Evaluating Empowerment in Genetic Counseling Using Patient Reported Outcomes

Running head: Evaluating empowerment arising from cancer genetic counseling

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Conflict of Interests

All authors have no conflicts of interest to declare.

Data Availability

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

Data about patient-reported outcomes from cancer genetics services (CGS) are lacking but are essential to guide service evaluation and improvements. We measured improvement in empowerment, following genetic counseling in Singapore using a culturally-adapted version of the Genetic Counseling Outcome Scale (GCOS-24); and sought to identify factors associated with change in empowerment. The GCOS-24 was administered to 155 patients of the CGS, at pre- and post-counseling or testing timepoints. Of which, 110 patients underwent genetic testing. Individual pre- and post-counseling responses were subjected to Rasch analysis; the scale was subsequently split into Cognitive Control (CC) and Emotional Control (EC) domains. Associations of baseline characteristics with changes in pre- and post-CC and EC scores were assessed using multiple regression analysis. Both CC and EC scores showed significant improvement following genetic counseling and testing. While all items in the CC domain of being showed increases at follow-up, aspects of EC related to alleviating negative emotions ($p = 0.88$) and hopelessness ($p = 0.2$) did not demonstrate significant improvement. Our study revealed significant improvement in patient empowerment in patients who have received cancer genetic counselling, while revealing a need to cultivate hope and facilitate the alleviation of negative emotions in patients during genetic counselling.

Word count: 200

Keywords: Genetic Counseling, Genetic Testing, Genetic Services, Rasch Analysis, Regression Analysis, Hope
INTRODUCTION

Genetic counseling is a communication process which aims to help individuals and families understand and adapt to the medical, psychological, familial and reproductive implications of a heritable genetic condition.\textsuperscript{1,2} Though the practice of clinical genetics was established in the 1970s,\textsuperscript{3} evaluation of its impact on patient-reported outcomes has been lacking, due to the paucity in the availability of robust outcomes.\textsuperscript{4,5}

In Singapore, there is a growing demand for cancer genetic services\textsuperscript{6,7} and, as such, it is a priority to evaluate patient-reported outcomes from genetic counseling and testing. The lack of such information impedes progress in the field, as evidence-based improvements cannot be made.

The Genetic Counseling Outcome Scale (GCOS-24) (Supplementary Materials 1) is a validated genetics-specific Patient Reported Outcome Measure (PROM) and assesses patient-reported outcomes from genetic counseling and testing.\textsuperscript{8} It captures a construct coined ‘empowerment’, defined as ‘a set of beliefs that enable a person from a family affected by a genetic condition to feel that they have some control over and hope for the future’.\textsuperscript{9} It encompasses components of decisional control, cognitive control, behavioural control, emotional regulation and hope. Furthermore, the GCOS-24 has demonstrated utility in service evaluation\textsuperscript{10} and quality improvement\textsuperscript{11} in genetic counseling services.

It has been used in a study of 42 patients from a cardiology setting in USA by Ishon et al\textsuperscript{12}, which demonstrated significant improvement in empowerment scores, which consequentially led to a greater awareness for surveillance recommendations in patients following genetic counseling. In the psychiatric context, a recent publication which used the GCOS-24 on a larger sample size showed an increase in empowerment following genetic counseling.\textsuperscript{13} Similar increases in empowerment were observed in Danish\textsuperscript{14}, Dutch\textsuperscript{15} and Spanish\textsuperscript{11} validations of the GCOS-24.

However, one limitation of the GCOS-24 is that psychometric evaluation of GCOS-24 has largely involved classical test theory to date. Rasch analysis, a form of item response theory, provides significant insight into the psychometric properties of a scale,\textsuperscript{16,17} including: appropriate use of response categories; measurement precision; how well items ‘fit’ the underlying trait; how well the items measure a specific construct.
(unidimensionality); targeting of item difficulty to participants’ ability; and differential
item functioning (DIF; item bias). Rasch analysis has used by Grant et al\textsuperscript{18} to develop
a short-form of the GCOS-24, to create a less burdensome scale for respondents that
is similarly capable of capturing genetic counseling and testing-derived empowerment.

The aims of this study were threefold. Firstly, we aimed to measure the improvement
in patient empowerment, if any, following cancer genetic counseling using the GCOS-
24; with the secondary intention to identify and understand the factors associated with
change in empowerment. Finally, we aimed to evaluate the psychometric properties
of the GCOS-24 using Rasch analysis.

MATERIALS AND METHODS

Study Design

This was a single arm, pre-post counseling (intervention) study conducted between
May 2016 and May 2017 at the Cancer Genetics Service (CGS) at the National Cancer
Centre Singapore (NCCS). We represent a specialized cancer genetics service with
master’s level trained genetic counsellors working under a model of care adapted from
the United States.\textsuperscript{19} The CGS sees predominantly Singaporean Chinese, Malay, and
Indian patients with a personal and/or family history of cancer referred from general,
surgical, oncologic and gynecological specialties. The GCOS-24 was offered to
English-speaking, adult (≥21 years old) participants attending the CGS for the first
time. Individuals with significant hearing impairment (questionnaire administration
could take place over the telephone), cognitive impairment or any physical disability
that prevented them from participating in the study were excluded. Written informed
consent was obtained from all participants prior to the study and the study protocol
was approved by the SingHealth Centralised Institutional Review Board (CIRB number
2016/2367).

Study procedure

Participants were recruited face-to-face at the clinic, and after informed consent was
obtained, they were asked to complete the pre-counseling GCOS-24 prior to their first
genetic counseling session. The recruitment process was conducted by a research
coordinator (HS). The pre-test genetic counseling session was led by a genetic
counsellor (STL, EC, or YC) or a clinical cancer geneticist (JN). It typically included a
verbal discussion, with the use of visual aids, to provide information on the suspected
genetic condition and cancer risk assessment based on personal and family history.
Counselling skills are applied to facilitate coping and adaptation to the knowledge of a
possible hereditary condition that runs in the family. The goal of the session is to reach
a shared decision for genetic testing between the participant and their families, that is
aligned with clinical recommendations. These sessions generally lasted between 30
to 45 minutes. Participants had the option of completing the post-counseling GCOS-
24 via telephone, mail (written) or online methods (via Google survey), which was
facilitated by a research coordinator (HS).

During the counselling session, patients who met clinical testing criteria were offered
genetic testing to understand if they carried a pathogenic variant that predisposes
them to cancer. There were also asymptomatic patients who came for genetic
counseling as they were considering predictive testing for a familial condition. For
patients who declined genetic testing, the post-counseling GCOS-24 was conducted
2 weeks after their most recent counseling session. They were subsequently given an
open date appointment. For patients who elected to undergo genetic testing, an in-
person result disclosure appointment (with STL, EC, YC or JN) was scheduled 2 to 6
weeks after, dependent on turnaround time for testing ordered. These appointments
typically last for 15 to 45 minutes, dependent on the type of result that was returned.
The post-counseling GCOS-24 was administered 2 weeks after results disclosure (i.e.
4 to 8 weeks after they completed the pre-counseling GCOS-24).

Participants also completed a sociodemographic questionnaire which captured
information about their gender, age, ethnicity, education status, genetic testing subsidy
eligibility (eligible <SGD$1,800 monthly household income per person) and personal
and family history of cancer. All data collected were anonymized.

Cultural Adaptation of GCOS-24
The GCOS-24 scale comprises 24 items across five domains (decisional control: three
items, cognitive control: six items, behavioral control: eight items, emotional regulation:
three items and hope: four items) which are rated on a seven-point Likert-type
response scale ranging from ‘strongly agree’ to ‘strongly disagree’. Scores are
summed to provide an overall ‘empowerment’ and domain scores, where higher scores equal higher levels of empowerment.

Because the GCOS-24 was developed in the UK, we first conducted some cognitive interviews with patients to assess the clarity and cross-cultural applicability of the GCOS-24 items for eliciting the required information. English-speaking patients aged 21 and above, who had previously received genetic counseling at the CGS (n=12), were interviewed by trained interviewers. Interviews were audio-recorded and noted on standardized interview forms (Supplementary Materials 2). Responses were reviewed iteratively by the study team (JY, EF, MM & JN), and were used to guide edits to the GCOS-24 to improve clarity and comprehensibility of the items (Supplementary Materials 1). There were no edits that changed the original meaning of items made (Supplementary Materials 1). Item 6 was modified to ‘I can see that good things (e.g. early detection & personalized screening) have come from having this condition in my family.’, where the examples of ‘early detection & personalized screening’ were added for better comprehension of what ‘good things’ might refer to. Item 10 was edited to ‘I don’t know what could be gained from each of the options (e.g. genetic testing) available to me.’, where the example of ‘genetic testing’ was included to explain what ‘options’ might refer to.

Other Modifications of the GCOS-24
The response scale was modified to include a ‘not applicable’ option for items relating to children (items 3, 13, 19, 21, 24) to provide an appropriate response for those participants who did not have and were not considering children in the future. This was an outcome of participant feedback we received from the cognitive interview exercise. While the addition of a ‘not applicable’ option response may influence the psychometric properties of the instrument as it creates the potential for missing data. Unlike in classical test theory, where missing data is a problem, Rasch analysis does not require complete data in order to generate person measure estimates. Therefore, the addition of a ‘not applicable’ option instead has improved the psychometric properties of the GCOS-24 as participants are not forced to answer items irrelevant to them, and any ensuing missing data will not affect the score provided by Rasch analysis.
Psychometric Assessment of the GCOS-24

Rasch analysis was used to assess the psychometric properties of the adapted GCOS-24 using the Andrich rating scale model\textsuperscript{23} with Winsteps software (version 3.92.1), Chicago, Illinois, USA.\textsuperscript{24} Rasch analysis transformed the ordinal ratings of the questionnaire into estimates of interval measures (expressed in log of the odds units, or logits) to allow for parametric testing.\textsuperscript{25} Item bias, thereby DIF was assessed for gender, age, educational status and presence of strong cancer family history to establish possible associations between baseline patient characteristics with magnitude of change in empowerment. To ensure that differences between the pre- and post counseling GCOS-24 scores were valid indicators of changes over time, pre- counseling and post-counseling GCOS-24 data were stacked and DIF for time points was assessed. Absence of DIF was considered evidence of invariance over time.

The adapted GCOS-24 displayed good precision (person separation index (PSI) $> 2.0$) and targeting (difference between person and item means $< 1.0$) and no DIF for age, gender or time (Table 1). However, there was evidence of multidimensionality within the scale, with the eigenvalue for the first contrast $> 2.0$, the variance explained by the first factor $< 50\%$ and 3 mis-fitting items. Moreover, inspection of the standardized residual loadings for items indicated that 6 items were all relating to cognitive, behavioral or decisional control, loaded together. Therefore, based on this and the domain structure posited in the paper by Tirado et al,\textsuperscript{11} this supported the splitting of GCOS-24 into two discrete scales which were analyzed separately: 1) 'Cognitive control' [CC] (items 1-3, 5, 7, 10, 12-18, 23 and 24), which encompassed making informed decisions about the future, forward planning, decision-making, the utilization of socioeconomic and health-related resources and systems and the integration and contextualization into one’s own healthcare blueprint; and 2) 'Emotional control' [EC] (items 4, 6, 8, 9, 11, 19-22), which encompassed hope and emotional regulation.

The CC scale initially displayed disordered thresholds (meaning that some of the response categories were not being used as intended) and multidimensionality with a high eigenvalue, low variance explained for the first contrast and two mis-fitting items (Table 1). However, upon iterative removal of items 13, 12, 18 and 5, measurement precision increased and the disordered thresholds and multidimensionality were largely resolved. The emotional domain had suboptimal precision (PSI $< 2.0$) and
possible evidence of multidimensionality (eigenvalue of first contrast >2.0) (Table 1).

However, only three items (4, 11 and 21) loaded together, which was not enough to
form a separate scale; therefore, no further splitting was applied.

**Statistical Analysis**

Responses of participants who failed to complete the post-counseling GCOS-24 were
excluded from analysis. The patient sample was characterized using mean (standard
deviation [SD]) and median (interquartile range [IQR]) for description of normally and
non-normally distributed data respectively.

Wilcoxon signed-rank test was used to determine significant differences in the CC and
EC domains post-intervention. We also present an item-by-item analysis as well as for
the overall score. Additionally, effect sizes (ES; calculated as the difference in the
mean scores between the baseline and follow-up examinations divided by the
standard deviation (SD) of the scores for the baseline group) were utilized to determine
clinically significant pre-post changes.\(^{26}\) An ES of 0.20-0.49 was considered small,
0.50-0.79 as moderate and ≥0.80 as large.\(^{27}\)

The association of baseline characteristics with pre-post counseling changes in
cognitive and emotional control domains were assessed by multiple regression models
fit using the baseline characteristics as independent variables, with changes in CC
and EC scores between baseline and post-counselling scores as dependent variables,
respectively. For each model, variable selection was conducted via best subsets
selection using the Akaike Information Criterion (AIC), leading to a final reduced
model. All analyses were performed using Stata 15.0 (Statacorp LP, College Station,
TX, USA), and statistical significance was defined as \(p < 0.05\).
RESULTS

Baseline Characteristics

Of the 208 participants who were invited to participate in this study, 155 completed the GCOS-24 at both time-points and were included in the analysis (response rate: 74.5%). Most were female (n=136, 87.7%), median age was 46 (18-71) years old, and majority were Chinese (n=111, 71.6%) (Table 2). Most patients (n=84, 54.2%) had a personal history of breast and/or ovarian cancer. Most patients had a personal (n=115, 74.2%) and/or family history of cancer (n=109; 70.3%). The majority (n=110, 71.0%) of participants opted to proceed with genetic testing after counseling, where most consented to a multi-gene diagnostic test (n=96; 61.9%), while the remainder consented to a predictive test for a known familial pathogenic variant (n=14; 9.0%). Majority of our participants (n=79; 50.0%) received a negative or a variant of uncertain significance (VUS) genetic test result, others (n=21; 20.0%) received a positive genetic test result, while a minority (n=45; 29.0%) of participants declined genetic testing.

GCOS-24 Scores Pre- and Post-Intervention

Scores in both domains (CC and EC) (Supplementary Materials 3) increased following genetic counseling (Table 3). Overall post-intervention CC score [median 1.23, IQR (-0.33 - 6.16)] was significantly higher (p<0.001) than the pre-intervention score [median 0.46, IQR (-1.10 - 3.55)]. A similar significant trend was noted for overall post-intervention EC scores [median 0.99, IQR (-1.14 - 6.41)] versus pre-intervention scores [median 0.61, IQR (-1.14 - 3.90)].

In our item by item analysis for CC, several items showed substantial increases post-intervention. For example, item 1 “I am clear … why I am attending the clinical genetics service” [pre-intervention: median 0.56, IQR (-2.33 - 3.55); post-intervention: median 2.74, IQR (-1.01 - 6.16); p<0.001], item 7 “I can control how this condition affects my family”, item 10 “I don’t know what could be gained of the options (e.g. genetic testing) available to me”, [pre-intervention: median 0.36, IQR (-2.74 - 4.11); post-intervention: median 1.93, IQR (-2.74 - 6.16); p<0.001], and item 17 “I don’t know what I can do to change how to condition affects me / my children”, [pre-intervention: median 0.55, IQR (-2.55 - 4.30); post-intervention: median 2.12, IQR (-0.77 - 6.16); p<0.001] (Table 3) demonstrated the largest effect sizes.
The overall increase in scores for the EC domain was largely attributed to feeling more capable of coping with the condition post-counseling (item 9) [pre-intervention: median 0.98, IQR (-2.04 - 3.90); post-intervention: median 0.98, IQR (-2.04 - 6.41), p=0.046], being more positive about the future (item 20) [pre-intervention: median 0.90, IQR (-2.12 - 3.90); post-intervention: median 0.90, IQR (-2.12 – 6.41); p=0.024], and learning the positive aspects of having such a diagnosis (item 6) [pre-intervention: median 0.69, IQR (-2.33 – 3.90); post-intervention: median 0.69, IQR (-2.33 – 6.41); p=0.014]. However, it was notable that genetic counseling had little to no impact on participants’ feelings of being upset (item 4) and hopefulness for their children to have a rewarding family life (item 9).

Baseline factors associated with change in CC and EC scores

Compared to those without a family history of cancer, participants with a family history of cancer **were** significantly associated with a smaller increment in CC scores ($\beta$: 0, -0.56; CI: -0.99, -0.03; p value = 0.036) (Table 4). Females were significantly associated with a greater increment in EC scores than males ($\beta$: 0, -0.61; CI: -1.17, -0.05; p value =0.033) (Table 5). Compared to participants who did not proceed with genetic testing, those who received a negative or VUS result were significantly associated with a greater increment in CC scores ($\beta$: 0, 0.76; CI: 0.28, 1.24; p value =0.002) (Table 4) and EC scores ($\beta$: 0, 0.78; CI: 0.35, 1.21; p value <0.001) (Table 5). Similarly, participants who received a positive result were associated with greater increments in CC scores ($\beta$: 0, 0.81; CI: 0.21, 1.42; p value =0.009) (Table 4) and EC scores ($\beta$: 0, 0.64; CI: 0.10, 1.19; p value = 0.02) (Table 5) than participants who did not proceed with genetic testing. Of those who underwent genetic testing, the extent to which CC scores (Table 4) and EC scores (Table 5) increased were largely similar between participants who received positive results and those who received negative or VUS results.
DISCUSSION

Our study explored the impact of cancer genetic counseling provided by the NCCS CGS on patient empowerment using the culturally-adapted GCOS-24 instrument. We found a statistically significant increase in EC and CC scores following genetic counseling and testing (in patients who underwent genetic testing). These findings provide empirical evidence that genetic counseling provided by the CGS improves patient empowerment, thus highlighting its value in the delivery of genetics services in Singapore. Secondly, our psychometric analysis of the adapted GCOS-24 found that while the instrument as a whole was multidimensional, two key domains, namely CC and EC, were valid measures to assess the extent of patient empowerment arising from genetic counseling and testing.

Our study found that CC and EC were significantly improved post-genetic counseling and testing, and with the magnitude of improvement greater for the CC domain. These findings are concordant with recent systematic reviews of clinical genetics outcome research which have concluded that patients benefit from genetic counseling and testing, particularly in the areas of knowledge, ‘perceived personal control’ (PPC), improved risk perception accuracy, and reduced anxiety. Our findings were largely concordant with that of Tirado et al, who found that the overall GCOS-24 score improved post-counseling and testing, specifically the cognitive domain. This is consistent with our findings that patients were in a better position to establish control over their conditions, namely by managing how it affects their families. We also found that patients felt better equipped to navigate educational, financial and social resources available to consequentially make better autonomous decisions that are potentially life-altering for them and their descendants. Genetic counseling and testing was also observed to improve patients’ knowledge of what they could do to change the impact of the condition.

With a growing demand for cancer genetic services in Singapore and as the inclusion of genetic counsellors in patient care is increasingly found to be cost-effective, our study demonstrates that this model of care is beneficial for patients in the Asian context, where patients benefit from increased empowerment following genetic counseling and testing. Genetic counseling has been found to provide patients with a better knowledge of surveillance and risk-reducing options, which was subsequently
reported to empower patients in their decision-making regarding genetic testing by Augestad et al.\textsuperscript{32}

Notably, there were items pertaining to feelings of sadness and hopelessness in the EC domain in which no statistically significant improvement was reflected. This is similar to Tirado et al\textsuperscript{11} who highlighted a lack of significant improvement in the emotional regulation domain of the GCOS-24 (items 4, 11, and 21), which overlaps with the EC category defined here. These findings suggest a place for hope-based inventions, warranting research to understand how hope can be appropriately introduced during genetic counseling. Hope-based interventions, in the form of group therapy sessions where psychological questionnaires were administered, were found to be effective in allaying anxiety of patients with a predisposition to psychological conditions who were undergoing genetic testing.\textsuperscript{33} Hope-based interventions, focuses on prioritizing hope in patients and encourages \textit{goal-directed thinking},\textsuperscript{34} which enable recipients to achieve a higher dispositional hope. When achieved, patients benefit from greater psychological well-being, improved health knowledge, adoption of preventive health behaviors and adaptation to chronic illnesses. In the same vein, the reciprocal engagement model (REM) for genetic counseling provides a useful framework for the design of counseling strategies for the delivery of genetic results. These strategies have been proven to personalize the result communication and risk counseling process,\textsuperscript{35} which could be a way to improve emotional control in patients receiving genetic results. The incorporation of such interventions in genetic counseling practice may promote the delivery of holistic care, whilst presenting a systematic approach to instilling and improving emotional regulation in patients. Our findings highlight the growing importance of addressing emotional issues in genetic counseling. This is consistent with a review of genetic risk communication measures, which found emotional counseling elements to confer more benefit than informational elements.\textsuperscript{36}

In our study, higher empowerment levels were observed in patients who elected to proceed with genetic testing over patients who declined testing, suggesting that patients who underwent testing possessed a better understanding of their condition, as well as medical and non-medical resources available. Furthermore, they were also the group identified with higher emotional control levels, which meant they could cope better with new information that genetic testing provides them with. A better
understanding of the motivations and deterrents for genetic testing in at-risk patients is also warranted.

Rasch analysis was used to optimize the psychometric properties of the GCOS-24, which found that the scale was multidimensional in its overall form. Multidimensionality is problematic as patients respond differently to subsets of items and, if an overall score is used, true changes in sub-domains may be masked or neutralized, thus affecting the study conclusions. Therefore, we recommend that an overall score be avoided for the GCOS-24 and that separate CC and EC domain scores should be reported instead. Our findings demonstrate the importance of using Rasch analysis to verify and optimize the psychometric properties of PROMs in clinical research and our application of Rasch analysis to the GCOS-24 represents a useful contribution to clinicians and researchers hoping to measure patient-reported outcomes such as patient empowerment following genetic counseling. However, given our relatively small sample size in a culturally-diverse Asian population, further studies of similar design are required to confirm our findings. Recently, another PROM has been developed for the measurement of outcomes research related to risk communication in genetic counseling as part of the FOCUS-GC (Framework for Outcomes of Clinical Communication Services in Genetic Counseling). Further psychometric evaluation would be useful to determine if it is a useful PROM for measuring clinically significant changes in empowerment.

Strengths of our study include a cross-cultural adaptation of the GCOS-24 in an Asian population and our use of Rasch analysis to optimize the psychometric properties of the scale and enhance measurement precision and improve the robustness of our results; a well-characterized cohort with an equal distribution in age and a variety of cancers with suspicions for hereditary conditions, such as breast, ovarian, and colorectal cancers; inclusion of unaffected patients allowed us to measure genetic counseling-derived empowerment in individuals with a family history suggestive of a genetic condition.

There are several limitations to this study. Firstly, patient empowerment as operationalized in the GCOS-24 may not capture certain important patient reported outcomes that result from genetic counseling. This is complicated by the lack of consensus on tools reliable for such an assessment and what constitutes genetic
counseling-derived benefits.\textsuperscript{4} The study conducted in U.K. utilized the EQ-5D scale and an internal audit survey tool for comparison, while our study was limited to the GCOS-24. Without EQ-5D, the calculation of Quality Adjusted Life Years (QALY) delivered to patients seen by the CGS was unattainable, which demonstrates the limitation of the GCOS-24 for use in economic evaluation of a service. However, in a study comparing EQ-5D against the GCOS-24,\textsuperscript{11} EQ-5D was found to have problematic ceiling effects, with no detectable pre-post changes in scores, as it fails to capture patient outcomes of clinical genetics. Second, the post- GCOS-24 was administered relatively quickly after their genetic counseling session, which denied a longitudinal follow up of the patient’s emotional status (including that of hope), which might prove more effective in capturing patients with a reduced ability for emotional regulation. It would be meaningful to clarify if poor emotional control is attributed to pre-existing conditions or in fact exacerbated by genetic counseling. Studies have identified several risk factors that predispose patients to long-term post-testing distress, namely a pre-existing history of anxiety, depression, or psychiatric conditions,\textsuperscript{38-44} as well as pre-existing heightened cancer worry, elevated cancer risk perception, poor support networks, and an unfavorable test result. Voorwinden, Jaspers \textsuperscript{45} Screening for patients who demonstrate these prognostic variables for increased psychological distress from genetic testing, would allow for the personalization of a counseling program for them, thereby facilitating better psychological adaptation to their condition.

Third, neither the CC or EC domains achieved perfect fit to the Rasch model, both demonstrating some evidence of multidimensionality, while the EC domain demonstrated suboptimal precision. Therefore, the results should be interpreted with caution and future studies with larger sample sizes are required to confirm our domain structure.

**CONCLUSION**

In conclusion, our study revealed that patients who received cancer genetic counseling by trained genetics clinicians experienced a significant improvement in empowerment. However, more emphasis must be placed on cultivating hope and alleviating emotions of distress in patients during genetic counseling. Finally, our study demonstrated the utility of Rasch analysis in revealing multidimensionality of the GCOS-24, for which
scores for cognitive control and emotional regulation should be reported separately.
**Authorship Contributions**

JY was responsible for design, acquisition, analysis and interpretation of data, as well as drafting of the manuscript for publication.

SYL was involved in the analysis and interpretation of the data as well as drafting of the manuscript for publication.

EC was involved in the analysis and interpretation of the data, and critically reviewed the manuscript for publication.

JL was involved in the analysis of the data and critically reviewed the manuscript for publication.

HS was involved in the acquisition of the data and critically reviewed the manuscript for publication.

STL was involved in the acquisition of the data and critically reviewed the manuscript for publication.

YC was involved in the acquisition of the data and critically reviewed the manuscript for publication.

MM was involved in the conception and design of the study, and critically reviewed the manuscript for publication.

EKF was involved in the design and data analysis of the study, and critically reviewed the manuscript for publication.

JN was responsible for the conception, design and progress of the study, critically reviewed the study at the stages of data acquisition, interpretation and manuscript drafting.
Ethics Approval

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2000 (5). This study was approved by the SingHealth Centralised Institutional Review Board (CIRB number 2016/2367).

Patient Consent and Confidentiality

Informed consent was obtained from all patients included in the study.
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