Article

The positive predictive value of asymmetrical skin creases in the diagnosis of pathological developmental dysplasia of the hip

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Abstract	## Aims The aim of this study was to identify the association between asymmetrical skin creases of the thigh, buttock or inguinal region and pathological developmental dysplasia of the hip (DDH). ## Patients and Methods Between 1 January 1996 and 31 December 2016, all patients referred to our unit from primary or secondary care with risk factors for DDH were assessed in a "one stop" clinic. All had clinical and sonographic assessment by the senior author (RWP) with the results being recorded prospectively. The inclusion criteria for this study were babies and children referred with asymmetrical skin creases. Those with a neurological cause of DDH were excluded. The positive predictive value (PPV) for pathological DDH was calculated. ## Results A total of 105 patients met the inclusion criteria. There were 71 girls and 34 boys. Only two were found to have pathological DDH. Both also had unilateral limited abduction of the hip in flexion and a positive Galeazzi sign with apparent leg-length discrepancy. Thus, if the specialist examination of a patient with asymmetrical skin creases was normal, the PPV for DDH was 0%. ## Conclusion Isolated asymmetrical skin creases are an unreliable clinical sign in the diagnosis of pathological DDH. Greater emphasis should be placed on the presence of additional clinical signs to guide radiological screening in babies and children. Key messages: - This study confirms that asymmetrical skin creases are an unreliable clinical sign in the diagnosis of pathological developmental dysplasia of the hip. If the hip joint is clinically normal, it is highly unlikely there will be an association with pathological developmental dysplasia of the hip. Routine radiological and sonographic imaging of the hip joints is unnecessary if there is no limitation of hip abduction and normal leg lengths (negative Galleazzi). Newborn and infant physical examination guidelines should reflect this. Cite this article: *Bone Joint J* 2018;100-B:??-??.
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We have found the following papers that may be helpful to reference, including two papers written by the senior author. If you would like to cite any of these papers, please indicate where the citation(s) should be inserted. We will then renumber all references in-house.

Nie K, Rymaruk S, Paton RW. Clicky hip alone is not a true risk factor for developmental dysplasia of the hip. *Bone Joint J* 2017;99-B:1533-1536.

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Massa BSF, Guarniero R, Godoy RM Jr, et al. Use of inlet radiographs in the assessment of reduction after the surgical treatment of developmental dysplasia of the hip. *Bone Joint J* 2017;99-B:697-701.

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Developmental dysplasia of the hip joint (DDH) is a dynamic condition, in which minor dysplasia may deteriorate to an irreducible dislocation, or an unstable hip may stabilize within a few weeks.[[1-6]] The abnormality may be physiological or pathological. Pathological DDH is defined on ultrasound as either Graf Type III or IV, or an irreducible dislocation. Risk factors conventionally associated with pathological DDH include female gender, a family history, breech presentation, oligohydramnios, postural and fixed deformities of the foot, caesarean section and torticollis.[[8,9]] However, recent authors have only identified an association with female gender, breech presentation and a family history.[[10-12]] Non-idiopathic instability of the hip can also occur in children with neuromuscular conditions and is not described as DDH.[[7]]

In the United Kingdom, all new born babies undergo clinical assessment for instability of the hip with the Ortolani or Barlow manoeuvres shortly after birth, usually at the place of birth, and six to eight weeks later, usually within primary care. Other clinical signs known to have diagnostic value in pathological DDH are unilateral limitation of abduction of the hip in flexion[[11]] and an apparent limb length discrepancy, which can be seen as a short femur and a positive Galeazzi sign.[[8]] Asymmetrical skin creases can be defined as “asymmetry of the junction of the thigh to the trunk as viewed from the front and of the skin creases on the inside of the thigh".[[8]] They have been reported to be an important clinical sign for DDH since the 1969 Standing Medical Advisory Committee (SMAC) guidelines for hip screening in the United Kingdom. The more recent Newborn and Infant Physical Examination (NIPE) guidelines from Public Health England state that asymmetrical creases are a “screen positive sign" and affected children should be referred for expert assessment to exclude DDH.[[12]] Experts in Europe and the United States have, however, questioned the association between asymmetrical skin creases and DDH in the

absence of unilateral limitation of abduction of the hip in flexion.[[15-17]] The aim of this study was to investigate this association.

Patients and Methods

We established a 'one-stop' DDH screening programme in 1992. All babies referred with either risk factors or a clinical suspicion of DDH were assessed by the senior author (RWP) clinically and sonographically. Initially our catchment population included the areas of Blackburn, Darwen, Ribble Valley, and Hyndburn. From 2007 this expanded to include Burnley, Rossendale, and Pendle. Within the clinic, the source and reason for the referral, the age of the child and clinical and sonographic findings were prospectively recorded in a database (Microsoft Excel 2016, Microsoft, Redmond, Washington), which was used to identify the children for this study.

The inclusion criteria were all babies and children referred with asymmetrical skin creases of either the inguinal, adductor or gluteal folds between 1 January 1996 and 31 December 2016. Exclusion criteria included neurological or syndromic aetiology. During the period of the study, there were 113 741 births within the catchment area and 7187 referrals to the clinic, of which 105 met the inclusion criteria.

Clinical examination included the Ortolani and Barlow manoeuvres and assessment for unilateral limitation of abduction of the hip in flexion or apparent leg length discrepancy using the Galeazzi sign to examine for a short femur and assessment of leg length.[[8]] A normal examination was defined as a negative Ortolani and Barlow test, a full range of movement of the hips and equal leg lengths. The presence of asymmetrical creases were noted in all cases. Ultrasound imaging was performed with the baby on their side using modified Harcke dynamic and modified Graf static methods and a 5-7.5 MHz linear array transducer (Dornier AL 2200 prior to 2001, Sonosite 180 between 2001 and 2015, and Sonosite M Turbo from 2016).[[9,18]] A pathological hip was defined as a modified Graf

type III or IV hip, as advocated by Rosendahl et al.[[18,19]] Babies aged under six months with a pathological hip were treated in a Pavlik harness. Those with modified Graf type II hips were classified as physiological and observed with serial sonography and only underwent treatment if the hip progressed to a modified Graf Type III or IV or to an irreducible dislocation. For the purpose of this study, if a Graf Type II hip progressed to a more severe type of dysplasia, the most severe sonographic classification was documented. Those with a Graf Type I hip and a normal examination were discharged.

Statistical analysis

The results were analysed using sensitivity and positive predictive values (PPV), calculated using 2 × 2 contingency tables (Microsoft Excel 2016).

Results

As shown in Table I, there was a trend towards increased annual referral numbers for babies and children with suspected DDH and asymmetrical skin creases. This was particularly evident after 2008, when there was also a paradoxical decline in live birth rates.

[[TblCap]]**Table I.** The total live birth rate, total number of referrals to the clinic and number referred with asymmetrical skin creases (ASC) between 1996 and 2016. After 1996, the catchment area included Blackburn, Darwen, Ribble Valley, and Hyndburn; after 2007, it also included Burnley, Rossendale, and Pendle

Year	Total live births	Total referrals	ASC referrals
1996	3964	272	1
1997	3767	238	0
1998	3802	273	2
1999	3681	235	4
2000	3692	269	1
2001	3605	353	1
2002	3611	330	2
2003	3785	269	2

2004	3810	271	2
2005	3882	274	2
2006	3992	323	0
2007	7292	325	2
2008	7554	329	3
2009	7483	320	4
2010	7366	372	8
2011	7280	450	9
2012	7263	424	3
2013	7122	461	7
2014	7017	427	13
2015	6839	445	21
2016	6934	527	18
Total	113 741	7187	105

There were 71 girls (68%) and 34 boys (32%), of whom 94 (90%) were referred from primary care or community practitioners, the remainder from the new born checks. The age at the time of clinical assessment was bimodal in distribution, with peaks at three months and nine months. Apart from female gender, other recognized risk factors for DDH included one baby with a sibling affected by DDH and four with a breech presentation. None of these were diagnosed with DDH.

The clinical and sonographic findings are shown in Tables II and III. Clinical examination was normal in 84 babies (80%), all of whom had Graf Type I hips on sonographic imaging. A further 19 (18%) were found to have another significant clinical finding but had normal sonographic imaging (Graf Type I). Only two (2%) had pathological DDH, with unilateral dislocation of the hip being diagnosed on sonography. Both were referred from primary care: one boy, aged three months and one girl aged ten months. Both had unilateral limitation of abduction of the hip in flexion and an apparent leg-length discrepancy with a positive Galeazzi sign. The referral letter for the girl did not record

additional abnormal findings, however, the boy was also referred with an apparent leg length discrepancy of > 1 cm (Galeazzi positive) and decreased abduction in flexion. Both had otherwise normal development and no risk factors for DDH, and both were treated successfully with EUA, arthrogram, closed reduction and a hip spica.

[[TblCap]]**Table II.** Clinical findings for developmental dysplasia of the hip (DDH) and associated positive predictive value (PPV)

Clinical finding
84 (80%) had isolated asymmetrical skin creases (ASC) (of which 0 had DDH, PPV 0%)
19 (18%) had ASC plus one other finding (of which 0 had DDH, PPV 0%)
8 had ASC and limited hip abduction in flexion
11 had ASC and leg length discrepancy
2 (2%) had ASC plus 2 other factors, both of which were confirmed to have DDH (PPV 100%)
2 had ASC, limited abduction in flexion and a leg length discrepancy

[[TblCap]]**Table III.** Results of sonographic assessment; the alpha angle was unavailable for one baby in the series of 105

Alpha angle	**Baby's left hip, n**	**Baby's right hip, n**
Graf I (> 60°)	101	100
Graf IIa & IIb (50° to 60°)	**2 -** 1 had a normal clinical examination, discharged. baby had reduced abduction on initial examination. Reviewed at 6 mths, when the asymmetrical crease remained, but clinical examination was normal, thus discharged	**4 -** 3 had an alpha angle of 60°; discharged after normal clinical examination. 1 had an alpha angle of 53° and underwent examination under anaesthetic, arthrography, closed reduction and a hip spica
Graf IIc (43° to 49°)	0	0
Graf III & IV (< 43°)	**1** had an alpha angle of 40°; had an examination under anaesthetic, arthrogram, closed reduction and a hip spica	0

Thus, in babies with isolated asymmetrical skin creases, or asymmetrical creases with only one other clinical finding, as assessed by an experienced specialist, statistical analysis found a sensitivity for dislocation of the hip and a PPV both of 0%. Also, in those with asymmetrical creases and both limited abduction in flexion and an apparent leg-length discrepancy, the sensitivity and PPV were both 100% (Microsoft Excel 2016).

Discussion

The true prevalence of asymmetrical skin creases is unknown, although published figures suggest they are present in 20% to 25% of babies with normal hip joints.[[20-22]] If all babies found to have asymmetrical creases are referred for assessment, as the current NIPE guidelines suggest,[[12]] the minimum expected number of referrals to our service during the 20-year period would have been 22 748. As we only received 105, it is probable that many babies were examined by experienced paediatricians and primary care professionals and considered to have a normal hip. However, it highlights an alarming variation in practice. Furthermore, of the 105 referrals, 94 (90%) were from primary care or the community, which suggests that asymmetrical creases are either dismissed or are not apparent in the neonatal population.

When the rate of late diagnosed DDH was not reduced following the introduction of the Standing Medical Advisory Committee National screening programme[[13]] in 1969, one theory was that the examiners were poorly trained and inexperienced.[[26]] Thus in 2008, the United Kingdom National Screening Committee launched The National Health Service Newborn and Infant Physical Examination programme (NIPE),[[27]] which aimed to ensure that all healthcare professionals conducting examinations were fully trained and competent. It has been repeatedly shown that small groups of dedicated, well-trained examiners have an improved rate of detection of DDH, a reduction in false positive diagnosis and treatment, and a decrease in the rate of late diagnosis.[[23-25]] We acknowledge that there is justification

for a referral if the primary healthcare professional is not confident enough to exclude DDH. However, individuals tasked to assess hips for DDH clinically should have the appropriate training to identify those hips with known risk factors, such as unilateral limitation of abduction of the hip in flexion or a positive Galeazzi sign.

The NIPE 2008 guidelines paid specific attention to examination of the hips and stated the following: “Check symmetry of the limbs and skin folds. Perform Barlow and Ortolani manoeuvres”. Guidelines in the United Kingdom, including the most recent guidance from Public Health England for 2016 to 2017, continue to state the importance of assessing the symmetry of skin creases.[[8,12,27]]

Our study found that following the introduction of the 2008 NIPE guidelines, referrals for asymmetrical skin creases increased, despite a decline in live births within the catchment population (Table I). While it is imperative to maintain a high level of clinical suspicion for DDH, the evidence linking asymmetrical skin creases and DDH, in the absence of associated clinical findings and risk factors, remains weak. Palmen et al[[15]] studied 500 newborns and noted that 27% had no skin creases, 40% had symmetrical creases, and 33% had asymmetrical creases. Four had an abnormal provocative test of hip stability, of which two had symmetrical creases. Barlow,[[28]] in his study of 1962, examined over 9000 infants and found asymmetrical creases in < 50% of those diagnosed with DDH, and the great majority of those with asymmetrical creases had normal hips.

Omeroğlu and Koparal[[22]] reviewed 188 babies with suspected DDH, and found that the rate of DDH was 38% in those with asymetrical creases, compared with 10% in those without ($p < 0.05$). In this series five children with asymmetrical creases and abnormal ultrasounds had no other positive clinical finding. However, there was no mention of associated risk factors. Hassan and Shannak[[29]] reviewed 370 babies with confirmed DDH, of whom 83% had asymmetrical creases and at least one other significant clinical finding.

Stein-Zamir et al[[30]] reviewed 51 babies, several of whom had more than one positive clinical finding, with a PPV of 50% for asymmetrical creases. While these papers all conclude that asymmetrical creases are an important clinical finding associated with DDH,[[22,29,30]] they present insufficient data to conclude that asymmetrical creases are of clinical significance in the absence of associated clinical findings and risk factors.

The authors of two major systematic review articles found that it is difficult to conclude that asymmetrical skin creases are a useful clinical finding, there being scarce and unsupportive evidence linking asymmetrical creases with pathological DDH.[[17,31]] Our study is unique in its long period of observation and prospective data collection from one experienced clinician (RWP), making it more objective than previous studies. The undisputed clinical signs associated with pathological DDH are unilateral limitation of abduction in flexion after the age of two months, and a positive Galeazzi sign. If these signs were positive, the PPV was 100%. If a baby with asymmetrical creases has either of these clinical signs, radiological or sonographic assessment of the hip joint is imperative. In a baby with asymmetrical creases, normal movement of the hip and equal leg lengths, we found the PPV of DDH is 0%. This would suggest that Public Health England should review their guidelines, as the present recommended ‘screen positive’ guidance is not evidence-based. Further research in the form of a meta-analysis is warranted.

In conclusion, this study confirms that asymmetrical skin creases are an unreliable clinical sign in the diagnosis of pathological DDH and that most of these babies have a normal hip. If the hip is clinically normal, it is highly unlikely that there will be associated pathological DDH. Routine radiological and sonographic imaging of the hips appears unnecessary if there is no limitation of abduction in flexion and normal leg lengths (negative Galeazzi sign). National guidelines should reflect this.

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