



## Case Report

# Anterior sacral meningocele complicated by rectothecal fistula and rectorrhoea: A Case report and review of the literature

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## ABSTRACT

**Background:** Anterior sacral meningocele (ASM) is a rare congenital anomaly. It is characterized by herniation of the dura through a defect in the anterior sacrum. Rarely, however, it may extend to the rectal area through a rectothecal fistula with or without rectorrhoea.

**Case Description:** Here, we present a case of ASM associated with a rectothecal fistula and rectorrhoea. Surgical closure of the ostium of the cyst through a posterior approach resulted in long-term improvement

**Conclusion:** An ASM with both rectothecal fistula and rectorrhoea is extremely rare.

**Keywords:** Anterior sacral meningocele, *En block* sacrum laminotomy, Mini plate, MRI, Rectothecal fistula, Sacrum reconstruction

## INTRODUCTION

Anterior sacral meningocele (ASM) is a rare congenital anomaly characterized by dural herniation through a defect in the anterior sacrum.<sup>[1,3-8]</sup> This congenital pathology may be rarely complicated by an accompanying rectothecal fistula and cerebrospinal fluid (CSF) leakage in or around the anus known as rectorrhoea.<sup>[1,3-8]</sup> This was first reported in 1999,<sup>[3]</sup> and since then, only six additional cases, including this case, have been published [Table 1].<sup>[1,4,5,7,8]</sup>

Here, we present a 24-year-old female with ASM, a rectothecal fistula, and rectorrhoea, who was successfully treated with a single-stage posterior approach.

## CASE REPORT

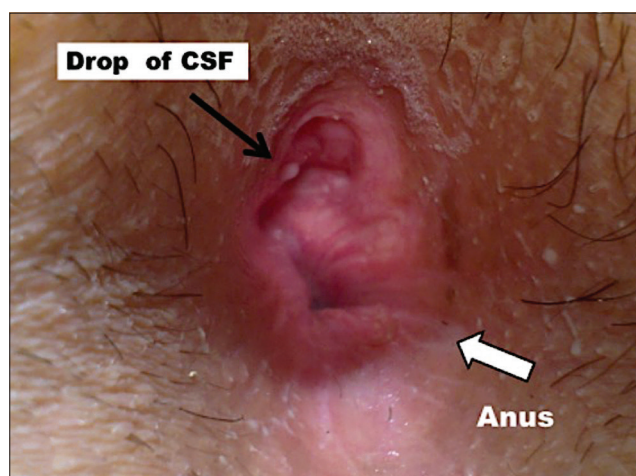
A 24-year-old female complained of intermittent watery discharge from her anus over the past 12 months. On examination, she had a fistula posterior to the anus through which a drop of clear watery liquid extruded utilizing a Valsalva maneuver [Figure 1]. Ultrasound revealed a presacral cystic mass. On pelvic radiography, a scimitar defect was observed involving the left caudal side of the sacrum [Figure 2]. The lumbosacral 3D computerized tomography (CT) showed the scimitar

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**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 <i>et al.</i> <sup>[3]</sup>	1999	Female	31	Meningitis, rectal discharge	Posterior approach, excision fistula	Good
Phillips <i>et al.</i> <sup>[7]</sup>	2006	Male	48	Meningitis, rectorrhea	Laparotomy, excision fistula	Good
Sánchez <i>et al.</i> <sup>[8]</sup>	2008	Male	64	Meningitis, rectorrhea	Laparotomy, excision fistula, colostomy	Good
Bergeron <i>et al.</i> <sup>[1]</sup>	2010	Female	40	Meningitis, rectorrhea	Laparotomy, closure fistula, colostomy	Good
Koksal <i>et al.</i> <sup>[4]</sup>	2011	Female	44	Recurrent meningitis, rectorrhea	Posterior, after Ventricular drainage	Good
Mankotia, <i>et al.</i> <sup>[5]</sup>	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.

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.



**Figure 2:** Radiograph of the sacrum, AP view shows the scimitar defect on the left side.

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].

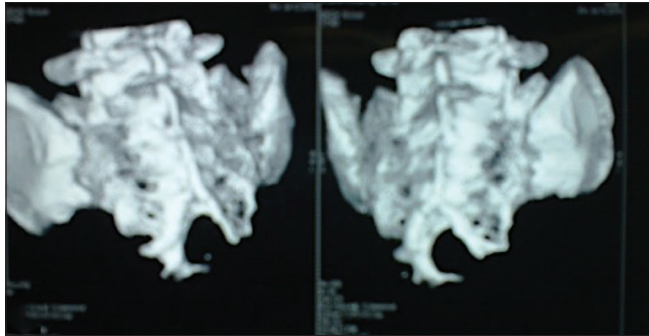
### DISCUSSION

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

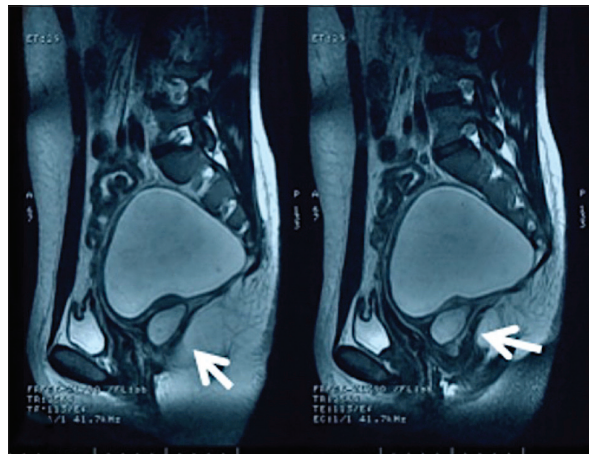
defect.<sup>[1,3-8]</sup> ASM is more common in white females and usually becomes symptomatic in the second or third decades of life (e.g., at childbearing age).<sup>[1,3-8]</sup>

### Currarino syndrome

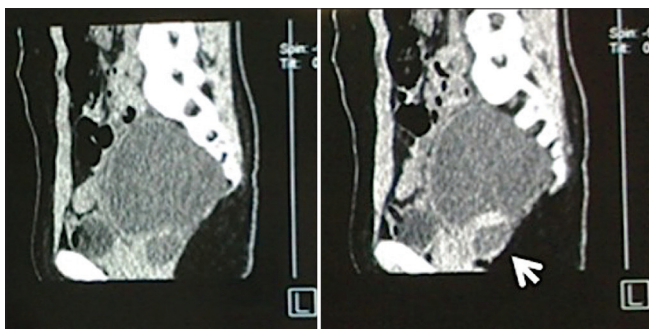
This congenital anomaly may be a part of Currarino's triad, in which ASM coexists with sacral agenesis and anorectal malformation. Furthermore, associated with this anomaly



**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).

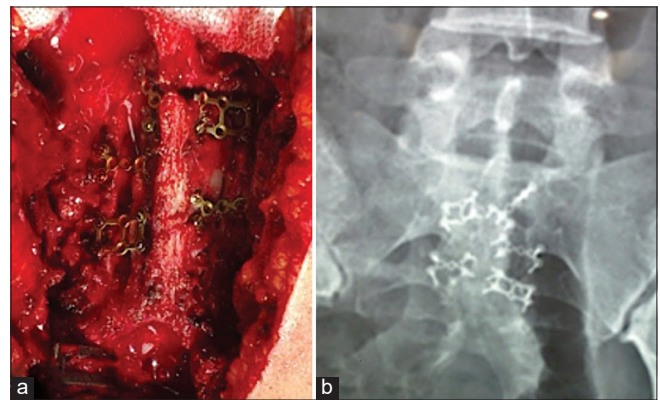


**Figure 5:** CT metrizamide myelography shows the cyst and its extension to the anorectal region.

are the following; duplication of the uterus, epidermoid or dermoid cysts, lipomas, and teratomas.<sup>[1,3-8]</sup>

### 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).<sup>[1,3-8]</sup> Most ASMs are diagnosed incidentally while performing pelvic sonography, lumbosacral MRI scans, or during exploratory laparotomy.<sup>[1,3-8]</sup> 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 rectorrhhea.<sup>[1,3-5,7,8]</sup>



**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 rectorrhoea, leakage of watery fluid in the anorectal region may point to the proper diagnosis.<sup>[1,3,5,7,8]</sup> Rarely, intestinal bacteria may contaminate the CSF through the rectothecal fistula resulting in bacterial meningitis.<sup>[1,3,7,8]</sup>

### Imaging

On plain radiographs, the scimitar sacrum, seen in 50% of cases, is the pathognomonic finding for ASM.<sup>[1,3-8]</sup> This is further confirmed utilizing 3D CT. Continuity of the cyst with the thecal is best demonstrated utilizing metrizamide CT myelography.<sup>[1,3-8]</sup> 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.<sup>[1,3-8]</sup> The aim of surgery is to obliterate the communication between the subarachnoid space of the thecal sac and herniated cyst.<sup>[1,3-5,7,8]</sup> In uncomplicated cases, a dorsal transsacral approach with the closure of the pedicle of the meningocele is safe.<sup>[5]</sup> Even, in complicated cases with a history of meningitis, after eradication of the infection, the posterior approach is usually successful.<sup>[1,4]</sup> 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.<sup>[3,5]</sup> The one stage, retro, or transperitoneal approach have mostly been done by the colorectal surgeons.<sup>[1,7,8]</sup> Recently, transperitoneal transcystic laparoscopic closure of the orifice of the meningocele by plastic clips has also been described.<sup>[2]</sup>

### CONCLUSION

ASM with a rectothecal fistula with or without rectorrhoea 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.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

### REFERENCES

- 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.
- Clatterbuck RE, Jackman SV, Kavoussi LR, Long DM. Laparoscopic treatment of an anterior sacral meningocele. Case illustration. *J Neurosurg* 2000;92:246.
- 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.
- 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.
- 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.
- Massimi L, Calisti A, Koutzoglou M, Di Rocco C. Giant anterior sacral meningocele and posterior sagittal approach. *Childs Nerv Syst* 2003;19:722-8.
- 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.
- 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.

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