■ CHILDREN'S ORTHOPAEDICS

Spontaneous recovery in the majority of stable dysplastic hips treated with active surveillance

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Aims

Worldwide controversy exists on the optimal treatment of stable dysplastic hips. The most common treatment options are abduction brace treatment and active surveillance. The primary aim of this study was to assess the effect of active surveillance in stable hip dysplasia, by investigating the percentage of Graf IIb stable dysplastic hips that recover spontaneously without abduction brace treatment. The second aim was to identify prognostic factors for spontaneous recovery of stable dysplastic hips.

Methods

A single-centre, prospective cohort study was conducted at the Máxima Medical Center between 1 March 2019 and 1 March 2023. Infants aged three to 4.5 months at the first outpatient clinic visit, diagnosed with Graf IIb hip dysplasia, and treated with active surveillance were included. Spontaneous recovery was defined as infants who had a normalized hip on ultrasound (α-angle ≥ 60°) after six weeks of active surveillance without receiving abduction brace treatment. Baseline infant characteristics and ultrasound measurements were used as potential predictor variables for spontaneous recovery in logistic regression analyses.

Results

A total of 508 infants with Graf IIb stable dysplastic hips were included. Overall, 473 infants (93.1%) recovered spontaneously with active surveillance. Of the remaining 35 infants, 25 infants (4.9%) received six weeks and ten infants (2.0%) received 12 weeks of additional Pavlik harness treatment until hip normalization. In univariate and multivariate analyses, an impaired hip abduction was negatively associated (OR 0.43 (95% CI 0.20 to 0.93); p = 0.033) and being a firstborn child positively associated (OR 2.20 (95% CI 1.04 to 4.63); p = 0.042) with spontaneous recovery.

Conclusion

The majority of Graf IIb stable dysplastic hips recover spontaneously in infants aged three to 4.5 months after six weeks of active surveillance. We recommend active surveillance with ultrasound as primary treatment for these infants. However, care must be taken with active surveillance in infants with a limited hip abduction.

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Introduction

Developmental dysplasia of the hip (DDH) is one of the most common musculoskeletal disorders in newborns.[1] DDH covers a broad spectrum of anatomical disorders of the paediatric hip joint, ranging from stable hip dysplasia to unstable hip dysplasia and hip dislocations.[2] Incidence is 3.7% in children up to six months of age in the Netherlands.[3] If left untreated, hip dysplasia may lead to pain, gait problems, and early adulthood hip osteoarthritis. DDH underlies 9% of all total hip arthroplasties and 29% of total hip arthroplasties in people aged up to 60 years.[4]

Abduction brace treatment is the cornerstone of DDH treatment. The Pavlik harness is the most commonly used abduction device for infants up

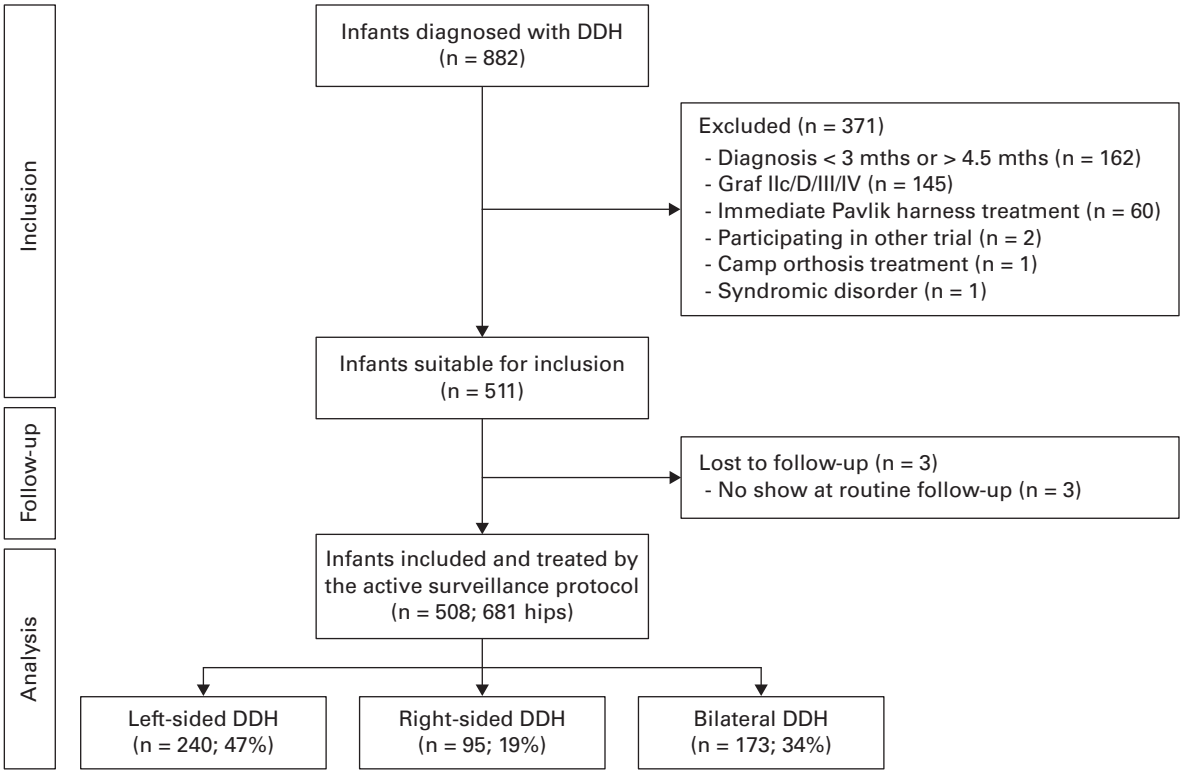

Fig. 1

Flowchart of included infants with Graf IIb developmental dysplasia of the hip (DDH) and treated with active surveillance.

to the age of six months.[5,6] Although abduction brace treatment has gained widespread use, discrepancies in treatment outcomes exist. Treatment success rates differ from 95% to 100% in dysplastic hips, and 80% to 88% in dislocated hips.[5,7,8] Although rare, Pavlik harness treatment is linked to abduction brace related complications (femoral nerve palsy and avascular necrosis), and is thus not entirely harmless.[9] Another drawback of the Pavlik harness is the substantial strain it places on parents and caregivers.[10] A high level of emotional burden, difficulties with parenting, and parental treatment concerns of a child in a Pavlik harness have been reported.[10-13]

Despite the high success rate of abduction brace treatment, controversy exists on the necessity for Pavlik harness treatment in the mildest form of DDH; the so-called stable dysplastic hips (Graf IIb). This is underlined by a recent British study on the management of DDH under the age of three months, where no consensus was reached regarding the treatment of Graf IIb dysplastic hips.[14] Active surveillance, defined as closely monitoring the development of the infant's hip with ultrasound imaging and intervening when necessary, is potentially a more suitable and cost-effective treatment strategy for these hips.[15-17] Scientific evidence for active surveillance of stable dysplastic hips though is sparse. Pollet et al[16] found no difference between Pavlik harness treatment and active surveillance in children aged three to four months with Graf IIb and Graf IIc dysplastic hips at the age of six and ten months. Based on 49 infants, 80% of these hips recovered spontaneously without Pavlik harness treatment.[16] A recent systematic review found no difference between abduction brace treatment and active surveillance in infants with stable DDH under the age of four months.[15] A drawback of the six studies included is the substantial methodological heterogeneity. The finding that 12% of the infants did receive abduction brace treatment in the second instance in this review shows that not all stable dysplastic hips spontaneously develop into normal hips.[15] Considering the promising results of active surveillance in previous studies, the question arises whether stable dysplastic hips that do need abduction brace treatment can be distinguished from those that can do without.

The primary aim of this study was to assess the effect of active surveillance in stable dysplastic hips, by investigating the percentage of Graf IIb stable dysplastic hips in infants aged three to 4.5 months that recover spontaneously without abduction brace treatment. As a secondary aim, this study attempted to determine prognostic factors for spontaneous recovery in stable dysplastic hips.

Methods

Study design. A single-centre, prospective cohort study was performed at the Máxima Medical Center (MMC) between 1 March 2019 and 31 March 2023. The MMC is a tertiary referral centre for DDH in the Netherlands. Informed consent was obtained from all parents. The Medical Research Ethics Committee at our institution approved the execution of this study (N19.022/L19.027). The study was reported according to the STROBE guidelines.[18]

Table I. Baseline infant characteristics. Missing data < 2% for all parameters.

Characteristic	Data
Infants, n	508
Hips, n	681
Mean age at start of active surveillance, mths (SD)	3.7 (0.3)
Sex, n (%)	
Male	157 (31)
Female	351 (69)
Affected side, n (%)	
Left	240 (47)
Right	95 (19)
Bilateral	173 (34)
Risk factor, n (%)	
Family history of hip dysplasia*	239 (47)
Firstborn child	246 (48)
Breech position	248 (49)
Swaddling	142 (28)
Twin	8 (1.6)
Physical examination, n (%)	
Galeazzi test positive	4 (0.8)
Barlow test positive	1 (0.2)
Ortolani test positive	0 (0)
Impaired hip abduction†	96 (19)
Hip ultrasound	
Mean α-angle affected hip, ° (SD)	56.3 (2.1)

*Defined as ≥ 1 first- or second-degree family member with DDH or hip osteoarthritis at age < 50 years.
†Impaired hip abduction defined as hip abduction ≤ 70° and/or side-to-side abduction difference ≥ 20°.
DDH, developmental dysplasia of the hip.

Study population. Infants aged three to 4.5 months at the first outpatient clinic, diagnosed with Graf IIb hip dysplasia on ultrasound imaging and primary treatment of active surveillance for six weeks, were included. Infants who were primarily treated with an abduction device, had syndromic disorders, and those who participated in other trials or had incomplete follow-up data were excluded. When there was a premature birth (< 37 weeks of pregnancy), the referral age was corrected by calculating the chronological age in weeks minus the number of weeks of prematurity.[19]

A total of 882 infants with hip dysplasia were part of the prospective cohort of the MMC between 1 March 2019 and 31 March 2023. Of this series, 508 infants (57.6%) met the inclusion and exclusion criteria (Figure 1). This corresponds with 681 hips, as 173 infants had bilateral DDH.

The baseline characteristics of infants treated according to the active surveillance protocol are depicted in Table I. The mean age at start of active surveillance was 3.7 months (SD 0.3), and 69% (n = 351) of the infants were female. The amount of missing data was < 2% for all parameters (Table I) and classified as missing completely at random. Therefore, a complete case analysis was performed.[20]

Screening and treatment protocol. Worldwide, screening programmes for hip dysplasia vary from ultrasound screening of all newborns to clinical screening of all infants combined with selective ultrasound screening based on risk factors for DDH.[21,22] In the Netherlands, clinical screening of all infants combined with selective ultrasound screening based on risk factors for DDH at the age of three months is an integral part of the national surveillance programme for DDH.[23] Therefore in the current study, infants with risk factors for DDH (positive family history of DDH, positive family history of hip osteoarthritis at age < 50 years in first- or second-degree relatives, or breech position > 32 weeks of pregnancy) were evaluated around the age of three months by a hip ultrasound according to the Graf classification.[23] In addition, infants with abnormalities on physical examination (hip abduction < 70°, side-to-side hip abduction difference ≥ 20°, or knee height difference), suggestive for hip dislocation, were evaluated by a hip ultrasound at less than three months of age. In those with an α-angle < 60° or hip dislocation on ultrasound imaging, the infant was referred to the paediatric orthopaedic surgeon.[23] In line with the 2021 Dutch national guideline on DDH, infants with stable hip dysplasia were treated by the active surveillance protocol.[24] This protocol specifies that stable dysplastic hips were closely monitored using ultrasound imaging following six weeks of active surveillance, without the application of an abduction brace during this initial period. When the α-angle < 60° or hip abduction ≤ 70° after six weeks of active surveillance, abduction brace treatment with a Pavlik harness was initiated (23 hours/day). Spontaneous recovery was defined as infants who had a sonographically normalized hip (α-angle ≥ 60°) during follow-up without receiving an abduction brace or other more invasive treatment.

Measurements. Infant characteristics (date of birth, sex, firstborn child, preterm birth < 37 weeks, and twin), affected hip (left, right, bilateral), DDH risk factors (breech position > 32 weeks of pregnancy, positive family history in first- or second-degree relative of DDH, or hip osteoarthritis at age < 50 years and swaddling of the child), and physical examination (hip abduction in degrees, Galeazzi test, Barlow and Ortolani sign) were routinely collected during the first outpatient clinic visit. Ultrasound imaging of the hip was performed by trained musculoskeletal radiologists according to the Graf criteria.[25] Ultrasound images were reviewed by the paediatric orthopaedic surgeons or a trained paediatric orthopaedic resident (WWEST, FQMPvD, ATB, JJT) based on the Graf classification.[25] Both α- and β-angle were measured. At routine follow-up after six weeks, ultrasound imaging was repeated and reviewed according to the above-mentioned criteria.

Statistical analysis. Statistical analysis was conducted using SPSS Statistics v. 22.0.0.0 (IBM, USA). Baseline infant characteristics were presented as mean (SD) for continuous variables and counts with percentages for discrete variables. Missing data were evaluated and handled according to the recommendation by Heymans and Twisk.[20] The percentage of spontaneous recovered Graf IIb stable dysplastic hips was calculated for all infants. Assessment for prognostic variables was performed by logistic regression analysis. Spontaneous recovery of hip dysplasia after six weeks of active surveillance on infant level as dichotomous variable was chosen as a dependent variable. As independent variables, we included: unilateral versus bilateral DDH, sex, positive family history in first- or second-degree relative, breech position, firstborn child, swaddling, twin, impaired hip abduction (abduction ≤ 70° and/or side-to-side abduction difference ≥ 20°), and α-angle at initial ultrasound.

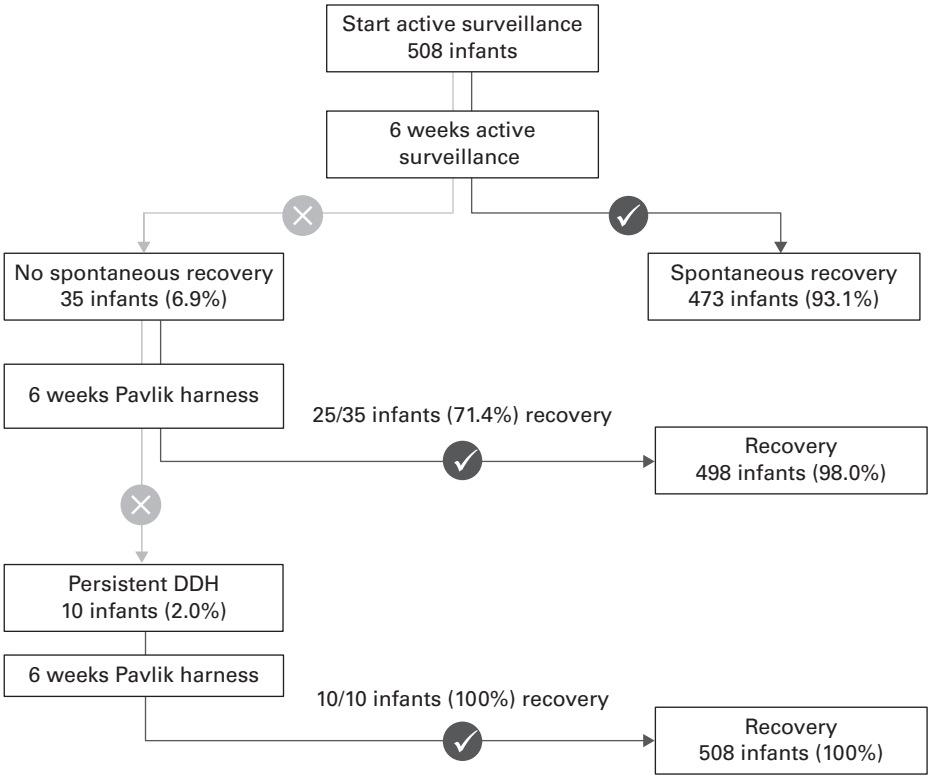

Fig. 2

Results of the Graf IIb stable dysplastic hips in the active surveillance treatment protocol. DDH, developmental dysplasia of the hip.

Univariate logistic regression analyses were performed, and variables with $p < 0.15$ were included in the multivariate logistic regression analysis. A level of statistical significance of $p < 0.05$ was applied in the multivariate logistic regression analysis.

Results

Spontaneous recovery of Graf IIb stable hip dysplasia was observed in 473 infants (93.1%) and in 637 hips (93.5%) (Figure 2). Of the 35 infants for whom Pavlik harness treatment was initiated after six weeks of active surveillance, 24 (68.6%) had an α-angle < 60°, four (11.4%) had a hip abduction ≤ 70°, and seven (20%) had a combination of both. Of those, 25 (71.4%) recovered after six weeks and ten (28.6%) after 12 weeks of additional Pavlik harness treatment. In the group of ten infants who underwent 12 weeks of Pavlik harness treatment, seven received only night-time abduction bracing for the last six weeks. No treatment other than a Pavlik harness was needed.

Of the nine potential predictor variables for spontaneous recovery, two were associated with spontaneous recovery in univariate logistic analyses (Table II): firstborn child (OR 2.10 (95% CI 1.00 to 4.41); p = 0.049) and impaired hip abduction (OR 0.48 (95% CI 0.23 to 1.01); p = 0.046). Both parameters remained significant predictors for spontaneous recovery according to the multivariate logistic regression analysis (Table II). Being a firstborn child was positively associated (OR 2.20 (95% CI 1.04 to 4.63); p = 0.042) with spontaneous recovery, and impaired hip abduction was negatively associated (OR 0.43 (95% CI 0.20 to 0.93); p = 0.033) with spontaneous recovery.

Firstborn infants had a chance of 95.5% (235/246) for spontaneous recovery. For non-firstborn infants, this chance was 90.8% (238/262). Infants with an impaired hip abduction had a chance of 88.5% (85/96) for spontaneous recovery, compared to 94.1% (386/410) for infants without impaired hip abduction. Of the 35 infants who did not recovery spontaneously after active surveillance in this prospective cohort study, 11 (31.4%) had a limited hip abduction. The multivariate model had a low predictive value for spontaneous recovery after active surveillance ($R^2 = 0.04$).

Discussion

Worldwide controversy exists on the optimal treatment of stable dysplastic hips, either abduction brace treatment or active surveillance. Our study showed promising short-term results of active surveillance of Graf IIb stable dysplastic hips in three- to 4.5-month-old infants. The hips of 93.1% of these infants developed spontaneously into normal hips after six weeks of active surveillance. Being a firstborn child is positively associated and an impaired hip abduction is negatively associated with spontaneous recovery of Graf IIb stable hip dysplasia.

The current study is the largest prospective cohort study that examined the effect of active surveillance in stable dysplastic hips. The high percentage of spontaneous recovery is in line with the results of the systematic review by Paulussen et al.[15] They reported a spontaneous recovery rate of 88% in 308

Table II. Univariate and multivariate logistic regression analyses on predictors for spontaneous recovery of stable developmental dysplasia of the hip.

Predictor variable	Odds ratio (95% CI)			
	Univariate regression analysis	p-value	Multivariate logistic analysis	p-value
Infant parameter				
Unilateral DDH	1.16 (0.57 to 2.36)	0.691		
Female sex	1.03 (0.49 to 2.15)	0.949		
Family history of DDH	0.65 (0.33 to 1.30)	0.221		
Breech position at birth	1.01 (0.51 to 2.01)	0.977		
Firstborn child	2.10 (1.00 to 4.41)	0.049*	2.20 (1.04 to 4.63)	0.042
Swaddling	0.73 (0.35 to 1.51)	0.388		
Twin	0.51 (0.06 to 4.30)	0.543		
Physical examination				
Impaired hip abduction	0.48 (0.23 to 1.01)	0.046*	0.43 (0.20 to 0.93)	0.033
Ultrasound parameter				
α-angle	0.99 (0.88 to 1.12)	0.888		

*Included in multivariate logistic regression analysis.
DDH, developmental dysplasia of the hip.

infants, despite the large methodological heterogeneity, variety of age of diagnosis, different abduction devices and variable criteria to initiate abduction brace treatment of studies reported in their systematic review.[15] A randomized controlled trial (RCT) by Pollet et al[16] on the effect of active surveillance of stable dysplastic hips reported a spontaneous recovery of 80%. This finding may be attributed to the inclusion of more severe cases of stable dysplastic hips in their study, suggesting that such cases are less likely to recover spontaneously following active surveillance. Nevertheless, the current study did not find an association between α-angle and spontaneous recovery. A substantially lower amount of spontaneous recovery (62%) was reported in a large retrospective study by Lankinen et al.[17] However, they studied a completely different subset of infants with DDH, namely the clinical unstable dysplastic hips in infants under two weeks of age.[17]

Despite the promising results of active surveillance of Graf IIb stable dysplastic hips, in our study in 6.9% of the infants (n = 35) normalization of the hip did not occur after six weeks, and secondary Pavlik harness treatment was initiated. Yet, 71.4% of these infants recovered after six weeks and 100% after 12 weeks of secondary Pavlik harness treatment. It is unclear whether these hips would have normalized to a Graf I hip with more time irrespective of Pavlik harness treatment. More research is needed before the decision is taken to completely refrain from treating stable dysplastic hips which are not spontaneously improving. Delaying treatment for a longer time period could also jeopardize the effectiveness of Pavlik harness treatment, as its efficacy is limited to the age of six months.[6,26] Furthermore, there is a paucity of evidence on which stable dysplastic hips spontaneously develop into normal hips and which benefit from secondary abduction brace treatment. The current research is the first study that examined multiple factors that may predict spontaneous recovery in three- to 4.5-month-old infants with stable hip dysplasia. The predictive value for spontaneous recovery in our multivariate model was low. A noteworthy finding of the present study is the negative association between impaired hip abduction and spontaneous recovery. Overall, 31.4% of the infants who had Pavlik harness treatment after active surveillance were diagnosed with a limited hip abduction. As far as we know, this is the first study that showed an association between impaired hip abduction and a lower chance of spontaneous recovery. We hypothesize that newborns with a limited hip abduction lack adequate mechanical stimulation required for normal acetabular development. Abduction brace treatment positions the infant's hip in a flexed and abducted position, directing the femoral heads towards the triradiate cartilage in the centre of the acetabulum, resulting in stimulation of acetabular development.[27] Therefore, we suggest that infants with a limited hip abduction may benefit from immediate abduction brace treatment. Being a firstborn child was positively associated with spontaneous recovery in our study. A protective association between firstborn children and residual dysplasia was reported in a previous study,[28] in which the authors argued that parity order is a risk factor for perinatal DDH, but not for hip dysplasia at an older age. We hypothesize that the restricted intrauterine space predisposes the firstborn children to DDH at birth,[29] who then tend to recover spontaneously after birth once the in utero movement restriction is no longer an issue.

Active surveillance of DDH has a number of advantages over Pavlik harness treatment. In the first place, in our study it reduced unnecessary treatment of 93.1% of Graf IIb stable dysplastic hips in three- to 4.5-month-old infants. Previous studies have shown that abduction brace treatment has a major impact on infant, parents, and caregivers, as well as being accompanied by serious emotional burden, parental treatment concerns, and difficulties with parenting.[10-13] Furthermore, Pavlik harness treatment carries a small risk of brace-related complications.[9] Another potential benefit of active surveillance is the reduction in healthcare costs, as no Pavlik harness device is needed and fewer outpatient clinic visits are indicated.[17]

The strengths of our study include the prospective study design, with a large number of infants with stable DDH, low rate of missing data, and extensive number of parameters in the prediction model. We acknowledge the potential limitations of the current study, mostly inherent to the prospective cohort study design that is integrated in a clinical care setting. In the first

place, selection bias may have occurred. Of the 60 infants who received immediate Pavlik harness treatment, a relatively large proportion were excluded from the analysis as they received immediate abduction brace treatment because of an impaired hip abduction. Based on the negative association between an impaired hip abduction and spontaneous recovery in the current study, immediate abduction brace treatment of this subgroup of infants is justifiable. Another potential limitation is that the screening, diagnosis, and treatment of DDH discussed in this paper are specific to the Dutch context, and there is considerable global variation in the age at which these processes are typically carried out. Most DDH screening policies advocate for screening, diagnosis, and treatment at an earlier age than presented in our study.[21] Despite these global differences, we think that the impact of active surveillance in stable dysplastic hips observed in this study is applicable beyond the Dutch healthcare system. In countries with early DDH screening protocols, hips classified as physiologically immature (Graf IIa) are typically diagnosed. A major part of these hips will mature spontaneously to a Graf I hip. However, 10% progress to a Graf IIb hip by three months, which would necessitate Pavlik harness treatment.[30] Our study indicates that even for a hip classified as Graf IIb at age three to 4.5 months, immediate Pavlik harness treatment is not necessary and active surveillance is a viable option. Therefore, our findings are applicable to countries with different screening, diagnosis, and treatment protocols, as well as to infants diagnosed and treated later than suggested by their national guidelines.

Although the current study shows promising short-term effects, future research should focus on the long-term effects of active surveillance of stable dysplastic hips. Knowledge of the amount of residual acetabular dysplasia after one year of age in infants treated by active surveillance is lacking. The prevalence of severe residual acetabular dysplasia in Pavlik harness treated hips is 10% at the age of two years and 1% to 3% at the age of five years.[31] The extent of residual acetabular dysplasia in infants treated by active surveillance is unknown, but it is crucial to gain a better insight into the long-term effects of active surveillance. Secondly, a gap in knowledge exists on the outcome of active surveillance of the more severe Graf IIc stable dysplastic hips, as these hips were excluded in this study. Furthermore, we used an α-angle ≥ 60° as a cut-off value for normalization of the dysplastic hip joint. It is questionable whether this cut-off value is truly normal in three- to 4.5-month-old infants, as Graf states that an α-angle ≥ 60° is normal for infants at the age of three months.[32] However, a study on the development pattern of 6,134 hips showed that the α-angle increases during the first three months of life and enters a plateau phase during the fourth to sixth month after birth.[33]

In conclusion, we found that the majority of infants aged three to 4.5 months with Graf IIb stable hip dysplasia develop spontaneously into normal hips after six weeks of active surveillance. Furthermore, we showed that an impaired hip abduction has a negative association and being a firstborn child has a positive association with spontaneous recovery. Based on our study findings, we recommend active surveillance by serial ultrasound imaging as primary treatment for all infants aged up to three to 4.5 months with Graf IIb stable hip dysplasia to avoid overtreatment, treatment burden for infant, parents, and caregivers, increased healthcare costs. Care must be taken with active surveillance of infants with a limited hip abduction, as this is negatively associated with spontaneous recovery.

Take home message

- The majority of stable dysplastic hips in three- to 4.5-month-old infants recover spontaneously after six weeks of active surveillance.
- A limited hip abduction is negatively associated with spontaneous recovery of stable dysplastic hips.
- Reducing unnecessary abduction brace treatment limits burden for infants, parents, and the healthcare system.

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