Radiology Case Reports 14 (2019) 112–115

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

Rectothecal fistula complicating anterior sacral meningocele repair

Patricia Ojeda, MD, Derek Khorsand, MD, Mazen Zawaideh, MD, Orpheus Kolokythas, MD*

Department of Radiology, University of Washington, 1959 NE Pacific St, Seattle, WA 98195, USA

ARTICLE INFO

Article history:
Received 3 September 2018
Revised 7 September 2018
Accepted 14 September 2018
Available online 26 October 2018

Keywords:
Iatrogenic rectothecal fistula
Meningocele
Sacral agenesis

ABSTRACT

We present a case of an iatrogenic rectothecal fistula in a 34-year-old man who underwent repair of a congenital anterior sacral meningocele, intraoperatively complicated by rectal perforation. Postoperatively, the patient developed symptoms of meningitis prompting concern for the cerebrospinal fluid leak. Subsequent workup with computed tomography (CT) and magnetic resonance imaging demonstrated a postoperative pseudomeningocele and fistulization with an abdominal fluid collection. CT myelography confirmed the fistulous connection was between the pseudomeningocele and the rectum. Clinical suspicion of a rectothecal communication should be elevated for patients who undergo anterior sacral meningocele repair and postoperatively develop symptoms concerning for meningitis. We suggest that CT myelography be considered in the evaluation of viscero-thecal fistulas if clinical or other initial radiologic evaluation suggests the possibility of this diagnosis.

Case report

A 34-year-old man with chronic back pain developed severe, left lower quadrant abdominal pain with focal swelling, prompting presentation to an outside institution. During his work-up there, imaging demonstrated sacral agenesis and an anterior sacral meningocele (ASM), and the patient was referred to our institution for neurosurgical repair of this congenital anomaly. Physical examination at the time of presentation at our institution was unremarkable, in particular the patient demonstrated no neurologic abnormalities and no abdominal distension or tenderness. The patient had a history of exploratory laparotomy 11 years prior due to stab wounds; however, there were reportedly no intraperitoneal injuries noted.

At our institution, the surgeons opted for an anterior approach repair through the patient's prior laparotomy site. The surgery was complicated by bowel adherence to the ASM and unintentional rectal laceration during adhesion take-down. The meningocele was repaired as originally planned followed by a diverting sigmoid colostomy to avoid infectious sequela from the injured rectum. The patient was discharged home on postoperative day 10 with plans for colostomy take-down in subsequent weeks. On postoperative day 14, the patient presented to the emergency department with severe headache,

Competing Interests: The authors have declared that no competing interests exist.

* Corresponding author.
E-mail addresses: pojeda@uw.edu (P. Ojeda), dkhor@uw.edu (D. Khorsand), mazen1@uw.edu (M. Zawaideh), orpheus@uw.edu (O. Kolokythas).

https://doi.org/10.1016/j.radcr.2018.09.011

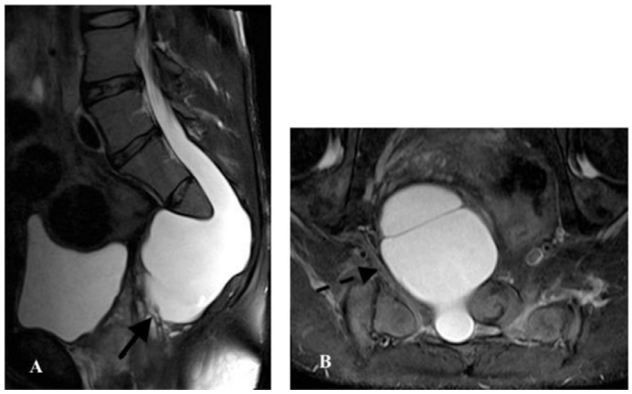

Fig. 1 – T2 fat-saturated MRI obtained at presentation. (A) Sagittal image shows a sacral defect through which a T2 hyperintense sac (solid arrow) extends from the spinal column into the posterior pelvis with a simple septation representing a meningocele. (B) Axial image shows intact meningocele wall (dotted arrow) without evidence of rupture or fistula.

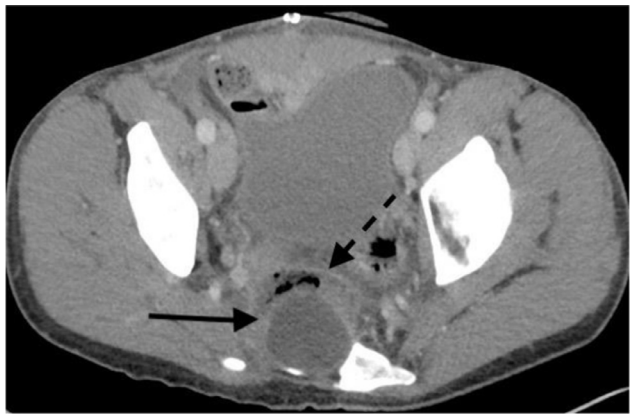

Fig. 2 – Axial CT pelvis with contrast obtained after patient reports severe headache on postoperative day 14 status postanterior meningocele repair. Image shows residual pseudomeningocele at the sacral deformity (solid arrow) with direct contact and mass effect on the anteriorly located rectum (dotted arrow).

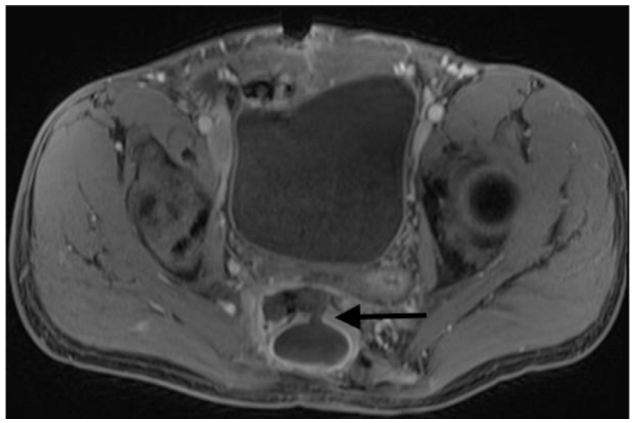

Fig. 3 – Axial fat suppressed T1 weighted gradient echo image with intravenous gadolinium of the pelvis obtained on postoperative day 19 after patient showed no signs of improvement after antibiotic treatment: image shows communication between the pseudomeningocele and an anterior fluid collection (arrow). No other fistula was identified.

nausea, and vomiting, with physical exam significant only for somnolence and photophobia. Laboratory evaluation showed an elevated WBC count of 13.08 thousand/μL (normal 4.3-10.0), elevated the cerebrospinal fluid (CSF) protein of 136 mg/dL (normal 15-45) and elevated CSF white blood cells 3915 cells/μL (normal 0-5) with normal CSF glucose levels. His presentation and lab results were concerning for bacterial meningitis. Given his recent surgery, CT abdomen/pelvis with IV contrast was performed which demonstrated a pseudomeningocele—an ectatic thecal sac forming from postoperative complication at the site of repair—which likely developed due to CSF leakage from the original ASM. The study also demonstrated a possible abscess anterior to this thecal outpouching (Fig. 1). A lumbar spine MRI was then performed due to concern for CSF leak (Fig. 2). This was not identified, however, the collection anterior to the pseudomeningocele was again noted to be suspicious for abscess. Antibiotics were empirically started for suspected meningitis. Although culture of cerebrospinal fluid showed no bacterial growth after several days, the patient failed to symptomatically improve on antibiosis, necessitating further work-up.

An MRI of the pelvis was performed on postoperative day 19, showing a fistulous connection between the pseudomeningocele and an anterior fluid collection (Fig. 3). Discussion ensued as to the most appropriate modality to evaluate for clinically suspected rectothecal fistula, with consideration of a barium enema, however, CT myelography was ultimately decided upon. On postoperative day 20, a lumbar spine CT myelogram unequivocally demonstrated a pseudomeningocele fistulization to the rectum (Fig. 4). The next day, a multidisciplinary team of surgeons intraoperatively identified the fistula at the site of the prior rectal tear and turbid fluid anterior to the pseudomeningocele. The defect was subsequently repaired and a rectus muscle flap was placed anterior to the sacrum for dural coverage. Follow-up CT lumbar myelogram 4 days following repair showed no evidence of the large fistulous tract.

Discussion

ASM is a rare congenital abnormality of dural herniation through an anterior sacral defect resulting in a sac of meninges and cerebrospinal fluid protruding into the presacral extraperitoneal space [1,2]. CSF pressure is hypothesized to cause dural ectasia at the site of an osseous defect and result in sac dilatation. The defect has a female predilection

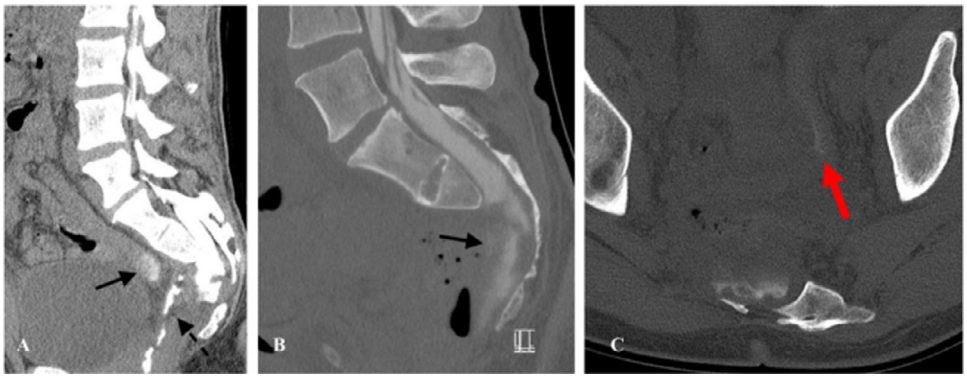

Fig. 4 – Lumbar spine CT myelogram obtained on postoperative day 20 due to clinical concern for suspected rectothecal fistula. (A) Sagittal reconstruction soft tissue window shows communication between the spinal column and rectum (dotted arrow) with contrast extending into the sigmoid colon (solid arrow). (B) Sagittal reconstruction bone window shows intrathecal contrast in the pseudomeningocele and extending anteriorly into the rectum (solid arrow). (C) Axial image shows residual contrast within sigmoid (thick arrow) and pseudomeningocele. Findings confirm rectothecal fistula.

and is oftentimes an incidental finding during pelvic or rectal examinations. Anterior meningoceles are usually asymptomatic; however, if symptoms develop, they are often due to mass effect on adjacent structures causing obstetrical complications, urinary incontinence, decreased rectal tone, perineal hyperalgesia, or sciatica, among others [1,3-5]. If patients acquire symptoms associated with the ASM, they usually do not develop until the third decade of life. ASM is more difficult to diagnose relative to posterior meningoceles, which can often be visualized on external examination. Thus, high clinical suspicion with corresponding symptomatology is necessary to prompt imaging evaluation and diagnosis of an ASM [6]. An ASM may be seen in conjunction with Curriano syndrome [7], characterized by the triad of ASM, partial sacral agenesis, and anorectal malformation.

Surgical intervention is the treatment of choice for symptomatic or rapidly growing meningoceles as they will not naturally regress. Occasionally, surgical repair for asymptomatic maternal ASMs is indicated if obstetrical complications are anticipated during pregnancy and childbirth. Repair involves aspiration of the meningocele and closure of the stalk to the thecal sac. Anterior or posterior approaches may be utilized for repair, the latter of which is associated with less complications but may only be used in uncomplicated cases as direct evaluation of the anterior visceral structures adjacent to the ASM is limited [1,3].

Rectothecal fistulas are extremely rare, with only 10 prior cases reported in the literature to-date. Although the etiology is unclear, it is hypothesized that spontaneous fistulas arise from fecal impaction and subsequent perforation of the intestinal wall [3,8]. These patients often present with meningitis due to enteric contamination and frequently undergo emergent surgical intervention after diagnosis [3-5]. Of the reported cases, we did not find any iatrogenic rectothecal fistulas as demonstrated in this case.

Several imaging modalities for the diagnosis of an ASM have been documented—intracavitary ultrasound for thecal sac identification, radiography for assessment of associated sacral dysgenesis, and CT for demonstration of the actual meningocele and its extent. However, MRI is regarded as the best modality due to superior delineation of soft tissue planes and clear visualization of the spinal canal and contents, thus facilitating superior surgical planning [3,9,10].

However, when determining the best imaging modality for evaluation of suspected rectothecal fistulas in the setting of ASM, although MRI provides better assessment of associated abscesses, CT myelography should be considered the gold standard for diagnosis. By facilitating direct visualization of the fistulous course, CT myelography enables succinct surgical planning and assessment of the integrity of surrounding neural anatomy. Although in the case of suspected rectothecal fistula, barium enema could be considered, as was the case for our patient, this technique poses the theoretical risk of facilitating retrograde enteric contamination of the thecal space and should be avoided unless there is an absolute contraindication to myelography. Due to this advantage of direct visualization of the cord and meninges, the American College of Radiology and American Society of Neuroradiology retain practice guidelines in which myelography is indicated for suspected CSF leak, particularly in the setting of postspinal surgery headache, and for diagnostic evaluation of spinal disease [11].

Clinical suspicion of a visceral-thecal communication should be elevated for patients who undergo ASM repair and postoperatively develop symptoms concerning for meningitis. We suggest that CT myelography be considered in the evaluation of viscero-thecal fistulas if clinical or other initial radiologic evaluation suggests the possibility of this diagnosis.

REFERENCES

[1] Ashley WW Jr, Wright NM. Resection of a giant anterior sacral meningocele via an anterior approach: case report and review of literature. Surg Neurol 2006;66(1):89–93 discussion 93.

[2] Villarejo F. Anterior sacral meningocele: review of the literature. Surg Neurol 1983;19(1):57–71.

[3] Phillips JT. 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(10):1633–5.
[4] Sanchez AA. Rectothecal fistula secondary to an anterior sacral meningocele. J Neurosurg Spine 2008;8(5):487–9.
[5] Koksal A. 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(7):528–31.
[6] Calihan RJ. Anterior sacral meningocele. Radiology 1952;58(1):104–8.
[7] Currarino G, Coln D, Votteler T. Triad of anorectal, sacral, and presacral anomalies. Am J Roentgenol 1981;137(2):395–8.
[8] Bergeron E. A 40-year-old woman with cauda equina syndrome caused by rectothecal fistula arising from an anterior sacral meningocele. Neurosurgery 2010;67(5):E1464–7 discussion E1467-8.
[9] Fitzpatrick MO, Taylor WA. Anterior sacral meningocele associated with a rectal fistula. Case report and review of the literature. J Neurosurg 1999;91(Suppl. 1):124–7.
[10] Lee KS. The role of MR imaging in the diagnosis and treatment of anterior sacral meningocele. Report of two cases. J Neurosurg 1988;69(4):628–31.
[11] American College of Radiology website. ACR-ASNR practice guideline for the performance of myelography and cisternography. 2008.