

Acute Gravid Uterine Torsion: Case Report about an Uncommon Obstetric Emergency

Keywords: Uterine torsion; Round ligament asymmetry; Postpartum hemorrhage

Abstract

Introduction: Uterine torsion is defined by a vertical rotation of more than 45 degrees around its cervical-isthmic axis. It is a rare emergency with serious complications that can be life-threatening for the fetus and the mother. Its pathophysiology has not yet been fully explained but this situation is generally the result of a several factors with mainly the asymmetry of the transverse diameter of the uterus and pelvic adhesions.

Case Presentation: We hereby report the case of a 26-year-old female patient, third gesture fourth pare without particular medical history, who presented to the emergency department at 37 weeks of amenorrhea of a twin pregnancy with severe abdominal pain of sudden onset. Initially mistaken for retroplacental hemorrhage or uterine rupture, it was only in the pre operator phase of an emergency cesarean section that the diagnosis of uterine torsion was made. The operation was complicated by postpartum hemorrhage due to uterine atony which was managed by prompt medical care and a triple Tsirulnikov ligation. She was discharged home with her two healthy newborns at D4 post-op.

Conclusion: Uterine torsion is an uncommon and serious obstetric complication of difficult diagnosis that can be life-threatening for the fetus and the mother. The specificity of our case is twofold. Firstly, it is the third case of uterine torsion in twin pregnancy reported in the literature to date. Secondly, it is the first time to our knowledge that the difference in length of the two round ligaments is observed as a possible factor in the pathophysiology of this complication.

Introduction

Uterine torsion is defined as a vertical rotation of the uterus more than 45 degrees around its axis at the junction of the cervix and the uterus. The first cases of uterine torsion in non-pregnant patients with fibroids were described by Times in 1861 and Virchowen in 1863 [1]. It was not until 1876 that Labbe described the first case of uterine torsion in a pregnant woman [2]. Since then, many cases have been described in the literature, notably in the report of 212 cases published by Jensen in 1992 [2]. The cases described in the published literature generally present up to 180 degrees of torsion, but some go up to 720 degrees [2]. This anomaly has only been observed in three cases of twin pregnancies [3]. Clinical symptoms range from asymptomatic to extreme abdominal pain. It is usually diagnosed late, at the time of caesarean section. We hereby describe the third case of uterine torsion in a twin pregnancy complicated by postpartum hemorrhage.

Case presentation

We hereby present the case of a 26-year-old female patient, third gesture fourth pare with two vaginal deliveries and without particular medical history, who presented to our emergency room with abdominopelvic pain of sudden onset at 37 weeks of amenorrhea



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while the course of her current twin pregnancy was without particularities. On admission, two hours after the onset of the pain, the patient presented with a cutaneo-mucosal heat, a conserved blood pressure (120/60) with tachycardia at 115 bpm. She described severe abdominal pain increased by uterine mobilization. No metrorrhagia or loss of amniotic fluid was observed. The active fetal movements were reduced and we noted the presence of uterine contracture. Obstetric ultrasound revealed a still-evolving twin pregnancy with bradycardia estimated at 80 bpm for the first twin in cephalic position and at 85 bpm for the second twin in seated position and a fundial placenta. The suspicion of uterine rupture or retroplacental hemorrhage led to an emergency cesarean section under general anesthesia. A non-bloody ascites of 300 cc was observed. The uterus appeared ischemic and the territory of the inferior segment was occupied by a vascular network consisting mainly of ecstasic veins. We then observed perioperatively a uterine torsion of 110 degrees to the right. A first attempt at intra-abdominal reduction was unsuccessful and the uterus was still presenting its left adnexa (Figure 1). A segmental Pfannenstiel incision was made and completed with Metzenbaum chisel allowing cephalic extraction of the first twin laterally to the abdomen and then podalic extraction of the second twin after reduction of the uterine



Figure 1: Intraoperative photograph of uterine torsion to the right showing the left adnexa.

torsion. The fetuses weighed 2560g and 2450g respectively and had an Apgar score of 3/5/10 and 4/6/10. The hysterorrhaphy was performed without difficulty. There were no myomas, cysts, adhesions nor malformations but the left round ligament (on the side contralateral to the direction of uterine torsion) was about 5 cm longer than the right. After easy extraction of both twins, the patient presented with heavy bleeding despite complete artificial delivery followed by uterine revision and rapid hysterography. Major uterine atony persisted despite the injection of 40 IU of oxytocin. A triple Tsirulnikov ligation was therefore performed, which made it possible to control the postpartum hemorrhage. We completed the operative procedure with a plication of the left round ligament. After her hemodynamic status was restored, the patient's postoperative course was simple. She was discharged home with her two healthy newborns at D4 post-op.

Discussion

Uterine torsion is defined by a rotation of the uterus by more than 45 degrees in regard to its longitudinal axis. In two thirds of cases, it is dextrorotatory exceeding the physiological dextrorotation [2]. There is only one review of the literature by Jensen et al. of 212 cases [2]. In this review, fetal death in occurred in 12% of cases. Fetal morbidity was reported, sometimes severe, but no maternal death. Most commonly, torsion occurs during labour. Symptomatology included metrorrhagia, cervical dystocia, uterine pain, hyperkinesia, hypertonia, and even hemorrhagic shock [3]. Jensen et al. found that the symptomatology is proportional to the importance of the torsion [2]. Painful symptomatology can be concealed by peridural analgesia. The indication for cesarean section is often based on stagnation of dilatation or abnormalities of the fetal heart rate. Factors favoring torsion are large uterine myomas, uterine malformations, multiple pregnancies, or oblique or transverse fetal presentations [2]. Parity, maternal age and gestational age do not appear to be risk factors. Our case is characterized by a spontaneous occurrence outside labour, in a twin pregnancy.

The etiologies that can explain this pathology are numerous and diverse [2,4]. Among the most widely found causes are four main categories. Firstly, the authors found situations causing an asymmetry of the transverse diameter of the uterus, such as a transverse presentation (22% of cases), the presence of lateralized fibroids (21% of cases), a uterine malformation such as a bicornuate or bifid uterus (11% of cases) and a multiple pregnancy like our case (1% of cases). In this category we can add the important unilateral elongation of the round ligament compared to the contralateral one as in our case. Secondly, we have ectopic pelvic tumors, especially ovarian (3% of cases), followed thirdly by postoperative or idiopathic pelvic adhesions (7% of cases) and finally fourthly by morphological abnormalities of the patient: loose abdominal wall (3% of cases), bone abnormalities of the spine and/or pelvis (1% of cases) [2,4]. In 16% of cases, no etiology was found [2,4]. Although all of these situations are present in many women, torsion remains a very uncommon obstetrical pathology. It is therefore legitimate to wonder whether a combination of events could be at the origin of such a complication. In 1931 Robinson and al. had already put forward this hypothesis followed by Nesbitt et al. in 1956 [5]. The elements defined as being the cause of this anomaly in a patient already predisposed would be a fetal hyperactivity, false maternal movements and postural abnormalities [2,5].

Intraoperative findings frequently include ascites and numerous varices around the isthmic portion [3]. When the torsion exceeds 180 degrees, the diagnosis is difficult to make and the hysterotomy may be inadvertently performed on the posterior aspect of the uterus. If the situation is recognized prior to hysterotomy, a reduction of the uterine torsion can be attempted in order to incise in classic territory as we have attempted. This most often involves uterine exteriorization [3]. If reduction is not possible, it is essential to perform the hysterotomy away from the lateral edges of the uterus to preserve the vascular pedicles and ureters. The choice of a vertical hysterotomy is then preferable [3]. The risk of postpartum hemorrhage as in our case may be increased by acute ischemia of the myometrium responsible for atony which may lead to hemostatic hysterectomy [3]. The literature seems to consider maternal death as exceptional but it is certain that the occurrence of complications such as retroplacental hemorrhage or severe postpartum hemorrhage threaten the maternal prognosis [2,7]. For the clinician, the diagnosis of severe forms of torsion remains difficult and the differential diagnosis is dominated by uterine rupture or retroplacental hemorrhage.

Some authors have attempted to confirm non-acute uterine torsion by imaging. On ultrasound, the placenta may change position to the point of facial inversion, sometimes with ovarian vascular anomalies [7]. Abrupt changes in placental position during extracorporeal circulation, with maternal pain and fetal bradycardia, have been reported as typical cases [8]. MRI can also provide another criterion (abnormal shape of the upper vagina), but is certainly not possible in emergency situations like our case [9]. To date, no study has assessed the risk of recurrence after torsion and determined the need for prophylactic uterine fixation. Fatih et al. published a case of posterior hysterotomy for torsion on the 22nd day of pregnancy without prior surgical intervention. Some authors recommend plication of the round ligament [10,11]. Nevertheless, the real usefulness of this plication remains debatable and should only be performed when there is direct observation of obvious elongation of a round ligament compared with the contralateral one. On the other hand, it seems reasonable to remove large myomas before allowing a new pregnancy.

Conclusion

Uterine torsion is an uncommon and serious obstetric complication of difficult diagnosis that can be life-threatening for the fetus and the mother. The specificity of our case is twofold. Firstly, it is the third case of uterine torsion in twin pregnancy reported in the literature to date. Secondly, it is the first time to our knowledge that the difference in length of the two round ligaments is observed as a possible factor in the pathophysiology of this complication.

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