



A Review of the use of Ultrasound Imaging of the Spine in Neonates with Sacral Cutaneous Findings

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Abstract

Background: Ultrasound imaging of the spine is frequently used to rule out underlying spinal dysraphisms in newborns with sacral cutaneous findings - majority of which are sacral dimples.

Aim: To assess the relationship between sacral findings on newborn examination and presence of spinal dysraphism, thereby optimizing the use of ultrasound imaging.

Methods: We reviewed the clinical records of newborns managed between January 2013 and December 2019, who were found to have sacral findings on newborn examination. The number and types of sacral cutaneous findings, results of ultrasound spine imaging if performed, follow up status and neurodevelopmental outcomes of these infants were examined.

Results: Out of 12227 newborns, 332 (2.7%) were found to have sacral findings on examination. Among which, 307 had single cutaneous lesion, while 25 had two cutaneous lesions. Majority (80%) of those with single lesion are isolated sacral dimples. Of those with single lesion, 129 (42%) were further evaluated with ultrasound imaging of the spine, all were normal except one was reported to have a low lying cord. Of those with two lesions, 15 (60%) were further evaluated and all had normal scans. None of the patients in our study population were found to have neurodevelopmental concerns associated with spinal dysraphism on follow up.

Conclusion: Ultrasound imaging is not routinely indicated in infants with an isolated sacral dimple. Larger population studies may be needed to determine whether there is significant association between multiple cutaneous lesions, or complex sacral dimples, and the presence of occult spinal dysraphism.

Keywords: Neonates; Ultrasound spine; Sacral cutaneous finding; Spinal dysraphism

Introduction

Ultrasonographic imaging of the spine is frequently used to look for underlying spinal dysraphism in newborns found to have cutaneous signs on spine examination. Spinal dysraphism occurs from the failure of fusion of underlying vertebral bodies due to abnormal fusion of the posterior vertebral arches [1]. This can result in herniation of the meninges, sometimes along with the spinal cord. In open spinal dysraphisms, herniated neural tissue may be exposed. In occult spinal dysraphisms, the overlying skin is intact and neural tissue remains unexposed [1]. A case series published by Soonawala et al. [2] of 47 patients with occult spinal dysraphisms and mean age of diagnosis at 2 year-old found that the commonest presentation were cutaneous lesions (60%) and symptoms of tethered cord neurological, urological or orthopedic (70%). Younger children tend to present with cutaneous lesions picked up in the post-natal period that led to further evaluation, while older children may present with either cutaneous lesions or neurological deficits [3].

In general, 4% to 7% of healthy newborns are found to have one or more of these cutaneous signs, of which approximately 75% of them are isolated sacral dimples [4]. The prevalence of sacral dimples in newborns has been reported to be around 2% to 5% [5]. Other cutaneous signs include tuft of hair, deviated gluteal fold, sacral mass, lipoma, hemangioma, skin discoloration and skin tags. Over the years, many studies have been done to look for associations between cutaneous lesions

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Received Date: 07 Dec 2020

Accepted Date: 08 Jan 2021

Published Date: 15 Jan 2021

Citation:

Cherie C, Grace Tan M, Cheo Lian Y. A Review of the use of Ultrasound Imaging of the Spine in Neonates with Sacral Cutaneous Findings. *Ann Pediatr Res.* 2021; 5(1): 1053.

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and presence of spinal dysraphism. There is evidence in literature showing that isolated sacral dimples are not associated with spinal dysraphisms on ultrasound, and many advocate against routine use of ultrasound imaging for these babies [6-10]. In 2018, the Choosing Wisely Canada Pediatric Neurosurgery committee published a list of five recommendations with the aim of improving quality of healthcare by reducing unnecessary resource utilization and healthcare costs [5]. One of the recommendations was to not image a midline dimple related to the coccyx in an asymptomatic infant or child, with evidence showing that there is no increased risk of occult spinal dysraphism compared to general population of infants cited. On the other hand, presence of 2 or more cutaneous lesions has been reported to be associated with a higher risk of spinal dysraphism [11,12], and ultrasound screening has been recommended in this group of infants.

In our center, depending on what the physical findings are, newborns found to have cutaneous lesions on spine examination are sometimes further evaluated with an ultrasound. We conducted a review on the (i) use of ultrasound imaging in these patients, (ii) results of the scans performed and (iii) neurodevelopmental outcomes of these patients, to identify any potential association between the presence of cutaneous lesions and underlying spinal dysraphism. With these results, we aim to optimize the use of radiological tests, reduce unnecessary healthcare costs, and avoid unwarranted parental anxiety; ultimately improving the quality of patient care.

Materials and Methods

This was a retrospective review that included all infants born in the Department of Neonatal and Developmental Medicine in Singapore General Hospital (SGH) between January 2013 and December 2019. Data of 332 patients who were found to have spinal cutaneous lesions on newborn screening examination were extracted from a secured online database (REDCap). Information obtained included their gestational age at birth, birth weight, maternal antenatal risk factors, cutaneous lesions recorded from examination, any ultrasound imaging of the spine performed, date and results of scan if performed. Follow up status and details of issues noted on follow up (including, but not limited to, any neurodevelopmental concerns) were gathered from electronic medical records found on secured platforms that are accessible across all public healthcare institutions. These, however, do not include records of visits to private healthcare facilities.

Results

Clinical characteristics

A total of 12227 infants were managed between January 2013 and December 2019 in our center. Of which, 332 (2.7%) were found to have cutaneous lesions on screening examination performed between 24 h to 48 h of life- 36 were preterm infants (less than 37 weeks' gestation at birth), and 296 were term. Out of these 332 infants, 307 had single cutaneous lesion, while 25 had two cutaneous lesions. None of our infants had more than 2 findings on examination.

Within the group of 307 infants with single cutaneous lesions, 246 had a sacral dimple, 34 had deviated gluteal cleft, 24 had tuft of hair, 1 had a sacral nevus, 1 had sacral puckering and 1 was described to have sacral fullness. The 129 (42%) out of 307 of these infants were further evaluated with ultrasound imaging of the spine. Figure 1 shows the number of patients within each of these groups who did and did not have ultrasound imaging done.

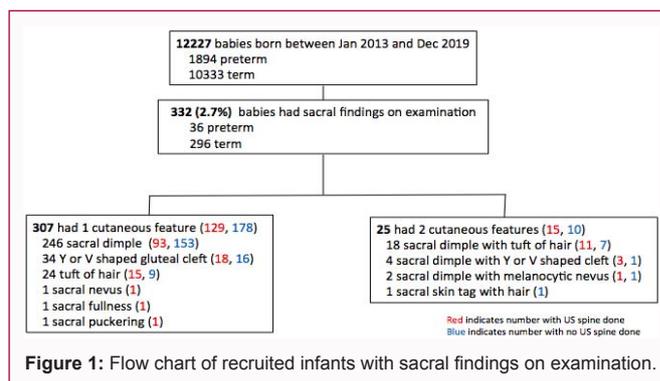


Figure 1: Flow chart of recruited infants with sacral findings on examination.

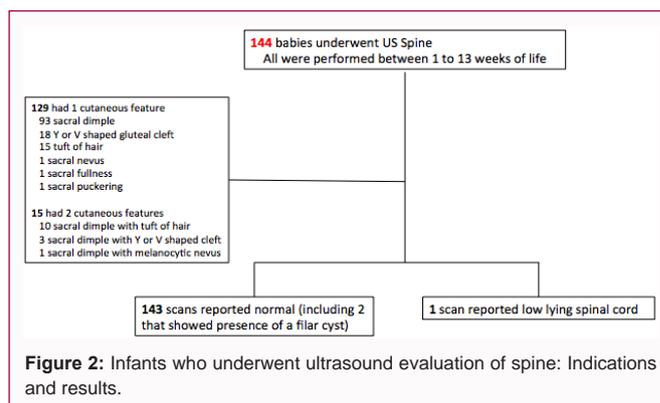


Figure 2: Infants who underwent ultrasound evaluation of spine: Indications and results.

Within the group of 25 infants with two cutaneous lesions, 18 had sacral dimple with a tuft of hair, 4 had sacral dimple with deviated gluteal cleft, 2 had sacral dimple with melanocytic nevus, and 1 had sacral skin tag with tuft of hair. Out of these infants, 15 (60%) were further evaluated with ultrasound imaging of the spine. Figure 1 shows the number of patients within this group who did and did not have ultrasound imaging done.

Ultrasound findings

Among the 332 infants who had cutaneous lesions found on examination, 144 (43.3%) underwent ultrasound imaging of the spine. All scans were performed between 1 to 13 weeks of life. Of those who had ultrasound imaging, 129 had single cutaneous lesion as the indication for scan, while 15 had two cutaneous lesions as the indication. Figure 2 shows the breakdown of various indications for those who had ultrasound imaging done. Majority of the scans (143 out of 144, 99.3%) were reported normal, including 2 that showed presence of a filar cyst. Only 1 scan was reported to show a low-lying spinal cord, this was in a patient who had an isolated sacral dimple. None of these patients required further ultrasound or magnetic resonance imaging.

Of infants with two cutaneous lesions, all 15 who had ultrasound evaluation showed normal scan findings.

Follow up outcomes

We reviewed the follow up status and outcomes of all patients with cutaneous lesions. Of 332 infants, 69 had no electronic medical records found indicative of follow up, 45 had routine well child follow up in public healthcare facilities and were discharged well before 2 years of age with no further healthcare visits, 218 had follow up or healthcare visits beyond 2 years of life. The 195 out of these 218 had no developmental concerns on follow up; while 23 had (7 were born preterm and 16 term). Details of the developmental outcomes of these infants can be found in Table 1. Within these 23 children, 10 had

Table 1: Outcomes of infants with developmental delay.

No	Gestational age (week + day)	Spinal Examination Findings	US Spine Findings	Neurodevelopmental outcome	Age at diagnosis (year/month)	Maternal/Antenatal risk factors
1	27+3	Sacral dimple	Normal	Global developmental delay	2y	Type 2 Diabetes Mellitus, Chronic renal failure, Anemia
2	27+5	Sacral dimple	Not done	Speech delay	1y 10m	-
3	31+0	Sacral dimple	Low lying spine	Global developmental delay	3y 3m	Twin to Twin transfusion syndrome
4	32+4	Sacral dimple	Not done	Pervasive developmental disorders	2y 11m	-
5	33+0	Sacral dimple	Not done	Speech delay	2y 9m	Gestational Diabetes on insulin
6	36+3	Sacral dimple	Normal	Global developmental delay	2y 5m	-
7	36+4	Sacral dimple	Not done	Speech delay	2y 6m	-
8	37+2	Sacral dimple	Normal	Selective mutism	3y 3m	Gestational Diabetes on insulin
9	37+6	Sacral dimple and Y shaped gluceal cleft	Normal	Autism spectrum disorder	2y 11m	Placenta previa major
10	38+0	Sacral dimple	Not done	Autism spectrum disorder	2y 2m	-
11	38+0	Sacral dimple	Normal	Global developmental delay	11m	-
12	38+1	Y shaped gluceal cleft	Not done	Speech articulation concern	4y	-
13	38+3	Y shaped gluceal cleft	Not done	Language Delay	3y 5m	Maternal thalassemia
14	39+0	Sacral dimple	Normal	Pervasive developmental disorders	4y 6m	-
15	39+0	Sacral dimple	Not done	Autism spectrum disorder	3y 1m	-
16	39+0	Sacral dimple	Not done	Speech and Language delay	5y	Maternal Hepatitis B carrier, anemia
17	39+0	Tuft of hair	Filar cyst	Autism spectrum disorder	3y	-
18	39+0	Sacral dimple	Not done	Global developmental delay	3y 1m	Antenatally diagnosed fetal hydrops, well at birth
19	39+3	Sacral dimple and Y shaped gluceal cleft	Normal	Autism spectrum disorder	2y 7m	Maternal hypothyroidism
20	40+0	Sacral dimple	Not done	Global developmental delay	4y 5m	-
21	40+0	Sacral dimple	Not done	Behavioural emotional disorder	5y 5m	-
22	40+0	Sacral dimple	Not done	Autism spectrum disorder and Global developmental delay	3y 2m	-
23	41+4	Sacral dimple with tuft of hair	Normal	Speech delay	3y	-

ultrasound imaging previously done (9 were normal and 1 had low lying cord), while 13 had no scans done.

We further reviewed the outcomes of the 13 patients with developmental delay and no previous ultrasound imaging done. The 5 had speech and language delay, 1 had speech articulation difficulties, 4 had autism spectrum disorder, 1 had global developmental delay and 1 had behavioral emotional disorder of childhood. None of these patients were seen or noted to have neurological, urological, bowel or orthopedic concerns relating to possible spinal cord pathology. We infer that the developmental concerns in these children are unlikely to be related to the cutaneous lesions of the spine.

The patient with abnormal ultrasound finding of low-lying spinal cord with global developmental delay was a preterm infant born at 31 weeks' gestation. There was no surgical intervention required for the spinal finding, and the delay in development may be attributable to prematurity.

Discussion

Isolated sacral dimple not indicative of increased risk of OSA (occult spinal dysraphism)

While various cutaneous stigmata have been reported in cases of spinal dysraphisms, there is an increasing amount of research evidence showing that infants with an isolated sacral dimple do not have an increased risk of spinal dysraphism. Studies published by Moore et al. [7], Beh et al. [6], Sneineh et al. [9] and Gibson et al.

[13] looked at the relationship between sacral examination findings and results on ultrasound imaging, and the common finding from all these papers was that none of the infants with isolated sacral dimples were found to have spinal dysraphism on ultrasound. Liat et al. [8] assessed the utility of ultrasound imaging in 157 neonates found to have either a sacral dimple or deviated gluteal fold and similarly found that none of them demonstrated any clinically significant pathological findings. Authors from these studies proposed that routine use of ultrasound screening is not indicated in babies with isolated sacral dimples and many called for revised guidelines to optimize resource utilization.

Results from our study similarly showed that isolated sacral dimples are not associated with spinal dysraphism, as all but one of our patients who had spinal imaging done for isolated sacral dimples had normal scan results. The remaining one patient was reported to have a low-lying cord that did not require surgical intervention nor resulted in any neurological disability.

Presence of 2 or more cutaneous lesions a stronger marker of OSD

McGovern et al. [11] analyzed data of 216 patients with sacral findings referred for ultrasound imaging and found that there was no significant correlation between the presence of a sacral dimple and presence of dysraphism. Instead, it found that infants with more than one clinical marker on examination were 6 times more likely to have dysraphism compared to those imaged based on a single marker (OR 6.0, 95% CI 1.289 to 27.922, p=0.022). Along the same vein, a

review of 54 cases published by Guggisberg et al. [12] detected Occult Spinal Dysraphism (OSD) in 3 of 36 (8.3%) patients with isolated congenital midline lesion, in contrast to 11 of 18 (61.1%) patients with combination of 2 or more different skin lesions supporting the hypothesis that a combination of 2 or more congenital midline lesion is a stronger marker of OSD.

In our study, all patients who had ultrasound screening done for 2 cutaneous lesions had normal ultrasound finding. Those who had 2 cutaneous lesions but not evaluated further with ultrasound imaging were not found to have any follow up concerns, up to the time this study was conducted (patient ages between 6 months to 7.5 years old), related to possible missed spinal dysraphism. In other words, from our data, infants with 2 cutaneous lesions were not found to be at higher risk of spinal dysraphism. However, the limitation of our study is the small sample size; and we did not have infants with more than 2 cutaneous lesions for further analysis.

Complex dimples higher risk of OSD than simple dimples

A study published by Kriss et al. [14] in 1998 categorized sacral dimples into simple and complex. Complex dimples are large (>5 mm in diameter), high (>2.5 cm from anus), or appeared in combination with lesions. In absence of these features, a sacral dimple would be considered simple. In this study, 8 (40%) out of 20 of those with complex dimples were positive for spinal dysraphism, while none of the neonates with a simple dimple had spinal dysraphism.

In our center, findings of multiple cutaneous lesions are recorded, but measurement of the size of a sacral dimple and its distance from anus is not a routine practice. There would be concerns of inter-observer variability and accuracy of measurement. From our review of the current literature, there are limited information available comparing complex and simple sacral dimples. Further prospective studies may be needed to help determine if the above characteristics makes a sacral dimple more likely to be associated with spinal dysraphism.

Identifying patients who may benefit from ultrasound screening to rule out OSD

The main finding from our retrospective review is consistent with that of other published studies: The presence of an isolated sacral dimple is not associated with increased risk of occult spinal dysraphism and ultrasound imaging is unlikely to pick up clinically significant pathology in these cases. All, but one infant, with isolated sacral dimples and further evaluated with ultrasound had normal scan findings. In that one case, ultrasound showed a low-lying spinal cord which did not warrant further investigations or surgical intervention. Thus, in infants with isolated sacral dimple and who are otherwise healthy (no concerns of congenital syndromes or neurological deficits), we propose that parents be assured and ultrasound evaluation is not needed [15,16].

Our study also did not find an increased risk of spinal dysraphisms in infants with 2 cutaneous lesions. However in view of our small sample size and results from other studies that have shown significant association between the two (although sample sizes in these studies are not large), we would still recommend for ultrasound evaluation for cases with multiple cutaneous lesions, until there is more evidence to safely absolve this group of infants from routine ultrasound evaluation.

Ultrasound evaluation should still be performed in infants with: Antenatal scans showing abnormal spine anatomy, presence of other

congenital anomalies that can be associated with spinal dysraphism (e.g. VACTERL), or neurological deficits that localize to a spinal pathology.

Limitation of Study

The limitations of our study include a small sample size, as well as the lack of information on visits to private healthcare institutions due to issues regarding accessibility to those medical records. However, it is unlikely that those who visited private healthcare institutions would make up a significant proportion of the whole population. Given that vast majority of children in Singapore are reviewed in public healthcare institutions for developmental assessment and immunization for first two years of life, and under the purview of School Health Screening services when they start elementary school, it is again unlikely that the diagnosis of spinal dysraphism is not captured in the medical records.

Conclusion

In developing a cost-effective workflow that allows better utilization of resources, reduction in healthcare costs and reduction in unnecessary parental anxiety, we recommend against routine ultrasound imaging of infants with isolated sacral dimples. Larger population studies may be needed to determine relationships between other perceived risk factors (e.g. multiple cutaneous lesions or complex sacral dimple) and the presence of OSD.

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