Distal femoral extension and shortening osteotomy as a part of multilevel surgery in children with cerebral palsy

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Background: **There are several reports describing an increase in anterior pelvic tilt after hamstring lengthening in children with cerebral palsy (CP). Distal femoral extension and shortening osteotomy (DFESO) is an alternative treatment for correction of flexed knee gait, but investigations analyzing outcome and influence on adjacent joint are few in the literature. The purpose of this study was to analyze the influence of DFESO on knee and pelvis in children with CP. Furthermore, it was of interest if an additional patellar tendon advancement (PA) influences outcome.**

Methods: **In this retrospective study, 31 limbs of 22 children (GMFCS I-III; mean age: 12.1±3.1 years), who received DFESO were included and kinematic parameters (knee, pelvis) measured by 3-D-gait analysis were compared before and at least 1 year after surgery (mean follow-up period: 15.6 months).**

Results: **After surgery, during stance phase minimum knee flexion improved significantly by 20.5° (*P*<0.001) and mean anterior pelvic tilt increased by 4.0 degrees (*P*=0.045). In 16 limbs, the postoperative increase in maximum anterior pelvic tilt was more than 5°. Limbs who received an additional PA showed the biggest increase in anterior pelvic tilt.**

Conclusions: **DFESO is an effective method for correction of flexed knee gait in children with CP. Furthermore, the results of this study indicate that DFESO may lead to an increase in anterior pelvic tilt, which may lead to a recurrence of flexed knee gait. In this context, PA seemed to aggravate the effect on the pelvis.**

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Introduction

In children with cerebral palsy (CP), flexed knee gait is a common gait abnormality.[1,2] In the literature, several factors are mentioned contributing to this gait abnormality. In this context, an abnormal shortness of the hamstrings and lever-arm dysfunctions are most important as underlying factors.[1,3] Due to collapse of the moments for hip extension, knee extension and ankle stabilization upright gait is impaired or impossible.[4] Furthermore, an abnormal tone and a shortness of the hamstrings will also increase knee flexion during walking.[1,5] As contractures of the hamstrings and lever arm dysfunctions are less frequent in early childhood, flexed knee gait often develops after the age of ten years.[1,5] Flexed knee gait impairs mobility and independency. Furthermore, it leads to an increased energy consumption during gait. As a consequence of patella alta and increased joint moments, knee flexion gait, degeneration of cartilage and soft tissue occur over time.[5] Therefore, the treatment of flexed knee gait plays a central role in the treatment of gait abnormalities in children and adolescents with cerebral palsy.

Apart from clinical examination, three-dimensional gait analysis is an important tool to identify flexed knee gait and its underlying pathologies. Furthermore, it is useful for planning and monitoring the treatment.[1] Several methods including physiotherapy, orthotic treatment and surgical correction are useful for treatment of flexed knee gait.[4,6] In case of flexed knee gait due to structural deformities, usually a surgical correction is done during a single event multilevel surgery (SEMLS).[5,7]

One common procedure for the correction of flexed knee gait is hamstring lengthening (open or percutaneous techniques).[7] In the literature studies reported encouraging short-term and intermediate outcomes with an improvement of knee function, independency and pain relief after lengthening of the hamstrings in patients with crouch gait.[5,8-10] Nevertheless, after hamstring lengthening, there are also reports describing controversial outcome especially concerning increased anterior pelvic tilt, which is a common finding after hamstring lengthening.[8,11] In a long-term outcome study, Dreher et al[7] showed a deterioration and a persistent increased anterior pelvic tilt eight years after hamstring lengthening. In addition to that, conversion of the hamstrings to monoarticular muscles (maintaining their function to stabilize the pelvis) did not show superior long-term follow-up outcomes compared with conventional hamstring lengthening.[7]

A more recent method for correction of flexed knee gait is distal femoral extension osteotomy (DFEO). This method was shown to lead to a similar short-term outcome compared with hamstring lengthening.[3,12-16] During DFEO, most of the pediatric centers perform a femoral shortening, which might have a relevant impact on anterior pelvic tilt. Femoral shortening leads to an elongation of the hamstrings, which on the other hand results in an increased anterior pelvic tilt. However, studies investigating changes of the anterior pelvic tilt after DFEO are still rare in the literature.[12] The purpose of this study was to investigate the effects of distal femoral extension and shortening osteotomy (DFESO), as a part of SEMLS, on functional knee and pelvic position in children with bilateral spastic cerebral palsy (BSCP) during walking. A further intent was to assess, if the severity of knee flexion gait influences the outcome after DFESO and if the improvement of knee extensions correlates with an increase in anterior pelvic tilt. Last, it was of interest, if additional patella tendon advancement has an influence on knee and pelvis.

Methods

Subjects, inclusion and exclusion criteria

According to specific inclusion criteria [BSCP, age 6-16 years, flexed knee gait, ability to ambulate (GMFCS Level I-III), three-dimensional (3-D) gait analysis within three months before (E0) and at least twelve months after surgery], children who underwent DFESO as a part of multilevel surgery were selected from our motion laboratory data base.

Flexed knee gait was defined as minimum knee flexion of more than one standard deviation of an aged matched control group during stance. According to that, definition limbs showing a minimum knee flexion of more than eight degrees were included.

Patients with previous surgery at lower limbs, previous interventions like botulinum toxin A injection (or serial castings) within the last six month were excluded from this study.

Finally, in this retrospective study, 22 children (14 males, 8 females; 31 limbs), with a mean age of 12.1 years (min/max: 6/16 years; SD: 3.1 years) were included. There were 2 GMFCS I patients (4 limbs), 9 GMFCS II patients (10 limbs) and 11 GMFCS III patients (17 limbs). The study was approved by the local ethical committee.

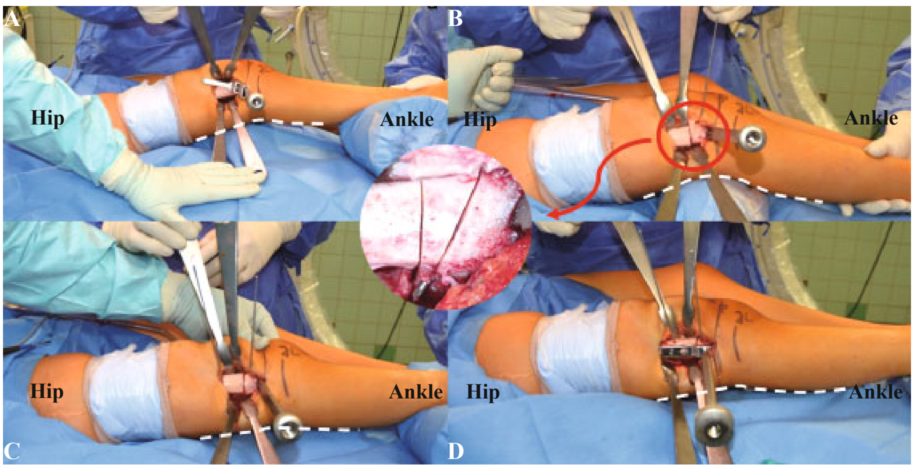

Fig. 1. The femur of a 12-year-old girl with bilateral spastic cerebral palsy and flexed knee gait. A distal femoral extension and shortening osteotomy as well as a derotation osteotomy were performed. A chisel for lateral plate implantation was driven into the femoral bone perpendicular to the long axis of the tibia (**A**). Afterwards two osteotomies were performed: a distal one parallel to the chisel guide and a proximal one perpendicular to the longitudinal axis of the femur (**B**). A trapezoid shaped bony wedge (5 mm anterior based) was removed to avoid stretching neurovascular structures and to relax knee flexor muscles (**C**). After removal of the wedge, the osteotomy was reduced and fixated with an angle-stable locking plate (**D**). The white scattered line illustrates the achievement of total knee extension after removal of the bone wedge. Furthermore, hip and ankle are highlighted for orientation.

Distal femoral extension and shortening osteotomy

The DFESO was performed by a senior surgeon as reported by Novacheck et al[12] and Brunner et al.[17] An anterior based trapezoid shaped bone fragment was removed of the distal femur for extension (Fig. 1).

The amount of extension and shortening of the femur, which was done in every child, was planned according to the knee flexion contracture measured in clinical examination. In 27 limbs 90 degrees blade plates (Implantcast, Buxtehude, Germany), and in four limbs locking plates of DePuySynthes (DePuySynthes, Westchester, USA) were used for fixation after DFESO. The used devices were approved by the Food and Drug Administration. Concomitant surgery included foot stabilization, hamstring lengthening (in case of short hamstrings measured by popliteal angle with the contralateral hip flexed) and patella advancement in cases of patella alta (Table 1).

In addition to that, standard anteroposterior and lateral view radiographs of the knee and the distal femur were done before and after (2 days, 6 weeks, at final follow up examination) surgery. In this study, the radiographs were evaluated concerning positioning of the plate and screws (osteolysis, implant failure) as well as the bony healing (non-union, cortical hypertrophy). After surgery, bilateral shank plaster casts and thermoplastic tigh splints were applied. The casts were connected with a rod to fix the limbs in 10-15 degrees of external rotation. During the first three days after surgery, the knees were fixed in ten degrees of knee flexion (Fig. 2). Epidural anesthesia was used for seven days after surgery. Physiotherapists applied early passive and active mobilization of the knee and hip joints starting at the first day after surgery. During the first two weeks after surgery, knee flexion was limited to 40 degrees. After the second postoperative week the limit of knee flexion was increased to 60 degrees for further two weeks, if an additional patellar

Table 1. The children included in this study and the performed procedures

Age (y)	GMFCS	Body side	Max FU in mon	Pelvic/hip osteotomy	Soft tissue surgery of the hip	Rectus femoris transposition	Hamstring lengthening	DFESO	Distal femur or tibia derotation osteotomy	Patella tendon advancement	Equinus correction*	Hind foot reconstruction†
15	I	Right	12	-	+	+	+	+	-	-	-	-
15	I	Left	12	-	+	-	+	+	-	-	-	-
6	I	Right	12	-	-	-	-	+	+	-	+	-
6	I	Left	12	-	-	-	-	+	+	-	+	-
11	II	Right	23	-	-	+	+	+	+	-	+	+
14	II	Right	14	+	-	-	+	+	-	-	+	+
12	III	Right	16	-	-	-	+	+	-	-	-	+
12	III	Left	16	-	-	-	+	+	-	-	-	+
15	III	Right	16	+	-	+	-	+	+	+	-	+
16	III	Right	15	-	-	-	-	+	+	+	-	+
16	III	Left	15	-	-	-	-	+	-	+	-	+
13	II	Left	13	-	-	-	-	+	+	-	+	-
13	II	Left	17	-	+	+	-	+	+	-	+	-
13	II	Left	14	-	-	+	+	+	+	+	+	+
14	II	Right	23	-	-	+	-	+	+	-	+	+
7	III	Right	15	-	+	+	+	+	-	-	+	+
7	III	Left	15	-	-	+	+	+	-	-	+	+
16	III	Left	20	-	-	+	-	+	+	-	-	+
8	III	Left	16	-	+	-	+	+	-	-	+	-
15	III	Left	20	-	+	-	-	+	+	+	-	-
9	III	Right	15	-	-	+	-	+	+	-	+	-
9	II	Right	23	-	-	+	-	+	+	+	+	+
16	II	Right	12	-	-	-	-	+	-	-	+	-
16	II	Left	12	-	-	-	-	+	-	-	+	-
12	II	Right	13	-	-	-	-	+	+	+	-	+
12	III	Right	23	-	-	-	-	+	+	+	+	-
12	III	Left	23	-	-	-	-	+	+	+	+	-
9	III	Right	12	-	+	-	-	+	+	-	+	+
9	III	Left	12	-	+	-	-	+	+	-	+	+
13	III	Right	12	-	-	-	-	+	+	-	-	-
13	III	Left	12	-	-	-	-	+	-	-	-	-

GMFCS: gross motor function classification system; FU: follow up; DFESO: distal femoral extension and shortening osteotomy. *: equinus correction was done using an aponeurotic lengthening of the calf muscles or an Achilles tendon lengthening; †: hind foot reconstruction included tendon transpositions, osteotomies and/or arthrodeses.

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advancement (PA) was performed during the SELMS. The children began to use full weight bearing four weeks after surgery.

Three-dimensional gait analysis

Gait analysis was performed using a conventional 3-D motion capture (Vicon® camera system, Oxford Metrics, Oxford, United Kingdom) as reported before.[18] Skin mounted markers were applied to bony landmarks of the patients according to a standard protocol and kinematics were calculated according to a standard software procedure (Plugin Gait; Oxford Metrics, Oxford, United Kingdom) based on Kadaba et al.[19] The same physiotherapist and study nurse with special education in pediatric neuro-developmental therapy and gait analysis carried out pre- and post-examinations including documentation of the popliteal angle.[20] Patients were asked to walk barefoot along a 7-m walkway at self-selected speed. At least five representative strides were averaged for further analysis. Eleven patients used four-point crutches or a walker for assistance during 3-D-motion analysis (GMFCS III), therefore in these patients kinetics could not be recorded.

Data analysis

The clinical, kinematic and kinetic data were evaluated before surgery (0 to 3 months before surgery: E0) and at 12 to 24 months after surgery (E1). The kinematics of the pelvis, the knee and ankle were analyzed in the sagittal plane. The Shapiro-Wilk test was applied to test for normal distribution. Afterwards the kinematics before and after the surgery were compared using Student's *t* test and Mann-Whitney *U* Test. Pearson's test was used for correlations. Minimum knee flexion before surgery during stance was correlated with the improvement in knee extension after surgery. In a second step, the improvement in minimum knee flexion measured by motion analysis was correlated with the postoperative change in anterior pelvic tilt. Last, an individual analysis was conducted to assess the influence of an additional patellar tendon advancement on knee extension and pelvic tilt: Two subgroups were formed and the values of each group were compared before and after surgery. The level of significant was set at $P<0.05$. SPSS (IBM SPSS Statistics 19, IBM Ehningen, Germany) was used for statistical analysis.

Results

Outcome

The mean follow up period was 15.6 months (SD: 3.9 months, minimum: 12 months, maximum: 23 months), (Table 1). No patient needed revision surgery and there was no iatrogenic nerve lesion. Radiographic evaluation showed completed consolidation at follow-up in all patients (Fig. 3). Furthermore, in none of the cases an osteolysis or a failure of the implant occurred.

Clinical exam

Knee flexion contracture decreased significantly ($P<0.001$) from 19.0 degrees (SD: 7.2°) before to -2.3 degrees (SD: 5.8°) after surgery. Four limbs showed a knee extension of more than zero degrees after surgery. Furthermore, the popliteal angle decreased significantly after surgery ($P<0.001$; preoperative: 61.6° (SD: 16.7°), postoperative: 42.3° (SD: 17.9°).

Knee kinematics

During stance mean knee flexion was 46.2° before and 26.6° after surgery. This decrease of 19.6 degrees was statistical significant. There was also a significant improvement in minimum knee flexion of 20.5° ($P<0.001$) and in knee range of motion of 8.1° (min-max; $P=0.007$) during stance phase (Table 2), (Fig 4).

Ankle kinematics

There was a significant decrease in mean ($P=0.017$) and maximum ($P=0.013$) ankle plantarflexion during stance phase after surgery. Furthermore, the point of maximum ankle plantarflexion during stance change from 56.2% of gait cycle to 29.1% of gait cycle significantly ($P=0.002$) (Table 2).

Pelvic tilt

There was a slight increase of 4.0° in mean anterior pelvic tilt, which was statistically significant ($P=0.045$) after surgery (Fig. 4). In contrast, the maximum anterior pelvic tilt did not change significantly ($P=0.072$). Nevertheless, after surgery 20 limbs showed an increase in mean anterior pelvic tilt and 19 limbs showed an increase in maximum anterior pelvic tilt of more than one standard deviation (1.5°). Sixteen limbs

Table 2. The kinematics of the pelvis, knee and ankle in degrees

Variables		E0	E1	*P* value
Pelvic Tilt (0-100% GC)	Mean	13.4 (1.5)	17.4 (1.6)	0.045
	Max	17.4 (1.5)	21.5 (1.5)	0.072
	Max@%GC	52.1 (57.9)	51.7 (56.4)	0.866
	Range	8.3 (1.5)	8.5 (1.6)	0.735
Knee FlexEx	Mean (ST)	46.2 (2.4)	26.6 (2.6)	<0.001
	Min (0-100% GC)	40.0 (2.4)	19.2 (2.4)	<0.001
	Range (0-100% GC)	24.6 (2.5)	32.7 (2.8)	0.007
DorsiPlanFlex	Mean (ST)	0.9 (2.5)	9.6 (2.0)	0.017
	Min (ST)	-17.2 (3.4)	-3.8 (2.4)	0.013
	Min@ST	56.2 (20.5)	29.1 (29.6)	0.002

The standard deviations are illustrated in brackets. The values of E0 and E1 were compared using Student's *t* test and Mann-Whitney *U* Test.

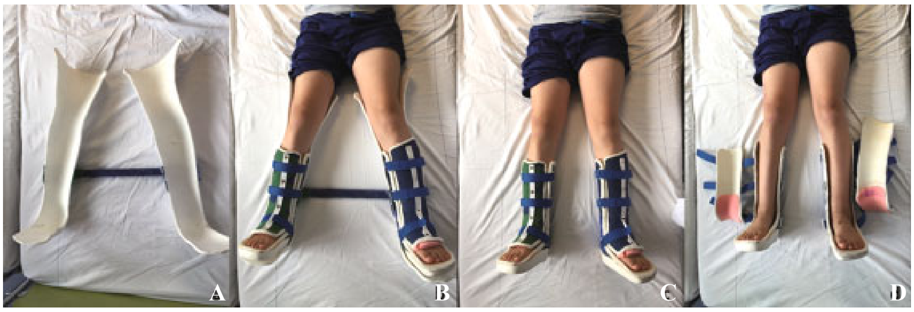

Fig. 2. The cast construct of a patient four weeks after distal femoral extension and shortening osteotomy. Shank plaster casts connected by a wooden rod (**A, B**) were used to restrict movement after surgery. Nevertheless, the casts of the thigh could be removed for mobilization (**C, D**).

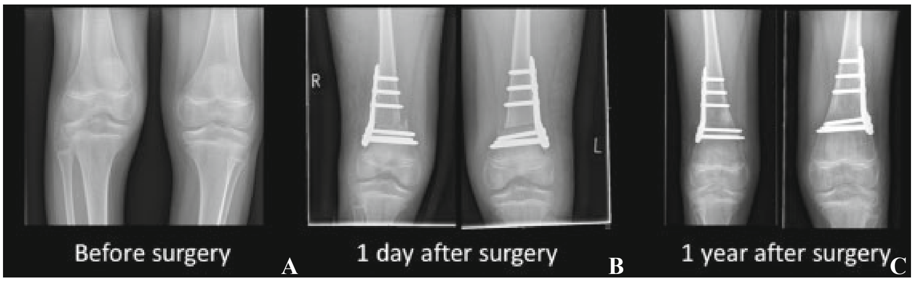

Fig. 3. Anteroposterior X-rays of the knee of a 9-year boy preoperative (**A**), one day (**B**) and one year after distal femoral extension and shortening osteotomy (**C**).

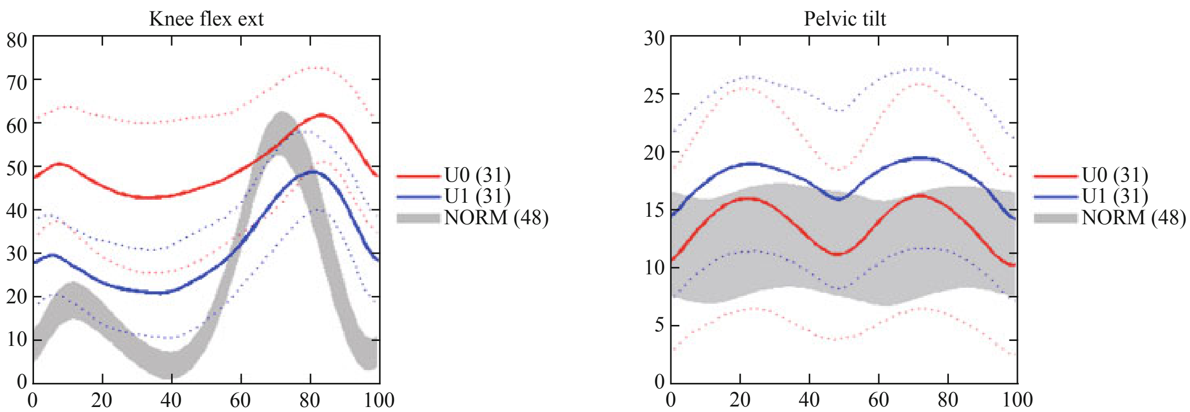

Fig. 4. Sagittal plane kinematic mass graphs of the knee (left) and the pelvis (right) before (U0, red line) and one year (U1, blue line) after distal femoral extension and shortening osteotomy. The kinematics of an age matched control group are also illustrated (grey). The *x*-axis marks the gait cycle in percent. The *y*-axis marks the range of motion in degrees.

even showed an increase of more than five degrees in maximum anterior pelvic tilt. A connection between lengthening of the hamstrings and increase in mean or maximum anterior pelvic tilt could not been found. In addition there was also no significant change in anterior to posterior pelvic range of motion (min-max; P=0.735) (Table 2).

The minimal clinical importance difference (MCID)[21] and an individual analyses was calculated to evaluate if the change in mean anterior pelvic tilt of 4.0° was clinical important. Out of the 31 limbs 20 limbs showed an increase in mean anterior pelvic tilt of more than 1.5 standard deviations. Seven limbs showed a decrease in mean anterior pelvic tilt of more than 1.5 standard deviations after surgery. The MCID was 3.1 degrees.

Correlations

There was a significant correlation between the minimum knee flexion during stance phase (3-D gait analysis) before surgery and its improvement after surgery (Pearson correlation coefficient: 0.75, P<0.001).

The correlation of improvement in minimum knee flexion during stance phase (3-D gait analysis) and increase in anterior pelvic tilt did not show any significant correlation (Pearson correlation coefficient: 0.27, $P>0.05$).

Patellar tendon advancement

Before surgery, there were no significant differences in mean anterior pelvic tilt and mean/min knee flexion (3-D gait analysis) between limbs who received PA (9 limbs) and those who did not receive PA (22 limbs). Both groups showed a significant increase in minimum knee flexion ($P<0.001$) and mean anterior pelvic tilt ($P<0.05$) after surgery. The increase in maximum anterior pelvic tilt was higher in children who received PA and this was statistically significant (PA: increase of 5.6°; no PA: increase of 3.5°; $P=0.027$).

Discussion

Several authors described an increase in anterior pelvic tilt and deterioration of flexed knee gait after hamstring lengthening after mid- and long term follow-up.[7,8,11] Another method for correction of flexed knee gait is DFESO, which is recently becoming more popular for the treatment of flexed knee gait in cerebral palsy.[3,12-15] This study intended to investigate the influence of DFESO on pelvis and knee in children with bilateral cerebral palsy and flexed knee gait. The results of this study showed a significant ($P<0.001$) decrease in minimum knee flexion by 19.6 degrees during stance phase at a mean follow up period of 15.6 months after DFESO.

There were no complications, which led to revision surgery. Especially there was no iatrogenic nerve palsy, which probably was caused by the protective effect of femoral shortening during the DFEO.[15]

Overall, the findings of this study were in accordance with reports of other authors, who presented similar short term results after DFESO[3,12-16] and hamstring lengthening.[7,9] Nevertheless, in contrast to Novacheck et al,[12] who showed an amplifying effect of PA on the reduction in knee flexion gait, in this study there was no significant relationship between reduction in mean knee flexion and PA. In this study nine limbs showed a patella alta and received PA in addition to the DFESO. Despite there was no statistic significant difference, those limbs showed slightly increased values for knee extension during stance compared with the other limbs, who did not receive PA. The missing significance might be attributed to the limited number of subjects in this investigation.

The results of this study corroborate the findings of previous studies,[3,12,13] which demonstrated that DFESO is a powerful method for the correction of flexed knee gait in cerebral palsy. Furthermore, it is a very effective and safe method to correct knee flexion contracture. Children with a severe knee flexion contracture tended to profit most from the procedure concerning the reduction of flexed knee gait. Compared with a previous work,[7] the effect of DFESO on the knee seems to be comparable with the effect of hamstring lengthening.

An increase in anterior pelvic tilt is a well-known side effect after hamstring lengthening.[7,11] Therefore, many surgeons sought for alternative treatment methods. DFEO was found to be a potential alternative to hamstring lengthening in cases of flexed knee gait and reports presenting short term outcome were presented in the literature.[3,12,13,16] As there are only few reports in the literature, the knowledge about the influence of DFEO or DFESO on pelvic tilt is still poor. Therefore, the intent of this study was to evaluate the influence of DFESO and PA on pelvic tilt. In this study mean anterior pelvic tilt increased significantly by 4.0 degrees ($P=0.045$) after DFESO. As the change in mean anterior pelvic tilt of 4.0° was significant, but seemed to be few, the MCID was calculated. This analysis showed a MCID of 3.1°, which illustrated that the change in mean anterior pelvic tilt seemed to be clinical relevant after DFESO. Furthermore, 20 out of the 31 limbs showed an increase of more than 1.5 SD in mean anterior pelvic tilt.

The DFESO was performed during a single-event multilevel surgery. Therefore, other performed procedures might have influenced the effect of the DFESO. In ten limbs, also a moderate lengthening of the hamstrings was conducted. Nevertheless, the findings of this study are comparable to other reports, which presented changes in pelvic tilt after DFEO.[3] As other authors reported a similar increase after DFESO[3,12,14] and after hamstring lengthening,[7,9] the effect of hamstring lengthening and DFESO on anterior pelvic tilt seems to be comparable. Stout et al[3] demonstrated the greatest increase in anterior pelvic tilt in patients, who were treated with a DFESO and patella advancement. In ours study, nine limbs received a concomitant patellar tendon shortening due to patella alta. Corroborating the findings of Stout et al,[3] limbs who were treated with concomitant PA did show a significant ($P=0.027$) increase in maximum anterior pelvic tilt compared with limbs who did not receive PA in our study. Novacheck et al[12] postulated that a hip flexion contracture and rectus femoris spasticity or contracture also may lead to an increased anterior pelvic tilt after DFESO and therefore, in those cases an additional rectus femoris transposition should be performed. In this study, in eleven limbs a transposition of the rectus femoris muscle was accomplished (Table 1). However, this did not prevent for an increase in anterior pelvic tilt in those patients. Therefore, in case of spasticity or contracture of the rectus femoris, a transposition of the muscle might decrease the amount of increase in pelvic tilt, but it seemed that it was not able to prevent an increase in anterior pelvic tilt in general after DFESO.

In case of hamstring lengthening, an increased anterior pelvic tilt is suspected to lead to recurrence of flexed knee gait in long term follow up.[7] Recently, there are no studies investigating medium- or long-term outcome after DFESO. This is a subject remained to be investigated.

This study has several limitations. In eight limbs soft tissue surgery of the hip (lengthening of the psoas and/or adductor longus) was performed. A lengthening of the psoas muscle might have an impact on the anterior pelvic tilt. In ten limbs an additional lengthening of the hamstrings, which might had a relevant impact on minimum knee flexion and pelvic tilt, was performed. The study included few subgroup analyses, whose power was low due to the small sample size. Further studies with an enhanced sample size are needed to address this issue and allow different subgroup analyses.

In conclusions, this study showed encouraging results after DFESO in children with bilateral spastic CP and flexed knee gait. DFESO is an effective and safe procedure and leads to comparable functional outcome compared to hamstring lengthening. However, it seems that increase in anterior pelvic tilt is also a problem.

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Contributors: Klotz M contributed to the study design, data analysis, interpretation and drafting manuscript; Hirsch K contributed to acquisition of data and data analysis; Heitzmann D contributed to acquisition of data and data analysis; Hagmann S contributed to data analysis and revising manuscript; Maier M contributed to data analysis and revising manuscript; Dreher T contributed to the study design, interpretation, drafting & revising manuscript.

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