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Validation of the Individualized Neuromuscular Quality of Life Questionnaire in Korean Patients With Genetic Neuromuscular Diseases

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Background and Purpose The Individualized Neuromuscular Quality of Life questionnaire (INQoL) is a widely used measure of the quality of life in patients with neuromuscular diseases. The purpose of this study was to translate and validate the Korean version of INQoL in Korean patients with neuromuscular diseases.

Methods We translated the original INQoL version into Korean while applying appropriate language adaptations. The internal consistency, known-group validity, and test-retest reliability were also assessed. Construct validity was measured using the modified Rankin Scale (mRS) score and the manual muscle testing (MMT)-sum score based on the Medical Research Council scale, and concurrent validity was measured using the 36-item Short Form Survey (SF-36) questionnaire.

Results This study enrolled 193 patients. The coefficients for internal consistency (Cronbach's α=0.805 to 0.987) and test-retest reliability (Spearman's ρ=0.453 to 0.886) were adequately high for all subscales except in the 'treatment effects' dimension. INQoL subscales other than those for locking, droopy eyelids, double vision, and swallowing difficulties were significantly associated with their relevant SF-36 domains (Spearman's ρ=-0.274 to -0.833). Functional status and muscle strength were most strongly associated with independence (Spearman's ρ=0.753 and p<0.001 for mRS score, Spearman's ρ=-0.741 and p<0.001 for MMT-sum score).

Conclusions The Korean INQoL is a reliable and validated measurement tool for Korean patients with neuromuscular diseases.

Keywords neuromuscular diseases; quality of life; validation study.

INTRODUCTION

Genetic neuromuscular diseases are heterogeneous groups of genetic diseases that primarily affect the muscles, peripheral nerves, or neuromuscular junctions. Approximately 600 causative genes of genetic neuromuscular diseases have been identified.[1] These neuromuscular diseases share the common feature of motor weakness, but the age at symptom onset, rate of disease progression, and pattern of involvement vary widely according to the genetic causes. Many studies have investigated the pathogenic mechanisms and therapeutic candidates of genetic neuromuscular diseases, but a cure remains elusive. There is also an increasing need for reliable biomarkers that assess the severity of disability.

The health-related quality of life (HRQoL) is defined as the perceived physical and mental health of an individual or group.[2] The concept of HRQoL is particularly emphasized in patients with chronic diseases such as genetic neuromuscular diseases, because most of these patients have disabilities and require lifelong management. Therefore, improving

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HRQoL is considered one of the most important therapeutic goals in patients with chronic diseases. HRQoL is often used as an outcome of clinical trials for determining therapeutic benefits.[3] HRQoL is usually measured by patient-reported questionnaires, which are classified into two categories: generic and disease-specific. Disease-specific questionnaires are developed for patients with a specific disease, which allows the HRQoL to be measured more precisely since these questionnaires are better able to reflect disease characteristics such as muscle weakness, pain, or spasticity.

The Individualized Neuromuscular Quality of Life questionnaire (INQoL) is a neuromuscular disease-specific questionnaire that was originally developed in the United Kingdom.[4] It is currently used to assess the disease burden in patients with various neuromuscular diseases.[5,6] INQoL has been translated and validated in several countries, including the United States, Italy, the Netherlands, Serbia, Spain, and Japan.[7-12]

This study aimed to translate INQoL into Korean and validate the new version for Korean patients with neuromuscular diseases.

METHODS

INQoL

INQoL (version 2.0) consists of 45 items within 11 subscales, which are classified into 3 dimensions.[12] The 'symptoms' dimension comprises seven subscales focusing on specific symptoms of neuromuscular diseases: weakness, locking, pain, fatigue, droopy eyelids, double vision, and swallowing difficulties. The 'life domains' dimension comprises five subscales that relate to how much their muscle condition affects the personal lives of patients: activities, independence, social relationships, emotions, and body image. The 'treatment effects' dimension comprises two subscales asking about the positive and negative effects of treatment: perceived treatment effects and expected treatment effects. The quality of life (QoL) score is calculated from the items in the 'life domains' dimension, and represents the level of the patient's overall HRQoL.

All INQoL items are scored on a 7-point Likert scale from 0 to 6 or from 1 to 7. A higher subscale score indicates a greater impact of the disease on the patient's symptoms or life domains, and a higher QoL score indicates a worse overall HRQoL of the patient.

Translation and cultural adaptation

INQoL was translated in accordance with previously reported guidelines.[13,14] Prior to initiating this study, we received approval from the MAPI Research Trust (Lyon), a nonprofit research institute hosting INQoL. We developed the Korean version of INQoL (version 2.0) according to the MAPI guidelines for linguistic validation of a clinical outcome assessment. The forward translation (from English to Korean) of INQoL was executed independently by two neurologists. One neurologist was a researcher who knew the concepts of INQoL, and the other was naïve about them. Differences between the two translations were reconciled through discussion by these two translators plus a senior neurologist.

The draft version of the Korean INQoL was then translated into English by a native English speaker who was bilingual in Korean. The backward-translated version of the questionnaire was reviewed by the forward translators and the original author (M.R. Rose). After several discussions about discordance between the original version and the back-translated version, the revised version of the Korean INQoL was produced. The revised version of the questionnaire was tested on ten patients before making appropriate modifications to produce the final version of the questionnaire.

Patient recruitment and data collection

Patients were recruited at Gangnam Severance Hospital. The inclusion criteria were 1) having a genetic neuromuscular disease, 2) aged >19 years, and 3) able to communicate in Korean. Our study included patients who were able to write or speak despite receiving mechanical ventilation. However, patients who had cognitive impairment or drowsiness were excluded due to the patient-reported nature of the questionnaire. We obtained information regarding sex, age at symptom onset, age at the examination, muscle strength, and genetic causes by reviewing the medical records of the patients and also interviewing them. Muscle strength was assessed using manual muscle testing (MMT) scores based on the Medical Research Council scale (0, 1, 2, 3-, 3, 3+, 4-, 4, 4+, 5-, and 5), which were then converted into an 11-point scale from 0 to 10.[15] The MMT-sum score was the sum of the following 30 strength values: neck flexion, neck extension, and bilateral shoulder abduction, elbow extension, elbow flexion, wrist extension, wrist flexion, finger extension, finger flexion, hip flexion, hip extension, hip abduction, knee extension, knee flexion, ankle dorsiflexion, and ankle plantarflexion. The functional status of the patients was assessed using the modified Rankin Scale (mRS).

The patients were also assessed using the 36-item Short Form Survey (SF-36) for concurrent validation of INQoL. The SF-36 is the most commonly used generic measurement of HRQoL. The Korean version of the SF-36 questionnaire was previously validated in a general population.[16,17] All assessments were performed over a 1-day period in the hospital. Patients who visited the hospital twice within 6 months

repeated the questionnaire to evaluate test-retest reliability.

Statistical analyses

Nonparametric tests were used to assess the association and group differences for INQoL, because most of the INQoL domain scores in the current sample were not consistent with the assumption of normality. Reliability was assessed by the internal consistency, item-total correlation, and test-retest reliability. The internal consistency of items within each subscale was measured using Cronbach's α. Spearman's correlation coefficients were used to assess the test-retest reliability of the questionnaire, and the item-total correlation was assessed between the INQoL subscales of the life domains and the QoL score. Validity was assessed using construct validation (with the mRS and MMT-sum scores) and concurrent validation (with the SF-36 questionnaire). Correlations between the two groups were assessed using Spearman's correlation analysis. The Mann-Whitney U test was used to identify the differences in the INQoL subscales between males and females, with differences considered statistically signif-

Table 1. Genetic profiles of 193 Korean patients with genetic neuromuscular diseases

Disease name	Gene symbol	Total patients (*n*=193)	Retested patients (*n*=20)
Genetically confirmed myopathy		149 (77)	18 (90)
Dystrophinopathy	*DMD*	17 (9)	3 (15)
Myotonic dystrophy	*DMPK*	24 (12)	4 (20)
Facioscapulohumeral muscular dystrophy type 1	*DUX4**	27 (14)	3 (15)
Limb-girdle muscular dystrophy R2	*DYSF*	20 (10)	3 (15)
GNE myopathy	*GNE*	8 (4)	2 (10)
ADSS1 myopathy	*ADSS1*	7 (4)	0
Limb-girdle muscular dystrophy R1	*CAPN3*	6 (3)	1 (5)
Collagen-VI-related myopathy	*COL6A1*	5 (3)	0
Glycogen storage disease type 2	*GAA*	5 (3)	1 (5)
Myotonia congenita	*CLCN1*	4 (2)	0
Distal myopathy with nebulin defect	*NEB*	3 (2)	0
TTN-mutation-related myopathy	*TTN*	3 (2)	0
Progressive external ophthalmoplegia, autosomal dominant 1	*POLG*	2 (1)	0
Myopathy, congenital, with fiber-type disproportion	*RYR1*	2 (1)	0
Limb-girdle muscular dystrophy R4	*SGCB*	2 (1)	0
Myopathy, congenital, with fiber-type disproportion	*TPM3*	2 (1)	0
Very-long-chain acyl-coenzyme A dehydrogenase deficiency	*ACADVL*	1 (1)	1 (5)
Myopathy, distal, Tateyama type	*CAV3*	1 (1)	0
Myasthenic syndrome, congenital, 1A, slow-channel	*CHRNA1*	1 (1)	0
Limb-girdle muscular dystrophy D1	*DNAJB6*	1 (1)	0
Limb-girdle muscular dystrophy R9	*FKRP*	1 (1)	0
Limb-girdle muscular dystrophy R16	*DAG1*	1 (1)	0
Limb-girdle muscular dystrophy R17	*PLEC1*	1 (1)	0
Myasthenia, congenital, 12, with tubular aggregates	*GFPT1*	1 (1)	0
Emery-Dreifuss muscular dystrophy 2, autosomal dominant	*LMNA*	1 (1)	0
Rigid spine muscular dystrophy 1	*SELENON*	1 (1)	0
Facioscapulohumeral muscular dystrophy type 2	*SMCHD1*	1 (1)	0
Inclusion-body myopathy with early-onset Paget disease with or without frontotemporal dementia 1	*VCP*	1 (1)	0
Genetic confirmed neuropathy		18 (9)	1 (5)
Spinal muscular dystrophy	*SMN1*	9 (5)	1 (5)
Spinal and bulbar muscular atrophy	*AR*	7 (4)	0
Charcot-Marie-Tooth disease, type 1A	*PMP22*	1 (1)	0
Charcot-Marie-Tooth disease, type 20	*DYNC1H1*	1 (1)	0
Undiagnosed genetic myopathy		26 (14)	1 (5)

Data are presented as *n* (%).
*Inappropriate reactivation

icant at $p \leq 0.05$. SPSS Statistics software (version 25, IBM Corporation, Armonk, NY, USA) was used for the statistical analyses.

Ethical considerations

This study was approved by the Institutional Review Board (IRB) of Gangnam Severance Hospital, Korea (approval number: 3-2020-0467). Written informed consent was obtained from all patients according to the protocol of the IRB.

RESULTS

Translation and cultural adaptation

Linguistic and cultural differences between English and Korean produced several difficulties in the translation process. Translating 'myotonia' was difficult because there is no such terminology in Korean that is familiar to the general public. Moreover, there were some cultural differences in the selection of the terms, such as 'at the moment' and 'in the face of my condition' in Korean, but these did not cause practical problems or misunderstandings about the meaning of each item.

Patient characteristics

This study enrolled 193 patients (141 males and 52 females) with genetic neuromuscular diseases, of which 175 (91.0%) and 18 (9.0%) had genetic myopathy and neuropathy, respectively (Table 1). The most common disease was facioscapulohumeral muscular dystrophy type 1 (n=27), followed by myotonic dystrophy (n=24), dysferlinopathy (n=20), Becker muscular dystrophy (n=17), and spinal muscular atrophy (n=9). The disease was not confirmed genetically in 26 patients (14.0%). The median ages at symptom onset and the examination were 18.0 years (interquartile range [IQR]=8.0–30.0 years) and 38.0 years (IQR=30.5–46.0 years), respectively. An invasive or noninvasive mechanical ventilator was used in 28 patients (15.0%). The median mRS and MMT-sum scores were 2 (IQR=1.5–3.0) and 230.0 (IQR=176.0–270.0), respectively.

All patients had genetic myopathy (Table 1). The median ages at symptom onset and the examination were 16.0 years (IQR=10.0–23.0 years) and 30.0 years (IQR=22.0–37.0 years), respectively. An invasive or noninvasive mechanical ventilator was used in one patient (7.0%). The median mRS and MMT-sum scores were 2.0 (IQR=1.0–3.0) and 244.0 (IQR=180.0–286.0), respectively.

Descriptive statistics of the INQoL subscales are presented in Table 2. In the 'symptoms' dimension, the most frequently experienced symptom was weakness (94.3%), and the least common symptom was double vision (5.2%). The median score was highest on the weakness subscale (78.9, IQR=63.2–86.8) and lowest on the subscales for droopy eyelids, double vision, and swallowing difficulties (all 0.0, IQR=0.0–0.0). In the 'life domains' dimension, the median score was highest on the activities subscale (75.0, IQR=52.3–90.7) and lowest on the social relationships subscale (50.9, IQR=23.6–71.8). The median QoL score was 66.1 (IQR=48.3–78.6).

Reliability and validity of the Korean INQoL

Table 3 presents the findings for the internal consistency of each subscale. Cronbach's α values other than for expected treatment effects exceeded 0.8, ranging from 0.805 for perceived treatment effects to 0.987 for locking. The average item-scale correlation within each subscale was also strong, with

Table 2. Descriptive statistics of the INQoL

INQoL subscale	Symptom present, *n* (%)	Median [interquartile range]	Range
Muscle weakness	182 (94.3)	78.9 [63.2–86.8]	0–100
Pain	113 (58.5)	36.8 [0.0–68.4]	0–100
Fatigue	169 (87.6)	68.4 [47.4–84.2]	0–100
Locking	77 (39.9)	0.0 [0.0–68.4]	0–100
Droopy eyelids	25 (13.0)	0.0 [0.0–0.0]	0–100
Double vision	10 (5.2)	0.0 [0.0–0.0]	0–100
Swallowing difficulties	29 (15.0)	0.0 [0.0–0.0]	0–100
Activities		75.0 [52.3–90.7]	0–100
Independence		55.6 [22.2–86.1]	0–100
Social relationships		50.9 [23.6–71.8]	0–100
Emotions		63.9 [41.7–80.6]	0–100
Body image		66.7 [47.2–83.3]	0–100
Perceived treatment effect		0.0 [0.0–16.7]	-33.3 to 91.7
Expected treatment effect		0.0 [0.0–16.7]	-50.0 to 100.0
QoL score		66.1 [48.3–78.6]	0–94.4

INQoL, Individualized Neuromuscular Quality of Life questionnaire; QoL, quality of life.

Table 3. Reliability as measured using the internal consistency of items

Subscale	Internal consistency (*n*=193)			Test-retest reliability (*n*=20)	
	Number of items	Range of item-scale correlations	Cronbach's α	Correlation coefficient	*p*
Muscle weakness	3	0.883–0.911	0.924	0.772	<0.001
Pain	3	0.952–0.971	0.973	0.453	0.045
Fatigue	3	0.937–0.962	0.963	0.791	<0.001
Locking	3	0.978–0.984	0.987	0.517	0.020
Droopy eyelids	3	0.974–0.986	0.984	0.886	<0.001
Double vision	3	0.968–0.988	0.985	0.838	<0.001
Swallowing difficulties	3	0.942–0.968	0.971	0.805	<0.001
Activities of daily living	5	0.848–0.886	0.886	0.774	<0.001
Independence	3	0.750–0.923	0.882	0.859	<0.001
Social relationships	10	0.946–0.955	0.954	0.595	0.006
Emotions	6	0.901–0.930	0.927	0.638	0.002
Body image	3	0.788–0.851	0.871	0.678	0.001
Perceived treatment effects	3	0.617–0.801	0.805	0.348	0.133
Expected treatment effects	3	0.348–0.812	0.577	0.224	0.342
QoL score	14	0.925–0.933	0.933	0.840	<0.001

QoL, quality of life.

the correlation coefficient ranging from 0.715 for perceived treatment to 0.990 for locking. Twenty patients completed INQoL twice for assessing the test-retest reliability. All subscales except the 'treatment effects' dimension were significantly correlated (Table 3). Spearman's correlation coefficients ranged from 0.453 for pain to 0.886 for droopy eyelids. All subscales in the 'life domains' dimension were significantly associated with each other, and their Spearman's correlation coefficients ranged from 0.506 for emotions and independence to 0.816 for QoL score and social relationships (Supplementary Table 1 in the online-only Data Supplement). In particular, the Spearman's coefficients for the correlations between the QoL score and all other subscales exceeded 0.7 (Supplementary Table 1 in the online-only Data Supplement).

Table 4 presents the concurrent validity as assessed by evaluating correlations between the INQoL subscales and their relevant domains in the SF-36. The INQoL subscales other than those for locking, droopy eyelids, double vision, and swallowing difficulties were significantly associated with their relevant SF-36 domains (Spearman's ρ=-0.274 to -0.833).

Table 5 indicates that the INQoL subscales other than those for locking, droopy eyelids, double vision, and swallowing difficulties were significantly correlated with the mRS and MMT-sum scores. Among the significantly associated subscales, clinical severity was associated most strongly with independence (Spearman's ρ=0.753 and p<0.001 for mRS score, Spearman's ρ=-0.741 and p<0.001 for MMT-sum score) and most weakly with pain (Spearman's ρ=0.215 and p=0.003 for mRS score, Spearman's ρ=-0.277 and p<0.001 for MMT-sum score). Patients who were older at the examination had lower scores on INQoL subscales such as muscle weakness (Spearman's ρ=0.311, p<0.001), fatigue (Spearman's ρ=0.202, p=0.005), swallowing difficulties (Spearman's ρ=0.213, p=0.003), activities (Spearman's ρ=0.298, p<0.001), independence (Spearman's ρ=0.332, p<0.001), social relationships (Spearman's ρ=0.341, p<0.001), emotions (Spearman's ρ=0.145, p=0.045), body image (Spearman's ρ=0.213, p=0.003), and QoL score (Spearman's ρ=0.300, p<0.001) (Supplementary Table 2 in the online-only Data Supplement). Females had higher scores on the fatigue subscale (Mann-Whitney U=2852.5, p=0.018) (Supplementary Table 2 in the online-only Data Supplement). The scores on the INQoL subscales were not associated with the age at symptom onset (Supplementary Table 2 in the online-only Data Supplement).

DISCUSSION

The present results indicate that the Korean INQoL has adequate reliability and validity for Korean patients with neuromuscular diseases. The present Korean patients had difficulty understanding the term myotonia, which was also found in a Dutch study,[7] but all of them fully understood the other contents of the Korean INQoL despite the presence of cultural differences. Our study also included patients with various neuromuscular diseases. INQoL was initially developed for a wide range of neuromuscular diseases,[18] but many previous studies primarily involved patients with myopathies.[7,8,11,12,19] Therefore, one strength of this study is that it has expanded

the scope of the diseases covered by INQoL, including genetic neuropathies.

The internal consistency and test-retest reliability were high except for the 'treatment effects' dimension, which might be because that dimension is intended to evaluate how patients perceive their ongoing treatment. Moreover, this study investigated patients with genetic neuromuscular diseases. Although gene therapy is being investigated widely at present, most interventions are symptom-relieving treatments rather than disease-modifying treatments. Therefore, the concept of treatment effects might be ambiguous for patients, and they might feel differently about treatment effects depending on their condition or mood at the time when they complete a questionnaire. These findings were similar to those of previous validation studies.[8,12] Thus, the 'treatment effects' dimension may be inadequate for clinical usage.[10-12] Our results for the item-total correlations between INQoL subscales of the life domains and QoL score in the Korean population were similar to some previous results[8,12] and moreover were strongly correlated with the Dutch results.[7]

The INQoL subscales other than those for locking, droopy eyelids, double vision, and swallowing difficulties had moderately strong correlations with the SF-36 items in Korean patients with genetic neuromuscular diseases. These findings were similar to those of a Japanese study.[12] The weak associations of SF-36 items with four disease-specific symptoms indicate that they have been ignored in other generic questionnaires. Therefore, our study emphasized the importance of disease-specific questionnaires such as INQoL in patients with neuromuscular diseases. The INQoL subscales other than those for locking, droopy eyelids, double vision, and swallowing difficulties were negatively affected by older age and severe disabilities (based on mRS and MMT-sum scores). Functional status and muscle strength generally affect the QoL, and previous studies have demonstrated that the mRS

Table 4. Concurrent validity, assessed using the correlations between INQoL subscales and items on the SF-36

INQoL subscale	SF-36 item	Correlation coefficient	*p*
Weakness	Physical functioning	-0.635	<0.001
	Role physical	-0.619	<0.001
Pain	Pain	-0.561	<0.001
Fatigue	Energy/fatigue	-0.571	<0.001
Locking	Physical functioning	-0.112	0.120
	Role physical	-0.215	0.003
Droopy eyelids	Physical functioning	-0.124	0.086
	Role physical	-0.106	0.144
Double vision	Physical functioning	-0.148	0.039
	Role physical	-0.122	0.092
Swallowing difficulties	Physical functioning	-0.143	0.048
	Role physical	-0.138	0.056
Activities	Physical functioning	-0.698	<0.001
	Role physical	-0.677	<0.001
	Social functioning	-0.622	<0.001
Independence	Physical functioning	-0.833	<0.001
	Role physical	-0.603	<0.001
Social relationships	Social functioning	-0.714	<0.001
Emotions	Role emotional	-0.449	<0.001
	Emotional well-being	-0.611	<0.001
Body image	Role physical	-0.387	<0.001
	Role emotional	-0.274	<0.001
QoL score	General health	-0.531	<0.001
	Health change	-0.514	<0.001

INQoL, Individualized Neuromuscular Quality of Life questionnaire; SF-36, 36-item Short Form Survey; QoL, quality of life.

Table 5. Known-group validity, assessed using the correlations between INQoL subscales and clinical severity as measured using the mRS score and the MMT-sum score based on the Medical Research Council scale

INQoL subscale	mRS score		MMT-sum score	
	Correlation coefficient	*p*	Correlation coefficient	*p*
Weakness	0.523	<0.001	-0.566	<0.001
Pain	0.215	0.003	-0.277	<0.001
Fatigue	0.325	<0.001	-0.316	<0.001
Locking	0.061	0.397	-0.142	0.049
Droopy eyelids	-0.038	0.605	-0.060	0.409
Double vision	0.093	0.201	-0.139	0.055
Swallowing difficulties	0.031	0.673	-0.115	0.110
Activities	0.560	<0.001	-0.546	<0.001
Independence	0.753	<0.001	-0.741	<0.001
Social relationships	0.413	<0.001	-0.418	<0.001
Emotions	0.258	<0.001	-0.296	<0.001
Body image	0.369	<0.001	-0.391	<0.001
QoL score	0.473	<0.001	-0.466	<0.001

INQoL, Individualized Neuromuscular Quality of Life questionnaire; MMT, manual muscle testing; mRS, modified Rankin Scale; QoL, quality of life.

score and measured muscle strength are correlated with the INQoL subscales.[7,8,12] However, locking, droopy eyelids, double vision, and swallowing difficulties are affected more by the presence of a specific disease than by the functional status due to the disease severity. We also found that the INQoL subscales were negatively affected by older age at the time of the examination. This is similar to the findings of a Japanese study, and may be associated with disease progression.[12]

Our study had some limitations. First, the study population was relatively small. However, the subject-to-item ratio was 4.29, which could be considered fair,[20] and the total sample size and subgroup sizes (for test-retest reliability) of the study were not inferior to those in other studies validating INQoL in Japan and Serbia.[11,12] The number of hospital visits during the study period was also relatively small, which is probably due to most genetic neuromuscular diseases being chronic and disabling. Completing the questionnaire over the phone or on the internet might be useful for overcoming these accessibility limitations. Second, the time interval between the two tests when assessing the test-retest reliability was both inconsistent and slightly longer than those in previous studies. Even though we only included patients with chronic and genetic neuromuscular diseases, this inconsistency might have underestimated the test-retest reliability. Third, this study did not assess the responsiveness of the questionnaire, and so this should be evaluated in future studies with long-term observations.

In conclusion, the Korean INQoL is a reliable and validated measurement tool for Korean patients with neuromuscular diseases. Further studies of its responsiveness and subgroup analyses are needed. This questionnaire could be widely used as a unified tool to assess the clinical course and treatment response of patients with neuromuscular diseases in Korea.

Supplementary Materials

The online-only Data Supplement is available with this article at https://doi.org/10.3988/jcn.2022.18.5.514.

Availability of Data and Material

The datasets generated or analyzed during the study are available from the corresponding author on reasonable request.

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Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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