Development of a Novel Orofacial Motor Function Assessment Scale for Children With Cerebral Palsy

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ABSTRACT

Purpose: The aim of this study was to develop the orofacial motor function assessment scale (OFMFAS) and appraise its performance in terms of validity and reliability in a cohort of 116 cerebral palsy patients.

Methods: The scale was developed according to a process derived from the theory of measurement and scale development previously described in the dental literature. The final version of the OFMFAS comprises 13 items that have 30 subitems. Results were statistically analyzed by Bland-Altman for: (1) intrarater consistency; (2) kappa coefficient for inter-rater agreement; and (3) Cronbach alpha for internal consistency reliability.

Results: The majority of OFMFAS items showed a very good agreement between the raters (κ>0.75). Cronbach alpha for the 13-item scale was 0.93, indicating excellent internal consistency reliability.

Conclusion: This study shows that the oralfacial motor function assessment scale is the first statistically-based scale for the quantitative assessment of oral-motor skills in cerebral palsy children. It is an easy-to-use, accurate, and valid method of assessment. (J Dent Child 2005;72:113-118)

KEYWORDS: CEREBRAL PALSY, ORAL-MOTOR DYSFUNCTION, VALIDITY, RELIABILITY, CHILDREN

The 4 main requirements for oral-motor development are: (1) stability and mobility of the ingestive system; (2) rhythmicity; (3) sensation; and (4) oral-motor efficiency and economy.[1] The oral preparatory phase of swallowing involves a complex range of activities that include food-getting, mastication, bolus management, and transport.[2] Marked impairments in the development of motor coordination interfere with daily living activities, including all normal oral-motor functions.

The behavioral expression of the many different pathologies falls into 3 categories:

1. resistance to accepting food orally;
2. lack of energy and endurance to do the "work" of eating;
3. oral-motor disabilities, resulting in an inability to produce the necessary motor skills for ingestion.[1]

Cerebral palsy (CP), defined as a range of nonprogressive syndromes of posture and motor impairment, is a common cause of disability in childhood.[3,4] This disorder results from various insults to different areas within the developing central nervous system, which partially explains the variability of clinical findings.[5] Clinical patterns of involvement described in cerebral palsy include: diplegia, hemiplegia, quadriplegia and double hemiplegia. Movement disorders can coexist with the clinical patterns of involvement, and there can be spasticity, rigidity, hypotonia, dystonia, or a mixture of these disorders.[6]

CP's clinical classification has been based upon a description of the motor deficits. Pure forms of motor dysfunction rarely occur, if ever, and classification by description of motor deficits suffers from low interobserver agreement.[7]

CP children have a high prevalence of feeding problems, often resulting in malnutrition.[8] A major cause of these problems is oral-motor dysfunction (ie, difficulty in chewing and swallowing.[9]) Orofacial dysfunctions are a severe health problem, with feeding problems being the most common feature

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presented by these children.[10] The child with oral-motor impairment has a great difficulty getting food into the mouth and, additionally, there are frequent problems with food loss due to excessive drooling, coughing, choking, and poor motor control.[11]

Oral-motor skills are often dysfunctional in CP patients. Lingual dysfunction, specifically tongue thrust, is consistently observed. Other patterns of oral dysfunction include hyperactive and hypoactive gag reflexes, oral hypersensitivity, and prolonged and exaggerated bite reflexes. Inadequate function of cheek and lip musculature may prevent formation of an adequate oral lip seal, leading to food/liquid loss and inhibiting distal propulsion of an organized bolus.[12-15]

Primitive reflexes of the normal infant such as suckle-swallow, rooting, gagging, and biting are essential for infant survival and are part of the normal development.[16] The persistence of these and other primitive reflexes such as the asymmetrical tonic neck reflex (ATNR), often observed in patients with developmental delay, cerebral palsy, or head injury, however, can interfere with the patient's feeding skills.[17-19] Abnormal responses such as the biting and suckle-swallow reflexes, lack of tongue lateralization, jaw instability, and phasic biting can severely limit an individual's ability to masticate, position, and swallow a food bolus safely. Mastication may be limited to the basic jaw opening and closing movements with no jaw lateralization.[2]

There are no known dental literature studies that have evaluated CP patients' orofacial motor function, without determining food texture or fluid intake, for the purpose of establishing a CP population assessment standard based on clinical patterns, movement disorders, and oral-motor involvement.

The purpose of this study was to describe the development of the orofacial motor function assessment scale (OFMFAS) and its performance in terms of validity and reliability in a cohort of 116 CP patients. The development and performance of the OFMFAS are being reported in an additional paper.

METHODS

MEASUREMENT GOALS

In developing the OFMFAS, the authors aimed to generate a scale applicable to CP children who present a wide range of associated disorders mediated by the central nervous system. This means that the OFMFAS needed to be sensitive to the functional oral-motor abilities of these individuals. Considering that the OFMFAS was intended to be used as an outcome measurement in intervention studies, it needed to be sensitive to changes in addition to being valid and reliable.

DEVELOPMENT PROCESS

The OFMFAS was constructed according to a process derived from the theory of measurement and scale development previously described in the dental literature.[20,21] This study's protocol was approved by the Ethics Committee of the Universidade Federal de Sao Paulo-Escola Paulista de Medicina, Brazil. After being informed about the aims of the investigation, a written consent was obtained from the responsible adult for each child participating in the study.

STEPS TO DEVELOP THE OFMFAS

Three steps were used to develop the OFMFAS: (1) a literature review; (2) a new assessment scale; and (3) validity and reliability testing.

LITERATURE REVIEW

A search strategy, identified from Medline and comprising the period between 1974 to 2004, was used to identify relevant studies reflecting methods used for: (1) reflex; (2) oral reflexes; (3) oral-motor patterns; (4) oral-motor skills; and (5) oral assessment. This search was matched with CP, and a few studies were found.[1-3,15,17,19,22,23]

GENERATION OF A NEW ASSESSMENT SCALE

The OFMFAS' conceptual framework for the OFMFAS was developed through a review of the concepts of motor oral function. The OFMFAS items were generated in 2 steps:

1. A preliminary pool of 35 items was developed by abstracting items from existing questionnaires.
2. A face and content validation study was conducted involving 2 dentists who assessed 12 CP patients. In the second step, the preliminary item pool was reviewed for its comprehensiveness, relevance, and clarity. Based on scores and comments, a modified pool was developed by excluding irrelevant items, writing of additional items, and combining items. The OFMFAS final version consisted of 13 items with 11 jaw movement subitems, 2 facial movement subitems, 3 lip movement subitems, 1 glossopharyngeal and vagal motor movement subitem, 1 palatal movement subitem, 8 tongue movement subitems, and 4 oral reflex subitems—totaling 30 subitems

A Likert scale ranging from 0 (unable to perform or determine; inconsistent) to 2 (performed adequately) was used to fill out the scale by 2 different dentists.

VALIDITY AND RELIABILITY TESTING

The performance of OFMFAS was assessed through a validity and reliability study. Despite OFMFAS being been developed to assess oral-motor skills in CP patients, it was initially applied by 2 dentists to 20 normal individuals (ie, with no neurological damage) who were within the same age range of patients in the study. The objective was to establish the maximum and minimum scores obtained for normal individuals.

During data collection, 2 other dentists recruited and assessed a new sample of 116 CP children attending a special grade school and the odontological unit of Lar Escola Sao Francisco Rehabilitation Center, Universidade Federal de Sao Paulo, Brazil. For the test-retest reliability assessment, the OFMFAS was applied twice to all patients by both raters within a 2-week interval. As reliability has been defined as the ratio between-subject variance and total variance,[24] the OFMFAS total and subtotal scores were generated by adding the numerical response codes.

To evaluate intrarater consistency of each rater's test-retest measurement, a Bland Altman[25] analysis was used in the total score measurement. Each test item of the final version of the OFMFAS was examined statistically for raters' agreement using the kappa coefficient[26] for inter-rater agreement.

Internal consistency reliability of the total scale and subtotal scales of OFMFAS was assessed by means of Cronbach alpha.[27] Significance test level was set at $P<.05$.

RESULTS

CLINICAL FEATURES OF PATIENTS

The results of OFMFAS assessments in individuals with no neurological damage had maximum and minimum scores of 60 and 57, respectively, for both examiners (means±SD=59.3±0.92 and 59.5±0.61, respectively).

A total of 116 CP patients with cerebral palsy were enrolled in the main study to assess validity and internal consistency reliability. The clinical features of all patients regarding gender, age, movement disorder, and dysfunction patterns are shown in Table 1.

Table 1. Characteristics of the 116 Cerebral Palsy Children Evaluated by the OFMFAS

Movement Disorder	Clinical Patterns	Male (N=70) %	Female (N=46) %	Total (N=116) %
Spasticity	Diplegia	16 (23)	14(30)	30 (26)
	Hemiplegia	10 (14)	0	10 (9)
	Quadriplegia	16 (23)	24 (52)	40 (35)
	Double hemiplegia	2 (3)	6 (13)	8 (7)
Dystonia		20 (29)	0	20 (17)
Mixed		6 (9)	2 (4)	8 (7)
Mean age		10.7±3.2	10.3±2.6	10.5±2.9

The total scale score ranged from 0 to 60 (mean ±SD=29.45±15.36 and 29.55±15.35 for the first and the second examiner, respectively).

There was a significant correlation between scale total score for test and retest for both raters ($r=0.997$, $P<.001$). Using Bland-Altman plot, the repeatability coefficients showed a very good agreement between the results obtained at the first and second assessments for each rater. The difference mean was:

1. 0.09, with a 95% confidence interval (range=2.4 - 2.2) for the first rater;
2. 0.07, with a 95% confidence interval (range=2.5 - 2.6) for the second rater.

Each test item of OFMFAS' final version was examined statistically for rater agreement using a kappa coefficient. They were calculated for each pair of raters for each of the 30 subitems. Data are shown in Table 2.

The authors observed that, in the majority of the items, there was a very good agreement between the raters ($\kappa>0.75$). Only items 6 and 7 showed a good agreement ($0.45<\kappa<0.75$).

RELIABILITY

Internal consistency reliability was assessed by means of Cronbach alpha, which was 0.93 for the 13-item scale—indicating excellent internal consistency reliability. This was not improved by the deletion of any item. All item-total score correlations were significant at $P<.001$ level.

Finally, based on score descriptive analyses (with the scores varying from 2 to 58), the authors divided the scores in quartiles according to the severity of the condition observed in the OFMFAS: (1) first quartile=≤19 (severely impaired); (2) second quartile=20 to 31 (moderately impaired); (3) third quartile=32 to 41 (slightly impaired); and (4) fourth quartile=≥42 (very slightly impaired).

The data of the 116 CP patients assessed via the OFMFAS are presented in Table 3.

Table 2. Kappa Coefficient for Each Pair of Raters for Each of the 13 Items, With Respective Subitems

Item	κ a	b	c	d
Jaw mobility	0.939	0.869	0.960	0.957
Voluntary jaw protrusion	1.000	0.908		
Voluntary lateral jaw movements	1.000	0.928	1.000	
Rapid coordinated jaw movements	0.801	0.800		
Voluntary facial movements	0.875	0.897		
Lip muscle strength: puffed-out cheeks/maintain pressure	0.713			
Rapid coordinated lip movements	0.711	0.871		
Glossopharyngeal and vagal motor	0.811			
Rapid coordinated palatal movements	0.821			
Hypoglossal motor: voluntary tongue movements	1.000	0.938		
Voluntary elevation and lateralization of tongue	0.877	0.942	0.956	0.952
Rapid coordinated movements of tongue	0.909	0.749		
Oral abnormal reflexes	1.000	1.000	1.000	0.880

DISCUSSION

This study describes the development and evaluation of the OFMFAS in CP children. No known dental literature studies exist that have assessed CP patients' orofacial motor function, without determining food texture or fluid intake, in order to establish a CP population assessment standard based on clinical patterns, movement disorders, and oral-motor involvement.

Since the ultimate goal was to develop a measurement system that could be used in clinical trials and evaluation research, the scale was de-

Table 3. Cerebral Palsy Patients' Distribution According to Oral-Facial Motor Function Assessment Scale (OFMFAS): Degrees of Impairment

OFMFAS	Movement Disorder	Clinical Patterns	Distribution(N)
≤19=severely impaired (N=30)	Spasticity (N=28)	Diplegia	6
		Hemiplegia	-
		Quadriplegia	18
		Double hemiplegia	4
	Mixed (N=2)	Quadriplegia	2
20-31=moderately impaired (N=28)	Spasticity (N=20)	Diplegia	4
		Hemiplegia	2
		Quadriplegia	12
		Double hemiplegia	2
	Dystonia (N=4)	-	4
	Mixed (N=4)	Quadriplegia	4
32-41=slightly impaired (N=32)	Spasticity (N=20)	Diplegia	10
		Hemiplegia	2
		Quadriplegia	6
		Double hemiplegia	2
	Dystonia (N=10)	-	10
	Mixed (N=2)	Hemiplegia	2
≥42=very slightly impaired (N=26)	Spasticity (N=20)	Diplegia	10
		Hemiplegia	6
		Quadriplegia	4
		Double hemiplegia	-
	Dystonia (N=6)	-	6

veloped using a process previously described[20,21] consisting of a description of the most frequent oral-motor problems that affect CP patients.

Therefore, the most important motor functions involved in the mastication, swallowing, and phonation processes[2,11,17] were included in the scale as an instrument to assess these patients' oral-motor capacity. The presence of pathological oral reflexes was also included, since they are observed in a large number of individuals with neurological lesions.

When considering a measurement system for use in clinical practice or clinical trials, clinicians and investigators should look for evidence that is pertinent to these individuals and, in fact, important for the target population.[28]

A content-valid protocol for the assessment of oral-motor skills must have enough items to satisfy the full domain of content relevant to the assessment. It is impossible to determine the exact number of items required to satisfy the domain of content, but it is always better to begin with the construction of too many test items rather than too few, since inadequate items can always be eliminated later.[29] A total of 5 items were discarded from the pilot study and others were altered or reworded as a result of kappa coefficient analyses. The final version of the OFMFAS, which was applied, is shown in Figure 1.

The selection of the items included in this scale allowed for the assessment of the oral functions that are more frequently impaired in CP patients. This enabled the detection of the alterations and, consequently, the classification of the individuals according to their oral-motor capacities, and not just as CP patients.

The gross motor performance assessment and the pediatric disability evaluation index are considered to be the best standard measurements, because they are complementary and test different function features also used for CP children.[30] Nevertheless, these 2 measurement tools cannot assess the oral-motor skills in these patients. The authors also had to include the great variability of clinical findings in CP children[5] and interobserver variation in the diagnosis, since they are important sources of bias in studies of CP patients.[7]

The distribution of the most common clinical patterns (ie, diplegia, quadriplegia, and hemiplegia) is difficult to assess, because specific diagnoses are not available.[5] In this study, the authors used the movement disorder and the clinical patterns, since all of the patients were enrolled in a rehabilitation program and the CP diagnosis was medical. For dentists who do not work together with a multidisciplinary team, however, such a clinical pattern of distribution is not readily accessible at patients' dental evaluation

This study's results show that OFMFAS is a reliable assessment protocol for CP children with varied degrees of involvement regarding their oral-motor skills. Furthermore, only 7 to 10 minutes are required to complete a patient's assessment sheet, since it does not involve feeding patients a range of food textures and fluids, as in the schedule for oral-motor assessment.[23]

The very good rater agreement shows that the OFMFAS has been successfully designed to be used by different professionals who treat CP patients.

Each OFMFAS item was statistical analyzed. The high inter-rater agreement of 0.93 for the total of 30 subitems demonstrates OFMFAS reliability between the raters.

The assessment of physically and/or mentally handicapped subjects is difficult, and the subjects of this study were no exception. Those who have worked with such patients understand the difficulties an investigator must deal with in a clini-

cal study that assesses performance skills. Decreased motor control, sensory-motor deficits, communication difficulties, and altered cognitive functions challenge the investigator's ability to assess these individuals accurately.[2] The variability in their OFMFAS performance is an example of the degrees of functional oral levels of these patients.

Conflicting results regarding caries, oral hygiene, flow rate, and malocclusion found in literature may be due to the fact that these patients are identified only by the nonspecific code G80.0 and G80.9 from the International Classification of Diseases (ICD 10) and not taking into account the real oral-motor conditions.

This study's results showed that it was possible to identify quadriplegic and diplegic CP patients in all quartiles, which allowed the authors to infer that the global motor involvement is not directly related to oral performance. The gastrotomized patients who aspirate and present significantly poorer oral-motor skills in spoon-feeding, biting, chewing, and swallowing[22] showed the lowest OFMFAS scores.

This protocol's reliability is important in that it reflects the consistency of results with repeated measurements. To date, there is no gold standard for oral-motor skill assessment protocols in CP patients. The improvement in oral-motor skills may help these children ingest food more competently (ie, with less spillage[22]), enhance oral hygiene performance, and decreasing the risk of oral diseases.

The analysis presented here demonstrates that the OFMFAS has good construction validity and good internal consistency reliability. This scale, when used by dentists or other professionals who work with a CP population, can help to establish an adequate and real odontological treatment program based on oral-motor limitations, thus contributing to an improvement in the quality of life of CP patients.

CONCLUSIONS

This study shows that the orofacial motor function assessment scale is the first statistically-based scale for the quantitative assessment of oral-motor skills in children with cerebral palsy. It is an easy-to-use, accurate, and valid means of oral-motor skill assessment in these children.

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