Original Clinical Article

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Effect of the Sharrard procedure on hip instability in children with Down syndrome: a retrospective study

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Abstract

Purpose The aim of this study was to retrospectively analyze the effect of the Sharrard procedure on hip instability in children with Down syndrome (DS), as measured by the migration index.

Methods In total, 17 children (21 hips) were included from six hospitals in the Netherlands between 2003 and 2019. The primary outcome, hip instability, was assessed with the Reimers' migration index on preoperative and postoperative plain anteroposterior pelvic radiographs. The mean age at surgery was 8.1 years, the majority of children were male (64.7%) and the mean follow-up time was 7.3 years.

Results The mean preoperative migration index was 46% (sd 23.5) and the mean postoperative migration index was 37% (sd 28.4). The mean Delta migration index (the difference in pre-operative migration index and most recent post-operative migration index) showed an improvement of 9.3% (sd 22.7). An improvement in migration index was observed in 52%, no change in 29% and deterioration in 19% of hips. No (re)dislocations occurred in 91% of the hips. No major complications were observed during the follow-up period.

Conclusion Early intervention is warranted in children with DS showing hip instability or hip migration, in order to succeed with less complex procedures. The Sharrard procedure should be considered in children with DS showing hip instability or hip migration, since it aims to rebalance the muscles of the hip joint, is less complex than bony procedures of the femur and acetabulum, surgery time is often shorter, there are fewer major complications and the rehabilitation period is shorter.

Level of Evidence: IV - retrospective case series

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Introduction

Down syndrome (DS), also known as trisomy 21, is the most common symptomatic chromosomal abnormality with survival into adulthood and is associated with several musculoskeletal abnormalities, including hip instability.[1,2] Approximately 7% to 30% of children with DS present with subluxation and dislocation of the hip.[3] Life expectancy of patients with DS has increased from nine years to 55 years and continuing growth is expected, which might result in an increased incidence of painful arthritis, osteoporosis, bone fragility and related problems in the future.[1,4]

Patients with DS show normal hip development *in utero*, but can develop hip instability during childhood, unlike children with developmental dysplasia of the hip.[1,5] Literature suggests that patients with DS show posterior acetabular wall deficiency, increased acetabular retroversion, increased femoral anteversion and a normal neck-shaft angle.[5] Whereas some studies have proposed joint laxity as a possible cause of hip instability,[5,6] others did not find a relationship between joint laxity and hip migration in DS patients.[3,7] Another plausible aetiological factor of hip instability might be muscular inbalance.[3] Children with DS portray delayed motor development, associated with muscle hypotonia, ligament laxity and cerebellar dysfunction.[8] Characteristics of motor dysfunction are longer reaction times, longer movement times and co-contraction of agonist-antagonist muscles.[8,9] It is hypothesized that children with DS use increased co-contraction to increase stiffness and compensate for joint laxity.[10] Gait analysis shows that

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JOURNAL OF CHILDREN'S ORTHOPAEDICS

children with DS portray increased external hip rotation, prolonged hip flexion and increased knee flexion/extension. These alterations in kinematic parameters are also believed to affect stiffness.[8,10] Also, weakness of hip extensors results in a walking pattern consisting of hip flexor muscles and anterior pelvic tilt, thereby creating a point of gravity behind the hip axis to prevent forward falling.[11] This difference in muscle balance is believed to affect the shape and growth of the hip joint, which might cause developmental hip disorders.[12]

Multiple interventions for hip instability in children with DS, including closed reduction and several surgical techniques, have been described over the years, often showing suboptimal outcomes and considerable complication rates.[13-22] In 1959 and 1964, Sharrard [23,24] described a posterior iliopsoas transfer of the tendon insertion for the treatment of paralytic dislocation of the hip in patients with meningomyelocele and cerebral palsy, showing positive results in 17/22 patients. The Sharrard procedure transfers the iliopsoas muscle tendon insertion from the trochanter minor to the trochanter major through a foramen in the ala iliaca (Fig. 1). The aim of this procedure is to restore muscle balance by reducing the flexion position of the hip during walking by strengthening the extension and abduction of the hip joint.[24] Based on the hypothesis that hip instability in children with DS is caused by muscle imbalance, it was believed the Sharrard procedure could prove to be beneficial in treating hip instability in children with DS. In the Netherlands, several children with DS and hip instability have received operative therapy according to the Sharrard procedure.

The aim of this study is to retrospectively analyze the effect of the Sharrard procedure on hip instability in children with DS in the Netherlands. To our knowledge, this is the first study to analyze the results of the Sharrard procedure in children with DS.

Materials and methods

Study design and participants

This is a multicentre, retrospective study analyzing the effect of the Sharrard procedure on hip instability in children with DS. All children with DS ≤ 18 years who underwent a Sharrard procedure between 1st January 2003 and 31st December 2019 in the Netherlands were identified and included. In total, 18 children (23 hips) in six hospitals were identified: Medisch Spectrum Twente (Enschede), Ziekenhuisgroep Twente (Hengelo), University Medical Centre Utrecht (Utrecht), Máxima Medical Centre (Veldhoven), University Medical Centre Groningen (Groningen) and Isala Hospital (Zwolle). Informed consent was obtained from all participants. Demographic characteristics and radiographic data were retrospectively collected from patient medical files.

Treatment

In total, 13/18 children underwent a unilateral Sharrard procedure and 5/18 children underwent a bilateral procedure. All Sharrard procedures were performed by paediatric orthopaedic surgeons and were equivalent to the surgery Sharrard described in 1964,[24] namely a transfer of the iliopsoas muscle tendon insertion from trochanter minor to trochanter major through an artificial foramen in the ala iliaca. Patients were identified for surgery by plain radiographs and physical examination. Inclusion criteria for operative intervention with the Sharrard procedure were a migration index ≥ 30%[2] on plain radiographs before puberty or habitual luxation as assessed during physical examination and determined preoperatively with an image intensifier. Additionally, the multidisciplinary Down-team must expect that the child will be able to continue to walk after puberty. Exclusion criteria were comorbidities prohibiting surgery and the inability of the child to rehabilitate postoperatively. An additional Pemberton

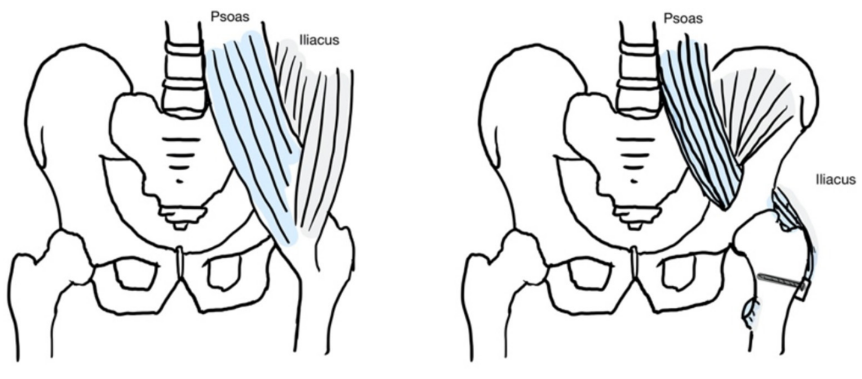

Fig. 1 a) Normal anatomy of the musculus psoas and musculus iliacus, with the tendon insertion at the trochanter minor and; b) transposition of the tendon insertion of the m. psoas and m. iliacus from trochanter minor to trochanter major through a foramen in the ala iliaca.

osteotomy was performed in patients who showed secondary acetabular pathophysiology (acetabular dysplasia), mostly due to delayed referral to our medical centres. Postoperatively all children followed a six-week recovery plan; a spica-cast for the first three weeks and slowly increased walking exercises in the last three weeks. Additionally, patients followed a rehabilitation programme, after which walking/climbing stairs/biking is expected to reach a preoperative level.

Measurements

The primary outcome measurement, hip instability, was assessed using the Reimers' migration index[25] in plain anteroposterior pelvic radiographs. The Reimers migration index divides the part of the femoral head outside Perkin's line by the total width of the femoral head (multiplied by 100%), resulting in a migration percentage. This measurement is often used in literature studying hip dislocation and has been validated in children with cerebral palsy.[26,27] The migration index is the benchmark to identify hip instability in cerebral palsy, is easily applicable, has excellent inter- and intraobserver reliability and is able to assess the effect of therapeutic interventions.[28,29] Additionally, habitual instability was determined preoperatively by the orthopaedic surgeon by physical examination using an image intensifier. Preoperative radiographs and the most recent postoperative radiographs were examined by AZ, FM and HP. Preoperative and most recent postoperative migration indexes were compared (Delta migration index: the difference in pre-operative migration index and most recent post-operative migration index [(= post-operative migration index – pre-operative migration index]) to study the effect of the Sharrard procedure on the migration index and thus hip instability. A positive Delta migration index ≥ 6% (postoperative migration index – preoperative migration index) shows deterioration of the migration index after surgery, while a negative Delta migration index ≤ -6% shows an improvement in the migration index after surgery.

Statistical analysis

Patients were excluded from the analyses in case of one missing radiograph data point. Demographic variables were tabulated. Differences in preoperative and postoperative migration indexes (Delta migration index) were analyzed with a paired *t*-test. The influence of age (five or less years *versus* more than five years, six or less years *versus* more than six years, seven or less years *versus* more than seven years), preoperative migration index (≤ 40% *versus* > 40%, ≤ 30% *versus* > 30%) and having performed a simultaneous Pemberton osteotomy (yes/no) on postoperative migration index were studied with an independent *t*-test. The cut-off point of 40% was chosen, since the Dutch paediatric cerebral palsy guidelines argue for surgical intervention when the migration index is > 40%.[30] A Kaplan Meier curve was created, showing the number of events over time, with events defined as migration indexes > 40%. Also, the differences in survival curves of children with DS aged five or less years *versus* more than five years, six or less years *versus* more than six years and seven or less years *versus* more than seven years were compared and tested for statistical significance with the Log Rank test. The statistical analyses were performed with SPSS version 25 (IBM SPSS Statistics for Windows; Armonk, New York: IBM Corp.). An overview of results of published studies in English on interventions for hip instability in children with DS was created based on a literature study on PubMed (n = 8).

Results

Data of 18 children with DS were collected. One female patient (n = 2 hips) was excluded from the analyses, since the preoperative plain radiograph was not available. Therefore, 17 children (21 hips) were included in the analyses. The majority of children were male (64.7%) and the mean age at surgery was 8.1 years (SD 2.9). The right hip was affected in 76%, the left hip in 0% and both hips in 24%. The mean follow-up time from the preoperative to postoperative radiograph was 7.3 years (median 6.6; 2.6 to 14.8). In 6/21 hips (29%), a simultaneous acetabular osteotomy (Pemberton osteotomy) was performed due to acquired dysplasia of the acetabular wall. All demographic characteristics and migration indexes are shown in Table 1.

The mean preoperative migration index (n = 21 hips) was 46% (SD 23.5; 10% to 100%). The mean postoperative migration index was 37% (SD 28.4; 0% to 100%). The difference in migration index between the postoperative migration index and preoperative migration index (Delta migration index) was -9.3% (SD 22.7; p = 0.076). An improvement in migration index was observed in 11/21 hips (52%), six hips showed no change (29%) and 4/21 hips (19%) showed a deterioration. The mean Delta migration index in the improved group was -25% (SD 19.1; -67 to -7) and in the deteriorated group 19% (SD 4.5; 14 to 25). There were no (re)dislocations during follow-up in 91% of the hips (19/21 hips). Demographic characteristics of the improved and deteriorated group are shown in Table 2.

A significant difference in postoperative migration index was found between children with DS with a preoperative migration index ≤ 40% (mean postoperative migration index = 23%; SD 13.4) compared with those with a preoperative migration index > 40% (mean postoperative migration index = 52%; SD 10.6), using an independent *t*-test (p = 0.027). A significant difference in postoperative migration index was also found between children with DS with a preoperative migration index ≤ 30% (mean post-

Table 1 Demographic variables and migration indexes of 17 children with Down syndrome (n = 21 hips)

Patient	Sex	Age at surgery, yrs	Hip	Follow-up time, yrs	Migration index preoperative, %	Migration index postoperative, %	Delta migration index, %
1	Male	9	R	14.8	*70*	*84*	*14*
2	Male	6	R	8.3	*29*	*20*	*-9*
3	Male	9	R	6.6	*40*	*44*	*4*
4a	Male	4	R	8.2	*10*	*29*	*19*
4b	Male	4	L	8.2	*24*	*10*	*-14*
5	Male	12	R	6.5	*33*	*35*	*2*
6	Male	11	R	8.6	*43*	*31*	*-12*
7	Female	6	R	5.9	*19*	*16*	*-3*
8	Male	6	R	12.2	*34*	*30*	*-4*
9	Male	12	R	3.0	*54*	*0*	*-54*
10	Female	7	R	7.0	*54*	*36*	*-18*
11	Female	10	R	2.6	*41*	*60*	*19*
12	Female	8	R	5.9	*35*	*39*	*4*
13a	Female	7	R	2.6	*100*	*100*	*0*
13b	Female	7	L	2.6	*75*	*100*	*25*
14	Male	7	R	6.6	*45*	*30*	*-15*
15	Male	5	R	3.7	*30*	*0*	*-30*
16a	Male	14	R	8.1	*40*	*14*	*-26*
16b	Male	14	L	8.1	*50*	*43*	*-7*
17a	Female	4	R	13.3	*100*	*33*	*-67*
17b	Female	4	L	13.3	*40*	*17*	*-23*

Follow-up time from preoperative plain radiograph to most recent postoperative plain radiograph
R, right; L, left

Table 2 Demographic variables in the improved and deteriorated Delta migration index groups

	Improved (n = 11)	Deteriorated (n = 4)
Sex male, %	*73*	*50*
Average age at surgery, mean yrs, median yrs, SD	8.0, 7.0, 4.0	7.5, 8.0, 2.6
Right hips (n, %)	8.0 (73)	3.0 (75)
Mean follow-up time,* yrs (SD)	8.0 (3.2)	7.0 (3.1)
Pemberton hips (n, % with Pemberton)	3.0 (27)	1.0 (25)

*Follow-up time from preoperative plain radiograph to most recent postoperative plain radiograph

operative migration index = 15%; SD 10.9) compared with those with a preoperative migration index > 30% (mean postoperative migration index = 44%; SD 28.9), using an independent *t*-test (p = 0.047).

Additionally, a significant difference in postoperative migration index was found between children aged six or less years at time of surgery (mean postoperative migration index = 19%; SD 11.2) compared with children aged more than six years at the time of surgery (mean postoperative migration index = 47%; SD 30.8) (p = 0.009). No differences were found for the age categories five or less years *versus* more than five years and seven or less years *versus* more than seven years at the time of surgery. Also, no significant difference in postoperative migration index was found for having performed a simultaneous Pemberton osteotomy or not. A Kaplan Meier curve, with events defined as migration indexes > 40%, is shown in Figure 2. The Log Rank test showed a significant difference (p = 0.039) in survival curve for children with DS six or less years and children with DS more than six years at time of surgery. No significant differences in survival curves were found for children aged five or less years compared with more than five years and seven or less years compared with more than seven years.

No major complications, such as stress fractures, non-union or infections have been observed in this population during the complete follow-up duration. An overview of published studies on (non-)operative techniques for hip instability and complications in children with DS is summarized in Table 3.

Discussion

Children with DS show a considerable societal burden, consisting of high societal costs for patients with DS[30] and a lower quality of life for children with DS compared with typically developed children.[32,33] Previous studies have demonstrated that untreated hip instability might result in a painful hip, decreased ambulation and wheelchair use.[1] This is the first study to analyze the effect of the Sharrard procedure on hip migration index. Dislocations were defined as a migration score of 100%, according to the Dutch paediatric cerebral palsy guidelines.[30] This study showed no (re)dislocations during follow-up in 91% of the hips (19/21 hips). Postoperative luxation was observed in both hips of one patient, showing immediate postoperative failure. The two hips were not excluded from the analyses in order to provide a complete overview of all operated hips in the Netherlands. No major complications were observed during the total follow-up period. Multiple interventions, operative and nonoperative, for hip instability in children with DS have been described in the literature (Table 3). In all studies study population

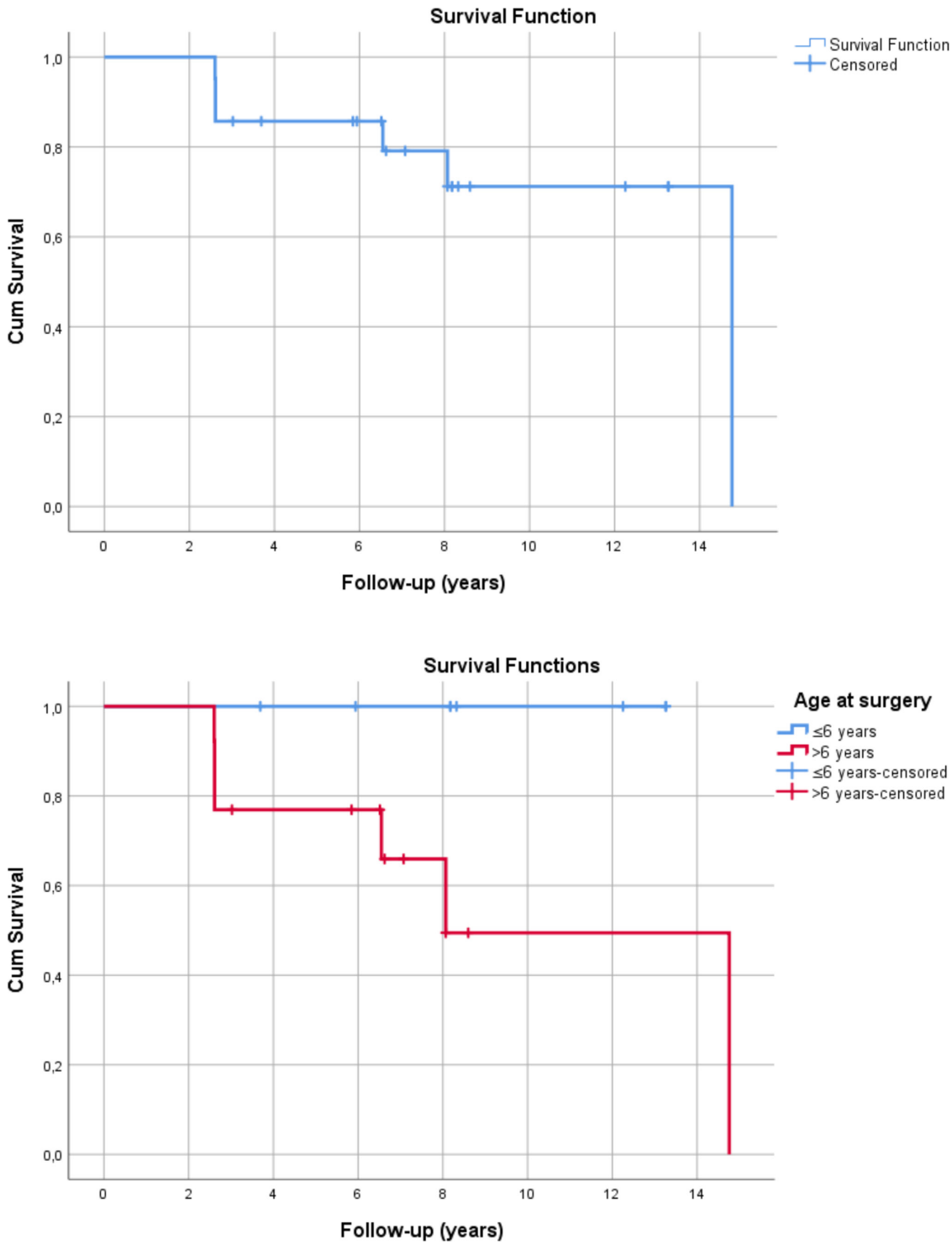

Fig. 2 a) Kaplan Meier curve, with events defined as preoperative migration index > 40%, for the total population and; b) Kaplan Meier curves comparing children with Down syndrome (DS) six or less years and children with DS more than six years. Follow-up defined as time from preoperative plain radiograph to most recent postoperative plain radiograph

sizes are small and range from two to 23 patients. These studies show varying results for comparable operative approaches and it appears that studies that show better results on hip stability are often accompanied by a considerable number of comorbidities. An overview of these studies, including outcomes and comorbidities, are found in Table 3. The Sharrard procedure shows comparable (re) dislocation numbers compared to bony surgical proce-

Table 3 Overview of published studies on multiple (non-)operative techniques and complications of hip instability in children with Down syndrome

Author	Year	n	Intervention	Outcome	Complications
Bennet et al[13]	1982	18 patients, 26 hips	Closed reduction and casting	4/5 persistent hip instability	19% infection rate in the operated group
			Capsular plication	50% hip instability	
			Femoral osteotomy with capsular plication	3/3 hip stabilization	
			Innominate osteotomy with open reduction and capsular plication	3/4 hip stabilization	
			Chiari osteotomy	4/4 subluxations or redislocations	
			Schanz osteotomy	2/2 hip stabilization	
			Femoral osteotomy	5/9 redislocations	
Aprin et al[16]	1985	6 patients, 10 hips	Isolated or combined femoral varus osteotomy, Salter pelvic osteotomy and capsular plication	3/10 persistent hip instability	Osteomyelitis of the ilium, fracture of the proximal femur, leg discrepancy
Greene [14]	1998	2 patients, 3 hips	Closed reduction and casting	2/2 hip stabilization	None
Beguiristain et al[15]	2001	5 patients, 7 hips	Femoral derotational osteotomy (3 patients, 4 hips)	1/4 progressive subluxation	None
Woolf and Gross[21]	2003	2 patients	Modified Pemberton osteotomy with increased posterior coverage	2/2 hip stabilization	None
Knight et al[17]	2011	9 patients, 16 hips	Femoral varus osteotomy	14/16 hip stabilization	12% peri-implant femoral fractures, 88% varus deformity of the femur, 13% superficial wound infections, 6% hip arthritis, 6% persistent waddling gait
Sankar et al[22]	2011	23 patients, 35 hips (18 hips gross instability)	Periacetabular osteotomy	11/12 hip stabilization	Reoperation rate of 12%, postoperative hematoma, femoroacetabular impingement, asymptomatic stress fracture, asymptomatic ischial nonunion
			Femoral osteotomy	3/6 hip stabilization	
Aly and Al-kersh[18]	2018	7 patients, 10 hips	Femoral varus osteotomy and Dega pelvic osteotomy	10/10 hip stabilization	Limb-length discrepancies in unilateral cases
Maranho et al[19]	2018	16 patients, 21 hips	Anteverting triple periacetabular osteotomy	20/21 hip stabilization	5% superficial infection, 24% nonunion of the pubic and ischial osteotomies, 14% stress fractures, 33% stress reaction ischiopubic synchondrosis, 5% deep infection

dures. The Sharrard procedure should be considered in children with DS with instable hips showing hip migration, since the Sharrard procedure aims to rebalance the muscles of the hip joint, is less complex than bony procedures of femur and acetabulum, surgery time is often shorter than bony procedures, there are fewer major complications and the rehabilitation period is shorter. Remarkably, the right hip or bilateral hips were affected in all children, whereas the left hip was never solely affected. This finding, or a possible explanation, has not been previously described in literature.

The Dutch paediatric cerebral palsy guidelines argue for surgical intervention when the migration index is > 40% in children with cerebral palsy.[30] For children with DS and hip instability, the results of this study show that children with a preoperative migration index ≤ 40% show better postoperative results compared with children with a preoperative migration index > 40%. Similar results were found for a preoperative migration index ≤ 30% compared to > 30%. This suggests that the advice of the Dutch paediatric cerebral palsy guideline on the cut-off point of 40% might not be applicable to hip instability in children with DS. Based on the results of this study, in combination with the knowledge that hip migration in unstable hips of children with DS will progress and might lead to permanent secondary acetabular pathophysiology,[2] it could be argued that orthopaedic surgeons should not wait for a migration index > 40% but should intervene sooner in case of hip instability in children with DS, when treatment might be less complex.

Additionally, the Dutch guidelines for DS suggest regular follow-up of hip dislocation via plain pelvic radiographs every two years from the age of four till 14.[34] A significant difference in postoperative migration index was found between age at surgery categories six or less years and more than six years. Survival analysis showed significantly different survival curves for these categories. No (re)dislocations were observed in children six or less years of age at surgery. Also, children aged six or less years appeared to show larger Delta migration indexes and lower preoperative migration indexes. Based on these findings in combination with the knowledge that early intervention is warranted to prevent secondary acetabular pathophysiology, we argue to provide yearly follow-up of the hips in children aged four to six years and two-yearly follow-up in children aged seven to 14 years in order to detect hip instability and migration at an early stage and succeed with less complex procedures.

The Dutch paediatric cerebral palsy guidelines advise surgery to be preferable before the age of five years (when the migration index is > 40%).[30] Our findings appear to corroborate intervention at an early age, since children aged six or less years at surgery show better postoperative outcomes than children aged more than six years at surgery. The survival curves of children aged five or less years compared with more than five showed a similar survival curve compared with the categories six or less years and more than six years. Although this difference (five or less years *versus* more than five years) was not statistically significant, it could be argued that this is due to the low power of this study. However, a precondition for operative intervention is a well-developed walking pattern, which is known to be delayed in children with DS, starting between the age of nine to 15 months in typically developed children and approximately one year later in children with DS.[10] Taking into account our findings and previous literature, we advise early intervention when the migration index is > 30% or habitual dislocation is present and the child has a well-developed walking pattern.

A limitation of this study is the retrospective design, chosen due to the low prevalence of children with DS and hip instability. Therefore, children have distinct follow-up durations and measurements are performed at different points in time. Second, a limited number of patients (21 hips) were included. However, the study population is comprised of all patients that have been operated upon according to the Sharrard procedure in the Netherlands. Due to the small sample size, no additional subgroup analyses could be performed. Lastly, even though a moderate follow-up duration was included, long-term results are warranted.

In conclusion, we believe that early intervention is warranted in children with DS showing hip instability or hip migration, in order to succeed with less complex procedures. The Sharrard procedure should be considered in children with DS showing hip instability and migration, since the Sharrard procedure aims to rebalance the muscles of the hip joint, is less complex than well-known bony procedures of the femur and acetabulum, surgery time is often shorter than bony procedures, there are fewer major complications and the rehabilitation period is shorter. In the future, long-term prospective cohort studies should replicate these findings in a larger population. Also, future studies should evaluate the migration index at multiple measurement points to study trends in migration index.

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COMPLIANCE WITH ETHICAL STANDARDS

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OA LICENCE TEXT

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ETHICAL STATEMENT

Ethical approval: All procedures performed in studies involving human participants 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.

Informed consent: Informed consent was obtained from all individual participants included in the study.

ICMJE CONFLICT OF INTEREST STATEMENT

The authors declare that they have no conflict of interest.

AUTHOR CONTRIBUTIONS

FM: Data analysis, Writing of the manuscript.
LB: Data collection, Writing of the manuscript.
FvD: Data collection, Writing of the manuscript.
HP: Data collection, Data analysis, Writing of the manuscript.
AZ: Data collection, Data analysis, Writing of the manuscript.

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