CASE REPORTS AND TECHNICAL NOTES

CONGENITAL DERMAL (PILONIDAL) SINUS WITH DURAL CONNECTION

CASE REPORT AND DISCUSSION

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Congenital dermal sinuses (pilonidal sinuses) in the sacrococcygeal region are not uncommon. However, cases of dermal sinus in communication with the dura are rare. To date, we believe, 15 such cases have been reported; the present case would make 16.

Walker and Bucy compiled the previously reported cases, and added 8 cases in 1934. Kooistra again reviewed the 13 previously reported cases and added one of his own in 1942. Since that time we believe the only case reported has been that of Shenkin, Hunt and Horn. Because of the previous adequate reviews, we shall not repeat the description of the cases reported by Clark, Moise, Ripley and Thompson, W. and N. Sharpe, Ottonello, Hipsley, Hamby, Stammers, and Boldrey and Elvidge.

The present case differs in several respects from those previously reported. The patient is the oldest of any described, 32 years (see Table 1). He had minimal symptoms as compared to previous patients, and he had no serious complications of the lesion.

CASE REPORT

A 22-year-old Sergeant was admitted to Wakeman General Hospital on 20 December 1945, as a direct admission from the Camp Atterbury Separation Center. His chief complaint was increasing stiffness of the right leg, associated with right sciatic pain.

Past history was non-contributory with the exception that he had always been aware of a small dimple over the sacrum, but this had never troubled him, had never drained, and he had not restricted his activities as a student because of it until the onset of the present illness in April 1943. In March 1943, he was inducted into the Army, and it was while he was engaged in basic training that he developed his first symptoms, which consisted of undue stiffness and soreness in the right lower extremity. This continued intermittently with increasing severity, and he reported to sick call from time to time and was given some tablets, with no relief. He was able to remain on active duty, which included several months overseas in the European theater. Over a period of several months, he gradually developed a moderate chronic low back pain superimposed on that in the lower extremity.

In July 1945, he noticed considerable tenderness in the lower lumbar region and within a few days developed inflammation in this area with localized redness, pain, and a slight amount of discharge from the sinus. He again went on sick call, and treatment consisted of hot wet dressings. The soreness cleared spontaneously, but recurred in October 1945, when the same type of treatment cleared the inflammation. Neither of these attacks was severe enough to keep the patient in bed, nor were they associated with much generalized malaise, stiff neck, or other signs of meningeval irritation. He was told, at that time, that he had a pilonidal cyst that should be removed.

Since the second attack of inflammation, he had suffered daily from pain, beginning in the right buttock and sacral region, radiating down the back of the thigh to the knee, and associated with increasing soreness and stiffness, both in the back and in the right leg. All symptoms were made worse by any exertion, especially bending or lifting, or walking for any distance. On admission to this hospital, he had such pain and disability that he was unable to do

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any consistent or sustained work, and was desirous of any operation which might offer some amelioration of his symptoms. He had never noticed any significant degree of weakness or sensory changes. He maintained that there had never been any drainage from the sinus except on the two occasions when it became inflamed.

*Examination.* He was a well-developed young man, who did not appear ill. There was an obvious dermal sinus, with a small tuft of protruding hair, in the midline at the level of the 2nd sacral vertebra, higher than the usual pilonidal sinus. There were no signs of inflammation in this area. The patient stood with a slight scoliosis convex to the left, and placed more of the body weight on the left leg than he did on the right. Forward bending was limited to about 40° so that the finger tips failed to touch the floor by 24 in. Motions of the spine in other directions were impaired to a somewhat lesser extent. The right thigh was slightly smaller than the left, measuring 17 in. in circumference at a point 10 in. distal to the anterior-superior iliac spine, and the left thigh measured 18 in. at the same level. There was no demonstrable atrophy of the lower leg. There was a slight weakness of the extensors and flexors of the right knee, as compared to the left, but this was not enough to cause a demonstrable limp. There was a positive Lasègue sign bilaterally with straight leg-raising limited to 20° on the right and 60° on the left. Knee and ankle jerks were equal and active bilaterally and the plantar responses were normal. Sensory examination revealed no abnormal sensory changes anywhere in the body.

X-rays of the spine revealed spina bifida occulta of the entire sacrum (Fig. 1). X-rays of the pelvis were otherwise negative and those of the knee were negative. Because of the possibility that the sinus might extend into the spinal canal, a pantopaque myelogram was performed 25 January 1946. The needle was inserted in the 3rd lumbar interspace and a free flow of crystal-clear fluid was obtained at a pressure of 6 mm. of mercury. The Queckenstedt test was negative. Analysis of the cerebrospinal fluid thus obtained revealed no cells, a negative

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**Fig. 1.** Preoperative X-rays revealing spina bifida. The sinus passed through just above the separate spine segment in region of S-2 in the area of the notch.