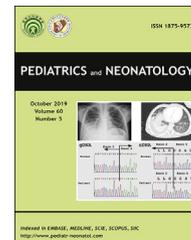




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Original Article

Clinical significance of asymmetric skin folds in the medial thigh for the infantile screening of developmental dysplasia of the hip



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Key Words

congenital;
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Background: The presence of asymmetric skin folds in the medial thigh (ASM) is often used as one of the screening items for developmental dysplasia of the hip (DDH). However, few studies have evaluated the clinical relevance of ASM in DDH.

Methods: We reviewed the data of 584 infants aged 4–12 months who were referred to our institute because of suspicious findings in periodic infantile DDH screenings. The reasons for referral were classified into ASM, gluteal asymmetry, limb-length discrepancy, limitation of hip abduction, and click/clunk of the hip. A simple radiograph and hip examination including stability assessment were evaluated in all the infants by an experienced pediatric orthopedic surgeon.

Results: Most of the referred infants presented with ASM only (78.4%, 458/584), but none of them were diagnosed with DDH. On the other hand, about 18.3% of infants referred for other reason(s) were diagnosed with DDH.

Conclusion: Although the presence of ASM was the most common reason for referral, it had little clinical significance as a screening item for DDH in the infants aged 4–12 months. Therefore, ASM alone is not a clinical indicator of DDH.

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1. Introduction

Developmental dysplasia of the hip (DDH) is a spectrum of disease that affects the developing hip joint ranging from minor looseness of the ligament to complete dislocation.^{1,2} The prognosis is unpredictable,¹ but patients who are diagnosed and treated late have poorer outcomes, and increased rates of surgery and complications.^{3–6} Therefore, many countries recommend periodic infantile hip screening for DDH even though there are no suspicious findings during newborn period.^{5,7–9}

The physical findings of patients with DDH vary by age, and infantile DDH screening also reflects these changes.^{7,8} While hip clunk (Ortolani or Barlow sign) is the most important suggestive finding for DDH in neonates, physical signs such as Galeazzi sign or limb-length discrepancy (LLD), asymmetry of the gluteal, thigh or labial folds, and limited hip abduction are more indicative in infants older than 3–4 months.^{7,8}

Although there is debate,¹⁰ an asymmetric skin fold in the medial thigh (ASM) has been described as one of suggestive findings for infantile DDH in many textbooks.^{11,12} Therefore, the presence of ASM has been thought to be an important finding for infantile DDH screening in many countries.^{1,7} In many Asian countries, ASM is considered to be an important suggestive finding, and health screening systems officially recommend referral of the infants with ASM to pediatric orthopaedic surgeons.¹³ In our experience, the presence of ASM seems to be the most common referral reason of infantile DDH screening, because of its relative convenience for pediatricians and primary physicians (Fig. 1); however, contrary to expectations, few studies have actually evaluated its clinical significance in DDH.



Figure 1 An asymmetric skin fold in the medial thigh (ASM). A 6-month-old girl referred for the presence of ASM. After physical and radiological evaluations, she was diagnosed as not having DDH. It is easy and relatively objective to detect the presence of this kind of asymmetry, but few studies support its clinical relevance in DDH.

Pursuant to these findings, we evaluated infants who were referred due to clinical suspicion of DDH upon screening, and we investigated the referral reasons, especially focusing on ASM, and the clinical significance of ASM as a diagnostic indicator of DDH. In addition, we evaluated the clinical course of infants who had a stable hip but mild acetabular dysplasia.

2. Methods

2.1. Authors' protocol for the infants referred under the suspicion of DDH

Our hospital is a tertiary referral hospital with a specialist team for pediatric orthopedic surgery. Since 2009, we have prospectively gathered data from neonates and infants (aged 0–12 months) who were referred for suspicion of DDH. The data included referral reasons, physical findings, radiographic data, and diagnoses. Referral reasons were recorded in medical charts by special typists during the preliminary medical interview. One surgeon, a professor of pediatric orthopedic surgery with more than fifteen years of experience, performed all of the physical and radiographic examinations and diagnosed all of the infants. For infants aged 4 months or older, we routinely took a simple radiograph of the anteroposterior (AP) view of both hips.

2.2. Study population

The study protocol was approved by the institutional review board of our institute (IRB No. 2015-1081). Informed consent was waived by the IRB. We retrospectively reviewed the prospectively-gathered data of 662 infants, aged 12 months or younger, who were referred to our institute from January 2010 to December 2014 because of abnormal findings on the DDH screening test. There were no cases of teratologic, neuromuscular, and/or syndromic hip dysplasia, possibly because the screening was aimed at normal infants. Among them, 78 infants aged <4 months were excluded because of lack of consistency/reliability of the radiographic evaluation and physical examinations originating from invisible femoral epiphyseal nucleus^{14,15} and their inborn ligamentous laxity.⁷ Finally, 584 referred infants were analyzed, and there were no missing data or radiographs in any of the included cases.

2.3. Investigated variables

The referral reasons were categorized into ASM, gluteal asymmetry, LLD, limited hip abduction, and hip click/clunk. These characteristics were selected based on previous studies^{5,7–9,16} and were modified based on discussions between two experienced pediatric orthopedic surgeons working in a tertiary general hospital. Other referral reasons that did not seem to be clinically significant, such as habitual posture, suspicion of thick extremities, or delayed development, were ignored. 'ASM' included asymmetric skin fold in the medial thigh at any level (proximal/middle/distal) (Fig. 1). 'Gluteal asymmetry' included asymmetric inguinal/gluteal/ischiogluteal skin fold and suspicion of

differences in the size and shape of the buttocks. 'LLD' included suspicion of LLD and a positive Galeazzi/Allis sign. 'Limited hip abduction' included any suspicion of limited motion of the hip regardless of the degree and the position. Difficulty diapering was also included in this category. Any snapping or clicking/clunking sounds/feeling in the hip, and suspicion of a positive Barlow/Ortolani sign were included in the 'hip click/clunk' category. We included hip click in the category although hip click alone is not considered significant^{9,17} because we realized that many primary physicians seemed to be confused about the difference between hip click and clunk during the review.

Radiological parameters associated with DDH such as intactness of Shenton line, position/size of the femoral epiphyseal nucleus, and the acetabular index (AI) were evaluated and measured using AP radiographs of the hip. Those parameters were used as an adjunct for diagnosing DDH. For infants aged 4–6 months who were suspicious of DDH or present ambiguous radiographic findings, ultrasonography was also performed adjunctively for diagnosis. However, no strict guideline was used for indication of ultrasonography. For the stable hip with other parameters intact, AI was measured to determine the presence of acetabular dysplasia.^{14,15,18} In those cases, we used age-adjusted AI; AIs $\geq 32^\circ$ for infants aged 4 months, $\geq 28^\circ$ for those aged 5–6 months and $\geq 25^\circ$ for those aged 7–12 months were regarded as high or increased AI.¹⁹ If high/increased AI decreased and scored within the above-mentioned range during follow-up, we used the term normalization of AI or normalized AI. For clarity, we referred to AI as age-adjusted AI when relative AI such as high, increased, or normalized was used. To test interobserver reliability for assessment of the AI, two observers performed assessments in 30 randomly selected cases. The interobserver reliabilities of the measurements were evaluated using intraclass correlation coefficients (ICCs). The ICC of interobserver reliabilities for the measurements was over 0.9, representing satisfactory agreement, which was in accordance with data from a previous study.¹⁴ Thus, measurements recorded in the medical chart, which were measured by a single investigator in the outpatient clinic, were used for the analyses.

2.4. Diagnosis/classification and follow-up

The referred infants were diagnosed based on physical findings and radiological examinations, and the diagnosis was assigned into three categories: DDH, mild acetabular dysplasia, or normal. Cases with an indeterminable diagnosis were followed up after 1–3 months, with physical examination and radiographs if necessary. Among the various conditions associated with DDH, diagnosis of 'DDH' was only confirmed for the subluxatable/dislocatable/dislocated types,⁹ and individualized treatments were performed for those patients. 'Mild acetabular dysplasia' was diagnosed for cases with increased AI but stable hip joint and intact Shenton line and other radiological parameters. Their parents were shown how to position the infantile hip in abduction if possible, and the infants were followed up every 3–6 months with physical examinations and radiological evaluations until normalization of AI. Infants who

showed no abnormalities on physical and radiological examinations were regarded as 'normal' and returned to their primary physician for periodic screening examinations.

2.5. Statistical analysis

The differences in frequencies were examined using the Fisher's exact test. Statistical analyses were conducted using SPSS for Windows statistical software (version 21; IBM Co., Armonk, NY). P values < 0.05 were considered statistically significant.

3. Results

3.1. Demographics, referral reasons, and diagnosis

The demographics, referral reasons, and diagnoses of the 584 infants are presented in Table 1. The presence of ASM was the most common referral reason ($n = 538$, 92.1%), and it was the only reason for referral in 78.4% (458 of 584).

3.2. Clinical significance of ASM as an indicator of DDH

Table 2 lists the diagnoses and referral reasons for patients with ASM. None of the 458 infants (0%) who presented with ASM as the only referral reason were diagnosed with DDH (Table 2) in contrast to those who were referred for other referral reason(s) with ASM (12.5%, Table 2) or without ASM (28.3%, Table 3). Overall, 18.3% (23/126) of infants who presented with other referral reason(s) were diagnosed with DDH.

3.3. Clinical course of infants with mild acetabular dysplasia

The clinical course of infants who showed mild acetabular dysplasia (stable hip but increased AI) is shown in Fig. 2. Among them, all the infants who were followed up showed improvement of the AI during follow-ups. The mean time for normalization of the AI was approximately 14 months.

4. Discussion

Although many physicians do not believe ASM to be clinically significant,¹⁰ the presence of ASM has been described as one of the significant suggestive findings for DDH,^{11,12} and it is currently used as one of the significant suggestive findings for DDH.^{1,7} In many Asian countries, ASM is still officially regarded as one of the significant check points for DDH screening as well as the other findings such as the limitation of abduction in flexion, family history related with hip disorders, female gender, and breech position at birth (<http://www.jpoa.org>, date late assessed January 09, 2016, and <http://health.mw.go.kr>, date late assessed January 09, 2016). However, few studies have evaluated its clinical significance in DDH. To the best of our knowledge, this is the first study to assess the clinical significance of ASM in infantile DDH, and to present the clinical course of infants with a stable hip but increased

Table 1 Demographics, referral reasons, and diagnosis.

Characteristics	number or mean \pm SD	proportion or range
Gender		
Boy	235	40.2%
Girl	349	59.8%
Age at initial presentation (months)	6.6 \pm 2.0	4–12
Referral reason (duplicated)		
ASM	538	92.1%
ASM as the only referral reason	458	78.4% ^a
Gluteal asymmetry	20	3.4%
LLD	56	9.6%
Limited hip abduction	22	3.8%
Hip click/clunk	35	6.0%
Follow-up duration (months) ^b	13.0 \pm 7.1	5–30
Diagnosis		
DDH	23	3.9%
Boy: Girl (proportion of girl)	3:20	(Girl 87.0%)
Age at initial presentation (months)	7.3 \pm 2.8	4–12
Both: Right: Left	3:11:9	13.0%: 47.8%: 39.1%
ASM as a referral reason	10	43.5%
Mild acetabular dysplasia	71	12.2%
Boy: Girl (proportion of girl)	8:63	(Girl 88.7%)
Age at initial presentation (months)	6.4 \pm 1.5	4–12
Both: Right: Left	41:13:17	57.7%: 18.3%: 23.9%
ASM as a referral reason	67	94.4%
Normal	490	83.9%
Boy: Girl (proportion of girl)	224: 266	(Girl 54.3%)
Age at initial presentation (months)	6.6 \pm 2.0	4–12
ASM as a referral reason	461	94.1%

SD, standard deviation; ASM asymmetric skin fold in the medial thigh; LLD, limb-length discrepancy; DDH, developmental dysplasia of the hip.

^a Among the whole 584 infants who were referred.

^b Applicable only for the infants with mild acetabular dysplasia.

AI. Additionally, although the data were retrospective, this study was planned prospectively, and initial physical and radiological evaluations were performed in all of the infants (no missed cases and no missed data).

Most of the infants (92.1%) in our current study were referred for the presence of ASM. The infants who presented with ASM as the only reason for referral comprised 78.4% of the study population, but none of those infants were diagnosed with DDH (Table 2). In contrast, about 18% of infants who were referred for other reasons were diagnosed with DDH (Tables 2 and 3). Although ASM constituted the majority of referral reasons, it appears to have little clinical significance as a diagnostic indicator of DDH, especially in infants aged 4–12 months. For infants who showed mild acetabular dysplasia (stable hip but increased AI), their follow-up data showed satisfactory clinical courses; all of the AIs normalized in a mean time of 14 months, and none of the infants showed development of instability or deterioration of AI.

We postulated the rationale for why ASM has been regarded as a significant indicator of DDH screening. When hip dislocation occurs, the femoral head moves upward so that the relative length of femur becomes shorter, thereby

possibly causing ASM. However, because this is theoretically caused by relative shortening of the femur, LLD or Galeazzi sign should be preceded and pronounced. Moreover, if dislocation occurs, asymmetric inguinal skin fold and ASM should accompany this because of superior migration of the femoral head. This supports the fact that ASM alone has little clinical significance.

We presumed that the presence of ASM was the most common reason for referral because of its relative objectivity and convenience for pediatricians, primary physicians, and even parents (Fig. 1). Some physical examinations, such as Ortolani/Barlow maneuvers, need training and experience, but ASM requires neither. Even parents can easily determine the presence/absence of ASM. The growth of the Internet has added a new dimension to the spread of worry among parents about DDH when their babies present with ASM. Therefore, the primary physician who screens infants with ASM may be forced to refer them to higher referral centers, even if the physician believes that it has little clinical significance.

The results of our study showed that ASM alone is not a clinical indicator of DDH; thus, this finding alone should not necessitate further evaluation for DDH. The results of the

Table 2 Referral reasons according to diagnosis in 538 infants who presented with ASM. There was no patient with DDH among the infants who were referred for ASM ALONE (in bold type). However, 12.5% (10/80) of the infants who were referred for ASM and other reason(s) were diagnosed with DDH (0% vs. 12.5%, $p < 0.001$).

Diagnosis (number, %)	Referral reason ^a	Number (%)
Without other referral reason (N = 458, 85.1%)^b		
DDH (n = 0, 0.0%)		
Mild acetabular dysplasia (n = 57, 12.4%) ^c		
Normal (n = 401, 87.6%)		
With other referral reason(s) (N = 80, 14.9%)		
DDH (n = 10, 12.5%)	Gluteal asymmetry	2 (20.0%)
	LLD	7 (70.0%)
	Limited hip abduction	1 (10.0%)
	Hip click/clunk	3 (30.0%)
Mild acetabular dysplasia (n = 10, 12.5%) ^d	Gluteal asymmetry	3 (30.0%)
	LLD	3 (30.0%)
	Limited hip abduction	1 (10.0%)
	Hip click/clunk	3 (30.0%)
Normal (n = 60, 75.0%)	Gluteal asymmetry	4 (6.7%)
	LLD	33 (55.0%)
	Limited hip abduction	9 (15.0%)
	Hip click/clunk	14 (23.3%)

% indicates the ratio of the just above categories.

^a Any referral reason other than ASM. Some cases are duplicated.

^b Among the 538 infants who were referred as ASM.

^c 47 patients (82.5%) were followed up, and their AI ultimately normalized (13.8 ± 7.6 months [5–30]). Four patients (7.0%) are still being followed up with (7.3 ± 2.2 months [5–10]), and six patients (10.5%) were lost to follow-up.

^d Four patients were followed up (6, 12, 18, and 19 months) and confirmed to have normalized AI. Four patients were followed (10.5 ± 5.7 months [6–18]) and showed improved, but still high AI; therefore they need more follow-up. The other two patients were lost to follow-up.

present study showed no different frequencies of ASM between the infants with mild acetabular dysplasia and a normal hip (Table 1). This result also supports that ASM alone rarely even indicates hip dysplasia unless it is associated with other findings. Some may argue that it is more important to be certain that the patient does not have DDH, because delayed diagnosis of this disease is generally associated with poor outcomes.^{5,20} Authors also agree that the sensitivity of DDH screening is more important than the specificity or effectiveness. However, none of the 458 infants who were referred for ASM alone were diagnosed with DDH in our study. The problem of low incidence of DDH can be raised in this regard, but we believe that its low incidence cannot decrease the significance of this result because they are referred population (not a normal population) from the screening. About 18% of the 126 infants who were referred for reasons other than ASM, however, were diagnosed with DDH. Additionally, the authors want to emphasize that ASM should not be confused with asymmetric gluteal folds, which were regarded as 'gluteal asymmetry' in the present study. The authors believe that asymmetric gluteal folds are an important finding suggesting hip dysplasia in infants and further studies such as ultrasound or simple radiographs are needed for further evaluation.

In our study, the infants with other physical findings but without ASM were more frequently diagnosed as DDH (28.3%) than those with other physical findings and ASM (12.5%). From a statistical point of view, ASM can be

misunderstood as having some protective roles against DDH. However, this might be because its presence caused the primary physician to be more sensitive towards ambiguous findings. For example, the physician may be more likely to suspect abnormal clicking, LLD, or limited hip motion in the presence of ASM. Therefore, some normal infants who only have ASM may more likely be suspected to have other abnormal findings, and this could lead to a decreased rate of DDH in the infants with ASM and other physical findings. Likewise, although about 95% of the infants without DDH presented ASM, less than half the infants with DDH presented ASM, which would result in bias.

There are some considerations when interpreting the results of this study. First, the diagnostic criteria for DDH should be noted. DDH refers to a spectrum of anatomical abnormalities ranging from dysplasia to a dislocated hip.^{5,9} Among various spectrum of DDH, only subluxatable/dislocatable/dislocated conditions were defined as DDH in this study. Second, we only included infants aged 4–12 months. Physical findings vary according to age. In newborns, the diagnosis is very ambiguous due to physiologic instability originating from the inborn laxity.^{1,7,8} Radiographic evaluations in infants under 4 months of age are also reported to be unreliable.^{14,15} Therefore, the authors believe that excluding the infants aged <4 months would contribute to more reliability and consistency in the present study. Third, we did not fully assess the risk factors for DDH such as female gender, family history, and breech position,^{7,8,21} which are considered important for screening.^{1,7–9}

Table 3 Referral reasons according to diagnosis in 46 infants who did not present with ASM. About 28% of them were diagnosed with DDH. It was also statistically different from that of the infants with ASM only (28.3% vs. 0%, $p < 0.001$).

Diagnosis (number, %)	Referral reason ^a	Number (%)
DDH (n = 13, 28.3%)	Gluteal asymmetry	5 (38.5%)
	LLD	3 (23.1%)
	Limited hip abduction	6 (46.2%)
	Hip click/clunk	7 (53.8%)
Mild acetabular dysplasia (n = 4, 8.7%) ^b	LLD	3 (75.0%)
	Hip click/clunk	1 (25.0%)
Normal (n = 29, 63.0%)	Gluteal asymmetry	6 (20.7%)
	LLD	7 (24.1%)
	Limited hip abduction	5 (17.2%)
	Hip click/clunk	7 (24.1%)

% indicates the ratio of the just above categories.

^a Any referral reason other than ASM. Some cases are duplicated.

^b Three patients were followed up (9, 9 and 15 months) and confirmed to have normalized AI. One patient was lost to follow-up.

However, the risk factors become of less value for diagnosing DDH if an infant already presents with positive findings on screening²² because the incidence of DDH in infants with those risk factors is also very low.⁷ Careful physical examination by experienced specialists and radiological evaluation are of diagnostic value for those infants.²² Therefore, the authors believe that the lack of data about risk factors would not substantially affect the results of our study. Fourth, the screening method was not regulated and may be inconsistent. The results of DDH screening are influenced by the experience and training of the examiner,⁷ but we could not regulate the consistency of

the screening methods or the quality of the examiner. We merely investigated the referral reason and its association with DDH. Although the quality of the examiner could not be assessed objectively, screenings were performed mostly by pediatricians and occasionally family medicine doctors.

In conclusion, although the presence of ASM was the most common reason for referral, it had little clinical significance as a diagnostic indicator of DDH, at least in infants aged 4–12 months. Infants with mild acetabular dysplasia and a stable hip are thought to have satisfactory clinical courses. This is the first study to assess the clinical significance of ASM as a screening tool for DDH, and to present the clinical course of infants with mild acetabular dysplasia but a stable hip.

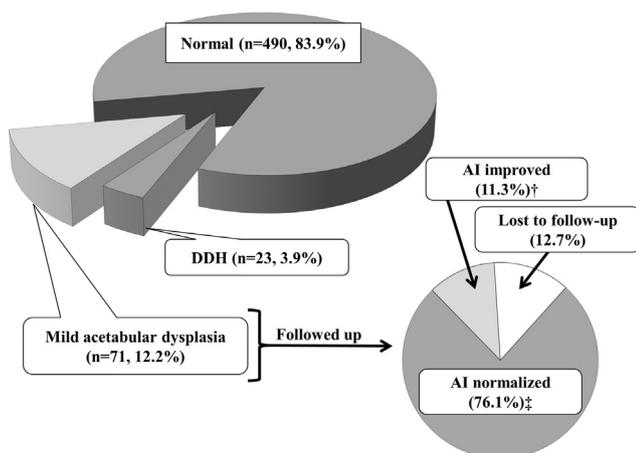


Figure 2 Clinical courses of referred infants. Of the 584 referred infants, 71 (12.2%) presented with mild acetabular dysplasia (increased AI, but a stable hip). Among them, 76.1% were followed up until there was satisfactory improvement of AI (mean 13.6 ± 7.3 months, 5–30 months); 11.3% showed improvement of AI that was still not sufficient (mean 8.9 ± 4.4 months, 5–18 months); and the remaining 12.7% were lost to follow-up. All of the infants with stable hips and increased AI showed improvement over time. † AI was improved during follow-up but is still high and needs further follow-up. ‡ The infants showed normalized AI and stable hip at the last follow-up.

Conflict of interest

All authors declare no conflicts of interest.

Acknowledgments

We have not received substantial contributions from non-authors.

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