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MANAGEMENT AND HISTOPATHOLOGICAL FINDINGS OF AN ATYPICAL PLACENTAL SITE NODULE IN A UTERINE NICHE POST-MULTIPLE CESAREAN SECTIONS: A CASE REPORT AND REVIEW OF LITERATURE

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Abstract
Uterine niche, a cesarean scar defect, is an iatrogenic complication following cesarean sections, often asymptomatic but associated with complications like abnormal uterine bleeding, pelvic pain, and reduced fertility. This case report describes a 33-year-old woman with a history of three cesarean sections presenting with chronic pelvic pain and intermenstrual bleeding for three years post her last cesarean. Diagnostic imaging, including transvaginal ultrasound and MRI, revealed a uterine niche with significant myometrial thinning. Hysteroscopy confirmed the defect, and laparotomy facilitated complete excision and repair of the niche. Histopathological examination identified an atypical placental site nodule (APSN), prompting beta-HCG surveillance due to potential malignancy risks. The patient remained asymptomatic during a six-month follow-up, with negative beta-HCG levels and normal ultrasound findings. This case highlights the importance of comprehensive imaging, surgical intervention, and histopathological evaluation in managing symptomatic uterine niches, particularly with atypical findings like APSN. It underscores the need for long-term follow-up to monitor healing and prevent obstetric complications in future pregnancies.
Keywords: Uterine niche, Cesarean scar defect, Atypical placental site nodule, Intermenstrual bleeding, Chronic pelvic pain, Hysteroscopy

INTRODUCTION

Sacculation, uterine diverticulum, isthmocele, or cesarean scar defect is a pouch-like iatrogenic defect that develops at the location of a prior cesarean section scar as a result of inadequate tissue recovery. In 2019, the European Niche Taskforce defined a niche as "an indentation of the uterine myometrium at the site of the CS scar with a depth of at least 2 mm," and then classified them into three types: basic, simple with a branch, and complicated forms. [1]. As many as 70% of women have uterine niches with previous cesarean sections, though most of the cases remain asymptomatic. However, the prevalence varies significantly based on the population characteristics and diagnostic method used. [2–4].

Risk factors for developing a uterine niche include surgical considerations like the cesarean incision's timing and procedure, and patient factors like maternal obesity, gestational diabetes, and pre-eclampsia, which may impede proper wound healing. [5–9]. Accurate diagnosis of a niche can be challenging due to these variables and the need for comprehensive imaging techniques. Saline-infused sonohysterography (SHG) is preferred over transvaginal sonography (TVS) due to its higher sensitivity in detecting niche depth and residual myometrial thickness (RMT), which are critical for planning surgical interventions. [4].

Decreased fertility, higher miscarriage rates, and a higher probability of obstetrical problems like scar ectopic pregnancy, placenta accreta, scar dehiscence, & rupture of the uterus in subsequent pregnancies are the major implications of uterine niche. [10–14]. These risks imply regular follow-up care and precise imaging techniques for high-risk women to monitor and manage these defects effectively. [15–17].

A lady with a history of repeated cesarean sections is described in this case report. She is 33 years old and presented with chronic pelvic pain and intermenstrual bleeding for 3 years, developing after her last cesarean section.

Case Presentation

Intermenstrual bleeding and persistent pelvic discomfort were symptoms that brought a 33-year-old lady with a history of three cesarean deliveries to the clinic. Her gravida score was 3, para 4.The onset of her complaints began three years ago following her last cesarean section. She described the pelvic pain as moderate, colicky, and only partially responsive to analgesics. For her intermenstrual bleeding, she received oral contraceptive pills (OCP) for one year;

however, she reported no improvement in her symptoms. Her medical history includes diabetes mellitus, which she has had for the past seven years, managed with metformin and insulin.

Clinical assessment

The patient had standard laboratory testing, including a full blood count, and a normal physical examination, blood glucose levels, and inflammatory markers, which did not reveal any significant findings. Ultrasound of the pelvis showed a hypoechoic area on the lowest part of the uterus. Magnetic resonance imaging (MRI) revealed a defect in the ventral wall of the uterus, measuring approximately 1.3 cm in width, with significant thinning of the overlying myometrium. This defect, consistent with a uterine niche, was filled with blood. The endometrium and cervix appeared normal. See Figure 1 for MRI findings.

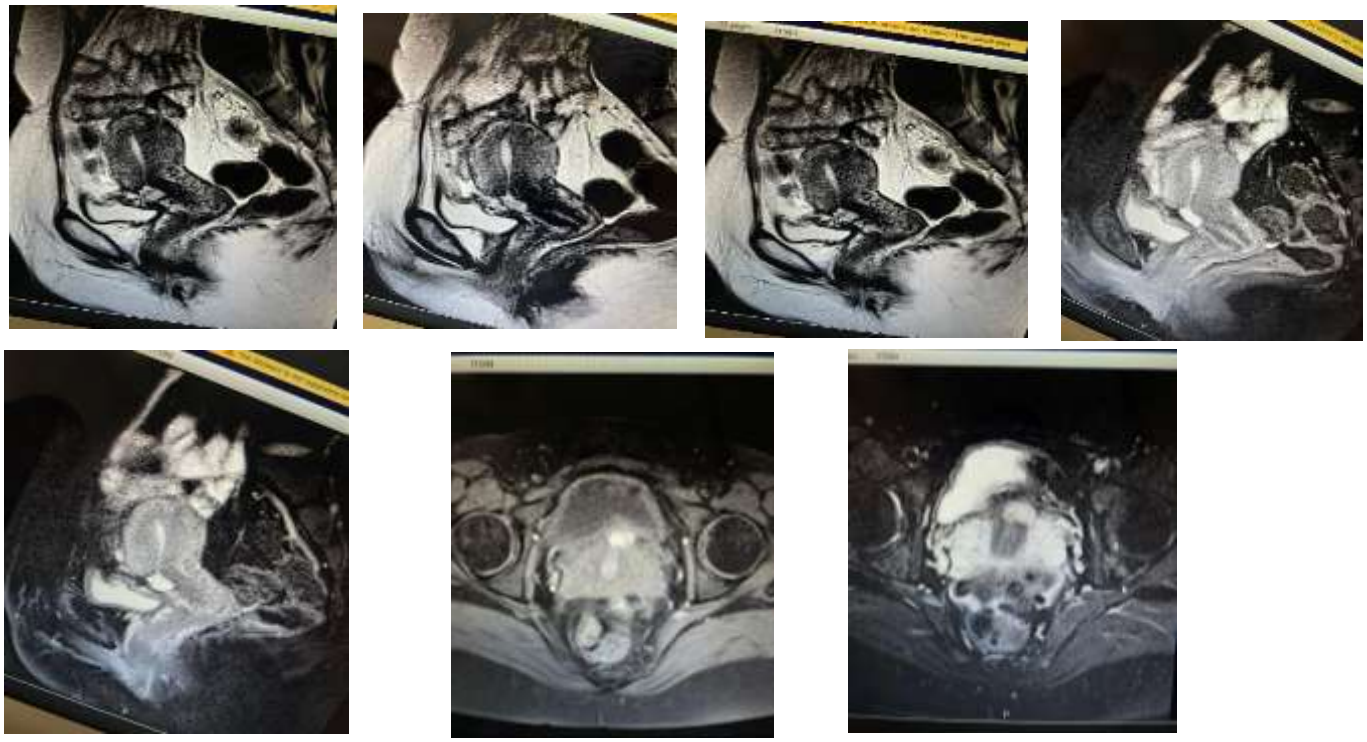


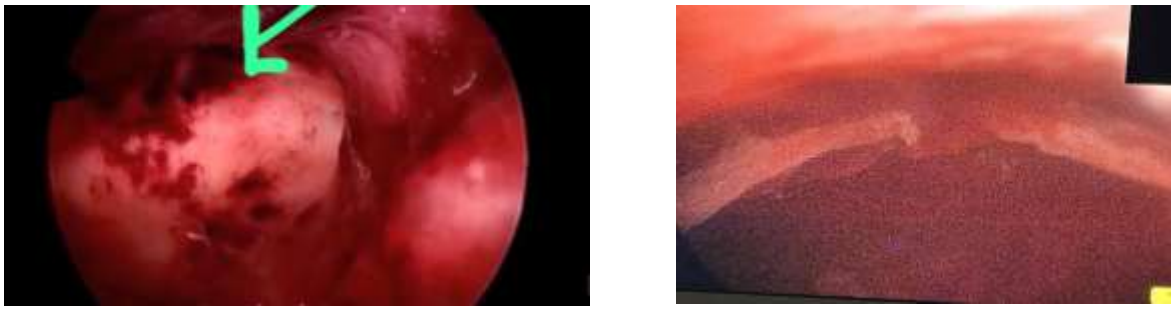
Figure1. MRI photos

Management

On August 18th, 2023, the patient underwent a diagnostic hysteroscopy, which confirmed a normal endometrial cavity, but did find a little hole in the uterine anterior wall. The defect was visible and accessible during the hysteroscopy. See Figure 2 for Hysteroscopy findings, with arrows indicating the abnormalities.

Subsequently, a laparotomy was performed. During the procedure, the bladder, which was adherent due to previous cesarean sections, was meticulously removed from the uterine anterior wall to reveal the isthmocele, which measured approximately 1 cm and communicated with the endometrial cavity. Complete excision of the niche was followed by refreshing the edges and repairing the site in two layers using continuous non-locked sutures with Vicryl 1-0. See Figure 3 for Intraoperative findings during the procedure. The patient's surgical recuperation was uneventful, and they were released from the hospital in good health.

Figure 2. Hysteroscopy findings



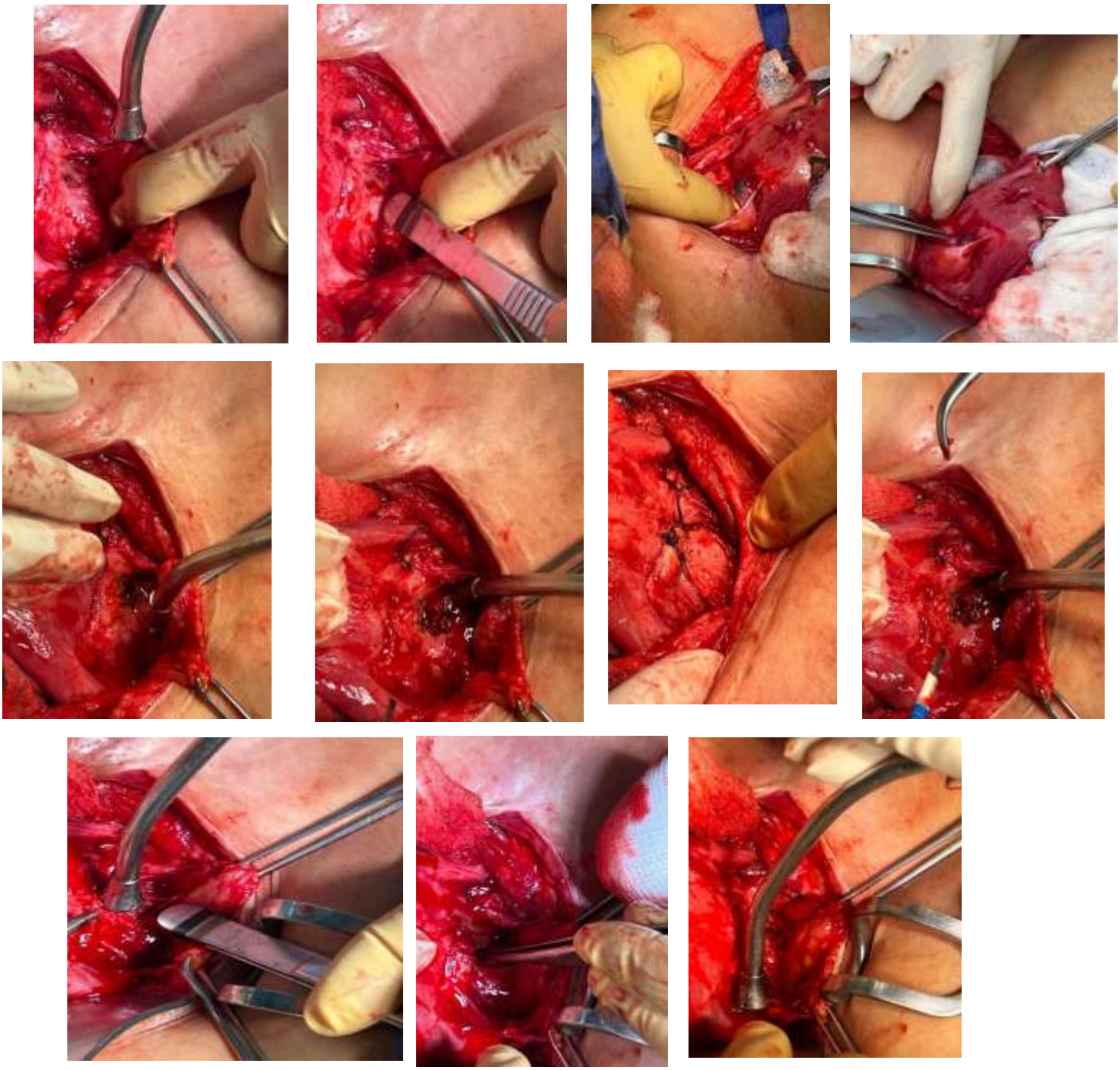


Figure 3. Intraoperative findings in the procedure

Histopathology

Histopathological examination identified an atypical placental site nodule (APSN). Given the potential association of these nodules with epithelioid trophoblastic tumors (ETT), surveillance of beta-HCG levels and close clinical follow-up were recommended.

Follow-up

The follow-up period was extended for six months postoperatively, during which the patient remained asymptomatic. Serum beta-HCG levels were negative, indicating no immediate evidence of malignancy. Additional follow-up measures included a pelvic ultrasound every three months to monitor the healing process. The patient was advised not

to conceive for at least two years to ensure better healing of the surgical site and was counseled about the potential risks of future pregnancies, including scar ectopic pregnancy and placenta accreta spectrum.

DISCUSSION

Case Summary

A 33-year-old lady who has had many caesarean sections is the subject of this case report, who presented with chronic pelvic pain and intermenstrual bleeding three years after her last cesarean section. The patient underwent successful surgical exploration and repair, followed by a histopathological examination. The diagnosis was confirmed as a uterine niche with an atypical placental site nodule. The case underscores the significance of recognizing and managing post-cesarean complications, the importance of histopathological examination for atypical findings, and the necessity of comprehensive follow-up to monitor malignancy and ensure optimal recovery. This report contributes to the limited literature on APSN in uterine niches and provides valuable insights for clinical practice.

Definition of uterine niche

Defective tissue recovery after a cesarean section (CS) scar may lead to an iatrogenic pouch-like abnormality called a uterine niche. A niche was defined in 2019 by the European Niche Taskforce as "an indentation of the uterine myometrium at the site of the CS scar with a depth of at least 2 mm." Simplicity, with a branch, and complexity were the three categories into which niches were categorized [18]. This disorder is also known as uterine isthmocele, diverticulum, uterine dehiscence, or caesarean scar deformity.

Prevalence

As many as 70 percent of women who have had a caesarean section in the past have uterine niche, however, just thirty percent of those women have any symptoms. Different diagnostic imaging methods, defining criteria, and populations provide different reported prevalence rates. A niche, which is characterized as any abnormality in the anterior part of the wall at the level of the cervical isthmus, was discovered in seventy-five percent of the women who sought hysteroscopic sterilization. [2].

There is a strong correlation between the diagnostic procedure and prevalence rates. According to saline sonohysterography (SHG), the incidence is between 56% and 84%, but transvaginal sonography (TVS) shows a prevalence between 24% and 70% [2]. On a randomly selected group of women who had had a caesarean section. Between thirteen percent to eighty-four percent of niches were identified by TVS in a randomly selected group of mothers who had had a caesarean section. Instances of SHG use ranged from 42% to 84%. Assuming a 2 mm depth for the niche, the prevalence ranged from 13–75% in the same group [3]. SHG has a better chance of detecting uterine niches. Using a depth of 2 mm as a criterion, Van der Voet et al. found that 64.5% of SHG samples had niches, whereas 49.6% of TVS samples had them. Because of this, many niches go unnoticed when TVS is used without gel or saline [4]. In women who have had one or more caesarean procedures, MRI may also detect uterine niches, which occur in 6% to 50% of cases [19].

Potential risk factors

Risk factors are divided into surgical factors and patient factors. One of the most important surgical factors is lower uterine incisions, because the cervical glands create mucus, which obstructs the myometrial approximation. Nabothian cysts and other big, mucus-filled niches form lower in the uterus as a consequence of this process. [5]. Furthermore, the timing of the incision is also a very important factor. It was observed that uterine niches increase in frequency when the cesarean incision is made at a cervical dilation of 5 cm or more [6] or after cervical effacement [7]. It is possible to interrupt myometrial development and create a niche if the deeper uterine muscle layer is not included. Therefore, endometrial preservation methods, suturing that is not perpendicular or tangential, and other similar procedures may induce niche growth [5]. The occurrence of defects, rupture, and dehiscence was determined to be the same when comparing single and double layer closures. Nonetheless, ultrasonography revealed a much smaller RMT in patients undergoing single-layer closure. [8].

In the present case, obstetrical parameters and perioperative data from the last cesarean section were not available to determine whether such risk factors apply. Nevertheless, the presence of multiple uterine scars likely contributed to these surgical factors.

Infection, inadequate hemostasis, devascularization, ischemia, and poor wound healing may all play a role in the disease [5]. This might lead to adhesion development between the CS site and the anterior abdominal wall. Lastly, there may be a correlation with patient variables that are related to wound healing. Uterine niche development was linked to preeclampsia, gestational diabetes, and maternal obesity because these conditions increased the likelihood of the CS incision not healing completely. [9].

Clinical Presentation

The vast majority of niche-using women have no symptoms whatsoever. When experiencing symptoms, the most common gynecological ailment is a prolonged menstrual cycle, followed by spotting, persistent pelvic discomfort, abnormal uterine bleeding (AUB), and dysmenorrhea [3,20,21]. This fits the bill for our patient, who began

experiencing persistent pelvic discomfort and heavy menstruation shortly after our previous cesarean surgery and persisted throughout the postpartum period.

Women who have a niche often have menstrual cramps that last anywhere from eleven to sixteen days [22]. Another common sign of post-caesarean section is bleeding that occurs after a woman's period ends. Intermenstrual spotting for at least two days or brownish discharge for at least two days after the end of menstruation, in the case of bleeding lasting more than seven days, constitutes this condition [23]. Random samples of women who have had a caesarean section with a niche had a higher incidence of postmenstrual spotting (20% to 60.8%), compared to 8.3% of women who did not have a niche [21]. Additionally, it may continue in specific cases even after therapy has ended [24]. The ratio of the depth of the niche to the thickness of the surrounding wall, as measured by TVS, is a good predictor of how symptoms would develop about the anatomical defect. [4].

Pain could also be an alarming sign for the niche. Women with a niche may present with dysmenorrhea (7.4–42.7%) or dyspareunia (18%), or other less specific pain manifestations such as chronic pelvic pain or suprapubic pain. [25]. The odds of chronic pelvic pain vary greatly and are higher in women with a niche, reaching up to 30%, compared to 5–7.4% in women without a niche. [3,26]. Abnormal myocontraction that empties the niche contents may be the cause of pain, which can cause physiological irregularities and continuous uterine efforts to expel trapped material [27].

Niches can also present with a complication. One of the symptoms and complications of a uterine niche is reduced fertility. Pregnancy and live birth rates are lower for women who have had a prior cesarean section compared to those who have had a vaginal delivery; this disparity becomes much more noticeable when a niche is present [10]. One possible cause is the buildup of peri-ovulatory fluid or persistent inflammation caused by leftover blood. This may impede sperm penetration, fertilization, and implantation. [11].

Reported findings about the link between abortions and niches are contradictory. Some research found that niche-carrying women had a greater risk of miscarriage than niche-free women [12], whereas other investigations found the opposite [10,13]. There is a slight association between scar ectopic pregnancy and future obstetric complications such as placenta accreta, scar dehiscence, uterine rupture, and scar ectopic pregnancy. [14].

Diagnosis

Diagnosing a uterine niche can be challenging, as demonstrated by the present case and literature. The main challenge in our case was the accurate identification and assessment of the niche on initial imaging, complicated by the presence of multiple uterine scars. This may lead to small defects being missed in imaging diagnosis. Confirmation required hysteroscopy, which revealed a small defect, and the diagnosis was ultimately established through laparotomy and histopathology. Additionally, the discovery of APSN underscores the need for vigilant histopathological examination to rule out associated malignancies.

In a non-pregnant condition, niche may be seen via tvs, sonography, magnetic resonance imaging (MRI), hysteroscopy, or 3-D ultrasound. With nothing in the way of training required, TVS and SHG are the most practical, accessible, and economical options [28]. Confirmation of the diagnosis is provided by the presence of an anechoic space, either with or without fluid, at a depth of at least 1 mm in the myometrium at the location of the cesarean scar, and by a depth of at least 2 mm in the same region. [3,29,30].

Vertically measuring the distance from the uterine serosa to the apex of the defect is known as residual myometrial thickness (RMT). SHG is preferred over TVS because it typically shows a larger niche depth and thinner RMT, without the use of gel or saline implantation, which might lead to an underestimation [4]. This has important practical implications since a sufficiently thick residual myometrium is required for hysteroscopic niche resection (HNR), which is a method for treating bleeding that originates in the niche. Thus, the benefits of SHG for niche detection surpass the drawbacks, such as the prolonged examination time and relative patient discomfort. [29].

Accordingly, when contrasted with TVS, SHG provides a more accurate diagnosis of niches, revealing a greater frequency, improved identification of bigger niches, and thinner remaining myometrium. Because of this, TVS outperforms SHG in terms of sensitivity and specificity [2-4]. Typically, TVS misses minor but potentially clinically relevant niches. Without intrauterine fluid or when just the sagittal plane is used for examination, they could go unnoticed [30]. Due to the horizontal nature of the uterine incision, it is essential to use the transversal plane in addition to the mid-sagittal plane in order to avoid missing niches that are placed laterally. However, 3D TVS can rebuild and show selected areas within the volume dataset post hoc, making it a replicable tool for niche assessment. Nevertheless, it requires specialized education, more time spent on examinations, and more costly 3D ultrasound equipment [31].

While evaluating the niche using imaging, it is important to look for other anatomical abnormalities such concavity, aberrant vascularity, visible serosa, and formations that resemble cysts or polyps [32,33].

No universally accepted definition of a huge literary niche has been proposed as of yet. On the other hand, the majority of research have used alternative criteria to categorize a big niche, such as a dehiscence risk coefficient (the ratio of RMT to absolute myometrial thickness, or AMT) below 0.25, a depth/AMT ratio over 0.50, or an RMT/AMT ratio below 0.50. According to some sources, a big niche is defined as one with a depth of at least 0.50 cm, a range of red blood cell transfusion (RMT) values between 0.21 cm and 0.56, or no remaining myometrium at all (a "total defect").

Management

There are no set standards for treating niches at the moment. Secondary infertility, recurrent miscarriage, AUB, postmenstrual spotting, prior scar ectopic pregnancies, and other symptomatic conditions may be managed clinically with medication treatment and surgical procedures. It is not advisable to routinely correct niches that are discovered by accident unless there are intentions to have children in the future. The symptoms of AUB may be alleviated with medical treatment, including hormone therapy. Although the levonorgestrel IUS does not shorten menstrual cycles, oral contraceptives are useful in cases when pregnancy prevention is not a priority. The inability to repair uterine architecture, high recurrence rates upon medication withdrawal, and substantial adverse effects from prolonged usage are some of the limits of medical treatment [37].

Transvaginal, laparoscopic, and transabdominal repair procedures are all part of the surgical repair toolbox. The use of a natural opening makes transvaginal repair the least intrusive procedure; there is no scarring of the abdomen following surgery. In addition, transvaginal repair allows for full-thickness closure, direct access to the niche cavity, and total excision of tissues around the niche, which improves quality of life and symptoms connected to the niche [38]. There is a significant decrease in spotting after surgery, whether the procedure is performed transvaginally or hysteroscopically. Further benefits of the vaginal approach include access to inflammatory and fibrous tissues around the niche cavity, the ability to adjust the uterine flexion posture, and suturing for lower uterine segment structural restoration. Hysteroscopic resection, on the other hand, requires less time in the operating room and less time in the hospital, and there are less problems and lower expenses associated with the procedure [37]. Success rates range from 92% to 100% after hysteroscopic resection, and from 100% following laparoscopic and vaginal repair, all with minor complications, according to a meta-analysis [4].

To minimize the danger of bladder damage and ensure a successful niche resection, it is advisable to leave 2.5-4 mm of remaining myometrium. This will help prevent complications such as bladder perforation. In addition, cervical incompetence is more likely to occur after proximal resection [39].

Prevention of niche formation

The primary goal of uterine niche prevention is to lower the rate of cesarean sections. The use of proper surgical procedures to ensure thicker residual myometrium and robust scar will, among other secondary prevention strategies, continues to be the cornerstone in niche prevention [17]. Uterine closure utilizing the far-far-near-near double-layer unlocking approach may help reduce isthmocele development and provide adequate residual myometrium, according to a recent randomized controlled trial including 138 women [40].

Due to the hypothetically higher risk of uterine rupture, further study suggests that planned cesarean sections should not be delayed beyond 38 weeks of gestation [41]. On the other hand, after niche management, there is no consensus on the best time and method of distribution [1].

Patient Education

Along with other potential risks, informing patients about the need for a cesarean section should include the long-term prospect of establishing a niche. Important uterine niche problems that patients should be aware of include amniotic tube rupture (AUB), pelvic discomfort, and secondary infertility. Further patient counseling includes discussing the other immediate benefits of a cesarean section alongside long-term risks [15]. It is essential to emphasize the importance of regular follow-up care to monitor for niche development, particularly in symptomatic women. Therefore, educating patients for the symptoms and importance of consulting the physician is crucial, in addition to the eventual treatment options. Additionally, patients should be informed about using TVS with saline or gel to ensure precise niche assessment, guiding subsequent management decisions [15–17].

CONCLUSION

Diagnosing post-caesarean uterine niche demands a high index of suspicion since it is a relatively novel clinical phenomenon. Although subfertility may need surgical repair, hormonal therapy is the preferred method for hemorrhage symptoms. Smaller niches with an RMT more than 3 mm are best removed by hysteroscopic methods; niches lower in the body may be treated transvaginally; major defects or women hoping to conceive in the future are best removed by a transabdominal technique.

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