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Original Article

Translation, cultural adaptation, and validation of the Indonesian version of *Pediatric Quality of Life Inventory Rheumatology Module* (PedsQL-RM) questionnaire for children with rheumatic diseases

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Abstract

Background Quality of life is an important outcome in the management of children with chronic conditions such as rheumatic diseases. *The Pediatric Quality of Life Inventory Rheumatology Module* (PedsQL-RM) questionnaire has been proven valid and reliable, but a validated Indonesian version of the questionnaire is not yet available.

Objective To translate the PedsQL-RM into the Indonesian language, perform a transcultural adaptation, and assess its validity.

Methods A cross-sectional study was performed in patients aged 2 to 18 years with systemic lupus erythematosus (SLE) or juvenile idiopathic arthritis (JIA) in Dr. Cipto Mangunkusumo Hospital, Jakarta. The initial phase of the study consisted of forward translation from the original English version into Indonesian, synthesis by experts, backward translation, and cognitive debriefing, resulting in the final version of the questionnaire. The second phase was testing the final questionnaire on patients in each age group and their parents. Tests were carried out in two sessions with an interval of 2 to 4 weeks. The questionnaire consisted of a child report and a parent report, each measuring five dimensions: pain and hurt, activities, treatment, worry, and communication. We subsequently assessed validity and reliability of each dimension in the child and parent reports for the child and teen age groups. Validity was expressed as correlation coefficient (r) between dimension scores with the total score.

Results The finalized Indonesian questionnaire was completed by 53 children aged 2-18 years with SLE or JIA and their parents. Due to small numbers of subjects in the younger age ranges, analysis was only performed in the 8-to-18-year age group. Validity varied from good to very good (r=0.437 to 0.910) for the child report and from poor to good (r=0.153 to 0.808) for the parent report. The questionnaire was deemed reliable, with a Cronbach's alpha of 0.755 to 0.785.

Conclusion The Indonesian version of the PedsQL-RM is valid and reliable for assessing quality of life in children aged 8 to 18 years with rheumatologic disease. Further study is needed to assess the validity and reliability of the tool for children aged 2 to 7 years. **[Paediatr Indones. 2023;63:139-45; DOI: https://doi.org/10.14238/pi63.2.2023.139-45**].

Keywords: *quality of life; PEDSQL-RM; validity; reliability*

Rheumatic diseases in children consist of several autoimmune and inflammatory conditions.[1] The most common are systemic lupus erythematosus (SLE) and juvenile idiopathic arthritis (JIA). SLE can affect all organ systems and has a wide range of clinical manifestations.[2] In JIA, inflammation mainly affects joints, causing chronic pain and limitations in movement.[3] To date, there is no cure for rheumatic diseases, but remission can be achieved with treatment. Treatment goals include inflammation reduction, pain management, quality of life optimization, and, whenever possible, disease remission. Management consists of medical therapy, physical activities, and nutritional support.[1]

Rheumatic disease in children can cause high morbidity and mortality from both the disease and the effects of treatment. Since the disease can be progressive, quality of life is a major aspect of management.[4] Many studies have reported that rheumatic disease negatively affects quality of life.[5] Despite improvement in treatment quality and survival in SLE patients, their quality of life remains low.[6] Around 65% of SLE patients have chronic

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fatigue, 40% experience pain, 37% have anxiety, and 30% are depressed.[7] A Swedish study in children with JIA also reported a suboptimal level of quality of life.[8]

It is important to appropriately measure quality of life (QoL) in the management of children with rheumatic diseases using a valid and reliable instrument. *The Pediatric Quality of Life Inventory – Generic Core* (PedsQL-GC) Module is commonly used in Indonesia to assess the QoL of children in general. This instrument is widely accepted, can be used in both healthy and sick children, but it is not designed for specific conditions or diseases.

Rheumatic diseases possess typical characteristics, such as chronic onset, fluctuating disease activity, and long-term treatment.[4] It is important to assess QoL using a specific tool to produce a more accurate measurement. *The Pediatric Quality of Life Inventory Rheumatology Module* (PedsQL-RM) questionnaire specifically measures QoL in children with rheumatologic diseases. The questionnaire has been widely used in many countries and its validity and reliability have been well established.[9] Since a validated Indonesian version is not yet available, we aimed to translate and assess the validity and reliability of an Indonesian version of the PedsQL-RM. An Indonesian version of the questionnaire will be useful for children with rheumatologic diseases and their parents whose primary language is Indonesian. Accurate and objective scores can be used as reference to arrange for better management for the patient in the hope of increasing QoL and yielding better outcomes.

Methods

This validity study was performed on patients aged 2 to 18 years diagnosed with SLE or JIA at Dr. Cipto Mangunkusumo Hospital from January to April 2020. Exclusion criteria were intellectual disability, autism, Down syndrome, cerebral palsy, or parental illiteracy.

The initial phase of the study consisted of forward translation of the Peds-QL-RM from the original English version to Indonesian, synthesis by experts, backward translation, and cognitive debriefing, resulting in the final version of the questionnaire. Synthesis was done through discussions on the questions included in the translated questionnaire. In cognitive debriefing, we discussed the Indonesian version that had been tested by a small number of users. During this process, word choice and ambiguity are evaluated. The distributor, *Mapi Research Trust* (Lyon, France), has granted permission to translate and culturally adapt the tool. The steps for translation and adaptation are shown in **Table 1**.[10] The second phase aimed to test the final questionnaire in patients in each age group and their respective parents. The test was carried out in two sessions with an interval of two to four weeks between the test and retest.

The questionnaire is available for toddlers (aged two to four years), young children (aged five to seven years), children (8-12 years), and teens (13-18 years), and needs to be completed by both the patient and their parents.[2] For each age group, there is a child report and a parent report. Each report is scored separately and measures five dimensions: pain and hurt (four items), activities (five items), treatment (seven items), worry (three items), and

Table 1. Translation and adaptation of the questionnaire[10]

Step	Translator	Results
First translation	Two native Indonesians; one with a medical background who understood the questionnaire material and one layperson.	Translated version A and B
Synthesis of the translation and expert review	An allergy and immunology expert, methodology experts, and language and cultural experts reviewed and synthesized version 1translated versions A and B into one version.	Version 1 questionnaire
Backward translation	Native layperson English speakers translated the Indonesian version 1 back to English. They compared the resulting translation to the original.	Version 2 questionnaire
Cognitive debriefing	Version 2 of the questionnaire in Indonesian was tested on 10 children with similar characteristics to the study subjects.	Revised questionnaire (pre-final)
Final review by experts	Review by medical and language/cultural experts of the revised questionnaire to produce the final version.	Final questionnaire in Indonesian

communication (three items). Items were scored on a five-point Likert scale from 0 (never) to 4 (almost always), then reverse-linearly transformed to a 0-to-100 scale as follows: 0=100, 1=75, 2=50, 3=25, and 4=0. The dimension score was the mean of the transformed item scores in the dimension.[9]

Data were processed to produce validity and reliability scores. We used *SPSS version 27.0* software (*IBM*, Armonk, New York) to help with data analysis. Cronbach's alpha coefficient and intraclass correlation coefficient (ICC) were used to measure inter-observer reliability. Cronbach's alpha scores of 0 to 0.2 were defined as unreliable, 0.21 to 0.40 as poorly reliable, 0.41 to 0.60 as moderately reliable, 0.61 to 0.80 as reliable, and 0.81 to 1 as excellently reliable. An instrument with a minimum Cronbach's alpha of 0.6 and ICC of 0.5 to 0.75 was deemed reliable. Internal consistency was measured by ICC for each version (children's and parents'), with scores classifed as poor if ICC was <0.5, moderate if it was 0.5 to 0.75, good if it was 0.75 to 0.9, and excellent if it was >0.9.[11]

For purposes of this study, we calculated a total score that was the sum of all domain scores. We performed Pearson's correlation between each domain score and the total score. The questionnaire was considered valid for a dimension if there was a good, statistically significant correlation between the dimension score and the total score. A correlation coefficient (r) of 0 to 0.2 showed poor correlation, 0.21 to 0.4 low correlation, 0.41 to 0.6 good correlation, 0.61 to 0.8 strong correlation, and 0.81 to 1 very strong correlation.[12] We also assessed the correlation between the child and parent reports using Spearman's rank correlation. The 95% confidence interval (95%CI) was reported where appropriate. A P value of <0.05 was considered statistically significant. This study protocol had been approved by the Medical Research Ethics Committee of Universitas Indonesia.

Results

A total of 53 subjects were included in the study, which consisted of five patients in the toddler group, six in the young child group, 17 in the child group, and 25 in the teen group. The majority of subjects were girls (83%) with SLE (79.2%) and had already been diagnosed more than six months prior to the study period. Due to small numbers of subjects in the toddler and young child groups, statistical analysis was performed only on the child and teen groups. Subject characteristics can be seen in **Table 2**.

In the child age group, the child report had correlation coefficients that ranged between 0.437 to 0.919 (good to very strong correlation). We found statistically significant correlations between the dimension and total scores for activities, treatment, worry, and communication parameters. In the teen group, the correlation coefficients ranged between 0.451 to 0.807 (good to strong correlation), with P<0.05 for all parameters.

The validity of the parent report varied between poor to good (r=0.153 to 0.808). In the child age group, the parent report had good to strong correlations between dimension and total scores (r=0.577 to 0.808), with P<0.05 for pain and hurt, treatment, worry, and communication. In the teen age group, the parent report showed good to strong correlation in all dimensions (r=0.533 to 0.714), with P<0.05. The correlations between dimension and total scores for both the child and parent reports are listed in **Table 3**. Cronbach's alpha was 0.755 to 0.785, indicating that the questionnaire was reliable, as shown in **Table 4**.

For the child report, ICC was good and excellent in all parameters except for daily activities in the teen age group (ICC=0.251). However, for the parent report, most ICC scores were <0.5 (poor) (**Table 5**).

We assessed for correlations between the child and parent reports for each dimension (**Table 6**). Correlations for pain and hurt were good in all age groups (rho=0.604, P=0.01 the child age group and rho=0.692, P<0.001 for the teen age group). The correlation for the "Treatment" dimension in the child age group was good (rho=0.605, P=0.01), but correlations between child and parent reports for all other parameters were low and statistically not significant.

Discussion

While the majority of our subjects were SLE patients (79.2%), similar studies in California and Italy had only 9.1% of subjects with SLE,[9] with most of their subjects being children with fibromyalgia.[13]

Table 2. Patient's characteristics

Characteristics	2-4 years (n=5)	5-7 years (n=6)	8-12 years (n=17)	13-18 years (n=25)	Total (N=53)
Gender, n(%)					
Male	2	2	3	2	9 (17.0)
Female	3	4	14	23	44 (83.0)
Diagnosis, n(%)					
SLE	0	4	14	24	42 (79.2)
JIA	5	2	3	1	11(20.8)
Father's occupation, n(%)					
Employee	4	4	10	15	33 (62.2)
Others	1	2	7	10	20 (37.8)
Mother's occupation, n(%)					
Housewife	3	5	14	20	42 (79.2)
Working mother	2	1	3	5	11 (20.8)
Father's education, n(%)					
Primary-junior high school	3	1	5	8	17 (32.1)
Senior high school	2	2	11	15	30 (56.6)
University	0	3	1	2	6(11.3)
Mother's education, n(%)					
Primary-junior high school	1	2	6	9	18 (34.0)
Senior high school	4	2	10	13	29 (54.7)
University	0	2	1	3	6 (11.3)
Onset of disease, n(%)					
<6 months	2	3	4	2	11 (20.8)
>6months	3	3	13	23	42 (79.2)

Table 3. Validity in children and parents' domains

Domain	Children		Parents	
	Children group	Teen group	Children group	Teen group
Pain and hurt	0.437	0.772*	0.622*	0.596*
Activities	0.622*	0.451*	0.153	0.533*
Treatment	0.910*	0.769*	0.787*	0.714*
Worry	0.501*	0.807*	0.808*	0.767*
Communication	0.756*	0.757*	0.577*	0.654*

*statistically significant

Table 4. Reliability of the Indonesian version of the questionnaire (Cronbach's alpha)

Variables	Children group	Teen group
Children's total score	0.759	0.785
Parents' total score	0.760	0.755

Other rheumatic diseases included in prior studies on this questionnaire were dermatomyositis and spondyloarthritis.[9] Around 83% of our subjects were girls, similar to the study in California (76.1%).[9] The incidence of rheumatologic disease was much higher in girls, as it is associated with the role of estrogen in the development of autoimmune diseases. The predilection of rheumatic disease in girls vs. boys is estimated to be 3 to 6.6:1 for JIA and 3-5 : 1 in SLE.[14]

In our study, 42 patients were aged >8 years. All children <5 years of age had a diagnosis of JIA. Our findings were in agreement with a previous study which found that the incidence of rheumatic diseases, especially SLE, was highest during puberty, while JIA was more common in younger aged children.[15]

The majority of subjects had been diagnosed six months or more prior to the study, so parents were already quite familiar with the disease and management. Most parents had graduated from senior high school, while the lowest educational level was primary school graduate. All respondents

Table 5. Intraclass correlation coefficient (95%CI)

Domain	Report			
	Children's		Parents'	
	Children group	Teen group	Children group	Teen group
Pain and hurt	0.749 (0.43-0.901)	0.883 (0.752-0.946)	0.053 (0.042-0.508)	0.294 (0.107-6.12)
Activities	0.983 (0.952-0.994)	0.251 (-0.152-0.582)	0.028 (-0.490-0.446)	0.318(-0.547-0.700)
Treatment	0.957 (0.886-0.984)	0.752 (0.551-0.883)	0.012 (-0.459-0.478)	0.009 (-0.396-0.381)
Worry	0.834 (0.601-0.851)	0.878 (0.744-0.853)	0.442 (-0.147-0.799)	0.020 (-0.405-0.371)
Communication	0.840 (0.614-0.939)	0.968 (0.929-0.986)	0.085 (-0.609-0.491)	0.099 (-0.301-0.469)

Table 6. Correlations between children's and parents' reports

Domain	Age group			
	Children group		Teen group	
	r	P value	r	P value
Pain and hurt	0.604*	0,010	0.692*	<0,001
Activities	0.446	0,073	0.382	0,059
Treatment	0.605*	0,010	0.177	0,396
Worry	0.432	0,083	0.230	0,270
Communication	0.035	0,895	0.151	0,471

*Spearman<0.05

completed the questionnaire, which indicated that the questionnaire was generally easy to understand, similar to a previous study which reported missing data in less than 2% of subjects.[16]

Validity is a reflection of the accuracy of an instrument to measure certain values.[17] We assessed the validity of the Indonesian version of the questionnaire by assessing the correlations between each dimension score and the total score for both the child and parent report in each age group. The validity of the children's domain varied (r=0.437 to 0.910) with good to strong correlations. Our findings indicate that all translated items are suitable and appropriate to represent the dimension.

Validity of the parent report varied between poor to good (0.153-0.808). In the child age group, the dimensions of pain and hurt, treatment, worry, and communication were all good and had strong, statistically significant correlations with the total score (r=0.577 to 0.808). In the teen age group, all dimensions had good to strong correlations with the total score (r=0.533 to 0.767), indicating that all five dimensions in the parent report were well-described by the translated questions, despite having lower correlation coefficients compared to those of the child report.

A poor correlation coefficient was found in the activities dimension of the child age group parent report, although the finding was not statistically significant. Items in the dimension explored obstacles in daily activities from parents' perspective. The items' sentence substance and structure can lead to different perspectives and views on the topic, so answers might have varied. While translating and adapting the items, we incorporated cultural values without changing the construction or material from the original questionnaire, so the poor correlation in this dimension needs further examination. Other factors that may have contributed to the result, such as educational background, was already minimized by the fact that the parents' lowest level of education was elementary school graduate. Overall, our results were in agreement with another validation study performed in California, which found good to strong correlation.[9]

We also assessed correlations between child and parent reports for each dimension. A strong and significant correlation was found in the pain and hurt dimension in both age groups, and in the treatment parameter in the child age group. However, poor correlations were noted in the communication dimension in both age groups and in the treatment dimension in the teens group. Similarly, a study found

that the correlation was strongest for the pain and hurt dimension (0.744) and weakest for the communication dimension (0.285).[16] The items for the communication and treatment dimensions are subjective, so there can be differences between children's and their parents' answers, in relation to parental perspectives and children's openness to parents. Parents and children do not always share similar perspectives on QoL.[14] In self-reported questionnaires, subjects tend to give socially acceptable responses based on an impression they want to create, so the QoL assessment of the PedsQL questionnaire is highly recommended for both parents and children, in order to yield more comprehensive views.[18]

Reliability refers to the instrument's ability to consistently produce similar results in repeated measurements. Low reliability can be caused by disagreement between the raters or the instability of the subject being measured. There are three main measurements for instrument reliability: test-retest reliability, alternate-form reliability, and internal consistency reliability. Internal consistency can be measured using Cronbach's Alpha.[15,17] Although reliability is important, its role is insufficient if not supported by good validity.[11]

The Indonesian version of the questionnaire was found reliable, with Cronbach's alpha values ranging from 0.755 to 0.785. Our findings show that all items yielded the same result in repeated measurements, hence rendering the questionnaire a consistent instrument to assess QoL. This result was similar to those of several previous studies. In the California study, the questionnaire was deemed reliable with a Cronbach's alpha of 0.75 to 0.86 for the child report and 0.82 to 0.91 for the parent report.[9] A previous study noted good reliability, with Cronbach's Alpha of 0.68-0.86.[16] An Italian study reported a Cronbach's alpha of 0.7 for both the child and parent reports.[11]

The ICC is a common index to describe consistency in test-retest reliability. The test-retest was performed in 2-to-4-week intervals. For the child report, ICC was good and excellent in all domains, except for daily activities for the teen group. However, the parent report resulted in a low ICC. These findings are similar to those of a study in 24 children with SLE, which reported an ICC of 0.5 (95%CI 0.04 to 0.7).[4] This difference might have been due to changes in patient within the time interval, either clinically or psychologically. The PedsQL-RM questionnaire is known to be sensitive in detecting changes over time.[19] The ICC is actually difficult to apply to this questionnaire because of its high sensitivity to any clinical changes.[20]

This is the first translation, cultural adaptation, and validation of PedsQL-RM into Indonesian. The questionnaire is easy to understand, has a low risk of missing data, an can be completed in a short time, hence, it is applicable to both inpatients and outpatients. The Indonesian version of the PedsQL-RM is a reliable and valid instrument for QoL measurement in children with rheumatic diseases. Further study is needed to validate the questionnaire in younger children (2-7 years). Using this version of the tool, the QoL of children with rheumatic diseases in Indonesia can be measured more accurately and may inform patient management decisions.

Conflict of interest

None declared.

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