Successful management of a giant anterior sacral meningocele with an endoscopic cutting stapler: case report

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Anterior sacral meningoceles (ASMs) are rare defects in the sacrum with thecal sac herniations and symptoms that commonly include constipation, dysmenorrhea, and urinary disturbances. An ASM causing hydronephrosis and acute renal failure from compression of the lower portion of the urinary tract is a rare clinical entity. Only one other case has been reported. The authors present the case of a 37-year-old man admitted for obstructive renal failure and hydronephrosis due to a giant ASM that measured 25 × 12 × 18 cm and compressed the ureters and bladder. The ASM was successfully treated via an anterior transabdominal approach in which the authors used a novel technique for watertight closure of the meningocele pedicle with an endoscopic cutting stapler. The authors review the literature and discuss the surgical options for the treatment of ASMs, specifically the management of ASMs in the context of obstructive renal failure and hydronephrosis.

http://thejns.org/doi/abs/10.3171/2015.8.SPINE15129

KEY WORDS meningocele; spina bifida; hydronephrosis; renal failure; congenital

Case Report

Initial Presentation and Examination

A 37-year-old man with spina bifida occulta, initially diagnosed at birth, was followed up by the neurosurgery service for a large ASM associated with neurogenic bladder and recurrent urinary tract infections. As the ASM was initially considered asymptomatic, the patient was followed up serially until a urinary tract infection progressed to the extent of sepsis that required hospitalization and admission to the intensive care unit. The size of the meningocele progressed, obstructing both ureters and causing hydronephrosis and acute renal failure, for which ureteral stents were placed by cystoscopy. Serum creatinine levels had increased to 150–180 μmol/L (1.7–2.0 mg/dL) (normal 50–115 μmol/L or 0.6–1.3 mg/dL).

On examination, he had spastic limbs bilaterally; the strength of the distal muscle groups was normal except he had reduced left plantar flexion resulting from a prior foot surgery. A large mass was both palpable and visible in the midabdominal area. MRI of the lumbar spine and abdomen demonstrated an expansive ASM measuring 25 × 12 × 18 cm, grossly displacing the abdominal structures (Fig. 1).
Operation

The primary surgical objective was the closure of the communication between the meningocele and subarachnoid space. This involved direct visualization of the anomaly, aspiration of the meningocele contents, spinal cord detethering, meningocele pedicle ligation, and watertight closure.22

Repair of the meningocele was first attempted through a sacral laminectomy. Operative findings revealed that the spinal cord was tethered anteriorly at the sacrum with nerve tissue extending into the abdominal portion of the meningocele (confirmed by intraoperative stimulation). With regard to this, it was felt the risks of neurological deterioration were too great with a posterior approach alone. Thus, the procedure was aborted and the meningocele would be approached anteriorly in a second operative procedure after discussion with the patient.

One week later, in collaboration with the general surgeon, the patient was brought back to the operating room and the meningocele was approached via a midline transabdominal approach. After dissection of the ASM from the surrounding abdominal structures, the retroperitoneum was opened to reach the base of the ASM at the sacrum. The ASM was opened to dissect the adherent nerve tissue from the dura mater. Watertight closure of the pedicle was achieved with a 45-mm articulating endoscopic linear stapler/cutter (Endopath ETS Compact-Flex 45, Ethicon) with 2.5-mm-height staples and 6 staple rows. Despite the absence of leakage, the staple line was reinforced with additional suture line and fibrin glue (Fig. 2).

Postoperative Course

The postoperative course was uneventful and creatinine levels rapidly dropped to 115–128 μmol/L (1.3–1.4 mg/dL). The patient has been followed up for a period of 9 months with improvement of urinary symptoms and stable MRI findings (Fig. 3); his leg spasticity, however, has remained unchanged.

Discussion

The pathogenesis of ASMs is thought to be one of a congenital neural tube defect arising during embryonic development at the stage of neurulation with a defect in the anterior sacral wall. Meningocele development is explained by CSF pulsation eroding the weakened sacral wall and leading to out-pouching.18,22 ASM formation has
FIG. 3. Postoperative sagittal (A) and axial (B) T2-weighted images of the ASM showing net size reduction from preoperative imaging, and showing stability 8 months postoperatively (C and D).

also been described in the context of minor trauma to the anterior sacrum, through the same proposed mechanism of CSF pulsation and subsequent erosion.9 Otherwise, some clinical conditions such as Currarino’s triad and Marfan’s have been associated with ASMs. In the present report, our patient did not have any of these associated conditions.10,22

The presentation of ASMs is highly variable; it is believed that the true incidence is underestimated due to their relatively asymptomatic nature. Approximately 300 cases of ASM have been described in the literature.7 The diagnosis is incidental in the context of gynecological investigations or pregnancy in otherwise asymptomatic patients.6,10,27 When present, symptoms are mostly related to the mass effects over surrounding organs in the pelvis. The local pressure on the rectum, urinary bladder, female genital organs, and sacral nerve roots produces a predictable array of symptoms—chronic constipation,1,14,19 urinary incontinence, dysmenorrhea, and chronic back pain,19 among others, are all well recognized. In extreme cases, fistulization between the meningocele and intestines can occur.9 Indeed, ASMs may present with bacterial meningitis, presumably due to a communication between the CSF and an enteric structure.24 Finally, death is also a dreaded complication of ASM, since rare cases of fatal spontaneous rupture have been reported during labor.27

Management of ASMs reported in the literature has been both surgical and observational. It has been acknowledged that conservative management is an appropriate option for small, uncomplicated ASMs, without associated tumor or in the context of pregnancy. However, spontaneous regression of ASMs has not been observed and life-threatening complications such as fistulization, meningitis, and rupture have been described.5,6,21,22 Conversely, a surgical approach is indicated in the case of symptomatic compression of pelvic structures or fistulization. In the present case, obstructive renal failure and hydronephrosis were related to the mass effect of the ASM, and it was felt that the obstructive renal failure would continue to evolve without surgical intervention. Various surgical approaches have been proposed for the treatment of symptomatic ASMs. Early reports have noted transrectal or transvaginal aspiration of cyst contents. However, these approaches were associated with infectious complications, early recurrence, and death.18 Various different conventional surgical approaches exist for the treatment of ASMs, but decisions are made on a case-by-case basis. Principal factors that influence surgical approach include size of the ASM, size of the ostium, and presence of tumors.25 The anterior transabdominal approach was first introduced as a means of obtaining direct access to the ASM; however, surgical risks of trauma to adjacent structures and high rates of complications due to incomplete “watertight” dural closure have limited acceptance of this approach. However, this technique is highly applicable in the case of large ASMs or large ostium, and with careful dissection of visceral structures, risks can be minimized.2,22,25 The posterior approach, in which sacral laminectomy and ligation of the neck of the meningocele are performed, was introduced in 1938 and was associated with fewer complications than the anterior operation.11,22 In this approach, the goal is to close the communication between the subarachnoid space and meningocele, and spontaneous regression occurs in most cases.2,22 The posterior sagittal approach has been used frequently in the treatment of pediatric colorectal disorders, and it allows good exposure to the pelvic floor while minimizing potential neurological complications.18 The inferior presacral approach has a high risk of visceral injury and offers poor visualization of neural structures. The obliteration of the ASMs, but as is the case when shunting in other contexts, lumboperitoneal shunts can be employed as an adjuvant treatment in the case of failed primary treatment of ASMs, but as is the case when shunting in other contexts, shunt malfunction is a limitation.4,22

Acute urinary complications related to the direct com-
pression of an anterior sacral meningocele have been rarely reported in the literature.4,7,8,16,17 (Table 1). In the 4 reported cases, symptoms were explained by the mass effect exerted on the bladder, and on initial physical examination, the meningocele could be mistaken for a palpable globe.4,7 Hydronephrosis related to compressive ASM was described in only one other case.7 Various treatment modalities have been reported, but in the majority of circumstances, surgical intervention was undertaken with resolution of the urinary symptoms. In only one case, described by Lefere and colleagues, an ASM recurred after surgical management, and symptoms resolved once a lumbar peritoneal shunt was placed.17 In these cases, there were no patients with genetic syndromes.

In the present case, obstructive renal failure and hydronephrosis were related to the mass effect of the ASM, and it was felt that the symptoms would continue to evolve in the absence of surgical intervention. We proceeded with a dorsal transsacral approach, but after the sacral laminectomy was done, we noted that the cord and the nerve tissue had extended beyond the level of the sacrum and into the intraabdominal part of the ASM. Based on evaluation of the imaging, it would have been appropriate to proceed primarily with the transabdominal approach; however, given the advantages of the transsacral approach, we felt it offered advantages. As we continued in the transabdominal approach, we noted, after opening of the ASM, that the neural tissue had migrated to the lower third of the bladder, and on initial physical examination, the meningocele could be mistaken for a palpable globe.4

The endoscopic articulating cutting stapler was used to address the neural tissue had migrated to the lower third of the bladder, and on initial physical examination, the meningocele could be mistaken for a palpable globe.4

This device allowed for watertight closure in a time-efficient manner (Fig. 2). With further reinforcement of the staple line with additional sutures and fibrin glue, this closure method, we believe, offers advantages in the closure of the meningocele pedicle and removal of the redundant tissue. The use of gastrointestinal anastomosis devices has been previously suggested as a successful means of ensuring watertight dural closure in a time-efficient manner.3,12 However, we highlight the advantages of an endoscopic articulating stapler for closing dural pedicles in difficultly accessible situations and with complex conformation.

This report presents an important limitation. Indeed the follow-up duration was limited (9 months), and the slowly progressive nature of this disease does not prevent late recurrence. A close and prolonged monitoring is then highly suitable. As reported by Lefere and colleagues, a shunt could be proposed in such a situation. This treatment option has not been proposed as first line given the size of the lesion and the severity of compression, which favored an open surgery to obtain a rapid and significant reduction of the mass effect.

However, this case is remarkable in many aspects. This is only the second reported case of hydronephrosis due to a giant ASM. It also highlights the difficulty of the decision making, with the need to preserve functional neurological structures to protect the patient’s neurological status. Finally, it demonstrates the value of a novel dural closure technique that provided effective watertight closure, making the anterior transperitoneal approach safer.

**References**

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Disclosures
Dr. Daniel Shedid reports being a consultant for DePuy Synthes and receiving fellowship support from Medtronic and DePuy Synthes. Dr. Westwick reports being a consultant for DePuy Synthes and receiving fellowship support from DePuy Synthes.

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Conception and design: Shedid. Acquisition of data: Sunna, Westwick, Berania. Analysis and interpretation of data: Westwick, Zairi, Berania. Drafting the article: Sunna, Westwick, Berania. Critically revising the article: Shedid, Sunna, Westwick, Zairi. Reviewed submitted version of manuscript: Shedid, Sunna, Westwick, Zairi. Approved the final version of the manuscript on behalf of all authors: Shedid. Study supervision: Shedid.

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