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Pediatrics 2003;112;641-647

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ABSTRACT. *Objective.* **Dermal sinus tracts are an uncommon form of spinal dysraphism often presenting in childhood with skin findings, neurologic deficit, or infection. We reviewed our surgical experience, examining presenting symptomatology, operative findings, and patient outcomes.**

Methods. **A retrospective analysis of operated dermal sinus tract cases by the senior author (A.H.M.) from 1970 to present was made.**

Results. **Twenty-eight patients were identified; 17 female and 11 male. Five cervical, 4 thoracic, 9 lumbar, and 9 lumbosacral tracts were explored. Sixteen patients presented at <1 year of age, and 12 were >1 year. Reasons for referral included cutaneous findings (15), neurologic deficit (8), foot abnormalities (4), infection (3), pain (2), and scoliosis (1). Our initial examination revealed cutaneous findings (eg, sinus ostea, pigmentation changes, erythema, skin tags, subcutaneous masses) in 27 patients and neurologic deficit in 19. Age-related differences were apparent. Patients >1 year were more likely to have neurologic deficit (92%) as compared with those <1 year (50%). Bifid spinous processes were noted at dural tract entry in 17 patients. Twenty-two tethered cords, 14 inclusion tumors, and 6 patients with evidence of arachnoiditis were found intraoperatively. Mean follow-up was 33 months. Eleven (39%) remained neurologically intact, 12 (43%) improved, 2 (7%) were unchanged, and 3 (11%) were worse with 2 having decreased perianal sensation and 1 slightly worsened motor function postoperatively.**

Conclusions. **Although most patients were referred for cutaneous stigmata evaluation, >50% had neurologic deficit, intradural tumors, or tethered cords. Skin findings identification should initiate prompt radiologic evaluation and neurosurgical intervention with intradural exploration. Timely intervention may preserve or improve neurologic function in these patients.** *Pediatrics* **2003;112:641–647;** ***dermal sinus tract, cervical, thoracic, lumbar, split cord malformation, intraspinal masses, meningitis.***

ABBREVIATIONS. DST, dermal sinus tract; MRI, magnetic resonance imaging.

Dermal sinus tracts (DSTs) are an uncommon form of spinal dysraphism whose cause is attributed to a failure of dysjunction during fetal development. Normally, the cutaneous ectoderm that ultimately forms the skin and dermal appendages separates from the neuroectoderm, which forms the spinal cord sometime between the third and eighth week of gestation.[1–3] This process, referred to as dysjunction, allows for the insertion of mesoderm, which then forms the vertebral column and underlying musculature that separate the skin from the spinal cord. When a focal failure of dysjunction occurs, a persistent connection, or tract, between the skin elements and the underlying neural structures is established. This tract is lined by stratified squamous epithelium encased in dermal tissue and terminates on or near neural structures.[4] The dysjunction theory supports Mount's observation in 1949 that the dermatomal level of the defect often corresponds to the neural level of the central nervous system structures with which the tract communicates.[5]

DSTs have been reported all along the midline neuroaxis, from the nasion and occiput down to the lumbar and sacral regions. A review of all published cases of congenital spinal DSTs in 1990 reported that 1% of all tracts along the spine were cervical, 10% were thoracic, 41% were lumbar, and 35% were lumbosacral.[6] Although most clinicians would agree that the preponderance of DSTs occur in the lumbo-sacral region, it is probable that several of these earlier case reports and series include coccygeal pits in their reports.

Coccygeal pits and DSTs are distinctly different clinical entities.[7] DSTs are located above the gluteal cleft, have a cephalically oriented tract, and are often associated with intradural pathology. In contrast, coccygeal pits are located within the gluteal cleft, are oriented caudally or straight down, and are not associated with intradural pathology.[7] Coccygeal pits may occur as frequently as 4% in the general population.[8] The incidence of DSTs is usually reported as 1 in 2500 live births.[8] However, this may not be representative of the true incidence of DSTs, as skewed patient populations and lack of follow-up flaw the studies cited as the basis for the derivation of this figure.[9,10] In addition, as these studies were performed in a pre-magnetic resonance imaging (MRI) era, it is likely that they include a number of coccygeal pits in their series. In conclusion, the incidence of true DSTs, although certainly uncommon, remains unknown.[11]

Historically, DSTs have generally come to clinical attention with 1 of 3 symptom constellations. Earlier in the last century, skin findings often did not prompt evaluation, and patients presented later in life with either infection[12,13] or neurologic deficit.[3,4] More recently, increased attention has been paid to

From the Department of Neurosurgery, University of Iowa Hospitals and Clinics, Iowa City, Iowa.

Received for publication Jun 19, 2002; accepted Apr 4, 2003.

Reprint requests to (A.H.M.) Department of Neurosurgery, 1849 JPP, University of Iowa Hospitals and Clinics, Iowa City, IA 52242. E-mail: arnold-menezes@uiowa.edu

early evaluation of cutaneous findings overlying the midline neuroaxis. These findings may present as a sinus ostium with a cephalically oriented tract, angiomata, hypertrichosis, skin tags, abnormal pigmentation, subcutaneous lipomas, or symptoms of local infection such as erythema or induration[7,8] (Table 1). Cutaneous findings have on occasion obscured the skin opening or ostium[14,15]; thus, neuroimaging of all cutaneous findings over the midline neuroaxis above the gluteal cleft is warranted. In addition, DSTs are seen in association with other pathology, such as inclusion tumors (eg, dermoid, epidermoid, teratoma),[1,3,4,16,17–20] split cord malformations (formerly diastematomyelia),[21] and tethered cords.[22] Their presence is often revealed by neuroimaging studies, particularly MRI.

Today, the widespread availability of neuroimaging coupled with increased attention to skin findings over the neuroaxis has led to earlier diagnosis and treatment of DSTs. We chose to review our operative experience with these unusual entities, examining specifically the reason for patient referral, presenting symptoms, physical examination findings, presence of associated pathology, and patient outcomes.

METHODS

The institutional review board of the University of Iowa Hospitals and Clinics gave administrative approval for this study. Retrospective reviews of operative records of all patients who were operated on by the senior author (A.H.M.) from 1970 to the present were examined to identify subjects. Charts, radiographs, and operative notes were obtained and reviewed. Each record was abstracted by a single investigator (L.L.A.). Information was collected regarding demographic variables such as age, sex, timing and mode of presentation, symptom(s) prompting neurosurgical referral, initial neurologic and physical examination findings, radiographic evaluation, operative findings, and patient outcome. Patients who underwent surgical exploration of sacral dimples or coccygeal pits were excluded from analysis; only subjects who were found to have a true DST were included in this study.

RESULTS

Demographics

Twenty-eight patients with DSTs were identified during the review and were included in the study. Seventeen patients were female, and 11 were male. Age at presentation ranged from day of life 1 to 55 years of age. Sixteen patients had presented by 1 year of age, and another 12 did not present until after their first birthday. Five cases were located in the cervical spine, 4 were thoracic, 9 were lumbar, and 9 were located at or below the lumbosacral junction. An additional case had a tract terminating at L3 along with a coccygeal pit at L5-S1. Three patients had previously undergone surgical procedures at the level of the sinus tract. All of these cases were cervical. This information is summarized in Table 2.

TABLE 1. Cutaneous Findings Associated With Dermal Sinus

Sinus ostea with a cephalically oriented tract
Angiomata
Hypertrichosis
Skin tags
Abnormal pigmentation
Subcutaneous lipomas
Symptoms of infection such as erythema or induration

Reason for Referral

The most common reasons prompting a patient's referral were recognition of skin findings (15 patients) or neurologic deficit (8 patients). Other conditions that prompted referral included foot abnormalities (4 patients), infection (3 patients), pain (2 patients), and scoliosis (1 patient). An additional patient was referred because of continued constipation of unknown cause in conjunction with a skin dimple.

When the reason for referral was examined in relation to the patient's age at presentation, trends in presentation as a function of age became apparent (Table 3). Twelve of 16 patients who were younger than 1 year at the time of presentation were referred for evaluation of skin findings, as opposed to 3 of the 12 patients who were older than 1 year at presentation. In contrast, 7 patients who were older than 1 year were referred for evaluation of neurologic deficit, as opposed to only 1 patient who was younger than 1 year. Three of the 4 patients with foot abnormalities were infants, and all 3 infections occurred in infants. Complaints of pain, not surprising, were present only in those who were older than 1 year. The case that was referred after recognition of scoliosis was also in a child who was older than 1 year.

Findings on Initial Neurosurgical Evaluation

Patient physical examination findings appreciated on the initial neurosurgical visit are summarized in Table 3. As was the case with reason for referral, again, age-related differences were appreciated in the frequency of physical examination findings.

Cutaneous Findings

Twenty-seven of 28 patients had cutaneous findings recognized on initial neurosurgical evaluation. Findings included simple dimples, pigment changes, raised plaques, skin tags, and angiomata (Fig 1). Sizes of these lesions ranged from a few millimeters to a 4.5 × 7-cm lesion with a central dimple and surrounding telangiectasia. As previously mentioned, 1 patient had 2 lesions: a DST at L3 in combination with a coccygeal pit at the lumbo-sacral junction. A second patient had 2 cutaneous openings overlying a single tract.

Neurologic Deficit

Although only 8 patients were recognized as having neurologic deficit at the time of referral, 19 were found to have neurologic deficit on the initial neurosurgical examination. Findings included motor weakness in 11 patients, sensory change in 7, reflex changes in 15, gait changes in 5, decreased sphincter tone in 6, and difficulty with bowel and bladder function in 4. In patients who were younger than 1 year, 50%, or 8 of 16 patients, had neurologic deficit at the time of presentation. In contrast, 92%, or 11 of 12 patients, who were older than 1 year had neurologic deficit at the time of referral.

TABLE 2. Demographics ($N = 28$)

Patient No.	Age at Presentation	Sex	Previous Operation	Tract Level	Reason For Referral
1	1 d	Male	No	L4-5	Skin defect
2	3 d	Female	No	T3-T5	Skin defect
3	3 d	Male	No	S1	Infected pilonidal cyst
4	2 mo	Male	No	T5-T7	Skin defect
5	2 mo	Female	No	L3-4	Skin defect
6	2 mo	Female	No	L2-3	Skin defect
7	7 wk	Male	No	L3 & L5-S1	Skin defect
8	3 mo	Female	No	L3	Skin defect
9	3 mo	Male	No	S2-3	Skin defect
10	4 mo	Male	No	L3	Skin defect
11	6 mo	Female	No	L3-4	Neurologic deficit
12	6 mo	Female	No	L5-S1	Skin defect
13	6 mo	Female	No	C4	Skin defect
14	8 mo	Female	No	S2	Skin defect, foot abnormality
15	9 mo	Female	No	C7	Infected "pimple" on back
16	9 mo	Male	No	L4	Recurrent meningitis/skin boil
17	16 mo	Female	No	L5-S1	Foot abnormality
18	2 y	Male	No	S2	Neurologic deficit, foot abnormality
19	3 y	Male	No	L2-3	Skin defect
20	3 y	Female	No	T8-T9	Scoliosis
21	4 y	Female	No	S1	Skin defect
22	8 y	Female	No	L5-S1	Constipation, skin defect
23	8 y	Female	Yes	C5-C6	Neurologic deficit
24	9 y	Male	No	S5	Foot abnormality
25	16 y	Female	No	L4	Neurologic deficit
26	16 y	Female	Yes	C5-6	Neurologic deficit
27	41 y	Female	Yes	C2	Neurologic deficit and pain
28	55 y	Male	No	T11	Neurologic deficit and pain

TABLE 3. Reason for Referral and Findings on Initial Examination

	Entire Group ($N = 28$)	<1 Year ($N = 16$)	>1 Year ($N = 12$)
Reason for referral			
Cutaneous findings	15	12 (75%)	3 (25%)
Neurologic deficit	8	1 (6%)	7 (58%)
Foot abnormalities	4	3 (19%)	1 (8%)
Infection	3	3 (19%)	0
Pain	2	0	2 (17%)
Scoliosis	1	0	1 (8%)
Findings on initial examination			
Cutaneous findings	27	16 (100%)	11 (92%)
Neurologic deficit	19	8 (50%)	11 (92%)
Motor weakness	11	3 (19%)	8 (67%)
Sensory change	7	0	7 (58%)
Reflex change	15	6 (37.5%)	9 (75%)
Decreased sphincter tone	6	1 (6%)	5 (42%)
Altered bowel/bladder function	4	0	4 (33%)
Gait changes	5	0	5 (42%)
Foot abnormalities	4	3 (19%)	1 (8%)
Infection	3	3 (19%)	0
Pain	3	0	3 (25%)
Scoliosis	1	0	1 (8%)

Infection

Only 3 patients had evidence of infection or a history of meningitis in this series; all were infants. Two had infected skin lesions overlying the tract entry site, and another had a history of discharge from the sinus tract. One of the infants with a skin lesion also had a history of *Haemophilus influenzae* meningitis. In each of these 3 cases, surgical exploration was performed after treatment of the infection. Patients were afebrile without laboratory evidence of infection at the time of surgery, so as to avoid additional complications such as overt meningitis or arachnoiditis and scarring.

Pain

Two patients had been referred with a complaint of pain, but on our initial evaluation, a third patient also reported discomfort. It is interesting that these 3 patients, a 16-year-old girl, a 41-year-old woman, and a 55-year-old man, were the oldest patients in the series. The very young age and nonverbal status of many subjects most likely contributed to the paucity of complaints in this group.

Radiographic and Operative Findings

Nineteen patients had vertebral abnormalities demonstrated on radiographic studies. These included bifid lamina (often overlying the site of tract entry) in 17 patients and fused or "block" vertebrae in 3 patients. In addition to the 1 patient who was referred with scoliosis, 3 patients were found to have scoliosis on initial radiographs. Twenty-four of 29 patients had preoperative MRIs; however, only 10 patients had sinus tracts appreciated preoperatively. Intraoperative findings included 22 patients (79%) with tethered cords and 3 with cerebrospinal fluid leaks from the DST encountered at the time of surgery. Three patients had split cord malformations (formerly diastematomyelia), and 2 patients had nerve roots herniated into the sinus tract. Six patients had arachnoid changes including opacity of the normally clear-appearing structure, arachnoid adhe-

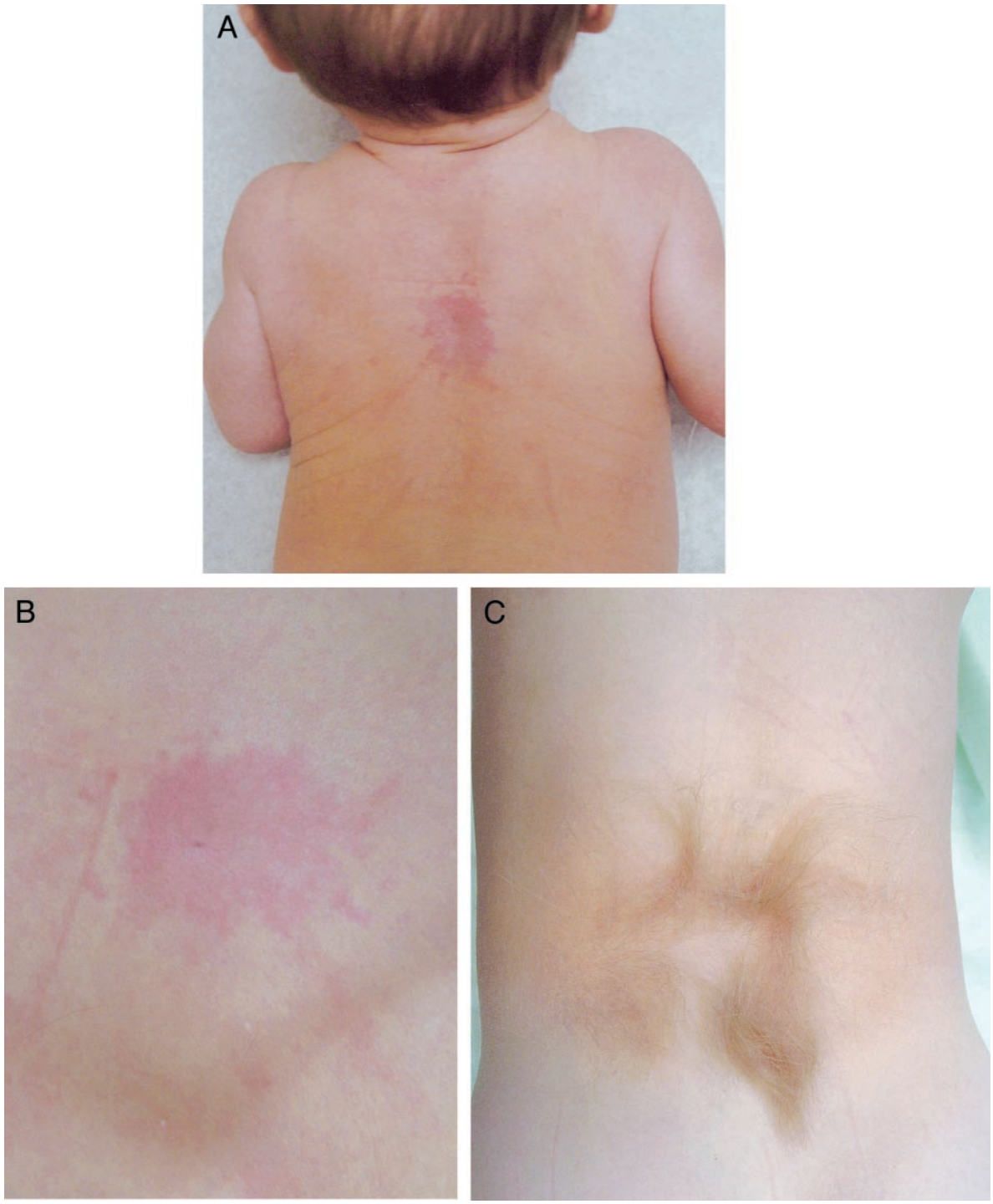

Fig 1. A, Photograph of an infant demonstrating pigmentation change and a visible sinus opening. B, Close-up photograph of the same infant. C, Hypertrichosis with underlying pigmentation change.

sions, and frank arachnoiditis. It is interesting that only 1 of the 3 patients identified preoperatively with a history of infection had these findings. The other 5 occurred in patients with no known history of infection.

Fourteen patients (50%) had mass lesions or tumor associated with the DST. These included 8 lipomas, 3 dermoids, 2 epidermal inclusion cysts, and 1 endodermal cyst. This information is summarized in Table 4.

Patient Outcome and Follow-up

Mean duration of follow-up was 33 months (range: 3–154 months). Eleven patients who were neurologically intact before surgery remained so postoperatively (Table 5). Twelve patients demonstrated im-

TABLE 4. Radiographic and Operative Findings ($N = 28$)

Vertebral abnormalities	
Bifid laminae overlying tract entry site	17
Block vertebrae	3
Scoliosis	4
Intraoperative findings	
Tethered cords	22
Mass lesions	14
Lipoma	8
Dermoid	3
Epidermal inclusion cyst	2
Endodermal cyst	1
Arachnoid changes	6
CSF leaks from the sinus tract	3
Split cord malformation	3
Nerve root herniation into sinus tract	2

CSF indicates cerebrospinal fluid.

TABLE 5. Patient Outcome ($N = 28$)

Neurologically intact pre- and postoperatively	11 (39%)
Neurologically improved postoperatively	12 (43%)
Improved to normal examination	5
Improved as compared with preoperative examination	7
Neurologically unchanged	2 (7%)
Neurologically worsened	3 (11%)

provement in their neurologic examination, including 5 patients who improved to a normal neurologic examination. Two patients were neurologically unchanged with a stable neurologic deficit. These deficits included motor weakness and a lax anal sphincter in a 2-year-old boy and continued motor weakness, sensory deficit, sphincter laxity, and reflex changes in a 16-month-old girl. Three patients were slightly worse postoperatively. Two had decreased perianal sensation that had not been documented preoperatively. Both of these patients had intradural pathology (lipoma and epidermoid). One patient had worsened motor function. This patient was initially referred with a question of infection, and Gram-positive cocci were seen on Gram stain at the time of surgery, although subsequent cultures were negative. Additionally complicating surgical resection in this case was the presence of an epidural lipoma, which blended into a large, dysplastic, sacrally located conus. No neurologic deficit was appreciated preoperatively, but this patient had no motor function with ventral stimulation at L5, S1, or S2 at the time of surgery and currently at 19 months of follow-up demonstrates an L4 motor/sensory level.

DISCUSSION

Certainly, progress has been made in educating health care providers to further evaluate patients with cutaneous findings over the neuroaxis. The more widespread availability of advanced neuroimaging techniques undoubtedly has aided this as well. The trend toward earlier diagnosis and referral has allowed for neurosurgical intervention in a more timely manner and has most likely decreased the development of complications relating to a more delayed presentation secondary to infection or tumor progression, which are usually associated with a worse outcome. In French's[6] exhaustive review of the world literature on DSTs in 1990, 61% of patients presented with infection as opposed to only 11% in our series. It remains interesting, however, that although the most frequent reason for referral to a neurosurgeon was recognition of skin findings, a significant number of patients had serious pathology and/or neurologic deficit on initial evaluation (Fig 2). Seventy-nine percent of our series had tethered cords, 68% had neurologic examination abnormalities on initial examination, and 50% had intradural mass lesions or tumor. Clearly, a delay in diagnosis seems to allow for development of neurologic sequelae as evidenced by the differences in findings when examined as a function of age. Patients who were younger than 1 year were more likely to be neurologically intact (50%) and were more often referred for evaluation of skin findings only (75%). This is in stark comparison to those who were older than 1 year, among whom only 8% of patients were neurologically intact. In this group, only 25% were referred for skin findings only, as many patients presented with varying degrees of neurologic deficit instead.

It is important for the primary care provider to keep several tenets in mind when initially evaluating patients with DSTs. First, the patient may have a low-lying conus because of tethering; therefore, lumbar puncture should be avoided if possible to prevent inadvertently entering cord structures. Likewise, introducing a probe into a sinus tract to explore its termination carries a risk of injuring neural structures as well as that of introducing bacteria and skin contaminants intradurally.[7]

Anterior-posterior and lateral radiographs of the spine along with MRI are indicated in evaluation of a possible DST. Plain radiographs are most useful in demonstrating dysraphic changes such as laminar defects. However, absence of these changes does not exclude the diagnosis of a sinus tract. Plain films should not be used as the sole screening tool but rather to assist in preoperative planning. MRI is the diagnostic tool of choice, as it allows for visualization of the tract as well as associated pathology such as tethered cord, inclusion tumor, syrinx, or more rarely a split cord malformation (Fig 3). If the MRI is normal and the tract is above the gluteal crease, then surgical exploration is still recommended as the tract may be missed if its trajectory is out of the imaging plane.[7] This was the case for 14 patients in our series who did not have sinus tracts appreciated on their preoperative MRI. In our series, all patients who were explored on the basis of skin findings had pathology; there were no negative explorations in our series. It has been our experience that even when the MRI demonstrates a tract, it is not always predictive of the full extent of the pathology appreciated at the time of operation, making surgical exploration mandatory. This is particularly well illustrated in the case of patient 8, whose MRI and intraoperative photographs are shown in Fig 4.

In our series, 82% of patients either improved or

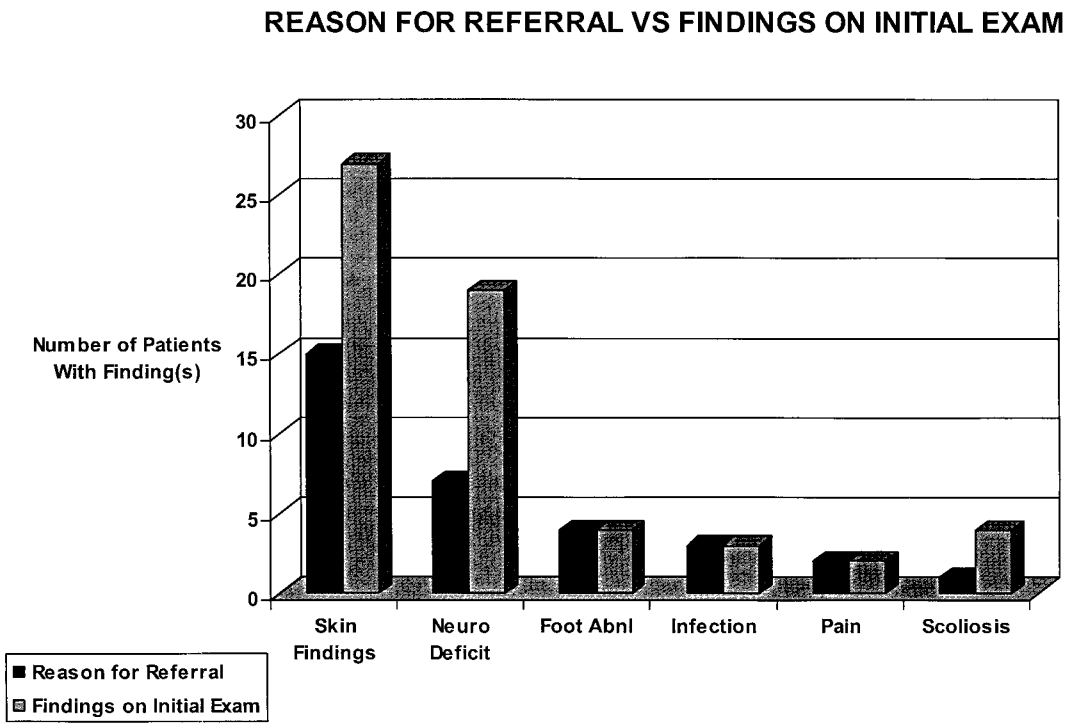

Fig 2. Graph demonstrating the reason prompting referral to a neurosurgeon (black bars) as compared with actual physical examination findings appreciated on the initial examination (N = 28). Foot Abnl indicates foot abnormalities.

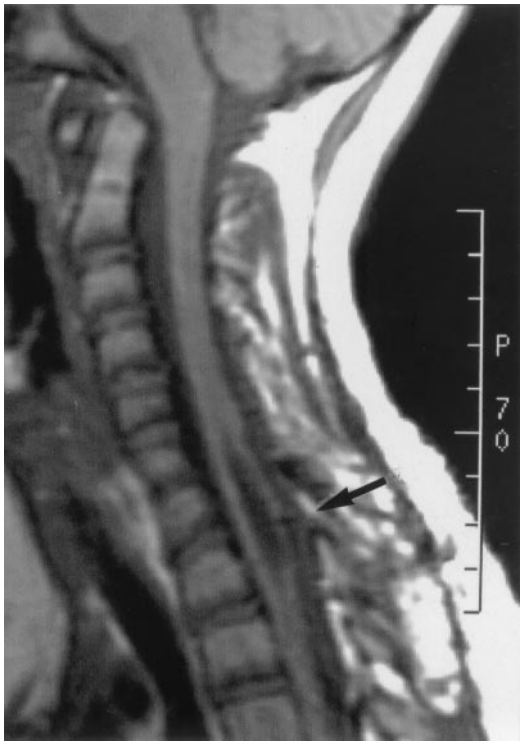

Fig 3. T1-weighted sagittal cervical-thoracic MRI from patient 23 illustrating a DST with associated syrinx. The tract seems to enter dural and cord structures at C6–7 (arrow); however, at the time of surgery, it was found to terminate at C5–6. Note that the sinus tract dorsally tethers the spinal cord.

remained neurologically intact after surgery, and 7% were neurologically unchanged. No patient who was neurologically intact preoperatively had worsening of his or her neurologic status after surgery. One could speculate that without prompt, timely diagnosis and intervention, these patients might have developed neurologic changes over the intervening months, with subsequent less favorable prognoses and outcomes. Delayed diagnosis and intervention exposes the patient to multiple risks, including infection (meningitis or abscess formation), increased tumor size secondary to continued tumor growth coupled with an increased degree of difficulty of resection, and the risk of development of neurologic deficit secondary to either of the aforementioned causes.

Three patients in our series were reoperations. It is important to recognize that simple excision of a cutaneous mass will not obviate intradural pathology, which is frequently present. It may also make subsequent reoperation more difficult because of the presence of scar tissue or loss of definition of tissue planes. Patients should continue to be followed through puberty, with regularly scheduled detailed neurologic examinations. Radiographs should also be obtained periodically to screen for the development or progression of scoliosis. A change in the neurologic examination should prompt the clinician to entertain the possibility of a delayed retethering or incomplete tract resection and possible tumor growth.[23,24] Reimaging with both plain films and MRI is indicated in this scenario.

CONCLUSIONS

The presence of cutaneous stigmata over the midline neural axis should not be considered benign and must initiate prompt radiologic evaluation and neurosurgical referral. It is important for clinicians to be able to distinguish between the benign coccygeal pits

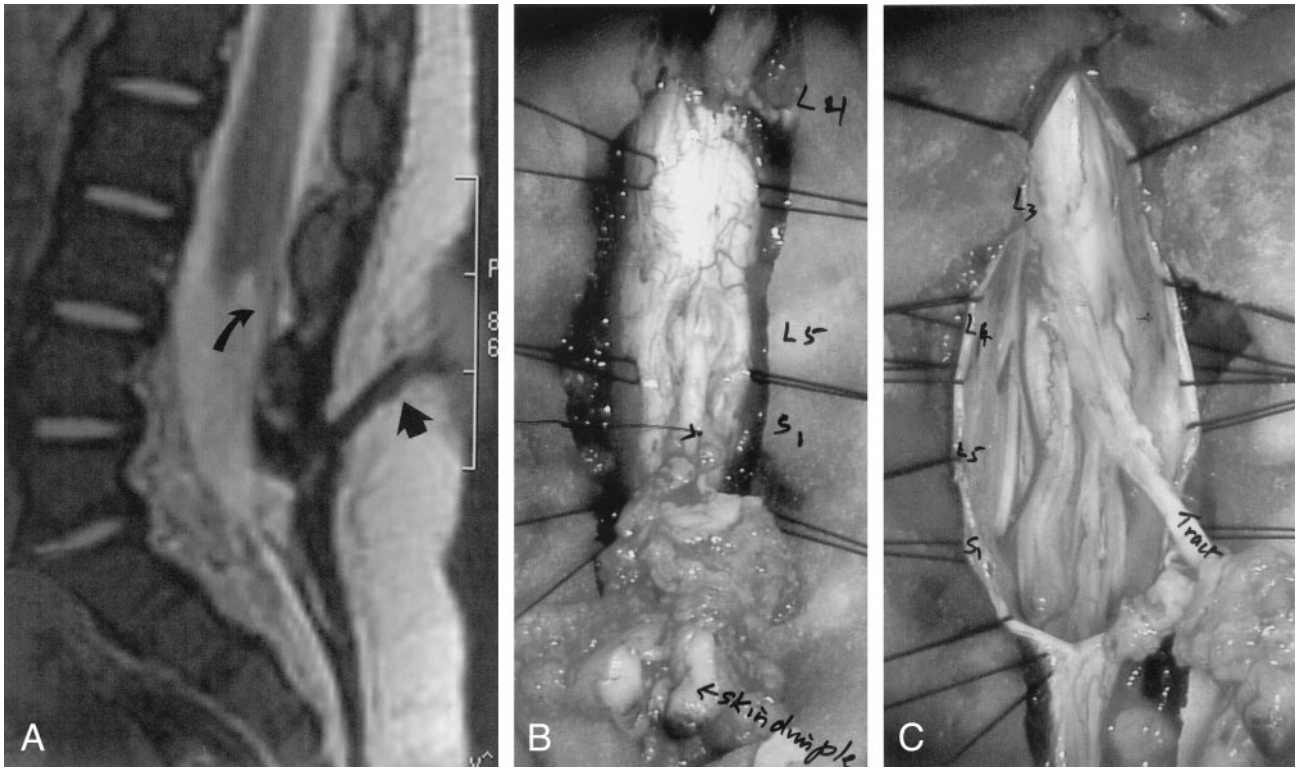

Fig 4. A, T2-weighted sagittal lumbar MRI from patient 8 demonstrating a sinus tract that seems to enter the dura at L5-S1 (arrowhead). This dural entry point was confirmed at the time of surgery; however, on intradural exploration, the tract was found to extend to L3. In retrospect, the intradural extension of the tract at L3 is suggested by the MRI (curved arrow). B, Photograph through the operating microscope of the same patient after L3-S1 laminectomy. The externally recognized skin dimple is dissected free of surrounding tissue at the bottom of the photograph. The tract (arrow) seems to enter the dura at L5. C, A later photograph from the same patient after the dura was opened. Note that the tract (which entered the dura at L5) actually terminated on the low-lying conus at L3.

and the potentially more ominous DSTs. Timely, definitive operative intervention with intradural exploration can preserve or improve neurologic function for many in this patient population.

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