Introduction

Anterior sacral meningocele (ASM) is a rare congenital anomaly characterized by dural herniation through a defect in the anterior sacrum. This congenital pathology may be rarely complicated by an accompanying rectothecal fistula and cerebrospinal fluid (CSF) leakage in or around the anus known as rectorrhea. This was first reported in 1999, and since then, only six additional cases, including this case, have been published. Here, we present a 24-year-old female with ASM, a rectothecal fistula, and rectorrhea, who was successfully treated with a single-stage posterior approach.

Case Report

A 24-year-old female complained of intermittent watery discharge from her anus over the past 12 months. On examination, she had a fistula posterior to the anus through which a drop of clear watery liquid extruded utilizing a Valsalva maneuver. Ultrasound revealed a presacral cystic mass. On pelvic radiography, a scimitar defect was observed involving the left caudal side of the sacrum. The lumbosacral 3D computerized tomography (CT) showed the scimitar...
defect more clearly [Figure 3]. On MRI, the cystic mass in front of the sacrum was hypointense on T1 and hyperintense on T2-weighted images; these findings were compatible with an ASM [Figure 4]. The lumbosacral CT metrizamide myelogram confirmed communication between the presacral cyst and the thecal sac. Further extension of the meningocele toward the anal region was also suspected [Figure 5].

**Surgery**

Through a midline posterior incision from S1 to sacral hiatus, en block laminotomies were performed. At the caudal side of the thecal sac and the level of the scimitar defect, a narrow-necked ostium of the meningocele was identified, explored, ligated, and sectioned. Before the closure of the neck of the meningocele, CSF was aspirated.

The bony defect was covered with the bone flap and affixed with mini plates [Figure 6]. Exploration and closure of the fistula through a secondary anterior approach were not attempted.

**Postoperative course**

The postoperative course was uneventful; there were no complications. At 10-year follow-up, the rectal examination disclosed spontaneous closure of the fistula. The MRI also confirmed the complete resolution of the previously noted meningocele/fistula [Figure 7].

**DISCUSSION**

ASM is a rare congenital anomaly characterized by herniation of a meningocele sac into the presacral space through a sacral

---

### Table 1: Detailed information about all seven cases including the presented case.

| Authors       | Year | Sex  | Age | Clinical picture                      | Closure of the pedicle of the sac                  | Outcome |
|---------------|------|------|-----|---------------------------------------|---------------------------------------------------|---------|
| Fitzpatrick et al. [3] | 1999 | Female | 31 | Meningitis, rectal discharge          | Posterior approach, excision fistula               | Good    |
| Phillips et al. [7]     | 2006 | Male  | 48 | Meningitis, rectorrhea               | Laparotomy, excision fistula                       | Good    |
| Sánchez et al. [8]     | 2008 | Male  | 64 | Meningitis, rectorrhea               | Laparotomy, excision fistula                       | Good    |
| Bergeron et al. [1]    | 2010 | Female | 40 | Meningitis, rectorrhea               | Laparotomy, closure fistula, colostomy             | Good    |
| Koksal et al. [4]      | 2011 | Female | 44 | Recurrent meningitis, rectorrhea      | Posterior, after Ventricular drainage              | Good    |
| Mankotia, et al. [5]   | 2018 | Female | 52 | Only rectorrhea                      | Posterior, closure fistula                         | Good    |
| Current case           | 2020 | Female | 24 | Only rectorrhea                      | Posterior approach, fistula left untreated         | Good    |

**Figure 1:** Photograph of the patient in the prone position, note the fistula posterior to the anus, and a drop of watery liquid.

**Figure 2:** Radiograph of the sacrum, AP view shows the scimitar defect on the left side.
defect.\textsuperscript{[1,3-8]} ASM is more common in white females and usually becomes symptomatic in the second or third decades of life (e.g., at childbearing age).\textsuperscript{[1,3-8]}

**Currarino syndrome**

This congenital anomaly may be a part of Currarino’s triad, in which ASM coexists with sacral agenesis and anorectal malformation. Furthermore, associated with this anomaly are the following; duplication of the uterus, epidermoid or dermoid cysts, lipomas, and teratomas.\textsuperscript{[1,3-8]}

**Clinical picture**

As ASM is not typically associated with cutaneous abnormalities or posterior spina bifida, many patients with ASM remain asymptomatic or show minor symptoms (e.g., constipation, dysuria, and dysmenorrhea pressure on the rectum, urinary bladder/female genital organ findings).\textsuperscript{[1,3-8]} Most ASMs are diagnosed incidentally while performing pelvic sonography, lumbosacral MRI scans, or during exploratory laparotomy.\textsuperscript{[1,3-8]} On rare occasions, the constant pressure of the cyst on the rectum in association with CSF pulsations may lead to erosion and fistula formation at the anorectal region with or without rectorrhea.\textsuperscript{[1,3-5,7,8]}

**Figure 3:** Reconstructed 3D CT shows the scimitar defect more clearly.

**Figure 4:** Sagittal T2-weighted MR image demonstrates a cystic mass in presacral region, note an extension of the cyst toward the anorectal region. (white arrow).

**Figure 5:** CT metrizamide myelography shows the cyst and its extension to the anorectal region.

**Figure 6:** (a) Intraoperative photograph showing reconstruction of the sacrum with mini plate, (b) postoperative sacral X-ray ap view shows the reconstructed sacrum with mini plates.

**Figure 7:** Postoperative sagittal 2-weighted MRI 10 years after surgery; no cystic mass anymore, note the posterior aspect of the sacrum indicating mini plates.
In such cases, with rectothecal fistula and rectorrhea, leakage of watery fluid in the anorectal region may point to the proper diagnosis.\textsuperscript{[1,3,5,7,8]} Rarely, intestinal bacteria may contaminate the CSF through the rectothecal fistula resulting in bacterial meningitis.\textsuperscript{[1,7,8]}

**Imaging**

On plain radiographs, the scimitar sacrum, seen in 50% of cases, is the pathognomonic finding for ASM.\textsuperscript{[1,3-8]} This is further confirmed utilizing 3D CT. Continuity of the cyst with the thecal is best demonstrated utilizing metrizamide CT myelography.\textsuperscript{[1,3-8]} MRI Imaging studies may also show the scimitar sacrum with herniation of the meningocele through the defect and may also demonstrated, whether there is an accompanying tethered cord (e.g., low-lying conus and/or thick fatty filum).

**Management**

Surgical intervention for ASM is typically warranted.\textsuperscript{[1,3-8]} The aim of surgery is to obliterate the communication between the subarachnoid space of the thecal sac and herniated cyst.\textsuperscript{[1,3-5,7,8]} In uncomplicated cases, a dorsal transsacral approach with the closure of the pedicle of the meningocele is safe.\textsuperscript{[5]} Even, in complicated cases with a history of meningitis, after eradication of the infection, the posterior approach is usually successful.\textsuperscript{[1,4]} In the event that a low-lying conus exists, detethering the spinal cord with resection of the filum may be required; the resultant bony defect from the sacral laminectomy can be covered with an appropriate size titanium mesh. In patients who have undergone en block laminotomies, the bony flap can be replaced and fixed with the use of mini plates.

In addition to the posterior approach, an aggressive anterior surgical procedure may be warranted to address life-threatening meningitis and/or gross contamination.\textsuperscript{[1,5]} The one stage, retro, or transperitoneal approach have mostly been done by the colorectal surgeons.\textsuperscript{[1,7,8]} Recently, transperitoneal transcystic laparoscopic closure of the orifice of the meningocele by plastic clips has also been described.\textsuperscript{[2]}

**CONCLUSION**

ASM with a rectothecal fistula with or without rectorrhea is extremely rare and should be fully evaluated preoperatively with MR/3D CT and myelo-CT studies, so they can be effectively and safely surgically corrected.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**REFERENCES**

1. Bergeron E, Roux A, Demers J, Vanier LE, Moore L. A 40-year-old woman with cauda equina syndrome caused by rectothecal fistula arising from an anterior sacral meningocele. Neurosurgery 2010;67:E1464-75.
2. Clatterbuck RE, Jackman SV, Kavoussi LR, Long DM. Laparoscopic treatment of an anterior sacral meningocele. Case illustration. J Neurosurg 2000;92:246.
3. Fitzpatrick MO, Taylor WA. Anterior sacral meningocele associated with a rectal fistula. Case report and review of the literature. J Neurosurg 1999;91:124-7.
4. Koksal A, Canyigit M, Kara T, Ulus A, Gokbayir H, Sarisahin M. Unusual presentation of an anterior sacral meningocele: Magnetic resonance imaging, multidetector computed tomography, and fistulography findings of bacterial meningitis secondary to a rectothecal fistula. Jpn J Radiol 2011;29:528.
5. Mankotia DS, Sawarkar DP, Singh PK, Kumar A, Verma SK, Chandra PS, et al. Rare case of cerebrospinal fluid proctorrhea caused by anterior sacral meningocele with rectothecal fistula. World Neurosurg 2018;114:323-5.
6. Massimi L, Calisti A, Koutzoglou M, Di Rocco C. Giant anterior sacral meningocele and posterior sagittal approach. Childs Nerv Syst 2003;19:722-8.
7. Phillips JT, Brown SR, Mitchell P, Shorthouse AJ. Anaerobic meningitis secondary to a rectothecal fistula arising from an anterior sacral meningocele: Report of a case and review of the literature. Dis Colon Rectum 2006;49:1633-5.
8. Sánchez AA, Iglesias CD, López CD, Cecilia DM, Gómez JA, Barbadillo JG, et al. Rectothecal fistula secondary to an anterior sacral meningocele. J Neurosurg Spine 2008;8:487-9.

**How to cite this article:** Rahimizadeh A, Ehteshami S, Rahimizadeh A, Karimi M. Anterior sacral meningocele complicated by rectothecal fistula and rectorrhea: A case report and review of the literature. Surg Neurol Int 2020;11:117.