The Enigmatic Sacro-Coccygeal Dimple: To Ignore or Explore?

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In everyday practice, pediatricians routinely encounter congenital midline coccygeal and sacral dimples (Figures 1-6). These cutaneous coccygeal and sacral stigmas, most of which are below the intragluteal crease, occur in as many as 4.8% of all children.¹ Yet the incidence of the “true” problematic lesions related to these dimples, such as spinal dysraphism, is only about 1 in every 2,500 births; spinal lipoma, which occurs in 1 of 4,000 births; and dermal sinuses, which occur in 1 of 2,500 births.² More importantly, as explained below, the more common coccygeal dimples seem to be uniformly benign. So, what is all the fuss about? And why are so many babies undergoing expensive evaluations?

Unfortunately, another study of nearly 2,000 consecutive neonates found that as many as 3% were observed to have a significant paraspinal abnormality above the intergluteal crease.³ These findings are among the more worrisome observations, and definitely require further evaluation. However, this high incidence for abnormal sacral dimples seems to be relegated merely to this one study from 40 years ago. Additionally, practitioners seem to be lumping together the rates from each dimple region — those above (worrisome) with those below (usually innocuous) the intergluteal crease.

Therefore, apparently many practitioners feel compelled to further evaluate the mere simple dimple in the sacro-coccygeal area, at least by ultrasound in most of these children. During their training, they have been instilled with a persuasive fear that these sacro-coccygeal dimples may be the only manifestation of an occult spinal dermal sinus tract, tethered cord, spinal dysraphism, etc. Once they decide to perform the additional imaging, they must then explain their fear to the family in order to obtain permission to perform the test.

If as many as 4.8% of normal infants have this physical manifestation of the

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Figure 1. A 6-month-old male who had an ultrasound of the sacro-coccygeal area performed at 2 days of life. Would you have performed the ultrasound for this apparent shallow coccygeal dimple?
coccyx area, these usually unnecessary evaluations can become very expensive for the family, as well as from a public health perspective. The potential clinical ramifications for the infant also become manifested by terrifying the parents and extended family of this potential newborn problem, once they learn your rationale for this evaluation. “You mean my baby could become paralyzed, never obtain bladder control, or require major neurosurgery on the spinal cord?” they’ll ask.

We need to do a better job deciding when to further evaluate the differentiating features of sacral and coccygeal dimples and other associated cutaneous stigmas in this region.

**DORSAL SPINAL SINUS TRACTS**

These sacral dermal tracts can be associated with a heterogeneous set of five major problems in children.

1. Tethering of the spinal cord.
2. Bacterial meningitis secondary to bacteria entering the connecting dermal tract.
3. Aseptic (chemical) meningitis secondary to debris from the spinal tract entering the spinal canal.
4. Compression of the spinal cord or nerve roots from a dermoid cyst.
5. Diastematomyelia.

Other than the very rare case of meningitis, the most commonly observed signs related to these “true” abnormalities include focal neurologic abnormalities, which may develop in 50% and 92% of children aged younger than 12 months and older than 12 months, respectively. In addition, these children may also develop scoliosis, back pain, and orthopedic foot abnormalities — especially very tight heel cords and persistent toe walking.

**RELATIONSHIP TO PILONIDAL CYSTS**

Are sacro-coccygeal dimples related to later pilonidal cysts? It is arguable as to whether any of these deeper non-contiguous sacro-coccygeal dimples could possibly lead to a later pilonidal cyst, a condition commonly encountered in the adolescent and young adult population, with a peak incidence of 19 years and 21 years for females and males, respectively. However, pilonidal cysts are thought to be acquired rather than congenital. The documented risk factors for pilonidal disease are as follows:

- Overweight/obesity.
- Local trauma or irritation.
- Sedentary lifestyle or prolonged sitting.
- Family history.
• Deep natal cleft.

Thus, the congenital dimple may rarely become susceptible to recurrent infection from either trauma or ingrown hairs.

"SIMPLE DIMPLE RULES" FOR SACRAL DIMPLES

The following parameters define which sacral dimples are high risk:6,7

• Larger than 0.5 cm in size.
• Located more than 2.5 cm cephalad to the anal verge.
• Associated with overlying cutaneous markers:
  - True hypertrichosis, or hairs within the dimple (distinctly different than the mild hairiness seen in Figure 6).
  - Skin tags.
  - Telangiectasia or hemangioma (Figure 7).
  - Subcutaneous mass or lump.
  - Apparent aplasia cutis.
  - Abnormal pigmentation.
• Bifurcation (fork) or asymmetry of the superior gluteal crease (Figure 8).

Therefore, the patients in Figures 1-6 would not warrant further evaluation.

Kriss and Desai1 observed that none of the 207 neonates with a sacral dimple who did not meet any of the first three criteria above, had spinal dysraphism. By contrast, spinal dysraphism was present in 40% of the 20 neonates who met any one or more of the first three criteria.

As Drolet8 reported, “Most sacral dimples that fall within the gluteal crease are healthy.” Furthermore, according to Mark S. Dias, MD, FAAP,9 chair of the American Academy of Pediatrics’ section on Neurosurgery, the depth of the tract is also probably irrelevant. It is the associated additional cutaneous abnormality, such as a hemangioma or additional dimples observed more cephalad, that requires further evaluation. Overlying café au lait spots, flammeus nevus (stork bite), and Mongolian spots (Figure 2) are not considered abnormal.10

Any skin lesion near the midline, such as that seen in Figures 7 and 9, is worrisome. A questionable lesion, as seen in Figure 9, should declare itself with mere observation during the course of a few days to a week or so. This lesion was not aplasia cutis, and it appeared to be an early impetigo sore that responded to a course of clindamycin orally. Thus, an ultrasound of the sacral spine area was unnecessary.

Figure 4. A 12-day-old white female with two small, shallow, almost midline sacral and coccygeal dimples. Does the presence of a second dimple warrant newborn ultrasound of the area?

Figure 5. A 5-month-old male infant who has a small coccygeal shallow dimple along with some hairiness of the region. Is this true hypertrichosis, or merely a typically hairy baby? Does he warrant a further evaluation of the area?

Figure 6. A 5-month-old male infant who has a small coccygeal shallow dimple along with some hairiness of the region. Is this true hypertrichosis, or merely a typically hairy baby? Does he warrant a further evaluation of the area?
COCCYGEAL DIMPLES

Weprin and Oakes\(^5\) conducted a thorough English language literature search of cutaneous coccygeal abnormalities. Only seven cases were reported to have developed an intraspinal abnormality or neurologic infection. Five of these cases had ancillary abnormal findings, including overlying hemangioma or additional cephalad dimples. Furthermore, when the authors examined the records of 1,000 patients — mostly younger than 6 months old — in their own neurosurgical clinic, no patients with an isolated coccygeal pit were observed or identified as having or developing spinal dysraphism or complications. Even when thorough radiologic evaluations of all children with coccygeal dimples were performed in several large series, the researchers failed to identify any abnormalities, even occult spinal dysraphism.\(^5\) Thus, the odds of missing any pathology hiding behind a coccygeal dimple seem quite remote.

TRUE INCIDENCE OF ABNORMAL SACRAL DIMPLES

In contrast to the older study by Powell et al.\(^3\) discussed above, a more recent 2003 retrospective study of 5,440 neonates by Lee and colleagues\(^11\) found that only 0.5% of 200 neonates had an abnormal finding with the sacral dimple confirmed by ultrasound. Note, however, that similar to other studies, this child also had additional overlying abnormal cutaneous findings and an abnormally cephalad dimple. The “Simple Dimple rules” continue to predict abnormal dimples.

GLUTEAL CLEFTS

Although the Nelson Textbook\(^7\) states that imaging requirement is considered “uncertain” for gluteal fold deviations, several experts have said that an asymmetrical or bifurcated gluteal cleft may be a fairly good harbinger of occult spinal dysraphism.\(^10\) Albright,\(^12\) a neurosurgeon from Wisconsin, estimated a notably high association (approaching 30%) between tethered cord and bifurcated or angulated gluteal cleft. In fact, Figure 8 shows our lone recent case of ultrasound-documented tethered cord with a conus medullaris at L2 spine, which was related to a sacrococcygeal finding. Note that this child had a bifurcated gluteal cleft. The child’s condition will need to be confirmed by MRI and repaired after age 8 months per our neurosurgery consultant.

FURTHER EVALUATION OF THE ABNORMAL SACRAL DIMPLE

Instrument probing of any deep dimple is not advised, as one could...
introduce bacteria or debris into the spinal canal if the tract is already open. Firm compression of the adjacent region, as shown in Figures 3B and 10B, may be worthwhile to help delineate the depth and extent of the dimple. I also recommend demonstrating this technique to show the parents how to clean a deeper dimple (Figures 3B and 10B). This may be beneficial in preventing a secondary nidus of infection within the dimple.

ULTRASOUND SCREENING

Ultrasound screening seems to be the imaging evaluation of choice for most practitioners. It can be used to screen the neonate at lower risk or marginal risk for spinal dysraphism, such as those in Figures 1 to 6 — if you still insist on screening them. The procedure must be performed before the child is 4 months of age, and it does not require sedation. However, ultrasonography may miss smaller spinal cord lesions, many lipomas, and especially dermal sinus tracts. This technique is also highly operator-sensitive for accuracy, and I would recommend typically performing it inside a major hospital with higher-level neonatal capabilities. Our small community hospital, with its limited neonatal ultrasound experience, would not be a prudent choice for this procedure.

MRI SCAN

For dimples that are higher risk, more suspicious, or with multiple overlying cutaneous stigma, MRI is the preferred initial imaging method. Any abnormalities on ultrasound should also be confirmed with MRI to determine the full extent of the lesion. However, MRI requires sedation, a significant issue with neonates and infants. It can also occasionally miss smaller lesions. If you have any lingering suspicions about the dimple, neurosurgical consultation is recommended regardless. And remember that if you also find an incidental, asymptomatic, or occult spinal dysraphism in any pediatric patient, the child may need further investigation of the entire spinal cord by MRI, as well. This lesion has been associated with more severe spinal abnormalities such as syringomyelia, diastematomyelia, and even tethered cord.

CONCLUSION

Entirely too many expensive and anxiety-provoking imaging evaluations for sacral and coccygeal dimples are being performed in otherwise healthy neonates. Almost no coccygeal dimple

Figure 9. A 5-day-old white female with a near midline small ulcerated, denuded lesion of the coccygeal area. Could this be aplasia cutis? A possible laterally displaced meningocele? Fetal-monitor burn? Or early staphylococcal superficial skin infection?

Figure 10. (A) A 4-week-old infant who had an ultrasound of this coccygeal dimple performed in the newborn nursery due to the depth of the dimple. (B) Further firm compression of the dimple with two thumbs shows that it has a “bottom.” The primary recommendation should be routine deep cleaning of the debris within the dimple using this technique.
requires further evaluation, unless additional overlying dermatologic abnormalities are also seen, such as those described in the “simple dimple” rules. Furthermore, sacral dimples that do not meet the “simple dimple” criteria rarely ever need further evaluation. Be aware of the high association of bifurcated or asymmetric creases with a tethered cord. When imaging evaluation is undertaken, ultrasound is a reasonable screen for low-risk lesions, but MRI is preferred for either higher-risk lesions or for confirmation of an abnormal ultrasound finding.

If you are still uncertain or suspicious of the sacral dimple, consult your neighborhood neurosurgeon.

**REFERENCES**