CASE REPORTS AND TECHNICAL NOTES

ANTERIOR SACRAL MENINGOCELE*

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Anterior sacral meningocele is a meningeal cyst which presents anteriorly through a unilateral congenital defect in the sacrum, in contrast to the usual posteriorly situated midline meningocele.

In recent years the condition has aroused increasing interest due to its rarity and to the unhappy complications resulting from incorrect diagnosis and treatment. In their extensive review of the literature Coller and Jackson1 uncovered 23 cases, of which 18 were treated surgically with a case mortality rate of 44 per cent, and eight patients of this group were cured. Shidler and Richards2 recently have contributed one additional cure, and this report adds a tenth successful surgical result to the literature. This presentation is intended to aid in standardizing the operative management of this unusual lesion in the light of our own and the reported experience of others.

The procedure of choice appears to be a posterior transsacral approach, excision of the cyst, and ligation or suture of the pedicle. For additional bony protection it is preferable to transfix the pedicle within the sacral canal beneath the most caudally placed intact neural arch. Other adjuncts to the surgical management are closure without drainage, obliteration of the dead space by soft part suture or postoperative elastic pressure, and the employment of prophylactic chemotherapy. The abdominal and the transvaginal routes of approach to expose an anterior meningocele have proven almost uniformly unsuccessful and often fatal. The technique of posterior drainage and packing of a dead space that leads to the spinal theca and opens immediately contiguous to the anus invites disaster in the form of retrograde contamination.

A survey of our own and the available cases in the literature immediately suggests that the presenting symptoms may be developed along three general lines: (1) Local manifestations due to pressure of the cyst on the rectum and adjacent pelvic viscera; (2) neurological deficit in the sacral segments; and (3) disturbances resulting from intermittent increase in intracranial pressure by direct hydraulic action. The size of both cyst and pedicle, the direction taken by the enlarging mass, and the degree to which neural elements are incorporated within the sac are the determining variables. As growth occurs the cyst may enlarge anteriorly, lateral to the rectum, and enter the true pelvis with resulting constipation, menstrual disturbances and, at times, soft part dystocia. Sphincteric disturbances and varying grades of sacral anesthesia are the usual neurological manifestations. Pressure on a cyst (as in sitting) that has a broadly patent communication to the terminal theca will be immediately transmitted to the cerebrospinal system, giving rise to headache, and the usual train of symptoms associated with increased intracranial pressure; whereas, if the pedicle is markedly attenuated this will not be observed.

CASE REPORT

A 26-year-old white female was admitted to the Out Patient’s Hospital of the United States Naval Hospital, Oceanside, California, on January 22, 1945. Her chief complaint was severe, aching pain in the sacral region of four years’ duration. The pain was intermittent in occurrence, but in recent months had recurred with increasing intensity and radiated into the left sciatic distribution. The discomfort was particularly acute in the sitting position, which could be endured for only 15-minute periods. Associated with sacral pain were episodes

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of bitemporal headache, nausea, vomiting and photophobia. The patient had been aware of borderline incontinence all her life, with occasional urinary or fecal soiling on laughter or unusual exertion. Her past history was significant in two respects: (1) She had suffered several attacks of pyelitis, and urograms taken elsewhere revealed a coexistent congenital anomaly in bilateral bifid renal pelves; (2) the first pregnancy was stillborn following high forceps extraction, and two subsequent deliveries were carried out by Caesarian section. She dated the onset of sacral pain to the first instrumental delivery four years ago.

Examination. There were no abnormalities in the general condition. There were well-healed Caesarian section scars. Local inspection revealed an aberrantly formed sacrum having a large left-sided bony defect with the intact portion deviating to the right. On bimanual palpation an exquisitely tender, fluctuant mass was outlined occupying the sacral defect and

![Image](image.png)

Fig. 1. Flat x-ray film of the lumbosacral spine and coccyx taken during urographic studies shows the characteristic “scimitar sacrum.”

situated slightly to the left of the midline. Neurological examination indicated hypesthesia in the left second and third sacral segments and relaxation of the anal sphincter. Funduscopic inspection was normal and the routine urine, blood and spinal fluid analyses gave no unusual findings.

Roentgenograms of the lumbosacral spine and coccyx presented the characteristic “scimitar sacrum” associated with anterior meningocele (Fig. 1). There was a large semicircular defect of bone in the left half of the third, fourth, and fifth sacral segments, with a curving deviation of the remaining sacrum and coccyx to the right, and fusion of the coccygeal segments. Pantopaque was then inserted into the lumbar canal and myelograms demonstrated free communication between an extended terminal theca and the large meningocele sac occupying this defect. The communication occurred by means of a pear-shaped pedicle beneath the lamina of the second sacral segment (Figs. 2 and 3).

Transsacral exploration was done through a posterior midline incision extending from the fifth lumbar spine to the coccyx. The aponeurotic attachments of the sacrospinalis and gluteus maximus were incised and retracted, uncovering a bluish, loculated meningocele sac, which measured approximately 5 cm. in diameter and 7 cm. in length to its tapering pedicle beneath the second sacral neural arch. The cyst incompletely filled the dead space formed by the sacral defect, the remaining area containing an excess of fibro-fatty tissue. On the left posterior aspect of the sac was a small dermoid containing sebaceous material. The cyst was mobilized and then opened for examination with the patient in Trendelenburg's position. Caudal nerves were identified high on the right posterior wall of the pedicle; no neural elements were incorporated within the sac itself. Following resection of the cyst it was found that the caudal roots could be spared by a slight tenting of the pedicle closure. Accordingly, a double row of interrupted silk sutures, longitudinally directed in the median plane, with interposed muscle strips for reinforcement, were used in plastic closure of the pedicle. A V-shaped flap of dura was employed to form an artificial root pouch on the right. The completed suture line was entirely protected by the bony arch of the second sacral canal. The wound was then closed in layers without drainage.

Course. Chemotherapy was instituted immediately following surgery, employing penicillin (100,000 units) and sulfadiazine (90 gr.) daily for one week. The postoperative course was smooth, the incision healing by primary intention, and, apart from a temperature elevation to 100° on the first postoperative day, entirely afebrile. She voided normally on the day following surgery, and urination subsequently has been normal. The neurological status was unchanged. The patient was kept flat in bed in mild Trendelenburg position until February
25, 1945, when activity was gradually resumed. Recheck pantopaque myelograms then showed a minute leak at the left angle of the suture line into the dead space beneath. This continued until March 20, 1945, with reabsorption of the small amount of spinal fluid occurring within the dead space without external fistula formation. Elastic pressure, employing seasponge, was then applied for two weeks; recheck myelograms on April 1, 1945 demonstrated that the leak had entirely sealed (Fig. 4). In retrospect it was felt that external pres-

**Fig. 4.** The completed suture line showing slight tenting at the right angle at the site of artificial root pouch formation.

sure might have been employed at an earlier date as soon as the danger of incisional necrosis had passed. At the time of discharge on April 6, 1945 the patient was free of headache and sciatica, and incisional tenderness over the sacrum was progressively subsiding. When seen in the Out Patient Clinic early in May she had returned to normal activity and was housekeeping for her family.

**Pathological Report.** The sac was a fibrous membrane containing strands of smooth muscle and numerous venous channels. There was an occasional collection of xanthoma cells in the wall. One area showed a surface lining of transitional columnar type epithelium similar to bladder epithelium. In other zones the sac had a distinct squamous epithelial lining and the wall contained sympathetic nerve tissue.

Diagnosis: Meningocele, showing dermoid characteristics and containing sympathetic nerve cells.

**SUMMARY**

The characteristic clinical and roentgenographic features of anterior sacral meningocele have been noted with a discussion of the operative management of a successfully treated case. The technical aspects of primary excision and closure have been presented with a view to standardizing the surgical treatment of this unusual lesion.

**REFERENCES**