

The Application of Goal Attainment Scaling in Cervical Dystonia – An Exploratory Observational Pilot Study

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Abstract: Background: Due to its heterogeneous manifestation an individualized approach to reach therapeutic goals in cervical dystonia (CD) is advantageous.

Objectives: The aim of the current study was to adapt goal attainment scaling (GAS) to drive the management of CD.

Methods: 38 patients with CD, regularly treated with botulinum neurotoxin (BoNT), were involved in the current exploratory observational pilot study. GAS, including domains of motor, pain, disability, and psychiatric features, was applied to set up individualized goals with the calculation of initial GAS T-scores. Following at least 4 BoNT injection cycles, patients were reassessed whether they reached the pre-set goals.

Results: The initial GAS T-scores (median: 36.9, range: 22.8–40) significantly improved ($P < 0.001$) to the end of the study (the median of final GAS T-scores: 50, range: 25.5–63.6).

Conclusions: The applicability of GAS in CD patients was confirmed, but further large-scale studies are needed refining this innovative approach.

Cervical dystonia (CD) is the most common form of focal dystonias, but its heterogeneous manifestation makes the diagnosis difficult and delayed in a substantial number of cases.^{1,2} Although the gold standard botulinum neurotoxin (BoNT) treatment can relieve numerous related symptoms, the identification of the involved muscles to be injected and appropriate dose selection are challenging and need expertise.³ However, the application of the collum-caput (COL-CAP) concept and the utilization of imaging tools and electromyography (EMG) may considerably help determine the best strategy.^{4–7} Although several rating scales were developed for patient follow-up in CD,^{8–10} overall, they did not yield an appropriate individualized approach for the management of heterogeneous symptoms and the evaluation of change.

Goal attainment scaling (GAS) has been frequently used for assessing outcomes in many different healthcare programs such as rehabilitation of patients with spasticity.¹¹ The GAS approach, composed of six steps (File S1), provides a useful framework to evaluate goal attainment, and measures achievement of expectation instead of measuring of

function.¹² Healthcare system is moving the focus of care from disease-oriented toward patient-oriented medicine, and there is growing evidence that goals are more likely to be achieved if patients are involved in setting them.^{13,14} Using this concept in the management of CD via the application of GAS for outcome measure would be a novel approach.

The primary aim of our study was to utilize GAS to measure the degree of change in the scale associated with BoNT treatment in CD. The secondary aim was to determine the influencing factors of baseline measure and its change.

Patients and Methods

CD patients who were diagnosed by movement disorder specialists based on the phenomenology and receiving at least three

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Keywords: cervical dystonia, botulinum neurotoxin, ultrasound, COL-CAP, goal attainment scaling.

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Received 11 August 2023; revised 29 March 2024; accepted 13 May 2024.

Published online 00 Month 2024 in Wiley Online Library (wileyonlinelibrary.com). DOI: 10.1002/mdc3.14118

consecutive previous BoNT injection cycles, out of which at least one was performed using the COL-CAP concept AND ultrasound (US) guidance at the outpatient BoNT clinic of the Department of Neurology, University of Szeged, were involved in the current study from November 2020 until September 2022. For the development of GAS in CD, a practical approach recommended in rehabilitation¹² (<https://www.kcl.ac.uk/nmpc/assets/rehab/gas-goal-attainment-scaling-in-rehabilitation-a-practical-guide.pdf>) was adapted as a first step. Next, keeping the original structural build-up, the following major domains were applied: 1. motor, 2. pain, 3. disability, 4. psychiatric features. The further developmental steps and the method of baseline and final GAS T-score (a derived numerical value which should be improved to ≥ 50 in mean if the goals are appropriately achieved) calculations are described in File S2 in details, whereas the final version of the 5-item Likert scale of grading for each subdomain and the record sheet for goals are presented in Files S3 and S4, respectively. Initially, the target date of reassessment (at the day of BoNT injection and before that) was determined as the fourth BoNT injection following study inclusion, but due to COVID-19 pandemic, that was modified to at least the fourth BoNT injection following study inclusion. All the involved patients successfully completed the study.

The factors assessed for their influencing (or confounding) effect on either the initial GAS T-score or GAS T-score change, or on both are presented in File S5, along with the factors studied for their association with the goal, ie, whether the patient reached at least GAS T-score of 50.

All statistical calculations were performed with the freely available R software (R Development Core Team, <https://www.r-project.org/>). The detailed description of statistical analysis is presented in File S6. Data reporting was performed according to the STROBE checklist for observational studies.

Results

The baseline and final demographics and treatment characteristics of the study population ($n = 38$) are presented in Table 1.

The change in GAS T-score (12.6; -0.8 – 32.3 , ± 7.6 ; 95% confidence interval: 10.1–15.1) was significant (paired t test: $P < 0.0001$; Cohen's d : 1.65 (large); Figure 1).

The influencing (or confounding) effects of the assessed factors on either the initial GAS T-score or GAS T-score change, or on both, or on global goal achievement are presented in Files S7–S13. In summary, the most important findings were that the

TABLE 1 Baseline and final demographics and injection characteristics of the study population

Variable	Baseline ($n = 38$)	Till/by final ($n = 38$)	P value
Age (years)	56.6 (24–79, ± 12.9)	58.4 (25–81, ± 12.9)	<0.0001
Gender (M/F)	12/26	12/26	1
Tremor	14	13	1
Number of COL-CAP forms per patient	2 (1–4, 1)	2 (1–5, 1)	1
Number of injected muscles per patient	3 (2–8, 1)	4 (1–8, 1)	0.7768
Change in the injected muscles	-	12	n.a.
Number of injection cycles	23 (3–99, 46.8)	30 (10–105, 47.5)	<0.0001
Number of injection cycles during the study period	-	7 (4–7, 1)	n.a.
Injection cycles according to the COL-CAP concept	7 (3–15, 7.8)	13 (10–22, 7.8)	<0.0001
US-guided injection cycles	5.5 (1–14, 1.8)	12 (8–21, 2)	<0.0001
Rate of injection cycles according to the COL-CAP concept AND US guidance	0.3 (0.02–1, 0.6)	0.5 (0.1–1, 0.6)	<0.0001
At least 50% injections according to the COL-CAP concept AND US guidance (finally reached/not reached GAS T-score of 50)	17 (14/3)	20 (17/3)	0.2482
Distribution of BoNT-A formulations (A/I/O)	18/12/8	18/12/8	1
Change in BoNT dose (increase/decrease)	-	7/10	n.a.
Corrected BoNT dose (unit)	110.5 (62.2–272.7, 45.8)	110.2 (41.4–207.2, 47.9)	0.7583
GAS T-score	36.9 (22.8–40, 1.4)	50 (25.5–63.6, 5.9)	<0.0001

Note: Data are presented as mean (minimum–maximum, standard deviation) or median (minimum–maximum, interquartile range). To reveal the differences in data count, McNemar test was applied, otherwise the Wilcoxon signed-rank test was used for group comparisons, except age and GAS T-score where paired t test was utilized. Abbreviations: A, aboBoNT; BoNT, botulinum neurotoxin; COL-CAP, collum-caput; F, female; GAS, goal attainment scaling; I, incoBoNT; M, male; n.a., not applicable; O, onaBoNT; US, ultrasound.

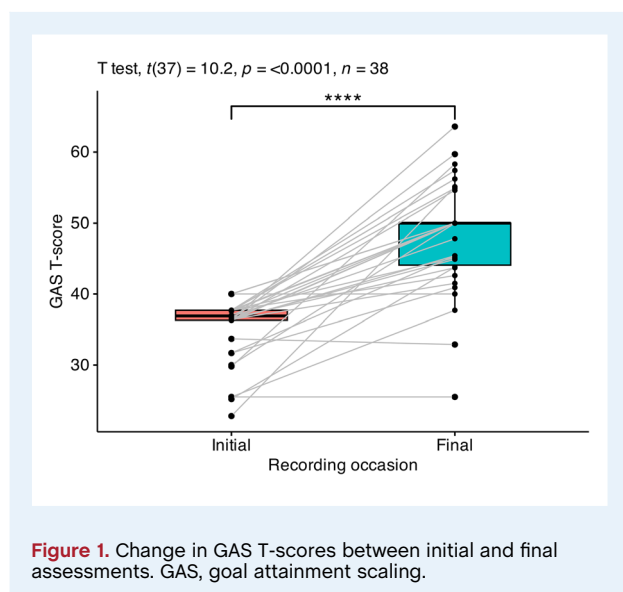


Figure 1. Change in GAS T-scores between initial and final assessments. GAS, goal attainment scaling.

presence of tremor resulted in significantly lower initial GAS T-scores, the main subtypes according to the COL-CAP concept significantly influenced GAS T-score change, and the correlation of this latter with the total number of injection cycles was caused by spuriousness. Furthermore, the GAS T-score change was significantly higher when at least 50% of injections were performed according to the COL-CAP concept and US guidance.

The results of the deeper analysis of primary and secondary goals regarding their effect on the initial GAS T-score and GAS T-score change are presented in Files S14 and S15. The major finding in this context was that the change in weighted scores was significantly higher when the goal was set as primary.

Discussion

The experience and satisfaction of patients are perhaps the most important measures of the quality of care.¹⁵ Accordingly, a patient driven journey map was developed in CD as well to draw attention to the significance of patient involvement in the design of healthcare services to improve their quality.^{16,17} However, currently there is no appropriate tool for measuring the achievement of individual expectations in CD, so its development may be essential to improve the quality of care. The easiest way would be the adaptation of such a widely applied tool from a related area, eg, the management of spasticity. Accordingly, the aim of this study was to utilize GAS for that purpose.

The initially calculated GAS T-scores were only affected by the presence of tremor (it was observed in 36.84% of cases) which resulted in significantly lower scores. This is in line with data from the literature that the presence of tremor in focal dystonias resulted in lower quality of life (QoL) by mainly affecting the physical role functioning.¹⁸ However, the robust improvement in GAS T-scores was no longer influenced by tremor.

The magnitude of GAS T-score change was slightly affected by the initial GAS T-scores, ie, the change was a little larger in patients with lower initial scores. However, this is not a robust finding, so it needs further confirmation in future studies. The results demonstrated that patients with at least 50% BoNT injection cycles according to the COL-CAP concept AND US guidance have a more than 10 times higher odds ratio to reach the therapeutic aim, ie, GAS T-score of 50. So the predominating approach (ie, the best possible selection of muscles and the use of targeting devices) probably matters more than the pure number of injection cycles. Furthermore, following a given period with conventional anatomical-based injections and muscle selection paradigm, patients may present less therapeutic response even if they are shifted to the management with novel approaches. Finally, the significant influence of the main subtypes according to the COL-CAP concept on the GAS T-score change may be resulted from their uneven distribution between the separated two groups (torticaput: 41.67%; torticollis: 44.44%, retrocaput: 25%, shift: 50%, laterocaput: 75% in the less responsive group; retrocollis: only one patient) as well, but propensity score matching cannot be carried out for such a data sample with $n = 38$.

The lack of difference between the three subdomains with the highest sample number (movement, pain, and tremor) in the weighted scores demonstrates that the applicability of GAS would be uniform regardless the type of major complaint. The finding that the magnitude of change was significantly higher when a goal was set as primary rather than secondary indicates that the focus was mainly kept on the primary goal to improve. The motor predominance of primary goals is not surprising, but since the strongest predictors regarding the QoL were demonstrated to be anxiety and depression,¹⁹ the psychiatric features were underrepresented when setting goals by the patients (only two primary and four secondary goals out of the 38 and 34, respectively). The reason for this avoidance may be the stigmatizing effect of having psychiatric problems, which is a quite common phenomenon in Hungary.

Only one abstract has been published regarding the use of GAS in CD.²⁰ In that study, 22 patients were recruited with three goals for each patient. Six weeks following BoNT treatment the patients were reassessed, and GAS T-scores were calculated again. 59.09% of patients reported GAS T-score of >50, and the mean change was 41.62%. Head tremor was the most concerned symptom (in 45.45% of cases). The current study focused on the long-term (following at least four with the median of seven BoNT injections) effects of treatment excluding acute treatment effects. Thereby only 23.68% of patients reported GAS T-score of >50, but 60.5% reached the therapeutic aim, ie, GAS T-score of ≥ 50 . Accordingly, the change in percentage was also lower (34.15%) in the current study. Similar to the previously reported findings, a motor feature (movement — 39.47%) predominated the selected goals, but not the tremor (18.42%). One possible reason behind this would be the relatively low frequency (36.84%) of tremor in the current sample compared to international data.²¹ Accordingly, the second most common primary complaint was pain.

The following major study limitations could be identified: 1. No CD specific scales or measures of QoL were applied to

validate the changes in GAS T-scores (a future consensus is needed on the composition of test battery covering all the corresponding domains — motor, non-motor, activities of daily living, and functioning — of CD). 2. The relatively low case number limited the interpretation of the results of subgroup analyses (eg, studying the effect of main subtype according to the COL-CAP concept or the difference between the characteristics of primary and secondary goals), and no propensity score matching could be carried out in case of unequal data distribution. Furthermore, appropriate model construction (either logistic or linear) to predict the magnitude of change with pre-specified initial parameters cannot be implemented either. 3. The study is lacking BoNT-naïve patients initially starting with the application of the COL-CAP AND US guidance.

Despite these limitations and its pilot nature the current study clearly demonstrated the applicability of GAS in setting up individualized therapeutic aims in CD in a measurable way, drawing attention to the utility of the simultaneous use of the COL-CAP concept and US guidance as well. However, further well-designed prospective multicentric studies with large (at least several hundreds of patients) sample numbers are needed implementing, but not restricted to the following approaches: 1. the determination of the magnitude of GAS T-score change in BoNT naïve patients; 2. the validation of GAS-T score change with selected battery of CD specific scales and measures of QoL; 3. the analysis of each type of isolated CD according to the currently published diagnostic criteria¹ with the inclusion of subgroup analysis regarding genetic forms²²; 4. the inclusion of other device-aided (eg, EMG) AND/OR rehabilitation techniques.²³ Finally, based on the obtained data the construction of robust models to predict the magnitude of therapeutic response may be achieved as well.

Author Roles

(1) Research project: A. Conception, B. Organization, C. Execution; (2) Statistical analysis: A. Design, B. Execution, C. Review and Critique; (3) Manuscript preparation: A. Writing of the first draft, B. Review and Critique.

M.Sz.: 1C, 2A, 2C, 3A
 G.G.: 1B, 1C, 2C, 3A
 L.Sz.: 1B, 2C, 3B
 A.S.: 1B, 2C, 3B
 T.G.: 1B, 2B, 3B
 P.K.: 1B, 2C, 3B
 D.Z.: 1A, 1B, 1C, 2A, 2B, 3A

Disclosures

Ethical Compliance Statement: All procedures performed in this study involving human participants were approved by the Regional Human Biomedical Research Ethics Committee of the University of Szeged and were in accordance with the 1964 Helsinki declaration and its later amendments or comparable

ethical standards. Written informed consent was obtained from all individual participants included in the study (Regional Human Biomedical Research Ethics Committee of the University of Szeged registration number is 22/2021). We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this work is consistent with those guidelines.

Funding Sources and Conflict of Interest: Open access publication was funded by the University of Szeged Open Access Fund, grant number: 6471. PK was supported by the Ministry of Innovation and Technology of Hungary from the National Research, Development and Innovation Fund (TKP2021-EGA-32) and Hetényi Géza grant. The authors declare that there are no conflicts of interest relevant to this work.

Financial Disclosures for the Previous 12 Months: MSz has nothing to disclose. GG received travel expenses, conference registration fees, and educational grants from Ipsen, Medis, Medtronic, and Sandoz. LSz received travel expenses, conference registration fees, and educational grants from Abbott, Abbvie, Medtronic, and Sandoz. AS received travel expenses, conference registration fees, and educational grants from Abbott, Orion, and Sandoz. TG has nothing to disclose. PK received speaker's honoraria, travel expenses, conference registration fees, and educational grants from Abbvie, EVER Neuro Pharma GmbH, Ipsen, Medtronic, Orion, and Richter Gedeon Plc. DZ received speaker's honoraria, travel expenses, conference registration fees, and educational grants from Abbvie, EVER Neuro Pharma GmbH, Ipsen, Medis, Medtronic, Orion, and Richter Gedeon Plc.

Data Availability Statement

The data that support the findings of this study are available from the corresponding author upon reasonable request. ■

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Supporting Information

Supporting information may be found in the online version of this article.

Supplemental file S1. Six steps of GAS approach.

Supplemental file S2. Some developmental steps of goal attainment scaling and the method of baseline and final GAS T-score calculations.

Supplemental file S3. Goal Attainment Scaling — applied domains with Likert scale of grading. VAS, visual analogue scale.

Supplemental file S4. Goal Attainment Scaling — record sheet. DOB, date of birth; SSN, social security number; w_i , the weight assigned to the particular goal; x_i , the attainment score for each goal.

Supplemental file S5. The factors assessed for their influencing (or confounding) effect on either the initial GAS T-score or GAS T-score change, or on both, and the factors studied for their association with the goal, ie, whether the patient reached at least GAS T-score of 50.

Supplemental file S6. The detailed description of statistical analysis.

Supplemental file S7. The influencing (or confounding) effects of the assessed factors on either the initial GAS T-score or GAS T-score change, or on both, or on global goal achievement.

Supplemental file S8. The effect of the presence of tremor on initial GAS T-score.

Supplemental file S9. The effect of main subtypes of CD according to the COL-CAP concept on GAS T-score change.

Supplemental file S10. Correlation of the change in GAS T-score with the total number of injection cycles (linear approach). Regression line (blue) with 95% confidence interval (green).

Supplemental file S11. Results of the partitioning cluster analysis of the change in GAS T-score with the total number of injection cycles.

Supplemental file S12. The effect of the percentage of US AND COL-CAP application on GAS T-score change.

Supplemental file S13. Possible confounders of initial GAS T-score and GAS T-score change. The initial GAS T-score was not influenced by the total number of previous injection cycles, rate of previous injection cycles according to the COL-CAP concept AND US guidance, and the main subtypes of CD according to the COL-CAP concept (A). However, the change in scores were influenced by these factors, including a spurious correlation with the total number of injection cycles (B). The initial GAS T-score was only influenced by the accompanying tremor (C), not affecting its change (D). Blue lines (green area: 95% confidence interval for the regression line) indicate fits for the whole data population, whereas red and black lines (red areas: 95% confidence intervals for the regression lines) that of only for the color-coded subpopulations according to the GAM. GAM, generalized additive model.

Supplemental file S14. The analysis of primary and secondary goals regarding their effect on the initial GAS T-score and GAS T-score change.

Supplemental file S15. The effect of goal classification on the change in the weighted scores.