



HAL
open science

Feasibility of a shorter Goal Attainment Scaling method for a pediatric spasticity clinic - The 3-milestones GAS

A. Krasny-Pacini, F. Pauly, J. Hiebel, S. Godon, M. Isner-Horobeti, M. Chevignard

► To cite this version:

A. Krasny-Pacini, F. Pauly, J. Hiebel, S. Godon, M. Isner-Horobeti, et al.. Feasibility of a shorter Goal Attainment Scaling method for a pediatric spasticity clinic - The 3-milestones GAS. *Annals of Physical and Rehabilitation Medicine*, 2017, 10.1016/j.rehab.2017.01.005 . hal-01503046

HAL Id: hal-01503046

<https://hal.sorbonne-universite.fr/hal-01503046>

Submitted on 6 Apr 2017

HAL is a multi-disciplinary open access archive for the deposit and dissemination of scientific research documents, whether they are published or not. The documents may come from teaching and research institutions in France or abroad, or from public or private research centers.

L'archive ouverte pluridisciplinaire **HAL**, est destinée au dépôt et à la diffusion de documents scientifiques de niveau recherche, publiés ou non, émanant des établissements d'enseignement et de recherche français ou étrangers, des laboratoires publics ou privés.

FEASIBILITY OF A SHORTER GOAL ATTAINMENT SCALING METHOD FOR
A PEDIATRIC SPASTICITY CLINIC —

THE 3-MILESTONES GAS

Authors

Agata Krasny-Pacini^{1,2,4}

Fernand Pauly^{3,4}

Jean Hiebel^{1,4}

Sandrine Godon⁴

Marie-Eve Isner-Horobeti^{1,5}

Mathilde Chevignard^{2,6}

1. Institut Universitaire de Réadaptation Clemenceau-Strasbourg, 45 bd Clemenceau, Strasbourg. France
2. Sorbonne Universités, UPMC Univ Paris 06, CNRS, INSERM, Laboratoire d'Imagerie Biomédicale (LIB), Paris, France
3. Service de MPR et pédiatrie, CH du Luxembourg, Luxembourg,
4. Service de Chirurgie Orthopédique Infantile, hôpital de Hautepierre, CHU de Strasbourg, avenue Molière, 67098 Strasbourg cedex
5. Strasbourg University, Fédération de Médecine Translationnelle de Strasbourg (FMTS), EA 3072 "Mitochondrie, stress oxydant et protection musculaire", France.
6. Hôpitaux de Saint Maurice, Service de Rééducation des Pathologies Neurologiques Acquises de l'Enfant, F-94410, Saint Maurice, France

Abstract

Background. Goal Attainment Scaling (GAS) is a method for writing personalized evaluation scales to quantify progress toward defined goals. It is useful in rehabilitation but is hampered by the experience required to adequately “predict” the possible outcomes relating to a

particular goal before treatment and the time needed to describe all 5 levels of the scale. Here we aimed to investigate the feasibility of using GAS in a clinical setting of a pediatric spasticity clinic with a shorter method, the “3-milestones” GAS (goal setting with 3 levels and goal rating with the classical 5 levels). Secondary aims were to 1) analyze the types of goals children’s therapists set for botulinum toxin treatment and 2) compare the score distribution (and therefore the ability to predict outcome) by goal type.

Methods. Therapists were trained in GAS writing and prepared GAS scales in the regional spasticity management clinic they attended with their patients and families. The study included all GAS scales written during a 2-year period. GAS score distribution across the 5 GAS levels was examined to assess whether the therapist could reliably predict outcome and whether the 3-milestones GAS yielded similar distributions as the original GAS method.

Results. In total, 541 GAS scales were written and showed the expected score distribution. Most scales (55%) referred to movement quality goals and fewer (29%) to family goals and activity domains.

Conclusion. The 3-milestones GAS method was feasible within the time constraints of the spasticity clinic and could be used by local therapists in cooperation with the hospital team.

Introduction

Goal Attainment Scaling (GAS)¹ is a method for writing personalized evaluation scales to quantify progress toward defined goals (see practical guidelines²⁻⁴ and literature reviews⁵⁻⁷). GAS produces an individualized, criterion-referenced measure of a person’s goal achievement. GAS scales are written by “predicting” the possible outcomes relating to a particular goal and precisely defining goal attainment levels before treatment. One GAS scale is written for each identified rehabilitation goal, with an emphasis on the family/child’s participation in goal selection when possible. Success of the intervention is then quantified on a 5-point ordinal scale, typically ranging from -2 to +2, the middle point “0” corresponding to the most probable expected outcome after treatment/rehabilitation. Scores can be aggregated to quantify the extent to which a group of children who are receiving the same type of intervention achieve their personalized rehabilitation goals⁸⁻¹⁰, although such aggregation of scores is not necessarily needed and has been criticized¹¹.

GAS has been used in various pediatric rehabilitation research fields ¹²⁻²⁰, including treatment with botulinum toxin (BoNT) ^{8-10,21-24}. The clinimetric properties are idiosyncratic, ²⁵ but rehabilitation professionals can write reliable ²⁶ and valid ^{27,28} scales that are more responsive to change than are usual standardized measures ²⁹ (see review ⁶ for pediatric rehabilitation). However, few studies have investigated the use of GAS in clinical settings.

In addition, the feasibility of the method in clinical practice is questionable for 3 main reasons. First, GAS is time-consuming: writing one GAS scale requires about 12 to 49 min ³¹. In addition, how many levels of attainment should be described is unclear, but the number may affect the time needed to write the GAS scale. Kiresuk and Sherman ¹ suggested that at least 2 levels should be described and the others “deduced”. Many teams follow this suggestion in clinical practice. For example, Turner-Stokes et al. ⁴ recommends precisely describing the patient’s initial level (-2) and the expected outcome (0) ⁴; goal attainment is then subjectively scored by judging whether a goal was attained “as expected” (0), “better” than expected (+1), or “much better” than expected (+2) etc. The Turner-Stokes method is probably quick, but 1) it uses subjective adjectives for “how much” a goal has been attained, which are not reliable, observable, and pre-determined measures to characterize each level; 2) it does not require reflecting on different possible goal outcome levels before treatment; 3) it does not require predicting what exactly would be a “much better” than expected (+2) level; and 4) to our knowledge, its inter-rater reliability has not been tested. These methodological and practical issues have been recently detailed ³. At the other extreme, Steenbeek et al. ³¹ and many authors precisely describe each of the 5 levels of goal attainment. This method takes more time (45-49 min per scale) but has good and well-documented inter-rater reliability ^{23,26}.

A second feasibility issue is that training in GAS is recommended, ^{15,31} but few professionals have access to training, although successful training programs for pediatric teams have been published ³¹.

Finally, ^{5,32} a major drawback is that the method highly depends on the ability of the GAS-setting team or staff member to generate valid, reliable and meaningful scales. Indeed, GAS has been considered more a measure of how adequately a therapist can foresee an outcome than an outcome measure itself ^{5,33,34,35,36}. Whether teams other than highly specialized research teams or therapists can foresee an outcome precisely enough to write adequate scales is unknown. Furthermore, in pediatrics, a child’s prognosis with specific goals is often not known, even for knowledgeable therapists. This situation is a major limitation in the use of

GAS, especially in research: GAS scales will determine the efficacy or inefficacy of a treatment, but in fact the conclusions may relate more to inadequate appreciation of possible progress at the level of the individual child.

Because describing all 5 GAS levels is time-consuming and describing only 2 levels seems to lead to more subjective scaling, we aimed to find a quicker (than Steenbeek's) but more precise (than Turner-Stokes') way of writing GAS, reflecting on different levels of goal attainment before treatment. The shortened GAS method had to be feasible in a routine spasticity-management clinic for children. The secondary aims were to 1) analyze the types of goals set with the shortened GAS method for BoNT A (BoNT-A) treatment for spasticity and 2) compare the score distribution (and therefore the ability to predict outcome) by goal type.

Methods

Preliminary intervention: training therapists in GAS

In our region, children are referred to the university hospital for BoNT-A injections, but most pre-BoNT-A assessments and all rehabilitation sessions following injections are performed by the child's usual therapists, either in private practice or in institutions or the special school the child attends. The child's rehabilitation therapists who have the best perspective of the child's possibilities and factors impeding progress because they see the child in daily life, communicate with families regularly and follow the child's progress over prolonged periods. Therefore, our intervention aimed at teaching GAS to children's usual therapists, who are independent of the hospital team for the specialized spasticity-management clinic, so that they can write GAS scales for their clients but in cooperation with the spasticity team.

The first step was to introduce GAS in one-evening information and teaching group seminars, presenting 1) the general GAS method, relying heavily on the Steenbeek et al.³¹ examples from an interdisciplinary team training, 2) discussing published GAS examples^{23,26,27,31,37}, 3) presenting BoNT-A action and BoNT-A-related treatment goals and 4) presenting the shortened GAS method.

Following this introductory theoretical training, rehabilitation therapists were encouraged to write GAS scales according to the 3-milestones method (see below), for their BoNT-A-related goals, including goals meaningful for families ("family" including the child). Therapists accompanied the child to the pre-injection clinic, to discuss treatment goals and to

adjust, check, correct or change the GAS scales they had written according to specific, measurable, attainable, relevant and time-specific (SMART) GAS writing rules^{2,37,38}: each GAS level had to be specific, measurable, attainable, and realistic, and a pre-determined delay for scoring had to be agreed upon (time-specific). If parents had not participated in the therapists' GAS writing process, further GAS scales were written, as appropriate, focusing on family goals during the pre-injection clinic.

After 6 months of such practical use of GAS by children's therapists in the spasticity clinic, all children referred for BoNT-A injection during their regular clinical care were included in a study that tested the 3-milestones GAS, following the same procedure as the training phase. All therapists' GAS scales were discussed with the spasticity-management clinic team and changed or developed further if needed, and further GAS scales could be written for additional goals identified by the team and/or the family.

Study design

The study was approved by the hospital ethics committee (CPP IDF VI). All children who received BoNT-A injections in the upper and/or lower limbs between May 2009 and June 2011 in our pediatric clinic, regardless of etiology of brain lesion or degenerative disease etc., could potentially be included. Our clinic tends to follow up children until early adulthood, and therefore young adults cared for by the pediatric team were also included. If a child needed BoNT-A injections several times during the study period, each treatment was considered separately, which resulted in more treatment sessions than children.

The following data were collected before treatment: etiology of brain lesion (cerebral palsy versus other diagnosis not meeting the Bax et al. criteria³⁹ for cerebral palsy [CP]); type of CP (spastic, dystonic, ataxic, choreo-athetotic or mixed [see⁴⁰⁻⁴² for descriptions]), and Gross Motor Function Classification System (GMFCS)⁴³ functional level (used for only children with CP).

Treatment goals were defined by clinical examination, visual (and sometimes video) movement analysis, parents' requests (e.g., nursing difficulty due to spastic adductors), the health condition's known prognostic factors (e.g., risk of hip migration, progressive worsening of ulnar deviation), or functional goals of the child's physiotherapist and occupational therapist (e.g., shoulder abduction limitation hampering crawling).

Shortened GAS writing method: the 3-milestones GAS

For each goal, a GAS scale was written, precisely describing 3 levels: -2 (initial level of the child in relation to the goal), 0 ("expected" level, i.e., the level the child would most likely attain at 8 weeks post-treatment), and +2 (best-expected but realistic outcome). These 3 levels represented the 3 GAS "milestones". Intermediate points -1 and +1 were not described but were intermediate (levels -2, 0 and 2 were milestones that indicated the transition to the next level). Examples are provided in Table 1. Contrary to Turner-Stokes' guide⁴, the initial level was *always* -2 even if a child's level could worsen after treatment, because assigning an initial level to -1 does not allow for detecting progress when the goal has not been attained as expected. In clinical practice, we therefore adopted the Steenbeek^{23,26,29} methodology that used "3" to score worsening. GAS scales were adjusted, corrected or changed after discussion between the family, the child's therapist(s) and the spasticity-management clinic team, according to SMART GAS writing rules.

BoNT-A treatment is a standard treatment for spasticity in children with CP. Combined with other treatments (occupational therapy, casts, splints, walking aids etc.), it can reduce impairment and improve activity^{22,44,45}. Evidence of improvement in participation and quality of life is still uncertain. Assuming that BoNT-A treatment associated with rehabilitation and other treatments is effective, we expected that if the teams had understood how to write GAS scales and how to express the levels -2, 0 and +2 so that these levels were milestones, we would see mostly 0 scores (about 43% of all scores), followed by -1 and +1 scores (about 21% each) and a few extreme -2 and + 2 scores (about 7% each), as in the theoretical GAS score distribution proposed in the original method⁴⁶. Hence, adequate score distribution across the 5 GAS levels was used as a feasibility criterion to show that a shortened GAS writing method yields the same pattern of distribution as classical GAS writing methods.

GAS level -2	GAS level 0	GAS level +2
Cannot ride a horse because of hip adduction and hamstrings spasticity that does not allow sufficient leg opening (too painful even with stirrup adaptations).	Can ride a horse but the stirrups has to be placed higher to allow the important knee flexion accompanying the hip abduction.	Can ride a horse with stirrups placed in a normal position and sits comfortably on the horse's back.
Cannot be left alone on the toilet: requires an adult to maintain sufficient hip abduction, so the urine does not wet the clothes.	Can be left alone on the toilet, but requires an adult to maintain sufficient hip abduction so the child can wipe herself	Can manage the toilets alone (even if she needs help for the transfer). Has sufficient hip abduction to avoid wetting the clothes and to wipe herself.
Starts to eat a bowl of mashed potatoes unaided but cannot finish it	Eats a bowl of mashed potatoes in 11 to 15 minutes, aided by an anti-ulnar deviation splint	Using splint, eats at normal speed (similar to the family's other children)
Cannot eat alone because the right upper limb activity during the meal is impeded by dystonic left upper limb movements into the right functional space.	The dystonic left upper limb continues to enter the right functional space, however the child manages to eat alone, in the absence of stimulation (talking, TV, noise...) increasing dystonic movements.	The dystonic left upper limb does no longer enter the right functional space and the child can eat alone, in the presence of other siblings and regular environmental stimulations.

Table 1 : Examples of 3 milestones GAS's

For each BoNT-A treatment, at least one GAS scale was written, but the maximum number of scales was not limited, which led to as many scales as goals chosen for treatment. For each child, at least one "family GAS" goal was recommended. When the parents did not have a specific goal in mind, they were encouraged to identify problematic situations and to participate in the goal-setting procedure in a guided way. The spasticity-management team and the child's usual therapists helped the families to choose which goal-attainment criteria could be observably scored (e.g., if the parents' goal was "that my child walks better," goal attainment levels could be defined by using the physical aids needed to walk, the distance the child could walk, the number of falls, endurance, or the type of surface the child could walk on [slope, uneven land, etc.]). Families chose the family GAS levels of goal attainment, guided by the spasticity-management team. Families were asked for a precise description of the current problem (e.g., if the problem was difficulty dressing their child, they were asked about the time needed, the child's pain, their evaluation of overall difficulty rated from 0 and

10, their own possible pain in the arms to maintain sufficient abduction or knee flexion, the type of clothes they chose to make the dressing easier, etc.) until a clear way of scoring future change could be chosen (e.g., improvement in terms of time, difficulty rated from 0 to 10, etc.). The -2 level was described by using terms that were chosen to score future change. To define the 0 GAS level, families were asked what they would consider a meaningful improvement for the problem. Finally, to determine the +2 GAS level, they were asked what would be the best-expected outcome for this goal. Because family wishes were sometimes unrealistic, the best-expected outcome was discussed with them until agreement was reached. A similar method was used if the GAS goals proposed by therapists were not clearly formulated.

Goals were classified into 4 categories, predetermined before the start of the study: 1) “family GAS” goals, relating to goals explicitly formulated with/by families (e.g., horseback-riding, facilitating diaper change); 2) “functional GAS” goals, relating to activity and participation domains of the International Classification of Functioning (ICF)⁴⁷¹ (e.g., walking on uneven ground in the neighboring park, opening a jam jar); 3) “quality of active movement GAS” goals (referring to goals such as gait analysis parameters, active range of motion of the upper limb) relating to body structure and function domains of the ICF; and 4) “joint mobility GAS” goals (including passive range of motion and tone), also relating to body structure and function domains of the ICF. This last category included goals for which there was no immediate functional criterion and no active movement progress (e.g., reducing adductor muscle tone to decrease the risk of hip dislocation in the longer term but not leading to any functional change at the 8-week post BoNT-A injection clinic).

Six to 10 weeks after the injection (ideally 8 weeks), children were re-examined by the spasticity-management team and GAS goals were scored, taking into account 1) the clinical examination with visual (and sometimes video) movement analysis but no use of standardized scales or gait analysis, 2) the child’s rehabilitation therapists’ reports and/or discussion with them if they were present, and 3) perspectives of the child and parents. Family GAS goals were always rated directly by the parents (and the child if possible). Quality of active movement GAS and joint mobility GAS goals were scored by using examination and child performance during the clinic (often aided by video analysis of movement). For goals other than “family GAS” goals and goals that could not be directly tested during the clinic (especially some functional GAS goals), the child’s therapists were asked to rate the GAS

goal they had proposed and families were also asked for their GAS rating if the goal could be observed at home. In case of disagreement between the therapist and family, the child's abilities and goal were discussed in terms of context influencing outcome. If an agreement could not be reached, the GAS scale was duplicated and 2 different scores were given: one for the school/therapy context and one for the home context.

GAS were scored on the 5 levels of GAS. Each GAS scale resulted in a raw score with the possible values -2, -1, 0, +1 and +2. Because GAS scales are ordinal data, we adopted Tennant's view⁴⁸ that an arithmetic operation such as T-score calculation and use of parametric statistics is inappropriate for GAS data analysis^{3,48-50} (for variants and debates on GAS methodology, especially the use of T-scores versus raw scores, the weighting of goals according to their importance or difficulty, for example, see Krasny-Pacini et al.^{3,11}). Furthermore, a growing number of studies of children with CP have avoided the use of T-scores (e.g.,^{16,19,20,23,51,52}).

For each child, we obtained as many raw scores as GAS scales, and no global calculation was derived to sum all the raw scores, similar to the method used by Steenbeek²³. Each level was considered as a category — -2, -1, 0, 1 and 2 — and not as a continuous variable. The distribution of raw scores by the 5 levels was analyzed visually and by chi-square test. We considered an adequate score distribution across the 5 GAS levels⁴⁶ as a feasibility criterion to show that a shortened GAS writing method yields the same pattern of distribution as classical GAS writing methods and is a possible alternative to describing all 5 GAS levels in clinical practice. However, because the theoretical GAS score distribution published by Kiresuk did not include a -3 level, we analyzed -3 scores (3 GAS scales scored -3 in our sample) together with -2 scores. Distributions were also analyzed for all the GAS scales but also according to goal type.

Results

In a 2-year period, 158 BoNT-A treatment sessions were performed (Fig. 1). GAS data were not available for 19 sessions because the family did not come to the post-injection clinic, the GAS scoring sheet was lost, or the GAS scale had not been written or scored. Therefore 139 treatment sessions were analyzed. Because each child had several goals (and related GAS scales) per treatment session, the 139 treatment sessions yielded a total of 541 GAS scales (see Fig. 1). These 541 GAS scales related to 93 different children because some children had

undergone more than 1 treatment session during the inclusion period (see Table 2). We could not score 4 of the 541 scales because they referred to goals that were not scorable (e.g., hamstring injection for parental goal of dressing their daughter in fashionable tight jeans because summer came and the girl wore only skirts; adductor injection for a child's goal to more comfortably ride her horse, but horseback riding lessons stopped). Therefore, 537 GAS scales were scored and analyzed for the study. Seven children had a single scale, 14 had 2 scales, 32 had 3 scales, 47 had 4 scales, 22 had 5 scales, 11 had 6 scales and 6 children had 7 scales.

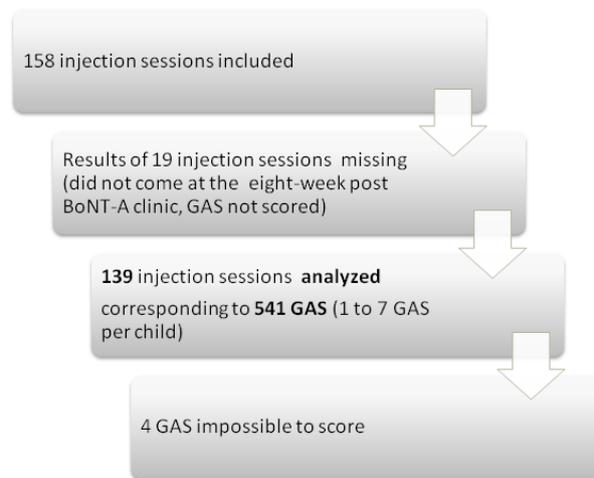


Figure 1: Flow chart of inclusions

Gender	
Boys	50%
Girls	50%
Etiology	
Cerebral Palsy	80%
Other known diagnosis	9%
Unknown diagnosis	11%
Gross Motor Function Classification System (GMFCS) *	
I	17%
II	17%
III	29%
IV	21%
V	16%
Clinical form	
Pure Spastic	69%
Spastic-Dyskinetic	25%
Spastic-Ataxic	4%
Other	2%
Number of treatment sessions for the same child during the study period	
1	63
2	17
3	9
4	4

Table 2: Characteristics of included children

* Only children diagnosed with CP (80%) were classified according to the GMFCS

Patients' mean age was 7.6 ± 4.5 years (range 1-24). Patient characteristics are described in Table 2. Most injections were performed for children with CP as defined by the Executive Committee for Definition of Cerebral Palsy³⁹. The children who did not meet the definition of CP (20%), presented spasticity or dystonia of various causes, mostly degenerative diseases, epileptic encephalopathy or brain lesions after 18 months of age (for 2 children). Half of these

children without CP had unknown etiology of the spasticity. Most children had spasticity alone, but one quarter also showed ataxia, dyskinesia (dystonia or choreo-athetosis) (Table 2).

For all children, at least one GAS scale could be written during the usual spasticity clinic (45 min per child). We could not determine the exact time needed to write each GAS scale per child because the GAS writing process occurred during the clinic and was not circumscribed (i.e., while discussing a BoNT-A goal, discussion could drift to rehabilitation, school issues, need for a sitting device, other medications or exploration of disability etiology, etc.). Because the 45-min clinic dealt with a large variety of issues, including determining GAS goals but not exclusively, the overall time needed was less than in previous studies of the classical GAS method^{30,31}.

Most goals (55%, 297 GAS scales) referred to movement quality (e.g., equinofoot, wrist hyperflexion during grasping) and the 3 other categories (functional, family, joint mobility) were equally represented (14-16%) (Fig. 2): 80 GAS scales related to family goals, 77 to functional goals and 83 to body structure domains.

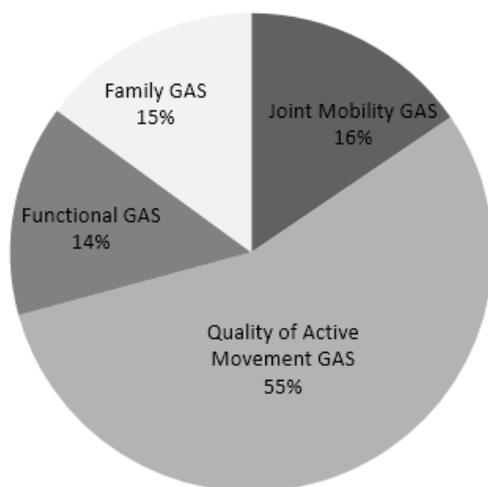


Figure 2: Distribution of GAS goals according to the four predetermined GAS categories

Post-treatment GAS scores were distributed approximately as expected (Fig. 3), with most being 0 scores (expected level of attainment), with a decrease toward extreme scores (more -1 and +1 than -2 and +2 scores). The median GAS score was 0. However, the distribution differed significantly from the theoretical distribution ($p=0.002$), and post-treatment results were significantly better than expected, with more raw scores > 0 (46%) than < 0 (18%)

($p=0.001$). Overall, family goals and functional goals were achieved (83% of family goals and 79% of functional goals scoring 0, +1 or +2). The functional GAS score distribution did not differ from the expected GAS distribution ($p=0.17$) (Fig. 4), but the distribution of family GAS scores differed from the expected pattern of distribution ($p=0.001$) (Fig. 5): mostly 0 scores (goal attained as expected), but extreme scores -2 and +2 were more frequent than intermediate results (-1 and +1) ($p=0.002$) and the distribution was skewed toward better scores (>0) ($p=0.005$).

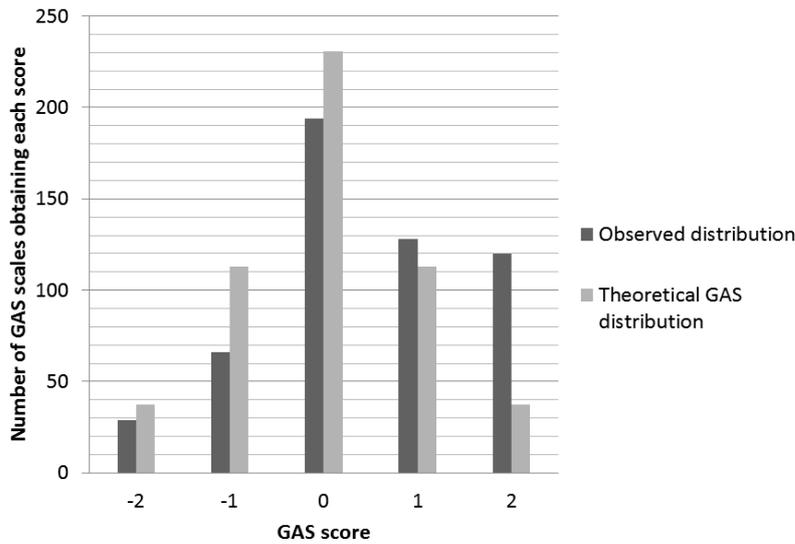


Figure 3: Distribution of all post-treatment GAS scores

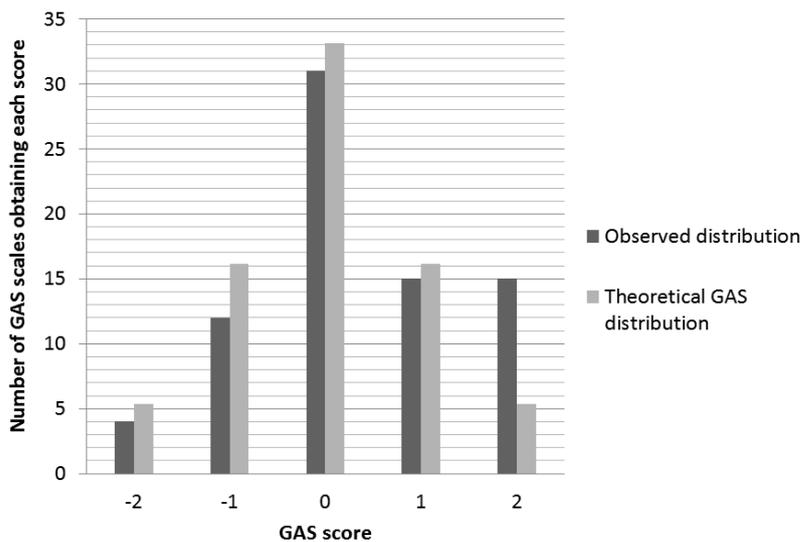


Figure 4: Distribution of post-treatment GAS scores for "functional GAS" category

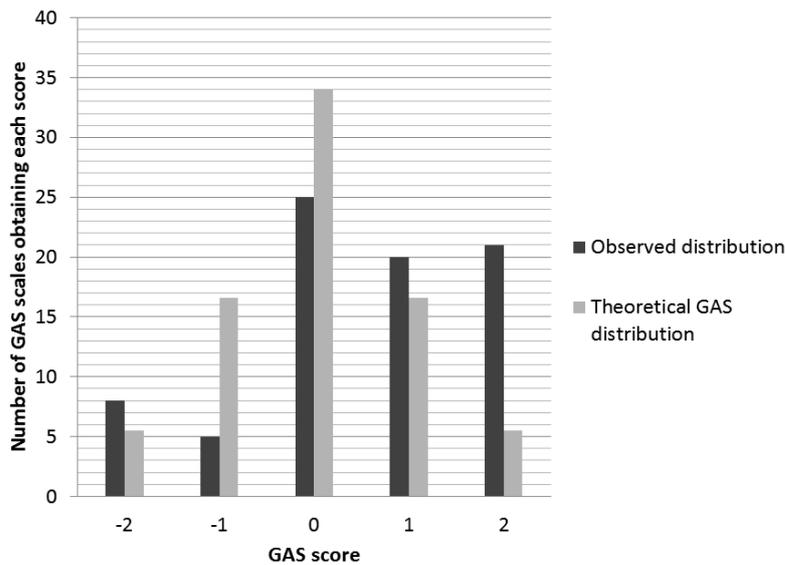


Figure 5: Distribution of post-treatment GAS scores for “family GAS” category

Discussion

Here we aimed to investigate the feasibility of using GAS in a clinical setting of pediatric spasticity with a shorter method, the 3-milestones GAS (goal setting with 3 levels and goal rating with the classical 5 levels), than the original method. The 3-milestones GAS method, as expected, showed a median score of 0 after BoNT-A treatment. The distribution pattern of the scores from all GAS scales on one hand and functional GAS scales on the other, showed a modal value of 0 scores and decreased frequency toward extreme scores, as expected. Most GAS scales referred to active-movement quality goals and fewer to family goals and activity domains. GAS could be used routinely for most BoNT-A treatment sessions. Only 4 GAS scales could not be scored and not because of inadequate formulation but because the goal was no longer an issue.

Therapists could determine goals and predict expected treatment outcomes as well as the possible levels of goal achievement. These findings suggest that community therapists, after appropriate training, in association with spasticity-clinic therapists, had knowledge of the BoNT-A usefulness and adequate skills to set realistic goals and choose adequate levels for GAS scales. However, the GAS scales in this study were discussed, checked, corrected, or changed at the pre-injection clinic visit. Hence, our results represent the joint combination of the spasticity-clinic team’s knowledge of BoNT-A treatment and the local therapists’ knowledge of the child’s possibilities and enabling/constraining factors (motivation, pain, family support, etc.). Thus, our results should not be generalized to contexts in which local

therapists or spasticity clinic teams alone create the GAS scales without their complementary knowledge of the child's possible progress. An interesting approach would have been to compare GAS scales set by the child's local therapist and those set by the spasticity clinic team, but this was not possible because of time constraints of the clinical setting. Indeed, if the spasticity clinic had to write their own GAS scales (to be compared with those from local therapist), from scratch, the clinic time would not have been sufficient. Instead, time was preferentially used to discuss and set collaboratively the most specific, measurable, attainable, and relevant (SMART) goals possible.

Despite the precise description of only 3 milestones, 5 levels could easily be used for scoring. We found significantly more scores corresponding to a better than expected outcome level of goal attainment than below the expected level of goal attainment: the effect of treatment was relatively underestimated when writing GAS scales. This problem is inherent with measures such as GAS: high scores can always be interpreted as a particularly effective treatment or as too easy goals for treatment^{6,11,533}. When all GAS scales were taken together, the distribution differed from the classical GAS distribution, but the functional GAS distribution followed the expected distribution. Precisely comparing our results with studies of GAS with BoNT-A in CP is difficult because the GAS score distribution is usually not reported in studies. Such studies report heterogeneous results with 1) a median of 0 scores²³ and T-scores around 50,²⁴ 2) a majority of -1 scores and T-scores < 50,^{24,51} or 3) a majority of +1 and T-scores > 50^{8,9}.

The discussion of BoNT-A efficacy is beyond the aim of this study; however, of note is the GAS methodological problem that higher scores may be due to a highly effective treatment or inadequate prediction of GAS levels (i.e., too easy levels that can be attained without the BoNT-A treatment [see Krasny-Pacini et al.¹¹ for a discussion of the impact of erroneous GAS levels on the judgment of an intervention efficacy]). The only study that reported full GAS score distribution in CP was the Löwing et al. study,⁵² testing goal-directed therapy with all GAS goals set with the family (equivalent to our "family GAS" category). The authors found a marked skew toward higher scores and a mode of +1, similar to our study, with family goals attained better than expected (although in our study the mode remained 0). This finding suggests that families are 1) better able to understand what the possible outcomes are and thus more likely to report those outcomes achieved after injection or 2) influenced by the process of the injections and because of their investment in the treatment are more likely to

report positive outcomes, perhaps even if they are not occurring or are not related to the injections.

The 3-milestones GAS for family goals did not yield similar results as the original GAS method: extreme scores (-2 and +2) were more frequent than intermediate scores (-1 and +1). The most probable explanation is that the levels of attainment corresponding to the 3 milestones described all possible outcomes and there was not enough “room” for intermediate conditions (especially the -1 level). Rating categories without “anchors” (i.e., 1 and -1 in this GAS approach) were less likely to be selected.

The 3-milestones approach may not be appropriate when writing family-focused goals, or the guidance the families received to define the -2, 0 and +2 levels may not have been adequate. We chose to describe the distribution of the most relevant GAS categories only (functional and family GAS), because these two categories are the only ones for which GAS should be used. There are other measures for body structures and functions, and it did not seem relevant (or would be even confusing) to analyze score distribution of categories for which GAS is not an adequate method.

We found a disproportion among goal types: quality of active movement goals were 3 times more numerous than all other types of goals (joint mobility, functional and family goals). In total, more than half of all goals corresponded to a quality of movement goal. Because goal themes were freely chosen, few goals (15%) corresponded to joint mobility (relating to reduction of spasticity or increased range of motion), which indicates that these days, therapists consider that a treatment goal should not only aim at attenuating impairments but also increase activity and participation. However, functional and family GAS goals represented only 30% of goals, although the aim of the goal-setting process was to promote a family-centered and functional approach. This finding can be explained in several ways. First, a functional or a family GAS is more difficult and takes longer to write because therapists cannot rely on the usual classical clinical examination (range of motion measurement, muscle tone and visual analysis of movement), but they need to listen to and discuss with the child and family to “know, uncover and understand what is meaningful,”⁵⁴ which takes time. Second, the goals relating to movement analysis, range of motion, tone, and spasticity fit in the traditional approach of “improving the quality of movement”, often used in our country, but family and functional GAS goals represent the concepts of family-centered practice⁵⁵, functional therapies^{18,56,57} and context-focused^{58,59} approaches. Finally the disproportion in

goals is consistent with the literature on BoNT-A efficacy, which shows its effectiveness for spasticity management but with less evidence of effectiveness in activity domains ⁴⁵.

A major limitation of this study but also of the therapists' culture in spasticity management in our country is the insufficient collaboration with families in goal setting and in intervention planning. That only 15% of goals were set collaboratively with the families reflects the still long way to go to reach the family-centered practice recommended in the literature. GAS training did not teach any methods of collaborative goal setting, motivational interviewing or family empowering. Future training in GAS should include and emphasize collaborative goal setting with families. Past research has shown the utility of the Canadian Occupational Performance Measure (COPM)⁶⁰, as a starting point for setting goals ⁶¹. Using that COPM rather than open-ended questions can increase the proportion of goals pertaining to activity domains ⁶².

The growing enthusiasm of clinicians for GAS is often hampered by the time needed for the development and writing of GAS scales. Our study shows the feasibility of GAS scales developed using a precise description of 3 levels only. Although the mean time needed to write one GAS scale could not be determined, a 45-min clinic was sufficient to write 1 to 7 GAS scales, as well as discuss parents' questions and needs regarding rehabilitation, the need for devices, medications, spasticity treatment. This study was designed as a feasibility study. The GAS score distribution corresponded approximately to our initial assumptions. We assume our method is more precise than the Turner-Stokes method, but it should be assessed further, especially regarding inter-rater reliability, before claiming that its psychometric properties are close to Steenbeek's reliable but time-consuming method. Nonetheless, our study shows that in a pediatric spasticity-management clinic, using a goal-based dialogue, physicians and therapists working in BoNT-A therapy, child's rehabilitation therapists, and children and family members can work together, set realistic goals and predict outcome after treatment.

A major drawback of GAS methodology is that it highly depends on the ability of the GAS setting team or staff member to generate valid, reliable and meaningful scales. Indeed, GAS may be more a measure of how adequately a therapist can foresee outcome than an outcome measure itself ^{5,33,35,36}. A child may show progress on GAS scales due to a measurement error or on a GAS scale that is not reliable because of too easy goals, has unequal distances between GAS levels or uses subjective criteria for goal attainment^{25,53}. Conversely, progress

may not be detected if the scale is written with levels of goal attainment that are too difficult for the child. Whatever the GAS method used, 3 milestones or other, therapists should be aware of this inherent problem of GAS.

The limitations of the study are that 1) the writing and the scoring of GAS scales were performed by the same people (children's therapists and spasticity-management team), 2) the GMFCS was used to classify all children with CP, including those older than 18 years, 3) we did not assess inter-rater reliability of the 3-milestones GAS, and 4) we did not compare GAS scales written by the local therapists and by the spasticity team (but reported GAS scales generated by their joint reflexion). These issues should be addressed in future studies. Another method to assess the 3-milestones GAS reliability would be to explore its "alternate-form reliability"¹ (i.e., the interrater reliability when one assessor uses one GAS form, here a form containing a precise description of all 5 GAS levels, and the other a 3-milestones GAS, here containing the precise description of 3 levels only), similar to Steenbeek's approach²⁶. The aim of the 3-milestone GAS is to be practical for use in clinical practice by therapists and it is not intended for research. When GAS is used as an outcome measure in research on treatment efficacy, GAS should meet a series of criteria¹¹ that are not easily applicable in routine clinical practice. As such, the present study can serve for clinical purposes but should by no means serve as a training model when GAS is used as an outcome measure to determine the efficacy of a new intervention. Such training should be supported by the recent publication of Krasny-Pacini et al.¹¹.

Conclusions

In collaboration with a pediatric spasticity-management clinic team, children's therapists can be trained to use the 3-milestones GAS to create and assess goals. This GAS method is feasible within the time constraints of the clinic. Therapists can determine realistic goals and predict expected treatment outcomes as well as the possible levels of goal achievement. Apart from the family GAS goals, the 3-milestones GAS scores had a distribution similar to the classical 5-level GAS, so it may be a relevant option for GAS in clinical settings, although inter-rater reliability needs to be explored. Most GAS scales (55%) referred to movement quality goals and fewer (29%) related to family goals and activity domains, which emphasizes the progress, but future work is still needed to focus more on function and less on quality of movement and to move to more functional and context-based goals and therapies after BoNT-

A. More collaboration with families in goal setting is the key challenge of the future use of GAS in clinical practice.

Conflict of interest. The spasticity pediatric clinic team has been provided with a video analysis software by Allergan, commercializing BoNT-A. Allergan also financed anatomy training for members of the spasticity-management team. The opinions expressed in this paper are the authors' own and are unrelated to Allergan policy because the aim of the study was not to study BoNT-A efficacy.

Legends

Figure 1. Flow of inclusion of Goal Attainment Scaling (GAS) scale. BoNT-A, botulinum toxin A

Figure 2. Distribution of GAS goals by 4 predetermined GAS categories.

Figure 3. Distribution of all post-treatment GAS scores.

Figure 4. Distribution of all post-treatment GAS scores for functional GAS goals.

Figure 5. Distribution of all post-treatment GAS scores for family GAS goals.

Acknowledgements

We thank all the occupational therapists and physiotherapists of Alsace region who accompanied the children to the spasticity clinic and who wrote GAS scales for their patients during the study. A special thanks to the occupational therapists and physiotherapists of ARAHM institutes "Iris", "Grillons" and "SSESD" in Strasbourg and for their enthusiasm for GAS and their valuable views about meaningful goals.

References

1. Kiresuk TJ, Sherman R. Goal Attainment Scaling: A general method for evaluating comprehensive community mental health programs. *Comm Mental Health J.* 1968;4(6):443-453.
2. Bovend'Eerdt TJ, Botell RE, Wade DT. Writing SMART rehabilitation goals and achieving goal attainment scaling: a practical guide. *Clin Rehabil.* 2009;23(4):352-361. doi:10.1177/0269215508101741.

3. Krasny-Pacini A, Hiebel J, Pauly F, Godon S, Chevignard M. Goal Attainment Scaling in rehabilitation: A literature-based update. *Annals of Physical and Rehabilitation Medicine*. 2013;56(3):212-230. doi:10.1016/j.rehab.2013.02.002.
4. Turner-Stokes L. Goal attainment scaling (GAS) in rehabilitation: a practical guide. *Clin Rehabil*. 2009;23(4):362-370. doi:10.1177/0269215508101742.
5. Schlosser RW. Goal attainment scaling as a clinical measurement technique in communication disorders: a critical review. *J Commun Disord*. 2004;37(3):217-239. doi:10.1016/j.jcomdis.2003.09.003.
6. Steenbeek D, Ketelaar M, Galama K, Gorter JW. Goal attainment scaling in paediatric rehabilitation: a critical review of the literature. *Dev Med Child Neurol*. 2007;49(7):550-556. doi:10.1111/j.1469-8749.2007.00550.x.
7. Vu M, Law AV. Goal-attainment scaling: a review and applications to pharmacy practice. *Res Social Adm Pharm*. 2012;8(2):102-121. doi:10.1016/j.sapharm.2011.01.003.
8. Desloovere K, Schörkhuber V, Fagard K, et al. Botulinum toxin type A treatment in children with cerebral palsy: evaluation of treatment success or failure by means of goal attainment scaling. *Eur J Paediatr Neurol*. 2012;16(3):229-236. doi:10.1016/j.ejpn.2010.09.006.
9. Lowe K, Novak I, Cusick A. Repeat injection of botulinum toxin A is safe and effective for upper limb movement and function in children with cerebral palsy. *Dev Med Child Neurol*. 2007;49(11):823-829. doi:10.1111/j.1469-8749.2007.00823.x.
10. Wallen MA, O'Flaherty SJ, Waugh M-CA. Functional outcomes of intramuscular botulinum toxin type a in the upper limbs of children with cerebral palsy: a phase II trial. *Archives of Physical Medicine and Rehabilitation*. 2004;85(2):192-200. doi:10.1016/j.apmr.2003.05.008.
11. Krasny-Pacini A, Evans J, Sohlberg MM, Chevignard M. Proposed Criteria for Appraising Goal Attainment Scales Used as Outcome Measures in Rehabilitation Research. *Archives of Physical Medicine and Rehabilitation*. 2016;97(1):157-170. doi:10.1016/j.apmr.2015.08.424.
12. Dreiling DS, Bundy AC. A comparison of consultative model and direct–indirect intervention with preschoolers. *American Journal of Occupational Therapy*. 2003;57(5):566–569.
13. Gharebaghy S, Rassafiani M, Cameron D. Effect of Cognitive Intervention on Children with ADHD. *Physical & Occupational Therapy In Pediatrics*. 2015;35(1):13-23. doi:10.3109/01942638.2014.957428.
14. Krasny-Pacini A, Limond J, Evans J, Hiebel J, Bendjelida K, Chevignard M. Context-sensitive goal management training for everyday executive dysfunction in children after severe traumatic brain injury. *J Head Trauma Rehabil*. 2014;29(5):E49-64. doi:10.1097/HTR.0000000000000015.

15. Mailloux Z, May-Benson TA, Summers CA, et al. Goal attainment scaling as a measure of meaningful outcomes for children with sensory integration disorders. *Am J Occup Ther.* 2007;61(2):254-259.
16. Mayson TA, Ward VJ, Harris SR. Parent goals as outcome measures for children receiving treadmill training: a series of case reports. *Dev Neurorehabil.* 2012;15(3):219-222. doi:10.3109/17518423.2011.654282.
17. Rosenberg L, Maeir A, Yochman A, Dahan I, Hirsch I. Effectiveness of a Cognitive-Functional Group Intervention Among Preschoolers With Attention Deficit Hyperactivity Disorder: A Pilot Study. *American Journal of Occupational Therapy.* 2015;69(3):6903220040p1. doi:10.5014/ajot.2015.014795.
18. Sorsdahl AB, Moe-Nilssen R, Kaale HK, Rieber J, Strand LI. Change in basic motor abilities, quality of movement and everyday activities following intensive, goal-directed, activity-focused physiotherapy in a group setting for children with cerebral palsy. *BMC Pediatrics.* 2010;10(1):26. doi:10.1186/1471-2431-10-26.
19. Ten Berge SR, Boonstra AM, Dijkstra PU, Hadders-Algra M, Haga N, Maathuis CGB. A systematic evaluation of the effect of thumb opponens splints on hand function in children with unilateral spastic cerebral palsy. *Clin Rehabil.* 2012;26(4):362-371. doi:10.1177/0269215511411936.
20. Wallen M, Ziviani J, Naylor O, Evans R, Novak I, Herbert RD. Modified constraint-induced therapy for children with hemiplegic cerebral palsy: a randomized trial. *Dev Med Child Neurol.* 2011;53(12):1091-1099. doi:10.1111/j.1469-8749.2011.04086.x.
21. Bjornson K, Hays R, Graubert C, et al. Botulinum toxin for spasticity in children with cerebral palsy: a comprehensive evaluation. *Pediatrics.* 2007;120(1):49-58. doi:10.1542/peds.2007-0016.
22. Hoare BJ, Wallen MA, Imms C, Villanueva E, Rawicki HB, Carey L. Botulinum toxin A as an adjunct to treatment in the management of the upper limb in children with spastic cerebral palsy (UPDATE). *Cochrane Database Syst Rev.* 2010;(1):CD003469. doi:10.1002/14651858.CD003469.pub4.
23. Steenbeek D, Meester-Delver A, Becher JG, Lankhorst GJ. The effect of botulinum toxin type A treatment of the lower extremity on the level of functional abilities in children with cerebral palsy: evaluation with goal attainment scaling. *Clin Rehabil.* 2005;19(3):274-282. doi:10.1191/0269215505cr859oa.
24. Wallen M, O'Flaherty SJ, Waugh M-CA. Functional Outcomes of Intramuscular Botulinum Toxin Type A and Occupational Therapy in the Upper Limbs of Children With Cerebral Palsy: A Randomized Controlled Trial. *Archives of Physical Medicine and Rehabilitation.* 2007;88(1):1-10. doi:10.1016/j.apmr.2006.10.017.
25. Krasny-Pacini A, Evans J, Sohlberg MM, Chevignard M. Proposal of criteria for appraising Goal Attainment Scales used as outcome measures in rehabilitation research. *Archives of Physical Medicine and Rehabilitation.* 0(0). doi:10.1016/j.apmr.2015.08.424.

26. Steenbeek D, Ketelaar M, Lindeman E, Galama K, Gorter JW. Interrater reliability of goal attainment scaling in rehabilitation of children with cerebral palsy. *Arch Phys Med Rehabil*. 2010;91(3):429-435. doi:10.1016/j.apmr.2009.10.013.
27. Palisano RJ. Validity of goal attainment scaling in infants with motor delays. *Phys Ther*. 1993;73(10):651-658-660.
28. Palisano RJ, Haley SM, Brown DA. Goal attainment scaling as a measure of change in infants with motor delays. *Phys Ther*. 1992;72(6):432-437.
29. Steenbeek D, Gorter JW, Ketelaar M, Galama K, Lindeman E. Responsiveness of Goal Attainment Scaling in comparison to two standardized measures in outcome evaluation of children with cerebral palsy. *Clin Rehabil*. 2011;25(12):1128-1139. doi:10.1177/0269215511407220.
30. Cusick A, McIntyre S, Novak I, Lannin N, Lowe K. A comparison of goal attainment scaling and the Canadian occupational performance measure for paediatric rehabilitation research. *Developmental Neurorehabilitation*. 2006;9(2):149-157. doi:10.1080/13638490500235581.
31. Steenbeek D, Ketelaar M, Galama K, Gorter JW. Goal Attainment Scaling in paediatric rehabilitation: a report on the clinical training of an interdisciplinary team. *Child Care Health Dev*. 2008;34(4):521-529. doi:10.1111/j.1365-2214.2008.00841.x.
32. McLaren C, Rodger S. Goal attainment scaling: Clinical implications for paediatric occupational therapy practice. *Australian Occupational Therapy Journal*. 2003;50(4):216-224. doi:10.1046/j.1440-1630.2003.00379.x.
33. Turner-Stokes L, Baguley IJ, De Graaff S, et al. Goal attainment scaling in the evaluation of treatment of upper limb spasticity with botulinum toxin: a secondary analysis from a double-blind placebo-controlled randomized clinical trial. *J Rehabil Med*. 2010;42(1):81-89. doi:10.2340/16501977-0474.
34. Cytrynbaum S, Ginath Y, Birdwell J, Brandt L. Goal Attainment Scaling A Critical Review. *Eval Rev*. 1979;3(1):5-40. doi:10.1177/0193841X7900300102.
35. Simeonsson RJ, Jr DBB, Huntington GS, Brandon L. Scaling and attainment of goals in family-focused early intervention. *Community Ment Health J*. 1991;27(1):77-83. doi:10.1007/BF00752717.
36. Ertzgaard P, Ward AB, Wissel J, Borg J. Practical considerations for goal attainment scaling during rehabilitation following acquired brain injury. *J Rehabil Med*. 2011;43(1):8-14. doi:10.2340/16501977-0664.
37. King GA, McDougall J, Palisano RJ, Gritzan J, Tucker MA. Goal Attainment Scaling: its use in evaluating pediatric therapy programs. *Physical & Occupational Therapy in Pediatrics*. 2000;19(2):31-52. doi:10.1080/J006v19n02_03.
38. Schut H, Stam H. Goals in rehabilitation teamwork, Disability and Rehabilitation, Informa Healthcare. *Disability & Rehabilitation*. 1994;16:223-226.

39. Bax M, Goldstein M, Rosenbaum P, et al. Proposed definition and classification of cerebral palsy, April 2005. *Dev Med Child Neurol*. 2005;47(8):571-576.
40. Leonard CT. Childhood Motor Disorders. *Pediatrics*. 2003;112(6):1462-1463.
41. Paneth N. Establishing the diagnosis of cerebral palsy. *Clin Obstet Gynecol*. 2008;51(4):742-748. doi:10.1097/GRF.0b013e318187081a.
42. Sanger TD, Delgado MR, Gaebler-Spira D, Hallett M, Mink JW. Classification and definition of disorders causing hypertonia in childhood. *Pediatrics*. 2003;111(1):e89-97.
43. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. *Dev Med Child Neurol*. 2008;50(10):744-750. doi:10.1111/j.1469-8749.2008.03089.x.
44. Lukban M, Rosales R, Dressler D. Effectiveness of botulinum toxin A for upper and lower limb spasticity in children with cerebral palsy: a summary of evidence. *Journal of Neural Transmission*. 2009;116(3):319-331. doi:10.1007/s00702-008-0175-8.
45. Novak I, McIntyre S, Morgan C, et al. A systematic review of interventions for children with cerebral palsy: state of the evidence. *Developmental Medicine & Child Neurology*. 2013;55(10):885-910. doi:10.1111/dmcn.12246.
46. Cardillo JE, Smith A. Psychometric issues. In: Kiresuk TJ, Smith A, Cardillo J, eds. *Goal Attainment Scaling: Applications, Theory, and Measurement*. Vol xviii. Hillsdale, NJ, England: Lawrence Erlbaum Associates, Inc; 1994:199.
47. World Health Organization. WHO | International Classification of Functioning, Disability and Health (ICF). WHO. <http://www.who.int/classifications/icf/en/>. Published 2001. Accessed December 29, 2015.
48. Tennant A. Goal attainment scaling: current methodological challenges. *Disabil Rehabil*. 2007;29(20-21):1583-1588. doi:10.1080/09638280701618828.
49. Merbitz C, Morris J, Grip JC. Ordinal scales and foundations of misinference. *Arch Phys Med Rehabil*. 1989;70(4):308-312.
50. Stucki G, Daltroy L, Katz JN, Johannesson M, Liang MH. Interpretation of change scores in ordinal clinical scales and health status measures: The whole may not equal the sum of the parts. *Journal of Clinical Epidemiology*. 1996;49(7):711-717. doi:10.1016/0895-4356(96)00016-9.
51. Mall V, Heinen F, Siebel A, et al. Treatment of adductor spasticity with BTX-A in children with CP: a randomized, double-blind, placebo-controlled study. *Dev Med Child Neurol*. 2006;48(1):10-13. doi:10.1017/S0012162206000041.
52. Löwing K, Bexelius A, Brogren Carlberg E. Activity focused and goal directed therapy for children with cerebral palsy – Do goals make a difference? *Disability and Rehabilitation*. 2009;31(22):1808-1816. doi:10.1080/09638280902822278.

53. Ruble L, McGrew JH, Toland MD. Goal Attainment Scaling as an Outcome Measure in Randomized Controlled Trials of Psychosocial Interventions in Autism. *J Autism Dev Disord.* 2012;42(9):1974-1983. doi:10.1007/s10803-012-1446-7.
54. Bright FAS, Boland P, Rutherford SJ, Kayes NM, McPherson KM. Implementing a client-centred approach in rehabilitation: an autoethnography. *Disabil Rehabil.* 2012;34(12):997-1004. doi:10.3109/09638288.2011.629712.
55. King S, Teplicky R, King G, Rosenbaum P. Family-centered service for children with cerebral palsy and their families: a review of the literature. *Semin Pediatr Neurol.* 2004;11(1):78-86.
56. Ahl LE, Johansson E, Granat T, Carlberg EB. Functional therapy for children with cerebral palsy: an ecological approach. *Developmental Medicine & Child Neurology.* 2005;47(9):613-619. doi:10.1111/j.1469-8749.2005.tb01213.x.
57. Ketelaar M, Vermeer A, Hart H 't, Beek E van P, Helders PJ. Effects of a Functional Therapy Program on Motor Abilities of Children With Cerebral Palsy. *PHYS THER.* 2001;81(9):1534-1545.
58. Darrah J, LAW MC, POLLOCK N, et al. Context therapy: a new intervention approach for children with cerebral palsy. *Dev Med Child Neurol.* 2011;53(7):615-620. doi:10.1111/j.1469-8749.2011.03959.x.
59. Law MC, Darrah J, Pollock N, et al. Focus on function: a cluster, randomized controlled trial comparing child- versus context-focused intervention for young children with cerebral palsy. *Developmental Medicine & Child Neurology.* 2011;53(7):621-629. doi:10.1111/j.1469-8749.2011.03962.x.
60. Law MC, Baptiste S, Carswell A, McColl MA, Polatajko H, Pollock N. *Canadian Occupational Performance Measure.* SLACK Incorporated; 1998.
61. Ostensjø S, Oien I, Fallang B. Goal-oriented rehabilitation of preschoolers with cerebral palsy--a multi-case study of combined use of the Canadian Occupational Performance Measure (COPM) and the Goal Attainment Scaling (GAS). *Dev Neurorehabil.* 2008;11(4):252-259. doi:10.1080/17518420802525500.
62. Verkerk GJQ, Wolf MJMAG, Louwers AM, Meester-Delver A, Nollet F. The reproducibility and validity of the Canadian Occupational Performance Measure in parents of children with disabilities. *Clin Rehabil.* 2006;20(11):980-988. doi:10.1177/0269215506070703.