

Coccygeal Pits

Bradley E. Weprin, MD*†§, and W. Jerry Oakes, MD||¶

ABSTRACT. *Background.* Congenital dermal sinuses represent cutaneous depressions or tracts that are lined by stratified squamous epithelium. They communicate between the surface of the skin and deeper structures and may occur anywhere along the craniospinal axis. These sinuses are thought to result from abnormal separation of the cutaneous and neural ectoderm between the third and fifth week of intrauterine life. They may be often accompanied by other cutaneous stigmata, various dysraphic abnormalities, or intraspinal tumors.

In the sacrococcygeal area, cutaneous congenital abnormalities are relatively common. It is estimated that 2% to 4% of children harbor intergluteal dorsal dermal sinuses. These intergluteal sinuses in the perianal region are frequently referred to as pits or dimples. Their cause is considered similar to other congenital dermal sinuses and appears unrelated to acquired pilonidal conditions observed in adults. They may become susceptible to local recurrent infection from trauma or hirsutism.

Controversy regarding the evaluation and management of cutaneous defects in the coccygeal region exists.

Methods. Both a literature review and a career review of clinical material were performed. Databases for articles published in English were surveyed for key words relating to coccygeal sinuses using standard computerized search techniques. The medical records of children presenting to our neurosurgical clinic for evaluation of dorsal dermal sinuses were reviewed to identify those with intergluteal sinuses.

Results. In the evaluation of reported cases and of our own, we were unable to identify any children with coccygeal sinuses without other cutaneous markers other than hair with findings suggestive of intraspinal communication.

Conclusions. Intergluteal dorsal dermal sinuses are relatively common lesions that frequently come to neurosurgical attention. They do not seem to be associated with significant risk of spinal cord and intraspinal anomalies. Simple intergluteal dorsal dermal sinuses without other cutaneous findings do not require radiographic or surgical evaluation and treatment. If other markers or neurologic symptoms are present, however, radiographic evaluation may be indicated. *Pediatrics* 2000;105(5). URL: <http://www.pediatrics.org/cgi/content/full/105/5/e69>; *occult spinal dysraphism, spina bifida occulta, dermal sinus, pilonidal sinus.*

ABBREVIATION. OSD, occult spinal dysraphism.

Cutaneous abnormalities of the back may represent underlying malformations of the spine. One such anomaly, the congenital dermal sinus, is a superficial depression or tract in the skin that is lined by stratified squamous epithelium. Its appearance can signify the presence of an abnormal connection between the skin surface and subarachnoid space and/or an occult dysraphic state. This potential communication places the child at additional neurologic risk from meningitis, which can sometimes be recurrent. These congenital dermal sinuses are frequently associated with other cutaneous signatures, occult dysraphic lesions, or intraspinal tumors. The natural history of such occult spinal dysraphic abnormalities is variable and often unpredictable. Although some individuals remain asymptomatic throughout adulthood, others may develop progressive dysfunction of the lower limbs and bladder. The insidious fashion in which such complications develop may lead to irreversible damage before any symptomatic manifestation. The risk of neurologic deterioration exists at all ages and increases with time and is frequently progressive.¹⁻⁸ The detection of such a subtle cutaneous anomaly in a child may be crucial to future neurologic, urologic, and orthopedic development.

Congenital dermal sinuses may be difficult to identify. They can be located anywhere along the craniospinal axis. Embryologically, the lesions are thought to develop from faulty neurulation. The neural ectoderm incompletely separates from the cutaneous surface ectoderm, a term referred to as incomplete dysjunction.⁹ Histologically, the sinus tract is lined by stratified squamous epithelium with surrounding dermal tissue. The majority of these lesions occur in the lumbar or lumbosacral region followed by the occipital and thoracic regions, respectively. They may extend rostral a considerable distance to terminate several spinal segments above the cutaneous ostium.¹⁰ The dermal sinus tract may actually end blindly in the subcutaneous tissue or it may extend into the spinal canal, as it does in nearly one half of cases. They are infrequently associated with complex vertebral abnormalities unless other forms of occult spinal dysraphism (OSD) are present.

In the coccygeal region cutaneous, congenital abnormalities are relatively common (Fig 1). They are frequently referred to by multiple names (Table 1). It has been determined that ~2% to 4% of children harbor intergluteal dorsal dermal sinuses.¹¹⁻¹⁴ These

From *Children's Medical Center of Dallas, Dallas, Texas; †Neurosurgeons for Children, Dallas, Texas; §Department of Neurological Surgery, University of Texas-Southwestern Medical School, Dallas, Texas; ||Department of Surgery, University of Alabama; and ¶Children's Hospital of Alabama, Birmingham, Alabama.

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Fig 1. Photograph of the low back and buttocks of an infant. A coccygeal pit is appreciated within the intergluteal crease.

sinuses that occur below the natal cleft in the perianal region are frequently referred to as pits or dimples. They may become susceptible to local recurrent infection from trauma or hirsutism. They are not related to acquired pilonidal conditions observed in adults.¹² Their cause is not entirely understood.

Controversy regarding an association between coccygeal pits and spina bifida or any communication with the subarachnoid space exists in the literature. Some authors argue that the presence of any cutaneous abnormality in the gluteal region warrants radiographic and/or surgical evaluation because of a suspected association with abnormal communications with or various abnormalities of the contents of the intraspinal cavity.^{13,15-18} Some clinicians suggest that the respective appearance of the lesion should determine its further work-up. Lesions are inspected for the ability to discern the cutaneous base or for the presence of hair.¹³ Others, however, believe that all coccygeal dimples or sinuses are innocent and warrant no additional evaluation other than physical examination.¹⁹⁻²³ Hence, the proper evaluation and management of these isolated cutaneous defects in the coccygeal region are relatively uncertain.

Given the common occurrence of these cutaneous abnormalities in children, any statement requiring diagnostic evaluation and/or surgical exploration for all coccygeal pits is of a public health concern. Do patients with coccygeal pits warrant investigation or treatment for possible intraspinal anomalies or infection? In an attempt to determine the appropriate therapeutic assessment of these intergluteal abnormalities, 2 tasks were performed.

METHODS

We searched the medical literature for published studies concerning the association between coccygeal pits and spinal dysraphism and/or infection. Using standard computer search techniques, articles written in English containing the following key words were reviewed: dermal sinus, pilonidal sinus, spina bifida occulta, OSD, congenital dermal sinus, and sacrococcygeal dermal sinus. Original and review abstracts and articles were evaluated. The bibliographies of the relevant articles were examined to identify additional studies of association. The 2 investigators reviewed all reports.

In addition, the medical records of all children presenting to our neurosurgical clinic for evaluation of dorsal dermal sinuses between July 1978 and July 1998 were reviewed. The clinic

spanned 2 academic institutions during the study. The clinical presentation, radiographic evaluation, and subsequent management of patients were studied to identify appropriate individuals for inclusion. The clinical evaluation consisted of a detailed neurological and general physical examination in all patients.

A uniform definition was applied to the diagnosis of an isolated coccygeal pit: a cutaneous pit, dimple, or sinus located below the level of a symmetric intergluteal crease that is without the associated presence of any additional cutaneous anomaly. Children were excluded from additional review if hemangiomas, abnormal tufts of hair, areas of cutaneous hypo- or hyper-pigmentation, sinuses, dimples, or subcutaneous masses were identified anywhere on the back in addition to the presumed coccygeal lesion (Fig 2). Children were also excluded by the presence of an asymmetric gluteal cleft.

RESULTS

After an extensive and critical review of the English literature, only 7 cases of cutaneous, coccygeal abnormalities associated with abnormalities of or abnormal communications with intraspinal contents were identified (Table 1). These 7 individuals formed the basis of 5 reports.^{13,15,24-26} Their clinical presentation varied. Six individuals presented with a neurologic infection, bacterial meningitis affected 5, and a spinal epidural abscess occurred in another. The final patient was neurologically normal and without history of antecedent infection but underwent prophylactic surgical exploration. An intradural dermoid tumor was identified.

Our literature review suggests that the relative risk of associated neurologic infection or deficit is exceedingly rare. Only 7 individuals have been reported in the English literature to exhibit findings suggestive of coccygeal pit in association with an intraspinal abnormality or neurologic infection. Careful inspection of these published reports may reduce this small number even further. In 5 cases, the coccygeal abnormality was not in isolation.^{13,15,24} Additional dimples and/or sinuses above the intergluteal crease and hemangiomas were documented. The risk of associated OSD and neurologic infection has been clearly demonstrated for such cutaneous abnormalities. The presence of coccygeal pit, shown to be quite common, may have been incidentally present in these patients. The serendipitous presence of the coccygeal anomaly may have had nothing to do with the associated neurologic abnormality.

Similarly, the description of exact location is inconclusive in the reports of 2 additional patients.^{25,26} The terminology used for location description is inconsistent and photographic documentation is lacking with these respective reports. The sinuses described in the reports by Ripley and Thompson²⁵ and by Stammers²⁶ may actually be located above the natal cleft of the buttocks representative of well-characterized cutaneous signatures of OSD.

After a comprehensive review of the medical records of individuals evaluated in our neurosurgical clinic during a 20-year interval and exclusion of those who exhibited additional cutaneous abnormalities, a total of 1000 patients with simple coccygeal pits were identified. Nearly all patients were below 6 months of age. Evaluation was limited to clinical examination and history. Radiographic imaging studies were not routinely obtained unless per-

TABLE 1. Literature Search of Patients With a Presumed Association Between a Coccygeal Pit and Either Neurologic Infection or Intradural Pathology^{13,15,24–26}

Authors (Year)	Number of Patients	Clinical Presentation
Ripley and Thompson ²⁵ (1928)	1	Meningitis
Stammers ²⁶ (1938)	1	Meningitis
Shenkin et al ²⁴ (1944)	1	Meningitis
Haworth and Zachary ¹³ (1955)	1	Spinal epidural abscess
Kajiwara et al ¹⁵ (1985)	3	Meningitis (2) and dermoid (1)

formed before referral. The patient ages ranged between 1 week and 20 years. No patient was found to exhibit any history of neurologic infection or neurologic deficit on either their initial evaluation or follow-up.

DISCUSSION

The general terms spina bifida and spinal dysraphism refer to those malformations involving any or all the tissues on the midline of the back. They are used to designate those spinal anomalies that possess an incomplete or an inadequate fusion of dorsal midline structures of the developing embryo.²⁷ They represent a spectrum of deformities that include abnormalities of the skin, vertebral column, meninges, or neural elements that may occur alone or in combination.^{28–30} The extent of the malformations may be of mild, moderate, or severe degree. Vertebral column abnormalities are invariably present with involvement of the spinal cord and meninges. Abnormalities of the skin are also common in such instances.²⁹ Hence, the detection of a subtle cutaneous anomaly in a child may be crucial to future neurologic, urologic, and orthopedic development.

OSD refers to lesions that are concealed without exposure of neural tissue or cystic masses. The location and nature of the neural malformation is less obvious on physical examination than overt forms of open spina bifida. They are a heterogeneous group of conditions that are categorized together because of their common embryological origin and the tendency for multiple pathologic entities to be expressed simultaneously in a single individual. Examples include the tight filum terminale, intraspinal lipoma, split cord malformation, dermal sinus and inclusion tumor/cyst, neurenteric cyst, meningocele manque, and myelocystocele. The exact incidence of OSD in the general population is not entirely clear. Many defects remain undiscovered and persist without evidence suggestive of neurologic, musculoskeletal, or urologic impairment into adult life. These occult forms of spinal bifida are much more common than are those that are open. The natural history of OSD is variable and often unpredictable. Although some individuals remain asymptomatic throughout adulthood, others may develop progressive dysfunction of the lower limbs and bladder. The insidious fashion in which such complications develop may lead to irreversible damage before any symptomatic manifestation. The risk of neurologic deterioration exists at all ages. It increases with time and is frequently progressive.^{1–8} Neurosurgical intervention has been demonstrated to halt progression of neurologic deficits.

The optimal management for the multiple abnormalities of OSD includes early diagnosis, neurosurgical referral, and surgical intervention. The primary problem with these conditions is not the risk of intervention, but actually the identification of which individuals are at risk for neurologic compromise and the recognition of the earliest possible clinical manifestations that will provide their detection. Clinical abnormalities may vary according to age. They may bear no obvious relationship to the nervous system. In addition, monitoring the bowel and bladder function in a young child is difficult and too often postponed until an age consistent with urinary continence is reached and irreversible deficits are already present.³¹

Cutaneous signatures are often the initial marker of congenital spine abnormalities and are the most common finding leading to investigation.^{1,32} It is estimated that over one half of individuals with OSD exhibit such stigmata at presentation.^{8,20,31,33,34} They tend to occur in the midline of the back and are often located at the level of the intraspinal abnormality. They are most commonly identified in the lumbosacral region. Numerous cutaneous lesions have been described that may occur singularly or in combination.²⁰ Superficial lesions include areas of abnormal or unusual patterns of hair growth, hemangiomas, paraspinal telangiectasias, areas of hyper- or hypopigmentation, lobulated fatty subcutaneous masses, skin tags or tails, asymmetrical gluteal creases, and dermal sinuses or dimples.

Congenital dermal sinuses are cutaneous depressions or tracts that are lined by stratified squamous epithelium. They can signify both the occult dysraphic state and the presence of a connection between the skin surface and subarachnoid space. They may be difficult to identify and can be located anywhere along the craniospinal axis. They are thought to develop in response to an abnormal separation of the cutaneous and the neural ectoderm between the third and fifth weeks of intrauterine life. They are frequently associated with other cutaneous abnormalities, various dysraphic lesions, or intraspinal tumors.

In the sacrococcygeal region, cutaneous congenital abnormalities are common. In a prospective search for congenital dermal abnormalities of the craniospinal axis, Powell et al¹¹ examined 1997 consecutive newborns delivered at a single institution during a 1-year period. Approximately 3% of the neonates exhibited significant paraspinal abnormalities above the intergluteal crease, while 4.3% of children exhibited coccygeal pits.¹¹ Hence, these intergluteal abnormalities are not infrequent.

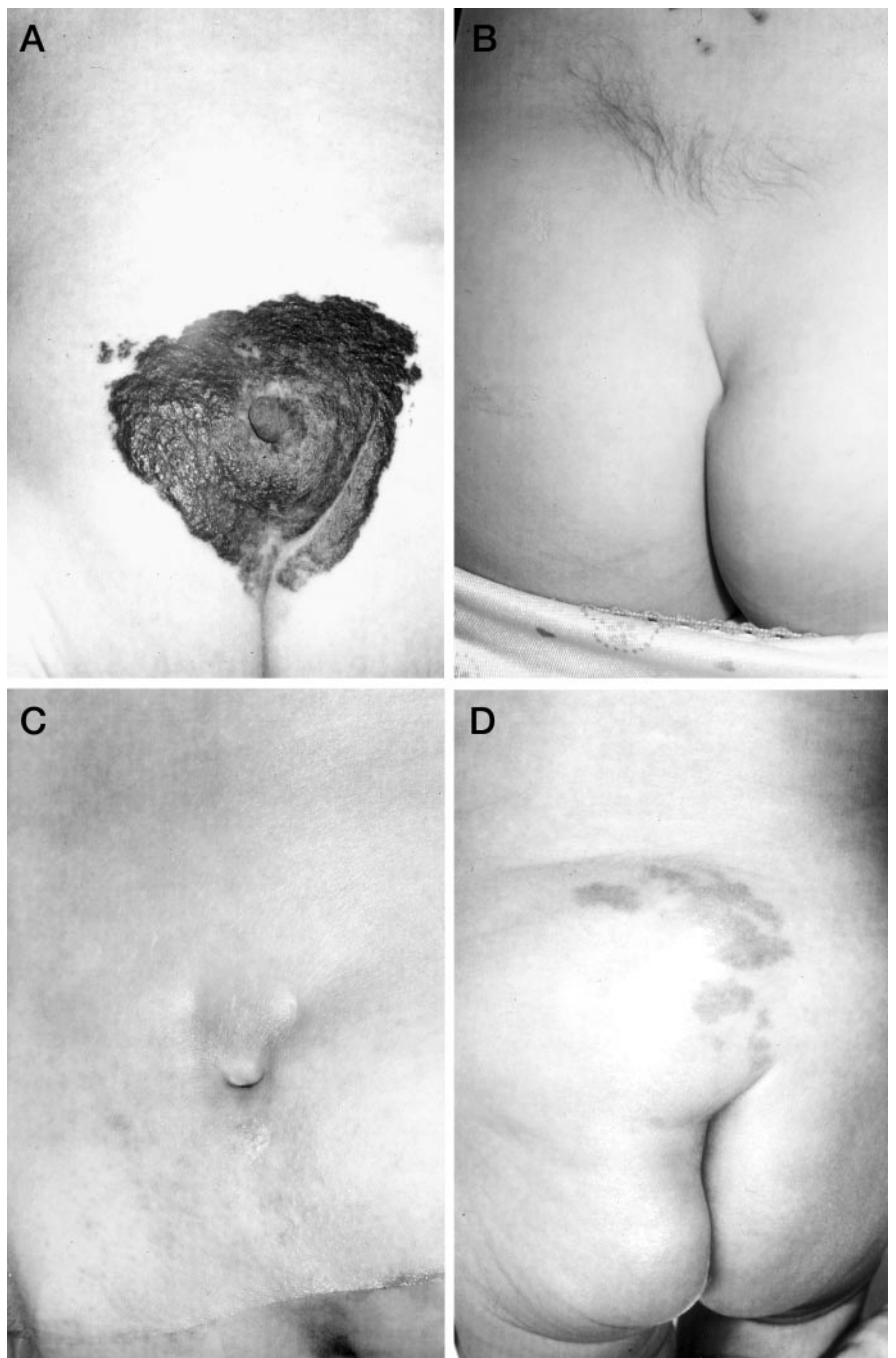


Fig 2. Photographs of the low back and buttocks of infants excluded from this study. All children possessed a coccygeal pit in addition to a variety of cutaneous signatures for OSD. A exhibits cutaneous hemangioma; B, abnormal tuft of hair; C, an additional dermal sinus, above the intergluteal crease; and D, cutaneous hemangioma and subcutaneous lobulated fatty mass.

They may become susceptible to local recurrent infection from trauma or hirsutism. They are not related to acquired pilonidal conditions observed in adults.¹² Their cause is not clear. Controversy regarding an association with OSD and the proper evaluation and management of isolated cutaneous defects in the coccygeal region exists.^{13,15-23} Given the relative frequency of these cutaneous abnormalities, any statement requiring diagnostic evaluation is of public health concern. Based on our studies, it becomes difficult to recommend surgical treatment or even radiographic evaluation for isolated coccygeal pits.

Retrospective review of our own patient data supports the innocence of coccygeal pits. Although such data can be criticized for lacking radiographic documentation, others have already demonstrated evi-

dence of radiographic benignity. Herman et al³⁵ performed spinal ultrasound on 53 infants with coccygeal pits. The average age of those studied was 24 days. The location of the conus medullaris was found to be between T12 and L1 in 13%, behind the L1 vertebral body in 20%, and behind the L2 vertebral body in 67%. No intraspinal anomalies were identified.³⁵ Gibson et al³⁶ prospectively examined 95 neonates harboring cutaneous abnormalities of the back with ultrasound. Seventy-five of the 95 children had isolated coccygeal pits. No abnormality of the spinal axis was identified in those with coccygeal pits.³⁶ The radiographic data appear to correlate and follow our clinical impressions that isolated coccygeal pits are benign. Hence, the burden of proof is not with us to radiographically demonstrate that simple

coccygeal pits are benign. The burden of proof must rest with those who mandate radiographic or even surgical investigation. To this, there seems to be no justification.

CONCLUSIONS

Coccygeal pits are very common abnormalities of the skin. Lesions in isolation are associated with a small incidence for associated neurologic infection or neurologic deterioration. Therapeutic evaluation may be limited to physical examination. Lesions in association with other well-defined cutaneous stigmata of OSD warrant further radiographic and/or surgical inspection.

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