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ORIGINAL ARTICLE

Reliability and validity of the March of dimes preconception/prenatal family health history questionnaire: The Persian version

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ABSTRACT **In recent years, there has been a remarkable gap between rapid advancements in genetic technology and public health practice. Looking at the familial health history may bridge this gap for easier and cheaper diagnosis and prevention of congenital anomalies.The aim of this study was to validate and culturally adapt the March of Dimes Preconception/Prenatal Family Health History Questionnaire for the Iranian population.After obtaining written permission from March of Dimes, the translation–back translation of the original questionnaire was performed. The content validity was assessed by a team of 12 experts. Based on a sample of 50 general practitioners and 100 subjects referred to health centers from September to November 2014 in Tabriz, Iran, test-retest reliability and inter-rater reliability were evaluated by Kappa and Intra-class Correlation Coefficient (ICC). Content validity of the Persian version of the questionnaire was confirmed according to the modified kappa value above 0.76 for all the items included in this tool. Inter-rater reliability assessment yielded a kappa value between 0.62 and 0.92 for variables with dichotomous measurement scales and ICC ranged from 0.6 to 0.9 for variables with numeric scales. Test–retest re-administration produced kappa ranging from 0.62 to 0.92 for variables with dichotomous measurement scales and ICC from 0.6 to 0.9 for variables with numeric scales. The Persian version of the March of Dimes preconception/prenatal family health history questionnaire showed acceptable reliability and validity and may be used as a simple tool for the detection of risk factors of birth defects in Iranian population.**

Key Words: family history, questionnaire, reliability, validity

INTRODUCTION

"Congenital anomalies affect 1 in 33 infants with 3.2 million birth defect-related disabilities every year in the globe. They may result in long-term disability with significant impacts on individuals, families, health care systems and communities" (WHO 2014). Birth defects are the first leading causes of prenatal mortality and childhood morbidity and disability in many countries (Dastgiri et al. 2011). A recent report by the March of Dimes showed that, worldwide, an estimated 6% of births or 7.9 million children are born annually with a major birth defect of genetic or partially genetic origin (Romitti 2007). Congenital anomalies are the most common causes of death in children (1–59 months) in Iran (Rahbar et al. 2013). Total prevalence of congenital anomalies was 1.9 per 100 births between 2000 and 2011 in east Azerbaijan, northwest of Iran (Bateni et al. 2013).

"The wide range of causes of birth defects means that a portfolio of prevention approaches is needed. The prevention of these disorders is available in 60% of cases" (Czeizel 1993; Czeizel et al. 1993).This needs however epidemiological information.

Genetic achievements can be applied to public health programs by taking family history, even though the advent of genomics and consequently its domination challenged us to examine how we can apply the assumptions of genetic knowledge to public health practice (Khoury 2003). Meanwhile, such a big challenge can also serve as an opportunity to target health promotion activities to high risk populations in a more effective and efficient manner. Thus, family health can be regarded as a unique tool to grab this opportunity because it covers such genetic and environmental components of the diseases as shared cultural and behavioral risks. (Khoury 2003; Yoon et al. 2003).

The Centers for Disease Control and Prevention (CDC) in their 2006 report offered 10 recommendations for improving preconception health. One of the elements of these recommendations is obtaining preconception family health history (Johnson et al. 2006).

Family history can also lead to early diagnosis during pregnancy, which allows for secondary interventions in decision-making during pregnancy, including location and mode of delivery and tertiary interventions in medical care during the newborn period and childhood (Dolan and Moore 2007).

Various tools for assessing preconception family health history have been developed and validated but there is currently no validated instrument designed specifically for Iranian people focusing on birth defects.

The aim of this study was to validate and culturally adapt the March of Dimes Preconception/Prenatal Family Health History Questionnaire for the Iranian population.

MATERIALS AND METHODS

Subjects

This study was carried out in the Tabriz district, in the northwest of Iran. The study consisted of 100 married female subjects who were recruited from a rural population of the Tabriz district and referred to health centers from September to November 2014. Rural population in the region was 158 731 people who received their primary health care from 17 health centers. The inclusion criteria included married female populations, aged 15–49.

Using Microsoft Excel 2010 eligible persons were selected randomly. Each woman had a unique code in the rural health care system.

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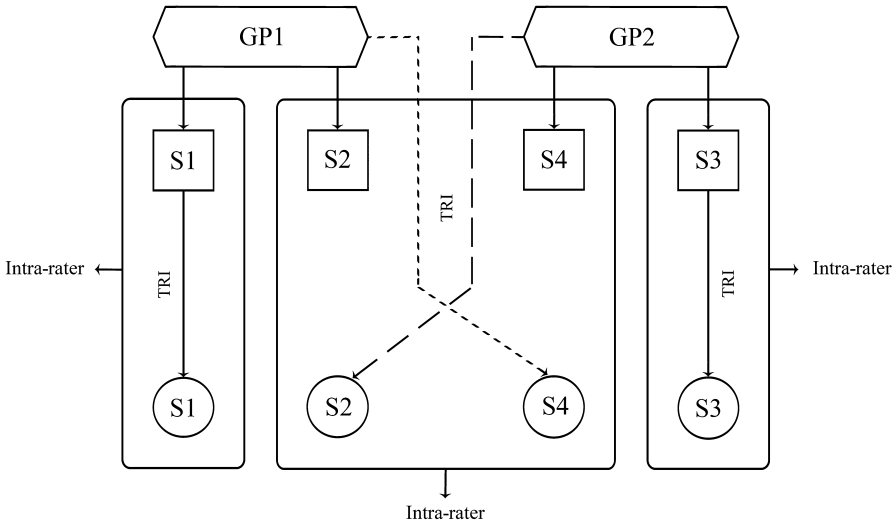

Fig. 1 Allocation diagram for assessing inter-rater and intra-rater reliability of the questionnaire.
GP, General Practitioner; S, Subject; TRI, Test Retest Interval = 4 weeks.
□, Test phase; ○, Retest phase.

All 50 general practitioners (GPs) who worked in rural health centers of the Tabriz district were invited to participate in this study to complete the questionnaires for the subjects. These GPs were grouped randomly to 25 pairs. Each pair of GPs was randomly allocated to assess four subjects who were randomly selected from the 100 sample participants. As illustrated in the Figure 1, the questionnaire was completed for all four subjects twice with the time interval of 4 weeks. Both two phases of administration and re-administration of the questionnaire were performed by the same GPs in the paired group for two subjects in order to assess intra-rater reliability. To investigate inter-rater reliability the re-administration phases were performed independently by both the GPs for two remaining subjects of the group (dashed lines in Figure 1). The duration of each data collection was approximately 15–20 min. All data were collected during a 2-month period in 2014. Neither the physicians nor the study subjects knew about the second interview in advance.

Selection of the questionnaire

We reviewed the literature to find the birth defects-specific tool for obtaining preconception family health history. Among methods such as Becoming a Parent, 2nd Edition–Wisconsin Association for Perinatal Care (Committee, 2007), Comprehensive Perinatal Services Program–Initial Combined Assessment, California (Program, n. d.), Women's Health Questionnaire–Boston Healthy Start Initiative (Commission, n. d.), PKC Preconception Guidance Tool (Tool 2006), The Preconception/Prenatal Family History Questionnaire–The March of Dimes (Foundation, 2008), met all of the requirements. We got permission from the Associate Director, Health Information Delivery Pregnancy & Health Education Center of March of Dimes Foundation to start the validation study of this tool.

Measurements

This tool was initially designed by March of Dimes as a highly valid and reliable measure of preconception family health history. The tool was designed for use in the clinical care setting to screen for potential risks of families associated with birth defects.

This checklist was developed to gather information about the married women and their spouses. The questions cover basic demographic information, ethnic background, past medical and developmental history, and current medical issues, exposure to risk factors and health behaviors for both the wife and her spouse. The questionnaire also includes data such as number of pregnancies, number of full-term and preterm births and number of stillbirths, we labeled this section as "pregnancy background". It also includes past medical history of particular diseases, for example, thyroid diseases, diabetes and seizures, which were referred to as "particular diseases history". A wide range of specific conditions about couples and their extended families are covered through the past medical and developmental history section. Two more questions include having had a genetic testing and being blood relatives of couples, which we considered as the category of "others".

A small space for "office use only" was replaced in the tool to write significant findings and recommendations as well as the date discussed with family, the health care provider's signature, and the patient signature.

Preparation of the Persian versions

The English version of the questionnaire was translated into Persian by two bilingual experts providing the first draft of the Persian version of the instrument after reaching dual agreement on the translation of English text. The ethnic background items changed totally according to the existing ethnic groups of the Iranian population. The draft was then checked by an expert panel of two pediatricians, one epidemiologist and an obstetrician. The Persian translation agreed by the expert panel was then back-translated into English by another bilingual person and was compared for compatibility with the original version.

Content validity

The content validity was assessed by a team of 12 professional experts including pediatricians, gynecologists, obstetricians, public health practitioners and clinical geneticists. In the qualitative assessment, experts provided written feedback on the clarity and relevance of the content of the questions to the Iranian culture. To ensure valid applicability and prevent loss of reliability due to potentially various understandings of the items in different cultural settings, a process of cultural adaptation was followed through investigating and discussing all the items in a panel of experts and making modifications or adding descriptions to those items needing to be clarified or explained. For instance, the ethnic background items were substituted by an existing ethnic group of the country. A

brief description was provided for the questions related to specific medical conditions such as Canavan disease, Phenylketonnuria, Gaucher disease by a team of professional experts. Quantitative evaluation was performed by administering a questionnaire for each expert panel to ask the necessity, relevancy, simplicity and clarity of each item based on a 4-point scale response to each question.

Ethical considerations

All eligible subjects were asked to complete written informed consent to participate in the study. All subjects' information remained confidential. This survey received ethics approval from the committee of ethics in Tabriz University of Medical Sciences.

Statistical analysis

The content validity statistic used in this study was the modified content validity index (modified kappa). This index is preferred to traditional I-CVI because it also measures the chance agreement (Polit et al. 2007).

The reliability for variables with dichotomous measurement scales such as past medical and developmental history was assessed using kappa statistic (Sim and Wright 2005). Kappa values of 0.80 and above represented excellent agreement, values between 0.61 and 0.80 represented substantial agreement, 0.41 to 0.61 represented moderate agreement, and values below 0.40 suggested fair to poor agreement (Landis and Koch 1977). The Intra-class Correlation Coefficient (ICC) was used to assess the reliability for variables with numeric scales such as pregnancy background. ICCs ≤ 0.4 were considered poor to fair, 0.41–0.60 moderate, 0.61–0.80 good and >0.80 excellent (Bartko 1966).

Data were analyzed using the STATA 11 statistical software package (STATA Corporation, College Station, TX, USA).

RESULTS

Sample characteristics

A total of 190 completed questionnaires were collected for data analysis, with less than 5% missing data. The mean age of participants was 32.4 (SD = 10.3). Seventy-four subjects had an under diploma education (74%), few had finished high school (total 10 persons and 10%) and 16 (16%) subjects were illiterate. Eighty-three persons were housekeepers without an income and 17 worked as carpet weavers with an income level of less than $US200 per month.

Mean age of the GPs who completed the questionnaires for subjects was 37 (SD = 5). Thirty-three GPs (66%) were female and 17 (34%) were male. Mean length of working experience of GPs in the health centers was 8 years (SD = 3).

Content validity

The content validity of the questionnaire was assessed by a team of 12 professional experts. Except for few minor changes, no major change was applied to the original instrument. From the first step of the assessment of the tool, five items of the 134 questions were revised according to the quantitative evaluation and qualitative recommendations of the experts. Content validity of the Persian version of the questionnaire was confirmed according to the modified kappa value above 0.76 for all items included in the tool.

Intra-rater reliability

For the intra-rater component of the study, questionnaires were administered and re-administered for 96 subjects with the time interval of 4 weeks. Across the items with dichotomous measurement scales, kappa varied between 0.68 and 0.94. Table 1 presents intra-rater reliability for various category of the questionnaire.

Test-retest reliability for the category of pregnancy background was assessed using the intra-class correlation coefficient, which ranged from 0.74 for "Miscarriages" and "Preterm labor" to 0.96 for "Number of Pregnancies". Sixty-seven percent of the items of the pregnancy background category yielded excellent agreement and the remaining 33% yielded good agreement.

Inter-rater reliability

As expected, inter-rater agreement measures were slightly lower than those for intra-rater agreement which were calculated for 94 questionnaires. As indicated in Table 1, kappa values showed substantial to excellent agreements across the items with dichotomous measurement scales ranging from 0.62 to 0.92.

Inter-rater agreement was then examined for variables with numeric scales through pregnancy background category using ICC. Fifty-five percent of items had ICC values greater than 0.8, showing that inter-rater agreement was excellent and 45% ones produced ICC over 0.61, suggesting that inter-rater reliability was good.

DISCUSSION

"Obtaining a family history remains an inexpensive and basic approach to identify individuals at risk for genetic disorders. Family history is a way to reach those at higher risk and to target resources to get them into screening. A family history can establish patterns of

Table 1 Inter-rater and intra-rater reliability statistics for the various categories of the questionnaire

	Inter-rater reliability (Kappa)			Intra-rater reliability (Kappa)		
	n (%)	*n* (%)	*n* (%)	*n* (%)	*n* (%)	*n* (%)
Category (number of items)	≥0.80	0.61–0.80	≤.60	≥0.80	0.61–0.80	≤0.60
Past medical history (8)	6 (75)	2 (25)	0	7 (87)	1 (13)	0
Ethnic background (2)	2 (100)	0	0	2 (100)	0	0
Health behaviors (10)	7 (70)	3 (30)	0	8 (80)	2 (20)	0
Particular diseases history (11)	5 (45)	6 (55)	0	7 (64)	4 (36)	0
Exposure to risk factors (4)	3 (75)	1 (24)	0	2 (50)	2 (2)	0
Past medical and developmental history of families (74)	52 (70)	22 (30)	0	61 (82)	13 (18)	0
Others (7)	6 (86)	1 (14)	0	5 (71)	2 (29)	0

inheritance and serve as a guide to diagnostic, therapeutic, and preventive approaches" (Malarcher et al. 2002). In this study, we aimed to evaluate the validity and reliability of the March of Dimes Preconception/Prenatal Family Health History Questionnaire for the Iranian population. According to the findings of this study, the Persian version of the questionnaire showed good content validity and sufficient comprehensiveness. The data of the study also proved that the results were replicable over a 4-week period.

To our knowledge, reliability and validity of this tool have not been examined yet in Iran and other countries; therefore, data collected through this study were not comparable with similar studies.

In terms of comprehensiveness, according to the feedback given by the close scrutiny of 12 professional experts and their remarkable approval of the validity variables, it can be inferred that the questionnaire includes the critical and essential points to investigate risk factors for birth defects. The relevancy and clarity criteria were also verified by the reviewers with the expression that all items could be understood clearly without any unnecessary questions included. To assess the content validity based on the expert views, a traditional consensus-based content validity index is being used in most studies. However, we used an alternative modified measure that takes into account the consistency of agreements (Polit et al. 2007).

Intra-class correlation coefficients for numeric variables and kappa statistics for dichotomous variables showed acceptable test-retest reliability and inter-rater reliability for the questionnaire items. The majority of items (92 of 116, 80%) had kappa values greater than 0.8 and 20% of items showed kappa between 0.61 and 0.8, suggesting that intra-rater agreement was substantial to excellent for variables with dichotomous measurement scales. ICC for variables with numeric scales ranged from 0.74 to 0.96 indicating good to excellent test-retest reliability. Inter-rater reliability also was promising, which yielded kappa between 0.62 and 0.92 for variables with dichotomous measurement scales and ICC ranged from 0.6 to 0.9 for variables with numeric scales.

Considering the role of screening programs in the prevention of birth defects especially in the developing countries such as Iran indicates the necessity of applying them in their health system. Accordingly there is a need for a valid and reliable tool to set up a prevention program for congenital anomalies in Iran. Iran has a very efficient and potential primary health system network that has recently been more activated by applying the "Family Physician" project. So there is a sufficient opportunity to design a surveillance system for congenital anomalies. A valid and reliable questionnaire as a risk assessment tool can play a key role in early detection of birth defects in such a surveillance system.

The Persian version of the March of Dimes preconception/prenatal family health history questionnaire showed acceptable reliability and validity and may be used as a simple tool for the detection of risk factors of birth defects in the Iranian population.

Some points should be taken into account about the application of this tool.

The March of Dimes Preconception/Prenatal Family Health History Questionnaire is a risk assessment tool. As a risk assessment tool, the use of such a questionnaire will be helpful for increasing the chance of detecting those at higher risks of genetic disorders (Frezzo et al. 2003); however, some limitations should also be considered, such as the fact that this tool has not been validated and widely used over various settings or populations worldwide and its real value is not well documented. No doubt cost-effectiveness needs also to be assessed before recommending its use widely. In this research study we only assessed the validity and inter- and intra-rater reliability of this tool in the study setting. So applicability of this questionnaire in the whole population of the country needs to be investigated in other cost effectiveness studies.

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DISCLOSURE

None.

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