Table 1. Quality methodology.

| Author (year) ^{ref} | Åkerstedt (2009) 14 | Khan (2007) ¹⁵ | Patikas (2006) 16 | Patikas (2006) 17 | Seniorou (2007) ¹⁸ | Thomason (2011) ¹⁹ |
|------------------------------|--|---------------------------|---------------------|-------------------|-------------------------------|-------------------------------|
| Design | gn Prospectief Cohort Retrospectief Cohort | | RCT | RCT | RCT | RCT |
| internal validity | | | | | | |
| Randomly divided | dna | dna | yes | yes | yes | yes |
| blind randomized | dna | dna | yes | yes | no | yes |
| defined study | yes | no | yes | yes | yes | yes |
| Groups of identical baseline | dna | dna | yes(SC) | yes (SC) | yes (SC) | yes (SC) |
| blind subjects | dna | dna | no | no | no | no |
| therapists blind | dna | dna | no | no | no | no |
| testers blind | ? | no | no | no | no | no |
| Confouders / bias | ? | yes | ? | ? | ? | ? |
| Use valid, reproducible | yes | yes | yes | yes | yes | yes |
| measuring instruments | | | | | | |
| defining outcome | yes | yes | Doubtful | Doubtful | Doubtful | Doubtful |
| Intention to treat | dna | dna | yes | no | no | yes |
| external validity | • | | | • | | |
| Inclusion/exclusion | yes | yes | yes | yes | yes | yes |
| numbers (n) | 11 | 85 | 39 | 39 | 20 | 19 |
| Discription physicaltherapy | no | no | no | no | no | no |
| intervention | | | | | | |
| Check co- intervention | no | yes | no | no | no | yes |
| Follow- up duration enough | yes | yes | yes | yes | yes | yes |
| Loss to follow- up< no0% | yes | yes | yes | yes | yes | yes |
| results | | | | | | |
| Mean | yes | no | yes | yes | yes | yes |
| Standaard deviation | no | no | yes | yes | yes | yes |
| 95 % confidence interval | no | no | yes | yes | no | yes |
| statistical analysis | - Descriptive statistics | no | - Within and | - ANCOVA | - within subject | - ANCOVA |
| | - Visual Analysis | | between factor | - ANOVA | standard deviation | - lineair regresion |
| | | | - ANCOVA | | - CV % | |
| | | | - Unpaired t- tests | | - paired sample t-tests | |
| | | | - Chi- square | | - ANCOVA | |
| | | | nonpara. stat. | | | |

ANCOVA: analysis of covariance, ANOVA: analysis of variance, CV: coefficients of variation, SC: statistical correction, dna: does not apply, ?: uncertain.

Table2. Data-extraction; study, population, surgery procedures and follow-up.

| ot described. |
|---------------|
|) |

| STUDY | POPULATION | | | SEMLS ASSESMENT | | | |
|--|---------------------|--|--|-----------------|--------------------|--|---|
| Author (year) ^{ref} | N (male/ female) | N control/ intervention (male/ female) | Mean Age in years (y) (SD) [Range] | CP type | GMFCS | Surgery procedures | Follow up/ Period after surgery |
| Åkerstedt (2009) ¹⁴ | 11 (10/1) | | 13.8 [9-18] | bi / uni | I, II, III | CE, DFO, GE, HE RecFT | T0= before SUR T1= 1 year T2= 2 year |
| Khan (2007) ¹⁵ | 85 (53/32) | | 8.5 [5-12] | bi | IV, V ^a | STP= 79% BP= 21% | T0= before SUR T1= mean 3.5 year (range 2-5) |
| Patikas extension torq (2006) ¹⁶ | 39 (27/12) | CG=20 (14/6) EG=19 (13/6) | 9.7 (2.8) | bi | 1,11,111 | CG= 144 STP 56 BP EG= 127 STP 65 BP | T0= before SUR T1= 6 months T2= 1 year |
| Patikas (2006) ¹⁷ walking | 39 (27/12) | CG=20 (14/6) EG=19 (13/6) | 9.7 (2.8) | bi | I, II, III | CG= 144 STP 56 BP EG= 127 STP 65 BP | T0= before SUR T1= 6 months T2= 1 year T3= 2 year (Only gait analysis) |
| Seniorou (2007) ¹⁸ | 20 (10/10) | AE= 9 PRT= 11 | 13.0 (2.0) [7-16] | bi | I, II, III | AdE, CE, CLO, DFO, DTO, GE, HE, PsE, RecFT, SPLAT TPE | T0= before SUR T1= 6 month T2= 7.5 month T3= 1 year |
| Thomason (2011) ¹⁹ | 19 (12/7): | SEMLS= 11 PRT=8 | 9.8 [6-12] | bi | II, II | TT, RO, SH, MtE, SF | T0= before SUR/PRT T1= 1 year T2= 2 year |

AdE: adductor muscles extension; AE: active exercises; bi: bilateral; BP: bony procedure; CE: calcaneus extension; CG: control group; CLO: calcaneal lengthening osteotomy; CP: cerebral palsy; DFO: derotating femoral osteotomy; DTO: derotating tibia osteotomy; EG: exercise group; GMFCS: Gross Motor Function Classification System; GE: gastrocnemius extension; HE: hamstrings extension; MtE: muscle tendon extension; N: number; PRT: progressive resistance training; PsE: psoas extension; RecFT: rectus femoris transfer; RO: rotatie osteotomy; SD: standard deviation; SEMLS: single event multilevel surgery; SF: stabilization foot according to directive; SH: stabilization hip according to directive; SPLAT: split tibilalis anterior transfer; STP: soft tissue procedure;; SUR: surgery; TPE: tibialis posterior extension; TT: tendon transfer; Uni: unilateral.

Appendix 1 Searchstring PubMed

#1 Disease

"Cerebral Palsy" [Mesh] OR "Cerebral Palsy" [tw]

#2 Population

"Child" [Mesh] OR "Child, Preschool" [Mesh] OR "Adolescent" [Mesh] OR "Pediatrics" [Mesh] OR child [tw] OR schoolchild [tw] OR "child, preschool" [tw] OR adolescent [tw] OR pediatrics [tw] OR paediatrics [tw] OR boy [tw] OR boys [tw] OR boyhood [tw] OR girl [tw] OR girls [tw] OR girlhood [tw] OR youth [tw] OR youths [tw] OR teen [tw] OR teens [tw] OR teenager [tw] OR children [tw]

3 Physical therapy

"Physical Therapy Modalities" [Mesh] OR "Physical Therapy (Specialty)" [Mesh] OR "Physical Therapy" [tw] OR physiotherapy [tw] OR Rehabilitation [MeSH] OR Rehabilitation [Subheading] OR Rehabilitation [tw] OR "Exercise" [Mesh] OR "Muscle Stretching Exercises" [Mesh] OR "Motion Therapy, Continuous Passive" [Mesh] OR "Exercise Therapy" [Mesh] OR "Resistance Training" [Mesh] OR "Exercise" [tw] OR "Muscle Stretching Exercises" [tw] OR "Motion Therapy, Continuous Passive" [tw] OR "Exercise Therapy" [tw] OR "Resistance Training" [tw] OR "motor control" [tw] OR "motor learning" [tw]

#4 Surgery

"General Surgery" [Mesh] OR "Orthopedics" [Mesh] OR "Surgery" [tiab]

#5 Combined searchstring

#1 AND #2 AND #3 AND #4

Appendix 2

2

1 *Protocol design of PTT program in children with CP after SEMLS.*

It is important to realize that a standard SEMLS patient does not exist. Depending on the different
SEMLS surgery procedures and protocols it is not possible to describe one type of fixed
treatment protocol or one type of patient. Therefore, it is important to consider the following
factors:

Surgery: a different progression during PTT will be seen after SEMLS depending on
 whether one or both legs are operated and whether two or three levels of surgery (ankle, knee,
 and hip) was needed.

GMFCS level: children with CP GMFCS I and II show more selectivity and are able to
 perform the exercises more easily compared to children with GMFCS III.

Other factors: Cognitive level, the ability to use two hands and the support of the child's
 system (parents, teachers etc.) have a crucial influence on the performance and progress of the
 treatment.

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16 During the period of immobilization, exercises and specific instructions are given according to the protocols of the orthopedic surgeon. In order to make adequate use of the new alignment and 17 18 gait opportunities (possibilities), co-interventions such as orthosis, plaster and devices are needed in the post-surgery intervention plan.¹⁻³ Adequate planning of the co-interventions is essential in 19 20 order to start PPT. This care preferably will be organized in a specialized multidisciplinary team that will be coordinated by the rehabilitation physician. Four to six weeks after surgery, X-rays 21 will be taken and the orthopedic surgeon will decide if mobilization can be started with full body 22 weight, using temporarily ankle and knee immobilizers to guarantee the safety of the child during 23 24 standing. When 100% weight bearing is not allowed the child need to stand with support of a stander and walk with crutches if possible combined with the non-weight-bearing exercises and 25 26 instructions.

The care for these patients needs to be individually tailored and patients and their parents shouldbe accompanied by the expert team both pre- and postoperatively.

We present a framework of important elements of the PTT after SEMLS at the point that the child has permission from the orthopedic surgeon to bear full weight with ankle and knee immobilizers, typically 4-6 weeks after surgery:

32 1. *Goal*:

From completely inactive (bedridden) to fully active on all levels (depending on the rehabilitation goals) of ICF-CY in which pain and fatigue are crucial factors to be

considered during treatment, because the child did not bear weight on their legs for 6 1 weeks. With guidance from a physical therapist, the child learns to regain strength within 2 the new ROM after SEMLS and the child needs to learn a new pattern for standing and 3 walking activities in daily live. 4

- 2. Frequency and duration 4-6 weeks post-operatively till 24 months
 - 4 to 6 weeks post-surgery, depending on the consolidation of the bones, the intense 4 weeks of daily PTT starts till 8 to 10 weeks for 1.5-2 hours, combined with 1.5 to 2.5 hours independent performance of instructed exercises by child and parents.
 - From week 8 to 10 weeks till 6 months weeks; 3-5 times a week 1 hour a day PTT and • 1-2 hours a home program
 - 6-12 months; 2-4 times a week 1 hour a day PTT and 0.5-1 hours a home program
 - 12-24 months; 1-2 times a week $\frac{1}{2}$ -1 hour a day PTT •
- See table 1.
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Table 1. SEMLS treatment schedule

| Surgery | Post- | Time line | | Frequency | Duration | Duration | Duration |
|---------|----------|-----------|--------|-----------|----------|-------------|-------------|
| | surgery | | | Per week | PTT | independent | independent |
| | recovery | | | | | exercises | Program at |
| | time | Start | End | | | Institute | home |
| SEMLS | | 4-6 | 8-10 | | | | |
| | 4-6 | Weeks | weeks | Daily | 1.5-2 h | 1.5-2.5 h | |
| | weeks | 8-10 | 6 | | | | |
| | | weeks | months | 3-5 | 1 h | | 1-2 h |
| | | 6 | 12 | | | | |
| | | Weeks | months | 2-4 | 1 h | | 0.5-1 h |
| | | 12 | 24 | | | | |
| | | months | month | 1-2 | 0.5-1 | | |

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Note: More or less PTT is possible depending on the policy of the orthopedic surgeon

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3. Intensity and method of treatment

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4 to 6 weeks post-surgery: It is essential for the start of the PTT to manufacture optimal ankle and knee immobilizers. From day one the child will be placed in a standing 22 23 position with the immobilizers to support standing and to guarantee the safety of the child. 24 Within the treatment during the transition from standing to walking, it is important to align the orthoses with shoes. 25

26 From day one the child starts with strength training 3-4 times a week from unloaded to functional loaded exercises according to the method of progressive resistance exercise training using the 27

repetition maximum method.⁴ This means 1-3 sets of each exercise and within each set, muscle 28

fatigue is reached between 6-12 repetitions. Criteria for quality of moving are leading during
muscle strength exercises to increase the load. The following muscle groups are trained
specifically: hip extensors, hip abductors, knee extensors, abdominal muscles and when possible
plantar flexors depending on the use of ankle foot orthoses (AFO's).

First, the aspect of the quality of movement while standing and walking is essential. The 5 focus on the gait pattern is heel strike, extension of the knee and hip during midstance with a 6 7 minimal pelvic drop and keep extend the knee and hip throughout the standing phase in order to facilitate knee flexion during swing phase. The child needs to adapt to the weight on their heels 8 during standing and walking, which is a new condition, as before SEMLS, the child did not bear 9 weight on their heels. Use of manual and verbal feedback ⁵, walking aids and technologies, such 10 as body weight support treadmill training and body weight support over ground training (Zero-11 G), are beneficial in learning a new gait pattern. $^{6;7}$ 12

The child starts always in the walkway and weight supported treadmill training will be used along with crutches, tripods or a backward rollator. If possible, Zero-G training will be used. The bodyweight supported treadmill training will start daily when knee flexion reaches 80 degrees. The speed starts from 0.1 to 0.5 km/h, with 30-50% bodyweight support and 2-3 x 2 minutes walking. After 4 weeks the speeds vary from 0.5 to 1.5km/h, with 10-30% bodyweight support and 3 x 3-6 minutes walking. The amount of time the patient will use a walking device will depend on individually recover time.

When the SEMLS is performed on one leg and the child has an optimal hand function, crutches are used. When the SEMLS is performed on two legs and the child has an optimal hand function, tripods are used. When indicators are described of a restricted cognitive level are present, a limited use of two hands and the support of the child's system is confined, a backward rollator will be considered. The child leaves the rehabilitation center with the aid of a walking device. The decision of which walking aid to use will depends on the progress of the child.

It is important to keep the active and passive ROM obtained by SEMLS. During the immobilization period, knee immobilizers are worn and the knee flexion is limited. ROM exercises and variation of posture will be provided including; the hip flexors, hip adductors, knee flexors, knee-extensors and the m. gastrocnemius. Variation of posture will be offered during the day by having the child lying in prone, sitting with straight legs, standing in a stander, biking on a Motomed or a special hometrainer to optimize the active and passive ROM and will be used daily1-2 times for 10-30 minutes.

Balance training will be started in standing position with knee and ankle immobilizers.Exercises start with weight shifting from one leg to the other, balance while walking with

(minimal) walking aids and to training for falls. For children that have SEMLS performed on one leg, it is possible to stand without the knee immobilizers at the end of the first week or beginning of the second week of the PTT. For children that have SEMLS performed on two legs, it is possible to stand without the knee immobilizers in the end of the second week or beginning of the third week of the PTT. Practice based observation indicates that this has been the case for the vast majority of children.

From the beginning of the rehabilitation, a daily, individually structured home program will be made of the four elements described above, including variation of posture and exercises for: strength, gait, the active and passive ROM and balance. However, recovery time for the child is also an essential part. Activity of daily living (transfers in- and out of bed, dressing, toilet etcetera) are part of daily training, preferably during daily care using devices when necessary guided by an occupational therapist. A multidisciplinary approach is necessary to maintain the methods and the quality of exercise training to ensure the quality of the treatment.

After 4 weeks of training in a rehabilitation center, the treatment is transferred to a private practice where the patient was previously being treated, coached by the physical therapist of the rehab center. Focal points of treatment will be gait training, strength training, balance training, maintaining the active and passive ROM related to meaningful and functional activities. Outlining of the orthosis remains a point of attention.

19 2-6 months: In this phase it is likely possible to work on the specific activities that 20 are part of the child's request, because the cardiovascular and strength condition on body and functioning level is still insufficient. Functional (supported) gait training to learn a new walking 21 22 pattern and preservation of the new active ROM is continued. Functional muscle strength training is intensified in load. Orthoses are used and the use of a walking aid is reduced depending on the 23 24 abilities of the child. Based on our clinical experience we advise to use an assistive device until 25 the patient is strong enough to overcome trunk sway (leaning to one side when lifting the 26 opposite leg) or a minimal pelvic drop. Balance and gait training on the GRAIL (Gait Real-time 27 Analysis Interactive Lab) is optional.

• 6 to 12 months: The patient's needs are now prioritized, taking into account the post-surgery recovery. In the first half year, the focus was therefore more on function and activity level (ICF-CY) and from this period on the emphasis is more on participation level. The walking is optimized and functional muscle strength training is intensified in load with more complex exercises and combined with aerobic/anaerobic endurance training. The support during gait training is further minimalized during daily activity, depending on the child's progress with the training. In this phase the initial request and goals of the child are more within reach and a high frequency and intensity of the PTT is required due to the need to improve muscle strength, as the condition of the body is still recovering from SEMLS. Balance and gait training on the GRAIL is optional.

12-24 months: The emphasis is on fine-tuning of daily life activities and sports
activities, which includes functional strength training combined with the emphasis on
aerobic/anaerobic endurance training. The frequency of physical therapy is dependent on the goal
of the patient in this phase. Children with GMFCS I and II walk without walking devices. Most
children will go through the pubertal growth spurt with marked changes in height, weight and
sometimes in body mass index ² and therefore monitoring is important.

Evaluations by the rehabilitation center take place 25 weeks, 1 year, 2 and 5 years postoperatively as seen in the literature. ^{8;9} Twelve weeks post-operatively, the child is seen by the orthopedic surgeon for monitoring the progression of the consolidation of the child's bones.

15 Suggestions for measuring instruments for evaluation

Pre- and post-measurements are depending on the goals of the child and the parents, for the
 SEMLS and the PTT. We advise the following measurements on the different ICF-CY levels:¹⁰⁻²¹

Function level: 18 • 19 o physical examination of lower extremity (mobility, selectivity, strength, spasticity) 3 D Gait analysis (step-length, walking speed, kinematics lower limbs) 20 0 10 meter timed walking test 21 0 1 minute walking test 22 0 Activity and participation level: 23 • Canadian Occupational Performance Measure 24 0 The Goal Attainment Scaling 25 0 Gross Motor Function Measure (Domain D and E) 26 0 Functional Mobility Scale 27 0 28 0 Mobility questionnaire Quality of life Questionnaire to be determined 29 0 30 31

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|------------------------|------|--|--|--|--|--|
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