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Rare Case of Large Anterior Meningocele Associated with Uterine Didelphys

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Abstract

We report a 27-year-old female who presented with abdominal distension, saddle anesthesia, and lower back 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. Anterior approach was chosen, uterine didelphys was identified and mobilized, meningocele identified and gentle drainage with subsequent resection of the cyst wall and watertight closure was achieved. Subsequent MRI demonstrated resolution of most the sacral meningocele. Patient symptoms improved and are doing well.

Keywords

Sacral Agenesis, Curarino Triad, Large Meningocele

1. Introduction

The anterior sacral meningocele is defined as herniation of the meningeal sac due to a developmental bone defect in the front of the sacrum bone [1]. It is reported that an anterior sacral meningocele was first described in 1837 [2] [3]. It may be congenital or acquired. While anterior sacral meningocele usually occurs as a congenital defect, most acquired sacral meningoceles occur due to duralectasia associated with neurofibromatosis, Marfan's syndrome, and Ehlers-Danlos syndrome [4]. The clinical presentation can vary from asymptomatic to the presence of nonspecific symptoms like constipation, dyspareunia, dysmenorrhea, urinary incontinence, urinary retention, dysuria, polyuria, radiculopathy, and/or paresthesia related to genitourinary, neurological, reproductive, or colo-

rectal dysfunction due to mass effect on the abdominal viscera. Most patients present with symptoms that are due to compression of the adjacent organs, and the most common symptom is constipation [5].

In this article, we present the radiological findings of sacral agenesis, large anterior sacral meningocele, and uterine didelphys in the case of afemale patient.

2. Case Report

We report a 27-year-old female who presented with abdominal distension, saddle anesthesia, and lower back pain. We obtained written informed consent for the publication of this case report. 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 (**Figure 1**). Also a dermoid cyst was noted.

We used an anterior approach, whereby uterine didelphys was identified and mobilized, meningocele was exposed and gentle drainage was done, followed by resection of its wall (**Figure 2**). Afterwards watertight closure was achieved. A dermoid cyst was found and excised as well, it contained hair follicles. Layers were closed and patient was doing well postoperatively.

MRI was performed day 3 postoperatively showing significant reduction in size of the meningocele (**Figure 3**). The patient made full recovery and was discharged 1 week postoperatively.

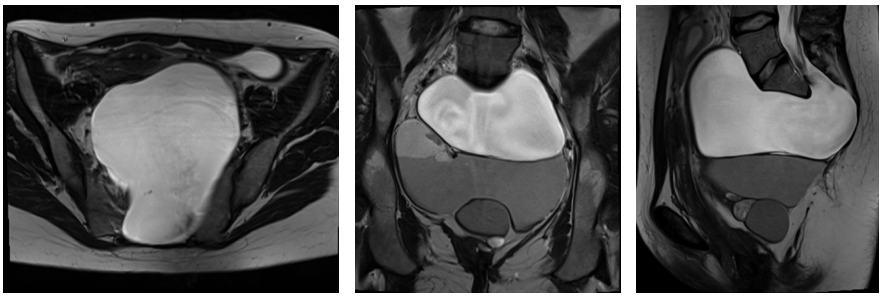
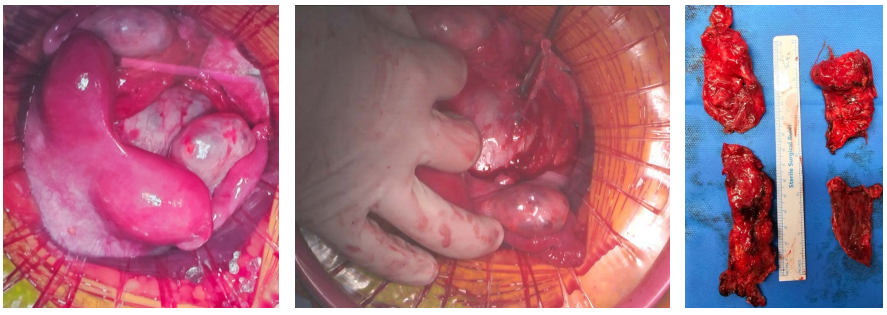


Figure 1. Preoperative MRI imaging, T2 sequence, showing the meningocele in three planes axial, coronal and sagittal (from left to right).



(a) (b) (c)

Figure 2. (a): Intraoperative findings of uterine didelphys and meningocele; (b): Drainage of meningocele; (c): resected walls of meningocele.

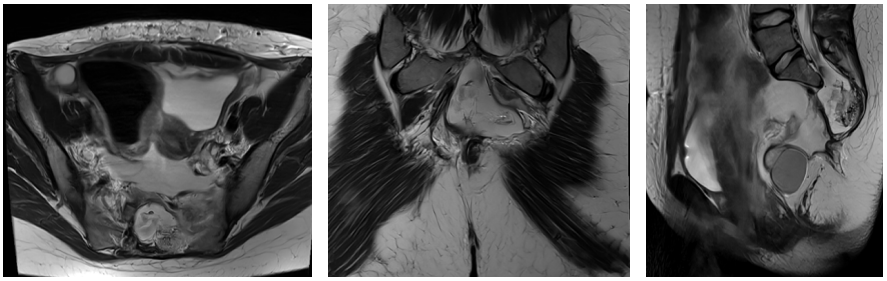


Figure 3. Postoperative MRI MRI imaging, T2 sequence in three planes axial, coronal and sagittal (from left to right).

3. Discussion

The anterior sacral meningocele is due to an anterior sacral defect or a herniation of the meningeal sac. The possible mechanisms for the formation of the sacral meningocele include congenital defects, traumatic, iatrogenic, or degenerative. The congenital form of the condition can vary from minor sacral defects to complete sacral agenesis. The anterior sacral meningocele may be associated with other congenital abnormalities or may be isolated. Anorectal malformations, lipomas, urogenital tract and uterine anomalies, sacrococcygealteratoma, epidermoidtumor, and dermoid cysts may accompany the anterior sacral meningocele [4] [6]. The anorectal anomaly, ventral sacral defect, and presacral mass (meningocele, teratoma or enteric cyst) are known collectively as "Currarino's triad" [6].

When compared to posterior and lateral meningoceles, anterior sacral meningocele has a lower incidence, a little more than 250 scientific papers are written on this pathology since its first description in 1837 [2] [3].

The anterior sacral meningoceles are usually diagnosed in the second or third decades with an increased prevalence in women. They may be asymptomatic or may present as nonspecific symptoms like long-term constipation, urinary dysfunction, lower back pain, dysmenorrhea, or perineal hypoalgesia [1] [4] [5]. These symptoms may occur as a result of direct compression of the herniated meningeal sac, sacral nerve root compression, or spinal cord tethering. Also, congenital defects that may occur in autonomic innervation of the bladder and anal sphincter might be associated with constipation and urinary dysfunction [4] [5].

The anterior sacral meningocele does not regress [2]. Even though, it may be asymptomatic, some authors have described infectious complications such as meningitis. Thus, surgical treatment is recommended [5] [7]. Other physicians have recommended surgical treatment when there is an increase in lesion size or when the sacral meningocele is symptomatic [2].

Uterine didelphys is one of the rare Mullerian duct anomalies which are congenital defects that occur during embryological development. They are associated with poorer outcomes of pregnancies such as reduced live births, premature labor, increased chances of spontaneous abortions [8] [9] [10].

4. Conclusion

Since the anterior sacral meningocele is a rare entity, it may be confused with more common cystic lesions of gynecologic origin during routine examinations of female patients. Surgical treatment is recommended especially when the patient is symptomatic or the lesion is growing in size.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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