

### Anterior Sacral Meningocele: An Uncommon Cause of Constipation in Early Childhood

N.K. Bedi  
R. Chadha  
D. Bagga  
R. Anand  
A. Mohta

Anterior sacral meningocele is an uncommon congenital anomaly in which herniation of the meninges occurs through an anterior sacral defect producing a meninges-lined cystic lesion in the presacral space. Clinically, most cases are detected in the third decade of life and are more common in females in ratio of 4 : 1(1). Diagnosis in the pediatric age group is uncommon. We report a case of anterior sacral meningocele in a 1½-year-old female child whose primary complaint was constipation and in whom the correct diagnosis was unsuspected for 6 months.

#### Case Report

A 1½-year-old female child, was

---

*From the Departments of Pediatric Surgery and Radio-Diagnosis, Lady Hardinge Medical College and Associated Kalawati Saran Children's Hospital, New Delhi 110 001.*

*Reprint requests: Dr. Rajiv Chadha, G-123, Vikaspuri, New Delhi 110 018.*

*Received for publication: October 30, 1991;*

*Accepted: January 16, 1992*

referred with a 6 month history of constipation and gradually increasing abdominal distension. The child also had increased frequency of micturition. During this 6 month period, she had been prescribed various laxatives and enemas by her attending physicians with no lasting relief.

Physical examination showed a thin-built infant, weighing 6.4 kg with mild pallor. The abdomen was distended and fecoliths were palpable in the lower abdomen. A per-rectal examination revealed a cystic mass in the pre-sacral space with marked anterior displacement and compression of the rectum. There was no neurological deficit. A barium enema examination done 3 months prior to referral, revealed a bony sacral defect with the pathognomonic 'scimitar sign' of an anterior sacral meningocele along with narrowing of the rectum and lower sigmoid colon (Figs. 1 & 2). Ultrasonography confirmed the presence of a cystic presacral mass with a narrow stalk.

On surgical exploration through abdominal route, an 8×8 cm cystic meningocele sac was found, adherent to the posterior rectal wall and projecting between the leaves of the sigmoid mesentery (Fig. 3). After dissection of the sac from its attachments, the narrow stalk was exposed and ligated with 1-0 nylon. The abdomen was closed without drainage. The post-operative course was uneventful and the child was discharged 7 days after surgery. At 3 months follow-up the child is asymptomatic.

#### Discussion

Anterior sacral meningocele is a rare

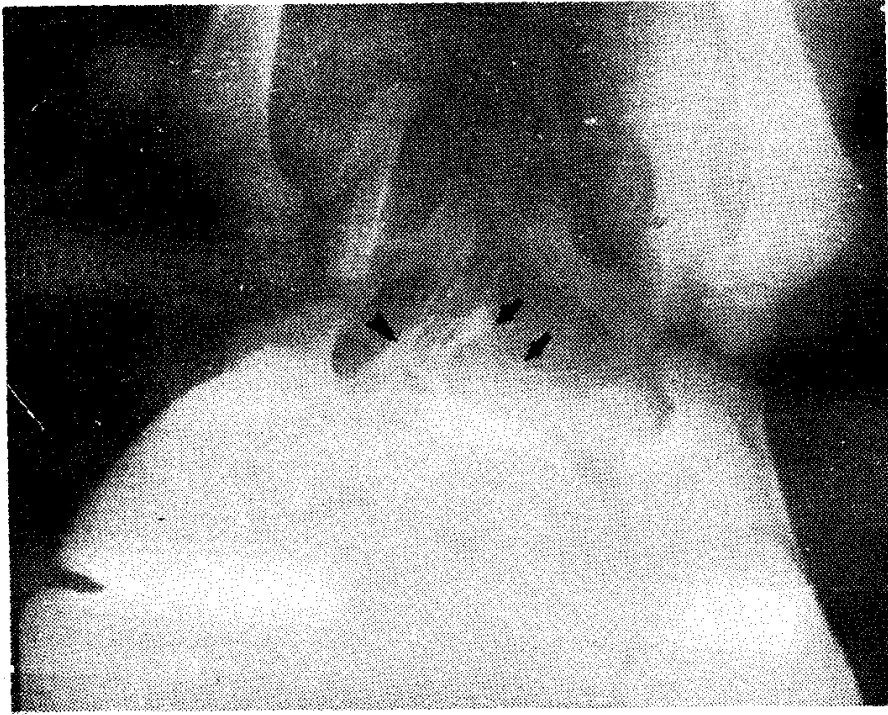


Fig. 1. Barium enema showing characteristic 'scimitar' appearance of the sacrum (small arrows). Narrowing and displacement of the rectum and lower sigmoid colon is also seen (arrow-head).

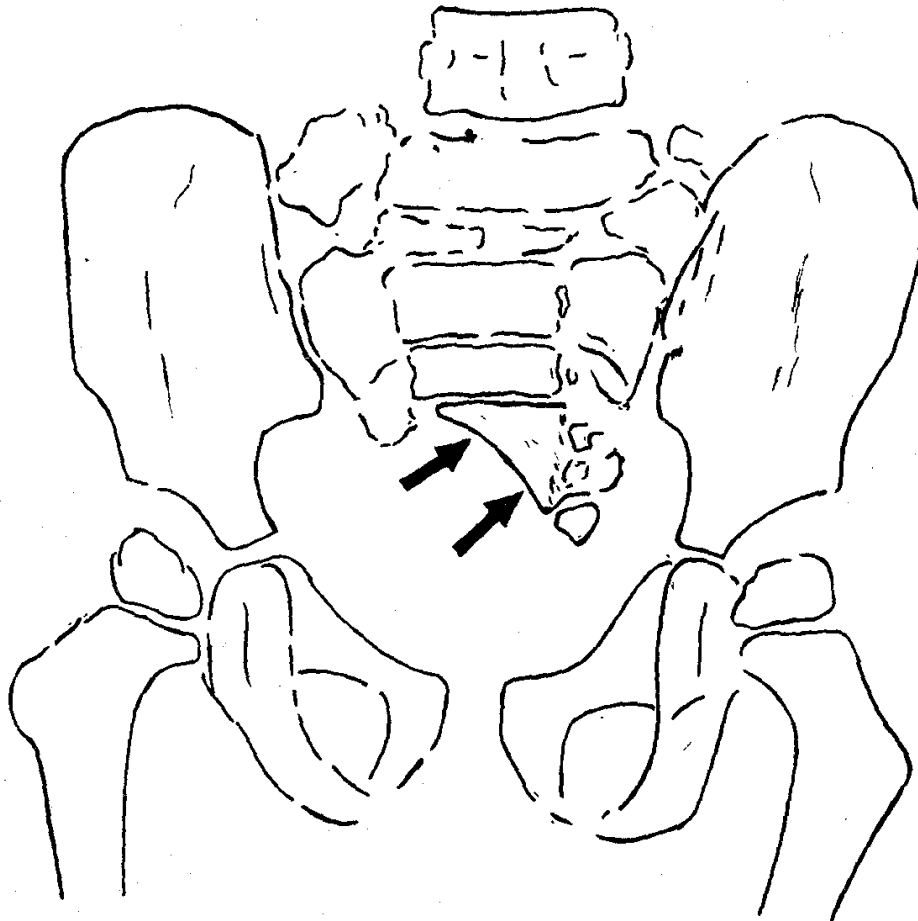


Fig. 2. Line diagram of the sacral deformity with the 'scimitar sign' as seen on the radiograph.

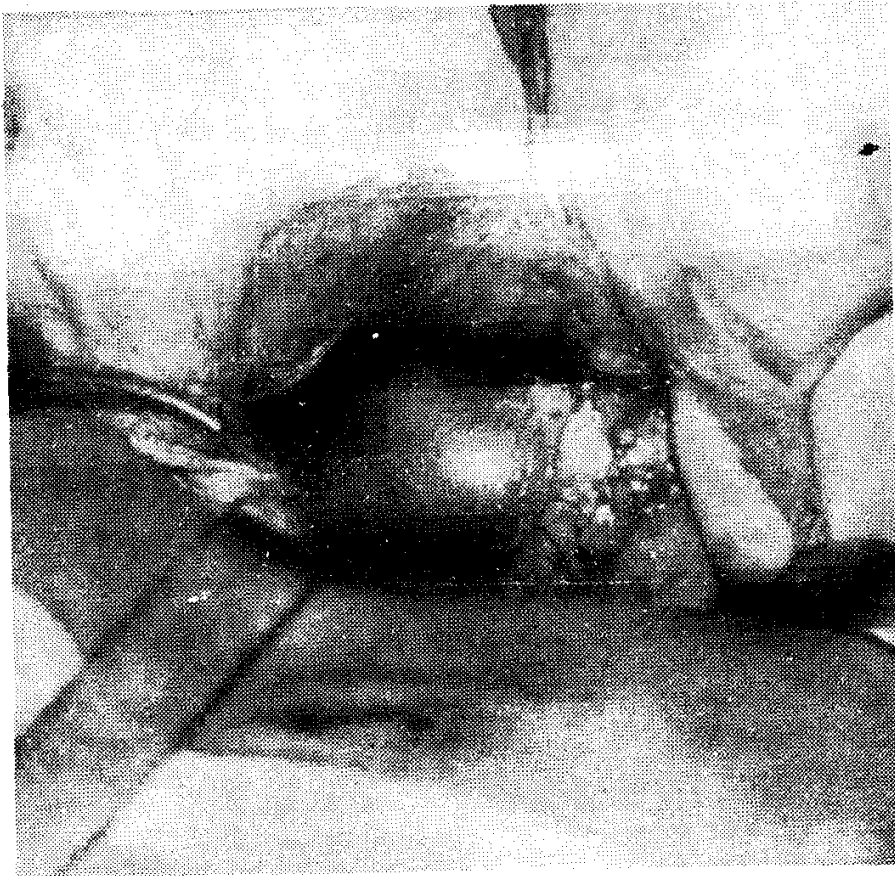


Fig. 3. Photograph taken at surgery showing the anterior sacral meningocele with adjacent sigmoid colon.

condition which was first described in 1837(2). It produces symptoms related to compression of adjacent pelvic structures and sacral nerve roots by the meningocele sac. Association with other anomalies such as bicornuate uterus, duplication of the vagina and anal atresia or stenosis has been reported(1). The condition has also been associated with Marfan's syndrome and neurofibromatosis(1).

Anterior sacral meningoceles are relatively uncommon in the pediatric age group and a review of 148 cases by Villarejo *et al.* revealed that only 28 cases had been reported in children less than 5 years of age(3). These included 9 male and 19 female patients. The most frequently reported presenting symptom in this age group was constipation.

Presacral masses including anterior sacral meningocele are relatively uncommon and may remain unsuspected leading to a delay in diagnosis as in our patient. In all the 3 infants with anterior sacral meningocele reported by Anderson and Burke(4), diagnosis was delayed for the same reason. In this context, the value of a simple digital per-rectal examination in every child with constipation cannot be overemphasized. The finding of a cystic presacral mass on per-rectal examination is a strong pointer to the diagnosis of anterior sacral meningocele or other cystic presacral lesions such as a cystic teratoma or retro-rectal duplication cyst. The additional finding of the pathognomonic 'scimitar sign', sacral deformity on a radiograph of the pelvis as in our case would confirm

the diagnosis of anterior sacral meningocele.

The 'Scimitar sign' seen in approximately half the cases of anterior sacral meningocele is produced by a unilateral sacral defect with a smooth margin simulating the shape of the old Arabic sabre(5). Around 20% of patients, however, have a midline sacral defect with the lateral wings of the sacrum draping either side of the defect(6). Ultrasonography, CT scan and magnetic resonance imaging (MRI) may be used to demonstrate the extent of the meningocele, characteristics of the sac and the width of the stalk(1,7).

The surgical approaches to presacral meningocele include the posterior trans-sacral by laminectomy, the perineal and the anterior transabdominal approach. The posterior approach allows identification and preservation of the nerve roots but water tight closure of the pedicle may not be possible(7). The anterior trans-abdominal approach is more straightforward and offers better access to the neck of the sac although there is more potential risk of damage to nerve roots. In our patient exposure as well as excision of the meningocele was entirely satisfactory using the trans-abdominal approach.

In conclusion, although anterior sacral meningocele is an uncommon cause of constipation in early childhood it should be kept in the differential diagnosis whenever a child presents with constipation. The simple but often omitted per-rectal examination and a plain skiagram showing the characteristic sacral bony deformity are sufficient to diagnose this unusual congenital condition.

#### REFERENCES

1. Green WJ, Green AE. Large pelvic mass in a patient with neurofibromatosis. *Inv Radiol* 1988, 23: 772-774.
2. Bryant T. Case of deficiency of the anterior part of the sacrum, with a thecal sac in the pelvis, similar to the tumor of spina bifida. *Lancet* 1837-1838, i: 358-360.
3. Villarejo F, Scavone C, Blazquez MG, Pascual-Castroviejo I, Perez-Higueras A. Anterior sacral meningocele: Review of the literature. *Surg Neurol* 1983, 19: 57-71.
4. Anderson FM, Burke BL. Anterior sacral meningocele. A presentation of three cases. *JAMA* 1977, 237: 39-42.
5. Jabre A, Ball JB Jr, Tew JM Jr. Anterior sacral meningocele: Current diagnosis. *Surg Neurol* 1985, 23: 9-13.
6. French BN. Midline fusion defects and defects of formation. *In: Neurological Surgery, Vol 3, Ed Youmans JR. Philadelphia, WB Saunders, 1982, pp 1236-1380.*
7. Simpson BA, Glass RE, Mann CV. Anterior sacral meningocele: Magnetic resonance imaging and surgical management. *Br J Surg* 1987, 74: 1185.

---

## Congenital Hemihypertrophy and Wilms' Tumor

S. Sarkar  
D. Prakash  
R.K. Marwaha  
R. Samujh  
K.L.N. Rao

Congenital idiopathic hemihypertrophy has been reported to be associated with a

---

*From the Departments of Pediatrics and Pediatric Surgery, Postgraduate Institute of Medical Education and Research, Chandigarh 160 012.*

*Reprint requests: Dr. R.K. Marwaha, Associate Professor, Department of Pediatrics, PGIMER, Chandigarh 160 012.*

*Received for publication: August 9, 1991;*

*Accepted: January 16, 1992*