

# Anterior sacral meningocele mimicking an adnexal mass in an infertile woman: a report of an extremely unusual case

İnfertil bir kadında adneksiyal kitleyi taklit eden anterior sakral meningesel: çok nadir görülen bir olgu sunumu

Hüseyin Çağlayan Özcan<sup>1</sup>, Mehmet Alptekin<sup>2</sup>, Berna Kaya Uğur<sup>3</sup>, Mete Gürol Uğur<sup>1</sup>, İbrahim Erkuşlu<sup>2</sup>, İrfan Kutlar<sup>1</sup>

<sup>1</sup>Department of Obstetrics and Gynecology, Gaziantep University School of Medicine, Gaziantep, Turkey

<sup>2</sup>Department of Neurosurgery, Gaziantep University School of Medicine, Gaziantep, Turkey

<sup>3</sup>Department of Anesthesiology and Reanimation, Gaziantep University School of Medicine, Gaziantep, Turkey

## ABSTRACT

The differential diagnosis of cystic adnexal masses, some placed retroperitoneally, includes various pathologies. Preoperative diagnosis is important to prevent major neurological deficits during surgery. Herein, we present a case of anterior sacral meningocele diagnosed in an infertile woman using magnetic resonance imaging (MRI) and eliminated with surgery. An MRI performed to determine the origin of retroperitoneal fixated masses helps direct the patient to the right consultant department and may avoid an inappropriate surgical approach, which may lead to a failure. A retroperitoneal mass, such as anterior sacral meningocele, may contribute to the pathogenesis of infertility by distorting the utero-tuboovarian anatomy.

**Keywords:** Adnexal cyst, anterior sacral meningocele, retroperitoneal mass

## ÖZ

Kistik adneksiyal kitlelerin ayırıcı tanısında bazıları retroperitoneal olan çeşitli patolojiler düşünülmelidir. Preoperatif tanı operasyon sırasında gelişebilecek nörolojik sorunları önlemek için önemlidir. Makalemizde, infertil bir kadında manyetik rezonans görüntüleme (MRI) yöntemiyle teşhis edilen ve opere edilen anterior sakral meningesel vakasını sunduk. Retroperitoneal bölgeye fikse olmuş kitlelerinin kökenini bulmak için yapılan MRI hastaya daha sonra büyük sorunlara neden olabilecek yetersiz bir cerrahi işlem uygulamayı önlemenin yanında, hastanın uygun bir bölüme yönlendirilmesine yardımcı olur. Anterior sakral meningesel gibi retroperitoneal bölgeye yerleşmiş bir kitle utero-tuboovarian anatomiyi bozarak infertilite gelişmesine yol açabilir.

**Anahtar kelimeler:** Adneksiyel kist, anterior sakral meningesel, retroperitoneal kitle

## INTRODUCTION

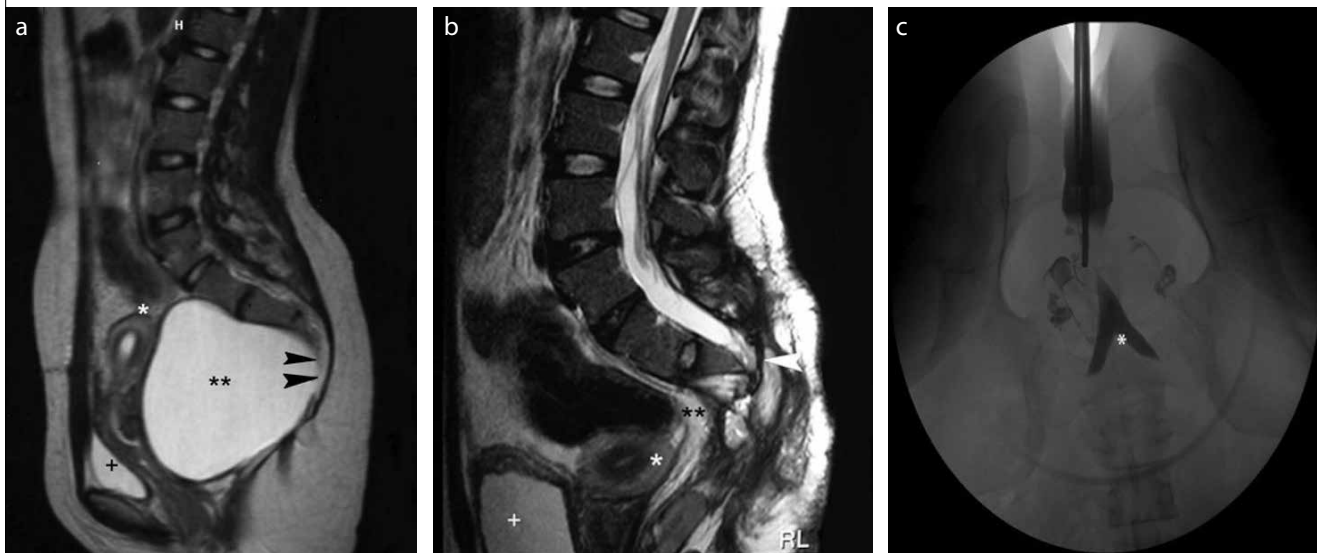
An adnexal mass with a completely cystic appearance is a frequently encountered clinical entity and most often arises from the ovaries. The differential diagnosis includes various pathologies, some of which are very rare and are placed retroperitoneally. The differential diagnosis of retroperitoneal cystic masses includes arachnoid cyst, anterior sacral meningomyelocele (ASM), schwannoma, teratoma, hamartoma, neuroenteric cyst, adrenal cyst, or primary mucinous cystic tumor of the retroperitoneum (1). Retroperitoneal masses should be considered particularly when dealing with uncertain or unusual pelvic symptoms and physical findings (2). In this report, we present a very rare case of ASM mimicking a cystic adnexal mass in a primary infertile woman.

## CASE PRESENTATION

A 26-year-old null gravid woman, who was married for 6 years, had been admitted to a state hospital complaining about infertility. She also had complaints of dyspareunia, lower back pain with numbness, and pain in both legs over a period of 20 years. Her pain was increased when she bent over or walked a short distance. The pain was relieved after resting. She previously had undergone an elective laparotomy in a state hospital due to a cystic adnexal mass located posterior to the uterus, but intraoperative observation revealed that the cyst was retroperitoneal in origin. Two months later, the patient was referred to our clinic without any further intervention.

The physical examination of the patient revealed an immobile mass of 10×10 cm in diameter with a soft consistency, which

Figure 1. a-c. (a) Preoperative sagittal T2-weighted magnetic resonance image demonstrating an anterior sacral meningocele (\*\*). Note the uterus (\*) compressed between the meningocele and the bladder (+). A large passage between the subarachnoid space and the meningocele (black arrowheads). (b) Postoperative sagittal magnetic resonance image showing normally positioned uterus (\*) and bladder (+). The absence of meningocele (\*\*) indirectly suggests the complete obliteration of the anterior meningocele defect with fascia substitute (white arrowhead) (c) Hysterosalpingography image demonstrating incomplete uterine septum (\*).



was localized posteriorly to the uterus. The uterus was displaced more superiorly in the pelvis due to this mass, which led to difficult palpation and observation of the cervix during the speculum examination. An ultrasonography examination revealed a completely cystic mass of 11×10 cm in diameter localized posteriorly to the uterus and displacing it upwards. We also visualized an incomplete uterine septum and planned to perform hysterosalpingography (HSG) after an appropriate management of the mass. Magnetic resonance imaging (MRI) was then performed because of the possible retroperitoneal origin of the mass. MRI revealed an anterior sacral meningocele (Figure 1a). The patient was then referred to the department of neurosurgery with a preliminary diagnosis of ASM.

The neurosurgeons have preferred a posterior surgical approach. A mid-sagittal skin incision was made between L5-S4 levels. The closure of this area was impossible without dural substitute. First, the arachnoid membrane around the nerves was secured. Then, the communication between the ASM and the dura mater space was disconnected by tethering the neck of the meningocele. Postoperative control with MRI revealed that the uterus and bladder were returned to their normal position (Figure 1b). Two months after surgery, the planned HSG that revealed incomplete uterine septum was performed (Figure 1c). The patient had undergone hysteroscopic resection.

#### DISCUSSION

Retroperitoneal masses should almost always be considered in differential diagnosis of pelvic masses. One of the very rare types of these masses is ASM. ASM is a unilocular or multilocular enlargement of the meninges from the sacral spinal canal to the retroperitoneal presacral space through sacral bony defects and is characterized by a communication with the subarachnoid

space (3, 4). Congenital and acquired are the major types of ASM in literature (5). Congenital ASM is usually accompanied by urological and gynecological pathologies; however, it can rarely occur alone (6). In our case, we detected an incomplete uterine septum, and this is as far as we know, the first report of ASM with uterine septum in an infertile woman.

Currarino syndrome is a well-known autosomal-dominant disease, which includes sacral bony defect, anorectal malformation, and presacral mass. This syndrome is most often presented with congenital ASM (7). Acquired ASM develops due to preceding enlargement of the dura mater and arachnoid membrane that can or cannot result from connective tissue disorders (8).

In two-thirds of all cases, localized symptoms emerge (7). These symptoms occur as a result of pressure on the sacral nerve roots, rectum, bladder, or genitalia. Lower back and pelvic pain, constipation, difficulties in defecation, dysmenorrhea, dyspareunia, and urinary incontinence are the most common symptoms. In the differential diagnosis of retroperitoneal masses, ASMs should always be considered (6, 9). A fixed pelvic mass located posteriorly to the uterus, which is not mobile during a bimanual pelvic examination, should also give the clinician a suspicion of a retroperitoneal pathology and a need for more definitive imaging techniques, such as MRI, other than pelvic ultrasonography. Because of the poor visualization resolution, such as revealing connection between the sacral spinal canal and the pathology, ultrasonography should not be considered primarily (8). MRI is the gold standard for the diagnosis of ASM, which allows for the most accurate preoperative imaging to determine the surgical course (3, 10). ASMs can be misdiagnosed as an adnexal mass in gynecological practice; however, it is usually located posteriorly and both ovaries may be visualized separately (11). Due to the

retroperitoneal location of the ASM, diagnosis may also be made during the surgery (6, 12). Preoperative diagnosis is important to prevent major neurological deficits during surgery. A retroperitoneal mass, which is found unexpectedly at an operation due to the other pelvic diseases, should not be resected (2). Also, any diagnostic intervention, such as transrectal or transvaginal aspiration or biopsy, should be avoided to prevent meningitis, which can result from rectal injuries during the course of anterior sacral meningocele. If it occurs, it may lead to severe morbidity and mortality (13).

Anterior sacral meningomyelocele can be symptomatic or asymptomatic. When it is symptomatic, surgical approach is necessary for relieving the patient's complaints. Traditionally, neurosurgeons intend to extirpate the relation between the subarachnoid space and the meningocele (14). Anterior, posterior, and endoscopic approaches are the surgical choices. Easy ligation of the neck of lesion can be achieved and any associated spinal cord problem can be determined by the posterior approach (10).

## CONCLUSION

As in our case, a large retroperitoneal mass may contribute to the pathogenesis of infertility by distorting the utero-tuboovarian relation. Gynecologists have to take into account that gynecologic anomalies may accompany ASMs and consider further evaluation in these patients. An MRI performed to determine the origin of retroperitoneally fixated masses helps direct the patient to the right consultant department and may avoid an inappropriate surgical approach, which can lead to a failure. Also, a multidisciplinary surgical approach with a neurosurgeon to such patients will help to prevent possible complications.

**Informed Consent:** Written informed consent was obtained from our patient who participated in this study.

**Peer-review:** Externally peer-reviewed.

**Author Contributions:** Concept - İ.K.; Design - M.A.; Supervision - İ.E.; Data Collection and/or Processing - B.K.U.; Analysis and/or Interpretation - H.Ç.Ö.; Writing Manuscript - H.Ç.Ö.; Critical Review - M.G.U.

**Conflict of Interest:** No conflict of interest was declared by the authors.

**Financial Disclosure:** The authors declared that this study has received no financial support.

**Hasta Onamı:** Yazılı hasta onamı bu çalışmaya katılan hastadan alınmıştır.

**Hakem Değerlendirmesi:** Dış bağımsız.

**Yazar Katkıları:** Fikir - İ.K.; Tasarım - M.A.; Denetleme - İ.E.; Veri Toplanması ve/veya İşlemesi - B.K.U.; Analiz ve/veya Yorum - H.Ç.Ö.; Yazıyı Yazan - H.Ç.Ö.; Eleştirel İnceleme - M.G.U.

**Çıkar Çatışması:** Yazarlar çıkar çatışması bildirmemişlerdir.

**Finansal Destek:** Yazarlar bu çalışma için finansal destek almadıklarını beyan etmişlerdir.

## REFERENCES

- Balat O, Aydın A, Sirikci A, Kutlar I, Aksoy F. Huge primary mucinous cystadenoma of the retroperitoneum mimicking a left ovarian tumor. *Eur J Gynaecol Oncol* 2001; 22: 454-5.
- Sharp KW, Reddy VS. Nongynecologic conditions encountered by the gynecologic surgeon In: Rock JA, Jones III HW, editors. *Te Linde's Operative Gynecology*, ninth edition. Philadelphia: Lippincott Williams & Wilkins; 2003. p.1281-84.
- Tostain J, Perraud Y, Preynat P, Duthel R. Anterior sacral meningocele with urologic manifestations. Report of 3 cases. *Prog Urol* 1992; 2: 464-71.
- Franck B, Dourlhes N, Ali BM, Heurtebise F, Ravon R, Moreau JJ. The anterior sacral meningocele. *Rachis* 1993; 5: 147-52.
- Haga Y, Cho H, Shinoda S, Masuzawa T. Recurrent meningitis associated with complete Currarino Triad in an adult. *Neurol Med Chir* 2003; 43: 505-8. [\[CrossRef\]](#)
- Khan AK, Deb S, Ray DK, Nag BK. The coperitoneal shunt in case of symptomatic anterior sacral meningocele. *Neurol India* 2004; 52: 403-4.
- Lynch S, Wang Y, Strachan T, Burn J, Lindsay S. Autosomal dominant sacral agenesis: Currarino syndrome. *J Med Genet* 2000; 37: 561-6. [\[CrossRef\]](#)
- Shetty DS, Lakhar BN. Lateral sacral lipomyelomeningocele: a rare anomaly. *Neurol India* 2002; 50: 204-6.
- Klenerman L, Merrick MV. Anterior sacral meningocele occurring in a family. *J Bone Joint Surg Br* 1973; 55: 331-4.
- Tani S, Okuda Y, Abe T. Surgical strategy for anterior sacral meningocele: two cases reports. *Neurol Med Chir* 2003; 43: 204-9. [\[CrossRef\]](#)
- Gardner PA, Albright AL. "Like mother, like son:" hereditary anterior sacral meningocele. Case report and review of the literature. *J Neurosurg* 2006; 104: 138-42.
- Forsnes EV, Segna RA. Dural ectasia presenting as an adnexal mass. *Obstet Gynecol* 1996; 88: 712-3. [\[CrossRef\]](#)
- Bal S, Kurtulmus S, Kocyigit H, Gurgan A. A case with cauda equina syndrome due to bacterial meningitis of anterior sacral meningocele. *Spine* 2004; 29: 298-9. [\[CrossRef\]](#)
- Hanna AS, Morandi X. Anterior sacral meningocele in pregnancy. Case report. *J Neurosurg Spine* 2001; 94: 162-4. [\[CrossRef\]](#)

## How to cite:

Özcan HÇ, Alptekin M, Kaya Uğur B, Uğur MG, Erkutlu İ, Kutlar İ. Anterior sacral meningocele mimicking an adnexal mass in an infertile woman: a report of an extremely unusual case. *Eur J Ther* 2017; 23: 184–6.