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ORIGINAL PAPER

Variables influencing the pelvic radiological evaluation in children with developmental dysplasia of the hip managed by closed reduction: a multicentre investigation

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Abstract

Purpose This study aims to evaluate (1) the probability to achieve normal pelvic radiographs in children with developmental dysplasia of the hip (DDH) treated by closed reduction and (2) the amount of time needed to achieve normal pelvic radiographs and to assess what factors influence both probability and time to achieve normal radiographic parameters following CR and spica cast immobilization for DDH.

Methods We retrospectively reviewed 436 patients (393 girls, 43 boys; 507 hips) with DDH treated by closed reduction (CR). Tönnis grade, AVN, acetabular index (AI), centre-edge angle (CEA), and Severin radiographic grade were evaluated on plain radiographs. Criteria to rate pelvis radiographs as normal were established. Cox regression was used to evaluate the factors influencing the probability and the time to achieve normal radiographs.

Results According to our criteria, 167 hips (32.9%) achieved normal radiographic parameters during follow-up. The overall amount of time to achieve normal pelvis radiographs was 36.1 ± 15.5 months. Patients older than 24 months of age at the time of CR needed longer time to achieve normal radiographic parameters (55.2 ± 28 months) compared with other age groups. Cox regression analysis suggested the overall cumulative probability of recovery increased by 46% at five years following CR, then it tended to plateau with an annual increase less than 5%. Age older than 24 months, bilateral dislocation, pre-operative AI greater than 40°, and AVN were risk factors for reduced probability of achieving normal radiographic parameters.

Conclusions The cumulative probability of achieving normal pelvis radiographs increases linearly during the first five years following CR, then it tends to plateau. Age older than 24 months and Tönnis grade III and IV are associated with longer time to achieve normal radiographic parameters. Age older than 24 months, bilateral dislocation, pre-operative AI greater than 40°, and AVN are risk factors for reduced probability of achieving normal radiographic parameters in children with DDH treated by closed means.

Keywords Developmental dysplasia of the hip · Closed reduction · Acetabular index · Avascular necrosis of femoral head · Normal pelvis radiographs

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Introduction

Developmental dysplasia of the hip (DDH) is a congenital defect of the hip joint characterized by an abnormal anatomical relationship between the femoral head and the acetabulum, leading to acetabular dysplasia, subluxation, and complete dislocation of the femoral head [1, 2]. The aim of treatment for DDH is to obtain a stable and concentric reduction of the hip as early as possible and avoid the occurrence of avascular necrosis of the proximal femoral epiphysis (AVN). At present, closed reduction (CR) and spica cast immobilization under general anaesthesia are the treatment of choice for children younger than 24 months of age [1–3].

It has been reported that about 70% of children with DDH managed by CR can achieve satisfactory radiographic outcome, while the remaining one-third develops residual acetabular dysplasia [4, 5]. The rate of Severin's grade I and II hips following closed reduction and spica cast immobilization reported by different researchers varies between 62 and 93.6% [6–10]. Although most patients can achieve satisfactory radiographic outcome, not all radiographs can be considered normal, especially in patients with Severin type II hips. Additionally, some controversy still exists whether the Severin grading system can be used in children younger than four to six years of age. Additionally, no study has investigated the factors influencing the probability to achieve normal radiographs and the time it takes for radiographs to become normal.

The aim of this study was to retrospectively review all patients with DDH treated by CR in three institutions, to evaluate (1) the probability to achieve normal pelvic radiographs in children with DDH treated by closed reduction and (2) the amount of time needed to achieve normal pelvic radiographs and to assess what factors influence both probability and time to achieve normal radiographic parameters following CR and spica cast immobilization for DDH.

Materials and methods

The medical records of 848 children (3 institutions) with late detected DDH treated by CR during the period 2004–2015 were collected and retrospectively reviewed.

The inclusion criteria were (1) confirmed diagnosis of late-presenting DDH treated by CR and cast immobilization; (2) complete clinical and radiographic data; (3) follow-up longer than 24 months.

A total of 412 out of 848 patients (48.6%) were excluded from the analysis due to (a) incomplete radiological data and follow-up shorter than 24 months (288 patients; 69.9%); (b) concomitant diagnosis, such as cerebral palsy, tethered cord syndrome, meningomyelocele, arthrogryposis multiplex congenita, or other neuromuscular diseases (83 patients; 20.1%); (c) treatment by open reduction due to redislocation or widened medial joint space (41 patients, 10%).

Four hundred and thirty-six patients (393 girls, 43 boys; 507 hips) met the inclusion criteria. The left hip was involved in 217 patients (49.8%), the right in 141 patients (32.3%), and both hips in 71 patients (16.3%); another seven patients (1.6%) had bilateral dislocation but only one hip could be included due to redislocation of the contralateral one. The average age at the time of CR was 16.3 ± 5.1 months (range, 6–35). The mean length of follow-up was 48.1 ± 20.6 months (range, 24–139).

CR was performed under general anesthesia, plus adductor tenotomy if the adductor was considered to hinder reduction. Arthrography was performed in order to evaluate the position of the hip and assist reduction. After CR, an abduction cast (spica cast or dynamic cast [8]) was applied to maintain reduction for three to four months. The cast was changed once six weeks after initial CR. After the cast removal, patients were placed in an abduction brace for three months full-time, followed by night-time use only until a stable reduction was achieved.

Radiographic assessment

All patients were followed up for at least 24 months. Anteroposterior (AP) pelvic radiographs of the hip were obtained at each follow-up visit. The acetabular index (AI) prior to CR was measured on AP pelvic radiographs. The dislocation of the hip was also assessed on AP pelvic radiographs according to the Tönnis grading system [11]. Following CR, the AI and centre-edge angle of Wiberg (CEA) were also measured at each follow-up visit. The measurement of AI and CEA was performed with the Picture Archiving and Communication System (PACS) by two observers who were blinded to the final Severin classification (TRY and LYQ).

AVN of the femoral head was identified according to the criteria described by Salter et al. [12] and was graded according to Bucholz and Ogden's method [13]. We grouped type I AVN with the normal hip category because type I AVN is a transient ischemia of the femoral head which can resolve completely [14, 15]. Two independent raters (LYQ and FC) evaluated AVN. If they could not come to an agreement on the type of AVN, a discussion with at least three other senior paediatric orthopaedic surgeons was performed. Final radiographic outcomes of the hips were graded according to the criteria described by Severin [14].

Normal pelvic radiographs criteria

At each follow up visit, all hips were evaluated to determine whether or not the hip achieved normal pelvic radiographic parameters.

To be considered normal, the hip had to meet the following criteria: (1) normal AI and CEA (no more than one standard deviation above the mean value for normal age matched

children) as reported by Shi et al. [16, 17]; (2) normal appearance of the teardrop [18] and normal morphology of the acetabulum; (3) no signs of AVN and ossific centre of the affected hip of similar shape and size compared with the unaffected side. Moreover, to be considered normal, each hip had to meet the above criteria in two consecutive assessments (at 1-year interval). The time elapsed between CR and normalization of all radiographic parameters was defined as "recovery time" and it was expressed in months calculated.

Statistical analysis

All statistical analyses were performed using the statistics package SPSS 22.0 (SPSS, Chicago, IL, USA). Data were expressed as numerical variable, frequencies, and percentages, with the means and standard deviations. Cox regression analysis was used to evaluate the factors influencing the probability and the time to reach normal radiographic parameters. When performing Cox regression analysis, the final event was defined as achieving normal pelvis radiographs. For patients achieving normal radiographic parameters, the variable "time" was the "recovery time." On the other hand, patients not achieving normal radiographic parameters at final follow-up were recorded as censoring and, for them, the variable "time" corresponded to the follow-up time. Chi-square test, one-way analysis of variance (ANOVA), or Student's *t* test were also used to assist further analysis. The level of statistical significance was set at $P < 0.05$.

Results

Radiographic measurements showed good to excellent inter-observer reliability for AI (intraclass correlation coefficient, ICC = 0.934) and CEA (ICC = 0.862).

The mean preoperative AI was 35.4° ± 4.4°. Among the included 436 patients (507 hips), 235 hips (46.4%) were Tönnis grade II, 239 (47.1%) were grade III, and 33 (6.5%) were grade IV. The ossific nucleus of the femoral head was presented in 407 hips (80.3%).

At final follow-up, the mean AI and CEA were 21.8° ± 6.1° and 23.6° ± 8.9°, respectively. AVN occurred in 66 hips (13%); there were 55 type II AVN (77.3%), 12 type III (18.2%), and three type IV (4.5%). According to the Severin classification, 331 (65.3%) hips were graded as Severin I, 60 (11.8%) as Severin II, 113 (22.3%) as Severin III, and three (0.6%) hips as Severin IV.

Overall, 167 hips out of 507 (32.9%) achieved normal radiographs during follow-up: 81.4% and 92.8% of the hips recovered within 48 and 60 months post CR, respectively (Fig. 1).

Twenty-four out of 167 hips underwent secondary pelvic osteotomy. Among the 143 hips (29%) that achieved normal radiographic parameters without pelvic osteotomy, 83.2% and 93% of the hips recovered within 48 and 60 months post CR, respectively (Fig. 2).

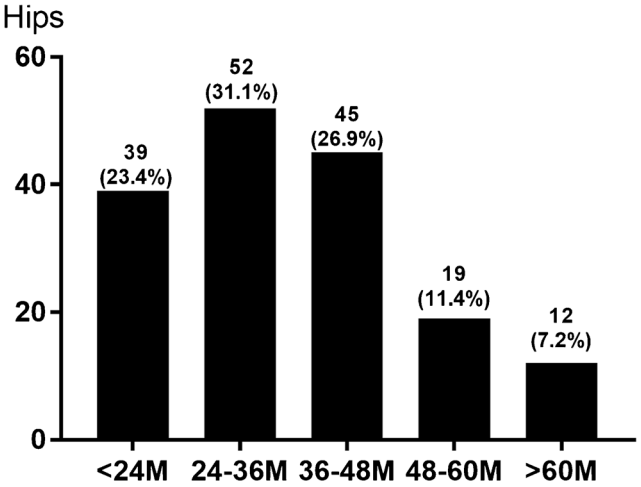

Fig. 1 Hips achieved normal radiographs in different age groups

The mean recovery time was 36.1 ± 15.5 months ($n = 167$). Hips were divided into four groups according to age at the time of CR: (a) age less than 12 months; (b) age between 12 and 18 months; (c) age between 18 and 24 months; (d) age over 24 months.

The mean "recovery time" of patients younger than 12 months (30.1 ± 18.3 months) was significantly shorter compared with that of other age groups ($P < 0.05$); on the other hand, the mean "recovery time" of patients older than 24 months (55.2 ± 28 months) was significantly longer compared with that of other age groups ($P < 0.05$) (Table 1).

The "recovery time" of Tönnis type II hips (32.5 ± 17.2 months) was significantly shorter compared with that of Tönnis III (39.6 ± 13.2 months) and IV (40.8 ± 9.6 months) ($P < 0.05$). However, there was no significant difference on

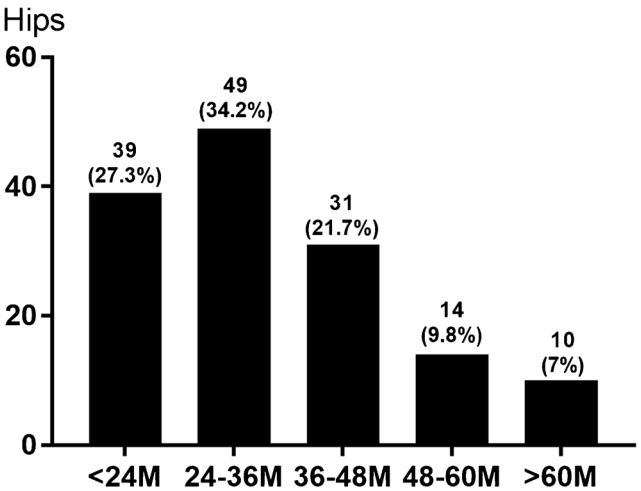

Fig. 2 Hips achieved normal radiographs in different age groups (excluded the hips underwent secondary pelvic osteotomy)

Table 1 The rate and time to achieve normal pelvis radiographs among four age groups

		< 12 M	12–18 M	18–24 M	≥ 24 M	F/χ^2	*P*
Normal pelvis radiographs	No	52	151	108	29	13.716	0.003
	Yes	37 (41.6%)	85 (36%)	42 (28%)	3 (9.4%)		
Time (M)		30.1 ± 18.3	36.4 ± 13.0	39.4 ± 15.1	55.2 ± 28	4.211	0.007

M months

"recovery time" among left, right, and bilateral side ($F = 0.967$, $P = 0.382$).

Cox regression analysis

Cox regression analysis found the overall probability of achieving normal radiographic parameters increased with follow-up time (Fig. 3). The cumulative probability of achieving normal pelvis radiographs was 10%, 21%, 36%, and 46%, two, three, four and five years post CR, respectively. After the fifth year, the probability to have normal radiographs tended to plateau with an annual increase less than 5% (Fig. 3). Age, sides, preoperative AI, and AVN were the risk factors influencing the probability of achieving normal radiographic parameters (Table 2).

Cox regression analysis also indicated that the cumulative probability of achieving normal radiographic parameters in patients older than 24 months was significantly lower compared with that in the other three age groups ($P < 0.05$); no differences were observed among patients younger than 24 months of age (< 12, 12–18, and 18–24 months; $P > 0.05$) (Fig. 4). Similar results were confirmed by chi-square test assessment (Table 1).

The cumulative probability of achieving normal radiographic parameters in patients with bilateral dislocation was significantly lower compared with that in patients with unilateral involvement ($P = 0.046$); no significant differences were observed between left and right side ($P = 0.613$) (Fig. 5).

The cumulative probability of achieving normal pelvis radiographs in patients with preoperative AI greater than 40° was significantly lower compared with that in patients with pre-operative AI less than 30° and AI between 30° and 40° ($P = 0.03$); no significant differences were observed between patients with pre-operative AI less than 30° and AI between 30° and 40° ($P = 0.331$) (Fig. 6).

The cumulative probability of achieving normal pelvis radiographs in patients without AVN (no AVN and type I AVN) was significantly higher compared with patients with type II, III, and IV AVN ($P < 0.001$); no significant differences were observed between patients without AVN and patients with type I AVN ($P = 0.135$) (Fig. 7).

Discussion

The present study found that although 65.3% of the hips were Severin grade I, only 32.9% of the hips achieved normal pelvic radiographs, meaning that half of the patients with excellent radiographic outcome had abnormal pelvis radiographs. Severin classification is widely used to evaluate the

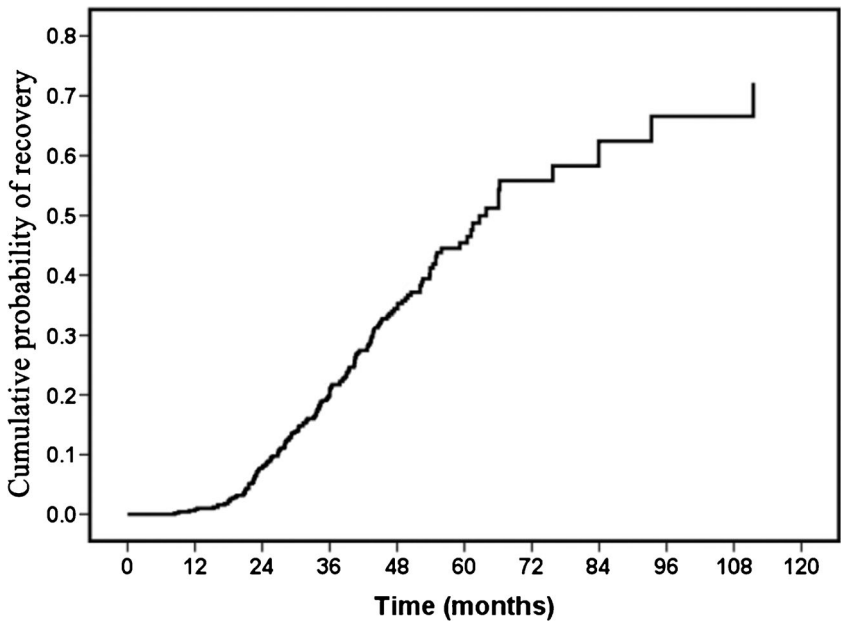

Fig. 3 Overall probability of the hips achieving normal pelvic radiographs under Cox proportional regression model

Table 2 Predictors of achieving normal pelvis radiographs by Cox regression

	Coefficient	SE	Wald	*P*	RR	95% CI of RR
Age	− 0.057	0.018	10.476	0.001	0.944	0.912, 0.978
Gender	0.207	0.258	0.645	0.422	1.230	0.742, 2.041
Sides	− 0.225	0.095	5.579	0.018	0.798	0.662, 0.962
Tönnis grade	− 0.044	0.140	0.100	0.752	0.957	0.727, 1.258
Ossific nucleus	0.375	0.228	2.701	0.100	1.454	0.930, 2.273
Initial AI	− 0.048	0.018	7.195	0.007	0.953	0.921, 0.987
AVN	− 1.449	0.297	23.740	0.000	0.235	0.131, 0.421

CI confidence interval, *RR* relative risk, *SE* standard error

radiographic outcome of patients with DDH treated by closed or open reduction and it utilizes the CEA as the main parameter to assess the radiographic outcome [14]. However, studies have shown poor intra-observer and inter-observer reliability of Severin classification [19, 20]. Additionally, Li et al. have shown that AI, and not CEA, is the best predictor of late residual acetabular dysplasia after closed reduction in developmental dysplasia of the hip [6]. In order to identify what hips achieved to normal radiographic parameters, we evaluated AI, CEA [16, 17], and the morphology of the hip joint.

Our study showed that during the first five years following CR, the probability of achieving normal pelvic radiographs increased by 46% (Fig. 3). On the other hand, after the fifth year, the cumulative probability tended to plateau, with an annual increase less than 5% (Fig. 3). Among the 167 hips with normal pelvic radiographs, 93% normalized within the first five years post CR (Figs. 1 and 2). It has been shown that the most rapid ossification period of the acetabulum in normal children occurs during the first fourtofive years of life; after this period, the annual growth of the acetabulum decreases [17, 21–23]. More recently, Li et al. reported the growth of the acetabulum can continue until seven to eight years following CR in patients with satisfactory radiographic outcome [6, 24]. Albinana et al. [4] and Fu et al. [25] also found AI stops to improve six years post CR in patients with Severin I/II hips.

Our study also showed that age is an important factor influencing the cumulative probability and the time to achieve normal pelvis radiographs (Fig. 4). In particular, patients older than 24 months of age had lower chances and needed more time to have normal pelvis radiographs compared with younger patients. Several studies have shown that older age is a key factor leading to poor outcome and to secondary pelvic osteotomy in patients with DDH treated by CR [24, 26–28]. Li et al. found that patients aged 24–36 months had a significantly lower rate of Severin type I hips (39.2%) compared with patients younger than 24 months [8, 29, 30]. This is probably related to the decreased growth potential of the acetabulum in older patients [17, 21–23].

Our results also found bilateral hip dislocation decreases the cumulative probability to achieve normal pelvis

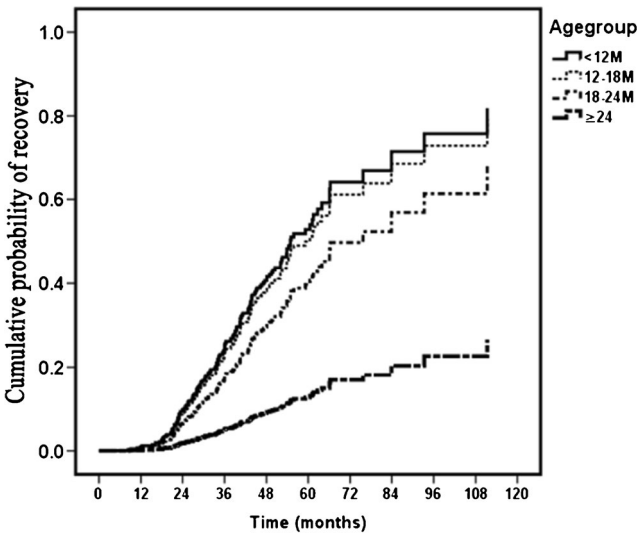

Fig. 4 Cumulative probability of the hips achieving normal pelvic radiographs in different age groups under Cox proportional regression model. An age older than 24 months significantly decreased the probability of achieving normal pelvic radiographs

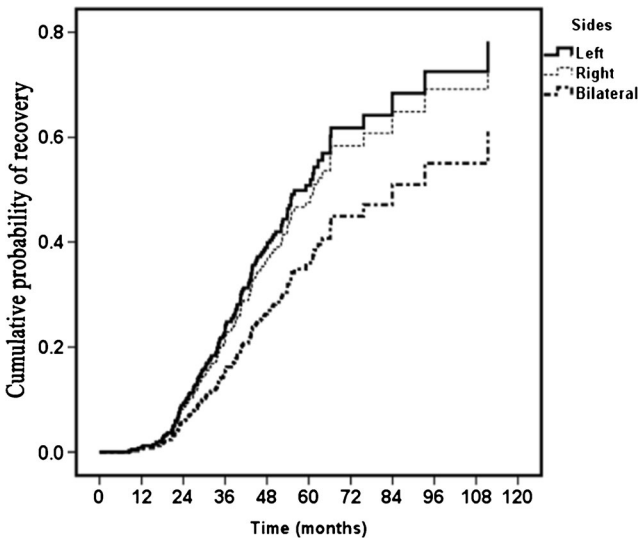

Fig. 5 Cumulative probability of the hips achieving normal pelvic radiographs in different sides under Cox proportional regression model. Bilateral dislocation significantly decreased the probability of achieving normal pelvic radiographs

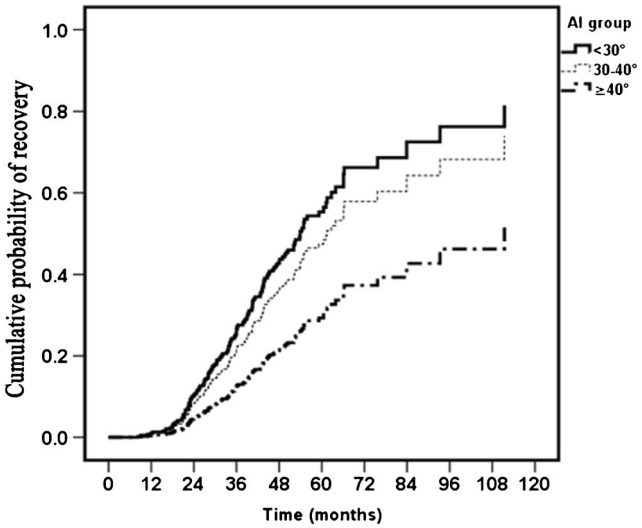

Fig. 6 Cumulative probability of the hips achieving normal pelvic radiographs in different AI groups under Cox proportional regression model. A pre-operative AI greater than 40° significantly decreased the probability of achieving normal pelvic radiographs

radiographs. At present, there is still controversy if bilateral DDH is a risk factor for poor outcome. Kitoh et al. reviewed 45 hips with DDH treated by closed reduction; they found bilateral DDH showed significantly poorer radiographic outcome than unilateral [7]. However, a recent long-term follow-up study by Terjesen could not identify any significant difference on radiographic outcome between unilateral and bilateral DDH [31]. Eamsobhana et al., Kaneko et al., and Sankar et al. also found bilateral dislocation did not lead to poor radiographic outcome [10, 26, 32]. The number of bilateral cases

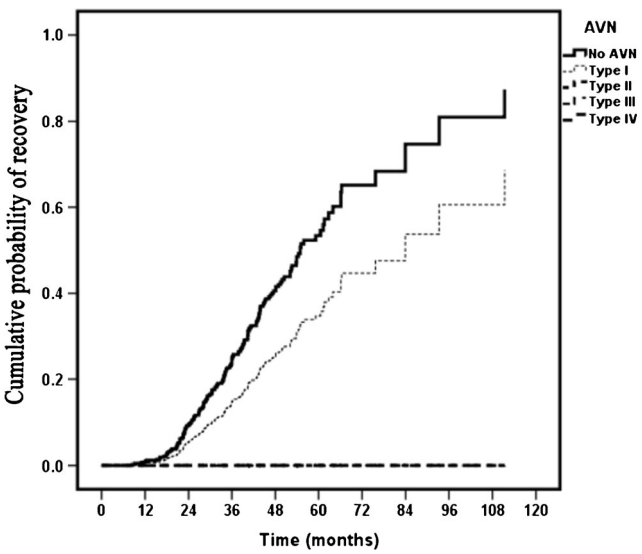

Fig. 7 Cumulative probability of the hips achieving normal pelvic radiographs in different AVN groups under Cox proportional regression model. No hips with type II, III, or IV of AVN achieved normal pelvic radiographs

included in our study (71 patients; 142 hips) was higher compared with that in previously published works that included less than 14 bilateral patients, and it may contribute to strengthen our findings [7, 10, 26, 31, 32].

Interestingly, we also found that pre-operative AI greater than 40° decreased the cumulative probability to achieve normal pelvis radiographs. This result was similar to previously published studies [24, 33–35]. In particular, Li et al. reported that the best AI improvement should be expected during the first four years following CR (35% in patients younger than 12 months). Therefore, hips with AI over 40° are unlikely to achieve normal radiographic parameters even if 35% remodeling rate should occur.

We suggest the best time for secondary pelvic osteotomy should be around five years following CR, as also reported by previous studies [7, 10, 36]. In particular, we found that patients with DDH treated by CR have 46% probability to achieve normal pelvic radiographs without the need of subsequent pelvic surgery; unnecessary surgeries can potentially be avoided if pelvic osteotomy is performed five years after CR. However, patients with preoperative AI greater than 40° and older than 24 months of age or with bilateral involvement, secondary surgery should be performed earlier due to the reduced probability to achieve normal radiographic parameters.

Tönnis grade III and IV dislocation did not influence the cumulative probability to achieve normal pelvis radiographs although it required more time; in particular, Tönnis grade III or IV hips needed eight months more to achieve normal pelvis radiographs compared with Tönnis grade II hips. Previous studies have reported that severity of dislocation is a risk factor for poor radiographic outcome and secondary pelvic osteotomy [35, 37–39].

In the present study, we found the overall AVN rate was 13%. Importantly, the cumulative probability of achieving normal pelvis radiographs in patients without AVN or type I AVN was significantly higher compared with patients with type II, III, and IV AVN. Our results are in agreement with previous long-term follow-up studies having demonstrated AVN is a risk factor for poor outcome. In hips with AVN, the morphology of the femoral epiphysis and neck and the growth of the proximal femoral physis are disturbed with reduced remodeling capacity [27, 31, 40].

In conclusion, the cumulative probability of achieving normal pelvis radiographs increases linearly during the first five years following CR, then it tends to plateau. Among the hips with normal radiographic findings at last follow-up visit, 93% normalized within the first five years post CR. Age older than 24 months and Tönnis grade III and IV are associated with longer time to achieve normal radiographic parameters.

Age older than 24 months, bilateral dislocation, pre-operative AI greater than 40°, and AVN are risk factors for reduced probability of achieving normal radiographic parameters in children with DDH treated by closed means.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict interests.

Ethical approval informed consent All procedures were performed in studies involving human participants and were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards. This is a retrospective study of patients' data, and an IRB approval was obtained (GZWCMC 2015020904).

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