



Anterior sacral meningocele presenting as acute urinary retention A case report

Mathieu LEFERE, Norbert VERLEYEN, Hans FEYS, Jan F. A. SOMERS

From Jan Yperman Hospital, Ypres, Belgium

Anterior sacral meningocele is caused by a congenital hiatus in the anterior sacrum. We report a very rare case which presented as acute urinary retention. The common findings of anterior sacral meningocele include atypical low back pain, urological and gynaecological symptoms. Acute urinary retention as a presenting symptom does not appear to have been mentioned in the English literature.

Keywords : anterior sacral meningocele ; Currarino syndrome ; acute urinary retention.

INTRODUCTION

Anterior sacral meningocele (ASM) was first described in 1837. The entity consists of a congenital hiatus in the anterior sacrum, through which the meninges (dura and arachnoidea mater) gradually herniate. This movement is thought to be driven by cerebrospinal fluid pulsations (1). The onset of symptoms is usually in early adult life, when the mass has grown large enough to exert pressure on nearby organs, such as the bladder, rectum and uterus. Usually symptoms (orthopaedic, gynaecological, abdominal, urological or neurological) appear gradually and are chronic in nature. To our knowledge, this is the first case report in the English literature where an ASM presents as an acute urinary retention in a seemingly healthy young male adult.

CASE REPORT

A 22-year-old male patient presented to our emergency department with lower abdominal pain. He had been unable to pass urine since the night before. Physical examination revealed a large suprapubic mass. Its estimated volume on echographic investigation was more than one liter. This acute urinary retention was treated by way of transurethral catheterisation, after which the patient was discharged from the hospital. Further follow-up was planned on an ambulatory basis. Urological evaluation consisted of urinary flow measurement. The maximum flow rate was far lower than expected in a patient of his age. We found a large volume on postmicturitional echographic evaluation of the bladder, along with a thickened bladder wall. The

-
- Mathieu Lefere, 6th-year medical student.
Catholic University Leuven, Leuven, Belgium.
 - Norbert Verleyen, MD, Consultant Urology Surgeon.
Department of Surgery, Jan Yperman Hospital, Ypres, Belgium.
 - Hans Feys, MD, Consultant Orthopaedic Surgeon.
 - Jan F. A. Somers, MD, Consultant Orthopaedic Surgeon.
Department of Orthopaedic Surgery, Jan Yperman Hospital, Ypres, Belgium.
- Correspondence : Dr. Mathieu Lefere, Zandvoordestraat 2b, 7780 Komen, Belgium.
E-mail : mathieu_lefere@hotmail.com
© 2009, Acta Orthopædica Belgica.
-

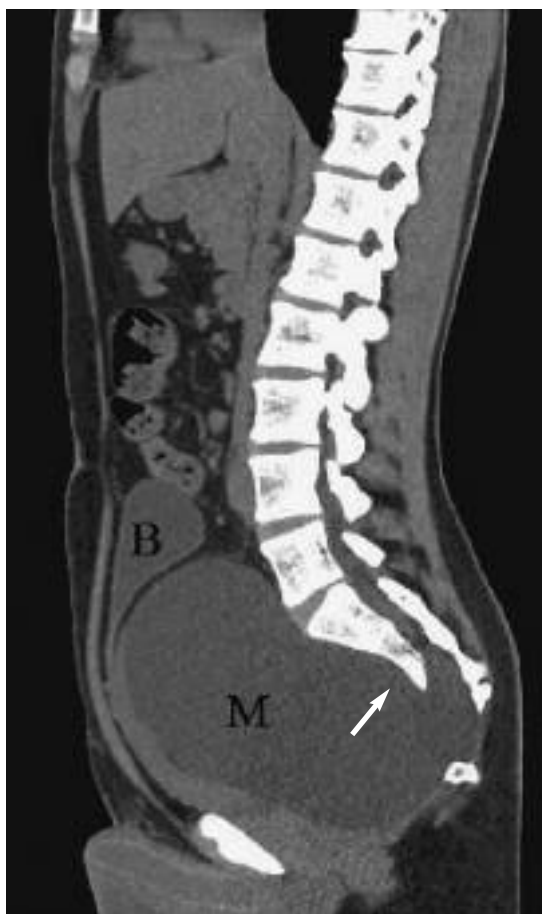


Fig. 1. — CT-scan of the abdomen and pelvis, showing a large pelvic mass (M) in direct communication with the spinal canal through a defect in the anterior sacrum (arrow). The bladder (B) is pushed anteriorly.

patient was then referred for a computerized tomographic (CT) scan of the abdomen and pelvis (fig 1). The CT image showed a large pelvic mass (largest diameter of 17 cm), in direct communication with the spinal canal through a defect in the anterior sacrum. Bladder and rectum, both pushed aside by the mass, appeared normal. Magnetic Resonance Imaging (MRI) of the same region was compatible with an anterior sacral meningocele. Definitive treatment consisted of closure of the meningocele sac and the sacral defect by a muscle flap. Despite this surgical attempt a recurrence of the cystic structure was noted, along with bladder compression. A symptomatic relief was finally provided with the insertion of a peritoneal shunt.

DISCUSSION

No more than about 200 cases of ASM have been reported since it was first described in 1837. The entity consists of a congenital hiatus in the anterior sacrum, through which the meninges (dura and arachnoidea) gradually herniate. This movement is thought to be driven by cerebrospinal fluid pulsations (1). The onset of symptoms is usually in early adult life, when the mass has grown large enough to exert pressure on nearby organs, such as the bladder, rectum and uterus. In our patient, the pressure on the bladder neck had risen to such a level as to make voiding impossible. Other presenting symptoms reported in previous cases included back pain, acute meningitis (6), chronic constipation (4), recurrent headaches and obstetrical problems (5). Other urologic symptoms may include urinary incontinence or urgency. Upon clinical gynaecological examination of female patients, the meningocele can be mistaken for an ovarian cyst (2). ASM is sometimes part of a hereditary syndrome called the Currarino triad. This combination of a presacral mass, anorectal malformation and hemisacrum was described by Currarino *et al* in 1981 (3). It may also be related to Marfan's syndrome (7). Careful analysis of the paediatric medical files of the patient revealed a tendency to anal atresia in the postnatal period, for which dilations were performed. We could therefore consider this a Currarino syndrome, although there were presently no signs of permanent anorectal malformation. To our knowledge, this is the first case report in the English literature where an ASM presents as acute urinary retention in a seemingly healthy young male adult. It shows the importance of thorough further investigation in young patients presenting with these symptoms. Our patient might have developed an acute meningitis, had a suprapubic puncture been performed as treatment of the urinary retention in the emergency setting.

REFERENCES

1. Ashley WW, Wright NM. Resection of a giant anterior sacral meningocele via an anterior approach : case report and review of the literature. *Surg Neurology* 2006 ; 66 : 89-93.

2. **Benes V, Barsa P, Plny R, Suchomel P.** Anterior sacral meningocele misdiagnosed for an ovarian cyst. *Eur J Obstet Gynecol Reprod Biol* 2008 ; 138 : 249-250.
3. **Currarino G, Coln D, Votteler T.** Triad of anorectal, sacral, and presacral anomaly. *AJR Am J Roentgenol* 1981 ; 137 : 395-398.
4. **Gardner PA, Albright AL.** "Like mother, like son": hereditary anterior sacral meningocele. *J Neurosurg* 2006 ; 104 : 138-142.
5. **Manson F, Comalli-Dillon K, Moriaux A.** Anterior sacral meningocele : management in gynecological practice. *Ultrasound Obstet Gynecol* 2007 ; 30 : 893-896.
6. **Philips 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 : 1633-1635.
7. **Raftopoulos C, Delecluse F, Braude P et al.** Anterior sacral meningocele and Marfan syndrome : a review. *Acta Chir Belg* 1993 ; 93 : 1-7.