The natural history of developmental dysplasia of the hip: sonographic findings in infants of 1–3 months of age

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The natural history of sonographic developmental dysplasia of the hip was determined in a population-based study in which 5170 infants were screened by ultrasound using Graf's method. Of the normal hips at the age of 1 month, 99.6% were still normal at the age of 3 months. Of the immature type IIa/IIa+ and type IIa− hips, if untreated, 95.3 and 84.4% had become normal, respectively. Of the infants with type IIc, D and III/IV hips at the age of 1 month 70, 58.3 and 90.9% were treated, respectively. This study shows that normal hips remain normal in nearly 100% of cases, but development to abnormality is possible. It shows also that most of the immature hips at the age of 1 month become normal without treatment. Although treatment seems to be indicated in the majority of sonographically abnormal hips, the occurrence of overtreatment could not be excluded in our study. *J Pediatr Orthop B* 14:325–330 © 2005 Lippincott Williams & Wilkins.

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Introduction

One of the prerequisites for introduction of a screening program is that the natural history of the condition, including the development from latent to declared disease, is adequately understood [1]. In the early 1980s, Graf [2] developed and introduced ultrasonography as a diagnostic tool for developmental dysplasia of the hip (DDH). On the basis of Graf's ultrasonographic classification, ultrasound screening for DDH has been recommended. An ultrasonography-guided treatment protocol was designed for all type IIa− hips or worse [3–5]. However, as recognized by Terjesen *et al.* [6] and Teanby and Paton [7], the natural history of sonographically detected DDH is still not well described.

Several studies have followed up infants with neonatal immature hips and found that most of these hips normalized without treatment (83–98%), but it is unclear at what age they normalized [4,8–15]. Few studies have addressed the natural history of DDH in the sonographically abnormal hip. In the study of Castelein *et al.* [16], only four of the 101 neonatal immature or abnormal hips had developed definite dysplasia at the age of 6 months. In the study of Gardiner and Dunn [17], 42% of the infants with neonatal abnormal hips (type IIc or worse) randomized to sonographic surveillance did not need treatment. Two other studies found that about 80% of the sonographically abnormal hips normalized spontaneously [8,14]. These findings are in contrast with the opinion of Graf that spontaneous maturation of type IIc hips or worse is not to be expected [18]. Regarding the variety of opinions on the timing of ultrasound screening, it is essential to gain a better understanding of the natural history of DDH [19–22].

We performed a population-based study (the Soundchec study), in which infants were sonographically screened at the age of 1, 2 and 3 months at the child health care (CHC) centre. Infants were referred to a hospital for diagnostic evaluation according to a standardized referral protocol. The decision to treat was left to the discretion of the attending orthopaedic surgeon. Since not all infants with immature and abnormal hips were referred or treated immediately after referral, it became possible to follow up on the development of the hip in sonographically normal, immature and abnormal hips after the neonatal period.

Materials and method

Between October 1998 and July 2000, 5170 unselected infants were included in the Soundchec Study. Informed parental consent was obtained prior to participation in the study. The infants were screened by an ultrasound examination of the hip at the age of 1, 2 and 3 months at the CHC centre. The examinations were performed according to Graf's principles using a linear array 7.5 or 5 MHz transducer by diagnostic radiographers under the supervision of the project team (radiologist, orthopaedic

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Table 1 Classification of ultrasonographic hip types according to Graf [23]

Type		Age (weeks)	Bony roof	Bony rim	Cartilaginous roof	Alpha (°)	Beta (°)
I	Mature hip joint	Any age	Good	Angular/Blunt	Covering	>60	
IIa	Physiologically immature	0–5	Sufficient	Round	Covering	50–59	
IIa+	Appropriate for age	6–12	Sufficient	Round	Covering	50–59	
IIa–	Maturity deficit	6–12	Deficient	Round	Covering	50–59	
IIb	Delay of ossification	>12	Deficient	Round	Covering	50–59	
IIc	Critical range	Any age	Severely deficient	Round to flat	Covering	43–49	<77
D	Decentering hip	Any age	Severely deficient	Round to flat	Displaced	43–49	>77
IIIa	Decentered hip	Any age	Poor	Flat	Displaced cranially, without structural alterations	<43	
IIIb	Decentred hip	Any age	Poor	Flat	Displaced cranially, with structural alterations	<43	
IV	Decentered hip	Any age	Poor	Flat	Displaced inferomedially	<43	

resident, orthopaedic surgeon and CHC physician) [23]. For the purpose of evaluation, the children received an ultrasound reference test at the age of 8 months. The hips were graded according to Graf's classification (Table 1). The study was approved by the Ethics Committee of the Hospital Medisch Spectrum Twente in Enschede, The Netherlands.

The referral protocol prescribed that at the first screening only infants with decentered hips (type D or worse) should be referred for diagnostic evaluation. At the second and third screening, severe immature (type IIa–) and abnormal hips (type IIc and worse) were indications for referral. In practice, however, the referral protocol was not always strictly adhered to.

Of the children with type IIa and IIa+ hips, 2.5% were referred at the first screening. The hips of these infants showed a significantly lower α-angle than the hips of the non-referred infants (53.3° versus 57.9°). It was also noted that slightly more of the girls and slightly more of the infants with a family history of DDH in third- or fourth-degree relatives were referred. As a result of this, the groups of referred and non-referred infants were not completely comparable. The natural history data of the non-referred infants with type IIa and IIa+ hips should therefore be interpreted with some caution.

Of the children with type IIa– hips, 28.8% were referred at the age of 1 month. The α-angle of the hips of referred and non-referred infants did not differ significantly. There was also no significant difference with regard to age, sex, birth rank, breech position, family history of DDH and results of a preceding physical examination of the hips at the CHC centre. Therefore, the natural history of type IIa– hips determined in the infants who were not referred provided a valid approximation for the natural history in unselected infants with type IIa– hips.

Infants with type IIc hips or worse were almost always referred immediately, but a 'wait–and–see' approach was adopted regularly. For these hips, the treatment rate was determined to get an impression of the natural history.

All infants who were first screened at the age of 1 month were selected for the analysis of the natural history of sonographic hip types. A radiologist and two orthopaedic surgeons reviewed all abnormal sonographic findings (type IIc–IV) at the age of 1 month and reached consensus on diagnosis. In case the quality of the ultrasound image was inadequate, the infant was excluded from the study. The study results were based on the worst affected hip. If the hips were classified equal at the age of 1 month, the left hip was included. Distribution of the hip types at the age of 2 months was based on the infants who were not referred at the first screening. Non-referred infants who attended the third screening provided the data on the distribution of the hip types at the age of 3 months. Given the fact that all infants with type IIc hips or worse at the age of 2 months had been referred, the assumption was made that the hips of these infants would also have been abnormal at the third screening. To represent the group as a whole, weighted proportions were calculated to determine the distribution of the hip types at the age of 3 months. Confidence intervals (CI) for weighted proportions were calculated as recommended by Waller *et al.* [24]: the method of combining binomial variances and exact limits when a zero prevalence occurred in any stratum.

Results

At the first screening 4473 infants were screened. The median age of these infants was 5 weeks (range 2–7 weeks). Of these infants, 62.2% had normal hips, 36.4% had immature and 1.5% had abnormal hips according to the worst affected hip (Table 2). At the second and third screening 3790 and 3604 non-referred infants were screened at a median age of 9 weeks (range 8–11 weeks) and 14 weeks (range 12–24 weeks), respectively.

Type I hips

At the age of 2 months, 2388 infants of the 2781 infants with normal hips at the first screening were screened again (Fig. 1). Of them, 97.5% (95% CI, 96.8–98.1%) had type I hips, 2.2% (95% CI, 1.6–2.9%) had type IIa+ hips and 0.3% (95% CI, 0.1–0.6%) had type IIa– hips. At 3

months of age, 2324 of the 2388 infants were screened for the third time. Calculation of weighted proportions showed that of the infants with normal hips at the age of 1 month, 99.6% (95% CI, 99.4–99.9%) had normal and 0.4% (95% CI, 0.1–0.8%) had dysplastic hips at the age of 3 months (Fig. 2a).

Type IIa/IIa+ hips

Of the 1560 infants with type IIa/IIa+ hips at screening one, 1356 were again examined at the second screening. In 79.1% (95% CI, 76.8–81.2%) the hips had normalized. In 14.9% (95% CI, 13.0–16.9%) the hips were still typed as IIa+, in 4.6% (95% CI, 3.6–5.9%) the hips had become type IIa– hips and in 1.4% (95% CI, 0.9–2.2%) the hips had become abnormal. At the age of 3 months, 95.3% (95% CI, 94.2–96.6%) of the infants with immature hips at the age of 1 month had developed normal hips whereas the hips of 4.7% (95% CI, 3.6–5.8%) of the infants had become abnormal (Fig. 2b).

Type IIa– hips

Of all infants at the first screening, 66 had type IIa– hips. At the age of 2 months, 61.0% (95% CI, 44.5–75.8%) of the re-screened infants had normal and 12.2% (95% CI, 4.1–26.2%) had abnormal hips. Immature hips were seen in 26.8% (95% CI, 14.2–42.9%) of the infants with type IIa– hips at the first screening, 14.6% (95% CI, 5.6–29.2%) still having type IIa– hips. Calculation of weighted proportions demonstrated that the hips of 85.4% (95% CI, 80.7–96.4%) of the infants with type IIa- hips at the first screening had become normal at the age of 3 months while 14.6% (95% CI, 10.0–34.5%) had abnormal hips by that time (Fig. 2c).

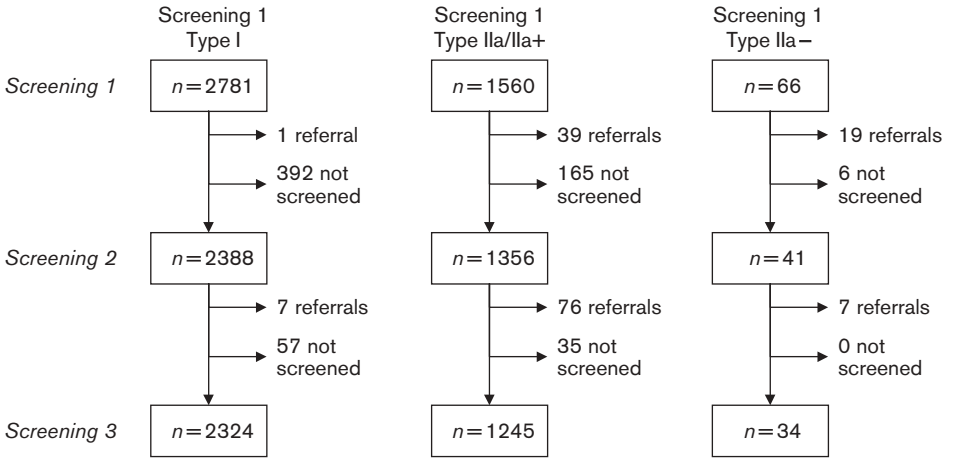
Table 2 **Sonographic screening results at the age of 1 month and final treatment rate per hip type in 4473 infants screened by ultrasound**

Sonographic hip type (worst affected hip)	Screening results		Children treated		
	n	%	*n*	%	95% CI (%)
I	2781	62.2	22	0.8	0.5–1.2
IIa/IIa+	1560	34.9	128	8.2	6.9–9.7
IIa–	66	1.5	21	31.8	20.9–44.4
IIc	20	0.4	14	70.0	45.7–88.1
D	24	0.5	14	58.3	36.6–77.9
III/IV	22	0.5	20	90.9	70.8–98.9
Total	4473	100	219	4.9	4.3–5.6

Treatment rates

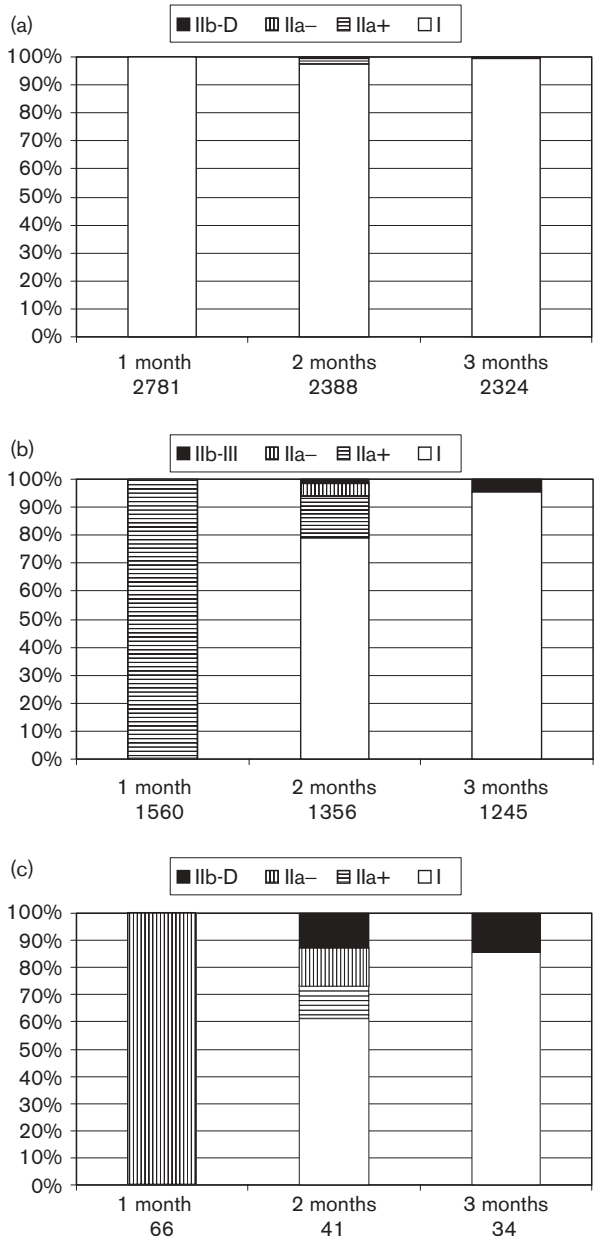
The actual treatment rates for infants with type I, IIa/IIa+ and IIa– hips at the first screening were higher than the corresponding weighted proportions of abnormal hips at the age of 3 months. However, except for the type IIa/IIa+ hips the confidence intervals are overlapping (Table 2). The treatment rates for abnormal hips at the age of 1 month were 70.0% for type IIc hips and 58.3% for type D hips. All infants were treated with an abduction splint or a Pavlik harness except for two infants who were treated by traction and one infant who required a surgical intervention. Of the 16 infants with type IIc or D hips who were not treated, 12 had both normal radiographs and a normal ultrasound reference examination at the age of 8 months. Two had normal findings on pelvic radiograph and did not attend the ultrasound reference examination. Two had a normal ultrasound screening examination at the age of 3 months and a normal ultrasound reference examination at the age of 8 months. Of the 22 infants with type III or type IV hips, two were not treated. In these two infants, a policy of watchful waiting was applied, and their hips showed normal findings on radiography at the age of 16 and 17 weeks, respectively (Fig. 3).

Fig. 1



Flowchart of the ultrasound screening examinations for infants with normal and immature hips at the age of 1 month.

Fig. 2



Schematic representation of the natural history of developmental dysplasia of the hip in infants screened by ultrasound at 1–3 months of age. (a) Normal hips at the age of 1 month. (b) Type IIa/IIa+ hips at the age of 1 month. (c) Type IIa– hips at the age of one month.

Discussion

The natural history of DDH was studied using the results of a population-based screening program. In this program, a large group of infants was screened by ultrasound at the age of 1, 2 and 3 months. Hips that were normal at the age of 1 month remained normal in 99.6% of the infants. Type IIa/IIa+ hips became normal in 95.3% and type IIa- hips in 85.4%. It was not possible to determine the normalization rate in infants with sonographically abnormal hips (type IIc and worse) because of the low number of infants being screened again at the age of 3 months. However, treatment rates of 70 and 58% for type IIc and D hips seem to support the hypothesis that a substantial number of sonographically abnormal hips do normalize without treatment.

A considerable number of infants were not screened at the age of 2 months. This can be explained by a temporarily limited personnel capacity resulting in the second examination being performed just after the age of 2 months. In addition, parents sometimes cancelled their appointments due to illness, lack of time and vacation. Given this explanation, it is assumed that these examinations were missing at random. In other words, there is no reason to assume that selection bias has occurred due to these missing values.

The analysis of the natural history of DDH rests on the assumption that no selection has occurred in referring infants for diagnostic work-up. This is not self evident because one would expect hips of referred infants to be more severely dysplastic than hips of non-referred infants. Indeed, the proportion of infants actually treated suggests that the referred infants were more severely affected. However, the distribution of risk factors and the results of the physical examination of the hip at the CHC centre of the referred and non-referred infants were remarkably similar and support the assumption that no selection had occurred. Since ultrasound screening for DDH has been known to carry the risk of overtreatment, the higher treatment rate suggests that at least in some children overtreatment has occurred [25,26].

The natural history of DDH in this study was based on the worst affected hip of infants while other investigators presented natural history data that were based on both hips. However, including both hips in the analysis produces too small confidence intervals due to the dependence of the hips within an infant. The normalization rates of IIa hips reported in other studies ranges from 83 to 98% [4,8–15]. Our results are in line with these findings. Yet, in our study, more infants with abnormal hips were treated than in the studies of Rosendahl *et al.* [14] and Bialik *et al.* [8], who followed a policy of watchful waiting. Since, in our study, treatment was started at the discretion of the attending orthopaedic surgeon upon referral, we cannot exclude the possibility that the higher treatment rate in our population reflects some form of overtreatment. In addition, the ultrasound screening in our study was performed at the age of 1 month and not in the first days of life. This might also explain the higher treatment rate of abnormal hips because the probability that hips of a specific hip type resolve without treatment decreases with age. Pauer *et al.* [12] described this phenomenon for immature hips in which the normalization rate decreased from 94% in

Fig. 3

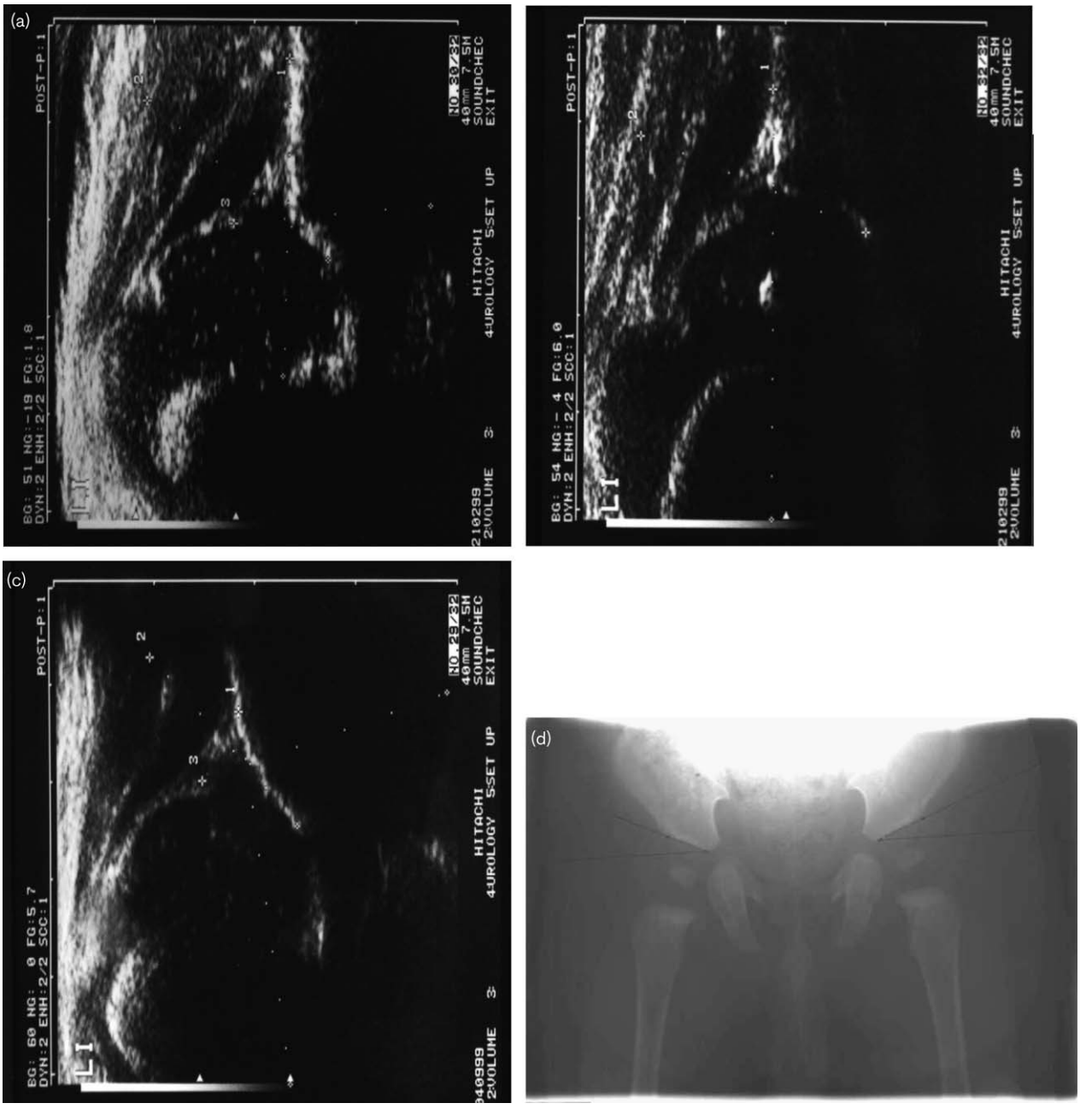


Illustration of the natural history of developmental dysplasia of the hip in two children with type III hips at the age of 1 month. Sonograms of the left hip of the same child at the age of (a) 1 month (type III) and (b) 8 months (type I). (c) Sonogram of the left hip and (d) radiograph of both hips of the same child at the age of 1 month (type III) and 4 months (normal), respectively.

immature hips detected in the first 10 days of life to 83% in those detected between 36 and 56 days.

On the basis of the results of a study on reproducibility described elsewhere, the supposed deterioration of normal hips can, at least partly, be explained by observer variation [27]. It means that normal hips did not deteriorate, but were initially incorrectly typed as normal. If this is the case in our study, it will be in concordance with the findings of Graf and others that normal hips remain normal [4,28,29]. However, deterioration of initially type I hips has also been described in other studies [8,12,30]. Since an expert panel agreed on the classification of the abnormal hips, it is unlikely that observer variation (i.e. incorrectly typed as abnormal) had an influence on the normalization rate of the abnormal hips.

In conclusion, this study shows that normal hips remain normal in nearly 100%. Most of the sonographically immature hips at the age of 1 month become normal without treatment at the age of 2 or 3 months. Type IIc and D hips may also normalize without treatment, but in the majority of these hips treatment seems to be indicated. Controlled trials comparing a wait-and-see regimen and abduction treatment for immature hips and IIc and D hips are indicated in order to develop a treatment algorithm and to prevent overtreatment.

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