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A case of adenomyosis in a noncommunicating functional rudimentary horn mimicking a subserous myoma

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Abstract:

This report discusses a rare case of adenomyosis in a noncommunicating functional rudimentary horn, initially described as a subserous myoma on imaging. A 35-year-old nulligravid presented with dysmenorrhea since menarche, acute right lower quadrant pain, and a palpable right pelvic mass. A myomectomy was initially planned, but during the laparoscopic procedure, a rudimentary horn adjacent to a unicornuate uterus was discovered. The patient underwent a second procedure for diagnostic hysteroscopy, diaphonoscopy, chromotubation, laparoscopic excision of the rudimentary horn, and right salpingectomy, recovering without complications. Histopathology revealed diffuse adenomyosis with proliferative endometrium. The case emphasizes the challenges in diagnosing such conditions through imaging. It highlights the importance of considering Müllerian anomalies in pelvic mass diagnoses, particularly when atypical symptoms or imaging findings are present.

Keywords:

Adenomyosis, pelvic mass, rudimentary horn, subserous leiomyoma, unicornuate uterus

Introduction

Congenital anomalies of the female urogenital tract are rare deviations from normal anatomy, often affecting reproductive function. The American Society for Reproductive Medicine (ASRM) provides a standardized framework for categorizing Mullerian anomalies into several classes based on uterine anatomical deviations and embryological origins.[1] A unicornuate uterus is formed due to the underdevelopment of one Mullerian duct, sometimes associated with a rudimentary horn in 65% of cases.[2] This presence of a rudimentary horn can complicate the clinical presentation and imaging interpretation of pelvic masses, particularly when associated with pathologic conditions that involve the uterine myometrium such as adenomyosis.

This paper presents a rare case of adenomyosis in a noncommunicating functional rudimentary horn, detailing the presentation, diagnostic challenges, and surgical management. It also explores the embryological development of this anomaly.

Case Report

A 35-year-old nulligravid presented with a history of dysmenorrhea since menarche and severe pelvic pain of 3 months' duration, with no associated abnormal uterine bleeding. She had no relevant comorbidities or family history. Pelvic examination revealed a normal vagina and cervix with a palpable mass at the right adnexa. An initial transvaginal ultrasound showed a heterogeneous mass at the right lateral portion of the uterus described as a subserous myoma measuring

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4.0 cm × 4.9 cm × 4.4 cm [Figure 1]. She was then referred to the section for minimally invasive gynecologic surgery for laparoscopic myomectomy.

On laparoscopy, the previously identified subserous myoma measured approximately 5 cm × 5 cm and was fused to the right lateral portion of a unicornuate uterus. Attached laterally was a normal right fallopian tube and right ovary, which appeared anatomically continuous with the ipsilateral broad ligament [Figure 2]. There were endometriotic implants on the bladder peritoneum. The planned myomectomy was abandoned, and further evaluation was performed postoperatively. Pelvic magnetic resonance imaging (MRI) with contrast was done, which described the presence of a right adnexal mass to consider uterine leiomyoma with subserous component and unremarkable bilateral kidneys [Figure 3].

The patient was advised and consented to surgical intervention consisting of combined laparoscopy and hysteroscopy, diaphonoscopy, chromotubation, excision of the right rudimentary horn, and right salpingectomy under general anesthesia.

With the patient in dorsal lithotomy position under general anesthesia, one 10-mm infraumbilical port and two 5-mm accessory ports lateral to the right and left inferior epigastric artery were created. Intraoperatively, dense adhesions were noted between the right lateral wall of the rudimentary horn and the pelvic sidewall [Figure 4]. Hysteroscopy showed a narrow and tubular uterine cavity with a normal left tubal ostium, characteristic of a left unicornuate uterus. The right tubal ostium was not seen [Figure 5]. Diaphanoscopy showed absent translucency of the hysteroscopic light at the rudimentary horn [Figure 6]. Bilateral ureters were identified. Adhesiolysis was done using blunt dissection and bipolar electrosurgery to free the rudimentary horn from the right pelvic sidewall [Figure 7]. The right uterotubal ligament, utero-ovarian ligament [Figure 8],

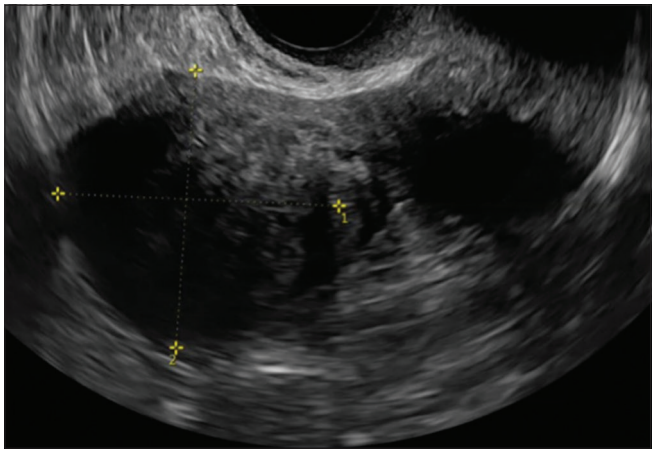

Figure 1: Transvaginal ultrasound in transverse view, showing a heterogeneous mass at the right posterolateral portion of the uterus described as a subserous myoma

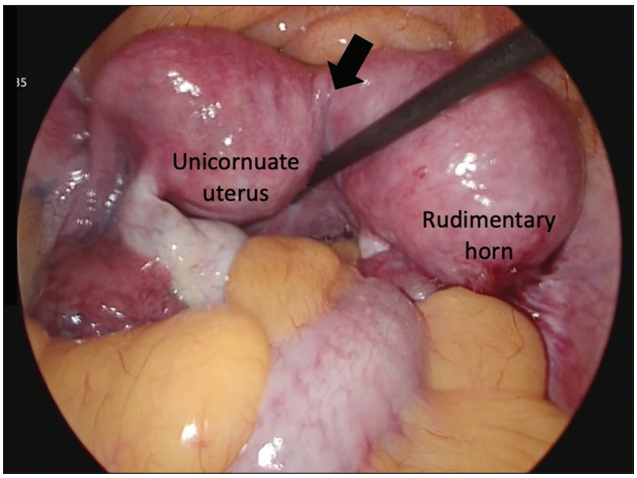

Figure 2: Left unicornuate uterus attached to a rudimentary horn by a fibrous band (black arrow)

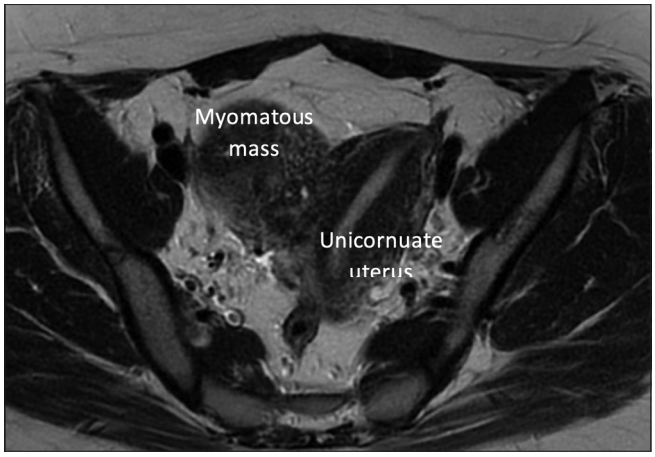

Figure 3: Pelvic magnetic resonance imaging with contrast describing a right adnexal mass, to consider a uterine leiomyoma with subserous component

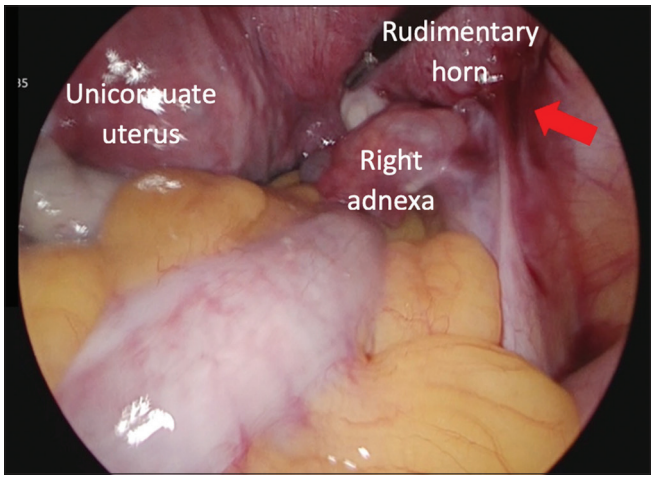

Figure 4: Rudimentary horn along with its attached ovary and fallopian tube, showing dense adhesions to the right pelvic sidewall (red arrow)

and right round ligament [Figure 9] were coagulated and transected using a curved bipolar device. The broad ligament and uterovesical fold were dissected up to the attachment site [Figure 10]. The uterine artery originating at the base of the fibrous band was identified, coagulated, and transected [Figure 11]. The rudimentary horn was excised carefully along a fibrous band [Figure 12]. The right mesosalpinx was coagulated and transected; the right salpingectomy was done [Figure 13]. Chromotubation with egress of dye on

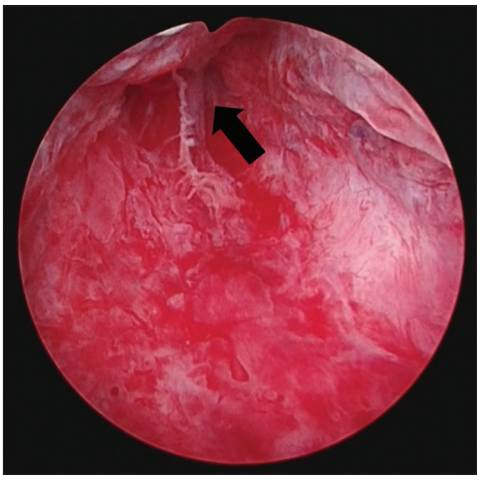

Figure 5: The hysteroscopic image displays a narrow and tubular uterine cavity with a visible left tubal ostium (black arrow), characteristic of a left unicornuate uterus. The right tubal ostium was not visualized

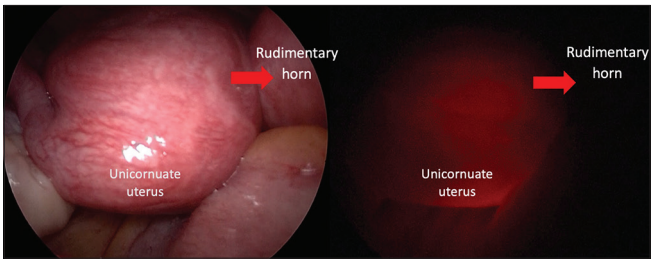

Figure 6: This image illustrates the absence of translucency of the hysteroscopic light at the rudimentary horn, as indicated by the red arrow, both before and after the laparoscopic light was turned off

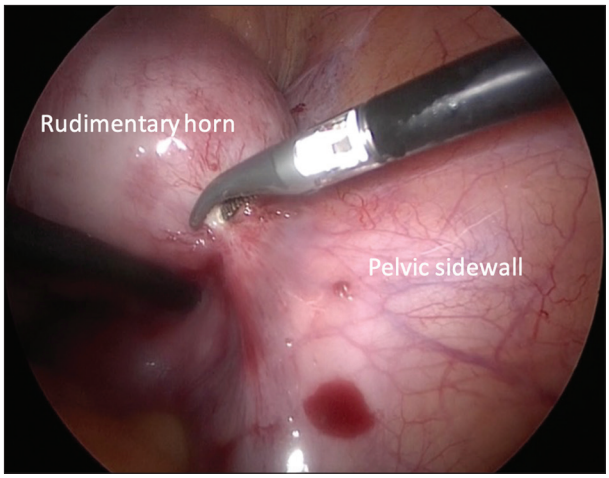

Figure 7: Adhesiolysis was done using blunt dissection and bipolar electrosurgery to free the rudimentary horn from the right pelvic sidewall

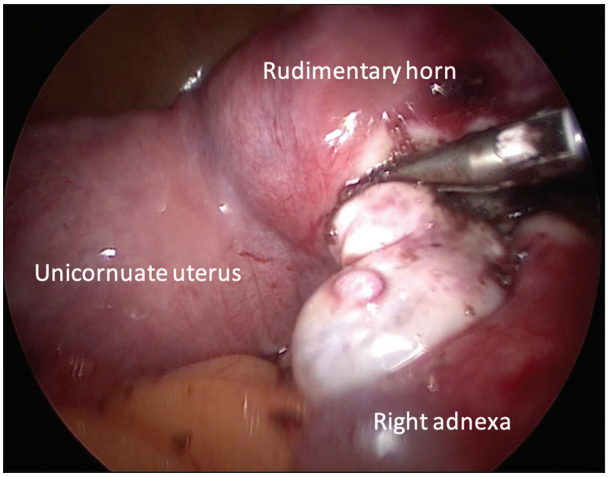

Figure 8: The right utero-ovarian and uterotubal ligament was transected using a curved bipolar device

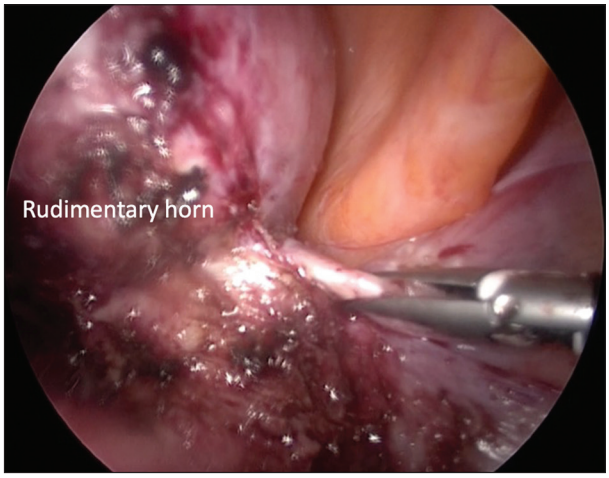

Figure 9: The right round ligament was coagulated and transected using a curved bipolar device

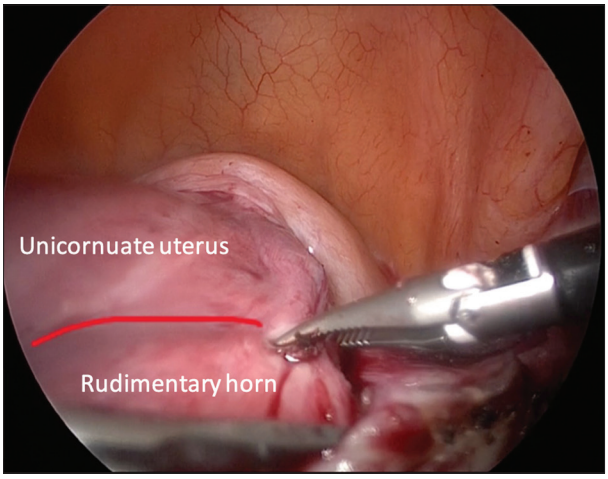

Figure 10: The dissection of the broad ligament and uterovesical fold up to the attachment site, indicated by the red line

the left fallopian tube was noted while no evidence of dye was seen at the site of resection [Figure 14]. The left lateral port was extended to 3 cm, and the specimen was retrieved. The postoperative period was uneventful, and the patient was discharged on the second postoperative day. Histopathological examination of the rudimentary horn revealed diffuse adenomyosis with proliferative endometrium, while the right fallopian tube was unremarkable. The final diagnosis was a unicornuate uterus with a functional noncommunicating right rudimentary horn with adenomyosis. Follow-up was done after 4 weeks, and the patient was symptom free.

Discussion

Mullerian anomalies are congenital malformations of the female reproductive tract that result from abnormal development of the Mullerian ducts during embryogenesis, with a prevalence of 0.5%–6.7% in the general population.[1] These anomalies can affect the uterus, cervix, and upper two-thirds of the vagina and often have a significant impact on reproductive health.

The ASRM categorizes these anomalies into different groups based on their anatomical deviations and embryological origins.[2] A unicornuate uterus results from the incomplete fusion of the two Müllerian ducts, occurring in approximately 0.4% of all women.[3] A rudimentary horn is a small, underdeveloped structure originating from the same Müllerian duct that failed to fully develop into the main uterus.[4] Notably, around 74%–90% of unicornuate uteri are associated with a contralateral rudimentary horn.[4]

The other female genital tract anomaly classification is the European Society of Human Reproduction and Embryology/European Society for Gynaecological Endoscopy system based on the main uterine findings (U0–U6) and cervical/vaginal anomalies (C0–C4; V0–V4); the present case was initially categorized as U4b C0 V0.[5]

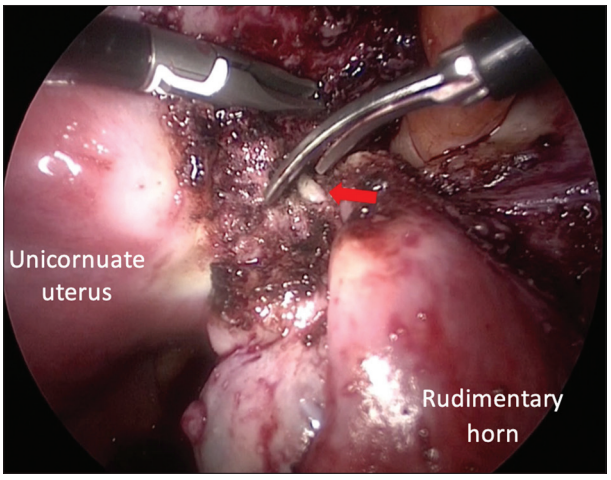

Figure 11: The uterine artery (red arrow), originating at the base of the fibrous band, which has been identified, coagulated, and transected

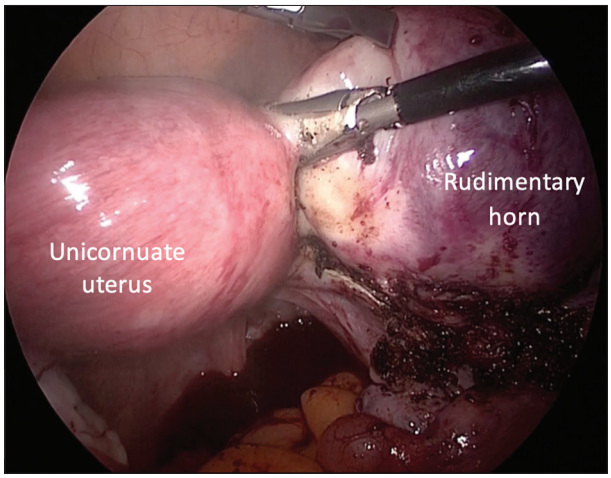

Figure 12: The excision of the rudimentary horn along a thick fibrous band using a curved bipolar device

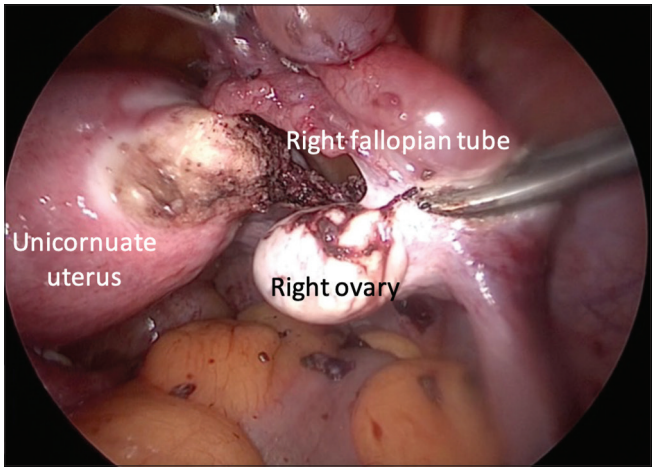

Figure 13: A right salpingectomy with coagulation and transection of the right mesosalpinx

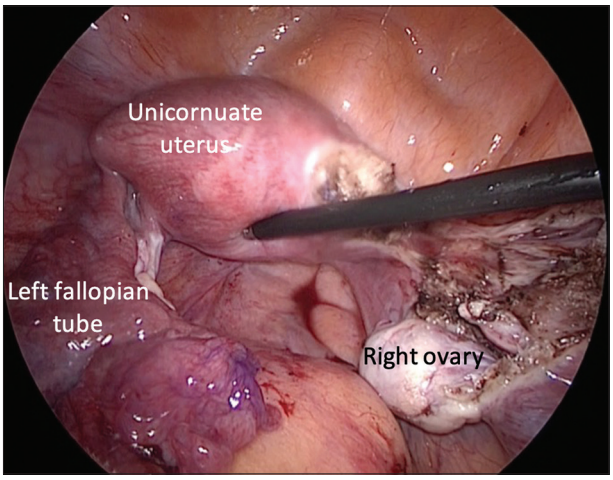

Figure 14: This postoperative image shows chromotubation, with dye egress through the left fallopian tube, and absence of dye at the resection site

Rudimentary horns may be functional or nonfunctional and communicating or noncommunicating. A functional rudimentary horn contains active endometrial tissue that responds to hormonal cycles, leading to menstrual bleeding, whereas a nonfunctional rudimentary horn lacks active endometrial tissue and does not undergo menstrual changes. A communicating rudimentary horn has a direct connection with the main uterine cavity, allowing passage of menstrual blood, whereas a noncommunicating rudimentary horn is isolated, leading to hematometra.[4]

About 55% of cases involve noncommunicating rudimentary horns with a functional endometrium that may fill with blood or fluid, causing obstruction and pain.[4] These patients present with different symptoms. In patients who have started menstruating, these anomalies often cause acute or chronic pelvic pain. It was previously believed that this was due to a blocked outflow for the endometrium, causing uterine distention, uterine contractions, compression of adjacent pelvic structures, and induction of an inflammatory process including endometriosis.[4] However, it has been found that even nonfunctional horns can still cause pain, and patients have reported relief after these horns are removed. It was hypothesized that the growth of leiomyomas and the presence of a degenerative process, such as fibroid degeneration, in a nonfunctional rudimentary horn, may cause pain and discomfort.[6] Fedele *et al*. analyzed 10 functional rudimentary uterine horns using light and scanning electron microscopy, revealing that abnormalities at the endomyometrial junction may limit endometrial shedding, leading to a decreased formation of hematometra. In addition, they observed that imperfect tissue differentiation in these horns contributes to adenomyosis, underscoring the distinct pathology of these structures.[7,8]

At the time of presentation, a pelvic mass may or may not be present, which is observed in approximately 20% of patients admitted to the hospital with similar conditions.[9] This variability in presentation is due to the nature of rudimentary horns, which can consist of solid muscle, areas of adenomyosis, or hypoplastic tissue.[8] In the discussed case, the presence of a palpable right pelvic mass was one of the key clinical findings, which, combined with the patient's history of dysmenorrhea and acute pain, initially led to a diagnosis of a subserous myoma.

Adenomyosis and leiomyoma are both benign conditions of the uterus that often result in symptoms that may mimic malignancy, such as heavy menstrual bleeding and pelvic pain.[10] Adenomyosis involves the presence of endometrial tissue within the myometrium, leading to symptoms from abnormal endometrial tissue growth and elevated levels of prostaglandins, which cause dysmenorrhea and heavy menstrual bleeding.[11] Reports describing adenomyosis in relation to Mullerian anomalies are rare, with one such case of rudimentary horn adenomyosis reported in a 20-year-old patient.[12]

The pathogenesis of adenomyosis remains complex, with two primary hypotheses: invagination and metaplasia. The invagination hypothesis suggests that adenomyosis results from repeated tissue injury and repair, leading to the endometrial layer's invagination into the myometrium. This theory is supported by the higher incidence of adenomyosis in individuals with high parity or a history of uterine surgery. Conversely, the metaplasia theory suggests that adenomyosis originates from the metaplasia of displaced pluripotent epithelial cells from the Müllerian ducts or remnants during their fusion, which could be relevant in the index patient's case.[13]

Diagnosing these conditions can be difficult and typically relies on imaging techniques. MRI is frequently used to diagnose adenomyosis, showing characteristic features such as poorly defined low-signal-intensity areas within the junctional zone.[14] However, transvaginal ultrasound is also a valuable option for diagnosing Müllerian anomalies due to its low cost and better patient tolerance. Ultrasound is typically employed for diagnosing leiomyomas and identifying distinct fibroid masses.[15] The overall sensitivity of ultrasonography in definitively diagnosing a rudimentary horn is 26%, with only 14% of cases being identified before the onset of symptoms.[5]

Adenomyosis and leiomyomas share several imaging similarities on MRI and ultrasound, which can complicate their differentiation. On MRI, both conditions may appear as areas of low signal intensity on T2-weighted images, exhibit junctional zone thickening, and show myometrial heterogeneity. They also present variable enhancement patterns after contrast administration.[14] Ultrasound imaging reveals that both adenomyosis and submucous myomas can display heterogeneous echotexture and ill-defined margins.[11] Posterior acoustic shadowing is common in both, although more frequently observed with myomas, and small cystic areas within the myometrium can also be present in both conditions.[11] These overlapping imaging features often require additional clinical correlation or histopathological examination for a definitive diagnosis. In the reported case, the rudimentary horn was described as a subserous myoma on both ultrasound and MRI. The discovery of adenomyosis within the rudimentary horn, confirmed through histopathological examination, underscores the complexities in diagnosing Müllerian anomalies. This case highlights that imaging alone may not always provide a definitive diagnosis, emphasizing

the critical role of histopathology in reaching an accurate conclusion.

Preoperative considerations involve renal imaging to rule out any renal anomalies, assessing the degree of myometrial connection between the uterine horns to anticipate the need for potential suture repair of the myometrial defect after resection, evaluating the presence of uterine masses, checking for abnormalities in vaginal or cervical anatomy, considering the duration of symptoms, and reviewing any history of prior abdominal surgery. These factors help to prepare for the complexity of the procedure.[16] In the reported case, these preoperative considerations would have been invaluable for planning the surgical approach, enabling the surgical team to anticipate the procedure's complexities and optimize the chances of a successful outcome.

Excision of the uterine horn is the standard treatment for noncommunicating or obstructing uterine horns.[16] The primary reasons for this procedure include alleviating dysmenorrhea, preventing endometriosis caused by transtubal menstrual reflux, and avoiding the risk of pregnancy implantation in a functional rudimentary horn. These goals aim to alleviate symptoms, while conserving and optimizing fertility.[16]

The key principle for surgically resecting a uterine horn is to clearly define the Müllerian structures and their connections. To verify the noncommunicating side, chromotubation can be utilized by injecting methylene blue dye through the cervix. The dye will flow from the fallopian tube on the patent side, while no dye will appear on the obstructed side.[16] Hysteroscopic transillumination, or diaphanoscopy, can be employed to guide laparoscopic dissection by clearly identifying the dissection plane between the rudimentary horn and the uterus. In the index case, these techniques were utilized, enabling the surgical team to perform the procedure with greater precision.[17]

An evaluation of the relative position of the rudimentary horn in relation to the unicornuate uterus is essential. This assessment helps determine if the structures are separate, connected by a thin fibrous band, or fused. When a fibrous band is present, the primary blood supply to the rudimentary horn typically originates from the ipsilateral uterine artery and runs beneath the band, making it straightforward to coagulate or ligate.[4] In contrast, if the rudimentary horn is firmly attached to the unicornuate uterus, the blood supply generally runs laterally to the unicornuate uterus and below the uterine horn, complicating hemostasis. In such cases, the rudimentary horn often receives blood from both the ipsilateral uterine artery and the myometrial arcuate arteries of the contralateral uterine artery. These vessels will need to be coagulated before dissection.[4] In the index case, a thick fibrous band was identified, with the primary blood supply located beneath it, allowing for straightforward ligation.

The surgical procedure for resecting a rudimentary uterine horn is similar to a laparoscopic hysterectomy. Initially, the round and utero-ovarian ligaments, as well as the isthmic portion of the fallopian tube on the side of the functional, noncommunicating horn, are transected. The broad ligament is incised to access the retroperitoneal space and identify the ipsilateral ureter. The vascular pedicle of the uterine horn is then identified. Next, the uterovesical peritoneal fold is incised, and the bladder is reflected. The uterine vessels for the noncommunicating horn are coagulated and transected. The horn is then separated from the unicornuate uterus using various techniques, such as bipolar electrosurgery, endoscopic scissors, or the harmonic scalpel. The ipsilateral fallopian tube is also removed.[4] An ipsilateral salpingectomy is often performed during the resection of a rudimentary horn to prevent ectopic pregnancy, as the fallopian tube on the affected side is typically nonfunctional due to abnormal uterine anatomy. Removing the tube alongside the rudimentary horn eliminates this risk and avoids complications like recurrent pelvic pain or infection.[18]

Separating a densely fused horn from a rudimentary uterus is challenging. The goal is to preserve the functional hemiuterus, while excising all the tissues associated with the noncommunicating horn. Hysteroscopic transillumination can guide dissection in densely adherent cases. If the rudimentary horn is significantly fused to the unicornuate uterus at the cervical level, thermocoagulation can help prevent recurrences by coagulating any remaining functional cervical tissue.[4]

The specimen can be morcellated and removed from the abdomen, but if morcellation is unavailable, it can be removed intact through a minilaparotomy incision. Chromotubation can be used to check for myometrial defects at the resection site, which should be closed in layers if present.[4] In the index case, a 3-cm minilaparotomy incision was performed for specimen retrieval, as a morcellator was unavailable at the hospital at the time of surgery. Chromotubation revealed no dye passage through the myometrial defect, eliminating the need for suturing.

Noncommunicating rudimentary uterine horns are common findings associated with unicornuate uteri. When the horn has functional endometrium, patients typically present in the postmenarcheal period with pain symptoms secondary to the obstruction. The use of different imaging modalities to correctly diagnose the anomaly is crucial – as several other entities may be confused as a noncommunicating rudimentary horn.

Once diagnosed, surgical intervention is necessary to relieve pain and preserve fertility. Combining hysteroscopy and laparoscopy for diagnosis and treatment, using surgical techniques unique to this condition, can effectively manage patients with a noncommunicating rudimentary horn.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Authorship contributions

Dr. Mello Dee P. Monte - Involved in the conceptualization, writing of the original draft, review and editing.

Dr. Marie Janice Alcantara-Boquiren - Involved in the review, revision, commentary and editing of the draft.

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Conflicts of interest

There are no conflicts of interest.

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