Misdiagnosis of Accessory Cavitated Uterine Malformation: 2 Cases and Literature Review

Wei Bian[1], Jun Yuan[2], Zhi-Feng Wang[1], Li Ling[3], Jinjuan Feng[4], Hui Liu[1]*

[1]*Department of Radiology, Jiaxing Maternity and Child Health Care Hospital, College of Medicine, Jiaxing University, China*
[2]*Department of Pathology, Jiaxing Maternity and Child Health Care Hospital, College of Medicine, Jiaxing University, China*
[3]*Department of Ultrasound, Jiaxing Maternity and Child Health Care Hospital, College of Medicine, Jiaxing University, China*
[4]*Department of Radiology, Ningbo City Yinzhou District Third Hospital, China*

***Correspondence:** Hui Liu, Department of Radiology, Jiaxing Maternity and Child Health Care Hospital, College of Medicine, Jiaxing University, Jiaxing, China, Tel: 18668339003,

316770590@qq.com

Radiology Case. 2025 May; 19(5):1-8 :: DOI: 10.3941/jrcr.5572

HUMAN AND ANIMAL RIGHTS
All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000 (5). Informed consents were obtained from the patients for publication of this case report.The study was approved by the Ethical Committee of Jiaxing Maternity and Child Health Care Hospital under number 2024-Y-63.

Funding: This research received no external funding.

Data Availability Statement: The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

CONFLICTS OF INTEREST
The authors declare no conflicts of interest.
Statement: The material contained in this manuscript has not been previously published and is not being concurrently submitted elsewhere.

ABSTRACT

Infectious aneurysm related to endocarditis is a complication that usually involves the distal middle cerebral artery branches. There is paucity in the literature of cases involving the common or external carotid arteries, which are extremely rare. We report the case of a previously healthy 30 years old female with bacterial endocarditis-related infected skin lesions, complicated with infectious aneurysm involving a distal segment of the frontal branch of the right superficial temporal artery, intraparenchymal hematoma and septic embolisms. To our knowledge, this is the first case reported of an infectious aneurysm involving a distal external carotid artery branch in the context of proven bacterial endocarditis.

CASE REPORT

BACKGROUND

Accessory cavitated uterine malformation (ACUM) is a rare and frequently misdiagnosed Müllerian duct anomaly due to its nonspecific clinical manifestations and low clinical awareness. This study presents two misdiagnosed ACUM cases, highlighting the crucial diagnostic role of MRI through characteristic imaging features such as the "Oreo cookie sign." These cases contribute significantly by enhancing diagnostic awareness, validating imaging biomarkers, guiding clinical management, and addressing knowledge gaps in ACUM research.

The findings reinforce laparoscopic excision as the gold standard treatment while emphasizing the importance of preoperative imaging evaluation. Although ACUM has been included in the 2022 Chinese Expert Consensus, related research remains insufficient. By integrating its pathogenesis, imaging characteristics and pathological features, this study promotes interdisciplinary collaboration among radiology, pathology and gynecology departments, advancing precise diagnosis and treatment for young patients with refractory dysmenorrhea, demonstrating substantial clinical value.

INTRODUCTION

Accessory cavitated uterine malformation (ACUM) is a rarely encountered, unclassified Müllerian duct-derived anomaly, representing an obstructive reproductive tract malformation [1]. The clinical manifestations often include progressive dysmenorrhea or pelvic and abdominal pain. Due to the rarity of this disease and the limited knowledge among clinicians and radiologists, ACUM is frequently misdiagnosed,

thereby impacting patient management.

Here, we present two cases of ACUM patients who were misdiagnosed at the Jiaxing Maternal and Child Health Hospital between June 2021 and May 2024. We describe the pathogenesis, clinical features, imaging characteristics, treatment, and prognosis of this condition, with the aim of improving the understanding of ACUM among radiologists, pathologists, and gynecologists.

CASE PRESENTATION

Case 1

A 30-year-old female patient presented with a uterine mass that had been detected 4 years prior. The clinical manifestation was lower abdominal pain during the first 1-2 days of menstruation, and the gynecological examination was unremarkable. Transvaginal ultrasound revealed a hypoechoic lesion measuring approximately 39 mm × 37 mm × 36 mm in the left uterine wall, with a 13 mm × 8 mm anechoic component containing thick fluid and a linear vascular signal (Figure 1A). Both ovaries appeared normal. The preliminary diagnosis was Uterine leiomyoma with degeneration in the left uterine wall.

Magnetic resonance imaging (MRI) showed a round, mass-like lesion in the left uterine wall, measuring around 40 mm in diameter. The lesion exhibited heterogeneous signal intensity, with the majority of the lesion showing signal similar to the myometrium, and a high-signal-intensity focus in the center on both T1-weighted and T2-weighted images. The lesion was not communicating with the uterine cavity (Figure 1B-1F). The imaging diagnosis was a probable adenomyoma or degenerated leiomyoma in the left uterine wall, with a remnant horn uterus to be excluded.

Intraoperatively, laparoscopic examination revealed a prominent, thickened lesion in the left posterior uterine wall near the left uterine cornu, with an ill-defined border. Under the guidance of ultrasonography, the lesion was completely excised, and brown fluid was observed upon incision. Both uterine cornua, fallopian tubes, and ovaries were normal. The initial pathological diagnosis was a left uterine wall adenomyoma.

However, upon further review by a senior pathologist, the lesion was found to consist of endometrial glands and stroma surrounded by circularly distributed uterine smooth muscle tissue, forming an organoid structure, which was distinct from the typical distribution seen in adenomyosis, and was consistent with the pathological characteristics of ACUM (Figure 1G-1I).

At the 3-month follow-up after discharge, the patient had regular menstrual cycles without dysmenorrhea.

CASE 2

A 20-year-old female patient presented with uterine polyp and adenomyosis in the right uterine wall identified during routine ultrasound screening. Transvaginal sonography showed an 8mm×6mm×4mm hyperechoic lesion within the uterine cavity. The right uterine wall was slightly bulging, with a 37mm×36mm heterogeneous hypoechoic area (adjacent to the serosa and endometrium), with indistinct borders, and a 20mm×14mm hypoechoic area within, with linear flow signals around the periphery. Transvaginal ultrasound diagnosis was:1) Endometrial polyp; 2) Focal adenomyosis of the right uterine wall (Figure 2A). MRI examination revealed a well-circumscribed, round-shaped lesion in the right uterine wall, measuring approximately 38 mm × 35 mm × 39 mm, with isointense signal on T1WI and hypointense signal on T2WI. A round-shaped lesion of approximately 14 mm in diameter with heterogeneous hyperintense signal on both T1WI and T2WI was observed in the center of the lesion, which was clearly demarcated from the uterine cavity. The endometrium was thickened to about 10 mm, with a linear, hypointense T2 signal lesion of approximately 5 mm (Figure 2A-2F). The morphology and signal of both uterine horns and ovaries were unremarkable. The radiological diagnosis was: 1) adenomyoma in the right uterine wall; 2) possible endometrial polyp.

Hysteroscopy revealed a 12 mm × 10 mm × 8 mm, pale red, polypoid lesion near the internal cervical os on the left uterine wall. The endometrium was normal, and the bilateral tubal ostia were visible. The polypoid lesion was gradually resected using an electrosurgical loop, and the specimen was confirmed as an endometrial polyp on pathological examination. The lesion in the right uterine wall was not intervened.

After comprehensive review of the MRI images by multiple radiologists and literature review, a consensus diagnosis of ACUM in the right uterine wall was established. The patient was followed up for 1 year, with no reported discomfort or abnormal symptoms.

DISCUSSION

Pathogenesis of ACUM

ACUM is a congenital developmental anomaly, and its exact pathogenesis and incidence remain unclear. Past studies have referred to this lesion using various terms, such as "Accessory uterine cavity", "Cystic myometrial lesion", "Isolated cystic adenomyoma", and "Juvenile cystic adenomyoma"[2-5]. In 2010, the renowned expert in female genital tract malformations, Acién, proposed a hypothesis regarding ACUM based on similar previous cases[6]. In 2012, he further summarized the clinical characteristics of these patients and first coined the term "accessory cavitated uterine mass" (ACUM) [7]. It is currently believed that ACUM is caused by the persistence and duplication of Müllerian duct tissue in the attachment area of the round ligament, which may be related to a dysfunction in the peritoneal traction mechanism during round ligament development [6-8]. Due to the lack of comprehensive understanding of ACUM in the past, the accurate clinical diagnosis rate was relatively low, which affected patient management. In recent years, ACUM has gained widespread recognition, and the "Chinese

Expert Consensus on Nomenclature and Definition of Female Genital Tract Malformations (2022 Edition)" has specifically included ACUM as a distinct entity [9]. However, the literature on this condition remains limited, and further interdisciplinary collaboration is needed to improve the understanding of this disease.

Clinical manifestations of ACUM

ACUM typically lacks specific clinical presentations and often occurs in young female patients. The most common symptom is progressive and severe dysmenorrhea, followed by pelvic pain or abdominal cramping, with some patients being asymptomatic [10]. The pain may be attributed to the accumulation of menstrual blood within the accessory cavity, leading to increased intracavitary pressure and compression of the sensory nerve fibers. The pain can occur throughout the entire menstrual cycle and may last for more than 20 days, often unresponsive to conventional non-steroidal anti-inflammatory medications [11]. The menstrual cycle, duration, and volume are typically normal in these patients.

Diagnosis and Differential Diagnosis of ACUM

Diagnostic Criteria: In 2010, Acién proposed the diagnostic criteria for ACUM [6]: 1) an isolated accessory cavitary mass located below the round ligament, not communicating with the fallopian tube; 2) normal development of the uterus, fallopian tubes, and ovaries (with occasional exceptions [7]); 3) the cavity lined with functional endometrium, including glands and stroma, and containing chocolate-like fluid contents; 4) no adenomyosis or adenomyotic cysts in the rest of the uterus, but small foci of adenomyosis may be present in the myometrium adjacent to the cavity.

Histopathological Characteristics of ACUM: Histologically, ACUM is characterized by: 1) the cavity being lined with functional endometrium with evidence of bleeding; 2) an outer layer of well-organized smooth muscle tissue [10]. Immunohistochemical staining shows positive expression of CD10, estrogen receptor (ER), and progesterone receptor (PR) in the normal endometrium lining the cavity. The surrounding smooth muscle tissue also stains positive for desmin, ER, and PR-4 [12]. Due to the increased intracavitary pressure caused by accumulated menstrual blood, focal adenomyosis-like changes may be observed in the myometrium adjacent to ACUM, while the rest of the uterus is free of adenomyosis. The histopathological features of the case 2 reported in this paper are consistent with the literature.

Ultrasonographic Features of ACUM: Pelvic or transvaginal ultrasound is a simple and effective initial imaging modality for evaluating the uterus and adnexa, with transvaginal ultrasound providing better visualization of the lesion. Typically, ACUM appears as an isolated cystic lesion within the myometrium of the anterior-lateral uterine wall, below the attachment of the round ligament. The cystic area is usually round, with a ground-glass echogenicity and a smooth lining. The cystic lesion is surrounded by a thick, hypoechoic rim of myometrium, and the Doppler flow within the lesion is not significantly different from the surrounding myometrium [13,14]. Additionally, three-dimensional (3D) ultrasound in the coronal plane can provide clearer visualization of the specific anatomical location and characteristic imaging features of ACUM, further aiding in the diagnosis [14,15].

The Advantages of MRI in Diagnosing ACUM

While transvaginal ultrasound is typically the initial imaging modality for ACUM, many patients are relatively young (possibly virgins) and unable to undergo transvaginal examination. Pelvic MRI avoids invasive procedures and can provide important details, allowing the identification of the cystic structure within the myometrium as ACUM. On MRI, ACUM appears as an isolated, round, functional cavity within the myometrium of the anterior-lateral uterine wall, below the attachment of the round ligament, with a non-communicating appearance (i.e., not connected to the uterine cavity or fallopian tubes), while the rest of the uterine structure is normal. The MRI findings of the cavity correlate well with the surgical and pathological results [8,11]. In 95.5% of ACUM cases, the cavity contains blood, which appears hyperintense on both T2-weighted and T1-weighted imaging16. The cavity is surrounded by a circular low signal intensity on T2-weighted imaging, corresponding to the smooth muscle tissue. Although the second case in this report was not confirmed by pathology, the MRI findings and hysteroscopic visualization allowed for a diagnosis of ACUM in the right uterine wall. The heterogeneous hyperintensity within the cavity in this case may be related to different stages of bleeding.Our analysis revealed that the low signal intensity of the smooth muscle tissue surrounding the cavity on T2-weighted imaging (T2WI) was equal to or lower than the normal myometrial signal, and similar to the signal characteristics of the junctional zone. Integrating the case analysis and literature review, we found that the planar T2WI appearance of the ACUM lesion resembled an "open Oreo cookie". The central high signal intensity component corresponds to the "cream filling" of the Oreo cookie, while the surrounding low signal intensity rim represents the "cookie" portion. This distinct MRI signal pattern is a helpful diagnostic feature of the ACUM lesion (Figure 3). Furthermore, we believe that if reconstructed, the T2-weighted three-dimensional (3D) images may demonstrate a morphological appearance akin to a round cream-filled chocolate candy, reminiscent of the "Malteser" brand. This unique imaging feature can serve as a useful mnemonic device to help radiologists recognize and remember the distinctive imaging features of the ACUM lesion.

MRI is a reliable tool for the accurate diagnosis of ACUM. More importantly, it can distinguish ACUM from other causes of chronic pelvic pain, such as adenomyosis and endometriosis, as well as other imaging differentials like adenomyoma, degenerated leiomyoma, and rudimentary uterine horn [16]. Uterine leiomyomas and adenomyosis/adenomyoma typically

occur in women aged 30-50 and can be found in any part of the uterus. Adenomyosis/adenomyoma is characterized by the invasion of endometrial glands and stroma into the myometrium, leading to hypertrophy of the surrounding myometrium. On MRI, it appears as ill-defined areas of low signal intensity on T2-weighted imaging with scattered foci of high signal intensity on T2-weighted and/or T1-weighted imaging. Degeneration of uterine leiomyomas often follows a certain process, from the initial detection of the leiomyoma to the development of degeneration, which can cause rapid growth of the leiomyoma and lead to symptoms such as heavy or prolonged menstrual bleeding or irregular vaginal bleeding. Rudimentary uterine horn is often associated with unicornuate uterus, and on the underdeveloped side, soft tissue signal similar to the myometrium can be seen, with or without a uterine cavity and endometrial signal, and if the cavity is present, it may be connected to the cavity of the unicornuate uterus or just attached by a fibrous band.

The T2-weighted MRI image demonstrates the characteristic appearance of the ACUM lesion, which is reminiscent of an "open Oreo cookie". The central high signal intensity component corresponds to the "cream filling" of the Oreo cookie, while the surrounding low signal intensity rim represents the "cookie" portion. This distinct MRI signal pattern is a helpful diagnostic feature of the ACUM lesion.

MRI helps with timely diagnosis, allowing patients to receive prompt treatment to alleviate their pain symptoms, and can guide appropriate, fertility-sparing laparoscopic excision [11].

Treatment of ACUM

Laparoscopic excision of ACUM is the most common and effective treatment approach, as the lesion is located in the superficial myometrium near the attachment of the round ligament, making the surgical procedure relatively straightforward [1,17]. Laparoscopic removal of ACUM can achieve complete symptom resolution with a low risk of recurrence. However, for small ACUM lesions located deep within the myometrium, the laparoscopic approach can be more challenging, as it may be difficult to intraoperatively locate the cystic cavity. Therefore, preoperative assessment of the position and depth of the ACUM relative to the myometrium is crucial for successful surgical management. In such cases, intraoperative ultrasound guidance can be very helpful in guiding the complete excision of the lesion. Accurate surgical management can minimize the risk of uterine scarring and future uterine rupture during pregnancy.

In recent years, ultrasound-guided sclerotherapy using ethanol and ultrasound-guided drainage have also been used to treat ACUM, as these techniques can help relieve the intracavitary pressure and alleviate the patient's pain symptoms [14], 16. Previous studies have reported that medical treatments, such as oral contraceptives or gonadotropin-releasing hormone agonists (GnRH-a), can provide some pain relief, but they do not offer a definitive cure, and surgical excision is ultimately required for symptom resolution [4,5,11].

CONCLUSIONS

The etiology of adenomyotic cystic uterine mass (ACUM) remains incompletely understood, but it is likely associated with Müllerian duct anomalies and myometrial developmental defects. The primary clinical manifestations include progressively worsening dysmenorrhea, persistent pelvic pain, or colicky abdominal pain. Imaging examinations, particularly pelvic magnetic resonance imaging (MRI), can aid in the diagnosis, but the definitive diagnosis relies on surgical pathological examination. Early and accurate diagnosis of ACUM is crucial for timely treatment. Surgical excision is currently the widely accepted first-line treatment approach. Laparoscopic removal of the lesion can achieve complete symptom relief and has a low risk of recurrence, though the procedure may be more challenging for lesions located deep within the myometrium. In such cases, preoperative assessment of the position and depth of the ACUM, as well as the use of intraoperative ultrasound guidance, can facilitate successful surgical management and minimize the risks of uterine scarring and future uterine rupture during pregnancy.

TEACHING POINT

Accessory Cavitated Uterine Malformation (AUCM) is a rare condition that is often misdiagnosed due to lack of awareness among radiologists, pathologists, and gynecologists.

Understanding the pathogenesis, clinical presentation, and imaging features of AUCM is crucial for making an accurate and timely diagnosis.

Specific MRI T2WI findings, such as the "Oreo" and "Marly" signs, can help radiologists identify AUCM and guide clinicians in appropriate treatment planning.

Early recognition of AUCM is important, as it can assist clinicians in formulating the most suitable treatment strategies for the patient.

Increased awareness and education about AUCM among healthcare professionals involved in its management can help reduce the risk of misdiagnosis and improve patient outcomes.

QUESTIONS

Which of the following answer choices is false?

1. ACUM is a congenital developmental anomaly.
2. ACUM often occurs in young female patients.
3. The definitive diagnosis relies on pelvic magnetic resonance imagings. (applies)
4. The "Oreo" and "Marly" signs in MRI findings, can help radiologists identify AUCM
5. Surgical excision is treatment approach.

Explanation

1. ACUM is a congenital developmental anomaly. [Accessory cavitated uterine malformation (ACUM) is a rarely encountered, unclassified Müllerian duct-derived anomaly, representing an obstructive reproductive tract malformation]

2. ACUM often occurs in young female patients. [ACUM typically lacks specific clinical presentations and often occurs in young female patients. The most common symptom is progressive and severe dysmenorrhea, followed by pelvic pain or abdominal cramping, with some patients being asymptomatic.]

3. The definitive diagnosis relies on pathological examination. [Imaging examinations, particularly pelvic magnetic resonance imaging (MRI), can aid in the diagnosis, but the definitive diagnosis relies on surgical pathological examination.]

4. The images of AUCM have distinct characteristic features. Specifically, the "Oreo" and "Marly" signs on MRI T2-weighted imaging can help radiologists identify AUCM. [The planar T2WI appearance of the ACUM lesion resembled an "open Oreo cookie". The central high signal intensity component corresponds to the "cream filling" of the Oreo cookie, while the surrounding low signal intensity rim represents the "cookie" portion. This distinct MRI signal pattern is a helpful diagnostic feature of the ACUM lesion（Figure 3）. Furthermore, we believe that if reconstructed, the T2-weighted three-dimensional (3D) images may demonstrate a morphological appearance akin to a round cream-filled chocolate candy, reminiscent of the "Malteser" brand.]

5. AUCM is ususlly removed by Surgical excision. [Surgical excision is currently the widely accepted first-line treatment approach. Laparoscopic removal of the lesion can achieve complete symptom relief and has a low risk of recurrence, though the procedure may be more challenging for lesions located deep within the myometrium.]

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FIGURES

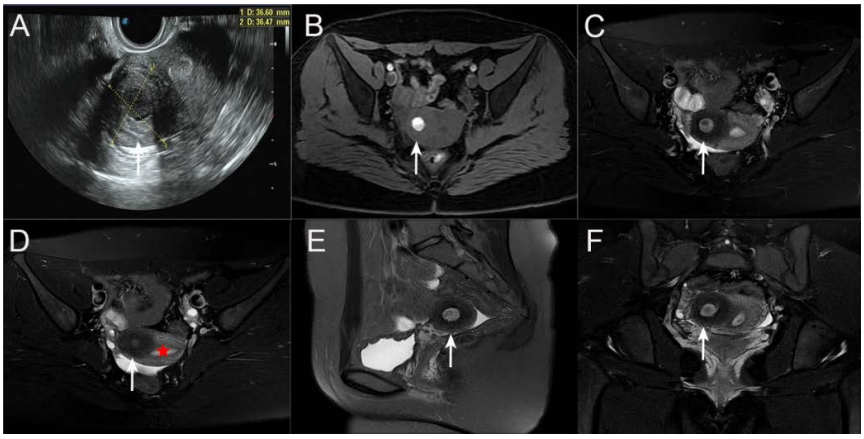

Figure 1: Ultrasonographic, MRI, and pathologic images of an accessory cavitated uterine malformation (ACUM).

The white arrows indicate the ACUM. A) Transvaginal ultrasound shows a hypoechoic, well-defined mass measuring approximately 39 mm × 37 mm in the left uterine wall, with anechoic components. B-F) MRI demonstrates a round, heterogeneous mass within the left uterine wall, approximately 40 mm in diameter. The central portion of the lesion exhibits high signal intensity on both T1-weighted and T2-weighted images, while the surrounding tissue has a similar signal to the normal myometrium. The uterine horns are visible on both sides of the lesion (red □), and an additional adenomyosis focus is seen superior to the main lesion (red arrow). G) Low-power microscopic examination reveals endometrial glands and stroma surrounded by a well-organized, concentric layer of smooth muscle tissue, forming an organ-like structure, which is distinct from the typical distribution seen in adenomyosis. H) High-power view of the endometrial glands and stroma. I) The surrounding myometrium shows the presence of inflammatory cells and hemosiderin deposits, indicative of a reparative and reactive tissue change due to obstructed blood outflow.

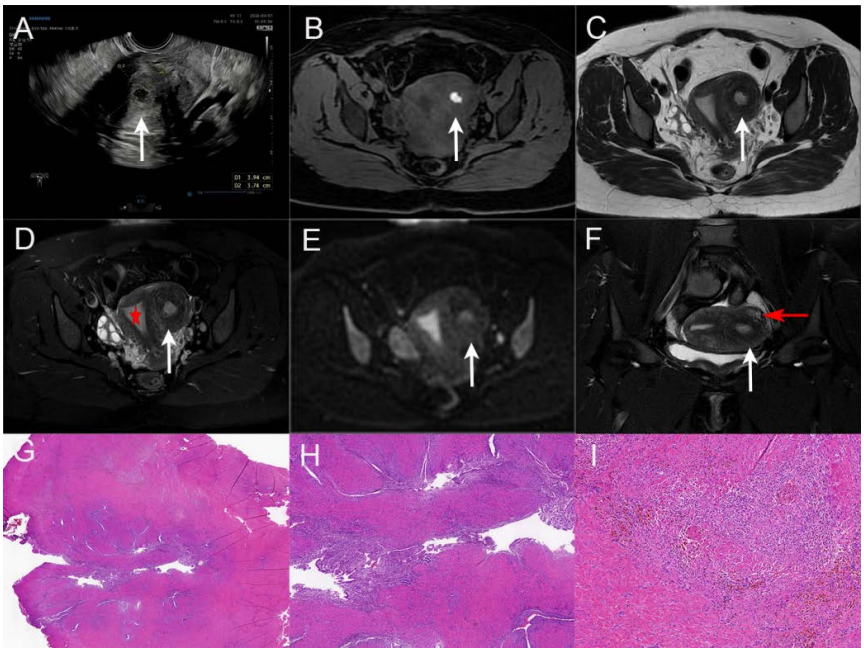

Figure 2: Ultrasonographic and MRI images of an accessory cavitated uterine malformation (ACUM). The white arrows indicate the ACUM. A) Transvaginal ultrasound shows a heterogeneous, hypoechoic mass measuring approximately 37 mm × 36 mm in the right uterine wall, with an indistinct border and a central area of relatively decreased echogenicity. B-F) MRI examination demonstrates a well-defined, round-shaped mass in the right uterine wall, measuring approximately 38 mm × 35 mm × 39 mm. The mass exhibits isointense signal on T1-weighted images and hypointense signal on T2-weighted images, with a central round focus of heterogeneous high signal intensity on both T1-weighted and T2-weighted sequences. The lesion is clearly demarcated from the uterine cavity, and the bilateral uterine horns appear normal (red □).

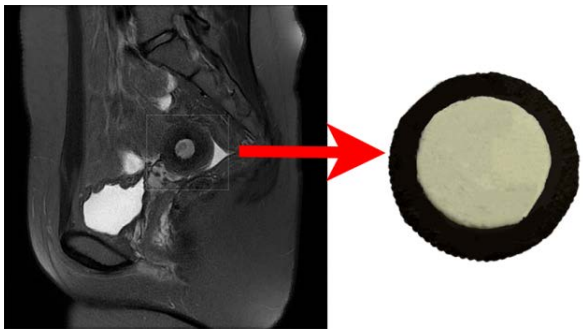

Figure 3: MRI appearance of the ACUM lesion resembles the "open Oreo cookie" sign.

KEYWORDS

Accessory cavitated uterine malformation; Magnetic Resonance Imaging (MRI); Adenomyosis

ABBREVIATIONS

AUCM = Accessory Cavitated Uterine Malformation
MRI = Magnetic Resonance Imaging
T1WI = T1-Weighted Imaging
T2WI = T2-Weighted Imaging
ER = Estrogen Receptor
PR = Progesterone Receptor
GnRH-a = Gonadotropin-Releasing Hormone Agonists