Development of a Spanish (Castillian) version of the Childhood Health Assessment Questionnaire. Measurement of health status in children with juvenile chronic arthritis

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Abstract

Objective
To demonstrate that the Spanish (Castillian) version of the Childhood Health Assessment Questionnaire (cHAQ-S) is a valid and reliable instrument for measuring the health status of children with juvenile chronic (or rheumatoid) arthritis (JCA) and is sensitive to change.

Methods
A conceptual translation of the original questionnaire into Spanish and two back-translations were performed. The cHAQ-S was completed by the parents of young children (aged 1 to 19 years) affected by JCA, and additionally by those children aged over 9. A second cHAQ-S was administered at least 15 months after the first one.

Results
The cHAQ-S was administered to 79 parents of patients affected by JCA. The test-retest reliability was evaluated among 16 patients, and no significant differences between the first and second administration were found (0.88 versus 0.84; p > 0.6; intraclass correlation coefficient R = 0.94). The Cronbach’s alpha coefficient was 0.948, indicating an excellent internal reliability with a mean correlation between the different components of the questionnaire varying from 0.3557 to 0.7831. For the between-observer reliability, an intraclass correlation coefficient of 0.96 was obtained. Correlations between DI (Disability Index) and several measures of disease activity were all statistically significant (Spearman’s R ranged from 0.42 to 0.87; p < 0.005). Patients who improved showed similar improvement in the DI (p = 0.015), while patients who worsened showed a worsening of the DI (p = 0.1) and patients whose condition was stable showed no change in DI (p = 0.6).

Conclusion
The cHAQ-S is a feasible, reliable and valid instrument for the determination of the health status of Spanish children suffering from JCA. It is also sensitive to changes in the child’s health status.

Key words
Juvenile rheumatoid arthritis, disability evaluation, quality of life, questionnaires, outcome assessment, sensitivity to change.

Introduction

Due to its chronic and variable course, juvenile chronic (or rheumatoid) arthritis (JCA) requires periodic check-ups by a pediatric rheumatologist. The evaluation and control of the patient suffering from JCA has been based, until now, on clinical and laboratory parameters. Both measurements have the disadvantage of only partially evaluating the patient’s disease status, ignoring such important aspects as the ability to carry out daily activities, the presence of pain, or the patient’s own evaluation of his/her state of health. These traditional measurements are usually more concerned with quantifying disease activity than the impact that inflammation specifically, and the disease process in general, may have on the health status of the sufferer. Indeed, some of these measurements have poor sensitivity, making the detection of changes in the health status all the more difficult (1, 2).

As a result of these difficulties, several new instruments have been developed with the aim of quantifying the consequences of this disease and evaluating the health status of children with JCA. They are specific questionnaires developed for children with rheumatic diseases that are comprised of a series of items that have to be completed by the patient and/or his/her parents. None of these instruments have been validated in our setting.

There have been several specific instruments developed with the aim of evaluating the health of children with JCA (3), such as the Pediatric Arthritis Impact Measurement Scales (pediatric AIMS) (4), the Juvenile Arthritis Functional Assessment Report (JAFAR) (5), and the Childhood Health Assessment Questionnaire (cHAQ) (6) which is a modification of the Health Assessment Questionnaire (HAQ) designed for adults (7).

The cHAQ is a questionnaire that can be self-administered in older children or can be completed by the parents in the case of younger children. It has been shown to be suitable in patients aged between 1 and 19 years. It is short and easy to use; it does not require an interviewer and can be filled out in under 10 minutes. This makes it a useful instrument for day-to-day clinical practice.

Another advantage is that the questionnaire allows the pediatrician to undertake follow-up of the rheumatic patient from infancy to adulthood using the same standardised instrument. The cHAQ has been shown to have excellent reliability and validity, higher than that of any of the other instruments developed for assessing JCA (3). Currently, in addition to the original American-English questionnaire, versions of the cHAQ have been validated in Brazilian-Portuguese (8), Swedish (9) Italian (10) and Norwegian (11), all with excellent results. Recently, a further three Hispanic versions have been validated: in Costa Rican Spanish (12), Mexican Spanish (13) and Argentinean Spanish (14).

These versions have been semantically and culturally adapted to their respective countries. They contain, however, expressions that would be strange and not easily understood in our setting, meaning that they should not be applied in Spain (15).

The aim of this study was to develop a Spanish (Castillian) version of the Childhood Health Assessment Questionnaire (cHAQ-S) and demonstrate that it is a valid and reliable instrument for measuring the health status of children with JCA and that it is a feasible procedure for day-to-day clinical practice. A secondary aim was to assess the sensitivity to change of the cHAQ-S.

Materials and methods

Translation of the questionnaire

The original questionnaire was translated into Spanish by a group of clinicians made up of two paediatricians, a paediatric-rheumatologist, a neuro-paediatrician and a rehabilitation specialist. These professionals were instructed to undertake a conceptual translation rather than a literal one, and they were allowed to change any items should they feel that the item did not correspond to the habits and customs in our country. Once consensus was obtained, a professional English-Spanish translator translated this first Spanish version back into English. After modifying discrepancies between the two versions and clearing up any remaining doubts, a second Spanish version was elaborated and was once again translated back into English, this time by
Spanish cHAQ validation / J.-J. García-García et al.

an English speaking epidemiologist who was unaware of the original questionnaire.

The items were then classified into three categories according to their agreement between the two versions, using the method proposed by Alonso and Antó (16). Items in category A were those with "a very satisfactory degree of agreement", those in B had "an almost satisfactory degree of agreement", and those in C had "a less satisfactory degree of agreement". Alternative translations for the items in category C were obtained from the original version and were translated back into English.

In addition to the version designed to be filled out by the parents, another version with only minimal idiomatic differences was developed to be filled out directly by children with JCA aged over 9 years. In order to confirm that the resulting instrument corresponded with the original, the author of the original questionnaire (G. Singh) reviewed the Spanish version as well as the back-translated version in English. Like the original cHAQ, the cHAQ-S is divided into 3 sections: Disability Index (DI), Pain, and Global Assessment (6).

Study population

Patients aged between 1 and 19 years being treated at the Hospital Sant Joan de Déu and diagnosed with JCA according to the EULAR criteria (17) were included. The patients’ parents were consecutively informed of the study and asked to participate during routine check-ups at the outpatient clinic of the rheumatology unit. The health questionnaire was administered only to the patient’s parents (parent version) for those patients under 9 years of age with adequate reading skills and all the parents were asked to fill out the child and parent versions of the cHAQ-S questionnaire, respectively. The first 16 consecutively recruited patients, from whom 16 parent questionnaires and 8 child questionnaires were obtained, were given a further questionnaire to be filled out 7 days later at home and to be returned to the hospital using a stamped addressed envelope. A new questionnaire was sent to all the parents included in the study 15 months after the last patient had been enrolled. They were asked to fill it out at home and then return it.

The following clinical variables were also studied directly:
1) The number of involved joints; these were classified as joints that were swollen, that hurt when moved, or that hurt with impaired movement (18).
2) Functional status according to the ARA classification (19).
3) Functional status according Steinbroker’s Functional Class (20).

Statistical analysis

The statistical analysis was performed using SPSS for Windows version 6.0. The DI, pain and Global Assessment were tested against the onset form of JCA using a non-parametric test (Kruskall-Wallis). Similarly, correlations between DI and age, and DI and the length of evolution were checked using Spearman’s rank correlation coefficient.

The test-retest reliability of the DI was studied using paired T-tests and by calculating the intra-class correlation coefficients (21). The internal reliability was evaluated using Cronbach’s alpha (22). The between observer reliability to determine differences between the child and parent responses was studied using the intra-class correlation coefficient and paired T-tests. The validity of the DI from the cHAQ-S was evaluated from the correlation, using Spearman’s rank correlation coefficient, between the questionnaire score and other measures of disease.

The patients were divided into 3 groups according to the changes in their Parent’s Global Assessment (improvement, stable or worsening). This was used to study the sensitivity to change. The mean change in DI between each questionnaire was calculated for each of the above-mentioned groups, from which the standardised size of the observed differences (effect size) were calculated by dividing the mean change by its standard deviation. An effect size of around 0.20 was considered to be small, one of around 0.50 was classified as moderate, and an effect size was deemed to be large if its value exceeded 0.80 (23).

A principal components analysis was undertaken to check the scoring system of the cHAQ-S.

Lastly, forward step-wise multiple regression models were used to study the effect on the Parent’s Global Assessment and the Physician’s Global Assessment of the other two sections of the questionnaire, namely the DI and Pain.

Results

The final Spanish (Castillian) version of the Childhood Heath Assessment Questionnaire (see Appendix) is made up of 23 items in category A (76%), 5 in category B (17%), and 2 in category C (7%). The sample was made up of 87 patients (33 boys and 54 girls). A total of 79 questionnaires were completed by the patients’ parents using the specifically adapted version for parents; 8 were not completed because 5 children were accompanied by an adult who did not usually live with the patient, 2 children attended the visit alone, and 1 mother was illiterate. The results of the parental questionnaires are here described; the results of the child version were very similar and have been omitted.

A total of 56 out of the 79 patients suffering JCA were pauciarticular at onset, 16 had a systemic disease onset and 7
were polyarticular. Since its onset, the disease had become polyarticular in 17 patients (21.5%). The mean disease duration was 4.8 years [standard deviation (SD) = 4.1 years; range: 3 months to 18 years]. The mean age of the patient when the questionnaire was administered was 9.7 years (SD = 4.9 years; range: 1 year to 19.25 years). At the time of the visit, the disease was inactive in 37 patients, in 19 it was slightly active, in 18 it was moderate, and in 5 it was severely active. According to the Steinbroker Classification, 42 patients were in class I, 30 were in class II, 4 were in class III and 3 were in class IV.

With regard to the questionnaire variables, the mean value for the DI was 0.5 (SD = 0.6; median = 0.125), for Pain it was 0.6 (SD = 0.8; median = 0.3), and for the Global Assessment it was 0.55 (SD = 0.7; median = 0.2). The mean values for each variable by JCA type are presented in Table I. No relationship was found between DI and the patient’s age (Spearman’s rank correlation coefficient R = -0.05; p > 0.6) nor between DI and disease duration (Spearman’s R = -0.03; p > 0.7).

The test-retest reliability was calculated based on data from 16 patients; the mean length of time that elapsed before the second questionnaire was returned was 12.5 days (SD = 7). The mean DI value for the first questionnaire was 0.88 (SD = 0.92; median = 0.75) and for the second it was 0.84 (SD = 0.87; median = 0.87); the paired T-test did not reveal a significant difference (p > 0.6). The intra-class correlation coefficient between the two scores was 0.94.

Cronbach’s alpha, the measure of internal reliability, gave a value of 0.948. Removal of any of the areas produced a reduction in the alpha coefficient. The mean correlation between the areas (inter-item) was 0.62 (SD = 0.12; range 0.3557 to 0.7831) indicating moderate correlation. The correlation coefficients between DI and the other areas ranged from 0.66 to 0.85 (mean = 0.77; SD = 0.07).

The between-observer reliability was determined for a total of 33 child-parent pairs, with the questionnaire being completed by both parties during the same visit. The mean DI was 0.42 (SD = 0.7; median = 0.125) according to the parents’ questionnaire, and 0.38 (SD = 0.7; median = 0.125) according to the children. The difference was not statistically significant (p > 0.2) and the intra-class correlation was 0.96.

In the absence of a gold standard, validity was studied by correlating DI with a series of clinical and laboratory measurements related to the activity and severity of JCA (Table II).

Of the 79 parents of patients recruited in the initial sample, 50 replied to the second questionnaire. The mean time lapse between the completion of the two questionnaires was 20 months (SD = 0.37; range: 14.5 - 24 months). Using the Parent’s Global Assessment to evaluate the 50 patients, it was determined that 29 had improved, 13 remained unchanged, and 8 had worsened. In the group of improved patients, the DI value had also significantly improved (p = 0.017) in the period between the two questionnaires: according to the DI, 16 patients improved and 8 worsened. For the 8 patients whose Parent’s Global Assessment indicated a worsening of the child’s condition, the DI was marginally worse (p = 0.1): 4 had worsened, one improved, and 3 unchanged. Lastly, among the 13 children whose condition had remained stable, the DI similarly showed no significant change (p = 0.6): specifically, one child improved, 2 worsened and for the remaining 10 children the DI was unaltered. An analysis of the effect size showed moderate differences between the two questionnaires both for the group that improved and the group that worsened.

| Table I. Description of the three main variables of the cHAQ-S by disease severity at onset (expressed as the mean ± SD, with the median shown between parentheses). |
|---------------------------|-----------------|-----------------|-----------------|-----------------|----------------|
| Index Value               | Pauciarticular  | Systemic        | Polyarticular   | Total           | p-value*       |
| Disability index (DI)     | 0.2 ± 0.3       | 1.1 ± 1.0       | 1.5 ± 1.1       | 0.5 ± 0.8       | p < 0.001      |
| Pain                      | 0.5 ± 0.7       | 0.8 ± 0.8       | 1.1 ± 0.9       | 0.6 ± 0.8       | p < 0.05       |
| Global assessment         | 0.3 ± 0.5       | 0.9 ± 0.8       | 1.4 ± 0.9       | 0.6 ± 0.7       | p < 0.001      |

*From Kruskall-Wallis tests for differences in the index value between the 3 diseases.

| Table II. Convergent validity of the cHAQ-S. Correlation between the DI and several conventional measures of disease activity or laboratory parameters. |
|-----------------------------|--------------------------|
| Measure of disease activity/parameter | Spearman’s rank correlation coefficient* |
| Steinbroker’s functional class | 0.87                  |
| Number of involved joints    | 0.86                  |
| ARA functional class         | 0.79                  |
| Physician’s global assessment| 0.86                  |
| Disease activity             | 0.79                  |
| Morning stiffness            | 0.68                  |
| Erythrocyte sedimentation rate| 0.59                  |
| C-reactive protein           | 0.67                  |
| Haemoglobin                  | -0.58                 |
| Platelets                    | 0.42                  |

* All correlations were statistically significant (i.e., non-zero); p < 0.005.
The following results: a stronger relationship with the assessment score than Pain (B = 0.32; 95% CI 0.17 - 0.48), although this latter association was still considered important. The model including both variables explained a total of 65% of the variance in the Physician’s Global Assessment.

Discussion
The use of measures of patient well-being as a basic part of the clinical assessment is becoming increasingly frequent in the medical literature. Certain measures are replacing the more traditional methods, whilst other act as complements. This change in emphasis reflects the fact that the basic wish of a patient is not to reduce the levels of certain laboratory parameters or to be able to have an extra 10% of movement in his/her articulations. Instead, the patients have other aims, such as being able to undertake day-to-day activities on their own, to be in less pain, to be able to engage in their favourite sport or to receive normal schooling. It was in order to cope with these demands that clinicians have been obliged to develop a series of measurements derived from the subjective perspective of the patient. These measurements in turn should provide a more objective parameter that will be as applicable in clinical disease control as well as in the research for different and new treatment regimes. In this way we will be able to get closer to the patients and help them to speak to us about their worries, their preferences and their physical limitations.

In the area of pediatric rheumatology, one of most widely used instruments designed for this purpose is the Childhood Health Assessment Questionnaire (cHAQ). In the present study we have demonstrated that the Spanish-Castillian version of the questionnaire is both feasible and easy to use, and has a high degree of validity and reliability as a measure of the health status of children suffering from JCA. The questionnaire will not only be useful in the follow-up of such patients, but will also permit participation in international multicentre studies in which the cHAQ is one of the principal measures (24, 25). Other instruments aimed at obtaining a complete or integral assessment have been developed recently (26-29). These collect information not usually taken into account in shorter instruments, but they are extensive questionnaires that are not easily applicable to general clinical practice or in the area of clinical trials. In spite of this, they are useful as they can identify specific problems in certain patients.

It is important that any process of cross-cultural adaptation follow a valid scientific methodology with regard to the translation of the instrument, so that a maximum degree of equivalence is achieved (30). A semantically and conceptually equivalent translation is ideal, although if one has to choose between one of the two we believe that conceptual equivalence is the better. It is for this reason that in the case of the cHAQ-S some items have been removed (e.g., “yardwork”) or replaced by others (“school bus” by “bus”) and some points have been added to increase the patient’s understanding of certain concepts.

There have been a number of recent publications concerning the transcultural adaptation of the cHAQ into Spanish, such as the Costa Rican version and the Mexican version. These adaptations differ greatly from each other and from the cHAQ-S. For instance, the Mexican version has only 6 items that are identical, grammatically and syntactically, with the Spanish cHAQ-S. For instance, the Mexican version. These adaptations differ greatly from each other and from the cHAQ-S. For instance, the Mexican version has only 6 items that are identical, grammatically and syntactically, with the Spanish version (items 3, 4, 8, 11, 16 and 30). Another 6 were regarded as being highly similar (2, 5, 9, 12, 15 and 22), and the remaining 18 items showed differences of varying degrees of importance. The Costa Rican version showed even more semantic differences with the cHAQ-S, in that only 3 items were completely identical (2, 4, and 12). Four were highly similar (7, 15, 16, and 21) and the remaining 23 showed such notable differences that some were difficult to understand. On the other hand, between the Costa Rican and the Mexican questionnaires only one question was identical (26), 7 were highly similar (4, 6, 11, 12,
From the results we can deduce that the DI is the most appropriate index for the measurement of disability, since the 8 areas of the questionnaire had similar weights in one single factor, the first component. Also, the fact that the magnitudes of all the components were greater than 0.79 indicates that all the elements of the questionnaire measure something very specific (disability) and not other potential parameters. The cHAQ-S is suitable for use at any age, as demonstrated by the fact that none of the patients answered “Not applicable” to all of the questions within a given area and the DI was shown to be independent of the patient’s age. The internal reliability, as measured by Cronbach’s alpha coefficient, is considered acceptable if above the value of 0.8. As the coefficient resulting from this study was appreciably above this threshold, one can assume that all the areas that make up the DI are effectively measuring the same phenomenon. Furthermore, the removal of any of the components produced a reduction in the alpha coefficient, indicating that all of the areas are necessary and relevant in the calculation of the DI. Likewise, the moderate levels of correlation between the areas indicate that none of the areas are redundant even though they are all measuring the same underlying construct, and that none of the areas are so independent from the rest that they could be considered as not belonging to the DI. Similar inter-item correlations have been described elsewhere for the adult version (HAQ) (7) and for the original American cHAQ (6).

With regard to the convergent validity analysis, it should be noted that almost all of the correlations were classified as moderate or high. It can be deduced from this that the DI section of the cHAQ-S measures what it was designed to measure and also that the DI is effectively correlated with the other measures used in the assessment of disease. From the results of the comparison of the DI between the two cHAQ-S questionnaires administered some 20 months apart, and by studying the effect size measurements, we can deduce that the cHAQ-S is an effective instrument for the detection of changes in clinical status over time. This characteristic is of great importance as it allows an objective measurement of clinical follow-up by the use of an instrument specifically designed to measure the disability that the child may suffer during the evolution of his/her disease. Sensitivity to change had only been previously evaluated in the Norwegian and Italian versions of the cHAQ (11, 31) and in preliminary studies of the original American version (32). This evaluation should be undertaken for all versions of the cHAQ. We decided to use the Parent’s Global Assessment as the standard for functional status, as this is the parameter that best reflects the health status subjectively perceived by the parent/patient, and therefore the parameter that we should be concerned about and ready to act upon. In the Italian study, the physician’s and parents’ global assessments were found to be the most responsive measures, whilst functional ability (measured by the cHAQ) showed only moderate responsiveness (31).

Several relevant comments can be made from the regression model. On the one hand, the high percentage of variance in the Global Assessment score explained by the DI implies that there was a high degree of agreement between what the physician and the parents intuitively evaluated and what the cHAQ-S measured. On the other hand, a different pattern of evaluation between the parents and the physician was observed: for the parents the concept of Pain was of greater importance in the Global Assessment of health than it was for the physicians (although disability was the main parameter for both groups), whilst the physicians lent greater importance to the degree of disability than did the parents. We can conclude that the Spanish (Castilian) version of the cHAQ is a feasible, valid and reliable tool for determining the state of health of Spanish children suffering from JCA. It is also sensitive to changes in the child’s health status. Further specific questionnaires should be developed in our setting that are more extensive and whose aim is to cover other aspects related to the patients’ quality of life, so as to provide a greater insight into the special needs of each child, thus helping us, the clinicians, to act appropriately.
APPENDIX. Items from the Spanish-cHAQ (full version is available on request).

VESTIRSE Y ASEARSE
¿Su hijo es capaz de:
- Vestirse, incluyendo abrocharse los botones y atarse los cordones de los zapatos?
- Enjabonarse el pelo?
- Quitarse los calcetines?
- Cortarse las uñas?

LEVANTARSE
¿Su hijo es capaz de:
- Levantarse de una silla baja o del suelo?
- Acostarse y levantarse de la cama o ponerse de pie en la cuna?

COMER
¿Su hijo es capaz de:
- Cortar la carne con el cuchillo?
- Llevar una taza o un vaso a la boca?
- Abrir una caja de cereales nueva?

CAMPAR:
¿Su hijo es capaz de:
- Caminar al aire libre en terreno llano?
- Subir cinco escalones?

HIGIENE PERSONAL
¿Su hijo es capaz de:
- Lavarse y secarse todo el cuerpo?
- Entrar y salir de la bañera?
- Sentarse y levantarse del vater o de un orinal?
- cepillarse los dientes?
- Peinarse o cepillarse el pelo?

DESTREZA
¿Su hijo es capaz de:
- Coger y bajar de una estantería, situada justo por encima de la cabeza, un objeto pesado como un libro o una caja de juegos?
- Agacharse para recoger un papel del suelo?
- Ponerse un jersey por la cabeza?
- Girar la cabeza y mirar hacia atrás por encima del hombro?

PRENSIÓN
¿Su hijo es capaz de:
- Escribir o hacer garabatos con un lápiz?
- Abrir la puerta de un coche?
- Abrir un tarro abierto previamente?
- Abrir y cerrar un grifo?
- Girar el pomo de una puerta y empujarla para abrirla?

ACTIVIDADES
¿Su hijo es capaz de:
- Hacer recados o comprar?
- Subir y bajar de un coche, de un coche de juguete o de un autobús?
- Montar en bicicleta o en triciclo?
- Hacer tareas domésticas como lavar los platos, sacar la basura, hacerse la cama o limpiar su habitación?
- Correr y jugar?

Grades: 0: Sin ninguna dificultad [Without any difficulty]; 1: Con alguna dificultad [With some difficulty]; 2: Con mucha dificultad [With a lot of difficulty]; 3: Incapaz de realizar [Unable to do]; No procede [Not applicable].

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