Intraspinal lesions associated with sacrococcygeal dimples

Clinical article

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Object. Sacrococcygeal dimples in the gluteal fold, also known as coccygeal pits, are observed in 2%–4% of newborns. Sacrococcygeal dimples are not generally considered to be associated with a significant risk of intraspinal anomalies and therefore are not thought to require further radiographic evaluation. Accordingly, the precise incidence and nature of intraspinal lesions that may be associated with sacrococcygeal dimples is unclear. This study was conducted to determine the incidence of intraspinal lesions in patients with intergluteal dimples.

Methods. In this study, the authors used MRI to evaluate 103 children who were seen at the Niigata University Medical and Dental Hospital between 2006 and 2011 because of skin abnormalities in the lumbosacral region. Of these children, 14 were excluded as having a subcutaneous fatty mass, and 5 were excluded because the dimples were above the gluteal fold or did not end at the coccyx. The remaining 84 patients were classified according to whether the bottom of the dimple was visible (shallow) or not (deep). The authors also retrospectively examined other skin abnormalities and coexisting anomalies.

Results. The mean age at the time of MRI evaluation was 11.7 months. Magnetic resonance imaging led to the identification of fibrolipoma of the terminal filum (FTF) in 14 cases (16.7%); 6 of these patients also had a low conus. Classified by depth, there were 58 cases with shallow and 26 with deep dimples. Fibrolipoma of the terminal filum was found in significantly more patients with deep dimples (9 [34.6%]) than in those with shallow dimples (5 [8.6%]). The frequency of other congenital anomalies was significantly higher in patients with FTF-associated dimples (6 [42.9%] of 14) than in those with dimples that were not associated with FTF (9 [12.9%] of 70).

Conclusions. Fibrolipoma of the terminal filum was identified by MRI in 16.7% of patients with sacrococcygeal dimples. The risk of FTF increased when the dimples were deeply excavated or were accompanied by congenital anomalies. Magnetic resonance imaging should be performed to identify intraspinal lesions when there are high risk factors for intraspinal abnormalities, or when an ultrasound screening suggests intraspinal abnormalities.

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KEY WORDS • coccygeal pit • sacrococcygeal dimple • low conus •
MRI • spine • fibrolipoma of the terminal filum • pediatric neurosurgery

DORSAL midline skin abnormalities are often an initial marker of spinal malformation and are the most common finding prompting further investigation. Over half of the patients with occult spinal dysraphism exhibit such stigmata at presentation.^{12,17} Cutaneous lesions may include abnormal hair growth, hemangiomas, hyper- or hypopigmentation, fatty subcutaneous masses, skin tags, asymmetrical gluteal creases, and dermal sinuses or dimples. A congenital dermal sinus, which is a cutaneous tract connecting the skin surface and the subarachnoid space, can be located anywhere along the craniospinal axis. In the gluteal fold, these dermal sinuses, frequently referred to as sacrococcygeal dimples or pits, project to the coccyx and occur in 2%–4% of all newborns. Several lines of evidence indicate that sacro-

coccygeal dimples are not associated with a significant risk of intraspinal anomalies.^{9,10,14,16,20} However, whether there is any association between coccygeal pits and intraspinal lesions, or with any other communication with the subarachnoid space,^{1,5,9,14,16} is controversial. Many studies that have used ultrasound to investigate intraspinal lesions occurring with sacrococcygeal dimples have reported that sacrococcygeal dimples are not associated with intraspinal abnormalities.^{1,6,12,15,17,20} However, ultrasound screening is limited both by the skill of the technician and by the amount of vertebral ossification, and a small lipoma or dermal sinus tract can be overlooked.^{1,5,11,12} In the present study, we used MRI to evaluate the intraspinal abnormalities associated with intergluteal dimples.

Methods

We retrospectively reviewed the cases of 103 patients who were seen at Niigata University Hospital between

Abbreviation used in this paper: FTF = fibrolipoma of the terminal filum.

2006 and 2011 for skin abnormalities in the lumbosacral region. Of these, we excluded 14 cases of subcutaneous fatty mass, indicating intraspinal lipoma, and 5 cases with a dimple above the gluteal fold, indicating dermal sinus tract. The remaining 84 patients with intergluteal dimples that ended at the coccyx were examined in this study. One of the patients was referred after filar lipoma was diagnosed on MRI. The remaining 83 patients were referred without any previous imaging evaluations such as MRI or ultrasound. Clinical information was obtained from radiology reports and the clinical charts of the pediatric neurosurgeons. We evaluated the frequency of intraspinal lesion or a low-lying conus using a 1.5-T MRI scanner. A low conus was defined as a spinal cord terminating below the L-3 vertebral body. We obtained T1- and T2-weighted sagittal and axial images as far up as the thoracic spine. We evaluated not only the dimple depth but also the pigmentation and asymmetry of gluteal folds. Dimple depth was classified by whether the bottom of the dimple was visible (shallow) or not (deep) (Fig. 1). We also compared the frequency of other skin abnormalities and congenital anomalies between patients with and without intraspinal abnormalities. All statistical comparisons were assessed using 2-tailed tests.

Results

This study evaluated 84 patients, 42 boys and 42 girls. The mean age at the time of MRI evaluation was 11.7 months (range 3–39 months). Magnetic resonance imaging indicated fibrolipoma of the terminal filum (FTF) or fatty filum in 14 cases (16.7%), 6 of which also showed a low conus (Table 1 and Fig. 2). There were no cases of dermal sinus connecting the skin and the subarachnoid space. Grouped by depth, 58 patients had shallow and 26 had deep dimples. Fibrolipoma of the terminal filum was significantly more prevalent in subjects with deep dimples (9 [34.6%]) than in those with shallow dimples (5 [8.6%]; $p = 0.0036$, Fisher's exact test) (Fig. 2). Only 1 patient (Case 1) had a hemangioma and deviated gluteal folds, and he also had FTF. Six patients had an abnormal gluteal fold. Patients with FTF (3 [21.4%] of 14; Cases 1, 4, and 14

[Table 1]) were somewhat more likely to have abnormal gluteal folds than those without FTF (3 [4.3%] of 70; $p = 0.0553$, Fisher's exact test). Patients with FTF-associated dimples (6 [42.9%] of 14) (Table 1) had a significantly higher incidence of other congenital abnormalities than those with dimples not associated with FTF (9 [12.9%] of 70) (Table 2) ($p = 0.016$, Fisher's exact test) (Fig. 3).

Discussion

Intergluteal dermal sinuses, frequently referred to as coccygeal pits or dimples, are considered to fall on the low-risk end of the spectrum of skin stigmata. In an international survey conducted by Ponger et al., 48% of pediatric neurosurgeons did not recommend any imaging evaluation for neonates with simple dimples, which were defined as a soft-tissue depression appearing up to 2.5 cm above the anus or within coccygeal proximity of the coccyx.¹³ However, 52% of those surveyed recommended screening—30% recommended ultrasound screening and 22% recommended MRI—indicating some controversy as to whether imaging tests are indicated for simple dimples. Although almost half of the pediatric neurosurgeons who responded to the survey did not consider sacrococcygeal dimples to indicate a particular risk of intraspinal lesions, the present study found that 16.7% of sacrococcygeal dimples were associated with FTF. In contrast, Brown et al.³ observed that FTF occurred in 4% of a random adult population examined by spinal MRI ($n = 100$). We cannot directly compare the frequency of FTF between their study and ours, however, because the population in each study was different (adults vs infants), and their study did not examine whether dimples or other sacrococcygeal skin lesions were involved.

Table 3 summarizes representative studies of the association between sacrococcygeal dimples and intraspinal lesions. Ben-Sira et al. used ultrasound to screen 254 neonates with midline skin stigmata¹ and found no pathological findings in any of the 109 cases with simple dimples. Other studies have also reported normal ultrasound results in most patients with sacrococcygeal dimples^{6,7,12,15} and suggested that ultrasound screening is not

FIG. 1. Dimple classification. In shallow dimples (left), the bottom of the dimple is visible; in deep dimples (right), the bottom of the dimple is not visible.

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TABLE 1: Summary of patients with FTF*

No.	Type	Low Conus	Conus Level	Associated Anomalies†	Other Skin Lesions‡
1	D	yes	L-4 mid		hemangiomas, DGFs
2	D	yes	L-4 mid		
3	D	yes	L-4 mid		
4	D	yes	L3–4	hypospadias	DGFs
5	D	yes	L3–4		
6	D	no	L-2 mid	duplicated thumb	
7	D	no	L-1 low		
8	D	no	L-2 mid		
9§	D	no	L-1 mid		
10	S	yes	L-5 mid	tetralogy of Fallot	
11	S	no	L-2 low	hypospadias	
12	S	no	L-2 mid		
13	S	no	L-2 low	floating kidney	
14	S	no	L-2 low	cryptorchism	DGFs

* D = deep; DGF = deviated gluteal fold; S = shallow.

† Frequency of associated congenital anomalies: 42.9%.

‡ Frequency of associated other skin lesions: 21.4%.

§ Referred after MRI.

necessary for infants with low-risk lesions, such as simple dimples. This finding contrasts with our study, in which 16.7% of the patients with simple dimples (both deep and shallow) had FTF, and 7% also had a low conus. Note that our definition of low conus was fairly strict. In an MRI study, Wilson and Prince concluded that a conus level up to L-3 might be within the normal range for children.²¹ Therefore, we strictly defined a low conus as a spinal cord terminus below the L-3 vertebral body. We performed MRI after 3 months of age when the conus has generally reached its final position, as suggested by previous reports. One study recommended surgically untethering an abnormal conus medullaris position associated with FTF, since there is a very low rate of surgical complications.^{2,19} We performed surgery to untether the spinal cord in all the cases of low conus; these patients were asymptomatic prior to surgery and remained so after surgery. Figure 4

shows a typical case of a patient treated surgically. In this case, the MRI results indicated FTF and showed the conus in the middle of the L-4 vertebral body.

Although ultrasound is noninvasive and readily available, its effectiveness in detecting intraspinal lesions is limited by the amount of vertebral ossification, especially in patients older than 6 months of age, and by the skill of the technician. Ultrasound screening can also lead to false positives or negatives in diagnosing FTF because of the difficulty of distinguishing lesions from the surrounding echogenic cauda equina.^{5,11,12} Thus, our use of MRI may account for our study's higher frequency of intraspinal lesions associated with simple dimples, compared with previous results. Supporting this view, Sasani et al. used ultrasound, MRI, or both to examine 264 subjects with sacral dimples (Table 3) and found 41 cases (15.5%) of tethered cord, including 21 with thickened and fatty

Fig. 2. Incidence of FTF in patients with sacroccygeal dimples. Patients with deep dimples were significantly more likely to have FTF than patients with shallow dimples.

Fig. 3. Incidence of other congenital anomalies. Patients with FTF-associated dimples had a significantly higher incidence of having other congenital abnormalities than patients with dimples that were not associated with FTF.

terminal fila, 16 with dermal sinuses, 3 with diastematomyelia, and 1 with lipomyelomeningocele.¹⁶ There was a discordance of 16.6% between ultrasound and MRI, with ultrasound often failing to detect abnormalities. Thus, the authors concluded that MRI is preferable for evaluating spinal disorders in neonates.

Ultrasound is also less sensitive than MRI for detecting dermal sinus tracts,^{1,5,11,12} which are sometimes misidentified as sacrococcygeal dimples (Table 3).^{10,16} In clinical practice, however, it is important to distinguish between dermal sinus tracts and sacrococcygeal dimples, as they are treated differently. The dural sac ends at S-2,⁸ and sacrococcygeal dimples do not extend into the intradural space. We differentiated between the 2 pathologies by confirming that the dimples were in the gluteal fold and that the base of the dimples projected to the coccygeal tip.¹⁷ In our study, the dimples were not associated with dermal sinus tracts in any of the subjects. This was because the dimple position was checked carefully, and patients with dimples above the gluteal fold were excluded from the study.

The recognition of risk factors for intraspinal lesions

TABLE 2: Number of associated anomalies in cases without intraspinal lesions*

Anomaly	No. of Cases
hypospadias	2
vesicoureteral reflux	1
hypoplastic kidney, chronic renal failure, inguinal hernia	1
polydactyly, syndactyly, microphthalmia	1
inguinal hernia	1
polydactyly, syndactyly	1
subependymal heterotopic gray matter	1
ventricular septal defect	1

* Frequency of associated congenital anomalies: 12.9%. Frequency of other skin lesions: 4.3%.

associated with dimples is increasing. Kriss and Desai defined high-risk dimples as being large (≥ 5 mm in diameter), localized above the gluteal fold or high on the back (≥ 2.5 cm from the anus), or accompanied by other skin stigmata.¹² Our study included patients with a double gluteal fold, a deviated gluteal fold, or pigmented lesions, but excluded those with a fatty mass or those suspected of having a dermal sinus tract above the gluteal fold. Under these conditions, the frequency of intraspinal lesions was significantly higher in patients with deep dimples than in those with shallow dimples. It should be noted, however, that shallow dimples can be associated with filar lipoma (8.6%), so dimple depth alone is not a perfect indicator of intraspinal lesions. Moreover, it is our experience that the gluteal fold shape and the dimple depth can change with age, especially during the first months of life. Based on this experience, we determined the dimple depth after 3 months of age in all cases. Regarding the gluteal folds, a deviated gluteal fold is considered a sign of an underlying subcutaneous lipoma. However, an isolated deviated gluteal fold is not associated with intraspinal lesions.^{1,7} In the present study, the frequency of deviated gluteal folds in patients with FTF was significantly higher than in patients without FTF. We need to evaluate the angle and shape of the gluteal folds precisely before we can conclude that a deviated gluteal fold associated simple dimple is another risk factor for intraspinal lesions. Tubbs et al. reported that hemangioma is another risk factor of intraspinal lesions.¹⁸ In our study, there was only 1 case of hemangioma with a deep dimple and deviated gluteal folds, and this patient also had an FTF (Case 1). However, as the number of cases with hemangiomas was very few, we cannot draw any conclusions about the correlation between hemangiomas and intraspinal lesions here.

We also found that the presence of other congenital anomalies increased the risk of intraspinal lesions. Chern et al. recently investigated the frequency of occult spinal dysraphism in 943 infants with cutaneous markers and reported abnormal ultrasound findings in 58 cases (6.2%).⁴ This frequency was markedly lower than that obtained

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TABLE 3: Representative studies on the association of sacrococcygeal dimples with intraspinal lesions

Authors & Year	Method	No. w/ Dimples	No. w/ Other Abnormalities
Chern et al., 2012	ultrasound	638	38
	MRI	38	5
Ben-Sira et al., 2009	ultrasound	109	1 (conus borderline L2–3)
	MRI	8	0
Henriques et al., 2005	ultrasound	41	4*
	MRI	4	3*
Kriss & Desai, 1998	ultrasound	160	0
Guggisberg et al., 2004	ultrasound or MRI	5	0
Sasani et al., 2008	ultrasound &/or MRI	264	41†
Robinson et al., 2005	ultrasound	86	0
Gibson et al., 1995	ultrasound	75	1 (low conus L-3)
	MRI	1	1 (same case)

* Dermal sinus tract.

† Of these patients, 21 had thickened and fatty terminal fila, 3 had diastematomyelia, 1 had alipomyelomeningocele, and 16 had dermal sinus tracts.

for cases with congenital anomalies, in which abnormalities were found on the ultrasound studies from 27.7% of the subjects (48 of 173). These data suggest that the presence of congenital anomalies is a more reliable predictor of intraspinal lesions than cutaneous markers. Our study likewise indicated that intraspinal lesions were significantly more frequent when the dimples were accompanied by other congenital anomalies.

Although many clinicians do not consider simple dimples to require radiographic evaluation,^{1,5–7,12,13,15,20} in

the present study, MRI revealed that 16.7% of the dimples were associated with FTF. Moreover, a low conus was found in 42.9% of the cases with FTF-associated dimples. We consider ultrasound to be useful as a first screen for simple dimples, owing largely to its ease of use. However, if an ultrasound study detects potential abnormalities, if it is hampered by vertebral ossification, or if the subject has other risk factors for intraspinal abnormalities, MRI should be used to accurately diagnose and evaluate intraspinal lesions.

FIG. 4. Case 2. Typical case of a dimple associated with FTF. **A:** Photograph of the deep dimple. **B:** Intraoperative photograph showing FTF. **C:** Sagittal T1-weighted MR image showing FTF (arrow). **D:** Sagittal T2-weighted image indicating a low conus at the L-4 vertebral body (arrowhead).

Conclusions

Our study identified FTF in 16.7% of patients with simple dimples. The risk factors for associated intraspinal lesions were deep dimples and the presence of other congenital anomalies. The existence of skin abnormalities such as asymmetrical gluteal folds may also be a risk factor. Further study is necessary to determine whether dimple depth is an independent predictor of intraspinal lesions. Ultrasound can be used to screen patients quickly; however, when ultrasound findings are equivocal or subjects carry a high risk factor, MRI can be used to accurately evaluate intraspinal lesions.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author contributions to the study and manuscript preparation include the following. Conception and design: Harada, Nishiyama. Acquisition of data: Harada. Analysis and interpretation of data: Harada. Drafting the article: Harada, Nishiyama. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Harada. Statistical analysis: Harada. Study supervision: Fujii.

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