DOI: https://doi.org/10.53350/pjmhs22167482

ORIGINAL ARTICLE

Validation of Pedsql™ Duchenne Muscular Dystrophy Module (Acute Version 3.0), in Young Children (Age 5-7)

ANAS ALI[1], MUHAMMAD ASIF JAVED[2], HAMZA SHAHZAD[3], ABDUL MAJID SHAHID[4], SHAMOOS SAQIB[5], AYESHA MALIK[6]
[1]*Clinical physiotherapist Rising Sun Institute for special children*
[2]*Assistant professor Riphah international university Lahore*
[3]*Physiotherapist Rising sun institute for special children Lahore. hamzash.17@gmail.com*
[4]*Consultant physiotherapist Boston physiotherapy and wellness clinic.*
[5]*Physiotherapist/Content Director at Tibbi shamoossaqib@gmail.com*
[6]*Physiotherapist Amna Memorial medical center near UBL, main GT road rachna town Shahdara Lahore*
Corresponding author: Anas Ali, Email: Anasali424@yahoo.com, Cell: 03314456918

ABSTRACT

Objective: The goal of this project was to accurately translate the "PedsQLTM 3.0 DMD" Module into Urdu and to assess its validity in health-related quality of life in the Duchenne muscular dystrophy (DMD)" children age 5 to 7 years reported by their parents.

Method: The study was conducted with the parents/caregivers of DMD children in Rising Sun Institute, PSRD, Hope,Gosha e Shifa, Special gems, Ghazali Foundation Rawalpindi and DHQ hospital Jhang. Study was cross cultural validation including 56 primary care givers. First of all questionnaire of primary care giver was translated in Urdu by using forward translation, the second step was reconciliation of items; in this the best Urdu translation was selected. Backward translation was completed in the third phase, and a review was conducted to evaluate the full forward-backward procedure in order to produce a final forward translation. Validity & Reliabity were measured. To determine in the internal consistency Cronbach's Alpha was used Test-Retest reliability was checked. Content Validity Index and Content Validity Ratio were measured through 5 experts. To analyze the collected data SPSS 26 was used.

Results: The results showed that in both genders from age 5 to 7 years DMD affects the quality of life of child as reported by parents. The value of Cronbach's alpha was 0.913.After factor analysis KMO was 0.793 and because the p value was less than 0.05, the test was significant.

Conclusion: Urdu version of "PedsQL™ Duchenne Muscular Dystrophy Acute Version 3.0(age 5-7) Parent report for young children" is a stable and consistent instrument for assessing quality of life measures of the DMD children and can be applied in Urdu speaking population throughout the world.

Keywords: Duchenne Muscular Dystrophy, "PedsQL™ 3.0 DMD Module", Urdu translation, validity, quality of life.

INTRODUCTION

PedsQL™ 3.0 DMD Module" is required for the examination of the quality of life in children aged 5 to 25 who have DMD. The "PedsQL™ DMD 3.0 module" has four scales that analyze the child's "Daily Operations," "Therapeutic Obstacles," "Worry," and "Interaction" over the preceding seven days in the age group 5-7. It usually includes Parent reports as well as DMD child self-reports. "The Pediatric Quality of Life Inventory™ (PedsQL™) 3.0 DMD Module" was created to evaluate the standard of living in individuals with DMD aged 5 to 18 (1). \

Variations found in the DMD gene on the X chromosome induce Duchenne Muscular Dystrophy The dystrophin located on the inner side of the membrane (sarcolemma) of skeletal and cardiac muscle cells is controlled by this gene (2). The absence of dystrophin leads to destrutctions of muscle fibers during contraction. The chronic nature of this destruction will cause inflammation, which in turn hinders muscle fiber regeneration. Eventually, these changes lead to the substitution of muscle by fibrotic and adipose tissue through a process called fibrosis (3). DMD is X linked recessive inheritance disease, but more mutations are possible and two-thirds of sufferers do not have a strong family history too (4). DMD is a devastating kind of neurological disease that affects one out of every 3,500 to 6,000 male infants. DMD is normally begins between the ages of three and six (5). Initial observations in children with DMD could include delays in attaining developmental stages such as sitting or standing without aid; toe walking; an odd ambling gait; difficulty ascending stairs or rising from a seated posture (Gower's sign); and recurring falls. Due to muscle damage, babies and young children may seem awkward and clunky, with unnatural expansion of the calves (pseudohypertrophy). Global learning disability is another sign. Fatigue is also very common in DMD which leads to irreversible muscular weakness. Due to fatigue, DMD children may not show interest in daily functional activities which limits their social participation. It also leads them towards dependence on caregivers. Some DMDs children may also have memory issues (cognitive impairment) and learning problems (6). Children with Duchenne acquire significant walking and breathing problems in the long run, and the muscles that help them breathe and the heart eventually cease working, resulting in premature mortality, usually in their second to third decade of life(7). Mostly DMD patients have 85 intelligence quotients (IQ). Activity Cognition is lower than verbal intelligence. Intellectual impairment has no relation with muscular weaknesses or other symptoms. ADHD and mental disorders are also common in males with DMD (8).

Parent feedback in form of a report or questionnaire is very important because it tells the health status during daily activities information in Duchenne muscular dystrophy children (1). "The Pediatric Quality of Life Inventory™ (PedsQL™) 3.0 DMD Module" was created for the evaluation of the standard of living in individuals with DMD aged 5 to 18 (9). In Thailand, researchers have conducted studies on establishing a Thailand's version of "PedsQL 3.0."According to them DMD is a chronic illness which have an effect on the children's physical health, intellectual, emotional, and recreational activities, but also disturbs their relative's lives. PedsQL™ 3.0 DMD Module" is require for examination of quality of life in children aged 5 to 25 who have DMD. The "PedsQL™ DMD 3.0 module" has four scales that analyze the child's "Daily Operations," "Therapeutic Obstacles," "Worry," and "Interaction" over the preceding seven days in the age group 5-7. It usually includes Parents reports as well as DMD child self-reports. In Past few years many studies have attempted QOL as an assessment tool in studies of children with Cerebral palsy but for DMD no suitable measure to check the Quality of life has been designed Quality of life of children with DMD in several countries, cultures and medical setups should be done by using the different questionnaires to improve life quality and expectancy. Whenever feasible, the World Health Organization (WHO) encourages that child's life standard be studied via self-reporting (10). Children, even the tiniest ones, have been found capable of selfreporting. However, ideas suggest that testimonies may be misleading due to instability, immaturity, intensity of illness or disease, and cognitive impairment. When children are unable to self-report, their caregivers can conduct a level of health evaluation

since it is assumed that they have a high knowledge of their kids and could provide truthful data regarding them. It has been well acknowledged that the both self-reporting and parent reports contribute valuable supplementary data about a child's quality of life (11).

The purpose of the current project was to translate "PedsQL™ Duchene Muscular Dystrophy module Acute Version 3.0, Parent report for young children (age 57)" and measure its reliability & validity so that it could be used by the physical therapists to sort out the problems of DMD children with the help of Urdu speaking parents/caregivers.

METHOD

The study was conducted with the parents/caregivers of DMD children in Rising Sun Institute,

PSRD, Hope, Gosha e Shifa, Special gems, Ghazali Foundation Rawalpindi and DHQ hospital Jhang. Study was cross cultural validation of pedsQL™ Duchenne muscular dystrophy Module (acute version 3.0), in young children (age 5-7).

First of all questionnaire of primary care giver was translated in Urdu by using forward translation, the second step was reconciliation of items; in this the best Urdu translation was selected. Backward translation was completed in the third phase, and a review was conducted to evaluate the full forward-backward procedure in order to produce a final forward translation.The Urdu translation of the PedsQL™ Duchenne Muscular Dystrophy Module (Acute version 3.0), Parent Report for Children (Age 5-7) questionnaire was carried out after obtaining permission from the original scale's designer. Translation was done is following steps:

Forward Translation: Two individuals, one of whom had adequate understanding of all aspects of physiotherapy, independently translated the English Questionnaires into URDU.

Differences were then evaluated by comparing both Urdu translations. A consensus was obtained, and two distinct translations were combined into a single joint translation.

Expert Panel Back Translation: Two bilinguals who were also fluent in Urdu independently transcribed the Urdu questionnaire into English and were unaware of the questionnaire

Contrasts between the new back-translated questionnaire and basic questionnaire were assessed.

The committee's final version of the questionnaire was administered to five persons, and the questionnaire's functionality was evaluated throughout the application

Caregivers/Parents were recruited based on inclusion and exclusion criteria which was following

Inclusion Criteria: Parents of children aged 5 to 7 years confirmed with DMD by Neuro Physician.

Parents who were capable of delivering data or comprehending the questionnaire's subject explanations.

Parents who were familiar and could spoke Urdu Language.

The caregivers who were capable of fulfilling out the surveys on their own.

Exclusion Criteria: Parents of children with some other neurological disorders e.g. (Autism, Dyspraxia, Cerebral palsy, ADHD, Epilepsy and Dyslexia) or psychological disorders were not acceptable.

Care givers/Parents that weren't able to spoke Urdu.

Sample was taken by convenient sampling technique were questioned if they wanted to take part in the research or not, then the sample of 56 willing parents were elaborated and given Urdu

Translated "PedsQL™ Questionnaire" to mark the activities performed then Scoring was done according to activities performed. 56 parents filled the questionnaires. Questionnaire was again filled by the same Parents of DMD children after 7 days to check Test–retest reliability which was checked by ICC.

RESULTS

Results showed that out of 28 DMD children 14 were boys and 14 were girls whose parents filled the questionnaire. According to results 12 children had age 5 years, 11 children had age 6 years and 5 children had age 7 years whose parents participated in study. Cronbach's alpha for "PedsQL Questionnaire" was 0.913 showing that this scale is reliable tool for assessing the quality of life of DMD Children reported by their parents. Interclass correlation coefficient ICC showed good reliability with value Single Measure 0.370 and Average Measure 0.91.

Questionnaire was again filled by the same Parents of DMD children after 7 days. Test–retest reliability which was checked by ICC ranged between .877 to.943 for "Urdu translation and Validation of PedsQL Duchene Muscular Dystrophy module Acute Version 3.0 Parent report for young children (age 5-7)" values of less than 0.4 indicates the fair reliability .50 to .69 moderate reliability, 0.70 to 0.79 shows good reliability and value that greater than 0.80 shows excellent reliability.

Questionnaire was rated by 5 experts for measuring content validity index on four different grounds which were simplicity, clarity, ambiguity & relevance four point ordinal scales. Average of each scale was measured four each item. Then average for each question rated by expert was calculated for CVI to find out the content validity of 18 items of "PedsQL Urdu" version.

Formula for content validity ratio was CVR= (N_e-N/2)/ (N/2) in which N represents the total number of panelists whereas N_e was represents the number of panelists that indicating essentials By using Lawshe the numeric value of content validity ratio was calculated.

To determine the discriminant validity independent t test was used Independent t-test indicates differences in means of all variables for both groups. Result showed that statistics showed satisfactory discriminant validity of both groups shown in table-1.

Table-1: Discriminant validity of Urdu Translation of PedsQL™ Duchenne Muscular Dystrophy Module (Acute version 3.0), Parent Report for Children (Age 5-7) Questionnaire: Group Statistics:

	Male	Female
Daily Activities (PROBLEM WITH)		
Q1: Trouble eating with a fork and knife.	2.05±1.13	1.83±0.98
Q2: Difficulty writing or draw with a pen or pencil	2.27±0.98	2.00±1.26
Q3: Difficulty putting his or her own clothes.	2.73±0.83	2.33±0.82
Q4: Using the toilet without assistance.	3.36±0.58	2.50±0.84
Q5: Need more time than others to complete tasks.	3.32±0.89	3.17±0.98
Treatment (PROBLEM WITH)		
Q6: Taking medicines	1.95±1.13	2.50±0.55
Q7: Physical therapy or daily medicines Causing pain	2.23±1.15	2.67±0.52
Q8: Difficulty being responsible for his or her medicines or physical therapy.	2.50±1.22	2.50±0.55
Q9: Difficulty managing his or her muscle problem.	3.09±1.15	3.17±0.41
Worry (PROBLEM WITH)		
Q10: Worrying about his or her muscle problem.	2.86±1.08	3.00±0.63
Q11: Worrying about whether or not my medicines are working	2.73±1.08	3.00±0.63
Q12: Worrying about his or her family	2.77±1.19	3.33±0.52
Q13: Worrying about needing help from others	3.23±0.75	3.17±0.41
Q14: Worrying about not being accepted from others.	3.09±0.81	3.00±0.63
Q15: Worrying about being treated differently from others my age	2.59±0.80	2.67±0.82
Communication (PROBLEM WITH)		
Q16: Difficulty telling the doctors and nurses how he/she feel.	2.82±0.91	3.00±0.00
Q17: Difficulty asking the doctors and nurses questions	2.73±0.88	3.00±0.63
Q18: Difficulty Explaining his/her muscle problem to others the doctors and nurses how I feel	2.91±0.97	3.00±0.63

After factor analysis of sample of 56 parents of DMD children KMO was 0.793 and the p value was less than 0.05 which showed

significance of the test there was no Sphericity issue (Bartlett's test 0.01). Three variables were responsible for the variance in data. Result showed that data had maximum variance in 3 domains. Factor 1 had 44.364% variance, factor 2 had 15.727% variance and factor 3 had 12.462% variance. There were 3 factors on the plot which had value greater than 1 out of 18 items in all domains, which showed that these components were those over which all items would be loaded shown in figure-1.

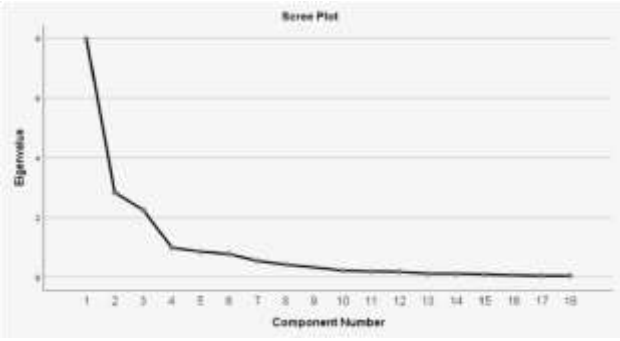

Figure-1: Discriminant validity of Urdu Translation of PedsQL™ Duchenne

DISCUSSION

Duchene Muscular Dystrophy lead to the chronic disabilities and restricted the daily life activities of the young children .They didn't participate or showed interest in any of the chores as the symptoms of the dystrophy progresses (11). DMD also effected the parent quality of life, they had to stick with the child to help them in doing house hold chores or child's personal work (12).

Current study tried to translate a tool which was intended to have the parents/caregivers feedback of DMD children, how they were living and what problems parents as well as children are facing in routine days. This Scale in Urdu helped the parents to give their responses what issues they were facing in treating the DMD child as well as their own health. Urdu version of "PedsQL™ Duchenne Muscular Dystrophy Acute Version 3.0(age 57) Parent report for young children" would also help experts to provide facilities for parents/caregivers of DMD children and decide the best interventions which could help them to play some role in society.

All the researches in which PedsQL™ had been translated into local languages had commuted these variables of tool and the current study was very close to them. This study found satisfactory internal consistency having the Cronbach's Alpha coefficient value of 0.91, and majority of studies had Cronbach's Alpha mean value that was similar to the current study. Original Scale was also translated into the UK-English language and in comparison to current study results of internal reliability i.e. 0.90 was same as its results. Children suffering from chronic illnesses had more problems with their parents in comparison to DMD children. When "PedsQL™ 4.0 Parent report" was used in healthy population children its Overall Scale Score had an internal consistency reliability of = 0.90, which was the same as the current study (13).

In another study in which of "PedsQL™ Duchenne Muscular Dystrophy Acute Version 3.0(age 57) Parent report for young children" was translated into Chinese the Chronbach alpha for parents was 0.86 nearer to the current study (14). In article "The PedsQLy* 4.0 as a Pediatric Population Health Measure: Feasibility, Reliability, and Validity" the results were similar to current study as population taken was general pediatric population and their parent's reports (15).

The measuring features of "PedsQL 3.0 Generic Core Scales" in DMD population health were presented in current study. Current findings confirmed "PedsQL 3.0's" reliability and validity as parent-reported standard of living related to health measurement instrument for DMD young children aged 5 to 7.

It had been shown via all of the discussion that Urdu translated "PedsQL Duchenne Muscular Dystrophy Acute Version 3.0(age 5-7) The Parent Report for Young. Children" questionnaire was a relevant and trustworthy tool that could be used in health care settings throughout the whole country to provide accurate diagnostic outcomes related to general population quality of life.

CONCLUSION

"PedsQL™ Duchenne Muscular Dystrophy Acute Version 3.0(age 5-7) Parent report for young children" was translated in Urdu with forward and backward translation protocols. Analysis of the collected data through parents showed that it was a reliable and valid questionnaire to determine the Quality of life of DMD children.

REFERENCES

1. medicine WGJP. Development of the World Health Organization WHOQOL-BREF quality of life assessment. 1998;28(3):551-8.
2. Karalaki M, Fili S, Philippou A, Koutsilieris MJIv. Muscle regeneration: cellular and molecular events. 2009;23(5):779-96.
3. Yiu EM, Kornberg AJJNI. Duchenne muscular dystrophy. 2008;56(3):236.
4. Fernandes LA, Caromano FA, Hukuda ME, Escorcio R, Carvalho EVJBJoPT. Elaboration and reliability of functional evaluation on going up and downstairs scale for Duchenne Muscular Dystrophy. 2010;14:518-26.
5. Choi Y-A, Shin H-I, Shin HIJBmd. Scoliosis in Duchenne muscular dystrophy children is fully reducible in the initial stage, and becomes structural over time. 2019;20(1):1-8.
6. Birnkrant DJ, Bushby K, Bann CM, Apkon SD, Blackwell A, Brumbaugh D, et al. Diagnosis and management of Duchenne muscular dystrophy, part 1: diagnosis, and neuromuscular, rehabilitation, endocrine, and gastrointestinal and nutritional management. 2018;17(3):251-67.
7. Hendriksen JG, Vles JSJJocn. Neuropsychiatric disorders in males with duchenne muscular dystrophy: frequency rate of attention-deficit hyperactivity disorder (ADHD), autism spectrum disorder, and obsessive—compulsive disorder. 2008;23(5):477-81.
8. Gravelle AMJJoan. Caring for a child with a progressive illness during the complex chronic phase: parents' experience of facing adversity. 1997;25(4):738-45. 12. Thongsing A, Likasitwattanakul S, Sanmaneechai OJH, Outcomes QoL. Reliability and validity of the Thai version of the Pediatric Quality of Life inventory™ 3.0 Duchenne Muscular Dystrophy module in Thai children with Duchenne Muscular Dystrophy. 2019;17(1):18.
9. Whitney N. Health-Related Quality of Life of Children with Duchenne Muscular Dystrophy: University of Northern Colorado; 2021.
10. Deeken JF, Taylor KL, Mangan P, Yabroff KR, Ingham JMJJop, management s. Care for the caregivers: a review of self-report instruments developed to measure the burden, needs, and quality of life of informal caregivers. 2003;26(4):922-53.
11. Kohler M, Clarenbach CF, Bahler C, Brack T, Russi EW, Bloch KEJJoN, Neurosurgery, et al. Disability and survival in Duchenne muscular dystrophy. 2009;80(3):320-5.
12. Baiardini I, Minetti C, Bonifacino S, Porcu A, Klersy C, Petralia P, et al. Quality of life in Duchenne muscular dystrophy: the subjective impact on children and parents. 2011;26(6):707-13.
13. Upton P, Eiser C, Cheung I, Hutchings HA, Jenney M, Maddocks A, et al. Measurement properties of the UK-English version of the Pediatric Quality of Life Inventory™ 4.0 (PedsQL™) generic core scales. 2005;3(1):1-7.
14. Hu J, Jiang L, Hong S, Cheng L, Kong M, Ye YJH, et al. Reliability and validity of the Chinese version of the pediatric quality of life inventory TM (PedsQL TM) 3.0 neuromuscular module in children with Duchenne muscular dystrophy. 2013;11(1):1-9.
15. Varni JW, Seid M, Kurtin PSJMc. PedsQL™ 4.0: Reliability and validity of the Pediatric Quality of Life Inventory™ Version 4.0 Generic Core Scales in healthy and patient populations. 2001:80012.