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CROSS-CULTURAL VALIDATION OF THE ACTIVITIES SCALE FOR KIDS PERFORMANCE VERSION: PROOF OF RELIABILITY AND VALIDITY IN ITALIAN CHILDREN WITH CEREBRAL PALSY

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To those who value the journey
more than the destination.

To Valerio, for his full support
and infinite patience.

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CANDIDATE PROFILE

Stefania Costi was born in Sassuolo (Modena), Italy, on May 15, 1971.

She has a Bachelor of Science degree in Physiotherapy (1996), a Master of Science degree in Management of Health Education (2000), a Master of Science degree in Assistive Technology (2005), a Master of Science degree in Rehabilitation Science (2006), a Master of Science degree in Management of Allied Health Professions, and a Master of Science degree in Research Methodology (2009).

She also completed a clinical research fellowship training program in the field of Pulmonary Rehabilitation at West Park Healthcare Centre in Toronto (Ontario), Canada (2004-2005).

She has been working as a physiotherapist since 1996 and has been the Coordinator of the Bachelor of Science in Physiotherapy degree program of the University of Modena and Reggio Emilia since 2014. She also has been a lecturer in Research Methodology for the same degree program.

Further, she has been a Researcher (Track A) in the field of physiotherapy (SSD MED/48) at the University of Modena and Reggio Emilia since 2016.

Her didactic, clinical, and scientific interests concern the field of research methodology in physiotherapy, with a special emphasis on:

- cerebral palsy
- pulmonary rehabilitation
- cancer rehabilitation
- development of academic learning skills

Ph.D. SCIENTIFIC ACTIVITIES REPORT (2016-2019)

The following reports the main Ph.D. research activities conducted during the 3-year course. Part I describes the scientific activities and projects dealing with the main topic of the Ph.D. project. It also contains a summary of additional scientific activities carried out. Part II contains a list of the dissemination activities of the results of the research carried out in the timeframe 2016-2019.

Part I: Main Scientific Activities

YEAR 1

In the first year, much of my time and energy focused on developing the research project and on its submission to the Local Ethics Committee, and on recruiting patients with cerebral palsy. I supervised and coordinated the work of the co-authors, especially those involved in data collection. Moreover, I contacted all the elementary and middle schools of the city of Reggio Emilia in order to inform them about the project and ask for their collaboration.

As for additional scientific activities, I continued working on a research project on pulmonary rehabilitation in patients undergoing surgical resection for lung cancer, writing the protocol of a clinical trial that has been published (Appendix V).

Moreover, I continued working on a research project regarding the return to work of cancer survivors: I actively participated in the selection and appraisal of the literature in this field and participated in the analyses of the data collected in the Province of Reggio Emilia.

YEAR 2

Much of my work during the second year focused on completing the data collection on healthy children, continuing the data collection on children with cerebral palsy, and supervising and coordinating the co-authors' work. I also performed preliminary analyses of data collected on both healthy children and on children with cerebral palsy. I also submitted the final part of the research project regarding the assessment of convergent validity of the ASKp to a call for research proposals (FAR 2017) and obtained the financial support necessary to complete data collection and to disseminate the results.

YEAR 3

Most of my work focused on completing data collection, analyzing the data, and writing three manuscripts, which are currently under submission and constitute the results of this research project.

Preliminary data on the reliability and validity of the ASKp in healthy children were presented in a poster at the XXXI Conference entitled "Asfissia perinatale ed encefalopatia ipossico-ischemica: prevenzione, diagnosi, terapia e riabilitazione", held in Modena (Italy) on 20-22 March 2019 (Appendix II).

Data on the reliability and validity of the ASKp in children with cerebral palsy will be presented as a poster at the Conference and Scientific Event Annual Meeting "Health 4.0: Designing Tomorrow's Healthcare," which should have taken place on 19-21 March 2020 in Coimbra (PT) (Appendix III) but, due to the COVID-19 pandemic, it has been postponed to 25-27 June 2020.

As for additional scientific activities, I collaborated on a research project on occupational therapy in complex patients, as well writing the protocol of a clinical trial that has been accepted for publication in January 2020 (Appendix IV).

Part II: Dissemination activities

Published manuscripts

Fugazzaro S, Costi S, Mainini C, Kopliku B, Rapicetta C, Piro R, Bardelli R, Rebelo PFS, Galeone C, Sgarbi G, Lococo F, Paci M, Ricchetti T, Cavuto S, Merlo DF, Tenconi S. PUREAIR protocol: randomized controlled trial of intensive pulmonary rehabilitation versus standard care in patients undergoing surgical resection for lung cancer. BMC Cancer. 2017 Jul 31;17(1):508. doi: 10.1186/s12885-017-3479-y. (Appendix V)

Costi S, Pellegrini M, Cavuto S, Fugazzaro S. Occupational Therapy in Rehabilitation of Complex Patients: Protocol for a Superiority Randomized Controlled Trial. Accepted on Jan, 2020 by the Journal of Interprofessional Care. doi: 10.1080/13561820.2020.1711720. (Appendix IV)

Costi S, Bressi B, Cavuto S, Braglia L, Ferrari A, Pelosin E. Cross-cultural Validation of Activities Scale for Kids: The Performance of Healthy Italian Children. Accepted for publication by The Journal of Sports Medicine and Physical Fitness.

Costi S, Mecugni D, Alboresi S, Beccani L, Bressi B, Paltrinieri S, Ferrari A, and Pelosin E. Construct validity of the Activities Scale for Kids performance in children with cerebral palsy: brief report. Accepted for publication by Developmental Neurorehabilitation.

Submitted manuscripts

Costi S, Filippi MC, Braglia L, Beccani L, Corradi I, Bruzzi E, Signorelli C, Pelosin E. Reliability and Construct Validity of the Activity Scale for Kids in Italian Children with Cerebral Palsy. Under review by Physical & Occupational Therapy in Pediatrics.

Further publications

Messina R, Dallolio L, Fugazzaro S, Rucci P, Iommi M, Bardelli R, Costi S, Denti M, Accogli MA, Cavalli E, Pagliacci D, Fantinia MP, Taricco M, on behalf of LAY Project. The Look After Yourself (LAY) intervention to improve self-management in stroke survivors: results from a quasi-experimental study. Accepted by Patient Education and Counseling in Jan, 2020. doi: 10.1016/j.pec.2020.01.004

Costi S, Crisafulli E, Trianni L, Beghè B, Faverzani S, Scopelliti G, Chetta A, Clini E. Baseline Exercise Tolerance and Perceived Dyspnea to Identify the Ideal Candidate to Pulmonary Rehabilitation: A Risk Chart in COPD Patients. Int J Chron Obstruct Pulmon Dis. 2019 Dec 27;14:3017-3023. doi: 10.2147/COPD.S223038. eCollection 2019. PubMed PMID: 31920298.

Marchioni A, Tonelli R, Sdanganelli A, Gozzi F, Musarò L, Fantini R, Tabbì L, Andreani A, Cappiello G, Costi S, Castaniere I, Clini E. Prevalence and development of chronic critical illness in acute patients admitted to a respiratory intensive care setting. Pulmonology. 2019 Oct 28. pii: S2531-0437(19)30176-X. doi: 10.1016/j.pulmoe.2019.09.006. [Epub ahead of print] PubMed PMID: 31672594.

Errante A, Bozzetti F, Sghedoni S, Bressi B, Costi S, Crisi G, Ferrari A, Fogassi L. Explicit Motor Imagery for Grasping Actions in Children With Spastic Unilateral Cerebral Palsy. Front Neurol. 2019 Aug 7;10:837. doi: 10.3389/fneur.2019.00837. eCollection 2019. PubMed PMID: 31447762.

Paltrinieri S, Vicentini M, Mazzini E, Ricchi E, Fugazzaro S, Mancuso P, Giorgi Rossi P, Costi S. Factors influencing return to work of cancer survivors: a population-based study in Italy. Support Care Cancer. 2020 Feb;28(2):701-712. doi: 10.1007/s00520-019-04868-0. Epub 2019 May 25. PubMed PMID: 31129762.

Errante A, Di Cesare G, Pinardi C, Fasano F, Sghedoni S, Costi S, Ferrari A, Fogassi L. Mirror Neuron System Activation in Children With Unilateral Cerebral Palsy During Observation of Actions Performed by a Pathological Model. Neurorehabil Neural Repair. 2019 Jun;33(6):419-431. doi: 10.1177/1545968319847964. Epub 2019 May 10. PubMed PMID: 31072215.

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Alboresi S, Sghedoni A, Borelli G, Costi S, Beccani L, Neviani R, Ferrari A. Are perceptual disorder signs in diplegic cerebral palsied children stable over time? A retrospective cohort analysis. Minerva Pediatr. 2019 Apr 16. doi: 10.23736/S0026-4946.18.05237-4. [Epub ahead of print] PubMed PMID: 30994318.

Pellegrini M, Formisano D, Bucciarelli V, Schiavi M, Fugazzaro S, Costi S. Occupational Therapy in Complex Patients: A Pilot Randomized Controlled Trial. Occup Ther Int. 2018 Sep 3;2018:3081094. doi: 10.1155/2018/3081094. eCollection 2018. PubMed PMID: 30250407.

Paltrinieri S, Fugazzaro S, Bertozzi L, Bassi MC, Pellegrini M, Vicentini M, Mazzini E, Costi S. Return to work in European Cancer survivors: a systematic review. Support Care Cancer. 2018 Sep;26(9):2983-2994. doi: 10.1007/s00520-018-4270-6. Epub 2018 May 29. PubMed PMID: 29845421.

Schiavi M, Costi S, Pellegrini M, Formisano D, Borghi S, Fugazzaro S. Occupational therapy for complex inpatients with stroke: identification of occupational needs in post-acute rehabilitation setting. Disabil Rehabil. 2018 May;40(9):1026-1032. doi:

10.1080/09638288.2017.1283449. Epub 2017 Feb 7. PubMed PMID: 28264614.

Conference Proceedings

Related to the topic of this Ph.D. research study:

Costi S, Filippi MC, Beccani L, Cavuto S, Braglia L, Pelosin E. Activities Scale for Kids performance: stima dell'affidabilità nella popolazione di bambini italiani con Paralisi Cerebrale Infantile. Poster presentation at XXXI Corso di aggiornamento "Asfissia perinatale ed encefalopatia ipossico-ischemica: prevenzione, diagnosi, terapia e riabilitazione", Modena, 20-22 marzo 2019. (Appendix II)

Costi S, Alboresi S, Mecugni D, Ferrari A, Boggiani E, Pelosin E. Assessment of Physical Functioning in Children with Cerebral Palsy: data of Convergent Validity between the Activities Scale for Kids and the Gross Motor Function Measure. Poster presentation at the Conference and Scientific Event Annual Meeting "Health 4.0: Designing Tomorrow's Healthcare," Coimbra, 19-21 Marzo 2020. (Appendix III)

Not related to the topic of this Ph.D. research study:

Paltrinieri S, Vicentini M, Mazzini E, Ricchi E, Fugazzaro S, Mancuso P, Giorgi Rossi P, Costi S. Indagine sui fattori lavorativi che condizionano il rientro al lavoro dei pazienti oncologici: studio epidemiologico sulla provincia di Reggio Emilia. Oral communication at the 43° Convegno AIE tenutosi a Catania nel 23-25 ottobre 2019.

Costi S, Paltrinieri S, Fugazzaro S, Bertozzi L, Bassi C, Pellegrini M, Vicentini M, Mazzini E. Employment rate and predictors of return to work in European Cancer survivors:

a systematic review. Poster presentation at the 26th International Conference on Health promoting Hospitals and Health Services, Bologna, 6-8 Giugno 2018

Paltrinieri S, Rondini E, Fugazzaro S, Gozzi C, Costi S, Mazzini E. Return to work in Italian cancer survivors: the innovative social-health care network. Oral communication at the 26th International Conference on Health promoting Hospitals and Health Services, Bologna, 6-8 Giugno 2018

Paltrinieri S, Fugazzaro S, Vicentini M, Mancuso P, Giorgi Rossi P, Mangone L, Ricchi E, Mazzini E, Costi S. I fattori che influiscono sul rientro al lavoro dei pazienti oncologici: studio epidemiologico condotto a Reggio Emilia. Poster presentation at the XLII Convegno AIE, Lecce, 24-26 Ottobre 2018

Pellegrini M, Fugazzaro S, Costi S, Schiavi M. Home-based occupational therapy to reduce disability and enhance social reintegration in complex patients in Reggio Emilia. Oral communication at the 26th International Conference on Health promoting Hospitals and Health Services, Bologna, 6-8 Giugno 2018

Accogli MA, Denti M, Costi S, Fugazzaro S. Therapeutic patient’s education program in addition to physical activity to engage onco-heamatological patients in managing cancer-related fatigue. Oral communication at the 26th International Conference on Health promoting Hospitals and Health Services, Bologna, 6-8 Giugno 2018

Cantarelli L, Mainini c; Bardelli R, Sobral rebelo PF, Tenconi S, Rapicetta C, Piro R, Costi S, Galeone C, Ruggiero P, Tedeschi C, Fugazzaro S, Kopliku B. Resezione polmonare per tumore al polmone e riabilitazione intensiva: revisione sistematica e definizione del protocollo PUREAIR. Poster presentation at the 45° CONGRESSO NAZIONALE SIMFER, Genova, 22-25 ottobre 2017.

Kopliku B, Mainini C, Bardelli R, Sobral Rebelo PF, Cantarelli L, Tenconi S, Rapicetta C, Piro R, Costi S, Galeone C, Ruggiero P, Tedeschi C, Fugazzaro S. Riabilitazione polmonare perioperatoria per pazienti con tumore al polmone trattati chirurgicamente: dati preliminari dell'RCT PUREAIR. Poster presentation at the 45° CONGRESSO NAZIONALE SIMFER, Genova, 22-25 ottobre 2017.

Mainini C. Bardelli R, Kopliku B, Sobral Rebelo PF, Cantarelli L, Tenconi S, Rapicetta C, Piro R, Costi S, Fugazzaro S. Preliminary data of an early pulmonary rehabilitation programme in surgically treated lung cancer patients. Poster presentation at the 11th International Society of Physical and Rehabilitation Medicine (ISPRM) World Congress, Buenos Aires, 30 Aprile – 4 Maggio 2017

Ferrari F, Montermini C, Ghirotto L, Mainini C, Fugazzaro S, Costi S. Partecipare ad uno studio riabilitativo di ricerca per pazienti con NSCLC: GT sul processo psico-sociale alla base delle motivazioni. Poster presentation at the Congresso Scientifico Internazionale AIFI 2017, Roma, 12-13 Ottobre 2017

Paltrinieri S, Fugazzaro S, Mazzini E, Pellegrini M, Bassi MC, Tedeschi C, Fugazzaro S, Costi S. Employment rate and predictors of return to work in people treated for oncologic diseases: a systematic review. Poster presentation at the 3rd World Congress on Cancer and Prevention Methods, Barcellona, 5-7 aprile 2017

FOREWORD

I consider obtaining my doctoral degree the most important achievement in my professional career as a physiotherapist, and I consider it the sine qua non for an academic career with all the necessary credentials. In my career as a physiotherapist I have cared for children with cerebral palsy; their ability to come up with solutions to adapt to the demands of everyday life has surprised and amazed me again and again. Moreover, I have realized that the objective assessments used in healthcare do not always completely describe these children's ability to carry out their activities in their own environment, often through adaptive strategies. For these reasons, I started to look for an evaluation tool that was able to illustrate the motor performance of a child during daily activities as perceived by the child him/herself, something the tools usually administered by healthcare professionals do not.

The research path I have followed in these last three years has allowed me to fully understand all the steps necessary for the cross-cultural validation of an evaluation tool. I have become aware of, and now fully appreciate, the complexity of the validation process of those assessment tools we clinicians use every day. The results of this Ph.D. research project provide physiotherapists and other allied healthcare professionals with an evaluation tool for use in the Italian pediatric population that complements those already in use, resulting in a more complete evaluation of the child.

Future studies will complete the validation process of this tool by investigating all its psychometric properties.

My deepest thanks to my mentor, Prof. Elisa Pelosin, for helping and guiding me during the realization of this Ph.D. project and for her immense trust in me.

I am grateful to Prof. Adriano Ferrari for the example he sets in always seeking to gain a deeper understanding of the phenomena that surround children with cerebral palsy. This example has been crucial to my professional growth.

Special gratitude goes to my colleagues Mariacristina Filippi, Barbara Bressi, Sara Paltrinieri, Laura Beccani, Silvio Cavuto, and Luca Braglia for their constructive feedback and for their support during all stages of my Ph.D. program.

Immense appreciation to Jacqueline M. Costa, who went far beyond helping me in the editing of this doctoral research thesis.

Finally, a heartfelt thanks to Sandra, Sonia, Vittoria, Ilenia, and Rita, my colleagues at the Bachelor of Science in Physiotherapy degree program, whose reliability and competence in taking on much of the course workload have allowed me to undertake this journey.

INTRODUCTION

Activities limitations in cerebral palsy

Cerebral palsy (CP) affects about 1 in 500 new live births worldwide, with an estimated prevalence of 17 million people, based on recent studies [Oskoui, 2013; Graham, 2016]. It is the most common disease that leads to physical disability and activity limitation in children living in industrialized countries [Reddihough & Collins, 2003].

CP describes a group of permanent disorders of the development of movement and posture that are attributed to a non-progressive disturbance in the fetal or infant brain; the motor disorders are often accompanied by epilepsy and disturbances of sensation, perception, cognition, communication, and behavior [Rosenbaum, 2006]. Motor impairment is the hallmark of CP, in most cases manifesting as unilateral or bilateral spastic CP [Shevell, 2009]. As children with CP grow, the effects of the brain lesion and spasticity extend to most parts of the musculoskeletal system and lead to secondary impairments, including abnormalities of the foot, patella alta, hip displacement, and pelvic obliquity [Gajdosik & Cicirello, 2002]. Children and adolescents with CP, particularly those with bilateral manifestations, are characterized by higher energy expenditure while walking, higher body fat percentage, and lower physical fitness than their healthy peers [Odding, 2006]. Moreover, sensory impairments affect about half of individuals with CP, more frequently those with unilateral manifestations [Odding, 2006]. This framework of impairments leads to restrictions in physical function, i.e., the ability to use the body to interact with the environment and to perform meaningful activities. As a result, children with CP frequently cannot perform age-related chores in a way that is considered “normal” for their healthy peers [Kerr, 2007]. Thus, although the relationship between impairment and disability in CP is not linear, the restrictions in physical function lead to a condition of physical disability that affects the performance of activities of daily living (ADL) and participation in this population [Kerr, 2007],

The *activities and participation* domain of the International Classification of Functioning, Disability and Health (ICF) [WHO, 2001] defines the ADL as “the execution of specific life tasks by individuals”. These life tasks can be classified as personal ADL, which are oriented towards self-care (e.g., grooming, bathing), or instrumental ADL, which are oriented towards personal independence (e.g., preparing meals, doing schoolwork). During development, tasks such as playing and performing age-related chores are essential to supporting participation in life situations; their accomplishment contributes to the child’s health, functioning, and creation of a personal biography [WHO, 2007]. Participation in life activities is reduced in children with CP [Majnemer, 2008; Orlin, 2010] in proportion to their level of physical disability and is mediated by personal and environmental factors [Morris, 2006]. Not surprisingly, participation in ADL is associated with crucial outcomes, such as physical and psychosocial well-being [Shikako-Thomas, 2012]. Given the value attributed to promoting activities and participation for successfully living with CP, ADL execution should always be accurately assessed and appropriately addressed in the rehabilitation of children with CP.

The assessment of ADL execution in cerebral palsy

Rehabilitation healthcare professionals should assess relevant activities with robust outcome measures to enable intervention planning and to accurately appraise results.

Activities can be assessed in terms of performance, capacity, or capability:

performance describes what a person does in the typical circumstances of his/her daily life, capacity describes what a person can do in a standardized, controlled environment, and capability describes what a person can do in his/her daily environment [Holsbeeke, 2009]. Performance measures can differ among environmental setting [Tieman, 2004] and tend to be less favorable than capacity and capability measures as performance captures and illustrates children's everyday habitual functioning in a real-world setting [Holsbeeke, 2009], making performance measures probably the most relevant. [Young, 1996].

It is likely that ADL execution differs based on the nature of various developmental disabilities (e.g., autism, spina bifida) [Mâsse, 2013]. As CP is a complex condition characterized by motor impairment and associated disturbances that can progressively influence ADL execution [Bartlett, 2019], professionals should rely on outcome measures that are validated for children with CP.

However, the choice of the most appropriate measure depends not only on its content and psychometric properties but also on the purpose of the assessment, the availability of a version suitable to the context, and the child's and child's family's specific goals. To date, nine outcome measures are psychometrically sound and thus appropriate to assess ADL execution in school-aged children with CP [Sakzewski, 2007; James, 2014; Schiariti, 2014]. Of these tools, the ABILHAND-Kids [Arnould, 2004], the Children's Hand-use Experience Questionnaire (CHEQ) [Sköld, 2011], the Pediatric Evaluation of Disability Inventory (PEDI) [Haley, 1992], and the Vineland Adaptive Behavior Scales (VABS) [Sparrow, 2005; Sparrow, 1984] are measures of capability and are administered via proxy report [Arnould, 2004; Haley, 1992; Sparrow, 2005; Sparrow, 1984] or by self-administration [Sköld, 2011]. The PEDI seems to be the best measure of ADL capability, with restriction to elementary school-aged children. The Functional Independence Measure for Children

(WeeFIM) [UDS, 1993] measures the child's degree of independence and need for assistance in 18 functions across the three general domains of *self-care*, *mobility*, and *cognition*. This tool is administered through direct observation by a trained healthcare professional. The few tools that measure ADL performance in children with CP are the Assessment of Motor and Process Skills (AMPS) [Fisher & Jones, 2010], the Klein-Bell ADL scale [Klein & Bell, 1979], the Assessment of Life Habits (LIFE-H) [Lepage, 1998], and the School Function Assessment (SFA) [Coster, 1998]. Both the AMPS and the Klein-Bell ADL scale are administered through direct observation by a healthcare professional, with formal training required to administer the AMPS. Instead, the LIFE-H and the SFA are administered by proxy report; the former can also be self-administered.

The AMPS is suitable for all ages, and it permits linking the ability to perform ADL with the child's underlying motor and process difficulties. It is noteworthy that this tool has been validated on thousands of individuals with different diagnoses (including CP), although less than 10% of these individuals were under age 16 [Fisher & Jones, 2010]. While its validity has been confirmed in typically developing children [Poulson, 1996], further validation studies on the population with CP are strongly recommended [James, 2014]. The same conclusion can be drawn for the LIFE-H, which has only been validated on 48 children with CP [Lepage, 1998]. The Klein-Bell ADL scale investigates basic self-care activities, but its long administration time (60-180 minutes) significantly reduces its practicality in the clinical setting. The SFA contains a comprehensive set of activities that can be scored separately from the other domains tested, but its focus is on measuring the student's performance on school-related tasks typical of an elementary school curriculum, not on general ADL.

To summarize, there are currently several outcome measures that assess ADL execution in children with CP, but only a few of them assess performance, and those few have limited acceptability or would require further validation studies.

Notably, as both the United Nations and the World Health Organization endorse client-centered/family-centered care [Lansdown, 2011; WHO, 2007], professionals should use tools in clinical practice that include families and children in goal setting. This can be achieved by including children and/or families in the development phase of the assessment tool and by adopting measures that collect the client's opinion. In this regard, it is to be noted that the activities investigated by all the above-mentioned outcome measures were chosen either through expert consent and/or by reviewing the literature and adapting tools already in use, many of which were designed for adults. During the developmental phase of the CHEQ, families were interviewed to identify activities they deemed relevant to their child [Sköld, 2011]; their children's opinions, however, were not collected. Furthermore, as already pointed out, only few outcome measures allow for self-administration by children [Sköld, 2011; Lepage, 1998]. Thus, the clients' opinion did not contribute to the design of any of these ADL assessment tools and is rarely collected directly in their administration phase.

To conclude, activities play a pivotal role in supporting participation and health in children, and professionals must assess this domain of the ICF in their clinical practice, paying specific attention to the construct of activity performance. Several outcome measures are already available for this purpose, but none can be considered exhaustive [Sakzewski, 2007; James, 2014]. Further, none was created to capture the child's perception of his/her functioning in everyday life. So, although the PEDI is a sound tool to measure capability in ADL for elementary school-aged children, there is no outcome measure to date that can be considered a gold standard for measuring physical disability applied to the construct of activity performance in children and adolescents with CP.

The Activities Scale for Kids

In the field of pediatric rehabilitation, several rating scales have been developed, most of which have not been formally translated into Italian and validated in this cultural context. One of these rating scales is the Activities Scale for Kids (ASK), developed in Canada in the 1990s by N.L. Young [Young, 1995].

The ASK is a self-report measure of physical disability designed for children and teenagers from ages five to 15 years who have limitations in ADL activities due to a broad range of musculoskeletal disorders. This is a unique client- and family-centered measure, generated from qualitative research (interviews) with numerous children and their parents, supplemented by recommendations from expert clinicians and data from the literature. The phrases children used to describe their limitations in activities were collected during the interviews and used to formulate the items of this scale. Furthermore, the ASK relies on a child's self-reporting (proxy report if self-report is unfeasible) of his/her limitations in activities, which is of the utmost importance when trying to help children express their needs and facilitate their empowerment [Missiuna & Pollock, 2000].

The ASK can be used both to describe the child's status at a single point in time or to monitor changes that may take place over time. Thus, the ASK is suitable as an outcome measure for therapeutic interventions [www.activitiesscaleforkids.com].

There are two versions of the scale: ASK performance (ASKp) and ASK capability (ASKc). The performance version measures what the child "did do" during the previous week, while the capability version measures what the child could have done in the same timeframe [Young, 1996]. Clinicians can choose to administer either version alone or concomitantly, based on the purpose of the assessment.

Both versions contain an initial section with instructions, followed by 30 multiple-choice questions (items) testing different common activities the child may have conceivably done in the preceding week. The 30 activity items are grouped into seven subdomains, not independently validated: personal care (3 items) such as “I put toothpaste on my toothbrush, then brushed my teeth by myself,” dressing (4 items) such as “I put my shirt on by myself,” other skills (4 items) such as “I made a snack (or prepared breakfast or lunch) by myself,” locomotion (7 items) such as “I got around inside my home without anyone to help me,” play (2 items) such as “I played sports by myself or with a few friends,” standing skills (5 items) such as “I got through heavy doors by myself,” and transfers (5 items) such as “I got down onto the floor from standing, and got back up again by myself.” Some of these subdomains aim at measuring the capability or performance in basic and instrumental ADL, while others aim at capturing the motor skills that are functional to the performance of ADL and thus may be useful when exploring the nature of activity limitations. Each of the 30 activity items is answered on a 5-point ordinal scale and then aggregated into one overall summary score, which is calculated as the average of all the items completed and is expressed as a zero-to-100 score. The summary score is calculated on a minimum of 23 answers [Young, 2007]. Higher scores indicate lower levels of limitation in activities and functioning.

The ASK also includes two questions that investigate the aids used by children while performing the activities, and a third question asks about the degree of assistance required during self-administration of the scale. None of these three questions contributes to the final score.

To the best of my knowledge, the ASK is unique since it collects the child’s perspectives on his/her capability or performance in executing meaningful activities, without mediation by a third party [Young, 1995], and it requires no special training or equipment [Young, 1995; Young, 1996].

Thus, integrating the ASK within the global assessment of children with CP is of paramount importance when planning the rehabilitation process because it helps orient treatments towards children’s real and perceived needs.

For this reason, rehabilitation professionals frequently use the performance version of the ASK in clinical practice and research when caring for children with CP [Morris, 2005; Harvey, 2008; Capio, 2010; www.activitiesscaleforkids.com]. The ASKp was developed and validated on a population of children with mixed diagnoses who were currently experiencing activity limitations due to musculoskeletal disorders of various severity and nature. While amputations, fractures, muscular dystrophy, spina bifida, and other disorders were included in the validation samples, CP was not widely represented [Young, 1995; Young, 2000]. As CP differs substantially from primary peripheral diseases and from orthopedic disorders in terms of its nature and natural history, it is not possible to take for granted that its influence on activity performance is similar to that of distinct developmental disabilities [Mâsse, 2013]. We thus considered it appropriate to verify the psychometric properties of the ASKp in a large population of children with CP.

Psychometric properties of the ASK

In order to be valid and useful, outcome measures must provide clinicians and researchers with clinically meaningful and accurate results. The term *meaningful* means that a measure’s score must provide a vivid representation of a patient’s current health status, and the term *accurate* means that the measure must demonstrate some psychometric properties, such as reliability, validity, and responsiveness [Prinsen, 2018].

Researchers are unanimous in considering reliability and validity as the chief psychometric properties of a measure [de Souza, 2017].

Reliability is the degree to which an outcome measure produces consistent results on different administrations, when all relevant conditions remain constant [Prinsen, 2018]. In other words, reliability is the extent to which the scores obtained for a patient who has not changed remain the same despite repeated measurements, without any measurement errors. The following properties pertain to reliability:

a) internal consistency (or homogeneity), a prerequisite to rule out any major sources of measurement errors, shows whether all the items of a measure assess the same construct, assuming the scale to be unidimensional. This form of reliability is often associated with questionnaires composed of multiple items that are aggregated into a summary score, as the ASK is. More precisely, internal consistency is a measure of the extent to which individual items of the questionnaire are related to the overall summary score. High correlations show that the items are measuring the same construct. A widespread measure of internal consistency is Cronbach's alpha coefficient, which demonstrates the covariance level between the items of a measure and thus the interrelatedness among the items [Streiner, 2015; Prinsen, 2018]. Values of Cronbach's alpha coefficients close to 0.6 are satisfactory, and values higher than 0.7 are ideal [Streiner, 2003]. The internal consistency of the ASK was originally tested on 28 children and resulted excellent (Cronbach's alpha 0.99) [Young, 2007].

b) stability (or reproducibility), which measures how similar scores are when the measure is administered at two different times (test-retest), assuming that the object of assessment has not changed over this period. The time span between the test and retest can influence the result: in questionnaires, a time span of 10-14 days is judged appropriate to measure stability [Keszei, 2010]. Stability over time is of chief importance when a measure is intended to make comparisons between conditions that can change over time, as the ASK does. In continuous variables (linear data), the interclass correlation coefficient (ICC) is the most used index of stability because it reflects both the degree of correlation and the agreement between repeated measurements. In other words, it is capable of excluding systematic differences between test and retest from the measurement error [de Vet, 2006, Brooks, 2002]. Minimum ICC values of 0.7 are considered satisfactory [Streiner, 2003]. The stability of the ASK was originally tested on 18 children and resulted excellent for both the performance (ICC 0.97) and the capability (ICC 0.98) versions [Young, 2007].

c) concordance degree is the degree of concordance between two or more observers administering the measure on the same patient and occasion (inter-rater), or the degree of concordance of the same observer (responder if self-administered) administering the measure on the same patient on different occasions (intra-rater) [Prinsen, 2018]. Concordance degree is of utmost importance when raters are part of the measurement process, for example, when the assessment requires hands-on or observational skills. Several parameters exist for calculating concordance, ranging from simple (e.g., percent agreement) to more complex (e.g., Cohen’s Kappa and ICC). Again, the ICC is widely used as an index of concordance, with values close to 1 meaning total concordance, values close to 0 meaning lack of concordance, and scores above 0.7 considered satisfactory [Streiner, 2003]. Inter-rater reliability of the ASK was demonstrated by the high degree of agreement between the parents’ scores and those of their children (ICC 0.96 for the ASKp and 0.98 for the ASKc) [Young, 2007].

For a scale to be useful, the user must be able to draw accurate conclusions about the presence or absence of the attribute being measured. This is the domain of validity, which is the degree to which an outcome measure measures exactly what it claims to measure [Prinsen, 2018]. That is, a questionnaire is valid when its items are appropriate, meaningful, and useful so that accurate inferences can be made with respect to the construct it is intended to measure. Unlike reliability, validity is not a characteristic of the tool and must be determined regarding a specific construct with reference to a defined population. Validation is not a single-step process; it occurs through the accumulation of evidence across different data sets and comparisons to verify some fundamental properties. The following properties pertain to validity:

a) content validity, which is the degree to which the tool's content adequately reflects the construct that is being measured [Streiner, 2015; Prinsen, 2018]. In other words, it pertains to the extent to which the items of a questionnaire represent an adequate and relevant sampling of a defined universe (content domain), for example, meaningful activities in children. There is no specific statistical test to quantify content validity. Face validity, a judgment of whether the items of a tool seem to be an adequate representation of the construct intended to be measured, is a simple qualitative method that is widely used to estimate content validity [Prinsen, 2018]. This judgment can be delivered by a committee of experts (professionals), but also by clients on whom the scale will be used. This qualitative judgment can be strengthened by measuring the degree of concordance between the members of the committee on the relevance and reflectiveness of the items with respect to the construct. The degree of concordance can be quantified by a simple concordance agreement with K coefficient, or by a more complex content validity index (CVI), whose threshold of acceptability was set by experts at 0.80 [Lawshe, 1975].

As previously stated, the ASK items were generated from the pertinent literature and from input from children, parents, and professionals and were subsequently selected based on the frequency and relevance of the activity. The concordance between the items generated by children and their parents was good (85% agreement, K = 0.70). The children's input was highly valued in the process of generating items; further, children contributed extremely relevant information on how to word the questions [Young, 1995].

b) criterion validity is the degree to which the target assessment tool's score corresponds to an external criterion [Prinsen, 2018]. This criterion must be a valid and widely accepted measure of the construct to be measured in the population of interest. That is, it must be considered an outcome measure or criterion that is the "gold standard." If the target tool measures what is intended to be measured, then its results must agree with those of the gold standard. Criterion validity is calculated by correlation coefficients: the higher the correlation, the higher the criterion validity. The criterion validity is a challenge for researchers because an outcome measure widely accepted as the gold standard cannot easily be found for each construct of health. On the other hand, where there is a gold standard, the issue is what the benefit is (e.g., ease of use, cost) of validating a new tool for the same construct.

As discussed in the previous paragraphs, there is no outcome measure that can be considered the gold standard for measuring physical disability in children with CP. To test the criterion validity of the ASK, a convenience sample of 24 children completed a random sample of 15 items extracted from the ASKc, then children were asked to demonstrate the same 15 activities in a clinical setting and in front of two clinicians, who independently rated the child's ability using the ASKc. Criterion validity was tested only for the ASKc because the presence of observers and the artificial environment were not considered coherent with the concept of performance. The correlation between the ASKc child-reported score and the clinician-reported score was high (Spearman's coefficient = 0.92, 95% CI 0.82-0.97). The inter-rater reliability between clinicians was extremely high (ICC = 0.99, 95% CI lower limit 0.98) [Young, 2000].

c) construct validity is the degree to which the score obtained from a tool concurs with the theoretical construct to be measured and is consistent with the theory and hypothesis underlying the construct [Prinsen, 2018]. Therefore, in order to establish the construct validity, it is essential that it be based on a theory and that some predictions must be made and verified. As an example, if a questionnaire is to measure physical disability in terms of limitations in activity performance, one should expect that its score will differ between healthy and disabled children. Recent approaches to psychometry affirm that there are not different 'types' of validity: rather, they are all various aspects of construct validity [Streiner, 2015]. Specifically, construct validity is verified by determining whether an outcome measure displays: a) strong correlations with similar measures (convergent validity); b) weak correlations with unrelated measures (divergent validity); c) ability to separate individuals into groups based on the level of the attribute measured (discriminant validity). Since construct validity is a multifactorial concept that cannot be verified with a single step, the statistical approaches vary, depending on what is being measured.

Previous studies ascertained the construct validity of the original version of the ASK (both performance and capability versions) in terms of convergent, divergent, and discriminant validity on a population of 200 children with a mean age of 10.1 years (SD 3.1, range 5-15 years) [Young, 1997; Young, 2000; Young, 2007]. Convergent validity was tested by comparing the ASK with the Childhood Health Assessment Questionnaire (CHAQ), a valid questionnaire that measures physical disability in children with arthritic conditions [Feldman, 1995]. Thus, while the CHAQ could not be considered a criterion for the general population with musculoskeletal disorders, the comparison between the two measures was evidence of construct convergent validity for the ASK. Spearman’s correlation between the ASK and the CHAQ was 0.82 (95% CI 0.75-0.87) for the ASKp and 0.85 (95% CI 0.79-0.89) for the ASKc [Young, 2007]. Further evidence of good to excellent convergent validity was collected when looking at the correlation between the ASKp score and the following: a) the parent-report format of the Pediatric Outcomes Data Collection Instrument (PODCI) ($r \geq 0.78$) [Pencharz, 2001], which is a valid measure of functional status in pediatric populations undergoing orthopedic surgery for a broad range of diagnoses; b) the summary score and the scores of the physical independence and mobility domains of the Lifestyle Assessment Questionnaire in Cerebral Palsy (LAQ-CP) (all $r \geq -0.90$) [Morris, 2006], which measures the impact of disability on the family life of children with CP; c) a subset of items of the Gross Motor Function Measure-66 (GMFM-66-IS) ($r = 0.83$) [Bjorson, 2013], which is a measure of motor function in children with CP; d) the Gross Motor Function Classification System (GMFCS) (r =

-0.90) [Morris, 2006], which classifies children with CP based on clinically meaningful differences in their motor function. Divergent validity was tested comparing the ASK with the subdomains *emotion* and *speech* of the Health Utilities Index Mark 3 (HUI_3) [Boyle, 1995]. The hypothesis was that the correlation would be weak as those constructs are dissimilar from physical disability. Spearman's correlation between these divergent constructs were 0.15 (*emotions*) and 0.09 (*speech*) for the ASKp and -0.12 and 0.08 for the ASKc [Young, 2007]. Finally, discriminant validity was ascertained by comparing the ASK summary score with the clinician's global rating of disability (mild, moderate, severe), which was available for 28 children. Statistically significant differences (one-way ANOVA $p < 0.0001$) in the average ASK scores (both performance and capability) were found between the three disability ratings. In other words, children classified as severely disabled by their clinician typically had lower scores [Young, 1997]. Also, the ASKp has shown that it is able to distinguish between healthy children and children with mild musculoskeletal disabilities of various etiology [Plint, 2003].

Altogether, these results argue in favor of the construct validity of the original version of the ASK [Young, 2007].

In the clinical setting, outcome measures are frequently applied to detect any meaningful changes over time, which may be due to the natural history of a condition or may be subsequent to treatment. This property is called sensitivity to change, or responsiveness: the ability of an outcome measure to quantify clinically important changes in the construct to be measured and to distinguish it from fluctuation in the results due to any error of measurement [Prinsen, 2018]. Responsiveness is determined by measuring the relationship between changes in the score of the target outcome measure over time, providing that a clinically meaningful change has in fact occurred.

Responsiveness is usually represented as a coefficient or a series of coefficients, or by means of formal statistical tests [Brooks, 2002]. The responsiveness of the ASK was tested in a study conducted on a subsample of 22 children, chosen from a larger sample of 200, whose condition was expected to change in the coming months. At the beginning of the study and again after 6 months, the CHAQ and both versions of the ASK were administered to the children in the study, and their parents were asked whether the expected change had occurred. Responsiveness was assessed by calculating the standardized response mean (SRM), which is obtained by dividing the mean change by the standard deviations of the score change [Liang, 1985]. The SRM of both the ASKp (1.1) and the ASKc (0.94) were compared to those observed for the CHAQ (0.96); the relative responsiveness was satisfactory in both cases (1.16 for ASKp, 0.98 for ASKc) [Young, 2007]. This means that the sensitivity to change of the ASK is equal to, or even greater than, the reference chosen (CHAQ).

Finally, it is worth noting that the ASK seems to have adequate acceptability; 99% of the questionnaires administered during validation studies were fully completed [Pencharz, 2001]. Also, little time is required to complete it (10 to 30 minutes) [Young, 2007], and the performance version resulted suitable for administration both in its web-based format and mailed paper format [Morris, 2005; Young, 2009]. Therefore, the ASKp is highly feasible for self-assessment and is cost-effective as well.

To conclude this section on the properties of the original version of the ASKp, it should be noted that the psychometric properties of any outcome measure, even when presented separately, are not independent. It is widely accepted that reliability is necessary but not enough to guarantee validity [Guyatt, 1987], and that responsiveness is strictly connected to validity, so much so that some authors sustain it is a component of validity [Hays & Hadorn, 1992]. Thus, an outcome measure can be reliable although not valid and not responsive with respect to a specific construct. This is possible because consistent (reliable) measurements do not necessarily imply that a specific, desired construct is being measured, nor that the change in this construct can be detected. On the other hand, a measure that yields inconsistent (unreliable) results cannot yield valid results [Brooks, 2002]. Also, reliability and validity are not fixed properties as they can vary based on the circumstances and population under study [de Souza, 2017]. For this reason, the psychometric properties of a measure should be verified when the circumstances change. This is the case even when an outcome measure is used in a cultural context different from that of the original validation. In fact, while assessment tools are usually developed and validated for specific contexts, their use is often extended to areas that differ greatly from the original cultural, linguistic, and ethnic backgrounds. In addition to the formal translation of the tool, these differences imply the need for a linguistic, cultural, and semantic adaptation to the context in which it will be applied, without changing its original measurement purpose [Prinsen, 2018]. Furthermore, the robustness of its psychometric properties should be confirmed in the new target population [Prinsen, 2018; Sousa & Rojjanasrirat, 2011].

The Italian cross-cultural validation process of the ASKp

According to data collected in 2011 on Medline by the Italian Society of Physiotherapy, only few (48 out of 237) rehabilitation outcome measures used both for children and adults have been formally validated in Italian [www.sif-fisioterapia.it]. Although this data collection is not up-to-date and not even exhaustive – only one database was consulted – it is clear that there is a great need to proceed with local cross-cultural validation of outcome measures that are already successfully used at the international level. As stated in the previous paragraphs, the ASKp has characteristics that distinguish it from, and in some respects make it preferable to, other outcome measures in the field of pediatric rehabilitation. In the Children Rehabilitation Unit for Severe Developmental Disabilities of Reggio Emilia (Italy), about 2,800 consultations take place every year, of which 2,000 on children with a disability due to CP. In this context, the need to assess children's abilities to carry out their daily life activities emerged; after examining some available tools, the clinicians' interest focused on the ASK. Thus, in 2014, we decided to undertake the cross-cultural validation process of the ASK performance version. The decision to proceed with the performance version rather than the capability version was based on the opinion of several rehabilitation professionals in this field, who felt the ASKp better satisfied their information needs regarding children with CP [Fabbri, 2016].

We immediately asked Dr. N.L. Young, the original developer of the copyrighted ASK, for permission to use the ASK for research purposes, which Dr. Young granted. The first step of the cross-cultural validation was conducted by following the guidelines of Sousa and Rojjanasrirat [Sousa & Rojjanasrirat, 2011]. Very briefly, the ASKp had been translated and blindly back-translated, the opportune comparisons were made as recommended, and the pre-final version was pilot tested for clarity and relevance in a sample of children with musculoskeletal disorders (n.12), their parents (n.12), and experienced physiotherapists (n.11). Based on the results of this study, the instructions, the three questions investigating the use of assistive devices and the degree of assistance required for activities, and the answer options reached an inter-assessor agreement ≥ 80% by children, parents, and physiotherapists participating in the study. The 30 multiple-choice items of the ASKp were judged clearly understandable by all the children and the physiotherapists, with an overall inter-assessor agreement ≥ 80%. Parents judged 29 items clearly understandable (inter-assessor agreement ≥ 80%); one item (n.13) reached an inter-assessor agreement of 75% because one word was judged ambiguous and was therefore substituted with a suggested synonym.

Hence, the Italian version of ASKp was judged by all the interviewed participants to be understandable and clear overall in terms of meaning and word choice.

The evaluation of relevance, or face validity, was judged by the experienced physiotherapists and showed that the minimum value of the Item-CVI scored in four items was equal to 0.63, slightly higher than the critical value of 0.56 determined by Lawshe on the basis of the number of judges involved [Lawshe, 1975]. CVI-Universal Agreement, i.e., the proportion of items deemed relevant by all the judges with respect to the total number of items rated, was equal to 0.76 [Lawshe, 1975; Polit & Beck, 2006; Chiorri, 2010]. The average value of CVI at the scale level was 0.93, which is higher than the cutoff indicated by experts [Lawshe, 1975; Sousa & Rojjanasrirat, 2011].

Finally, the panel of physiotherapist-researchers analyzed the suggestions made by the participants; through the process of formal consent, the panel agreed on a culturally and linguistically adapted Italian version of ASKp [Fabbri, 2016], which is now being tested for further psychometric properties.

Purpose and general organization of the research project

The main goal of this Ph.D. research project is to carry forward the validation process of the Italian version of the ASKp in children with CP. The culturally adapted Italian ASKp was achieved in 2016 by a group of researchers led by the Ph.D. candidate Stefania Costi [Fabbri, 2016] (Appendix I), following a recommended translation/back-translation process [Sousa & Rojjanasrirat, 2011]. The same study also confirmed the face and content validity of this assessment tool in children with CP. This research project continued the validation process of the culturally adapted Italian ASKp by verifying its reliability (internal consistency) and construct validity in the target population. With regard to the latter, as the ASKp aims to measure the theoretical construct of physical disability, three hypotheses that are consistent with this construct were set a priori. If the ASKp is to measure physical disability, a) it should be able to discriminate healthy children from children with CP; b) it should be able to discriminate between children with CP with different levels of disability; c) its results should be convergent with those obtained with valid measures of motor function in CP, as motor function is a construct strictly linked to physical disability.

Three studies were conducted during the 3-year Ph.D. research course (2016-2019). The results are reported in the chapters of the present dissertation.

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CHAPTER I

The ASKp in healthy Italian children: first evidence of construct validity.

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Cross-cultural Validation of Activities Scale for Kids: The Performance of Healthy Italian Children

by

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ABSTRACT

Background: The assessment of physical performance is a key element in the rehabilitation of individuals with disabilities, and understanding patients' perception of their abilities is of foremost importance for the success of the whole process. The Activities Scale for Kids (ASK) is one of the few reliable and responsive outcome measures that allows children and adolescents to accurately report their physical functioning in typical activities for this age. The performance version of the ASK has recently been translated and culturally adapted to the Italian context.

Methods: This cross-sectional study has been implemented to describe the ASKp score distribution in a population of Italian school aged individuals.

Results: A population of 209 Italian children with a mean age of 10.96 years (SD 2.92, range 5-15) participated in this study during the academic year 2017/18. The ASKp score ranged from 52.58 to 100, with a mean value of 89.07 points (SD 10.57). Scores greater than 90 were attained for 57.4% of participants, confirming the ceiling effect of this assessment tool when administered in healthy children (95% CI: 50.4% - 64.2%). The ASKp was able to differentiate between different levels of physical performance according to different developmental ages (p<0.001), and it confirmed the ability to distinguish between children with typical development and those with disability (p<0.001).

Conclusions: The results of this study provide the first evidence of construct validity of the Italian version of the ASKp. These results will help clinicians to interpret ASKp scores of children with musculoskeletal limitations.

Trial registration: ClinicalTrials.gov Identifier: NCT03325842

Key words: Reference Values, Outcome Assessment, Child, Cross-cultural Comparison, Activities Scale for Kids, Activity of Daily Living

Introduction

Child and adolescent health are associated with stage of development, and limitations in abilities in this age differ from those in adults in terms of their nature, intensity and impact [1].

Therefore, specific tools are needed to fully capture the impact of limitations in the lives of younger individuals, and several outcome measures have been developed with this purpose. In the developmental age, activities like walking, playing or performing age-appropriate chores influence physical, social and psychological development and are strictly connected to participation [1].

One of the most widely used measures of physical functioning in usual abilities in pediatrics is the performance version of the Activities Scale for Kids (ASKp) [S1], a self-report questionnaire for children and adolescents aged 5 – 15 years with musculoskeletal disorders, which requires no special training or equipment [2,3]. It covers seven out of the nine International Classification of Functioning domains of activities and participation, and focuses on performance, measuring what the child usually does [4,5].

A clear advantage of using ASKp in clinical practice is that it captures the child's perspectives on the impact that disabilities or limitations have on daily life, without mediation by a third party [2,6]. The integration of this perspective is crucial when planning rehabilitation interventions because it permits focusing on the child's needs, monitoring changes over time. Thus, rehabilitation professionals frequently use this scale in research and in clinical practice [6,7].

It is well known that outcome measures must possess some basic psychometric requirements and that they should be validated in the cultural context of use. The ASKp has been tested for its validity through appropriate studies [2,3,8,9], and has shown excellent reliability [6-8] and good content, concurrent, construct and discriminative validity [6-8,10] in children and adolescents with activity limitations due to musculoskeletal disorders.

A cross-cultural validation study has been recently performed to obtain the Italian version of the ASKp [11]. As part of the cultural adaptation process, it is useful to know how the ASKp performs, on average, in a population of healthy individuals living in the context of application, and an analysis of healthy children had been completed in Canada, where the ASK was originally developed [9]. This facilitates the interpretation of the ASKp score obtained in individuals with disabilities living in the same context.

To date, no data have been published on ASKp in healthy Italian children.

The aim of this descriptive study is to determine the ASKp score in a population of Italian children without musculoskeletal disorders. The results of this study will help clinicians in interpret ASKp scores of children with musculoskeletal limitations.

Materials and Methods

Participants and setting

This study involved Italian individuals aged 5 to 15 with no skeletal limitations attending schools in Reggio Emilia, a city of 150,000 inhabitants in northern-central of Italy.

Study design

This independent cross-sectional study provides for a single self-administration of the ASKp.

It was conducted in accordance with the principles of the Helsinki Declaration and was approved by the Ethics Committee of the Province of Reggio Emilia (Protocol n.

2017/0004096, chairperson: Dott. Satolli, approved on the 15/02/2017). Children and their parents gave their informed consent for participation in this study.

The Authors state they do not have any conflict of interests or bias in this study.

During the academic year 2017/18 the principals of the 12 public elementary and middle schools of Reggio Emilia were contacted by e-mail. In the e-mail they received information regarding the purpose of the study and were asked for an appointment in order to define the most feasible study procedures. During the appointment, the school principal and the teachers were shown the Italian version of the ASKp and the written information regarding the study together with the consent to participate in it. Then, the most feasible ASKp administration procedure was defined.

The ASKp

The ASKp is a questionnaire asking children what they "did do" during the previous week in 30 usual activities across seven subdomains of physical functioning (standing skills, personal care, dressing, locomotion, transfers, play, other skills). Each of the 30 activity items is answered by the child using a 5-point ordinal scale, corresponding to a score from 0 to 4. For any single item, the lower the score, the higher the perceived physical functioning related to the activity tested. However, in order to keep the data entry process simple and limit the possibility of data entry errors, when data are to be entered into the Excel spreadsheet provided by the developers of the ASK©, as was the case in this study, the raw values to enter are 1 for the 1st response option, 2 for the 2nd, and so on. The Excel spreadsheet was designed to work with raw scores in this format and results in a single summary score from 0 to 100, with higher scores indicative of perception of full physical function.

Besides the 30 activity items that contribute to the final score, the ASKp also asks about assistive devices in use for indoor and outdoor mobility, and the degree of assistance required in completing the questionnaire. This questionnaire takes approximately 5 to 12 minutes to complete [10].

Data analysis

Based on the study conducted by Plint and colleagues [9], we considered that a minimum of 100 children would have to be recruited to allow for an accurate estimate of the ASKp score in healthy Italian children. This sample size was estimated by assuming that the Pediatric Outcomes Data Collection Instrument (PODCI) scale standard deviation measured in healthy children would be a good proxy of the ASK standard deviation in the same population, since the PODCI scale and the ASKp have the same range of possible score and correlate very well [10].

Descriptive statistics were used to represent the sample characteristics and the response rates. Means were compared by independent samples T tests (or by one-way ANOVA analyses where there were more than two groups), variances by F test; percentages were accompanied by 95% Wilson confidence interval. Data entry was performed using Microsoft Excel 2010; statistical analysis with Excel and R version 3.3.3 [S2].

Results

Two principals gave their consent to meet a researcher together with the school coordinator. In both cases, the school coordinator and some of the teachers collaborated in the study by explaining the study to the parents of the eligible children and giving them an envelope containing the written information regarding the study, the consent form for their child’s participation, the consent form to process the child’s data, and a copy of the Italian version of the ASKp. Overall, 233 (183+50) envelopes were distributed to the two schools (J.F. Kennedy and M.E. Lepido, both in Reggio Emilia). Two hundred and five parents gave consent for their child to participate in the study. In most cases (n.113), the ASKp was self-administered at home, with the assistance of a parent if needed, and then returned to the teacher. In other cases, the teacher dedicated time to allow for the self-administration of the ASKp at school (n.92). At the end of this administration procedure, 197 ASKp were completed and entered into the database. We then checked whether all ages included in the range from 5 to 15 were represented. As the extreme ages, namely 5, 6, and 15 years, were proportionally less represented then the others, in order to sample at least ten children for each age between 5 and 15, the researchers recruited a further convenience sample among children of their relatives or colleagues (n.18). In all these cases, the ASKp was self-administered at home.

Altogether, 215 ASKp were collected (figure 1).

Participants were a mean age of 10.96 years (SD 2.92) and were balanced for sex distribution (49% of females versus 51% of males). As expected, none of the participants used aids to move inside or outside the home.

The majority of participants (73%), especially among adolescents, completed the questionnaire on their own, while a certain amount of help was necessary for younger children (Table I).

There were 75 missing data, half of which were retrieved in the questionnaires completed by younger children (37 out of 75). Only six questionnaires were insufficiently completed to compute the final score (one 9-year-old, one 12-year-old, two 10-year-olds, and two 13-year-olds), confirming the acceptability of this assessment tool [10].

Thus, the ASKp summary score was computed on a total of 209 children, and ranged from 52.58 to 100, with a mean of 89.06 (SD 10.57), significantly higher ($p < 0.001$) than the mean value previously recorded in the population of children with musculoskeletal disorders (68.2 ± 24.12) [8] (see Table II). The scores for the various age classes were significantly different: from the mean value of 84.79 for the 5-10-year-olds to 95.01 for the 14-15-year-olds, ($p < 0.001$). As expected, the average ASKp score increased with age, showing a greater level of physical functioning in adolescents than in younger children. This is even more evident in figure 2, which represents the box plot of the ASKp summary score distribution: children from 5 to 10 years perceived their performance as poorer than that of adolescents.

This demonstrates that in healthy children, the ASKp can distinguish between different levels of physical performance, at different developmental stages.

However, as shown in figure 3, ASKp summary scores greater than 90 were attained for most participants (n. 120, 57.4%, 95% CI: 50.4% - 64.2%), confirming the ceiling effect of this assessment tool when administered to healthy children, as already suggested by Plint and colleagues [9].

Looking at each single item, the median value attributed by participant was 1 for each, representing a perception of full physical functionality in all the activities tested (see Supplementary Digital Table 1 for comprehensive data analysis). Thus, the average value attributed by participants to all 30 items was 1.59 (± 0.44) points. The items with the highest mean score, representing the most critical activities in the sample investigated, were “I took care of my medical needs” (3.74), “I made a snack by myself” (2.58), and “I did my usual job or chores” (2.45).

Discussion

This descriptive study shows that the Italian version of the ASKp, which is in the process of being validated, is likely to differentiate between the level of the physical performance of healthy children and that of children with musculoskeletal disorders.

Comparing our sample with that of healthy Canadian children previously collected by Plint and colleagues [9], it is worth noting that the average age of the two samples is almost identical, but the mean ASKp score in the Italian children is slightly lower (89.07 versus 93.12, $p<0.001$). Even more evident is the difference between the variability of the score distribution in the two healthy populations, which is higher in the Italian sample (SD 10.57 versus 6.45, $p<0.001$). Therefore, it is possible that the perception of physical performance in activities typical of this age is relatively variable in the population of healthy Italian children, and also that lower performance in younger children may be possible. Therefore, when the ASKp is used in the Italian context, clinicians should bear in mind that a low score in a child or young adolescent (≤13 years) does not necessarily reflect a deviation from the norm for his/her age class.

This is precisely the purpose of this study: to see how the ASKp works in the healthy population in order to better interpret the scores collected when this questionnaire is administered to individuals with musculoskeletal disorders. This fundamental step should never be overlooked in the validation process of any assessment tool.

In this perspective, our study confirms that the ASKp can differentiate between children at different ages and thus between different levels of physical performance in activities. Moreover, the Italian version of the ASKp seems to distinguish between healthy children and children with musculoskeletal disability [8], even though this assertion can only be verified once the ASKp has been administered to a representative sample of Italian children with disability, whose scores might differ from those of Canadians. This is even truer when dealing with individuals with milder degrees of disability, whose score may overlap with that of younger healthy children [8]. Future studies on Italian children with musculoskeletal disability will be of help in clarifying this issue.

Finally, as already highlighted in the results, three items, namely “I took care of my medical needs,” “I made a snack by myself,” and “I did my usual job or chores,” scored considerably above the average score of the whole set of items of the ASKp (Supplementary Digital Table 1). These three items could represent activities that the child, especially if very young, does not perform independently in the Italian context.

Most of the data presented in this study were collected from children attending schools in two neighborhoods of one mid-sized northern Italian city with a high socioeconomic level. We cannot therefore rule out that young healthy Italians living in different contexts (e.g., a rural or a poorer context) may show different levels of functionality in the same activities. However, we believe it is unlikely that this type of recruitment strongly biased the results of this study.

A strong point highlighted by our results is the high proportion of complete self-administration of the ASKp. This is very important because the ASKp was developed based on the words used by children to describe their disability, with the precise aim of capturing the children’s perspective regarding their physical limitation and of reflecting changes that are clinically relevant for their everyday life [2].

Conclusions

This study shows that the self-administration of the ASKp is feasible in healthy Italian children. Further, the expected ceiling effect in this population, close to that already seen in a different context, is confirmed. However, the Italian version of the ASKp can differentiate between different levels of perceived physical performance per different age classes, providing the first evidence for its construct validity. Although future studies must add insight on its ability to reveal mild disability, especially among the youngest Italians, the ASKp seems to be a promising assessment tool which, together with other tools of the same construct, will help clinicians to evaluate the child’s level of functional performance and perhaps the effects of the rehabilitative therapies implemented as well.

NOTES

Conflicts of Interest

The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

Funding Statement

This work was not supported by funding.

Author's contribution

Stefania Costi gave substantial contributions to the conception and design of the work, as well as the acquisition, analysis, and interpretation of data; she also drafted the work and approved the final version to be published. She agrees to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Barbara Bressi gave substantial contributions to the acquisition, analysis, and interpretation of data; she also drafted the work and approved the final version to be published. She agrees to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Silvio Cavuto and Luca Braglia gave substantial contributions to the conception of the work, as well as the analysis and interpretation of data; they revised the draft of the manuscript critically for important intellectual content and approved the final version to be published. They agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Adriano Ferrari and Elisa Pelosin gave substantial contributions to the conception and design of the work, as well as the interpretation of data; they revised the draft of the manuscript critically for important intellectual content and approved the final version to be published. They agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Acknowledgments

We especially want to thank the children of the J.F. Kennedy Elementary School and the M.E. Lepido Middle School of Reggio Emilia and their parents, as well as the teachers and the principals of the two schools. Without their free commitment, this study would not have been possible.
We thank Jacqueline M. Costa for the English language editing.

Data Availability

The demographics and clinical data collected to support the findings of this study are restricted by the Ethics Committee of the Province of Reggio Emilia (Italy) in order to protect patient privacy. The data that support the findings of this study are available from the corresponding author (S.C.), upon reasonable request, with the permission of Azienda USL-IRCCS of Reggio Emilia, Italy.

TABLES

Table I. - Help needed in completing the questionnaire

	5-10		11-13		14-15		Total	
	N	%	N	%	N	%	N	%
Alone	58	52.25	30	90.90	69	97.18	157	73.02
Help reading the questions	25	22.52	1	3.03	0	0	26	12.09
Help with some answers	19	17.11	1	3.03	2	2.81	22	10.23
Help with most answers	5	4.50	0	0	0	0	5	2.32
Other	4	3.60	1	3.03	0	0	5	2.32

Table note: N=number

Table II. - ASKp score distribution for age class

	N	Mean	SD	Min	1st Q	Median	3rd Q	Max
5-10	108	84.79	10.96	52.58	78.12	86.78	93.33	100
11-13	30	90.67	10.60	54.31	87.12	94.58	97.61	100
14-15	71	95.01	5.95	75.00	93.96	96.66	99.16	100
Total	**209**	**89.06**	**10.57**	**52.58**	**82.50**	**93.33**	**97.41**	**100**

Table note: N=number; NA=not applicable; SD=standard deviation; Min=minimum; Q=quartile; Max=Maximum

Figure 1. Flowchart of the ASKp data collection

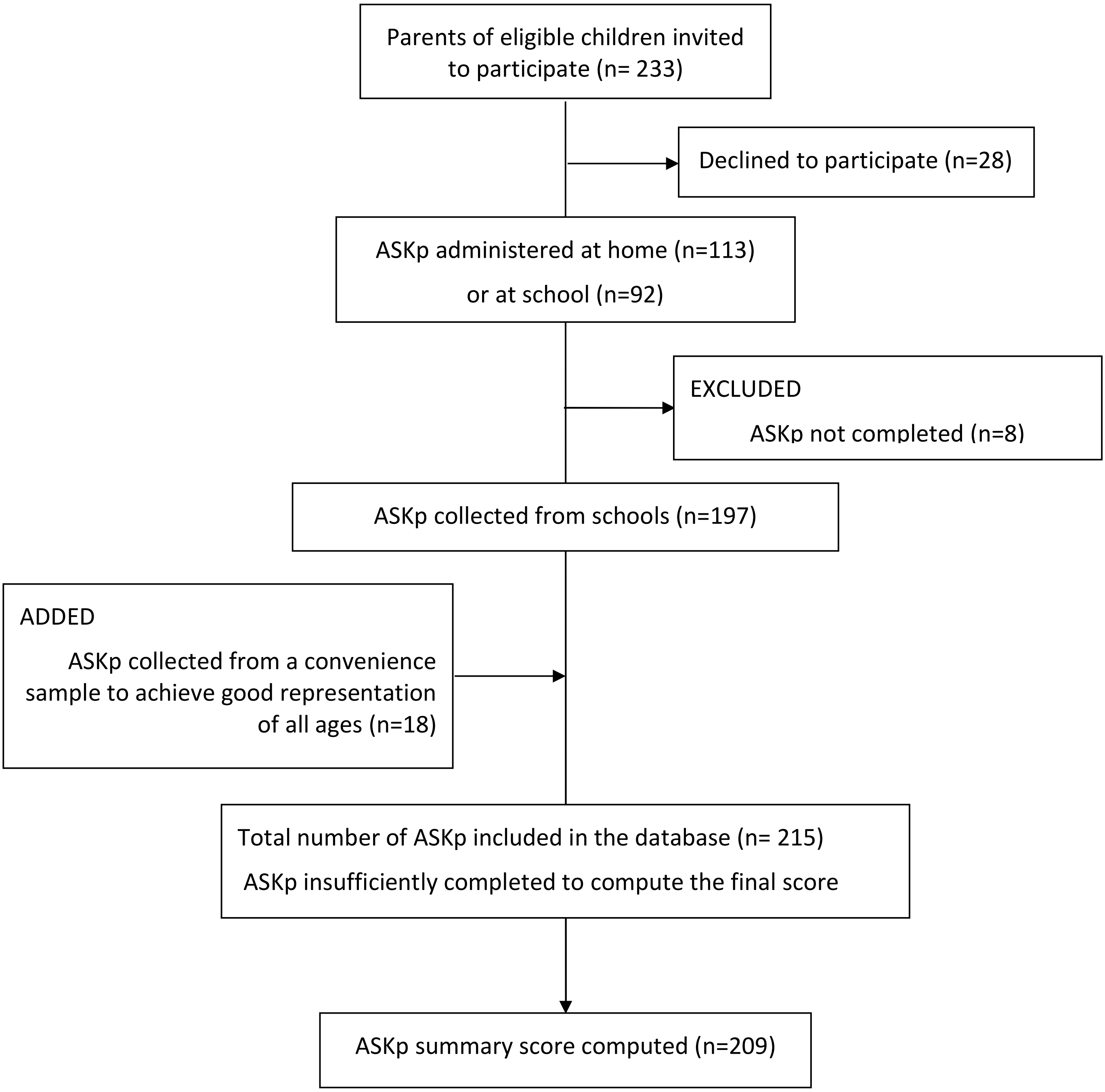


Figure 2. ASKp score distribution for age class

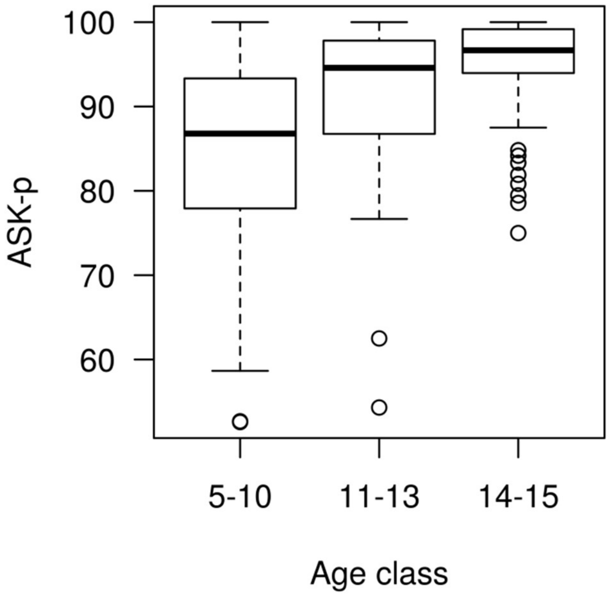
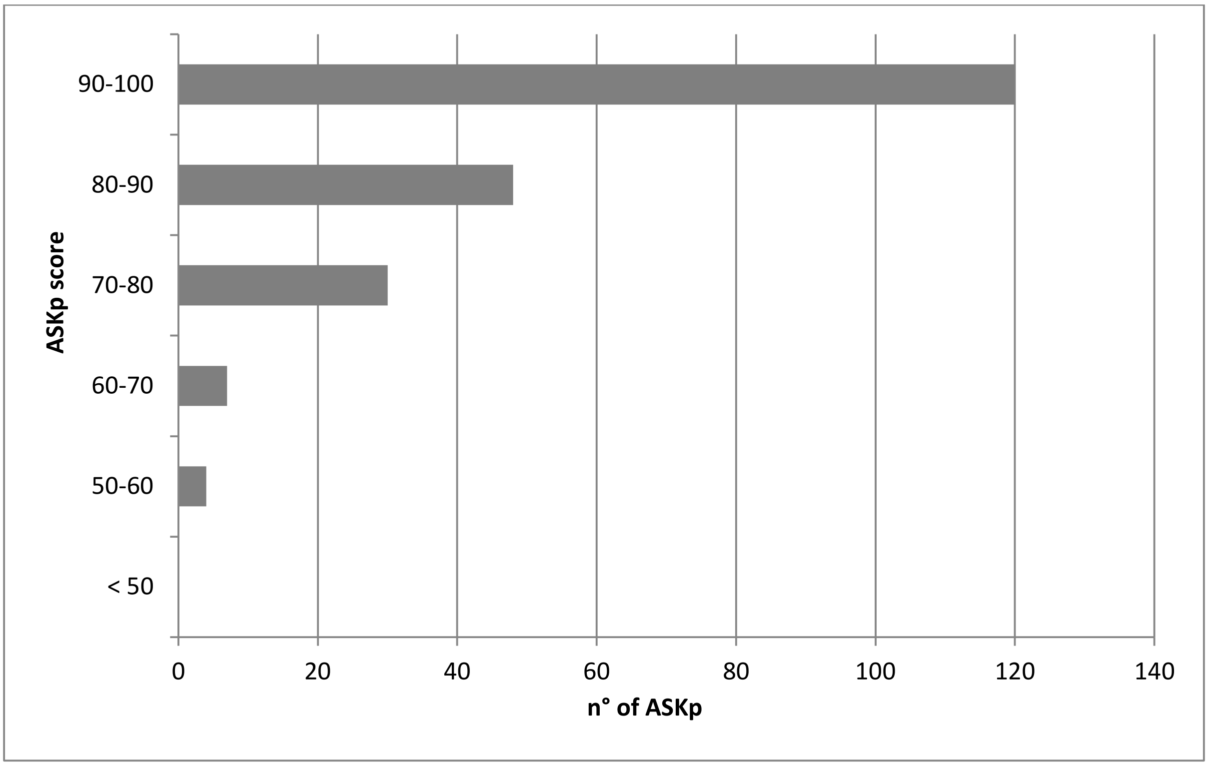


Figure 3. ASKp score distribution in the sample



Supplementary Table. Point estimates and distribution of scores obtained for each item in the sample examined.

	1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**	**11**	**12**	**13**	**14**	**15**
Mean	1.23	1.04	1.27	1.18	1.15	1.43	1.33	2.58	2.45	3.74	1.56	1.58	1.13	1.15	1.80
SD	0.63	0.25	0.77	0.55	0.50	1.11	0.93	1.51	1.62	1.86	1.18	1.24	0.68	0.76	1.48
Min	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00
1st Q	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	2.00	1.00	1.00	1.00	1.00	1.00
Median	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	2.00	1.00	1.00	1.00	1.00	1.00
3rd Q	1.00	1.00	1.00	1.00	1.00	1.00	1.00	4.00	3.00	6.00	1.00	1.00	1.00	1.00	2.00
Max	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00

item N	**16**	**17**	**18**	**19**	**20**	**21**	**22**	**23**	**24**	**25**	**26**	**27**	**28**	**29**	**30**	**Total**
Mean	1.50	1.84	1.83	1.61	1.44	1.44	1.29	2.09	1.62	1.21	1.15	1.05	2.31	1.26	1.49	1.59
SD	1.31	1.50	1.48	1.28	0.91	1.11	0.74	1.66	1.24	0.80	0.58	0.42	1.77	0.78	1.41	0.44
Min	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	
1st Q	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	
Median	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	
3rd Q	1.00	2.00	2.00	2.00	1.00	1.00	1.00	3.00	2.00	1.00	1.00	1.00	3.00	1.00	1.00	
Max	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	1.00	

Legend: N=number; SD=standard deviation; Min=minimum; Q=quartile; Max=maximum

Data entered into the Excel spreadsheet provided by the developers of the ASK©. In order to keep the data entry process simple, the Excel spreadsheet was designed to work with raw scores in the following format: 1 for the 1st response option, meaning “I did it every time I needed to,” 2 for the 2nd, 3 for the 3rd, 4 for the 4th, 5 for the 5th, meaning “I didn’t do it at all when I needed to,” and 6 for the not applicable option, if present. Therefore, for each item, the lower the score, the higher the perceived physical functioning related to the activity tested.

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CHAPTER II

The ASKp in children with cerebral palsy: evidence of reliability and construct validity

Submitted as:

Reliability and Construct Validity of the Activity Scale for Kids in Italian Children with Cerebral Palsy

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ABSTRACT

Aims: Physical functioning is crucial to participation in life activities. It should thus always be accurately assessed in children with cerebral palsy (CP). The Activities Scale for Kids performance (ASKp) is currently used for this purpose, although its psychometric properties have never been investigated in a wide population of children with CP, the principal cause of disability in industrialized countries.

Methods: This cross-sectional study provides for a single self-administration of the Italian ASKp to 206 children with CP to test its internal consistency and construct validity in this population.

Results: In children with CP, the average ASKp score was 75.5±18.1, which deviates from normative data. The ASKp showed substantial stability and good internal consistency when applied to the target population (Chronbach's α 0.91, 95% CI 0.89-0.93). It distinguished adolescents from children ($p<0.001$), children with different clinical manifestations of CP ($p<0.002$), children with or without intellectual deficit ($p<0.001$), and children who needed assistive devices for indoor mobility from those who did not.

Conclusions: Although the validation of the Italian ASKp in children with CP requires further confirmation, these results support its use to properly guide treatment that integrates children's perspective in their rehabilitation process.

Trial registration: ClinicalTrials.gov Identifier: NCT03325842

Key words: construct validity; outcome assessment; child; cross-cultural comparison; Activities Scale for Kids; activities of daily living

Cerebral Palsy (CP) describes a group of permanent disorders of the development of movement and posture that affects about 1 in 500 new live births worldwide, with an estimated prevalence of 17 million people (Oskoui, 2013; Graham, 2016). The hallmark of CP is motor impairment, which in most cases manifests itself as unilateral or bilateral spastic CP (Shevell, 2009) and is associated with other disturbances, such as sensation, perception, cognition, communication, behavior, and epilepsy, (Rosenbaum, 2006). For children living in industrialized countries, CP is the most common disease that leads to disability, i.e., limitations in physical functioning and restrictions in everyday activities (Reddihough et al. 2003). The activities of daily living (ADL) can be defined as specific, age-related life tasks oriented towards self-care, such as grooming or bathing (personal ADL), or oriented towards personal independence, such as preparing meals or doing homework (instrumental ADL) (WHO, 2007). During development, these tasks are essential to supporting participation in life situations, such as playing and performing age-related chores, whose accomplishment contributes to the child's health, functioning, and creation of personal biography (WHO, 2007). Participation in life activities is reduced in children with CP (Majnemer, 2008; Orlin, 2010) in proportion to their level physical functioning and mediated by personal and environmental factors (Morris, 2006). Thus, physical functioning and ADL performance should always be accurately assessed and appropriately addressed in the rehabilitation of children with CP.

One of the most widely used measures of pediatric physical functioning in ADL is the performance version of the Activities Scale for Kids (ASKp) (http://www.activitiesscaleforkids.com), a self-report questionnaire for children and adolescents aged 5-15 years with musculoskeletal disorders that takes 10 to 30 minutes to complete and requires no special training or equipment (Young, 1995; Young, 1996.) The

ASKp covers seven of the nine International Classification of Functioning, Disability and Health domains of activities and participation, and it measures performance, i.e., what the child usually does (McConachie, 2006; Chien, 2014).

The ASKp is unique because it captures the child's perspectives on the impact that disability has on daily life, without mediation by a third party (Young, 1995; Harvey, 2008). The integration of this perspective is crucial when planning rehabilitation interventions because it permits focusing on the child's needs and monitoring changes over time. Thus, rehabilitation professionals frequently use this scale in research and in clinical practice (Harvey, 2008; Capio, 2010).

The ASKp has been tested for its validity through appropriate studies (Young, 1995; Young, 1996; Young, 2000; Plint, 2003), showing excellent reliability (Harvey, 2008; Capio, 2010; Young, 2000) and good content, concurrent, construct, and discriminative validity (Harvey, 2008; Capio, 2010; Young, 2000; Pencharz, 2001). The validity of the ASKp had been tested on several populations of children and adolescents with musculoskeletal disorders of various nature, but CP was not widely represented in these populations (Young, 2000; Pencharz, 2001). As CP is a complex condition, it is likely that ADL execution is affected differently in children with this disorder compared to others (e.g., autism, spina bifida). It therefore seems appropriate to ascertain the validity of the ASKp in a large population of children with CP.

The aim of this study was to collect the first evidence on the reliability and construct validity of the ASKp in a population of Italian children with CP. We planned to ascertain internal consistency of the Italian ASKp and, if this requirement was satisfied, we planned to proceed to testing the following a priori assumptions:

(1) Since the age range of the population tested was quite broad, adolescents were expected to perceive fewer limitations in the activities tested than would younger individuals, as reflected by the ASKp.

(2) Since unilateral and bilateral spastic CP are substantially different clinical manifestations of CP, we expected that these two populations would perceive different levels of physical function.

(3) We expected children with no intellectual deficit to perceive fewer limitations in the activities tested.

(4) We expected that children who did not need assistive devices for mobility would perceive fewer limitations than those who did need them.

Methods

Design

This independent cross-sectional study provided for a single self-administration of the Italian version of the ASKp to children with CP.

Participants (Subjects)

Between 2014 and 2019, we recruited 220 children with CP (122 males; range 5-15 years old; mean age 10.6 ± 2.9 years) at the Children Rehabilitation Unit for Severe Developmental Disabilities, of the Azienda Unità Sanitaria Locale – IRCCS of Reggio Emilia (Italy). Children were included according to the following inclusion criteria: a) confirmed diagnosis of CP based on diagnostic criteria (MRI and clinical history); b) age 5-15 at time of recruitment; c) clinical manifestation of unilateral or bilateral spastic CP. We excluded children whose moderate or severe intellectual deficit was based on their documented regular attendance of and appropriate scholastic performance in a mainstream primary school, on their lack of need for a learning support teacher, and on the clinical judgment of their referring physician.

Children and their parents gave their informed consent for participation in this study, which was approved by the Ethics Committee of the Province of Reggio Emilia (Protocol n. 2017/0004096).

Measures (Instrumentation)

The Italian version of the ASKp was cross-culturally adapted and tested for content validity in a population of 35 children with CP, their parents, and pediatric physical therapists (Fabbri, 2016). As does the original questionnaire, the Italian ASKp asks what children “did do” during the previous week, referring to 30 usual activities that cover seven subdomains of physical functioning (standing skills, personal care, dressing, locomotion, mobility, play, other skills). The child answers each of the 30 activity items using a 5-point ordinal scale. For any single item, the lower the score, the higher the perceived physical functioning related to the activity tested. Data are entered into a spreadsheet provided by the developers of the ASK©, and results are presented in a single summary score from 0 to 100, with higher scores indicative of perception of full physical function.

Besides the 30 activity items that contribute to the final score, the ASKp also inquiries about the assistive devices used for indoor and outdoor mobility and the degree of assistance required to complete the questionnaire.

The Italian ASKp was also tested in a population of healthy Italian children, confirming its high acceptability and providing the first evidence of its construct validity (data submitted).

Procedure

The physician’s agenda was checked weekly to identify the children with CP for whom a medical appointment was scheduled. Eligibility criteria were verified by checking the medical record. In case of doubt, the referring physician was asked for clarification. Parents of eligible children were then contacted to ask their permission to interview their child about his/her ADL for research purposes. Those parents that agreed were asked to come to the next scheduled appointment one hour earlier.

The day of the appointment, the research aim and procedure were explained to the parents and their children in more detail; if they agreed to participate, written informed consent was collected and the ASKp was self-administered to the child right away, in a separate quiet room. A researcher was present to help children younger than 9 years or those who asked for support to complete the questionnaire. For those under age 9, the parents’ presence in the room was granted provided that they did not intervene in any way so as not to influence the child's answers.

If the child was worried because of the doctor’s appointment, the ASKp was administered afterwards, when feasible. In a few cases, when this was not possible due to organizational reasons or to the child’s apprehension, parents were asked to administer the ASKp at home in the following few days and to send it back to the researchers. In those cases, precise indications were given to the parents to guarantee reliable self-administration (i.e., quiet room, no distractions, so as not to influence the answer).

Data Analysis

A priori, we established recruiting 200 individuals based on the subject-to-item ratio and based on data from the initial study of psychometric properties of the original ASKp, which was tested for validity on 200 children (Young, 2000). The subject-to-item ratio is a commonly recommended approach to determine sample size in validation studies, but this ratio can vary depending on the approaches followed (Anthoine, 2014). Recommendations range from 2 to 20 subjects per item (Hair, 1995; Kline, 1979), with an absolute minimum of 100-250 subjects (Everitt, 1975; Gorsuch, 1983; Cattell, 1978).

Descriptive statistics were used to summarize clinical and demographic data.

The analysis of the internal consistency was carried out by calculating Cronbach's alpha, both overall and by the 1-item out analysis, in order to verify the possible presence of high-impact items on internal consistency of the instrument. Several alpha thresholds have been reported in the literature for interpreting the degree of consistency but, typically, a positive result ranges from 0.70 (acceptable) to 0.95 (very good) (Ponterotto, 2007; Tavakol, 2011).

Finally, a box plot distribution of the ASK-p score was made for the main demographic and clinical characteristics of the sample (sex, age class, diagnosis, presence of mild intellectual deficit, and need for assistive devices for mobility) to visually check for any possible trend; distributions were then further formally compared using Kruskal-Wallis test in order to detect a positional shift in any of the groups considered. Confidence intervals were two-tailed and calculated considering a 0.95 confidence level. The tests performed were considered statistically significant if the *p* values were <0.05. Statistical analyses were performed using R 3.5.2.

Results

In this study 220 ASKp were administered to participants, returned to researchers, and included in the analysis.

Males made up 55% of the sample, and bilateral CP was more prevalent (69%). All children fell into categories I to III of the Gross Motor Function Classification System (GMFCS), meaning that they were ambulant children, some of whom might need assistive devices for indoor and outdoor mobility. Few children (10%) manifested mild intellectual deficit. Table 1 shows the participants' characteristics.

The ASKp summary score distribution in the sample is summarized in Table 2. Fourteen children answered fewer than 23 questions; obtaining a summary score for them was therefore impossible. The distribution of the ASKp summary score is presented for the whole sample of 206 children and separately for sex, for age class (namely those attending elementary, middle, or high school), and for the main clinical characteristics. Figure 1 represents the frequency of distribution of the ASKp score. Fifty-seven participants (29%) obtained a score of ≥ 90, meaning complete or almost complete physical function in usual activities; nonetheless, 150 participants (73%) perceived limitations, in part very severe (24 with score < 50).

Table 3 reports the ASKp score distribution (frequency and percentage) and missing data for each of the 30 questionnaire items.

The average Cronbach's α, calculated on the 206 questionnaires for which the summary score was obtained, is 0.91 (95% CI 0.89-0.93), showing a good internal consistency for the Italian ASKp. The 1-item out analysis confirmed the questionnaire's substantial stability, with Cronbach's α values equal to 0.90 or 0.91 for all the analyses performed (Appendix 1).

Finally, figure 2 represents the box plot of the ASKp score distribution for the main demographic and clinical characteristics of the sample. As expected, the ASKp score did not differ for sex. However, it did distinguish different levels of physical functioning in ADL related to different age classes, confirming that adolescents perceived fewer limitations in the activities tested than did children ($p < 0.001$), as also shown in the healthy population (data submitted). Also, as hypothesized, the ASKp was able to differentiate between individuals with different clinical manifestation of CP: in fact, individuals with bilateral CP perceived higher levels of physical limitations in ADL than did those with unilateral CP ($p < 0.002$); individuals without intellectual deficit perceived fewer limitations than did those with mild deficit ($p < 0.001$), and individuals who did not need assistive devices for indoor mobility perceived fewer limitations than did those that needed them ($p < 0.001$).

Discussion

The data collected confirm the internal consistency and stability of the Italian version of the ASKp, and all the assumptions postulated a priori to verify its construct validity were confirmed. Therefore, this study represents the first evidence of validity of the ASKp specifically applied to children with CP.

Considering that CP is a condition substantially different from those mostly represented in the sample of the original ASKp validation study (neuromuscular, orthopedic and arthritic conditions, autism, etc.) (Young, 2000), it is important to check the psychometric properties of the ASKp in this specific group of children. As expected, our study confirmed that the ASKp measures pediatric physical functioning in ADL and does not contain items that weaken the reliability of the score. Having confirmed unidimensionality and acceptable stability in children with CP, we addressed the traditional measurement properties for construct validity, meaning that we tested and verified some hypotheses that concur with the theoretical construct the ASKp was designed to measure. To our knowledge, no previous validation study of ASKp tested its ability to identify different physical functioning levels associated with different age classes. However, since the population tested has quite a wide age range, it is plausible to assume that the performance of daily activities (especially in domestic or community-based) develops with age (Van Gorp, 2018). Consequently, the ASKp score should reflect the individual child's developmental stage. Indeed, the average summary score varied based on different age-related levels of physical functioning in ADL.

Regarding the ability to identify subjects with limitations, the ASKp Italian version, like the original one, could differentiate between the population of children with CP and the healthy one, as the average score of 75.5 (± 18.1) is substantially different from the average of healthy Canadian (93.2 ± 6.45) or Italian children (89.06 ± 10.57) (Plint, 2003; data submitted). More interestingly, in a sample of children with CP with mild to moderate gross motor limitations (GMFCS categories I to III), the ASKp was able to distinguish between different levels of physical functioning related to the two main clinical manifestations of CP (82.5 ± 13.6 of unilateral versus 72.4 ± 19.0 of bilateral). This result confirms its clinical validity and is coherent with the results obtained in children with neuromuscular disorders of various etiology (Young, 2000), where the ASKp distinguished between children with mild, moderate, or severe disability. As expected, in our population of ambulant children, the ASKp highlighted a lower level of physical functioning perceived by those who needed assistive devices for indoor mobility; it was also able to differentiate between children with and without mild intellectual deficit, confirming the role of intellectual disability in affecting the development of daily activity performance (Van Gorp, 2018).

To summarize, our data confirm the construct validity of the Italian version of the ASKp also in the population with CP.

However, some limitations of the study design suggest caution in interpreting these results. First, as the sample included only children with mild to moderate gross motor limitations (GMFCS I to III), any inference cannot be extended to the entire population with CP. For example, as the ASK is self-administered, children with the most severe disability were not invited to participate in the study so as to spare them any unnecessary emotional suffering. Second, the classification of children according to the presence of intellectual deficit was based only on clinical evaluation because we applied the ASKp in a pragmatic clinical context. Our choice was borne out by the results we obtained, in line with those from other studies (van Gorp, 2018; Vos, 2013). However, it would be prudent to check whether the self-administration of the ASKp in these children is reliable enough, as already demonstrated in children without intellectual disability (Young, 1995). Therefore, future study designs could compare the ASKp scores obtained by children with mild cognitive impairment to reference scores (e.g., that of physical therapist and/ or parent). Finally, in the absence of a gold standard that evaluates exactly the same construct, we would suggest conducting studies that examine the convergence of the ASKp results with those of another outcome measure that investigates similar attributes.

Despite the need for further investigations, this study, conducted on a large population of children with CP, provides clinicians and researchers with a solid interpretative basis useful when evaluating their patients. This is of the utmost importance given that the ASKp is widely used to evaluate physical functioning in children with CP (Morris, 2005; Harvey, 2008; Capio, 2010).

Conclusions

To conclude, the results obtained confirm the fundamental psychometric properties of the Italian ASKp when applied to the population with CP. Although the validation process of the Italian ASKp requires further work, this assessment tool can support clinicians in integrating the child's perspective into the priority aims of rehabilitation. Given that CP is a complex pathology causing lifelong disability with strong repercussions on autonomy and that the quality of life of children with CP is closely linked to being able to participate in everyday life situations, this is fundamental (WHO, 2007).

Disclosure statement

The authors report no conflict of interest.

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Table 1. Sample characteristics distribution.

Table 2. ASKp summary score distribution

Table 3. Score distribution for the 30 items of the ASKp

Figure 1. Frequency of distribution of the ASKp score.

Figure 2. Box plot of the ASKp score distribution

Appendix 1. Results of the 1-item out analysis

Table 1. Distribution of sample characteristics

Age	Years	10.6 (± 2.9)
Sex	Female	98 (45%)
	Male	122 (55%)
Clinical manifestation of CP	Unilateral CP	68 (31%)
	Bilateral CP	152 (69%)
Intellectual deficit	None	198 (90%)
	Mild	22 (10%)
Assistive devices	None	86 (39%)
	Outdoor only	40 (18%)
	Indoor and outdoor	93 (42%)

Legend: CP= Cerebral Palsy

NOTE: 1 participant did not answer the question regarding assistive devices in use.

Table 2. ASKp summary score distribution

		N	Min	IQ	Median	Mean	IIIQ	Max	SD
	Total sample	**206**	**26**	**64**	**80**	**75.5**	**91**	**100**	**18.1**
Sex	Female	92	32	63	78	74.5	91	100	18.5
	Male	114	26	65	80	76.4	91	100	17.8
Age	5-10 years	112	35	60	74	70.3	83	97	16.6
	11-13 years	36	34	68	85	79.7	93	99	17.9
	14-15 years	58	26	72	90	83.0	96	100	18.0
Clinical manifestation of CP	Unilateral	63	36	76	85	82.5	93	100	13.6
	Bilateral	143	26	61	74	72.4	89	100	19.0
Intellectual deficit	None	186	32	68	81	77.2	91	100	17.3
	Mild	20	26	49	59	60.3	77	92	18.3
Assistive devices	None	81	46	78	85	83.6	94	100	12.8
	Outdoor	37	36	73	85	80.4	94	100	17.4
	Ind. & Out.	88	26	52	68	66.1	81	99	18.3

Legend: N= Number; Min= Minimum; IQ= 1st Quartile; IIIQ= 3rd Quartile; Max= Maximum; SD= Standard Deviation; Ind.= Indoor; Out.= Outdoor

Table 3. Score distribution for the 30 ASKp items

	1	%	2	%	3	%	4	%	5	%	Missing	Total
I_01	136	62.4	22	10.1	25	11.5	11	5.0	24	11.0	2	220
I_02	175	79.5	15	6.8	10	4.5	5	2.3	15	6.8	0	220
I_03	106	48.2	18	8.2	32	14.5	17	7.7	47	21.4	0	220
I_04	128	58.2	27	12.3	23	10.5	16	7.3	26	11.8	0	220
I_05	112	50.9	26	11.8	31	14.1	16	7.3	35	15.9	0	220
I_06	83	38.2	29	13.4	43	19.8	26	12.0	36	16.6	3	220
I_07	85	38.8	28	12.8	29	13.2	16	7.3	61	27.9	1	220
I_08	55	28.8	17	8.9	22	11.5	15	7.9	82	42.9	29	220
I_09	137	67.2	26	12.7	20	9.8	14	6.9	7	3.4	16	220
I_10	118	55.4	11	5.2	18	8.5	14	6.6	52	24.4	7	220
I_11	105	49.8	25	11.8	30	14.2	21	10.0	30	14.2	9	220
I_12	79	42.2	20	10.7	37	19.8	24	12.8	27	14.4	33	220
I_13	184	84.4	14	6.4	7	3.2	2	0.9	11	5.0	2	220
I_14	151	75.5	17	8.5	16	8.0	10	5.0	6	3.0	20	220
I_15	151	71.6	15	7.1	19	9.0	5	2.4	21	10.0	9	220
I_16	142	68.9	19	9.2	15	7.3	5	2.4	25	12.1	14	220
I_17	125	60.4	29	14.0	20	9.7	11	5.3	22	10.6	13	220
I_18	93	50.3	31	16.8	20	10.8	18	9.7	23	12.4	35	220
I_19	87	42.6	42	20.6	34	16.7	18	8.8	23	11.3	16	220
I_20	162	74.7	13	6.0	13	6.0	15	6.9	14	6.5	3	220
I_21	117	63.9	23	12.6	14	7.7	8	4.4	21	11.5	37	220
I_22	173	80.1	17	7.9	6	2.8	11	5.1	9	4.2	4	220
I_23	134	69.4	15	7.8	11	5.7	8	4.1	25	13.0	27	220
I_24	120	59.7	14	7.0	22	10.9	14	7.0	31	15.4	19	220
I_25	136	75.6	11	6.1	11	6.1	6	3.3	16	8.9	40	220
I_26	93	57.1	21	12.9	25	15.3	6	3.7	18	11.0	57	220
I_27	147	71.0	22	10.6	19	9.2	3	1.4	16	7.7	13	220
I_28	122	63.9	19	9.9	18	9.4	13	6.8	19	9.9	29	220

I_29	90	48.1	27	14.4	25	13.4	19	10.2	26	13.9	33	220
I_30	155	72.4	15	7.0	12	5.6	14	6.5	18	8.4	6	220
30 items	3701	60.4	628	10.3	627	10.2	381	6.2	786	12.8	477	6600

Legend: I=item

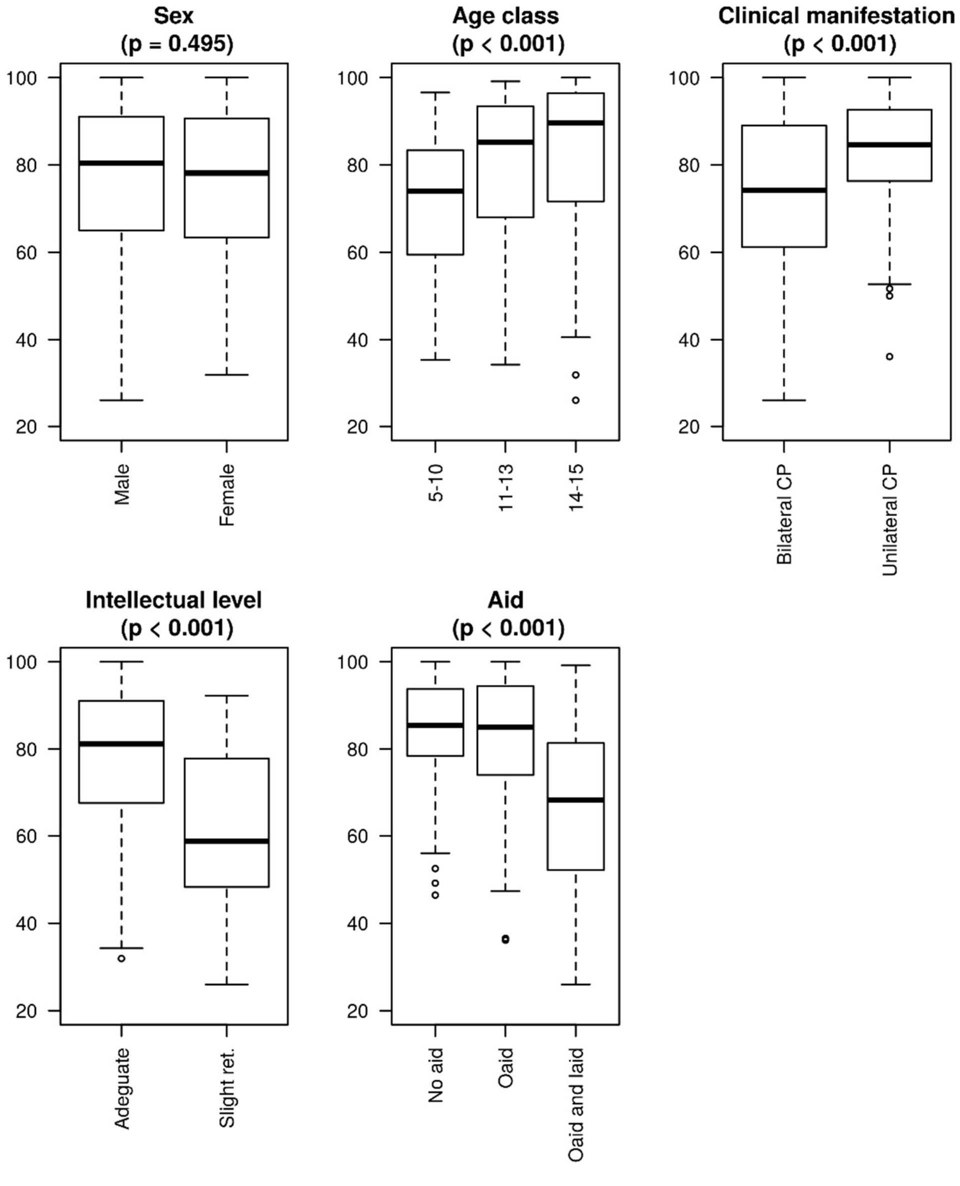

Figure 1. Frequency of distribution of the ASKp score.

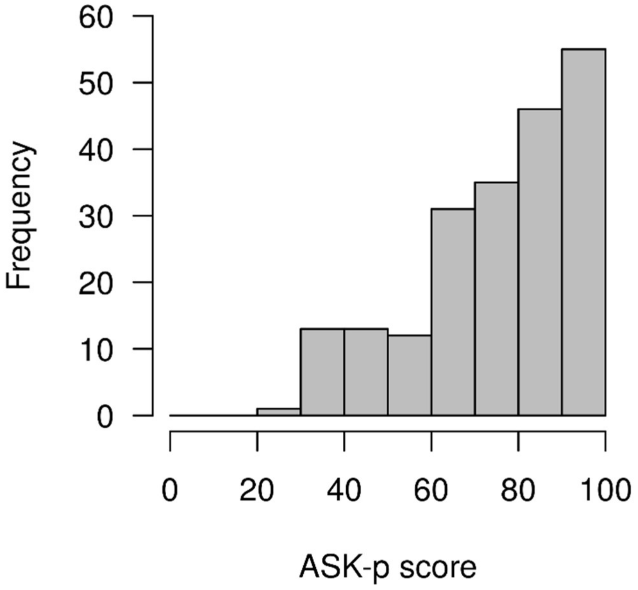

Figure 2. Box plot of the ASKp score distribution

Appendix 1. Results of the 1-item out analysis

	raw_alpha
Q01_denti	0,90
Q02_gabinetto	0,91
Q03_pulizia	0,90
Q04_maglietta	0,90
Q05_pantaloni	0,90
Q06_abbottonarsi	0,90
Q07_scarpe_messe	0,90
Q08_cura	0,91
Q09_scrittura	0,91
Q11_spuntino	0,91
Q12_lavoretti	0,90
Q10_camminare	0,90
Q13_mobilità_casa	0,91
Q14_camminare_luog_affol	0,91
Q15_scale	0,91
Q25_muoversi_aperto	0,90
Q27_superf_scivolose	0,91
Q28_salita	0,91
Q26_fare_sport	0,90
Q29_passo_amici	0,91
Q16_in_piedi	0,90
Q17_trasporto	0,91
Q18_allungarsi	0,90
Q19_portare_cibo	0,91
Q24_aprire_porte	0,90
Q20_sedersi_alzarsi_sedia	0,90
Q21_sedersi_pavimento	0,91
Q22_alzarsi_letto	0,91
Q23_abbassarsi_pavimento	0,90
Q30_entr_esc_auto	0,90

Legend: I=item

CHAPTER III

Convergent validity with measures of motor function in children with cerebral palsy.

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Construct validity of the Activities Scale for Kids performance in children with cerebral palsy: brief report

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in

Developmental Neurorehabilitation

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ABSTRACT

Aims: This study collects evidence of construct convergent validity of the ASKp, comparing its results with the 66-item Gross Motor Function Measure (GMFM-66) and with the Gross Motor Function Classification System (GMFCS) and testing the ASKp's ability to discriminate between individuals with different functional capabilities.

Methods: This cross-sectional study involved 60 children with CP assessed with the GMFM-66 who self-administered the Italian version of the ASKp.

Results: Children were 10.9 (±3) years old with GMFCS Level I-III. Moderate correlations were found between GMFM and ASKp scores ($r = 0.577$; $p < 0.001$), and between GMFCS levels and ASKp score ($r_S = -0.541$, $p < 0.001$). The ASKp discriminated between children with different functional capabilities, as determined by the GMFCS ($F = 18.2$, $p < 0.001$).

Conclusions: The ASKp is valid to assess physical functioning, a crucial domain in the rehabilitation of children with CP.

Trial registration: ClinicalTrials.gov Identifier: NCT03325842

Keywords: cerebral palsy; outcome assessment; construct validity; child; Activities Scale for Kids

Introduction

In industrialized countries, cerebral palsy (CP) is the most common disease that leads to childhood disability;[1] the limitations in motor capacity, a hallmark of CP, lead to restrictions in physical functioning and everyday activities.[2] Thus, the assessment, goal setting and treatment trajectories of CP should be framed within the International Classification of Functioning, Disability and Health (ICF) model,[3] which illustrates how the body's function and structures are reciprocally associated with activities and participation and how this dynamic relation is mediated by personal and contextual factors. Focusing on activities and participation, the ability to perform activities is a vital outcome for children with CP and other chronic disabilities as it supports participation in life situations and contributes to the child's health, functioning and creation of personal biography.[4] Therefore, motor capacity must clearly be preserved or restored in children with CP because it is positively associated with physical functioning and activity performance.[5,6] For this reason, the 66-item Gross Motor Function Measure (GMFM-66)[7] is universally used to measure the severity of motor disability in this population and the Gross Motor Function Classification System (GMFCS)[8] is used to categorize children with CP based on their functional capability. So, as the GMFM-66 measures the execution of skills in a standardized environment and the target of GMFCS is capability, neither of these tools can fully describe a child's habitual behavior.[9] Thus, there is a need for tools in the rehabilitation of children with CP that measure pediatric physical functioning while performing activities in the context of daily life.

One of the most widely used measures of pediatric physical functioning is the performance version of the Activities Scale for Kids (ASKp),[10] which covers seven of the nine ICF domains of activities and participation and measures performance, i.e. what the child usually does.[11,12] Rehabilitation professionals involved in the care of children with CP frequently use this feasible self-report questionnaire both in research and in clinical settings.[13,14] However, the psychometric properties of the ASKp have never been tested on a large population of children with CP; the original validation studies were conducted on a large sample with mixed diagnoses, such as neuromuscular diseases or dysmorphisms.[10,15] As CP is a complex condition and its prognosis is different from that of other developmental diseases, the ASKp has recently been cross-culturally validated by our group,[16] and its fundamental psychometric properties have been tested in a large population of children and adolescents with CP [data submitted].

This study aimed to collect evidence of construct validity of the ASKp. We tested the ASKp convergent validity with the GMFM-66 and the GMFCS in a sample of Italian children with CP, and we tested the ASKp's ability to discriminate between individuals with different levels of functional capability. Thus, as gross motor capacity and functional capability are among the determinants of activity performance, the following a priori assumptions were to be verified: (1) we expected a positive correlation between the GMFM-66 and the ASKp scores; (2) we expected a negative correlation between the levels of GMFCS, where higher scores indicate more severe disability, and the ASKp score; (3) accordingly, we expected that the average ASKp score of children with different levels of functional capability would differ, demonstrating higher levels of physical performance in children with lower disability.

Materials and Methods

Design

This cross-sectional study was conducted with the formal approval of the local Ethics Committee (Province of Reggio Emilia, Protocol n. 2017/0004096). The study consisted of a single self-administration of the Italian version of the ASKp to children with CP who, for clinical reasons, were simultaneously assessed with the GMFM-66.

Study sample

Children with CP referred to the Children Rehabilitation Unit for Severe Developmental Disabilities of the Azienda Unità Sanitaria Locale – IRCCS of Reggio Emilia (Italy) from 2017 to 2019 were screened for eligibility in this study. Inclusion criteria were:

- confirmed diagnosis of CP based on diagnostic criteria (MRI and clinical history)
- age 5-15 years
- in need of clinical assessment of gross motor function using the GMFM-66.

We excluded children with moderate or severe intellectual deficit based on their documented regular attendance of and appropriate scholastic performance in a mainstream primary school, on their lack of need for a learning support teacher and on the clinical judgment of their referring physician.

Children and their parents gave their informed consent for participation in this study.

Procedures

During a routine appointment, the research aim and procedure were illustrated to parents of eligible children, and written informed consent was collected from families who agreed to participate. As part of the clinical evaluation, participating children were assessed with dimensions D and E of the GMFM-66 following precise guidelines,[17] and categorized based on their functional capability using the GMFCS.

Whenever possible, the cross-culturally adapted Italian version of the ASKp[16] was self-administered right away in a quiet room, with assistance provided by a physiotherapist to those children who requested it. The presence of a parent was allowed for children under age 9, provided that they did not intervene in any way so as not to influence the child's answers. If self-administration was unfeasible during the appointment, parents were asked to provide for a reliable self-administration of the ASKp at home (i.e. a quiet room with no distractions, so as not to influence the answer) and to send it back to the researchers as soon as possible. In these cases, if the ASKp was returned more than three months after GMFM-66 administration, they were excluded.

The ASKp inquiries about a child’s performance during the previous week of 30 usual activities that refer to seven subdomains of physical functioning (standing skills, personal care, dressing, locomotion, mobility, play, other skills). Results are summarized in a single score from 0 to 100, with higher scores indicating the perception of full physical function. Three further items that do not contribute to the final score inquire about the degree of assistance needed to complete the questionnaire and the assistive devices used for indoor and outdoor mobility.

Data Analysis

Descriptive statistics were used to summarize clinical and demographic data. Mean and standard deviation (SD) were used to summarize continuous variables, whereas counts and frequencies were used to summarize categorical variables. The strength and direction of associations were analyzed using parametric and nonparametric correlation coefficients (the Pearson r and the Spearman $r_{s,}$ respectively). The ASKp scores were compared with functional capability levels of GMFCS using ANCOVA, correcting for the influence of age, performing Tukey test for post-hoc comparisons and using the Bonferroni correction for multiple comparisons.

The tests performed were considered statistically significant if the *p* values were < 0.05. Statistical analyses were performed using Excel 16.0 and Jamovi 1.0.7.0.

Results and Discussion

Seventy-six children were included in the study. However, the data of 16 of these children were not included in the analysis because: a) two children did not answer at least 23 questions from the ASKp, which represents the minimum number of answers to compute the overall score; b) in twelve cases it was not possible to test all the items of the D and E dimensions of the GMFM-66, making it impossible to calculate the overall score; c) in two cases the ASKp was self-administered at home and returned later than three months after the GMFM-66 assessment.

The study sample included 60 children (52% males) aged 10.9 years (min.5 – max 15) with bilateral or unilateral spastic diplegia (70% and 30%, respectively), classified in the Levels I (37%), II (45%) and III (18%) of the GMFCS. The average GMFM-66 score in the whole sample was 73.3 (± 8.9) and the average ASKp score was 75.3 (± 17.5). Most children used an assistive device for indoor and/or outdoor mobility (50% indoor and outdoor, 20% outdoor), and only a few manifested mild intellectual deficit (13%).

As hypothesized, a moderate positive correlation between the GMFM-66 and the ASKp scores and a moderate negative correlation between the levels of GMFCS and the ASKp score were found ($r = 0.577$; $p < 0.001$ and $r_s = -0.541$, $p < 0.001$, respectively), confirming that the ASKp is a valid measure of physical functioning, a construct related to gross motor capacity.

Further, the average level of physical performance measured by the ASKp differed based on the GMFCS level ($F = 10.1$, $p < 0.001$), confirming the hypothesis that the ASKp can discriminate between children with different levels of functional capability (Table 1 and Figure 1). The post hoc comparisons showed a statistically significant difference between the average ASKp score of both children in GMFCS Level I and Level II compared to children in Level III ($p < 0.001$ and $p = 0.013$, respectively).

Taken together, these results add evidence to confirming the construct validity of the ASKp, and support its use in the assessment of the physical performance and limitations in activities of children and adolescents with CP. This is clinically relevant, given that the ASKp is, to our knowledge, one of the few scales that capture the child's perception of his / her abilities to carry out usual activities.

A limitation of the study is certainly the absence of an outcome measure that can be considered a criterion for the measurement of physical performance in this population. However, the construct assessed by the GMFM is a determinant of physical performance and the GMFM-66 is a valid and reliable tool used worldwide to assess it.[18] Although not perfect, the strength of the association between these two variables confirms the construct validity of the ASKp.

Another limit is certainly the absence of children with severe motor disabilities due to CP, namely those classified as level IV or V of the GMFCS. This is because the clinical routine in the context in which the research was carried out foresees the frequent administration of sections D and E of the GMFM-66 to ambulant children with CP. This limit, dictated by feasibility aspects of the study, is also present in previous studies.[19] Future research should thus verify whether the demonstrated correlations are confirmed in children with CP with lower functional capability.

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Declaration of interests

The authors report no conflict of interest.

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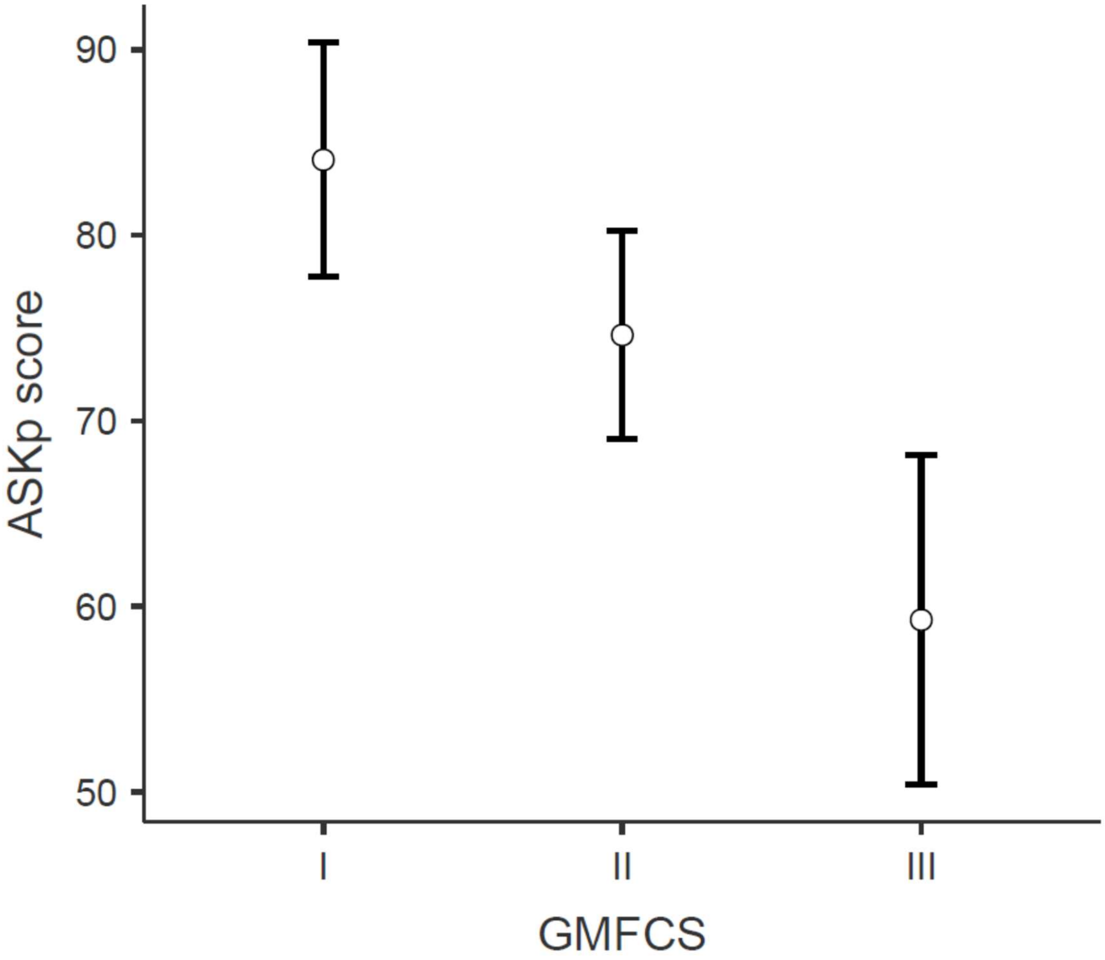
Table 1. Distribution of sample characteristics per GMFCS Level

	I	II	III
n.	22	27	11
Age (SD)	11.7 (3.1)	10.6 (3.1)	9.9 (2.4)
ASKp score (SD)	85.1 (13.6)	74.3 (17)	58.0 (11.2)
GMFM-66 score (SD)	81.5 (5.5)	72.6 (7.4)	63.6 (4.5)
Bilateral/Unilateral CP	9/13	22/5	11/0
Intellectual deficit Yes/No	1/22	3/24	4/7
Assistive device Yes/No*	13/9	19/8	10/1

*as self-reported by children participating in this study

Legend: GMFCS= Gross Motor Function Classification System; SD= Standard deviation; ASKp= Activities Scale for Kids performance; GMFM-66= Gross Motor Function Measure 66 items CP= Cerebral Palsy;

Figure 1. Level of physical performance based on GMFCS Level



ASKp= Activities Scale for Kids performance; GMFM-66= 66-item Gross Motor Function Measure

DISCUSSION

Implications for rehabilitation of children with cerebral palsy

This project has achieved its dual purpose: to verify the basic psychometric properties of the Italian version of the ASKp and to verify this tool's validity in the population of children with CP.

The clinical and research implications of this work are relevant because, as already argued in the Introduction, there is a lack of rehabilitation outcome measures formally validated in Italian [www.sif-fisioterapia.it]. Moreover, despite the fact that the ASKp is widely used to evaluate physical disability in children with CP, its psychometric properties have never been studied in a large population of children with sequelae of this pathology.

Outcome measures in rehabilitation encompass a plethora of relevant constructs and can be used to enhance clinical decision making at the level of individual, group, and client programs. Therefore, it is very important to define a priori what to evaluate and why and to choose the most appropriate measure. Besides quality of life, rehabilitation may target impairment, activity limitations, participation in life activities, and environmental factors. As measurements of impairment do not correlate strongly with activity or with participation [Kerr, 2007; Hazard, 1994], a first fundamental assumption in rehabilitation is that the outcomes selected at the level of the individual should reflect that individual's ability to perform an activity or to participate in a desired social role. Outcome measures able to determine the individual's ability to be active and participate in life are likely to be more relevant both to the client and to the rehabilitation professional. At the group level, rehabilitation outcome measures may be designed to discriminate between individuals on a specific construct or may be useful to predict a future effect based on data collected at an earlier point in time. These outcomes allow better organization of social-healthcare services and rehabilitation programs. Finally, measures may be designed to evaluate the effect of a rehabilitation program quantifying changes over time at the individual or group level; achieving this aim is vital to clinical practice and research.

However, both in rehabilitation research and in clinical practice, several of the variables of interest and relevant outcomes correspond to abstract theoretical constructs; thus, measuring these variables with valid and reliable tools is a crucial component in the quality of care and in research. This is even more true when a variable is measured by means of self-reporting, as in the case of the ASKp, due to the risks of measurement error and of conscious bias. However, self-assessment captures a subjective and unique view of health status and quality of life issues that are not necessarily apparent, and thus becomes essential to the therapy process as it facilitates both the definition of goals that are relevant to the individual and to patient empowerment, even in children [Missiuna & Pollock, 2000; Porter, 2010]. The fundamental role of self-reporting is even more evident in the comprehensive assessment of patients with long-term conditions, where disability and symptoms usually require complex interventions through a client-centered approach that fosters self-management beyond ongoing medical support and periodic monitoring of function, pain, and quality of life [Peters, 2014].

This is certainly the case with CP, whose chronic nature impacts all aspects of a child’s life. It is therefore of paramount importance that the child’s perspective contributes to setting relevant goals and is represented in measuring the effectiveness of interventions implemented to improve the life of this young patient [Morris, 2005; Harvey, 2008].

Although considerable attention has been paid to patient-reported outcome measures since the 1990s, and although it has been verified that very young children hold a unique view of themselves that is valid and stable over time [Sturgess, 2002], as already discussed in the Introduction, to date very few self-assessment tools are available for children with CP. In most clinical fields, including the rehabilitation of children with CP, self-assessment is primarily used to describe the multidimensional construct of quality of life [Mpundu-Kaambwa, 2018]. However, to set relevant and achievable rehabilitation goals for CP, it is important to know the patient's perception of more specific constructs that contribute to quality of life [Vargus-Adams, 2011].

The ASKp addresses the construct of physical disability related to the performance of day-to-day activities. This construct can easily be traced back to the *activities and participation* domain of the ICF, as *activity* is the execution of tasks in daily life situations [WHO, 2001; WHO, 2007]. Children with CP show activity limitations and experience restrictions in participation: these restrictions can have serious consequences on psychosocial well-being of children with CP compared to their peers [Shikako-Thomas, 2012]. Therefore, it is essential to use valid outcome measures that describe the impact of physical disability on activities and participation and that contribute to reliably depicting the child's quality of life [Park, 2016]. The ASKp can contribute to this; it was designed to reliably collect self-reporting from children about the impact of their physical disability on activity performance [Sturgess, 2002]. Further, the ASKp allows a better understanding of the everyday challenges that children and adolescents with CP face.

Proof of reliability and validity of the ASKp in children with cerebral palsy

Overall, the results of this project affirm the psychometric properties of the ASKp in children with CP and confirm that this tool can be used to quantify physical disability with negligible errors in this population.

Regarding the reliability of this assessment tool, the internal consistency of the Italian version of the ASKp was verified in a sample of individuals with CP whose size was large enough to yield reliable results; it registered a Cronbach's alpha of 0.91 (95% CI 0.89-0.93), which is good but not as high as those recorded in the validation processes of the original version and of the Portuguese version of the ASKp (both 0.99) [Young, 2007; Paixao, 2016]. It must be noted that that the alpha level depends not only on the magnitude of the correlation between the items of a scale but also on the number of the items tested: the alpha therefore commonly exceeds 0.90 when a large number of items are tested. This is because if we assume that every response to an item has some associated error of measurement, then by averaging responses over a series of questions, we can reduce this error. On the other hand, when the alpha is too high, this may suggest a high level of item redundancy, i.e., several items asking the same question in slightly different ways [Streiner, 2015]. For an assessment tool, therefore, a good level of internal consistency, compared to an excellent one, could even demonstrate a better balance between the desired homogeneity of the items and the need to evaluate as many aspects as possible of the same construct.

It might be thought that this small difference in the values of internal consistency of different versions of the ASKp depended on the target population, as the homogeneity of both the original and of the Portuguese versions of the ASKp was tested on children with a broad range of musculoskeletal disorders, while we tested it on children with CP. However, this is not likely as an identical internal consistency (Cronbach's alpha 0.91) was also registered for the Italian version of the ASKp on children with diseases other than CP. In a well-conducted master's thesis [Cerisola & Fioravanti, 2014], the homogeneity of the ASKp was tested on a sample of 260 children (56% female) aged 5 to 15 years with musculoskeletal disability resulting from the following diseases: a) congenital or acquired orthopedic diseases (87%), including dysmorphisms, scoliosis, and fractures; b) neuromuscular diseases (7%), such as peripheral neuropathies and spina bifida; and c) rheumatological diseases (6%), such as juvenile idiopathic arthritis or scleroderma. Therefore, this small difference is more likely to be peculiar to the Italian context of application, indicating slight cultural differences in the interpretation of the construct assessed; the need to reverify the homogeneity of an assessment tool when applied in different cultural contexts is thus justified.

As regards validity, it must be remembered that it is a characteristic related to the specific construct to be assessed and that it can change in relation to the target population. Given that the ASKp is often used to assess physical disability in children with CP [Morris, 2005; Harvey, 2008; Capio, 2010; www.activitiesscaleforkids.com] and that its validity has never been quantified specifically in this population, this research project has begun to fill this gap. Once the content validity of the ASKp was verified [Fabbri, 2016], as there is no tool that can be considered the gold standard against which the validity of the ASKp can be estimated, the validation process continued through the verification of three a priori hypotheses. Altogether, these hypotheses support the theory that the ASKp is a valid measure of physical disability in children with CP.

The first hypothesis was that the ASKp would be able to discriminate between healthy children and children with CP. This hypothesis was verified, as the ASKp scored 89.06 (±10.57) in healthy children and 75.5 (±18.8) in children with CP, rejecting the hypothesis that the two samples were extracted from the same population (p<0.001). It is noteworthy that the results obtained in the healthy sample were consistent with those of Plint and colleagues [Plint, 2003], which proved the ability of the ASKp to distinguish between healthy children and children with musculoskeletal disabilities of various etiology. Moreover, the results obtained in the healthy Italian sample showed the ability of the ASKp to detect higher levels of activity performance in adolescents compared to those in younger children. This is coherent with the longitudinal trajectories of reference values for activities tracked in typically developing individuals, which reveal how children and adolescents reach 90% of their maximum performance level of self-care, community, and domestic activities at about 7, 15, and 18 years of age, respectively [Sparrow, 1984]. The comparison with reference values also explains the ceiling effect of the ASKp in adolescents, as self-care activities are the first to be fully performed by children and are also more highly represented in the ASKp than are domestic ones. Not only, the ceiling effect was also confirmed in children with CP; again, and in line with other investigations, the ASKp detected higher levels of activity performance as age increased [van Gorp, 2018]. In the absence of intellectual deficit, the longitudinal trajectories of reference values for activities are only slightly delayed in children with CP compared to the values of typically developing children, and the maximum performance level is reached for several activities in adolescence [Vos, 2013], whereas motor capacity plateaued at a younger age [Smits, 2013].

The second hypothesis was that the ASKp would discriminate between children with CP with different levels of disability. This hypothesis was verified by several comparisons since, on average, the ASKp score differed in children with and without mild intellectual deficit, in children with and without the need for assistive devices for indoor mobility, in children with unilateral or bilateral manifestation of CP, and in children with clinically meaningful differences in motor function, i.e., those classified as Level III of the GMFCS [Palisano, 1997] compared to both Levels II and I. As expected, and in accordance with previous studies, children with CP and intellectual deficit showed lower levels of activity performance compared to those without this deficit [Vos, 2013; van Gorp, 2018]. However, this research project also showed that this difference was already evident for mild levels of intellectual deficit, i.e., children who were still capable of self-assessment, reporting a further disadvantage compared to their peers with CP. In my opinion, this data point should be investigated further, with the aim to understand how much of this further disadvantage could be mitigated by compensatory measures and by exploiting the resources of the child, the family, and the social context. The ability of the ASKp to distinguish between children with different levels of gross motor function and with different needs for assistive devices for mobility is easily explained and confirms the hypotheses underlying the theoretical construct. Of note, in 2007, Palisano and colleagues classified a cohort of adolescents with CP on the basis of their motor function, assessed their activity performance using a modified 38-item version of the ASKp, and tested the hypothesis that performance would differ based on the GMFCS Level. Their results confirmed the discriminatory capacity of this modified ASKp version [Palisano, 2007]. However, other fundamental psychometric properties of the tool have never been examined before. Finally, the ability to distinguish between children with different topographical manifestations of CP merits reflection; our results showed a major impact of bilateral CP on activity performance, but it is also well known that performance in ADL can be strongly affected in children with unilateral CP [James, 2015]. However, few of the items of the ASKp investigate activities that require fine and bimanual motor skills, while several items investigate motor abilities related to mobility or standing skills, which are definitely more affected in bilateral than in unilateral CP [Gorter, 2004].

The third hypothesis was that the ASKp score in children with CP would be convergent with the score of the Gross Motor Function Measure-66 (GMFM-66), a valid measure of gross motor function in CP [Russell, 1989], a construct strictly linked to physical disability. A moderate positive correlation was found between those measurements, confirming the hypothesis. It must be noted that this correlation was weaker than that between the ASKp and a subset of items of the GMFM-66 [Bjorson, 2013] (r = 0.54 versus r = 0.83). Possible explanations for this difference in the strength of the correlation might be that, in this research project, children with CP were classified as Levels I-III of the GMFCS and were consistently assessed with dimensions D and E of the GMFM, whereas Bjorson and colleagues included children on the whole spectrum of GMFCS and used subsets of items that were selected from all five dimensions of the GMFM. Therefore, the two studies actually investigated slightly different samples using subsets of items that might have differed substantially. This is a possible explanation, but further evidence is required to more fully understand the strength of this correlation. Finally, to verify the third a priori hypothesis, we also looked at any convergence between the ASKp and the GMFCS scores. As expected, a moderate negative correlation was found. Again, although this correlation verified the hypothesis, it was weaker than that previously reported in the literature between the same variables [Morris, 2006] (r = -0.54 versus r = -0.90). Again, Morris and colleagues' sample had an overall higher level of disability (GMFCS Levels I-V) than did ours. However, this alone is unlikely to explain such a different strength of correlation. Pending any evidence that may clarify this phenomenon, the moderate strength of both the ASKp - GMFM and the ASKp – GMFCS correlations we registered might also be due to the fact that, as already argued, neither the GMFM-66 nor the GMFCS can fully describe a child's habitual behavior, despite their both being tools of undisputed value in quantifying motor function and classifying children in clinically meaningful categories on this basis [Gorter, 2004]. In fact, besides the hallmark movement and posture disorders of CP, sensation, perception, cognition, communication, and behavior disturbances and epilepsy also affect activity performance, the degree of limitation being in relation to the level of motor disability [Bartlett, 2019]. Therefore, a correlation that verifies the convergence of moderate strength between these tools might be entirely plausible; a correlation between the GMFM-88 and the LAQ-CP was found whose strength ($r = -0.52$, $p < 0.001$) [Kerr, 2007] was very similar to that in this research project. The LAQ-CP is a measure of participation restrictions in children with CP which has also been found to strongly correlate with the ASKp ($r = -0.96$) [Morris, 2006]. This further evidence of convergent validity confirms the dynamic association between motor function and the *activity and participation* domain of the ICF, which is mediated by personal and environmental factors.

Last but not least, only 7% of children with CP delivered incomplete ASKp (< 23 answers), demonstrating a high acceptability of this questionnaire.

Implications for future studies

The cross-cultural validation process of scales requires that the psychometric properties of the translated and culturally adapted instrument be subjected to verification of its reliability and validity properties [Sousa & Rojjanasrirat, 2011]. As already stated, validation is a process that takes place through several steps: with regard to reliability, we had not yet verified the stability of the ASKp when we administered it to Italian children with CP. This verification is currently underway through the Bachelor's thesis of a physiotherapy student at the University of Modena and Reggio Emilia (IT). The hypothesis currently being tested in that thesis is that the ASKp has sufficient stability between two subsequent administrations, which occur at a proper time interval. Of particular importance will be the data provided by the subgroup of patients with mild intellectual deficit to understanding how stable the results obtained through self-administration of the ASKp in this group are. In fact, as intellectual impairment is a common comorbidity affecting nearly 50% of children with CP [Novak, 2012], in this Ph.D. research project we chose to include children with mild intellectual disability in the sample of individuals to help us to validate the Italian ASKp. The results obtained are in line with the theoretical construct that we wanted to verify. However, it is not known whether patient-reported outcome measures such as the ASKp can be used reliably with children with mild intellectual deficit. For this reason, these children may be at risk of exclusion from routine patient monitoring, research, and quality improvement schemes, thus increasing potential health inequalities [Jahagirdar, 2012]. Therefore, future studies should verify the acceptability of the ASKp self-administration and reproducibility of its results in this specific population. Further, the degree of concordance between the answers given by children with intellectual deficit and those of their parents should also be verified, bearing in mind that incomplete agreement would not necessarily mean poor instrument reliability, as children and parents may have different perceptions regarding the construct investigated.

With regard to validity, the absence of a gold standard that could act as a criterion prevents us from verifying this type of property, which, for feasibility reasons, was not verified even in the validation process of the original ASK; it was tested only for the capability version [Young, 2000]. A possible criterion to be used should be sought in the evaluation made by teachers, family members, or healthcare professionals who habitually observe the child's performance in the context of life without intervening in any way. However, this type of study would likely pose further sources of bias and, considering that several proofs of construct validity have already been provided by this research project, we believe that healthcare professionals can already use the ASKp with confidence. Yet we still do not know whether the Italian version of the ASKp is responsive to changes in the population of children with CP. This is certainly an aspect to be investigated in future studies, which makes sense only now that we have confirmation of the fundamental psychometric properties of the Italian ASKp.

Finally, the ASKp is validated for children and adolescents from 5 to 15 years of age. While development of motor skills is usually completed by adolescence performance can still progress as it depends strongly on personal and environmental factors [van Gorp, 2018]. Moreover, if we can expect the development of self-care activities to be completed in adolescence, other domestic or community activities continue to develop for years [van Gorp, 2018]. The ASKp investigates only a few of these domestic and community activities, which should be investigated in a more extensive way over a long period time, because complete social inclusion, the ultimate goal of the rehabilitation process, can be critical during and beyond adolescence [Wintels, 2018].

Limitations and strengths of this Ph.D. research project

The results of this research project must be interpreted in the light of several limitations.

First, the children and adolescents included, both healthy and with CP, were not completely representative of their reference population. Healthy children were recruited in a limited area of only one midsize, highly industrialized Italian city. As performance partly depends on the sociocultural and environmental context, a sample of a more heterogeneous origin would have allowed a greater generalization of the results. The children with CP, instead, were recruited in a highly specialized referral center for children from all over Italy. However, we chose not to include children with higher levels of functional limitation (GMFCS Levels IV and V) because we did not want to cause them any unnecessary frustration that might have arisen during the self-administration of the questionnaire, thereby inadvertently highlighting the several limitations they suffer. Plus, in the sample selected, the proportion of children classified as Level I and those as Level II of GMFCS was slightly different from that of the target population of children with CP (sample: Level I 37%, Level II 45%; target population: Level I 48%, Level II 36%) [Reid, 2011].

Moreover, an irreconcilable limitation of this study design is that children with more severe intellectual deficit were completely unrepresented because they could not reliably complete the ASKp. Further, the sample of children with CP for whom the GMFM-66 and the GMFCS were both available included only 60 individuals, with only 11 categorized as Level III of GMFCS. This sample could not therefore adequately represent the reference population.

Finally, the absence of a gold standard for the measurement of activity performance prevented us from validating the ASKp against a widely accepted criterion.

In the absence of that, we preferred to give priority to the verification of assumptions underlying a solid theoretical construct, which is a strength of this research project, as is the large size of the samples of healthy children and of children with CP on whom the ASKp was tested. Worthy of note, the results of this research project are consistent with those of previous studies; the small differences found concern the strength of the results obtained but they never denied the hypotheses tested. Another strength of this research project is that it described the distribution of the ASKp score in a wide sample of healthy children. This step is often overlooked in cross-cultural validation processes. However, it is always important to know the distribution of scores of a self-administered questionnaire in the healthy population to provide our patients with the most useful information. However, the greatest strength of this study lies in the usefulness of this measure, which was chosen among those available by clinicians of two highly specialized centers [Fabbri, 2016] to quantify the activity performance of children with CP. Since, as previously argued, there are relatively few culturally adapted and validated outcome measures in contexts other than the one of origin, and since it is necessary to have tools that allow for valid assessments and international comparisons, I believe that the results of this work may be of help to both clinicians and researchers involved in CP rehabilitation.

Conclusion

This Ph.D. research project continued the validation process of the culturally adapted Italian ASKp, focusing on the population of children with CP. The Italian ASKp corresponds to the original in terms of language and content. In Italian children and adolescents with CP, it had good reliability and acceptable values of validity to measure physical disability applied to the construct of activity performance. Moreover, it was considered clear and acceptable to a pediatric population. The results of this Ph.D. research project represent a small but clinically relevant contribution to Italian rehabilitation professionals, who do not always have a sufficient range of validated measures available for their clinical practice and research. Worthy of note, in the care of patients with chronic and complex diseases such as CP, it is important to assess all the ICF domains. As motor impairment is a hallmark of CP, reliable and valid motor capacity measurements have been developed and spread over the years. However, the carryover of motor skills into life situations also depends on personal and environmental factors and is not always easy to observe and measure. For these reasons it is important to develop outcome measures addressing the *activity and participation* domain and targeting the population with CP.

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APPENDICES

APPENDIX I

Transcultural validation of Activities Scale for Kids (ASK): translation and pilot test

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ABSTRACT

Background and aim of the work: The Activities Scale for Kids performance (ASKp) is one of the few self-assessment questionnaires in pediatric rehabilitation that measures child perception in performance of daily routine activities. ASKp is composed of 30 questions designed to explore activities and participation in children and teenagers with musculoskeletal disorders. Scores assess level of physical ability, identify appropriate treatment and monitor changes over time. We undertook this cross-cultural validation study to achieve a culturally adapted Italian version of ASKp, to be tested on Italian children.

Methods: ASKp was converted into Italian by two independent certified translators, supported by two Italian research physiotherapists. Twelve children with musculoskeletal disorders, their parents and eleven expert pediatric physiotherapists independently assessed clarity of language and semantics of each item. Physiotherapists also evaluated content validity of each item.

Results: One item out of 30 did not reach inter-rater agreement ≥80% for clarity and was modified according to suggestions given. A panel of three research physiotherapists improved clarity of some other items based on suggestions from study participants. Regarding content validity of ASKp, ICVI was ≥0.59 for all items and overall ASKp was deemed valid (S-CVI/Ave 0.93).

Conclusions: The Italian version of ASKp is linguistically clear and culturally coherent with a high content validity. It can be extremely useful in assessing activity limitation perspectives in Italian children and adolescent ranging from five to 15 affected by musculoskeletal disorders.

Key words: cross-cultural comparison, disability evaluation, self-assessment, child, adolescent

Background

The International Classification of Functioning Disability and Health defines activities as execution of specific tasks by individuals. During development, these tasks include walking, playing or accomplishment to basic activities or age-related chores [1]. The prevalence of limitation in activities among Italian children is not clearly established, but can be estimated approximately at 4,6% [2]. Hence, measurement of these phenomena is crucial to quantify limitations of activity dimension and to quantify results of interventions. Measurement must rely on robust assessment tools and procedures [3].

In order to be valid and useful in a clinical setting, assessment tools must possess some basic psychometric requirements, such as reliability, validity, sensitivity to change (responsiveness) and clinical relevance. Furthermore, a very important aspect to consider when choosing an assessment tool is its validation in a given cultural context. In fact, assessment tools are usually developed and validated for specific contexts, but their use is often extended to areas that differ greatly from their original cultural, linguistic and ethnic background. These differences imply the need for instruments which are linguistically, culturally and semantically adapted to target populations. This adaptation process provides, in addition to language translation, a review and cross-cultural validation that tailors the instrument to the context in which it will be applied, without changing its original measurement purpose [4].

According to research recently conducted by the Italian Society of Physiotherapy, only 48 out of a total of 237 rehabilitation rating scales used both for children and adults have been formally validated in Italian (www.sif-fisioterapia.it).

In the field of pediatric rehabilitation, several rating scales have been developed, most of which have not been formally translated and validated in Italian. One of these is the Activities Scale for Kids (ASK), developed in Canada in the 90s by NL Young [5].

ASK is a child self-report measure of physical disability. It is designed for children and teenagers from five to 15 years, who experience limitations in daily routine activities due to musculoskeletal disorders. This measure, unique among others, had been generated based on interviews with numerous children and their parents, supplemented by the recommendations from expert clinicians and a review of the literature. Phrases used by children to describe their disability during the interviews were used to formulate the questions. ASK may be used to assess child status at a single point in time or monitor changes over time or be associated with therapeutic intervention outcomes (www.activitiesscaleforkids.com).

This assessment tool consists of an initial instruction followed by 30 multiply-choice questions (items) (Table 1, column A). The 30 items, divided into seven sub-domains, are: personal care (3 items) such as "I put toothpaste on my toothbrush then brushed my teeth by myself", dressing (4 items) such as "I put my shirt on by myself", other skills (4 items) such as "I made a snack (or prepared breakfast or lunch) by myself", locomotion (7 items) such as "I got around inside my home without anyone to help me", play (2 items) such as "I played sports by myself or with a few friends", standing skills (5 items) such as "I got through heavy doors by myself" and transfers (5 items) such as "I got down onto the floor from standing, and got back up again by myself". These sub-domains may be useful for exploring the nature of activity limitations, but have not been independently validated. All the items are aggregated into one summary score.

ASK also includes three questions that investigate aid devices used by children and degree of assistance required in performing activities. Multiple-choice answers are given for each question.

There are two versions of the scale: ASK performance (ASKp) and ASK capability (ASKc). The performance version measures what the child 'did do', whereas the capability version measures what the child 'could do' during the previous week. So, ASKp is suitable to measure what children usually do in their habitual context, whereas ASKc measures what children can do in a defined situation, apart from real life [6]. Clinicians may choose to administer either version alone or concurrently, depending on assessment objectives.

To date, ASK is one of the few questionnaires which measures child perspectives on disability and limitation, provides an option of examining performance and/or capability and requires no special training or equipment [5,6].

The original English version of ASK has been tested for its validity through appropriate studies [5,7,8], showing excellent reliability [7] and good content, concurrent, construct and discriminative validity [7,9] even when administered via web or by mail [10,11]. Two recent systematic reviews confirmed the high reliability and validity of ASKp in children ranging from five to 15 [12,13]. Moreover, compared to other physical activity rating scales, ASK showed robust psychometric properties in most areas [13] and ASKp was judged more reliable and valid when applied in field-based studies in children with physical disabilities [12].

A clear advantage of using ASKp in clinical practice is that subjective judgments by young patients on impact that disabilities or limitations have on daily life can be assessed directly, and not mediated by a third party [5,13]. Integration of this data is of paramount importance in rehabilitation planning, because it directs treatment to the real needs of children.

For this reason, rehabilitation professionals frequently use this scale in clinical practice and research [12,13]. This is also the case in Italy, despite the fact that ASKp has never been previously validated in Italian.

Thus, even if further studies are suggested [14], we choose to formally validate ASKp to verify its psychometric properties and make possible its use in the Italian context.

Aims

This study provides the basis for validation of the Italian version of ASKp, in order to respond to the need of having cross-culturally validated tools that contribute to the development of evidence-based pediatric rehabilitation.

The aim of this study is to attain a culturally and linguistically adapted Italian version of ASKp, which will subsequently be tested for its comprehensive psychometric characteristics.

Methods

Before starting the study, we individually consulted a convenience sample composed of twelve rehabilitation professionals (physiotherapists and physicians) who worked in two Italian child rehabilitation centers (Unit for Severe Disability of the Developmental Age – Hospital Santa Maria Nuova of Reggio Emilia and Pediatric Hospital Meyer of Florence) and routinely used ASK in their clinical practice. When asked for their opinion about the usefulness of ASK, they affirmed that children can provide more reliable information if they are asked to report on activities they actually carry out (performance, assessed by ASKp), rather than on activities they think they would be capable of doing (capability, assessed by ASKc). They also stated they frequently use the ASKp to measure physical disability in their practice because, in their habitual population of chronically disabled children, the intent of treatment is primarily to improve the quality of existing functions. Consequently, a direct assessment of community function and limitations of direct relevance to patients is needed. Therefore, they suggested that validation of ASKp would be clinically meaningful.

Participants

In order to carry out this study we selected the following convenience samples:

- 12 children and teenagers with musculoskeletal disorders, similar to the sample of the original validation study [4], referred to the Hospital Meyer of Florence from January 2013 to March 2013 for rehabilitation;
- one parent for each child/teenager included in the study;
- 11 physiotherapists in pediatric field, working or attending as a consultant the Unit of Child Rehabilitation at the Hospital Meyer of Florence from January 2013 to March 2013;
- two certified translators.

Inclusion criteria for children and teenagers were the following:

- five to 15 years old;
- musculoskeletal disorders (neuromuscular diseases, rheumatic diseases, fractures, diseases of the peripheral nervous system, spina bifida);

- native Italian speaker;
- who had at least one Italian parent.

We excluded children and teenagers suffering from cognitive impairment, based on data reported in medical record.

Inclusion criteria for physiotherapists were the following:

- professional experience of at least five years in pediatric physical therapy;
- native Italian speaker.

To be included in the study, parents had to be Italian and native Italian speakers, whereas the two certified translators had to be experts in medical terminology.

Study design

This transcultural validation study received a favorable opinion from the Ethics Committee of the Hospital Meyer of Florence. We also got the permission to use the original ASKp for research purposes from its Author. We undertook the following steps to obtain cross-cultural validation of the Italian version of ASKp:

a) translation of assessment tool from original language to target language by a certified translator; back-translation of the tool from target language to original language by a certified translator, who worked independently; comparison of the two versions in the original language (the former validated one in English and the latter back-translation) and resolution of any discrepancies by way of agreement among translators and two Italian research physiotherapists, skilled in pediatrics and authors of the study;

b) assessment of ASKp for linguistic and semantic clarity of each component of the tool in target language, namely instructions, 30 items concerning activities, three questions regarding aid devices and degree of assistance required by child and answer options. This assessment was carried out independently by participants (children/teenagers, parents and physiotherapists) using a dichotomous scale. Study participants could also provide suggestions to improve clarity of text;

- assessment of ASKp for relevance of questions and answers in target language. This assessment was carried out independently by the physiotherapists who, for this purpose, used a score scale of 1 to 4, where 1= not relevant; 2= little relevant; 3= quite relevant; 4= very relevant [15];

c) examination of suggestions provided by study participants by a panel composed of three research physiotherapists (skilled in pediatrics and authors of the study) and consensus regarding changes to be made to the Italian version of ASKp, through formal consent.

Data analysis

Data collected were statistically processed to determine the following measures:

- linguistic and semantic clarity of the instrument, by calculating percentage of analyzed components judged unambiguous and explicitly clear. Level of inter-rater agreement had to be at least 80% [4];
- degree of relevance assigned to each question of the instrument, by calculating the Item-Content Validity Index (I-CVI). I-CVI represents the proportion of experts who attribute a score of 3 or 4 to each analyzed item. For the purpose of this study, critical value for I-CVI is 0.59. This value is due to the number of judges employed in validation process [16,17,18];
- degree of global relevance of the instrument, by calculating the Scale-Content Validity Index/Average (S-CVI/Ave), which represents the average value of individual I-CVI. A degree of global relevance ≥0.80 is considered good and values ≥0.90 as excellent [17];
- degree of universal agreement on relevance, by calculating the Content Validity Index Universal Agreement (CVI-UA). CVI-UA is the proportion of items that receive a relevancy score of 3 or 4 by judges, with respect to the total number of items of the instrument. In literature, there is no clear agreement on the critical value to interpret this parameter, which is contingent on the number of judges involved in the validation process [17].

Results

During a routine session, a physiotherapist explained the purpose of the study to eligible children and parents and asked for consent to participate.

Thus, the Italian version of ASKp was then administered to twelve enrolled children and one of their parents of Italian nationality, who were native speakers (Table 1, column B) [insert Table 1 about here]. Of the twelve children, eight were female and four were male, with a mean age of 11 years (min.6 – max.15). The participating children had the following diseases: Duchenne muscular dystrophy (n.2), rheumatoid arthritis (n.2), functional limitations resulting from fractures (n.2), spina bifida (n.4), hereditary sensory motor neuropathy (disease of Charcot-Marie-Tooth) (n.1) and obstetric brachial plexus injury (n.1). Of the twelve parents, nine were mothers and three were fathers with a mean age of 39 years (min. 25 – max 56).

The Italian version of ASKp was given to both children and parents independently, asking them to indicate the linguistic and semantic clarity of each component of the instrument. Children and parents could provide suggestions for improving clarity of the final Italian version.

In parallel, eleven physiotherapists eligible for the study were contacted and asked to contribute to the study at the Hospital Meyer of Florence (Table 2). So, the Italian version of ASKp was distributed to physiotherapists experienced in pediatric area who worked in the Unit of Child Rehabilitation at the Hospital Meyer of Florence (n.8), or at the Local Health Units of Ravenna and Imola (n.3). At the time of this study, these physiotherapists were attending Hospital Meyer as consultants. The physiotherapists included in the study were all female, mean age 38 (min.32 – max.51), with an average of 8 years' experience in pediatrics (SD ± 3.2). Of the eleven physiotherapists in the study, nine have completed or were attending a post-graduate program in Pediatric Physiotherapy.

The physiotherapists were asked to indicate linguistic and conceptual clarity of each component of the Italian version of ASKp, to provide suggestions to improve comprehensibility of text and to assess relevance of each question and answer options.

Data regarding clarity of assessment tool showed that:

1) 30 multiply-choice questions were judged clearly understandable by all children and physiotherapists, with an overall inter-assessor agreement ≥80%.

In particular, children reported a complete inter-assessor agreement for 25 questions, an inter-assessor agreement equal to 92% for three questions (n.12, n.13, n.25) and an inter-assessor agreement equal to 83% for the remaining two questions (n.8, n.14) (Figure 1) [insert Figure 1 about here].

Expert physiotherapists reported complete agreement for twenty questions, an inter-assessor agreement equal to 91% for seven questions (n.7, n.10, n.12, n.15, n.16, n.17, n.18) and an inter-assessor agreement equal to 82% for the remaining three questions (n.8, n.9, n.23) (Figure 2) [insert Figure 2 about here].

According to the parents, 29 questions were judged clearly understandable, registering an inter-assessor agreement ≥80%. Among these, 21 questions obtained complete agreement, six questions obtained an inter-assessor agreement equal to 92% (n.7, n.16, n.23, n.25, n.27, n.28) and the remaining two questions obtained an inter-assessor agreement equal to 83% (n.8, n.12) (Figure 3) [insert Figure 3 about here]. Question n.13, which states, “I got around my home without anyone helping me” reached an inter-assessor agreement of only 75%. Parents deemed the Italian translation unclear because the Italian word “*mosso*” translated from the English “got around” was judged to be ambiguous. They suggested replacing it with the synonym “*spostato*”.

2) the instructions, the three questions investigating use of aid devices and the degree of assistance for activities and the answer options reached an inter-assessor agreement ≥80% by both children, parents and physiotherapists participating in the study.

Hence, the Italian version of ASKp was judged overall understandable and clear in terms of semantic and language for all interviewed participants.

Even if they had been judged clear, some participants recommended improving comprehensibility of the following questions:

- question n.7 “I put my shoes on and did them up by myself” was judged clear by 92% of the parents and 91% of the physiotherapists. However, four physiotherapists and one parent suggested a structural change to the question because the two activities listed, being very different, should be treated as two distinct items. However, we decided not to integrate this suggestion because it would introduce a structural change with respect to the original valid ASKp.

- question n.8 “I took care of my medical needs – Examples: put on splints or took medication” was judged clear by 83% of both children and parents and by 82% of physiotherapists. It is interesting to note that among the ten children who judged this question clear, four reported that they understood it by only after reading the example. Moreover, five physiotherapists reported that the example is of fundamental importance for understanding the meaning of the question. Two children judged the Italian translation of this question unclear and reported that the Italian word "*curato*", translated from the English “took care”, is difficult to understand. Some parents and physiotherapists suggested replacing it with a synonym (*preso cura*), which was deemed too broad and vague by the translators. Therefore, we decided not to act on this suggestion because we believed it could lead to a misinterpretation of the question.

- question n. 9 “I did my printing (or script writing) by myself” was declared clear by 82% of the physiotherapists. One physiotherapist who judged the question unclear, suggested changing its content to “I did my homework alone”, while the other reported that the two activities, printing and writing, are different from each other, but did not suggest any changes. We did not modify this item because all children and parents, to whom it is addressed the ASKp, judged it clear.

- question n. 12 “I did my usual job or chores – Examples: paper route, babysitting, or doing the dishes” was judged clear by 83% of both children and parents. One child who judged the question unclear reported that he did not understand the examples while another reported that he was not allowed to babysit. Parents who judged the question as unclear, pointed out that in Italy it is not usual for children to perform chores reported in the examples. They advised replacing them with activities more suited to Italian culture, such as “I've tidied my room, I set the table or I washed the dishes”. Thus, we decided to change the examples as suggested.

- question n. 23 “I got down onto the floor from standing, and got back up again by myself” was judged clear by nine physiotherapists (82%). Two physiotherapists, who judged the question unclear, reported that the technical Italian translation for the English “from standing” (*dalla posizione eretta*) may be difficult to understand, especially for younger children, and suggested changing it to a more common lay term (*da in piedi*). Three out of nine physiotherapists, who judged the question clear, suggested the same change. So, we adapted the item as suggested since, even though in Italian both these terms mean standing position, only the latter is used in everyday communication.

Also, as advocated, we changed the original Anglo-Saxon names showed in the instructions with Italian ones.

Evaluation of relevance, or content validity, shows that the minimum value of I-CVI scored in four items was equal to 0.63, slightly higher than the critical value of 0.56 determined by Lawshe [16] on the basis of the number of judges. CVI-UA, i.e. the proportion of items deemed relevant by all judges with respect to the total number of items rated, was equal to 0.76 [17]. The value of S-CVI/Ave was 0.93, which is higher than the cut-off indicated by guidelines [4].

Finally, the panel of research physiotherapists analyzed suggestions provided by participants in this study and, through the process of formal consent, agreed on a culturally and linguistically adapted Italian version of ASKp (www.activitiesscaleforkids.com) which, in the near future, will be further tested for complete psychometric characteristics.

Discussion

The aims of this study were to cross-culturally adapt the ASKp questionnaire to Italian and to determine content validity of this version in young patients with musculoskeletal dysfunction. In pediatric rehabilitation, there is a lack of suitable and reliable assessment tools validated in the Italian language. Thus, professionals often rely on non-validated instruments, among which the ASKp, that has previously proven to be valid, reliable, responsive to change and showed minimal ceiling effects and no floor effects. Therefore, we consider appropriate to validate this tool to evaluate physical disability in the population of Italian children and adolescents.

ASKp for Italian patients was cross-culturally adapted to the Italian context following a systematic standardized approach [4]. This approach required a forward and back-translation, a review by experts and testing of the semi-final version to ensure that the meaning of original items regarding idiomatic expressions and content was maintained. Consequently, the final version of this assessment tool met all the original aims.

Overall, no difficulties were encountered in translating the questionnaire and the back-translation corresponded accurately to the original version. The ASKp reached an inter-assessor agreement ≥80% for clarity.

Only one item, n.13 “I got around inside my home without anyone to help me”, did not reach the cut-off requested for clarity because the first Italian translation for the English “got around” (*mosso*) was considered ambiguous and unclear in this context; hence, we agreed to replace it with a synonym (*spostato*). In Italian, both terms mean moved around, although they are used in different contexts.

All other components of ASKp were judged clear. However, we found some critical issues in the linguistic and semantic translation of some entries, which in Italian culture were uncommon or difficult to understand. Thus, in accordance with the translators, we decided to accept some participants’ suggestions and deemed clarity of the text improved (items n.12 and n.23).

As for the relevance, or validity of content, we highlight that all the components of the Italian version of ASKp exceeded the cut-off required by guidelines; in fact, percentages of agreement and both I-CVI and S-CVI/Ave exceeded critical values reported in literature. To our knowledge, the resulting value of CVI-UA reflects an overall high degree of content validity, taking into consideration that using eleven judges makes total agreement for this parameter very difficult to achieve.

This study represents the first step required to correctly extend the use the ASKp in the Italian clinical and research contexts. To our knowledge, there are no other validated Italian instruments specifically addressed to self-report the activities habitually carried out by children in their context of life. This is of great importance considering that the perspectives of children have been further overlooked in pediatric physical disability assessment. In fact, pediatric physical disability has most often been measured either by clinician observation in a clinical setting or by proxy report, even if previous research demonstrated that children are competent reporters of physical activities [5] and their opinion should be consequently considered the gold standard to measure disability. Furthermore, self-report is desirable to appropriately address the intervention, because abilities observed in clinical setting do not consistently reflects abilities performed in the community and in daily life. Concerns about child self-report might arise from beliefs that cognition is not completely developed in children. However, ASK questionnaire may be unique in that the items were worded specifically for children. Likewise, in this cross-cultural adaptation a large sample of children assessed the clarity of the Italian adaptation and prompted more appropriate words from their perspective.

The strengths of this study include standardized methods used for all procedures and a sufficiently large sample for a preliminary test. We interviewed three different population (children, parents and physiotherapists) to assess clarity of the instruments. Moreover, a wide variety of musculoskeletal conditions was included, similar to the validation process of the original assessment tool [7].

In the future, we recommend further investigation to document all the psychometric properties of the Italian version of ASKp and to determine its responsivity to clinical changes in children with musculoskeletal conditions similar to those involved in the development of the original assessment tool. Additionally, as ASK is widely used among patients with conditions different from the spectrum for which it was originally developed [11,12,13], we suggest to document measurement properties of the Italian version of ASKp also in children with Cerebral Palsy.

In conclusion, the Italian version of ASKp is a clear and culturally adapted tool addressed to young Italian patients and their parents. Furthermore, it has good content validity and is helpful in assessing and measuring activity limitation in children with musculoskeletal dysfunction.

Limits of the study

There are several potential limitations associated with this study. First, children and parents included were recruited from a single hospital and their cultural backgrounds may not fully reflect the wide-ranging Italian culture. Nevertheless, AOU Meyer is a reference center for the treatment of pediatric diseases for patients coming from all Italy. In addition, its central geographic position is in Florence, which is considered “the cradle of the Italian language” by linguistic scholars. Hence, we can say with a fair degree of confidence that the Italian version of ASKp can be easily understood throughout the Italian peninsula.

A second potential limitation is that our sample of health professionals included only physiotherapists but, in fact, this constraint reflects the current approach to physical activity assessment in Italy, which is predominantly performed by physiotherapists.

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Table 1: sample of items of the ASKp original version and Italian version

AREA	Column A: ORIGINAL VERSION	Column B: ITALIAN VERSION
PERSONAL CARE	I put toothpaste on my toothbrush then brushed my teeth by myself	Ho messo il dentifricio sullo spazzolino e mi sono lavato i denti da solo
	I used the toilet at home by myself	Ho usato il gabinetto di casa da solo
DRESSING	I put my shirt on by myself	Mi sono messo la maglietta da solo
	I fastened my clothes by myself	Mi sono chiuso i vestiti da solo
OTHER SKILLS	I took care of my medical needs	Mi sono curato da solo
	I made a snack (or prepared breakfast or lunch) by myself	Mi sono preparato uno spuntino da solo (oppure ho preparato la colazione o il pranzo)
LOCOMOTION	I got around inside my home without anyone to help me	Mi sono spostato in casa senza l’aiuto di nessuno
	I walked (or wheeled) in crowded areas	Ho camminato (oppure usato la sedia a rotelle) in luoghi affollati
	I got around outside without anyone to help me	Mi sono mosso all’aperto senza l’aiuto di nessuno
PLAY	I played sports by myself or with a few friends	Ho fatto sport da solo o con degli amici
STANDING SKILLS	I carried things in 2 hands by myself	Ho trasportato delle cose da solo usando 2 mani
	I got through heavy doors by myself	Ho aperto porte pesanti da solo

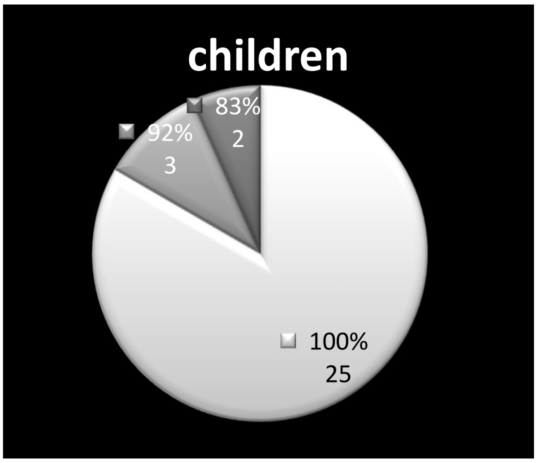
TRANSFERS	I got down onto the floor from standing, and got back up again by myself	Mi sono abbassato al pavimento da in piedi e poi mi sono rialzato da solo
	I got in and out an automobile by myself	Sono entrato e uscito da un’automobile da solo

Table 2: Characteristics of the sample

	n.	average age (range)	gender (F/M)	characteristics
Children	12	11 (6-15)	8/4	DMD (n.2), RA (n.2), functional limitations due to fractures (n.2), SB (n.4), C-M-T (n.1) OBPI (n.1)
Parents	12	39 (25-56)	9/3	n/a
Physiotherapists	11	38 (32-51)	11/0	experience in pediatrics 8 years (SD ± 3.2)

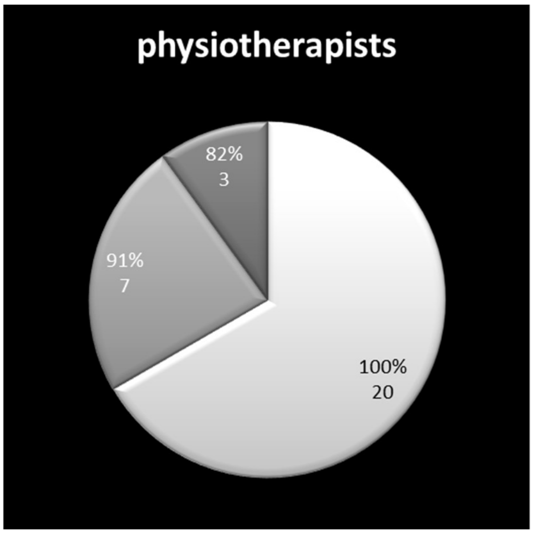
n.=number; F=female; M=male; DMD=Duchenne muscular dystrophy; RA=rheumatoid arthritis; SB=spina bifida, C-M-T=Charcot-Marie-Tooth; OBPI=obstetric brachial plexus injury; n/a=not applicable; SD=standard deviation.

Figure 1: Child report



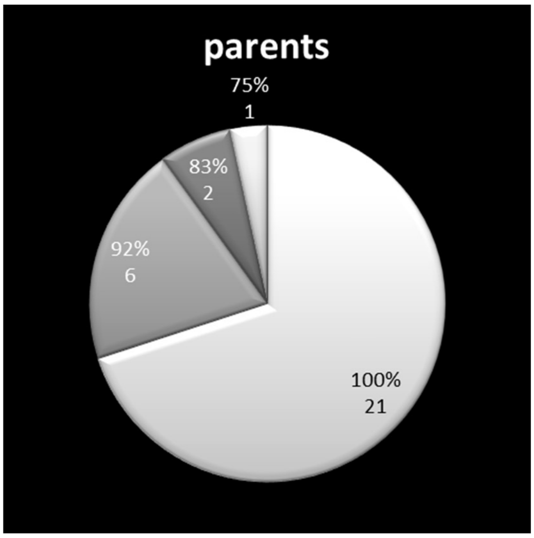
Level of inter-assessor agreement expressed by children

Figure 2: Physiotherapist report



Level of inter-assessor agreement expressed by physiotherapists

Figure 3: Parent report



Level of inter-assessor agreement expressed by parents

APPENDIX II

Activities Scale for Kids performance: stima dell'affidabilità nella popolazione di bambini italiani con Paralisi Cerebrale Infantile

Poster presentation

Costi S, Filippi MC, Beccani L, Cavuto S, Braglia L, Pelosin E. Presentato al XXXI Corso di aggiornamento "Asfissia perinatale ed encefalopatia ipossico-ischemica: prevenzione, diagnosi, terapia e riabilitazione", Modena, 20-22 Marzo 2019.

SERVIZIO SANITARIO REGIONALE
EMILIA-ROMAGNA
Azienda Unità Sanitaria Locale di Reggio Emilia
IRCCS Istituto in tecnologie avanzate e modelli assistenziali in oncologia

Activities Scale for Kids performance: stima dell'affidabilità nella popolazione di bambini italiani con Paralisi Cerebrale Infantile

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Background

La paralisi cerebrale infantile (PCI) è la più comune causa di disabilità dell'infanzia. I bambini con PCI sperimentano spesso difficoltà nelle attività della vita quotidiana, pertanto la misurazione delle loro performance in queste attività diviene un aspetto prioritario dell'intervento riabilitativo. La Activities Scale for Kids performance (ASKp) è una scala validata su bambini con disturbi muscoloscheletrici (Young, 2000), il cui utilizzo è diffuso anche nella popolazione con PCI (James, 2014), in quanto ha il vantaggio di cogliere la percezione del paziente rispetto alla propria abilità (Young, 1995). L'obiettivo di questo studio è ottenere una stima dell'affidabilità della versione italiana della scala ASKp (Fabbri, 2016) nella popolazione con PCI.

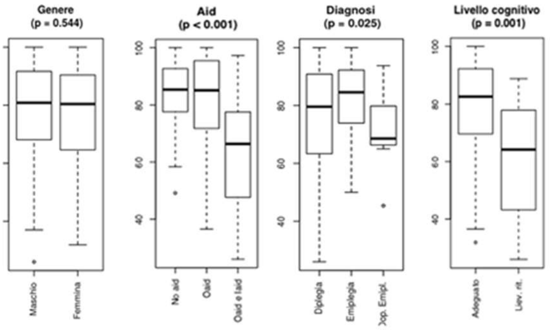
Materiali e metodi

Il disegno di studio cross-sectional prevede la somministrazione della scala ASKp ad un campione di bambini/adolescenti di età compresa fra i 5 e i 15 anni, con emiplegia o diplegia quale esito di PCI, buona comprensione della lingua italiana e assenza di deficit cognitivi di livello moderato o grave. L'elaborazione statistica effettuata sui dati raccolti consiste nell'analisi della coerenza interna della scala.

Risultati

Sono stati inclusi consecutivamente nello studio 205 bambini/adolescenti con diagnosi di PCI. Presentiamo i dati dell'analisi preliminare effettuata su i primi 139 partecipanti allo studio. Il campione incluso ha età media 10 anni e 6 mesi. E' rappresentato in maggioranza da maschi (58%) con diagnosi prevalente di diplegia (57%) e livello cognitivo adeguato (90%). Il 60% necessita di un ausilio per deambulare.

Il punteggio medio ottenuto nella ASKp è 77.14 (min 26.00-max 100.00). Il valore α di Cronbach è risultato pari a 0.91 (IC 0.89-0.93), dimostrando l'elevata coerenza interna della ASKp. La ASKp si è dimostrata inoltre in grado di distinguere sottogruppi con caratteristiche cliniche differenti: doppie emiplegie verso diplegie ed emiplegie (p=0.025), lieve ritardo mentale verso assenza di ritardo (p=0.001) e necessità di ausilio nella deambulazione indoor verso ausilio solo outdoor o nessun ausilio (p<0.001).



Conclusioni

La ASKp appare valida al suo interno quando applicata alla popolazione di bambini con PCI. Ad oggi, la ASKp è l'unica scala di autovalutazione delle abilità nelle ADL in ambito infantile validata in lingua italiana e può rappresentare un utile strumento di misura per la valutazione funzionale e la pianificazione dell'intervento riabilitativo in età evolutiva.

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APPENDIX III

Assessment of Physical Function in Children with Cerebral Palsy: data of Convergent Validity between the Activities Scale for Kids and the Gross Motor Function Measure

Poster presentation

Costi S, Alboresi S, Mecugni D, Ferrari A, Boggiani E, Pelosin E. Sarà presentato alla Conference and Scientific Event Annual Meeting "Health 4.0: Designing Tomorrow's Healthcare," Coimbra, 19-21 Marzo 2020.

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Assessment of Physical Function in Children with Cerebral Palsy: Convergent Validity between the Activities Scale for Kids and the Gross Motor Function Measure

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Introduction

Physical function is crucial to participation in life activities and must always be accurately assessed in children with cerebral palsy (CP)[1]. The Activities Scale for Kids performance (ASKp) is currently used for this purpose, but its psychometric properties have not yet been completely verified in children with CP[2], the principal cause of disability in industrialized countries[3]. Recently, the reliability of the Italian version of the scale was confirmed in this population and initial proof of validity was collected. However, there is no gold standard that can serve as a criterion. We verified the ASKp convergent validity with respect to the Gross Motor Function Measure-66 (GMFM-66), the gold standard in the assessment of gross motor abilities in this population

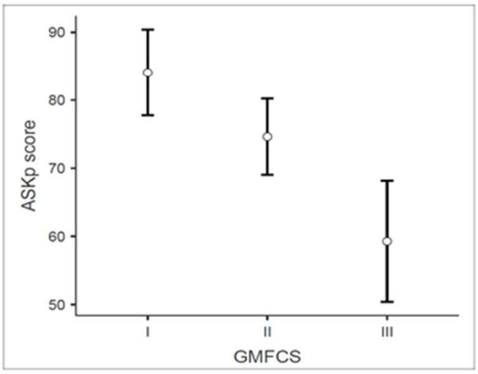
Methods

In this cross-sectional study we included 60 children with CP aged 5-15 years who were classified for functional capability using the GMFCS and assessed for severity of motor disability using the GMFM-66. Children self-administered the Italian version of the ASKp

Results

Sample characteristics	
M/F	31/29
Age (SD)	10.9 (3.0)
ASKp score (SD)	75.3 (17.5)
GMFM-66 score (SD)	73.3 (8.9)
Bilateral/Unilateral CP	42/18
Intellectual deficit Yes/No	8/52
Assistive devices Yes/No	42/18

The ASKp correlated moderately with both the GMFM-66 ($r = 0.577$; $p < 0.001$) and the GMFCS ($r = -0.541$; $p < 0.001$) and was able to discriminate between children with different levels of functional capability



Post hoc comparisons showed a statistically significant difference between the average ASKp score of both children in GMFCS Levels I and II compared to children in Level III ($p < 0.001$ and $p = 0.013$, respectively)

Conclusions

These results confirm the construct validity of the ASKp and contribute to the evidence that supports the ASKp as a valid tool to assess physical function, a crucial domain in the rehabilitation of children with CP

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APPENDIX IV

Occupational Therapy in Rehabilitation of Complex Patients: Protocol for a Superiority Randomized Controlled Trial Transcultural validation of Activities Scale for Kids (ASK): translation and pilot test

Preprint version of the manuscript published as

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Abstract:

Patient-centered occupational therapy is feasible in complex patients in the early phase of rehabilitation, and it contributes to meet needs in the domains of self-care, productivity, and leisure, promoting social role. We planned this single-center single-blind two-arm parallel individual patient randomized controlled trial, to verify the superiority of experimental occupational therapy (EOT) compared to standard rehabilitation (SR) in producing higher levels of patients' social participation assessed by the Reintegration to Normal Living Index. EOT is a patient-centered, hospital/home-based rehabilitation intervention based on the Canadian Model of Occupational Performance and Engagement. This appropriately powered study will provide evidence regarding the effectiveness of EOT on the recovery of social participation in the critical transition of complex patients from hospital to the home setting.

Implications for Rehabilitation

- Complex patients in the early rehabilitation phase manifest needs in the self-care domain, as well as in those of productivity and leisure areas, strictly related to their social role.
- Patient-centered occupational therapy interventions in complex patients are feasible and may improve performance and satisfaction related to meaningful activities.
- This RCT will assess the effectiveness of patient-centered, hospital/home-based experimental occupational therapy compared to standard rehabilitation care in producing higher levels of social participation in complex patients.

Trial registration: ClinicalTrials.gov Identifier: NCT03668938 (first posted date 13/09/2018)

Key Words: Social Participation, Occupational Therapy, Complex Patients, Canadian Model of Occupational Performance and Engagement, Randomized Controlled Trial.

Introduction

Better diagnostic work-up and improved therapies have enhanced life expectancy over the last few decades, but individuals affected by pathological events can survive for years with severe disabilities (Global Burden of Disease Study 2013 Collaborators, 2015) that cause restrictions in activities of daily living and social life.

In view of the increasingly advanced age and comorbidities of individuals, the implementation of biopsychosocial and patient-centered rehabilitation approaches focusing on individual health status rather than on diseases is recommended (World Health Organization. International Classification of Functioning, Disability and Health [WHO], 2001). Patient-centered care is a complex biopsychosocial approach that establishes a partnership between healthcare professionals, patients, and their families to ensure that the care delivered focuses on the patients' needs, values, and preferences (IOM, 2001). Although it is still difficult to quantify the benefits of patient-centered care on a patient's health status, the adoption of such an approach is particularly suggested in rehabilitation to guide careful ethical reasoning in particularly complex or challenging matters (Hunt & Ells, 2013). Multidisciplinary rehabilitation aims to recover functions and independence in daily living, thereby allowing individuals with limitations to return to their occupational role and to social participation; the final goal is to restore satisfactory quality of life.

Thus far, studies on the effects of occupational therapy have focused on populations with specific diseases (Kristensen, Persson, Nygren, Boll & Matzen, 2011; Phipps & Richardson, 2007; Simon Crompton, 2004); to the best of our knowledge, no study has been conducted to establish the effects of occupational therapy on the care needs of patients with disabilities due to a variety of events, regardless of the underlying pathology that caused hospitalization. However, unlike most medical specializations, rehabilitation deals with patients with transient or definitive disability due to a number of different pathological events. Thus, in accordance with the International Classification of Functioning, Disability and Health (ICF) aim, we conducted an observational study in 2014 to describe the occupational needs of complex inpatients who required high-level care (Schiavi et al., 2018). In accordance with Maslow's hierarchy (Maslow, 1943), the results of that study showed that, despite initial concern in basic functions, what patients cared about most, just a few weeks after an acute event, was recovering their social role. Consistent with other populations of patients (Harris & Eng, 2004; Müller, 2011), complex patients considered recovering occupations in productivity and leisure areas very important right from the hospitalization phase, and their aim was to achieve a good level of social participation as quickly as possible.

Based on the results of that observational study, in 2016 we conducted a pilot randomized controlled trial (RCT) to assess the feasibility and estimate the effect size of an experimental patient-centered occupational therapy intervention for complex patients in the early phase of rehabilitation, from admission to the ward until the first few weeks after discharge (Pellegrini et al., 2018). The experimental intervention proved to be feasible and resulted in clinically relevant improvements in the performance score of the Canadian Occupational Performance Measure (COPM) (Law et al.,2014), the primary outcome measure of that study, as well as in important secondary outcomes measures. Regarding these latter, the experimental group showed a clear trend towards gains in the return to normal living as assessed through the Reintegration to Normal Living Index (RNLI) (Wood-Dauphinee, Opzoomer, Williams, Marchand & Spitzer, 1988), although between-group differences did not reach statistical significance.

Rationale

Complex patients in the early phase of rehabilitation manifest needs in the domain of self-care, but also in that of productivity and of leisure areas, being as they are strictly related to the patient's social role. Our hypothesis is that patient-centered occupational therapy may bring about higher levels of social participation than standard rehabilitation care in complex patients. Since the feasibility of occupational therapy interventions in complex comorbid patients with impairments of the nervous or the musculoskeletal system has been ascertained (Pellegrini et al. 2018), it is time to conduct a clinical trial to investigate the effects of patient-centered occupational therapy on complex patients' needs.

Thus, we designed this RCT to answer the following question, "Is experimental patient-centered occupational therapy intervention (EOT) superior to the standard rehabilitation care (SR) in producing higher levels of social participation in complex patients referred to inpatient rehabilitation?"

Materials and Methods

Study design

A single-center 1:1 single-blind, individually randomized controlled trial with two parallel arms, powered for superiority, was designed in accordance with the SPIRIT statement. (Figure 1).

Ethics

This independent study has been approved by the local Ethics committee [n. 573/2018] and funded by the local health authority Azienda USL-IRCCS di Reggio Emilia, Italy. All participants will provide written informed consent.

Setting

This study is conducted at the Physical Medicine and Rehabilitation (PMR) Unit of the Arcispedale Santa Maria Nuova of Reggio Emilia from October 2018. This is a public hospital with a catchment area of about 535,000 inhabitants and the PMR ward performs an average of 350 admissions per year. Complex patients are about 250 of the annual admissions.

Participants

All adult patients admitted to the PMR ward are screened for complexity criteria, regardless of the diagnosis of access, using the Rehabilitation Complexity Scale Extended (RCS-E) (Turner-Stokes, Scott, Williams & Siegert, 2012; Rodà et al.,2015). The RCS-E assesses dependence in daily activities, special nursing care needs, daily medical monitoring, and the need for rehabilitation and aids. Patients with an RCS-E score ≥ 9 are deemed complex and are screened for eligibility by the multiprofessional rehabilitation team.

Exclusion criteria for this study are:

- individuals who are resident at nursing home facilities
- individuals for whom, upon admission to PMR, home discharge is not expected due to their clinical and social conditions.
- Mini-Mental State Examination <19
- clinical conditions that could prevent patient's participation in the EOT intervention (e.g., psychiatric disorders, severe heart failure, advanced cancer)
- communication barriers that do not allow a reliable evaluation and an active collaboration during treatment (e.g., severe aphasia, foreign individuals)

Eligible patients are referred to the occupational therapist (OT), who informs them in detail about the purposes and methods of the study. Patients who agree to participate are recruited into the study.

Details of the experimental intervention and active control

After recruitment and baseline assessments, patients are randomized to receive SR in the PMR ward or EOT, provided in addition to SR.

Standard rehabilitation

SR of complex patients is provided by a multiprofessional team composed of physicians, nurses, physiotherapists, speech therapists, psychologists, and social workers, and actively involves the patient and caregiver(s). It is performed in hospital and takes place twice a day – two hours in the morning and one hour in the afternoon – six days per week. Treatment aims are defined through team discussion of baseline assessments and the proposal of realistic goals to be achieved in the short-medium term. Regular team meetings allow for adapting the program to each patient's progress.

SR is task-oriented and centered on the recovery of independence in the basic activities of daily living (B-ADL) in order to obtain patients' discharge to their usual domicile.

Patients and their caregivers are trained in the self-management of the disease, its residual limitations, and the aids provided (wheelchair, etc.), and they and instructed in the prevention of secondary impairments (risk of falls, wounds, muscle shortening, etc.).

Patients are discharged from hospital when the following conditions have been met: clinical stability, safety in transfers (e.g., bed-chair, wheelchair-toilet), walking, and climbing stairs (if possible), provision of the prescribed aids at home. Close to discharge, brief return-home permits are proposed to allow patients to practice the B-ADL at home.

For individuals with permanent disability, environmental barriers at home are assessed on site to make the house accessible.

Upon discharge, patients are informed of their rights in case of disability as well as of the benefits of a healthy lifestyle to prevent further episodes of the underlying disease. Further, they are encouraged to take part in regular physical activity, even when a permanent disability is present (e.g., attending adapted physical activity programs) (Calugi et al., 2016; Desnoyers, Riesco, Fülöp & Pavic, 2016). If deemed necessary, an appropriate number of outpatient rehabilitation treatments (e.g., physiotherapy and/or speech therapy) can be provided.

Experimental patient-centered occupational therapy intervention (EOT)

EOT is personalized and provided by the OT in charge of the study. We adopted the Canadian Model of Occupational Performance and Engagement to implement a patient-centered experimental intervention (Townsend & Polatajko, 2007). Accordingly, we use the COPM to define the patients' priority and to orient the EOT intervention, adapting it to functional recovery and to the different settings in which the study takes place (hospital and home). Thus, the COPM is administered twice during the study: the first time at baseline, and a second time immediately before discharge (T1).

At baseline, the OT visits the places routinely frequented by the patient in his/her life for productive or leisure activities (community centers, associations, supermarkets, gyms, etc.) in order to detect opportunities and barriers to participation. This information is reported to the multiprofessional team meeting, providing rehabilitation intervention with individualized specific information collected in the context of everyday life.

Based on previous knowledge in this field (Schiavi et al., 2018; Pellegrini et al., 2018), EOT is applied according to the following sequential phases:
- the in-hospital phase, which aims to achieve the goals defined through the first COPM administration, mostly attributable to the domain of self-care but also to that of productivity and of leisure. Although personalized in frequency and duration, in this phase EOT is usually held daily and focuses both on issues strictly related to independence in the B-ADL (e.g., dressing, washing, breakfast) and on issues related to more advanced daily activities (e.g., cooking, cleaning, sending text messages, managing money, etc.).

- the home-based phase, which aims to achieve the new goals defined through the second COPM administration. The typical goals of this phase are related to advanced daily activities performed at home and in everyday environments (e.g., doing light housework, mobility in the community, shopping, return to usual activities). If deemed necessary, any residual objective in the domain of self-care can also be faced. The OT teaches strategies, recommends aids and provides personalized information regarding local social resources that can support patients and caregivers to perform the activity of interest to the best of their abilities. The frequency of the home-based EOT intervention is personalized and includes supervised sessions and repetitions of activities to be carried out autonomously by patients and their caregivers, at the frequency recommended by the OT. Any difficulty perceived by patients or caregivers during repetitions of activities is addressed in the subsequent supervised session in order to achieve the highest level of self-management of the residual limitation (Appendix – Homework).

To encourage patients' social participation and their return to everyday life, which is a clinically meaningful goal of rehabilitation and the primary aim of this study, the OT collects information about the networks and social support agencies available in the community that may be useful to most patients, and may even look for more specific ones in response to individual needs. The OT then encourages the patient and caregiver(s) to refer to the appropriate service(s) that can facilitate achieving that patient's goals in social participation. For example, patients might need assistance in the return-to-work process (e.g., UNAMANO project https://una-mano.webnode.it/), or peer support and encouragement in resuming activities of interest (e.g., InformaSalute Service http://biblioteca.asmn.re.it/informasalute) and, when possible, undertaking new, health-promoting activities.

Assessment points and study aims

Assessment points are presented in Table 1. Patients included in the study are assessed at baseline (T0), which takes place within 7 days from admission to the PMR ward, immediately before randomization. Afterwards, they are assessed within 3 days before discharge (T1), and at follow up (T2), 90 days (±15 days) after discharge.

The primary aim of this study is to investigate the effectiveness of the EOT on participation in everyday life of complex patients three months after discharge.

The primary outcome measure to assess the superiority of EOT over SR is the change (post-pre difference) in the RNLI score, measured at follow up and compared with that at discharge. This outcome is not collected at baseline as this construct can be reasonably quantified in the appropriate setting.

The secondary aims of this study are to investigate the effectiveness of EOT on the patient's perceived performance and satisfaction in carrying out his/her priority occupational activities, independence in activities of daily living and in instrumental activities, mood disturbances, and quality of life.

The secondary outcome measures are:

- changes (post-pre differences) in COPM performance score and satisfaction score. The COPM is administered twice during the study: a first COPM is administered at baseline and its scoring takes place at discharge; a second COPM is administered at discharge, and its scoring takes place at follow up. As already mentioned, this is to implement a patient-centered experimental intervention.

- change in the Modified Barthel Index (MBI) score (Shah, Cooper & Maas, 1992), change in the Instrumental Activities of Daily Living (IADL) score (Lawton & Brody, 1969), and change in the Hospital Anxiety and Depression Scale (HADS) score (Zigmond & Snaith, 1983). These outcomes are measured at each assessment point, and the three timeframes are used for analysis (T2-T0, T2-T1 and T1-T0).

- change (post-pre difference) in the Short-Form 12 (SF-12) score (Apolone et al., 2005), measured at follow up and compared to that at discharge. Similar to the RNLI, we do not collect this outcome at baseline as several items of the SF-12 pertain to activities and roles that are unlikely to be carried out in the hospital.

Additional measurements

In order to ensure a global view of the patient's health status, the simultaneous presence of multiple pathologies is assessed in all patients included in the study at baseline using the Charlson Comorbidity Index (Charlson, Szatrowski, Peterson & Gold, 1994).

Furthermore, data to confirm the feasibility of the EOT in the target population are collected. For this purpose, we consider recruitment and adherence rates. Recruitment rate is defined as the ratio of patients who agree to participate in the study to those who are eligible. Adherence rate, which is measured only in the experimental group, is the ratio of completed EOT sessions to the number of planned EOT sessions. As patient adherence to protocol is crucial in rehabilitation, this cutoff has been set at ≥ 75% (Williams et al., 2014; Hayden, van Tulder & Tomlinson, 2005). Adherence to the protocol is recorded by the OT in charge of the study, who lists the sessions carried out in the hospital; in the home-based setting, patients are requested to register the activities performed in the Homework log (Appendix - Homework), and data are reviewed weekly to increase completion rates (Frost et al., 2017).

Data on the length of hospital stay, the level of assistance needed at home, and the need for further outpatient rehabilitation treatments (e.g., physiotherapy and/or speech therapy) after hospital discharge will be collected in both groups.

Treatment-related side effects are collected throughout the study in both groups. We consider as minor side effects self-reported pain in joints or muscles involved in the activities; major side effects include any accidental injury occurring during rehabilitation activities which requires medical intervention.

Masking and blinding

In such rehabilitation intervention, blinding is not possible for patients and therapists. To limit bias, we ensure that the main outcome measure (RNLI) and some secondary measures (MBI and IADL) are collected by a blinded second OT, trained in the use of this tools.

The COPM is collected by the OT in charge of the study, because its administration entails the creation of a partnership between the patient and the healthcare professionals, the foundation for occupational therapy interventions. The HADS and the SF-12 are self-administered outcome measures.

Withdrawal from trial

Participation in the study will be withdrawn if any of the following occurs:

- patient discharged to a nursing home

- serious adverse events which result in death or require prolongation of hospitalization (e.g., fracture, stroke relapse, etc.) as well as a new medical judgment on suitability to continue in the study and a comprehensive clinical reassessment;

- patient lost to follow up.

All withdrawals will be recorded with specific reason.

Statistical consideration

Sample size

Based on the pilot study by Pellegrini et al. (2018), we estimate that the mean difference between the two groups in the T2-T1 interval for the main outcome measure of the study is 14 (sd = 21) points, which reflects a clinically relevant gain (Wood-Dauphinee et al., 1988). We expect a dropout rate of about 20% of patients who consent to the study due to the aforementioned reasons. Accordingly, assuming 5% type I error and 80% power for this study, we plan to enroll 92 patients (about 46 patients in each arm). The sample size calculation was made by the software nQuery Advisor 7.0, sheet MTT0.

Randomization procedure

After the baseline assessment, the OT telephones the local Clinical Trials and Statistics Unit, which generated the concealed allocation sequence; the responder simultaneously enters the patient's identity code and uses a predefined randomization list to make the assignment. Group allocation is revealed to healthcare professionals performing interventions and to patients after baseline evaluations are completed.

Statistical analysis

Demographic and clinical characteristics of the participants in the study will be presented with descriptive statistics.

Statistical analyses will be performed according to the intention-to-treat approach. Five percent significance will be used to assess the p-values and 95% two-sided confidence intervals will be provided for each tested parameter; the confidence intervals will be calculated assuming normal distribution for the related estimators.

Statistical techniques will compare the mean of the change between groups in the outcome measures as follows:

- t-test for independent samples to compare the mean of the changes (defined as T2 – T1) in RNLI and in SF-12.

- t-test for independent samples to compare the mean of the changes (defined as T2 – T0 and T1 – T0) in the HADS, MBI, and IADL.
- t-test for independent samples to compare the mean of the changes of scores obtained in the first COPM (T1 – T0) and in the second COPM (T2 – T1) administered.

For all the aforementioned statistics, heteroscedasticity will be checked by Levene's test and, if necessary, the Satterthwaite adjustment will be used. Furthermore, as a secondary analysis, the comparisons previously listed will also be adjusted for the baseline values.

Statistical calculations will be performed by the local Clinical Trials and Statistics Unit using SAS System release 9.2 or later, R release 3.3.3 or later, SPSS release 23 or later, according to the availability at the time of the data analysis.

Data regarding the treatment-related side effects, length of hospital stay, the level of assistance required and the need for further outpatient rehabilitation treatment after discharge, the adherence to the protocol, and the feasibility of the experimental intervention EOT are represented by descriptive statistics.

Although it has not been plainly demonstrated, non-adherence to protocols could in principle impact the outcomes of rehabilitation treatments (Hayden et al, 2005; Vermeire, Hearnshaw, Van Royen & Denekens, 2001; World Health Organization [WHO], 2003); thus, if patient adherence to the protocol proves to be less than 75% (Hayden et al., 2005), we will perform a secondary per-protocol analysis.

Duration and time frame

The study started enrolling in October 2018. As we expect the number of the population eligible for this study to be 5 patients/month, the total duration will be 24 months.

Discussion

The main aim of this RCT is to assess the effectiveness of personalized occupational therapy on top of standard rehabilitation on complex patients' level of social participation and return to everyday life. Previous evidence has confirmed that an evidence-based rehabilitation approach in this population should be complex, long-term, continuous, and tailored to the patient's needs (Woodman, Riazi, Pereira & Jones, 2014). Environmental factors must always be assessed and integrated into rehabilitation to break down barriers; also, all the social resources available in the family and in the community must be used (Jellema et al, 2016; Walsh, Galvin, Loughnane, Macey & Horgan, 2015). Furthermore, it has also been demonstrated that the need for social participation, namely productive and leisure activities, emerge early and increase progressively during the rehabilitation process (Schiavi et al. 2018). From an ICF perspective, social participation is considered a pivotal goal of every rehabilitative intervention (Noreau et al.,2004). However, this need is not always satisfied (Eriksson, Aasnes, Tistad, Guidetti & von Koch, 2012; Eriksson, Kottorp, Borg & Tham, 2009), which can lead to the risk of social isolation and lower level of life satisfaction (Eriksson et al., 2009; Bergström, Guidetti, Tham & Eriksson, 2017).

A pilot study showed that occupational therapy added to standard rehabilitation was feasible and safe in complex patients in a hospital/home-based rehabilitation setting (Pellegrini et al., 2018). Such intervention improved both patients' performance and satisfaction in carrying out meaningful activities and improved independence in instrumental ADL. However, these gains did not lead to a marked progress in participation in everyday life.

Thus, this study protocol aims to test an occupational therapy intervention focusing on achieving social participation. To do this, we have chosen to adapt treatment objectives during rehabilitation by administering two COPMs. The rehabilitation objectives will be adapted to the functional level, to the patient's interests, and to the intervention setting, which changes over the course of the rehabilitation process. During the hospital-based phase, patients focus primarily on achieving a good level of independence in B-ADL, without which it is not possible to achieve good levels of social participation (Eriksson et al., 2012). In the subsequent home-based phase, the experimental intervention clearly addresses the participation of the individual in the community. To do this, the OT develops a personalized intervention focused at enabling the patient to perform his/her everyday activities in their ecological environment, including through the support of organizations or facilities operating in any patient-specific context. Thus, the OT creates the appropriate network to support the patient's recovery of his/her social life. If the effects of this experimental approach are confirmed, patients' transition from the hospital to the home setting should be facilitated, with a potential reduction of risk of life disruption and social isolation both for patients and their caregivers (Coleman, 2003; Lou, Carstensen, Jørgensen & Nielsen, 2017).

Limitations of the study

A possible limitation of this study is that it is conducted in a single center. This choice, necessary for logistical and economic reasons, has allowed us to obtain more interpretable results. As known, usual care in rehabilitation may encompass a wide variety of practice approaches, which can vary considerably from one facility to another. Since this study is conducted in real clinical practice with the aim of determining whether EOT can improve current practice, defining what has been the usual care applied to controls in the experimental setting was essential. Thus, we chose not to carry out a multicentric study at this stage, because it could prevent us from seeing the effect of EOT in the specific context of this study.

Another limitation of this study, intrinsic to the type of intervention carried out, is the impossibility of eliminating the risk of performance bias: in trials conducted in the rehabilitation field, it is almost always impossible to guarantee the blinding of healthcare professionals and patients to group allocation.

Finally, the COPM is administered open label. However, in this trial the COPM is mainly used to define the relevant activities and consequent treatment goals, and only serves as a secondary outcome measure.

Despite these limitations, the study design is ambitious, as it has the aim of demonstrating the superiority of the experimental treatment compared to a standard care that already complies with the most recent guidelines on the treatment of complex patients who routinely access rehabilitation (CBO, 20012; Winstein et al., 2016; SPREAD, 2007).

Moreover, this trial is based on an important body of evidence (Schiavi et al., 2018; Harris & Eng, 2004; Müller et al., 2011; Pellegrini et al., 2018) and has been powered to give valid answers to the hypothesis tested.

Conclusions

Individuals with complex care needs frequently require continuous care in multiple settings. This protocol investigates the effect of an innovative rehabilitation approach, EOT, whose aim is to satisfy patients' needs in a hospital and home-based setting. If the results of this trial confirm the superiority of this approach, it may become a model of rehabilitative transitional care not yet explored (Hand et al., 2017). As this approach, extended to all complex patients, is highly individualized and embedded in patients' life, it should theoretically be highly generalizable to the population of interest.

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Declaration of interest

The authors report no conflicts of interest.

Confidentiality

The demographics and clinical data collected to support the findings of this study are restricted by the Ethics Committee of the Province of Reggio Emilia (Italy) in order to protect patient privacy. The data that support the findings of this study are available from the corresponding author (M.P.), upon reasonable request, with the permission of Azienda USL-IRCCS of Reggio Emilia, Italy.

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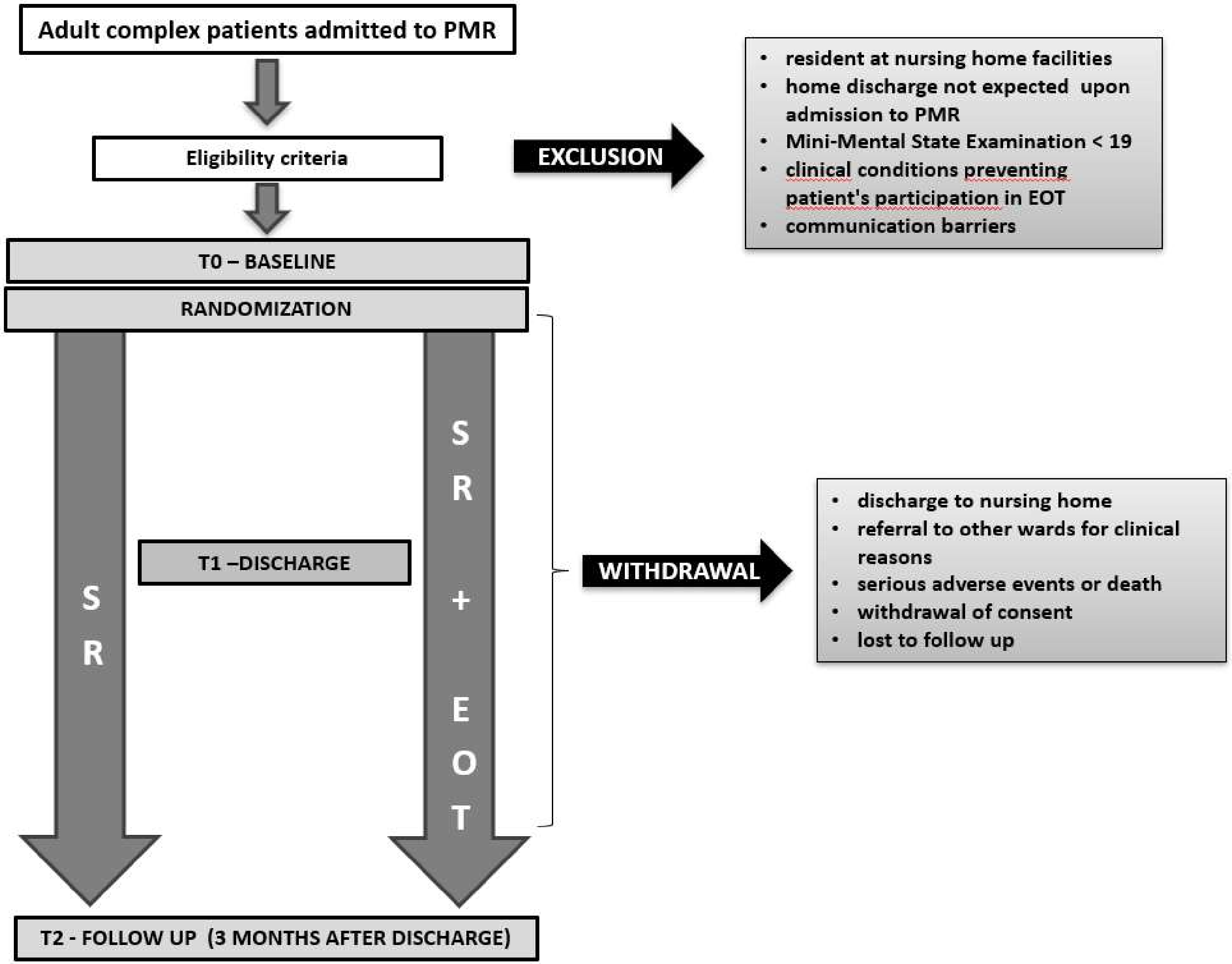
Adult complex patients admitted to PMR
Eligibility criteria
EXCLUSION
resident at nursing home facilities
home discharge not expected upon admission to PMR
Mini-Mental State Examination < 19
clinical conditions preventing patient's participation in EOT
communication barriers
T0 – BASELINE
RANDOMIZATION
SR
SR + EOT
T1 –DISCHARGE
WITHDRAWAL
discharge to nursing home
referral to other wards for clinical reasons
serious adverse events or death
withdrawal of consent
lost to follow up
T2 - FOLLOW UP (3 MONTHS AFTER DISCHARGE)

Table 1. Assessment points

	T0 - baseline (within 7 days from PMR admission)		**T1 - discharge** (within 3 days before PMR discharge)	**T2 – follow up** (90 ± 15 days from discharge)
Simultaneous presence of multiple pathologies (Charlson's combined Comorbidity Index[22])	X	**RANDOMIZA**		
Mood disturbances (HADS[20])	X		X	X
Independence in activities of daily living (MBI[18])	X		X	X
Independence in instrumental activities (I-ADL[19])	X		X	X
Patient's performance and satisfaction in carrying out priority occupational activities (first COPM[11])	X		X	
Patient's performance and satisfaction in carrying out			X	X

priority occupational activities (second COPM)	T I		
Length of stay	O	X	
Return to normal life after the event (RNLI[12])	N	X	X
Quality of life (SF-12[21])		X	X
Level of assistance needed at home		X	
Outpatient rehabilitation treatments		X	
Adherence		Throughout the study in experimental group	
Treatment-related side effects		Throughout the study	

Legend: PMR = Physical Medicine and Rehabilitation; HADS = Hospital Anxiety and depression Scale; MBI = Modified Barthel Index; IADL = Instrumental Activity of Daily Living; COPM = Canadian Occupational Performance Measure; RNLI = Reintegration to Normal Living Index; SF-12 = Short Form 12.

Appendix – Homework

Homework			Date
Activity			
Degree of client's autonomy observed by the OT	☐ Autonomous	☐ Supervision	☐ Minimal help
	☐ Moderate help	☐ Intensive help	
Caregiver			
Directions to the caregiver			
Number of repetitions			
Expected re-evaluation date			
Homework			Date
Degree of client's autonomy observed by the OT	☐ Autonomous	☐ Supervision	☐ Minimal help
	☐ Moderate help	☐ Intensive help	
Caregiver			
Directions to the caregiver	Followed? ☐ Yes ☐ No If no, why?		
Verification of the number of repetitions			

APPENDIX V

PUREAIR protocol: randomized controlled trial of intensive pulmonary rehabilitation versus standard care in patients undergoing surgical resection for lung cancer.

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