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## CASE REPORT

# Uterine torsion and subsequent rupture in a gravid bicornuate uterus associated with an elevated alpha-fetoprotein

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**SUMMARY**

Uterine torsion is a rare obstetric complication with a non-specific presentation. We describe a patient with a bicornuate uterus and a pregnancy complicated by a markedly elevated second-trimester maternal serum alpha-fetoprotein (MSAFP), intermittent pelvic pain and fetal growth restriction. At 24 weeks gestational age, she presented to labour and delivery with an acute abdomen. A subsequent exploratory laparotomy revealed torsion and rupture of her right uterine horn. Uterine torsion can be difficult to diagnose because it is not associated with specific symptoms or characteristic imaging findings. In patients with a bicornuate uterus who present with abdominal pain, an elevated mid-trimester MSAFP may be a harbinger of placental ischaemia as a result of uterine torsion.

**BACKGROUND**

Uterine torsion is a rare obstetrical emergency. Although the cause of uterine torsion is unknown, risk factors include uterine anomalies such as bicornuate uterus, fibroids, pelvic adhesions, ovarian cysts and fetal malpresentation.<sup>1</sup> While the complication is not associated with a specific biomarker elevation, certain biomarkers have been associated with adverse obstetrical outcomes in general. Maternal serum alpha-fetoprotein (MSAFP) screening was originally used to detect aneuploidy and fetal malformations such as neural tube and abdominal wall defects. Subsequently, it was discovered that unexplained MSAFP elevation in the second trimester is associated with increased risk of pregnancy complications and adverse obstetrical outcomes including pre-eclampsia, preterm labour, intrauterine growth restriction (IUGR) and intrauterine fetal demise.<sup>2</sup> We describe a case of uterine torsion in a bicornuate uterus initially presenting as an elevated MSAFP.

**CASE PRESENTATION**

A 26-year-old gravida 2 para 1001 at 24 weeks of gestation, dated by last menstrual period consistent with 8 week ultrasound, presented to labour and delivery with 2 hours of severe, constant, right lower-quadrant pain as well as urinary retention, vomiting and syncope. She denied vaginal bleeding or amniorrhaxis. She reported intermittent episodes of similar abdominal pain throughout the pregnancy, including an instance at 22 weeks

where she presented to labour and delivery for sharp right lower-quadrant abdominal pain and vomiting that woke her from sleep. At that time, she had normal vital signs, fetal heart tones and bedside ultrasound. Labs were notable only for an elevated white cell count (WCC) and she was discharged with strict return precautions. Her obstetric history was significant for a bicornuate uterus diagnosed in her previous pregnancy for which she required a low transverse caesarean delivery secondary to malpresentation. Her first pregnancy was in her left uterine horn, this pregnancy was in the right.

Her prenatal course was notable for markedly elevated alpha-fetoprotein (AFP) at 20.91 multiples of the median (MoM) at 17 weeks of gestational age. At 19 6/7 weeks of gestation, a fetal anatomic ultrasound demonstrated no structural abnormalities, specifically no neural tube or abdominal wall defects. But the fetus had asymmetric growth restriction. An amniocentesis for chromosome analysis was performed, which resulted in a 46 XY karyotype. The AFP in the amniotic fluid was within normal range at 1.78 MoM and no acetylcholinesterase was detected. At 23 5/7 weeks of gestation, just 2 days prior to admission, the estimated fetal weight was 397g, below the 10th percentile for gestational age.

At the time of arrival to the hospital, the patient was ill, appearing with rigours but afebrile and haemodynamically stable. The patient's respiratory and cardiac examinations were normal. She had right lower quadrant tenderness without guarding or rebound and her vaginal examination demonstrated a long closed cervix but marked tenderness in the posterior fornix.

Electronic fetal monitoring demonstrated a baseline of 150 beats per minute with minimal variability, no accelerations and no decelerations. Her haemoglobin was 14.8 g/dL and WCC was  $18 \times 10^9/l$ . An abdominal ultrasound revealed an indeterminate tissue mass posterior to the uterus. Immediately following the ultrasound, an MRI of the abdomen/pelvis confirmed ill-defined soft tissue mass with free fluid. Over the next 5 hours, the patient's vital signs did not change, but she declined clinically. She had worsening abdominal pain and distention. Then she developed abdominal guarding and rebound tenderness. At this point, variable decelerations were noted in the fetal heart tracings. Given her deterioration, the decision was



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made to perform an emergency exploratory laparotomy. General surgery was consulted in the event the source of her pain was not pregnancy related.

In the operating room, a vertical midline skin incision was made, and 1.5 L of blood was evacuated from the peritoneal cavity. The right horn of the uterus was torsed and necrotic. Bleeding was noted from an area of rupture on the lateral aspect of the uterine horn. The right fallopian tube and ovary were also necrotic. A hysterotomy was performed through a fundal incision to deliver the fetus followed by amputation of the right uterine horn and right salpingo-oophorectomy. An appendectomy was performed at the recommendation of general surgery. We speculate that the surgeon believed the appendix appeared inflamed with possible necrosis, and an appendectomy was performed to remove all doubt that appendicitis could be contributing to her symptoms. The baby was born alive with a heart rate in the 70s, appeared grossly normal, but weighed 338 g. The patient declined aggressive interventions and elected to focus on the neonate's comfort given fetal weight and gestational age. The infant expired after 45 min of life in the neonatal intensive care unit.

### OUTCOME AND FOLLOW-UP

Approximately 15 months later, she became pregnant in her remaining left uterine horn. This pregnancy was uncomplicated. The AFP was normal, the fetus grew appropriately, and the patient delivered a healthy infant by scheduled caesarean section at 37 weeks of gestation.

### DISCUSSION

The prevalence of uterine malformations due to müllerian defects has been reported to range from 4% to 7%, the most common anomaly being bicornuate uterus.<sup>3</sup> This uterine anomaly is associated with increased incidence of fetal malpresentation, preterm deliveries, miscarriages and fetal growth restriction.<sup>3</sup> A bicornuate uterus is also a risk factor for uterine torsion.<sup>4-6</sup> This may be due to unilateral muscular attachments providing decreased stability and allowing increased mobility which puts the uterus at increased risk of torsion.<sup>5</sup>

A PubMed search demonstrates that this is the sixth reported case of uterine torsion in a bicornuate uterus reported in the literature. In contrast to our case, the patients in the other cases presented with absent fetal heart tones. But consistent with our case, the torsion occurred between 21 and 33 weeks of gestation.<sup>4-6</sup> Thus, it seems that women with bicornuate uteri are at greatest risk of torsion during the mid-trimester.

Elevated mid-trimester MSAFP has been associated with IUGR.<sup>7</sup> As such, a proposed explanation for this finding in the context of a structurally normal fetus includes fetal-placental insufficiency.<sup>7,8</sup> One case-control study found that mid-trimester levels of amniotic fluid angiogenin, a known marker of tissue ischaemia, were significantly elevated in patients with elevated mid-trimester MSAFP levels.<sup>8</sup> Our case of uterine torsion is unique in that the patient demonstrated elevated MSAFP leading up to the time of torsion, the fetus was growth restricted, and the patient had intermittent abdominal pain throughout pregnancy. We hypothesise that the patient's uterus torsed intermittently throughout the first 24 weeks of pregnancy, constricting uterine blood flow which led to placental and myometrial ischaemia. Indeed, this mechanism would account for the patient's pain, the

elevated MSAFP, poor fetal growth and ultimately the uterine rupture.

In conclusion, uterine torsion is a rare but morbid complication in a pregnant patient with a bicornuate uterus. The presentation can be variable and the diagnosis is difficult to make. Our case is particularly unique because the torsion was associated with a markedly elevated MSAFP.

### Learning points

- ▶ A bicornuate uterus has decreased stability and increased mobility which increases the risk of torsion.
- ▶ Uterine torsion should be considered in the differential diagnosis for acute abdominal pain during pregnancy.
- ▶ In the right setting, an elevated mid-trimester alpha-fetoprotein may be a harbinger of placental ischaemia associated with uterine torsion.
- ▶ Uterine torsion is mostly likely to occur between 21 and 33 weeks of gestation.

**Contributors** JL conducted the initial literature review and produced the initial draft of the case report. WY contributed with further literature review, the design of the case report and addition of important intellectual content. They worked together to revise the work. Both authors gave final approval for the published version and agreed to be accountable for all aspects of the work to ensure its accuracy and integrity.

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