Is secondary radiological follow-up of infants with a family history of developmental dysplasia of the hip necessary?

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The practice of regular radiological follow-up of infants with a positive family history of developmental dysplasia of the hip is based on the widespread belief that primary acetabular dysplasia is a genetic disorder which can occur in the absence of frank subluxation or dislocation. We reviewed all infants who were involved in our screening programme for developmental dysplasia of the hip, between November 2002 and January 2004, and who had a normal clinical and ultrasound examination of the hip at six to eight weeks of age, but who, because of a family history of developmental dysplasia of the hip, had undergone further radiography after an interval of 6 to 12 months. The radiographs of 89 infants were analysed for signs of late dysplasia of the hip and assessed independently by three observers to allow for variability of measurement. There were 11 infants (11%) lost to follow-up.

All the patients had normal radiographs at the final follow-up and none required any intervention. We therefore question the need for routine radiological follow-up of infants with a positive family history of developmental dysplasia of the hip, but who are normal on clinical examination and assessment by ultrasound screening when six to eight weeks old.

It is common practice to screen the hips of infants with a positive family history of developmental dysplasia of the hip (DDH), clinically and ultrasonographically, in view of the increased risk of the condition in this group. If these examinations are normal, uncertainty remains as to whether the patient can simply be discharged or should be followed up for a further period with radiographs taken after six or 12 months. It has been our policy to obtain a radiograph after 6 to 12 months in such patients, but this approach has not been conclusively supported in the literature.

For orthopaedic surgeons, early diagnosis of DDH results in a successful outcome in most patients. However, the need for a more sensitive means of early diagnosis has led to the development of targeted ultrasound screening¹⁻⁵ of infants with at-risk factors for DDH, in addition to the required clinical screening programme. After the age of four months an anteroposterior pelvic radiograph is the method of choice for the assessment of DDH.⁶

Risk factors for DDH include a positive family history, breech presentation, torticollis and congenital foot deformities. Garvey et al,⁷ in a study of radiological screening at four months of infants with one or more risk fac-

tors, found an increased incidence for DDH in 26 per 1000 live births. Wynne-Davies⁸ believed that acetabular dysplasia was a genetic disorder which could be present without frank dislocation or subluxation of the hip. For this reason children with a positive family history are often subject to closer scrutiny by ultrasound and sometimes radiography.

Previous studies^{3,7,8} have demonstrated a small but definite incidence of dysplasia of the hip presenting late in infants whose hips were clinically normal at initial presentation. Popple et al⁹ described dysplasia presenting late in infants whose hips were both clinically and ultrasonographically normal when first examined.

Our aim was to investigate the incidence of late dysplasia of the hip in association with a positive family history but initially normal clinical and ultrasonographic examination, and to assess the necessity for radiological follow-up of such infants.

Patients and Methods

Between November 2002 and January 2004, 732 infants were referred to our DDH screening clinic. Referrals were from paediatricians, general practitioners and health visitors because of a clinical suspicion of DDH or one

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Table I. Reason for referral and degree of family relationship in infants with a positive family history of developmental dysplasia of the hip (DDH)

Reason for referral	Family member with DDH		
	First degree*	Second degree†	Third degree‡
Family history only	52	26	2
Family history and breech	5	3	0
Family history and clicking hips	4	8	0

* eg, parents, siblings

† eg, aunts, uncles, half-siblings, grandparents

‡ eg, first cousins

Table II. The acetabular index (mean for each group (mths) in comparison with the mean reference values within 1 SD according to Tonnis¹²) for right (R) and left (L) hips

	5 to 6		7 to 9		10 to 12		13 to 15	
	R	L	R	L	R	L	R	L
Girls								
Our series	22.5	24.08	21.4	21.8	22.2	22.5	24.5	26.55
Tonnis ¹²	27.3	29.3	25.3	26.6	24.7	27.1	24.6	26.9
Boys								
Our series	22.3	21.9	20.5	19.2	20.8	21.3	23	22.6
Tonnis ¹²	24.2	26.8	24.6	25.4	23.2	25.2	23.1	24

of the following risk factors: 1) a positive family history (first-, second-, or third-degree relatives); 2) clicking of the hip; 3) breech presentation; 4) congenital foot deformity or 5) torticollis.

Of 732 consecutive referrals, 100 showed a family history of DDH. We have reviewed these 100 infants retrospectively. There were 43 boys and 57 girls at six to eight weeks of age. All were normal on the initial clinical examination by a senior author (NKG) and all had normal hips on static ultrasound assessment carried out by specially-trained radiographers (JS, SC) (Graf type 1 = 85; Graf type 2A = 15¹⁰). Of these, 11 failed to attend follow-up at six or 12 months and therefore 89 were available for the final review.

These infants were seen again at between six and eight months of age when a plain anteroposterior radiograph of the pelvis was obtained. Seven were reviewed between 10 and 13 months of age for a variety of unrelated reasons. Those with an acetabular index at the upper limit of normal for their age were reviewed again six months later.

The following radiological parameters were assessed by three observers (PK, NKG, JS): 1) the acetabular index; 2) the presence of the proximal ossific nucleus and 3) the shape of the teardrop.

After agreeing on the landmarks to be used for each observation and possible errors in measurement,¹¹ the assessments were performed independently. The limits between normal and abnormal acetabular indices were defined by the age of the infant at the time of the radiograph, as outlined by Tonnis.¹² Inter-observer variation for the values of the acetabular index between the three observers was calcu-

lated using Lin's concordance test.¹³ The correlation coefficient for continuous variables (ρ_c) was calculated using a web-based statistical calculator.¹⁴

We were also interested in the current practice across the United Kingdom and therefore a questionnaire was sent to all members of The British Society for Children's Orthopaedic Surgery to determine whether or not infants with a positive family history of DDH had radiographs taken at six to 12 months if their initial clinical and ultrasound examination had been normal. We obtained the Society's permission to do this survey.

Results

The reason for referral and the degree of family relationship are given in Table I. The mean values for the acetabular index on each side measured in boys and girls are shown in Table II. The mean reference values for the acetabular index¹² are provided for comparison.

Of the 89 infants, 71 had normal radiographs at the age of six to eight months and were therefore discharged. Seven patients did not attend their original follow-up appointments, but were seen after recall at a mean of two years of age. All had normal indices and were discharged. The remaining 11 had acetabular indices at the upper end of the normal range for age and were reviewed again with further radiographs at 12 months. At this stage, ten were normal and were discharged. The remaining patient was reviewed at 18 and 24 months when radiographs were normal and the child was discharged.

The proximal femoral ossific nucleus was present bilaterally on the initial radiograph in 80 infants, absent bilaterally in eight and absent on one side only in one. All had normal ossific nuclei bilaterally on follow-up radiographs. The teardrop was 'U'-shaped bilaterally in 42, unilaterally in four and had not yet developed on either side in 43 infants at the time at which the radiographs were taken. We did not find a 'V'-shaped teardrop on any radiograph.

A series of pelvic radiographs of a child at 6, 14 and 24 months is shown in Figure 1. The value of the acetabular index was initially at the upper limit of normal for age (within 1 SD of normal), but reached a normal value at the final follow-up.

There was a 'substantial' to 'almost-perfect' correlation between the three observers for values of the acetabular index. The concordance correlation coefficient¹³ (ρ_c) ranged between 0.7918 and 0.9257 (lower one-sided 95% confidence limits (CI) were 0.7 to 0.9). Based on this series, the 95% CI for the true prevalence of acetabular dysplasia in the presence of a normal hip ultrasound was 0% to 4%.

In our survey of the British Society for Children's Orthopaedic Surgery, 106 of 172 members (62%) returned their questionnaires; 17 were not in active practice and therefore declined to comment. Of the 89 active respondents, 31 (35%) would perform a follow-up radiograph and 58 (65%) said that in the presence of a normal clinical and ultrasound examination, they would not.

Fig. 1a

Fig. 1b

Fig. 1c

Anteroposterior pelvic radiographs of an infant at a) six, b) 14 and c) 24 months of age. At 6 and 14 months the acetabular angle was at the upper limit of normal for age (right 29°, 28°; left 28°, 27.6°, respectively), but at 24 months it was well within the normal range for age (right 22.3°; left 22°).

Discussion

Infants with a family history of DDH form a considerable proportion of all referrals to developmental dysplasia of the hip screening clinics. Clinical examination by experienced personnel is the required standard for screening in neonates. Additional ultrasound examination of the hip is also common practice and is considered to be a safe, effective and reliable method of assessment.^{1-3,11} All the infants in our study were assessed by experienced orthopaedic surgeons.

Family history, breech presentation and clicking of the hip are considered to be risk factors for DDH. An isolated clicking hip is of doubtful significance and in the presence of a normal ultrasound examination in the neonatal period the finding can be disregarded.¹⁵ In our series, eight infants were reviewed because of breech presentation and 12 had a clicking hip as well as a positive family history. All of these infants had a normal ultrasound scan.

Yamamuro and Chene¹⁶ demonstrated the wide variability in age range at which the proximal femoral ossific nucleus first appears (1 to 10 months). Asymmetry is a common finding in up to 16% of normal hips. This resolves spontaneously and is not considered to be clinically significant.¹⁷ There is a similar variability in age at which the acetabular 'teardrop' normally appears (3 to 24 months).^{17,18} Our study showed similar variations in the appearance of the proximal femoral ossific nucleus and the teardrop. A triangular or 'V'-shaped teardrop is an indicator of acetabular dysplasia,¹ but none of our patients demonstrated this.

Previous studies^{3,7-9} have shown an incidence of late-onset dysplasia of the hip in children with a positive family history. The practice of routine radiological follow-up at four months and later of infants at risk of DDH, is supported by the studies of Garvey et al⁷ and Popple et al.⁹ In the first study, the infants were only examined clinically and

none had ultrasound of the hip at the initial screening. Late dysplasia was detected radiologically in 18% of infants.⁷ In our view, clinical examination is too insensitive to ensure that these hips were normal even in the hands of experienced clinicians and the results of this study should be viewed with some scepticism.

In the series of Popple et al,⁹ late dysplasia was reported to be present in 2.5% of infants at follow-up until 12 months of age. The authors did not state whether any of their patients required any intervention and were unable to determine if the dysplastic hips did or did not finally become normal. The significance of the reported dysplasia is therefore uncertain.

We acknowledge the limitations of our study since 11% of our patients failed to attend for follow-up radiographs and we could not rule out the possibility of acetabular dysplasia in the non-attenders. However, we are a tertiary-level referral centre and the only paediatric orthopaedic hospital in our catchment area. We would therefore expect any problems to be referred to us, although some patients may have moved away and late dysplasia may not present to us for some years to come.

Wynne-Davies⁸ believed that primary acetabular dysplasia was an inherited characteristic and was the main cause in infants diagnosed late with DDH. However, Hoaglund and Healey¹⁹ in an analysis of 408 relatives of 78 children with DDH found that the acetabular development in the family members who did not have congenital dysplasia of the hip did not differ from that of control subjects. They suggested that acetabular dysplasia, rather than being an inherited abnormality, was secondary to subluxation or dislocation.

Our findings suggest that in children with a positive family history, normal clinical and ultrasound findings at the screening examination at six to eight weeks is sufficient to

exclude most cases of DDH without the need for later radiographs. In our practice to date, later follow-up radiographs have failed to identify any patient with an abnormality, although we accept that this may occur in our non-attending patients.

As a result of this study we have decided to discontinue routine radiological examination of infants with a family history of DDH if the initial clinical and ultrasound examination is normal. We believe that our current practice is not cost-effective. This change brings us in line with 65% of our colleagues in the British Society for Children's Orthopaedic Surgery. There is nevertheless some lingering uncertainty regarding the actual incidence of late dysplasia in children with a family history of DDH whose initial ultrasound examination is normal, but it must be small, and based on the 95% CI of our study it is unlikely to be more than 4%.

Supplementary Material

A further opinion by Dr Amaka Offiah is available with the electronic version of this article on our website at www.jbjs.org.uk

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