SACRAL PERINEURIAL CYSTS

With Case Report

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THE diagnosis of a sacral perineurial cyst may be difficult to establish because of the location caudad to the tip of the dural sac and because the cysts do not communicate with the subarachnoid space. Spinal fluid manometrics and analysis, as well as intradural contrast myelography, have proved of no value. The progressive development of a clinical picture pointing to the sacral portion of the cauda equina is perhaps the best indication. The entity should be suspected and searched for in the occasional patient when, at laminectomy for sciatica, the protruding disk is not disclosed.

The patient described showed clinical findings of a lesion compressing the sacral portion of the cauda equina which was found at operation to be an extradural cyst.

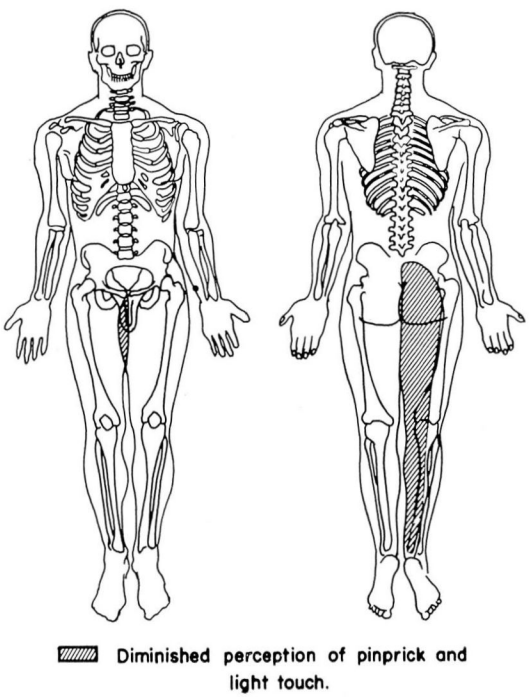

Fig. 1. Sensory deficit in right saddle area starting with S_2 dermatome. (Author's case).

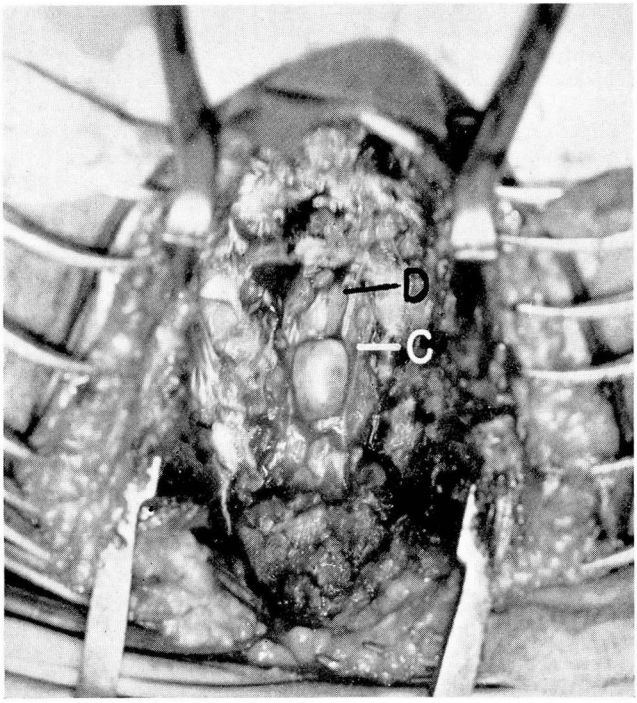

Fig. 2. Operative exposure showing tip of dural sac (d) with perineurial cyst (c) presenting just below.

Case Report

A 40 year old white housewife was first seen in the Department of Urology at the Cleveland Clinic in August, 1949 because of headaches and urinary retention. Four years previously she first experienced increasing frequency, nocturia, and a suprapubic bearing-down sensation when reclining. There had been hesitancy, small urinary stream, and even with straining, incomplete emptying of the bladder. In November 1949, complete retention of urine developed. The patient was hospitalized elsewhere for 10 days for repeated catheterization, after which she was able to void with difficulty. Three days before admission complete retention again occurred. During the past 3 months she had progressive occipital and retro-orbital headache, more severe when reclining. In the past 2 months a dull, aching pain in the anterior aspect of the right and then the left thigh appeared, as well as a deep, steady, sacral backache, both of which were intensified when in the supine position.

On admission, she was found to have 600 cc. of residual urine without, however, the expected bladder neck obstruction. Neurologic examination (fig. 1) was completely negative with the exception of a relative right saddle hypalgesia extending down the posterior aspect of the right leg in the second sacral dermatome. The patellar and Achilles reflexes were equal and active. Jugular compression caused mild increase in the sacral backache. Lumbar puncture, performed at the lumbosacral interspace, revealed clear, colorless cerebrospinal fluid with normal hydrodynamics. The fluid contained 1 cell per cu. mm., a negative Pandy reaction, 33 mg. total protein, and negative Wassermann, Kahn, and colloidal gold reactions. A roentgenogram of the lumbosacral spine revealed no abnormalities.

Pelvic examination was normal. The patient was slightly obese, weighing 151 pounds, and was 5′ 3″ tall. Blood pressure was 110/80. The remainder of the general physical examination was satisfactory. Blood sugar level was 99 mg. per cent, 5-¾ hours postprandially, and the urea, 21 mg. per cent.

In view of the sensory changes, a lesion compressing S-2 was suspected and sacral laminectomy was performed on September 9, 1949. The Hudson drill with a large burr was used to penetrate the posterior sacral plate at the lower border of the S-2 segment, slightly to the right of the midline. As the bone was penetrated, a thin, translucent, tense, cystic membrane bulged through the opening. No epidural fat was visible. Rongeurs were used to enlarge the bony exposure to include the second sacral lamina, the lower half of the first, and the upper half of the third sacral segments. In the cephalad portion of the exposure (fig. 2) the caudal end of the dural sac was visualized with the normal epidural fat around and below except where the clear, cystic structure was apparent. The dural sac pulsated normally although the cyst did not. The dural sac was easily collapsed by gentle pressure; the cyst was extremely tense and unyielding. When the head of the table was lowered the dural sac collapsed, but the cyst remained tense. A hypodermic needle was introduced into the cyst and clear fluid obtained. The cyst was then removed carefully by dissection and appeared to originate from a small nerve filament in the exact midline arising at the extreme tip of the dural sac in the region of the filum terminale. The removal of the cyst left a concave bed formed by compressed right sacral nerve roots. Hemostasis was assured and the incision was closed in the usual manner.

By the third postoperative day the patient was voiding spontaneously and the saddle hypalgesia was diminishing. She was discharged on the ninth postoperative day, at which time there was complete relief of all subjective urinary symptoms. The hypalgesia extending into the right leg and in the right saddle area could not be elicited.

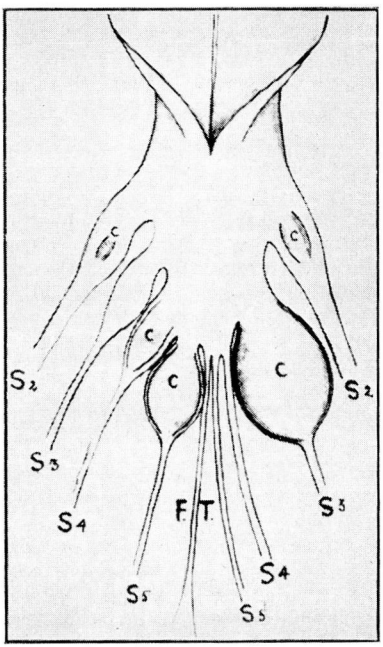

Fig. 3. Composite drawing showing position and size of cysts at cadaver dissection. From Tarlov, I. M.: Arch. Neurol. and Psychiat. 40:1067 (Dec.) 1938.

She was last examined 3½ months after operation, at which time she stated that the headaches, urinary symptoms, leg and low back pain had been completely relieved. However she still retained a small amount of residual urine which diminished at each succeeding examination. Neurologic examination revealed only normal findings.

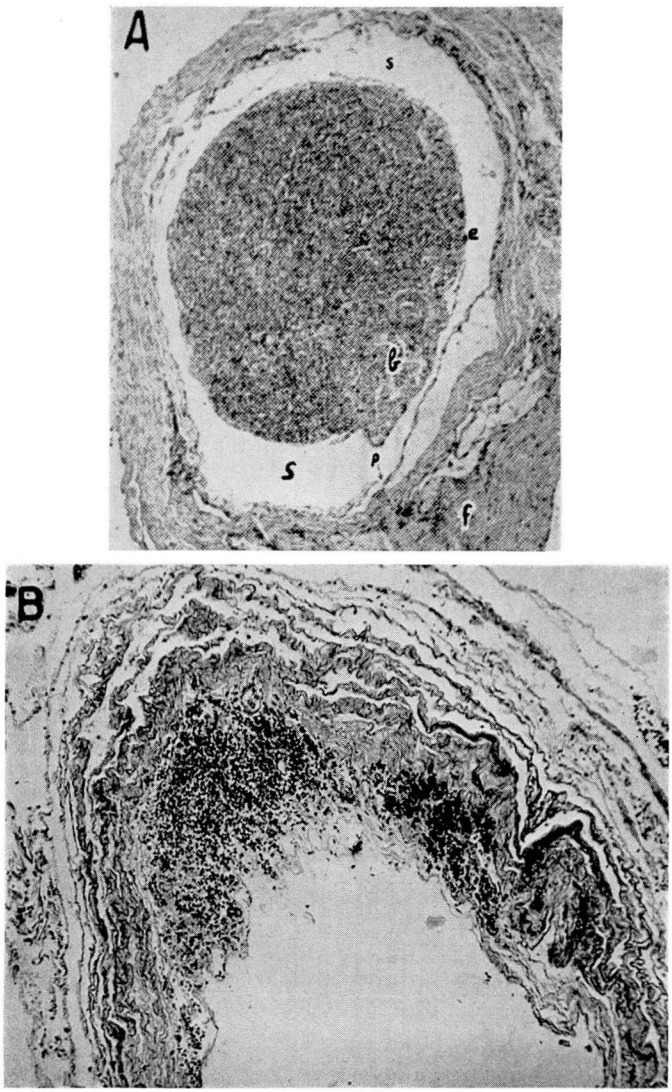

Fig. 4. (a) Cyst beginning in the perineurial space (S) between the endoneurium (e) and the perineurium (P). Epineurium (f) surrounds the perineurium (x-35). From Tarlov, I. M.: Arch. Neurol. and Psychiat. **40**:1067 (Dec.) 1938; (b) Author's case showing analagous microscopic appearance to Fig. 4a (x-70).

Discussion

One other case report of a similar lesion has come to my attention. In 1948, Tarlov[1] described a perineurial cyst attached to the posterior root of the right second sacral nerve in a 42 year old woman with sciatica. This patient exhibited a reduced lumbar lordosis, local pain on pressure over the fifth lumbar spinous process, pain down the posterior aspect of the right leg on hyperextension and right lateral flexion of the back, weakness of dorsiflexion of the toes, hypalgesia along the outer aspect of the dorsum of the right foot, and an absent right Achilles reflex. When the expected disk protrusion was not found at operation, his attention was called to an extrusion of fat from beneath the lamina of the sacrum. Further exposure disclosed the cyst. With its removal the patient experienced prompt relief of symptoms and rapid clearing of neurologic signs.

Interest was first focused upon this pathologic entity by Tarlov[2] in 1938 during dissection studies of the filum terminale. In 30 autopsies performed upon adults with no correlated symptoms, he found 5 instances of cysts attached to the posterior sacral or coccygeal nerves (fig. 3) or nerve roots on the extra-dural portion of the filum terminale. These cysts were neither parasitic nor neoplastic, and appeared first as a space developing between the continuation of the arachnoid covering the root, the perineurium, and the endoneurium – the underlying pial extension surrounding the nerve fibers (fig. 4a). Foci of lymphocytes and plasma cells are frequently present in the outer perineurial cyst wall as well as an increased number of relatively large blood vessels. In his study of the literature, Tarlov was able to find only 2 earlier articles; one, by Marburg,[3] describing 4 cysts of the spinal ganglia, and another by Hinrichs[4] reporting 1 case in which 3 cysts occurred on the posterior thoracic and lumbar roots near the ganglia. None of these autopsy cases had correlated symptoms, and the exact site of origin was not reported.

The incidence of these cysts is not as uncommon as the paucity of reports would indicate. Tarlov recorded 5 cases in which cysts were found out of 30 cadavers dissected, and 3 similar conditions in 39 later dissections.[5] Rexed[6] at the Karolinska Institute in Sweden has recently reported 8 instances out of 17 specimens examined in which he found arachnoidal proliferations with severe cystic changes at one or several levels of both posterior and ventral roots in the dorsal and lumbar areas at their entry into the intervertebral foramina. None of the patients from whom these specimens were derived had shown symptoms suggestive of the pathology found. The microscopic sections from this material showed a close resemblance to those described previously by Tarlov except for the additional findings of arachnoidal proliferation. These findings suggest that some patients with radicular pain at various levels presenting diagnostic problems have perineurial cysts. Further correlation of clinico-pathologic data in this regard is needed.

Summary

Sacral perineurial cysts are relatively common and can produce symptoms. The 2 cases discussed suggest that this condition should be suspected more often. When the diagnosis is made surgery offers good possibilities for complete relief of symptoms.

References

1. Tarlov, I. M.: Cysts (perineurial) of sacral roots; another cause (removable) of sciatic pain. J. A. M. A. **138**:740 (Nov. 6) 1948.
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3. Marburg, O.: Cited by Tarlov.[2]
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5. Tarlov, I. M.: Personal communication.
6. Rexed, B.: Arachnoidal proliferations with cyst formation in human spinal nerve roots at their entry into intervertebral foramina; preliminary report. J. Neurosurg. **4**:414 (Sept.) 1947.