CASE REPORT

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Uterine Mesothelial Cyst Arising from the Myometrium: A Rare Case Report

Myometriyumdan Kaynaklanan Uterin Mezotelyal Kist: Nadir Bir Olgu Sunumu

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Abstract

Intramyometrial mesothelial cysts of the uterus are extremely rare, with only a few cases described in the literature. This report aims to present a uterine mesothelial cyst originating from the myometrium and to highlight its clinical, radiological, and histopathological features. A 42-year-old asymptomatic woman who had been followed for approximately five years for a uterine cyst presented for routine evaluation. Transvaginal ultrasonography revealed a 5.7×3.2 cm bilobulated hypoechoic intramural cystic lesion in the uterine fundus. Pelvic magnetic resonance imaging demonstrated a 5×3.5 cm well-defined, septated intramyometrial cystic lesion that was hypointense on T1-weighted and hyperintense on T2-weighted images. Due to cyst enlargement, difficulty attending regular follow-ups, and anxiety regarding malignancy, total laparoscopic hysterectomy was performed. Gross pathology showed a septated cyst containing serous fluid. Microscopy revealed a cyst wall lined by benign cuboidal mesothelial cells without atypia. Immunohistochemistry showed strong human bone marrow endothelial marker-1 and PANCK positivity, weak D2-40 and CK5/6 positivity, and estrogen and progesterone receptor negativity, confirming a uterine mesothelial cyst. Intramyometrial mesothelial cysts can mimic other uterine cystic lesions clinically and radiologically. Definitive diagnosis requires histopathology supported by a broad immunohistochemical panel. Hysterectomy provides the lowest recurrence risk in women who have completed childbearing, whereas complete cyst excision is essential for fertility preservation. More case reports are needed to better understand their behavior.

Keywords: Human bone marrow endothelial marker-1 (HBME1), immunohistochemistry, intramyometrial mesothelial cyst

Öz

Uterusun intramiyometriyal yerleşimli mezotelyal kistleri son derece nadirdir ve literatürde yalnızca birkaç olgu bildirilmiştir. Bu çalışmada, intramiyometriyal kökenli uterin mezotelyal kist olgusunu sunarak klinik, radyolojik ve histopatolojik özellikleriyle farkındalık oluşturmayı amaçladık. Kırk iki yaşında, yaklaşık beş yıldır uterin kist nedeniyle izlenen asemptomatik kadın hastada, transvajinal ultrasonografide uterin fundusta 5,7×3,2 cm boyutunda bilobüle hipoekoik intramural kistik lezyon saptandı. Manyetik rezonans görüntüleme, T1'de hipointens, T2'de hiperintens, iyi sınırlı ve septalı 5×3,5 cm intramiyometriyal kist ile uyumlu bulgular gösterdi. Kistin zamanla büyümesi, hastanın düzenli takibe uyum zorluğu ve malignite endişesi nedeniyle total laparoskopik histerektomi yapıldı. Makroskopide septalı, seröz içerikli kistik yapı izlendi. Mikroskopide kist duvarının atipi içermeyen küboidal mezotelyal hücrelerle döşendiği görüldü. İmmünohistokimyasal incelemede *human bone marrow endothelial marker*-1 ve PANCK güçlü pozitif, D2-40 ve CK5/6 zayıf pozitif, östrojen ve progesteron reseptörü ise negatif bulundu. Bulgular uterin mezotelyal kist ile uyumluydu. İntramyometriyal mezotelyal kistler klinik ve radyolojik olarak diğer uterin kistik patolojileri taklit edebilir. Kesin tanı histopatoloji ve geniş immünohistokimyasal panel ile konulur. Fertilitesini tamamlamış hastalarda histerektomi en düşük nüks riskiyle tedavi seçeneğidir; fertilite isteği olanlarda total eksizyon önem taşır. Nadir görülmesi nedeniyle daha fazla olgu bildirimi gerekmektedir.

Anahtar kelimeler: *Human bone marrow endothelial marker*-1 (HBME1), immünohistokimya, intramiyometriyal mezotelyal kist

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Introduction

Uterine mesothelial cysts are very rare pathologies. Up to now, approximately 150 cases of mesothelial cysts have been reported in the literature, the majority of which have been seen in localizations such as round ligament, mesentery, peritoneum, adnexa, and only 6 cases have been reported in the uterus, and only 3 of them are myometrium-related (1). Since it is a rare pathology, there is no clear consensus in diagnosis and treatment (2). We aimed to increase awareness of this rare pathology by presenting a case of mesothelial cyst originating from the uterus myometrium.

Case Report

A 42-year-old asymptomatic woman with a 5-year history of a uterine cyst presented for routine follow-up. Obstetric history included two vaginal deliveries, one cesarean section, and three curettages. She had no additional comorbidities. Transvaginal ultrasonography revealed a 5.7×3.2 cm bilobulated hypoechoic intramural cystic lesion located in the uterine fundus, accompanied by adenomyosis. Adnexal structures were normal. Pelvic magnetic resonance imaging (MRI) demonstrated a 5×3.5 cm well-circumscribed, septated intramyometrial cystic lesion, hypointense on T1-weighted and hyperintense on T2-weighted images, without contrast enhancement. A previous MRI performed 5 years earlier had shown a 2.4×1.7 cm cyst in the same region (Figure 1). Laboratory tests and tumor markers were within normal limits. Due to difficulty attending regular follow-ups, lesion growth, and anxiety about malignancy, the patient underwent total laparoscopic hysterectomy with bilateral salpingectomy. Postoperative recovery was uneventful. Gross examination revealed a 5×3 cm well-defined, septated intramural cyst containing serous fluid in the uterine fundus (Figure 2). Microscopy showed a cyst wall lined by a single layer of benign cuboidal mesothelial cells without atypia, mitosis, or necrosis. Immunohistochemistry demonstrated strong positivity for human bone marrow endothelial marker-1 (HBME1) and PANCK, weak positivity for D2-40 and CK5/6, and negativity for estrogen and progesterone receptor (ER/PR) (Figure 3). These findings confirmed the diagnosis of a uterine mesothelial cyst. Written and verbal informed consent was obtained from the patient.

Discussion

Uterine mesothelial cysts are rare lesions originating from mesothelial cells and are mainly observed in women of reproductive age (3,4). They were first described by Plaut in 1928 (5). These cysts are categorized as congenital or acquired. Congenital forms arise from mesonephric or paramesonephric ducts, whereas acquired forms may develop secondary to leiomyoma degeneration, adenomyosis, cystic endometrial hyperplasia, or serosal inclusion cysts (6). Their pathogenesis remains unclear, and developmental anomalies, reactive processes, or neoplastic mechanisms have been proposed (1,2,7). Mesothelial cysts may also occur in the pelvis, upper abdomen, and retroperitoneum. The use of various terms—such as "benign multicystic mesothelioma," "cystic mesothelioma," and "peritoneal inclusion cyst"—reflects uncertainty regarding whether these lesions are reactive or neoplastic (5,8). They have been associated with

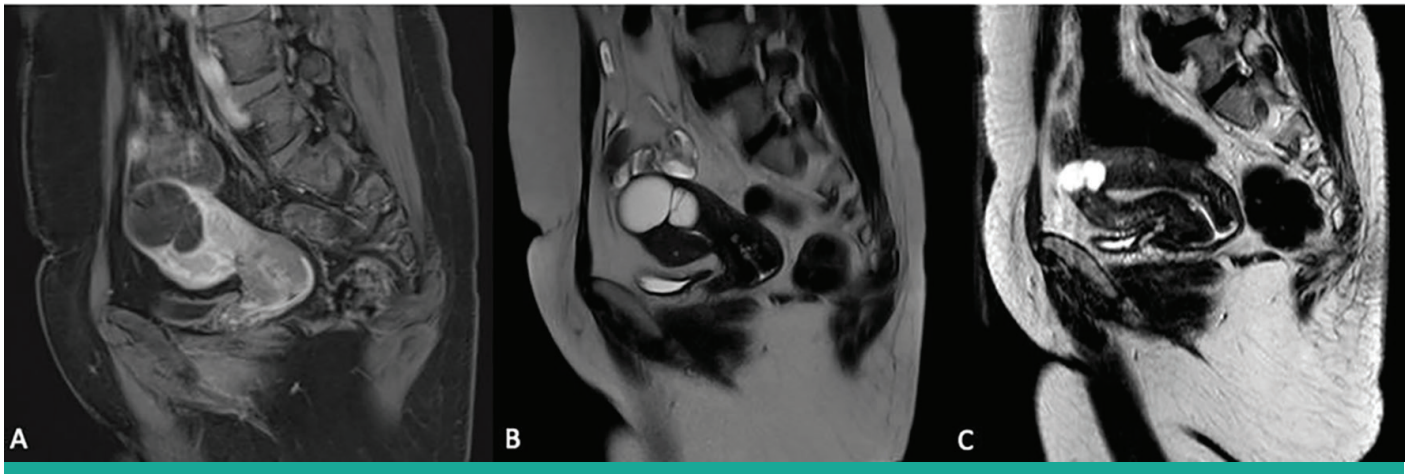

Figure 1. A mesothelial cyst with smooth borders that appears hypointense on T1-weighted MRI (A). and hyperintense on T2-weighted MRI (B). The patient's MRI image taken 5 years ago (C)

MRI: Magnetic resonance imaging

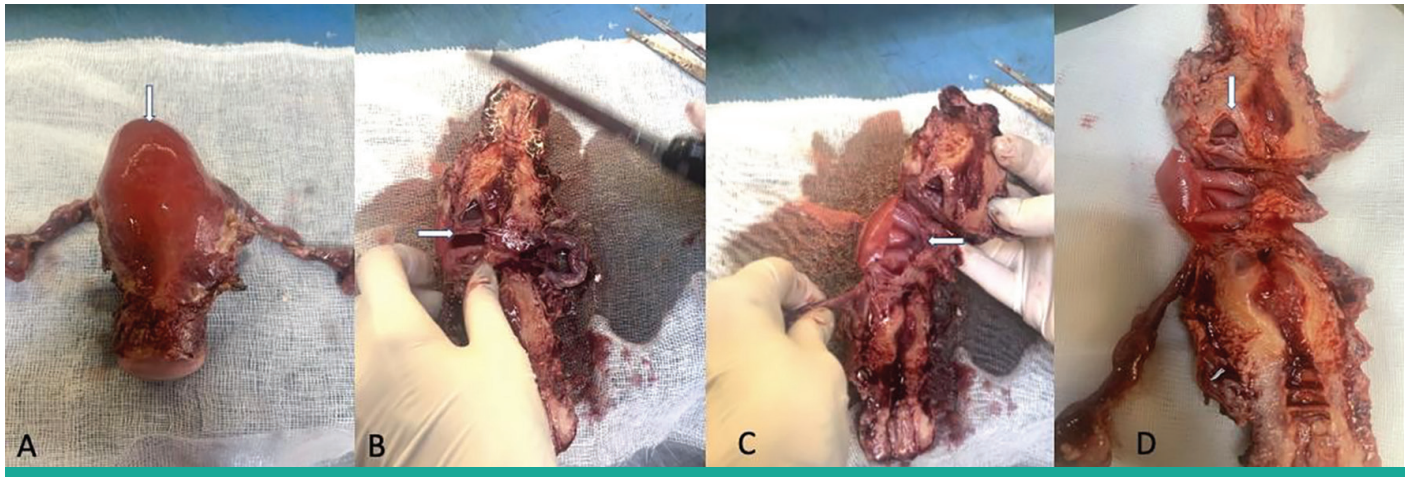

Figure 2. A: Localized mesothelial cyst in the uterine fundus. B: Septate appearance of intramural mesothelial cyst. C: The inner wall of the cyst is smooth and flat. D: The cyst has no connection with the endometrial cavity

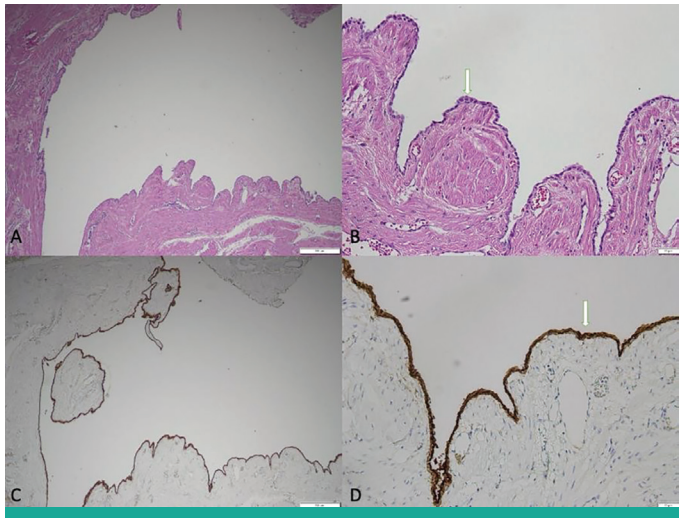

Figure 3. Cyst lined with cuboidal epithelium and stained positive for hematoxylin eosinophilia (A: Original magnification X4, B: Original magnification X20). Cyst wall stained strongly with human bone marrow endothelial marker-1 (HBME-1) in immunohistochemical staining (C: Original magnification X4, D: original magnification X20)

endometriosis, pelvic inflammatory disease, and previous abdominal surgery (1,2,7,8). Literature suggests that chronic peritoneal inflammation may trigger proliferation of mesothelial cells, promoting cyst formation (8). In our patient, although endometriosis or chronic inflammation was not evident, adenomyosis and previous cesarean delivery may have contributed to cyst development. Reported growth during reproductive years, pregnancy, and gonadotropin stimulation has raised the possibility of hormonal responsiveness, although ER/PR expression is mostly negative (1,2,9). ER/PR negativity in our case supports this.

Most uterine mesothelial cysts are asymptomatic and incidentally detected (10). Our patient remained asymptomatic for approximately five years, with gradual cyst enlargement. When cysts enlarge, abdominal pain or a palpable mass may occur; menorrhagia is rare (1,6,10). Ultrasonography is the initial imaging modality and typically shows a multicystic, non-calcified, vascular lesion (2). MRI provides superior anatomic detail but is not the first-line investigation due to cost. Mesothelial cysts typically appear hypointense on T1-weighted and hyperintense on T2-weighted sequences (11). Imaging findings in our case were consistent with these features. Tumor markers are usually normal (5), as in our patient. Histologically, mesothelial cysts are lined by a single layer of cuboidal or columnar mesothelial cells without atypia or mitotic activity, and the wall often consists of fibrous tissue (5,6). Diagnosis is confirmed by histopathology. Immunohistochemical markers such as calretinin, HBME1, WT1, CK5/6, D2-40, and mesothelin commonly show positivity; however, these markers may also stain adenocarcinomas and are not diagnostic alone (12). Calretinin has high sensitivity and specificity, whereas HBME1 shows lower sensitivity (13). Therefore, a broad immunohistochemical panel is recommended (13). In our case, strong HBME1 and PANCK staining and weak D2-40 and CK5/6 positivity, along with ER/PR negativity, were consistent with previous reports.

No standardized treatment protocol exists due to the rarity of mesothelial cysts (7). Asymptomatic patients may be managed with periodic ultrasonographic follow-up (2,11). Ultrasound-guided aspiration can relieve symptoms temporarily but is not recommended when malignancy is

suspected and is associated with rapid fluid reaccumulation (7-9). Laparoscopic excision is an option; however, complete removal may be difficult when the cyst wall is embedded within the myometrium, potentially increasing recurrence (6,7). Recurrences are usually local and, although extremely rare, malignant mesothelioma transformation has been reported (10). For patients without fertility desire or those with high recurrence risk, hysterectomy offers the lowest recurrence rate (1). In our case, total hysterectomy was preferred due to follow-up difficulties and cancer-related anxiety.

Conclusion

In conclusion, mesothelial cysts are uncommon lesions that may mimic other uterine cystic pathologies clinically and radiologically. Definitive diagnosis relies on histopathology supported by a broad immunohistochemical panel. Management should be individualized according to symptoms, reproductive plans, and cyst behavior. Observation with regular ultrasonography is appropriate for asymptomatic patients, whereas surgical excision may be required for symptomatic or recurrent cases. Hysterectomy provides the lowest recurrence risk in patients who have completed childbearing. Further case reports and larger series are needed to clarify the natural history and clinical implications of mesothelial cysts.

Ethics

Informed Consent: Written and verbal informed consent was obtained from the patient.

Footnotes

Authorship Contributions

Surgical and Medical Practices: G.Ö., H.G., Concept: G.Ö., H.G., Design: G.Ö., H.G., Data Collection or Processing: G.Ö., Analysis or Interpretation: G.Ö., H.G., Literature Search: G.Ö., Writing: G.Ö., H.G.

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