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Adaptation and preliminary validation of the genetic counseling outcome scale (GCOS-24) in
a Brazilian genetic counselling setting

ABSTRACT

Health professionals working in services providing genetic counseling need objective instruments to assess genetic counseling outcomes and also to “give a voice” to those using these services. Lack of knowledge regarding such outcomes may directly impact the effectiveness and the potential benefits of counseling, quality of life, health promotion, and empowerment of those receiving counseling. There are very few instruments available for most countries, however there are none in Brazil. In this context, this study aimed to adapt and preliminarily validate the Genetic Counseling Outcome Scale (GCOS-24), a Patient-Reported Outcome Measure (PROM), originally developed in British English. This methodological study recruited 278 individuals attending a medical genetic service at a Brazilian university hospital. We performed the translation, back-translation, semantic validation, pilot study and field study for testing of some psychometric properties. The instrument’s internal consistency and test-retest reliability (stability) were assessed using Cronbach’s alpha coefficient and Intraclass Correlation Coefficient, respectively. The **Brazilian version of the** GCOS-24 presented face and content validity, satisfactory internal consistency (Cronbach’s $\alpha=0.71$), and moderate stability (ICC=0.52). **It** was considered reliable, easily understood and relevant to assessing the genetic counseling outcomes for the study participants. Its construct validity still needs to be assessed to verify the instrument’s internal structure and its potential use to measure change in empowerment following genetic counseling provided by Brazilian clinical genetics services.

Keywords: Genetic Counseling; Patient Participation; Genetics Services; Validation Studies.

INTRODUCTION

Genetic counseling is conceived as the process of advising individuals to facilitate their understanding of and adaptation to the medical, psychological, and familial implications of genetic contributions to pathologies (Resta et al., 2006). Interest in assessing genetic counseling outcomes arose when the first clinical genetics services began providing such counseling, which indirectly may result in the assessment of services (Payne et al., 2008). Several instruments were developed for this purpose, however, the constructs used by them most frequently addressed knowledge of genetic risks, counselees' health status, and quality of life (Payne et al., 2008). These instruments have been criticized, mainly because they are similar to memory and comprehension tests, or because they measure psychological constructs such as anxiety and depression, rather than measuring counselees' benefits nor genetic counseling outcomes (McAllister et al., 2008a).

A new scale was proposed to assess the care provided by clinical genetics services, which has the potential to measure an essential benefit of genetic counseling that are valued by patients and providers of clinical genetics services, in other words, counselees' empowerment (McAllister et al., 2008b; McAllister et al., 2012). Empowerment is a new construct that emerged from extensive qualitative research with patients and genetics health professionals (McAllister et al., 2008a; McAllister et al., 2008b; McAllister et al., 2008c; McAllister et al., 2012). It has been recommended by the World Health Organization (Wallerstein, 2006) and is considered both a process as well as an outcome; clients may be empowered by the health staff or by themselves (Barr et al., 2015; Bravo et al., 2015; Castro et al., 2016). In the context of genetic counseling, an empowered client is an autonomous agent, with a certain control over his/her health and self-care instead of being a mere passive recipient of health care (McAllister et al., 2012).

The Genetic Counseling Outcome Scale (GCOS-24) has the potential to measure the empowerment construct by capturing five dimensions of empowerment: decisional control, cognitive control, behavioral control, emotional regulation and hope. It is efficacious to assess the genetic counseling process, as well as the services that provide it, “giving a voice” to patients and their families (McAllister et al., 2011). GCOS-24 is a patient-reported outcome measure (PROM), that is, a brief questionnaire with the potential to capture a patient’s self-reported health-related aspects without further interpretation on the part of a healthcare provider (Black, 2013). The GCOS-24 was translated and culturally adapted for use in Denmark and Spain and is also being used in studies conducted in Australia, as well as in the United Kingdom, where it was originally developed (McAllister, 2013; Diness et al., 2017; Muñoz-Cabello et al., 2017).

Because it is a short and simple instrument, focused on the needs of families, adequate for the non-directive genetic counseling model, and centered on psychosocial outcomes, the GCOS-24 has the potential to be used with Brazilian families receiving genetic counseling. Additionally, there is a lack of validated outcome assessment instruments specific to Brazilian individuals affected by genetic conditions. Therefore, this study’s aim is to verify whether the GCOS-24 presents evidence of reliability and validity to measure empowerment in Brazilian genetic counseling services, so that the health staff of these services can use a PROM among their clients.

This article reports the process of adaptation and preliminary validation of the Genetic Counseling Outcome Scale (GCOS-24) in a sample of Brazilian families receiving genetic counseling.

METHODS

It is a methodological study, conducted for fourteen months according to the stages recommended by the European group DISABKIDS® (DISABKIDS Group Europe, 2006): (1) translation to Brazilian Portuguese; (2) back-translation; (3) semantic validation; (4) pilot study; and (5) field study.

The author of the original scale (MM) has authorized and approved the adaptation and preliminary validation to the Brazilian Portuguese, as well as has participated in each of the above cited stages. All procedures complied with the ethical guidelines established by the Research Ethics Board of both the proposing and cooperating institutions, The University of São Paulo at Ribeirão Preto College of Nursing and The University of São Paulo Medical School at Ribeirão Preto Hospital and Clinics, respectively, which reviewed and approved the study (Opinion report No. 839.558). Additionally, all the participants signed free and informed consent forms according to Resolution 466/12 from the Brazilian National Council of Health.

The study's potential population was composed of 278 participants (patients or one of their family members) who were receiving genetic counseling in a medical genetic service at a Brazilian university hospital. The inclusion criteria were: be 18 years or older (age of majority in Brazil, when a person is considered an adult); be oriented in time and space; self-aware; be able to understand simple orders; and be able to express herself/himself in a coherent manner; verified by their ability to inform their age, address, day of the month and of the week (Fayers et al., 2007). Companions of patients and non-consanguineous relatives were excluded. The sample was composed by convenience, as the potential participants were invited to participate in the research at the waiting room of the genetics outpatient clinic, according to their order of arrival for their genetic counseling consultation with the geneticist doctors, who provided the role of genetic counselor.

All participants approached were aware of the reason for referral to clinical genetics services, as they had already been seen by the geneticist doctors, at least three times at the

genetic counseling service, thus having at least minimal knowledge about their genetic counseling process.

The GCOS-24 and other research questionnaires were applied before the genetic counseling consultation, since the participants used to leave the clinic immediately after their consultation, once the majority of them depended on public transportation with an established schedule. The first author and a rigorously trained research assistant invited the potential participants, who met the study's inclusion criteria to participate in the study. Those who agreed to participate signed the consent form and answered the instruments in the presence of the researchers, in a private environment according to the guidance of the author of the original scale and the study methodology. The participants answered the research instruments by themselves, without researcher's interference.

All the above-mentioned stages of the study followed the same recruitment and data collection process.

Genetic Counseling Outcome Scale (GCOS-24)

The GCOS-24 is composed of 24 items specifically designed to measure the empowerment construct among families and patients of services that provide genetic counseling. It includes a seven-point Likert scale ranging from “strongly agree” to “strongly disagree”. Its final score ranges from 24 to 168 points; a lower score represents less empowerment, while higher scores represent greater empowerment (McAllister et al., 2011).

Translation to Brazilian Portuguese and back-translation

The translation of the instrument from English to Brazilian Portuguese was independently performed by two bilingual individuals with knowledge of the theme and resulted in two versions in Brazilian Portuguese. These two versions were analyzed by the first

author and by a group of five nurses with expertise in genetic counseling, who reconciled both translations and reached a final consensual Brazilian Portuguese version.

The Brazilian Portuguese version was sent to two native English speakers, unaware of the study's objectives, who independently back-translated the instrument into English. The first author and the research group members reconciled these back translations and reached the final back-translated version. This reconciled version of the instrument was then sent to the author of the original scale, who compared it with the original GCOS-24, **found no significant differences, and gave her agreement to proceed. Thus,** a harmonized version was established to be used in the semantic validation process.

Semantic Validation

The semantic validation consisted of interviews intended to verify the participants' understanding of the instrument items meaning. This stage involved a number of participants recommend by DISABKIDS® method (DISABKIDS Group Europe, 2006). The following instruments were applied: the Brazilian Portuguese GCOS-24; a questionnaire addressing sociodemographic data; and two forms developed by the DISABKIDS® group (DISABKIDS Group Europe, 2006) intended to identify general and specific impressions of the scale's items. All the instruments enabled standardization and documentation of the semantic validation process, after which, the author of the original scale agreed with a version that was culturally and linguistically harmonized for Brazil. Participants were not later included in the pilot study.

Pilot Study

Following the semantic validation process, the first author conducted a pilot study to estimate the duration of the interviews, to provide preliminary analysis of the internal consistency of the GCOS-24 adapted version, and to simulate a field study. In order to

accomplish that, she applied a questionnaire addressing sociodemographic data and the semantically validated version of the GCOS-24.

Field Study

The adapted scale was answered by a representative sample of the target population to assess its preliminary psychometric properties.

The scale's internal consistency was assessed using the Cronbach's alpha coefficient, acceptable values of which are higher than 0.70 (de Vet et al., 2011). The scale's stability was assessed by test-retest. Thirty people from the total of participants of the field study were invited to participate in the test-retest (Johanson & Brooks, 2010). They answered the scale, for the first time in person, before their consultation with the geneticist doctor; and for the second time, via telephone, thirty days after the first application, according to literature recommendations (Gay, 1992; Ruscetta et al., 2005). **In terms of genetic counseling, there was no significant intervention (i.e. new information given, new developments about their genetic condition, test results, etc.) between the first and the second time participants answered the questionnaire.**

This second application enabled assessing the scale's reproducibility over time (de Vet et al., 2011), and statistical analysis was performed using the Intraclass Correlation Coefficient (ICC). Reference values were: $ICC < 0.40$ indicates weak correlation; $0.41 < ICC < 0.60$, moderate correlation; $0.61 < ICC < 0.80$, good or substantial correlation; and $ICC > 0.81$ almost perfect or very good correlation (de Vet et al., 2011; McDowell, 2006).

A database was structured in an Excel spreadsheet (Microsoft Office, 2010) using the double entry technique. The Statistical Analysis System (SAS), version 9.0 was used to calculate frequencies and percentages, and significance level was established at 5% ($\alpha=0.05$).

RESULTS

For the aforementioned stages, a total of 278 people was invited to participate in the study, during their genetic counseling process, that took 14 months in all. Among them, 22 refused to participate, therefore 256 participants composed the final sample. For the semantic validation 40 potential participants were approached, 36 agreed to participate, and four refused. For the pilot study, 58 people were approached, 52 agreed to participate and six refused. For the field study, 180 participants were approached, 168 agreed to participate and 12 refused.

Semantic Validation

The semantic validation phase occurred over a period of three months and included 36 participants, out of a total of 40 invited participants. The mean age of participants was 37.1 years ($SD=13.4$), and most reported: being caucasians (66.7%); married or in stable unions (63.9%); catholic (69.4%); and living in urban areas (91.7%). About education, 36.1% presented incomplete primary school; 44.4% were employed with a family income of 2.89 times the minimum wage; three individuals, on average, depended on this income. Non-geneticists doctors had referred the majority of them (86.1%) to the genetics service, and the others have searched for this service by themselves, spontaneously.

Face validity was evidenced through semantic validation. In general, the GCOS-24 adapted version was well accepted and considered easy to understand; that is, its items were easy to understand and considered relevant. In regard to the scale answers categories (strongly disagree=1; disagree=2; slightly disagree=3; neither disagree nor agree=4; slightly agree=5; agree=6; and strongly agree=7), 50% of the users had some difficulty in discriminating between them. They inquired whether so many categories were necessary to express their answers. In the scale's specific assessment, a few respondents presented doubts and questioned some items. In regard to the GCOS-24's relevance, 80.5% considered the items relevant and 16.6% considered one of the items to be "sometimes" relevant: "I don't know what could be gained

from each of the options available to me.” In regard to gender, men considered all the items to be relevant, while women seemed to make a more judicious analysis of the items and expressed some doubts regarding the relevance of one of the items: “Having this condition in my family makes me feel anxious.”

In regard to difficulty in understanding items, there was a problem with item 10: “I don’t know what could be gained from each of the options available to me.” This item generated the most doubts among the participants. One participant, who had a bachelor’s degree, considered this item unclear and inconsistent, because of an imprecise statement: “options available to me”. Most of the participants considered the options for answers to be clear and consistent. Therefore, to facilitate understanding of item 10, after the author of the original scale agreed with this change, item 10 was modified to: “I do not know what I can gain from each of the possibilities available to me in order to make an educated decision.” This change was intended to improve understanding of the item for the target population in Brazil. At the end of this step, a version adapted for Brazilian Portuguese was produced that could be used in the pilot study.

Pilot Study

The pilot study lasted three months and included 52 people, out of a total of 58 invited participants. The mean age of participants was 38.6 years ($SD=2.9$). The majority of respondents reported being caucasian (51.9%); married or in stable unions (78.8%); catholic (75%); living in urban areas (82.7%), being employed (65.4%) and having a family income between 1 and 2 times the minimum wage (51.9%), with three individuals depending on the wages, on average; 28.8% reported completing high school.

The interviews that included filling out all the instruments lasted 20 minutes, 10 (± 2 minutes) of which were used specifically to answer the GCOS-24. At this moment, the

preliminary internal consistency of the GCOS-24 adapted version was not satisfactory (Cronbach's alpha: 0.55).

Field Study

The aim of the field study was to analyze the performance of the scale in a representative sample of the target population, to refine the scale structure and obtain the preliminary psychometric properties of the semantically validated GCOS-24.

The field study lasted five months and 168 people agreed to participate, out of a total of 180 invited participants. The mean age of participants was 38.5 years ($SD=10.1$), and most reported: being caucasian (75%); married or in stable unions (70.2%); catholic (53.6%); and living in urban areas (95.8%). In terms of education, 44.6% presented some undergraduate study; 48.2% were employed; most had a family income between 1 and 2 times the minimum wage, with approximately three individuals depending on this income.

Internal consistency was considered satisfactory, with a Cronbach's alpha of 0.71.

For the test-retest, of the 168 participants in the field study, 30 were randomly selected and invited to answer the adapted instrument again, through telephone interviews, respecting an interval of 30 days after the first application of the instrument. All were contacted by phone and agreed to respond to GCOS-24 again. There were no difficulties in this procedure, as the participants already knew the instrument, as well as the researchers who were collecting the data. The test-retest presented an $ICC=0.52$, which is considered moderate. A difference of 0.02 was found between the scores that resulted from the first and second applications of the GCOS-24, confirming that there was agreement between the answers collected at different points in time (test-retest). This agreement is essential for the application of the instrument at the genetic counseling services, without compromising the analysis of the empowerment construct.

The average time to answer all the instruments were slightly shorter than the previous phase; taking 15 minutes, and the completion of the scale itself took approximately 8 minutes (± 2 minutes).

After the steps described above, a final version of GCOS-24 was generated.

DISCUSSION

This is the first Brazilian scale with evidence of reliability, showing the potential to measure empowerment in terms of genetic counseling outcomes. Internationally recommended procedures for the translation, adaptation and validation of instruments measuring subjective constructs were adopted (DISABKIDS Group Europe, 2006). The final sample was composed by 256 participants, affected or at risk of being affected by genetic conditions; this number was considered to be significative to compose the final sample of a scale adaptation study (Sapnas & Zeller, 2002).

Semantic validation of the Brazilian Portuguese GCOS-24 revealed that the items were considered easy to understand and relevant for the respondents' empowerment; however, some items presented difficulty to participants in discriminating between the seven categories of answers. This difficulty may be associated with the low educational level amongst participants, as well as poor economic conditions, which may also be reflected in the interpretation of some items. For instance, the answers to the items "I can see that good things have come from having this condition in my family"; "I feel positive about the future"; and "I am able to make plans for the future," were mostly based on the families' financial conditions, rather than on the fact an individual had a genetic disorder. Concerns regarding a family's economic condition often overlap health problems, which is an aspect also reported by other validation studies (Santos, 2013).

The Brazilian Portuguese GCOS-24 was well-accepted and easy to understand by

participants. The experience with the pilot study and field study showed that no changes were imposed on the daily routine of the genetic service where the instrument was applied, because it took just a few minutes to be completed, a fact that favors its future use in the routine of genetic counseling services. Another relevant aspect from the pilot study and field study is that the scale can be self-administered; thus, the constant presence of a health care worker is unnecessary to apply it, as its application is fast and the instrument is easily understood, enabling the normal flow of the genetics outpatient clinic consultations to be maintained.

The instrument's internal consistency was verified using the Cronbach's alpha coefficient, which presented a value of 0.71, which is considered satisfactory (de Vet et al., 2011). A Cronbach's alpha of 0.87 was obtained by the British English version (McAllister et al., 2011). The internal consistency of the Spanish version was good (Cronbach's $\alpha = 0.84$) (Muñoz-Cabello et al., 2017) and the Danish version was relatively good (Cronbach's $\alpha = 0.79$) (Diness et al., 2017). The value found in Brazil was slightly lower to that found for the United Kingdom, Spain and Denmark's population, but is still acceptable. It is essential to identify level of agreement between answers gathered at different points in time (test-retest) to verify an instrument's stability/reproducibility (McDowell, 2006). According to established criteria, the ICC of the Brazilian Portuguese GCOS-24 is moderate (Fayers et al., 2007; de Vet et al., 2011; McDowell, 2006). In general, the literature shows that physical domains tend to present a greater level of agreement in comparison to subjective domains, which comprise emotional and/or social aspects (de Soárez et al., 2009; Eiser et al., 2001; Creemens, Eiser & Blades, 2006).

In Brazil, however, there is a lack of standardized and updated instruments in the field addressed by the GCOS-24, which prevented any comparisons being included in this study. Similarly, there are limitations preventing comparisons with data from other populations receiving genetic counseling. Due to Brazil's continental proportions, its social diversity and

social, economic and cultural specificities, this sample does not represent the entire Brazilian population affected or at risk of developing or transmitting a genetic condition. Consequently, it remains a challenge to compare data obtained in this study with those collected from other populations. Additional studies addressing the instrument's semantic validation in Brazilian regions that are distant from that where the GCOS-24 was originally adapted would be helpful, considering Brazil's vast size and its mixture of different cultures and regional linguistic. The difficulty in obtaining samples from different regions of Brazil when conducting the validation of any instrument is one of the biggest challenges that researchers face (Deon et al., 2011).

Regarding the test-retest, a difference of 0.02 was found in the ICC between the first and the second applications of the scale. Perhaps, if the time between applications would be reduced better results could be obtained. Factor analyses would help to verify the dimensionality reflected in the instrument's items for the Brazilian population. Another limitation of this study was that neither construct validity nor responsiveness of the Brazilian Portuguese GCOS-24 was carried out.

The Brazilian Portuguese GCOS-24 is expected to contribute to clinical practice and research in Brazilian clinical genetics services, not least because it is an instrument that can give a voice to individuals who need to empower themselves in order to cope with the challenges posed by a genetic condition, challenges that can be explored during genetic counseling.

Brazilian Portuguese GCOS-24 has the potential to contribute to a more complete assessment of genetic counseling outcomes, going beyond the traditional biomedical model by adding the measurement of subjective aspects to clinical parameters. Thus, individuals affected or at risk of being affected by genetic conditions can become active empowered participants not only in the counseling process, but also in the follow-up process and during the progress of their genetic condition.

CONCLUSION

The English version of the GCOS-24 has been translated to Brazilian Portuguese, adapted and preliminarily validated for use with individuals affected by or at risk of developing and/or transmitting a genetic condition. The Brazilian version presents face and content validity and satisfactory internal consistency, though close to the limit of what is considered acceptable, and moderate stability/reproducibility. The adaptation and preliminary validation process has been completed and the same research group that conducted this preliminary psychometric validation, is finishing the second step of the research, which assessed responsiveness and construct validity, and expanded psychometric analyses using exploratory and confirmatory factor analysis and item response theory.

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