Case Report

Acute late presentation of a functioning non-communicating rudimentary uterine horn containing an adenomyosis: A case report

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Abstract

A functioning noncommunicating rudimentary horn is a rare uterine malformation. The presence of rudimentary uterine horn with adenomyosis is even rarer situation. Clinical presentation varies from mild pain that might present late in the clinical course with complications that can be gynecological such as pelvic pain and endometriosis or obstetrical such as preterm delivery, cesarean section, and ectopic pregnancy. We are reporting a case of a young woman who presented with acute abdominal pain that was superimposed by chronic pelvic pain due to endometriosis and deep pelvic vein thrombosis secondary to an enlarging noncommunicating rudimentary uterine horn containing extensive adenomyosis. With the help of MRI, initial diagnosis was given as rudimentary functioning horn containing fibroid and unilateral renal agenesis. The treatment comprised complete laparoscopic excision of the entire horn, and the patient reported significant improvement afterward. Final histopathology was rudimentary horn containing adenomyosis. Our paper is one of few papers reported adenomyosis in function noncommunication rudimentary horn.

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Introduction

A unicornuate uterus with a noncommunicating functioning horn is a challenging clinical situation in terms of its presentation, diagnosis, and management. Treatment is usually tailored to the case clinical presentation [1].

Unicornuate uterus is uncommon, with an estimated prevalence of 0.1% in the general population [2]. The prevalence of this condition in the reproductive group desiring pregnancy is even much lower approaching 0.06% [3]. In fertile women, the prevalence of unicornuate uterus with rudimentary horn is reported to be 1 in 100,000. Unicornuate uterus has 4 subtypes based on the American fertility

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society classification: up to 90% of cases have a rudimentary uterine horn, but only 25% of them are cavitated and noncommunicating [4,5].

Very few published papers are available in the literature regarding cases of adenomyosis in the rudimentary horn [6,7]. The presentation of this condition is variable. Due to obstruction in the uterus, hematomata may accumulate, causing mass and pain. However, the pain might be mild sometimes, leading to delayed diagnosis. The possibly associated retrograde menstruation may lead to endometriosis. As cases of rudimentary horn may present with obstetric and/or gynecological complications, early prophylactic surgical intervention has been advocated [5,2].

Case presentation

A 26-year-old woman who had previously 4 deliveries has attended the emergency department at King Abdulaziz medical city complaining of severe left-sided pelvic pain radiating to the left thigh. This characteristic of pain was known to the patient since her menarche at the age of 16 years with on and off exacerbation of severe attacks. Due to the severity of pain she usually requires repeated doses of several analgesia agents (ibuprofen 600 mg and 1000 mg paracetamol). The requirement usually starts 2 days before her menses and last during its first 2 days. She had never taken contraceptive or hormonal medication. Her cycles had always been regular. She had undergone 4 deliveries; the first one was a vaginal delivery followed by 3 cesarean sections. All her pregnancies were full term, except the last delivery, which was at 35 weeks. Her babies’ weight was ranging between 2.9 and 3.6 kg. The patient’s initial diagnosis was established in 2012 when a heterogeneous mass measuring (5 x 4.6 x 5.9 cm) was detected during a regular gynecological ultrasound scan (fig. 1). A hysterosalpingography was performed (Fig. 2) that revealed unicorunate uterus patent right tube. Subsequently, the patient did not report again till 2018 while pregnant at 27 weeks of gestation. At that time, the uterine mass had slightly increased in size to measure (9.8 x 9.4 x 8.8 cm). A cesarean section was performed at 35 weeks on an emergency bases. The intraoperative findings revealed a unicorunate uterus with rudimentary horn and severe adhesions between the uterus and the surrounding viscera. A postpartum follow-up to complete the patient’s assessment and plan her possible surgical intervention was initiated; unfortunately, the patient did not show up to her appointments after her postpartum period.

Approximately 28 months after her last delivery, she reported to the emergency room twice with increased pelvic pain and vomiting. Her pain score was 10/10, with no other gastrointestinal or urinary symptoms. On examination, her abdomen was soft with significant tenderness and rebound pain in the left lower quadrant. Vaginal examination revealed a normal vagina with the cervix shifted to the right side. The laboratory results were normal, and a serum pregnancy test was negative. Ultrasound and computed tomography scan (CT scan) showed a rudimentary uterine horn containing a fibroid. There was left gonadal vein thrombosis and left-sided renal agenesis (Fig. 3). Emergency laparoscopy was performed under general anesthesia. A large mass was detected occupying the left pelvic side, with endometriotic spots and hydros-
alpinx on the left side (Fig. 4). The unicornuate uterus was seen pushed towards the pelvic wall because of the rudimentary horn mass. The mass was mainly solid, containing what looked like a fibroid, with some cystic areas and was completely not communicating with the right side and attached to it with thick membranous tissue. Excision of the rudimentary horn with its attached mass was decided. The procedure was started by isolating the mass from the left pelvic side wall. The left ovary and tube were morbidly adherent to the posterior aspect of the rudimentary horn and were difficult to be separated from. The left uterine artery was skeletonized and cauterized, followed by aspiration of the cystic area content to shrink the mass size and ease morcellation. The aspiration yielded the presence of dark brown chocolate like fluid. Complete excision and morcellation of the rudimentary horn with the attached mass was then done. The entire procedure was laparoscopic right rudimentary horn excision with right salpingoophorectomy.

Histopathological examination revealed a benign fallopian tube with pseudoxanthomatous salpingiosis, endometriosis, and serosal fibrous adhesions. The benign uterine tissue showed secretory endometrium with stromal breakdown and myometrium with adenomyosis and serosal fibrous adhesions. Although images were showing what looks like fibroid but final histopathology confirmed presence of extensive adenomyosis with no presence of fibroid.

The patient reported improvement in pelvic pain immediately postoperatively; repeated CT scan postoperative showed resolution of the previously identified pelvic vein thrombosis. The patient followed up as an outpatient 10 months after surgery. She had no further pain attaches during menstruation and no further need for analgesia or hormonal treatment.
since surgery. Consent was taken from the patient to publish the case details.

## Discussion

The clinical presentations of rudimentary noncommunicating horn are variable [4]. It might be a late presentation, as in our case where the patient had chronic pelvic pain due to the increasing size of the obstructed horn and the sequelae of retrograde bleeding, such as endometriosis and adhesions. Up to 55% of patients with a non-communicating rudimentary horn are reported to develop active endometriosis [8]. The acute pain superimposed on her chronic pain at the time of her admission was most probably aggravated by the deep pelvic vein thrombosis. Poor pregnancy outcomes are reportedly linked to unicornuate uteri, with the risk of preterm delivery as high as 44% [9,2]. Fortunately, our patient had most of her deliveries at term or near term with relatively good outcomes. Although Impression of fibroid based on preoperative images and intraoperative findings bulky rudimentary horn was suspected.

Fibroids in uteri with rudimentary horns are extremely rare, with very few reported cases [10–12]. Furthermore presence of adenomyosis in rudimentary horn is been rarely reported in literature [6,7].

Our patient had ipsilateral renal agenesis, which is known to be associated with such malformations and has been reported in 50% of all cases of obstructive Mullerian anomalies [4].

Hysterosalpingogram, ultrasound, particularly 3-dimensional ultrasound are known to be helpful diagnostic tools. However, the gold standard tool for all uterine malformations is magnetic resonance imaging (MRI) [3,12].

As our case presented in the acute stage, MRI was not practical. For cases presenting in acute or emergency situations, ultrasound is the preferable initial diagnostics tool, as it has a sensitivity of 29%-33% for diagnosing rudimentary horn [5].

Surgical excision of the rudimentary horn was achieved, although we believe that early excision would have been better as prophylactic removal of a cavitary rudimentary horn can prevent known complications such as endometriosis, ectopic pregnancy, dysmenorrhea, or even life-threatening conditions such as rupture of rudimentary horn in pregnancy [4,8]. Furthermore, excision of the rudimentary horn early enough before adhesions formation could have saved the right ovary that has to be removed with the specimen due to morbid adhesions. Finally, quality of life for this patient could have been improved years prior to the time of the surgery in the presence of early surgical intervention.

In our case, the surgery was performed laparoscopically, which is a common approach for surgical excision and confirmation of diagnosis [6]. Laparotomy is an available option when surgical experience with laparoscopy is limited and/or if malignancy is suspected [13–15]. Though we are presenting a case report that has its own limitations, we believe that our case is one of very few cases of obliterated rudimentary horn that contains extensive adenomyosis, more cases and papers would benefit better understanding of this complex pathology.

### Conclusion

Very few cases of noncommunicating rudimentary horn are reported, especially of rudimentary horn containing extensive adenomyosis. Obstetric and gynecological complications frequently occur in such cases. Gynecologists need always to maintain a high index of suspicion and timely intervention.

### Patient consent

We are confirming that written, informed consent for publication of our case was obtained from the patient.

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