Management of Sacral Dimples Detected on Routine Newborn Examination: A Case Series and Review

ACW Lee, NS Kwong, YC Wong

Abstract
Isolated sacral dimples are a common occurrence in Caucasian newborns and there has been a concern about their association with occult spinal dysraphism. A retrospective study was carried out in which infants born in a regional hospital during the year 2003 with a diagnosis of sacral dimple were examined. Twenty-six infants (0.5%) were identified from 5,440 live births. There was a female predominance (61.5%) and all infants were born at term. A tuft of hair close to the dimple was described in 6 babies, but none had any neurological deficit. Only 4 infants underwent ultrasonography or magnetic resonance imaging. No abnormality was detected. None of the 16 children who had been followed up (median 25.7 months) had any neurological deficit. A review of the current literature strongly indicates that isolated sacral dimples are innocuous and imaging study for occult spinal dysraphism is not indicated. A diagnostic strategy on the selective use of ultrasonography as a screening examination in atypical cases is proposed.

Key words
Dermal sinus; Neonatal screening; Neural tube defects; Physical examination; Spinal dysraphism

Introduction
Sacral dimples, also known as sacrococcygeal or coccygeal dimples or pits, are the commonest cutaneous anomaly detected at neonatal spinal examination.1 They are defined as shallow or deep depressions occurring at the lower sacral region close to or within the natal cleft. Sacral dimples have been conventionally treated as similar to other cutaneous stigmata such as dimples, hair or pigmented lesions at a higher spinal level as clues of occult spinal dysraphism. Recent studies, however, have indicated that sacral dimples are benign variants of normality. This study was carried out to examine the current handling of sacral dimples and to suggest a rational management according to published data.

Patients and Methods
The study was carried out in a regional hospital where routine examination was provided for all neonates delivered in the hospital. Infants born in 2003 and had been diagnosed with sacral dimples were identified through the Clinical Data Analysis & Reporting System of the Hospital Authority by the ICD9CM code of 685.1 (that shared with "pilonidal cyst without mentioning of abscess"). Their clinical records were retrieved to examine their demographic and birth data, findings on clinical examination and imaging studies, and outcomes on discharge and subsequent follow-up. No standardised management protocol was available and the decision to investigate was at the discretion of the medical officer in charge on a case-by-case basis.
Results

Twenty-six newborn infants were identified, representing 0.5% of the 5,440 live births in the year of study. Sixteen (61.5%) were girls. Nineteen (73%) were ethnic Chinese, 6 (23%) were of South Asian origin, and the other was a Caucasian infant. All infants were born at term (gestation 37.3-42.0 weeks) with birth weights ranging from 2.46-3.64 kg. A patch of hair close to the dimple was found in 3 Chinese and 3 South Asian neonates. None of them had any demonstrable neurological abnormalities. All except one infant underwent imaging studies of the spine including plain radiograph alone (n=21), plain radiography plus ultrasound (n=2), ultrasound alone (n=1), and magnetic resonance imaging (MRI) alone (n=1). All plain radiographs were obtained during the neonatal period, while ultrasonography and MRI were performed on an elective basis depending on availability in the hospital. No abnormalities were detected in any of the studies. Sixteen patients had follow-up neurological examinations at more than 6 months after birth. With a median follow-up of 25.7 months (range, 8 to 38.1 months), none of them developed any neurological abnormalities.

Discussion

One of the purposes of routine examination of the spine in the newborn infant is to look for cutaneous stigmata of an underlying occult spinal dysraphism or neural tube defects. The failure of neural tube closure during embryogenesis may be associated with abnormalities in the overlying ectodermal tissue, such as haemangiomas, hairy patches, skin tags, dimples or sinus tracts, and subcutaneous masses. In addition to herniation of the intraspinal contents in the form of meningocele or myelomeningocele, tethering of the spinal cord to the extra-spinal lesion may lead to progressive neurological deficit during subsequent growth. Thus, early detection and surgical correction are of utmost importance under such circumstances.

Midline sacral dimples that are found at the end of the spinal column in the sacrococcygeal region are the commonest cutaneous anomaly detected in the newborn infant. Prevalence rates from 1-4% have been quoted. An underestimate of the prevalence in the present study is possible, as some of the mildest cases might not have been coded in the hospital information system. Controversies with respect to their association with underlying neural tube defects exist and clinical studies to examine their significance were not available prior to 1993. Five case series have examined a total of 1,374 children (see Table 1) and fail to identify any association between sacral dimples and occult spinal dysraphism. In addition, Weprin and Oakes were unable to find any clearly documented cases of occult spinal dysraphism associated with sacral dimple from a literature search. On the contrary, Gibson et al. found a case of occult meningocele with tethered spinal cord in one of the 105 control, supposedly normal, infants. Thus, the burden of evidence strongly indicates that infants with isolated sacral dimples are not at a higher risk for neural tube defects than otherwise healthy babies.

Kriss and Desai examined 160 neonates who had midline sacral dimples less than 5 mm in size and situated within 2.5 cm of the anus. None had any signs of spinal dysraphism on ultrasonography. On the other hand, eight of the 20 neonates with "atypical" dimples (larger than 5 mm in size, situated farther from the anus, or occurring with other cutaneous markers) were found to have occult

Table 1  Reported case series of isolated sacral dimples in infants and children.

<table>
<thead>
<tr>
<th>Series</th>
<th>No. of patients</th>
<th>Imaging</th>
<th>Findings</th>
</tr>
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<tbody>
<tr>
<td>Herman et al.</td>
<td>53 infants</td>
<td>Ultrasound</td>
<td>No abnormality detected</td>
</tr>
<tr>
<td>Gibson et al.</td>
<td>75 study infants</td>
<td>Ultrasound</td>
<td>1 study infant had abnormal ultrasound but normal finding on MRI</td>
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<tr>
<td>Robinson et al.</td>
<td>86 infants</td>
<td>Ultrasound</td>
<td>1 control infant had occult meningocele and tethered cord</td>
</tr>
<tr>
<td>Weprin &amp; Oakes</td>
<td>1,000 children</td>
<td>No detailed</td>
<td>No abnormality detected</td>
</tr>
<tr>
<td></td>
<td>(most &lt;6 months)</td>
<td>description; not</td>
<td></td>
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<tr>
<td></td>
<td>old)</td>
<td>routinely indicated</td>
<td></td>
</tr>
<tr>
<td>Kriss &amp; Desai</td>
<td>160 infants</td>
<td>Ultrasound</td>
<td>No abnormality detected</td>
</tr>
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MRI, magnetic resonance imaging
neural tube defects. The authors thus concluded that dimples that were bigger in size, located at a higher spinal level, or associated with other cutaneous stigmata should be investigated. Their findings form the basis of recommendation for investigation of atypical sacral dimples in a recent review. MRI remains the gold standard for diagnosis of occult spinal dysraphism, but it is expensive and requires sedation of the infant. Plain radiography is insensitive as bony defects are difficult to identify in the neonatal period when the posterior elements of the vertebrae are not well ossified. Ultrasonography has emerged as the screening modality of choice for the infant with suspected spinal dysraphism. The presence of an intra-spinal mass, abnormal position (dorsal location or location at L3 or lower) or shape (non-tapered appearance) of the conus, or a thick filum are indications for MRI study.

The present study indicates that the medical officers involved in the care of newborn infants were sufficiently concerned with the incidental finding of sacral dimples, but there was a lack of guidance on how this condition should be managed. As most infants did not undergo ultrasound or magnetic resonance imaging, the finding of this study should not be interpreted as all sacral dimples were innocuous. However, in the published literature, isolated sacral dimples are distinguished from other spinal cutaneous markers by their lack of association with occult spinal dysraphism. Hence, only infants with atypical symptoms or signs should be selected for imaging studies. Ultrasonography of the lumbosacral spine is the screening investigation of choice. A suggested scheme for the management of sacral dimple incidentally found on routine examination is depicted in Figure 1.

In summary, cutaneous sinuses, dimples and patches along the spine should be routinely searched in the examination of newborn as clues to an underlying occult spinal defect. However, simple sacral dimples are innocuous unless they are large, located farther away from the anus, or in association with other cutaneous stigmata.

References