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Uterine torsion and intrauterine growth restriction: Case report and systematic literature review

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Abstract

Purpose: Uterine torsion (UT) in pregnancy is a rare condition in obstetric practice. It is defined as a rotation of the uterus of more than 45° around its long axis. Presentations are varied and, most of the time, this condition is recognized at laparotomy or cesarean section (CS). The aim of this study is to summarize the latest evidence about UT in pregnancy.

Methods: A systematic research of the literature was conducted fetching all papers published from March 2006 to June 2020. We collected data regarding clinical features, treatment, and feto-maternal outcomes. Finally, we reported data of a case of UT associated with intrauterine growth restriction (IUGR) diagnosed and treated at our institution.

Results: According to our search strategy, 38 articles were included. In 66% of the cases, acute symptomatology was present at the onset, most frequently abdominal pain was reported. In one-third of the cases, UT was diagnosed during CS without clinical suspicion. Only in two cases, including our case, IUGR was reported. Most (66%) of the cases presented a 180° torsion. In the majority of the cases, a CS was performed also with a deliberate or accidental posterior hysterotomy. One and six cases of maternal and fetal death were, respectively, reported.

Conclusion: UT is an infrequent obstetric condition but should be considered in case of abdominal pain, vomiting, or shock presentation during pregnancy. It could lead to a reduction in uterine blood flow contributing to poor placental perfusion, even though more evidence is needed to clarify this link.

Key words: cesarean section, growth restriction, pregnant uterus, preterm birth, uterine torsion.

Introduction

Uterine torsion (UT) in pregnancy is a rare condition in obstetric practice and is defined as a rotation of the uterus of more than 45° around its long axis. UT can be a life-threatening disease and is observed in all age-groups of the reproductive period, in all parity groups and during all stages of pregnancy. The etiology is not clear but several risk factors have been identified including morphologic anomalies, myoma uteri, pelvic adhesion, previous pelvic surgery, pelvic masses, attempt of external cephalic versions, and abnormal fetal presentation.¹ The most common symptoms include birth obstruction, abdominal pain, vaginal bleeding, shock condition, urinary and intestinal symptoms, while less frequently there is an asymptomatic clinical onset. In the early stages of pregnancy, if UT is diagnosed the treatment of choice

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is immediate laparotomy, manual detorsion of the uterus, and, if possible, surgical correction of the associated anomalies. Cesarean section (CS) is necessary to treat UT in the final stages of pregnancy or during labor and most of the times is an unexpected intraoperative diagnosis. The aim of this study is to summarize the latest evidence regarding diagnosis and management of UT in pregnancy. Furthermore, we reported a case of a 39-year-old woman with singleton pregnancy complicated by preeclampsia and placenta previa who underwent a CS with an incidental intraoperative diagnosis of UT.

Material and Methods

Study design

This is a systematic review of the literature on the UT in pregnancy with a case report presentation.

Search strategy

For the purpose of this study, a literature search was performed from January 2006 to May 2020 in the electronic database Scopus, PubMed/MEDLINE, ScienceDirect, and the Cochrane Library. The search strategy included the combinations of the Medical terms "Uterine torsion" AND "pregnant uterus." The literature search aimed to identify all the papers published in English language reporting cases of UT during pregnancy. Given the rarity of the topic, we included case reports, case series, and literature reviews.

Study selection and data extraction

Titles and/or abstracts of identified studies were screened independently by three authors (FFA, BN, and SF). The full text of the potentially eligible studies was retrieved and independently assessed for eligibility by other review team members (LF, EN, and FO). Any disagreement over the eligibility of particular studies was resolved through discussion with author (FF). The reference lists of all identified studies were systematically revised to identify other eligible publications. The review was written following the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Figure 1).

Inclusion and exclusion criteria

The study aimed to ask the following PICOS questions:

• Population: patients with a diagnosis UT during pregnancy.

- Comparison: no comparisons are expected.
- Outcomes: entity of UT and feto-maternal outcomes.
- Study design: case reports, case series, and systematic reviews.

Objective of the systematic review

The aim of the present study was to summarize the most recent evidence in literature concerning the diagnosis and management of UT in pregnancy.

Results

Literature review

Four major literature reviews regarding UT during pregnancy have been published: Robinson and Duval in 1931,² Nesbitt and Corner in 1956,³ Jensen in 1992,¹ and the last by Wilson in 2006.⁴ From 1992, a total of 212 cases were reported and 38 cases were added by Wilson's review in 2006. The search strategy provided a total of 523 articles after removing duplicates. Thus, 436 abstracts were screened and 384 were then excluded. A total of 52 full-texts were initially eligible. Of these, 13 studies were subsequently excluded according to inclusion and exclusion criteria. Finally, 385-42 articles were included in the analysis. In our literature review, none UTs were reported in the first trimester of pregnancy, five (13%) cases³⁸⁻⁴² in the second trimester, and 33 (87%) cases^{10–42} occurred in the third trimester. Most of the cases concerned a singleton pregnancies and only four cases 6,9,30,37 (10.5%) a twin pregnancy. Clinical manifestation was acute in 25 (66%) cases, asymptomatic with intraoperative findings in 12 (31.5%) cases, and in one case the onset was unspecified. Abdominal pain was the main symptom in 17 (68%) cases of acute onset, vomiting was reported in five (20% of acute symptomatic torsion) cases, vaginal bleeding in one (4%) case, one (4%) case of urinary retention,³⁸ and a shock is reported in two (8% of acute symptomatic torsion) cases. In two cases, UT followed an attempt of external cephalic version, in four (10.5%) cases were reported uterine anatomical anomalies, in one case a patient previously underwent sacro-hysteropexy, and in one case congenital connective tissue disorder¹⁷ was hypothesized to be implicated in the pathogenesis. In 10 cases, fetuses were in cephalic presentation (26%), breech presentation was reported in 13 (34%) cases, four (10.5%) cases with transverse presentation, and 11 (29.5%) cases did not provide the fetal

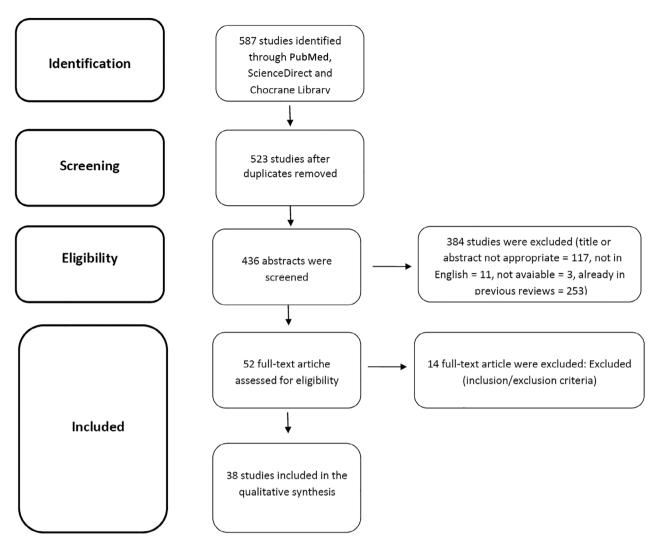


FIGURE 1 Systematic review flowchart following the PRISMA guideline

presentation. Different degrees of UT were reported: in 25 (66%) cases there was a 180° torsion, in five (13%) cases a torsion greater than 180°, and in eight (21%) cases the torsion was less than 18°. CS was performed in 35 (89.5%) cases, while vaginal birth at term was successful in two cases after laparotomic correction of UT in the second trimester. In one case, a dead fetus was left in utero and subsequent hysterectomy was accomplished (Table 2). Fetal death was reported in six (16%) cases and in one case a hysterectomy with dead fetus in situ was performed. Maternal death was reported in one (2.5%) case. Hysterectomy, B-Lynch ligation and hypogastric arteries ligation, uterine arteries ligation, and salpingo-oophorectomy were needed as additional surgery procedures, each in one case. In two cases, amputation of the twisted uterine horn in bicornuate uterus was necessary.

Case report

The patient was a 39-year-old gravida at 32 + 6 weeks of gestation age (GA) with a singleton pregnancy complicated by preeclampsia and placenta previa major. A retroverted and latero-deviated uterus was described since the fourteenth week of GA. Increased uterine arteries resistance was found during second trimester ultrasound screening; cervix was difficultly evaluable because of its position under the pubic symphysis. The patient was subsequently followed with ultrasound every month. An early intrauterine growth

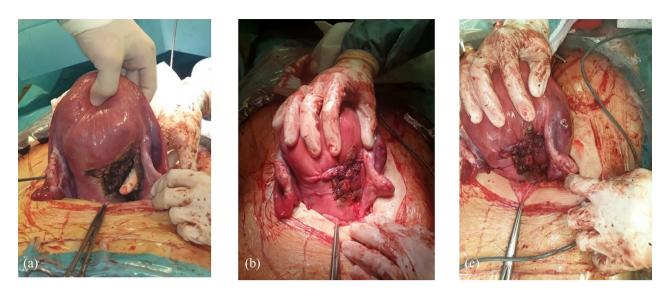


FIGURE 2 (a) Hysterotomy performed in a right lateral projection and large ligament lesion; (b) posterior view after uterine and large ligament suture; (c) right view after uterine and large ligament suture

restriction (IUGR) with impaired maternal and fetal Doppler velocimetry was found ultrasound evaluation at 25 weeks of GA. Placenta previa major was reported at 30 weeks of GA and 6 days later the patient was admitted to our Obstetric Unit for a worsening preeclampsia. During hospital stay periodic ultrasound Doppler velocimetry, cardiotocography, and biochemical preeclampsia screening tests were performed. At 32 + 6 weeks, the patient was urgently transferred to delivery room after the appearance of low variation and frequent decelerations of the fetal heart rate during cardiotocography monitoring. We decided to carry out an emergency CS and after abdominal wall opening a vascular plexus with approximately vessels of 15 mm of diameter was noted in the exposed part of the uterus. A transverse incision was performed where the cephalic portion of the fetus was evident. The fetus extraction was carried out as well as the afterbirth, and a 1202 g newborn was delivered. After uterus evacuation, a 90° –180° left rotation on the longitudinal axis was found. Because of uterus rotation, the incision was performed in the right and posterior part of the uterus causing a homolateral large ligament lesion (Figure 2a). After detorsion and exteriorization of the uterus, a doublelayer suture was performed to close the viscera (Figure 2b,c). The large ligament tear was closed and a pelvic drain was left in place. Estimated blood loss during surgery was 1000 mL and two blood cell units were administered. The drain collected 530 mL of hematic fluid in few hours after the surgery procedure and the blood tests showed values consistent with a condition of disseminated intravascular coagulation (hemoglobin: 10.4 vs. 12.2 g/dL; platelets 120 vs. 200×10^9 /L; PT 52% vs. 138%, INR 1.5 vs. 0.8, aPTT ratio 1.43 vs. 0.95, fibrinogen 51 vs. 439 mg/ dL). An abdominal CT scan reported a large parietal hematoma with a contrast die blush in the arterial phase suggesting an active bilateral bleeding from epigastric arteries. Endovascular approach was chosen to control the bleeding performing embolization of the epigastric arteries. The patient was then admitted to intensive care unit (ICU) for the following 2 days. On first postoperative day, anemia got worse despite blood cell transfusion and a new abdominal CT was carried out. A persistent bleeding from inferior epigastric artery was pointed out and a second endovascular procedure was decided. After 2 days, the patient was transferred to the Obstetric Unit with a progressive improvement of clinical condition and blood tests (hemoglobin 10.7 g/dL, platelets 105×10^9 / L, PT 126%, INR 0.8, aPTT ratio 0.97, fibrinogen 430 mg/dL). During hospital stay, hypertension was treated with nifedipine 30 mg daily until 9 days after surgery. At the time of discharge, all the drugs were interrupted. The baby was admitted to the intensive neonatal care unit for 13 days and then transferred to the Pediatric Unit until 36 days after birth.

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2006 ($N = 38$ cases)		
First trimester	0/38	0%
Second trimester	5/38	13%
Third trimester	33/38	87%
Singleton pregnancy	34/38	89.5%
Twin pregnancy	4/38	10.5%
Fetal presentation	10 (00	2.40/
Breech	13/38	34%
Cephalic	10/38	26%
Transverse	4/38	10.5%
Not reported	11/38	29.5%
Acute symptomatology	25/38	66%
Abdominal pain	$\frac{25}{50}$	68%
Vomiting	5/25	20%
Fetal distress	5/25	20%
Shock	2/25	8%
Vaginal bleeding	1/25	4%
Urinary retention	1/25 $1/25$	4%
Asymptomatic	$\frac{1}{23}$ $\frac{12}{38}$	31.5%
Clinical symptoms not	1/38	2.5%
mentioned	1750	2.370
incitabilea		
Anatomical abnormalities	4/38	10.5%
External cephalic version	2/38	5%
-		
Degrees of torsion		
180°	25/38	66%
$<\!180^{\circ}$	8/38	21%
$>180^{\circ}$	5/38	13%
	1 /00	0 50/
Maternal death	1/38	2.5%
Neonatal death	6/38	16%

TABLE 1 Characteristics of cases in the literature since 2006 (N = 38 cases)

Discussion

UT is defined as the torsion of the uterus around its longitudinal axis more than 45°1 and most torsions are approximately 180°, consistently with our review findings. Nonetheless, UTs from $60^{\circ 41}$ up to $720^{\circ 14}$ are reported. It is a rare obstetric complication, its prevalence is unknown and it occurs in women of all ages, parities, and gestations.¹ The etiology is not clear and various risk factors were reported such as fibroids, pelvic adhesions, ovarian cysts, anatomical alteration,^{1,3,4} sudden fetal or maternal movements, non-cephalic fetal presentation, external cephalic versions,^{15,36} multiple pregnancy,1 and finally smooth muscle abnormalities and ligamentous laxity may participate in abnormal uterine rotation.²⁴ In our literature review, four cases were twin pregnancies (10%) but they were not associated with worsening of fetal and maternal outcome;

, ,		
Cesarean section ^a	35	89.5%
Anterior hysterotomy	8	23%
Unintentional posterior hysterotomy	18	51%
Intentional posterior hysterotomy	8	23%
Fundal incision	1	3%
Hysterectomy with fetus in situ	1	2.5%
Vaginal birth	2	8.0%
Total	38	100%

^aAdditional surgery procedures performed as combined or single in three patients: B-lynch ligation (1), hypogastric arteries ligation (1), uterine arteries ligation (1), and salpingooophorectomy (1).

considering the incidence of multiple pregnancy and the rarity of UT relation was not clear. Four (10%) cases reported uterine congenital anatomical anomalies, 11,30,38,39 in one case the UT was in a patient with previous sacrohysteropexy⁸ and, in one case, congenital connective tissue disorder could be implicated in the pathogenesis.¹⁷ Despite the association of common pelvic pathology and uterine anomalies with UT, many case reports are available in literature without clear connection with identifiable pathology,⁴ suggesting that additional influences must be present. More frequently, UTs have an acute onset with a wide spectrum of symptoms such as abdominal pain, nausea and vomiting, birth obstruction, fetal distress up to an emergency situation of maternal shock (Table 1). Asymptomatic onset and accidentally diagnosis during laparotomy are less common but possible and it is reported in 31% of the cases. In our literature review, acute abdominal pain was reported in 68% of cases and the clinical finding imposes to always consider UT as a differential diagnosis in pregnancy. A dramatic situation of systemic shock is reported in 8% of cases since 2006 and a sudden worsening is possible requiring a prompt surgical management. Some authors reported cardiotocography abnormalities (20% of cases since 2006), probably due to the reduction of blood flow caused by the organ torsion.

UT is not an easy clinical or instrumental diagnosis. In literature, there are reported four pathognomonic clinical signs: spiral running of rectum or urethra, twisted vagina, and uterine artery pulse in anterior or posterior fornix.¹ Rarely, the diagnosis is suspected during pregnancy but an ultrasonography modification of placenta site or an abnormal course of ovarian vessel in front of the lower uterine segment could suggest the diagnosis.⁴³ Consistently with clinical findings reported by Jensen et al¹ some authors

propose the finding of X-shaped upper vagina on MRI (magnetic resonance imaging) as a sign of UT (the left lateral vaginal wall apex is twisted to the right side of the pelvis and vice versa, forming the X-shaped configuration).^{28,44} Nonetheless, the majority of the cases were diagnosed at laparotomy, as in our case report.

Three options are available after intraoperative finding: an attempt of manual uterine detorsion with careful evaluation of possible parametrial vessels damage, posterior lower segment incision, and vertical classic incision. In case of successful manual detorsion, an anterior lower segment incision is possible. Deliberate posterior cesarean hysterotomy is an option with irreducible torsion and sometimes is unaware performed. Additional round ligament plication was sometimes proposed in order to prevent recurrent torsion in the immediate puerperium.⁴⁵ Some authors report possible morbidity with a posterior incision with increased risk of surgical bleeding and uterine rupture in following pregnancies.^{13,24} In few cases, UT was suspected to reduce adnexal blood supply^{38,40} simulating hence an adnexal torsion⁴⁶ and requiring salpingo-oophorectomy.³⁸ Bilateral salpingectomy could be also safely performed for sterilization without increasing complication rate.47

To our knowledge, there are not enough cases available to determine the preferred management and different clinical approaches were reported in literature, as summarize in Table 2. We suggest that ideally the first surgical choice, when possible, is detorsion of the uterus and low anterior segment incision, in order to avoid associated complications of a posterior hysterotomy. Emergency laparotomy is the most frequent approach but in our review two cases of vaginal birth at term are found; in fact, Bukar et al⁴¹ reported a laparotomy approach with derotation at 16 weeks and Wang et al²⁹ published a case report of 22 weeks pregnant woman underwent an emergency laparotomy for torsion of myoma and rudimentary uterine horn. Laparoscopic surgery is not recommended for the management of UT despite recent advances⁴⁸ and wide applicability in challenging surgical cases⁴⁹.

UT is associated with significant morbidity and mortality.^{1,4} Overall maternal mortality rate in Nesbit and Corner³ review in 1956 was 13%, Jensen¹ reported in his review only one maternal death from 1960 to 1992. In our literature review, one maternal death was reported since 2006.⁶ UT could reduce the uterine blood flow leading to a decrease of placental perfusion²² and contributing to the process of IUGR

and Doppler-velocimetry alterations of the reported case report. To our knowledge, this is the second case in literature suggesting this correlation.³⁸ Color Doppler sonography in animal models documented a poor uterine perfusion in case of UT, reporting a high systolic flow and absent diastolic flow high systolic flow.⁵⁰ Nonetheless other factors, such as preeclampsia, might have play a role in the aforementioned case. Lack of previous ultrasound scans prevented to identify the precise torsion timing and further investigation is needed to understand the real clinical impact on uterine vascularization and placental perfusion. In our literature review, 13% of neonatal death was reported consistently with the findings of Jensen and Wilson.

Historically, the most significant factor affecting perinatal mortality was the degree of UT.^{1,3} Blood supply is decreased to the uterus when torsion occurs, venous blood flow was first reduced, and a consequent pressure raise in placental cotyledons leads to fetal distress and abruption. Fetal demise is possible if also arterial blood flow is barred. This pathologic process gives an account of the association with high degree torsion and worse clinical outcome. The literature review since 2006 reveals that torsion grades greater than 180° are associated with more surgical complications (60%), worse postoperative course, and higher incidence of fetal death (40%). Nonetheless, Toshinwal et al¹¹ reported a case of maternal shock and fetal death with a 90° UT underlining the unpredictability of the clinical development.

We analyzed the reported cases with an infaust outcome (maternal or fetal death) to identify similar demographics features or common symptoms of presentation. Two cases ended with fetal death reported a bicornuate uterus but relation with mullerian anomalies cannot be made. Literature demonstrated that fetal mortality and pregnancy outcomes are deeply influenced by a delayed management,⁴³ in fact, promptly intervention is essential to achieve good outcomes. In our literature review, clinical presentations were various and a key symptom was not identified. Nonetheless in all the five cases with poor outcome, vaginal bleeding or cardiotocographic patterns of fetal distress were absent, hence without the chance to provide early management given aspecific symptoms.

In conclusion, UT is an infrequent and potentially fatal complication of pregnancy, it occurs mainly in the third trimester and should be suspected in presence of abdominal pain, vomiting and shock in pregnant women especially if risk factors are known and in absence of a differential diagnosis. Rarely the diagnosis is done with an ultrasound evaluation and frequently UT is demonstrated intraoperatively. Cases of vaginal birth are sporadically reported in literature but emergency laparotomy is usually required. If manual detorsion and a classic anterior incision are not achievable, posterior hysterotomy could be performed taking into consideration possible surgical adverse events. The impact on uterine blood supply in case of chronic torsion could lead to poor placental perfusion, although further investigations are needed to assert evidence. UT has to be promptly managed in order to avoid the rare complication of feto-maternal exitus.

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Conflict of Interest

The authors declare that there is no conflict of interest.

Author Contributions

Federico Ferrari, Filippo Alberto Ferrari and Beatrice Negri contributed to the design and implementation of the research, to the analysis of the results and to the writing of the manuscript. Laura Franceschetti and Sara Forte partecipated to the literature search.

Both Franco Odicino and Enrico Sartori contributed to the final version of the manuscript and supervised the project.

Data Availability Statement

Data available on request from the authors.

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