■ CHILDREN'S ORTHOPAEDICS

Resolving residual acetabular dysplasia following successful brace treatment for developmental dysplasia of the hip in infants

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Aims

Radiological residual acetabular dysplasia (RAD) has been reported in up to 30% of children who had successful brace treatment of infant developmental dysplasia of the hip (DDH). Predicting those who will resolve and those who may need corrective surgery is important to optimize follow-up protocols. In this study we have aimed to identify the prevalence and predictors of RAD at two years and five years post-bracing.

Methods

This was a single-centre, prospective longitudinal cohort study of infants with DDH managed using a published, standardized Pavlik harness protocol between 2012 and 2016. RAD was measured at two years' mean follow-up using acetabular index-lateral edge (AI-L) and acetabular index-sourcil (AI-S), and at five years using AI-L, AI-S, centre-edge angle (CEA), and acetabular depth ratio (ADR). Each hip was classified based on published normative values for normal, borderline (1 to 2 standard deviation), or dysplastic (> 2 standard deviation) based on sex, age, and laterality.

Results

Of 202 infants who completed the protocol, 181 (90%) had two and five years' follow-up radiographs. At two years, in 304 initially pathological hips, the prevalence of RAD (dysplastic) was 10% and RAD (borderline) was 30%. At five years, RAD (dysplastic) decreased to 1% to 3% and RAD (borderline) decreased to < 1% to 2%. On logistic regression, no variables were predictive of RAD at two years. Only AI-L at two years was predictive of RAD at five years ($p < 0.001$). If both hips were normal at two years' follow-up (n = 96), all remained normal at five years. In those with bilateral borderline hips at two years (n = 21), only two were borderline at five years, none were dysplastic. In those with either borderline-dysplastic or bilateral dysplasia at two years (n = 26), three (12%) were dysplastic at five years.

Conclusion

The majority of patients with RAD at two years post-brace treatment, spontaneously resolved by five years. Therefore, children with normal radiographs at two years post-brace treatment can be discharged. Targeted follow-up for those with abnormal AI-L at two years will identify the few who may benefit from surgical correction at five years' follow-up.

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Introduction

Developmental dysplasia of the hip (DDH) is one of the most common musculoskeletal disorders in infants and represents a spectrum of pathology, from instability in 1 in 100 live births to dislocation in 1 in 1,000 live births.[1] The Pavlik harness is the most common bracing treatment for DDH in infancy with success rates of up to 95%, characterized by normal clinical examination and hip ultrasound, at the end of treatment.[2] Despite these findings, radiological evidence of residual acetabular dysplasia (RAD) has been reported in up to 30% of children who had DDH treated in infancy.[3,4]

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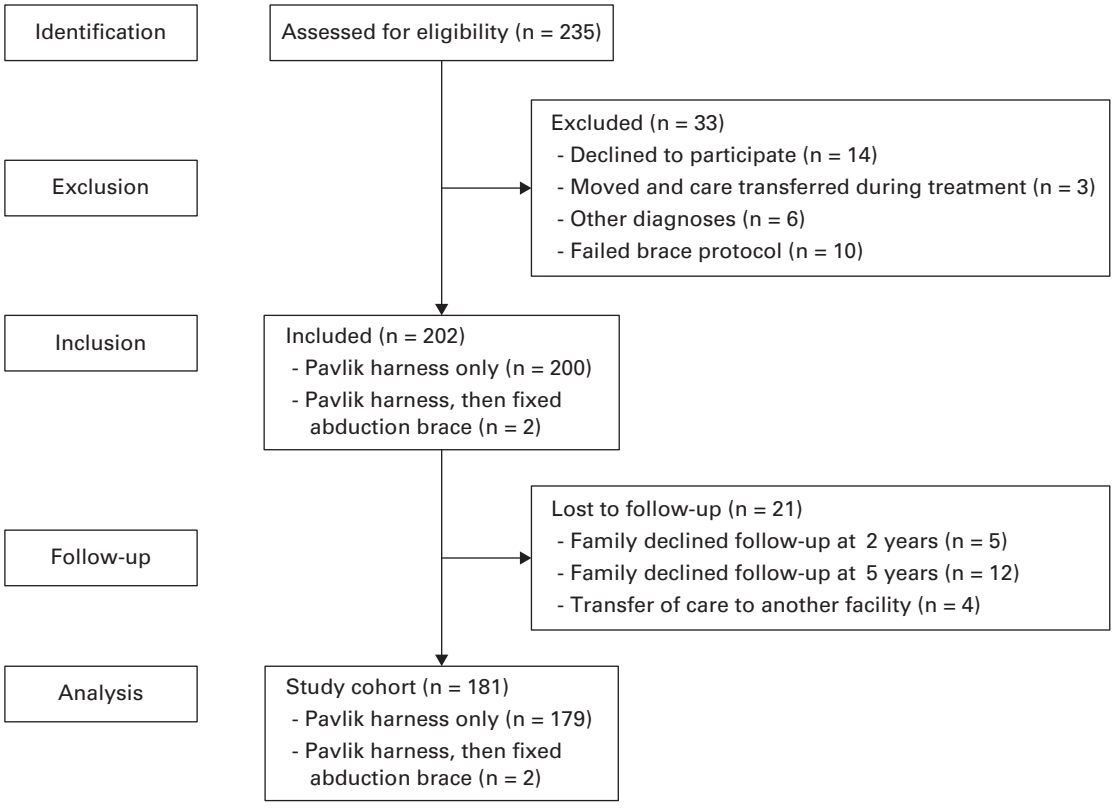


Fig. 1

Flowchart of participants through the study.

While typically benign in childhood, RAD at skeletal maturity is a deformity that leads to premature hip osteoarthritis as early as the fourth decade, and almost always by 65 years of age, suggesting that correction of RAD in childhood may be indicated to ensure long-term health of the hip joint.[5-7] However, the timing of surgical treatment of RAD in children is controversial as RAD often corrects spontaneously. Various studies have shown a higher prevalence of RAD at one to three years' follow-up compared to four to five years' follow-up, while the predictors and expected rate of spontaneous resolution remain unclear.[3,8,9]

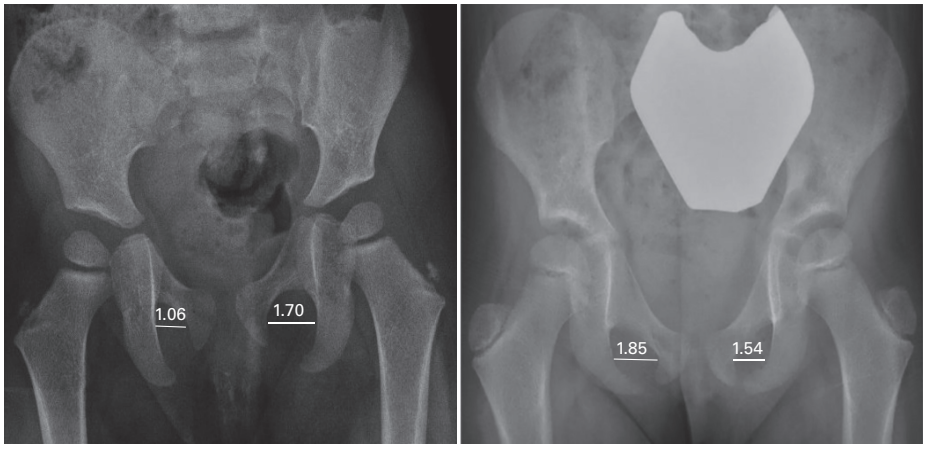
To identify those who may require surgical intervention for RAD, infants who have completed brace treatment for DDH are typically followed up in childhood by using hip radiographs, with some authors advocating annual follow-up.[6,10,11] However, it is reasonable to suggest that radiological follow-up could be more targeted to minimize unnecessary radiographs in those hips which are likely to spontaneously resolve. Studies have suggested that the incidence of RAD may be related to several factors including the severity of initial pathology, age at the initiation of treatment, and duration of wearing the Pavlik harness. No authors have comprehensively addressed these factors along with the rate of spontaneous resolution, and at what timepoints and for whom radiological follow-up can be discontinued. As such, the evidence base for specific follow-up recommendations following successful brace treatment is lacking.[12]

We have sought to identify the prevalence and predictors of RAD at two and five years following successful treatment for infant DDH using a standardized comprehensive bracing protocol. We also aimed to use our findings to inform practice on the need for, and duration of, radiological follow-up to eliminate unnecessary radiological exposure while capturing those who may benefit from further intervention.

Methods

This was a single-centre, prospective longitudinal cohort study of all eligible children who were managed using a published comprehensive nonoperative management protocol for infant DDH between January 2012 and December 2016.[2] The study protocol underwent institutional research ethics board approval (#1000080325). Patients aged six months or less were included in the study at the time of treatment initiation. All had radiological follow-up at a mean of two and five years post treatment. All successfully completed their course of bracing following the standardized protocol. Infants were excluded if they had other diagnoses (including neuromuscular conditions, teratological, or syndromic dislocations), if treatment was initiated elsewhere, or follow-up radiographs were incomplete.

Diagnosis and classification of dysplasia. The diagnosis and classification of DDH was made based the American Institute of Ultrasound in Medicine (AIUM) recommendations.[13] DDH was characterized by an α angle < 60°,[14] femoral head coverage



a b

Fig. 2

Example of obturator foramen diameter ratio for validating anteroposterior pelvis radiograph. Measurement was a) 0.62 at two years and b) 1.20 at five years.

(FHC) < 50%,[15] whether the hip was decentred on the static coronal view, and instability on stress testing in the transverse plane. Hips were considered decentred on ultrasound if there were interposed soft-tissue echoes between the base of the acetabulum and the femoral head, and if FHC was < 40%.[15,16] FHC cut-off was based on the upper limit of multiple published studies that report a range of < 23% to 40% FHC representing a hip dislocation on coronal view.[15,17-21] Using these parameters, hips were classified as normal, dysplastic, subluxable, subluxated, or dislocated.[2]

Management protocol. The protocol prescribes 12 weeks of Pavlik harness once the hips are centred in the brace on ultrasound imaging.[2] Success of brace treatment is determined by normal acetabular morphology (α angle > 60°, FHC > 50%), and hip stability on final ultrasound. At the end of treatment the brace was discontinued without a transitional period of partial use.

Validation of radiographs. A random sample of 10% of radiographs was evaluated using the obturator foramen diameter of Tönnis to ensure adequate positioning.[22] The Obturator Index is measured on the anteroposterior (AP) pelvic radiograph and provides a quotient of pelvic rotation by dividing the horizontal diameter of the obturator foramen of the right side and that of the left. In neutral rotation, the ratio is 1.0, but considered acceptable between 0.56 and 1.8.[22]

Study outcomes. At two and five years, RAD was defined as any abnormality on any measure with norm referencing at each timepoint to be most inclusive of abnormal findings. At two years, RAD was measured using acetabular index-lateral edge (AI-L) and acetabular index-sourcil (AI-S). At five years, AI-L and AI-S measurements were repeated with the addition of centre-edge angle (CEA) and acetabular depth ratio (ADR). International Hip Dysplasia Institute (IHDI) grade[23] and Bucholz and Ogden classification of avascular necrosis (AVN)[24] were also used at both timepoints. AI-L, AI-S, CEA, IHDI, and AVN have all been demonstrated to have good validity and reliability.[23-26]

At two and five years' follow-up, each hip was classified based on published population-based norm values as normal, borderline, or dysplastic based on sex, age, and right or left hip.[19,24,26-28] Borderline dysplasia on AI-L, AI-S, and ADR was defined as 1 to 2 standard deviations (SD) above the normative means, and dysplastic hips as > 2 SD. Reference values for CEA categorize ranges as normal, mild, and severe relative to sex, age, and laterality, which were renamed normal, borderline, and dysplastic for consistency.

Statistical analysis. Baseline characteristics including sex, birth position, birth order, family history of DDH, age at start of treatment, laterality, and initial severity of dysplasia were assessed using descriptive statistics. Fisher's exact test was used to compare categorical data, and independent-samples *t*-test was used for continuous data. The prevalence of RAD at two and five years in initially pathological hips was evaluated using descriptive statistics based on the transformed data relative to published norms. Logistic regression was performed to ascertain the effects of sex, fetal presentation, birth order, family history, laterality, and initial diagnosis of a centred versus decentred hip on the likelihood that patients would have any measure of residual dysplasia at two years' mean follow-up. Logistic regression was repeated to include only significant predictors at two years, and AI-L and AI-S at two years' mean follow-up, to determine the likelihood of RAD at five years' mean follow-up. Heat mapping was used to identify relative agreement between measures of RAD at two and five years' mean follow-up. Significance was set at an alpha < 0.05. Statistical analyses were completed using SPSS v29.0.1.0 (IBM, USA).

Results

A Strengthening the Reporting of Observational Studies in Epidemiology (STROBE)[29] diagram of participants is presented

Table I. Study cohort baseline demographic details.

Characteristic	Cohort value (n = 181)
Female, n (%)	159 (88)
Breech presentation, n (%)	85 (47)
First born, n (%)	126 (70)
Positive family history, n (%)	40 (22)
Mean age at the start of brace, wks (range; SD)	7.35 (0.5 to 22; 3.98)
Mean age at two-year radiograph, mths (range; SD)	25.62 (18 to 37; 2.84)
Mean age at five-year radiograph, mths (range; SD)	62.81 (49 to 83; 5.83)
Unilateral dislocation, n (%)	43 (24)
Bilateral dislocation, n (%)	20 (11)
Bilateral pathology, n (%)	123 (68)

SD, standard deviation.

Table III. Prevalence of residual acetabular dysplasia in initially pathological hips based on norm-referenced measures at mean two and five years' follow-up. All values are presented as absolute numbers and percentages.

Timepoint	Borderline or dysplastic combined (> 1 SD)	Borderline (1 to 2 SD)	Dysplastic (> 2 SD)
2 years*			
AI-L	110 (36)	80 (26)	30 (10)
AI-S	123 (40)	92 (30)	31 (10)
5 years*			
AI-L	13 (4)	8 (3)	5 (2)
AI-S	9 (3)	8 (3)	1 (< 1)
CEA	12 (4)	10 (3)	2 (< 1)
ADR	4 (1)	2 (< 1)	2 (< 1)

*n = 304 hips.
ADR, acetabular depth ratio; AI-L, acetabular index-lateral edge; AI-S, acetabular index-sourcil; CEA, centre-edge angle; SD, standard deviation.

in Figure 1. Of the 235 infants who began the bracing protocol, 202 infants (86%) were included in the final cohort. Of those, 181 infants (90%) successfully completed bracing and had two and five years' mean follow-up radiographs. There were no significant differences between the study cohort and those lost to follow-up (n = 21) (Supplementary Material 1).

The obturator indices of 20 randomly selected follow-up radiographs at two years and five years fell within the acceptable range of 0.56 to 1.8 (study sample 0.64 to 1.39) (Figure 2).

The demographic details, mean ages at follow-up, and pathology at initiation of treatment of the study cohort are presented in Table I and Table II.

The prevalence of RAD with the exclusion of initially normal hips (n = 58) from the norm-referenced measures are demonstrated in Table III. All hips were IHDI grade 1, and none had AVN at two years and five years' mean follow-up.

Using any single abnormality (borderline or dysplastic) as RAD in the logistic regression, no demographic variables were found to be predictive of RAD at two years' mean follow-up, nor severity of pathology at initiation of treatment. At five years, only AI-L at two years' mean follow-up was found to be predictive. We found that the logistic regression model was statistically significant ($X^2(4) = 28.964$; $p < 0.001$), which explained

Table II. Pathology at initiation of treatment.

Left hip, initial diagnosis	Right hip, initial diagnosis				
	Dislocated	Subluxated	Subluxatable	Stable dysplasia	Normal
Dislocated	20	8	9	2	14
Subluxated	6	23	9	1	7
Subluxatable	2	3	33	3	13
Stable dysplasia	1	0	1	2	5
Normal	1	5	10	3	X

20.9% (Nagelkerke R^2) of the overall variance in RAD, which in turn correctly classified 93.9% of the patients within the study. At two years, those with normal hips had no RAD at five years ($p = 0.002$), and those with measures of borderline or severe AI-L were 7.690 times ($p = 0.003$) and 16.094 times ($p < 0.001$) more likely to exhibit RAD at five years.

Table IV presents the heat mapping of hip presentations based on AI-L at two years and any measure (AI-L, AI-S, ADR, CEA) at five years. If both hips were normal at two years (n = 96 patients), all remained normal at five years. In patients where one or both hips were normal at two years (n = 134 patients), only one child presented with unilateral borderline findings at five years' follow-up, none had dysplasia. In those presenting with bilateral borderline pathology at two years (n = 21), only two had borderline findings of RAD at five years. In those with either borderline-dysplastic or bilateral dysplasia at two years (n = 26), features of dysplasia remained in 3/26 (12%) patients at five years.

Discussion

We prospectively studied a cohort of 181 children with DDH who had successfully completed a comprehensive nonoperative treatment protocol, with two and five years' radiological follow-up in order to evaluate RAD. In our cohort of 304 initially pathological hips, the prevalence of RAD at two years (borderline or dysplastic) was 43% (132 hips). At five years, the prevalence of RAD had reduced to 6% (17 hips), representing an 87% (115/132 hips) rate of spontaneous resolution between two and five years. No factors predictive of RAD at two-year follow-up were identified, and the only predictor of RAD at five years was AI-L at two years. All children with radiologically normal hips at two years' follow-up remained normal at mean five years' follow-up.

The reported rates of RAD following brace treatment for DDH varies widely in the literature from 2.4% to 30%,[4,10,30-32] making it difficult to provide recommendations for follow-up to identify hips for surgical intervention. Factors that influence the reporting of RAD include the presence or absence of a standardized treatment protocol, the age and severity of pathology at presentation, the length of follow-up (with longer typically showing less RAD), and marked variation on how RAD is defined.[4,10] We sought to clarify these issues through our study design with a prospective longitudinal cohort, a standardized validated treatment protocol, minimal loss to follow-up, validated follow-up radiographs at mean two and five years post

Table IV. Heat mapping of two years and five years norm-referenced measures based on right-left or left-right hip combinations (n = 181 patients).

5 years diagnostic group (all measures)	2 years diagnostic group (AI-L only)					
	Normal-normal	Normal-borderline	Normal-dysplastic	Borderline-borderline	Borderline-dysplastic	Dysplastic-dysplastic
Normal-normal	96	30	7	19	13	6
Normal-borderline	0	1	0	1	0	0
Normal-dysplastic	0	0	0	0	0	1
Borderline-borderline	0	0	0	1	3	0
Borderline-dysplastic	0	0	0	0	1	1
Dysplastic-dysplastic	0	0	0	0	1	0

Green: signs of improvement; Yellow: no change; Red: deterioration.
AI-L, acetabular index-lateral edge.

treatment, and the use of age- and sex-matched norm-referenced values to characterize RAD.[2]

Our overall findings are consistent with a systematic review by Shaw et al,[3] where only five articles over a 50-year period reported on the long-term outcomes of DDH after Pavlik harness treatment. The very limited number of studies that used a specified treatment protocol had lower rates of RAD (n = 1,861; 3.8%) in comparison to those without a standard protocol (n = 4,168; 17.6%).[10,30,31,33-39]

We found that baseline demographic details, including age at initiation of treatment, were not predictive of RAD. While Shaw et al[3] were unable to comment on this, Fujioka et al[40] demonstrated that there was no significant difference between age at initiation of treatment and CEA in 129 hips with 17.8% RAD at a mean follow-up of 20 years.

We also found that pathology at initiation of treatment was not predictive of RAD. Alexiev et al[30] previously noted three ultrasound parameters (dynamic coverage ≤ 22%, α angle < 43°, and abnormal echogenicity of the cartilaginous roof) were helpful in predicting RAD in 87 hips, with 6% at a mean of 5.3 years' follow-up. However, Sucato et al[37] reviewed 192 newborn hips that were stable on clinical examination but abnormal on ultrasound, and found no relationship between severity on ultrasound and RAD. Paranajape et al[4] reported a 30% rate of RAD in hips that were dislocated on initial presentation, but this finding may be confounded by how RAD is defined in the study using > 90th centile of population norms.

Cashman et al[10] demonstrated that almost all patients who showed late dysplasia could be identified by 18 months of age, and all of them were identified by five years.[10] In addition, they too reported AI-L as the most useful radiological predictor of RAD at five years' follow-up. Conversely, Modaressi et al[35] studied 150 patients who were treated and followed up until walking age. They found RAD among four females, necessitating surgery in early adolescence, and concluded that radiological follow-up should continue into adolescence. This and other studies encouraging long-term follow-up based on occasional late presentation of RAD tend to lack strict eligibility criteria, standard treatment algorithms, and failure of harness treatment and as such, recommendations should be viewed with caution.

We observed that children with normal radiographs at two years of age all had normal hips at five years' follow-up. Allington[33] published the retrospective results of 109 hips with successfully treated DDH using a Pavlik harness and a normal two-year radiograph. At a mean follow-up of ten years, all hips had normal clinical and radiological parameters. They concluded that, in a selected group of patients treated for DDH with a Pavlik harness, under their standardized protocol, and a normal radiograph at two years of age, long-term follow-up is not necessary, supporting the findings of our study. Our study also provides evidence to support the recent British Society for Children's Orthopaedic Surgery consensus recommending follow-up until two years for those children who have normal radiographs.[12]

To streamline the nonoperative care of DDH, we have translated our findings to perform radiological follow-up on every child at two years following brace treatment given that we found no predictors of RAD for that timepoint. Yet, at two years' follow-up, we now discharge those children with normal hips, as heat mapping showed that all hips that were normal at two years remained normal at five years. Those children who showed borderline or dysplastic hips at two year were eight and 16 times more likely to exhibit RAD at a mean of five years, and therefore are appropriate for further follow-up in order to identify the small proportion that may benefit from an acetabular osteotomy for dysplasia > 2 SD of norm reference values. Of note, and to date, every case of borderline dysplasia, we identified at five years following successful brace management in our practice, has spontaneously resolved by eight years.

We no longer perform annual follow-up radiographs between two and five years, as we feel that it exposes the child to unnecessary clinic visits and radiation, given that most hips spontaneously correct. Furthermore, surgical intervention prior to five years based on RAD will include some unnecessary operations. Based on our study cohort, these conservative recommendations would reduce follow-up visits at five years by 53%, leaving only 86 of 181 children for five-year follow-up. Even then, there is still only a 4.7% chance of finding RAD in this select group who would warrant surgery.

The limitations of our study are that it is single-centre using a standardized treatment protocol, and thus our results may not translate to other centres. We did not have a control group of non-treated hips, yet we did compare to validated norm-referenced values of acetabular development. Furthermore, to better assess the fate of RAD at two years, radiographs at skeletal maturity would be indicated, however we do not feel that continued radiological follow-up in a large group of children with corrected DDH is indeed ethical, given the need for imaging that uses ionizing radiation and such a low likelihood

in finding abnormalities. Finally, given the combinations of different patient characteristics and severities of RAD at two and five years' follow-up, confidence in subgroup analysis is reduced given the small numbers in each group.

In conclusion, we have undertaken a prospective longitudinal cohort of children successfully treated for DDH using a comprehensive brace protocol. We found that 43% of children demonstrated RAD at two years, yet 87% had resolved spontaneously by five years without further intervention. Therefore we recommend that patients with normal radiographs at two years can safely be discharged from care. Also, targeted follow-up for those patients with RAD at two years will identify the few who need surgical correction at five years' follow-up. We submit that our evidence-based pragmatic approach to DDH follow-up, will streamline care, minimize parental anxiety, and prevent unnecessary exposure to radiography.

Take home message

- We aimed to identify the prevalence and predictors of residual acetabular dysplasia (RAD) at two years and five years following successful brace treatment for infant developmental dysplasia of the hip (DDH).
- The majority of RAD at two years spontaneously resolved by five years, the only predictor of RAD at five years was an abnormal acetabular index at two years, and all children with radiologically normal hips at two years remained normal at mean five years' follow-up.
- We recommend that children with normal radiographs at two years can safely be discharged, and those with RAD at two years should be followed to identify those few who may need surgical correction at five years.

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Supplementary material

Table illustrating the comparison between the study cohort and those lost to follow-up.

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