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CASE REPORT

An elusive pelvic cyst: a case report of an anterior sacral meningocele

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Abstract We present an interesting case of a large anterior sacral meningocele. Despite its rarity, this anomaly can have serious complications and therefore should not be overlooked. A female presenting with a cystic presacral mass requires detailed imaging and a complete diagnostic workup prior to definitive treatment.

Keywords Laparoscopy · Pelvic cyst

Introduction

Anterior sacral meningocele (ASM) is a congenital lesion resulting from a herniation through a defect on the anterior surface of the sacral bone [1]. It is a rare anomaly and only about 200 cases have been reported in the literature since its first description in 2003 [2]. The rarity of this lesion means that it is often overlooked in the possible differential diagnoses of women undergoing surgery for pelvic cysts.

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Case report

A 37-year-old Para 2 Jehovah's Witness presented with a worsening history of chronic pelvic pain associated with dysmenorrhea, menorraghia, dyspareunia, shooting back pain and ultrasonic evidence of a right pelvic cyst. The patient had a background history of endometriosis and an ovarian cyst diagnosed at laparotomy at which time a cystectomy was performed. Two years later at 27 years of age, she underwent a laparoscopy to investigate recurrence of her pelvic pain and an ultrasound that had reported a 6-cm pelvic cyst. Endometriosis was again noted intra-operatively, but no cyst was found. At 28 years of age, she again underwent a laparoscopy for pelvic pain and the persistent ultrasound finding of a 6-cm pelvic cyst. Once more mild endometriosis was seen but the pelvic cyst remained elusive. The patient had delivered two children between her last laparoscopy and this presentation, one vaginally and one by caesarean section. No pelvic cyst was noted at the caesarean section.

In this presentation, a pelvic ultrasound was again performed, which indicated a pelvic cyst measuring approximately 8×5×7 cm, separate from the ovary. As medical therapy for her endometriosis was no longer effective in controlling her symptoms, a laparoscopy was scheduled to excise any possible endometriosis and perform a pelvic cystectomy.

A pelvic cyst had now been documented on ultrasound for at least 8 years despite failure to find this in three out of four of her previous operations. This cyst also now appeared to be enlarging. Tumour markers cancer antigen 125 (CA 125) and cancer antigen 19-9 (CA 19-9) were tested for pre-operatively and found to be within normal limits.

At laparoscopy there was some mild peritoneal endometriosis, which was excised. On initial inspection of the pelvis, no cyst was immediately obvious. Thorough

examination of the abdominal cavity and viscera were normal, and no mesenteric cyst was found. This had been thought to be a possible differential diagnosis preoperatively. On closer inspection on the right side of pelvis, there was a subtle fullness on the right pelvic sidewall, lateral to the uterosacral ligament and ventral to the hollow of scrum. In view of the patient being a Jehova's Witness, plus the fact that this 8-cm cyst would be in close proximity to deep pelvic vasculature, a vascular surgeon was consulted with regards to the best approach to this lesion. The vascular surgeon suggested no surgical intervention. In view of the possibility of this cyst being responsible for the patient's chronic symptoms and the fact that it now seemed to be enlarging, compounded by the potential to avoid further pelvic surgery through intervention, a decision was made to explore the cyst. Careful dissection was carried out. The fibrotic capsule was encountered and was not pulsatile. Aware of the large size of the cyst, and the close proximity to deep pelvic vessels, it was decided to de-roof, biopsy and marsupialise the cyst to prevent recollection. A drain was left in the pelvis. The cyst was marsupialised to the right uterosacral ligament with a non-absorbable suture (0 nylon). The cyst on laparoscopic inspection appeared simple; it contained clear fluid, had a smooth wall and single small septum with no vegetations. The histological report on the cyst-wall biopsy was of non-specific fibrous connective tissue with no evidence of malignancy.

The drain was removed on the first day post-operatively. The patient did describe some intermittent headaches in the peri-operative period, but despite these, opted for discharge on day 3. During the 2 weeks post-discharge, the patient's headaches became progressively worse. These were exacerbated by standing and alleviated by bed rest, and were not associated with fever or photophobia. She eventually attended the emergency department 14 days post-operatively for further investigation, and a neurological opinion was sought. There were no abnormal neurological findings on examination, and the patient was apyrexial. Blood tests including full blood count, urea/electrolytes/creatinine, and liver function tests were all normal.

In view of the severe postural headache, the patient was admitted and magnetic resonance imaging (MRI) organised. The MRI showed meningeal enhancement consistent with low pressure headache as well as low-lying cerebellar tonsils. Findings were consistent with cerebral hypotension. The patient was subsequently managed with strict bed rest, intravenous rehydration, caffeine and anti-thrombotic stockings to prevent deep vein thrombosis prophylaxis. In view of the symptoms of intracranial hypotension and radiological findings, the possibility that the pelvic cyst may have been of meningeal origin was suspected and a neurosurgical consultation requested. The neurosurgical team reviewed the patient and found her to be neurologically intact with no cutaneous stigmata of occult spinal dysphraphism. A pelvic computer tomography scan (CT) with both intravenous and oral contrast was ordered. The CT revealed a right-sided anterior sacral meningocele originating from the S3 neural exit foramen. It measured 4.5×4.5×3 cm. There was also an associated sacral deformity and hypoplasia of the coccyx (Figs. 1 and 2). In order to plan the neurosurgical approach, further imaging of her whole spine was performed to rule out any additional abnormalities (Fig. 3).

The neurosurgeon discussed the diagnosis and surgical approach comprehensively with the patient, who subsequently underwent a sacral laminectomy and repair of the anterior sacral meningocele. Intraoperatively, the caudal end of the dural sac was found to communicate with the large meningocele. Post-operatively, the patient was transferred to high dependency unit (HDU). She was nursed in the prone position for several days with a lumbar drain set to encourage draining 10 ml/h of CSF. She was also given intravenous antibiotics (keflin and gentamicin). The patient was discharged from the HDU on day 5 after the operation and went home the next day. Prior to discharge, she was counseled regarding the possibility of the hereditary nature of ASM [1] and screening of her child was recommended. At 6 weeks post-operatively, she had made an excellent recovery with only a small area of numbness on the left side of her buttock.

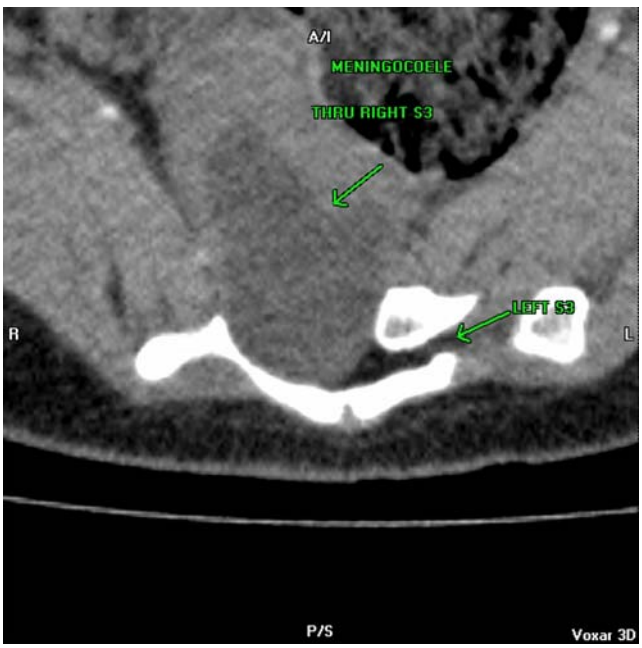

Fig. 1 Axial computed tomography through S3 level

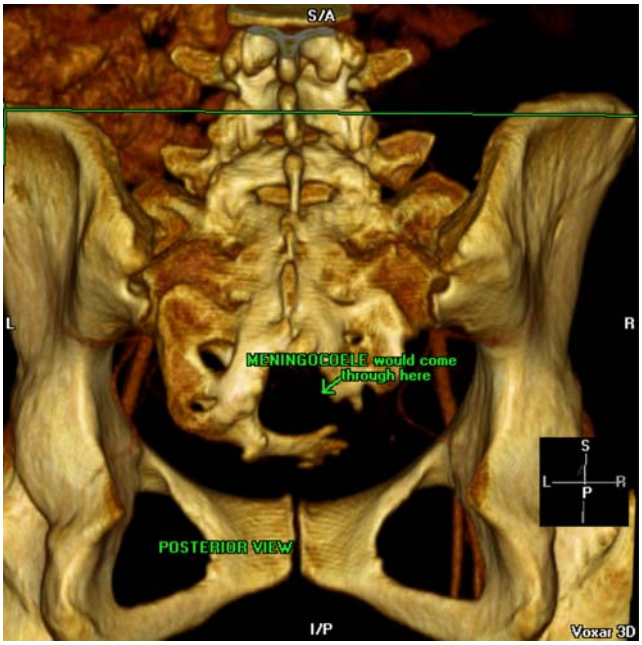

Fig. 2 Computer tomography 3D reconstruction of posterior view of sacral spine

Discussion

ASM is a rare congenital disorder. The first case report of an ASM was published in 1837, which resulted in an obstructed labour during childbirth and the subsequent death of the mother [3]. It seems that this condition is linked to gender with about 85% of the affected individuals being female [4]. However, some authors argue that the increased prevalence in females may be due to an increase in the recognition of this disorder, as women undergo regular abdominal and vaginal examination and manifest aggravated symptoms during pregnancy [5].

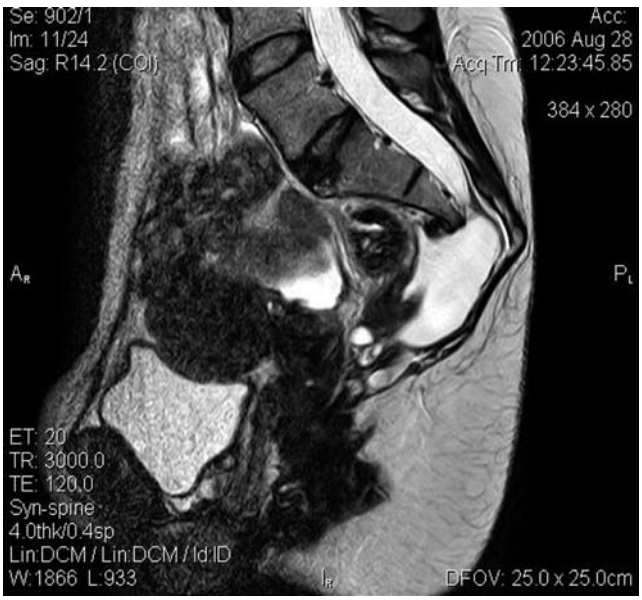

Fig. 3 Sagital T2-weighted MRI showing high-intensity signal from sacral meningocele at base of spinal cord

In ASM, the defect results from an abnormal formation of the caudal cell mass. Therefore patients affected by ASM often have other associated urogenital and rectal abnormalities [5]. It is sometimes associated with syndromes such as Currarino [6] and Marfan [7]. In those patients without any urogenital or rectal abnormalities or symptoms, ASM can be difficult to recognize. Most patients present with symptoms due to pressure on the surrounding organs including the bladder, uterus, rectum and sacral nerve roots. With age, the cyst tends to increase in size, producing symptoms such as constipation, urinary retention or frequency, and abdominal pain. In women, ASM can lead to birth dystocia [8]. Patients can also present with recurrent meningitis; headache during strain, defecation, and coitus [1] and low-pressure headache in response to a change in posture; or low-back pain [5].

It is extremely important to differentiate ASM from other presacral cysts. Before the 1960s, there was a very high surgical mortality reaching 40% due to mistakes in the preoperative diagnosis and limited imaging techniques [1]. As shown in our case report, one must remain vigilant when dealing with a presacral mass and always consider the possibility of ASM. Our patient did not have any of the above symptoms apart from a long-standing history of pelvic and shooting back pain. She also had normal vaginal deliveries after the initial diagosis of the pelvic cyst, so the lesion was not so significant to result in birth dystocia. Furthermore, her history of endometriosis inevitably diverted the surgeon's attention to its relevant pathology. Again this case highlights an important lesson. When a retroperitoneal pelvic cyst is diagnosed or suspected preoperatively, a radiological opinion should be sought and a CT or MRI performed to try and define the most likely underlying pathology.

Unlike the far more common posterior meningoceles, ASM usually presents in female children or young adults rather than in infancy. They are often found incidentally or detected as a soft mass on rectal or pelvic examination [6]. The sacral bone abnormality is often seen on AP radiographs [5]. Ultrasonography of the abdomen or pelvis can detect the meningocele [9]. MRI is currently the imaging modality of choice as it is excellent in displaying spinal and abdominal anatomy. It also reveals other associated abnormalities such as lipoma, dermoid, or other neural tube defects. Furthermore the contents of the meningocele can often be illustrated by an MRI scan [10]. In some cases, including our case, CT imaging may be helpful for further delineation of the relation of nervous structures to complex bony anatomy or for those unable to undergo MRI [11].

A review of the literature shows that surgery is usually indicated for ASMs due to their potential for significant complications [12]. In 1938, Adson introduced the posterior approach for closure of ASM, in which a laminectomy is performed with ligation of the area of communication between the proximal thecal sac and the meningocele [13].

Conclusion

We present an interesting case of a large anterior sacral meningocele. Despite its rarity, this anomaly can have serious complications and therefore should not be overlooked. A female presenting with a cystic presacral mass requires detailed imaging and a complete diagnostic workup prior to definitive treatment.

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