. However, she refused. Optimistically, there was no recurrence during the follow-up period of 6 months. We attribute this positive outcome partly to the low malignancy of the tumor and the standard and thorough operation carried out by a gynecologist with >30 years of experience, and partly to the short follow-up period.

5.4. Prognosis of ovarian dysgerminoma

Residual disease, tumor markers, the FIGO stage, and the volume of the residual tumor are all the critical factors of prognosis.[48] Besides, age over 45 years is also a significant predictor of recurrence.[49] In most cases, tumors are detected early, which contribute to accurate prognosis.[66] The prognosis of early-stage OD patients is excellent,[48,49,67] and the overall 5-year survival rate is approximately 100%.

6. Conclusion

In conclusion, features of OD, including age, symptoms, images date, and tumor marks, have non-specificity. However, these abnormal indicators may provide some evidence for accurate antenatal diagnosis. The management strategies should be considered comprehensively on an individual basis, and fertility-preserving surgery should be carried out in the second trimester if further pregnancy is desired. Adjuvant chemotherapy needs to be applied to the treatment of OD with FIGO stages II, III, and IV. If there are several weeks before the expected date of delivery, timely chemotherapy is indicated. The overall prognosis of OD patients is excellent.

Author contributions

Conceptualization: Tong Yu, Shu-Yan Liu, Man-Hua Cui.
Data curation: Xi-Wen Zhang, Li-Rong Zhai, Dong-Wei Huang, Zhen-De Jiang.
Formal analysis: Xi-Wen Zhang, Dong-Wei Huang, Zhen-De Jiang.
Methodology: Li-Rong Zhai, Zhen-De Jiang, Shu-Yan Liu.
Resources: Dong-Wei Huang.
Supervision: Tong Yu, Man-Hua Cui.
Writing – original draft: Xi-Wen Zhang, Li-Rong Zhai.
Writing – review & editing: Tong Yu, Shu-Yan Liu, Man-Hua Cui.

References

[1] Chen Y, Luo Y, Han C, et al. Ovarian dysgerminoma in pregnancy: a case report and literature review. Cancer Biol Ther 2018;19:649–58.
[2] Kodama M, Grubbs BH, Blake EA, et al. Feto-maternal outcomes of pregnancy complicated by ovarian malignant germ cell tumor: a systematic review of literature. Eur J Obstet Gynecol Reprod Biol 2014;181:145–56.
[3] Quirk JT, Natarajan N, Mettlin CJ. Age-specific ovarian cancer incidence rate patterns in the United States. Gynecol Oncol 2005;99: 248–50.
[4] Parker SL, Tong T, Bolden S, et al. Cancer statistics, 1997. CA Cancer J Clin 1997;47:5–27.
[5] Aggarwal P, Kehoe S. Ovarian tumours in pregnancy: a literature review. Eur J Obstet Gynecol Reprod Biol 2011;155:119–24.
[6] Gezginc K, Karatayli R, Yazici F, et al. Ovarian cancer during pregnancy. Int J Gynaecol Obstet 2011;115:140–3.
[7] Samiee-rad F, Zangivand AA. The coexistence of ovarian dysgerminoma and normal intra-uterine pregnancy presented with acute abdominal pain. Comparat Clin Pathol 2018;27:257–60.
[8] Ngwenya S. Ovarian dysgerminoma presenting as a pregnancy: case report. Trop Doct 2017;47:69–71.
[9] Gupta M, Jindal R, Saini V. An incidental finding of bilateral dysgerminoma during cesarean section: Dilemmas in management. J Clin Diagn Res 2016;10:QD04–5.
[10] Kamal NM, Khan U, Mirza S, et al. Ovarian dysgerminoma with normal serum tumour markers presenting in a child with precocious puberty. J Cancer Res Ther 2015;11:661.
[11] Smith HO, Berwick M, Verschraegen CF, et al. Incidence and survival rates for female malignant germ cell tumors. Obstet Gynecol 2006;107:1075–85.
[12] Dockerty MB, MacCarty WC. Dysgerminoma. With a report of nine cases, one associated with pregnancy. Am J Obstet Gynecol 1939;37:878–85.
[13] Schneider H, Vesell M. Dysgerminoma and pregnancy. Am J Obstet Gynecol 1947;53:688–91.
[14] Watson SL. Dysgerminoma complicating labor. Am J Obstet Gynecol 1956;72:1177–9.
[15] Kawahara H. Dysgerminoma complicating pregnancy. Am J Obstet Gynecol 1962;83:1531–3.
[16] Pece G. Dysgerminoma of the ovary in pregnancy: report of a case and review of the literature. Obstet Gynecol 1964;24:768–73.
[17] Smith AH, Ward SV. Dysgerminoma in pregnancy: report of a case. Obstet Gynecol 1966;28:502–4.
[18] Karlen JR, Akbari A, Cook WA. Dysgerminoma associated with pregnancy. Obstet Gynecol 1979;53:330–5.

[19] Lelle RJ, Majewski A. Dysgerminoma of the ovary in pregnancy - case report. Geburtshilfe und Frauenheilkunde 1985;45:815–6.
[20] Rabinowitz R, Granat M. Dysgerminoma of the ovary: incidental finding during cesarean section. Eur J Obstet Gynecol Reprod Biol 1985;19:105–8.
[21] Kulenthran A, Sivanesaratnam V. Dysgerminoma of the ovary associated with pregnancy. Asia Oceania J Obstet Gynaecol 1986;12:217–20.
[22] Buller RE, Manetta DV, Porto A, et al. Conservative surgical management of dysgerminoma concomitant with pregnancy. Obstet Gynecol 1992;79:887–90.
[23] Ueda M, Ueki M. Ovarian tumors associated with pregnancy. Int J Gynecol Obstet 1996;55:59–65.
[24] Sayedur Rahman M, Al-Sibai MH, Rahman J, et al. Ovarian carcinoma associated with pregnancy. A review of 9 cases. Acta Obstet GynecolScand 2002;81:260–4.
[25] Hubalek M, Smekal-Schindelwig C, Zeimet AG, et al. Chemotherapeutic treatment of a pregnant patient with ovarian dysgerminoma. Arch Gynecol Obstet 2007;276:179–83.
[26] GAUZA JE. Diagnosis of ovarian dysgerminoma during pregnancy. pdf>. 2010.
[27] Akhtar K, Shamshad Ahmad S, Kumar A, et al. Dysgerminoma with pregnancy and viable baby: a case report. Oman Med J 2011;26: 198–200.
[28] Gezginç K, Karatayli R, Yazici F, et al. Ovarian cancer during pregnancy. Int J Gynaecol Obstet 2011;115:140–3.
[29] Montesinos L, Acién P, Martínez-Beltrán M, et al. Ovarian dysgerminoma and synchronic contralateral tubal pregnancy followed by normal intra-uterine gestation: a case report. J Med Case Rep 2012;6:399.
[30] Luh LCPN, Mahendra INB, Suwiyoga K, et al. Management comprehensive multidisciplinary of malignant ovarian germ cell tumors and Feto - Maternal outcome: a case series report and literature review. Open Access Maced J Med Sci 2019;7:1174–9.
[31] Shaaban AM, Rezvani M, Elsayes KM, et al. Ovarian malignant germ cell tumors: cellular classification and clinical and imaging features. Radiographics 2014;34:777–801.
[32] Popović J, Pop-Trajković S, Stefanović M, et al. Dysgerminoma and pregnancy. Srpski Arhiv za Celokupno Lekarstvo 2017;145:526–9.
[33] Major T, Borsos A, Lampe L, et al. Ovarian malignancies in childhood and adolescence. Eur J Obstet Gynecol Reprod Biol 1995;63:65–8.
[34] Mahdi H, Kumar S, Seward S, et al. Prognostic impact of laterality in malignant ovarian germ cell tumors. Int J Gynecol Cancer 2011;21:257–62.
[35] Montesinos L, Acien P, Martinez-Beltran M, et al. Ovarian dysgerminoma and synchronic contralateral tubal pregnancy followed by normal intra-uterine gestation: a case report. J Med Case Rep 2012;6:399.
[36] Gupta M, Jindal R, Saini V. An incidental finding of bilateral dysgerminoma during cesarean section: dilemmas in management. J Clin Diagn Res 2016;10:QD04–5.
[37] Guerriero S, Testa AC, Timmerman D, et al. Imaging of gynecological disease (6): clinical and ultrasound characteristics of ovarian dysgerminoma. Ultrasound Obstet Gynecol 2011;37:596–602.
[38] Kim SH, Kang SB. Ovarian dysgerminoma: color Doppler ultrasonographic findings and comparison with CT and MR imaging findings. J Ultrasound Med 1995;14:843–8.
[39] Lazebnik N, Balog A, Bennett S, et al. Ovarian dysgerminoma: a challenging clinical and sonographic diagnosis. J Ultrasound Med 2009;28:1409–15.
[40] Tanaka YO, Kurosaki Y, Nishida M, et al. Ovarian dysgerminoma: MR and CT appearance. J Comput Assist Tomogr 1994;18:443–8.
[41] Kitajima K, Hayashi M, Kuwata Y, et al. MRI appearances of ovarian dysgerminoma. Eur J Radiol Extra 2007;61:23–5.
[42] Tatekawa Y, Kemmotsu H, Mouri T, et al. A case of pediatric ovarian dysgerminoma associated with high serum levels and positive immunohistochemical staining of neuron-specific enolase. J Pediatr Surg 2004;39:1437–9.
[43] Kawata M, Sekiya S, Hatakeyama R, et al. Neuron-specific enolase as a serum marker for immature teratoma and dysgerminoma. Gynecol Oncol 1989;32:191–7.
[44] Yoshida M, Koshiyama M, Konishi M, et al. Ovarian dysgerminoma showing high serum levels and positive immunostaining of placental alkaline phosphatase and neuron-specific enolase associated with elevation of serum prolactin level. Eur J Obstet Gynecol Reprod Biol 1998;81:123–8.
[45] Gemma R, Suzuki Y, Tanaka I, et al. Lactate dehydrogenase (LDH)-linked immunoglobulin in a patient with Graves' disease treated with methimazole. Intern Med 1992;31:377–9.
[46] Imaoka I, Wada A, Kaji Y, et al. Developing an MR imaging strategy for diagnosis of ovarian masses. Radiographics 2006;26:1431–48.
[47] Benedet JL, Bender H, Jones H3rd, et al. FIGO staging classifications and clinical practice guidelines in the management of gynecologic cancers. FIGO Committee on Gynecologic Oncology. Int J Gynaecol Obstet 2000;70:209–62.
[48] Gershenson DM. Management of ovarian germ cell tumors. J Clin Oncol 2007;25:2938–43.
[49] Gershenson DM. Current advances in the management of malignant germ cell and sex cord-stromal tumors of the ovary. Gynecol Oncol 2012;125:515–7.
[50] Pectasides D, Pectasides E, Kassanos D. Germ cell tumors of the ovary. Cancer Treat Rev 2008;34:427–41.
[51] Gauza JE, Reberti AG, Silva JC, et al. Diagnosis of ovarian dysgerminoma during pregnancy. Rev Assoc Med Bras (1992) 2010;56:517–9.
[52] Ruiz Ramos J, Roma E, Palomar L, et al. Paclitaxel and carboplatin treatment for advanced ovarian cancer during pregnancy. Chemotherapy 2013;59:344–5.
[53] Barut A, Arikan I, Barut F, et al. Ovarian cancer during pregnancy. J Pak Med Assoc 2011;61:914–6.
[54] Doi D, Boh Y, Konishi H, et al. Combined chemotherapy with paclitaxel and carboplatin for mucinous cystadenocarcinoma of the ovary during pregnancy. Arch Gynecol Obstet 2009;280:633–6.
[55] Motegi M, Takakura S, Takano H, et al. Adjuvant chemotherapy in a pregnant woman with endodermal sinus tumor of the ovary. Obstet Gynecol 2007;109(2 pt 2):537–40.
[56] Tabata T, Nishiura K, Tanida K, et al. Carboplatin chemotherapy in a pregnant patient with undifferentiated ovarian carcinoma: case report and review of the literature. Int J Gynecol Cancer 2008;18:181–4.
[57] Cardonick E, Usmani A, Ghaffar S. Perinatal outcomes of a pregnancy complicated by cancer, including neonatal follow-up after in utero exposure to chemotherapy: results of an international registry. Am J Clin Oncol 2010;33:221–8.
[58] Elit L, Bocking A, Kenyon C, et al. An endodermal sinus tumor diagnosed in pregnancy: case report and review of the literature. Gynecol Oncol 1999;72:123–7.
[59] Karimi Zarchi M, Behtash N, Modares Gilani M. Good pregnancy outcome after prenatal exposure to bleomycin, etoposide and cisplatin for ovarian immature teratoma: a case report and literature review. Arch Gynecol Obstet 2008;277:75–8.
[60] Kluetz PG, Edelman MJ. Successful treatment of small cell lung cancer during pregnancy. Lung Cancer 2008;61:129–30.
[61] Amant F, Halaska MJ, Fumagalli M, et al. Gynecologic cancers in pregnancy: guidelines of a second international consensus meeting. Int J Gynecol Cancer 2014;24:394–403.
[62] Doll DC, Ringenberg QS, Yarbro JW. Antineoplastic agents and pregnancy. Semin Oncol 1989;16:337–46.
[63] Zemlickis D, Lishner M, Degendorfer P, et al. Fetal outcome after in utero exposure to cancer chemotherapy. Arch Intern Med 1992;152: 573–6.
[64] Williams SF, Bitran JD. Cancer and pregnancy. Clin Perinatol 1985;12:609–23.
[65] Cardonick E, Iacobucci A. Use of chemotherapy during human pregnancy. Lancet Oncol 2004;5:283–91.
[66] Sayedur Rahman M, Al-Sibai MH, Rahman J, et al. Ovarian carcinoma associated with pregnancy. A review of 9 cases. Acta Obstet Gynecol Scand 2002;81:260–4.
[67] Dimopoulos MA, Papadimitriou C, Hamilos G, et al. Treatment of ovarian germ cell tumors with a 3-day bleomycin, etoposide, and cisplatin regimen: a prospective multicenter study. Gynecol Oncol 2004;95:695–700.