IMR Press

Original Research

Ultrasonic diagnosis and clinical management of singleton angular pregnancy: clinical study of a case series

Ling Zhang[1,*], Suhua Chen[1,*], Wei Li[1], Rong Liu[1], Mingfu Wu[1], Shuangmei Ye[1], Shuping Ding[1], Yihong Yang[1]

[1] *Department of Obstetrics and Gynecology, Tongji Hospital, Tongji Medical College, Huazhong University of Science and Technology, 430030 Wuhan, Hubei, China*

*Correspondence: zhanglingo709@tjh.tjmu.edu.cn (Ling Zhang); 13397182271@163.com (Suhua Chen)

DOI:10.31083/j.ceog4806212

Submitted: 31 March 2021 Revised: 14 May 2021 Accepted: 9 June 2021 Published: 15 December 2021

Background: To describe the fine ultrasonic diagnostic criteria and clinical management of different types of singleton angular pregnancy. *Methods*: Sixty cases of angular pregnancy were collected in a single Department of Obstetrics and Gynecology from January 2016 to July 2020. The general medical history, ultrasonic images, pregnancy outcomes, surgical records, clinical management, pathological examination results and postoperative ultrasound images were collected to analyze the related risk factors, clinical manifestation, fine ultrasonic diagnostic criteria, clinical management, outcomes, and complications. *Results*: Among the 60 cases, 46 cases (76.7%) had related risk factors and 14 (23.3%) did not. Twenty-five cases (41.6%) had clinical symptoms of vaginal bleeding with or without lower abdominal pain and 35 cases (58.4%) had no symptoms of an abnormal pregnancy. Fifty-nine cases (98.3%) were diagnosed as different types of angular pregnancy. The number of cases of type I, II and III angular pregnancy cases was 42 (71.2%), 13 (22.0%) and 4 (6.8%), according to the gold standard diagnosis of our research. Ultrasound sensitivity in the diagnosis of type I, II and III angular pregnancy in the first trimester was 83.3%, 69.2% and 50.0%. Fifty-six cases (93.3%) resulted in a favorable outcome, while 4 cases (6.7%) showed complications. *Conclusions*: The different types of angular pregnancy have variable pregnancy outcomes and risks requiring clinical management to be individualized. Fine ultrasonic diagnosis is both crucial and feasible.

Keywords

Angular pregnancy; Placental accrete; Uterine rupture; Ultrasonic diagnosis; Clinical management

1. Introduction

There exists wide-ranging debate over definitions of pregnancies located at the utero-tubal junction (angular, cornual, and the eccentric pregnancy) [1–3]. We adopted Williams' most current version [4], defining a cornual pregnancy as "a conception that develops in the rudimentary horn of a uterus with a Mullerian anomaly". Combining the literature [3, 5–7] with our clinical experience, we propose the term angular pregnancy to designate implantation of the embryo just medial to the utero-tubal junction at the lateral angle of the uterine cavity and inside of the round ligament.

We further propose a fine classification of angular pregnancy, dividing it into three types according to the location of implantation of the embryo and growing direction. Type I (endogenic type) is defined as implantation of the embryo partly in the uterine angular and mostly (>50%) growing toward the midline of the uterine cavity. Type II (exogenous type) is defined as implantation of the embryo completely in the uterine angular and growing outward but inside of the round ligament. Type III (angular and interstitial type) is defined as implantation of the embryo in both the angular and the opening of the interstitial portion of the fallopian tube and growing toward the interstitial portion of the fallopian tube. The gestational sac of type III angular pregnancy spans both sides of the round ligament. The coronal view and virtual partition of a normal uterus are shown in Fig. 1A–D. Angular pregnancy should be differentiated from tubal interstitial pregnancy, which is defined as implantation of the embryo completely in the interstitial part of the fallopian tube with outward growth. The implantation sites of three types of angular pregnancy are shown in Fig. 1E and Fig. 2A–D.

Angular pregnancy constitutes a high-risk pregnancy. If angular pregnancy is not diagnosed in the first trimester, it is more likely to be missed during the second and third trimesters. Angular rupture and massive hemorrhage may occur either prior to or during delivery, endangering the lives of the pregnant patient and her fetus. If prenatal diagnosis is missed, angular placenta implantation with uterine wall penetration may occur, resulting in obstetric complications such as postpartum hemorrhage and/or infection secondary to retained placental material.

The purpose of this study is to describe the related risk factors, clinical manifestation, ultrasonic diagnostic criteria, clinical management, outcomes, and complications of different types of uterine angular pregnancy.

2. Materials and methods

2.1 Study design and patients

This study is a retrospective and descriptive analysis of the data collected in routine pregnancy care. The cases feature patients with angular pregnancy who were treated in the De-

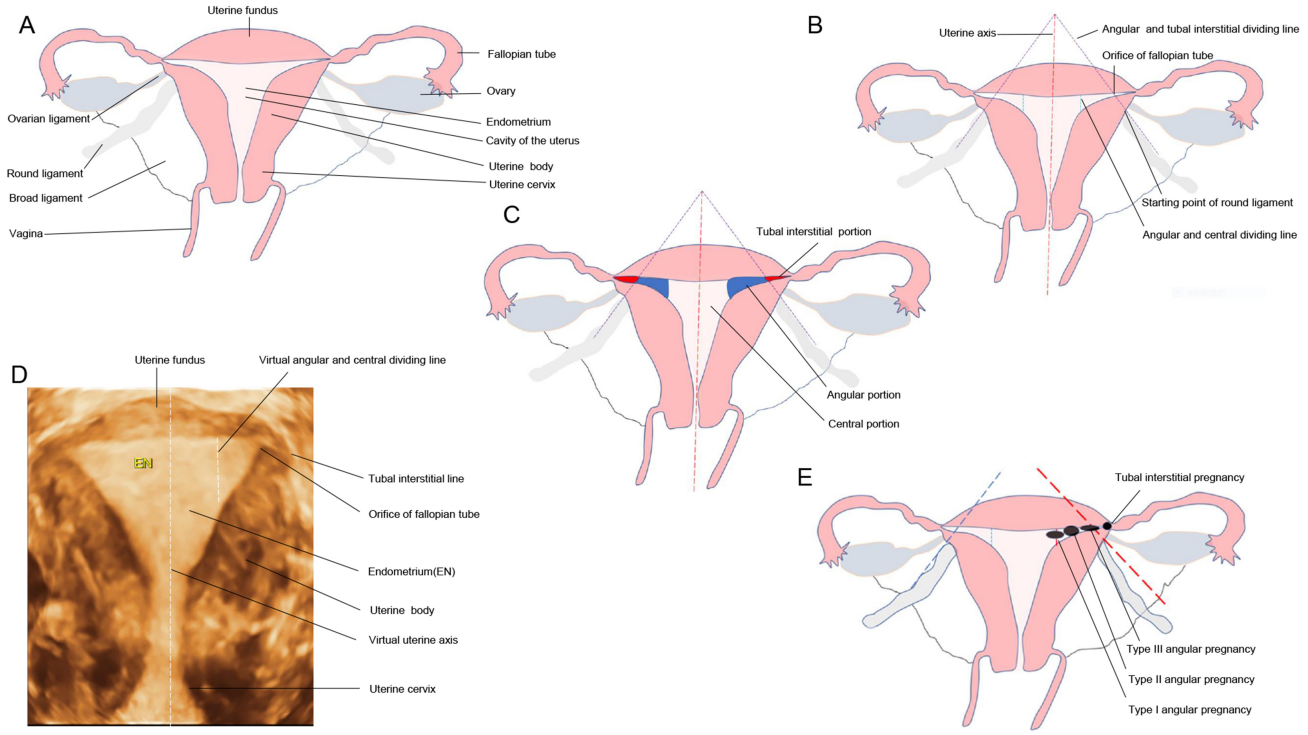

Fig. 1. The schematic diagrams and ultrasound image of virtual dividing lines and partitions of uterine cavity. Schematic diagrams: (A) Coronal view of uterus and adnexa. (B) Virtual dividing lines of uterine cavity. (C) Virtual partitions of uterine cavity. (E) The implantation sites of three types of angular pregnancy and tubal interstitial pregnancy (The black oval is the pregnancy sac.). Ultrasound render image: (D) Uterine coronal three-dimensional ultrasound render image, virtual dividing lines, and virtual partitions of uterine cavity.

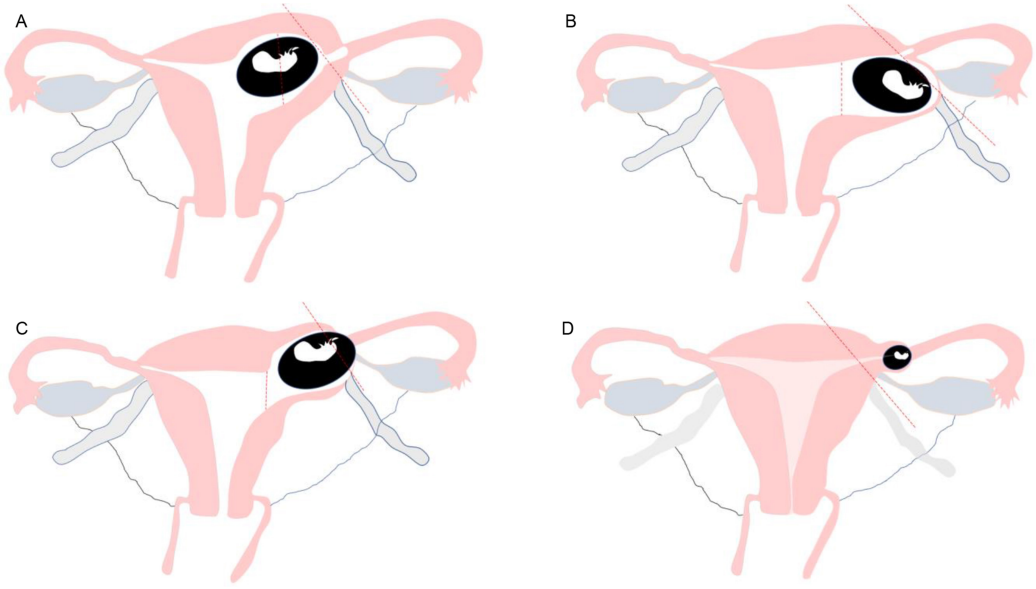

Fig. 2. Schematic diagrams of definitions and gold standard diagnostic criteria of three different types of angular pregnancy and tubal interstitial pregnancy (The short-dotted line is angle and central dividing line; the long-dotted line is angle and tubal interstitial dividing line; the black oval is the pregnancy sac.). Schematic diagrams: (A) Type I angular pregnancy (endogenic type). (B) Type II angular pregnancy (exogenous type). (C) Type III angular pregnancy (angular and interstitial type). (D) Tubal interstitial pregnancy.

partment of Obstetrics and Gynecology of Tongji Hospital affiliated with Tongji Medical College of Huazhong University of Science and Technology from January 2016 to July 2020. Inclusion criteria entailed the diagnosis of an angular pregnancy in early pregnancy by ultrasonic examination; the diagnosis of a normal early intrauterine pregnancy followed by the ultrasonic diagnosis of an angular pregnancy or angular placenta accrete during the middle or late pregnancy; or di-

agnosis postpartum by ultrasound of an angular pregnancy with portions of a retained placenta. Exclusion criteria entailed cases of angular pregnancy diagnosed by ultrasound in our institution but not observed, treated, or delivered in our hospital; malformed uterine anatomy; multiple gestation; or cases with absence of complete ultrasonic data.

2.2 Gold standard diagnosis

Diagnosis made by direct vision, postoperative pathological examination or clinical comprehensive diagnosis is regarded as the gold standard of our research and the gold standard of our center's clinical practice. Angular pregnancy has been divided into three types (See Fig. 2A–C): type I (endogenic type) with no or mild angular protrusion, wherein the majority (>50%) of the pregnancy sac is located in the uterine cavity; type II (exogenous type) with angular protrusion, wherein the majority (>50%) of the pregnancy sac (GS) is inside the round ligament and there is no fallopian tube abnormality; and type III (angular and interstitial type) with the protruding mass of pregnancy spanning both sides of the round ligament, wherein the outer boundary of the protruding mass extends to the opening of interstitial portion of fallopian tube with villi seen in angular, and the opening of interstitial portion of fallopian tube is evident via direct vision or pathological examination.

Type I angular pregnancy should be differentiated from normal intrauterine pregnancy, in which the GS is found in the endometrium of the body or fundus of the uterus without the diagnostic criteria of angular pregnancy at the time of surgery, delivery, or pathological examination of angular pregnancy. Type II and III angular pregnancy should be differentiated from tubal interstitial pregnancy. In tubal interstitial pregnancy, the pregnancy mass is found in the interstitial part of the fallopian tube outside of the round ligament, with villi seen in the interstitial portion of fallopian tube by direct vision of surgery and postoperative pathological examination (Fig. 2D). The surgical site of type II angular pregnancy is the angular, the surgical site of type III angular pregnancy is the angular and fallopian tube, and the surgical site of tubal interstitial pregnancy is the fallopian tube.

2.3 Ultrasonic diagnosis

During the ultrasound examination in the first trimester, our system recorded ultrasound images and reports that included the location of the gestational sac, whether the gestational sac is continuous with the endometrium at the fundus of the uterus, the presence of decidual wrapping sign, the presence of interstitial line sign [8, 9], the minimum thickness of the muscle wall from the outermost edge of the gestational sac to the serous layer at the angular, degree of protrusion of the gestational sac, boundary between the villi and the muscle wall of the gestational sac, and blood flow at the implantation site. If the gestational sac were located near the utero-tubal junction, a diagnosis of angular pregnancy was made and divided into three types according to the ultrasonic characteristics.

The ultrasonic diagnostic criteria of type I angular pregnancy include: (1) the vast majority (>50%) of the GS is located in the uterine cavity with a small part located in the angular; (2) O-shaped decidua wrapping sign can be seen around the GS; (3) without interstitial line sign; (4) without obvious protuberance of the angular, or with slight protuberance of the angular; (5) thickness of angular muscle wall is >3.5 mm. The ultrasonic diagnostic criteria of type II angular pregnancy include: (1) the vast majority (>50%) of the GS is located in the uterine angular with a small part located in the uterine cavity; (2) O-shaped decidua wrapping sign can be seen around the GS; (3) without interstitial line sign; (4) with obvious protuberance of the angular; (5) thickness of angular muscle wall is ≤3.5 mm. The ultrasonic diagnostic criteria of type III angular pregnancy include: (1) the GS is located in the uterine angular completely, extending to the interstitial part of the fallopian tube; (2) C-shaped decidua wrapping sign can be seen around the GS while the inner side of the GS is continuous with the decidua; (3) without interstitial line sign; (4) with obvious protuberance of the angular, and the vast majority (>50%) of the GS protrudes outward; (5) thickness of angular muscle wall is ≤1 mm. The ultrasonic characteristics of the three types of angular pregnancy in the uterine coronal three-dimensional ultrasound render image are shown in Fig. 3A–C.

Type I angular pregnancy should be differentiated from normal intrauterine pregnancy in which the GS is located in the endometrium of the body or fundus of the uterus without the ultrasonic diagnostic criteria in the first trimester. Type II and III angular pregnancy should be differentiated from tubal interstitial pregnancy. The ultrasonic diagnostic criteria of tubal interstitial pregnancy include: (1) the GS is located in the interstitial part of the fallopian tube; (2) the inner side of the GS is not continuous with the decidua; (3) with interstitial line sign; (4) the pregnancy mass is located in the interstitial part of the fallopian tube (Fig. 3D).

During the ultrasound examination in the second and third trimesters, the thickness of angular muscle wall, degree of protrusion and presence or absence of placenta accrete were recorded in cases of type I angular pregnancy. The residual placenta or villi and implantation at the angular site were recorded after delivery. The ultrasound scan was obtained by combined use of two-dimensional ultrasound, three-dimensional ultrasound, color Doppler and spectral Doppler. Measurement of the data was carried out on the two-dimensional image, while the three-dimensional image directly pinpointed the position of the pregnancy sac.

2.4 Statistical analyses

The statistical software package SPSS26.0 (IBM SPSS Statistics for Windows, 26.0 version, Armonk, NY, USA) was used for data analysis. The data of classified variables are represented by n (%) with the results of data analysis being shown by bar chart and the data of continuous variables being represented by M (25%–75%).

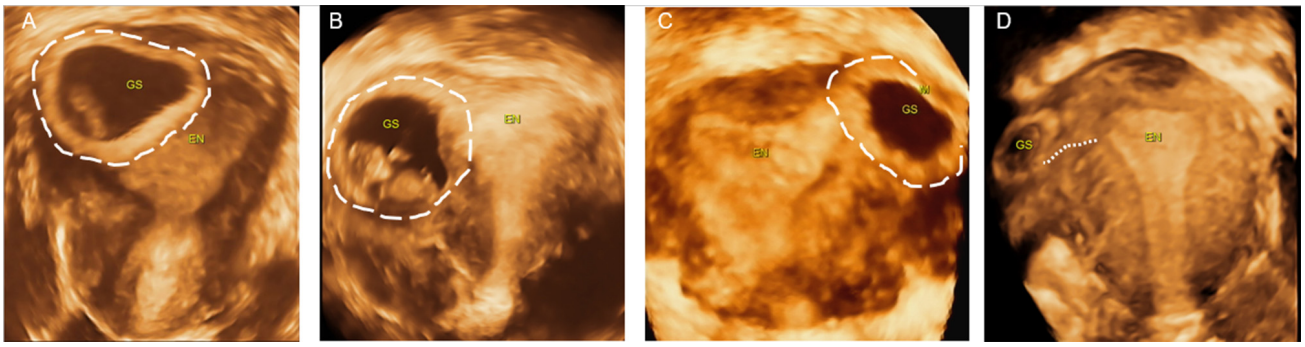

Fig. 3. The uterine coronal three-dimensional ultrasound render images of three types of angular pregnancy and tubal interstitial pregnancy (GS, gestational sac; EN, Endometrium; M, mass). (A) Type I angular pregnancy (Dotted line indicates O-shaped decidua wrapping sign). (B) Type II angular pregnancy (Dotted line indicates O-shaped decidua wrapping sign). (C) Type III angular pregnancy (Dotted line indicates C-shaped decidua wrapping sign). (D) Tubal interstitial pregnancy (Dotted line indicates interstitial line sign).

3. Results

A total of 60 cases were included in this study. The median age of pregnant women was 30 years (29–34), the median clinical gestational week was 7.5 weeks (7.0–8.9), and the median ultrasound gestational week was 6.5 weeks (5.8–7.8). The frequency and percentage of related clinical characteristics of cases are shown in Table 1. Among the 60 cases, 46 cases (76.7%) had related risk factors and 14 cases (23.3%) did not. Twenty-five cases (41.6%) had clinical symptoms of vaginal bleeding with or without lower abdominal pain and 35 cases (58.4%) had no symptoms of an abnormal pregnancy.

The ultrasonic diagnosis, clinical management, complication, pregnancy outcome and gold standard diagnosis of 60 cases are shown in Table 2. Among the 60 cases, 59 (98.3%) were diagnosed as different types of angular pregnancy according to the established gold criteria. The number of cases of type I, II and III angular pregnancies was 42 (71.2%), 13 (22.0%) and 4 (6.8%), respectively. The percentage of angular villi or placental accrete in type I, II and III angular pregnancy was 4.8% (2/42), 23.1% (3/13) and 100% (4/4). Seventy-five percent (3/4) of type III angular pregnancies had angular rupture with villi or placental tissue protruding outward.

The sensitivity of ultrasound in the first trimester diagnosis of type I, II and III angular pregnancy was 83.3% (35/42), 69.2% (9/13) and 50.0% (2/4). The sensitivity of ultrasound was calculated according to the ultrasonic diagnosis of early pregnancy in the first column vs. the gold standard diagnosis in the last column of Table 2. Because there were no true negative or false positive cases in this study, the diagnostic specificity is not calculated.

Frequencies of different clinical management without complications of various types of angular pregnancy based on the gold standard diagnosis are shown in Fig. 4. This analysis removed four cases of angular pregnancy with complications that included uterine rupture, inappropriate surgical methods or delivery mode resulting in residual villi or placental fragments in the angular region.

4. Discussion

Among the 60 cases, 59 cases (98.3%) were diagnosed as different types of angular pregnancy according to the established gold criteria. Different cases presented different risk factors, while varying types of angular pregnancy entailed different clinical manifestations, pregnancy outcomes and risks. Fine ultrasonic diagnosis proved to be feasible and of critical value. Clinical management of the different types of angular pregnancy require individualization. Correct clinical treatment and good clinical results were obtained in 55 cases. Complications occurred in 4 cases.

4.1 Risk factors

Studies have found that history of abortion, pelvic surgery, cesarean section, assisted reproductive technologies, pathological changes of the fallopian tube, endometriosis and luteal deficiency are all related risk factors [10, 11]. Our study found that previous history of delivery, manual abruption of uterine angular placenta, abortion, villi residual in angular region after surgical abortion, placenta residual in the angular region postpartum, ectopic pregnancy, salpingectomy, uterine angular operation, uterine cavity adhesion, assisted reproductive technologies, uterine leiomyoma and adenomyosis may be related risk factors for an angular pregnancy.

4.2 Clinical manifestations

The clinical manifestations of patients with an angular pregnancy include the absence of regular menstrual cycles with or without non-specific symptoms of vaginal bleeding, along with severe abdominal pain. Hemorrhagic shock may occur when the angular pregnancy ruptures, but most patients remain asymptomatic [3]. Angular pregnancy with placental accrete has no specific clinical symptoms but the placenta remains in the angular location during delivery and cannot be delivered naturally. Angular pregnancy with villi or placental implantation into the surrounding tissue has a higher risk of angular rupture.

Table 1. Clinical characteristics of cases (n = 60).

Clinical characteristics	n (%)
Gravidity and parity, pregnancy history	
Primigravid	24 (40.0%)
Vaginal delivery	15 (25.0%)
Cesarean section	8 (13.4%)
Induced labor	2 (3.3%)
Placental abruption	2 (3.3%)
Abortion	22 (36.7%)
Villus or retained placenta	2 (3.3%)
Ectopic pregnancy	4 (6.7%)
Salpingectomy	3 (5.0%)
History of uterine surgery or uterine complications	
Removal of endometrial polyps	2 (3.3%)
Lysis of uterine adhesions	2 (3.3%)
Adhesive band in the uterine cavity	4 (6.7%)
Uterine leiomyoma or adenomyoma	11 (18.3%)
Mode of conception	
Natural conception	48 (80.3%)
ART	12 (20.3%)
First pregnancy conceived naturally without any history of surgery or uterine complications	14 (23.3%)
Clinical symptoms	
Vaginal bleeding	17 (28.3%)
Lower abdominal pain	8 (13.3%)
Without any symptoms	35 (58.4%)

ART, assisted reproductive technology.

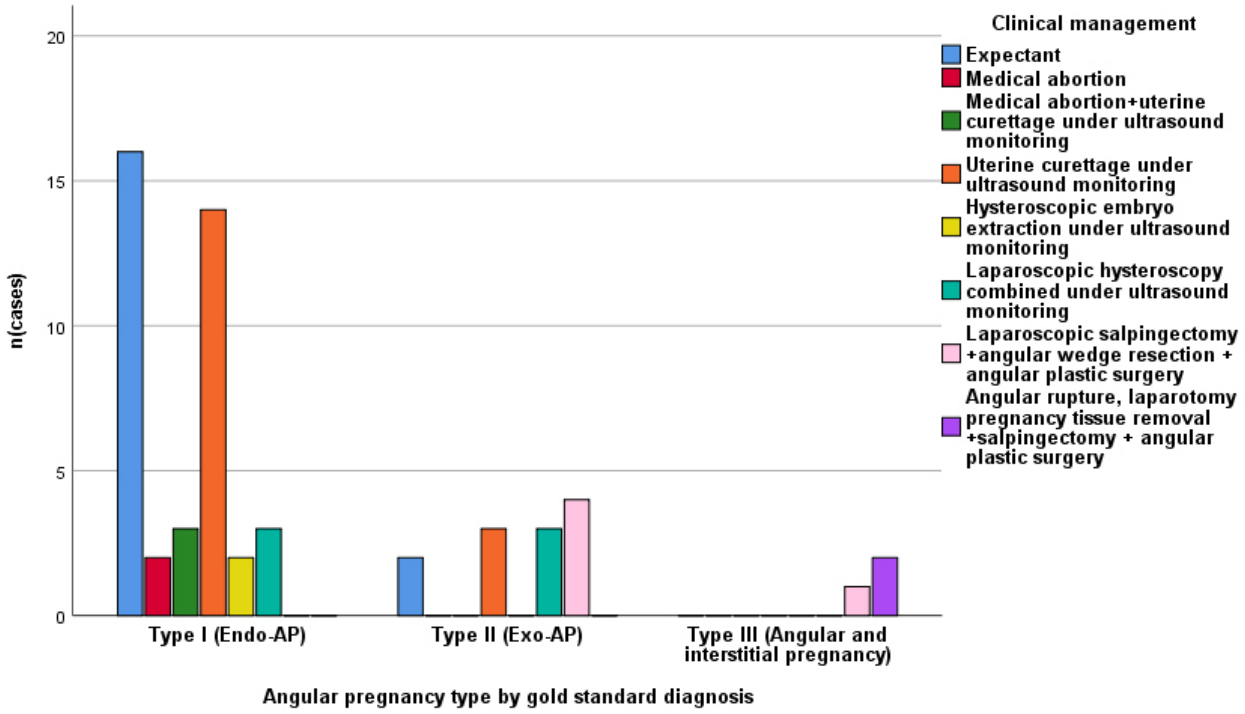

Fig. 4. Frequencies of different clinical management without complications of various types of angular pregnancy based on the gold standard diagnosis (n = 55).

Table 2. FTS diagnosis, clinical management, outcome, complication, and gold standard diagnosis (n = 60).

FTS (60, 100%)	Clinical management, outcome, and complication		GSD I (42, 71.2%) II (13, 22.0%) III (4, 6.8%) Interstitial (1, 1.7%)
normal IUP (4, 6.7%)	Expectant (2, 3.3%)	US at 32wks PP in the right angular, CD, PP in the right angular, MAP (1, 1.7%) (See Fig. 5)	I with PA (1, 1.7%)
		At 15 wks. US: right angular protruding; at 20wks, right lower abdominal pain, US: right angular and interstitial pregnancy, PA, right angular rupture. Emergency laparotomy, two lacerations and PA in the right angular, angular incision to take the dead fetus, salpingectomy + angular wedge resection + angular + angular plastic surgery (1, 1.7%) (See Fig. 6)	III with PA (1, 1.7%)
	Ask for termination (2, 3.3%)	Negative pressure uterine aspiration under UC, tissue residue in the angular, hysteroscopic tissue removal under UC (2, 3.3%)	I (2, 3.3%)
Type I AP (35, 58.3%)	Expectant (14, 23.3%)	Live birth without complications (8, 13.3%)	I (8, 13.3%)
		Live birth, angular placenta residue and PA (1, 1.7%)	I with PA (1, 1.7%)
		MA + UC (5, 8.3%)	I (5, 8.3%)
	Ask for termination (21, 35.0%)	Negative pressure uterine aspiration under UM (14, 23.3%)	I (14, 23.3%)
		MA + UC (3, 5.0%)	I (3, 5.0%)
		MA (2, 3.3%)	I (2, 3.3%)
		Hysteroscopic embryo extraction under UM (2, 3.3%)	I (2, 3.3%)
Type II AP (13, 21.7%)	Expectant (3, 5.0%)	The embryo stops developing or spontaneous abortion, hysteroscopic embryo extraction under UM (3, 5.0%)	I (1, 1.7%) II (2, 3.3%)
	Elective surgery (10, 16.7%)	Negative pressure uterine aspiration under UM (4, 6.7%)	II (4, 6.7%)
		Removal of pregnant tissue by laparoscopic hysteroscopy combined under UM (6, 10%)	I (3, 5.0%) II (3, 5.0%)
Type III AP (3, 5.0%)	Elective surgery (2, 3.3%)	Lower abdominal pain, laparoscopic salpingectomy + angular wedge resection + angular plastic surgery (1, 1.7%)	III with VA (1, 1.7%)
		Laparoscopic resection of tubal interstitial pregnancy (1, 1.7%)	Interstitial (1, 1.7%)
with VA (1, 1.7%)	Emergency operation (1, 1.7%)	Lower abdominal pain, angular rupture, pregnancy tissue removal + salpingectomy + angular plastic surgery (1, 1.7%)	III with VA (1, 1.7%)
Interstitial pregnancy (5, 8.3%) with VA (4, 6.7%)	Emergency operation (5, 8.3%)	Lower abdominal pain, laparoscopic resection of angular pregnancy (4, 6.7%)	II (4, 6.7%) with VA (3, 5.0%)
		Lower abdominal pain, angular rupture, laparotomy pregnancy tissue removal + salpingectomy + angular plastic surgery (1, 1.7%)	III with VA (1, 1.7%)

FTS, first-trimester screen; GSD, Gold standard diagnosis; US, Ultrasound; UM, ultrasound monitoring; PA, placenta accrete; PP, placental penetration; VA, villi accrete; IUP, intrauterine pregnancy; CD, cesarean delivery; MAP, manual abruption of placenta; AP, angular pregnancy; MA, medical abortion; UC, uterine curettage; wks, Weeks.

4.3 Diagnosis

In 1981, Janson *et al.* [7] put forward the following clinical diagnostic criteria for an angular pregnancy: (1) clinical presentation with painful asymmetric enlargement of the uterus; (2) directly observed (i.e., surgical) lateral distension of the uterus with displacement of the round ligament laterally; (3) retention of the placenta in the uterine angle. Angular pregnancy can be diagnosed in accordance with any of these criteria. The standard is clinical diagnosis based on gynecological examination and operative findings.

With the development of advanced medical imaging technology, current diagnostic methods for angular pregnancy include imaging diagnosis, laparoscopy, hysteroscopy, and laparotomy. Three-dimensional ultrasound is the first choice for the diagnosis of angular pregnancy with magnetic resonance imaging (MRI), feasible if necessary [12]. During the ultrasound examination in the first trimester, it is crucial to describe with accuracy the location of the gestational sac in the utero-tubal junction and to identify the fine type of an angular pregnancy in order to evaluate the risk of pregnancy and to formulate a customized management plan.

4.4 Clinical managements and outcomes

As the different types of angular pregnancy have varied pregnancy outcomes, clinicians should make personalized management plans accordingly.

Type I angular pregnancy grows toward the center of the uterine cavity, as reported by Fernandez *et al.* [13]. With a continuing pregnancy, some will result in a live birth, but there exists a high risk of miscarriage, rupture of uterine angular, placenta accrete and retained placenta. As the pregnancy progresses, the clinician should regularly monitor the degree of angular protrusion, the thickness of the muscle wall, the location of the placental attachment and whether there is evidence of placenta accrete. When a pregnant woman with type I angular pregnancy requests termination of pregnancy, negative pressure suction or drug abortion can be utilized as the majority of the gestational sac is in the uterine cavity. It is recommended to perform fixed-point clearance negative pressure aspiration under ultrasound or intrauterine visual system guidance and, if necessary, to visualize the uterus by laparoscopy (Fig. 4). Mollo *et al.* [14, 15] put forward that hysteroscopic intact removal of angular pregnancies may be used as a diagnostic and therapeutic tool for angular pregnancy, providing a unique image of the intact removal of the gestational sac and allowing a markedly less invasive approach. Our clinical practice supports this view.

In our study, a 31-year-old pregnant woman with a type I angular pregnancy had a previous live vaginal delivery 10 years prior without complications or other surgical history. She was diagnosed as having a normal intrauterine pregnancy during the first and second trimesters at another hospital. At 32 weeks gestation, ultrasound determined that the fetus had hydrocephalus and placental penetration at the uterine angular (Fig. 5A,B). The MRI scan confirmed the ultrasound examination findings. After informing the patient of these findings, she requested an induction of labor. Potassium chloride was injected into the fetal heart under ultrasound guidance and a stillborn fetus was delivered by a low transverse cesarean section. The placenta and fetal membranes were manually removed. The right uterine angular demonstrated a 50.0 mm × 40.0 mm × 25.0 mm sized blood sinus protruding from the uterine serosa and thick blood vessels on the uterine surface (Fig. 5C). Adnexa demonstrated no abnormalities. On the second postoperative day, her hemoglobin was 68.0 g/L and she was transfused 3.5 units. On the 6th postoperative day, color Doppler ultrasonography showed residual 14.0 mm × 14.0 mm placental tissue implantation in the right uterine angular (Fig. 5D). Menstruation returned to normal after conservative treatment.

With type II angular pregnancy, which is often associated with angular villi or placental accrete, the risk of angular rupture is very high. It is recommended to terminate the pregnancy after confirming the diagnosis, as shown in Fig. 4. The pregnancy sac of a type III angular pregnancy (uterine horn and interstitial type) is located at the beginning of the uterine horn adjacent to the interstitial portion of the fallopian tube. Following the diagnosis of a type III angular pregnancy, surgery should be performed as soon as possible. The mode of operation can be seen in Fig. 4. If the gestational age of the type II or III angular pregnancy is greater than 12 weeks at the time of diagnosis, the risk of uterine horn rupture and massive bleeding is high. If the uterine horn has ruptured at the time of diagnosis, emergency laparotomy is recommended. If the patient with the type II or III angular pregnancy is clinically stable and there is no embryo in the gestational sac, with the informed consent of the patient, the direct hysteroscopic ultrasound-guided injection of methotrexate around the gestational sac can be carried out to reduce surgical injury and increase the probability of preserving fertility, referencing the minimally invasive conservative treatment of tubal interstitial pregnancy reported by Leggieri *et al.* [16].

4.5 Complications

Complications of angular pregnancy include uterine rupture, residual villi, or placental fragments in the angular region. In our study, four cases involved complications caused by incorrect clinical management due to ultrasound misdiagnosis in early pregnancy.

One case was previously diagnosed as normal early intrauterine pregnancy in a separate hospital; however, the right angular protuberance without an angular pregnancy was identified during an ultrasound examination at the 15th week of pregnancy by a junior doctor at our institution. Five weeks later, the patient experienced pain in the right lower abdomen, with a clear ultrasound diagnosis by a senior doctor of a right angular and interstitial pregnancy with rupture of angular location. The patient developed hemorrhagic shock and accepted emergency laparotomy, with two lacerations (50.0 mm × 40.0 mm and 20.0 mm × 20.0 mm) and placenta accrete in the right angular, and numerous blood clots in the pelvis and abdominal cavity. Angular incision removed the

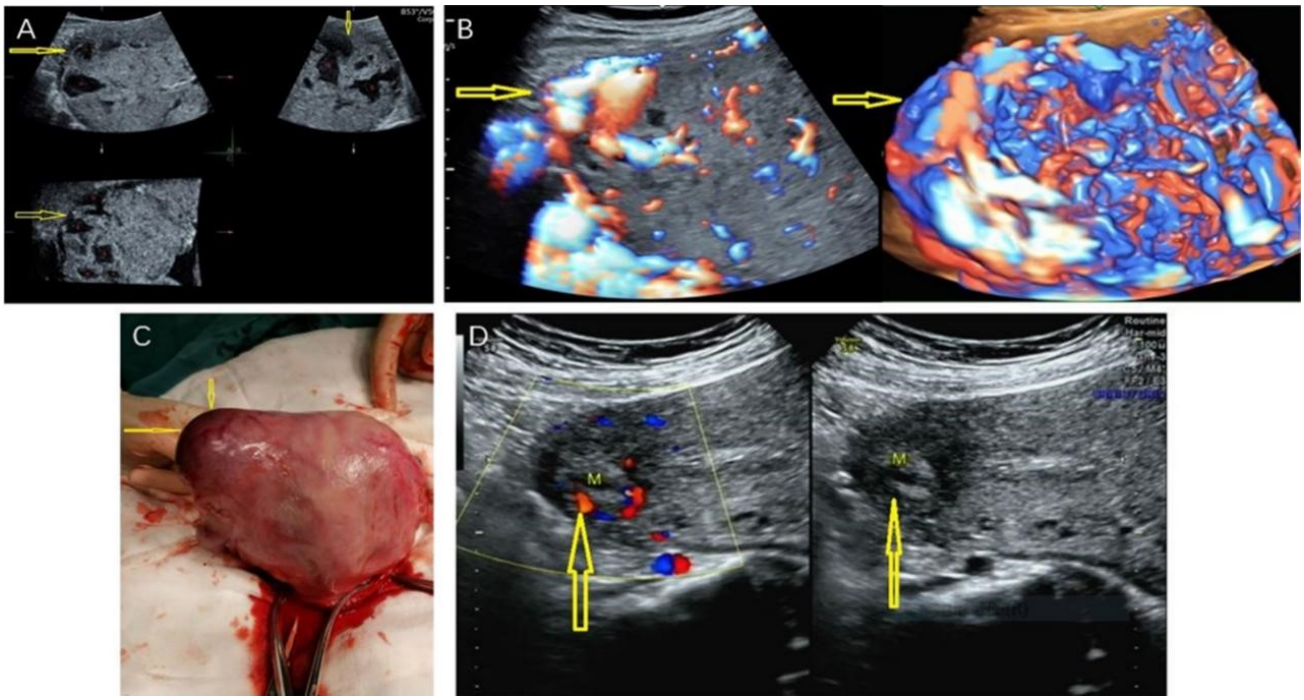

Fig. 5. The ultrasound images and intraoperative photos of a case of type I angular pregnancy with placental penetration in the right angular. (A) Multiplanar display of three-dimensional ultrasound at 32 weeks gestation. The right uterine angular was slightly protruding; the thickness of the uterine angular myometrium is 0 mm; the placenta is attached to the right angular, the right wall and the upper right posterior wall; and there was no boundary between the placenta and the muscle wall. Placental thickening with multiple lacunae and eddy current is noted (The yellow arrow indicates the disappearance of the normal muscle wall in the right angular with the placental tissue reaching the serosal layer and the red pentagram indicates multiple lacunae in the placenta). (B) Three-dimensional HD-Flow at 32 weeks of gestation (There are abundant and messy blood flow signals in the placenta and under the serosa of the placenta with the yellow arrow indicating that the type of blood flow in the right angular overflows the serosa). (C) A photo of uterus during low transverse uterine segment cesarean section (The yellow arrow indicates a 50.0 mm × 40.0 mm × 25.0 mm-sized blood sinus protruding from the uterine serosa; thick surface blood vessels can be seen in the right angular location). (D) Transverse section of the bottom of uterus by two-dimensional ultrasound on postoperative day 6 after cesarean section (The yellow arrow indicates that the retention of 14.0 mm × 14.0 mm placental tissue implantation seen in the right angular location).

dead fetus, accompanied by salpingectomy, angular wedge resection, and angular plastic surgery. She accepted infusion 1500 mL, concentrated red 11.5 units, plasma 400 mL and plasma substitute 2500 mL intraoperative and postoperative. Postoperative intestinal obstruction occurred, and following multidisciplinary consultation and active treatment, the patient was hospitalized for eight days and discharged. Postoperative pathological examination showed that angular placenta implantation and villi could be seen in fallopian tubes, consistent with type III angular pregnancy. Ultrasound and intraoperative images of this patient are shown in Fig. 6.

For two cases that were diagnosed as normal intrauterine pregnancy, the patients requested termination. They underwent negative pressure uterine aspiration under ultrasound monitoring. Following the operation, residual pregnancy tissue was noted in the angular region and hysteroscopic tissue removal was performed under ultrasound guidance. Type I angular pregnancy was postoperatively diagnosed.

The fourth case was diagnosed as an endogenic angular pregnancy by ultrasound during the first trimester, without diagnosis of angular placenta accrete during the second and third trimester; the patient carried the pregnancy to a term vaginal delivery. The placenta did not deliver spontaneously and residual placental tissue and accrete in angular location was diagnosed by ultrasound.

4.6 Strength and limitations

The main strength of our study is that the sample size is large, providing a sufficient number of cases of angular pregnancy with different high-risk factors, pregnancy outcomes and customized management. As a result, we were able to ascertain the high-risk factors of angular pregnancy, the value of ultrasound diagnosis and individualized management scheme.

This study has two limitations. One limitation is that ultrasound doctors have different levels of experience, accounting for potentially wide range of diagnosis and management. The other limitation concerns the pathological diagnosis of placenta accrete. Manual abruption of placenta or retained placenta *in situ* limits the accuracy of microscopic diagnosis or the lack of micropathological diagnosis during pathological examination.

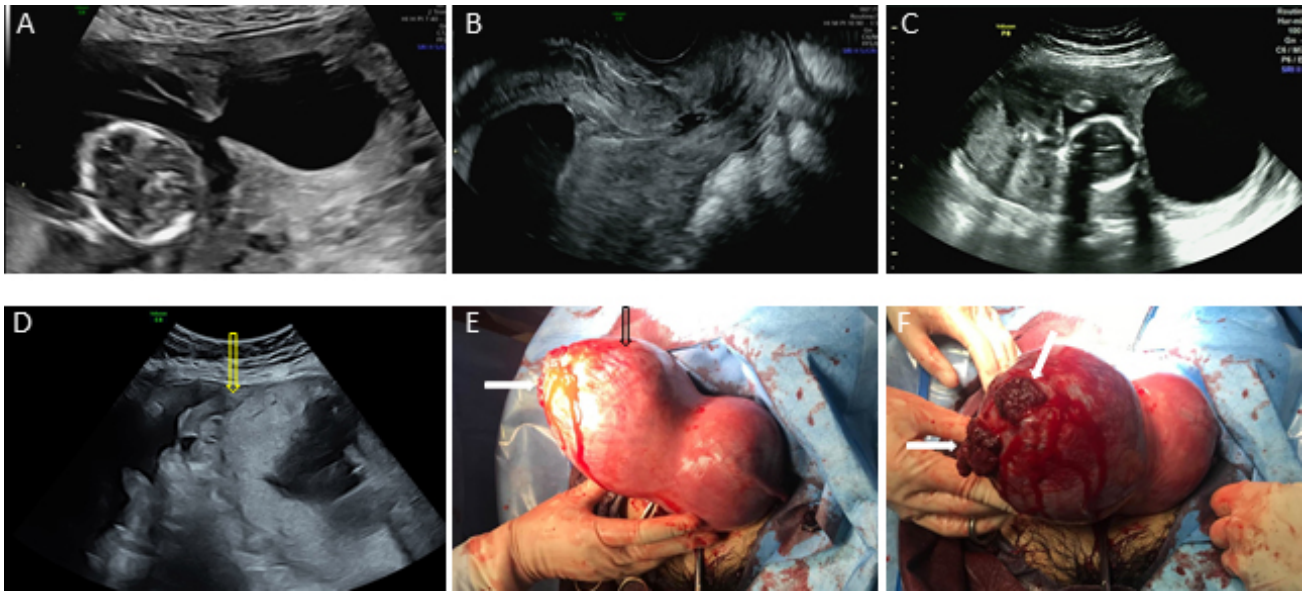

Fig. 6. The two-dimensional gray-scale ultrasound images and intraoperative photos of a case of type III angular pregnancy. Ultrasound examination at 15 weeks of gestation: (A) The fetus was found in the right angular, and there was an adhesive band between the right angular and the uterine cavity. (B) Only amniotic fluid without fetal structure was found in the middle and lower part of the uterine cavity. Ultrasound examination at 20 weeks of gestation: (C) The fetus was curled up in the right angular, and there was an adhesive band between the right angular and the uterine cavity. (D) Rupture of the right angular and hemoperitoneum (yellow arrow indicates the rupture of right angular). Intraoperative photos: (E) The black arrow indicates that the right angular is obviously protruding, and the white arrow indicates the rupture of the right angular. (F) The white arrow indicates that two lacerations of 50.0 mm × 40.0 mm-sized and 20.0 mm × 20.0 mm-sized can be seen in the right angular, the placental tissue protrudes outward, and the muscular tissue between the two ruptures is thin.

5. Conclusions

All providers caring for pregnant women should fully understand the ultrasonic diagnosis and classification criteria, clinical diagnosis criteria and pathologic diagnosis criteria for angular pregnancy. Different types of angular pregnancy entail different pregnancy outcomes and risks that require clinical management to be individualized for each patient. For the early identification of an angular pregnancy, ultrasound should be utilized to determine the type of angular pregnancy and to judge whether there is evidence for placenta accrete in the first trimester. Fine ultrasonic diagnosis is both feasible and crucial. This will allow the clinician to correctly determine risks to the pregnancy and to formulate a tailored management plan for the patient. This has the potential to reduce unnecessary medical termination of pregnancy and avoid adverse outcomes such as uterine rupture and massive hemorrhage. At the same time, ultrasonic diagnosis, and classification of placenta accrete during the second and third trimester in type I angular pregnancy is critical to the proper choice for time and mode of delivery.

Author contributions

LZ designed the research study. SHC gave guidance for the revision of the paper. LZ was responsible for ultrasonic diagnosis, collection, analysis of case data, writing and revision of the manuscript. SPD and YHY assisted in collecting ultrasound data. SHC and WL were responsible for obstetric patient management, operation, and data recording. RL, SMY and MFW were responsible for gynecologic patient management, operation, and data recording. Allauthors read and approved the final manuscript.

Ethics approval and consent to participate

All patients involved in this study signed an informed consent and power of attorney before accepting an operation. This study is a retrospective and descriptive analysis of the data collected prospectively for routine clinical services. The study protocol was approved by the ethics committee of our institution (approval number: TJ-C20210403).

Acknowledgment

We would like to express our gratitude to all those who helped during the writing of this manuscript, to the peer reviewers for their opinions and suggestions, the editors for their comments and guidance, and to Ms. Wang Xinmiao for her advice in creating the schematic diagram.

Funding

This research received no external funding.

Conflict of interest

The authors declare no conflict of interest.

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