Tesis doctoral:

Participación y calidad de vida en los niños y adolescentes con parálisis cerebral

Doctor Europeus

Autora: Egmar Longo Araújo de Melo

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PhD Thesis

PARTICIPATION AND QUALITY OF LIFE IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY

Doctor Europeus

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y
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INFORMAN

Que la Tesis Doctoral realizada bajo su dirección por **Dña. Egmar Longo Araújo de Melo,** con el título **"Participación y calidad de vida en los niños y adolescentes con parálisis cerebral",** reúne los requisitos de calidad, originalidad y presentación exigibles a una investigación científica, por lo que es susceptible de ser sometida a la valoración del Tribunal encargado de juzgarla.

Y para que conste a los efectos oportunos, firmamos la presente en Salamanca, a 28 de septiembre de 2012.

Fdo. Marta Badia Corbella Fdo. Mª Begoña Orgaz Baz

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CAPÍTULO 1

INTRODUCCIÓN

Definición de parálisis cerebral y prevalencia

La parálisis cerebral (PC) es la causa más frecuente de discapacidad física en la infancia, cuya prevalencia es de 2 a 2.5 por 1.000 nacimientos (McManus et al., 2006). Datos del proyecto Servillance apuntan que, a cada año, son registrados 10.000 nuevos casos de PC en Europa (Cans et al., 2007; Colver, 2006).

Según la nueva definición propuesta por la American Academy for Cerebral Palsy and Developmental Medicine (AACPDM), la PC describe un grupo de trastornos permanentes del desarrollo del movimiento y de la postura, que causan limitaciones en la actividad y que son atribuidos a alteraciones no progresivas ocurridas en el desarrollo cerebral del feto o del lactante. Los trastornos motores de la PC están a menudo acompañados por alteraciones de la sensación, percepción, cognición, comunicación y conducta, por epilepsia y por problemas musculo-esqueléticos secundarios (Rosenbaum et al., 2007).

Los niños y adolescentes con PC han sido tradicionalmente agrupados por el tipo de trastorno motor predominante, utilizando una categoría mixta para aquellos casos en que ningún tipo domina. Esta estrategia es la adoptada por el Sistema de Clasificación descrito en el Reference and Training Manual of Surveillance of Cerebral Palsy in Europe (SCPE), el cual divide la PC en tres grupos según las características neuromotoras predominantes (Cans et al., 2007). Todos los subtipos de PC tienen en común un patrón anormal del movimiento y de la postura. La PC espástica se caracteriza por aumento del tono muscular y por la presencia de reflejos patológicos (hiperreflexia u otros signos piramidales, por ejemplo respuesta de Babinski). La PC espástica puede ser bilateral o unilateral. La PC discinética se determina por movimientos involuntarios, incontrolados, recurrentes, ocasionalmente estereotipados con predominio de reflejos primitivos y tono muscular variable. Asimismo, puede ser distónica o coreo-atetósica. Por último, la PC atáxica incluye pérdida de la coordinación muscular ordenada, por lo que los movimientos se llevan a cabo con fuerza, ritmo y presión anormal.

El sistema clasificatorio de la función motora más utilizado en la PC es el Gross Motor Function Clasification System (GMFCS), lo cual clasifica la movilidad funcional o limitación de la actividad en cinco niveles de gravedad. Los niños con niveles I y II caminan de forma independiente; mientras que aquellos con nivel III requieren muletas o andadores para la locomoción, y por último, los niños con los niveles IV y V, por lo general, requieren una silla de ruedas (Palisano et al., 2000; Wood & Rosenbaum, 2000).

Modelos de comprensión de la discapacidad

En la actualidad, los investigadores y profesionales del ámbito de la educación, salud y de los servicios sociales están interesados no solamente en capturar el impacto de la discapacidad en la vida de los niños y adolescentes con PC, pero además están incorporando las medidas de calidad de vida (CDV) y de participación para evaluar la eficacia de las intervenciones

(Colver, 2009; Dahan-Oliel, Shikako-Thomas, & Majnemer, 2012; Shikako-Thomas et al., 2012).

De hecho, los modelos contemporáneos para la comprensión de la salud y de la discapacidad, tales como la Clasificación Internacional del Funcionamiento, de la Discapacidad y de Salud (CIF) y el modelo social de la discapacidad defienden que los resultados de las intervenciones deben optimizar la participación de los niños en el hogar, en la escuela y en la vida comunitaria (Colver, 2005). El modelo de la CIF reconoce que la discapacidad resulta de la interacción entre la persona y su ambiente, y el modelo social establece que la discapacidad resulta cuando las características del ambiente físico, social y actitudinal restringen la participación en las actividades que una persona necesita o quiere hacer (Colver 2005; Colver et al., 2012).

Los niños y adolescentes con discapacidad, de la misma manera que todos los niños y adolescentes, aspiran por salud, seguridad, respeto, educación, participación y posibilidad de contribuir a la vida de los otros (McConachie, Colver, Forsyth, Jarvis, & Parkinson, 2006). En este sentido, la Convención de las Naciones Unidas sobre los Derechos del Niño (United Nations, 2006), afirma que "el niño con una discapacidad física o mental deberá disfrutar de una vida plena y decente en condiciones que aseguren su dignidad, fomenten su autosuficiencia y faciliten la participación activa en la comunidad”.

Asimismo, la Convención sobre el Derechos de las Personas con Discapacidad (United Nations, 2006) afirma la obligación de los Estados de garantizar a las personas con discapacidad, en igualdad de condiciones con las demás, al entorno físico, transporte, la información y las comunicaciones. El contenido de ambos documentos abarca dos aspectos esenciales en el cuidado de los niños y adolescentes con discapacidad: información objetiva sobre qué hace el niño, es decir, participación, y qué siente el niño sobre su vida, es decir, CDV.

Pese los esfuerzos en el sentido de garantizar una vida digna y plena, la literatura apunta que los niños y adolescentes con PC aún no tienen las mismas oportunidades y se encuentran en situación de desventaja social cuando comparados con sus pares sin discapacidad (Beckung & Hagberg, 2002; Law et al., 2006; Michelsen et al., 2009).

Proyecto SPARCLE

La alta prevalencia de PC ha impulsado nuevas líneas de investigación acerca de la CDV y de la participación. A nivel europeo, el proyecto más prominente es el coordinado por el grupo SPARCLE (Estudio sobre la Participación de los Niños con Parálisis Cerebral que viven en Europa), cuyo objetivo ha sido examinar cómo la CDV y la participación de los niños y adolescentes con PC se relacionan con el ambiente (Colver, 2006).

La investigación ha sido desarrollada en nueve regiones de siete países de Europa: Inglaterra, Francia, Italia, Alemania, Irlanda, Dinamarca y Suecia.

Los resultados del estudio SPARCLE confirman el modelo social de la discapacidad, al demostrar que niños con niveles similares de función motora gruesa presentaban resultados distintos en cuanto a la participación y a la CDV, lo que subraya el efecto del ambiente físico, social y actitudinal (Colver et al., 2012; Fauconnier et al., 2009; Michelsen, et al., 2009).

Además, se comprobó que los niños con PC presentaban niveles de CDV muy similares a la de sus compañeros sin discapacidad. Los autores justifican estos hallazgos desde una perspectiva conocida como la "paradoja de la discapacidad", cuando un niño que ha nacido y se ha desarrollado con una discapacidad, la incorpora y vive la vida con la misma intensidad que la mayoría de los niños (Dickinson et al., 2007).

Participación

La participación, componente esencial de la CIF, es definida como el hecho de involucrarse en situaciones de la vida, lo que para los niños y adolescentes significa participar en las actividades de ocio y tiempo libre, así como en las actividades escolares y en el hogar (World Health Organization, 2001). La participación en las actividades de ocio es comprendida como vital para el desarrollo del niño, pues es justamente en este contexto donde ellos desarrollan habilidades y competencias, hacen amistades, alcanzan salud física y emocional, expresan creatividad, desarrollan su auto-identidad y determinan un significado y un propósito en la vida (King et al., 2003; Law 2002; Majnemer, 2009). Además, posibilita a los niños comprender las expectativas sociales y adquirir las competencias físicas y sociales necesarias para funcionar y prosperar en sus hogares y comunidades (Law et al., 2006).

En realidad, la participación se constituye un fenómeno nuevo y complejo, e incluye aspectos objetivos y subjetivos (Imms, Reilly, Carlin, & Dodd, 2008). Asimismo, puede ser desarrollada en contextos formales, que incluyen aquellas actividades estructuradas, con reglas o metas y con un líder, entrenador o instructor formalmente designado y que habitualmente exigen un planeamiento previo, e en contextos informales, que exigen poco o ningún planeamiento y frecuentemente son iniciadas por el propio niño (King, Petrenchik, Law, & Hurley, 2009).

Los modelos explicativos actuales sobre la participación no se centran de forma exclusiva en los factores del niño, sino que la describen desde distintas perspectivas o niveles (King et al., 2006; King et al., 2003; Palisano et al., 2011). King et al. (2006) proponen un modelo conceptual que incluye aspectos relacionados con el ambiente, con la familia y con el propio niño. Según apuntan los autores, hay factores que predicen directamente la participación, como la habilidad funcional del niño (motora, cognitiva y comunicativa), la participación de la familia en las actividades recreativas y de ocio, los valores familiares respecto a aspectos culturales e intelectuales y por último, la preferencia del niño por las actividades. Los factores que predicen indirectamente la participación son la percepción de los padres respecto a las barreras ambientales, la cohesión familiar, las relaciones de apoyo al niño, además de los ingresos de la familia. En definitiva, el modelo trata de analizar las relaciones complejas entre el niño y los contextos de la familia y de la comunidad (King et al., 2006).

Calidad de Vida

El concepto actual de calidad de vida (CDV) se relaciona con el concepto de salud de la Organización Mundial de la Salud (OMS)—bienestar físico, mental y social—que considera la percepción del propio individuo sobre su salud atendiendo a sus exigencias culturales, sistemas de valores, metas, expectativas y preocupaciones (Bullinger, Schmidt, & Petersen, 2002; Eiser & Morse, 2001; Skevington, Lotfy, & O'Connell, 2004).

CDV se refiere a la satisfacción de un individuo con todas las facetas de la vida, incluyendo el bienestar físico, social, económico y psicológico. Calidad de vida relacionada con la salud (CVRS) es un componente específico del constructo CDV y está centrada en el impacto de la salud sobre el bienestar del individuo (Hays et al., 1995; Upton, Lawford, & Eiser, 2008). En la presente tesis doctoral se utilizará el concepto amplio y genérico de CDV.

El interés en el estudio de la CVRS demuestra cierta limitación de los indicadores tradicionales usados en salud, tales como supervivencia y reducción de síntomas, y subraya la necesidad de investigar de manera más amplia los efectos de una enfermedad o de un tratamiento médico (Eiser, Hill, & Vance, 2000). Como consecuencia de este cambio de enfoque de la supervivencia hacia la CDV, CVRS se ha convertido en un importante medida de resultado en los últimos años (Bullinger, Schmidt, Petersen, & Ravens-Sieberer, 2006; Tebe et al., 2008). En el contexto infantil, el estudio de la CDV debe tener en cuenta la percepción del propio niño, considerando sus entornos sociales, tales como la familia, los amigos, la escuela y la comunidad, ya que todos estos aspectos ejercen una influencia importante sobre su CDV (Matza, Swensen, Flood, Secnik, & Leidy, 2004).

Mediante los esfuerzos, tanto en el sentido de la conceptualización del constructo CVRS, como para la obtención de instrumentos robustos, transculturales y fiables, se destaca, a nivel europeo, el proyecto KIDSCREEN en lo cual han participado 13 países, incluyendo España (Ravens-Sieberer et al., 2008). Según el grupo KIDSCREEN, la conceptualización de CVRS implica un modelo comprensivo de salud subjetivo y multidimensional, y puede ser vista como un constructo que describe aspectos físicos, psicológicos, mentales, sociales y funcionales del bienestar (Ravens-Sieberer et al., 2007).

La generación de los instrumentos KIDSCREEN se basó en revisiones de literatura, consulta con grupos de expertos y grupos de discusión de niños y adolescentes de todos los países participantes. El KIDSCREEN-52 abarca 10 dimensiones o dominios: Bienestar físico, Bienestar psicológico, Estado de ánimo y emociones, Auto-percepción, Autonomía, Relación con los padres y vida familiar, Escuela, Amigos y apoyo social, Rechazo social y Recursos económicos (Aymerich et al., 2005).

Estructura de la tesis

Aunque los trastornos motores sean predominantes en la PC, la presente tesis doctoral pretende ir más allá y contribuir para el conocimiento en el campo de la discapacidad infantil, comprendiendo la PC desde el modelo biopsicosocial de la discapacidad. Como España no fue incluida en el proyecto SPARCLE y teniendo el cuenta la prevalencia de PC, cuyos últimos datos oficiales indican que en España hay 82.200 personas con PC mayores de 6 años, y aproximadamente 2.000 entre 0-5 años (Instituto Nacional de Estadística, 2008), esta investigación fue desarrollada siguiendo el modelo del grupo SPARCLE y evalúa la participación y la CDV de los niños y adolescentes con PC que viven en España.

En la actualidad existe una tendencia creciente hacia la investigación de los constructos participación y CDV. Sin embargo, no hay datos sobre qué hacen y qué sienten los niños y adolescentes españoles con PC. En el marco de la presente tesis, el trabajo que se presenta a continuación tiene como finalidad analizar la participación en las actividades de ocio y la CDV de los niños y adolescentes con PC en España. Inicialmente, y para posibilitar la evaluación de la participación, se ha llevado a cabo la traducción y adaptación del Children's Assessment of Participation and Enjoyment (CAPE) al contexto español. A continuación se presenta el perfil y predictores de participación en las actividades de ocio de los niños y adolescentes con PC en España, considerando los factores relacionados con el niño, con la familia y con su ambiente. Además, se analiza el efecto de la participación sobre la CDV; y se evalúa el grado de acuerdo entre las percepciones de CDV de los niños y adolescentes con PC y sus padres en cuanto a la CDV de los niños. Últimamente, y siguiendo la tendencia actual en analizar la influencia del ambiente sobre la participación, a través de diseños transculturales, se ha desarrollado un estudio comparativo acerca de la participación de los niños y adolescentes con PC que viven en España y Holanda.

La tesis doctoral es una compilación de cinco artículos, uno de ellos publicado, otro aceptado para publicación y tres sometidos a revistas científicas en el campo de la discapacidad. Cada artículo compone un distinto estudio empírico; sin embargo todos están dirigidos hacia la participación y la CDV de los niños y adolescentes con PC en España.

En el primer artículo "**Cross-cultural validation of the Children's Assessment of Participation and Enjoyment (CAPE) questionnaire in Spain"** se presenta el proceso de traducción, adaptación y validación del CAPE al contexto español. Se analizan las propiedades psicométricas de la versión española del CAPE, la validez de constructo y discriminante, y fiabilidad test-retest.

El segundo artículo "**Patterns and determinants of participation in leisure activities outside school in children and adolescents with cerebral palsy"** proporciona un perfil de la participación en las actividades de ocio de los niños y adolescentes con PC en España, en cuanto a la diversidad, intensidad y disfrute en la participación, así como los predictores de participación considerando las variables del niño, de la familia y del ambiente.

El tercer artículo "**The influence of participation in leisure activities on quality of life in children and adolescents with cerebral palsy"** explora la influencia de la diversidad, intensidad y disfrute de participación en las actividades de ocio sobre los distintos dominios de CDV en los niños y adolescentes con PC.

El cuarto artículo "**Insights on quality of life of children and adolescents with cerebral palsy: comparing child and parent reports**" investiga el grado de acuerdo entre las percepciones de los niños y adolescentes con PC y de sus padres en cuanto a la CDV de los niños, y determina los factores relacionados al grado de acuerdo entre padres e hijos.

El quinto y último artículo "**Participation in leisure activities of children and adolescents with and without cerebral palsy: Comparing Spain and the Netherlands**" expone el estudio desarrollado por la doctoranda durante su estancia de 3 meses (septiembre a diciembre de 2011) en el Rehabilitation Center De Hoogstraat – University Medical Center, Utrecht, the Netherlands, como pre-requisito para obtención del doctorado europeo.

Por último, el séptimo capítulo abarca las conclusiones generales de los cinco estudios, las limitaciones, recomendaciones futuras e implicaciones prácticas.

Definition and predominance of cerebral palsy

Cerebral palsy (CP) is the most common cause of physical disability in childhood, whose predominance is 2 to 2.5 per 1,000 births (McManus et al., 2006). Servillance Project data suggest that each year, 10,000 new cases are registered in Europe for PC (Cans et al., 2007; Colver, 2006).

Under the new definition proposed by the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM), CP describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, behaviour, by epilepsy and by secondary musculoskeletal problems (Rosenbaum et al., 2007).

Children and adolescents with CP are traditionally grouped by the type of predominant motor disorder, using a mixed category for those cases where no dominant. This strategy adopted by the classification system described in the Reference and Training Manual of Surveillance of Cerebral Palsy in Europe (SCPE), divides CP in three groups according to the predominant neuromotor characteristics (Cans et al., 2007). All subtypes of PC have an abnormal pattern of movement and posture in common. Spastic CP is characterized by increased muscle tone and by the presence of pathological reflexes (hyperreflexia or other pyramidal signs, for example Babinski response). Spastic CP can be bilateral (CP-EB) or unilateral (CP-EU). Dyskinetic CP is determined by involuntary, uncontrolled, recurring, occasionally stereotyped predominantly primitive reflexes and muscle tone variable. It can also be dystonic or choreo-athetoid. Finally, Ataxic CP includes muscular incoordination order, which means the movements are performed with strength, rhythm and abnormal pressure.

The classification system of motor function most used in the CP is the Gross Motor Function Classification System (GMFCS), which classifies functional mobility or activity limitation in five severity levels. Children with levels I and II of GMFCS walk independently, while those with level III require crutches or walkers for locomotion, and finally, children with levels IV and V, usually require a wheelchair (Palisano, et al., 2000; Wood & Rosenbaum, 2000).

Models of understanding disability

Currently, researchers and professionals in the field of education, health, social service and are interested not only in capturing the impact of disability on the lives of children and adolescents with CP, but also are incorporating quality of life (QoL) and participation measures to assess the effectiveness of interventions (Colver, 2009; Dahan-Oliel et al., 2012; Shikako-Thomas et al., 2012).

Indeed, contemporary models for understanding health and disability, such as the International Classification of Functioning, Disability and Health

(ICF) and the social model of disability argue that interventions results should optimize participation of children in home, school and community life (Colver, 2005). The ICF model recognizes that disability results from the interaction between people and their environment, and the social model states that disability results when the characteristics of the physical, social and attitudinal environment restricting participation in the activities that a person needs or wants to do (Colver, 2005; Colver et al., 2012).

Children and adolescents with disabilities, in the same way that all children and adolescents, aspire for health, safety, respect, education, participation and ability to contribute to the lives of others (McConachie, Colver, Forsyth, Jarvis, & Parkinson, 2006). In this regard, the United Nations Convention on the Rights of the Child (United Nations, 2006) states that "the child with a physical or mental disability should enjoy a full and decent life in conditions which ensure dignity, promote self-sufficiency and facilitate active participation in the community".

Despite efforts in the direction of ensuring a life of dignity and fulfilment, the literature indicates that children and adolescents with CP still do not have equal opportunities and are at a social disadvantage when compared with their peers without disabilities (Beckung & Hagberg, 2002; Law et al., 2006; Michelsen et al., 2009).

The SPARCLE project

The high prevalence of CP has prompted new research, mainly on QoL and participation. At European level, the most prominent project is coordinated by the SPARCLE Group (Study on the Participation of Children with Cerebral Palsy living in Europe), whose objective is to examine how the quality of life and participation of children and adolescents PC relate with the environment (Colver, 2006). The research was carried out in nine regions of seven European countries: England, France, Italy, Germany, Ireland, Denmark and Sweden. The SPARCLE study results confirm a social model of disability by showing that children with similar levels of gross motor function had different results in terms of participation and QoL, highlighting the effect of the physical, social and attitudinal environment (Colver et al., 2012; Fauconnier et al., 2009; Michelsen et al., 2009). Furthermore, it was found that children with CP had similar levels of QOL than that of their peers without disabilities. The authors justify these findings from a disability paradox perspective, when a child is born and develops with a disability; he or she incorporates this and lives life in the same intensity of majority of children (Dickinson et al., 2007).

Participation

Participation, an essential component of the ICF, is defined by the ICF as involvement in life situations, which for children and adolescents means participating in recreational and leisure activities as well as school and home activities (WHO, 2001). Participation in leisure activities is understood as vital to a child's development, it is precisely in this context that they develop skills and competencies, make friends, achieve physical and emotional health, express creativity, develop their self-identity and determine a meaning and purpose in life (King et al., 2003; Law, 2002; Majnemer, 2009). It also enables children to understand social expectations and gain physical and social skills necessary to function and thrive in their homes and communities (Law et al, 2006).

Indeed, participation is a complex and new phenomenon, and includes both objective and subjective aspects (Imms, Reilly, Carlin, & Dodd, 2008). In addition, participation can be developed in formal settings, which include those structured activities with rules or goals and a leader, coach or instructor formally designated and usually require advance planning; and in informal contexts, which require little or no planning and often are initiated by the child (King, Petrenchik, Law, & Hurley, 2009).

The current explanatory models of participation do not focus exclusively on the factors of the child, but they described it from different perspectives or levels (King et al., 2003; King et al., 2006; Palisano et al., 2011). King et al. (2006) propose a conceptual model that includes aspects related to the environment, the family and the child. As the authors note, there are factors that predict participation directly, such as the child's functional ability (motor, cognitive and communicative), family participation in recreational and leisure activities, family values regarding cultural and intellectual aspects and finally, the child's preference for the activities. Factors that predict participation indirectly are the perceptions of parents regarding environmental barriers, family cohesion, relations child support, in addition to the family income. In conclusion, the model attempts to analyze the complex relationships between the child and the contexts of family and community (King et al., 2006).

Quality of Life

The current concept of quality of life (QoL) is related to the concept of health of the World Health Organization (WHO), which is physical, mental and social well-being, and considers the individual's own perception of their health meeting their cultural needs, value systems, goals, expectations and concerns (Bullinger, Schmidt, & Petersen, 2002; Eiser & Morse, 2001; Skevington, Lotfy, & O'Connell, 2004).

QoL refers to an individual's satisfaction with all facets of life, including physical, social, economic and psychological well being. Quality of life related to health (HRQoL) is a specific component of the QoL construct and focuses on the impact of health on the welfare of the individual (Hays et al., 1995, Upton, Lawford & Eiser, 2008). In this thesis the broad concept and generic QoL will be used.

The interest in the study of HRQoL demonstrates some limitations of traditional indicators used in health, such as survival and symptom reduction, and stresses the need to investigate more broadly the effects of an illness or medical treatment (Eiser, Hill & Vance, 2000). As a result of this shift in focusing on survival towards QoL, HRQoL has become an important measurement in recent years (Bullinger, Schmidt, Petersen, & Ravens-Sieberer, 2006; Tebe et al., 2008). In the child context, the QoL study should take into account the perception of the child, considering their social environments, such as family, friends, school and the community, as all of these have an important influence on their QoL (Matza et al., 2004).

Through the efforts of the sense of the conceptualization of HRQoL construct, as for obtaining robust, reliable and cross cultural tools, the KIDSCREEN project is highlighted at European level, in which 13 countries participated, including Spain. According to the KIDSCREEN group, the conceptualization of HRQoL involves a comprehensive multidimensional model of subjective health and can be seen as a construct that describes the physical, psychological, mental, social and functional well-being aspects (Ravens-Sieberer et al., 2007).

Generating KIDSCREEN instruments was based on literature reviews, consultation with expert groups and discussion groups of children and adolescents of all participating countries. The KIDSCREEN-52 includes 10 dimensions or domains: Physical well-being, Psychological well-being, Moods & emotions, Self-perception, Autonomy, Parental relations, Social support & peers, School environment, Social acceptance (bullying), and Financial resources. (Aymerich et al., 2005).

Structure of the thesis

Although motor disorders are prevalent in CP, the present thesis aims to go further and contribute to knowledge in the field of childhood disability, understanding the CP from the biopsychosocial model of disability. As Spain was not included in the SPARCLE project and taking into account the prevalence of CP, whose latest official data indicated that in Spain there are 82,200 people with CP over 6 years, and approximately 2,000 0-5 years (Instituto Nacional de Estadística, 2008), this research was developed following the SPARCLE Group's the model and evaluates participation and QoL of children and adolescents with CP living in Spain.

There is now a growing trend towards investigating the constructs participation and QoL, what are the determinants of participation and the participation effect on QoL in children and adolescents with CP. Despite the growing interest in the topic in the international scientific community, there is no data in Spain about what children and adolescents with CP do and how they feel. As part of this thesis, the study presented here is to analyze the participation in leisure activities and QoL of children and adolescents with CP in Spain. Initially, and to enable the evaluation of participation, translation and adaptation had to be undertaken of the Children's Assessment of Participation and Enjoyment (CAPE) to the Spanish context. After, the profile and predictors of participation in leisure activities of children and adolescents with CP in Spain were demonstrated, considering factors related to the child, with family and with their environment. Moreover, it has analyzed the effect of participation on QoL, and has assessed the degree of agreement between the perceptions of QoL of children and adolescents with CP and their parents regarding child's QoL. Lastly, following the current trend to analyze the influence of the environment on participation through cross cultural designs, we have developed a comparative study on the participation of children and adolescents with CP living in Spain and the Netherlands.

The doctoral thesis is a compilation of five articles, one of them published, another accepted for publication and three which have been submitted for publication in scientific journals in the field of disability. Each section comprises a different empirical study, but all are directed towards participation and quality of life of children and adolescents with CP in Spain.

The first article "Cross-cultural validation of the Children's Assessment of Participation and Enjoyment (CAPE) questionnaire in Spain" presents the process of translation, adaptation and validation of the CAPE into the Spanish context. The psychometric properties of the Spanish version of CAPE, construct validity and discriminant, and test-retest reliability were analyzed.

The second article "Patterns and predictors of participation in leisure activities outside school in children and adolescents with cerebral palsy" provides a profile of participation in leisure activities of children and adolescents with CP in Spain, in terms of diversity, intensity participation and enjoyment and predictors of participation considering the variables of the child, family and environment.

The third article, "The Influence of participation in leisure activities on quality of life in children and adolescents with cerebral palsy" explores the influence of diversity, intensity and enjoyment of participation in leisure activities on the various domains of QoL in children and adolescents with CP.

The fourth article, "Insights on quality of life of children and adolescents with cerebral palsy: comparing child and parent reports" investigates the level of agreement between the perceptions of children and adolescents with CP and their parents regarding QoL of children, and determines the factors related to the degree of agreement between parents and children.

The fifth and last article "Participation in leisure activities of children and adolescents with and without cerebral palsy: comparing Spain and the Netherlands" describes the study developed by PhD student during her stay of three months (September to December 2011) at the Rehabilitation Center De Hoogstraat – University Medical Center, Utrecht, the Netherlands, as a prerequisite to obtaining the European doctorate.

Finally, the seventh chapter covers the general findings of the five studies, limitations, future research lines and practical implications.

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CAPÍTULO 2

CROSS-CULTURAL VALIDATION OF THE CHILDREN'S ASSESSMENT OF PARTICIPATION AND ENJOYMENT (CAPE) QUESTIONNAIRE IN SPAIN

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ABSTRACT

Despite growing interest in the topic of participation, the construct has not yet been assessed in children and adolescents with and without Cerebral Palsy (CP) in Spain. As there are no available instruments to measure participation in leisure activities which have been adapted in this country, the goal of this study was to validate a Spanish version of the Children's Assessment of Participation and Enjoyment (CAPE). The sample comprised 199 children and adolescents with CP and 199 without CP, between 8-18 years of age, from 7 regions in Spain. The adaptation of the original version of CAPE was carried out through translation and backward translation, and the validity of the instrument was analyzed. Construct validity was assessed through the correlation of the diverse CAPE domains and the quality of life (QoL) domains (KIDSCREEN questionnaire). Discriminant validity was established by comparing children and adolescents with CP and typically developing children and adolescents. For test-retest reliability, the children and adolescents with and without CP completed the CAPE twice within 4 weeks. The correlations found between the CAPE domains and the QoL domains show that the CAPE presents construct validity. The CAPE discriminated children and adolescents with CP from those without any disability in the results of participation. According to most CAPE scales, typically developing children and adolescents engage in a greater number of activities than children and adolescents with CP. Test-retest reliability for the Spanish version of CAPE was adequate. The study provides a valid instrument to assess the participation of children and adolescents with and without CP who live in Spain.

Keywords: participation; leisure, children, adolescents, cerebral palsy, measurement.

INTRODUCTION

The current perspective of intervention aimed at people with cerebral palsy (CP) focuses on promoting participation in leisure activities, because this variable has become a firm indicator of good levels of quality of life (QoL) and health (Law, 2002; Vargus-Adams & Martin, 2011).

The International Classification Functioning Disability and Health (ICF), defines participation as the fact of engaging in life situations (WHO 2001). It is a complex phenomenon that needs to be understood and measured, taking into account both its objective and subjective components (Coster & Khetani, 2008; Imms, 2008; McConachie, Colver, Forsyth, Jarvis, & Parkinson, 2006).

Participation in leisure activities is considered vital for children's development, because it is precisely in this context where they develop skills and competences, make friends, achieve physical and emotional health, express creativity, develop their self-identity, and determine the meaning and purpose of their lives (Barletta & Loy, 2006; Dijkers et al., 2000; King et al., 2003; Law, 2002; Specht et al.. 2002). Leisure is defined as an activity in which an individual freely chooses to participate during their spare time because they find such activities enjoyable (Majnemer et al., 2008).

As evident in current publications (Fauconnier et al., 2009; Imms et al., 2009; Kang et al., 2010; King et al., 2006a; Klaas et al., 2010; Law et al., 2006; Law et al., 2011; Majnemer et al., 2010; Michelsen et al., 2009; Michielsen et al., 2011; Orlin et al., 2010; Palisano et al., 2009; Palisano et al., 2011), the Children's Assessment of Participation and Enjoyment (CAPE) is the most extensively used instrument to measure participation in leisure activities from the child’s perspective and adolescents with CP and other developmental disabilities, and it can also be applied to the able-bodied population. It assesses the diversity and intensity of participation, with whom the child participates, where the activity is performed, and the enjoyment levels (King et al., 2006a).

The original version of CAPE presents adequate psychometric properties and was validated in a broad longitudinal study with Canadian children and adolescents with physical impairments, and the instrument was found to correlate with other measures of diverse constructs (King et al., 2004; King et al., 2006a).

Actually, two translations of CAPE are available, 1 in Spanish for the Spanish-speaking population in Puerto Rico (a bilingual country: English and Spanish), and another in Dutch for the population in the Netherlands. In both studies children and adolescents with and without disabilities took part and provided data confirming the construct validity and test–retest and inter-interviewer reliability (Bult et al., 2010; Colón et al., 2008).

Despite increasing growing interest in the construct of participation and Cerebral palsy being one of the most common causes of disability in children, with 10,000 new cases occurring each year in the European Union (McManus, Corcoran, & Perry, 2006), there is no instrument aimed at assessing participation in children and adolescents with CP that is validated in Spain.

Hence, the goal of this study is to present the results of the process of translation, adaptation, and validation of CAPE to the Spanish context, more specifically to examine the construct and discriminant validity, and test-retest reliability.

As there was no other questionnaire available to measure participation that was validated in Spanish population, with regard to construct validity, we took into account that the constructs QoL and participation were correlated (McManus et al., 2008). Therefore, we expected that the CAPE domains scores would correlate positively with the QoL domains of KIDSCREEN.

Regarding discriminant validity, taking into account previous investigations (Bult et al., 2010; Imms et al., 2008; King et al., 2006a; Law et al., 2006; Majnemer et al., 2008), the following research hypotheses were formulated: 1) Children and adolescents with CP present a lower frequency (intensity) of participation in leisure activities than the typically developing children and adolescents; and 2) There are significant differences in the diversity of participation in the formal and informal domains of CAPE, and in the 5 types of activity that make up the test.

Lastly, we expect the results of test-retest reliability of the Spanish version of the instrument would be satisfactory.

METHODS

Participants

A convenience sample of 199 children and adolescents with CP was obtained from various centers that were located in diverse regions of Spain and which are members of the Spanish Confederation of Federations and Associations for the Care for People with Cerebral Palsy and Related Disabilities (ASPACE), to achieve a heterogeneous and representative sample of the population of children and adolescents with CP. The representatives of the organizations promoted the information about the investigation and a total of 16 centers responded to the request to participate in the study. The recruitment criteria included being between 8 and 18 years of age and having a confirmed diagnosis of CP. An exclusion criterion for the CP group was the presence of an autism spectrum disorder.

The CP group comprised 113 boys and 83 girls with an average age of 12.12 (SD = 3.02, range = 8-18 years) who attended 16 ASPACE centers from 7 regions of Spain.

The control group comprised 199 typically developing children and adolescents (48.7% male and 51.3% female), mean age 13.21 years (SD = 3.13; range 8-18 years), from 2 regular schools from 2 regions in Spain (see Table 1).

The research received ethical approval from the Bioethical Committee of the University of Salamanca. All parents gave written consent, and all children with sufficient intellectual capacity gave written consent or communicated consent if unable to write.

Table 1. Characteristics of the participants

Characteristics		With CP (N=199) N (%)	Without CP (N=199) N (%)
Gender	**Male**	113 (56.8)	97 (48.7)
	Female	86 (43.2)	102 (51.3)
Age	**Years**	Mean=12.12 (SD=3,02; Range=8-18)	Mean=13.21 (SD=3.13; Range=8-18)
GMFCS	**Level I**	50 (25.1)	-
	Level II	34 (17.1)	-
	Level III	37 (18.6)	-
	Level IV	27 (13.6)	-
	Level V	51 (25.6)	-
	None	38 (19.1)	-
Intellectual Impairment	**Mild**	43 (21.6)	-
	Moderate	33 (16.6)	-
	Severe	85* (42.7)	-

*In these cases (severe intellectual impairment) CAPE was completed by the father or mother

Instruments

Gross Motor Function Classification System (GMFCS). The GMFCS is an age-specific scheme designed for children with CP based on five levels of gross motor function, ranging from Level I (most able) to Level V (least able). The psychometric properties of GMFCS have been thoroughly tested and reported, and include evidence supporting its content validity, construct validity, inter-rater reliability and test–retest reliability (Palisano et al., 1997; Palisano et al., 2000; Wood & Rosenbaum, 2000).

Children´s Assessment of Participation and Enjoyment (CAPE). This scale assesses participation in leisure activities in children and adolescents between

6 and 21 years of age, with and without disability. It has 55 items, diverse out-of-school activities, in which are presented by means of illustrations or figures. These items are classified by domains: formal (15 items) or informal (40 items) and by type of activity: based on skill (10 items), physical activity (13 items), recreation (12 items), social (10 items), and self-improvement (10 items). For each item, 5 questions are posed: whether they carried out the activity in the past four months—which allows us to assess the diversity of activities in which they participate; if they answer affirmatively, they should indicate the frequency (intensity), with whom and where they carried out the activity, and, lastly, whether they enjoyed performing the activity.

High scores indicate that the child participates in many different activities and low scores indicate that the child participates in few activities. Intensity scores represent the average frequency of the total number of possible activities, rather than the average frequency of the activities the child actually performs. Overall, it represents the average extent of participation and must be interpreted along with the diversity score. The "with whom" score is an average measure of whether the child participates in activities alone, with family, friends, etc. Low scores indicate that the child's participation tends to be solitary or occurs with the immediate family. High scores indicate a more social orientation with activities carried out with many types of people. The "where" score is similar to the "with whom" score, so low scores indicate that activities are performed at home, and higher scores indicate that activities tend to be carried out in the child's neighborhood or broader community. Enjoyment scores reflect the child's average enjoyment of across the activities and high scores indicate high enjoyment (King et al., 2004).

The process of translation, adaptation, and validation of the CAPE questionnaire to the Spanish context was carried out following the recommendations for the adaptation of tests proposed by the International Test Commission (ITC) (Hambleton, 1994; Tanzer & Sim, 1999; Van de Vyjver & Hambleton, 1996). The study was developed in 5 phases: (a) translation by 3 translators; (b) synthesis of the translations by consensus of the 3 translators, which produced the first Spanish version of the original instrument; (c) selection of 2 focal groups of 7 children and adolescents with and without CP, aged between 8 and 18 years (the participants had verbal and communication skills that allowed them to understand and respond to the questions in order to analyze the cultural acceptability of the items or pictures). The information collected by the participants of the two groups showed that no item or picture needed changes; (d) concordance and synthesis was carried out by a committee of experts comprising seven professionals from the area of childhood disability, to achieve semantic, idiomatic, experiential, and conceptual equivalence. According to the experts, none of the items of CAPE had to be removed or replaced, so all the original items were kept; and (e) backward translation was performed by 2 translators, whose maternal language was English and who were bilingual in Spanish, who met with the research team of the study and together they compared the differences or discrepancies of both back-translated versions with the original version, obtaining as a result the definite version of CAPE.

An additional instrument was used to determine the validity of CAPE: the Spanish version of KIDSCREEN, a generic health-related QoL questionnaire for children (8–18 years of age). KIDSCREEN was chosen because it is a modern instrument with excellent psychometric properties, which uses questions derived from focus-group work with children across Europe to assess the QoL of children and young people. The instrument has a parent/proxy version with 52 items covering 10 domains, namely, physical well-being, psychological well-being, moods & emotions, self-perception, autonomy, parent relation & home life, financial resources, social support & peers, school environment, and social acceptance (Aymerich et al., 2005). The questionnaire uses 5-point Likert scales and each domain includes 3 to 7 items. Item responses are added to give a score of 0 to 100 for each domain, with higher scores indicating better QoL. Starting with the premise that participation in leisure activities in children with CP is related to their perception of QoL (McManus et al., 2008), we used the scores in this instrument to explore the validity of CAPE. As shown earlier, we expected to obtain positive correlations between the CAPE domains (diversity, intensity and enjoyment) and the domains of QoL, specifically: physical well-being, psychological well-being, moods & emotions, self-perception, autonomy and social support & peers, where, the higher CAPE scores, corresponded to the higher levels of quality of life (Dahan-Oliel et al., 2012; McManus et al., 2008).

Gender and age of the children and adolescents with CP were collected by a socio-demographic questionnaire.

The level of intellectual impairment was obtained from the psychological records of the center where the children or adolescents received services. A Psychologist reported on each child considering the following criteria intellectual impairment: none or mild (IQ>70); moderate (IQ 50-70), and severe (IQ<50).

Procedure

Data of children and adolescents with CP were collected by the first author and by researcher assistants (one at each ASPACE), who previously received formal training from the research team. Children and adolescents answered the CAPE questionnaire and parental assistance or proxy was used as necessary. Of the 199 children and adolescents with CP, 114 (57.3%) completed the CAPE questionnaire independently or received some assistance. In the cases of severe intellectual impairment (42.7%), the parents completed the CAPE questionnaire by proxy. In the typically developing children and adolescents group, CAPE was self-completed in all cases. A convenience sub-sample of 27 children completed the CAPE questionnaire again with the same interviewer after 4 weeks to assess test–retest reliability. Children with and without CP were included in these analyses (10 children without CP and 17 children with CP who were grouped into GMFCS levels as followed: 5 children in level I; 5 children in level II; 4 children in level II and 3 children in level IV).

The psychologist, social worker, or physical therapist of each center completed the socio-demographic questionnaire and the participants' GMFCS.

The KIDSCREEN parents` version was completed by a parent as the respondent. The data of the KIDSCREEN children´ version was collected through individual interviews carried out by the same researcher assistant, after the CAPE interview. Of 182 participants with CP evaluated for QoL through the parents' version, only 74 gained the child report, with or without assistance from the examiner.

Statistical Analyses

Firstly, descriptive analyses (means and standard deviations) were conducted for each CAPE domain in both groups.

For the validation process of CAPE, a detailed analysis of the psychometric characteristics of the instrument reliability and validity—construct and discriminant validity was performed.

In order to determine construct validity, Pearson's product moment correlation was used to analyze the correlations between the scores obtained in the domains of the KIDSCREEN parents' version and children's version and the CAPE domains, taking into account that the KIDSCREEN was developed under a rigorous methodological process, and it has been shown to have construct validity (Aymerich et al., 2005).

Regarding discriminant validity, t-tests were conducted to compare children and adolescents with CP and typically developing children and adolescents and to test the above-mentioned hypotheses.

To assess test-retest reliability, the intraclass correlation coefficients (ICC) was used (two-way mixed, with absolute agreement). In order to determine whether the measuring instrument can be considered reliable, the criteria of Fleiss (1981) was taken into account, which considers that values of ICC > 0.75 are excellent; between 0.60 and 0.74, good; between 0.40 and 0.59, moderate, and values lower than 0.40 poor. Also, the standard error of measurement (SEM) and the smallest difference in measurement (SDC) were calculated considering the recommendations established by de Vet et al. (2006).

To perform these analyses, the SPSS statistical package version 17 was used, and in all cases the level of significance of 0.05 was used.

RESULTS

Descriptive Statistics

Table 2 shows the means and standard deviations for each cape domain in both groups. Regarding diversity of participation, out of the 55 activities proposed, the group with CP participated in an average of 20.4 activities, and the typically developing children and adolescents group in 26.1. Participation intensity in the group with CP was 1.8, whereas the group without CP was 2.3. With reference to with whom they carried out the activity, the mean of 2.5 reflects that both groups carried out the activities mainly with their parents or other relatives. Concerning where they performed the activities, the means indicated that these activities were carried out at home, or in relatives' or neighbors' homes. Lastly, the mean level of enjoyment of both groups was between 3.5 and 3.9.

Table 2. Descriptive Statistics for CAPE´s domains for group with and without CP

Domains (Max. Scores)	CP group (N=199) Mean (SD)	Without CP group (N=199) Mean (SD)
Diversity (0-55)	20.4 (8.6)	26.1 (7.7)
Intensity (0-7)	1.8 (0.7)	2.3 (0.7)
With Whom (0-5)	2.5 (0.4)	2.5 (0.4)
Where (0-6)	2.7 (0.6)	2.9 (0.6)
Enjoyment (0-5)	3.9 (0.5)	3.5 (0.6)

Construct Validity of the Spanish Version of CAPE

We expected to obtain positive correlations between the CAPE domains (diversity, intensity and enjoyment) and the domains of quality of life: the higher the CAPE scores, the higher the levels of quality of life where, the higher CAPE scores, corresponded to the higher levels of quality of life.

Significant relations were obtained between the three domains of CAPE—diversity, intensity, and enjoyment—and most of the QoL domains assessed by the KIDSCREEN questionnaire (see Table 3).

Regarding the diversity domain, significant positive relations in the parents' version with the QoL domains of physical well-being, social support & peers, financial resources and psychological well-being were obtained; in the children's version, significant positive relations were only obtained with financial resources, indicating that high diversity of participation were associated with high QoL in these domains. However, significant negative relations of diversity with the domains of self-perception and mood & emotions were found, but only in the parents' version.

The intensity domain was correlated positively and significantly with the domains of physical well-being, social support & peers, and financial resources both for the parents' and the children's version, indicating that high intensity of participation were associated with high QoL in these domains. Moreover, in the case of the parents' version, it also was correlated positively with psychological well-being and autonomy, and negatively with self-perception (see Table 3).

Lastly, in the enjoyment domain (see Table 3), also were found positive and significant correlations with the domains of physical well-being, psychological well-being (parents' and children's version), and with the domains parent relations & home life, self-perception, autonomy, and school environment in the parents' version. Higher scores in these domains indicated higher levels of enjoyment.

Table 3. Correlation coefficients between CAPE domains and KIDSCREEN domains (parents and children version)

QoL Domains KIDSCREEN	Diversity		Intensity		Enjoyment	
	Parents	Children	Parents	Children	Parents	Children
Physical Well-being	.415**	.182	.438**	.275*	.243**	.360**
Psychological Well-being	.155*	.140	.216**	.176	.383**	.413**
Moods & Emotions	-.210**	.059	-.127	.137	.149	.122
Self-Perception	-.291**	-.082	-.262**	-.026	.165*	.164
Autonomy	.169*	.027	.243**	.135	.156*	.098
Parent Relations & Home Life	-.007	.026	.061	.140	.242**	.079
Social Support & Peers	.410**	.215	.456**	.308**	.140	.148
School Environment	.010	.074	.052	.045	.159*	.187
Social Acceptance	-.057	.039	-.024	.142	.084	-.005
Financial Resources	.261**	.327**	.251**	.358**	-.037	.119

Parents version: N=182. Children version: N= 74
Pearson´s product moment correlation was used in these analyzes.
***p<.001, **p<.01, *p<.05

Discriminant Validity of the Spanish Version of CAPE

Regarding discriminant validity, significant differences favoring the typically developing children and adolescents in the domains of diversity and intensity were observed. Specifically, in the diversity domain, differences in recreational, physical, and self-improvement activities and, in formal and informal domains were found (see Table 4). Typically developing children and adolescents participated significantly in more recreational, physical, and self-improvement activities, both in formal and informal domains.

Concerning intensity, the differences were statistically significant for physical, social, and self-improvement activities, and also in formal and informal domains (see Table 4). Typically developing children and adolescents participated more frequently in physical, social, and self-improvement activities, both in formal and informal domains.

Table 4. Mean participation diversity and intensity scores (standard deviation) by activity type

Domain/Activity type	Diversity		Intensity	
	Without CP	With CP	Without CP	With CP
Recreational	7.0 ** (2.5)	6.3 (2.8)	2.8 (1.2)	2.6 (1.2)
Active physical	4.4*** (2.3)	2.8 (1.8)	1.6*** (.89)	1.0 (.75)
Social	6.8 (1.8)	6.5 (2.0)	3.1** (.99)	2.9 (.93)
Skill-based	2.9 (1.6)	2.9 (1.7)	1.5 (.86)	1.4 (.88)
Self-improvement	5.2*** (1.9)	3.9 (2.0)	2.7 *** (1.0)	2.0 (1.0)
Formal	4.6*** (2.3)	3,6 (2.3)	1.5 *** (.78)	1.2 (.76)
Informal	22 *** (6.1)	17 (7.0)	2.6 *** (.78)	2.0 (.86)

With CP: N=199. Without CP: N= 199
t-tests were used to compare the two groups.
***p<.001, **p<.01, *p<.05

Test-retest reliability

The mean time between the assessments for test-retest reliability was 28 days (SD = 4). After calculating test-retest reliability for the 5 domains analyzed, the values of the ICC ranged between 0.54 and 0.80. Reliability for the CAPE domains were as followed: “enjoyment” and “where” excellent, “intensity” and “with whom” good and “diversity” moderate. Likewise, the standard measurement errors were low and confirmed the accuracy of the instrument. Lastly, the values found in the smallest difference in measurement indicated that the instrument can detect small changes (see Table 5). Therefore, the Spanish version of CAPE presents positive results regarding test-retest reliability.

Table 5. Test-Retest Reliability CAPE (N=27)

DOMAINS	ICC	95% C I	SEM	SDC
Diversity	0.54	0.04-0.78	3.26	9.01
Intensity	0.74	0.61-0.85	0.71	1.96
With Whom	0.71	0.57-0.84	0.89	2.46
Where	0.76	0.63-0.87	1.00	2.76
Enjoyment	0.80	0.69-0.89	0.86	2.38

The intraclass correlation coefficients (ICC), the standard error of measurement (SEM) and Smallest detectable change (SDC) were used in these analyzes.
SEMs and SDCs are expressed in the units as followed: diversity (0-55), intensity (0-7), with whom (0-5), where (0-6) and enjoyment (0-5).
CI (Confidence interval).

DISCUSSION

This study analyzed the validity of the Spanish version of CAPE, an instrument for assessing the participation in leisure activities from the children's perspective. Through participation in leisure activities, children acquire new skills, increase their physical, emotional, and social well-being and enhance their QoL (Hoogsteen & Woodgate, 2010). Being able to participate fully in society is particularly important for children to ensure their successful transition to adulthood and, ultimately, their independent living (Parkes et al., 2010).

To our knowledge, this is the first study that attempted to assess the participation of children and adolescents with CP living in Spain, using the Spanish version of CAPE. It is currently one of the most extensively used instruments because it provides valuable information about participation from the perspective of the children and adolescents (Clarke et al., 2011; Imms, 2008; King et al., 2011; Klaas et al., 2010; Majnemer et al., 2008; Michielson et al., 2011; Orlin et al., 2010; Palisano et al., 2009; Palisano et al., 2011).

The low results in participation of the Spanish sample of children and adolescents with CP regarding diversity and intensity were also found in studies using CAPE in Canada, the United States, Israel, and Australia (Imms, 2008; Majnemer et al., 2008; Orlin et al., 2010). Specifically, concerning the data on intensity, Imms (2008) adds that this score provides a relative indicator of participation frequency rather than an absolute measure and should be interpreted along with the diversity score. Imms et al. (2008) showed that Australian children with CP tended to participate in activities on their own or with family members or relatives, suggesting that the burden of providing support for activities lies within the family. Regarding where they carry out the activities, the means indicated that these activities are carried out at home, or in relatives' or neighbors' homes. The physical restrictions of children and adolescents with CP may limit them to performing these activities in special environments (King et al., 2011; Palisano et al., 2009). Also, they might participate more at home because their parents can help them (Imms, 2008).

The correlations found between the CAPE domains and the QoL domains assessed through the KIDSCREEN showed that CAPE presents construct validity.

The positive and significant correlation between diversity of participation and the domains of physical well-being, social support and peers was expected because engaging in more activities can facilitate the development of new social links and improve the feeling of physical well-being. Although there is no published evidence, the positive correlation between diversity and intensity of participation and domain of financial resources is not an unexpected factor because the availability of funds may facilitate participation in a greater number of activities.

Some studies have shown the influence of leisure activities in promoting quality of life of people with and without intellectual disabilities (Duvdevany & Arar, 2004; Duvdevany, 2008; Iwasaki, 2007). McManus et al. (2008) showed that independently of gender, age, and level of impairment, overall participation in everyday activities, evaluated by frequency of participation questionnaire (FPQ), had a significant effect on the QoL domains of Irish children with CP. An increase of 1 unit in participation was associated with increases of 7.8 and 13.9 in QoL related to physical well-being and social support & peers, respectively.

In spite of being unexpected our results indicated that, according to parents perception, there were a negative correlation between participation in leisure activities and QoL in the self-perception and mood & emotions domains, indicating that high levels of participation were associated with low QoL in these 2 domains. We believe that these negative correlations may be explained taking into account that the system of care predominant in Spain is focused on the family. When parents judge their children's' self-perception and mood and emotions as low, this leads them to promote higher diversity and intensity of participation. Consistent with this theory, it has been shown that parents developed a number of strategies and made extraordinary efforts to promote the social life of their children and adolescents who have a disability (Antle et al., 2007; Bedell & Dumas, 2004; King et al., 2006b).

Another important point is that these negative and significant correlations between the 2 domains of KIDSCREEN (moods & emotions and self-perception) appeared solely in the parents' version. Discrepancy between self-reports and parent-proxy reports of children and adolescents health-related quality of life (HRQoL) have repeatedly been acknowledged in the literature as 'the proxy problem' (Sattoe et al., 2012). In this sense we agree with the current tendency of to incorporate children's perceptions in the studies of QoL and participation in leisure activities (White-Koning et al., 2007), and for this reason we have considered both the children report and parent reports, to assess construct validity.

CAPE in its Spanish version discriminated children and adolescents with CP from those without any disability in the results of participation. In most CAPE domains, typically developing children and adolescents engaged in a greater number of activities than children and adolescents with CP. The best results were observed in favor of the group without CP in the diversity domain in recreational activity, physical activity, and self-improvement, and in the informal domain in another study using the CAPE (Engel-Yeger et al., 2009). The statistically significant differences found in physical, social and self-improvement activities were also observed in the validation study of the CAPE in the Netherlands (Bult et al., 2010).

These results indicate that Spanish children and adolescents with CP are still at a social disadvantage when compared to typically developing children and adolescents. Participation includes children's interactions with their parents, teachers, peers, classmates, siblings, and other family members within the accessible contexts of school, home, and community. In this sense, Simeonsson et al. (2001) asserted that participation may be facilitated or restricted by the nature and accessibility of the environment for children with disabilities. It is confirmed that improvement of participation in leisure activities must include changes in the environment (Badia et al., 2011).

Finally, the values of test-retest reliability were fairly acceptable and the results concerning intensity were quite similar to those obtained by the sample of children and adolescents with and without disability in the validation study of CAPE in the Netherlands (Bult et al., 2010).

The main contribution of the study was to make available a valid and reliable version of CAPE for its use in Spain. This has allowed us to know the participation profile of Spanish children and adolescents with CP with all GMFCS levels represented, and varying levels of intellectual impairment.

The cross-cultural adaptation of CAPE allows us to carry out comparative studies and to exchange information in other languages, countries, and cultures that also have some version of the instrument. Even considering the existence of a version in Spanish of CAPE (Colón et al., 2008), we underline that the Puerto Rico version was validated for the Spanish-speaking population in Puerto Rico, a country with markedly different cultural and geographical aspects from Spain, and hence, the need to obtain a version of CAPE validated in the Spanish context.

One of the limitations of the study is the use of a convenience sample, which, although heterogeneous and representative of the population of children and adolescents with CP in Spain, does not proceed from a study of the population record of CP. Failure to have included children and adolescents with other disabilities may be considered a limitation of the study, even though CP is most common physical disability in childhood and which predominates in rehabilitation centers in Spain. In addition, although we found positive correlations between participation and QoL, providing construct validity to the Spanish version of CAPE, is the first time that is used CAPE as a measuring instrument, correlating participation and QoL results and we have not investigated whether this pattern played in the group of typically developing children. In this sense it is essential to develop future research including children with and without disabilities to assess whether these findings are confirmed in different populations. Another limitation is that we did not provide information about the Preference for Activities of Children (PAC), a complementary tool of CAPE, which is used to measure activity preference (King et al., 2006a).

Neither could we establish correlations between the participation in leisure activities and the results of adaptive behavior or environmental needs, because these parameters were not assessed in the sample, as was done in other studies (Bult et al., 2010; King et al., 2003; King et al., 2011; Palisano et al., 2011).

CONCLUSION

The results obtained in this study support the validity of the Spanish version of CAPE to assess the participation in leisure activities in the Spanish context. It contributes to the advancement of the study of participation in leisure activities from the viewpoint of children and adolescents with and without CP and from a multidimensional perspective of the construct participation. This study falls within the framework in the incipient efforts to develop robust measurement instruments, based on the social model of disability, and to provide information about participation in leisure activities of children and adolescents with CP, which will serve to plan interventions and guide the processes of changing and improving the provision of such services.

Key messages

- The Spanish version of CAPE is a valid and reliable instrument to assess the participation in leisure activities out-of-school in Spain in children and adolescents with and without CP between aged 8 to 18 years old.
- Children and adolescents with CP in Spain participate less in out-of-school activities than typically developing children and adolescents.
- Participation is an important outcome measure, which should be taken into account when planning services targeting children and adolescents with and without CP.

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CAPÍTULO 3

PATTERNS AND PREDICTORS OF PARTICIPATION IN LEISURE ACTIVITIES OUTSIDE OF SCHOOL IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY

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ABSTRACT

This study analyzed the patterns and predictors of participation in leisure activities outside of school of Spanish children and adolescents with Cerebral Palsy (CP). Children and adolescents with CP (n = 199; 113 males and 86 females) participated in this cross-sectional study. Their mean age was 12.11 years (SD = 3.02; range 8-18 years), and they were evaluated using the Spanish version of the Children's Assessment of Participation and Enjoyment (CAPE). Means, standard deviations and percentages were used to characterize the profile of participation, and linear regression analyses were employed to assess associations between the variables (child, family and environmental factors) and the diversity, intensity and enjoyment of participation. Children and adolescents with CP reported low diversity and intensity of participation and high levels of enjoyment. Participation in leisure activities outside of school was determined more by child and environmental factors than by family ones.

Key words: participation; leisure activities; cerebral palsy; children; adolescents

INTRODUCTION

Cerebral palsy (CP) is the most common cause of disability in children, and it is estimated that there were 400,000 people living with CP in the European Union before its recent enlargement, with 10,000 new cases occurring each year (McManus et al., 2006). Because of their motor problems, children and adolescents with CP experience participation restrictions and limitations of physical activities (Donkervoort, Roebroeck, Wiegerink, van der Heijden-Maessen, & Stam, 2007; Imms, Reilly, Carlin, & Dodd, 2008).

Growing interest has emerged among rehabilitation professionals in moving beyond simply improving bodily functions and functional abilities in children with disabilities, and services now attempt to optimize social participation as one of the major goals (Hammal, Jarvis, & Colver, 2004; Simeonsson, Carlson, Huntington, McMillen, & Brent, 2001).

The International Classification of Functioning, Disability and Health (ICF) (World Health Organization, 2001) defines participation as involvement in life situations and defines environmental factors as the social, attitudinal and physical environments in which people live. When these factors have a positive influence on an individual's participation, they are called facilitators, and when they have a negative influence, they are called barriers (Lawlor, Mihaylov, Welsh, Jarvis, & Colver, 2006).

The UN Convention on the Rights of the Child, to which all European Union countries are signatories, states that disabled children should enjoy a full and decent life, in conditions which ensure dignity, promote self-reliance and facilitate the child's active participation in the community (article 23). The child shall not be denied the right, in community with other members of his or her group, to enjoy his or her own culture (article 30) (United Nations, 2006).

However, in comparison with peers without disabilities, adolescents and young adults with disabilities display less diverse participation, greater participation in passive recreational activities (such as watching television), and less social participation (King, Petrenchik, DeWit, McDougall, Hurley, & Law, 2010).

It is widely acknowledged that participation in leisure activities contributes to the physical, mental, and social health of individuals, families, and communities. Leisure has been defined as the time designated for freely chosen activities, performed when not involved in self-care, school or work, because they are enjoyable (Majnemer et al., 2010). Participation in leisure activities leads to positive self-esteem, a sense of competence, and higher academic achievement. In the appositive, low levels of participation at a young age can adversely impact motivation to participate at later ages, and the risk for developing obesity is higher among children who are physically inactive (Annesi, 2004; Kristensen et al., 2008; Parfitt & Eston, 2005).

It is important to understand the relative importance of the factors that may influence the leisure and recreation participation of children with physical disabilities and the paths by which these factors have their effects. Recent studies have highlighted several possible predictors of children's participation: environmental factors, such as physically accessible and welcoming environments; family factors, such as income and family functioning; and child factors, such as cognitive ability, preferences and social skills (King, Lawm, King, Rosenbaum, Kertoy, & Young, 2003; Shikako-Thomas, Majnemer, Law, & Lach, 2008).

In Spain, the latest official data indicate that there are 82,200 people with CP aged over 6 years, and approximately 2,000 between 0-5 years (Instituto Nacional de Estadistica, 2008). In Spain, models for the care of people with disabilities are not based on a medical model but instead are based on biopsychosocial models that orient services and public care for people with disabilities. For children with CP, it is vital to assess not only the child's disability but also the extent to which the child is able to and enjoys participating in meaningful domains, including leisure activities (Parush & Rihtman, 2008). In Spain, as in some other European countries, the US and Canada, rehabilitation services have considered other outcomes, such as participation and quality of life, because they are potentially modifiable variables.

The purpose of this study is to evaluate the patterns of participation in leisure activities outside of school of Spanish children and adolescents with CP, aged between 8-18 years. Specifically, this study has three objectives: 1) to describe comprehensively the leisure activities outside of school in which Spanish children and adolescents with CP participate (diversity), how often they participate (intensity) and whether they enjoy these activities; 2) to analyze the percentage of activities in which the children participate in each domain and the type of activity; and 3) to identify variables related to participation in leisure activities outside of school of children and adolescents with CP who live in Spain.

METHODS

Participants

A cross-sectional analytical design was used. Children and adolescents with CP of any severity of motor function, aged between 8 and 18 years old, were recruited from 16 Associations Caring for People with Cerebral Palsy and Related Disabilities (ASPACE) that were located in diverse regions of Spain.

One hundred and ninety nine children and adolescents participated. One hundred and thirteen were males (56.8%) and 86 were females (43.2%), with a mean age of 12.11 years (SD = 3.02, range 8-18 years). Of the total sample, 60.8% walked with or without support and 39.2% used a manual wheelchair or electric wheelchair. While 19.1% of the sample did not show Intellectual Impairment, 42.7% had severe impairment. Most children (72.4%) attended regular school. The characteristics of the family and context are shown in Table 1.

Table 1. Characteristics of the sample of children and adolescents with CP (n=199)

Variable	n (% of valid n)
Gender	
Male	113 (56.8)
Female	86 (43.2)
Age (M = 12.11, SD = 3.02, range = 8–18 years)	
8-12	115 (57.8)
13-18	84 (42.2)
GMFCS	
Level I	50 (25.1)
Level II	34 (17.1)
Level III	37 (18.6)
Level IV	27 (13.6)
Level V	51 (25.6)
Intellectual Impairment	
None	38 (19.1)
Mild	43 (21.6)
Moderate	33 (16.6)
Severe	85 (42.7)
Type of schooling	
Regular	144 (7.4)
Special	52 (26.1)
Missing data	3 (1.5)
Gender of caregiver	
Male	58 (29.1)
Female	138 (69.3)
Missing data	3 (1.5)
Age of caregiver (M = 43.34, SD = 5.86, range = 28–61 years)	
28-40	61 (30.7)
41-61	123 (61.8)
Missing data	15 (7.5)
Family type	
Two-parent	165 (82.9)
Single-parent	30 (15.1)
Missing data	4 (2.0)
Number of siblings	
None	49 (24.6)
1	93 (46.7)
2	44 (22.1)
3 or more	9 (4.5)
Missing data	4 (2.0)
Parental educational level	
Elementary school	54 (27.1)
High School	70 (35.2)
University	68 (34.2)
PhD	3 (1.5)
Missing data	4 (2.0)
Total family income (Euros € per month)	
Less than 1000 €	29 (14.6)
Between 1000-2000 €	86 (43.2)
More than 2000 €	77 (38.7)
Missing data	7 (3.5)
Area of domicile	
Population of less than 1000	22 (11.1)
1000-5000	24 (12.1)
Population of more than 5000	149 (74.9)
Missing data	4 (2.0)

Measures

Spanish version of Children´s Assessment of Participation and Enjoyment (CAPE)

The Children´s Assessment of Participation and Enjoyment (CAPE) (King et al., 2007) questionnaire was used to assess the profile of children's and adolescents' participation in leisure and recreation activities outside of mandated school activities. It is a reliable and valid self-report measure of participation for children and youth ages 6 to 21 that includes both formal and informal domains and five activity types: recreational, active physical, social, skill-based, and self-improvement activities (King et al., 2004; King et al., 2006).

CAPE includes 55 activities and is completed as a questionnaire or by interview. The child is asked whether an activity was performed during the past 4 months (diversity), and if so, how often (intensity), with whom, where and how much the child enjoyed that activity (enjoyment of participation). This instrument provides three levels of scoring: (1) overall participation scores, (2) domain scores reflecting participation in formal (15 items) and informal (40 items) activities, and (3) scores reflecting participation in recreational (12 items), active physical (13 items), social (10 items), skill-based (10 items), and self-improvement (10 items) activity types.

The diversity score is calculated by adding the number of activities performed. The 7 response options for how often an activity was performed (intensity score) range from one time in the past 4 months (score 1) to one time a day or more (score 7) and is calculated by adding the 'how often' scores and dividing by the total number of possible activities (King et al., 2004). Intensity scores represent the average frequency of the total number of activities possible, rather than the average frequency of the activities in which the child actually takes part, and provides a relative indicator of participation frequency, which is useful for comparing participation across activity types, groups of individuals, etc. (Imms et al., 2008). Enjoyment is measured on a five point scale ranging from one (not at all) to five (love it) (King et al., 2004). For the purpose of the present study, the diversity, intensity and enjoyment CAPE scores were used.

Similar to the original version of the CAPE, the Spanish version has demonstrated good psychometric properties (Badia et al., 2011). The Internal consistency in the Spanish version measured by alpha values ranges from 0.54 to 0.69 for the five CAPE activity types, which is considered moderate, and these scores are actually superior to the ones found in the original CAPE version (King et al., 2004).

Gross Motor Function Classification System (GMFCS)

GMFCS was used to categorize gross motor function. It is an age-specific scheme designed for children with CP that describes their motor performance on the basis of their functional abilities and their need for assistive technology and wheeled mobility. Functional levels range from I (independent gross motor function with few limitations) to V (complete dependence for all motor activities). The psychometric properties of GMFCS have been thoroughly tested and reported and include evidence supporting their content validity, construct validity, inter-rater reliability and test–retest reliability (Palisano et al., 1997, 2000; Wood & Rosenbaum, 2000).

Questionnaires on demographic variables

The following child factors were examined: sex (boys vs. girls), age (children: 8-12 years old; adolescents: 13-18 years old) and intellectual ability. Intellectual impairment was classified as follows: none or mild (IQ>70), moderate (IQ 50-70), and severe (IQ<50).

The following family and environmental factors were examined: type of school (regular vs. special), family cohesion (biparental vs. monoparental), Gender of caregiver (male vs. female), family income, parental educational level, number of siblings and area of domicile.

Procedure

Ethical approval for the study was obtained from the Bioethical Committee of the University of Salamanca. Written informed consent was obtained from all parents and from all children aged 12 years and above. Children and parents were assured that all necessary privacy measures would be taken during the use and storage of their data.

Before collecting the data, two research team members visited each ASPACE center and distributed information about the research. In those centers that expressed interest in collaborating, a research assistant (psychologist or social worker experienced in providing services to children and families with CP) was selected. This research assistant received formal training from one of the members of the research team.

The research assistants contacted the chosen families, and, after both the children and parents had signed and returned the written consent form, made an appointment for the interview at the ASPACE center. Data collection took place between December 2008 and June 2010.

A research assistant conducted a semi-structured interview with the child and parent. The CAPE was completed by the child/adolescent or by a parent if the child/adolescent was unable to contribute. The duration of the CAPE interview was approximately 1 hour.

Of the 199 children and adolescents with CP, 114 (57.3%) completed the CAPE questionnaire independently or received some assistance. In cases of severe intellectual impairment (42.7%), the parents completed the CAPE by proxy. Additionally, parents were asked to complete demographic questionnaires.

In each ASPACE center, the psychologist provided information about the intellectual ability of the child, and the physical therapists provided information about the GMFCS level of each child/adolescent.

Statistical analyses

Statistical analyses were performed using the SPSS for Windows software program, version 17.0 with a significance level of $p < .05$.

To determine diversity scores, we calculated the percentage of activities in which the children participated in relation to the number of activities of the domain, taking into consideration that the number of activities in the CAPE domains is different, and then made appropriate comparisons. In addition, to determine which activities had the highest levels of participation, the percentage of the sample that participated in each activity in the formal and informal CAPE domains was calculated.

Subsequently, we performed corresponding ANOVA or T tests separately for each CAPE domain and type of activity to test whether there were differences in diversity, intensity or enjoyment in participation.

To determine the factors that most influenced participation, three multiple regression analyses were conducted separately for each domain and type of activity. In the first regression model, child factors (sex, age, GMFCS level, intellectual impairment) were included. In the second regression model, family factors (age, gender, and educational level of the caregiver, type of family and number of siblings) were included. In the third regression model, environmental factors (income, type of school, size of domicile) were included.

RESULTS

The results are divided into two main sections. In the first section, we present a profile of the participation in leisure activities outside of school of children and adolescents with CP. We include information regarding how many leisure activities children participated in, in which types of leisure activities they participated, how often and how much enjoyment they reported, and finally, activities that had the highest participation levels. In the second section, we determine predictors of participation and individual differences in diversity, intensity and enjoyment. Finally, we determine whether child, family or environmental factors best predicted child participation.

Profile of participation in leisure activities

Overall, Spanish children and adolescents with CP participated in a mean of 20.93 activities (SD=8.91, range of 2 to 41), which represented 38% of the 55 CAPE activities.

Table 2. Descriptive results in Diversity, Intensity and Enjoyment of participation in CAPE domains (N=199)

CAPE Domain	Diversity Mean (SD) Proportion (SD)	Intensity Mean (SD)	Enjoyment Mean (SD)
General (55 items)	20.93 (8.91) 0.38 (0.16)	1.79 (0.77)	3.89 (0.56)
Formal (15 items)	3.42 (2.35) 0.23 (0.16)	1.17 (0.76)	3.83 (0.80)
Informal (40 items)	17.50 (7.241) 0.44 (0.18)	2.04 (0.86)	3.89 (0.57)
Recreational (12 items)	6.25 (2.87) 0.52 (0.24)	2.63 (1.18)	3.91 (0.65)
Social (10 items)	7.01 (2.35) 0.70 (.25)	2.87 (0.94)	4.04 (0.67)
Self-improvement (10 items)	3.07 (2.37) 0.31 (0.24)	2.00 (1.04)	3.35 (0.85)
Skill-based (10 items)	2.54 (1.88) 0.25 (0.19)	1.41 (0.88)	3.93 (0.88)
Active physical (13 items)	2.06 (2.02) 0.16 (0.16)	1.03 (0.75)	4.03 (0.81)

CAPE Scoring: Diversity (number of different activities)= number of activities involved compared to the total number of activities (check in manual). Intensity (how often)= 1- 1x/4 months, 2- 2x/4 months, 3- 1x/month, 4- 2-3x/month, 5- f1x/week, 6- 2-3x/week, 7- daily. Enjoyment (how much do you enjoy)= 1- not at all, 2- somewhat, 3- pretty much, 4- very much, 5- love it.

For diversity, results indicated proportionately greater participation [t(198)= 20.11, p<.001] in informal activities (44% of a possible 40 activities) than in formal activities (23% of a possible 15 activities). Children and adolescents with CP participated in an average of approximately 3 formal activities and 17 informal activities (see Table 2). Across the five activity types, we also obtained significant differences [F (4.792) = 402.70, p <.001]. Children took part in a greater proportion of social activities (70% of a possible 10 activities) and recreational activities (52% of a possible 12 activities) than in less skill-based activities (25% of a possible 10 activities), self-improvement activities (31% of a possible 9 activities) or active physical activities (16% of a possible 13 activities). In the last 4 months, children and adolescents with CP had participated in an average of 7 social activities, 6 recreational activities, 3 self-improvement and 2 skill-based and active physical activities (see Table 2).

For intensity of participation, the results showed a mean of approximately 2 points (twice in the last four months). Differences between the two types of activities (formal and informal) were also significant [t (186) = 16.54, p <.001]. The intensity of participation in formal activities was circa 1 point (once in the last four months), and the intensity of participation in informal activities was approximately 2 (twice in the last four months) (see Table 2). With respect to the five activity types, significant differences were also obtained [F (4,480) = 168.63, p <.001]. Mean intensity was significantly higher for social and recreational than for other types of activities. As demonstrated in Table 2, while the mean intensity in the social and recreational activities was 3 (once a month), the rest of the activity types received scores equal to or less than 2 (twice in the last four months or less).

With regard to enjoyment, Table 2 shows that, in general, children and adolescents enjoyed the activities in which they participated, with a mean score of 4 points (very much). No significant differences in the levels of enjoyment of formal and informal activities were found [t (186) = 1.31, p = .19]. With regard to the five activity types, significant differences were obtained [F (4,480) = 33.43, p <.001]. Levels of enjoyment were significantly lower for self-improvement activities than for other types of activities. While in all other activity types the level of enjoyment was around 4 points (very much), scores for enjoyment of self-improvement activities averaged around 3 points (pretty much).

When we analyzed the percentages of participation in different activities, we found that among the informal activities (see Table 3), more than half of the sample participated in 8 specific social activities (Hanging out, Visiting, Entertaining others, Listening to music, Going to a party, Talking on the phone, Going on a full day outing, Going to a live event), 6 specific recreational activities (Watching TV or a rented movie, Going for a walk or hike, Playing with things or toys, Doing crafts, drawing or coloring, Playing board or card games, Playing computer or video games) and 2 specific self-improvement activities (Shopping, Doing homework).

Table 3. Percentage of the children and adolescents participating in informal activities (n=199)

Informal Activities	Type of Activity	Percentage
Hanging out	Social	92.5
Visiting	Social	87.9
Entertaining others	Social	85.9
Watching TV or a rented movie	Recreational	83.4
Going for a walk or hike	Recreational	81.4
Listening to music	Social	80.4
Going to a party	Social	78.9
Playing with things or toys	Recreational	75.4
Doing crafts, drawing or coloring	Recreational	65.3
Shopping	Self-improvement	59.8
Playing board or card games	Recreational	58.8
Talking on the phone	Social	57.8
Going on a full-day outing	Social	56.8
Playing computer or video games	Recreational	54.8
Doing homework	Self-improvement	54.8
Going to a live event	Social	51.8
Going to the movies	Social	48.2
Reading	Self-improvement	44.2
Playing with pets	Recreational	40.2
Doing puzzle	Recreational	39.7
Playing on equipment	Recreational	37.7
Writing letters	Self-improvement	34.7
Collecting things	Recreational	33.2
Playing games	Active Physical	32.2
Doing pretend or imaginary play	Recreational	31.2
Dancing	Skill-based	30.7
Doing a chore	Self-improvement	26.1
Bicycling	Active Physical	24.1
Taking care of a pet	Recreational	23.6
Going to the public library	Self-improvement	22.1
Doing individual physical activities	Active Physical	22.1
Doing water sports	Active Physical	20.6
Playing non-team sports	Active Physical	13.1
Writing a story	Self-improvement	12.6
Making food	Social	12.1
Gardening	Active Physical	9
Doing snow sports	Active Physical	8
Doing voluntary work	Self-improvement	5
Fishing	Active Physical	4.5
Doing a paid job	Active Physical	1.5

More than 50% of the sample also participated in 2 formal skill-based activities (Doing Gymnastics and Swimming), and less than 10% participated in 1 of the formal skill-based activities (Taking art lessons) and in 1of the active physical activities (Doing martial arts) (see Table 4).

Table 4. Percentage of the children and adolescents participating in formal activities (n=199)

Formal Activities	Type of Activity	Percentage
Doing gymnastics	Skill-based	59.8
Swimming	Skill-based	55.8
Participating in school clubs	Active Physical	28.6
Doing team sports	Active Physical	27.6
Doing a religious activity	Self-improvement	27.1
Playing a musical instrument	Self-improvement	25.6
Horseback riding	Skill-based	23.1
Getting extra help for schoolwork	Self-improvement	20.6
Participating in community organizations	Skill-based	19.6
Learning to sing	Skill-based	12.1
Taking music lessons	Skill-based	10.6
Racing or track and field	Active Physical	10.6
Learning to dance	Skill-based	10.6
Taking art lessons	Skill-based	6.5
Doing martial arts	Active Physical	4

Predictors of the participation in leisure activities outside of school

Regression analyses revealed that the best predictors for overall diversity of participation were the child factors [F (4,193) = 36.96, p<.001], which explained 43% of the score differences. A model that included environmental factors was also significant [F (3.186) = 22.66, p <.001], although it explained only 27% of the score variability. Family factors had practically no impact on diversity of participation (R^2 between .01 and .07).

Table 5. Regression analysis including child, family and environmental variables that predict the diversity of participation

	Overall	Formal	Informal	Recreational	Social	Self-improvement	Physical	Skill-based
Child factors	β	β	β	β	β	β	β	β
Gender (0: boy, 1: girl)	.04	.06	.04	.04	.04	.02	-.04	.11
Age	-.10	-.03	-.11	-.22***	-.03	.04	-.02	-.10
Intellectual impairment	-.42***	-.26***	-.44***	-.40***	-.25**	-.43***	-.21**	-.09
GMFCS	-.39***	-.30***	-.37***	-.32***	-.09	-.31***	-.46***	-.26**
R²	.43	.20	.43	.38	.09	.33	.28	.10
Family factors	β	β	β	β	β	β	β	β
Age's caregiver	-.09	-.06	-08	-.16	-.02	.08	.02	-.06
Caregiver's gender (0: Male, 1: Female)	-.01	.02	-.01	-09	-.02	.06	-.05	.04
Parental Educational level	.04	.08	.02	.06	-.01	.09	.08	.15
Family cohesion (0: biparental, 1: monoparental)	.07	.07	.06	.07	.01	.09	.06	.12
Number of siblings	-.07	-.12	-.06	-.01	-.10	-.01	-.10	-.13
R²	.02	.03	.02	.03	.01	.03	.02	.07
Environmental factors	β	β	β	β	β	β	β	β
Income	-.03	.05	-.05	-.06	.02	.00	-.01	.05
Size of domicile	.08	.17*	.04	.01	.11	.08	.05	.13
Type of school (0: Regular, 1: Special)	-.51***	-.31***	-.52***	-.52***	-.23**	-.45***	-.21*	-.12
R²	.27	.14	.28	.28	.07	.22	.05	.04

***p<.001, **p<.01, *p<.05

Moreover, child-related variables, unlike environmental variables, explained differences in informal, recreational and self-improvement activities (see Table 5). Specifically, the GMFCS level and Intellectual impairment significantly predicted differences in the diversity of participation in all domains and in the types of activities - except social activities (GMFCS level) and skill-based activities (Intellectual impairment). Children and adolescents with lower levels of gross motor function and with less intellectual impairment had the highest mean diversity scores. Furthermore, children and adolescents who attended a regular school participated in more activities of all types than participants who attended a special school.

The results for intensity of participation (table 6) were quite similar to those for diversity of participation. Again, the child factors were the strongest predictors, explaining 40% of the variance in the overall intensity [F (4,193) = 32.01, p< .001]. A model that included environmental factors was also significant [F (3.186) = 19.66, p <.001], although only 24% of the variability was explained in this case. Again, family factors had practically no impact on the intensity of participation (R^2 between .02 and .05).

Finally, child-related variables explained differences in the intensity of participation in informal activities (37%) and in self-improvement activities (36%), whereas environmental variables explained 24% and 27% of the variance in these respective domains (see Table 6). Specifically, the GMFCS and Intellectual impairment significantly accounted for different intensities of participation in all domains and types of activities except social activities (GMFCS level) and skill-based (Intellectual impairment). Again, children and adolescents with lower levels of gross motor function and with less intellectual impairment had the highest mean intensity scores. Further, children and adolescents who attended a regular school participated with higher intensity in all the types of activities than did participants who attended a special school.

Table 6. Regression analysis including child, family and environmental variables that predict the intensity of participation

	Overall	Formal	Informal	Recreational	Social	Self-improvement	Physical	Skill-based
Child factors	β	β	β	β	β	β	β	β
Gender (0: boy , 1: girl)	.01	.02	.00	.01	.00	.00	-.10	.06
Age	-.10	-.01	-.12*	-.24***	-.03	.04	.00	-.09
Intellectual impairment	-.37***	-.25***	-.37***	-.28***	-.18*	-.44***	-.18*	-.11
GMFCS	-.40***	-.32***	-.37***	-.31***	-.09	-.34***	-.47***	-.24**
R^2	.40	.21	.37	.28	.05	.36	.28	.10
Family factors	β	β	β	β	β	β	β	β
Caregiver's age	-.11	-.03	-.12	-.19*	-.04	.05	.00	-.08
Caregiver's gender (0: Male, 1: Female)	-.01	.06	-.03	-.08	-.02	.03	-.07	.08
Parental Educational level	.04	.09	.01	.05	-.04	.09	.07	.15
Family cohesion (0: biparental, 1: monoparental)	.06	.02	.08	.04	-.05	.09	.10	.05
Number of siblings	-.08	-.14	-.06	-.03	-.07	-.05	-.13	-.10
R^2	.03	.03	.03	.04	.02	.02	.03	.05
Environmental factors	β	β	β	β	β	β	β	β
Income	-.09	.02	-.12	-.10	-.06	-.07	-.07	.01
Size of domicile	.09	.18*	.05	.00	.13	.12	.01	.16
Type of school (0: Regular, 1: Special)	-47***	-.32**	-.47***	-.40***	-.16*	-.50***	-.18*	-.17*
R^2	.24	.15	.24	.18	.04	.27	.03	.06

***p<.001, **p<.01, *p<.05

Concerning enjoyment, models with the same variables used to predict intensity and diversity of participation (child, family and environmental) explained only very low percentages of the variance (between 1% and 6%) in all domains and types of activity in the CAPE. Regression results for enjoyment are available in the online supporting Information.

DISCUSSION

The aim of this study was to find patterns of participation in leisure activities outside of school in a sample of Spanish children and adolescents with CP. In addition, we examined the percentage of activities in which the children participated and predictors of participation in leisure activities including child, family and environmental factors.

Our findings suggest that the Spanish children and adolescents with CP have low diversity and intensity of participation; however, they reported high levels of enjoyment. This is consistent with other studies that suggest that children with CP and other physical disabilities experience lower levels of participation in extracurricular activities when compared to their peers, which may have implications for skill development, fitness, acceptance within peer groups, developing friendships, self-confidence and autonomy (Engel-Yeger, Jarus, Anaby, & Law, 2009; Fauconnier et al., 2009; Imms et al., 2008; King et al., 2006; King, Petrenchik, Law, & Hurley, 2009; Law et al., 2006; Michelsen et al., 2009; Pratt, Baker, & Gaebler-Spira, 2008; Scehenker, Coster, & Parush, 2005).

In this study, children and adolescents with CP participated more frequently and in a wider range of informal than formal activities and participated mainly in social and recreational activities. Concurrent with these results, previous studies have shown that children and young people with disabilities participate less often in structured activities and more often in quiet, sedentary activities (Imms et al., 2008; King et al., 2009; Klaas, Kelly, Gorzkowski, Homko, & Vogel, 2010; Law et al., 2006). In fact, participation in informal activities is less likely to be adversely affected by environmental barriers, which may explain our results. The involvement of the Spanish children and adolescents with CP in active-physical, self-improvement and skill-based activities was diminished compared with other leisure activities, a pattern found in various studies (Bult et al., 2010; Imms et al., 2008; Law et al., 2006; Majnemer et al., 2008; Orlin et al., 2010). One Spanish study recently showed that youngsters and adults with developmental disabilities, including CP, participated to a greater extent in social activities and to a lesser extent in physical activities, confirming that this pattern may be observed not only during childhood and adolescence but also throughout life (Badia, Orgaz, Verdugo, & Ullán, 2012). However, although the diversity and intensity of participation of Spanish children and adolescents with CP was reduced, their levels of enjoyment were high for both formal and informal activities, as they were for all types of activities except for self-improvement. Similar results have been reported by an Australian study in which children with CP, aged between 10 and 12 years, presented high levels of enjoyment for all types of activity (Imms et al., 2008).

When we analyzed the percentage of participation in formal and informal activities, we observed that "Hanging out" and "Visiting" were engaged in by a higher percentage of children, which could reflect cultural norms. Published reports of the activities of typically developing Spanish children and adolescents suggest that such activities are also commonly performed by typically developing children (Marí-Klose, Marí-Klose, Vaquera, & Argeseanu, 2010). Hanging out also achieved the highest participation rate among Spanish youngsters and adults with Developmental Disabilities, including CP (Badia et al., 2012). The SPARCLE study, which included a large sample of children with CP, found substantial variations in the patterns of participation between children in seven European countries (Fauconnier et al., 2009).

The identification of predictors of participation is important because it helps guide effective health promotion strategies and policy initiatives that enable children with CP to participate more fully in society and to achieve greater life satisfaction. This study has shown that predictors of the diversity and intensity of participation in leisure activities of children and adolescents with CP are mainly variables related to child-disability - specific levels of gross motor function and Intellectual ability - and environmental factors associated with the type of school. Our family variables did not play a significant role.

Children with better motor and intellectual functioning who attend regular school participate in a wider range of activities and with a higher intensity. The strong associations between severity of motor impairment or intellectual impairment and lower participation support the results of other studies (Beckung & Hagberg, 2002; Fauconnier et al., 2009; Kerr, McDowell, & McDonough, 2007; Maher, Williams, Olds, & Lane, 2007; Majnemer et al., 2008; Morris, Kurinczuk, Fitzpatrick, & Rosenbaum, 2006). It has been suggested that the inclusion of children with disabilities in non-segregated schools could lead to improved participation in leisure activities and more social support networks outside of school (Scehenker et al., 2005). In fact, the majority of the children and adolescents with CP in our study attended regular school, confirming that education of students with special educational needs in Spain can take place within regular school.

Surprisingly, family factors were not strongly associated with participation, as has been previously reported (Palisano et al., 2011). However, in our study, we considered only socio-demographic data from the family, and we did not include variables such as family cohesion, parenting stress or coping styles. Indeed, the high proportion of the variance not explained by our models may indicate that some important predictors may not have been included in the analyses.

Finally, none of the predictors included in our study determined the degree of enjoyment of activities. Despite a growing literature on the subject of participation in children with CP, little attention has been devoted to factors related to levels of enjoyment of these activities, perhaps confirming that participation is a complex and multi-faceted construct (Bult, Verschuren, Jongmans, Lindeman, & Ketelaar, 2011). Only one study mentions predictors of enjoyment levels in children with CP (Majnemer et al., 2008). According to these authors, higher IQ was found to predict reduced enjoyment in leisure activities. A higher IQ might lead to a greater awareness of difficulties and thus result in decreased enjoyment from the activity itself (Parush & Rithman, 2008).

In summary, the results of this study indicate that, although children and adolescents with CP in Spain participated in few leisure activities with low frequency, they exhibited high levels of enjoyment. In fact, a major proportion of our sample had severe levels of motor and intellectual impairment, which may indicate that the severity of the disability is not an important predictor of the satisfaction and pleasure received from participating in leisure activities. In this sense, our results are encouraging, and they seem to point out that low diversity and intensity scores by themselves do not necessarily mean that participation is restricted. Instead, a child may successfully participate in only a few preferred activities that they actually enjoy.

One of the limitations of our study is that we did not analyze how an unsupportive environment may hinder participation in leisure activities, which may be an important predictor of participation (Badia et al., 2012). Additionally, we did not describe preferences for leisure activities. Knowledge of when and how often children are not engaged in preferred activities can be used to explore why and to identify strategies for helping families to access preferred activities or to select alternatives (King et al., 2004). Future research could identify preferences for activities in Spanish children and adolescents with CP, as well as barriers and environmental facilitators for participation, in order to encourage the fundamental right of children with disabilities to participate in their preferred leisure activities.

Finally, lobbying for 'disability friendly' policies, which guarantee that environments are accessible, will ensure that children and adolescents with CP are able to gain greater access to activities in the community setting. Such policies could help children and adolescents with CP to participate in meaningful activities of their own choosing, which is essential for promoting their health, skill competencies, personal autonomy, community inclusion and life satisfaction.

Highlights

- This study analyzed participation in leisure activities outside of school of Spanish children and adolescents with CP.
- Children and adolescents with CP participated in few activities with low frequency; however, they reported high levels of enjoyment.
- GMFCS level, intellectual impairment and type of school were the variables most related to various activity types across participation diversity and intensity.

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Declaration of interest: The authors declare no conflict of interest.

Online supporting information

Regression analysis including child, family and environmental variables that predict the enjoyment of participation

	Overall	**Formal**	**Informal**	**Recreational**	**Social**	**Self-improvement**	**Physical**	**Skill-based**
Child factors	β	β	β	β	β	β	β	β
Gender (0: boy , 1: girl)	.02	-.06	.03	-.07	.11	.07	-.14	-.01
Age	-.08	-.03	-.08	-.12	-.07	.00	.02	-.06
Intellectual impairment	.01	.20**	-.03	-.11	-.09	.13	-.01	.17*
GMFCS	-.05	-.08	-.03	-.02	-.04	.04	-.12	-.12
R²	.01	.04	.01	.01	.03	.03	.03	.04
Family factors	β	β	β	β	β	β	β	β
Caregiver's age	.01	-.04	.01	-.02	.05	-01	.00	-.04
Caregiver's gender (0: Male , 1: Female)	-.01	-.04	.01	-.02	-.02	.02	.06	-.06
Parental Educational level	.18*	.18*	.15	.11	.14	.19*	.06	.22**
Family cohesion (0: biparental, 1:monoparental)	.04	.06	.02	.01	-.03	.03	.08	.11
Number of siblings	.09	.14	.05	.06	-.01	.09	.05	.09
R²	.04	.05	.03	.01	.03	.04	.02	.06
Environmental factors	β	β	β	β	β	β	β	β
Income	-.03	.07	-.04	-.04	-.03	.06	-.07	.08
Size of domicile	.12	.10	.11	.16*	.05	.04	.09	.21**
Type of school (0: Regular, 1: Special)	-.06	.14	-.09	-.12	-.17*	.17*	-.12	.08
R²	.02	.03	.02	.04	.03	.03	.02	.02

***p<.001, **p<.01, *p<.05

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CAPÍTULO 4

THE INFLUENCE OF PARTICIPATION IN LEISURE ACTIVITIES ON QUALITY OF LIFE IN CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY

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Submmited for publicaction

ABSTRACT

Background: Participation is increasingly considered as one of the primary aims of paediatric rehabilitation and is believed to contribute to quality of life (QoL) Aims: The purpose of this study was to confirm if participation in leisure activities affects the QoL domains in children and adolescents with cerebral palsy (CP) in Spain. Methods: Participation: 206 children and adolescents with CP (and their parents), 115 boys and 91 girls, mean age of 11.96 years (SD = 3.00; range 8-18 years). Distribution according to the Gross Motor Function Classification System (GMFCS) was 24.3% Level I, 18% Level II, 18% Level III, 12.6% Level IV, and 27.2% Level V. Participation in leisure activities was assessed using the Spanish version of Children's Assessment of Participation and Enjoyment (CAPE) and QoL using the KIDSCREEN parents' version. Results: Diversity, intensity and enjoyment of participation explained the levels of QoL in the physical well-being, psychological well-being, autonomy, parent relation & home life and social support & peers domains, independently of gender, age and level of impairment (motor and intellectual). We also found that intensity and enjoyment of participation in informal activities had more influence on the different QoL domains. The most influenced domain by the enjoyment of participation in all domains and types of CAPE activities was psychological well-being. Conclusion: The participation in leisure activities had a positive effect on the QoL of the Spanish children and adolescents with CP.

Keywords: participation; quality of life; cerebral palsy; children; adolescents.

INTRODUCTION

Disabled children have the same aspirations as all children, hoping for health, security, respect, opportunities to learn skills, meaningful occupation and the possibility of contributing to the lives of others (McConachie, Colver, Forsty, Jarvis, & Parkinson, 2006). In this sense, the UN Convention on the Rights of the Child states that disabled children should enjoy a full and decent life, in conditions which ensure dignity, promote self-reliance and facilitate the child's active participation in the community (article 23) (United Nations, 2006). The content of this statement cover two elements considered essential in the current perspective of caring for children and adolescents with disabilities: objective information of what the child does, i.e. participation, and what child feels about their life, i.e. quality of life (QoL).

Participation is defined by the International Classification of Functioning, Disability and Health (ICF-CY) as involvement in life situations (World Health Organization, 2004). Typical examples of life roles that children participate in include social relationships with family and friends, domestic chores, learning at school, and leisure and recreational activities at home and in the community (Majnemer et al., 2010). Adolescents with physical disabilities have described participation as having fun, feeling successful, doing and being with others and doing things by themselves, highlighting both subjective and objective components of participation as key to their experiences (Heah, Case, McGuire, & Law, 2007). Participation leads to life satisfaction as well as a sense of competence and is essential for psychological and emotional well-being and skill development (Engel-Yeger, Jarus, Anaby, & Law, 2009). Involvement in a variety of activities, especially leisure activities of their own choosing, has been emphasized by adolescents with Cerebral Palsy (CP) as contributing to a good QoL (Shikako-Thomas et al., 2009).

In the same way as participation, QoL is increasingly included as an outcome variable to evaluate the effectiveness of interventions for children with CP. QoL is a multidimensional construct, which incorporates objective and subjective accounts of personal feelings, social relationships, local environment, societal values, political institutions, economic conditions, and international relations (Bjornson & McLaughlin, 2001; Colver, 2009).

Rosenbaum (2008) reported that to the extent that people are able to do the things that are important to them, to participate in whatever ways enable "engagement", their quality of life is probably better than were they thwarted in "participating". Furthermore, he highlighted the need to pay more attention to this dimension of children's lives with chronic illnesses and disabilities, including promoting participation as an important determinant of QoL (Rosenbaum, 2008).

Even considering the idea that participation may be an important determinant or component of QoL, there is a paucity of literature on quality of life and participation in leisure activities in children and adolescents with cerebral palsy. One of the few studies presently available was developed in Ireland (McManus, Corcoran, & Perry, 2008), where parents of children with CP gave information about their children`s participation and QoL. McManus and colleagues (2008) found that participation affects QoL domains physical well-being, social support & peers and mood & emotions. However, the Irish study only considers the perspective of parents, both in the evaluation of participation, and the QoL. Although in the child QoL literature, it is proposed that because of children's cognitive immaturity, limited social experience, and continued dependency, parents may be more able to report some aspects of their child's QoL (Davis et al., 2007; Waters et al., 2007), both assessment variants, i.e., self-reports and parent/proxy reports, should be included to gain as much information as possible and to identify the factors that influence quality of life (Neuner et al., 2011). On the other hand, the authors of the Irish study have assessed only participation frequency. Important aspects like the type of activity (diversity) and the level of enjoyment not been investigated, remaining unclear whether these factors also affect QoL of Children with CP.

Moreover, one revision recently published aiming to understand the influence of participation in leisure and social activities on the different dimensions of QoL has shown that the participation in physical and everyday activities enhance physical well-being (Dahan-Oliel, Mazer, & Majnemer, 2012). Also, that the participation in leisure activities has an effect on self-perception, can promote emotional and social QoL, and may contribute to happiness and enjoyment, and at least, that participating in chosen, preferred activities, promotes overall well-being. However, the review also highlighted some negative impacts of involvement leisure activities on QoL, mostly mediated by environmental constraints related to accessibility or lack of adaptations.

Because of scarce of information, we were interested in to investigate if the participation in leisure activities outside school influence QoL in children and adolescents with CP, considering objective (diversity and intensity) and subjective (enjoyment) aspects of the participation construct. Specifically, the aims of this study were:

1. To investigate if the participation in leisure activities affect QoL after adjusting for age, gender and severity of impairment (motor and intellectual).
2. To study the contribution of participation on QoL, according to the different type of activities (formal, informal, recreational, active physical social, skill-based and self-improvement activities).

METHODS

Participants

This paper used data from one large sample of CP children and adolescents from across Spain collected in the study on participation in leisure activities. Children and adolescents with CP were recruited from Associations Caring for People with Cerebral Palsy and Related Disabilities (ASPACE), between December 2008 and June 2010.

In order to achieve a heterogeneous and representative sample of the population of children and adolescents with CP, all ASPACE centers were invited to participate in this study. The 68 ASPACE centers are found in 17 regions in Spain, offering multi-disciplinary assessment services for 8.400

people with CP. Children and adolescents with CP, aged between 8 and 18 years, receiving services were eligible if they did not have a concomitant illness or health condition that might affect participation and QoL.

Measures

Participation was measured using the Spanish version of Children´s Assessment of Participation and Enjoyment (CAPE) (King et al., 2007), which assess the children's and adolescents´ participation in leisure and recreation activities outside of mandated school activities. The CAPE is designed to measure if and how children and youths aged between 6 to 21 participate in 55 informal and formal activities that measure participation in five dimensions: diversity, intensity, with whom, where and enjoyment. However, for the purpose of the present study the diversity, intensity and enjoyment CAPE scores were used. The Spanish version has demonstrated good psychometric properties (construct and discriminant validity, and test-retest reliability) (Longo, Badia, Orgaz-Baz, & Verdugo Alonso, 2012).

QoL was measured using KIDSCREEN, a 52- item generic health-related quality of life measure applicable to healthy and chronically ill children and adolescents aged 8–18 years and designed for child or parent report. The KIDSCREEN instrument was chosen because it has been well validated psychometrically with 22,110 European children from the general population, which made it possible to compare the Spanish children's results with those of children from other regions of Europe (Ravens-Sieberer et al., 2005). KIDSCREEN assesses 10 domains of QoL: physical wellbeing (5 items), psychological well-being (6 items), moods & emotions (7 items), self-perception (5 items), autonomy (5 items), parental relations & home life (6 items), financial resources (3 items), social support & peers (6 items), school environment (6 items) and social acceptance & bullying (3 items). The KIDSCREEN scores are converted into T-scores (mean = 50 and 1 standard deviation = 10), where higher values indicate a higher QoL.

The Gross Motor Function Classification System (GMFCS) was used to categorize gross motor function (Palisano et al., 1997). The GMFCS describes the motor performance of children with CP on the basis of their functional abilities and their need for assistive technology and wheeled mobility. Functional levels range from I (independent gross motor function with few limitations) to V (complete dependence for all motor activities).The psychometric properties of the GMFCS have been thoroughly tested and reported, and include evidence supporting its content validity, construct validity, inter-rater reliability and test–retest reliability (Wood & Rosenbaum, 2000).

A demographic questionnaire was completed by the parent or caregiver, providing information about the child and parent sex and age, number of parents in the family, parental educational level and family income.

Procedure

Ethical approval for the study was obtained from the Bioethical Committee of the University of Salamanca. Written, informed consent was obtained from all parents and from all children aged 12 years and above.

Two research team members visited each ASPACE center giving information about the investigation and 16 centers in 7 different regions of Spain responded to the request to participate. In each center a research assistant (psychologist or social worker experienced in providing services to children and families with CP) was formally trained by one of members of the research team.

The research assistants contacted the chosen families and an appointment was made for the interview at ASPACE center. The CAPE was completed by the child/adolescent in a face to face interview or completed by a parent if the child/adolescent was unable to contribute.

After the CAPE interview, a parent gave information on quality of life of her or his child completing the KIDSCREEN questionnaire parent version, and after that, completed the socio-demographic questionnaire. The duration of the CAPE interview took approximately 1 hour and KIDSCREEN 15-20 min.

In each ASPACE center psychologist provided information about the intellectual ability of each child/adolescent. The following criteria were considered as intellectual impairment: none or mild (IQ>70); moderate (IQ 50-70), and severe (IQ<50). Physical therapists were given information about the GMFCS level of each child/adolescent.

Statistical analyses

Statistical analyses were performed using the SPSS for Windows software program, version 17.0 considering a .01 level of significance.

Descriptive statistics (means and SD) were used to describe the sample. The children's diversity, intensity and enjoyment of participation in leisure activities and the QoL scores for each domain for parents' version were presented as mean and SD.

After that, the influence of the participation in leisure activities on the quality of life domains was evaluated controlling the affect of socio-demographic and disability- related variables. Hierarchical regression was executed for each domain of QoL, including in the first step socio-demographics variables (gender and age) and disability-related variables (GMFCS level and intellectual impairment level), and in the following step, the variables related to participation, (diversity, intensity and enjoyment).

Finally, to enhance interpretation and because the influence of different explanatory factors might differ depending on the type of activity, we also performed the above-mentioned regression analyses separately for each type of activities of CAPE (formal, informal, recreational, social, self-improvement, active physical and skill-based activities).

Diagnostic tests were used to check for violations of the assumptions inherent in linear regression models.

RESULTS

Group characteristics

Participated 206 children and adolescents with CP, 115 boys (55.8%) and 91 girls (44.2%), mean age of 11.96 years (SD = 3.00; range 8-18 years). Table 1 illustrates child characteristics. The sample was recruited from the 16 ASPACE centers in 7 different regions of Spain. Of the total sample, 112 children and adolescents with CP completed the CAPE independently or received some assistance. In the cases of severe intellectual impairment (43.7%), the parents completed the CAPE by proxy. Regarding the characteristics of the caregiver, the mean age was 43.16 years (SD = 5.77; range 28-61 years), and the majority was female (66.5%). In terms of family structure, in 85.2% of cases were bi parental and 14.8% single parent. The levels of family income were: low (less than 1000 €) in 16% of cases, medium (between 1000 and 2000 €) in 42% of cases, and high (more than 2000 €) in 46% of cases.

Table 1. Characteristics of the sample of children and adolescents with CP (n=206)

Variable	n (% of valid n)
Gender	
Male	115 (55.8)
Female	91 (44.2)
Age (M = 11.96, SD = 3.02, range = 8–18 years)	
8-12	126 (61.2)
13-18	80 (38.8)
GMFCS	
Level I	50 (24.3)
Level II	37 (18)
Level III	37 (18)
Level IV	26 (12.6)
Level V	56 (27.2)
Intellectual Impairment	
None	33 (16)
Mild	43 (20.9)
Moderate	39 (18.9)
Severe	90 (43.7)
Information not available	1 (.5)
Type of schooling	
Regular	113 (54.9)
Special	91 (44.2)
Information not available	2 (1)
Seizures	
No seizures	113 (54.9)
Seizures	90 (43.7)
Information not available	3 (1.5)
Hearing	
Does not need hearing aid	172 (83.5)
Profound/severe loss	15 (7.3)
Information not available	19 (9.2)
Vision	
Has useful vision	111 (53.9)
Blind or no useful vision	82 (39.8)
Information not available	13 (6.3)
Communication	
No difficulties	104 (52.5)
Difficulty (uses speech or communication aid)	94 (45.6)
Information not available	8 (3.9)

Participation and QoL results

Table 2 shows the means and standard deviations on participation and QoL measures. Children and adolescents with CP participated in a mean of 20.6 activities, and mostly in informal activities. Diversity of participation was lowest in active-physical, self-improvement, skill-based and formal activities. Intensity of participation was low; median scores ranged from 1.0 (active physical) to 2.9 (social activities). High levels of enjoyment for all types of activity were reported. A detailed description of participation in leisure activities for this population has been recently published (Longo, Badia, & Orgaz, 2012).

According to the parents, children and adolescents with CP showed low QoL for most domains, except for parent relation & home life and school environment. The lower results were obtained in physical well-being (where the mean was lower than 1 SD of the mean established by the instrument), social acceptance & bullying, social support & peers, and autonomy domains. On the other hand, the means in self perception, psychological well-being, moods & emotions, and financial resources domains were closer to the KIDSCREEN mean (50).

Table 2. Mean and standard deviations of the measures of participation and QoL

Measure	Mean (SD)
QoL KIDSCREEN parents´ version	
Physical well-being	38.8 (10.6)
Psychological well-being	48.4 (10.7)
Moods & emotions	48.3 (11.7)
Self perception	49.1 (10.0)
Autonomy	45.9 (10.8)
Parent relation & home life	52.7 (8.8)
Social support & peers	45.5 (13.4)
School environment	54.5 (9.9)
Social acceptance & bullying	43.1 (11.4)
Financial resources	48.0 (13.6)
Diversity of participation – CAPE	
Overall	20.6 (8.7)
Formal	3.6 (2.3)
Informal	17.1 (7.0)
Recreational	6.4 (2.9)
Active physical	2.8 (1.8)
Social	6.5 (2.9)
Skill-based	2.9 (1.7)
Self-improvement	3.9 (2.0)
Intensity of participation – CAPE	
Overall	1.8 (0.7)
Formal	1.2 (0.8)
Informal	2.1 (0.9)
Recreational	2.7 (1.2)
Active physical	1.0 (0.7)
Social	2.9 (0.9)
Skill-based	1.4 (0.9)
Self-improvement	2.0 (1.0)
Enjoyment of participation – CAPE	
Overall	3.9 (0.5)
Formal	3.9 (0.8)
Informal	3.9 (0.6)
Recreational	3.9 (0.7)
Active physical	4.1 (0.8)
Social	4.1 (0.7)
Skill-based	4.0 (0.9)
Self-improvement	3.4 (0.9)

The KIDSCREEN scales for each domain have mean=50 and SD=10 in the reference population. CAPE Scoring: Diversity (number of different activities)= number of activities involved in compared to total number of activities (check in manual). Intensity (how often)= 1- 1x/4 months, 2- 2x/4 months, 3- 1x/month, 4- 2-3x/month, 5- f1x/week, 6- 2-3x/week, 7- daily. Enjoyment (how much do you enjoy)= 1- not at all, 2- somewhat, sort of, 3- pretty much, 4- very much, 5- love it.

Influence of participation on the QoL domains

Regarding diversity of participation, when we control the effects of socio-demographic and disability-related variables, diversity of participation in leisure activities had a significant contribution to explaining the levels of QoL in the social support & peers domain (Table 3). The positive sign of the regression coefficient indicated that the greater the diversity of participation the more improved relationships with peers.

When analyzed separately the domains, we found that the diversity of participation in informal activities (p = .002) had a significant effect on the relationship with peers. In relation to the type of activity, we found that the diversity of participation in recreational activities (p = .006) was significantly contributing to explain the differences in quality of life in the domain social support & peers.

Table 3. Results of multiple linear regressions of quality of life domains on diversity participation in leisure activities, controlling the effect of gender, age, GMFCS level and intellectual impairment level variables

Quality of life domain	**F**	**Df**	**p-value**	**R^2 (%)**	**B**(1)	**95% CI**(1)	**t**(1)	**p-value**(1)
Physical well-being	15.35	5,153	<0.001	.33	.26	0.06-0.46	2.46	0.015
Psychological well-being	1.21	5,175	0.306	.03	.26	-0.13-0.65	2.12	0.035
Moods & emotions	2.13	5,156	0.060	.06	-.15	-0.42-0.12	-1.07	0.287
Self perception	6.13	5,155	<0.001	.17	-.28	-0.50- -0.06	-2.45	0.015
Autonomy	1.31	5,159	0.261	.04	.22	-0.04-0.48	1.72	0.088
Parent relation & home life	0.66	5,162	0.656	.02	.05	-0.17-0.27	0.46	0.648
Social support & peers	6.77	5,130	<0.001	.21	.49	0.20-0.78	3.23	**0.002**
School environment	1.63	5,171	0.155	.05	.18	-0.04-0.40	1.58	0.115
Social acceptance & bullying	3.75	5,163	0.003	.10	.13	-0.13-0.39	0.97	0.330
Financial resources	2.62	5,118	0.028	.10	.27	-0.08-0.62	1.53	0.129

(1)Unstandardised coefficient and associated statistics for diversity participation after adjustment for gender, age and level of impairment (GMFCS and Intellectual impairment).

In respect to intensity of participation, when we adjusted for gender, age, GMFCS level and intellectual impairment level, the intensity of participation significantly accounted for different in QoL in the physical well-being, psychological well-being, autonomy and social support & peers domains (Table 4).

Domain analysis indicates that participation more often in informal type activities is what explained the QoL in psychological well-being (p=.002), autonomy (p=.003) and social support & peers (p<.001) domains. A higher frequency of participation in recreational activities accounted for a significant contribution on the psychological well-being (p=.007) and social support & peers (p<.001) domains. At the same time, the intensity of participation in social activities contributed significantly to explain the differences in quality of life in the psychological well-being (p = .005) and social support & peers (p = .004) domains. Finally, a higher frequency of participation in skill-based activities improved QoL in the social support & peers (p=.003) domain.

Table 4. Results of multiple linear regressions of quality of life domains on intensity participation in leisure activities, controlling the effect of gender, age, GMFCS level and intellectual impairment level variables

Quality of life domain	F	Df	p-value	R^2 (%)	B⁽¹⁾	95% CI⁽¹⁾	t⁽¹⁾	p-value⁽¹⁾
Physical well-being	15.83	5,153	<0.001	.34	3.13	0.91-5.35	2.77	**0.006**
Psychological well-being	2.27	5,175	0.049	.06	4.03	1.50-6.56	3.12	**0.002**
Moods & emotions	1.90	5,156	0.098	.06	0.25	-2.73-3.23	0.17	0.869
Self perception	5.91	5,155	<0.001	.16	-2.75	-5.14- -0.36	2.25	0.026
Autonomy	2.43	5,159	0.038	.07	3.99	1.29-6.69	2.89	**0.004**
Parent relation & home life	1.07	5,162	0.379	.03	1.71	-0.52-3.94	1.50	0.137
Social support & peers	8.38	5,130	<0.001	.24	6.72	3.56-9.88	4.16	**<0.001**
School environment	2.02	5,171	0.078	.06	2.57	0.16-4.98	2.10	0.038
Social acceptance & bullying	4,10	5,163	0.002	.11	2.24	-0.52-5.00	1.58	0.116
Financial resources	2,65	5,118	0.026	.10	2.94	-0.73-6.61	1.57	0.119

(1)Unstandardised coefficient and associated statistics for intensity participation after adjustment for gender, age and level of impairment (GMFCS and Intellectual impairment).

Replicating the same type of analysis, the enjoyment of participation significantly accounted for different in QoL in the physical well-being, psychological well-being and parent relation & home life domains, indicating that higher levels of enjoyment in leisure activities, improved physical and psychological well-being, and parent relation (Table 5).

The detailed analysis indicated that higher levels of enjoyment improved QoL in the psychological well-being domain and that this association was found with the degree of enjoyment in the domains [formal (p =.001) and informal (p<.001)] and in all types of CAPE activities [social (p<.001), recreational (p<0.001), active physical (p<.001), self-improvement (p =.007) and skill-based (p=0.009)]. Moreover, the enjoyment in informal activities had a significant and positive effect on the physical well-being (p=.001) and the parent relation & home life (p=.001) domains. Also, higher levels of enjoyment in recreational activities improved results in the physical well-being (p=.008) and the parent relation (p=.003) domains. Enjoyment of physical activities improved physical well-being (p<.001) and enjoyment of social activities (p=.007) and self-improvement (p=.007) improved the parent relation.

Table 5. Results of multiple linear regressions of quality of life domains on enjoyment participation in leisure activities, controlling the effect of gender, age, GMFCS level and intellectual impairment level variables

Quality of life domain	F	Df	p-value	R^2 (%)	B⁽¹⁾	95% CI⁽¹⁾	t⁽¹⁾	p-value⁽¹⁾
Physical well-being	16.76	5,153	<0.001	.35	4.14	1.69-6.59	3.30	**0.001**
Psychological well-being	6.35	5,175	<0.001	.15	7.35	4.72-9.98	5.47	**<0.001**
Moods & emotions	2.71	5,156	0.002	.08	3.39	0.02-6.76	1.97	0.051
Self perception	5.79	5,155	<0.001	.16	2.90	0.93-5.57	2.14	0.034
Autonomy	1.55	5,159	0.177	.05	3.16	0.10-6.22	2.02	0.045
Parent relation & home life	2.72	5,162	0.022	.08	3.99	1.56-6.42	3.21	**0.002**
Social support & peers	5.03	5,130	<0.001	.16	3.31	-0.45-7.07	1.72	0.087
School environment	2.01	5,171	0.080	.06	2.86	0.17-5.55	2.09	0.038
Social acceptance & bullying	3.86	5,163	0.002	.11	1.89	-1.52-4.99	1.20	0.233
Financial resources	2.13	5,118	0.067	.08	-0.61	-4.73-3.52	-0.29	0.770

(1)Unstandardised coefficient and associated statistics for enjoyment participation after adjustment for gender, age and level of impairment (GMFCS and Intellectual impairment).

DISCUSSION

The aim of this paper has been to present an analysis of the influence of participation in leisure activities on the QoL domains in children and adolescents with CP in Spain, using child and parent assessed instruments. The World Health Organization (2001) stresses the importance of participation, in a full range of developmentally appropriate social, recreational and skill-building activities in determining QoL (WHO, 2001). Our results are encouraged and indicate that diversity, intensity and enjoyment of participation affect the QoL` results in 5 domains (physical well-being, psychological well-being, autonomy, social support & peers, and parent relation & home life).

The available research about the impact of participation in leisure activities on quality of life in children and adolescents with CP is limited. In congruence with our results, a recent study with 63 Canadian children with CP, utilizing the CAPE and the PedsQL have found that diversity and intensity of participation in leisure activities accounted for 32% of the variance for physical well-being QoL domain (Shikako-Thomas et al., 2012).

The literature attributes the relationship between leisure and well-being to several aspects including that leisure decreases the effects of negative events and enhances well-being by acting as a protective factor by providing social support, feelings of competency and relaxation (Caldwell, 2005; Dickinson et al., 2007; Poulsen, Ziviani, Johnson, & Cuskelly, 2008; Tedeschi & Calhoun, 2004).

In this study we found that the fact of participating more times in enjoying leisure activities increases the QoL in the physical well-being domain. McManus et al. (2008) also point out that increase in participation in everyday activities in children with CP in Ireland was associated with increase in QoL related to physical well-being, moods & emotions and social support & peers (McManus, Corcoran, & Perry, 2008). Active leisure may enhance children's well-being because social relationships can be developed and strengthened with physical activity, possibly by increasing self-confidence, and relationships are associated with children's happiness (Fredricks et al., 2002; Holder & Coleman, 2009).

According our results, high intensity of participation and enjoyment increase the QoL in the psychological well-being domain, suggesting that promoting participation may contribute, in part, to greater personal satisfaction with life. In fact, studies have highlighted that leisure enhances overall psychological well-being and this relation may be stronger than the relation between leisure and physical health (Caldwell, 2005; Sacker & Cable, 2006).

Also, we found that the enhance diversity and intensity of participation increase the QoL in the social support & peers domain. In this sense, studies have pointed out the importance of participating in a wide range of leisure activities, as participating in a greater number of leisure activities may provide additional benefits such as increasing diversity of children's experiences (Van Naarden Braun, Yeargin-Allsopp, & Lollar, 2006). A high intensity of participation also increases the QoL in the autonomy domain. Not needing to depend on another person's decision to participate and having enough time were found to facilitate level of participation in social activities (Badia, Orgaz, Verdugo, Ullán, & Martínez, 2011). Participating in recreational and social activities children may experience different situations with peers, from negotiating roles, to practicing social skills, and to integrating different life contexts (Shikako-Thomas et al., 2012).

Moreover, an increase in diversity, intensity and enjoyment in participation in recreational type activities, including activities such as watching TV and playing computer or video games, improves QoL in the psychological well being, autonomy, social support & peers and parent relation & home life domains. These results do not corroborate with findings from a study about leisure and children´s well-being in Canada that found that time spent on the computer was negatively correlated with all measures of children's well-being (Holder & Coleman, 2009). A possible explanation for these discordant results is that Holder & Coleman (2009) used measures of well-being and happiness, while in our study we used a HRQoL instrument. In addition, the sample only included Canadian children aged 8-12 years and without disabilities. Studies showed the differences between children with and without disabilities in relation to some participation patterns. For example, whereas typically developing children reported higher average levels of enjoyment for formal activities, children with disabilities did not differ in their enjoyment of formal versus informal activities (King, Petrenchik, Law, & Hurley, 2009).

Furthermore, our results indicate that higher enjoyment in active physical activities is associated with higher QoL in the psychological well-being domain. Shikako-Thomas et al. (2012) have shown a positive association between engagement in physical activities and both physical and psychosocial well-being, suggesting that according to parent-report, school-aged children with CP who participate more actively in physical activities feel better about their social life, school functioning, and their emotions. Participation in regular physical activity can foster independence, coping abilities, competitiveness, and teamwork among children with disabilities (Patel & Greydanus, 2002). Sports participation enhances the psychological wellbeing of children with disabilities through the provision of opportunities to form friendships, express creativity, develop a self-identity, and foster meaning and purpose in life (Dykens, Rosner, & Butterbaugh, 1998).

A recent research concluded that positive QoL outcomes were related to the child intrinsically, such as better physical well-being, self-perception, and emotional well-being while the negative ones were mostly related to environmental factors (Dahan-Oliel et al., 2012). Hence, one preliminary report on environmental needs and QoL in 104 Spanish children and adolescents with CP pointed out that 62.4% of the sample did not have access to adequate leisure facilities. Parents also reported that nearly 25% of cases, their children did not receive emotional support from their peers at school (Badia, Arias, Verdugo, Gómez, & González, 2012). This data calls for interventions in the sense of improving attitudes towards children with disabilities in the school environment. Therefore, it has been suggested that makes one wonder whether exposure to peers with functional differences in the preschool years might provide the best opportunities to help young children see and accept others as part of the social fabric of the community (Rosenbaum, 2010).

Studies on QoL are highly subjective and as such have inherent limitations, as we were aware of the limitation of using QoL evaluation being parent-based rather than child-based. Future studies are needed to validate these findings considering the child`s opinion. Also, further studies should be carried out to examine inter-relationships between the environmental variables and the participation in leisure activities and their influence on the QoL results in children and adolescents with CP in Spain.

CONCLUSIONS

In summary, this study has underlined the importance of participation in leisure activities on improving the QoL in physical well-being, psychological well-being, autonomy, parent relation & home life and, social support & peers of Spanish children and adolescents with CP. Besides, all the information discussed can support health care providers in promoting the participation of Spanish children and adolescents with CP in leisure activities. Unlike motor function, participation can be modified at any stage of life with adjustments in the physical, social and attitudinal and thereby improving QoL results. In fact, findings of a recent European study with children with CP confirmed the social model of disability and showed that the physical, social and attitudinal environment of disabled children influenced their participation in everyday activities and social roles (Colver et al., 2012). These arenas may be more amenable to intervention than motor function and may be the real keys to improved QoL.

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CAPÍTULO 5

INSIGHTS ON QUALITY OF LIFE OF CHILDREN AND ADOLESCENTS WITH CEREBRAL PALSY: COMPARING CHILD AND PARENT REPORTS

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ABSTRACT

Background: The World Health Organization (WHO) recommends that measures of quality of life (QoL) in children use self-reporting wherever possible. However, a high percentage of children with cerebral palsy (CP) and severe intellectual impairment are unable to self-report, which demands relying on parent reports. There is no information on research literature on the agreement between parent and child in reports of CP children's QoL in Spain Aim: The purpose of this study was to describe the level of agreement between reports of QoL obtained from children and adolescents with CP and their parents. We also examined the factors that may affect the level of agreement in the QoL between children with CP and their parental proxies. Methods: Sixty-six children and adolescents with CP (median age 12y 3mo, SD = 3y, range = 8–18 years) and their parents separately completed parallel forms of the KIDSCREEN questionnaire. Results: Correlation between child and parent scores was low in 8 domains, and moderate in only two domains. In 8 of 10 domains, children reported significantly better QoL than their parents did. The average frequency of disagreement (child-parent difference greater than half an SD of child scores) over all domains was 62.2%, with parents rating their child's QoL lower than the children themselves in 21% to 55% of child-parent pairs. Factors associated with a higher agreement between parent – child pairs were: age of the child (adolescents) gender of the child (female), parent and child (different genders) and not having siblings. Conclusions: This study demonstrated that, there discrepancies between parent and child reports and that assessing both parents and children may give additional insights into the QoL of children and adolescents with CP.

Key words: quality of life, child-parent agreement, children, adolescents, cerebral palsy

INTRODUCTION

"Theresa, 12 years old, had CP spastic bilateral, walked at home and used wheeled mobility in the community. When asked how she felt about her life, she said that all was fine because she had friends and had fun playing with them, enjoyed go to school and had independency moving in the community with her powered wheelchair. According her mother, Theresa was a sad and lonely child. Although the wheelchair allows her to go to school and around in the community independently, she didn´t had friends, occupied her time with computer games and rarely goes out." This divergence of opinion, no rarely observed between parents and children, is the central point of this paper: do parents know their children well enough to assess subjective aspects such as quality of life (QoL)?

In the field of medicine, conventional outcome measures such as reduction of symptoms is increasingly being supplemented by broader range outcomes such as QoL and health-related QoL (HRQoL). QoL, although variously defined, is generally conceptualized as a subjective and multidimensional construct encompassing several domains of life while HRQoL is considered to be a sub-domain of the more global construct of QoL (Bjornson & McLaughlin, 2001; Schipper, 1996).

Historically it was thought that children and adolescents could not reliably report their QoL, thus QoL was based on proxy-reports from parents (Ravens-Sieberer et al., 2005). Proxy reporters most often are parents, but occasionally others such as physician, nurse, or teacher informants may be used. In the child QoL literature, it is proposed that because of children's cognitive immaturity, limited social experience and continued dependency, parents may be more able to rate some aspects of their child's QoL (Eiser, Mohay, & Morse, 2000). In addition, parents are the principle decision makers regarding the child's treatment, at least until the child reaches the age of majority (Britto et al., 2004; Parsons, Barlow, Levy, Supran, & Kaplan, 1999). However, more recent research has shown that children' and their parents' perception of QoL can differ, and that children are able to reliably report their QoL provided the questionnaire is appropriate to their age and cognitive functioning (Ravens-Sieberer et al., 2005; Varni et al., 2005). Although the World Health Organization (WHO) to recommend that measures of QoL in children might use self-reporting wherever possible, some children with CP and severe intellectual impairment are unable to self-report (World Health Organization, 2001).

When using parent-reported HRQoL, either in addition to or in place of child-reported HRQoL, it is important to understand the areas of agreement and disagreement between the two (Sattoe, van Staa, & Moll, 2012). Several studies have reported low to moderate correlations between parent and child reports of HRQL (Chang & Yeh, 2005; Davis et al., 2007; Upton, Lawford, & Eiser, 2008) with generally stronger concordance noted on physical health domains (Eiser & Morse, 2001; Klassen, Miller, & Fine, 2006; Majnemer, Shevell, Law, Poulin, & Rosenbaum, 2008; Varni, Seid, & Kurtin, 2001). However, according Matza, Swensen, Flood, Secnik, & Leidy (2004), strong correlations do not necessarily indicate high agreement. Furthermore, he highlights that if parents consistently underestimate or overestimate HRQoL compared with the child's report, the 2 measures may be highly correlated despite large mean differences. Lastly, correlations provide a criterion of relative agreement that is a necessary, but not sufficient, indicator of source agreement (Matza, Swensen, Flood, Secnik, & Leidy, 2004).

Because a cure for CP is not achievable at this time, the goal of intervention is typically focused on promoting a child's QoL (Chambers & Sutherland, 1997; Chong, Mackey, Broadbent, & Stott, 2012). Nevertheless, the QoL of the children with CP in Spain, mainly with the most severe impairments, has been often overlooked. The purpose of this study was to determine if parent proxies can be used to assess children's QoL, especially because in many cases of children and adolescents with CP, proxy reports may be the only available source of data when children have severe intellectual impairment associated or are too young to understand the content of self-report measures. The specific aims of our study were: (1) to describe the level of agreement between reports of QoL obtained from children with CP and their parents and (2) to explore the factors that may affect the level of agreement in the QoL between children with CP and their parental proxies.

METHODS

Participants

The data analyzed in this paper were collected from a group of children and adolescents with CP (and their parents), who were participants in a larger study on participation in leisure activities of children and adolescents with CP in Spain. This study had been granted ethical approval from the Bioethical Committee of the University of Salamanca.

Participants were recruited from 11 Associations Caring for People with Cerebral Palsy and Related Disabilities (ASPACE) centers, in various regions of Spain, between December 2008 and June 2010. Families were eligible to participate in this study if they had a child with a diagnosis of CP, aged 8 to 18 years. Children who had another neuromuscular disorder, or were diagnosed with Attention Deficit Hyperactivity Disorder or Autism Spectrum Disorders were excluded from this study sample.

The definitive study sample consisted of 66 children and adolescents (and their parents), 40 boys (60.6%) and 26 girls (39.4%), mean age of 12.03 years (SD = 3.03), range 8-18 years. More than half of the sample had mild motor impairments (levels I and II GMFCS) and all attended to regular school. In 63.6% of the cases the questionnaires were completed by the mother and in 36.4% by the father. Table 1 illustrates child and parents characteristics.

Table 1. Socio-demographics characteristics (n=66 dyads)

Variable	n (%)
Gender	
Children	40 (60.6)
Girls	26 (39.4)
Age (M = 12.03, SD = 3.00, range = 8–18 years)	
8-11	34 (51.6)
12-18	32 (48.4)
GMFCS	
Level I	27 (40.9)
Level II	15 (22.7)
Level III	14 (21.2)
Level IV	7 (10.6)
Level V	3 (4.5)
Intellectual Impairment	
None	25 (37.9)
Mild	26 (39.4)
Moderate	13 (19.7)
Information not available	2 (3.0)
Type of schooling	
Regular	66 (100)
Special	0 (0)
Seizures	
No seizures	46 (69.7)
Seizures	18 (27.3)
Information not available	2 (3.0)
Communication	
No difficulties	50 (75.8)
Difficulty (uses speech or communication aid)	11 (16.7)
Information not available	5 (7.6)
Gender caregiver	
Male	24 (36.4)
Female	42 (63.6)
Age of caregiver (M = 43.16, SD = 5.16, range = 30–59 years)	
30-44	40 (60.6)
45-59	23 (34.8)
Information not available	3 (4.5)
Parental educational level	
Elementary school	19 (28.8)
High School	21 (31.8)
University or PhD	26 (39.4)
Family structure	
Single	7 (10.6)
Married or living with partner	59 (89.4)
Total family income (Euros € per month)	
Less 1000 €	9 (13.6)
Between 1000-2000 €	30 (45.5)
More 2000 €	26 (39.4)
Information not available	1 (1.5)
Siblings	
None	13 (19.7)
One	39 (59.1)
Two or more	14 (21.2)

Measures

Quality of Life (QoL) – Spanish version of the KIDSCREEN- 52

QoL was measured using KIDSCREEN, a 52- item generic health-related quality of life measure applicable to healthy and chronically ill children and adolescents aged 8–18 years (Ravens-Sieberer et al., 2005). The KIDSCREEN has child and parent/proxy versions with 52 items covering 10 domains: Physical well-being, Psychological well-being, Moods & emotions, Self-perception, Autonomy, Parental relations, Financial resources, Social support & peers, School environment and Social acceptance & bullying. The items have a five-point Likert scale with two sets of possible responses: never, seldom, quite often, very often, always and not at all, slightly, moderately, very, extremely. For each domain, the relevant items are summed and scaled to yield a score in the range 0–100 with higher scores indicating better quality of life. The child report version of the KIDSCREEN - 52 contains items identical to those in the parent report version, with the identifier "you" in place of "your child".

Gross Motor Function Classification System (GMFCS)

The GMFCS is a five-level system developed for children with CP to classify gross motor function based on usual performance in home, school and community settings. Distinctions between GMFCS levels are based on functional abilities and limitations, the need for assistive mobility devices, wheeled mobility and, to a lesser extent, quality of movement (Palisano et al., 1997). Functional levels range from I (independent gross motor function with few limitations) to V (complete dependence for all motor activities).The psychometric properties of the GMFCS have been thoroughly tested and reported, and include evidence supporting its content validity, construct validity, inter-rater reliability and test–retest reliability (Palisano et al., 1997; Wood & Rosenbaum, 2000).

Questionnaires on demographic variables

The parents completed a questionnaire providing information about their employment, educational qualifications, income, family cohesion, number of siblings, and about their child if his or her had seizures, which school he or she attended to.

Procedure

Two research team members visited the ASPACE centers giving information about the investigation. In each center a research assistant (psychologist or social worker experienced in providing services to children and families with CP) was formally trained by one of members of the research team.

A letter with information on the study and an invitation to participate were sent to the families chosen by the research assistants. Once consent was obtained, an appointment was made to carry out the interviews at ASPACE center. Written and informed consent was obtained from the parents of all children and adolescents who participated, and written assent was obtained from children and adolescents whenever possible.

Child and parent completed their QoL questionnaires in a face to face interview which was administrated by the same trained research assistant. Proxy raters were asked to respond the way they thought their child would score the individual items. The duration of the KIDSCREEN interview took approximately 15-20 min. After that, the parent completed the socio-demographic questionnaire.

In each ASPACE center psychologist provided information about the intellectual ability, and the physical therapists given information about the GMFCS level of each child/adolescent. The following criteria were considered as intellectual impairment: none or mild (IQ>70); moderate (IQ 50-70), and severe (IQ<50).

Statistical analyses

Descriptive statistics were used to characterize the sample and the outcomes. Firstly, the reliability (Cronbach's alpha) in each KIDSCREEN domain of parents and children questionnaires was examined.

In addition, the Pearson and intraclass correlations (ICC) between the children's and the parents' scores were analyzed. Correlations were interpreted according to the following criteria: low (inferior than 0.2); regular

(0.21-0.40); moderate (0.41-0.60); substantial (0.61-0.8), and excellent (superior than 0.8) (Szklo & Nieto, 2000).

Also, the means of directional differences (children score – parents score) were calculated and tested using paired Student's t tests. The mean difference score was standardized by relating this score to the average SDs of the child and parent scores (effect size). The values of the effect size (ES) were interpreted taking into account the criteria established by Cohen (1988): minimum between 0.2-0.5; moderate between 0.51-0.8 and important above 0.8.

The means of the absolute value of the differences between scores were calculated as indicators of agreement. We defined agreement when the absolute value was less than or equal to half of the SD of the children's scores, according to the broadly recognized definition of clinically meaningful difference in QoL.

Moreover, for each of these dyads, the proportion of the sample in which the scores between the parent and child were either equivalent or parent's score was lower, or child's score was lower, was calculated.

Later, we assessed the incidence of personal variables such as gender of the child, gender of the caregiver, the relationship between the gender of the child and the caregiver (same versus different), age (children versus adolescents), gross motor function (walking versus not walking), caregiver's educational level (primary, secondary and university), siblings (yes versus no) on the agreement and disagreement of their responses. We conducted two types of analysis: the first was the chi-square test to see if there were differences in the proportion of agreements between different groups; in the second, we analyzed the incidence of the degree of agreement using Student's t tests (for independent samples), considering the absolute differences.

Statistical analyses were performed using the SPSS for Windows software program, version 17.0 using a .05 level of significance.

RESULTS

Reliability

The KIDSCREEN parents and children versions proved to be reliable. The Cronbach's alpha coefficient range of the scales was .53-.92 in the case of children´s version and .59-.92 in the case of the parents´ version (see table 2). The means child self-reports and parent proxy reports for each domain of QoL also are shown in the table 2.

Table 2. Mean and Reliability of Child and Parent Reports dyads

Domain	Complete Pairs n (%)	Child's Report		Parent's Report	
		Mean (SD)	Cronbach`s alpha	Mean (SD)	Cronbach`s alpha
Physical well-being	60 (91)	46.88 (10.17)	.75	42.40 (11.05)	.81
Psychological well-being	66 (100)	54.01 (7.91)	.68	48.65 (9.19)	.90
Mood & emotions	63 (96)	51.03 (10.17)	.74	45.50 (12.15)	.85
Self-perception	64 (97)	53.95 (9.98)	.53	45.77 (9.50)	.59
Autonomy	65 (99)	51.53 (11.84)	.78	47.01 (9.31)	.81
Parental relation	65 (99)	54.41 (8.34)	.66	51.78 (8.71)	.63
Social support & peers	61 (92)	54.57 (11.85)	.81	48.47 (11.08)	.90
School environment	66 (100)	56.27 (10.83)	.80	51.38 (9.77)	.85
Social acceptance	65 (99)	45.34 (11.75)	.80	39.88 (11.81)	.72
Financial resources	56(85)	47.88 (12.71)	.92	51.59 (9.97)	.92

Level of agreement

Correlation between child and parent scores was low in 8 domains, and moderate in only two domains (Physical well-being and Social support & peers). The Pearson values ranged between -0.05 (Mood & emotions) and 0.60 (Social support & peers), while the ICC ones ranged between -0.04 for Mood & emotions and 0.53 for Social support & peers (see Table 3).

In 8 of 10 domains (Physical well-being, Psychological well-being, Moods & emotions, Self-perception, Autonomy, School environment, Social support & peers, and Social acceptance & bullying), children reported significantly better QoL than their parents did (see Table 3). In the Parental relations & home life domain children also reported better QoL than their parents did, but this difference was not significant. Financial resources was the only domain for which parents proxy-reported higher QoL than did the children themselves; however, this difference was not significant.

Table 3. Correlation, bias and agreement between child and parent reports

Domain	Correlation		Directional Difference[a]		Absolute Difference [b]
	Pearson	ICC	Mean (SD)	Effect Size [c]	Mean (SD)
Physical well-being	0.49***	0.45	4.48 (10.78)**	0.42	9.11 (7.23)
Psychological well-being	0.39**	0.33	5.36 (9.48) ***	0.63	8.21 (7.11)
Mood & emotions	-0.05	-0.04	5.53 (16.22)**	0.50	12.40 (11.75)
Self-perception	0.05	0.04	8.18 (13.42)***	0.84	12.34 (9.67)
Autonomy	0.20	0.18	4.52 (13.52)**	0.43	10.65 (9.39)
Parental relations	0.20	0.19	2.63 (10.78)	0.31	8.68 (6.84)
Social support & peers	0.60***	0.53	6.11 (10.23)***	0.53	9.34 (7.33)
School environment	0.21	0.19	4.90 (12.98)**	0.48	10.23 (9.30)
Social acceptance	0.24	0.22	5.47 (14.57)**	0.46	11.84 (10.02)
Financial resources	0.19	0.18	-3.71 (14.59)	0.33	11.59 (9.50)

ICC indicates intraclass correlation coefficient.

[a] Directional difference = child score- parent score

[b] Absolute difference = │child score- parent score│

[c] Effect size = │meana of directional differences│/[(SD_{child} + SD_{parent})/2]

*Two-sided p<.05

** Two-sided p<.01

*** Two-sided p<.001

The size of the effect was moderate (<0.50) in five domains (Physical well-being, Autonomy, School environment, Social acceptance & bullying, and Financial resources), and large (0.84) in the Self-perception domain (see Table 3).

The average rate of agreement over all domains was 37.8%. Specifically, rates of agreement ranged between 31% (for Self-perception and Autonomy domains) and 46% for Social acceptance domain (see Table 4). Disagreement, which consisted of parents rating lower than their children, ranged between 21% for Financial resources domain and 55% for Self-perception, while those related to parents rating higher than their children ranged between 8% for Psychological well-being domain and 41% for Financial resources domain. The average rate of disagreement over all domains was 62.2%. As shown in Table 4, the threshold for agreement was between 4 and 6 points.

Table 4. Agreement and disagreement between the parents and the children on the KIDSCREEN domains

Domain	Parent-child dyad		
	Agreement	Disagreement Parent < Child	Disagreement Parent > Child
Physical well-being	35%	50%	15%
Psychological well-being	35%	46%	19%
Moods & emotions	44%	48%	8%
Self-perception	31%	55%	14%
Autonomy	31%	51%	19%
Parental relation & home life	40%	42%	18%
Social support & peers	46%	44%	10%
School environment	44%	41%	15%
Social acceptance	34%	43%	23%
Financial resources	38%	21%	41%

Agreement= | child score - parent score | ≤ SD of child scores/2 (ie, threshold for each domain in the order specified above: Social support: 6 points; Psychological well-being: 4 points; Parental relations: 4 points; Physical well-being: 5 points; Self-perception: 5 points; Autonomy: 6 points; School: 5 points; Mood & emotions: 5 points; Financial resources: 6 points; Social acceptance: 6 points).

Factors affecting the degree of agreement and disagreement

Gender variable

The gender of the child only had an impact on the agreement in the Autonomy domain; the percentage of girls agreed with their parents (46.2%) was significantly higher than the proportion of boys (20.5%) [χ^2 (1) =4.82; p<.05] and no significant differences in the absolute difference was found.

The caregiver's gender had neither effect on the agreement or the difference.

Considering the relationship between the gender of the child and the caregiver, there was no effect on the agreement, but there were effects on the differences in the Self-perception domain. Differences in Self-perception domain were significantly higher (t (60.274) = 2.05, p<.05) between parents and children when they were of the same gender (mean absolute difference

= 14.48, SD =10.80) than when they were of different genders (mean absolute difference = 9.76, SD =7.50).

Age variable

There were significant effects of age in the Physical well-being domain, where the proportion of agreement was significantly higher in adolescents (51.7%) than children (19.4%) [χ^2 (1) =6.90; p=<.01]. In the Mood & emotions domain the agreement was also higher in adolescents (50.0%) than children (21.2%) [χ^2 (1) =5.73; p=<.05]. Regarding absolute differences, the pattern of results was similar; differences in Psychological well-being (t (64) =2.33, p <.05) were higher in the case of children (mean children = 10.12, SD =8.35 , mean adolescents= 6.18, SD =4.85) and also in Mood & emotions domain (mean =16.72 children, SD =13.38, mean adolescents = 7.65, SD =7.30) there were significantly higher differences in children (t (50.417) =3.38, p <.01).

Gross motor function variable

No significant differences in either the agreement or the degree of agreement were found.

Having siblings variable

There were significant differences in the Self-perception domain [χ^2 (1) = 3.88; p=<.05], where the proportion of agreement was higher when they had no siblings (53.8%) than when they had siblings (25.5%), and in the Autonomy domain [χ^2 (1) =7.22; p=<.01], where the proportion of agreement in those without siblings was 61.5% and in those with siblings was 23.1%. We also found differences in degree of agreement in the Parent relation domain (t (29.324) =1.84, p <.01), where the difference was greater in those with siblings (mean =9.45, SD =7.14) than in those without siblings (mean = 5.62, SD =4.47).

DISCUSSION

Despite the increase in the number of studies addressing the issue of parent–child agreement in assessment the QoL of children with chronic conditions, none has considered Spanish children and adolescents with CP. Our first aim was to determinate the level of agreement between child self-reports and parent-reports of QoL. We found that the correlations between child self-reports and parent proxy reports ranged between low and regular, except for Physical well-being and Social support & peers, where there was moderate correlation. These results indicate that parents of Spanish children and adolescents with CP were more aware on physical and social aspects of their children and are in line with previous researches that have found greater concordance between child and parent ratings for observable behavior as physical functioning (Eiser & Morse, 2001; Klassen et al., 2006; White-Koning et al., 2007).

According to Martin Bland & Altman (1986), strong correlations do not necessarily indicate high agreement. For example, if parents consistently underestimate or overestimate QoL compared with the child's report, the 2 measures may be highly correlated despite large mean differences (Martin Bland & Altman, 1986). In order to obtain a more reliable analysis of our results, we defined agreement in terms of a clinically meaningful difference in quality of life and we analyzed the rate of agreement and disagreement, considering the absolute values of the differences between parent and children (Norman, Sloan, & Wyrwich, 2003; Varni et al., 2005; White-Koning et al., 2007).

The average rate of agreement over all domains was 37.8% while the average rate of disagreement was 62.2%. These results were quite similar to those obtained in a major QoL study including children with CP from 7 European countries (White-Koning et al., 2007). Interestingly, when we analyzed in detail each domain, we found that parents and children had higher percentages of agreement in the Social support & peers (46%), Mood & emotions (44%) and School environment (44%) domains. Physical and Psychological well-being domains had the same rate of agreement (35%). Similar to our results, White-Koning et al. (2007) also had found higher percentages of agreement in the Social support & peers (55%) and Mood & emotions (44%) domains.

These results seem to confirm that correlation cannot be interpreted as indicating agreement. In our study, correlations (Pearson and ICC) were lowest in the Mood & emotions domain, which is consistent with the tendencies found in some studies on the basis of correlation, whereas this same domain had the second highest rate of agreement. However, the results in the Social support & peers domain were high both correlation and of agreement, and suggests that this is one of the KIDSCREEN domains considered more objective because it includes mostly factual questions, related to child's relationships with peers. Indeed, our results are in agreement with previous studies indicating lower parent–child agreement on the more abstract domains of QoL in children with chronic conditions (April, Feldman, Platt, & Duffy, 2006; Klassen et al., 2006; Majnemer et al., 2008; Theunissen et al., 1998; Yeh, Chang, & Chang, 2005).

Studies had suggested that when children and parents evaluate the child's QoL, they may draw on different values and factors that could result in discordance between raters (Eiser & Morse, 2001). While children living with a chronic illness may assess their own QoL dependent upon their subjective experience of how they feel mentally and physically, parents may vary in their awareness, sensitivity and tolerance of children's health concerns (Upton et al., 2008). On the other hand, when internal and experiential factors such as self-perception and experience of emotional support are incorporated into the measurement of QoL, agreement may be understandably less likely (Rosenbaum, 2009).

One important question is why parents underestimate their children's overall QoL, compared with the children themselves. Some authors justified this fact to so-called "disability paradox", i.e. some children with significant health problems are highly satisfied with at least some aspects of their lives, when it may not be apparent to their parents (Albrecht & Devlieger, 1999; Rosenbaum, 2009). Children tend to score themselves at the highest level and emphasize what they can do, whereas parents have the expectation that their child should be able to do everything able-bodied children can do, reflecting, a perspective more related to disability and emphasizing what the child cannot do (Oeffinger et al., 2007). In fact, parents may not adequately appreciate how well their child has adapted overall to their disability (Majnemer et al., 2008).

Our results are comparable to those of previous studies in which parents of children with chronic disorders scored worse HRQoL than children themselves. At the same time, the scores of reliability coefficients were same for both children and parents satisfactory, indicating the consistency of their answers. In our study the parents underestimated their children's QoL in 8 of the 10 domains of KIDSCREEN. Although children also rated their QoL below the cutoff point established by the KIDSCREEN in three domains

(Physical well-being, Support acceptance and Financial resources), in only one domain: Financial resources, parents have assessed QoL better than children. Indeed, the impact of the child's disease and actual problems may have an influence on the parents' experience of stress (Angold et al., 1998; Davis et al., 2007) and thus, their perception of the child's QoL. In this sense, a recent study has shown that parental depression is negatively related to parent proxy-reported QoL and that it may reflect that parents who are depressed view their child's life negatively, or parents of children with poor QoL tend to be depressed (Davis, Mackinnon, & Waters, 2011).

Finally, the second aim was to identify the factors that influence agreement in a sample of Spanish children with CP. We explored whether parent–child agreement was related to child, parental, socio-demographic, and clinical factors. Our results indicate that in the Physical well-being and Mood & emotions domains, adolescents agreed more with their parents than children, contrary to the development perspective, as it is expected that as children get older they spend more time away from parental supervision and share less their experiences with parents. In fact, studies that have compared the level of agreement between children of different ages and their parents report conflicting findings. Similar to our results, Varni et al. (2007) showed that the ICC's increased with the child's age across the scales and summary scores in a sample of 8,591 children ages 5 to 16 years using the PedsQL™ (Varni, Limbers, & Burwinkle, 2007). Cremeens, Eiser, & Blades (2006) and Majnemer et al. (2008) found that agreement increased with increasing age of adolescent, whereas Sattoe et al., (2012) found that adolescents agreeing with their parents were more often younger than the ones who rated their global HRQoL lower than did their parents. Our results can be justified by the fact that parents can be more attentive to the physical function of their adolescents, because, after developmental gains in childhood, persons with CP may decline in gross motor function as they move into adolescence (Bartlett, Hanna, Avery, Stevenson, & Galuppi, 2010), which can affect their mood and emotions.

The gender of caregiver (father or mother) and the motor function did not influence the agreement, which can be justified by the small sample size. The relatively low distribution of cases when comparing fathers and mothers, and in the same way in different GMFCS levels, although this variable was recoded into two groups: those who walk (GMFCS I & II) and those with wheelchair (GMFCS III, IV & V), may have compromised the potential in the statistical analyzes.

Studies had shown that the child's gender did not independently affect child-parent agreement (Britto et al., 2004; Janse, Uiterwaal, Gemke, Kimpen, & Sinnema, 2005; White-Koning et al., 2007), however, in our study, girls have agreed more with their parents than boys, but only in the Autonomy domain. Majnemer et al. (2008) also found that the child's gender influenced the degree of agreement, but in their study there were smaller differences in parent versus child scores of Psychosocial well-being and Emotional functioning domains if the child was male.

We have not found studies that have evaluated the effect of gender of the caregiver-child (both of the same or of different genders) in the degree of agreement, although in our study, parents and children of the same gender have agreed more in the Self-perception domain than parents and children of different genders. Similarly, we explored whether having siblings affected the degree of agreement and we found that in 3 domains (Self-perception, Autonomy and Parental relation & home life) children without siblings were consistent with their parents more than those with siblings. White-Koning et al. (2007) found that the association of not having siblings or have a disabled sibling influenced the disagreement (parent> child), but only in the Social support & peers domain.

LIMITATIONS & CONCLUSIONS

A number of study limitations should be highlighted. Firstly, the study comprised a small convenience sample of children and adolescents with CP that attended 11 of the 68 ASPACE centers found in Spain. Most children and adolescents with CP followed by ASPACE centers had serious impairments, and their level of intellectual impairment made it difficult for them to complete a questionnaire of HRQoL such as KIDSCREEN, making it impossible to have a larger number of participants. Another limitation is that we did not control some variables set by literature as important predictors of the level of agreement between parents and children, such as health status, welfare and parental stress (Davis et al., 2011; Waters et al., 2003; White-Koning et al., 2007). Further research is needed to obtain an even more comprehensive understanding of QoL, based in qualitative methods, which makes it possible, although it was a small sample, to obtain reliable data of children and adolescents with CP and their parents.

In conclusion, our study supports previous findings that parents have a tendency to underestimate their child's QoL (Rajmil et al., 2009; Russell, Hubley, Palepu, & Zumbo, 2006; Sheffler, Hanley, Bagley, Molitor, & James, 2009) and provides evidence that this tendency is valid to children and adolescents with CP in Spain. The children and adolescents included in this study had more optimistic ideas on their physical and psychological well-being, mood & emotions, autonomy, self-perception, school, social support and social acceptance than their parents did. The parents had shown better knowledge in aspects related to social support & peers, emotions and school, and therefore these aspects can more reliably answer, when the self-report of the children is not possible. Moreover, in general, parent-child agreement was higher in the case of adolescents versus children, girls against boys, and finally, for those who had no brothers or sisters.

Finally, we agree with Upton et al. (2008) and believe that disagreement between parents and children's reports of QoL found in our study in itself, unlikely to indicate that either is wrong or right but rather a consequence of each individual's beliefs about the child's health and well-being. Parental and child questionnaires could be measuring different realities, then, parental and child reports should be seen as complementary, and that useful information may be lost if parental reports are not obtained in addition to those provided by their children.

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CAPÍTULO 6

PARTICIPATION IN LEISURE ACTIVITIES OF CHILDREN AND ADOLESCENTS WITH AND WITHOUT CEREBRAL PALSY: COMPARING SPAIN AND THE NETHERLANDS

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ABSTRACT

Purpose: To assess participation in leisure activities of children and adolescents with Cerebral Palsy (CP) in Spain and the Netherlands, comparing findings with typically developing children and adolescents in both countries, and to identify variables related to participation in leisure activities for children and adolescents with CP. Method: The participants were 149 children and adolescents with CP (114 in Spain [43 girls and 71 boys, mean age 12.2 years, range 8-18 years] and 35 in the Netherlands [17 girls and 18 boys, mean age 13.1 years, range 8-18 years]), and 332 typically developing children and adolescents (199 in Spain [102 girls and 97 boys, mean age 13.2 years, range 8-18 years] and 133 in the Netherlands [71 girls and 62 boys, mean age 12.2 years, range 8-18 years]) who completed the Children´s Assessment of Participation and Enjoyment (CAPE) in its Spanish or Dutch version, respectively. Results: Spanish and Dutch children with CP had similar scores in overall diversity and intensity of participation, but there were significant differences in the formal domain and skill-based leisure activities, where the Spanish group scored significantly higher than the Dutch group. Similar results were found in the reference groups. The regression model for the children with CP that included the child variables, type of school and country explained 14% of the variance in overall diversity and intensity, and 28% in diversity in recreational and 22% in active physical activities. Conclusion: The differences between the two countries regarding participation in leisure activities of children with CP may be due to culture and environment.

Keywords: cerebral palsy, participation, leisure activities, children, CAPE.

INTRODUCTION

Over the last few years there has been a growing interest in participation of children and adolescents. Participation is defined by the International Classification of Functioning, Disability and Health (ICF-CY) as involvement in life situations (WHO, 2004).

Participation is important for all children to develop their identity and become active, independent members of society (Michelsen et al., 2009).

Leisure is defined as those activities the individual freely chooses to participate in during their spare time because they find such activities enjoyable (Majnemer et al., 2008). Through participation in leisure activities children develop communication skills, build friendships and gain autonomy and self-efficacy (Law et al., 2004). Thus, leisure activities are essential for the development of all children, particularly for children with disabilities.

Studies have shown that children and adolescents with Cerebral Palsy (CP) and other physical disabilities experience restrictions in participation, spend more time in quiet and sedentary activities and may suffer bullying and social exclusion (Donkervoort et al., 2007; Engel-Yeger et al., 2009; Imms, Reilly, Carlin, & Dodd, 2008; Lindsay & McPherson, 2012; Majnemer et al., 2008; Michelsen et al., 2009). It has been suggested that there is a negative effect of the physical, social, or attitudinal environment contributing to the participation restrictions (Forsyth, Colver, Alvanides, Woolley, & Lowe, 2007; Jönsson, Ekholm, & Schult, 2008; Majnemer et al., 2008¸ Rosenbaum, 2007).

The ICF-CY model states that personal and environmental factors influence participation either as a barrier or a facilitator (WHO, 2004). Given that the environment is a potentially modifiable factor, it is important to identify which features of the physical, social and attitudinal environments are barriers to participation. For example, Hammal, Jarvis, & Colver (2004) showed that level of participation of children with similar types and severity of CP varied according to their place of residence.

As participation is thought to be influenced by environmental factors such as where children live, findings may differ from one country to another. However, evidence about the differences between countries is scarce. One of the major studies presently available is the SPARCLE study (Fauconnier et al., 2009) in which participation in different European countries was assessed. However, Fauconnier et al. (2009) did not specifically focus on participation in leisure activities in children with CP and no comparison with healthy peers was made. Without this comparison it remains unclear whether the differences found between the countries are due to cultural influences or are specific for children with CP.

The present study aimed to identify if there are differences between the levels of leisure participation of children and adolescents with CP and typically developing children and adolescents who live in Spain and the Netherlands and obtain explicative models in both countries. Specifically, we intended:

1. To evaluate the diversity and intensity of leisure participation of children and adolescents with CP in Spain and the Netherlands, comparing findings with leisure participation of typically developing children in both countries (reference group);

2. To describe the type of leisure activities that the children and adolescents with CP and their typically developing peers participated in, in both countries;

3. To examine variables related to participation in leisure activities for children and adolescents with CP in both countries.

METHOD

This paper reports a cross-country study on participation in leisure activities in children and adolescents with and without CP in two countries, Spain and the Netherlands. The study made use of data collected in two earlier studies on the translation and validation of the Children´s Assessment of Participation and Enjoyment (CAPE) in both countries (Bult et al., 2010; Longo, Badia, Orgaz-Baz, & Verdugo Alonso, 2012).

Participants

Children and adolescents with CP, all levels of the Gross Motor Function Classification System (GMFCS) (Palisano et al., 1997); and aged between 8 and 18 years old were selected in both countries. Children and adolescents with low IQ scores (below 50) who could not answer the questions in the CAPE (independently or with assistance) were excluded. For this study, we defined children to be between 8-12 years old and adolescents between 13-18 years old.

The Spanish CP sample came from the 16 Associations Caring for People with Cerebral Palsy and Related Disabilities (ASPACE) in 7 different regions of Spain and from 3 regular schools. The Dutch CP sample consisted of children and adolescents from 7 schools (2 special and 5 regular) in 3 regions of the Netherlands.

In both countries children and adolescents with CP and their parents were contacted by a letter with information on the study. After both the children and the parents who wanted to participate had signed and returned the written consent form, the interviews were arranged.

A reference group of typically developing children (aged between 8 and 18 years old) was recruited in both countries, from regular schools in the same regions as the schools for special education and the ASPACE centers.

Procedure and measures

Ethical approval was provided in the two countries (Ethic Committee of the Universidad de Salamanca - Spain and the University Medical Center Utrecht - the Netherlands).

Participation was measured using the Children's Assessment of Participation and Enjoyment (CAPE). The CAPE is a measure for participation in leisure activities which is appropriate for children and young people aged 6 to 21 years, with or without disabilities (King et al., 2007). The children and adolescents report for each of the 55 items if the activity was performed in the previous 4 months (diversity), and if so, how often (intensity), with whom, where and how much they enjoyed that activity. For the purpose of the present study the diversity and intensity CAPE scores were used.

Participation can be evaluated using the overall scores, i.e., calculated for all 55 activities or can be evaluated using domain scores in formal activities (15 items) and informal activities (40 items). It is also possible to obtain scores for activities types: Recreational (12 items), Social (10 items), Active Physical (13 items), Skill-Based (10 items) and Self-Improvement (10 items).

Evidence of internal consistency, test–retest reliability, content validity, construct validity, convergent and discriminant validity of the CAPE`s original version have been reported (King et al., 2007). In this study two translated and validated versions of CAPE were used: the Dutch version (Bult et al., 2010) and the Spanish version (Longo et al., 2012).

For the samples of children with CP, the CAPE was completed by the child or adolescent in a one-on-one session which was administrated by trained research assistants in the ASPACE center (in Spain) or in the school (in the Netherlands). Parents provided information about the socio-demographics characteristics (as age and gender). Also, information about the GMFCS, the intellectual level (IQ) and the type of school (regular or special) was obtained from the medical records in the center or school which each child attended.

For the reference group, in each country the CAPE was completed by each child or adolescent in the school. For the Dutch children aged 8 to 12 years the CAPE was administered in a one-on-one session with a trained research assistant. Youth aged 12 years and older filled out the CAPE in the classroom after instruction by a trained research assistant. The research assistant was also present during the assessment to answer questions. For the Spanish children the CAPE was administered in a group session in the classroom after instruction by a research assistant and the teachers facilitated the distribution and collection of questionnaires. Each participant provided information about gender and age.

Statistical analysis

Descriptive statistics (frequency, means and standard deviations) were used to summarize the sample characteristics and to describe diversity and intensity of participation.

Between-country differences on diversity and intensity of participation were assessed using Student t-tests for independent samples. These analyses were utilized for the group of children with CP and for the reference group for all domains and activity types of the CAPE.

To gain more insight in the possible differences in diversity of participation between children in Spain and in the Netherlands, the percentages of the children that participated in each of the 55 activities of the CAPE were calculated, both in the CP and the reference groups.

In order to obtain parsimonious explicative models of the participation in leisure activities in children and adolescents with CP who live in both countries, multiple linear regression analysis - forward selection - was used.

Statistical analyses were carried out with SPSS, version 17. The level of statistical significance was set at 0.05.

RESULTS

Group Characteristics

The characteristics of the participants with CP and the reference group are summarized in Table 1. Data were available for a total of 149 children and adolescents with CP (114 in Spain and 35 in the Netherlands) and 332 typically developing children and adolescents (199 in Spain and 133 in the Netherlands). All variables considered were tested for significant differences (independent samples t-test). In the CP group we did not find any significant differences between both countries, except for type of school (97.4% of the Spanish children and adolescents attended regular school compared to 14.3% of the Dutch children and adolescents), and GMFCS levels (25.5% of the Spanish children and adolescents within GMFCS levels IV and V compared to 8.6% of the Dutch children and adolescents). In the reference group, both countries were similar concerning age and gender.

Table 1. Group characteristics of the children with CP and the Reference group in Spain and the Netherlands

Variables	Spain CP (n=114)	Spain Reference (n=199)	the Netherlands CP (n= 35)	the Netherlands Reference (n = 133)
Age (mean, SD, range)	12.2; 3.0; 8-18	13.2; 3.1; 8-18	13.1; 2.8; 8-18	12.2; 2.6; 8-18
Gender				
Female	43 (37.7%)	102 (51.3%)	17 (48.6%)	71 (53.4%)
Male	71 (62.3%)	97 (48.7%)	18 (51.4%)	62 (46.6%)
Intellectual Disability				
None-mild (IQ >70)	80 (70.2%)	-	27 (77.1%)	-
Moderate (IQ 50-70)	34 (29.8%)	-	6 (17.1%)	-
Missing	0 (0%)	-	2 (5.7%)	-
Kind of School				
Regular	111 (97.4%)	199 (100%)	5 (14.3%)	133 (100%)
Special	1 (0.9%)	0 (0%)	30 (85.7)	0 (0%)
Missing	2 (1.8%)	0 (0%)	0 (0%)	0 (0%)
GMFCS				
Level I	39 (34.2%)	-	18 (51.4%)	-
Level II	22 (19.3%)	-	7 (20.0%)	-
Level III	24 (21.1%)	-	7 (20.0%)	-
Level IV	19 (16.7%)	-	2 (5.7%)	-
Level V	10 (8.8%)	-	1 (2.9%)	-

Participation Diversity

Children with CP in Spain and in the Netherlands had comparable scores of overall diversity of participation, as outlined in Table 2. The Spanish children with CP reported higher scores in the formal domain and in the skill-based activities. No significance differences were found between the two countries in the scores of the other domains and activity types. The mean score of 4.3 obtained in the formal domain by the Spanish group and the mean score of 2.7 for the Dutch group represents, respectively 28.6% and 18% of the total formal activities included in the CAPE (15/55). On the other hand, the Spanish group reported a mean score of 20.6 and the Dutch group a mean score of 21.7, representing respectively 51.5% and 54.2% of total informal activities (40/55).

In the reference group, the children from the Netherlands reported higher scores on overall diversity, the informal domain and in social and recreational activities. As in the CP group, the Spanish reference group reported higher scores in the formal domain compared to the Dutch sample.

Table 2. CAPE diversity scores for children and adolescents with and without CP in Spain and the Netherlands

CAPE Domains (Max. Scores)	**Spain CP (N=114)**	**the Netherlands CP (N=35)**	**Spain Reference (N=199)**	**the Netherlands Reference (N=133)**
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
Diversity overall (55)	24.8 (6.5)	24.7 (7.6)	26.1 (7.7)	29.0 (5.4)*
Diversity formal (15)	4.3 (2.3)**	2.7 (2.5)	4.5 (2.2)***	3.6 (1.9)
Diversity informal (40)	20.6 (5.0)	21.7 (6.0)	21.2 (6.3)	24.3 (4.8)***
Diversity recreational (12)	7.6 (2.1)	7.8 (2.5)	7.0 (2.5)	7.9 (2.2)**
Diversity social (10)	7.0 (1.7)	7.1 (1.8)	6.9 (1.8)	7.4 (2.0)*
Diversity active physical (13)	3.2 (1.9)	3.4 (2.0)	4.4 (2.3)	4.6 (1.9)
Diversity skill-based (10)	3.2 (1.8)**	1.9 (1.9)	2.9 (1.6)	2.8 (1.6)
Diversity self-improvement (10)	4.6 (1.8)	4.3 (2.1)	5.2 (1.9)	5.2 (1.9)

SD: Standard Deviation.
***p<.001 **p<.01 *p<.05

Participation Intensity

In the CP group the intensity mean scores ranged from 0.7 (formal) to 3.2 (recreational) in the Netherlands and from 1.1 (active physical) to 3.1 (social) in Spain (Table 3). These results were not very different from those obtained with the reference groups, where mean scores ranged from 1.0 (formal) to 3.3 (recreational) in the Netherlands and from 1.5 (formal) to 3.2 (social) in Spain (Table 3).

Children with CP in the Netherlands participated less frequently than children with CP in Spain in the formal, social, skill-based and self-improvement activities of CAPE.

For the reference groups, the results were similar to the CP group in the formal and skill-based activities, where the Dutch children participated less frequently than Spanish children. However, Dutch children in the reference group participated more frequently in informal and in recreational activities compared to their Spanish peers.

Table 3. CAPE intensity scores for children and adolescents with and without CP in Spain and the Netherlands

CAPE Domains (Scores 1-7)	Spain CP (N=114)	the Netherlands CP (N=35)	Spain Reference (N=199)	the Netherlands Reference (N=133)
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)
Intensity overall	2.1 (0.5)	2.0 (0.6)	2.3 (0.7)	2.3 (0.5)
Intensity formal	1.4 (0.7)***	0.7 (0.5)	1.5 (0.7)***	1.0 (0.5)
Intensity informal	2.4 (0.6)	2.5 (0.7)	2.5 (0.7)	2.8 (0.6)**
Intensity recreational	3.0 (0.9)	3.2 (1.2)	2.8 (1.2)	3.3 (1.1)***
Intensity social	3.1 (0.8)*	2.7 (0.9)	3.2 (0.9)	3.0 (0.9)
Intensity active physical	1.1 (0.7)	1.1 (0.7)	1.6 (0.8)	1.7 (0.7)
Intensity skill-based	1.6 (0.9)***	0.8 (0.8)	1.5 (0.8)**	1.2 (0.8)
Intensity self-improvement	2.4 (0.8)**	1.9 (1.1)	2.7 (1.0)	2.5 (0.9)

SD: Standard Deviation.
***p<.001 **p<.01 *p<.05

Percentage of CAPE´s Activities

Table 4 shows differences between Spain and the Netherlands in 20 activities done most often in both groups of children with CP and the reference groups. Overall, there were many differences between the countries for the type of activities the children with CP participated in. These findings were comparable for the reference groups.

For 10 of the 20 activities there were differences between Spain and the Netherlands of more than 10% of children participating in that specific activity. Hanging out, for example, is the activity performed by most of the Spanish children in the CP group (97%), while this activity was 24th in the rank order of activities (49%) for the Dutch children with CP. Interestingly, the same differences were observed in the reference groups. The largest difference between the countries for the children with CP was observed in Doing gymnastics, which was performed by 71% of the children with CP in Spain (14th in rank order), while only 3% (55th in rank order) of the Dutch children with CP performed this activity. On the other hand, there were similarities between the countries in percentage of children with CP participating in activities, such as Watching TV (97%, 1th in rank order in the Netherlands and 94%, 2th in rank order in Spain), Going to a party (86%, 4th in rank order in the Netherlands and 85%, 5th in rank order in Spain), and Talking on the phone (77%, 11th in rank order in Spain and 74%, 12th in the rank order in the Netherlands).

Table 4. Percentage of children (CP and reference) participating in an activity: comparing countries

Item CAPE	Spain CP (Reference)		the Netherlands CP (Reference)	
	%	Rank	%	Rank
Hanging out* (Informal-Social)	97 (98)	1 (1)	49 (58)	24 (24)
Watching TV (Informal-Recreational)	94 (90)	2 (4)	97 (99)	1 (1)
Entertaining others* (Informal- Social)	90 (80)	3 (10)	74 (93)	13 (3)
Visiting (Informal-Social)	90 (91)	4 (3)	94 (92)	2 (5)
Going to a party (Informal-Social)	85 (78)	5 (12)	86 (90)	4 (6)
Doing homework* (Informal-Self-improvement)	85 (85)	6 (7)	63 (82)	15 (9)
Playing board or card games (Informal-Recreational)	83 (88)	7 (5)	74 (92)	11 (4)
Going for a walk or a hike* (Informal-Recreational)	81 (64)	8 (19)	54 (71)	20 (18)
Listening to music (Informal-Social)	80 (83)	9 (8)	86 (87)	5 (8)
Playing computer or video games* (Informal-Recreational)	80 (88)	10 (6)	91 (95)	2 (2)
Talking on the phone (Informal-Social)	77 (92)	11 (2)	74 (77)	12 (13)
Doing crafts, drawing or coloring (Informal-Recreational)	75 (76)	12 (14)	71 (76)	14 (14)
Playing with things or toys* (Informal-Recreational)	74 (49)	13 (23)	57 (57)	17 (25)
Doing gymnastics* (Formal-Skill-based)	71 (80)	14 (40)	3 (25)	55 (41)
Shopping* (Informal-Self-improvement)	68 (65)	15 (18)	83 (79)	6 (10)
Reading (Informal-Self-improvement)	66 (67)	16 (16)	57 (78)	18 (11)
Going on a full day outing* (Informal-Social)	61 (30)	17 (38)	80 (74)	9 (16)
Going to the movies* (Informal-Social)	60 (56)	18 (21)	80 (75)	8 (15)
Swimming (Formal-Skill-based)	56 (31)	19 (34)	49 (49)	25 (30)
Writing letters (Informal-Self-improvement)	55 (83)	20 (9)	54 (67)	19 (19)

Top 20 activities sorted in descending order for children with CP in Spain, comparing with children with CP in the Netherlands and with children in the reference groups in both countries. *Activities in which the difference in percentage of children performing the activity in the previous 4 months was at least 10%.

Determinants of Participation

The results of the multiple regression analyses in the CP group (N=149) are presented in Table 5 (Diversity) and Table 6 (Intensity).

Regarding diversity, the full model that included the child-related' variables age, gender, GMFCS, IQ level and type of school, and country, explained 14% of the variance in overall diversity, 28% in recreational and 22% in the active physical activity type. GMFCS level, gender and age contributed significantly to this model. Children with a better motor function participated more in overall diversity (β= −.275), the informal domain (β= −.235), self-improvement (β= −.183) and active physical activities (β= −.470). Girls participated more than boys in overall diversity (β= .160), recreational (β= .149) and social activities (β= .208). Children aged 8-12 years of age participated in more diverse activities than adolescents, 13-18 years of age (overall diversity β= −.199), in the informal domain (β= −.198) and in recreational activities (β= −.490). The country variable was significant only for the formal domain, where Spanish children with CP participated in more activities compared with the Dutch children with CP (β= −.352).

Table 5. Multiple Linear Regression for Diversity of Participation in children with CP (n=149)

	Diversity	Formal	Informal	Recreational	Social	Active Physical	Skill-based	Self-improvement
Variables	β	β	β	β	β	β	β	β
Country	,030	-,352*	,059	,018	,174	-,142	-,245	,223
Age	-,199*	-,075	-,198*	-,490***	-,029	-,026	-,151	,073
IQ level	,004	,051	-,022	-,047	,129	,018	,069	-,086
Gender	,160*	,129	,156	,149*	,208*	-,028	,134	,092
School	-,066	,080	-,013	,066	-,163	,158	-,044	-,376*
GMFCS	-,275**	-,095	-,235**	-,136	,062	-,470***	-,119	-,183*
R^2	**.14**	**.10**	**.13**	**.28**	**.06**	**.22**	**.13**	**.09**

Spain n=114; the Netherlands n=35

***p<.001 **p<.01 *p<.05

Variables: Country: 1 Spain, 2 the Netherlands; Age: 1 children, 2 adolescents; IQ level: 1 above 70, 2 50-70; Gender: 1 Boys, 2 Girls; School: 1 Regular, 2 Special. The GMFCS was used as a scale.

For the intensity of participation, the results were comparable to the diversity of participation. The model with the same variables explained 17% of the variance in formal, 21% in recreational and 21% in active physical.

In the same way, GMFCS level was the variable significantly related to most activity types, i.e., children with higher motor function had higher scores in overall intensity (β= −296), in the informal domain (β= −.225), and in self-improvement (β= −.223) and active physical activities (β= −.475). Gender and age were significantly related to recreational activities, where girls participated more frequently than boys (β= .159), and children participated more frequently than adolescents (β= −.414). Again the variable country was significantly related only to the formal activities domain, where the Spanish children with CP participated more often in these activities than the Dutch children with CP (β= −.437).

Table 6. Multiple Linear Regression for Intensity of Participation in children with CP (n=149)

	Intensity	**Formal**	**Informal**	**Recreational**	**Social**	**Active Physical**	**Skill-based**	**Self-improvement**
Variables	β	β	β	β	β	β	β	β
Country	-,066	-,437*	-,038	,120	-,059	-,186	-,229	,007
Age	-,140	-,073	-,147	-,414***	,095	,026	-,159	,091
IQ level	-,012	,004	-,021	,035	,091	-,024	,039	-,108
Gender	,144	,128	,109	,159*	,145	-,035	,131	,080
School	-,114	,045	,028	-,011	-,136	,121	-,116	-,323
GMFCS	-,296***	-,149	-,225**	-,107	,001	-,475***	-,106	-,223**
R^2	**.14**	**.17**	**.08**	**.21**	**.06**	**.21**	**.16**	**.13**

Spain n=114; the Netherlands n=35

***p<.001 **p<.01 *p<.05

Variables: Country: 1 Spain, 2 the Netherlands; Age: 1 children, 2 adolescents; IQ level: 1 above 70, 2 50-70; Gender: 1 Boys, 2 Girls; School: 1Regular, 2 Special. The GMFCS was used as a scale.

DISCUSSION

This study compared the participation of children and adolescents with CP in Spain and in the Netherlands. Spanish and Dutch children and adolescents with CP had similar scores in diversity of participation, but there were significant differences in the formal domain and skill-based activities, where Spain had more diverse scores.

The results regarding intensity of participation show that Spanish children and adolescents with CP, do formal activities and skill-based more often, indicating the same pattern of results found in diversity of participation. Alike results were shown with the reference group which confirms that the pattern is more culturally determined than related to disability.

It is important to emphasize that in both countries, children and adolescents with CP participated less often in formal activities compared to informal activities. The reduced participation in formal activities observed in children and adolescents with CP in this study and in other studies with different samples (Imms et al., 2008; Klaas, Kelly, Gorzkowski, Homko, & Vogel, 2010; Majnemer et al., 2008) is of concern because when young people with disabilities do participate in formal activities, they are more socially engaged, reinforcing the importance of this type of activity as a potential bridge to expanding social networks and experiences (Klaas et al., 2010). Additionally, Law et al. (2006) have showed that the informal activities may be more adversely affected by physical or institutional environmental barriers. The differences in engagement in formal activities found in the samples in the current study may reflect Spanish and Dutch cultural aspects of the two countries. In the same way, the culture and context may partially explain the differences found in skill-based activities. For example, one study with a large sample of children with CP - the SPARCLE study (Fauconnier et al., 2009) - found substantial variations in participation between children in countries in Europe, where children in Denmark showed higher participation levels compared to the others countries evaluated. According to the authors, these results might be explained by the regional policies, legislation and physical environment in the different countries. For example, in Denmark after school clubs are provided by the state which are attended every day by most of children up to age 12, with or without disability. In France and Ireland special transport for disabled children is only provided for going to school, in the UK it depends on the local authority, in Italy it is provided but subject to budgetary limitations, while in Denmark, Sweden and Germany transportation to school is extensively covered. Only Sweden and Denmark have central national resources which facilitate information to families with a child with disability about assistive technology. Moreover, Sweden and Denmark have a strong tradition of state provided services and care for children, while France, Germany, Ireland and Italy have a profile which focuses more on family care (Colver et al., 2011; Fauconnier et al., 2009; Michelsen et al., 2009). These findings suggest that some countries promote participation more than others through policies and regulation at national level (Fauconnier et al., 2009).

The fact that in the present study we were able to include data on participation in leisure activities of typically developing children in both Spain and the Netherlands, helps to expand the understanding of patterns of participation in children and adolescents with CP. For example, when analyzing the results of diversity in the formal activities domain in the reference groups, we observed the same differences between countries as in the group of children with CP, where the Spanish sample conducted more formal activities than the Dutch sample. A recent survey on 'Children and the Future' held in Spain indicates that it is common for parents to enroll their children in various types of formal activities and that children begin to participate in such activities at an early age (Marí-Klose P., Marí-Klose S., Vaquera, & Argeseanu, 2010). In the Netherlands engagement in organized activities is initiated by the parent and the child themselves. Although a lot of groups and clubs are available and it is common to enrol your child in one or two groups or classes, the results of the present study show that this happens less frequently for Dutch children compared to Spanish children. Although Dutch children participate in fewer activities and do so less frequently, they participate more and more frequently in informal activities. Apparently children in the Netherlands participate more in spontaneous activities in or around the house instead of organized activities.

Moreover, Spanish children and adolescents with CP take part more often in social and self-improvement activities than Dutch children and adolescents with CP, which was not observed in the reference groups. When considering the types of activities included in these two domains, one can argue that the type of school may be related to this. As in the Spanish sample 97.4% attends a regular school, while the majority of the children with CP in the Dutch sample (85.7%) attend a school for special education. Given that Dutch children with CP spend more time at school compared to typically development children (since special schools have a longer school day compared to regular schools), this results in less time for extracurricular activities such as hanging out, entertaining others, visiting, doing homework, going for a walk or a hike, shopping, etcetera. As the Spanish children with CP attend regular schools this allows them more free time to take part in such activities. Indeed, in the SPARCLE study children in special schools or special classes participated less frequently in activities as eating out, shopping, playing sports and watching sport events compared with children in regular schools (Michelsen et al., 2009).

At the same time, the type of school may have been a confounding variable, the question whether there were differences among children attending regular school and special school in the two countries could not be assessed due to the fact of the small sample size and the small variation within the groups in each country. Palisano et al. (2009) found that youths with CP in regular school were more likely to engage in activities with their friends than those in special educational programs. Likewise, Italian parents believed that their children´s integration in mainstream school was important to facilitate social interactions (McManus et al., 2006).

In order to enhance our understanding of the differences in diversity of participation in leisure activities between children and adolescents with and without CP in Spain and the Netherlands, we examined the 20 activities that most children and adolescents participated in. The results showed differences in patterns between Spanish and Dutch children and adolescents, regardless of the presence of CP. Some of these differences in participation between the Spanish and Dutch sample seem to reflect culturally determined patterns. For example, while 80% of the sample with CP in the Netherlands participated in the Dutch traditional activity of bicycling, only 34.2% of the Spanish children and adolescents with CP reported taking part in this activity. One study has shown that school-aged children in Ireland and England are more often physically active than children in Italy, France and Germany (Currie et al., 2004), and results of the SPARCLE study indicated that the frequency of children riding a bike was the highest in countries where there is a tradition of bicycling, such as Denmark and Germany (Michelsen et al., 2009). In analogy to the bicycling example, Spanish children and adolescents in the present study more frequently reported 'hanging out' as a favorite activity probably due to the fact that this leisure activity is very traditional in Spain. The results of a Canadian study showed that between 30% and 50% of the children with CP participated in bicycling, at least weekly, while, more than 50% of children with CP participated in hanging out (Majnemer et al., 2008). Interestingly, none of the two activities appeared in the 20 activities that most children participated in, in an Australian study which included 114 children with CP, aged between 10 years 9 months and 12 years 9 months (Imms et al., 2008).

However, there are activities that seem to be free from country differences, such as watching TV and talking on the phone, in which the results between both countries were similar in both the group of children with CP, as well as in the reference group. Watching TV was the main activity carried out by the Dutch children with CP and ranked second for the Spanish children with CP. This was true in other international studies (Canada and Australia) where watching TV ranked first in all activities of CAPE (Imms et al., 2008; Law et al., 2006).

In order to get more insight in the participation of children with disabilities, a key point is the identification of its determinants. Information gathered on determinants of participation might be helpful to assist families and professionals to guide health promotion strategies and policies aiming to optimize the involvement of children in activities at home, school and community. Also, knowledge of activities the child enjoys and family activity orientation has implications for identifying opportunities for participation (Palisano et al., 2009).

The full regression model including child variables in the present study explained 14% of the variance in scores of overall diversity and intensity, and 28% in diversity in recreational and 22% in active physical activities. This rather low level of explained variance supports previous studies that have shown that participation is complex and multidimensional (King et al., 2007; Palisano et al., 2009). The total of unexplained variance could be due to the fact that we were only able to control for environmental variables country and type of school. We agree, therefore, with recent calls in the literature to encourage researchers and rehabilitation professionals to consider environmental factors while assessing child participation (Rosenberg, Ratzon, Jarus, & Bart, 2011).

Not surprisingly, in the present study level of motor function of the child was the variable related to various activity types across the CAPE diversity and intensity scores. Findings of previous studies show that participation of children with CP differs based on their gross motor function (Bult, Verschuren, Jongmans, Lindeman, & Ketelaar, 2011; Imms et al., 2008; King et al., 2007; Majnemer et al., 2008; Orlin et al., 2010; Palisano et al., 2009). Parkes, McCullough, & Madden (2010) reported that gross motor function was a major determinant of participation in eight out of 11 domains of the Life-H questionnaire where increasing severity of impairment was related to a significantly reduced likelihood of high participation among children with CP in Ireland. In addition, intellectual level has been reported to be a determinant of participation of children with CP (Bult et al., 2011; Majnemer et al., 2008). However, we were unable to confirm this finding in our study, possibly because the limited sample size or limited variance within the scores.

Unfortunately, one of the main limitations of our study is that some variables considered important predictors of participation such as environmental and family variables could not be assessed because of the nature of the samples derived from the CAPE validation study in Spain and the Netherlands. Moreover, the different numbers of children in CP groups, and the differences found regarding the type of school and GMFCS levels between children in the two countries also may have affected the results. These factors should be considered in future research.

The strength of our study is that it is the first to compare the participation of two European samples of children and adolescents with and without CP utilizing the CAPE as measurement instrument, having carefully considered it to be the most appropriate tool to evaluate participation. Coinciding with this argument, The SPARCLE group recommends the use of the CAPE, which captures, among other aspects, frequency rather than difficulty of participation and does not incorporate assistance needed into the scoring system, as for example, the Life-H questionnaire (Fauconnier et al., 2009).

The findings of this study highlight that children and adolescents with CP who live in Spain and the Netherlands present similarities and differences in their participation in leisure activities. The differences can be partly accounted for by country specific aspects and partially by variables related to the child's disability. The findings in the reference group confirm the differences between Spain and the Netherlands. In conclusion, this study describes the participation of Spanish and Dutch children and adolescents with CP and with typical developing, contributing to the effort of broadening our knowledge about the participation, their patterns of results in children and adolescents with CP, valuing cross cultural studies.

Implications for Rehabilitation

• The Spanish children and adolescents with CP took part in more formal and skill-based activities than the Dutch children and adolescents with CP. The same differences were found for typically developing children and adolescents. Thus, these differences appear to be culturally determined.

• Gross motor functioning (measured by the GMFCS) was related to various leisure activity types across the CAPE diversity and intensity scores. Country of origin was related only to formal activities.

• This study considers participation as a multidimensional construct which is influenced by environmental aspects, for example, the country where the each child lives.

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CAPÍTULO 7

CONCLUSIONES

Conclusiones generales, limitaciones y futuras líneas de investigación, implicaciones prácticas

Los distintos artículos que forman parte de la presente tesis doctoral representan un punto de partida en el estudio de la participación en las actividades de ocio y de la CDV de los niños y adolescentes con PC en España. Desde esta perspectiva, la investigación desarrollada ha tenido como propósito analizar la participación en las actividades de ocio (diversidad, intensidad y disfrute) y su influencia sobre la CDV.

El primer estudio se justifica por la inexistencia de un instrumento para evaluar la participación en las actividades de ocio en niños y adolescentes adaptado al contexto español, lo que ha resultado en el proceso de traducción, adaptación y validación de la versión española del CAPE (King et al., 2007). En el primer artículo se analizaron las propiedades psicométricas de la versión española del CAPE, en lo cual se obtuvieron los siguientes resultados:

- El CAPE evalúa la participación siguiendo la tendencia actual de incorporar la percepción del propio niño en el desarrollo de instrumentos de medida dirigidos a población infantil (Imms, 2008; Imms et al., 2008). Además, abarca tanto aspectos objetivos como subjetivos del constructo participación, como propone el modelo de la CIF (Hammel et al., 2008). Es de fácil aplicación y permite establecer un "termómetro" de participación en las actividades de ocio, proporcionando información acerca de las diferentes actividades que hace el niño en los últimos 4 meses (formales, informales, sociales, recreativas, físicas, basadas en habilidad y de auto-superación). Además evalúa la frecuencia de participación, con quién realiza dichas actividades, dónde y el grado de disfrute (King et al., 2007).

- Los resultados han mostrado que la versión española del CAPE es un instrumento válido y fiable para evaluar la participación en las actividades de ocio en niños y adolescentes con y sin PC en España.

- La validez de constructo del CAPE fue evaluada considerando que los constructos participación y CDV están relacionados, como se ha demostrado a través de investigaciones recientes (Dahan-Oliel et al., 2012; McManus, Corcoran, & Perry, 2008; Shikako-Thomas et al., 2012). Los resultados de la correlación producto-momento de Pearson entre los dominios del CAPE (diversidad, intensidad y disfrute) y las dimensiones de CDV del KIDSCREEN tanto en la versión niños como en la versión padres demostraron que el instrumento es válido para evaluar la participación en las actividades de ocio.

- La validez discriminante, evaluada mediante pruebas t para muestras independientes, ha comparado los resultados de diversidad e intensidad de participación a través de los diferentes dominios y tipos de actividad del CAPE y se confirmaron las hipótesis de investigación, demostrando que el CAPE es capaz de discriminar los resultados de participación entre diferentes grupos.

- La fiabilidad test-retest analizada mediante la correlación intra-clase (ICC) entre las puntuaciones de diversidad, intensidad, con quién, dónde y disfrute, en la primera y segunda evaluación, demostró que se trata de un instrumento fiable.

Con la disponibilidad de la versión española del CAPE se ha podido llevar a cabo el segundo estudio lo que ha permitido insertar España en el panorama internacional actual en cuando a las investigaciones sobre participación en las actividades de ocio en los niños y adolescentes con PC. Por lo tanto, el propósito del segundo artículo fue establecer el perfil y los determinantes de participación en las actividades de ocio, considerando las variables relacionadas con el propio niño, con la familia y con algunos aspectos del ambiente. Las principales conclusiones del estudio fueron las siguientes:

- En general, los niños y adolescentes con PC en España, igualmente que aquellos que viven en otros países, presentan baja diversidad e intensidad de participación (Bult et al., 2010; Bult, Verschuren, Jongmans, Lindeman, & Ketelaar, 2011; Engel-Yeger, Jarus, Anaby, & Law, 2009; Fauconnier et al., 2009; Imms et al., 2008).

- En cuanto al perfil de participación, los niños y adolescentes con PC en España participaron en un promedio de 20 actividades, siendo 13 actividades del tipo informal y 3 del tipo formal. Además, llevaron a cabo más actividades del tipo social y recreativa. La más baja diversidad de participación en actividades del tipo física, de auto-superación y basada en habilidades encontrada en los niños y adolescentes españoles con PC también fue informada en otros estudios (Bult et al., 2010; Imms et al., 2008; Law et al., 2006; Shikako-Thomas, Majnemer, Law, & Lach, 2008).

- Los niños y adolescentes con PC también presentaron más alta intensidad de participación en actividades del tipo social y recreativa que en los otros tipos de actividad, cuyo promedio representa una frecuencia de una vez al mes.

- En cuanto al disfrute, los niños y adolescentes con PC aunque presenten baja diversidad e intensidad de participación, disfrutan mucho de las actividades en las cuales participan. Los niños y adolescentes con PC disfrutaron participar igualmente en las actividades del tipo formal e informal. En cuanto a los 5 tipos de actividad del CAPE, el nivel de disfrute fue más bajo solamente en las actividades de auto-superación.

- Los resultados demostraron que los factores del niño (edad, género, nivel de la función motora gruesa y nivel de discapacidad intelectual) influyeron sobre la diversidad e intensidad de participación en las actividades de ocio, explicando, respectivamente, un 43% y 40% de las diferencias en las puntuaciones generales de estos dos dominios. Así, por ejemplo, los niños y adolescentes con más limitaciones motoras e intelectual presentaron peores resultados en cuanto a la diversidad e intensidad de participación, lo que es consistente con investigaciones previas (Fauconnier et al., 2009; Kerr, McDowell, & McDonough, 2006; Maher, Williams, Olds, & Lane, 2007; Majnemer et al., 2008; Morris, Kurinczuk, Fitzpatrick, & Rosenbaum, 2006).

- Los factores ambientales estudiados (nivel de ingresos, área de domicilio y tipo de escuela) explicaron un 27% de las diferencias en las puntuaciones de diversidad y un 24% de las diferencias en las puntuaciones de intensidad de participación. Los niños y adolescentes que asistían a escuela regular presentaron más alta diversidad e intensidad de participación que aquellos que asistían a escuela especial.

- Las variables de la familia estudiadas como la edad y género del cuidador, el nivel educacional, cohesión familiar y número de hermanos apenas han explicado las diferencias encontradas en las puntuaciones de diversidad e intensidad de participación.

- Los modelos que incluyeron los factores del niño, de la familia y del ambiente tampoco explicaron las diferencias en las puntuaciones de disfrute, lo que confirma la naturaleza subjetiva del referido dominio (King et al., 2009; Majnemer et al., 2008).

El interés en identificar si la participación influía en la CDV de los niños y adolescentes con PC en España ha resultado en el tercer estudio. Los objetivos específicos fueron los que a continuación se formulan: (1) investigar si la participación afectaba la CDV, independiente del género, edad y grado de discapacidad (motora e intelectual) según la percepción de los padres y, (2) identificar la contribución de la participación en los diferentes tipos de actividad (formal, informal, recreativa, física, social, basada en habilidad y auto-superación) sobre los dominios de CDV, según la percepción de los padres, y se obtuvieron los siguientes resultados:

- Según la percepción de los padres, un aumento en la diversidad de participación mejoraba la CDV en el dominio Apoyo social y pares, cuando controlado el efecto de las variables: género, edad, nivel de la función motora gruesa y grado de discapacidad intelectual.
- Asimismo, un aumento en la intensidad de participación mejoraba la CDV en los siguientes dominios: Bienestar físico, Bienestar psicológico, Autonomía, y Apoyo social y pares, cuando controlado el efecto de las variables mencionadas anteriormente.
- Además, un aumento en el disfrute mejoraba la CDV en los dominios: Bienestar físico, Bienestar psicológico, y Relación con los padres y vida familiar, según la percepción de los padres.
- El análisis detallado por tipo de actividad del CAPE señaló que los niños y adolescentes con PC que llevaban a cabo más actividades (más alta diversidad) del tipo informal y

recreativa se sentían más satisfechos en cuanto al apoyo social y pares, según apuntaban sus padres.

- A parte de eso, los padres percibían que los niños y adolescentes con PC que participaban más frecuentemente en las actividades del tipo informal se sentían más satisfechos con su bienestar psicológico, autonomía y apoyo social y pares. Al mismo tiempo, aquellos que realizaban con más frecuencia las actividades del tipo social también se sentían más satisfechos en cuanto a su bienestar psicológico y en relación con sus pares. Por último, una alta intensidad de participación en actividades basada en habilidades también mejoraba la CDV en cuanto al apoyo social y pares, según apuntaban los padres.
- Por último, los niños y adolescentes que disfrutaban más en las actividades formales, informales, sociales, recreativas, físicas, basadas en habilidades y de auto-superación se sentían más satisfechos en cuanto a su bienestar psicológico, según la percepción de los padres. Asimismo, aquellos que disfrutaban más en las actividades informales y recreativas se sentían mejor físicamente y se relacionaban mejor con los padres. Igualmente, aquellos que disfrutaban más en las actividades físicas estaban más satisfechos en cuanto a su bienestar físico y, finalmente, aquellos que disfrutaban más

en las actividades sociales y de auto-superación se sentían más satisfechos en cuanto a la relación con sus padres.

El no poder haber considerado la perspectiva de los propios niños en cuanto a su CDV en el análisis de la influencia de la participación sobre la CDV ha motivado el desarrollo del cuarto estudio empírico de la presente tesis doctoral. Para ello, se trató de comparar las percepciones de CDV de los niños y adolescentes con PC y de sus padres, en cuanto a la CDV de los niños, evaluados a través del KIDSCREEN-52. Además, se investigó la influencia de las variables del niño y de la familia en el grado de acuerdo. El estudio ha incluido solamente aquellos niños y adolescentes con PC que han podido valorar su propia CDV, lo que ha resultado en una muestra de 66 díadas. Los principales resultados son presentados a continuación.

- Las correlaciones entre las percepciones de los niños y de sus padres fueron bajas en 8 dominios y moderadas en solamente 2 dominios (Bienestar físico y Apoyo social y pares).
- Los padres subestimaron la CDV de sus hijos en 8 de los 10 dominios del KIDSCREEN (Bienestar físico, Bienestar psicológico, Estado de ánimo y emociones, Auto-percepción, Autonomía, Escuela, Apoyo social y pares, y Aceptación social y bullying).

- Además, en el estudio se han considerado las medias de los valores absolutos de las diferencias entre las puntuaciones de los niños y de sus padres como indicadores de acuerdo, como recomienda la literatura actual (Sattoe, van Staa, & Moll, 2012; White-Koning et al., 2007; White-Koning, Grandjean, Colver, & Arnaud, 2008).
- La media de acuerdo considerando todos los dominios conjuntamente fue de 37.8%, variando entre 31% para los dominios Auto-percepción y Autonomía y 46% para Aceptación social y bullying.
- La media de desacuerdo considerando todos los dominios conjuntamente fue de 62.2%. La media de desacuerdo, cuando los padres subestimaron sus hijos, varió entre 21% para el dominio Recursos económicos y 55% para el dominio Auto-percepción. La media de desacuerdo, cuando los padres superestimaron sus hijos varió entre 8% para el dominio Bienestar psicológico y 41% para el dominio Recursos económicos.
- En cuanto a los factores que influían en el grado de acuerdo y desacuerdo, los resultados señalaron que las niñas estaban más de acuerdo con sus padres que los niños en el dominio Autonomía. Además, las diferencias en el dominio Autonomía fueron mayores cuando padres e hijos eran del

mismo sexo. Asimismo, los adolescentes estaban más de acuerdo con sus padres que los niños en los dominios Bienestar físico y Estado de ánimo y emociones. A parte de eso, hubo más diferencias entre padres e hijos en los dominios Bienestar psicológico y Estado de ánimo y emociones, cuando los hijos eran niños en comparación con los hijos adolescentes. Los niños y adolescentes que no tenían hermanos estaban más de acuerdo con sus padres que aquellos que tenían hermanos en los dominios Autonomía, Auto-percepción y Relación con los padres.

- Los resultados indicaron que los padres están más conscientes acerca de aspectos físicos y sociales de sus hijos con PC, lo que es consistente con hallazgos de la literatura (Eiser & Morse, 2001; Klassen, Miller, & Fine, 2006; White-Koning et al., 2008).
- Por último, los resultados sugieren que, aunque distintas, las percepciones de padres e hijos en cuanto a la CDV de los niños se pueden complementar y ambos pueden proporcionar informaciones válidas acerca de la CDV de los niños y adolescentes con PC en España.

El último de los estudios incluidos en esta tesis fue desarrollado en conjunto con el De Hoogstraat Rehabilitation Center - University Medical Center, Utrecht - the Netherlands. Teniendo en cuenta que la participación puede ser influenciada por factores ambientales como, por ejemplo, el país donde la persona vive, el estudio ha comparado los resultados de diversidad e intensidad de participación de los niños y adolescentes con PC que viven en España y Holanda. La inclusión de un grupo de referencia de niños y adolescentes sin discapacidad en ambos países ha posibilitado ampliar la comprensión de las diferencias en los resultados. Los principales hallazgos del estudio son descriptos a continuación:

- Los niños y adolescentes españoles y holandeses con PC presentaron resultados similares en cuanto a las puntuaciones generales de diversidad e intensidad de participación. Sin embargo, los niños y adolescentes españoles con PC presentaron más alta diversidad e intensidad de participación que los niños holandeses en las actividades formales y basadas en habilidad, siendo estas diferencias también encontradas en el grupo de referencia.
- En cuanto al ranking de las actividades que obtuvieron más alto porcentaje de participación en los dos países se pudo observar que en 10 actividades las diferencias fueron superiores al 10%. Así por ejemplo, mientras que Hanging out fue la actividad que ocupó el primer puesto entre los niños españoles, esta actividad ocupó el 24º puesto entre los

niños holandeses, siendo este mismo patrón de diferencia observado en el grupo de referencia.

- Con el objetivo de establecer modelos explicativos de participación para la muestra de niños y adolescentes con PC en los dos países, se llevó a cabo análisis de regresión incluyendo las variables: edad, género, GMFCS, nivel de discapacidad intelectual, tipo de escuela y país de residencia. El modelo ha explicado un 14% de las diferencias en las puntuaciones en cuanto a la diversidad e intensidad de participación y un 28% en diversidad de participación en las actividades recreativas.

- Los resultados del presente estudio demuestran que las diferencias observadas entre los niños y adolescentes con PC que viven en España y Holanda, en general., también son observadas en los niños y adolescentes sin discapacidad, lo que sugiere la influencia cultural en los resultados de participación.

Finalmente, con el desarrollo de los cinco estudios incluidos en la presente tesis se ha aportado información que contribuye al avance en el estudio de la participación en las actividades de ocio y de la CDV de los niños y adolescentes con PC que viven en España. En síntesis, esta tesis doctoral ha posibilitado el disponer en España de una herramienta, válida y fiable, para evaluar la participación en las actividades de ocio de los niños y adolescentes con y sin PC. Se ha identificado que, igualmente a lo que reporta la literatura internacional, la muestra española presenta baja diversidad e intensidad de participación; sin embargo exhibe altos niveles de disfrute, y que los factores relacionados con el niño y con su ambiente explican más las diferencias en las puntuaciones de diversidad e intensidad, que los factores relacionados con la familia. Además, se mostró que, según perciben los padres, la diversidad, intensidad y disfrute de participación influyen en cinco de los dominios de CDV de los niños y adolescentes con PC en España y que el hecho de participar es actividades específicas influye de manera distinta en los dominios de CDV. Asimismo, se averiguó que el nivel de acuerdo entre los niños y adolescentes con PC y sus padres en cuanto a la CDV de los niños es entre bajo y moderado; que los padres conocen mejor aspectos físicos y sociales de sus hijos; y que variables como género, edad y número de hermanos influyen en el grado de acuerdo. Por último, se identificó que los niños y adolescentes con PC que viven en España y en Holanda presentan diferencias y similitudes en cuanto a la participación en las actividades de ocio, que parecen estar más relacionadas con aspectos culturales de los dos países, que con aquellos relacionados con la discapacidad.

Limitaciones del estudio y recomendaciones futuras

Los resultados de los estudios incluidos en esta tesis han de interpretarse a la luz de algunas limitaciones.

En primer lugar, la versión española del CAPE ha sido validada con una muestra de niños y adolescentes con y sin PC (entre 8-18 años), mientras que el instrumento original va dirigido a niños, adolescentes y adultos jóvenes con edades comprendidas entre 6-21 años y también con otras discapacidades físicas, además de PC. Por lo tanto, sería conveniente analizar si el instrumento es válido y fiable para su uso en otras discapacidades y abarcando la franja de edad de los 6-21 años. Además, el instrumento utilizado en paralelo al CAPE: el Preferences for Activities of Children (PAC) no ha sido traducido al contexto español, lo que ha imposibilitado evaluar si los niños y adolescentes con PC en España participan en aquellas actividades que les gustarían realizar, y si los resultados de diversidad de participación reflejan sus elecciones o las de los padres o profesionales. El disponer de la versión española del PAC ayudará a comprender con más profundidad los resultados de participación.

Respecto al estudio de perfiles y determinantes de participación, no han sido incluidas algunas variables relacionadas con el ambiente físico, social y actitudinal. En este sentido se está desarrollando el estudio de la influencia del ambiente físico, social y actitudinal sobre la participación y la CDV de los niños y adolescentes con PC que viven en España. Para ello, se ha traducido y adaptado al contexto español el instrumento European Child Environment Questionnaire (ECEQ), desarrollado por el grupo SPARCLE, lo cual evalúa las necesidades y disponibilidad de aspectos del ambiente físico, social y actitudinal (Colver, 2006; Colver et al., 2011; Dickinson & Colver, 2009;

Forsyth, Colver, Alvanides, Woolley, & Lowe, 2007; Forsyth et al., 2010). En estos momentos se están procesando los datos de la versión española del ECEQ. Además, no se controlaron variables relacionadas con la familia como, por ejemplo, el nivel de stress de los padres, la preferencia de los padres por determinadas actividades de ocio, etc. Según reporta la literatura, dichos aspectos son importantes predictores de los resultados de participación (King et al., 2009; Klaas, Kelly, Gorzkowski, Homko, & Vogel, 2010; Majnemer et al., 2008; Palisano et al., 2011; Shikako-Thomas et al., 2008). Se recomienda también el desarrollo de investigaciones, tanto utilizando la metodología cuantitativa como cualitativa, en las cuales se puedan explorar estos aspectos con mayor profundidad.

La principal limitación relacionada con el tercer estudio fue la utilización de la versión padres del cuestionario de CDV KIDSCREEN-52 para evaluar la influencia de la participación en las actividades de ocio sobre la CDV. El hecho de no haber considerado la percepción de los propios niños puede representar un sesgo y, por lo tanto, los resultados deben ser interpretados con cautela. Sin embargo, el hecho de no obtener una muestra suficientemente amplia de niños y adolescentes con PC que podían contestar al KIDSCREEN-52 en su versión chicos impediría la realización de análisis más profundizados. Se recomienda el desarrollo de investigaciones futuras en las cuales se pueda considerar la percepción de los propios niños y adolescentes con PC respecto a su CDV.

Con referencia al estudio del grado de acuerdo entre padres e hijos en cuanto a la CDV de los niños y adolescentes con PC, las limitaciones están relacionadas al tamaño de la muestra. Se recomienda el desarrollo de estudios de naturaleza cualitativa, lo que permitirá, aunque utilizando muestras reducidas, comprender de manera más profundizada las semejanzas y diferencias entre las percepciones de CDV de los niños y adolescentes con PC y de sus padres. El bajo nivel de acuerdo encontrado en los dominios considerados más subjetivos de CDV puede simplemente reflejar que los padres perciben de manera distinta algunos aspectos de la vida de sus hijos (Eiser & Morse, 2001). Se recomienda capturar ambas percepciones, ya que la información aportada por los padres en determinadas cuestiones puede muy útil y será perdida en el caso de considerar únicamente la percepción del niño.

El estudio comparativo entre los resultados de participación de los niños y adolescentes con PC en España y Holanda también tuvo como principal limitación el tamaño de la muestra. El pequeño número de niños y adolescentes con PC de la muestra Holandesa ha imposibilitado hacer oportunos análisis, como por ejemplo, comparar los niños que asistían a escuela regular/especial en España y Holanda. Además, algunas variables consideradas como importantes predictores de participación no fueron controladas, como por ejemplo el nivel de ingresos de la familia y las barreras del ambiente físico, social y actitudinal. Se recomienda el desarrollo de talleres internacionales en los países que están utilizando el CAPE, para la discusión de diseños metodológicos comunes, lo que posibilitará, igualmente como ha logrado el grupo SPARCLE, comparar los resultados de participación con muestras numerosas de niños y adolescentes con PC y establecer modelos explicativos de participación que ayuden a guiar la políticas públicas de salud.

Implicaciones prácticas

La principal aportación de la presente tesis es abrir un camino incipiente hacia los procesos de intervención dirigidos a los niños y adolescentes con PC que viven en España, incorporándose a la tendencia internacional de cambio en los modelos de intervención centrados en la participación (qué hace el niño o adolescente) y en la calidad de vida (qué siente) más que en los procedimientos predominantes hasta ahora centrados en el trastorno motor; es decir, en enfoques cuyo objetivo era alcanzar un patrón motor normal, meta frecuentemente ilusoria. Los resultados de esta tesis puede representar una buena guía para orientar los proveedores de servicios los cambios necesarios en la planificación de recursos de apoyos para los niños y adolescentes con PC en España, cuya meta es garantizar la participación y la CDV.

Conclusions, limitations and future research, practical implications

The various papers that are part of this thesis represent a starting point in the study of participation in leisure activities and QoL of children and adolescents with CP in Spain. From this perspective, the research undertaken was purposely to analyze participation in leisure activities (diversity, intensity and enjoyment) and its influence on QoL.

The first study is justified by the lack of an instrument to assess participation in leisure activities in children and adolescents adapted to the Spanish context, which has resulted in the process of translation, adaptation and validation of the Spanish version of CAPE (King et al. 2007). In the first article we examined the psychometric properties of the Spanish version of CAPE, in which the following results were found:

- CAPE assesses participation following the current trend of incorporating perception of child himself in developing measurement instruments aimed at children (Imms, 2008; Imms et al., 2008). It also includes both the objective and subjective aspects of participation construct, as proposed by the ICF model (Hammel et al., 2008). It is easy to use and enables a "thermometer" of participation in leisure activities, providing information about the different activities that the child did in the last four months (formal, informal, social, recreational, physical, skill-based and self-improvement). It also assesses the frequency of participation, with whom performs such activities, where and the level of enjoyment (King et al., 2007).

- The results have shown that the Spanish version of CAPE is a valid and reliable instrument to assess participation in leisure activities in children and adolescents with and without PC in Spain.

- The construct validity of CAPE was assessed considering participation and QoL constructs are related, as demonstrated by recent research (Dahan-Oliel et al., 2012; McManus, Corcoran, & Perry, 2008; Shikako-Thomas et al., 2012). The results of the Pearson product-moment correlation coefficient among the domains of CAPE (diversity, intensity and enjoyment) and domains of QoL of the KIDSCREEN version both the children and parents versions showed that the instrument is valid for evaluating participation in leisure activities.

- The discriminant validity assessed by independent sample t tests, has been compared with the results of diversity and intensity of participation across different CAPE domains and types of activity and has confirmed the research hypothesis, showing that CAPE can discriminate participation results between different groups.

- The test-retest reliability analyzed by intra-class correlation (ICC) between the scores of diversity, intensity, with whom, where and enjoyment during the first and second evaluation confirmed that it is reliable instrument.

With the availability of the Spanish version of CAPE we were able to conduct the second study which has allowed insertion of Spain on the international scene where current research on participation in leisure activities in children and adolescents with CP are conducted. Therefore, the purpose of the second article was to establish the profile and determinants of participation in leisure activities, considering the variables related to the child himself, with family and with some aspects of the environment. The main conclusions of the study were:

- In general, the results showed that children and adolescents with CP in Spain, also have low diversity and intensity of participation as those living in other countries (Bult et al., 2010; Bult, Verschuren, Jongmans, Lindeman, & Ketelaar, 2011; Engel-Yeger, Jarus, Anaby, & Law, 2009; Fauconnier et al., 2009; Imms et al., 2008).

- Regarding the profile of participation, children and adolescents with CP in Spain participated in an average of 20 activities, 13 activities being informal and 3 formal. In addition, more social and recreational activities were carried out. The lowest diversity of participation in physical active, self-improvement and skill- based activities found in Spanish children and adolescents with CP has been also reported in other studies (Bult et al., 2010; Imms et al., 2008; Law et al., 2006; Shikako-Thomas, Majnemer, Law, & Lach, 2008).

- Children and adolescents with CP also had higher frequency of participation in social and recreational than in other types of activity, which represented an average frequency of once a month.

- Children and adolescents with CP even if they have low diversity and intensity of participation really enjoy the activities in which they participate in. Children and adolescents with CP enjoyed participating equally in formal and informal activities. As for the 5 types of activity of CAPE, the enjoyment level was lower only in the self-improvement activities.

- The results showed that child factors (age, gender, level of gross motor function and level of intellectual impairment) influenced the diversity and intensity of participation in leisure activities, explaining, respectively, 43% and 40% differences in the overall scores of these two domains. For example, children and adolescents with more motor and intellectual limitations had worse results in terms of the diversity and intensity of participation, which is consistent with previous research (Fauconnier et al., 2009; Kerr, McDowell, & McDonough, 2006; Maher, Williams, Olds, & Lane, 2007; Majnemer et al., 2008; Morris, Kurinczuk, Fitzpatrick, & Rosenbaum, 2006).

- The environmental factors studied (income level, area of residence and type of school) explained 27% of the differences in diversity scores and 24% of the differences in intensity of participation. Children and adolescents attending regular school showed higher diversity and intensity of participation than those attending special school.

- The family variables studied such as age and gender of the caregiver, educational level, family cohesion and number of siblings only explained the differences found in scores of diversity and intensity of participation.

- Models that included child, family and environment factors did not explain the differences in scores of enjoyment, which confirms the subjective nature of the construct (King et al., 2009; Majnemer et al., 2008).

The interest in identifying whether participation affected the QoL of children and adolescents with CP in Spain has resulted in the third study. The specific objectives of this study were the following: (1) investigate whether participation affected QoL, independent of gender, age and level of impairment (motor and intellectual) as perceived by parents and, (2) identify the contribution of participation in different types of activity (formal, informal, recreational, physical, social, skill-based and self-improvement) over the domains of QoL, as perceived by parents.

- The findings indicate that, as perceived by the parents, an increase in the diversity of participation improved QoL in the Social support & peers domain, when controlled by the effects of the variables: gender, age, gross motor function and intellectual impairment levels.

- Similarly, an increase in the intensity of participation improved QoL in the following domains: Physical well-being, Psychological well-being, Autonomy, and Social support & peers, when the same variables were controlled.

- Furthermore, an increased in the enjoyment improved QoL in the following domains: Physical well-being, Psychological well-being, Autonomy, and Social support & peers, when the same variables were controlled.

- The detailed analysis by type of CAPE activity showed that children and adolescents with CP who carried out more informal and recreational activities (higher diversity) felt more satisfied as to the social support & peers, as perceived by their parents.

- Apart from that, the parents perceived that children and adolescents with CP who participated more frequently in informal activities were more satisfied with their psychological, autonomy and social support & peers. At the same time, those who performed more often social activities also were more satisfied in their psychological well-being and relationship with peers. Finally, the high intensity of participation in skills-based activities also improved QoL regarding social support & peers, as pointed out by the parents.

- Lastly, children and adolescents who enjoyed more formal, informal, social, recreational, physical, skill-based and self-improvement activities were more satisfied in their psychological well-being, as perceived by parents. Likewise, those who enjoyed more informal and recreational activities felt better physically and interacted better with parents. Equally, those who enjoyed more physical activities were more satisfied in their physical and finally those who enjoyed more social and self-improvement activities were more satisfied in the relationship with their parents.

Failing to have considered the perspective of children themselves in their QoL in the analysis of the influence of participation on QoL has motivated the development of the fourth empirical study of this thesis. For this, we compared the perceptions of QoL of children and adolescents with CP and their parents, concerning the QoL of children, evaluated through KIDSCREEN-52. In addition, we investigated the influence of the variables of the child and caregiver in the degree of agreement. The study included only those children and adolescents with CP who were able to assess their own QoL, which has resulted in a sample of 66 days. The main results are as follows.

- The results showed that the correlations between the perceptions of children and their parents were low in eight domains and moderate in only 2 domains (Physical well-being and Social support & peers).

- Parents underestimated their children's QoL in 8 of the 10 domains of KIDSCREEN (Physical well-being, Psychological well-being, Mood & emotions, Self-perception, Autonomy, School, Social support & peers, and Social acceptance (bullying).

- We also considered the average of the absolute values of the differences between the scores of children and their parents as indicators of agreement, as recommended by current literature (Sattoe, van Staa, & Moll, 2012; White-Koning et al., 2007; White-Koning, Grandjean, Colver, & Arnaud, 2008).

- The mean agreement considering all the domains together was 37.8%, ranging from 31% for the Self-perception and Autonomy domains and 46% for the Social acceptance (bullying) domain.

- The mean disagreement considering all the domains together was 62.2%. The average disagreement (when parents underestimated their children) ranged from 21% for the Financial resources domain and 55% for the Self-perception domain. The mean disagreement, when parents overestimated their children ranged from 8% for the Psychological well-being domain and 41% for the Financial resources domain.

- Regarding the factors influencing the degree of agreement and disagreement, the results showed that girls were more in line with their parents than boys in the Autonomy domain. In addition, differences in the Autonomy domain were higher when parents and children were the same sex. Also, adolescents agreed more with their parents than children in the Physical well-being and Mood & emotions domains. Other than that, there were more differences between parents and children in the Psychological well-being and Mood & emotions domains, when the children were compared with adolescents. Children and adolescents without siblings agreed more with their parents than those who had siblings in the Autonomy, Self-perception and Parent relation domains.
- The results indicated that parents are more aware of social and physical aspects of their children with CP, which is consistent with findings from the literature (Eiser & Morse,

2001; Klassen, Miller, & Fine, 2006; White-Koning et al., 2008).

- Finally, the results suggest that, although different, perceptions of parents and children regarding the QoL of children can both complement and provide valid information about the QoL of children and adolescents with CP in Spain.

The last of the studies included in this thesis was developed in conjunction with the De Hoogstraat Rehabilitation Center - University Medical Center, Utrecht - The Netherlands. Since participation can be influenced by environmental factors such as the country where the person lives, the study compared the results of diversity and intensity of participation of children and adolescents with CP living in Spain and the Netherlands. The inclusion of a reference group of children and adolescents without disabilities in both countries has enabled a broader understanding of the differences in the results. The main findings of the study are:

- The Spanish and Dutch children and adolescents with CP showed similar results in terms of the overall scores of diversity and intensity of participation. However, Spanish children and adolescents with CP had higher diversity and intensity of participation than Dutch children in formal activities and skill-based, these differences were also found in the reference group.

- As for the ranking of the activities which had the highest percentage in the two countries, the differences in 10 activities were above 10%. For example, while Hanging out was ranked first among Spanish children with CP, this activity took the 24th place among Dutch children with CP and this same pattern of difference was observed in the reference group.

- In order to establish explanatory models of participation for the sample of children and adolescents with CP in the two countries, a regression analysis was conducted including the variables: age, gender, GMFCS, intellectual disability level, school type and country of residence. The model explained 14% of the differences in scores in terms of diversity and intensity of participation and 28% in recreational activities.

- The results of this study advocate that the observed differences between children and adolescents with CP living in Spain and the Netherlands, in general, are also observed in children and adolescents without disabilities, suggesting cultural influence on the results of participation.

Finally, with the development of the five studies included in this thesis we provided information that contributes to progress in the study of participation in leisure activities and QoL of children and adolescents with CP living in Spain. In summary, this thesis has enabled the availability of a valid and reliable instrument in Spain, to assess participation in leisure activities of children and adolescents with and without CP. Even though the Spanish sample was identified to have low diversity and intensity of participation, which was also reported in international literature, it still showed high levels of enjoyment, and that factors related to the child and their environment explained more the differences in diversity and intensity scores, than family-related factors. Furthermore, it was shown that the diversity, intensity and enjoyment of participation influenced five domains of QoL of children and adolescents with CP in Spain, and the fact that participated in specific activities influenced differently the domains of QoL. It was also found that the level of agreement between children and adolescents with CP and their parents regarding QoL of children is low to moderate, parents have are more aware about physical and social aspects of their children, and that variables such as gender, age and number of siblings influence the degree of agreement. Finally, it was found that children and adolescents with CP living in Spain and the Netherlands have differences and similarities in terms of participation in leisure activities which seem to be more related to cultural aspects of the two countries, than with those related to disability.

Study limitations and future recommendations

The results of the studies included in this thesis must be interpreted in light of some limitations.

Firstly, the Spanish version of CAPE has been validated with a sample of children and adolescents with and without CP (8-18 years), while the original instrument is aimed at children, adolescents and young adults aged 6-21 years and with other physical disabilities, as well as CP. Therefore, it would be appropriate to analyze whether the instrument is valid and reliable for use in other disabilities and covering the age range of 6-21 years. In addition, the instrument used in parallel to CAPE: the Preferences for Activities of Children (PAC) has not been translated into Spanish context, making it impossible to assess whether children and adolescents with CP in Spain participate in activities they would like to do, and if the results diversity of participation reflect their elections or those of parents or professionals. Having the Spanish version of the PAC will help us understand more fully the results of participation.

Regarding the study of profiles and predictors of participation, some variables related to the physical, social and attitudinal environment have not been included. In this sense the study of the influence of environment on participation and QoL of children and adolescents with CP living in Spain is being developed. For this the European Child Environment Questionnaire instrument (ECEQ), developed by the group SPARCLE, which assesses the needs and issues related to the availability of physical, social and attitudinal has been translated and adapted to the Spanish context (Colver, 2006; Colver et al., 2011; Dickinson & Colver, 2009; Forsyth, Colver, Alvanides, Woolley, & Lowe, 2007; Forsyth et al., 2010). Currently, the data of the Spanish version of ECEQ is being processed. Furthermore, family-related variables such as stress levels of parents, parents' preference for certain leisure activities, etc. have not been controlled. According to literature reports, these aspects are important predictors of the results of participation (King et al., 2009; Klaas, Kelly, Gorzkowski, Homko, & Vogel, 2010; Majnemer et al., 2008; Palisano et al., 2011; Shikako-Thomas et al., 2008). It also recommends the development of research, using both quantitative and qualitative methodology, in which to explore these issues in greater depth.

The main limitation related to the third study was the use of the parent version of the QoL questionnaire KIDSCREEN-52 to evaluate the influence of participation in leisure activities on QoL. The fact of not considering the perception of children themselves may represent a bias and, therefore, the results should be interpreted with caution. However, the failure to obtain a sufficiently large sample of children and adolescents with CP who could answer the KIDSCREEN-52 children version has impeded a more in-depth analysis. The development of future research into considering the perception of one's children and adolescents with CP in respect to their own QoL is recommended.

With reference to the study of the degree of agreement between parents and children regarding the QoL of children and adolescents with CP, limitations are related to sample size. The development of qualitative studies, allowing, albeit using small samples, a more in-depth understanding of the similarities and differences between the perceptions of QoL of children and adolescents with CP and their parents is recommended. The low level of agreement found in the more subjective QoL domains may simply reflect that parents perceive differently in some aspects of their children's lives (Eiser & Morse, 2001). Capturing both perceptions is recommended, as the information provided by the parents on specific issues may be very useful and will be lost in the case of considering only the child's perception.

The comparative study of the results of participation of children and adolescents with CP in Spain and the Netherlands also had the main limitation of the sample size. Appropriate analysis, such as comparing the children attending regular versus special school in Spain and the Netherlands was not possible due to the small number of children and adolescents with CP in the Dutch sample. In addition, some important variables considered as predictors of participation were not controlled, such as family income level and physical, social and attitudinal barriers. We recommend the development of international workshops in countries that are using the CAPE questionnaire, to discuss common methodological designs, which has been achieved by the SPARCLE group, will enable the comparison the results of participation with numerous samples of children and adolescents with CP and establish explanatory models of participation to help guide public health policies.

Practical implications

The main contribution of this thesis is to open a initial path towards intervention processes aimed at children and adolescents with CP living in Spain, joining the international trend of change in the intervention models that focus on participation (what the child or adolescent does) and QoL (what the child or adolescent feels) rather than predominant procedures concentrating on motor disorder which focuses on achieving a normal motor pattern, often an illusory goal. The results of this thesis can be a good guide to orient service providers the necessary changes in support resource planning for children and adolescents with CP in Spain, whose goal is to ensure participation and QoL.

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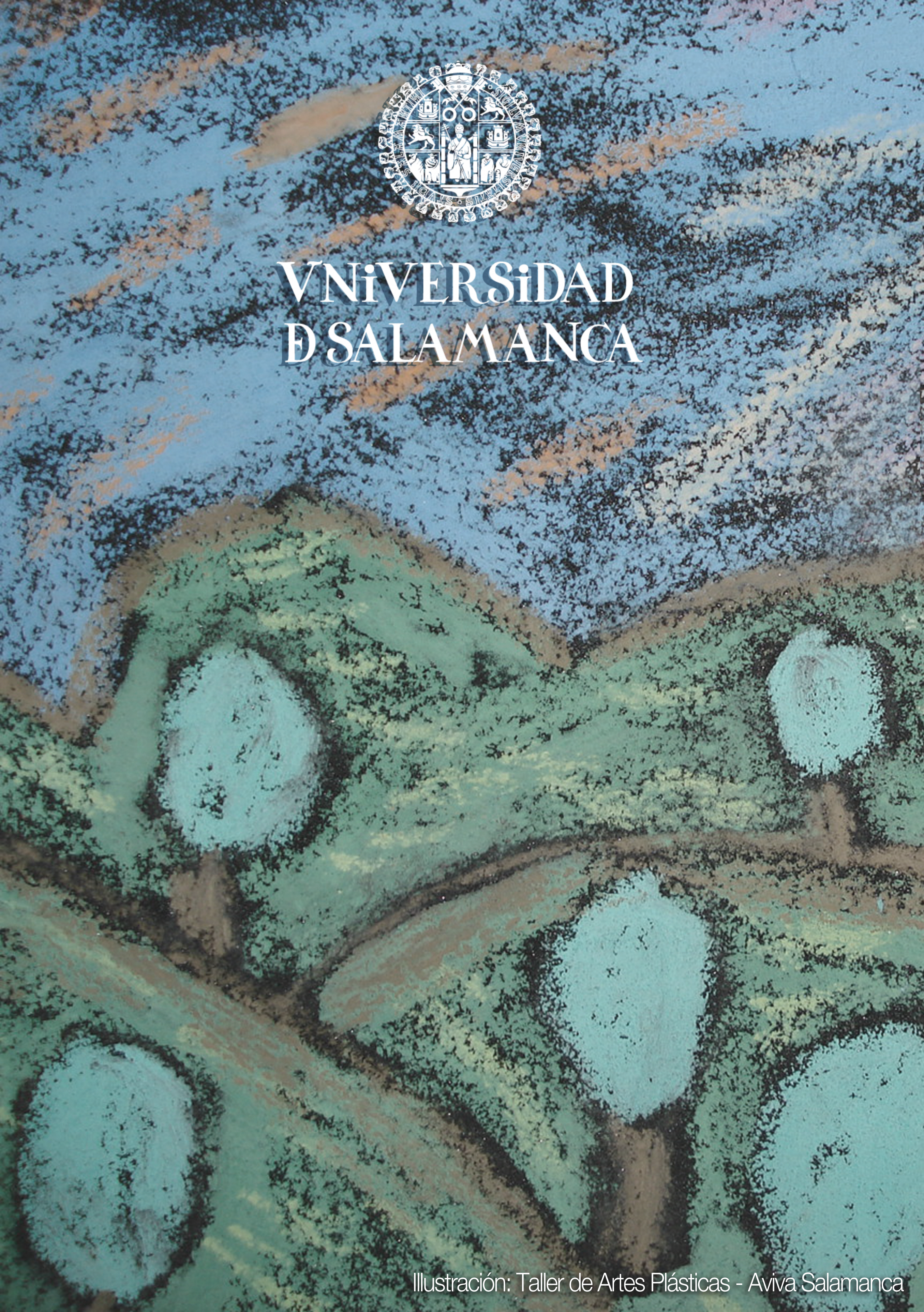

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