



Validity of a shoulder-specific quality of life questionnaire, the Western Ontario Rotator Cuff Index, for patients with scapula alata

Janne Gadsboell, PT, Sigrid Tibaek, PT, DMSc *

Department of Occupational Therapy and Physiotherapy, Rigshospitalet – Glostrup, University of Copenhagen, Glostrup, Denmark

ARTICLE INFO

Keywords:

Content validity
Scapula alata
Shoulder-specific QoL measure
Western Ontario Rotator Cuff Index
Content validity index
Modified kappa index

Level of evidence: Basic Science Study,
Development or Validation of Outcomes
Instruments

Background: No validated shoulder-specific quality of life measurements exist for patients with scapula alata (SA). The objective was to investigate the content validity of the Western Ontario Rotator Cuff (WORC) Index for SA patients.

Methods: Content validity was evaluated by an expert panel of medical doctors and physiotherapists ($n = 6$) and a sample of SA patients ($n = 44$). The Content Validity Index (CVI) and a modified kappa index (κ^*) assessed the relevance of WORC Index.

Results: The experts evaluated ten of 21 items as excellent for content validity ($I-CVI > 0.78$, $\kappa^* > 0.74$), five items as fair ($I-CVI < 0.78$, $0.40 < \kappa^* < 0.60$), and six were considered content invalid ($I-CVI < 0.78$, $\kappa^* < 0.40$). The average scale (S-CVI/Ave) for the entire WORC Index was 0.72.

The SA patients evaluated four of 21 items as excellent for content validity ($I-CVI > 0.78$, $\kappa^* > 0.74$), nine items as good ($I-CVI < 0.78$, $0.60 > \kappa^* < 0.74$), six as fair ($I-CVI < 0.78$, $0.40 < \kappa^* < 0.59$) and two were considered content invalid ($I-CVI < 0.78$, $\kappa^* < 0.40$). The S-CVI/Ave was 0.56.

Conclusion: This study is the first step evaluating content validity in the WORC Index for SA patients. The results indicated that half of the 21 items had excellent or good content validity. Several items need to be discussed by an SA team aiming to find consensus for changing or removing, leaving the possibility to develop a new quality of life measure, the first for SA patients.

© 2017 The Authors. Published by Elsevier Inc. on behalf of American Shoulder and Elbow Surgeons. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Scapula alata (SA) is a clinical condition in which the medial border and inferior angle of the scapula protrude prominently from the thorax.^{22,27,28} Velpeau first described it in 1826²⁵ and subsequently the condition has been reported in children¹⁸ and adults of all ages.^{21,23}

SA is a rare condition, difficult to identify and classify, and prevalence and incidence studies remain undescribed.^{19,21}

Multiple pathologies can lead to SA. Palsy of the serratus anterior muscle caused by injury to the long thoracic nerve is considered as the most common cause.²³ Injury to the nerve can be caused by infection, traction or compression of the nerve¹⁶ or damage to the nerve associated with surgery or accident.²³ The serratus anterior muscle is essential for the scapula motion and stability; palsy of the muscle causes misalignment and winging of the scapula re-

sulting in severe alteration of the scapula- and the shoulder/arm biomechanics. Consequently, the patient is unable to elevate the arm of the affected side above shoulder level. Most patients suffer from a sudden severe pain in the affected shoulder and arm. The pain typically subsides within a few weeks and subsequently the patients describe muscle weakness and fatigue. Eventually some are exposed to headache, pain in the shoulder and neck and/or paresthesia. Patients with SA have trouble with activities of daily living, need to stop sports activities and complain of major negative impact on quality of life (QoL). A significant proportion of patients require sick leave and, in the worst cases, lose their jobs as a consequence.

Some studies report spontaneous recovery following injury to the thoracic nerve; however full recovery, if it occurs, may take more than two years.^{7,13}

Various treatments have been proposed, ranging from conservative ones such as relative rest, physical therapy,⁷ scapular protecting brace,^{10,24} nonsteroidal anti-inflammatory drugs (NSAID) and steroid injections to various surgical interventions.^{27,28}

However, outcome-based treatment programs to guide clinicians are lacking, resulting in patients living with their disability handled incorrectly or not at all.

This manuscript is submitted with the permission of the co-author and the head of the Department of Occupational Therapy and Physiotherapy, Rigshospitalet – Glostrup, University of Copenhagen, Glostrup, Nordre Ringvej 57, DK-2600 Glostrup, Denmark.

* Corresponding author.

E-mail address: Sigrid@Tibaek.dk (S. Tibaek).

<http://dx.doi.org/10.1016/j.jses.2017.02.003>

2468-6026/© 2017 The Authors. Published by Elsevier Inc. on behalf of American Shoulder and Elbow Surgeons. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Recently, we presented in the detail a rehabilitation program²¹ and evaluated its impact on a patient reported tool, the Western Ontario Rotator Cuff (WORC) Index.⁸

The WORC Index was developed for patients with degeneration of the rotator cuff. It has been tested and found acceptable regarding reliability and validity in these patients.⁸

With the aim of measuring and evaluating the outcome of the SA rehabilitation program, we looked for a patient reported questionnaire concerning perceived QoL. However, we found only a generic patient administered QoL questionnaire, the World Health Organization Quality of Life assessment (100-item version) (WHOQOL-100)²⁰; but no disease-specific QoL tool was found for patients with SA. Consequently, we chose the WORC Index, the most recent of the tools, as we found it to represent adequately functional, social and emotional aspects.

In research as well as in clinical practice, it is recommended to have valid and reliable scoring measurements. The WORC Index has never been tested for reliability or validity in patients with SA.

The objective of this study was to investigate the content validity of the WORC Index for patients diagnosed with SA.

Materials and methods

The evaluation of WORC in terms of content validity was performed by an expert panel and by a sample of patients diagnosed with SA.

Experts

The expert panel consisted of six healthcare professionals: four medical doctors (each a specialist with experience in diagnosing SA) and two physiotherapists (each with significant clinical experience in rehabilitation of SA patients). The experts were recruited from Danish university hospitals, focusing on Departments of Orthopaedics, Sports Medicine, and Rheumatology, and from a Specialized Shoulder Rehabilitation Clinic and a private Rheumatology and Sports Medicine Clinic. The expert panel was recruited between 1st and 29th October 2013.

SA patients

A sample of 70 patients diagnosed with SA and receiving the physiotherapy rehabilitation program previously (<3 yr.) or currently was invited to evaluate the WORC Index in terms of content validity. The SA patients were recruited from an outpatient clinic, specialized in SA, at the Department of Occupational Therapy and Physiotherapy at a University Hospital from 1st August to 1st October 2015.

Method

The expert panel was invited to evaluate the relevance of the WORC Index to SA patients. They received a cover letter including a) information and instructions, b) a WORC Index Questionnaire, c) two evaluation sheets, one for each item and one for the total WORC Index, d) a demographic, medical and research education characteristics sheet, e) a stamped envelope for the reply. Additionally the experts were asked to write comments on each of the two judgement sheets.

In case of no reply reminders would be sent after two weeks and after five weeks.

The SA patients were likewise invited to evaluate the relevance of the WORC Index questionnaire to SA. They received a cover letter plus: a) information and instructions, b) two evaluation sheets, one for each item and one for the total WORC Index, c) a sheet for comments on each item, d) a sheet for demographic information and

e) a prepaid envelope. The SA patients too were asked to provide written, qualitative comments on each of the two evaluation sheets. They were asked to return the sheets preferably two weeks after receiving them, taking account of the summer vacation, October 1st, 2015 for their latest return.

Measurement

The WORC index questionnaire⁸

The WORC Index is a self-reported shoulder-specific QoL measure. The questionnaire is short, estimated to take 10 minutes to complete.

The WORC Index consists of 21 items grouped in five domains: Physical symptoms (items 1–6), sports/recreation (items 7–10), work (items 11–14), lifestyle (items 15–18), emotions (items 19–21).

The impact of each item is rated on a visual analogy scale (0–100 mm). A score of 0 point indicates no impact on QoL, while a score of 100 points indicates the worst-case scenario. Thus, it is possible to score from 0 to 2100 points. To present the score in a more clinically meaningful format, the score is reported as a percentage of normal by subtracting the total score from 2100, dividing by 2100, and multiplying by 100.

The WORC Index was translated into Danish in 2004 following international standards.

Content validity

The term content validity is defined as “The degree to which an instrument has the appropriate sample of items of the construct being measured.”¹⁴

The evaluation in terms of relevance of the WORC Index was assessed by the Content Validity Index (CVI),¹¹ which included:

- 1 Evaluation of each item in the WORC Index questionnaire; in terms of relevance to SA patients this was measured by Items-Content Validity Index (I-CVI).
- 2 Evaluation of the entire WORC Index questionnaire; in terms of relevance to SA patients this was measured by Scale-Content Validity Index (S-CVI).
- 3 The average deviation (AD) index was used as a measure of inter-rater agreement.²

The evaluation was rated as a number on a 4-point ordinal scale (1 = not relevant, 2 = somewhat relevant, 3 = quite relevant, 4 = highly relevant).⁴

Data analysis

The cut-off point for acceptable value for content value for each item was a rating of 3 or 4.

I-CVI was calculated as the percentage of experts and SA patients rating an item either 3 or 4, respectively. The S-CVI was calculated as the average of the I-CVIs for all items on the scale (S-CV I/Ave) rating the entire questionnaire by 3 or 4, respectively.

Any I-CVI rated greater than or equal to 0.78 by 6 or more experts is considered excellent. Subsequently Polit et al recommend 0.78 as excellent regardless of the number of experts. The recommendation for S-CVI/Ave is 0.90 or higher.^{15,26}

To counter the limitations of CVI, each I-CVI was adjusted for chance agreement by calculating the modified kappa statistic (κ^*).^{15,29} To compute the modified kappa, the probability of chance agreement was computed first: $P_c = (N/A) \times 0.5^N$ where N is the number of experts and A is the number of agreement of good relevance (rating 3 and 4). Then, the κ^* was calculated using the formula $\kappa^* = (I-CVI - P_c) / (1 - P_c)$.¹¹

According to the standards of Fleiss et al and Cicchetti and Sparrow the value of each κ^* was evaluated as: poor ($\kappa < 0.40$), fair ($0.40 < \kappa < 0.59$), good ($0.60 < \kappa < 0.74$) or excellent ($\kappa > 0.74$).^{3,5}

In this study, the experts and the SA patients were asked to evaluate the entire WORC Index overall as a QoL measurement for patients with SA using the same four-point scale, defined as S-CVI_{total}.

The AD index for Likert-type scales was used to measure inter-rater agreement.² The AD index measures the dispersion of responses about the median. This was calculated as the sum of differences from the median in absolute values divided by the number of experts. At 5% level of significance the cut-off limit was 0.37 for six raters. Values below 0.37 indicated acceptable and statistical significant agreement.²

Statistics

Statistical analysis was carried out using IBM SPSS (Statistical Package of Social Science) version 20.0 (Armonk, NY, USA). Median and interquartile range (IQR) are presented for the small sample and mean and standard deviation (SD) for the larger sample.

The level of statistical significance was set to $P < 0.05$.

Results

All six (100%) experts in the panel responded to the survey, five within two weeks and one after the first reminder.

In total, 45 (65%) SA patients responded to the survey, 30 (43%) within the first three weeks and the rest over the next five weeks.

The characteristics of the experts and the SA patients are presented in Table I and Table II.

The evaluations by the experts regarding each item of WORC Index (I-CVI) were all eligible while an evaluation by one SA patient was ineligible, leaving 44 evaluations for statistical analysis of I-CVI. Judgments regarding the entire WORC Index (S-CVI) from four SA patients were ineligible, leaving 41 evaluations for statistical analysis of S-CVI.

The experts

Ten (48%) (Table III) out of 21 items had an excellent content validity ($I-CVI > 0.78$, $\kappa^* > 0.74$), five (24%) items had a fair content validity ($I-CVI < 0.78$, $0.40 < \kappa^* < 0.60$) and six (28%) items were considered content invalid ($I-CVI < 0.78$, $\kappa^* < 0.40$).

The AD index was 0.50 (IQR 0.30–0.50) and 13/21 (61%) items did not met acceptable AD index. The average scale content validity (S-CVI/Ave) in the five domains was rated as follows: physical symptoms 0.42, sport/recreation 0.83, work 1.00, lifestyle 0.53 and emotions 0.83. Calculated for all five domains S-CVI/Ave was 0.72.

Table I
Characteristics of the expert panel

Experts, n = 6	No. (%)
Age, years ^a	53 (43–62)
Gender	
Women	2 (33)
Men	4 (67)
Medical education	
Orthopedic surgeon	1 (17)
Rheumatologist	3 (50)
Physiotherapist	2 (33)
Academic degree	
Ph.D.	1 (17)
Doctor of Medical Science	1 (17)
None	4 (66)
Clinical experiences with scapula alata patients, years ^a	15 (5–20)

^a Median and interquartile range (IQR).

Table II
Demographic and medical characteristics of patients with scapula alata

Patients with scapula alata, n = 45	No. (%)
Age, years ^a	52 (38–59)
Gender	
Women	26 (58)
Men	19 (42)
Educational level	
Short education	15 (33)
Medium education	16 (36)
Academic education	14 (31)
Employment status	
Working	33 (76)
Unemployed	1 (2)
Studying	4 (8)
Retired	7 (16)
Sick leave	0
Rehabilitation status	
Former	28 (62)
Current	17 (38)
Pathology	
N. Thoracicus affection	44 (98)
N. Accessorius affection	0 (0)
Both	1 (2)
Etiology	
Trauma	7 (16)
Mononeuritis	30 (67)
Surgery sequelae	0
Stretch/overload	3 (7)
Sequelae to infection	1 (2)
Other	4 (8)
Status after rehabilitation program	
Recovered	25 (56)
Recovering ongoing	18 (40)
No recovery	2 (4)
Affected side	
Left	3 (7)
Right	41 (91)
Both	1 (2)
Dominant hand	
Left	1 (2)
Right	41 (91)
No information	3 (7)

^a Median and interquartile range (IQR).

Five (83%) experts rated the total WORC Index as a relevant QoL measurement (range 0.83–1.00), resulting in S-CVI_{total} = 0.83.

Three (50%) experts commented that there were too many items not relevance for SA patients.

The SA patients

Four (9%) (Table III) out of 21 items had an excellent content validity ($I-CVI > 0.78$, $\kappa^* > 0.74$), nine (43%) items had a good content validity ($I-CVI < 0.78$, $0.60 > \kappa^* < 0.74$), six (28%) items had fair content validity ($I-CVI < 0.78$, $0.40 < \kappa^* < 0.59$) and two (9%) items were considered as content invalid ($I-CVI < 0.78$, $\kappa^* < 0.40$).

The average scale content validity (S-CVI/Ave) in the five domains was rated as follows: physical symptoms 0.58, sport/recreation 0.72, work 0.76, lifestyle 0.55 and emotions 0.63. Calculated for all five domains, the S-CVI/Ave was 0.56.

Thirty-six (88%) SA patients rated the total WORC Index as a relevant QoL measurement for patients with SA, resulting in S-CVI_{total} = 0.88.

Twenty-three (52%) SA patients had various comments such as concerning pain (items 1, 2 and 6) “Relevant from the very beginning because of the initial pain I experienced”; “I only experienced pain before I started the treatment” and “I miss a question about paresthesia in the arm, hand and fingers”.

Comments on the domain work (rated 0.78) SA patients commented “Very relevant with questions addressing daily living at home

Table III
Evaluation of content validity of the Western Ontario Rotator Cuff Index for patients with scapula alata

Item	Experts			SA-patients		
	I-CVI Experts n = 6	κ^*	Evaluation	I-CVI SA patients n = 44	κ^*	Evaluation
1. How much sharp pain do you experience in your shoulder?	0.50	0.27	Poor	0.64	0.63	Good
2. How much constant, nagging pain do you experience in your shoulder?	0.67	0.57	Fair	0.62	0.60	Good
3. How much weakness do you experience in your shoulder?	1.00	1.00	Excellent	0.84	0.84	Excellent
4. How much stiffness do you experience in your shoulder?	0.17	0.08	Poor	0.55	0.50	Fair
5. How much clicking, grinding or crunching do you experience in your shoulder?	0.00	0.00	Poor	0.51	0.44	Fair
6. How much discomfort do you experience in your neck because of your shoulder?	0.17	0.08	Poor	0.58	0.55	Fair
7. How much has your shoulder affected your fitness level?	1.00	1.00	Excellent	0.84	0.84	Excellent
8. How much has your shoulder affected your ability to throw hard or far?	1.00	1.00	Excellent	0.89	0.89	Excellent
9. How much difficulty do you have with someone or something coming in contact with your affected shoulder?	1.00	1.00	Excellent	0.71	0.71	Good
10. How much difficulty do you experience because of your shoulder in doing push-ups or other strenuous shoulder experiences?	0.33	0.12	Fair	0.42	0.38	Poor
11. How much difficulty do you experience in daily activities about the house or yard?	1.00	1.00	Excellent	0.73	0.73	Good
12. How much difficulty do you experience working above your head?	1.00	1.00	Excellent	0.84	0.84	Excellent
13. How much do you use your uninjured arm to compensate for your injured one?	1.00	1.00	Excellent	0.75	0.75	Excellent
14. How much difficulty do you experience lifting heavy objects from the ground or below shoulder level?	0.83	0.81	Excellent	0.73	0.73	Good
15. How much difficulty do you have sleeping because of your shoulder?	0.50	0.27	Poor	0.69	0.69	Good
16. How much difficulty have you experienced with styling your hair because of your shoulder?	0.67	0.57	Fair	0.51	0.44	Fair
17. How much difficulty do you have "being boisterous or horsing around" with family or friends?	0.33	0.12	Poor	0.44	0.39	Poor
18. How much difficulty do you have dressing or undressing?	0.67	0.57	Fair	0.55	0.50	Fair
19. How much frustration do you feel because of your shoulder?	0.83	0.81	Excellent	0.68	0.68	Good
20. How "down in the dumps" or depressed do you feel because of your shoulder?	0.67	0.57	Fair	0.60	0.58	Fair
21. How worried or concerned are you about the effect of your shoulder on your occupation or work?	1.00	1.00	Excellent	0.62	0.60	Good

I-CVI, Items Content Validity Index; κ^* , modified kappa index.

and at work in order to clarify the impact on quality of life". Another comment was: "I miss a question that elaborates the impact on working conditions such as sick leave and reorganization of work tasks".

The domain lifestyle (rated 0.55) had comments especially on items 16 and 17. The former concerns difficulties with hair styling. Item 17, concerning roughhousing or horsing around, was for most SA patients difficult to understand not a relevant activity.

Regarding the domain emotions, SA patients confirmed its relevance and one asks for "An extended emotional part".

Discussion

The present study is the first to investigate the content validity of the WORC Index for patients with SA. The WORC Index was evaluated in relation to relevance by an expert panel each with at least five years clinical experiences in the field of SA and with a sample of former or current SA patients, meaning that we used two heterogeneous expert panels.

The results indicated that approximately half of each item in WORC Index (I-CVI) had excellent or good content validity. The S-CVI/Ave was evaluated as not relevant according to the recommendation of Polit et al.¹⁵

Thus, several items may be discarded. There was agreement or near agreement between the two assessment groups on some of the issues; however, there was disagreement on others.

Concerning the domain physical symptoms (items 1–6), the experts rated three out of six items extremely low (I-CVI, range 0.00–0.17). Do these items need to be changed or removed? The SA patients rated the same items considerably higher (I-CVI, range 0.51–0.84). Likewise, their comments expressed the items as relevant. Experiences from SA patients based on practice in real-life environment are compelling, especially in regard to self-reported QoL measures.

Item 4 "How much stiffness do you experience in your shoulder?" was rated low (experts, I-CVI = 0.17 vs. SA patients I-CVI = 0.55).

Stiffness of the shoulder in SA patients is rarely a problem. However, lack of range of motion is an essential consequence. So an addition to item 4 may be considered, as Kirkley et al first presented item 4 in the WORC Index as "How much stiffness and lack of range of motion do you experience. . .?"⁹

Item 5 concerning the presence of noisy symptoms was rated not relevant by the experts (I-CVI = 0.00) and somewhat relevant by the participants (I-CVI = 0.51). That half of the patients rated the item relevant might be explained by the naturally increased attention to the shoulder and confused with the occurrence of harmless ordinary joint symptoms. The fact is that the noise from the shoulder, as well as from other parts of the musculoskeletal system, is a well-known phenomenon and does not necessarily have anything to do with the patient's SA. This symptom, just like stiffness (item 4), is a very unlikely complication, so the item may be considered redundant.

Items 1, 2 and 6 referring to sharp or constant, nagging pain of the shoulder and discomfort of the neck muscles, were rated differently among the experts as well as among the SA patients. The fact is that SA patients often experience pain in the acute phase of the condition. Then the pain disappears, in some cases however to return, albeit replaced by another type of pain. Regarding item 6 (experts, I-CVI = 0.17 vs. SA patients, I-CVI = 0.58) the rating also showed a rather significant difference. The question refers to discomfort of the neck muscles. Some patients experience pain and discomfort of the neck muscles and/or headaches in the later course because of the alteration of the muscle length and the muscles malalignment. The discomfort usually subsides once the treatment has started. Subsequently the presence or degree of pain varies from patient to patient. The difference between the two rating groups may be explained by the fact that the medical doctors usually see the patients only once or twice for diagnosis and referral to treatment. At that time, the focus is on the initial acute strong pain and the loss of shoulder function, which are significant symptoms for diagnosis. The PTs find the pain and discomfort subsiding rather fast as the treatment starts. However, patients having experienced

discomfort of the neck muscles find the item relevant. SA participant comments on pain and discomfort expressed relevance at the beginning of the course of disease and before treatment. One participant expressed: “Relevance of an item on paraesthesia in the arm” which is an obvious topic for some SA patients.

There was consensus among the two groups on the relevance of item 3, referring to weakness of the shoulder (experts, I-CVI = 1.0 vs. SA patients, I-CVI = 0.84), with comments like: “Very strong relevance because of reduced strength” and “Important because of decrease in both strength and endurance”.

Looking at the domain sports/recreation (items 7–10) there was agreement on relevance or high relevance of all items except item 9 referring to difficulties having something or someone coming in contact with the shoulder. Both the experts and the SA patients found the item not relevant (experts, I-CVI = 0.33 vs. SA patients, I-CVI = 0.42). There was only one comment on this item saying: “I had no problems in this respect”. Perhaps this item may be deleted or changed? However, the issues affecting loss of muscle strength and function are very relevant.

The domain work (items 11–14) was rated of relevance or high relevance by the experts, and relevant or somewhat relevant by the SA patients. Item 12, referring to difficulties working above shoulder level, was rated of high relevance by the experts (I-CVI = 1.0) and of relevance by the SA patients (I-CVI = 0.84). This emphasizes the relevance of items concerning range of motion such as the ability to use the arm above shoulder level. The overall rating showed that this domain in general is of relevance, as stated in the previously mentioned comments.

In the domain, lifestyle (items 15–18) three items were rated as somewhat relevant by each of the assessment groups. However item 16, referring to hair styling, was rated of no relevance by the experts (I-CVI = 0.33) and somewhat relevant by the SA patients (I-CVI = 0.51). Half of the patients found this item relevant. Twenty-three SA patients rated it relevant or highly relevant; looking into the gender distribution we found that 19 (83%) SA patients were women, showing that this item apparently is more relevant for women. An analysis for the expert group did not show this tendency though, so maybe a comparable analysis is not indicated in such a small sample size. The item might be considered redundant and therefore discarded.

Both groups rated the domain emotions (items 19–21) somewhat relevant on the two first items. Item 21, referring to concern about occupation or work, was rated highly relevant by the experts (I-CVI = 1.00) and somewhat relevant by the SA patients (I-CVI = 0.62). Some assessors in the SA patient group expressed high relevance of this domain in their comments. A factor analysis could be interesting in order to clarify the degree to which social and educational status and education attainment affect the ratings in this domain.

It seems that the domain referring to emotional issues was important to more than half of both the SA patients and experts. These items might be considered candidates for revision.

There were supplementary comments following this domain on the impact the brace has on QoL. These comments are informative; however, they are not useful as an issue in a questionnaire aimed at measuring QoL before and after treatment.

Methodological considerations

Several methodological issues in the current study need to be considered.

Experts should be selected with care, using well-defined criteria such as those proposed by Grant and Davis.⁶ Regarding the selection of SA patients, they were not randomly chosen in terms of different ages, sexes, disease severities, rehabilitation status, education and works. They were instead recruited successively from the Department of Occupational Therapy and Physiotherapy, with

the criteria of having SA diagnosed and having undergone the physiotherapy rehabilitation program²¹ previously or currently participating in it. This means that some of the patients experienced their burden of disability some time earlier as opposed to the group who experienced the impact on QoL currently or recently. A sample of SA patients with more differing demographic and physical characteristics would perhaps have been more representative.

Another issue is the small sample of experts; however, it does meet the recommended number¹¹ of a panel of between three and ten. However, Denmark, with approximately 5.5 million inhabitants, has only very few experts with thorough experiential knowledge of SA. The major portion (67%) of the six experts was doctors who see SA patients only once for diagnosis and then immediately refer them on, at the very beginning of the course of treatment.

Furthermore, the doctors were all men while the physiotherapists were all woman (33%). With this small sample size it was not possible to calculate in a statistical analysis the influences of factors such as sex and education. However, a panel consisting of experts with a more profound knowledge of SA patients, through significant clinical experience throughout the rehabilitation period, would certainly be desirable. Even so, it was obvious that we should use this heterogeneous assessment panel¹⁷ in order to achieve a more thorough content validity evaluation of WORC in SA issues. In future, it can lead to an instrument with greater relevance for SA patients.

A further issue is the method used to evaluate the relevance of WORC Index. Both assessment groups were asked to quantify content validity of WORC by using the CVI supplied by comments in writing. The CVI is an index of inter-rater agreement. There are alternative methods to CVI as described and discussed by Polit et al;¹⁵ however it seems that the CVI has been preferred in health-related fields as an indicator of content validity.

According to COSMIN criteria (Consensus-based Standards for the selection of health status Measurement Instruments),¹² domain validity includes three measurement properties: content validity, construct validity, and criterion validity. Moreover, the content validity should include face validity as a first aspect defined by Mokkink et al.¹² Face validity should be included in a second round of the study since it is regrettably missing in the current one.

Perspectives

This study is the first step in evaluating content validity of the WORC Index through a shoulder-specific QoL questionnaire for SA patients. The next step is a process where items, comments, and assessors are stringently sorted and discussed within an SA expert team, aiming to find consensus for revision, adding and/or eliminating some of the items in the possibility to develop a QoL measure for SA patients. Likewise, some assessors from the expert panel and from the panel of SA patients might be eliminated as described by Polit et al,¹⁵ or possibly replaced with more qualified assessors.

In general as well as in healthcare, there is an increasing awareness of the importance of integrating patient-reported QoL outcome measures.¹ The WORC Index in an adjusted version can become a valuable QoL outcome measurement tool for SA patients. It could eventually result in a self-report instrument for SA patients, assessing change over time, for use in future rehabilitation programs and in research.

Conclusion

The present study is the first step in evaluating the content validity of the WORC Index for SA patients. The results indicated that half of the 21 items were of good or excellent content validity. It seems that loss of range of motion, and of strength and endurance

are issues of great relevance. Several other items may not be as important in patients with SA.

Acknowledgements

The authors wish to thank the expert panel at the Department of Orthopaedics, Department of Sports Medicine, Department of Rheumatology, the Clinic of Specialized Shoulder Rehabilitation and a private Rheumatology and Clinic of Sports Medicine. Thanks are also due to the SA patients from the Department of Occupational Therapy and Physiotherapy, University of Copenhagen, Rigshospitalet – Glostrup. All patients signed consent forms.

References

- Anker SD, Agewall S, Borggrefe M, Calvert M, Jaime Caro J, Cowie MR, et al. The importance of patient-reported outcomes: a call for their comprehensive integration in cardiovascular clinical trials. *Eur Heart J* 2014;35:2001-9. <http://dx.doi.org/10.1093/eurheartj/ehu205>
- Burke MJ, Dunlap WP. Estimating interrater agreement with the average deviation index: a user's guide. *Org Res Methods* 2002;5:159-72. <https://doi.org/10.1177/1094428102005002002>
- Cicchetti DV, Sparrow SA. Developing criteria for establishing interrater reliability of specific items: applications to assessment of adaptive behavior. *Am J Ment Defic* 1981;86:127-37.
- Davis LL. Instrument review: getting the most from your panel of experts. *Appl Nurs Res* 1992;5:194-7.
- Fleiss JL, Levin B, Paik MC. Statistical methods for rates and proportions. Third ed. John Wiley & Sons, Inc.; 2004 ISBN 9780471526292.
- Grant JS, Davis LL. Selection and use of content experts for instrument development. *Res Nurs Health* 1997;20:269-74.
- Kauppila LI, Vastamaki M. Iatrogenic serratus anterior paralysis. Long-term outcome in 26 patients. *Chest* 1996;109:31-4.
- Kirkley A, Alvarez C, Griffin S. The development and evaluation of a disease-specific quality-of-life questionnaire for disorders of the rotator cuff: the Western Ontario rotator cuff index. *Clin J Sport Med* 2003;13:84-92.
- Kirkley A, Griffin S, McLintock H, Ng L. The development and evaluation of a disease-specific quality of life measurement tool for shoulder instability. The Western Ontario shoulder instability index (WOSI). *Am J Sports Med* 1998;26:764-72.
- Klebe TM, Dossing KV, Blenstrup T, Nielsen-Ferreira J, Rejsenhus L, Aalkjaer G, et al. [Scapulae alatae – angels' wings. A study of 64 patients treated with braces and physical therapy at the Viberg's hospital]. *Ugeskr Laeger* 2003;165:1779-82.
- Lynn MR. Determination and quantification of content validity. *Nurs Res* 1986;35:382-5.
- Mokkink LB, Terwee CB, Patrick DL, Alonso J, Stratford PW, Knol DL, et al. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *J Clin Epidemiol* 2010;63:737-45. <http://dx.doi.org/10.1016/j.jclinepi.2010.02.006>
- Pikkarainen V, Kettunen J, Vastamäki M. The natural course of serratus palsy at 2 to 31 years. *Clin Orthop Relat Res* 2013;471:1555-63. <http://dx.doi.org/10.1007/s11999-012-2723-7>
- Polit DF, Beck CT. The content validity index: are you sure you know what's being reported? Critique and recommendations. *Res Nurs Health* 2006;29:489-97. <http://dx.doi.org/10.1002/nur.20147>
- Polit DF, Beck CT, Owen SV. Is the CVI an acceptable indicator of content validity? Appraisal and recommendations. *Res Nurs Health* 2007;30:459-67. <http://dx.doi.org/10.1002/nur.20199>
- Safraan MR. Nerve injury about the shoulder in athletes, part 2: long thoracic nerve, spinal accessory nerve, burners/stingers, thoracic outlet syndrome. *Am J Sports Med* 2004;32:1063-76. <http://dx.doi.org/10.1177/0363546504265193>
- Schilling LS, Dixon JK, Knafel KA, Grey M, Ives B, Lynn MR. Determining content validity of a self-report instrument for adolescents using a heterogeneous expert panel. *Nurs Res* 2007;56:361-6. <http://dx.doi.org/10.1097/01.NNR.0000289505.30037.91>
- Scott DA, Alexander JR. Relapsing and remitting scapular winging in a pediatric patient. *Am J Phys Med Rehabil* 2010;89:505-8. <http://dx.doi.org/10.1097/PHM.0b013e3181cf55fa>
- Srikumaran U, Wells JH, Freehill MT, Tan EW, Higgins LD, Warner JJ. Scapular winging: a great masquerader of shoulder disorders: AAOS exhibit selection. *J Bone Joint Surg Am* 2014;96:e122. <http://dx.doi.org/10.2106/JBJS.M.01031>
- Szabo S. On behalf of the WHOQOL group: the World Health Organisation Quality of Life (WHOQOL) assessment instrument. In: Spiker B, editor. *Pharmacoeconomics in clinical trials*. Philadelphia, New York: Lippincott-Raven Publishers; 1996. p. 335-62.
- Tibaek S, Gadsboell J. Scapula alata: description of a physical therapy program and its effectiveness measured by a shoulder-specific quality-of-life measurement. *J Shoulder Elbow Surg* 2015;24:482-90. <http://dx.doi.org/10.1016/j.jse.2014.07.006>
- Vanderstraten J. Scapula alata. *La revue de la Médecine Générale* 2010;32-3.
- Vastamäki M, Kauppila LI. Etiologic factors in isolated paralysis of the serratus anterior muscle: a report of 197 cases. *J Shoulder Elbow Surg* 1993;2:240-3.
- Vastamäki M, Pikkarainen V, Vastamaki H, Ristolainen L. Scapular bracing is effective in some patients but symptoms persist in many despite bracing. *Clin Orthop Relat Res* 2015;473:2650-7. <http://dx.doi.org/10.1007/s11999-015-4310-1>
- Velpeau AALM. *Traité d'anatomie chirurgiale, ou Anatomie des Regions considérées dans ses rapports avec la chirurgie*. Paris: Chevot; 1826.
- Waltz CF, Strickland OL, Lenz E. *Measurement in nursing and health research*. Fourth ed. Springer Publishing Company; 2010 ISBN 978-0826105073.
- Warner JJ, Navarro RA. Serratus anterior dysfunction. Recognition and treatment. *Clin Orthop Relat Res* 1998;139-48.
- Wiater JM, Flatow EL. Long thoracic nerve injury. *Clin Orthop Relat Res* 1999;17-27.
- Wynd CA, Schmidt B, Schaefer MA. Two quantitative approaches for estimating content validity. *West J Nurs Res* 2003;25:508-18. <http://dx.doi.org/10.1177/0193945903252998>