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Validation of myasthenia gravis activity of daily living questionnaire: Persian version

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Abstract
Background: The 8-item myasthenia gravis activity of daily living (MG-ADL) questionnaire is a valid and reliable instrument for evaluating myasthenia gravis (MG)-associated disability. This study aims to assess its validity and reliability in the Iranian population.
Methods: A total number of 58 patients with MG were qualified for the examination. All 58 patients completed the Persian translation of 15-item Myasthenia Gravis Quality of Life (MG-QOL15) and MG-ADL questionnaires initially, and 30 filled out the MG-ADL questionnaire 2 to 4 weeks after the initial visit. Pearson correlation coefficient of questionnaires, internal consistency using Cronbach's alpha (α), and test-retest repeatability of the questionnaire were evaluated.
Results: The Pearson correlation coefficient of Persian versions of MG-QOL and MG-ADL was 0.93 (P < 0.01). The Persian MG-ADL showed satisfactory internal consistency (Cronbach's α = 0.89) and test-retest reliability (r = 0.99, P < 0.01).
Conclusion: The Persian MG-ADL is a valid and reliable questionnaire for the determination of activities of daily life in Iranian patients with MG.

Introduction

Myasthenia gravis (MG) is a chronic autoimmune disease characterized by intermittent weakness of voluntary muscles exacerbated by activity and partially cured by rest. In two-thirds of patients, the onset of the disease begins with the involvement of the ocular muscles, and as it progresses, the bulbar muscles and the muscles of the limbs become involved.[1]

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MG is an uncommon disease with an increasing prevalence of 20 per 100000 population in the United States (US). The prevalence of this disease is higher in women under 40 years of age and in men over 50 years of age and is equal in the age range of 40-50 years.[2] Although there are several effective treatments for this disease, none of them cause complete recovery of these patients, and their quality of life (QOL) decreases significantly over time.[2,3]

Due to the chronic nature of this disease and changes in patients' functional performance over time, functional tools are necessary to assess the course of the disease and its severity in patients. Since patients experience different fatigue levels and muscle weakness throughout the day and in different situations, it is impossible to assess the disease severity by clinical examination and electrodiagnostic evaluation.[4,5] Questionnaires have been prepared for this purpose that are filled by patients according to their condition.[6,7] The course of the disease and the response to treatment are obtained by comparing the calculated score from the questionnaire with the score obtained at previous visits. These comparisons have a significant impact on the evaluation of the patient's condition and prognosis.[4]

The most commonly used questionnaire for assessing the condition of patients is the myasthenia gravis activity of daily living (MG-ADL) questionnaire. The MG-ADL questionnaire is a simple 8-item questionnaire that is filled out by the patient. This questionnaire examines the function of the bulbar, respiratory, limb, and ocular muscles with a score of 0-3 for each item.[8] It is considered highly reliable and valid with the ability to discriminate between different severity of impairment and a good ability to predict clinical outcome improvement.[6,9,10] There is no validated MG-specific questionnaire in the Persian language. Due to the need to evaluate Iranian patients' quality and daily activities, we attempted to adapt the MG-ADL questionnaire and evaluate the Persian version of MG-ADL (MG-ADL-P) in terms of validity and reliability. This study is the first to assess the MG-ADL in the Iranian population to the best of our knowledge.

Materials and Methods

Patients: This study was conducted in the neuromuscular department of Shariati Hospital, Tehran University of Medical Sciences, Tehran, Iran, during 2018-2019. The Institutional Review Board approved this study at the Ethics Committee of Tehran University of Medical Sciences (Approval Number: IR.TUMS.MEDICINE.REC.1398.387). Written informed consent was obtained from all participants. Given eight items on the MG-ADL, the sample size was determined as 58. Fifty-eight consecutive outpatients referred to the neuromuscular clinic of Shariati Hospital were enrolled in this study. All patients, regardless of gender and age, with a confirmed diagnosis of MG of any type and severity except Myasthenia Gravis Foundation of America (MGFA) Class V, were enrolled in the study.

Measures: MG-ADL is an outcome measurement tool that evaluates the functional performance of daily activities of patients with MG. It consists of eight items that two items assess ocular, three bulbar, one respiratory, and two limb-related impairment. Each item is given a score between 0 and 3; thus, the total score range is from 0 to 24 points. A higher score demonstrates the patient's poor condition. The patients completed the 15-item Myasthenia Gravis Quality of Life (MG-QOL15) questionnaire to verify the construct validity of an MG-ADL-P scale. The MG-QOL15 is a valid and reliable scale in Iran (Persian language).[11] This scale consists of 15 items that investigate the health-related QOL. Each item is scored between 0 and 4 with the total score ranging from 0 to 60 points. Higher scores represent the poor QOL for the patients.

Translation: To adapt the MG-ADL questionnaire into Persian, we performed the 5-step method recommended by the World Health Organization (WHO). The translation was done with cross-cultural guidelines and the forward-backward method.[12,13] Initially, to translate the MG-ADL questionnaire, the original version of this questionnaire was translated by a neurologist. In the next step, a panel was formed with the translator, the relevant supervisor, and a faculty member of Tehran University of Medical Sciences who had experience in adapting questionnaires. In this session, the final version of the MG-ADL-P was obtained with the final editing to eliminate spelling and semantic problems and apply appropriate words and sentences. The developed Persian questionnaire was back-translated by another individual who was not aware of the content of original questionnaire. Finally, the original English version was compared with the translated version from Persian, and the translation process was approved as it transferred the general

concepts. Ten patients were enrolled in a pilot study using the latest version of MG-ADL-P to evaluate the comprehensibility of questions. The final version of the MG-ADL-P was then produced.

Statistical analysis was performed using SPSS software (version 21, IBM Corporation, Armonk, NY, USA), and a P < 0.05 was considered statistically significant.

Validity: Each patient filled both the MG-ADL-P and the previously adapted MG-QOL.[11] Then, the Pearson correlation coefficient of these data was calculated. The correlation coefficient can range between +1 (maximum correlation between data) and -1 (minimum correlation between data).

Reliability: The MG-ADL-P reliability was evaluated using Cronbach's alpha (α), inter-item correlations, and corrected item-total correlations. An α equivalent to 1 indicates perfect internal consistency, 0.9 excellent, 0.8 good, and 0.7 acceptable.[14]

Reproducibility: The reproducibility of the items and the questionnaire was obtained by the test-retest method. Twenty patients with relatively stable conditions answered the questionnaire twice at intervals of 2 to 4 weeks, and correlations between the results on the two visits were examined using Pearson correlation coefficients and intra-class correlation coefficient (ICC) between baseline and retest MG-ADL-P scores.

Results

Patients and clinical characteristics: A total of 58 patients with a mean age of 45.9 ± 7.2 years were included in the study. The duration of the disease varied from 2 months to 27 years, and the mean duration was 6.6 ± 5.9 years for patients. Among patients, 33 (56%) were men and 25 (44%) were women.

Translation: The MG-ADL-P questionnaire was translated successfully without any problems, considering the existing linguistic and cultural differences. All of the patients were able to answer the questionnaires fully, without any problem. One patient answered the questionnaire with the help of a researcher due to illiteracy and lack of reading ability. We found that one of the questions on the shortness of breath during activity in some older patients was mistaken with shortness of breath due to heart and respiratory diseases. Still, after consultation, we decided that it could not be replaced with any other phrases. Patients' shortness of breath could cause changes in their daily activities, and it was impossible to differentiate it from other diseases in a questionnaire. Table 1 demonstrates each item score of MG-ADL.

Reliability: In evaluating the reliability of the questionnaire, an acceptable internal consistency coefficient was obtained for the MG-ADL-P questionnaire. The obtained Cronbach's α coefficient was 0.89 and did not augment deleting any item. The mean of inter-item correlation was 0.54 and ranged between 0.29 and 0.84. All eight items of the MG-ADL-P scale exhibited good correlation, ranging from 0.62 to 0.86. Swallowing and double vision items showed the highest correlations with index scores (r = 0.79 and r = 0.86, respectively). Breathing and impairment of the ability to rise from chair items obtained less correlation with index scores (r = 0.62), and internal consistency was slightly higher when these items were deleted from the total score (α = 0.88).

Reproducibility: The reproducibility of the questionnaire was assessed by the test-retest method that was carried out on 20 patients. The Pearson correlation coefficient between the two visits of patients was 0.99 with P < 0.001. The positive Pearson correlation coefficient suggests a reasonably strong relationship for the test-retest questionnaire in MG-ADL-P.

Table 1. Reliability and test-retest correlation of the Iranian version of the myasthenia gravis activity of daily living (MG-ADL-P) questionnaire

MG-ADL-P items	Mean ± SD	Item-total correlation	Alpha if item deleted	Test-retest correlation
Talking	0.24 ± 0.54	0.68	0.88	0.83
Chewing	0.55 ± 0.65	0.64	0.88	0.95
Swallowing	0.40 ± 0.59	0.79	0.87	0.87
Breathing	0.24 ± 0.47	0.62	0.88	1.00
Impairment of ability to brush teeth or comb hair	0.09 ± 0.38	0.68	0.88	1.00
Impairment of ability to rise from a chair	0.40 ± 0.77	0.62	0.88	0.97
Double vision	0.38 ± 0.67	0.86	0.86	0.93
Eyelid drop	0.52 ± 0.92	0.68	0.88	1.00

MG-ADL: Myasthenia gravis activity of daily living; SD: Standard deviation

The analysis showed correlation coefficients varying between 0.83 and 1 at the item level and 0.99 for MG-ADL-P total score (all with P < 0.001); the ICC was 0.88 [95% confidence interval (CI): 0.83-0.92]. Figure 1 demonstrates the correlation between test and retest MG-ADL-P score. In addition, the mean difference between the test and the retest was 0.45 ± 0.60, and none of the 20 participants had a difference of higher than two points.
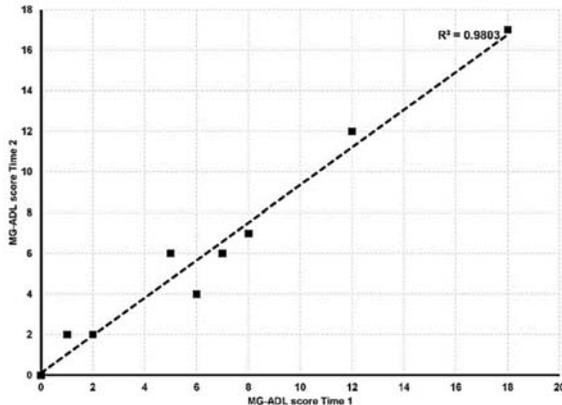

Figure 1. The correlation between test (Time 1) and retest (Time 2) of myasthenia gravis activity of daily living (MG-ADL) score

Validity: The mean scores of the MG-ADL-P and MG-QOL were 5.05 ± 8.39 and 2.81 ± 3.91, respectively. The Pearson correlation coefficients between the MG-ADL-P and MG-QOL15 were conducted to evaluate the validity of the questionnaire. The comparison of the MG-ADL-P questionnaire results showed a positive correlation with MG-QOL15 (r = 0.92, P < 0.001). Figure 2 shows the correlation plot between MG-ADL-P with MG-QOL15 scale.
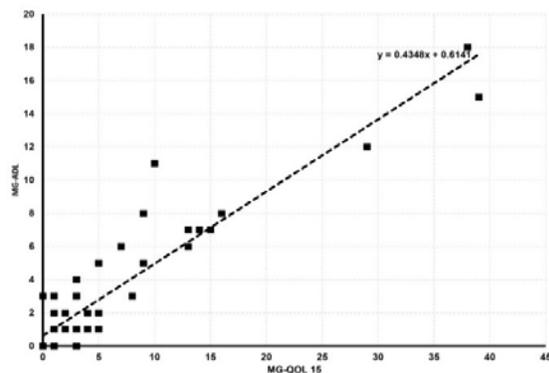

Figure 2. Correlation between myasthenia gravis activity of daily living (MG-ADL) with 15-item Myasthenia Gravis Quality of Life (MG-QOL15) scale

Discussion

The present study was designed to assess the reliability and validity of the the MG-ADL-P questionnaire in the patients with MG. The results of this study indicated good internal consistency, excellent test-retest reliability, and high validity of the MG-ADL-P scale. In this study, Cronbach's α was 0.89 that is a measure of internal consistency, i.e., how a collection of items is related to each other as a group. Several studies evaluated the reliability of the MG-ADL scale in different languages.[15-17] In the Mohd Thabit et al. study, the internal consistency was 0.67-0.69.[18] The internal consistency of the Italian, Arabic, and Turkish versions of MG-ADL was different.[15,16,19] The internal consistency of the Turkish version of MG-ADL was obtained 0.67, similar to the Mohd Thabit et al. study.

On the other hand, in Italian and Arabic versions, the internal consistency was attained as 0.77. In our study, high internal consistency has been obtained. Therefore, the MG-ADL-P is a valuable outcome measurement tool for evaluating clinical manifestations of patients with MG. It is known that MG-ADL assesses different aspects of the clinical impairment affected by MG. These different results in previous studies were predictable due to the multifaceted nature of the scale. The present study showed good test-retest reliability with an ICC of 0.88 that demonstrated the reproducibility of the questionnaire. Several studies have demonstrated excellent test-retest reliability in the different languages.[15-17,19]

The test-retest correlation of the MG-ADL-P scale was excellent for all the items similar to other studies.[9,19] Generally, on a reliable scale, all items should be correlated with the total score. We documented that all scale items were excellent except talking and swallowing items that represented a very good correlation with the total score. Item-total correlations are the correlations between each item and the sum score. In this study, all items showed an acceptable correlation with the total score. For assessment of the validity of MG-ADL-P scale, we evaluated its correlation with the MG-QOL scale. This study revealed a strong correlation between the MG-ADL-P and the MG-QOL scale (r = 0.92). Therefore, this instrument can be a useful tool for use in clinical practice. The previous study in Iran demonstrated excellent reliability and validity of the Persian version of the MG-QOL scale that had already been studied in Iran and showed remarkable internal

consistency.[11] In the previous study by Rozmilowska et al., a high degree of correlation was reported between MG-ADL-P and MG-QOL (r = 0.85).[17] Muppidi et al.[9] and Muppidi[10] found a strong positive correlation between the MG-ADL and the MG composite (MGC) (r = 0.85) as well as between the MG-ADL and the MG-QOL (r = 0.76). Alanazy et al.[15] and Raggi et al.[16] found a similar result as well.

These results indicate that MG-ADL-P, MG-QOL, and MGC scales have standard parameters for evaluating the clinical symptoms of patients with MG.

Conclusion

The MG-ADL questionnaire was translated into the Persian language (MG-ADL-P) and adapted by standard methods. Validity, reliability, and reproducibility were obtained in Persian-speaking patients. Due to the simplicity of this questionnaire, less time is needed to answer the questions compared to other questionnaires. Besides, due to high reliability and validity, this questionnaire can be used to record and follow the status of patients with MG during treatment.

Conflict of Interests

The authors declare no conflict of interest in this study.

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