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Brazilian Version of the Myelomeningocele Functional Classification (MMFC): Translation, Cultural Adaptation, and Psychometric Properties

Versão brasileira da classificação funcional de mielomeningocele (MMFC): Tradução, adaptação cultural e propriedades psicométricas

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Abstract

Objective: Perform the translation and cultural adaptation of the Myelomeningocele Functional Classification (MMFC) into Portuguese (Brazil) and study its psychometric properties.

Method: Validation study with translation, cultural adaptation and evaluation of psychometric properties: reliability, test-retest and convergent validity. Sample of 20 individuals with myelomeningocele with a median age of 10 (5 - 24.25) years, with a minimum of 3 and a maximum of 66 years. Reliability was determined by intra and interobserver agreement, using the results of the Intra-class Correlation Coefficient (ICC) and Confidence Interval 95% (IC - 95%). Convergent validity was performed using the Sharrard, Hoffer, Pediatric Evaluation of Disability Inventory (PEDI) and Functional Mobility Scale (FMS) classifications, and The Spearman Correlation Test was calculated.

Results: Intra (ICC range: 0.900-1.0) and interobserver (ICC: 0.936; IC - 95%: 0.839-0.975) reliability showed excellent levels of ICC. Convergent validity showed very strong correlations with FMS-5 ($r = 0.94$, $p = 0.00$) and FMS-50 ($r = 0.94$, $p = 0.00$); strong correlations with FMS-500 ($r = 0.87$, $p = 0.00$), Sharrard ($r = 0.76$, $p = 0.00$), Hoffer ($r = 0.83$, $p = 0.00$), PEDI Functional Skills: Mobility ($r = 0.84$, $p = 0.00$) and PEDI Caregiver Assistance: Mobility ($r = 0.77$, $p = 0.00$); and weak correlations with self care domain of PEDI ($r = 0.46$, $p = 0,04$). The test-retest showed ICC = 1.00.

Keywords
- classifications
- gait
- myelomeningocele
- prognosis
- validation study

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Conclusions: This study presents the psychometric properties of the MMFC, in addition to its translation and cultural adaptation into Portuguese, the native language of the author of the classification. MMFC demonstrates correlation with previously used myelomeningocele classifications. MMFC demonstrated good results in the psychometric properties evaluated. Thus, the MMFC seems adequate and applicable to individuals with myelomeningocele and valid for the Brazilian population.

Resumo

Objetivo: Realizar a tradução e adaptação cultural da Classificação Funcional da Mielomeningocele (MMFC) para o português (Brasil) e estudar suas propriedades psicométricas.

Método: Estudo de validação com tradução, adaptação cultural e avaliação das propriedades psicométricas: confiabilidade, teste-reteste e validade convergente. Amostra de 20 indivíduos com mielomeningocele e idade mediana de 10 (5 a 24,25) anos, com mínimo de 3 e máximo de 66 anos. A confiabilidade foi determinada pela concordância intra e interobservador, utilizando os resultados do coeficiente de correlação intraclasse (ICC) e o intervalo de confiança de 95% (IC - 95%). A validade convergente foi realizada por meio das classificações de Sharrard, Hoffer, Inventário de Avaliação Pediátrica de Incapacidade (*Pediatric Evaluation of Disability Inventory* [PEDI]) e Escala de Mobilidade Funcional (*Functional Mobility Scale* [FMS]). Além disso, o teste de correlação de Spearman foi realizado.

Resultados: A confiabilidade intraobservador (ICC: 0,900-1,0) e interobservador (ICC: 0,936; IC - 95%: 0,839-0,975) apresentou excelentes níveis de ICC. A validade convergente mostrou correlações muito fortes com FMS-5 ($r = 0,94$, $p = 0,00$) e FMS-50 ($r = 0,94$, $p = 0,00$); correlações fortes com FMS-500 ($r = 0,87$, $p = 0,00$), Sharrard ($r = 0,76$, $p = 0,00$), Hoffer ($r = 0,83$, $p = 0,00$) e Habilidades Funcionais: Mobilidade (PEDI) ($r = 0,84$, $p = 0,00$) e Assistência do Cuidador: Mobilidade (PEDI) ($r = 0,77$, $p = 0,00$); e fracas com o domínio autocuidado de PEDI ($r = 0,46$, $p = 0,04$). O teste-reteste revelou que ICC = 1,00.

Conclusões: Este estudo apresenta as propriedades psicométricas da MMFC, além de sua tradução e adaptação cultural para o português, língua nativa do autor da classificação. A MMFC demonstra correlação com classificações de mielomeningocele anteriormente utilizada. A MMFC teve bons resultados nas propriedades psicométricas avaliadas. Assim, a MMFC parece adequada e aplicável a indivíduos com mielomeningocele e é válida para a população brasileira.

Palavras-chave
- classificação
- estudo de validação
- marcha
- meningomielocele
- prognóstico

Introduction

Myelomeningocele (MM) is one of the congenital defects of neural tube closure in which the meninges, spinal cord and nerve roots are exposed, which causes motor and sensory losses in the lower limbs. Besides this, spinal abnormalities (hydromyelia, syringomyelia) or central (hydrocephalus, Arnold-Chiari) add motor and balance deficits.[1] The clinical and functional presentation varies depending mainly on the neurological level but being greatly influenced by the presence of spinal and lower limb deformities, neurological complications, obesity, motivation, among others.[2] Classifying each individual from a functional point of view is therefore very important, as it helps the anticipated necessary interventions and treatments and the prognosis regarding the degree of independence in daily life.

Classifying the different presentations in MM has already been the target of many researchers. Sharrard determined one of the best known classifications, taking into account the neurological level of the lesion (thoracic, lumbar, sacral).[3] Broughton proposed a modification in Sharrard's neurosegmental levels.[4] Hoffer classified patients according to their ambulatory capacity (non-ambulatory, non-functional walker, home walker and community walker).[5] McDonald et al.[6] made the classification based on the degree of muscle strength of the lower limbs, as did Asher et al.[7]; the first also aimed at the correlation with the prediction of walking ability. In Lindseth's classification,[8] the motor level is based on voluntary joint motor control. The comparative analysis of these studies, however, showed incompatibility between classifications based on anatomy and those based on functional and ambulation criteria,[9] essential for establishing treatment goals.

As in cerebral palsy, with the development of the Gross Motor Function Classification System (GMFCS)[10] and the Functional Mobility Scale (FMS)[11] a classification covering the various aspects of the clinical and functional presentation was developed for MM – the Myelomeningocele Functional Classification (MMFC).[12] The classification takes into account the muscular strength of the lower limbs, the type of external support and orthoses required for ambulation and the gait capacity, through the distance reached. Like the GMFCS and FMS classifications, it is an easy-to-interpret classification that has a prognostic value in terms of function, aiding in the therapeutic planning and communication between the team members that participate in the patient's treatment, in addition to expanding the understanding of its evolution. The use of this classification also allows for a more accurate assessment of treatment results.

The MMFC was published in English[12] and, understanding the importance of this comprehensive classification, this study proposes its translation, cultural adaptation and its psychometric validation, the latter not yet presented in the literature. Our purpose is to make its use more accessible in all services involved in the treatment of patients with MM in Brazil, standardizing data for the purpose of communication and presentation of results. Thus, the objectives of this study are: translation and cultural adaptation of the MMFC content into Portuguese (Brazil) and study the psychometric properties of the MMFC in the Brazilian version.

Method

Descriptive observational study, cross-sectional, divided into two phases: Phase 1- Translation and Cultural Adaptation of the MMFC into Portuguese; Phase 2–Psychometric Validation.

Phase 1, Translation and Cultural Adaptation of the MMFC into Portuguese, was developed following the stages proposed by Beaton et al.[13]: Stage 1: Translation by two translators English – Portuguese; Stage 2: Harmonization between both resulting in a single Portuguese version; Stage 3: Back-translation of the harmonized version initially by two Portuguese – English translators; Stage 4: Harmonization between both translators resulting in a single English version; Stage 5: International harmonization, where the versions resulting from the first and second harmonization are evaluated with the original author of the questionnaire.

For Phase 2, the sample was selected for convenience in rehabilitation clinics in southern Brazil. From 40 patients diagnosed with MM who were invited to participate, 22 accepted the invitation, and two were excluded due to eligibility criteria. Inclusion criteria were: Brazilian individuals with MM, with no age limit; and those of exclusion: those in the postoperative period of orthopedic surgery less than 6 months and with associated pathologies that affect motor function, such as cerebral palsy, among others. The RDNP author performed the evaluations of the 20 participants, and half of the participants came after 10 to 15 days for a new evaluation and stability evaluation (test-retest). The recruitment and data collection occurred from July 2021 to February 2022.

The psychometric properties evaluated were:

- Reliability

Intra-examiner reliability was determined by 15 professionals with experience in the field, selected for convenience (physiotherapists and orthopedists). They received by e-mail the MMFC classification in the Portuguese version and a video with the data to be analyzed for each patient. They classified the individuals on two occasions separated by two weeks, sending the result to the authors after each stage was completed. The data consisted of the case presentation (age, clinical characteristics), video demonstrating assessment of manual muscle strength of lower limbs (quadriceps, hamstrings, gluteus medius, gastrocnemius-soleus), video of gait for short distances (5 meters), including the use of orthoses and external devices, if used, and interview about walking capacity for medium (50 meters) and long distances (500 meters). The entire physical examination and interview were performed by the same researcher.

Inter-examiner reliability was assessed through the response of the first evaluation of the 15 professional examiners.

- Convergent validity

For convergent validity, one author classified the 20 individuals by MMFC, Hoffer, Sharrard and FMS and performed the manual assessment of muscle strength of lower and collected data of the Pediatric Evaluation of Disability Inventory (PEDI).[14–17]

Test Retest

The stability of the classification was evaluated by the test-retest, in which 10 individuals, after 10 to 15 days, were again evaluated by the same evaluator.

This study obtained ethical approval from a Ethics and Research Committee and the participants or their caregivers gave informed consent to the research and the publication of the results.

Statistical analysis was performed using the Statistical Package for the Social Sciences (SPSS) software, version 20.0 (SPSS Inc., Chicago, IL). In all cases, the significance level was set at 5%. Inter and intra-examiner reliability and repeatability were assessed using the results of the Intra-class Correlation Coefficient (ICC), considering a significance level of $p < 0.05$. An ICC value equal to 1 indicates that the values are identical in intra and inter-examiner comparisons or in the repeatability of the method. ICC values below 0.70 were considered unacceptable; between 0.71 and 0.79, acceptable; between 0.80 and 0.89, as very good; and above 0.90, excellent. The Spearman Correlation Test was used and the results classified according to the correlation coefficient (r): very strong ($r > 0.9$), strong (r from 0.7 to 0.9), moderate (r from 0.5 to 0.7) and weak (r from 0.3 to 0.5)

Results

In phase 1, translation and cultural adaptation, there were only grammatical and vocabulary disagreements between

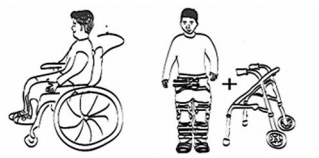
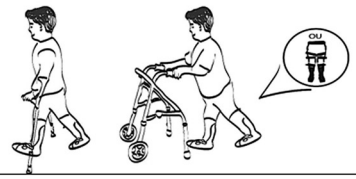
Functional classification of myelomeningocele				
Group	**Main functional feature**	**FMS Classification**	**Auxiliary Devices**	**Illustration**
MMFC1	It may or may not maintain psoas function. It has no function of the quadriceps.	Maximum 2/2/1	- Walker - Orthoses spanning the hip (reciprocal gait orthosis - RGO or long tutor with pelvic belt - HKAFO) - Wheelchair for long distances	
MMFC2	Maintains function of quadriceps and medial hamstrings. Does not have gluteus medius function.	Maximum 3/3/3	- Walker or crutches - Long brace with pelvic belt (HKAFO) or suropodal orthoses (AFO)	
MMFC3	Maintains quadriceps and gluteus medius function. Does not have gastrocnemiusoleus function	Maximum 5/5/5	- No external support - Suropodal orthoses (AFO) only	
MMFC4	Maintains quadriceps, gluteus medius and gastrocnemiusoleus function	Maximum 6/6/6	- No external support - Supramalleolar orthoses, insoles or nothing	

Fig. 1 Brazilian Version of Myelomeningocele Functional Classification (MMFC).

the translators, which did not affect the semantic equivalence of the content. They were discussed and harmonized and, due to the practical content of the classification, there were no problems of idiomatic and cultural equivalence (colloquial expressions). Only a small cultural adaptation was necessary, adding: 1- to the term Hip Knee Ankle Foot Orthosis (HKAFO) the term *tutor longo com cinto pélvico*, clinically used in Brazil, and 2- to the term Ankle Foot Orthosis (AFO), the term suropodalic orthosis was added. The Brazilian version of the MMFC is shown in ►**Fig. 1**.

Phase 2 - Psychometric Validation - involved a total of 20 individuals with MM, ranging in age from 3 to 66 years. ►**Table 1** presents the general characteristics of the sample. ►**Table 2** presents the neurosegmental and functional classifications of the individuals evaluated.

Table 1 Demographics and clinical characteristics of the sample of individuals with myelomeningocele

Variable	Total sample (n = 20)
Demographic Characteristic	
Age, years*	10 (5–24.25)
Sex, n (%)	
Male	7 (35)
Female	13 (65)
Race, n (%)	
White	19 (95)
Black	1 (5)
Clinical Characteristics	
Diagnosis, n (%)	
Pre-natal	13 (65)
Post-natal	7 (35)
Surgical repair, n (%)	
Intrauterine	1 (5)
24 hours	16 (80)
48 hours	2 (10)
1 week	1 (5)
Ventricular Peritoneal Shunt, n (%)	
Yes	16 (80)
No	4 (20)

*Age expressed by median (interquartile range). Other variables as absolute frequency (relative frequency) = n (%).

Table 2 Neurosegmental and functional classifications of the individuals with myelomeningocele evaluated

Variable	Total sample (n = 20)
Sharrard, n (%)	
Thoracic/ high lumbar	7 (35)
Low lumbar	8 (40)
Sacral	5 (25)
Hoffer, n (%)	
Community ambulator	12 (60)
Non-functional ambulator	2 (10)
Non ambulator	6 (30)
FMS 5 meters, n (%)	
1	7 (35)
2	3 (15)
3	3 (15)
4	1 (5)
5	5 (25)
6	1 (5)
FMS 50 meters, n (%)	
1	7 (35)
2	2 (10)
3	5 (25)
5	5 (25)
6	5 (5)
FMS 500 meters, n (%)	
1	10 (50)
2	2 (10)
3	2 (10)
5	5 (25)
6	1 (5)
MMFC, n (%)	
1	6 (30)
2	8 (40)
3	4 (20)
4	2 (10)

Abbreviations: FMS, *Functional Mobility Scale;* MMFC, *Myelomeningocele Functional Classification.*

Reliability

►**Table 3** presents the excellent levels of ICC in both interobserver and intraobserver reliability, and in intraobserver reliability identical values were observed in 60% of the examiners.

Convergent Validity

►**Table 4** shows the significant correlations of the MMFC with the functional scales evaluated.

Test-Retest

The MMFC reproducibility analyzed by the test-retest after 10 to 15 days of the first evaluation in half of the sample showed ICC =1.00.

Discussion

This is the first study to assess the psychometric properties of the MMFC. The present research translated and culturally adapted the classification into Brazilian Portuguese, the native language of the main author and of the author of the classification, and showed that the MMFC has excellent intra and inter-observer reliability, with excellent reproducibility and convergent validity with strong or very strong correlations with PEDI, FMS and the standard Hoffer and Sharrard classifications.

Many studies have concluded that the most important factor in determining the level of functionality of patients with MM is the level of neurological involvement.[3–5,7] One of the best-known classification of MM by neurological levels is Sharrard's. It has been used in many studies of evaluation and indication of treatment.[7] This classification, however, may not fully portray functional aspects as they can vary greatly within each level, depending on several factors, such as age, body mass index, motivation, presence of neurological conditions that interfere with balance, spinal and lower limb deformities that make standing and gait difficult, social aspects, type of orthosis and external support for gait, among others. Thus, the most complete classification is the one that also encompasses functional aspects and the pillars of the International Classification of Functioning Disability and Healthy (ICF) - Body functions and structures, Activities and Participation and Environmental Factors. Within this perspective, the MMFC represents an important classification tool, as it includes these factors. The addition of the FMS to this classification further details the functional and performance profile of the individual and the MMFC must always be applied together with FMS.

The evaluation of the quality of measurement instruments is important for the selection of instruments that provide valid and reliable measurements[18] The need for an adequate evaluation of the measurement properties in the evaluation instruments has been strongly recommended by the literature.[19] To our knowledge, the evaluation of the psychometric properties of the MMFC was not published, which is essential for its use not only in daily practice but also in future studies and publications.

The excellent results observed in the intra and interobserver reliability of the MMFC can not be compared, as there are no previous studies verifying this outcome in the literature. The reliability were performed by physical therapists and orthopedists in the area of neuro-orthopedic rehabilitation and they received a written explanation about the classification, showing the ease of understanding and performing it.

This study is the first presenting the convergent validity of the MMFC. A very strong to strong correlation was observed with the Sharrard and Hoffer classifications, PEDI and FMS

Table 3 Inter and Intraobserver reliability of Myelomeningocele Functional Classification (MMFC)

Observer	1ª evaluation*	ICC Inter (IC95%)&	2ª evaluation*	Absolute value of difference	ICC Intra (IC 95%)&
1	2.25 ± 0.83	0.936 (0.839-0.975)	2.35 ± 0.85	0.10	0.965 (0.913-0.986)
2	2.05 ± 0.86		2.00 ± 0.89	0.05	0.984 (0.961-0.994)
3	2.10 ± 0.94		2.10 ± 0.94	0	1.00
4	2.16 ± 0.81		2.00 ± 0.89	0.16	0.967 (0.917-0.987)
5	2.1 ± 0.89		2.1 ± 0.89	0	1.00
6	2.25 ± 1.04		2.35 ± 1.01	0.10	0.953 (0.883-0.981)
7	2.30 ± 1.31		2.10 ± 0.94	0.20	0.900 (0.752-0.960)
8	2.15 ± 0.91		2.15 ± 0.91	0	1.00
9	2.15 ± 1.01		2.15 ± 0.91	0	0.974 (0.933-0.990)
10	2.05 ± 0.92		2.05 ± 0.92	0	1.00
11	2.15 ± 0.85		2.15 ± 0.85	0	1.00
12	2.00 ± 0.89		2.15 ± 0.89	0.15	0.952 (0.873-0.981)
13	2.10 ± 0.89		2.10 ± 0.89	0	1.00
14	2.05 ± 0.86		2.05 ± 0.86	0	1.00
15	2.05 ± 0.92		2.05 ± 0.92	0	1.00

*Values for the first and second evaluation expressed as medium ± standard deviation of the total sample evaluated by each examiner.
&ICC= intraclass correlation coefficient (IC= confidence interval 95%).

Table 4 Spearman's Correlation showing concurrent validity of Myelomeningocele Functional Classification (MMFC)

Variables	r (p)	Magnitude
FMS 5m	0.94 (0.00)	Very strong
FMS 50m	0.94 (0.00)	Very strong
FMS 500m	0.87 (0.00)	Strong
Sharrard	0.76 (0.00)	Strong
Hoffer	0.83 (0.00)	Strong
PEDI Functional Skills: Mobility	0.84 (0.00)	Strong
PEDI Caregiver Assistance: Mobility	0.77 (0.00)	Strong
PEDI Caregiver Assistance: Self care	0.46 (0.04)	Weak
PEDI Functional Skills: Self care	0.46 (0.04)	Weak

Expressed as Spearman's correlation coefficient r (p value): Very Strong ($r > 0.9$), strong (r from 0.7 to 0.9), moderate (r from 0.5 to 0.7), and weak (r from 0.3 to 0.5).

mobility scales; and weak correlation with the PEDI self-care scale. Other studies have compared the various classifications of MM. Rethlefsen et al.[20] showed the relationship between the previous classification Dias Functional Classification of Myelomeningocele (FCM) - proposed by Dias et al.,[12] similar to the MMFC, but which did not include the use of the FMS - with other classifications of MM. The researchers analyzed 61 patients with spina bifida (77% MM and 23% lipomyelomeningocele) aged 6 to 16 years who were classified by FCM, FMS, Hoffer Classification, radiological neurological level (International Myelodysplasia Study Group - IMSG) and by muscle strength and ability to walk. They found an excellent correlation between FCM and FMS, for all distances, and a weak to moderate correlation between the Hoffer classification and the FCM.[20] The analysis between the FCM and the neurological level by muscle strength showed an excellent correlation.[20] The FCM did not show an excellent correlation with radiological classification.[20] In another study, Battibugli et al.[21] studied 161 patients with MM in order to examine the influence of the presence of ventriculoperitoneal shunt on the gait. They classified the patients using neurological level, lower extremity muscle strength and the type of external support for gait and FMS. Some of these patients were also assessed through temporal-spatial gait parameters of computerized gait analysis. The authors demonstrated that participants with a shunt had significantly lower FMS 500 and FMS 50 scores compared with participants without a shunt, but they did not find correlation with 5m score.[21] Despite a smaller number of patients analyzed, the present study included only patients with MM and covers different age groups and neurological levels, but still found results very similar to those depicted above. Regarding the excellent correlation of FCM between all FMS distances found by Rethlefsen et al.,[20] our results showed that the MMFC found excellent or very strong correlation with FMS 5 meters and FMS 50 meters. There was, however, a strong correlation with FMS 500 meters, possibly due to the older patients in our sample, in which more functional patients no longer had much independence over long distances, using wheelchair.

Another comparative study between the various classifications was published by Bartonek et al.,[9] where 73 patients were analyzed using the classifications of Sharrard, Hoffer, Lindseth, Broughton and Ferrari. They demonstrated that, based on the neurological level, it is not possible to identify or predict functional ability using any of the classifications. Tita et al.[22] also compared data taken from the medical records of 409 adult patients, classifying their neurological level using two versions of the National Spina Bifida Patient Registry Classification, the International Standards for Neurological Classification of Spinal Cord Injury motor level and the Broughton classification and comparing them with Hoffer's gait ability. They noted a significant correlation between the Hoffer classification and all neurological level scales evaluated, with the strongest correlation being with the Broughton classification.

As a differential in this study, we added the analysis of aspects of activity of ICF through the PEDI, and we obtained a strong correlation between the MMFC with mobility both for functional skills and with the amount of caregiver assistance and a weak correlation with self-care. We therefore show that the higher the MMFC, the greater the functionality of the individual, greater mobility and self-care and greater independence to walk. This demonstrates that the ability to stand up and have mobility is a determining factor for greater independence in day-to-day activities. The weak magnitude for the correlation with self-care is possibly due to the fact that the MMFC measures gait, mobility and self-care activities depend more on functional skills of the upper limbs.

This study has limitations, is a small sample from a specific geographic region, which can not reflect characteristics of the MM population; patients with prior orthopedic or neurosurgical procedures were included, which could have impacted muscle strength and its associations with other variables. We also did not control for other variables as access to rehabilitation programs, presence of ventriculoperitoneal shunt, etc which could have influenced the results. Also, there is a limitation in the age limits of some assessment instruments, since this study included adult patients.

Conclusion

The MMFC showed a strong correlation with the previously used MM classifications. The Brazilian version of the MMFC showed good results in the psychometric properties evaluated: intra and inter-observer reliability, test-retest and convergent validity. Thus, the MMFC seems adequate and applicable to individuals with MM and valid for the Brazilian population.

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Conflict of Interests
The authors declare no conflict of interest.

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