Asymptomatic Torsion of a Leiomyomatous Gravid Uterus at Term

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Abstract

Background: We are reporting a case of torsion of a gravid uterus. This is a rare occurrence and the non-specific clinical course of the condition make prospective diagnosis difficult and raises some management challenges; however, the diagnosis should be suspected if the uterus contains leiomyomata, especially if complications arise or if abnormal anatomy is detected during caesarean section. In this case, the distorted anatomy during an emergency caesarean section has guided the operator to the diagnosis of uterine leiomyomata causing asymptomatic 180 degrees uterine torsion in pregnancy, and led to the correction of the anomaly prior to the caesarean section.

Method: This is a retrospective and descriptive case report.

Conclusion: Uterine torsion can be asymptomatic. The operator should always ascertain the pelvic anatomy before incising the uterus. We hypothesize that the severe uterine torsion might have compromised the uterine and placental blood flow, and made the fetus not withstand the challenge of mild uterine contractions.

Keywords: Leiomyoma; Gravid uterus; Uterine rotation; Uterine torsion; Caesarean section

Introduction

The incidence of uterine leiomyomata in pregnancy is estimated to be between 0.1 to 3.9 % [1]. Complications arising on a leiomyoma during pregnancy are not common; nevertheless, there are many reported cases in the literature of red degeneration and severe abdominal pain, threatened preterm labour, preterm premature rupture of membranes, in utero growth retardation, placental abruption, placenta previa, postpartum hemorrhage, retained placenta, anaemia, rapid growth, pregnancy loss and rarely torsion of intra-mural leiomyoma [2,3]. The incidences of most of these complications are not significantly different from the general population [3], and the nature of the complication is determined by the location and size of the leiomyomata.

Uterine torsion is defined as rotation of the uterus of more than 45 degrees around its longitudinal axis [4], and uncomplicated uterine rotation during pregnancy is common but rarely exceeds 45 degrees and is mainly to the right [4- 6]. Conversely, uterine torsion of the gravid uterus of more than 180 degrees is a rare obstetrical emergency and might be associated with increased maternal and fetal morbidity and mortality, and is infrequently reported in the medical literature and it is possible that the uncomplicated cases are under reported.

Severe uterine torsion may decrease the uterine blood supply and may obstruct the venous return, and subsequently increasing the venous pressure in placental cotyledons and may eventually lead to placental separation and abortion and subsequently fetal distress [7], and if the obstruction progresses to occlusion of the uterine artery then the placental perfusion may become severely compromised, and may lead to fetal demise [4]. In contrast, the maternal mortality is extremely low and only one death has been reported since 1960 [8].

The non-specific clinical course and the rarity of this condition make prospective diagnosis difficult, if not impossible. Evidently, some obstetricians may not come across such a case during their entire carrier. Here, we present a rare case of leiomyomata causing asymptomatic 180 degrees uterine torsion in pregnancy, which was diagnosed and corrected intraoperative at term during an emergency caesarean section. We also hypothesize that the uterine torsion might have compromised
the uterus and subsequently the placental blood flow and made the fetus unable to tolerate the challenge of mild uterine contractions.

**Case Presentation**

A 37-years old primigravida with regular antenatal care had a booking ultrasound at 11 weeks gestation that showed two intra-mural fundal leiomyomata, with the largest diameter of the larger leiomyoma measuring 10 centimeters [CM] with features suggestive of cystic degeneration, the woman was asymptomatic. The pregnancy progressed satisfactorily and an ultrasound scan at 36 weeks showed that the diameter of the larger leiomyoma has increased to 15 cm. At 39 weeks gestation, the woman developed pre-eclampsia; hence, labour was induced with prostaglandins. Following the insertion of the first dose of the prostaglandin, PGE2, of 2 milligrams vaginally, the fetal heart trace became pathological showing variable decelerations and persistent bradycardia. There were mild uterine contractions, but no signs of labour or cervical dilatation; therefore, an emergency caesarean section was performed on the assumption that the cord was compressed and was not responding to routine measures. Intra operatively, the pelvic anatomy appeared distorted, which has led to the recognition of a180-degrees uterine rotation from right to left with the right ovary and right tube rotated to the left side. Two large uterine intra-mural leiomyomata were palpated in the right side of the fundus, and inadvertent uterine incision on the posterior wall was evaded by proper delineation of the anatomy. The uterine torsion was corrected by exteriorization of the uterus and a lower uterine segment cesarean section was performed. The woman gave birth to a healthy female baby weighing 3 kilograms. The post-operative period was uneventful; the woman was fully debriefed and was discharged home on the third post-operative day. A postnatal ultrasound scan showed the largest diameters of the two leiomyomata were 13 and 10 cm respectively and the patient was booked for myomectomy.

**Discussion**

Uterine torsion during pregnancy is a rare obstetrical emergency; nevertheless, it is associated with significant fetal mortality mounting to 12% and some maternal morbidity [4]. In contrast, maternal mortality is rare and only one maternal death due to uterine torsion has been reported since 1960 [8]. The condition is infrequently reported in the medical literature and it is not certain whether the condition is truly rare, or it is possible that low-risk, uncomplicated cases are underreported.

Uterine torsion occurs when the uterus rotates 45 degrees or more on its longitudinal axis and the rotation can range between 60 to 720 degrees [4] and more commonly to the right [8]. The exact mechanism of spontaneous uterine torsion is not known [4,5], as in most cases the anatomy was normal with no association to; age, parity or gestational age [4]. Though, an etiology may not be found some associated risk factors have been reported; leiomyoma, uterine malformations, pelvic adhesions, ovarian cysts, abnormal fetal presentation, or anomalies and maternal abnormalities of the spine and the pelvis [7]. In their review Nesbitt and Corner [1956] [9] reported an association with uterine leiomyomata in 31.8% of the cases, uterine anomalies in 14.9%, pelvic adhesion in 8.4%, ovarian cysts in 7%, abnormal presentation and fetal anomalies in 4.6% and abnormalities of the maternal pelvis and spine in 2.8%. Uterine torsion was also reported in association with External Cephalic Version (ECV) [10] and the authors have suggested adding this complication to counselling about ECV. Uterine torsion was also reported following maternal trauma and with messaging of a didelphys during labour [11,12].

The clinical presentation is variable and high-degree of suspicion is required, and many of the reported cases were diagnosed incidentally during surgery. Prospective diagnosis is made difficult by the absent and the non-specific symptoms [13,14]. As many as 11% of the cases were asymptomatic however, abdominal pain in association with tenderness on palpation, nausea and vomiting, obstructed labour, vaginal bleeding, shock and urinary and intestinal symptoms have all been described [4,5,7,15].

Fetal distress was suspected from the pathological fetal heart trace. Initially, the assumption was cord compression caused by uterine activity, which did not respond to routine measures; hence, an emergency caesarean section was performed. However, the subsequent intraoperative findings of severe uterine torsion would raise the possibility of the uterine blood supply being compromised by the severe rotation, which became overt with the onset of uterine contractions and reflected by the inability of the fetus to tolerate the challenge of uterine contractions generated by the prostaglandins. It appears that the likelihood of caesarean section is higher in the presence of leiomyomatous gravid uterus [3], and emergency laparotomy is invariably performed in the reported cases and the uterus is usually manually untwisted before delivery, although in some cases this was only possible after delivery. Additional procedures such as myomectomy, ovarian cystectomy or hysterectomy were performed in some cases to correct an encountered precipitating factor or to treat a complication [4,11,16].

When uterine torsion is recognized during laparotomy at term, manual correction followed by delivery of the fetus by a caesarean section is the treatment of choice. However, this is not always possible and there are cases were correction could not be accomplished, or the diagnosis was retrospective and a posterior hysterotomy has been performed either deliberately or unknowingly [6,12-14,16-19]. Both vertical and transverse posterior uterine incisions have been described [7]. In such cases elective section with future pregnancies is advisable. Although the risk of rupture of posterior transverse incision is theoretically less than a posterior vertical incision; nonetheless, the exact risk could not be ascertained [6].

To prevent recurrence of uterine torsion plication of the round ligament was reported [19] this and or the plication of the uterosacral ligament [20] were advocated with the assumption that the procedures will; keep the uterus in anteversion, reduce potential posterior uterine adhesions, reduce subsequent dyspareunia or any recurrence in the future; however, the validity of such procedures is questionable.

Prospective diagnosis is difficult, and some authors suggested that the modification of placental site, or any change in the position of a known leiomyomata on ultrasound images, or the abnormal position of ovarian and uterine vessels across the uterus on doppler should raise the suspicion of torsion [7,8]. Nicholson et al. [21] suggested that an X-shaped configuration of the upper vagina on a Magnetic Resonance Imaging (MRI) as a sign of uterine torsion. This is based upon the observation that the normal vagina appears as an H-shaped structure on MRI images, but with torsion of the uterus and upper vagina, the vagina appears as an X-shaped structure. The possibility of prospective suspicion or diagnosis of uterine torsion using modern imaging techniques in cases with risk factors is promising. Apparently, the MRI overcomes some of the limitations of ultrasonography, like the size of the gravid uterus and offers several advantages like a good overall topographic display, high intrinsic soft-tissue contrast, and less ionizing radiation [22].
Conclusion

Uncomplicated surgery and favourable outcomes are to be expected, if the anatomical landmarks are defined and checked prior to uterine incision during caesarean section; this should disclose any degree of torsion of the pregnant uterus and will minimize the risk of any inadvertent surgery causing damage to the uterus, blood vessels, or any other viscera.

We hypothesize that the severe uterine torsion might have compromised the uterine and placental blood flow, which made the fetus unable to tolerate the challenge of mild uterine contractions.

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References