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ORIGINAL RESEARCH ARTICLE

Linking Quality-of-Life Measures Using the International Classification of Functioning, Disability and Health and the International Classification of Functioning, Disability and Health—Children and Youth Version in Chronic Health Conditions

The Example of Young People with Hemophilia

ABSTRACT

Krasuska M, Riva S, Fava L, von Mackensen S, Bullinger M: Linking quality-of-life measures using the international classification of functioning, disability and health and the international classification of functioning, disability and health—children and youth version in chronic health conditions: the example of young people with hemophilia. *Am J Phys Med Rehabil* 2012;91(suppl):S74–S83.

Objective: This study aimed to measure the health and functioning of children with hemophilia in Europe using the International Classification of Functioning, Disability and Health (ICF) and the International Classification of Functioning, Disability and Health—Children and Youth Version (ICF-CY) as a frame of reference and items from health-related quality-of-life instruments as a measurement tool within a European data set.

Design: Based on the results of linkage of items from the hemophilia-specific health-related quality-of-life questionnaire for children and adolescents to ICF/ICF-CY, the categories most relevant for the description of health and functioning of children with hemophilia were identified for each domain of the ICF/ICF-CY. Using data from the European Study of Clinical, Health, Economic, and Quality-of-Life Outcomes of Hemophilia treatment on 446 children, the frequency of impairments in body structures and body functions, restrictions in activities and participation, and barriers in contextual factors were calculated.

Results: In general, the frequency of impairments, restrictions, and barriers was low in the studied population of children with hemophilia. However, the level of restriction as well as barriers was higher than the level of impairments. Older children, children receiving on-demand treatment, and children with severe hemophilia tended to have more problems compared with younger children, children receiving prophylaxis treatment, and children with mild to moderate hemophilia.

Conclusions: Using items from quality-of-life instruments with the ICF-CY as a frame of reference proved to be a useful approach for the assessment of health and functioning in children with hemophilia.

Key Words: ICF, ICF-CY, Quality-of-Life, Hemophilia, Children

The classification of health has undergone major developments in the past decades. Beyond the biomedical model of health, the physical, mental, and social well-being and, with it, the subjective perception of health has been acknowledged. In addition, functioning has been introduced as a major component of health. Both concepts, well-being and functioning, are now considered as essential aspects of human health, reflected in individual experience. They are also constituents of patients' health-related quality-of-life, which can best be understood through patient self-report. The current study describes and discusses health and functioning in children and adolescents with hemophilia in Europe based on the results of the linkage between the hemophilia-specific health-related quality-of-life questionnaire for children and adolescents (Haemo-QoL) and International Classification of Functioning, Disability and Health/International Classification of Functioning, Disability and Health—Children and Youth Version (ICF/ICF-CY) classification.

Hemophilia

Hemophilia is a lifelong genetic bleeding disorder caused by a deficiency of coagulation factor VIII (hemophilia A) or factor IX (hemophilia B), which affects mainly men.¹ It is classified as severe when the factor activity level is less than 1%, moderate when the factor activity level is between 1% and 5%, and mild when the activity is between 5% and 30%.² The major hallmark of severe hemophilia is spontaneous and repeated bleedings into joints, muscles, and soft tissues, with the first joint bleed usually occurring in the first years of life.³ Unprevented bleeds may result in the development of arthropathy, chronic pain, and muscle atrophy and ultimately leads to disability.¹

Hemophilia is mainly treated by replacing the missing coagulation factor using intravenous injections of factor concentrate to stop bleeding (on-demand treatment) or at regular intervals to prevent bleeding (prophylactic treatment).⁴ Prophylactic treatment is considered a gold standard for children with hemophilia, and it is recommended by the World Health Organization⁵ and by the World Federation of Haemophilia.⁵ Prophylactic treatment prevents the development of secondary health conditions such as arthropathy and enables boys with hemophilia to enjoy the same life as their healthy peers, with few restrictions related to the treatment administration. However, because of the high costs of prophylactic treatment, few countries provide this service to pe-

diatric patients. On-demand treatment remains a predominant approach, and in many areas around the world, children with hemophilia receive very little to no help.¹ Moreover, in Europe, treatment patterns for pediatric hemophilia patients differ across countries, with some countries reimbursing prophylaxis treatment for all children with diagnoses of hemophilia and others paying for on-demand treatment only.⁵ In case of chronic health conditions such as hemophilia, various nonmedical factors may strongly influence the patient's everyday functioning and well-being. These are, for example, living conditions of the family, support received from family members, friends, or coping strategies.⁶ The hemophilia management of a child remains a challenging task and involves health professionals, the child himself, his family, and his immediate social environment (school and peers).⁷

Health-Related Quality-of-Life

In the past four decades, the concept of health-related quality-of-life (HRQoL) has been increasingly acknowledged as an outcome criterion used in clinical trials and in the assessment of medical care.⁶

HRQoL instruments can relate to the health status in general (generic instruments) or to a particular health condition (disease-specific measures) from the individual's point of view (self-assessment) or that of an observer (proxy-assessment).⁸ HRQoL instruments may target child or adult populations and may be applicable to a single culture or internationally.⁹ Functioning is generally thought to be a more objective or observable category compared with the more subjective category of well-being. However, functioning may also be subjectively perceived, and well-being may be observable in behavior. Therefore, the perspective of who reports on functioning and well-being should be included in respective assessments.^{10,11} HRQoL measures are especially useful in the assessment of health outcomes in chronic disease such as hemophilia, for which no ultimate cure is available, and the improvement of a person's quality-of-life and functioning is the main focus of attention. In addition, the perceived burden of disease might extend beyond disease symptoms and inconveniences associated with treatment administration. Hemophilia is often associated with a stigma, and the behavior of the immediate social environment of a child (family and peers) can be negatively affected by the knowledge of the child's health condition. Parents might behave in an overprotective way,¹² and teachers might not allow a child to participate in a certain school event involving physical activity.¹³

The ICF and ICF-CY

The classification of health has undergone major developments in the last decades. In addition to the World Health Organization's International Classification of Disease,¹⁴ a standard diagnostic tool that classifies health from a biomedical perspective, the ICF has been developed.¹⁵ The ICF and its version for children and adolescents—the ICF-CY¹⁶—are universal conceptual frameworks that provide a globally accepted basis for understanding and studying health and health-related states as well as their outcomes and determinants.¹⁷ ICF was created with the aim of covering all aspects of human functioning and disability that are either aspects of health or are health-related, independent of diagnosis. The ICF-CY is a recently developed classification that aims to capture aspects of functioning relevant to children and adolescents (e.g., learning and playing). It was developed to be structurally consistent with the ICF for adults. ICF/ICF-CY can be applied in different areas such as clinical practice and research, demographics, education and social policy.

Since its introduction in 2001 the ICF has gained increasing attention in health science, especially in the field of measurement and health outcome research.¹⁷ The ICF can be used as a reference tool to report the level of functioning and disability of an individual or a selected population¹⁸ using ICF/ICF-CY categories and ICF/ICF-CY qualifiers. Categories are the basic units of ICF/ICF-CY classification, and they are grouped into four ICF/ICF-CY components: body functions and body structures, activities and participation, and contextual factors consisting of environmental and personal factors. There are no existing categories for personal factors because of a large cultural and social variance associated with them. ICF/ICF-CY and qualifiers cannot be directly applied in measurement process. Instead, appropriate assessment procedures, patient interviews, or laboratory tests need to be established and validated.¹⁸

One of the possible sources of information to quantify ICF/ICF-CY categories is items from HRQoL measures. HRQoL instruments provide reliable and valid self- (or proxy-) reported information on patient's well-being and functioning in relation to their health status. There are a large number of HRQoL measures that provide a variety of items, related to different health conditions, which can be linked to ICF categories. Two steps are required to translate information obtained from HRQoL instruments to ICF/ICF-CY categories and qualifiers. First, items from HRQoL measures need to be linked to the relevant ICF/ICF-CY categories; second, the response

scales of HRQoL measures need to be translated into ICF/ICF-CY qualifiers.

The Multidisciplinary Research Network on Health and Disability in Europe Project

The Multidisciplinary Research Network on Health and Disability in Europe (MURINET) project provided a unique opportunity to have a closer look at the role of the ICF/ICF-CY in the pediatric context. The MURINET project had several tasks represented by working groups from various countries. At the University of Hamburg, the goal was to identify relationships between the ICF/ICF-CY and HRQoL measures with regard to child health. The team at the Institute for Medical Psychology at the University Medical Centre Hamburg Eppendorf made use of data on quality-of-life in children and adolescents available from three studies: the European DISABKIDS study,¹⁹ a European Commission-funded project on the development of HRQoL tools and assessment of HRQoL in 1600 children with chronic conditions; the Haemo-QoL project²⁰ regarding the assessment of HRQoL in more than 300 children and adolescents with hemophilia; and the European Study of Clinical, Health, Economic, and Quality-of-life Outcomes of Haemophilia Treatment (ESCHQoL) project (www.eschqol.org), a European Commission-funded study on the health outcomes in more than 1400 persons with hemophilia; of these, more than 400 are children and adolescents.

The aim of the Haemo-QoL project was to develop and validate simultaneously in six European countries (France, Germany, Italy, the Netherlands, Spain, and United Kingdom) the Haemo-QoL questionnaire.²⁰ The Haemo-QoL is available as self-report and proxy version for three age groups (4–7, 8–12, and 13–16 yrs). Items from the Haemo-QoL questionnaire were linked to the ICF-CY categories within the MURINET project.²¹ The Haemo-QoL questionnaire was also included in the ESCHQoL study.

The aim of the ESCHQoL study was to assess outcomes in pediatric and adult hemophilia patients in a multinational and multidisciplinary approach, using patient-reported outcomes instruments for the assessment of quality-of-life, treatment satisfaction, health status, and healthcare consumption of patients with hemophilia in Europe. In total, 1424 patients from 21 European countries were enrolled in the study²²; among these were 446 children aged 4–16 yrs and their parents. Study participants and parents of young patients were given questionnaires containing questions about health care and instruments

assessing generic and disease-specific (Haemo-QoL questionnaire) HRQoL, treatment satisfaction, coping strategies, life satisfaction, and social support.

The methodology followed in the MURINET project was as follows: first, a linkage exercise was performed on the DISABKIDS chronic-generic and disease-specific modules and ICF classification.²³ Second, the linkage exercise was performed on the Haemo-QoL questionnaire.²¹ Third, the most relevant ICF/ICF-CY categories for the description of health and functioning of children with hemophilia were identified based on the results of the linkage of the Haemo-QoL questionnaire and the ICF/ICF-CY categories. Fourth, one item from the Haemo-QoL questionnaire was assigned to each of the previously identified categories. Fifth, the health and functioning of children with hemophilia in Europe was described using previously identified ICF/ICF-CY categories and the HRQoL data from the MURINET project. This article reports on the study comprising of the third, fourth, and fifth steps described above as part of the MURINET project.

When the DISABKIDS chronic-generic module and seven DISABKIDS disease-specific modules²⁴ were linked to the ICF classification, it was found that, on average, more than one ICF category can be assigned to the particular HRQoL item. This was summarized by the concept of “content density,” which shows an average number of ICF/ICF-CY categories that were assigned to one item in the questionnaire. Another important finding from this analysis was that, on average, different categories were represented in chronic-generic and in disease-specific modules of DISABKIDS questionnaire.

The primary aim of the current study was to describe the functioning status of children and adolescents with hemophilia in Europe using the ICF classification as a reference tool and the hemophilia-specific HRQoL questionnaire as a measurement tool, based on the outcomes from previous work performed by Fava et al.²³ and Riva et al.²¹ on linking HRQoL questionnaires to the ICF/ICF-CY classification.

METHODS

Haemo-QoL is the first HRQoL instrument developed specifically for children with hemophilia, which has been used in a number of clinical studies investigating pediatric hemophilia populations.²⁵ To respect the developmental challenges in time, the age versions differ in the number of items and domains. The Haemo-QoL version for 4- to 7-year-old children contains 21 items pertaining to eight domains, the version for 8- to 12-year-olds has 64 items

pertaining to ten domains, and the version for 13- to 16-year-olds includes 77 items from 12 domains. The eight domains common for all three age-related versions are as follows: “physical health,” “feelings,” “view of oneself,” “family,” “friends,” “others,” “sports and school,” and “treatment.” Additional domains for the version for 8- to 12-year-olds and 13- to 16-year-olds are “perceived support” and “dealing with hemophilia,” and the domains relevant only for version for the 13- to 16-year-olds are “future” and “relationships.” For the 4- to 7-year-olds’ version, a 3-point Likert response scale (never, sometimes, very often) is used, whereas a 5-point Likert scale (never, seldom, sometimes, often, all the time) is used for age group 8–12 yrs and 13–16 yrs. The Haemo-QoL is linguistically validated in 40 languages.²⁶

The ICF consists of four components: body functions and body structures relate to the physiologic functions of body systems (e.g., digestive functions or higher-level cognitive functions) and anatomical parts of body such as organs, limbs and their components. Activity and participation relates to the execution of a task or an action and to an involvement in a whole spectrum of life situations (e.g., dressing, preparing meals, or forming relationships). Environmental factors refer to the physical, social, and attitudinal environment in which people live (e.g., drugs or friends). Personal factors comprise features of an individual that are not part of a health condition or health state such as sex, race, age, lifestyle, and others. As mentioned, personal factors are not classified in the current version of ICF/ICF-CY. Categories are the basic units of ICF/ICF-CY classification and can be understood as “discrete, meaningful, universally shared, and understood elements which allow users to comprehensively classify and measure functioning of individuals and populations.”¹⁸ Each category starts with a letter (b, s, d, or e) followed by the chapter number (one digit) and, if a category refers to more specific aspects, the second-level number (two digits) and the third- and fourth-level numbers (one digit each). On the second-level categories, used in the current study, there are 362 categories. Each category can be assigned a qualifier that uses a generic 7-point scale (0, no problem; 1, mild problem; 2, moderate problem; 3, severe problem; 4, complete problem; 5, not specified; 6, not applicable).

With use of qualifiers, activity limitations and participation restrictions can be recognized within categories from activities and participation, impairments can be recognized in the areas of body structures and body functions, and facilitators and barriers are assigned to contextual factors.

To describe functioning using ICF categories, two steps are needed; first is linking the measurement tool to specific ICF categories following the linking rules,^{27,28} and second is transforming information obtained from the measurement tool into ICF qualifiers¹⁸ based on ICF check list.²⁹ Items from the Haemo-QoL questionnaire have been linked to the ICF questionnaire following the established rules²⁸; the linking process as well as its result have been presented elsewhere.²¹ In this study, we have used the results of the linkage process to select ICF/ICF-CY categories for the description of health and functioning of children with hemophilia and to choose one item from the Haemo-QoL that would represent each on the previously selected ICF/ICF-CY categories. We selected two categories from body function and body structures and from activities and participation and one category from the environmental factors based on how frequently each category was linked to the items from Haemo-QoL questionnaire, and those categories that were most frequently linked were selected. This was based on the assumption that the most frequently linked categories would also be the most relevant for the description of health and functioning of children with hemophilia.

It was then decided which item would represent the particular ICF/ICF-CY category. The following rules were followed: only items that were linked to one ICF/ICF-CY category were considered, and among the items confirming this requirement, the items that have been assigned to a particular ICF/ICF-CY category with the higher interrater agreement were selected.

The Likert-scaled response options from the Haemo-QoL were transformed into ICF/ICF-CY qualifiers to calculate the frequency of impairments, restrictions, and barriers in the study population, with the frequencies being an estimation of the prevalence of the above constructs. We decided not to include an age group (4–7 yrs) in the analysis because the response categories for this age group consisted of three categories only. For the two older age groups (8–12 and 13–16 yrs), the response scales ranged from 1 to 5, which coincides with the number of ICF/ICF-CY qualifiers. In the ICF check list,²⁹ the qualifiers are defined as follows: 0, no problem; 1, mild problem, which means that a problem is present less than 25% of the time; 2, moderate problem, which means that a problem that is present less than 50% of the time; 3, severe problem, which means that a problem is present more than 50% of the time; and 4, complete problem, which means that a problem is present more than 95% of the time. Following the similarities

between the definitions of qualifiers from the ICF checklist and the response scale from the Haemo-QoL questionnaire, responses from the Likert scale were transformed into ICF qualifiers as follows: “never,” 0 (no problem), “seldom,” 1 (mild problem); “sometimes,” 2 (moderate problem); “often,” 3 (severe problem); and “all the time,” 4 (complete problem). The frequencies of impairments, activity limitations, participation restrictions, and barriers for children with hemophilia in Europe were calculated using the data from the ESCHQoL study, in which 446 children participated. Additional clinical data was provided for each child. This enabled the comparisons of different groups in terms of severity of hemophilia and mode of treatment administration.

RESULTS

Overall, the most frequently linked category was b152 Emotional functions. Emotional function is defined as mental functions related to feelings and to the affective components of the processes of the mind; it was identified 12 times in the Haemo-QoL. Following Emotional functions, the second most frequently identified category in the body structures and body functions domains was b280 Sensation of pain, defined as the sensation of unpleasant feeling indicating potential or actual damage to some body structure. This category was identified three times. From the activities and participation component, two categories were identified most frequently (five times each), these were d920 Recreation and leisure and d750 Informal social relationship. d920 is defined as engaging in any form of play; recreational or leisure activity, such as informal or organized play and sports; programs of physical fitness; relaxation; amusement or diversion; going to art galleries, museums, cinemas, or theatres; engaging in crafts or hobbies; reading for enjoyment; playing musical instruments; and sightseeing, tourism, and traveling for pleasure. Category d750 is defined as entering into relationships with others, such as casual relationships with people living in the same community or residence or with coworkers, students, playmates, or people with similar backgrounds or professions. Finally the category most frequently (five times) identified in the environmental factors component was e410 Individual attitudes of immediate family members, defined as general or specific opinions and beliefs of immediate family members about the person or about other matters (e.g., social, political and economic issues) that influence individual behavior and actions.

For each of the five categories that were most frequently linked to the Haemo-QoL questionnaire,

TABLE 2 Frequency of reported restrictions in the area of sports-related activities and communication with friends in the population of children with hemophilia in Europe

ICD/ICF-CY Qualifier	Haemo-QoL Item "I was Worried Because of my Hemophilia" ICF-CY Code b152 Emotional Functions						Haemo-QoL Item "I had Pain in my Joints" ICF-CY Code b280 Sensation of Pain					
	Age Group		Treatment		Severity of Hemophilia		Age Group		Treatment		Severity of Hemophilia	
	8-12 yrs (n = 184)	13-16 yrs (n = 168)	On-Demand (n = 135)	Prophylaxis (n = 156)	Mild and Moderate (n = 98)	Severe (n = 193)	8-12 yrs (n = 186)	13-16 yrs (n = 159)	On-Demand (n = 134)	Prophylaxis (n = 154)	Mild and Moderate (n = 97)	Severe (n = 191)
0 (no restriction)	38.00%	41.70%	35.6%	48.1%	43.9%	41.5%	20.60%	23.90%	36.6%	26.0%	42.3%	25.1%
1 (mild restriction)	20.00%	14.91%	17.3%	17.4%	21.6%	12.6%	27.00%	11.90%	17.9%	21.4%	15.5%	22.00%
2 (moderate restriction)	19.00%	41.70%	35.6%	48.1%	43.9%	41.5%	20.60%	17.60%	14.9%	20.1%	11.3%	20.9%
3 (severe restriction)	14.10%	14.90%	14.8%	11.5%	12.2%	13.5%	24.20%	18.20%	9.0%	7.8%	5.2%	9.9%
4 (complete restriction)	13.00%	14.90%	16.3%	9.0%	13.3%	11.9%	32.80%	28.30%	21.6%	24.7%	25.8%	22.0%

ICF/ICF-CY, International Classification of Functioning, Disability and Health/International Classification of Functioning, Disability and Health-Children and Youth Version.

TABLE 3 Frequency of reported barriers in the area of parental protection and personal factors in the population of children with hemophilia in Europe

ICD/ICF-CY Qualifier	Haemo-QoL Item "I was Worried Because of my Hemophilia" ICF-CY Code b152 Emotional Functions						Haemo-QoL Item "I had Pain in my Joints" ICF-CY Code b280 Sensation of Pain					
	Age Group		Treatment		Severity of Hemophilia		Age Group		Treatment		Severity of Hemophilia	
	8-12 yrs (n = 184)	13-16 yrs (n = 161)	On-Demand (n = 132)	Prophylaxis (n = 154)	Mild and Moderate (n = 95)	Severe (n = 191)	8-12 yrs (n = 185)	13-16 yrs (n = 161)	On-Demand (n = 133)	Prophylaxis (n = 154)	Mild and Moderate (n = 96)	Severe (n = 191)
0 (no restriction)	28.80%	37.30%	29.5%	38.3%	40.0%	31.4%	47.03%	32.92%	41.4%	44.8%	53.1%	38.2%
1 (mild restriction)	18.50%	20.50%	18.9%	22.1%	22.1%	19.9%	22.70%	33.54%	24.8%	29.9%	25.0%	28.8%
2 (moderate restriction)	15.30%	18.60%	18.9%	15.6%	16.8%	17.3%	17.84%	16.77%	14.3%	16.2%	12.5%	16.8%
3 (severe restriction)	9.50%	11.20%	9.1%	9.1%	7.4%	9.9%	7.57%	8.07%	10.5%	5.8%	4.2%	9.9%
4 (complete restriction)	25.90%	12.40%	23.5%	14.9%	13.7%	21.5%	4.86%	8.70%	9.0%	3.2%	5.2%	6.3%

ICF/ICF-CY, International Classification of Functioning, Disability and Health/International Classification of Functioning, Disability and Health-Children and Youth Version.

receiving prophylaxis treatment, and children with severe hemophilia reported more participation restrictions in this area compared with children with mild or moderate hemophilia.

Finally, for the category e410 Individual attitudes of immediate family members, the item “my mother protected me too much because of my haemophilia” from the subscale Family was chosen, and for the personal factors domain, the item “Haemophilia made my life more difficult” from the View of yourself subscale was chosen. The frequency of reported barriers in the area of parental protection and personal factors is presented in Table 3

Children receiving on-demand treatment, children with severe hemophilia, and younger children reported higher level of barriers in the area of Individual attitudes of immediate family members compared with children receiving prophylaxis treatment, older children, and children with mild/moderate hemophilia. Younger children and children with mild or moderate hemophilia perceived that their hemophilia is making their lives difficult less frequently than older children and children with severe hemophilia.

DISCUSSION

To describe and analyze functioning and disability in a population of children with hemophilia in Europe, HRQoL items were mapped onto the ICF/ICF-CY categories.²¹ The frequencies of problems across ICF/ICF-CY categories were calculated based on the European data set on quality-of-life of children with hemophilia. Considering that categories from all ICF/ICF-CY domains enabled broad analyses of functioning and disability, including an analysis of the impact of, emotional functions and sensation of pain were the two most relevant categories within body functions and body structures in the population studied. In the activities and participation component, recreation and leisure as well as informal social relationships were the most important categories. Finally, Individual Attitudes of Immediate Family Members was the most important category representing the impact of environmental factors. From the considerable number of Haemo-QoL items linked to personal factors, one item (“Haemophilia made my life more difficult”) was selected to represent this area in the analyses.

More than 60% of the sample reported no or only minor impairment in the area of emotional functions, indicating high emotional well-being. More impairment was found in the area of pain. The major cause of pain in hemophilia is arthropathy in joints. As the level of arthropathy increases

with time and number of joint bleeds, older children and those receiving on-demand treatment are more impaired in this area. It may also be expected that children with severe hemophilia will experience more bleeds (leading to arthropathy) than children with mild hemophilia, although children with severe hemophilia tend to receive prophylaxis treatment preventing bleedings more often than children with mild/moderate haemophilia.

The restriction of functioning in relation to recreation and leisure seemed to be most affected by receiving on-demand treatment. This might be related to the fact that children receiving on-demand treatment might be restricted in strenuous free time activities to avoid bleedings. Interestingly, children receiving on-demand treatment seemed to have fewer restrictions in the area of informal social relationship compared with children receiving prophylaxis treatment. This might reflect the time needed and the potential family conflicts associated with the continuous administration of prophylactic treatment. This might also explain why younger children (8–12 yrs) perceive more barriers in relation to the individual attitudes of immediate family members than older children (13–16 yrs).

It can be noticed that the level of impairments in the body functions and body structures was lower than the level of participation restrictions in the area of activities and participation. This may indicate that the reason of participation restriction is not directly linked to impairments in body functions and body structures but is rather associated with other factors. These can be related to environmental factors such as attitudes of teacher, peers, and parents toward children with hemophilia and to the stigma associated with this disease. This might be particularly true for children receiving prophylaxis treatment because they do not develop arthropathy and the development of other complications related to hemophilia are very rare in prophylactic patients.

The study is subject to some limitations. One is that some more sophisticated methods, such as Rasch analysis,³⁰ could have been applied to the translation process of HRQoL responses into ICF/ICF-CY qualifiers. Further reflection on how to translate the HRQoL data into ICF/ICF-CY is needed. Secondly, only one disease-specific instrument measuring HRQoL in children with hemophilia was included in this study. The data on chronic generic HRQoL was available; however, the specific HRQoL used in the study was not linked to the ICF/ICF-CY categories, so the available data could not have been used to translate it into the ICF/ICF-CY qualifiers. Because different aspects of health and functioning

are covered in chronic-generic and disease-specific measures, some aspects of health and functioning of children with hemophilia might be missing in the analyses.²³

Comparison between the Haemo-QoL questionnaire and ICF/ICF-CY classification serves as an example of a cross-walk between HRQoL and the ICF/ICF-CY approach to measuring health, revealing the strengths and possible limitations of both approaches and providing arguments that support the validity of the ICF/ICF-CY classification. HRQoL instruments are constructed based on the subjective perception of health and quality-of-life of the persons in question (by means of focus groups or interviews). For example, similarities can be found between Haemo-QoL domains (e.g., friends, treatment) and ICF/ICF-CY higher-order categories (e.g. d750 Informal social relationship, e580 Health services, systems, and policies).

Contrary to the similarities found on the higher level, linking particular items to the ICF/ICF-CY categories revealed important differences between Haemo-QoL items and ICF/ICF-CY categories. In most cases, more than one category was assigned to a Haemo-QoL item, and often, it was impossible to assign any category to the Haemo-QoL item. This result was also found in linkage exercises with the DISABKIDS work.²³ This indicates some important differences between HRQoL items and ICF-categories, which should be taken into account when using items from HRQoL measures to report on health and functioning using ICF/ICF-CY classification.

Finally, it is important to mention that although the area of emotional functioning is widely represented in the Haemo-QoL instrument (as well as in HRQoL measures in general), on the second level, only one related category (b152 Emotional functions) can be found in the ICF/ICF-CY classification, and on the third level, additional five categories (b1520, b1522, b1528, and b1529) were found.

Therefore, the recommendation is that emotional aspects of health and functioning can be expanded in the future versions of ICF and ICF-CY.

More analyses should be performed (including chronic-generic measures) to identify conceptual similarities and differences between HRQoL and ICF/ICF-CY, to refine the focus of HRQoL items, and to expand the validity of ICF/ICF-CY classification in the assessment of health.

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