Translation and cross-cultural adaptation: Questionnaires for siblings of people with Trisomy 21

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Abstract. Trisomy 21 (T21), also known as Down syndrome (DS), is the most frequently diagnosed chromosomal abnormality. One of the main characteristics of people with T21 is intellectual deficit, accompanied by a global delay in development, including the areas of movement and language. When a child with a disability is born, such as T21, family relationships are impacted and there is a need to redefine roles and change the lifestyle of the family. The sibling of a person with a disability can often feel underserved due to the care and attention demands of the most vulnerable child. The objective of this study was to conduct the translation and cross-cultural adaptation of two questionnaires: "Questionnaire for brothers and sisters, from 9 to 11 years old" and "Questionnaire for brothers and sisters, from 12 years old", to assess the perspectives of siblings of people with T21 in the Brazilian population. Contact was made with the researcher who authored the translated instruments in order to request permission to prepare the Portuguese/Brazilian version of the instrument for use in this research; once the authorization for the translation of the instrument was obtained, the steps of translation, back-translation and cross-cultural adaptation were followed, obtaining satisfactory content validity indices from the evaluation of a group of judges.

Keywords. Down Syndrome, siblings, translation, instrument

1. Introduction

Trisomy of chromosome 21 (T21), also known as Down syndrome (DS)(1), is the most frequent chromosomal anomaly diagnosed (2–4). This condition was first reported by the English physician John Langdon Down in 1866, is characterized by the presence of an extra numerary chromosome in pair 21 and, according to the cytogenetic perspective, presents itself in three forms, being: Simple or free trisomy in 95% of cases, Mosaicism in 1 to 2% of individuals and Translocations between chromosomes 14 and 21, in about 3 to 4 % of cases (2,5,6).

One of the main characteristics of people with T21 is intellectual disability, accompanied by a global delay in development, including the areas of movement and language. The phenotypic characteristics are easy to recognize as: single palmar fold, brachycephaly, epicanthic folds, flattened nasal base, hypoplasia of the median region of the face, smaller front occipital diameter, short neck, protruding tongue, small and underdeveloped ears and hypotonia (2,4,5,7,8).

When a child with T21 is born, family relationships are impacted and there is a need to redefine roles and changes in the lifestyle not only of the parents, but also of the siblings and other members (9–13).

Studies on older siblings bring the consequences caused by the arrival of another child in the family. The firstborn suffers greater impact with the birth of other siblings (11,13–18)). The changes in family structure and dynamics resulting from the birth of a second child have repercussions on the first due to the imposition of new roles (11,15,19). Studies conducted with a group of older siblings of children with and without disabilities bring similarities in their results. Both groups of siblings are faced with the need for sharing, sharing of physical space, and parental attention to the younger sibling. It is also observed that the quality of the relationship and interaction between siblings is considered positive

in both groups (10,12,19).

Although similar, the experience and intensity of common situations when a sibling is born are different when it comes to the arrival of a child with a disability. Studies with groups of siblings of children without disabilities report the change in the division of parental attention, caused by the presence of another child in the home; intensification of routine and greater family unity. Similarly, in groups of siblings of children with disabilities, the impact felt by the changes in the organization and family structure may be greater, due to the need for more attention and spending of parents' time for medical follow-ups and treatments, as well as private experiences for this public, such as the discovery and acceptance of disability (11,13,20–22).

The sibling of a person with a disability may often feel underserved because of the care and attention demands of the most vulnerable child. The literature states that these findings can be considered in both Western and Eastern culture, as described by (21). Just like parents who had to adjust to the disability, siblings also go through the same emotional states, such as frustration, fear, anger, guilt, worry, among others (11,20,23). It is observed that the younger the age and the more restricted the information received about the disability, this child may have greater difficulty in facing his feelings about the needs of his brother (23).

The main difficulties characterized as stressors are related: to the increase of responsibilities and functions, both for the care of the brother with disabilities, as well as the activities of the house; feelings of loneliness and resentment due to the greater attention of parents and health professionals to the sibling with disabilities; lack of parental attention; jealousy when they perceive the differential treatment and favouring of the brother; negative feelings such as fear of not having the attention of the parents and that the sibling will die; guilt for having wished something bad to happen to the sibling or for not being affected by the same condition or disease; shame and embarrassment in the face of questions from other children about the physical or behavioural differences of the sibling and misinformation about the condition of the sibling, in general (24,25).

Resilience is one of the favourable points, pointed out as part of the gains to families who live the experience of having a child with special needs, being common the development of characteristics such as patience, understanding and altruism, as well as humanitarian attitudes and greater sense of autonomy and independence (24,26,27).

The presence of a sibling with a disability leads to an early maturation for siblings regardless of whether they are older or younger (9,11,13,26,28–30).

From the point of view of siblings, this factor may be caused by the perception of responsibility towards the sibling with disabilities, mainly aiming at his/her well-being (9,11–13,20,26,28,30). From the psychological point of view, such early maturation may not be beneficial for the child, because he fails to experience some stages necessary for a healthy emotional development, which may cause consequences in adult life (11,30).

A multicultural epidemiological survey conducted by Skotko, Levine and Goldstein, involving families of people with T21, resulted in three different studies, published in the American Journal of Medical Genetics (2011). The first evaluated 2,044 responses to questionnaires sent to parents and guardians of people with T21, the second involved questionnaires from 822 brothers and sisters of people with T21, aged 9 years or older, and the third study evaluated the survey responses of 284 people with Down syndrome (31–33).

The authors point out that, previously, there had not been a quantitative study with a large sample size that characterized the feelings and perceptions of siblings of people with T21, and studies with small samples had been carried out so far, compared to controls and other populations, but without direct questioning to the sibling regarding their perception (31).

This fact also occurs in studies conducted with the Brazilian population, as reported in the integrative review conducted by Brazilians authors, add that in studies with siblings of people with disabilities, parents are the main informants, however the perceptions of parents differ from those of children in the fraternal relationship (9,25,34).

Due to the scarcity of standardized instruments to address the sibling perception of T21 people, Skotko; Levine and Goldstein (31), developed a research instrument for this audience. Initially, ten siblings, recruited by the authors, participated in focus group tests for preliminary testing of the instrument. For the validity and reliability test of the questionnaires, 300 families associated with the Rhode Island Down Syndrome Society were invited to participate.

The questionnaire for siblings aged 9 to 11 years was elaborated from the following structure: Questions 1 to 12: Participant identification data; Questions from 13 to 26: 13 items with answers on a Likert scale of four points and two open questions. The questionnaire for siblings from 12 years of age follows the following structure: Questions 1 to 7 : Participant identification data; Questions from 8 to 20: 12 items with answers on a 7-point Likert scale; 04 (four) open questions and 06 (six questions) regarding the identification of the participant (31).

Considering the relevant experience of the authors Brian Skotko and Sue Levine in the theme about siblings of people with T21 and recognizing the scarcity of studies and instruments focusing on this family group, this study aimed to translate, make

the cross-cultural adaptation and seek evidence of validity of the "Questionnaire for brothers and sisters, from 9 to 11 years old" and "Questionnaire for brothers and sisters, from 12 years old"

2. Research Methods

The study was approved by the Research Ethics Committee of Universidade Presbiteriana Mackenzie under opinion n. 5.150.549.

The steps to reach the final version of a translation for the Brazilian version of the instrument followed the norms established Beaton and cols. (35) going through the phases of authorization of the authors, translation into Portuguese, evaluation of the translated version by a committee of expert judges, followed by back-translation and preparation of the final version for author's approval.

The steps were developed as follows:

Stage I – contact with the authors: Initially, a contact was made with the researcher Dr. Brian Skotko, author of the instruments that will be translated and used in this study, in order to request permission for the preparation of the version of the instrument in Portuguese/Brazil for use in this research.

Stage II - Translation: Once authorized, the translation was done by two bilingual researchers, born and literate in Brazil, independently, working in the area as translators of scientific material. Next, the synthesis of the two versions of the translations was performed by two specialists, born in Brazil, with clinical and teaching experience and experience in the subject addressed, to analyse discrepancies, arriving at the first version of the translation.

Stage III - Analysis of the committee of expert judges: The committee of judges was composed of six people with academic training with stricto sensu post-graduation and with experience in the studies of people with disabilities. The judges received the original English version of the instrument and the preliminary version to make considerations regarding semantic, idiomatic, experiential and conceptual equivalences and to evaluate the clarity of each of the items and need for adaptation and/or changes of terms, giving suggestions, if necessary.

Subsequently, the data of the form were analysed from the Content Validity Index (CVI) that measures the proportion or percentage of judges who are in agreement on certain aspects of the material produced and its items (36).

The six versions of the judges were compiled by the two experts (the same as in item III), arriving at the second preliminary version.

Stage IV - Back-translation and submission to the author for approval: The back-translation was then performed from the synthesized version, by a native translator of the English language, resident in Brazil, with knowledge of both languages. The verification of the back-translation with the original version provided the basis for the definitive version of the instrument in the Portuguese language of Brazil. After these steps, the definitive version was sent to the author for knowledge, adjustments and approval.

3. Results

3.1 Partial results

The representativeness of the answers was obtained from an evaluation based on a scale with four alternatives of the Likert type with scores from one to four. The index score was calculated by the sum of the items that were marked by the answers corresponding to "Adequate" and "Needs few changes" (equivalent to answers 1 and 2) by the specialists, eliminating the items that received scores "Needs many changes" and "Inadequate" (equivalent to answers 3 and 4), and finally for the calculation of CVI, The following formula was applied:

$$CVI = \frac{\text{Sum total of responses 1 or 2}}{\text{Total number of responses}}$$

Figure 1 - Content Validity Index

The CVI verified by the experts in the "Questionnaire for brothers and sisters aged 9 to 11 years" was 0.91, as shown below:

Items	**A**	**B**	**C**	**D**	**E**	**F**	
1	1	1	2	A	2	2	
2	1	1	2	A	2	3	
3	1	1	4	A	1	3	
4	1	1	1	A	1	1	
5	1	1	4	A	1	2	
6	1	1	1	A	2	2	
7	1	1	2	A	2	1	
8	1	1	1	A	1	1	
Total items	8	8	8	8	8	8	**48**
Answers 1 and 2	8	8	6	8	8	6	**44**

Table 1: Questionnaire for brothers and sisters aged 9 to 11 years.

The CVI verified by the specialists in the "Questionnaire for brothers and sisters from 12 years of age" was 0.85, as shown in the following

table:

Items	A	B	C	D	E	F	
1	1	1	2	1	1	2	
2	1	1	1	1	1	2	
3	1	1	1	1	1	1	
4	1	1	1	1	1	2	
5	1	1	1	1	1	1	
6	1	1	1	1	1	1	
7	4	3	4	1	2	3	
8	3	3	4	1	1	4	
9	3	1	4	1	1	2	
10	1	1	1	1	1	2	
11	1	1	1	1	1	1	
Total items	11	11	11	11	11	11	**66**
Answers 1 and 2	8	9	8	11	11	9	**56**

Table 2: Questionnaire for brothers and sisters age 12 and older

Thus, both questionnaires are considered representative when evaluated by a committee of six judges, since, for evaluation by this number of judges, it is expected that the CVI will reach an agreement rate higher than 0.78. In addition, in general, to verify the validity of new instruments, a minimum agreement of 0.80 is suggested.(36)

To facilitate data analysis, it was suggested that the questions of the questionnaires follow the same structure: Identification of the participant; items with Likert scale answers and open questions , so the author was proposed a new sequencing in the Questionnaire for brothers and sisters from 12 years of age, in which the questions numbered from 24 to 29 in the aforementioned table, which was after the open questions, were reassigned to the "identification of the participant", starting to have their numbering from 8 to 13, respectively.

In the Questionnaire for brothers and sisters aged 9 to 11 years, it was proposed to include a question covering the levels of education (question 8).

The final back-translated versions were sent to the author, with the respective suggestions for alteration, which were accepted by the author, being consolidated after a new revision of the questionnaire to be used in the collections in the next stage.

4. Conclusions

The theme about siblings of people with T21 is still little explored. Further research is important through instruments recognized and validated by the scientific community. The questionnaire proposed here for translation and cultural adaptation to the Portuguese language of Brazil was the only one found in the literature that is dedicated to the measurement of data related to the perspective of siblings with T21, proving necessary to search for evidence of validity for the Brazilian population. The content validity index (CVI) verified by the experts in the "Questionnaire for brothers and sisters aged 9 to 11 years" was 0.91 and in the "Questionnaire for brothers and sisters from 12 years of age, it was 0.85. These indexes can be considered satisfactory, confirming the content validity of the Brazilian version (36) Thus, the instrument can be used to generate foundations, so that the work of welcoming and supporting the sibling of the person with T21 in Brazil can be carried out with greater support from the literature.

5. References

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