Residual dysplasia of the hip after successful ultrasound-monitored treatment: how does an infant's hip evolve?

Daniel Dornacher, Bernd Lutz, Tobias Freitag, Mirco Sgroi, Rita Taurman and Heiko Reichel

Despite that normal values for the hip joint are reached at the end of ultrasound-monitored-treatment, the development of the acetabulum can be compromised during the growth phase. The acetabular index (AI) measured on a pelvic radiograph has been proven to be a reliable parameter. The aim of this study is to gain a better understanding of the dynamics of once-treated, residually dysplastic hips. This should be achieved by radiographically following these hips up to a milestone-examination at the end of preschool age. A total of 120 hips of consecutive 60 infants were included in this examination, each presenting with a residual developmental dysplasia of the hips (DDH) after successful ultrasound-monitored harness treatment. Radiographic follow-up was assessed retrospectively around 18 months, 3 years and 6 years of age, and the AI was measured. The age-dependent Tönnis classification was applied. The hips were assigned normal, mildly or severely dysplastic. Dependent *t*-test for paired samples indicated a highly significant improvement of the AI-values, including from the first to the second and from the second to the third follow-up. The percentage distribution into the Tönnis classification changed remarkably: in the first follow-up, 36 of the 120 hips were evaluated 'severely dysplastic', in the third follow-up only 1. On the other hand, three hips underwent acetabuloplasty. Even after normal values have been achieved at the end of ultrasound-monitored treatment, there remains a risk of residual dysplasia of the hips. Particularly, when the first radiographic examination shows nonphysiological findings, further close-meshed follow-up is recommended. Level of evidence: retrospective study of therapeutic outcome, consecutive patients, level II. *J Pediatr Orthop B* 31: 524–531 Copyright © 2022 Wolters Kluwer Health, Inc. All rights reserved.

Journal of Pediatric Orthopaedics B 2022, **31**:524–531

Keywords: developmental dysplasia of the hips, radiographic follow-up, residual dysplasia of the hips, ultrasound-monitored treatment

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Received 13 July 2021 **Accepted** 27 February 2022

Introduction

The introduction of general ultrasound screening according to Graf turned out to be a blessing for the treatment of developmental dysplasia of the hips (DDHs) [1]. The screening has led to a decrease in the incidence of late presenting hip dislocation and to a reduction of surgical procedures [2,3]. Despite normal values for the hip joint are reached at the end of ultrasound-monitored-treatment, the development of the acetabulum can be composed of endogenous factors during the growth phase [4–7]. In order to assess residual hip dysplasia after ultrasound-monitored harness treatment, the method of choice is an anterior–posterior (ap.) pelvic radiograph. For the monitoring of the acetabular development – in particular when residual dysplasia is present – the acetabular index (AI) has been proven to be a reliable parameter [8]. The necessity for the radiographic follow-up is widely accepted, but the frequency and the length of the radiographic follow-up are under discussion [9–15]. Repeatedly, adolescents and young adults are referred to our outpatient department, presenting with a symptomatic acetabular dysplasia and at the same time showing their ultrasound hip screening protocol with physiological images. These unexpected findings suggest that the penetration of an endogenous factor can deteriorate acetabular development to some period in skeletal growth. Given this, an adolescent-onset dysplasia is the subject of discussion [6]. Up to the present, comprehensive knowledge of acetabular development is still missing, and there is a lack of examinations with longitudinal character.

The aim of this study is to gain a better understanding of the dynamics of once-treated, residually dysplastic hips. This should be achieved by radiographically following these hips up to a milestone-examination at the end of preschool age. An insignificant decrease of the AI was proposed as null hypothesis.

Material and methods

Between January 2011 and December 2012, 358 infants with the diagnosis 'DDHs' were referred to our outpatient department of pediatric orthopedics. The newborns either were referred to our unit in the first few days after birth due to some risk factor for DDH (e.g.

 DOI: 10.1097/BPB.0000000000000984

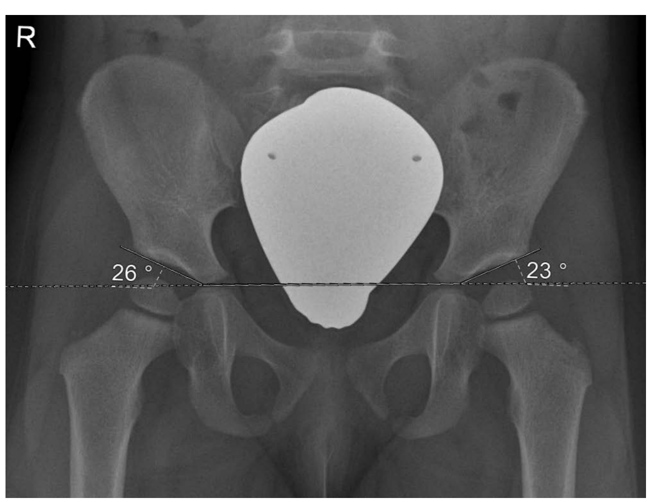
breech position and family history) or after a pathological ultrasound screening according to the German health care system, which is mandated at the age of 4–6 weeks. After ultrasound examination according the protocol described by Graf, 292 infants underwent ultrasound-monitored treatment. This comprised flexion-abduction splinting (Tübingen Splint, Pavlik-harness or Fettweis plaster cast), which followed the clinical practice guidelines recommended by Graf [1]. Particularly, in the Graf type III and IV hips, additional Vojta therapy was advised. The treatment was continued until physiological values were reached in the regularly performed ultrasound follow-up (α-angle above 64°). In all once-treated hip joints, radiographic follow-up was recommended to the time the children were able to walk. Then, an ap. pelvic radiograph was performed, and the AI was measured (Fig. 1). The AI was allocated according to the age-dependent classification system described by Tönnis (Tönnis 0 = below first SD (normal), Tönnis 1 = between first and second SD (mild residual dysplasia) and Tönnis 2 = beyond second SD (severe residual dysplasia) (Table 1) [8]. In the case of an insufficient radiograph (tilted or malrotated), for example, due to a recalcitrant infant, one more radiograph was performed. In the rare case of two insufficient images, the radiographic follow-up was rescheduled a few months later. When at least one of the hips did not show physiological values in the first pelvic radiograph, a second radiographic follow-up was recommended and scheduled at the end of the third year of life. It has to be underlined that from the first to the second radiographic follow-up, no treatment was performed. These were the patients of interest for this examination. Infants with bilaterally physiological hips in the first radiograph were not included. When a second radiograph was performed due to a residual dysplasia on the first radiograph, the measurement of AI was repeated in order to evaluate the dynamics of acetabular development. In these children, a third pelvic radiograph was recommended within the framework of a 'milestone-examination', approximately half a year before starting school. Again, around the age of 6, AI was measured.

The following exclusion criteria were applied: (a) relevant comorbidities or systemic diseases (e. g. neurological syndromes); (b) incomplete documentation of the ultrasound findings; (c) physiological presentation of both hips in the first pelvic radiograph following ultrasound-monitored treatment; (d) signs of avascular necrosis of the femoral head; (e) insufficient pelvic radiography (e.g. tilt and malrotation) and (f) early open reduction.

Statistics

Dependent *t*-test for paired samples was applied to assess an evolution of AI. The test was applied for the whole sample, then after distribution according to sex (girls vs. boys) and initial Graf type (I–IV in the first ultrasound examination). A correlation of the initial Graf classification (Graf type I–IV) with the AI at the first, second and third radiographic examination was calculated with the Kruskal–Wallis test. In beforehand, the normal distribution of the discrepancies of AI from the first to the second and from the second to the third follow-up was verified with the Kolmogorov–Smirnov test. Generalized estimating equation (GEE) was used to

Fig. 1



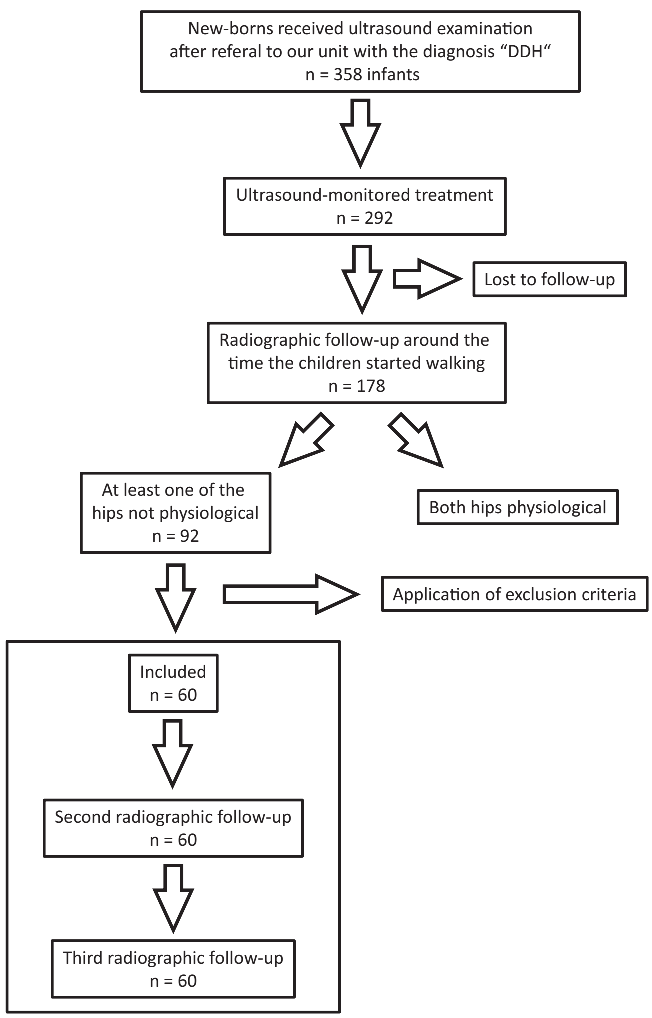
Anterior–posterior radiograph of a 26-month-old child, supine position, hips extended, patellae facing forward. The measurements of AI were performed using GE Centricity Universal Viewer Version 6.0 (General Electric Healthcare, Chalfort St Giles, UK). First, a horizontal line was drawn connecting the upper margins of the radiolucent triradiate cartilage (Hilgenreiner's line). Then a line was drawn through the medial edge of the sclerotic acetabular zone and tangential to the lateral sourcil. The angle between these two lines represented the AI. AI, acetabular index.

Table 1 **Age-dependent classification system for the acetabular index described by Tönnis [7]**

	Female				Male			
	Mild residual dysplasia (1s)		Severe residual dysplasia (2s)		Mild residual dysplasia (1s)		Severe residual dysplasia (2s)	
Age	Right	Left	Right	Left	Right	Left	Right	Left
10–12 months	24.5	27	29	31.5	23.5	25	27	29
13–15 months	24.5	27	29	31.5	23	24	27.5	27.5
16–18 months	24.5	26	29	30.5	23	24	26.5	27
19 months–2 years	24	25.5	28	30.5	21.5	23	26.5	27
2–3 years	22	23.5	25.5	27	21	22.5	25	27
3–5 years	18	21	22.5	25.5	19	20	23.5	24
5–7 years	18	20	23	23.5	17	19	21	23

Threshold values indicate mild or severe residual dysplasia.
1s, first SD; 2s, second SD.

Fig. 2



Patient selection after application of inclusion and exclusion criteria.

assess the statistical differences of the values of AI from the first to the second and from the second to the third radiographic follow-up. Significance was expressed by *P*-values. Data were processed using SPSS Statistics, Version 26 (IBM, Armonk, New York, USA).

This study was conducted retrospectively from data for clinical purposes. An IRB official waiver of ethical approval was granted from the local Ethics Committee of our institution (126/20 – FSt/Sta).

Results

Of the 292 infants undergoing ultrasound-monitored abduction treatment, 178 showed up around the time they started walking for a first radiographic follow-up. At this age, the rate of successfully obtained radiographs in the first attempt was 92% (in 164 of 178 infants). Six rather calcitrant infants did not allow sufficient radiographic imaging even in the second attempt. In these cases, no further attempt was forced and follow-up was postponed a few months. On 92 radiographic controls, at least one of the hips presented an AI classified Tönnis 1 or worse and, thus, were suitable for this study. Finally, after application of the exclusion criteria, 60 patients were included in this examination (46 girls and 14 boys), resulting in a total of 120 hips (Fig. 2). The mean age at the first follow-up was 15.6 months (range, 12–19 months). Consequently, all these infants received a second radiographic follow-up at the end of their third year of life (mean age 33.9 months, range, 26–45 months) and a third follow-up approximately half a year before starting school (mean age 5.9 years, range, 5.2–8 years).

For reasons of clarity, all the measurement results are presented in Table 2.

GEE confirmed the statistical difference of the values of AI from the first to the second and from the second to the third follow-up ($P < 0.001$, respectively).

Dependent *t*-test for paired samples indicated a highly significant improvement of the AI from the first to the second and from the second to the third follow-up. Null hypothesis (no difference of the values) was rejected ($P < 0.001$). The Kolmogorov–Smirnov test confirmed a normal distribution of the differences of the AI from the first to the second and from the second to the third follow-up ($P = 0.002$ and 0.000).

For the whole sample, the values of AI decreased from the first to the second follow-up by a mean of 5.6° (range, 1°–16°) and from the second to the third follow-up by 7.6° (range, 2°–28°).

The percentage distribution into the Tönnis classification system changed remarkably: For example, in the first follow-up, 36 of the 120 hips were evaluated 'severely dysplastic' (Tönnis 2), in the third follow-up there was only 1. Respectively, in the first follow-up, 44 of the 120 hips were normal (synonymous with the physiological hip in unilateral residual dysplasia, Tönnis 0) and 98 in the third follow-up (Table 2).

Kruskal–Wallis test showed no correlation between the initial Graf classification in ultrasound (Graf type I–IV) and the AI at the first, second or third radiographic follow-up. Categorized by the initial Graf type, on the first radiographic follow-up, AI for the initially Graf type I hips ranged from 22° to 32°, for the Graf type II hips from 20° to 33°, for the Graf type III hips from 23° to 33° and for the Graf type IV hips from 24° to 37°, respectively. The values of AI decreased for the Graf type I hips from the first to the second follow-up by a mean of 5.5° (range, 1°–14°), from the second to the third follow-up by 8.3° (range, 3°–20°). Respectively, the further data in brief: Graf type II: 5.5° (1°–13°) and 6.7 (1°–28°); Graf type III: 5.5° (1°–16°) and 8.6° (1°–22°); Graf type 4: 5.5° (1°–15°) and 10.8° (4°–23°). *t*-test for paired samples showed $P < 0.001$ (Fig. 3).

Categorized by sex, the values of AI for the girls decreased from the first to the second follow-up by a mean of 5.2° (range, 1°–15°) and from the second to the third follow-up by 8.0° (range, 1°–28°). Respectively, for the boys: 6.3° (2°–16°) and 6.8 (1°–22°). *t*-test for paired samples showed $P < 0.001$ (Fig. 4).

Discussion

The most important finding in this examination was the presence of a recognizable development for the residually dysplastic hips: Regarding the Tönnis classification system, in the first radiographic follow-up, two-thirds of the hips showed either mild or severe residual dysplasia, whereas in the third radiographic follow-up about four-fifths of the hips showed normal values. The null hypothesis was rejected: the values for AI decreased significantly

Table 2 Classification of the hips according to the Tönnis system, percentage-based frequency distribution

	First follow-up	Second follow-up	Third follow-up
Classification according the system described by Tönnis	Number of hips (*n*), percentage		
Tönnis 0 (normal)	44 (36.6%)	61 (50.8%)	98 (81.7%)
Tönnis 1 (mild residual dysplasia)	40 (33.3%)	45 (37.5%)	21 (17.5%)
Tönnis 2 (severe residual dysplasia)	36 (30%)	14 (11.7%)	1 (0.8%)

The amount of residually dysplastic hips decreased considerably from the first to the second and from the second to the third follow-up. At the third follow-up, only one hip was classified Tönnis 2.

from the first to the second and from the second to the third follow-up.

The second most important finding was the lacking correlation with the initial ultrasound presentation according to the Graf classification (Graf type I–IV) and the AI at each particular follow-up. For example, an initial Graf type 2a+ hip presented an AI above 30° in the first radiographic follow-up. On the other hand, an initial Graf type D hip presented with an AI of 22°. This underlines the necessity for a radiographic follow-up even after successful ultrasound-monitored treatment.

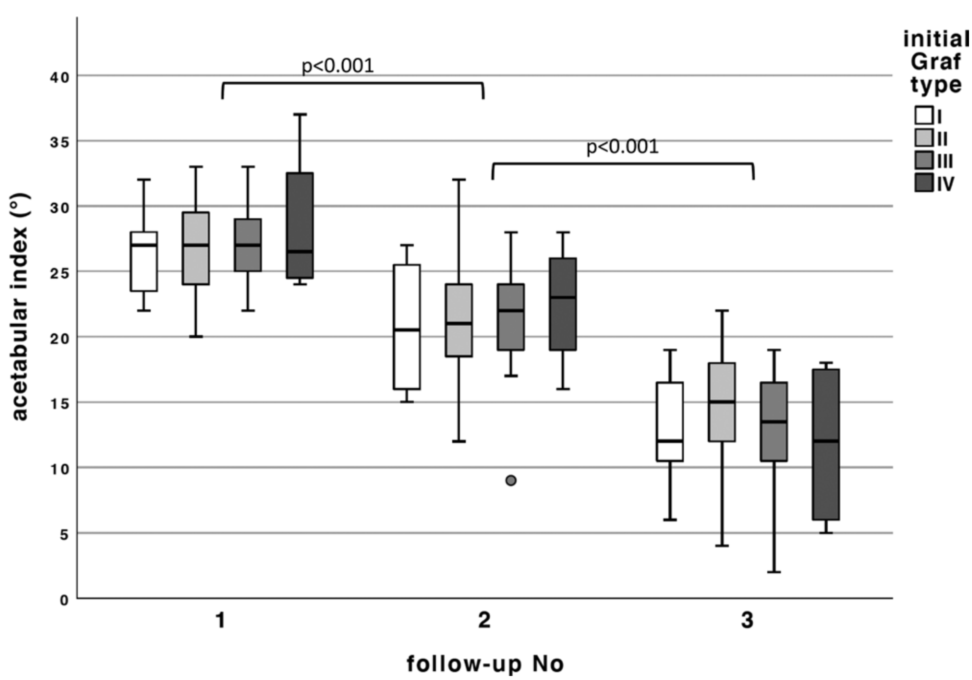
The presence of a residual dysplasia has already been described: Kubo *et al*. [12] conducted an examination on newborns with unstable hips (Graf type D, III and IV), all of which were treated with an abduction splint. This resulted in normal ultrasound findings at the end of the treatment (Graf type I). Despite this, the radiographic follow-up at 12–24months and 24–48 months revealed a considerable percentage of hips with a residual dysplasia according to the Tönnis classification. The authors describe mild residual dysplasia in 33.7% and severe residual dysplasia in 12% of the hips. This means that almost every second hip showed a pathology to the time of the first follow-up at 12–24 months [12]. In our study population, two-thirds of the hips showed residual dysplasia at the first follow-up. Kubo *et al*. [12] reported furthermore that to the time of the second radiographic follow-up at 24–48 months, the percentage of radiographically normal hips increased to a total of 73.5%. This resembles our findings. In contrast to the study of Kudo *et al*. [12], in our examination, the selection of the patients was not based on pathological findings in the initial sonographic presentation, but on the presence of a residual dysplasia in at least one of both hips in the first radiographical follow-up. This explains the higher percentage distribution of residually dysplastic hips in this examination.

In this context, Spaan *et al*. [16] concluded in their analysis that the findings in ultrasonographic and radiographic imaging in DDH cannot be translated. After the comparison of different parameters of ultrasound and radiographic imaging (including the Graf and Tönnis classification) acquired on the same day, no significant correlation was found [16].

Novais *et al*. [14] asked whether patient-specific factors and ultrasound values at initial examination translate to short-term residual dysplasia in patients successfully treated in a harness. Graf type IV was the only variable associated with the AI-values at a radiographic follow-up around the age of 12 months. The authors stated that risk of acetabular dysplasia following harness treatment is not negligible and radiographic surveillance is warranted [14].

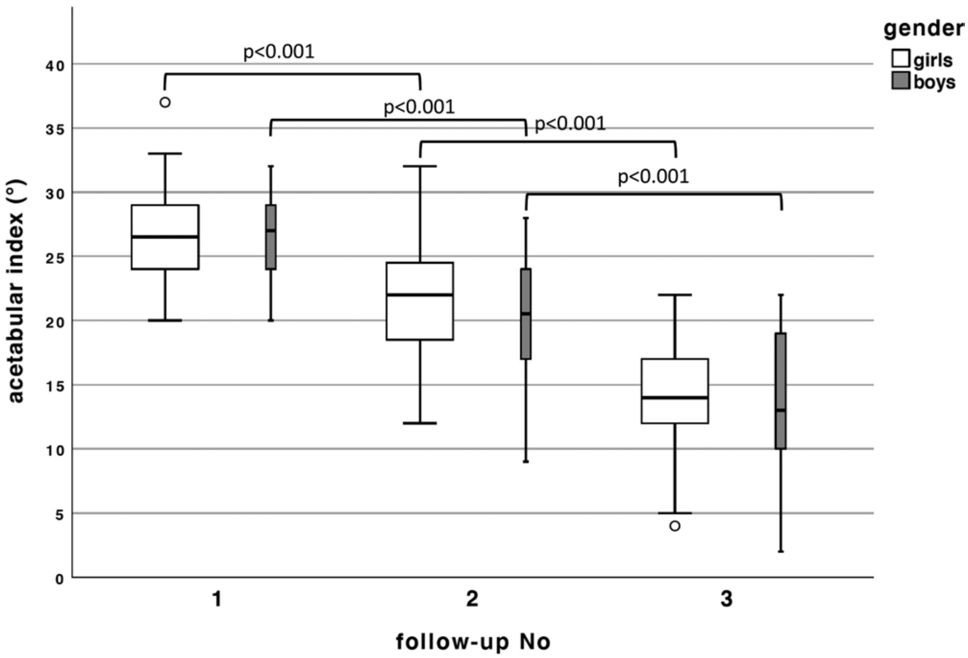
Utzschneider *et al*. [17] reported on the inappropriate correlation of sonographic and radiographic standard values concerning DDH. The authors used the Graf classification

Fig. 3



Development of the AI, categorized by the initial Graf type at the first ultrasound examination. The boxplots represent maximum, 1st quartile, median, 3rd quartile and minimum (top down). Kruskal–Wallis test showed no correlation of the Graf type (I–IV) and the AI. AI, acetabular index.

Fig. 4



Development of AI in female ($n = 92$) and male hips ($n = 28$). The boxplots represent maximum, 1st quartile, median, 3rd quartile and minimum (top down). The bar width represents the percentage distribution of girl's and boy's hips. AI, acetabular index.

for the sonographic findings and the AI for the radiographic evaluation. AI was categorized according to the Tönnis classification. A considerable statistical discrepancy between the sonographic findings at the end of ultrasound-monitored treatment and the radiographic findings at walking age was found. The authors stated that the sonographic and radiographic reference values should be subject to a critical reevaluation [17]. It has to be pointed out that the AI has been proven to be a reliable parameter with an inter- and an intraobserver reliability of ±3° to 4° (95% confidence interval) [18,19]. Utzschneider *et al.* [17] hypothesized that the widely used radiographic standard values according to the Tönnis classification might be too strict and, thus, recommend further examination [17]. The results of the present study underpin the hypothesis of Utzschneider *et al.* [17]: the allocation of AI according to the Tönnis classification measured in the first radiographic follow-up showed a rather high percentage portion of residually dysplastic hips. These findings were put into perspective by the Tönnis classification in the second and third radiographic follow-up, where the percentage portion of the residually dysplastic hips was remarkably lower. This leads to the conclusion that particularly in early childhood, the severity of residual hip dysplasia might be overestimated by the Tönnis classification system. The classification of AI should be reassessed critically.

In this examination, three hips in two patients showed severe residual dysplasia with an AI above 30° at the age of around 3 years. Indeed, in these hips, the dynamics of acetabular development from the first to the second radiographic follow-up was recognizable but insufficient to fall below a pragmatically chosen threshold of 30° AI. In these cases, Dega acetabuloplasty was recommended and performed. In this study, all patients presenting with a nonphysiological pelvic radiograph at the first follow-up received a rather short-term second follow-up at the age of around 3 years. This enabled an evaluation of the dynamics of acetabular development and – in persisting severe residual dysplasia – an early intervention at preschool age.

Allington *et al.* [9] retrospectively reviewed a consecutive series of infants, successfully treated with a Pavlik-harness and with a normal pelvic radiograph at 2 years of age. A total of 109 hips in 83 children were followed up for 10 years with the result of a clinically and radiographically normal examination. The authors concluded that in a selected group of patients treated for DDH with a Pavlik-harness, under a strict protocol and with a normal radiograph at 2 years of age, a long-term follow-up is not necessary [9].

However, Pun *et al.* [6] reported in their examination on an adolescent-onset acetabular dysplasia. It has been theorized that the development of adolescent-onset dysplasia involves delayed ossification in the triradiate cartilage and insufficient development of the lateral secondary ossification centers at the acetabular rim. The latter ossify during adolescence between 12 and 18 years [6]. A short-term follow-up as above-mentioned carries the risk of a rather late diagnosis in a young adult. With the aim of an intervention prior to irreversible damage, follow-up until skeletal maturity seems sensible.

Shaw *et al*. [20] lately stated in a systematic review of the literature that late acetabular dysplasia is not an uncommon clinical entity following successful treatment, with evidence occurring in 9.5% of hips and 4.1% requiring surgery. Particularly, children treated by institutions with a standard treatment protocol had significantly lower incidences of radiographic late dysplasia [20]. The same fundamental position is underlined by Multerer *et al*. [13] in their review of the current literature. The authors recommend a radiological follow-up of the affected as well as the unaffected side until the end of the growth phase due to the risk of residual dysplasia [13].

Additionally, in an outpatient department and examiners with different level of experience, a strictly standardized protocol has an advantage over an individualized regimen. This might imply a number of three pelvic radiographs in preschool age. Of course, needless radiographs should be avoided by all means. And radiation exposure is becoming a subject for an increasing number of parents. Sarkissian *et al*. [15] found that the notable incidence of residual hip dysplasia in the radiographic follow-up after previous DDH normalization in ultrasound outweighs the risk of radiation exposure, which amounts to an effective dose of less than 0.01 mSV. Radiographic follow-up at walking age allows timely diagnosis and early intervention in residual dysplasia [15].

This examination has several limitations. First, although parents were strongly recommended to make use of the radiographic follow-up after ultrasound-monitored treatment, more than a third of the infants were lost to follow-up. Unlike the ultrasound screening, the radiographic control is not mandatory in the German health care system, so the follow-up depends on the strong recommendation alone. In addition, our outpatient department has a supra-regional catchment area for the ultrasound-monitored treatment in DDH. After successful treatment, several infants might have received further follow-up close to their homes. Second, there were quite a number of drop-outs after application of the exclusion criteria. This makes aware of the fact that the establishment of a close-meshed follow-up does not come without difficulties. Third, three hips in two patients underwent pelvic osteotomy right after the second follow-up due to a severe residual dysplasia. Of course, in these three hips, the values for AI dropped dramatically. This affected the statistical data from the second to the third follow-up. Fourth, it would be of great interest to follow-up these cases until skeletal maturity to overview the potential of acetabular development. This is only partially possible with a follow-up until the end of preschool age. Fifth, it would have been useful to perform the first radiograph at the end of the abduction treatment in order to facilitate the comparison with the radiographic follow-up images. Since this is a retrospective examination, the timing of the first radiograph has been determined around walking age. This issue might be analyzed using a prospective study design. And sixth, in this examination, solely one radiographic parameter has been evaluated. The AI provided an objectifiable tool in the preset framework of this retrospective examination. In the day-by-day clinical practice and especially in the early stage, the radiographic assessment of an infant's hip requires a more differentiated set of additional radiographic parameters, for example, the Shenton's line, the extrusion index and the center-edge angle in older children.

Conclusion

Even after normal values have been achieved at the end of ultrasound-monitored treatment, there remains a risk of residual dysplasia of the hips. There is no correlation with the initial Graf type in the ultrasound screening and the AI in the particular radiographic follow-up. Based on our findings, we recommend a radiographic control shortly after the time children started walking (around 12–18 months). When the first pelvic radiograph shows features of residual dysplasia, a mid-term follow-up should be scheduled to evaluate the dynamics of acetabular development, for example, at the end of the third year of life. It has been demonstrated that the majority of the initially residually dysplastic hips reach normal values at the end of preschool age. In the absence of a sufficient development, acetabuloplasty should be considered.

In every once-treated hip and, in particular, when the radiographic examinations show nonphysiological findings, further follow-up is recommended. In order to gain comprehensive knowledge of the acetabular development, further examinations with a longitudinal character are necessary.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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