

CASE STUDY

Large anechogenic “Holes” in the uterus: The utility of contextual combination of ultrasound and hysteroscopy

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Cystic adenomyosis is a rare occurrence, especially in young patients. We report a challenging case of a 44-year-old African patient, in which the ultrasound described a large bilobate anechogenic cyst in the myometrium. The combination of ultrasound, contextual hysteroscopy, and subsequent magnetic resonance imaging helped to clarify the case, reducing the risk of complications for the patient, potentially due to misdiagnosis.

Keywords: hysteroscopy, cystic adenomyosis, ultrasound, sonohysteroscopy

Introduction

Uterine adenomyosis is characterized by the presence of heterotopic endometrial glands and stroma at about 2.5 mm in depth in the myometrium, or more than one microscopic field at 10 times magnification from the endometrium–myometrium junction, and a variable degree of adjacent myometrial hyperplasia (1).

This condition is related to dysmenorrhea and pelvic pain that can worsen the quality of life of patients.

A correct diagnosis was important to define the appropriate therapy. However, even if some peculiar sonographic characteristics are reported, the diagnosis is not always guaranteed. The combination of ultrasound and magnetic resonance imaging (MRI) could be useful to better define this condition (2).

In the case of cystic adenomyosis, a rare type of adenomyosis, diagnosis and management are extremely difficult (3).

Cystic adenomyosis is usually described in young patients, in whom the need to reduce symptoms is linked to the need for fertility sparing (4–8). However, elderly patients may also be suffering from cystic adenomyosis.

In these patients, even if there is no need for fertility sparing, difficulties may be related to other potential risks, which we discussed in our case report. In particular, the possible risk of uterine perforation following invasive procedures or the risk of adverse obstetrical outcomes, such as rupture of the uterus, in the case of pregnancy are remarkable.

Case presentation

A 44-year-old African woman had three vaginal deliveries, one voluntary termination of pregnancy, and one abortion a year earlier in Senegal. After this spontaneous miscarriage, which did not require hysterosuction, the patient chose a long-acting contraceptive and was administered quarterly intramuscular progesterone for three times consecutively. During this period, she was not subjected to any gynecological checks because she was in good health without any gynecological problems.

Just before the fourth progesterone-depo administration, the patient showed up in the emergency room complaining of irregular vaginal bleeding lasting two months. The beta-HCG was negative. The gynecological examination was regular.



FIGURE 1 | Transvaginal view of the uterus in subsequent sections, which showed the bilobate anechogenic myometrial cyst (marked with an asterisk).

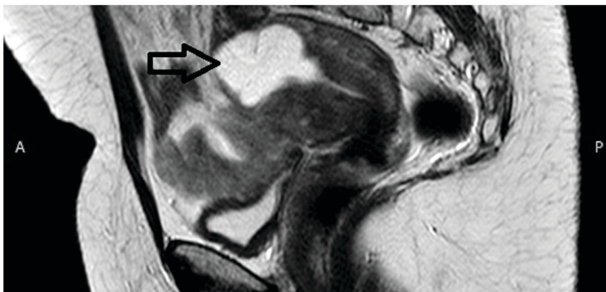


FIGURE 2 | Magnetic resonance image, which confirmed the diagnosis of cystic adenomyosis (marked with an arrow).

On ultrasound examination, she presented a non-univocal image, described as a big anechogenic and non-vascularized formation with subtle contours within the uterus (**Figure 1**). Adnexa was regular. Accordingly, she was referred for hysteroscopy.

At hysteroscopy, the cavity was larger than normal but without irregularities. The uterine fundus was distorted by a kind of “diverticulum.” Due to the difficulties in defining the picture, a second gynecologist performed a concomitant trans-abdominal ultrasound to guide hysteroscopy.

At the uterine fundus, a bilobate anechogenic formation was detected, and the suspicion of cystic adenomyosis has been raised. A biopsy of the endometrium was performed under ultrasound guidance. Histological examination described a secretive endometrium.

Since the effect of slow-release progesterone was almost over, the patient required sterilization. While waiting for the laparoscopic surgery, an MRI was prescribed. The MRI confirmed the presence of a cystic formation of the myometrium measuring $35 \times 26 \times 47$ mm, which reduced the muscular wall of the uterine fundus (**Figure 2**).

Laparoscopy, performed for tubal sterilization, confirmed the presence of an enlarged uterus of cystic consistency (**Figure 3**). The hypothesis to perform a hysterectomy was dismissed, with the patient preferring a less invasive surgery with maintenance of body image, also considering that the patient was young, sexually active, and asymptomatic.

The post-surgery course was regular. The patient was also scheduled for a follow-up to assess any

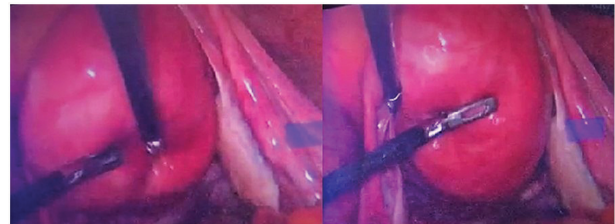


FIGURE 3 | Laparoscopic surgical view, which confirmed the cystic consistency of the uterine neoformation at instrument touch.

ultrasonographic changes that may have occurred following the discontinuation of progesterone therapy.

Conclusion

Although cystic adenomyosis is more common in young patients, it can be difficult to diagnose and manage in the elderly (4–8).

We presented our case, which demonstrated a non-univocal ultrasound image but with the potential for serious harm in the event of a misdiagnosis.

We hypothesized that the ultrasound picture could be correlated with a decidualization of the cystic adenomyoma due to the prolonged progesterone therapy, although we cannot confirm this hypothesis. However, we would like to underline that, in cases of hormonal therapy, the imaging could present atypical changes.

Moreover, in our case, only the combination of contextual hysteroscopy under ultrasound guidance could determine whether the adenomyosis was in continuity with the cavity. This combination of contextual procedures is not yet reported in literature with this intent. However, in our case, it allowed a better definition and reduced the risk of uterine perforation.

A correct definition of uterine structure is actually necessary in cases of large myometrial cysts, particularly in cases of invasive procedures such as hysterosuction, hysteroscopy, or even a simple endometrial biopsy.

We also used ultrasound and MRI to confirm the diagnosis afterward, as already suggested (2, 3).

In consideration of the patient’s request, we performed tubal sterilization; we chose not to associate hysterectomy or

cyst removal. We really didn't get histological confirmation of our imaging diagnosis. Laparoscopic sterilization, on the other hand, allowed for a less invasive surgery while maintaining body image and post-poning the risk of uterine rupture due to a thin uterine wall in the case of pregnancy.

In conclusion, the combination of ultrasound and contextual hysteroscopy helped to avoid potential complications related to invasive procedures or accidental pregnancies in this challenging case of cystic adenomyosis with a large bilobate myometrial cyst.

Author contributions

Material preparation and data collection were performed by MS, TT, SG, and AV. The first draft of the manuscript was written by PA, and all authors commented on previous versions of the manuscript. NP and MS revised the language and edited the manuscript. All authors contributed to the study and read and approved the final manuscript.

Conflict of interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships

that could be construed as a potential conflict of interest.

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