CASE REPORTS AND TECHNICAL NOTES

CONGENITAL DERMAL (PILONIDAL) SINUS WITH DURAL CONNECTION
CASE REPORT AND DISCUSSION

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(Received November 19, 1946)

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 3 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, Hippley, 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 30 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 meningeal 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

![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.](image-url)

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
Wassermann, a colloidal gold curve of 0000000000, no increase in globulin, total protein of less than 33 mgm. per cent and sugar of 45 mgm. per cent. Fluoroscopy and X-ray films presented a very interesting picture. As the opaque medium flowed caudally it split into two columns. The linear defect formed in the pantopaque column began to the right of the midline near the lower edge of the 5th lumbar vertebra and extended downward and medially to the midline over the 1st sacral segment.

Operation. On 14 February 1946, under avertin gas anesthesia, a midline incision was made from the level of the 4th lumbar vertebra to the 3rd sacral segment, surrounding the sinus opening over the 2nd sacral segment. Dissection was carried out, freeing the sinus completely to the point where it passed through the bony defect at the upper edge of the 2nd sacral segment. The free spinous process of the 2nd sacral segment was separated from the sinus tract and lifted out, after incising the fibrous tissue connecting it to the partially formed laminae. A partial laminectomy was performed to include the 5th lumbar and part of the 4th lumbar laminae (Fig. 2). After exposure of the dural sac the sinus was seen to enter the sac at the distal tip, near its posterior surface (Fig. 3). A probe dropped from the skin opening into the dural sac without any pressure. The dural sac was opened and a marked inflammatory reaction of all the nerve roots was noted. The roots were bound down to the anterior and lateral walls of the dura by firm adhesions. The inflammatory reaction was evidently old because several calcified plaques were present, the largest, about 10×4 mm., lying on and constricting the right 2nd and 3rd sacral roots. The flum terminale was bound down anteriorly in the region of the 4th lumbar vertebra and then coursed across the dural sac, becoming adherent to the posterior dura at the lower edge of the 5th lumbar vertebra, and ended in continuity with the opening of the sinus at the posterior edge of the tip of the dural sac. It was taut, and at the time of the operation it was felt this explained the defect seen in the myelogram (see discussion). Irrigation of the lumbar canal washed out several large lumps of soft filmy grayish-white material. These were sent to the pathologist for section; however, they dissolved com-

Fig. 2. Postoperative X-ray showing defect remaining after partial laminectomy.
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completely in the process of preparation. In view of the fact that the patient had such good function of the sphincters and such good strength in the legs, great care was used in freeing the roots and only the most seriously involved ones, including the right 2nd and 3rd sacral, were completely freed. Actually there was marked degeneration of these roots. The dural sac was closed with interrupted silk. The patient withstood the operation very well.

Course. For the first 2 or 3 days postoperatively, he had some hesitancy and difficulty in urination but this cleared spontaneously, and at no time was there any fecal incontinence. He was kept on bed rest for 12 days, and after gradual mobilization he was permitted to take a furlough on the 30th postoperative day.

The patient was seen and examined at 3 and 5 months after operation. He had remained free of pain in the leg, and there had been little or no residual backache. Moreover, there had been some improvement in the mobility of the low back since operation. There were no areas of sensory loss and the reflexes were equal and normal. A repeat myelogram on 4 June 1946 showed a residual defect (Fig. 4). He was separated from the service 5 months postoperative via Separation Center.

PATHOLOGY

The gross specimen included a small tab of epithelial tissue with a sinus orifice in its center. The sinus had been removed intact down to the dura, and is about 5 cm. in length. Cross section of the sinus tract reveals fatty tissue in the periphery, and as one approaches the sinus, there is an increased amount of fibrosis. The sinus measures 4×2 mm. in its widest diameter. It appears to be filled with a necrotic debris. Microscopic study reveals that the sinus tract is lined by stratified squamous epithelium, but there is little evidence of any active inflammatory process. The sinus tract is surrounded by a rather dense connective tissue in its immediate vicinity.

Fig. 3. Left: Sinus opening in ellipse of skin. The opened dura can be seen. Right: Showing sinus connecting dura with ellipse of skin and fat pad, which has been drawn downward with tip of dural sac. One of the calcified plaques can be seen between the two upper stay sutures and the adherent filum terminale can be seen attached to inside of dura near its edge.
Fig. 4. Pantopaque myelogram 5 months postoperatively, showing residual defect.

DISCUSSION

Communication of a congenital dermal sinus with the dura is rare, but the complications are so serious that it should always be considered as a possibility, especially if certain characteristics are noted. In all of the previously reported cases the patients have been younger than this man (see Table 1). In all instances the sinus opening has been above the usual position of

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| Cliffton & Rydell (1946)      | S2    | 22 yrs. | M | Yes (no block)
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pilonidal sinus openings, and in many cases it has been in the thoracic or cervical region (Table 1). Other ways in which these particular sinuses differ from the ordinary pilonidal sinus are that they usually have only one sinus opening and in several cases dermoid cysts have been present. In all cases reported there has been associated spina bifida.

Neurological signs, in most instances of a serious nature, have been present in all reported cases, but these have usually been due to the complications of the sinus, such as meningitis, tumor (dermoid) or abscess. The complications that may arise from communication between the dura and skin are numerous and serious. These range from escape of cerebrospinal fluid through all stages of inflammation to fulminating meningitis. Whether infection be spontaneous or induced by manipulation, such as probing or injection of various media, it has serious consequence. Severe meningitis occurred in at least 4 of the previously reported cases—those of Moise, Ripley and Thompson, one of Stammers and the case of Shenkin, Hunt and Horn. Two of these patients died. In this regard in treatment of congenital dermal sinuses, it is felt that if a single sinus opening, higher than usual, be present, with or without neurological signs, X-rays of the spine should be obtained for the presence of spina bifida. Myelograms should be made in each case. Any manipulation should be deprecated if there is any suspicion of communication with the dura.

In the cases of Ottonello, Hippley, and Kooistra myelograms were made, all of which showed complete block in the area of the sinus. The pantopaque myelogram in the present case revealed a very unusual finding, a slit-like defect in the pantopaque column in the region of the 5th lumbar and 1st sacral segments. In view of the operative finding, we considered this due to the thickened, taut, filum terminale drawn and held posteriorly by the inflammatory adhesions.

The sinus in this case was not proven to be continuous with the central canal by way of a patent filum terminale, as in the case of Shenkin, Hunt and Horn, but the possibility was certainly present. Further manipulation could not have been justified even if we had been aware at the time of their findings. Despite this lack, however, we feel our findings can be considered as confirmatory evidence of the opinion that these dermal sinuses arise from the vestiges of the central canal as suggested by Herrmann and Tourneux, Mallory, Gage, and Oehlecker and supported by Shenkin, Hunt and Horn. Whether all pilonidal sinuses owe their origin to this mechanism or arise as an infolding of the surface epithelium as maintained by Férec, Fox, and Stone, or in one of the many ways previously suggested, remains to be determined.

SUMMARY

1. A case of congenital sacral dermal sinus in communication with the dural sac and in direct continuity with the filum terminale is presented.
2. Unusual X-ray (myelogram) findings are described. There was a slit-like defect arising to the right of the midline of the lumbar 5 interspace and extending downward and medially over the 1st sacral segment.
3. The findings are considered evidence in support of the opinion that dermal sinuses in this area originate as malformations in development of the neural canal.
4. Pilonidal sinuses should always be viewed with suspicion, and manipulation of the sinus by probing, injection or other methods should be carried out with care and only after considering the possibility of an intradural connection of the sinus.

REFERENCES


**TWO TUMORS, MENINGIOMA AND GLOBLASTOMA MULTIFORME, IN ONE PATIENT**

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(Received for publication November 20, 1946)

A number of reports dealing with multiple intracranial tumors is available in the literature. If one excludes those cases that properly belong in the category of von Recklinghausen’s disease, the occurrence of such multiple tumors is decidedly infrequent. Out of a total of 295 cases, Cushing and Eisenhardt report but 3 instances of more than one meningioma in the same individual. An incidence of 1 in 75 cases is recorded by Frazier and Alpers. Somewhat higher is the figure listed by Horrax, 4 multiple meningiomas in a series of 60. Other such cases have been reported, most recently by Arieti and by Mufson and Davidoff. The reader is referred to these papers and to the monograph of Cushing and Eisenhardt for a more detailed perusal of the literature.

Multiple gliomas are also not unknown. Bailey and Cushing found 2 such instances in their collection of 412 gliomas, one a pinealoma and a glioma of the optic chiasm, the other a