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Bilateral massive leiomyomas in a bicornuate uterus, with torsion of the right horn

Valerie O'Besso ,[1] Adi Borovich,[1,2] Scott Chudnoff,[1] Justin To[1]

[1]Obstetrics and Gynecology, Maimonides Medical Center, Brooklyn, New York, USA
[2]Rabin Medical Center, Petah Tikva, Central, Israel

Correspondence to
Dr Valerie O'Besso;
valerie.obesso@gmail.com

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SUMMARY

Uterine leiomyomas are the most common benign tumours of the female reproductive system, often asymptomatic, but can cause significant clinical issues such as abdominal pain. This case report highlights the rare occurrence of a unilateral uterine horn torsion in a bicornuate uterus caused by a massive subserosal leiomyoma. Surgical intervention with myomectomy revealed a previously undiagnosed bicornuate uterus, and the patient's recovery was uneventful. This finding shows the need for clinicians to consider leiomyoma torsion as well as uterine horn torsion in their differential diagnosis of acute abdominal pain.

BACKGROUND

Uterine leiomyomas are the most common benign tumours of the female reproductive system, typically affecting women of reproductive age. While these tumours are often asymptomatic, their size, location and growth can result in various clinical presentations, which include pelvic pain, abnormal uterine bleeding and reproductive challenges.[1] Rarely reported cases of torsion of the stalk of a subserosal leiomyoma may occur, which can lead to acute abdominal pain and may require prompt surgical intervention.[2] Leiomyomas may also uncommonly be noted in patients with congenital uterine anomalies, such as a bicornuate uterus.[3] There are even fewer reported cases of uterine torsion in a bicornuate uterus, those mainly associated in an obstetric setting.[4] This case report highlights the rare combination of unilateral uterine horn torsion on a bicornuate uterus due to massive subserosal leiomyomas.

CASE PRESENTATION

An African American woman in her 30s presented to the emergency room with a chief complaint of acute right lower quadrant abdominal pain. The patient reported a progressive worsening of this pain over the past week, despite using over-the-counter medications. She described the pain as sharp, non-migrating and persistent, with no periods of relief. She also noted episodes of constipation, which she had previously attributed as the cause of her pain in the past. She revealed a 2-year history of intermittent abdominal pain, bloating and abdominal distension, which she assumed was related to weight gain. Additionally, she reported a long-standing history of constipation that was typically relieved with over-the-counter stool softeners. Over the past year, however, she had noticed a marked increase in abdominal distension accompanied by more frequent and severe episodes of pain. Although she had successfully managed her pain with over-the-counter medications such as acetaminophen in the past, this current episode had persisted and worsened despite its use. She denied a history of unintentional weight loss, loss of appetite or difficulty with urination. Her gynaecological history included a history of irregular menstrual cycles with occasional heavy bleeding, and she was previously diagnosed with polycystic ovarian syndrome in 2021. She endorsed a known history of leiomyomas since 2020 but was not aware of the size or any other uterine abnormalities. She denied the use of contraceptives and had never been pregnant or attempted pregnancy before. She denied any medical issues or prior abdominal surgery. She had a desire for future fertility.

On examination, the patient was alert and oriented, with some discomfort. Her vital signs were within the normal range. Her body mass index was 56.2 kg/m^2. Her abdominal examination was significant for large, non-mobile, solid, palpable masses on the left and right upper abdomen abutting the rib cage with tenderness to palpation and occupying most of the abdomen. The pregnancy test and the urinalysis were negative. She had a complete blood count showing a haemoglobin level of 10.4 g/dL and an unremarkable basic metabolic panel. Due to the size of the leiomyomas, the following tumour markers were obtained: cancer antigen (CA) 19–9, carcinoembryonic antigen, beta-human chorionic gonadotropin, inhibin B, alpha-fetoprotein and lactate dehydrogenase. Only the CA-125 level was elevated at 102.1 U/mL. The CT and MRI reports described two large dominant masses that appeared to arise from the uterus, measuring approximately 27 cm maximum dimension on the left and 24 cm maximum dimension on the right and favoured to be large myomas (figure 1). The patient was then hospitalised for pain optimisation with intravenous ketorolac and transitioned to oral acetaminophen and ibuprofen. After pain optimisation, the patient was counselled on her options, risks and benefits of treatment. She desired an abdominal myomectomy and was scheduled for surgery the following day.

The surgery was performed through a midline vertical incision extending to the umbilicus to access the abdomen. On abdominal cavity entry, the right-sided leiomyoma base, classified as International Federation of Gynaecology and

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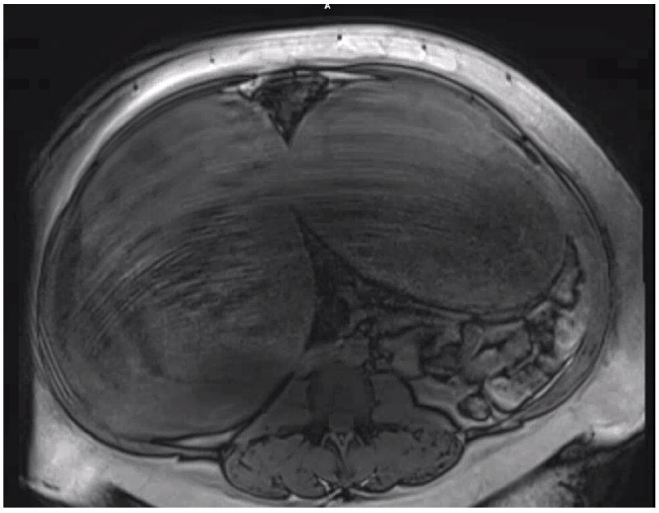

Figure 1 An MRI scan image demonstrating two large dominant masses filling the abdomen and pelvis, giving the impression of the leiomyomas. Axial T1-weighted MRI images of the pelvis reveal findings of two rounded heterogeneous leiomyomas, each one originating from the uterus.

Obstetrics (FIGO) stage 6, was noted to be torsed 360° and was starting to avulse. The leiomyoma was exteriorised, and its base, which was later identified as the right uterine horn, was successfully detorsed. Next, the second leiomyoma was delivered along with the uterus. On further inspection, both large-appearing myomas originated from the fundus of each horn of a bicornuate uterus and were both FIGO stage 6 in nature. Each myoma measured approximately 25 cm in diameter (figure 2). To reduce operative blood loss, a Penrose tourniquet was used to compress the uterine vessels. To perform this, a window was made in an avascular portion of the broad ligament using a Bovie electrocautery tip at the level of the internal os. The ends of the Penrose drain were threaded through the windows, pulled tight and secured with a Kelly clamp to compress the uterine vessels. Vasopressin was also injected at the base of the leiomyoma. Myomectomy was performed with Bovie electrocautery and blunt dissection. The incisions were closed in multiple layers using V-loc suture, and complete haemostasis was achieved. The Penrose tourniquet was removed, and during reperfusion, the incisions remained haemostatic. The bicornuate uterus was clearly identified after closure (figure 3). The patient had an uneventful recovery period. Postoperative haemoglobin level was 9.8 g/dL. Pathology confirmed the masses were benign leiomyomas, showing a typical whorled appearance with a

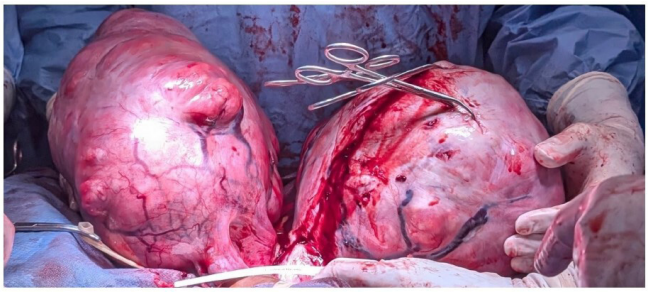

Figure 2 Intraoperative findings of two large subserosal leiomyomas with detorsion of the right uterine horn performed.

Figure 3 Picture of the removed leiomyomas from each of the uterine horns measuring a total of 9835 grams. Pathology report confirmed diagnosis of uterine masses as uterine leiomyomas with a moderate amount of tan-yellow calcifications noted.

moderate amount of cystic changes and tan-yellow calcification. The total weight of both leiomyomas on the pathology was 9835 grams (figure 4).

OUTCOME AND FOLLOW-UP

The patient returned for her postoperative appointment a month later with significant improvement in daily activities, less weight burden and the relief of knowing her diagnoses of both benign leiomyomas and a bicornuate uterus. Education on bicornuate uterus, including future fertility concerns, was provided. The patient was unable to attend further follow-up visits due to the inability to take off from work. However, our plan is to assess the uterine cavity with office hysteroscopy.

DISCUSSION

The case presented involves the rare occurrence of bilateral large subserosal leiomyomas located on each uterine horn of

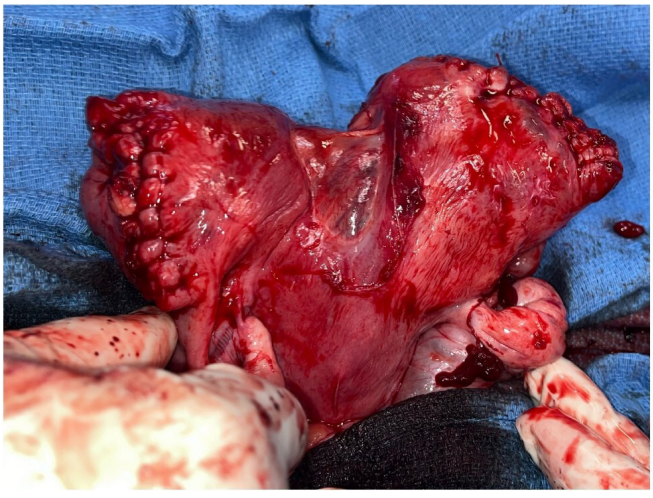

Figure 4 After myomectomy was performed, the finding of a bicornuate uterus was confirmed with serosal indentation seen with initial signs of avulsion secondary to its large subserosal leiomyoma on the medial aspect of the right uterine horn from torsion of the leiomyoma.

a bicornuate uterus, with an intraoperative finding of unilateral uterine horn torsion secondary to its leiomyoma.

Leiomyomas are benign smooth muscle tumours arising from the myometrium. The aetiology of leiomyomas is multifactorial, with genetic predisposition, hormonal factors and growth factors contributing to their development.[1] Torsion of pedunculated uterine leiomyomas is extremely rare, with a reported incidence of less than 0.25%. Torsion of a pedunculated leiomyoma may be considered a surgical emergency due to the risk of ischaemia and necrosis leading to reactive peritonitis and morbidity.[2 5] However, pedunculated subserosal uterine leiomyomas may also be asymptomatic or can be associated with varying degrees of pain. Although the diagnosis of torsion of a pedunculated subserosal uterine leiomyoma is typically confirmed intraoperatively in most cases, imaging modalities can provide valuable preoperative information. Doppler ultrasonography can raise suspicion of leiomyoma torsion by detecting reduced blood supply when the vascular pedicle is visible. In cases of inconclusive ultrasound findings, ideally, an MRI is performed for its higher sensitivity and specificity. The vascular pedicle of a subserosal leiomyoma is better appreciated on MRI in the form of T2 flow voids at the interface between the uterus and the mass indicating the 'bridging vessel sign', suggesting a uterine origin of the mass.[6] In our case, an MRI was performed, but neither torsion nor the uterine anomaly were identified, perhaps due to distortion by the leiomyomas.

This case also demonstrates the discovery of a bicornuate uterus intraoperatively with massive subserosal leiomyomas; our literature review on leiomyomas with congenital uterine anomalies showed very few results.[7] A bicornuate uterus is a uterine malformation produced due to abnormal fusion of the Müllerian ducts. It is a rare anomaly and associated with reproductive outcomes such as recurrent pregnancy loss and preterm labour.[8 9] Though the prior-reported cases of leiomyomas in bicornuate uteri have only been intramural and submucosal in location, our case presents the development of remarkably large subserosal leiomyomas, both of which were FIGO stage 6, on each uterine horn.

Though leiomyoma torsion itself is rare, our case shows that uterine horn torsion should also be on the differential of subserosal leiomyoma torsion. Some elements of uterine torsion can be seen in figure 4, where the medial aspect of the right uterine horn was starting to avulse. Due to the rarity of the co-occurrence of large leiomyomas located on each uterine horn of a bicornuate uterus, one may mistake the stalk of a leiomyoma for the uterine horn without careful dissection and adequate blood control. This case highlights the importance of preoperative surgical planning, review of imaging and adding the differential diagnosis of uterine torsion in the setting of large leiomyomas and unknown uterine anatomy prior to removal of leiomyomas.

In an acute setting of abdominal pain with a large uterine leiomyoma, leiomyoma and uterine horn torsion must be considered in the differential diagnoses. Though uterine anomalies are rare, patients with a history of abnormal uterine bleeding, recurrent pregnancy loss, preterm birth or infertility may have an underlying and undiagnosed diagnosis. Thus, comprehensive preoperative and intraoperative evaluation, including assessment of possible Müllerian anomalies and detailed leiomyoma mapping, is necessary to minimise inadvertent damage to a compromised uterus for optimal surgical outcomes and patient safety.

Patient's perspective

I knew about my fibroids back in 2020, but due to the COVID-19 pandemic, I had issues with follow-up visits and getting my MRI to see why my stomach felt so large, hard and painful at times. Initially, I wasn't even told about my fibroids when I got my imaging done, only that I had PCOS. Then, as time passed, I felt the pain gradually get worse, my clothes were starting to not fit, but my diet was always the same. Walking down the block or up the stairs felt like running a marathon while carrying extra baggage, and I would try to avoid stairs and take the elevators as much as possible. Eventually, the pain got so bad, I had to seek help. In the emergency room, I was glad to finally get some answers as to why I've been having pain this whole time and my stomach was getting so big.

Before surgery, I was excited to get these fibroids out, but at the same time, I was scared of losing my uterus. After the surgery, I woke up noticing a flatter stomach and feeling lighter. At my postop visit, I saw a photo of my fibroids that were removed, and they were huge! I am so thankful everything went great. I also now know that I have a heart-shaped uterus. To my surprise, the recovery period was quick, and I even went to work earlier than anticipated. I can finally walk long distances with no hesitation, and I can return to my old lifestyle that I haven't experienced in a long time.

Learning points

- Leiomyomas in bicornuate uteri are uncommon.
- In obese patients, large leiomyomas can be clinically missed.
- Coexistence of congenital uterine anomalies should be considered in patients with severely leiomyomatous uteri.
- Torsion of a single horn of a bicornuate uterus can occur, which may not be detected clinically.
- When assessing abdominal pain with imaging showing likely leiomyomas, the differential should include torsion of either the stalk of a leiomyoma or of a uterine horn.

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ORCID iD
Valerie O'Besso http://orcid.org/0000-0002-0596-1316

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