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Identification of relevant International Classification of Functioning Disability and Health (ICF) categories in patients with 22q11.2 Deletion Syndrome: a Delphi exercise

Identificação das categorias da Classificação Internacional de Funcionalidade, Incapacidade e Saúde (CIF) relevantes em pacientes com Síndrome de Deleção 22q11.2: um exercício Delph

Keywords

DiGeorge Syndrome
International Classification of Functioning
Disability and Health
Delphi technique
Quality of life
Rehabilitation

ABSTRACT

Purpose: The aim of this study was to identify the most typical and relevant categories of the International Classification of Functioning, Disability and Health (ICF) for patients with 22q11.2 Deletion Syndrome. **Methods:** Based on the Delphi technique an expert survey through e-mail was performed among health professionals' specialists in the 22q11.2DS. Data were collected in 2 rounds. Answers were analysed for the degree of consensus. **Results:** 7 Experts recruited through e-mail distribution lists of professional organizations and personal networks participated in the study. Categories in all ICF components that were considered typical and/or relevant by at least 80% of the responders were added to a pilot ICF instrument for children with 22q11.2DS, with a total of 145 ICF categories. **Conclusion:** a list of ICF categories that are considered relevant and typical for 22q11.2DS condition by international experts was created. This is an important step towards identifying ICF Core Sets for chronic paediatric conditions in Brazil.

Descritores

Síndrome de DiGeorge
Classificação Internacional de Funcionalidade
Incapacidade e Saúde
Técnica Delfos
Qualidade de Vida
Reabilitação

RESUMO

Objetivo: O objetivo deste estudo foi identificar as categorias mais típicas e relevantes da Classificação Internacional de Funcionalidade, Incapacidade e Saúde (CIF) para pacientes com Síndrome de Deleção 22q11.2. **Método:** Com base na técnica Delphi, foi realizada uma pesquisa por e-mail entre profissionais de saúde especialistas na Síndrome de Deleção 22q11.2. Os dados foram coletados em 2 rodadas. As respostas foram analisadas quanto ao grau de consenso. **Resultados:** 7 especialistas, recrutados por e-mail através de listas de distribuição de organizações profissionais e redes pessoais, participaram do estudo. As categorias em todos os componentes da CIF consideradas típicas e/ou relevantes por pelo menos 80% dos participantes foram adicionadas a um instrumento piloto da CIF para crianças com Síndrome de Deleção 22q11.2, com um total de 145 categorias da CIF. **Conclusão:** uma lista de categorias da CIF consideradas relevantes e típicas para a Síndrome de Deleção 22q11.2 foi criada por especialistas internacionais. Este é um passo importante para a identificação de Core Sets da CIF para condições pediátricas crônicas no Brasil.

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INTRODUCTION

The 22q11.2 Deletion Syndrome (22q11.2DS) is related to the deletion of a DNA segment, encompassing DiGeorge (DGS), velocardiofacial (VCFS) and facial and conotruncal anomalies syndromes[1]. With a prevalence of approximately 1: 4,000 live births[2] is the most common microdeletion identified in humans. With a broad spectrum, 22q11.2DS has more than 180 clinical manifestations, both physical and behavioural[3], being the second major cause of developmental delay and congenital heart defects, behind only Down Syndrome[4]. Health care management requires a multidisciplinary approach involving physicians, speech-language pathology and audiology, psychiatry, psychology, physical, occupational, and behavioural therapy, genetic counselling and others. Although common, lack of recognition of the condition and lack of familiarity with genetic testing methods, together with the wide variability of clinical manifestations, delays diagnosis.

Speech and language difficulties comprise the most distressing aspects for the parents of children with 22q11.2DS and can have a significant impact on daily functioning, especially regarding activity and participation, and on the quality of life of patients[5] which can result in high burden for the family. Providing adequate support to help optimize functioning and quality of life of the patient is essential[6].

Traditionally, speech-language pathologists and audiologists used a medical model to guide their assessment and treatment practices[7–9]. However, in recent years there has been a remarkable shift in practice beyond the medical model[10]; seeing the state of health with a broader and integrated vision between the disease and the social environment in which the individual is inserted. The World Health Organization (WHO) created a biopsychosocial model, the International Classification of Functioning, Disability and Health (ICF). ICF is a global and universal system that can integrate the medical and social models. Functioning and disability, according to the WHO, are related functions of the body's functions and structures, activity and participation, and the interaction with contextual factors on the individual health status[11]. Instruments that assess the impact on functioning in 22q11.2DS specifically are non-existent. Based on the ICF we can define a spectrum of functioning and health domains by using a globally agreed upon language of functioning and health for specific conditions. This is important because it provides a framework for assessment, data collection and quantification of clinical findings of a large number of pediatric patients[12].

The ICF classification offers a huge and exhaustive list of categories that can describe functioning across any health condition. Each ICF category is assigned a unique number and a definition. A major challenge when applying the ICF in practice is its length. The total ICF classification contains 1545 categories. In order to improve its applicability, it is necessary to develop a set of domains that describe the spectrum of functioning and health a specific health condition that can be used in clinical practice. Therefore, the objective of this study was to identify the most typical and relevant aspects of functioning in patients with 22q11.2DS using the ICF framework and classification by a survey of experts using the Delphi technique.

METHODS

Study design

A consensus-building, 2-round, e-mail survey with health experts in DiGeorge Syndrome, using the Delphi technique, was conducted from February to May 2019. The Delphi technique is a structured communication process with 4 key characteristics: anonymity, iteration with controlled feedback, statistical group response, and expert input (13). Figure 1 displays the Delphi rounds.

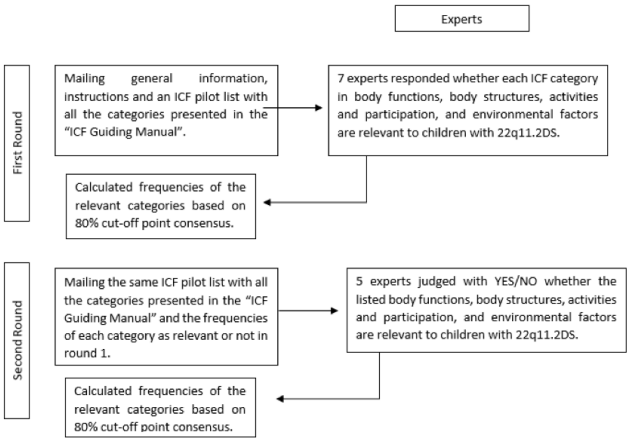

Figure 1. The course of the Delphi exercise

The present research was approved by the local Research Ethics Committee of The National Institute of Women, Children and Adolescents Health Fernandes Figueira/Oswaldo Cruz Foundation - IFF/Fiocruz (CAE 99733418.0.0000.5269).

Recruitment of participants

Since the ICF was developed to facilitate communication between different groups of people the aim was to include experts who work with patients with 22q11.2DS from different health professions. Potential participants who work in 22q11.2 reference centers were invited to participate via e-mail distribution from 2 states in Brazil.

Data collection and measures

A questionnaire based in the literature review using the "ICF Guiding Manual" issued by the Brazilian Federal Council of Speech-Language Pathology and Audiology(14), all categories presented in the Guiding Manual were added to create a pilot ICF list to be evaluated from the experts, which included: 57 body functions, 26 body structures, 84 activity and participation categories and 57 environmental factors categories.

In the first round a questionnaire based on the "ICF guiding Manual" with information letters were sent by e-mail to all identified experts. The letter included background information, purpose, ICF manual with the WHO definitions of body functions, body structures, activities and participation, and environmental factors and instructions with a detailed timeline. We also collected personal expert's information (gender,

professional training, academic titles, and highest degree). Experts who replied were contacted through the Google Forms platform and were invited to comprise a panel of experts to the study. The experts were presented to the ICF pilot list and were asked to judge each category's relevance concerning evaluation of children with 22q11.2DS in general, according to their clinical practice, since all of the experts had experience with the Syndrome and had previous knowledge of the ICF. The participants had 2 weeks to respond and reminders were sent out approximately 3 days before the deadline. The experts were blinded in the two rounds.

In the second round of the Delphi exercise the same questionnaire used in the round 1 was sent together with corresponding instructions. The participants were given the same categories with the percentage of all participants that had considered this ICF category as relevant and/or typical for the target condition in the round 1. The participants were asked to consider whether each ICF category is relevant and/or typical for patients with the target condition, taking their own and the answers of the group from the first round into account. Again, the participants had 2 weeks to respond, and reminders were sent out 3 days before the deadline. In both, first and second round, the experts could add any outcomes they considered as being relevant and/or comments.

Analyses

Descriptive statistics were used to analyze the response rates and personal characteristics of the participants. After each round of the Delphi exercise, the percentage of participants that considered an ICF category as relevant was calculated. Consensus threshold was set at 80% of the participants indicating the relevance of the category after the first and second round. Relative frequencies of ICF categories were calculated, and data analysis was performed. The agreement between the raters was analyzed by Cohen's Kappa and intraclass correlation (IC). Cohen's Kappa is used to measure how much agreement exists beyond the amount expected by chance alone. The results of the second round were further summarized and the ICF list is organized by components.

RESULTS

Participants and response rate

Experts from 2 different states in Brazil participated, that being São Paulo and Rio de Janeiro, two of the most important and influential states in Brazil. From the 7 experts, 6 were women. The level of education among the participants was high, where only one of them does not have a PhD degree. Although we had experts from different majors, the majority had continued their postgraduation studies in some area related to Genetics. Table 1 shows all the information related to the expert's profile that responded in at least 1 Delphi round.

Table 1. Professional profile of experts (n = 7)

Gender	Freq. (n)
Male	1
Female	6
Origin	
São Paulo	4
Rio de Janeiro	3
Academic Titles	
Speech- Language Pathologist and Audiologist	3
Physician	2
Biologist	1
Psychologist	1
Post-Graduation	
Doctorate in Communication Sciences and Disorders	1
Doctorate in Biological Sciences (emphasis in Biophysics)	1
Doctorate in Biological Sciences (emphasis in Genetics)	2
Doctorate in Rehabilitation Sciences (emphasis in Genetics)	1
Doctorate in Public Health	1
Master's in communication Sciences and Disorders	1
Professional Occupation	
Genetics	4
Language and communication disorders	3

Caption: Freq.= Frequency

In the first round of the study, 7 experts rated the ICF list. In the second round 5 answered the online form. The agreement on the level of the 4 ICF components for both first and second round of the study is presented in Table 2.

Table 2. Agreement between the raters in the ICF categories by components

ICF component	First Round	Second Round
Mean	73%	71%
Kappa	0.70	0.69
IC	0.65 – 0.74	0.63 – 0.75

Captions: ICF = International Classification of Functioning, Disability and Health

Relevant and/or typical ICF categories

The consensus frequency after the second round of the Delphi exercise is summarized in Table 3.

Table 3. Consensus frequency of the ICF components between the raters after the Second Round

ICF Component	Frequency (n)	Frequency (%)
Body Function (b)	28	19.3
Body Structure (s)	11	7.6
Activities and Participation (d)	70	48.3
Environment (e)	36	24.8
Total ICF categories	145	100

Captions: ICF = International Classification of Functioning, Disability and Health

In round 1 the participants named between 224 different ICF categories. In round 1, 123 ICF categories had a consensus equal or greater than 80%. In round 2 there was over 80% agreement for relevance in 145 ICF categories. The ICF categories in the 4 components considered relevant by at least 80% of the participants in round 2 were added to the final ICF list and are shown in Chart 1. The categories are presented in the order of the ICF.

In the component body functions, 28 different ICF categories reached a consensus of 80%. In the component body structures, 11 different ICF categories; activities and participation, 70 different ICF categories and in environmental factors, 36 different ICF categories.

Chart 1. ICF list after the Second Round of the Delphi exercise

BODY FUNCTIONS			
ICF Category	Description	ICF Category	Description
b110	Consciousness functions	*b167*	Mental functions of language
b114	Orientation functions	*b172*	Calculation functions
b117	Intellectual functions	*b180*	Experience of self and time functions
b122	Global psychosocial functions	*b189*	Specific mental functions, other specified and unspecified
b126	Temperament and personality functions	*b198*	Mental functions, other specified
b130	Energy and drive functions	*b230*	Hearing functions
b139	Global mental functions, other specified and unspecified	*b310*	Voice functions
b140	Attention functions	*b320*	Articulation functions
b144	Memory functions	*b330*	Fluency and rhythm of speech functions
b147	Psychomotor functions	*b340*	Alternative vocalization functions
b152	Emotional functions	*b398*	Voice and speech functions, other specified
b156	Perceptual functions	*b399*	Voice and speech functions, unspecified
b160	Thought functions	*b440*	Respiration functions
b164	Higher-level cognitive functions	*b735*	Muscle tone functions
BODY STRUCTURES			
ICF Category	Description	ICF Category	Description
s110	Structure of brain	*s330*	Structure of pharynx
s240	Structure of external ear	*s340*	Structure of larynx
s250	Structure of middle ear	*s398*	Structures involved in voice and speech, other specified
s260	Structure of inner ear	*s399*	Structures involved in voice and speech, unspecified
s310	Structure of nose	*s710*	Structure of head and neck region
s320	Structure of mouth		

Chart 1: Continuation...

ACTIVITIES AND PARTICIPATION			
ICF Category	Description	ICF Category	Description
d110	Watching	*d349*	Communication - producing, other specified and unspecified
d115	Listening	*d350*	Conversation
d120	Other purposeful sensing	*d355*	Discussion
d129	Purposeful sensory experiences, other specified and unspecified	*d360*	Using communication devices and techniques
d130	Copying	*d4*	Mobility
d135	Rehearsing	*d470*	Using transportation
d140	Learning to read	*d599*	Self-care, unspecified
d145	Learning to write	*d610*	Acquiring a place to live
d150	Learning to calculate	*d620*	Acquisition of goods and services
d155	Acquiring skills	*d629*	Acquisition of necessities, other specified and unspecified
d159	Basic learning, other specified and unspecified	*d710*	Basic interpersonal interactions
d160	Focusing attention	*d720*	Complex interpersonal interactions
d163	Thinking	*d729*	General interpersonal interactions, other specified and unspecified
d166	Reading	*d740*	Formal relationships
d170	Writing	*d750*	Informal social relationships
d172	Calculating	*d760*	Family relationships
d175	Solving problems	*d770*	Intimate relationships
d177	Making decisions	*d798*	Interpersonal interactions and relationships, other specified
d179	Applying knowledge, other specified and unspecified	*d810*	Informal education
d198	Learning and applying knowledge, other specified	*d815*	Preschool education
d199	Learning and applying knowledge, unspecified	*d820*	School education
d210	Undertaking a single task	*d825*	Vocational training
d220	Undertaking multiple tasks	*d830*	Higher education
d240	Handling stress and other psychological demands	*d839*	Education, other specified and unspecified
d298	General tasks and demands, other specified	*d840*	Apprenticeship (work preparation)
d299	General tasks and demands, unspecified	*d845*	Acquiring, keeping and terminating a job
d310	Communicating with - receiving - spoken messages	*d850*	Remunerative employment
d315	Communicating with - receiving - nonverbal messages	*d855*	Non-remunerative employment
d320	Communicating with - receiving - formal sign language messages	*d860*	Basic economic transactions
d325	Communicating with - receiving - written messages	*d870*	Economic self-sufficiency

Chart 1: Continuation...

d329	Communicating - receiving, other specified and unspecified	*d898*	Major life areas, other specified
d330	Speaking	*d910*	Community life
d335	Producing nonverbal messages	*d920*	Recreation and leisure
d340	Producing messages in formal sign language	*d940*	Human rights
d345	Writing messages	*d998*	Community, social and civic life, other specified
	CONTEXTUAL FACTORS		
e115	Products and technology for personal use in daily living	*e420*	Individual attitudes of friends
e125	Products and technology for communication	*e425*	Individual attitudes of acquaintances, peers, colleagues, neighbours and community members
e130	Products and technology for education	*e430*	Individual attitudes of people in positions of authority
e135	Products and technology for employment	*e435*	Individual attitudes of people in subordinate positions
e140	Products and technology for culture, recreation and sport	*e440*	Individual attitudes of personal care providers and personal assistants
e310	Immediate family	*e450*	Individual attitudes of health professionals
e315	Extended family	*e455*	Individual attitudes of other professionals
e320	Friends	*e460*	Societal attitudes
e325	Acquaintances, peers, colleagues, neighbours and community members	*e530*	Utilities services, systems and policies
e330	People in positions of authority	*e535*	Communication services, systems and policies
e340	Personal care providers and personal assistants	*e540*	Transportation services, systems and policies
e350	Domesticated animals	*e555*	Associations and organizational services, systems and policies
e355	Health professionals	*e560*	Media services, systems and policies
e360	Other professionals	*e570*	Social security services, systems and policies
e398	Support and relationships, other specified	*e575*	General social support services, systems and policies
e399	Support and relationships, unspecified	*e580*	Health services, systems and policies
e410	Individual attitudes of immediate family members	*e585*	Education and training services, systems and policies
e415	Individual attitudes of extended family members	*e590*	Labour and employment services, systems and policies

Captions: ICF = International Classification of Functioning, Disability and Health

DISCUSSION

This is the first research that explores aspects of functioning in children with 22q11.2DS in the perspective of ICF, previous studies show evidences in the use of ICF as a reference system for evaluation and standardized reporting of rehabilitation interventions[15,16]. The results found on this Delphi exercise showed that a consensus was reached on 145 ICF categories as relevant on functioning evaluation of patients with 22q11.2DS, according to 5 experts from different professional backgrounds, all of them have experience and work with 22q11.2DS and have previous knowledge about the ICF, although none of them are experts on its use. In the component Body functions many categories with a consensus of 80% or greater were from global mental functions (b110-b139), specific mental functions (b140-b189), and voice and speech functions (b310-b399). In addition, hearing functions (b230) and muscle tone functions (b735) was agreed as relevant for functioning in 22q11.2DS. Also, the relevance of respiratory functions in case of inhaling air into the lungs, the exchange of gases between air and blood, and exhaling air was recognized (b440). This goes in accordance with previous findings, since generalized motor delays, hypotonia, muscle fiber differences, impaired intellectual abilities, and brain abnormalities have been well reported before[17–20].

In the Body Structures component the agreed categories were related to the brain (s110), ear (s240, s250 and s260), nose (s310), mouth (s320), pharynx (s330), larynx (s340), structures of head and neck region (s710), and structures involved in voice and speech (s983 and s399); thus showing consistence with conditions associated with 22q11.2DS which includes palatal abnormalities, velopharyngeal insufficiency[21–23], altered structural brain and connectivity differences[24,25] and altered auditory processing[26].

The largest number of categories agreed upon was part of the component Activities and Participation, and most categories pertained to Learning and applying knowledge (Chapter 1), Communication (Chapter 3) and Major life areas (Chapter 8). In addition, >80% agreement was reached on the importance of restrictions in participation, including General tasks and demands (Chapter 2), Mobility (Chapter 4), self-care (Chapter 5) , Domestic life (Chapter 6), Interpersonal interactions and relationships (Chapter 7), Major life areas (Chapter 8) and Community, social and civic life (Chapter 9).

In the component Environmental factors, categories concerning support and relationships were perceived as most relevant and/or typical. Experts are also aware of the importance of products and technology (referring for personal use, communication, education, employment, culture, recreation, and sport), attitudes services, systems, and policies.

The identified categories reflect the experts' clinical experience with 22q11.2DS and their knowledge about the variety of manifestations of the disease that may not be as frequent but might have an important impact on functioning. For example, studies have reported that children with this syndrome experience mild to moderate intellectual disability, impairments in cognitive functioning and difficulties in a range of cognitive abilities, including cognitive control[27–31], and the agreed ICF categories related to global mental functions are likely relevant categories for limitations for most patients with 22q11.2DS, and can have important consequences, but will not affect a majority of patients.

Although the study was not designed to study differences across healthcare providers, interesting differences were seen. The Speech Language Pathologist and Audiologist who has a PhD in Genetics did not emphasize the importance of functions related to mental functions while this was not shared by the other Speech-Language Pathologist and Audiologist. Surprisingly, psychologist did not agree with all other experts regarding consciousness functions, orientation functions, and mental functions (other specified) as being relevant on functioning evaluation of children with 22q11.2DS. It could be that psychologists are used to treating patients and know better the conditions that result in more important participation restrictions. The largest differences were seen for the body functions and body structures, the opinion of the Speech-Language Pathologist and Audiologist who has a PhD in Communication Disorders was not shared in many categories by other health professionals. This might indicate different thoughts, but also experience in assessing such factors from clinical practice, since this Speech-Language Pathologist and Audiologist is the one who has the most years of practice working specifically with 22q11.2 patients nationally and internationally.

The differences between the relevant and typical ICF categories for evaluation in 22q11.2DS children points out to the need of a condition-oriented approach when defining ICF-based tools in clinical practice. Therefore, it may be possible to define a Generic list of ICF categories for all chronic conditions. The pattern of the consensus in the relevance of activities and participation and environmental factors on functioning evaluation of children with 22q11.2DS reflects the behavioural and psychological phenotype of the 22q11.2DS and the influence in their lives. Recent studies have been emphasizing the importance of studying factors that may influence the development of neuropsychiatric deficits in individuals with 22q11.2DS. It seems that psychotic disorders increase with age and follow a developmental pattern[32], demonstrating the need of careful support, assessment and monitoring of psychiatric symptoms and influences on developing such disorders since young age.

The largest number of ICF categories in Activities and Participation and Environmental factors in the final ICF list reflects the high contribution of the condition in the life of 22q11.2DS children and may also be related to the need of the experts in assessing influences in 22q11.2DS children's functioning in the new biopsychosocial model, where the experts see beyond the physical impact of the syndrome and seek to understand the real influence in patient's functioning. Few studies have been focusing on establish and measuring health related to quality of life as a clinical outcome in children with 22qDS; a recent study showed that children with 22qDS had a significantly poorer quality of life when compared to age-matched cohorts of healthy children and children with chronic disease[33].

As expected, because the ICF list in this study was made based in the literature review using the "ICF Guiding Manual" issued by the Brazilian Federal Council of Speech-Language

Pathology and Audiology, we can see that the largest set of relevant body functions and structures are related to Speech and Language functionalities. It is also not surprising that many of the relevant Activities and Participation categories are related to areas of the Speech-Language Pathologist and Audiologist practice, such as communication, learning and applying knowledge skills. Information regarding Personal factors are not currently classified in ICF and were not incorporated in this study.

Although much care was taken in the selection of experts and a relatively wide range of medical disciplines and health professions was achieved, most of them had the same level of education (doctorate) and area of post-graduation study (genetics). Thus, the selection of categories, as well as the importance accorded to some of them as reflected by the percentage of agreement, can be underestimated or overestimated. Participants were included based on expertise by verification via updated online Curriculum (curriculum lattes). Initially we had 7 experts participating in the study, and since the beginning of the study we noticed a certain difficulty from the participants in keeping the deadline, although we extended the period initially given in both rounds, and sent weekly reminders; after 4 weeks of waiting we finished round 2, with no answer from 2 raters, concluding the study with the answers we had collected, thus the second round of this study has a slightly small difference in the number of participants. It was noticed that in the course of the Delphi rounds those with the highest experience had the highest drop-out. The completion of each Delphi round was quite time consuming. However, none of the questionnaires returned were incomplete, suggesting responder fatigue did not play a major role.

In addition, experts agreed upon the relevance of many Environmental factors than were identified as facilitators or barriers by the patients. Experts also agreed on the importance of assistive products (e115, e125, e130, e135 and e140), support and relationships specified or not (e398 and e399), immediate and extended family, friends, acquaintances, peers, colleagues, neighbours and community members (e310, e15, e320 and e325), people in positions of authority (e330), personal care providers and personal assistants (e340), as well as domesticated animals (e350), health, and other professionals (e355 and e360); individual attitudes of immediate and extended family, friends, acquaintances, peers, colleagues, neighbours and community members, people in subordinate positions, people in positions of authority, personal care providers and personal assistants, health, and other professionals, and societal attitudes (e410, e415, e420, e425, e430, e435, e440, e450, e450, e455 and e460); and utilities services in systems and policies, communication, transportation, media, social security, general social support, health, education and training, and labour and employment (e530, e535, e540, e560, e570, e575, e580, e585 and e590).

The 80% cut-off point to express the consensus of the group of participants is based on the recognition that a higher cut-off point would generate very few items, while lower cut-off point would generate many items. Finally, we recognize that the generalizability of this Delphi exercise is limited due to the number and selection of experts. The amount of time that was necessary to answer round one, especially if a person did not know the concepts of the ICF before may have kept many experts away from participating in the second round of the study, which made the number of experts answering both phases slightly small in the end. As study limitations, the small number of experts and little geographic variety among them might have decreased the chance of detecting all relevant ICF categories in the evaluation of children with 22q11.2DS, even though we tried to recruit experts from a different majority of services and places, most of the participants experts came from Rio de Janeiro or São Paulo; and this may have reduced the generalizability of our results to other health services across the country. Previous studies have emphasized the need of including different experts from different parts of the world and different cultures in order to develop an ICF based practical tools[34,35].

CONCLUSION

Based on the consensus of experts, it was possible to come up with an ICF list of 145 categories that can serve as reference evaluation for children with 22q11.2DS. This study is an important step towards identifying Core Sets of ICF categories that can be used across chronic paediatric health conditions for research purposes, primarily. Therefore, this is a preliminary ICF list, and it should be tested in different regions and health services in order to improve the reliability and generalizability of these results to increase the validity and the acceptance among health professionals of future ICF based outcomes.

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Contribuição dos autores

APCC participou da idealização do estudo, coleta, análise, interpretação de dados, e redação do artigo. CTMR participou na condição de orientadora contribuindo para o delineamento do estudo, análise, interpretação dos dados, e redação final do artigo. DDGH e LCR participaram da análise, interpretação dos dados e redação final do artigo.