BRIEF REPORT

Assessing sensitivity to change of the genomics outcome scale (GOS)

Michael Sing Onn Ting | Angus Clarke | Marion McAllister

Centre for Medical Education, School of Medicine, Cardiff University, Cardiff, UK

Correspondence
Marion McAllister, Centre for Medical Education, School of Medicine, Cardiff University, Cardiff, CF14 4XN, UK
Email: mcallistermf@cardiff.ac.uk

Funding information
Unfunded study

Abstract
The Genetic Counseling Outcome Scale (GCOS-24) is a 24-item patient-reported outcome measure (PROM) that was developed to evaluate genetic counseling and testing services by measuring the construct of empowerment. The Genomics Outcome Scale (GOS) is a 6-item version of GCOS-24 that was designed to provide a PROM for use both within and outside clinical genetics services and reduce respondent burden. However, unlike GCOS-24, the sensitivity to change of the GOS has not yet been assessed in appropriate clinical settings. We carried out pre- and post-clinic surveys using the GOS to assess sensitivity to change of the GOS and produce before-and-after GOS data as part of a service evaluation. The survey was sent to patients attending the genetic counseling clinic for a first appointment at the All Wales Medical Genetic Service from 8 April 2019 to 18 September 2019. Patients attending disease management clinics, where genetic issues were not the primary concern, were excluded from this study. A total of 138 respondents were included in the final analysis. The result shows that empowerment scores, measured using the GOS, were significantly higher (p<0.05) after clinic attendance. The GOS shows good sensitivity to change, with a medium-to-large effect size (Cohen's $d = 0.73$). The result also shows that the service is delivering measurable benefits for its service users.

KEYWORDS
communication, genetic counseling, genetics services, patient-reported outcome measure

What is known about this topic
The Genomics Outcome Scale (GOS) is a six-item questionnaire that was designed as a shorter version of GCOS-24 to provide a PROM that can be used both within and outside of clinical genetics services and reduce respondent burden. Correlation between the two scales demonstrated that GOS maintains the ability of GCOS-24 to capture the construct of empowerment ($r = 0.838$).

What this paper adds to the topic
This study has shown good sensitivity to change of GOS, with a medium-to-large effect size (Cohen's $d = 0.73$). It also demonstrates that the All Wales Medical Genetics Service is delivering measurable benefits to its service users.

This is an open access article under the terms of the Creative Commons Attribution License, which permits use, distribution and reproduction in any medium, provided the original work is properly cited.
© 2021 The Authors. Journal of Genetic Counseling published by Wiley Periodicals LLC on behalf of National Society of Genetic Counselors
1 | INTRODUCTION

Patient-reported outcome measures (PROM) are increasingly implemented in healthcare systems to evaluate the effectiveness and quality of care (Meadows, 2011). The Genetic Counseling Outcome Scale (GCOS-24) is a 24-item PROM that was developed to evaluate genetic counseling and testing services (McAllister et al., 2011). GCOS-24 was designed to measure the construct of empowerment, comprising five sub-dimensions of outcomes valued by patients: cognitive, decisional and behavioral control, emotional regulation, and hope (McAllister et al., 2010, 2011). Each item on the GCOS-24 is rated on a 7-point Likert scale (1 = strongly disagree, 7 = strongly agree) (McAllister et al., 2011). Scores range from 24 to 168 with higher scores indicating higher levels of empowerment (McAllister et al., 2011). GCOS-24 has been demonstrated to have good test–retest reliability, sensitivity to change, and construct validity. (McAllister et al., 2011)

GCOS-24 has been translated into Danish, Spanish, Dutch, and Brazilian Portuguese (Diness et al., 2017; Muñoz-Cabello et al., 2017; Segundo-Ribeiro et al., 2020; Voorwinden et al., 2019). It is used extensively in clinical and research settings in both the UK and internationally. GCOS-24 was used in six of the 25 UK regional clinical genetics service in 2011–12 as part of a service evaluation exercise (McAllister, 2016). It has also been used to evaluate specialist genetic services, including psychiatric, cardiovascular, and cancer genetic services (Inglis et al., 2015; Ison et al., 2019; Yuen et al., 2020).

The Genomics Outcome Scale (GOS) was designed as a shorter version of GCOS-24 to provide a PROM that can be used both within and outside of clinical genetics services and reduce respondent burden (Grant et al., 2019). GOS was created through qualitative cognitive interviews, analysis of an existing data set of GCOS-24 responses using Samejima’s Graded Response Model (GRM), and Rasch modelling (Grant et al., 2019). Correlation between the two scales demonstrated that GOS maintains the ability of GCOS-24 to capture the construct of empowerment ($r = 0.838$) (Grant et al., 2019).

However, unlike GCOS-24, the construct validity and sensitivity to change of the GOS have not yet been assessed (Grant et al., 2019).

This study aims to (a) assess sensitivity to change of GOS and (b) provide before-and-after GOS data to the NHS All Wales Medical Genetics Service (AWMGS) as part of a service evaluation.

AWMGS is a clinical genetics service offering genetic counseling and testing in families where a genetic condition is suspected or known to be present, including autosomal dominant, recessive and X-linked conditions, learning disability, chromosome abnormalities, and developmental delay. Patient may be seen by a clinical geneticist or a genetic counselor. Previous research has demonstrated that patients attending AWMGS experience increased level of empowerment following clinic attendance, as captured by GCOS-24 (Costal Tirado et al., 2017), and so we would expect that if GOS is sensitive to changes in empowerment, that GOS would also capture increased level of empowerment amongst patients after AWMGS clinic attendance. From 8 April 2019 to 18 September 2019, patients attending the genetic counseling clinic for a first appointment at AWMGS were sent a pre-appointment GOS (Figure 1) by post, with completed pre-clinic questionnaires handed in on arrival at clinic. Patients who had forgotten to bring the completed pre-appointment GOS (Figure 1) were given a new pre-appointment GOS at the clinic to fill in on the spot. All patients completing and returning a pre-appointment GOS (Figure 1) were sent a post-appointment GOS together with a reply-paid envelope 4 weeks after clinic attendance. Patients attending disease management clinics, where genetic issues were not the primary concern, were excluded from this study.

Unfortunately, it has not been possible to calculate a participation rate or a completion rate. This is because there are so many locations across Wales in which clinics are held, with some in use every day and others only once per 1-2 months, and they have different secretarial and administrative staff involved. Practices have therefore diverged in terms of the proportion of patients given the questionnaires at each stage.

The before-and-after GOS data were inserted into a Microsoft Excel spreadsheet. The data were then analyzed using a paired samples t-test with IBM SPSS. After reversing the score for Question 3, the score of all six items was added up to provide a total empowerment score. We tested a hypothesis that empowerment scores would be significantly higher after clinic attendance. If the null hypothesis were rejected, this would show sensitivity to change of the

FIGURE 1 The genomics outcome scale (GOS)
were significantly different, we conducted a paired samples t-test \((t = 4.585)\) than post-clinic \((3.530)\).

(See Figure 3) The pre-clinic standard deviation was slightly wider on the high end of the scale compared to pre-appointment scores. Post-appointment scores tend to be distributed more powerment scores increased by an average of 2.97 points after the appointment.

Average empowerment (total) scores pre- and post-appointment.

Post-appointment empowerment scores were significantly higher for all items, except Question 3 (When I think about the condition in my family, I get upset).

Post-appointment empowerment scores were significantly higher than pre-appointment empowerment scores with an average increase of 3.26 points. This study has shown good sensitivity to change of GOS, with a medium-to-large effect size and some evidence of construct validity of the GOS. GOS has previously been shown to capture empowerment (Grant et al., 2019).

The present study has demonstrated not only that GOS is sensitive to change in empowerment levels, but has also demonstrated that patients attending AWMGS have higher levels of empowerment following clinical attendance. This therefore demonstrates that participants in this study have derived a measurable benefit from AWMGS attendance.

The effect size of this study \((d = 0.73)\) compares favorably with the original GCOS-24 study done in the UK \((d = 0.70)\) (McAllister et al., 2011). Published studies that have used the GCOS-24 have shown a wide range of effect size. Studies in psychiatric and cardiovascular genetic counseling demonstrated a large effect size \((d = 1.10\) and 0.94 respectively), whereas a 2018 Spanish study and a 2017 UK study showed medium-to-large effect size \((d = 0.70\) and 0.64 respectively) (Costal Tirado et al., 2017; Gerrard et al., 2020; Ison et al., 2019; Muñoz-Cabello et al., 2017). This variation in effect size could be due to variation in patient populations and practice models of genetic counseling in different centers.

Post-appointment empowerment scores were significantly higher for all questions, except Q3 (When I think about the condition in my family, I get upset), which addresses the subdomain of emotional regulation within the concept of empowerment (McAllister et al., 2011). This could signal a need for additional counseling at AWMGS to address deeper levels of emotional distress, or it could mean that emotional regulation is more intractable amongst patients of clinical genetics services because of the particular challenges that they face (Costal Tirado et al., 2017; Yuen et al., 2020). However, the GOS has been designed in such a way that it is the total score that is important, not the scores on the individual items.

There is currently conflicting evidence in the literature regarding the subdomain of emotional regulation of the GCOS-24. A study done in Vancouver, BC, has shown that the subdomains of empowerment on which psychiatric genetic counseling had the largest effect were powerlessness and emotional regulation (Gerrard et al., 2020).

\[ d = \frac{M_1 - M_2}{s_{pooled}} \]
This contrasts with the findings of studies in cancer genetics in Singapore and general clinical genetics in the UK, which showed no significant improvement in the emotional regulation sub-scale after genetic counseling (Costal Tirado et al., 2017; Yuen et al., 2020). The variability of the impact of genetic counseling on emotional regulation may be because of differences in the style of genetic counseling in different settings. As these studies were done in different countries, and with different types of patients, the variability may also be due to clinical and/or sociocultural differences between the participants. As Gerrard et al. (2020) commented, the differences in outcome observed between different patient populations would be a fruitful area for further research.

There were some important limitations to this study. Firstly, the extent to which the GOS survey was completed by all eligible patients is unknown. Whilst this is an important limitation, it does not significantly weaken the study findings because an adequate number of responses were collected to enable assessment of sensitivity to change of GOS. The study was also geographically limited to Wales, so the findings may not be reflective of genetic counseling services in other parts of the UK or internationally.

This study was also conducted purely in a clinical genetics setting. As the GOS is designed to be used in both clinical genetics settings and in other clinical specialties or research contexts where genetic counseling and/or testing are offered, it will be...
important to assess the usefulness of GOS in these other settings, for example, where genetic/genomic testing is offered in other medical specialties (‘mainstreaming’), for example, oncology, pediatrics and neurology. It would also be useful to conduct follow-up work to establish (i) the construct validity of GOS by testing hypotheses regarding how responses correlate / do not correlate with responses to other measures of similar constructs, that is, locus of control, depression, satisfaction of life (ii) the test-retest reliability and (iii) the minimum clinically important difference (MCID) for the GOS.

AUTHOR CONTRIBUTIONS
Michael Sing Onn Ting: Analysis and interpretation of data, writing of manuscript; Angus Clarke: Design of the study, data collection, supervision of Michael Sing Onn Ting in writing the manuscript, manuscript preparation; Marion McAllister: Design of the study, supervision of Michael Sing Onn Ting in the analysis of the data and in writing manuscript, manuscript preparation. Author Michael Sing Onn Ting confirms that he had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. All of the authors gave final approval of this version to be published and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

ACKNOWLEDGEMENTS
Dr. Melanie Myers served as Action Editor on the manuscript review process and publication decision. The authors thank the patients who agreed to participate in this survey and the clinical staff who contributed to data collection.

COMPLIANCE WITH ETHICAL STANDARDS

CONFLICT OF INTEREST
The authors have no conflict of interest to declare.

HUMAN STUDIES AND INFORMED CONSENT
All procedures performed in this study involving human participants were in accordance with the ethical standards of Cardiff & Vale University Health Board Research & Development Office and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study.

ANIMAL STUDIES
No non-human animal studies were carried out by the authors of this article.

DATA AVAILABILITY STATEMENT
Data available on request from the authors: The data that support the findings of this study are available from the corresponding author upon reasonable request.

ORCID
Marion McAllister https://orcid.org/0000-0001-7188-9255

REFERENCES
Benjamini, Y., & Hochberg, Y. (1995). Controlling the false discovery rate: a practical and powerful approach to multiple hypothesis testing. *Journal of the Royal Statistical Society: Series B*, 57, 289–300.
Costal Tirado, A., McDermott, A. M., Thomas, C., Ferrick, D., Harris, J., Edwards, A., & McAllister, M. (2017). Using patient-reported outcome measures for quality improvement in clinical genetics: An exploratory study. *Journal of Genetic Counseling*, 26(5), 1017–1028. https://doi.org/10.1007/s10897-017-0079-6
Diness, B., Overbeck, G., Hjortshøj, T., Hammer, T., Timshel, S., Sørensen, E., & McAllister, M. (2017). Translation and adaptation of the genetic counselling outcome scale (GCOS-24) for use in Denmark. *Journal of Genetic Counseling*, 26(5), 1080–1089. https://doi.org/10.1007/s10897-017-0086-7
Gerrard, S., Inglis, A., Morris, E., & Austin, J. (2020). Relationships between patient- and session-related variables and outcomes of psychiatric genetic counseling. *European Journal of Human Genetics : EJHG*, 28(7), 907–914. https://doi.org/10.1038/s41431-020-0592-1
Grant, P. E., Pampaka, M., Payne, K., Clarke, A., & McAllister, M. (2019). Developing a short-form of the genetic counselling outcome scale:
The genomics outcome scale. European Journal of Medical Genetics, 62(5), 324–334. https://doi.org/10.1016/j.ejmg.2018.11.015

Inglis, A., Koehn, D., McGillivray, B., Stewart, S. E., & Austin, J. (2015). Evaluating a unique, specialist psychiatric genetic counseling clinic: uptake and impact. Clinical Genetics, 87(3), 218–224. https://doi.org/10.1111/cge.12415

Ison, H. E., Ware, S. M., Schwantes-An, T. H., Freeze, S., Elmore, L., & Spoonamore, K. G. (2019). The impact of cardiovascular genetic counseling on patient empowerment. Journal of Genetic Counseling, 28(3), 570–577. https://doi.org/10.1002/jgc4.1050

McAllister, M. (2016). Genomics and patient empowerment. In D. Kumar, & R. Chadwick (Eds.), Genomics and society. Elsevier.

McAllister, M., Dunn, G., & Todd, C. (2010). Empowerment: qualitative underpinning of a new clinical genetics-specific patient-reported outcome. European Journal of Human Genetics, 19(2), 125–130. https://doi.org/10.1038/ejhg.2010.160

McAllister, M., Wood, A., Dunn, G., Shiloh, S., & Todd, C. (2011). The genetic counseling outcome scale: A new patient-reported outcome measure for clinical genetics services. Clinical Genetics, 79(5), 413–424. https://doi.org/10.1111/j.1399-0004.2011.01636.x

Meadows, K. (2011). Patient-reported outcome measures: an overview. British Journal of Community Nursing, 16(3), 146–151. https://doi.org/10.12968/bjcn.2011.16.3.146

Muñoz-Cabello, P., García-Miñáur, S., Espinel-Vallejo, M., Fernández-Franco, L., Stephens, A., Santos-Simarro, F., Lapunzina-Badia, P., & McAllister, M. (2017). Translation and cross-cultural adaptation with preliminary validation of GCOS-24 for use in Spain. Journal of Genetic Counseling, 27(3), 732–743. https://doi.org/10.1007/s10897-017-0154-z

Segundo-Ribeiro, M., Tavares Bacala, B., de Andrade Alvarenga, W., Castanheira Nascimento, L., McAllister, M., & Flória-Santos, M. (2020). Adaptation and preliminary validation of the genetic counseling outcome scale (GCOS-24) in a Brazilian genetic counseling setting. European Journal of Medical Genetics, 63(11), 104018. https://doi.org/10.1016/j.ejmg.2020.104018

Voorwinden, J., Plantinga, M., Krijnen, W., Ausems, M., Knoers, N., Velthuizen, M., Birnie, E., Lucassen, A. M., van Langen, I. M., & Ranchor, A. V. (2019). A validated PROM in genetic counselling: the psychometric properties of the Dutch version of the Genetic Counselling Outcome Scale. European Journal of Human Genetics, 27(5), 681–690. https://doi.org/10.1038/s41431-018-0318-9

Yuen, J., Lee, S. Y., Courtney, E., Lim, J., Soh, H., Li, S. T., Chen, Y., McAllister, M., Fenwick, E. K., & Ngeow, J. (2020). Evaluating empowerment in genetic counseling using patient-reported outcomes. Clinical Genetics, 97(2), 246–256. https://doi.org/10.1111/cge.13646

How to cite this article: Ting MSO, Clarke A, McAllister M. Assessing sensitivity to change of the genomics outcome scale (GOS). J Genet Couns. 2021;00:1–6. https://doi.org/10.1002/jgc4.1429