Pediatr Radiol
DOI 10.1007/s00247-014-3110-1

ORIGINAL ARTICLE

The simple sacral dimple: diagnostic yield of ultrasound in neonates

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Received: 2 February 2014 / Revised: 2 May 2014 / Accepted: 22 June 2014
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Abstract
Background Although spinal cord tethering is known to be associated with certain clinical syndromes and cutaneous stigmata, its incidence in healthy infants with simple sacral dimples has not been thoroughly evaluated.
Objective Our objective was to determine the frequency of tethered cord in otherwise healthy patients with simple sacral dimples.
Materials and methods We reviewed the lumbar spine US reports of all healthy neonates referred for a simple sacral dimple during a 12-year period at two children's hospitals. A sonogram was considered abnormal for a conus medullaris terminating below the L2–L3 disc space, decreased conus or nerve root motion, an abnormal filum terminale, or for the presence of an intraspinal mass, osseous dysraphism, or a sinus leading to the thecal sac. The medical records of patients with abnormal screening sonograms were reviewed to determine the final clinical outcome.
Results During the study period 3,991 infants underwent screening sonography. Of these, 107 were excluded because of the presence of other medical conditions. Of the remaining 3,884 healthy infants, 133 (3.4%) had an abnormal sonogram. Five (0.13%) of these infants were lost to follow-up; 52 subsequently had normal follow-up imaging; 49 had a low conus without other signs of tethering; 18 had a fatty filum; 2 had decreased conus motion; 2 had both a low conus and a fatty filum. None of these infants underwent surgery. Only the remaining 5/3,884 (0.13%) infants underwent surgical intervention (95% CI: 0–0.27%), and 4/5 were found to have a tethered cord intraoperatively.
Conclusion The risk of significant spinal malformations in asymptomatic, healthy infants with an isolated simple sacral dimple is exceedingly low.

Keywords Sacral dimple · Simple dimple · Tethered cord · Occult spinal dysraphism · Ultrasound · Infant · Neonate

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Introduction

The association of tethered cord syndrome with congenital anomalies and certain cutaneous stigmata such as hairy patches, subcutaneous lipomas, and rudimentary tails is well documented [1, 2]; however the association of a simple sacral dimple with tethered cord or other spine anomalies has not been clearly defined. Although cutaneous stigmata are reported in up to 86.3% of patients with occult spinal dysraphism [2], they are also present in 4.8–7.2% of healthy children, and 74% are simple sacral dimples [3, 4]. There is no clear consensus among clinicians on how to best work up an otherwise healthy child with an isolated simple sacral dimple [5].

Published online: 05 July 2014

A simple sacral dimple is defined as a solitary dimple less than 5 mm in diameter and less than 2.5 cm from the anus, midline in location, and without visible drainage or additional associated cutaneous stigmata such as a hairy tuft, hemangioma, skin tag or tail [3]. An example of a simple sacral dimple is shown in Fig. 1.

US imaging is an effective screening method for tethered cord, with a sensitivity of 96% and a specificity of 96% [6], and it has been reported to be a cost-effective screening tool for patients with simple sacral dimples [7]. However recent literature suggests that US screening may not be necessary in patients with isolated simple sacral dimples [3, 6, 8]. Despite the evolving guidelines for lumbar sonography [9], no large-series studies have been performed to confirm the negative association of simple sacral dimples with tethered cord.

The goal of our study was to determine the frequency of tethered cord or other cord anomalies in asymptomatic, healthy infants with an isolated simple sacral dimple. We hypothesized that the prevalence of underlying spine abnormalities in this population is negligible.

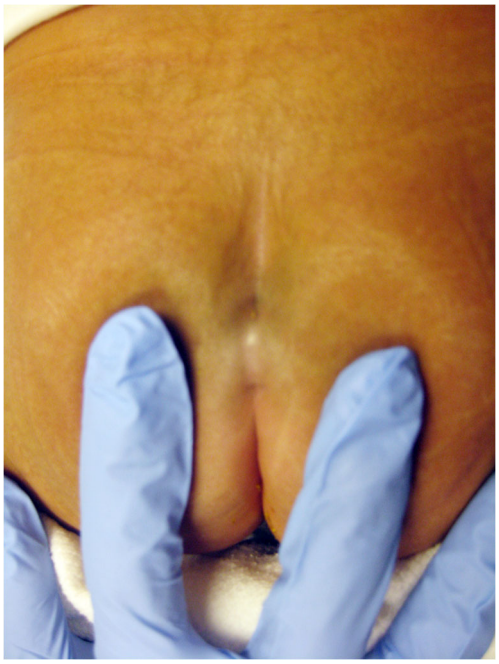

Fig. 1 Photograph of a simple sacral dimple in a 1-month-old boy

Materials and methods

We performed a multi-institutional cohort study after approval by the institutional review boards of both institutions. A medical word search was used to find all patients referred to Nationwide Children's Hospital (Columbus, OH) between Sept. 1, 2000, and Aug. 31, 2010, and Cincinnati Children's Hospital Medical Center (Cincinnati, OH) between Jan. 1, 2000, and July 31, 2012, for a screening lumbar spine ultrasound with the indication of a simple sacral dimple. While we had no strict age limitation for inclusion into the study, ultrasound of the lumbar spine is generally not performed at our institutions after the age of 6 months due to ossification of the posterior elements of the spine by this age. The medical record of each patient was reviewed to evaluate for the presence of other medical conditions; patients found to have other conditions such as congenital abnormalities, syndromes or chromosomal anomalies were excluded from the study.

All human studies have been approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its amendments. The legal guardian of the patient photographed gave informed consent to have the photograph published.

We reviewed the lumbar spine US reports of all children included in the study. At both institutions, standard US examinations of the lumbar spine were performed using high-resolution, 8- to 14-MHz linear-array transducers (variety of machines). A sonogram was considered abnormal for any of the following reasons: a conus medullaris terminating below the L2–L3 disc space, decreased motion of the conus or nerve roots, a fat-containing filum terminale or a filum thickened >2 mm, or the presence of an intraspinal mass, osseous dysraphism (best evaluated for on transverse US images), or a subcutaneous tract leading to the thecal sac. In children with transitional anatomy with a discrepancy between the level of the conus when counted from the last rib versus when counted from the sacrum, the lowest conus position was assumed. Normal variants such as a filar cyst, ventricularis terminalis, or a pilonidal tract (defined as a residual cord-like region composed of fibrous tissue extending from the skin dimple to the coccyx without connection to thecal sac) were not considered abnormal for the purposes of this study.

Additional spine imaging studies performed, including follow-up US or MRI, were reviewed in all children with an abnormal screening sonogram. When there was a discrepancy between the US and MRI findings, MRI was considered to be the gold standard. The medical records of these children were queried for neurosurgical consultation, physical examination findings, and operative reports.

We used descriptive analysis to calculate the percentages of children considered to have abnormalities, and 95% confidence intervals were also provided for the proportions.

Results

During the 12-year period of our study, 3,991 children underwent screening lumbar spine sonography for the indication of a simple sacral dimple. Of these, 107 (2.7%) were excluded because of the presence of congenital anomalies.

Of the remaining 3,884 healthy children, 3,751 (96.6%) had normal initial screening sonograms and 133 (3.4%) had abnormal sonograms. Five children with abnormal screening sonograms were lost to follow-up and did not undergo follow-up imaging or neurosurgical evaluation at either institution. The remaining 128 children underwent either follow-up US or MRI, with both the modality and timing of the follow-up imaging study determined by each child's ordering physician. Fifty-two children were subsequently shown to be normal on follow-up imaging, leaving only 76/3,884 (2.1%) children with abnormalities.

All 76 of the children with abnormalities underwent neurosurgical consultation. Seventy-one of these children were deemed normal by neurosurgery and did not undergo surgical intervention. The most common imaging finding in this group was an isolated low conus without other signs of tethering, in 49 children (Fig. 2). Of these children considered to have a low conus, 5/49 (10.2%) had transitional anatomy with six lumbar vertebrae. Other imaging findings in this group included 18 children with an isolated fatty filum (fibrolipoma) (Fig. 3), 2 with both a low-lying conus and a fatty filum, and 2 with subjective decreased motion of the conus but without other signs of tethering. Five children underwent surgical intervention. Figure 4 summarizes the imaging findings of healthy children who underwent screening lumbar spine sonography.

The incidence of surgical de-tethering in an otherwise healthy child presenting with an isolated simple sacral dimple was only 5/3,884 (0.13%, 95% CI: 0–0.27%). Of these five children, patient 1 demonstrated a nondependent conus at L2–L3 with decreased motion of the nerve roots and an enlarging ventricularis terminalis. This 9-month-old was asymptomatic at the time de-tethering surgery was performed. As an infant, patient 2 was shown to have transitional anatomy with limited motion of the conus, which terminated at L2–L3. At 6 years of age, this child underwent de-tethering for intermittent urinary and fecal incontinence, despite normal urodynamic studies, which did not indicate a neurogenic bladder. The child's symptoms did not improve after surgery. Patient 3 had transitional anatomy with a nondependent conus at L3 and a 3-mm filar lipoma. At the time of preventative de-tethering, this 1-year-old was asymptomatic with normal urodynamic testing. Patient 4 had a nondependent conus at L4–L5 and was also asymptomatic at the time of surgery. Patient 5 had a conus at L2–L3 with a 2.8-mm dilation of the central canal of the distal cord. This 5-month-old was asymptomatic at the time of de-tethering. Of the five children who underwent surgery, four were found at surgery to have a tethered cord. Table 1 summarizes the imaging and neurosurgical findings of these five children.

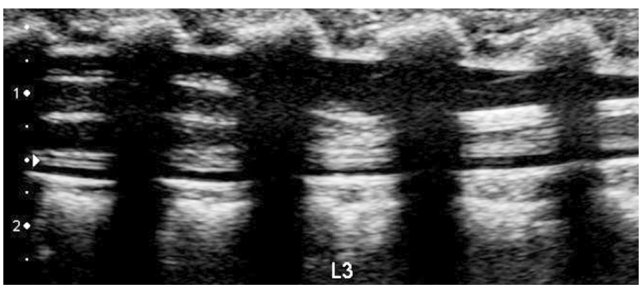

Fig. 2 Sonogram of a 2.5-month-old girl shows an isolated low conus, terminating at the mid-body of L3

Discussion

Simple sacral dimples identified in asymptomatic, healthy infants are infrequently associated with spinal dysraphism. From our large cohort of asymptomatic children with simple sacral dimples, only 0.13% of cases led to surgical intervention (with all children being essentially asymptomatic), confirming our hypothesis that the frequency of underlying spine abnormalities in this population is rare. Additionally, recent literature has shown that an isolated low conus between L2–L3 and mid L3 is seen in approximately 12% of screening lumbar spine sonograms and suggests that this finding is a normal variant [10]. Studies have also shown that small filar lipomas are an incidental finding in 4–6% of otherwise normal patients [11, 12].

Previously published studies regarding the association of simple sacral dimples and tethered cord support the data acquired from our study [1–4, 6, 8, 13–22]. Eight major studies have evaluated the relationship of a simple sacral dimple to tethered cord for a total of 2,357 patients, none of whom had imaging findings that led to surgical intervention [3, 6, 8, 13–17]. Two other studies did not distinguish between simple and more complex dimples [1, 4], including a recent article in which 5/638 patients with a dimple underwent surgery [1]. However, given that the methodology of this study did not define the location, nature or number of dimples in each patient, it is likely that some of these dimples would not have met the criteria for a simple sacral dimple. Furthermore, it is unclear what criteria were used for surgical intervention. However, even when the results of these two studies are combined with those of the other eight studies, the overall incidence of surgery in patients with dimples was still only

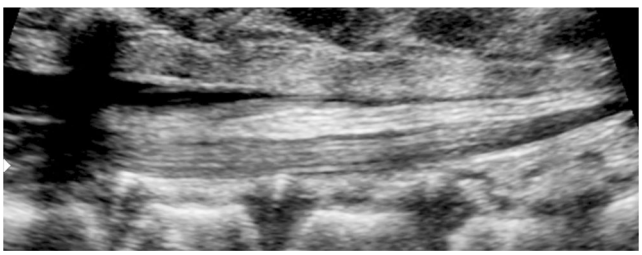

Fig. 3 Sonogram of a 2-week-old boy shows a fatty filum

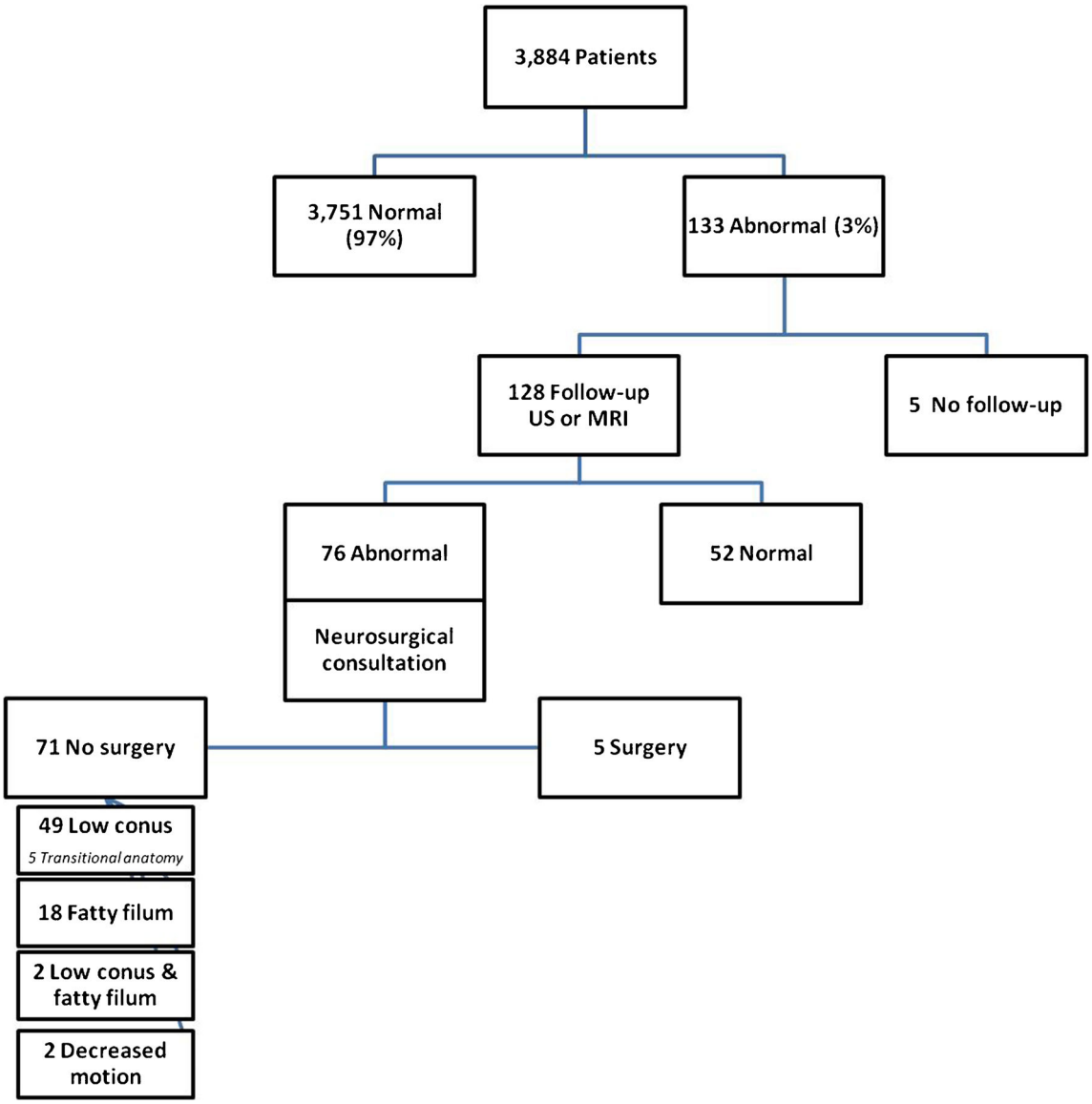

Fig. 4 Summary of results of screening spine sonography in healthy children

Table 1 Children who underwent de-tethering

	Age and symptoms at time of surgery	Imaging findings	Neurosurgical findings
Patient 1	9-month-old female Asymptomatic	Nondependent conus at L2–L3, decreased motion of nerve roots, enlarging ventricularis terminalis	Dilated ventricularis terminalis, tethered cord
Patient 2	6-year-old female Intermittent urinary and fecal incontinence, normal urodynamic studies, symptoms did not improve after surgery	Transitional anatomy, conus at L2–L3, decreased motion of conus	Mildly thickened and taut-appearing filum
Patient 3	1-year-old female Asymptomatic, normal urodynamic testing	Transitional anatomy, nondependent conus at L3, 3-mm filar lipoma	Tethered cord, fatty filum
Patient 4	1-year-old male Asymptomatic	Nondependent conus at L4	Tethered cord
Patient 5	5-month-old male Asymptomatic	Conus at L2–L3 2.8-mm dilated central canal of distal cord	Cord not tethered, arachnoid cyst associated with filum, peculiar brownish tissue along the filum with pathology showing pigmented cells and neurons

Table 2 Representative studies on the frequency of surgical intervention in children with sacral dimples

Author, year	Type of study	Criteria for simple sacral dimple used?	Number with dimples	Number requiring surgery	Abnormal findings on imaging
Ben-Sira et al. 2009 [6]	US	Yes	109	0	1 echogenic filum
Chern et al. 2012 [1]	US	**No**	638	5	3 fatty filum 1 dermal sinus tract 1 fibrous tract to muscle
Gibson et al. 1995 [17]	US	Yes	51	0	1 isolated low conus
Guggisberg et al. 2004 [16]	Various imaging modalities	Yes	3	0	–
Henriques et al. 2005 [4]	US	**No**	32	0	1 dermal sinus
Herman et al. 1993 [15]	US	Yes	53	0	–
	literature review[a] from surgical series	Yes	845	0	–
Kriss and Desai 1998 [3]	US	Yes	160	0	–
Robinson et al. 2005 [8]	US	Yes	86	0	–
Weprin and Oakes 2000 [14]	Physical exam and history	Yes	1,000	0	–
TOTAL			3,027	5	

[a] Literature review based on papers by Goodall [18], Manterola et al. [19], McLaren [20], Solla and Rothenberger [21] and Kooistra [22]

5/3,027 (0.17%), which is similar to the findings in our study. The sacral dimple literature is summarized in Table 2.

Although US imaging has been reported to be a cost-effective screening method in infants with suspected occult spinal dysraphism [7], it is interesting to note the potential cost savings in our study if US had not been performed on our healthy patient population with simple sacral dimples. The current fee for a spine sonogram at Cincinnati Children's Hospital Medical Center is $846 (technical fees = $525.65; professional fees = $320.35), with an average reimbursement of 55%. An average reimbursement of $465.30 multiplied by the 3,874 patients that eventually were deemed to be normal cost the health care system $1,802,572.20. This does not include the additional costs of follow-up sonography, MRI, sedation or neurosurgical consultation. Additionally, the parental stress that could have been avoided from these studies is priceless.

Our review of 3,884 healthy infants with simple sacral dimples is the largest study to date evaluating the relationship of a simple sacral dimple to tethered cord syndrome. However, there are a few limitations. First, five children with abnormal screening sonograms were lost to follow-up. However because Cincinnati Children's Hospital Medical Center and Nationwide Children's Hospital are the only subspecialty centers for pediatric neurosurgery in each of their respective metropolitan areas, it is highly likely that any child with tethered cord would have been operated upon at our institutions. Second, because of the retrospective nature of this study and the inability of the authors to examine the children's sacral dimples, we relied on the ordering clinician to determine whether the sacral dimple was simple in nature. Although it is possible that some of the children included in our study might not have met the criteria for a simple sacral dimple, elimination of these children may have actually strengthened our results, assuming they accounted for some of the abnormal sonograms. Additionally, inclusion of all children referred for simple sacral dimples as deemed by general pediatricians who may not have a particular area of expertise in cutaneous stigmata validates that the results of this study may be easily applied to general pediatric practices.

Further investigation and consideration of what constitutes an abnormal lumbar sonogram may be helpful. There are no clear-cut guidelines for the management of several of the minor abnormalities that can be seen on screening lumbar spine sonography, including an isolated low-lying conus and a fatty filum. Additionally, although approximately 16% of infants have been shown to have some type of transitional anatomy of the lumbosacral spine [23], no study has evaluated the significance of a low conus with a discrepant level when counting from the last rib versus from the sacrum. Thus the clinical significance of many of the patients considered to have an abnormal screening sonogram in our study is in question. Future work and guidelines in these areas may be helpful to guide the optimal management of these children.

Conclusion

Isolated simple sacral dimples in the asymptomatic healthy neonatal population are common. The risk of underlying significant spinal malformations in this patient population is exceedingly low.

Conflicts of interest None

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