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Evolution of gait in adolescents and young adults with spastic diplegia after selective dorsal rhizotomy in childhood: A 10 year follow-up study

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ABSTRACT

Background: Short-term benefit on gait of selective dorsal rhizotomy (SDR) surgery, which relieves spasticity of the lower extremities has been demonstrated in children with cerebral palsy (CP). However very little is known of the evolution of gait when patients become adolescents and young adults.

Research question: How does the gait pattern evolve in adolescents and young adults who underwent SDR during childhood?

Methods: A longitudinal study was performed including 19 ambulant patients with spastic diplegia due to CP or other causes (mean age at SDR: 6.6 ± 1.6 years) who were assessed four times: pre-SDR, 2 years post- SDR, 5 years post-SDR and at least 10 years post-SDR. From 2D video recordings, Edinburgh Visual Gait Score and lower limb joint kinematic parameters were calculated.

Results: Our data show that the improvement in the gait pattern obtained short-term after SDR continues during into adolescence and adulthood. Ten years after SDR all patients improved compared to baseline. Considering the lower limb joint kinematics, most notable improvements were found at knee and ankle joints. Compared to the evaluation before SDR, the range of motion of the knee increased: the knee was more extended at initial contact and knee flexion in midswing improved. Excessive ankle plantar flexion was reduced during the entire gait cycle. Only minor changes were found at hip and pelvis. Eight patients underwent additional orthopaedic surgery in the years after SDR, and the present findings should be considered as a combination of SDR, development and additional treatment.

Significance: We demonstrate lasting improvement of gait quality in ambulant patients with spastic diplegia who underwent SDR during childhood when they become adolescents and young adults.

1. Introduction

Selective dorsal rhizotomy (SDR) is a neurosurgical procedure aimed at eliminating spasticity in the lower limbs and thereby improving walking ability of patients with cerebral palsy (CP). Short-term benefits of SDR on the gait pattern have been demonstrated [1–5], with selection of suitable candidates for SDR and the definition of the individual goals being crucial to evaluate and interpret the outcomes [3,5].

Long-term follow-up studies have mainly focused on the effect of SDR on functional ability measured by the Gross Motor Function Measure (GMFM-66) [6–8] or questionnaires [9,10] and the results are controversial. Lundkvist Josenby et al. [6] found progressive improvement in gross motor function over 10 years after SDR, and Bolster et al. [8] reported that gross motor function in some children improved more than expected, noting that most of the patients underwent additional orthopaedic surgery. However, Tedroff et al. [11] concluded that despite the fact that the spasticity-reducing effect of SDR was maintained, this did not seem to improve long-term functioning or prevent contractures in patients with CP.

The number of studies that have evaluated the long-term (more than 10 years) effect of SDR on gait using kinematic parameters is limited. It has previously been reported that knee and hip joint motion during gait improved in patients affected with spastic CP 10 years after SDR [12] and 20 years after SDR [13]. Langerak et al. [14] compared gait kinematics of 31 adults who had undergone SDR during their childhood to age-matched healthy adults using 3D gait analysis (3DGA). The most relevant difference with the control group was that patients walked

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with a mild crouch gait pattern. Because of the cross-sectional nature of this study, they could not describe how gait developed over time. McFall et al. [15] analysed the changes in gait pattern in a group of 17 CP patients (Gross Motor Function Classification System [16] -GMFCS-level II—IV) who underwent SDR in their childhood and were assessed during their pre-adolescence (10–12 years of age) and post-adolescence (16–18 years). Exact term of follow up was not specified. Compared to pre-SDR, 3DGA revealed that patients walked faster and with longer steps, with better knee flexion in swing and more knee extension at initial contact, increased dorsiflexion at ankle and, as adverse effect, a small increase in anterior pelvic tilt. The improvements found in pre-adolescence didn't change in post-adolescence and the authors concluded that the initial improvement in gait pattern was followed by a functional plateau, which was maintained over time. Recently, Munger et al. [17] compared changes in gait kinematics of 13 spastic diplegic CP patients who underwent SDR and orthopaedic surgery to the changes in gait pattern of a matched group of 8 patients who underwent only orthopaedic surgery. The authors found that at follow-up gait significantly improved in both groups, but patients who did not undergo SDR showed more improvement. Thus, there are conflicting reports in the literature on the long-term effects of SDR on gait. Longitudinal studies with repeated assessments after SDR over time are limited and therefore it is not clear if the changes after SDR are maintained over time. In McFall et al.'s and Munger et al.'s study [15,17] only children with GMFCS level II, III and IV were included, while it has been previously reported that children with GMFCS I and II benefit more from SDR than children classified as level III and IV [6,7,18].

Therefore, the aim of the present study was to investigate the evolution of gait pattern over at least 10 years in ambulant adolescents and young adults with spastic diplegia due to CP or other causes, who underwent SDR during their childhood. Since 3DGA was not available at our institute when the first patients underwent SDR, the Edinburgh Visual Gait Score (EVGS) [19] was used to quantify the patient's overall gait quality from 2D video, in combination with a set of clinically relevant lower limb kinematic parameters.

2. Methods

From the database of our institute, children were included retrospectively if they satisfied the following inclusion criteria: affected with spastic diplegia due to CP or other diagnoses, having independent walking ability pre-SDR, having undergone SDR between 1998 and 2007, older than 4 years of age at SDR and at least four assessments including gait analysis, pre-SDR, 2 years post-SDR, 5 years post-SDR and at least 10 years after SDR. The study protocol was approved by the local medical ethics committee. All patients were operated by the same neurosurgeon (WvPO) at the VU University Medical Center (VUmc) Amsterdam, the Netherlands.

The children were indicated for SDR by a multidisciplinary team with a pediatric physiatrist and child neurologist, and complied with the following criteria: spastic diplegia, GMFCS Level I, II, or III; 2.5 years of age or older; spasticity (defined as velocity-dependent resistance to passive stretch) in at least six muscle groups in both legs; no serious deformities or contractures ($< 20^\circ$ flexion contracture in knee, popliteal angle $< 80^\circ$, at least 0° dorsal flexion in the ankle); at least able to crawl, sit independently > 10 s, maintain tall knee position, and squat at least seven times; good motivation and support and from parents/caregivers for the surgery and rehabilitation phase (see also van Schie et al. [4].), and no basal ganglia abnormalities on MRI (to exclude risk of developing dystonia).

The neurosurgeon performed the operation using the procedure described by Steinbok et al. [20]. During the operation, dorsal roots L2-S1 were exposed and separated into different rootlets after laminotomy of L2-L5 and opening of the dura. Transection of the rootlets was performed after electrostimulation, based on palpable muscle contraction and electromyographic response. No more than 50% of the rootlets were transected at each level. After the operation a period of 5 days of bed rest was prescribed, and patients were usually discharged 7 days after the operation. Children were all prescribed ankle foot orthoses to wear from the start of walking until skeletal maturation to prevent knee flexion contracture. Children received intensive physical therapy for 3 months, for 1 h a day, five times a week, followed by therapy three times a week until 6 months after surgery. After SDR all children were followed up yearly at the department of rehabilitation medicine of VUmc, and orthopaedic surgery or other treatments were performed if required.

In our study we included assessments at four time points: pre-SDR, 2 years post-SDR, 5 years post-SDR and ≥ 10 year post-SDR. Patients were classified using GMFCS levels. For non-CP patients the level of functioning was estimated based on their mobility. During each assessment patients were asked to walk at their comfortable walking speed on a 10 m walkway barefoot and with their own assistive devices, if needed. Videos on sagittal (left and right body side) and coronal (frontal and dorsal) planes were recorded. From sagittal and coronal video recordings, EVGS [19] was calculated. EVGS has been developed to measure the quality of gait in patients with CP based on 2D video recordings. EVGS reliability, repeatability, validity and sensitivity to change have been demonstrated previously [19,21,22]. The same operator (MR) calculated the EVGS for all the patients for all the assessments. She was blind to the identity of patients and before the analysis the video recordings were pooled together for all patients at the different time points.

Lower limb joint angles in the sagittal plane for pelvis, hip, knee and ankle at initial contact (IC), mid-stance (MST), opposite initial contact (OIC) and mid-swing (MSW) were quantified with a digital goniometer of custom made, open-source software (MoXie Viewer) previously validated [23]. For the dorsiflexion kinematics, the goniometer was aligned to the rear foot and especially in case of a severe deformity or midfootbreak, the forefoot was ignored. This was done to avoid overestimation of ankle dorsiflexion. Additionally, tibia inclination at opposite toe off (OTO), incomplete foot contact at MST, and range of motion of hip and knee joints during the gait cycle were calculated. For each subject, the mean EVGS value of the left and right leg was calculated. For analysis of the lower limb joint angles, the two patient's legs were considered separately.

EVGS was presented as mean and standard deviation for the whole group at each assessment and for subjects stratified by GMFCS level. Moreover, for each subject the difference between EVGS pre-SDR and 10y post-SDR was calculated. A change of 2.4 points was considered as minimal clinically important difference (MCID), where a decrease is indicative of improvement [24]. Statistical analysis for repeated measures (General Linear Model-GLM) and Bonferroni post-hoc analysis was performed, with time (pre-SDR, 2y, 5y and 10y post-SDR) as within-subjects effect, GMFCS as group effect, and time and GMFCS as interaction effect, using SPSS version 22. Kinematic parameters of lower limb joint were presented as mean and standard deviation for the whole group, and compared between pre-SDR and 2, 5 and 10y post-SDR using a repeated measures GLM and Bonferroni post-hoc analysis with time as within-subject effect. Additionally, in order to evaluate any differences among the three groups before surgery, pre-SDR data were compared between different GMFCS levels using ANOVA. Statistical significance was set at $p < 0.05$ for all tests.

3. Results

19 patients with spastic diplegia (5 females, 14 males) were included in this study (Table 1).

The mean children's age at SDR was 6.6 ± 1.6 years (range: 4–12 years). Eight patients were classified as GMFCS I, five as GMFCS II and six as GMFCS III. At the last assessment, mean patients' age was 19.2 ± 2.5 years (range between the pre-SDR assessment and the last