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ORIGINAL ARTICLE

Cross-cultural validation of the genetic counseling outcome scale in Korea

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Abstract

The Genetic Counseling Outcome Scale (GCOS-24) was developed to measure patient-reported outcomes to evaluate the effectiveness of genetic counseling and testing services. In the current study, the Korean version of GCOS (K-GCOS) was developed to reflect the sociocultural characteristics of Korea, and its clinical applicability was assessed. Overall, 231 Koreans, including patients with genetic diseases and their family members, participated and completed the K-GCOS, Hospital Anxiety and Depression Scale (HADS), Multidimensional Health Locus of Control (MHLC) scale, and Satisfaction with Life Scale (SWLS). Validity was examined by assessing the correlations between K-GCOS scores and other relevant scale scores. Reliability was confirmed using Cronbach's alpha and test–retest scores, measured over 2 weeks. We performed exploratory factor analysis of the five structures of GCOS-24. For K-GCOS, four-factor structures were identified: "cognitive–behavioral control," "uncertainty about control," "hope," and "emotional regulation." Four original GCOS-24 items were removed because of low factor loadings and small inter-item correlations. K-GCOS-20 scores were positively correlated with SWLS ($r = 0.456$) and MHLC-internal ($r = 0.213$) scores but negatively correlated with HADS (anxiety $r = -0.428$, depression $r = -0.469$) and MHLC-internal ($r = -0.278$) scores. These findings demonstrate that K-GCOS-20 is a reliable and valid tool for evaluating genetic counseling services in Korea.

KEYWORDS
empowerment, genetic counseling, genetic counselors, genetics services

1 | INTRODUCTION

In Korea, genetic counseling and testing services are primarily provided by tertiary general hospitals. Notably, although over 50,000 patients with rare diseases are newly registered in the country every year, 79.8% of them do not receive genetic counseling services (Choi et al., 2022). Therefore, the government is trying to introduce a genetics team involving medical geneticists and certified genetic counselors to facilitate the provision of genetic counseling services for patients with rare genetic diseases. However, for genetic counseling

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services to be recognized as an official medical practice in the health care system of Korea, it is necessary to evaluate whether these services can improve outcomes of the affected patients and their at-risk relatives. Genetic counseling improves patient satisfaction by improving knowledge, risk perception, and autonomy of patients while reducing the experience of stigmatization (McAllister & Dearing, 2015).

According to the U.S. Food and Drug Administration guidelines, patient-reported outcome (PRO) is defined as patients' self-reported outcomes (i.e., without interpretation by clinicians or third parties) based on their medical consultations or treatments. PROs are reported in terms of before–after changes in status, which distinguishes PROs from satisfaction (U.S. Department of Health and Human Services et al., 2006). PROs play a critical role in facilitating informed decision-making between patients and physicians regarding surgical procedures or clinical pathways and enabling comparison of individual patient outcomes, allowing evaluation of the effective allocation of medical resources (Choi et al., 2019). The active involvement of patients and their families in health care decision-making processes is a key factor in improving health care quality and performance evaluation (Choi et al., 2019).

The Genetic Counseling Outcome Scale (GCOS-24) can evaluate empowerment as an outcome of clinical genetic services (McAllister, Wood, et al., 2011). GCOS-24 can be used to evaluate the benefits of genetic counseling valued by patients and clinical genetic service providers, specifically the empowerment of counselors. Empowerment is 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" (McAllister, Wood, et al., 2011). This concept is pivotal in the health care domain, involving the acquisition of skills such as decision-making autonomy, utilization of support systems, and stress management, rather than passively adhering to expert guidance (Anderson & Funnell, 2005; Feste & Anderson, 1995; Gibson, 1991). As a patient-reported outcome measure (PROM), the GCOS-24 is designed to capture five domains: can make important life decisions (decisional control), has sufficient information about their family's condition (cognitive control), can manage one's feelings (behavioral control), can make effective use of the health care system (emotional regulation), and has hope for the future (hope) (McAllister, Wood, et al., 2011). In previous studies, this tool has been adapted and translated into various languages, including Danish, Spanish, Dutch, and Brazilian Portuguese, and its reliability and validity have been tested or observed (Diness et al., 2017; Muñoz-Cabello et al., 2018; Segundo-Ribeiro et al., 2020; Voorwinden et al., 2019). Moreover, it has been used to evaluate genetic counseling services for a range of conditions, including autism spectrum disorder, cardiovascular disease, psychiatric conditions, and cancer (Gerrard et al., 2020; Inglis et al., 2015; Ison et al., 2019; Yuen et al., 2020; Yusuf et al., 2021). Notably, GCOS-24 has good psychometric properties in different clinical settings, and its value as a PROM is recognized internationally.

This study developed a Korean version of the Genetic Counseling Outcome Scale (K-GCOS) that reflects Korean sociocultural characteristics. Additionally, we assessed the psychometric properties of the K-GCOS, including factor structure, reliability, internal consistency, and structural validity.

What is known about this topic

The Genetic Counseling Outcome Scale (GCOS-24) is used in various countries as a patient-reported outcome measure (PROM) for clinical genetic services. Although the GCOS-24 is an important tool for measuring patient empowerment, it has not yet been introduced in Korea.

What this paper adds to the topic

K-GCOS reflects the sociocultural characteristics of Korea. This study confirmed the reliability and validity of K-GCOS and may be used as basic data for assessing and improving Korean genetic counseling services.

2 | MATERIALS AND METHODS

2.1 | Study design

The current study was conducted in two phases: (1) Translation and cross-cultural adaptation and (2) assessment of the reliability and validity of K-GCOS.

2.2 | Phase 1: Translation and cross-cultural adaptation

2.2.1 | Translation

This study was conducted according to the International Society for Pharmacoeconomics and Outcomes Research (ISPOR) process for tool translation and cultural fit testing (Wild et al., 2005), which included forward translation, back translation, expert consultation, and pilot study, leading to the final version completion phase. The forward translation (English to Korean) of GCOS-24 was independently performed by three native English speakers who were bilingual in Korean and English. Any discrepancies between the translations were resolved via discussion by an interdisciplinary expert committee composed of seven experienced researchers (including three nursing professors, one professor of clinical research design and evaluation, one professor of genetic counseling, one professor of Korean language and literature, and one professor of gynecology and obstetrics). The resulting Korean version of the GCOS-24 was back-translated into English by an English first-language translator naive to the outcome measurement. Finally, the back-translated version of the questionnaire

was carefully reviewed by the research team and confirmed by the original author to finalize the draft items. The original English and Korean versions (forward and back translations) of the GCOS-24 are shown in Data S1 and S2, respectively.

2.2.2 | Content validation by experts

Draft items for K-GCOS derived via translation and back translation processes underwent content validation by experts. Notably, this expert group comprised 10 professionals, including one medical geneticist, two certified genetic counselors, one nursing professor, one professor of clinical research design and evaluation, one professor of gynecology and obstetrics, two physicians with ≥15 years of experience, one medical social worker, and one psychology professor. After the purpose of the study and item development processes were explained to the experts, their written consent was obtained. The panel of experts was instrumental in scoring the entire questionnaire and each item for relevancy, clarity, and cultural adaptability using a 4-point Likert scale (1 = strongly disagree, 2 = must be modified or disagree, 3 = agree after minor modification, and 4 = strongly agree).

2.2.3 | Pilot studies

The pilot study and cognitive interviews were conducted from March to April 2022, involving patients affected by or at risk of developing rare genetic diseases or hereditary cancer, as well as their families, from the medical genetics centers of tertiary general hospitals in the Seoul area and a cancer center in the Gyeonggi-do area. Participants were recruited using convenience sampling, and the inclusion criteria were the ability to read and express thoughts in Korean and aged ≥18 years.

The pilot study was conducted with 30 patients or their family members to evaluate the suitability of the questionnaire completed through translation and back translation. The pilot study assessed problems such as comprehension of questions, length and layout of the questionnaire, intended meaning of the questions, and time required for questionnaire completion.

Subsequently, cognitive interviews were conducted to assess whether discrepancies existed between the intended meaning of each item in the tool and respondent's interpretation of the items (Irwin et al., 2009). The first author conducted individual, face-to-face, semi-structured interviews with 15 participants diagnosed with or at risk of developing rare genetic diseases or hereditary cancer. Written informed consent was obtained from all participants, and cognitive interviews were conducted for 20 min to 1 h and 30 min for each patient. Cognitive interviews were conducted using a hybrid method of think aloud and verbal probing (Collins, 2003; Willis, 2005; Willis & Artino Jr., 2013), and the interviews were recorded and transcribed. Subsequently, the researchers qualitatively analyzed the participants' responses through thematic analysis to confirm the linguistic validity and acceptability of the questions (Collins, 2003). Unclear or difficult-to-understand words and sentences were then discussed among the expert committee members, and the final version was approved by consensus.

2.3 | Phase 2: Validation study

2.3.1 | Participants and data collection

This study recruited patients diagnosed with rare genetic diseases or hereditary cancer and their families who attended the medical genetics centers of tertiary general hospitals in the Seoul area and a cancer center in the Gyeonggi-do area between October and November 2022. Patients diagnosed with rare genetic diseases or hereditary cancers via genetic or chromosome testing were defined as those diagnosed with rare genetic disease or hereditary cancer. The inclusion criteria were as follows: patients with rare genetic conditions or relatives (parents, grandparents, and siblings); aged >18 years; and patients able to communicate, understand, and respond in Korean.

To assess test–retest reliability, internal consistent reliability, structural validity, and convergent/divergent validity, patients were recruited to complete a battery of questionnaires, including K-GCOS, K-HADS, K-MHLC, and K-SWLS. For test–retest reliability, 40 participants from the total validation study were asked to complete K-GCOS 14 days later. During the 14-day period, participants were not provided with any significant interventions likely to change their empowerment levels, such as new information, new developments regarding their genetic condition, or test results. All collected data were anonymized.

2.3.2 | Instruments

Korean version of the hospital anxiety and depression scale (K-HADS)

HADS—a self-reporting questionnaire—comprises 14 items, with seven items specifically addressing anxiety and seven focusing on depression (Zigmond & Snaith, 1983). Anxiety and depression levels are evaluated on a 4-point Likert scale. In this study, the Korean version of HADS was used after receiving approval for its use from GL Assessment, UK.

Korean version of the multidimensional health locus of control scale (K-MHLC)

The locus of control refers to expectations or beliefs about outcomes or reinforcement related to health. Notably, the MHLC assesses an individual's ability to control their own health and comprises three subfactors: internal factors, external factors, and chance (Wallston et al., 1978). The Korean version (K-MHLC) contains 17 items that are evaluated on a 6-point Likert scale (1 = Strongly disagree; 6 = Strongly agree) (Na, 1999).

Korean version of the satisfaction with life scale (K-SWLS)

SWLS is a tool used to measure the cognitive aspects of satisfaction with life (Diener et al., 1985). The Korean version (K-SWLS) comprises five items that are evaluated on a 7-point Likert scale (1 = Strongly disagree; 7 = Strongly agree) (Cho & Cha, 1998).

2.4 | Statistical analyses

SPSS WIN 26.0 (IBM Corp., Armonk, NY, USA) program was used to analyze the data obtained in this study. The general and disease characteristics of the participants were analyzed using descriptive statistical methods. Content validity was calculated using the scale CVI (S-CVI) and item CVI (I-CVI). The I-CVI was determined by dividing the number of experts with scores of 3 or 4 by the total number of participating experts. Similarly, S-CVI/Ave was calculated as the mean I-CVI for all scale entries. Generally, I-CVI ≥ 0.8 and S-CVI/Ave ≥ 0.9 were considered acceptable (Polit et al., 2007).

Exploratory factor analysis (EFA) was performed to evaluate construct validity. Kaiser–Meyer–Olkin values were calculated, and Bartlett's sphericity tests were performed to determine the feasibility of EFA for verifying construct validity. The varimax rotation method was used as the factor rotation method, and the factor loading standard was set at ≥0.40. The number of factors was determined by applying an eigenvalue >1.0 and a scree plot.

We tested the following hypotheses:

1. Empowerment is positively correlated with satisfaction with life
2. Empowerment is inversely correlated with (i) anxiety and (ii) depression
3. Empowerment is not correlated with the health locus of control

To test these hypotheses and assess their convergent validity, we used Pearson's correlation coefficients between K-GCOS and HADS, MHLC, and SWLS. Finally, internal consistency reliability was assessed by calculating Cronbach's alpha, and test–retest reliability was assessed by calculating the interclass correlation coefficients (ICCs) between the responses to the K-GCOS questionnaire consecutively administered, following a 2–4-week period during which levels of empowerment were not expected to change.

2.5 | Ethical considerations

Approval to conduct this research on human participants was obtained from the Institutional Review Board of the National Cancer Center on April 1, 2022, and Asan Medical Center on December 27, 2021 (IRB number: 2022-1250, NCC2022-0287). All procedures were conducted in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and the Helsinki Declaration of 1975, as revised in 2000. Informed consent was obtained from all patients prior to their inclusion in the study.

3 | RESULTS

3.1 | Translation and cross-cultural adaptation

During the translation and cultural adaptation process of GCOS-24, any discrepancies resulting from the English to Korean translation were discussed among expert committee members and resolved through consensus. They identified questions requiring modification because of cultural differences and discussed the meanings of the words "genetic condition," "clinical genetic service," "concerns," and "upset." Each item was adapted for culturally appropriate wording without altering the context.

We conducted a pilot study with 30 participants. The mean age of the participants was 41.5 (interquartile range [IQR] = 37–48, SD = ±10.87) years, and they had different places of residence and levels of education and employment status. The demographic and clinical characteristics of the participants are presented in Data S3. The questionnaire took an average of 10 min to complete, and it was generally well accepted and considered easy to understand. In the pilot studies, Questions 6, 7, and 15, which respondents found challenging, were rechecked, and modifications were made to the Korean expressions. Specifically, Question 6, originally phrased as "I can see that good things have come from having this condition in my family," was amended to "I think that my family's genetic condition offers advantages as well." Question 7, which initially stated, "how this condition affects my family," was revised to "the effects of my genetic condition on my family." Regarding Question 15, the content, initially "educational, financial, social support," was adjusted to "educational, economic, and social support, etc." Twenty-nine participants, excluding one, evaluated the scale answer categories (strongly disagree = 1; disagree = 2; slightly disagree = 3; neither disagree nor agree = 4; slightly agree = 5; agree = 6; and strongly agree = 7) as appropriate. The panel of expert committee members agreed to maintain the 7-point scale.

For cultural adaptation, we conducted cognitive interviews involving 15 participants with a mean age of 42 (IQR = 32–58, SD = 13.17) years. The interviews included five patients and 10 family members, and the interviews were conducted targeting various diseases. We identified the content aspects and level of understanding of these questions. The demographic and clinical characteristics of the participants in the cognitive interview and the cognitive interview analysis data are presented in Data S4. Specifically, the participants expressed difficulty understanding the term "genetic condition," and after discussion, the expert committee members agreed to add an explanation at the bottom of the questionnaire: "*A genetic condition refers to a condition in which a disease has occurred or is likely to occur in me or my family due to genetic causes (chromosomal or pathogenic variant)." All items from the original GCOS-24 were retained in the K-GCOS-24 without excluding any items.

3.2 | Validation study

3.2.1 | Characteristics of participants

Overall, 231 participants (164 women and 67 men), including patients with rare genetic diseases and their family members, were enrolled in this study (Table 1). The mean age of the participants was 42.0 (IQR = 35.0–47.0; SD = ±11.59) years, and 98 patients (42%), 94 mothers (41%), and 28 fathers (12%) were included. The patients were affected by hereditary cancer syndrome (101 participants, 43.7%), metabolic disorders (45 participants, 19.5%), or chromosomal anomalies (32 participants, 13.9%).

TABLE 1 Demographic and clinical characteristics of the participants ($n = 231$).

Characteristics	Categories	*n* (%)
Gender	Men	67 (29.0)
	Women	164 (71.0)
Age (years)	<20	1 (0.4)
	20–30	31 (13.4)
	30–40	64 (27.7)
	40–50	84 (36.4)
	50–60	31 (13.4)
	>60	20 (8.7)
Relationship with the patient	Patient themselves	98 (42.4)
	Mother	94 (40.7)
	Father	28 (12.1)
	Others (spouse, child, or siblings)	11 (4.7)
Marital status	Unmarried	52 (22.5)
	Married	171 (74.0)
	Divorced/separated	7 (3.0)
	Bereaved	1 (0.4)
Educational attainment	Elementary-school graduation	5 (2.1)
	Middle-school graduation	3 (1.3)
	High-school graduation	49 (21.2)
	Higher than college graduation	174 (75.3)
Number of patients in the family	1 person	177 (76.6)
	2 people	29 (12.6)
	3 people	17 (7.3)
	≥4	8 (3.4)
Type of diseases	Hereditary cancer syndrome	101 (43.7)
	Metabolic disorder	45 (19.5)
	Chromosomal anomalies	32 (13.9)
	Dysmorphic and congenital abnormality syndromes	22 (9.5)
	Cardiovascular disorders	19 (8.2)
	Neurology and neurodevelopmental disorders	8 (3.5)
	Others	4 (1.7)
Time elapsed since the diagnosis	<6 months	32 (13.9)
	6 months–1 year	14 (6.1)
	1–3 years	47 (20.3)
	3–10 years	82 (35.5)
	>10 years	56 (24.2)

3.2.2 | Reliability and validity of K-GCOS

Content validity

Ten experts assessed the content validity of K-GCOS, and the I-CVI for each item was found to be ≥0.80. The S-CVI was found to be 0.92. These results indicate that the experts confirmed the relevance and clarity of K-GCOS.

Construct validity

Using the data from the initial 24 items, EFA with varimax rotation was performed. The suitability of the data was verified using the Kaiser–Meyer–Olkin test result of 0.814 and Bartlett's sphericity test result of $\chi^2 = 1960.677$ ($p < 0.001$). Scree plot examination and EFA revealed multiple dimensions, indicating four or six primary factors. Using an iterative process, we explored four to six factor solutions. We established a stringent loading criterion of 0.40 and reviewed items for redundancy and phrasing. Using this iterative process, four items were removed. Notably, Item 24 had a multifactor loading of >0.20 for two factors (Factor 1 with 0.569 and Factor 5 with 0.412); given that item extraction should be based on a difference in interfactor loading of >0.20 (Kline, 1994), this item was excluded first. Moreover, Item 10 was excluded because it was loaded solely on Factor 6, and items 15 and 7 were excluded because of their low internal consistency (Cronbach's alpha coefficient = 0.51) despite being classified as Factor 5. Item 19 was extracted as a single factor; however, its factor loading was high for two factors, indicating that its content was more appropriate for the hope aspect related to Factor 3. In addition, Item 19 was not considered suitable as a single factor. Thus, it was classified as Factor 3. Finally, the 20 selected items were refactored following the same process, and a four-factor solution that best matched our underlying conceptualization was created.

Factor 1 was named "cognitive–behavioral control" because the responses "I know," "I understand~," and "I am able to explain~" were repeated. Factor 2 was named "uncertainty about control" because the uncertainties "I don't know~" and "I am not sure~" were evident. Factor 3 included items 6, 8, 19, 20, and 9 (ability to cope with family situation in the genetic context), which were classified as "hope" in the original instrument. All items, including Item 9, were labeled "hope" in the context of thinking positively about the future and looking forward to family life. Factor 4 was named "emotional regulation," which indicates that people can regulate their emotions related to the genetic status of their family members. These four factors had the following eigenvalues: cognitive–behavioral control, 4.83; uncertainty about control, 3.18; hope, 1.96; and emotional regulation, 1.28. These factors accounted for 16.20%, 14.37%, 20.44%, and 10.91% of the variance, respectively (total variance = 61.92%). Table 2 provides details of the abovementioned 20 items and 4 factors. The final form of the Korean version of the GCOS is presented in Table 2.

TABLE 2 Items in Genetic Counseling Outcome Scale in terms of the factors.

Item description (subscales)	Factor 1	2	3	4
Factor 1: Cognitive–behavioral control (6 items)				
1. I clearly know why I am receiving genetic condition counseling at the hospital	0.832			
2. I am able to explain the meaning of my genetic condition to anyone in the family who needs to know	0.760			
14. I understand why my doctor recommended genetic counseling to me	0.668			
23. I understand the concerns that led me to genetic counseling	0.664			
3. I know how my genetic condition may affect my children or the children I may have in future	0.621			
16. I am able to explain the implications of my genetic condition to nonfamily members who need to know (example: teachers and social workers)	0.617			
Factor 2: Uncertainty about control (6 items)				
18. I do not know who else in the family is likely to have the same genetic condition as me		0.754		
17. I do not know what I can do to change the effect that my genetic condition has on me and my children		0.710		
12. I am not sure how this genetic condition will affect my brothers, sisters or relatives (aunts, uncles, and cousins)		0.702		
22. I feel powerless that I am unable to do anything about my family's genetic condition		0.593		
13. Whatever decisions I make regarding my family's genetic condition will not change the future of my children or the children I may have in the future		0.589		
5. I do not know where to seek medical help for myself and for my family		0.529		
Factor 3: Hope (5 items)				
8. I think positively about the future			0.812	
20. I am able to plan for the future even with my genetic condition			0.748	
9. I am able to cope with my family situations with my genetic condition			0.744	
6. I think that my family's genetic condition offers advantages as well			0.658	
19. I hope that my children will be able to have a happy family life despite their genetic condition			0.406	
Factor 4: Emotional regulation (3 items)				
4. I get upset when I think about my family's genetic condition.				0.841
11. I am anxious that my family has this genetic condition				0.691
21. I feel guilty that I may pass on my genetic condition to my children				0.539
Removed items				
7. I think it is possible to control the effects of my genetic condition on my family				
10. Given my genetic condition, I do not know how I can benefit from the options (treatment and screening, etc.)				
15. I know how to get nonmedical help for myself and for my family (e.g., educational, economic, and social support, etc.)				
24. I believe I can make my own decisions about my genetic condition, which can change the future of my children or the children I may have in the future				

Convergent validity

We investigated the correlation between GCOS empowerment and its associated variables (i.e., anxiety, depression, quality of life, and health locus of control) (Table 3). The results revealed a significant negative correlation with anxiety ($r=-0.428$, $p<0.001$), depression ($r=-0.469$, $p<0.001$), and chance health locus of control ($r=-0.278$, $p<0.001$), whereas there was a significant positive correlation with life satisfaction ($r=0.456$, $p<0.001$) and internal health locus of control ($r=0.213$, $p<0.05$).

Reliability

Internal consistency and test–retest reliability for the K-GCOS scale are presented in Table 4. The Cronbach's alpha coefficient for the total scale was found to be 0.82, with each subfactor ranging between 0.69 and 0.80, indicating good internal consistency (cognitive–behavioral control = 0.80, uncertainty about control = 0.78, hope = 0.79, and emotional regulation = 0.69). Furthermore, to evaluate test–retest reliability, a survey of 40 participants was conducted 2 weeks after the initial assessment. The ICC ranged from 0.66 to 0.81, indicating temporal stability.

4 | DISCUSSION

This study verified the use of K-GCOS to evaluate the outcomes of genetic counseling services for patients with rare genetic diseases

and their families in Korea. To ensure the suitability of the tool for the Korean population, we performed translation and cross-cultural adaptation of GCOS into Korean using recognized international guidelines for patients with various diseases and their families, including those with chromosomal abnormalities, cardiovascular diseases, dysmorphic and congenital abnormality syndromes, hereditary cancer syndrome, metabolic disorders, and neurodevelopmental disorders.

The content validity of the Korean version was found to be excellent, indicating that all items of this tool are relevant for measuring the outcomes of genetic counseling. Moreover, the tool showed satisfactory psychometric characteristics, including reliability, construct validity, and convergent validity.

Content validity was considered appropriate when the I-CVI was ≥0.80 and S-CVI was ≥0.90 (Polit et al., 2007). In this study, the I-CVI and S-CVI were >0.80 and 0.92, respectively, indicating that the experts confirmed the relevance and clarity of K-GCOS.

The developers of the scale, McAllister et al. revealed that GCOS-24 has a 5-factor structure, which was determined through maximum likelihood EFA by promax rotation (McAllister, Dunn, & Todd, 2011; McAllister, Wood, et al., 2011). In this study, we performed EFA to investigate the construct validity of K-GCOS. Twenty items were extracted, leading to the formation of four factors (cognitive–behavioral control, uncertainty about control, hope, and emotional regulation), in contrast to the five-factor structure of the original GCOS-24. In particular, "uncertainty about control" included reverse coding items among the items of "cognitive control" and "behavioral control" in the original tool, and other items were included in "cognitive–behavioral control." "Hope" included the original tool items and Item 9 "I am able to cope with my family situations with my genetic condition", and "emotional regulation" was the same as the items of the original tool. The factor structure and number of items of K-GCOS differ from the original GCOS-24, likely because of the influence of the genetic counseling environment in Korea and Korean cultural factors. While verifying the cultural adaptation, reliability, and validity of GCOS, different factor structures and the number of items were also reported in the Netherlands and Spain. The Dutch and Spanish versions of GCOS-18 and GCOS-21 included six subfactors. Among the four factors identified in this study, two showed consistency with those in other versions. Notably, the factor "cognitive–behavioral control" aligned with the factors "Knowledge about condition" and "Knowledge about genetic services" from the Dutch version and the factors "Referral clarity" and "cognitive control" from the Spanish version.

The factor "uncertainty about control" (Items 18, 17, 12, 22, 13, and 5) in our study partially aligned with the factors "uncertainty about genetics" (Items 18 and 12) and "uncertainty about the treatment" (Items 10, 17, and 5) from the Dutch version. Item 10 "How to benefit from options when considering your genetic status" was excluded from our model and the Spanish version. Additionally, the factors "family impact" (Items 12, 18, and 21) and "decisional and behavioral control" (Items 24, 13, 17, 22, and 15) from the Spanish version partially aligned with the factor "uncertainty about control" in our study. In our model, items 24 "I believe I can make my own decisions about my genetic condition, which can change the future of my children or the children I may have in the future" and 15 "I know how to get nonmedical help for myself and for my family" were excluded, and they were also excluded in the Dutch version. Additionally, Item 21 "I feel guilty that I may pass on my genetic condition to my children" was included in the "emotional regulation" factor in our model as well as in the "negative emotions" factor in the Dutch version. Factor analysis of GCOS has low consistency across different languages (McAllister, Dunn, & Todd, 2011; McAllister, Wood, et al., 2011; Muñoz-Cabello et al., 2018; Voorwinden et al., 2019). This finding can be attributed to differences in the context of genetic counseling services and sample characteristics among countries as well as the effects of cultural factors.

TABLE 3 Convergent validity of the Korean version of the Genetic Counseling Outcome Scale.

Validation tool	Correlation	Significance probability (two-tailed)
HADS		
Anxiety	−0.428**	<0.0001
Depression	−0.469**	<0.0001
SWLS	0.456**	<0.0001
MHLC		
Internal	0.213*	<0.0001
Chance	−0.278**	<0.0001
Other	0.098	0.139

Abbreviations: HADS, hospital anxiety and depression scale; MHLC, multidimensional health locus of control scales; SWLS, satisfaction with life scale.

* $p < 0.05$ and ** $p < 0.01$.

TABLE 4 Reliability measured using the internal consistency of items.

	Internal consistency ($n = 231$)		Test–retest reliability ($n = 40$)	
Subscale	Number of items	Cronbach's α	Intraclass correlation coefficient	p-value
Cognitive–behavioral control	6	0.80	0.74	<0.001
Uncertainty about control	6	0.78	0.74	<0.001
Hope	5	0.79	0.81	<0.001
Emotional regulation	3	0.69	0.66	<0.001

The convergent validity of K-GCOS was evaluated by analyzing its correlation with empowerment-related factors, including anxiety, depression, quality of life, and health locus of control. According to previous studies, the original GCOS-24 showed positive correlations between empowerment and SWLS and internal MHLC, while showing negative correlations with anxiety, depression, and chance MHLC but no significant correlations with other MHLCs (McAllister, Wood, et al., 2011). The Spanish version of GCOS-24 showed positive correlations with SWLS and negative correlations with anxiety but no significant correlations with MHLC (Muñoz-Cabello et al., 2018). The Dutch version of GCOS-18 found negative correlations with anxiety and positive correlations with positive effect (Voorwinden et al., 2019). Similarly, this study found that empowerment experienced by patients with genetic diseases and their families through genetic counseling was positively correlated with SWLS and internal MHLC, whereas it was negatively correlated with the State–Trait Anxiety Inventory and chance MHLC. Furthermore, similar to GCOS-24, it did not show significant correlation with other MHLC scales (McAllister, Wood, et al., 2011). These results suggested a mutual correlation between empowerment and psychological variables, indicating that the provision of genetic counseling services for patients with genetic diseases and their families positively affected their quality of life (Ashtiani et al., 2014; Ison et al., 2019; McAllister & Dearing, 2015). Unexpectedly, empowerment was positively correlated with internal MHLC, indicating that a sense of health control is related to empowerment. Conversely, empowerment was negatively correlated with chance MHLC, indicating that beliefs in external factors controlling health are associated with lower empowerment (Muñoz-Cabello et al., 2018; Voorwinden et al., 2019). These differences could be due to variations in study populations, cultural differences, and differences in measurement tools, providing important insights into the complex interactions between various psychological constructs related to empowerment.

Regarding reliability, Cronbach's alpha coefficients were found to be 0.87, 0.79, 0.84, 0.71, and 0.77 for the original version GCOS-24, Denmark version of GCOS, Spain version of GCOS-21, Brazilian Portuguese version of GCOS-24, and Dutch version of GCOS-18, respectively (Diness et al., 2017; McAllister, Dunn, & Todd, 2011; McAllister, Wood, et al., 2011; Muñoz-Cabello et al., 2018; Segundo-Ribeiro et al., 2020; Voorwinden et al., 2019). In this study, Cronbach's alpha coefficient was 0.82, indicating that the scale had good internal consistency. In particular, the subscales "cognitive–behavioral control" (Cronbach's $\alpha = 0.80$), "uncertainty about control" (Cronbach's $\alpha = 0.78$), and "hope" (Cronbach's $\alpha = 0.79$) showed satisfactory internal consistency. However, "emotional regulation" (Cronbach's $\alpha = 0.69$) showed lower consistency than the other subscales, indicating that this subscale should be interpreted with caution. "Emotional regulation" showed slightly lower figures than the other factors because of the participant's response bias as the subfactors all consist of negative statements (Horan et al., 2003). While many researchers are seeking ways to reduce the participant's response bias for questionnaire configuration, this may also degrade the reliability of the tool at the same time. Nevertheless, the overall Cronbach's α value of 0.82 is within an acceptable range, supporting the internal consistency of the scale (DeVellis, 2017).

Furthermore, the test–retest reliability of K-GCOS was good. Consistent results were noted over a 2-week period, confirming the stable measurement of genetic counseling outcomes. According to the established criteria, the ICCs of the original version GCOS-24 (0.86) and Dutch version (0.75–0.92) were of reliable level, whereas the ICC of the Brazilian Portuguese version (0.52) was moderate (Landis & Koch, 1977; McAllister, Dunn, & Todd, 2011; McAllister, Wood, et al., 2011; Segundo-Ribeiro et al., 2020; Voorwinden et al., 2019). In this study, the total ICC of K-GCOS was found to be 0.66–0.81, and the correlations among scores were significant for all dimensions ($p < 0.01$). These results indicate that the psychometric properties of the scale are adequate in terms of internal consistency and stability over time. Therefore, K-GCOS is considered a tool with good validity and reliability for measuring genetic counseling outcomes.

This study has some limitations. First, convenience sampling was used to select study participants from two tertiary general hospitals; thus, the study sample may not represent all patients with genetic diseases and their families in Korea. Second, in this study, we could not conduct a confirmatory factor analysis to validate the results of the EFA, which revealed a four-factor structure for K-GCOS. Conventional practice recommends using separate samples for CFA and EFA to avoid potential biases (Fabrigar et al., 1999). However, owing to the limited sample size, it was impossible to verify the goodness-of-fit index and the extracted mean-variance and construct reliability through CFA after classifying the factors using EFA (Anderson & Gerbing, 1988; Hinkin, 1998). Furthermore, the factors of our study were extracted differently from those of GCOS-24. GCOS-24 was subjected to higher-order factor analysis in the initial validation study, which indicated that all five factors existed under the higher-order factor "empowerment," and the authors recommended using GCOS-24 as a one-dimensional scale (McAllister, Wood, et al., 2011). Further replication studies with larger sample sizes are warranted to test validity through various methods, including confirmatory factor analysis. Furthermore, the "emotional regulation" factor exhibited slightly lower internal consistency (Cronbach's $\alpha = 0.69$) than the other factors. Therefore, future research should consider validating this factor structure through CFA and exploring potential enhancements in internal consistency by adjusting or removing items. Finally, there was a limitation in the measurement of criterion validity, as no tool was available for assessing empowerment for patients with genetic diseases and their families in Korea. Further studies are needed to develop an empowerment intervention program by investigating the differences in empowerment before and after genetic counseling and by identifying influential factors related to the empowerment capabilities of patients with genetic diseases and their families.

In conclusion, K-GCOS is a clear, easy-to-understand, reliable, and valid PROM that can be used to assess genetic counseling outcomes in the Republic of Korea. In Korea, opinions have been

expressed regarding the importance of genetic counseling and the need for genetic counselors; however, no studies have evaluated the outcomes of genetic counseling. If genetic counseling in the clinical setting is strengthened through empowerment interventions based on the comprehensive understanding of patients with genetic diseases and their families, it will help these patients and their families to improve their understanding ability, leading to an improvement in their quality of life. In addition, K-GCOS can provide important data that can be used to design improvements to the quality of genetic counseling services and support the integration of quality genetic counseling services into the Korean health care system.

AUTHOR CONTRIBUTIONS

Sojin Yang: Conceptualization; data curation; formal analysis; investigation; methodology; validation; visualization; writing – original draft. **Yoon Jung Chang:** Conceptualization; formal analysis; funding acquisition; methodology; project administration; resources; supervision; validation; writing – review & editing. **Kyung Ok Kim:** Conceptualization; formal analysis; methodology; validation; writing – review & editing. **Beom Hee Lee:** Resources; writing – review & editing. **Sun-Young Kong:** Resources; writing – review & editing. **Marion McAllister:** Conceptualization; methodology; validation; writing – review & editing. **In Hee Choi:** Conceptualization; formal analysis; methodology; project administration; supervision; validation; visualization; writing – original draft; writing – review & editing. In Hee Choi confirms that they had complete access to all data in the study and take responsibility for the integrity of the data and accuracy of data analysis. All authors gave final approval for the publication of this version and agreed to be responsible for all aspects of the work to ensure that the questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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CONFLICT OF INTEREST STATEMENT

All authors declare that they have no conflicts of interest.

DATA AVAILABILITY STATEMENT

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

ETHICS STATEMENT

Human Studies and Informed Consent: Approval to conduct this research on human participants was obtained from the Institutional Review Board at National Cancer Center and Asan Medical Center (IRB number: 2022-1250, NCC2022-0287) approved on April 1, 2022 and December 27, 2021 respectively. All procedures were conducted 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. Informed consent was obtained from all patients for inclusion in this study.

Animal Studies: No non-human animal studies were carried out by the authors for this article.

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REFERENCES

Anderson, J. C., & Gerbing, D. W. (1988). Structural equation modeling in practice: A review and recommended two-step approach. *Psychological Bulletin*, *103*(3), 411–423. https://doi.org/10.1037/0033-2909.103.3.411

Anderson, R. M., & Funnell, M. M. (2005). Patient empowerment: Reflections on the challenge of fostering the adoption of a new paradigm. *Patient Education and Counseling*, *57*(2), 153–157. https://doi.org/10.1016/j.pec.2004.05.008

Ashtiani, S., Makela, N., Carrion, P., & Austin, J. (2014). Parents' experiences of receiving their child's genetic diagnosis: A qualitative study to inform clinical genetics practice. *American Journal of Medical Genetics. Part A*, *164*(6), 1496–1502. https://doi.org/10.1002/ajmg.a.36525

Cho, M. H., & Cha, K. H. (1998). *Cross-country comparisons of quality of life*. Jipmundang.

Choi, E. J., Park, S., & Chae, S. M. (2022). An exploratory analysis of unmet healthcare needs among patients with rare diseases and their caregivers. *Health and Social Welfare Review*, *42*, 141–157. https://doi.org/10.15709/hswr.2022.42.2.141

Choi, J.-S., Park, Y.-S., Kim, J.-A., & Park, C.-S. (2019). International trends on patient-reported outcome measures for improving care quality and its implication for South Korea: Focus on OECD PaRIS. *Quality Improvement in Health Care*, *25*(1), 11–28. https://doi.org/10.14371/QIH.2019.25.1.11

Collins, D. (2003). Pretesting survey instruments: An overview of cognitive methods. *Quality of Life Research: an International Journal of Quality of Life Aspects of Treatment, Care and Rehabilitation*, *12*(3), 229–238. https://doi.org/10.1023/a:1023254226592

DeVellis, R. F. (2017). *Scale development: Theory and applications*. SAGE.

Diener, E., Emmons, R. A., Larsen, R. J., & Griffin, S. (1985). The satisfaction with life scale. *Journal of Personality Assessment*, *49*(1), 71–75. https://doi.org/10.1207/s15327752jpa4901_13

Diness, B. R., Overbeck, G., Hjortshøj, T. D., Hammer, T. B., 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

Fabrigar, L. R., Wegener, D. T., MacCallum, R. C., & Strahan, E. J. (1999). Evaluating the use of exploratory factor analysis in psychological research. *Psychological Methods*, *4*(3), 272–299. https://doi.org/10.1037/1082-989X.4.3.272

Feste, C., & Anderson, R. M. (1995). Empowerment: From philosophy to practice. *Patient Education and Counseling*, *26*(1–3), 139–144. https://doi.org/10.1016/0738-3991(95)00730-n

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*, *28*(7), 907–914. https://doi.org/10.1038/s41431-020-0592-1

Gibson, C. H. (1991). A concept analysis of empowerment. *Journal of Advanced Nursing*, *16*(3), 354–361. https://doi.org/10.1111/j.1365-2648.1991.tb01660.x

Hinkin, T. R. (1998). A brief tutorial on the development of measures for use in survey questionnaires. *Organizational Research Methods*, *1*(1), 104–121. https://doi.org/10.1177/109442819800100106

Horan, P. M., DiStefano, C., & Motl, R. W. (2003). Wording effects in self-esteem scales: Methodological artifact or response style? *Structural Equation Modeling: A Multidisciplinary Journal*, *10*(3), 435–455. https://doi.org/10.1207/S15328007SEM1003_6

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

Irwin, D. E., Varni, J. W., Yeatts, K., & DeWalt, D. A. (2009). Cognitive interviewing methodology in the development of a pediatric item bank: A patient reported outcomes measurement information system (PROMIS) study. *Health and Quality of Life Outcomes*, *7*, 3. https://doi.org/10.1186/1477-7525-7-3

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

Kline, R. B. (1994). An easy guide to factor analysis and its remedies. *Survey Research*, *17*, 1–29.

Landis, J. R., & Koch, G. G. (1977). The measurement of observer agreement for categorical data. *Biometrics*, *33*(1), 159–174. https://doi.org/10.2307/2529310

McAllister, M., & Dearing, A. (2015). Patient reported outcomes and patient empowerment in clinical genetics services. *Clinical Genetics*, *88*(2), 114–121. https://doi.org/10.1111/cge.12520

McAllister, M., Dunn, G., & Todd, C. (2011). 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. M., 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

Muñoz-Cabello, P., García-Miñaúr, S., Espinel-Vallejo, M. E., Fernández-Franco, L., Stephens, A., Santos-Simarro, F., Lapunzina-Badía, P., & McAllister, M. (2018). 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

Na, A. J. (1999). *Analysis of variables related to the level of health promotion lifestyle in convalescent patients in military hospitals* (Unpublished Master's Thesis, Yonsei University).

Polit, D. F., Beck, C. T., & Owen, S. V. (2007). Is the CVI an acceptable indicator of content validity? Appraisal and recommendations. *Research in Nursing and Health*, *30*(4), 459–467. https://doi.org/10.1002/nur.20199

Segundo-Ribeiro, M., Bacalá, B. T., Alvarenga, W. A., Nascimento, L. C., 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

U.S. Department of Health and Human Services FDA Center for Drug Evaluation and Research, U.S. Department of Health and Human Services FDA Center for Biologics Evaluation and Research, & U.S. Department of Health and Human Services FDA Center for Devices and Radiological Health. (2006). Guidance for industry: Patient-reported outcome measures: Use in m+edical product development to support labeling claims: Draft guidance. *Health and Quality of Life Outcomes*, *4*, 79. https://doi.org/10.1186/1477-7525-4-79

Voorwinden, J. S., 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

Wallston, K. A., Wallston, B. S., & DeVellis, R. (1978). Development of the multidimensional health locus of control (MHLC) scales. *Health Education Monographs*, *6*(2), 160–170. https://doi.org/10.1177/109019817800600107

Wild, D., Grove, A., Martin, M., Eremenco, S., McElroy, S., Verjee-Lorenz, A., Erikson, P., & ISPOR Task Force for Translation and Cultural Adaptation. (2005). Principles of good practice for the translation and cultural adaptation process for patient-reported outcomes (PRO) measures: Report of the ISPOR task force for translation and cultural adaptation. *Value in Health*, *8*(2), 94–104. https://doi.org/10.1111/j.1524-4733.2005.04054.x

Willis, G. B. (2005). *Cognitive interviewing: A tool for improving questionnaire design*. SAGE.

Willis, G. B., & Artino, A. R., Jr. (2013). What do our respondents think We're asking? Using cognitive interviewing to improve medical education surveys. *Journal of Graduate Medical Education*, *5*(3), 353–356. https://doi.org/10.4300/JGME-D-13-00154.1

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

Yusuf, A., Peltekova, I., Savion-Lemieux, T., Frei, J., Joober, R., Howe, J., Scherer, S. W., & Elsabbagh, M. (2021). Adaptation and validation of the genetic counseling outcome scale for autism spectrum disorders and related conditions. *Journal of Genetic Counseling*, *30*(1), 305–318. https://doi.org/10.1002/jgc4.1323

Zigmond, A. S., & Snaith, R. P. (1983). The hospital anxiety and depression scale. *Acta Psychiatrica Scandinavica*, *67*(6), 361–370. https://doi.org/10.1111/j.1600-0447.1983.tb09716.x

SUPPORTING INFORMATION

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