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A Rare Case of Anterior Sacral Meningocele (ASM) in a Young Patient

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Abstract

We report a 24-year-old male who presented with abdominal distension, constipation and left sided groin pain. CT and MRI of the abdomen/spine were performed which showed a large anterior sacral meningocele occupying most of the pelvic and abdominal cavity and displacement of their respective contents. Initially a posterior approach with lumbosacral laminectomy (L5 - S4) was performed. The ostium of the meningocele was identified with several nerve roots identified passing through, adjacent and into the defect. As nerve roots traversed the ostium, watertight closure was not feasible without sacrificing nerve roots. Subsequent MRI demonstrated recurrence of approximately 60% of the anterior sacral meningocele. We therefore opted to approach the ASM anteriorly via an anterior approach with the help of colorectal surgical colleague. The ASM was completely embedded within the sigmoid and upper to mid rectal mesentry, with its own vascular supply to the thick walled capsule. This case highlights the need for a combined approach due to the incorporation of the pseudomeningocele into the omentum with the development of its own blood supply.

Keywords

Spina Bifida, Sacral, Meningocele, Abdominal Distension

1. Introduction

Anterior sacral meningocele (ASM) is an anomaly where the meninges protrude into retroperitoneal and presacral space through an anterior sacral defect. Most of the cases present in adulthood and diagnosis in childhood is rare. Common presentations include infection, meningitis and obstetric problems. We report a 24-year-old male who presented with abdominal distension. This case highlights the need for a combined approach due to the incorporation of the pseudomeningocele into the omentum with the development of its own blood supply.

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2. Case

2.1. Patient History

A 24-year-old male presented with a two-year history of gradually worsening abdominal distension. Acute symptoms were related to constipation and left sided groin pain. He reported no nausea, vomiting or weight loss. He did not have any other gastrological, urological or neurological symptoms or signs and was previously fit and well. Examination revealed generalized abdominal enlargement that was non-tender with bowel sounds present.

2.2. Findings

CT and MRI of the abdomen/spine were performed which showed a large anterior sacral meningocele occupying most of the pelvic and abdominal cavity and displacing their respective contents (**Figure 1**). The meningocele originated from the left anterolateral aspect of sacrum (S2 and S3) with no obvious neural structures passing through the defect on imaging.

CT brain revealed evidence of prosencephaly without hydrocephalus. There was no evidence of chiari malformation.

3. Surgical Approach

Due to the large size of the meningocele and displacement of the abdominal contents, and progressive symptoms, a surgical approach was favoured over conservative management. Initially a posterior approach with lumbosacral laminectomy (L5 - S4) was performed. The dura was opened in the midline and the thickened filum terminale was identified and divided. Thickened meninges were encountered with loculated, complex arachnoid layers surrounding the defect. The ostium of the meningocele was identified with several nerve roots identified passing through, adjacent and into the defect (**Figure 2**). As nerve roots traversed the ostium, watertight closure was not feasible without sacrificing nerve roots. The cerebrospinal fluid (CSF) was aspirated from the meningocele allowing it to collapse. Stay sutures were employed in the extremely thickened pseudomeningocele capsule. The ostium was then closed by laying artificial dural graft (Duragen) over the defect and sutured with 4.0 prolene. Wound was closed in standard fashion.

The patient did not have any post-operative complications, however, subsequent MRI demonstrated recurrence of approximately 60% of the anterior sacral meningocele. We therefore opted to approach the ASM anteriorly via an anterior approach with the help of colorectal surgical colleague. The ASM was completely embedded within the sigmoid and upper to mid rectal mesentry, with its own vascular supply to the thick walled capsule. The colonic and rectal mesentry was skeletonized off the wall of the ASM, with preservation of the colonic vascular supply, allowing resection of the ASM to the presacral space. The sac of the ASM was then opened, and almost all of the sac resected. The small residual sac was closed with a purse string suture (**Figure 3**).

4. Outcome

The patient made full recovery without deficit and was discharged home one-week post operatively. Follow up MRI at three months showed complete resolution of meningocele.

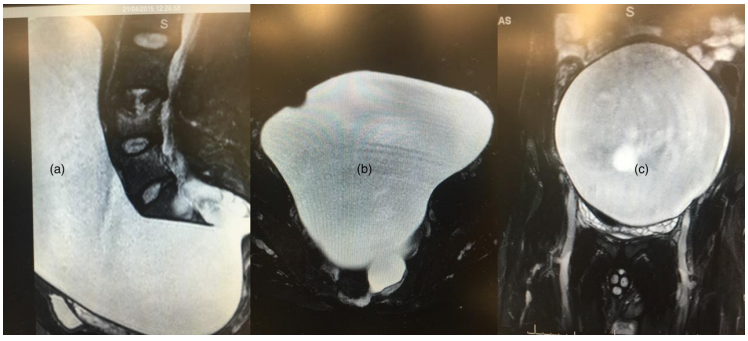

Figure 1. MRI abdomen showing large anterior sacral meningocele in sagittal (a), axial (b) and cornoal (c) orientation.

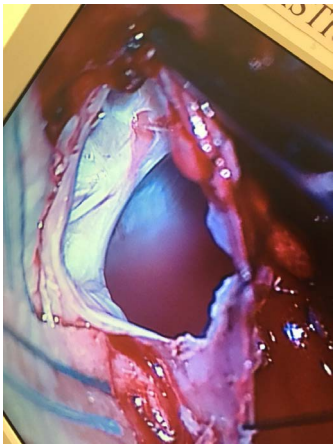

Figure 2. Demonstrating ostium of the anterior sacral meningocele after sacral laminectomy.

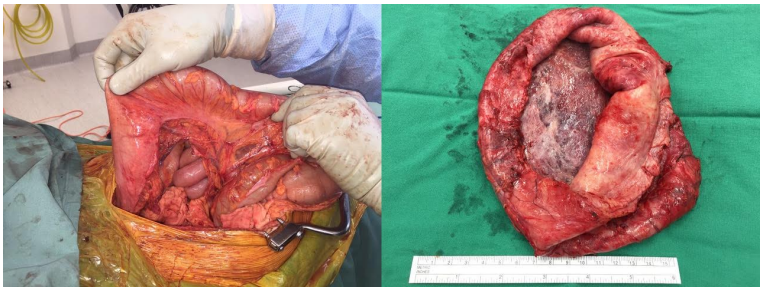

Figure 3. demonstrating meningocele adherent to omentum and the size post resection.

5. Discussion

ASM is a rare congenital anomaly which was first described in 1837 by Bryant [1]. The congenital malformation is usually associated with a sacrococcygeal bony defect in which there is a protrusion of dural sac anteriorly [2]. Fewer than 300 cases of ASM have been reported in literature. It may be associated with a variety of congenital disorders, most frequently the Currarino triad, consisting of congenital canal stenosis, scimitar sacrum and a presacral mass [3]. The onset of symptoms is usually in early adult life secondary to gradually increasing pressure on nearby pelvic structures resulting in urinary, bowel and obstetric problems. It has also been reported to present as cauda equina syndrome [4].

Surgical intervention is almost always necessary as ASM tends to enlarge with time secondary to CSF pulsations and causes progressive symptoms. If left untreated ASM can result in 30% mortality due to recurrent meningitis as well as obstetric and urinary tract complications [5]. There are no reports of spontaneous regression suggesting that the risks are unlikely to subside without treatment.

Early intervention is preferred as the meningocele is likely to be smaller, making surgical treatment easier and also, potentially reducing complications. The favoured approach for treatment of ASM is ligation of the ASM stalk through sacral laminectomy [6]. Plication of the dura or duraplasty with artificial dural graft may be necessary where nerve roots pass through the defect as it makes watertight closure impossible, a feature in about 20% of ASMs. Combination with anterior approach may be considered if the stalk is too wide to be ligated via laminectomy or if watertight closure cannot be accomplished due to ostium containing nerve roots, as performed in this case. Anterior approach may be preferred as a first line approach if there are other associated abdominal lesions, for example a tumour or a fistula that can be dealt with at the same time.

6. Conclusion

This case highlights the need for a combined approach due to the incorporation of the pseudomeningocele into

the omentum with the development of its own blood supply.

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