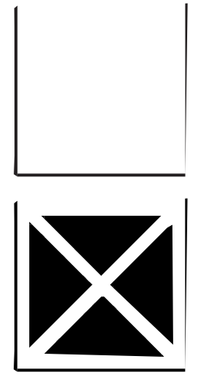
THE PSYCHOSOCIAL ADAPTATION OF CHILDREN AND ADOLESCENTS
WITH CEREBRAL PALSY AND THEIR PARENTS:
A DIFFERENT MATTER OR THE MATTER OF A DIFFERENCE?

Carlos Fernando de Paulo Carona

The University of Coimbra | 2013



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Title: The Psychosocial Adaptation of Children and Adolescents with Cerebral Palsy and their Parents – A Different Matter or the Matter of a Difference?

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Doctoral dissertation in Clinical Psychology, submitted to the Faculty of Psychology and Education Sciences of Coimbra University, under the supervision of Prof. Dr. Maria Cristina Canavarro and Prof. Dr. Martin Bax.

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You see someone on the street
and essentially what you notice about them is the flaw.

Diane Arbus

To my Godchildren,
Matias and Manuel

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In the course of my doctoral pursuit of nearly five years, especially during the phases of most solitary work, I was fortunate to count on the imponderable support provided by many people: some of them have been there since the beginning, others have come to be acquainted with me in the meantime, and others still have encountered me occasionally, both in joyful and in stressful times. Since this section of acknowledgments is not intended to be an exhaustive and flawless list of my enduring feelings of gratitude, I want to state clearly that all those many kindnesses have not been forgotten. Nevertheless, I am compelled to revisit here some of those whose memories have never left me – for the record, I mean.

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am sincerely thankful to Prof. Margarida Girão and Prof. Conceição Carvalho for their personal commitment with the on-time accomplishment of an adequate sample of controls. In addition, I must express my esteem for my work colleagues and coordinators at Coimbra Cerebral Palsy Association, and especially for Prof. Paulo Ferreira, whose collaboration in formatting and designing the Portuguese versions of Disabkids questionnaires and the present dissertation was basically irreprehensible.

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List of Abbreviations and Acronyms

APA | American Psychological Association

CCD | Committee on Children with Disabilities

CNS | Central Nervous System

CP | Cerebral Palsy

CPACFH | Committee on Psychosocial Aspects of Child and Family Health

DSM | Diagnostic and Statistical Manual of Mental Disorders

GMFCS | Gross Motor Function Classification System

HRQL | Health-related Quality of Life

ICD | International Classification of Diseases

IQ | Intelligence Quotient

INE | Instituto Nacional de Estatística [National Institute of Statistics]

INSA | Instituto Nacional de Saúde [National Institute of Health]

QL | Quality of Life

SCPE | Surveillance of Cerebral Palsy in Europe

SEM | Structural Equation Modeling

SES | Socio-economic Status

WHO | World Health Organization

WHOQOL | World Health Organization Quality of Life

Abstract

Background. The assessment of pediatric outcomes has been progressively moving from an exclusive focus on morbidity, mortality and function, to an overarching and subjective assessment that includes physical and psychosocial components of well-being, and values the patients and their families' perceptions. In this context, "The Disabkids Questionnaires" were developed to assess and promote the health-related quality of life (HRQL) of pediatric populations, as perceived by children and adolescents with chronic physical conditions and their parents/caregivers. As regards the specific population of pediatric cerebral palsy (CP), psychosocial research has developed slowly and it has mainly described impairments in the adaptation outcomes of children/adolescents with CP and their parents, while generally relying on parents' reports on their children's adjustment, and comparing children's outcomes to normative data. Moreover, there is limited research on potentially modifiable psychosocial factors and links related to those outcomes. Therefore, the objective of this research was two-folded: firstly and preliminarily, to develop the Portuguese versions of Disabkids-37 questionnaires (generic measures of pediatric HRQL); and secondly, to analyze adaptation outcomes, related psychosocial factors and potential mechanisms in children/adolescents with CP and their parents, mostly in comparison to typically developing children/adolescents and their parents.

Methods. This cross-sectional research project integrated two sequential phases. During a first preliminary phase, aimed at developing the Portuguese versions of Disabkids-37 questionnaires, 349 children/adolescents with chronic conditions (asthma and epilepsy) and one of their parents were administered clinical and socio-demographic datasheets, and the self and proxy-versions of Disabkids-37, Strengths & Difficulties Questionnaire (SDQ) and Kidscreen-10. The psychometric properties of Disabkids-37 were then analyzed in agreement with classical test theory. During the second phase, aimed at studying parent-child psychosocial adaptation in the context of pediatric CP, two independent samples with 223 dyads of parents and their children with and without CP, were administered the following measures (in addition to clinical and socio-demographic datasheets): for children/adolescents, Disabkids-37 (only for pediatric CP), SDQ, Kidscreen-10 and the Satisfaction with Social Support Scale for Children and Adolescents; for parents (in addition to proxy-reports on their children's adaptation outcomes), WHOQOL-BREF/EuroHis-QoL-8, The Revised Burden Measure, Mental Health Inventory – short form, and the Satisfaction with Social Support Scale. Univariate and multivariate analyses of covariance were conducted to examine differences in variables between subsamples. PROCESS

computational tool was used for path analysis-based mediation, moderation and moderated mediation analyses. SEM path models were also used for examining direct and indirect links between predictors and outcomes.

Results. The developmental adequacy and psychometric quality of the Portuguese versions of Disabkids-37 was ascertained, thus indicating this instrument as a valid and reliable measure to assess pediatric HRQL outcomes in Portugal, within a cross-cultural and developmental perspective. As regards the second, central phase of the research project, the following results are highlighted: (1) children/adolescents with CP reported lower levels of social support in comparison to their typically developing peers, but no significant differences emerged in terms of their psychological maladjustment; (2) internalizing and externalizing problems were found to mediate the link between social support and HRQL in children/adolescents with CP; (3) parents of these children/adolescents reported a similar quality of life (QL), but more subjective burden and less caregiving uplifts, in comparison to parents of typically developing children; (4) caregiving uplifts were found to moderate the associations between objective and relational burdens, and the Psychological and Social QL of parents of children with CP; in addition, differential main effects of burdens and uplifts on their QL were also observed; (5) caregiving burden was linked to parents and their children's psychological maladjustment and QL both directly (except for children's QL) and indirectly through social support; and finally, (6) these latest mediation effects were invariant in dyads of parents and their children with and without CP.

Conclusions. Disabkids-37 questionnaires are valid and reliable measures to incorporate a developmental approach in pediatric assessment routines. Interventions targeting perceived social support in children/adolescents with CP may improve their HRQL via the promotion of their psychological adjustment. A multidimensional assessment is to be preferred for understanding the caregiving experience of parents who have children with CP; in clinical interventions, the reduction of subjective burden may improve their overall QL, and the promotion of caregiving uplifts may attenuate the deleterious effects of burden or even exert direct beneficial effects on their psychological and social QL. Additionally, caregiving burden may be assumed as a strategic target for psychosocial interventions in the context of pediatric CP, since its reduction may be linked to improved psychological and QL outcomes in parents and children, via their enhanced perceptions of social support. Finally, it is suggested that clinical and social interventions should target condition-related challenges and specificities, whilst acknowledging and facilitating normative developmental/adaptation issues and processes in the context of pediatric.

Resumo

Introdução. A avaliação de resultados em saúde pediátrica tem estendido o seu foco para além dos temas da morbilidade, mortalidade e funcionalidade, de forma a incluir as mais recentes orientações de uma avaliação subjetiva, integrativa dos componentes físico, psicológico e social do bem-estar, e valorativa das perceções dos doentes e suas famílias. Neste contexto, os "Questionários Disabkids" foram desenvolvidos para avaliar e promover a qualidade de vida relacionada com a saúde (QVrS) em populações pediátricas, de acordo com as perceções das crianças/adolescentes com condições crónicas de saúde e suas famílias. Relativamente ao domínio da paralisia cerebral (PC) pediátrica, a investigação psicossocial tem vindo a desenvolver-se lentamente, e descrevendo sobretudo os resultados de adaptação de crianças/adolescentes com PC e dos seus pais, que são geralmente avaliados com recurso aos relatos dos pais sobre o ajustamento dos filhos, e tendo por base comparações com dados normativos. Além disso, os fatores psicossociais potencialmente modificáveis, bem como as suas relações com os resultados de adaptação, têm sido pouco estudados. Por conseguinte, o objetivo desta investigação foi duplo: primeira e preliminarmente, desenvolver as versões Portuguesas dos questionários Disabkids-37 (medidas genéricas da QVrS pediátrica); e em segundo lugar, analisar resultados, fatores associados e potenciais mecanismos da adaptação psicossocial de crianças/adolescentes com PC e seus pais, sobretudo em comparação com crianças/adolescentes sem alterações de desenvolvimento e seus pais.

Metodologia. Esta investigação de desenho transversal integrou duas fases sequenciais. Durante a primeira fase de estudos preliminares, direcionada para o desenvolvimento das versões portuguesas dos questionários Disabkids-37, 349 crianças/adolescentes com condições crónicas de saúde (asma e epilepsia), e um dos seus pais, preencheram fichas de dados clínicos e sociodemográficos, e as versões de auto e heterorrelato dos instrumentos Disabkids-37, Questionário de Capacidades e Dificuldades (SDQ) e Kidscreen-10. As propriedades dos questionários Disabkids-37 foram então analisadas em conformidade com a teoria clássica da validação psicométrica. Durante a segunda fase, dirigida ao estudo da adaptação psicossocial de pais e filhos no contexto da PC pediátrica, 223 díades de pais e seus filhos com/sem PC (duas amostras independentes) preencheram os seguintes instrumentos (para além das fichas de dados clínicos e sociodemográficos): no caso das crianças/adolescentes, o Disabkids-37 (apenas na amostra pediátrica), o SDQ, o Kidscreen-10, e a Escala de Satisfação com o Suporte Social para Crianças e Adolescentes; no caso dos pais (em acréscimo aos heterorrelatos sobre os resultados

de adaptação dos filhos), o WHOQOL-BREF/EuroHis-QoL-8, a Escala de Desgaste do Cuidador – Revista, o Inventário de Saúde Mental – abreviado, e a Escala de Satisfação com o Suporte Social. Ao nível do tratamento estatístico dos dados, foram realizadas análises univariadas e multivariadas de covariância, para avaliar diferenças nas variáveis entre amostras. O programa PROCESS foi utilizado para a realização de análises de mediação, moderação e mediação moderada baseadas na regressão estatística. Os modelos de equações estruturais foram igualmente aplicados à avaliação dos efeitos diretos e indiretos dos preditores sobre os resultados de adaptação.

Resultados. A adequação desenvolvimental e a qualidade psicométrica das versões Portuguesas dos questionários Disabkids-37 foram estabelecidas, indicando assim este instrumento como uma medida válida e fiável para avaliar resultados de QVrS pediátrica em Portugal, numa perspetiva transcultural e desenvolvimental. Por outro lado, foram observados os seguintes resultados principais, na subsequente fase nuclear da investigação: (1) as crianças/adolescentes com PC relataram níveis inferiores de apoio social em comparação com os seus pares sem alterações de desenvolvimento, mas não foram observadas diferenças significativas ao nível do seu ajustamento psicológico; (2) os problemas internalizantes e externalizantes funcionaram como mediadores na relação entre o apoio social e a QVrS de crianças/adolescentes com PC; (3) os pais destas crianças/adolescentes relataram uma qualidade de vida (QV) semelhante, mas mais desgaste subjetivo e menos gratificações na prestação de cuidados, em comparação com os pais de filhos sem alterações de desenvolvimento; (4) as gratificações na prestação de cuidados moderaram as relações entre os desgastes objetivo e relacional, e respetivamente, a QV psicológica e social dos pais de filhos com PC; adicionalmente foram observados efeitos diretos, diferenciados, dos diferentes tipos de desgaste e das gratificações, na QV destes pais; (5) o desgaste da prestação de cuidados associou-se com o desajustamento psicológico e a QV de pais e filhos (exceto com a QV de crianças), de forma direta, e indiretamente através do apoio social; e por fim, (6) estes últimos efeitos mediadores mostraram-se comuns em díades de pais e filhos com e sem PC.

Conclusões. Os questionários Disabkids-37 constituem-se como meios válidos e fiáveis para a incorporação de uma abordagem desenvolvimental nas rotinas de avaliação pediátricas. As intervenções dirigidas ao apoio social de crianças/adolescentes com PC podem melhorar a sua QVrS, através da promoção do seu ajustamento psicológico. A adoção de uma avaliação multidimensional é preferível para caraterizar a experiência de prestação de cuidados dos pais que têm filhos com PC; nas intervenções clínicas, a redução do desgaste subjetivo pode aumentar a sua QV em diferentes domínios, e a promoção de gratificações na prestação de cuidados pode

atenuar os efeitos deterioradores do desgaste, ou mesmo exercer efeitos diretos positivos na sua QV psicológica e social. Além disso, o desgaste na prestação de cuidados pode ser assumido como um alvo estratégico das intervenções psicossociais no contexto da PC pediátrica, uma vez que a sua redução pode estar associada a níveis superiores de QV e ajustamento psicológico nos pais e nos filhos, através do fortalecimento das suas perceções de apoio social. Por fim, sugere-se que as intervenções clínicas e sociais abordem na sua implementação as especificidades e desafios diretamente relacionados com a PC, ao mesmo tempo que reconheçam e facilitem temas e processos de adaptação e desenvolvimento normativos, no contexto da PC pediátrica.

INTRODUCTION |

The changing epidemiology of pediatric conditions has become particularly noticeable over the last decades: not only a number of chronic health conditions increased their prevalence among children and adolescents, but also the available treatments have improved considerably, thus extending the life expectancy for a number of those conditions (Bruil & Detmar, 2005). This changing epidemiology has sensibly implied a distinct pediatric healthcare perspective. Traditional endpoints, such as the reduction of symptoms and improved survival, became insufficient for assessing medical outcomes, and the consideration of more holistic, patient-oriented markers became warranted (Gerharz, Eiser, & Woodhouse, 2003). Therefore, **developmental contexts and multidimensional outcomes** emerged as crucial targets for pediatric assessment and intervention routines (Christakis, Johnston, & Connell, 2001). In contemporary pediatric settings, the promotion of positive health and adaptation outcomes assumes a remarkable prominence for two main reasons: on the one hand, reciprocal effects of chronic illness or disability and development have been widely acknowledged (Suris, Michaud, & Viner, 2004), and on the other hand, a significant amount of evidence on the associations between early development and later adaptation outcomes has been gathered (Coatsworth, 2010).

The healthcare scenario just described may be applied to the understanding of current clinical challenges in the context of **pediatric cerebral palsy** (CP). CP is a chronic disorder of movement and posture caused by a defect or lesion in the immature brain (Bax, 1964), and it has been reported as the most common physical disability in childhood (Stanley, Blair, & Alberman, 2000). CP frequently has significant effects on the daily life of children/adolescents and their families, and its care and treatment may be quite challenging in terms of time, financial expenses and stress (Vargus-Adams, 1995). Given its substantial variability in clinical manifestations (Liptak & Accardo, 2004), CP has been commented as an interesting prototype of childhood disability (Raina et al., 2004). In fact, most of the current pediatric healthcare issues outlined above are shared by CP cases, for which a life span approach has been recently advocated as an implication of their expanded life expectancy (Roebroeck, Jahnsen, Carona, Kent, & Chamberlain, 2009). Also in the clinical management of pediatric CP cases, there has been a shift from disability, impairment and functional markers, to a broadened assessment encompassing subjective and multidimensional outcome measures (Schneider, Gurucharri, Gutierrez, & Gaebler-Spira, 2001).

Despite this tendency, the **psychosocial adaptation** of children/adolescents with CP remains an underrepresented topic in literature (Vles, Hendriksen, Vles, Kessels, & Hendriksen, 2012). Nevertheless, in clinical and research settings, the adoption of multidimensional outcome

measures, such as **quality of life** (QL) and **health-related quality of life** (HRQL), necessarily implies an interdisciplinary approach to assessment and intervention procedures with pediatric CP cases (Liptak & Accardo, 2004; Vargus-Adams & Martin, 2009). Interestingly enough, **pediatric psychology** has been defined as an interdisciplinary field (cf. Roberts, LaGreca, & Harper, 1988) dealing with the psychosocial adaptation of children, adolescents and their families, in the context of pediatric health, illness and disability. Given the fact that the available evidence-based knowledge on those psychosocial topics is scarce (inasmuch as warranted) for pediatric CP, contributions derived from pediatric psychology research are of utmost importance to improve the understanding and outcomes in the clinical management of those cases.

At this point, it is worth mentioning that the development of current understanding on the psychosocial adaptation of individuals with CP and their families is equally needed and desirable for young adults and adults with CP (cf. Roebroeck et al., 2009), for children/adolescents who have CP and a comorbid learning disability (cf. Beckung & Hagberg, 2002), for "family units" (cf. Magill-Evans, Darrah, Pain, Adkins, & Krakochvil, 2001) and/or for family members other than the parents (cf. Barlow & Ellard, 2006). However, given the inherent (de)limitation of any research piece, it is worth noting that the present work was mainly developed with parents and their children (aged between 8 and 18 years old) with CP and no learning disability.

Throughout the research project, a developmental perspective was elected as the general theoretical framework to guide methodological and conceptual options, as well as to integrate research findings. This **developmental and ecological perspective** was based on the assumptions that characterize the macro-paradigm of **developmental psychopathology** (Achenbach, 1990; Cicchetti, 2006), and which have been highlighted in the specific context of pediatric psychology (Holmbeck, 2002a; Kazak, 1989). In addition to this macro-framework, the **"disability-stress-coping model"** (Wallander, Varni, Babani, Banis and Wilcox, 1989) was adopted to theoretically map parent-child adaptation mechanisms and process in the context of pediatric conditions. Following an extensive literature review on the topics of parent-child psychosocial adaptation to pediatric conditions in general, and CP in particular, this research project was aimed at filling some of the gaps in the existing literature, by systematically seeking the refinement of an answer (or "the answers") to the following core questions: "*How do children/adolescents with CP and their parents adapt to the challenges posed by that chronic physical condition?*"; and more specifically, "*Which are the psychosocial variables and mechanisms that may determine parent-child adaptation outcomes in the context of pediatric CP?*".

In the present dissertation, the main scientific outcomes from this research project are presented in the form of four empirical studies: three of those studies (Study I, Study III and Study IV) have been published in international periodicals and the other one (Study II) has been submitted to an international journal, thus awaiting decision. In addition to these four studies, a preliminary study on the cross-cultural adaptation of pediatric HRQL instruments is presented as an attachment to the dissertation (see attachment: "*Assessing pediatric health-related quality of life within a cross-cultural perspective: Semantic and pilot validation study of the Portuguese versions of DISABKIDS-37*").

This dissertation is organized in four chapters. **Chapter 1|Theoretical Framework** synthesizes the process and outcomes of the literature review that was performed throughout the project. This chapter begins with a series of brief sections on the themes of CP definition, diagnosis, classification, epidemiology and etiology, and then proceeds with the critical description of a theoretical framework on the psychosocial adaptation to chronic physical conditions in childhood and adolescence. Next, a specific section debates a number of terminological issues that became germane during the process of research development. The introductory chapter ends with the summary of the available research literature on parent-child psychosocial adaptation in the context of pediatric CP, followed by the signalization of current questions and challenges for that specific research topic.

In **Chapter 2|Research Aims and Methodology**, an operational description of the empirical component of the research project is provided. That description includes: the delineation of research aims and rationale; the justification of certain methodological options (e.g., research design, sampling procedures and adopted measures/instruments); and the integration of the different empirical studies within an overarching methodological framework. The last sections of this chapter comment the ethical considerations and the statistical options that underlay the research project development.

Chapter 3|Empirical Studies integrates the four empirical studies developed. Study I, entitled "*Examining a developmental approach to health-related quality of life assessment: Psychometric analysis of DISABKIDS generic module in a Portuguese sample*", sought to examine the psychometric adequacy of the Portuguese versions of Disabkids-37 questionnaires (generic module, long form) for HRQL assessment in children, adolescents and mixed age samples. In Study II, "*Social Support and Adaptation Outcomes in Children and Adolescents with Cerebral Palsy*", the psychological maladjustment and social support of children/adolescents with CP was characterized in comparison to their typically developing peers; in addition, the mediating effect of psychopathological dimensions (i.e., internalizing and externalizing problems) on the link between social support perceptions and

HRQL outcomes was assessed in a model accounting for potential age and gender specificities. Study III was called "*The Disability Paradox Revisited: Quality of Life and Family Caregiving in Pediatric Cerebral Palsy*", and aimed at understanding the nature and impact of the caregiving experience in parents of children/adolescents with CP, by describing their QL and their caregiving burden and uplifts in comparison to parents of typically developing children/adolescents; in this empirical study, the moderating role of caregiving uplifts in the associations between different types of burden and QL domains, was also examined. The last empirical study, Study IV, with the title "*Similarities Amid the Difference: Caregiving Burden and Adaptation Outcomes in Dyads of Parents and their Children with and without Cerebral Palsy*", examined the direct and indirect effects (i.e. via social support) of caregiving burden on the adaptation outcomes of children/adolescents with CP and their parents, and ascertained the invariance of such mechanisms in CP and non-CP samples.

Finally, **Chapter 4|Discussion** synthesizes main findings from the research project, and brings them together under the discussion of their theoretical integration. Complementarily, strengths and limitations of the research work performed are critically reviewed. Next, a selection of scientific implications and future research directions is briefly commented, followed by some concluding remarks on the clinical implications derived from the research project findings.

1 THEORETICAL FRAMEWORK

Cerebral palsy (CP): An overview
A historical perspective on the definitions of CP
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1. Cerebral Palsy (CP): An Overview

1.1. A historical perspective on the definitions of CP

Long before it was documented as an independent scientific object in clinical research and practice, CP was notably depicted in artistic terms by the Spanish tenebrist painter Jusepe de Ribera (1591-1652). "The Club-Footed Boy" (Ribera, 1642), an impressive oil on canvas painting that is currently housed in the Musée du Louvre, stands nowadays as a compassionate and picturesque portray of a young men with a hypothetical milder form of CP, and somehow embedded in a context of poverty and social exclusion, which might have been common for such cases in those days.

It was only in the mid-19th century that the English surgeon William Little first described "cerebral paralysis" as a result of brain damage related to preterm birth and perinatal asphyxia (Little, 1843). Following his seminal work, CP was known as "Little's disease" many years after. The plural form of the term "CP" was first used by the Canadian William Osler in his evidence-based monograph "The Cerebral Palsies of Children" (Osler, 1889).Therefore, Sigmund Freud was the first author who employed the term "CP" as a unifying nosographic category for a variety of infantile motor deficits of brain origin (Freud, 1968, as cited in Kavcic & Vodusek, 2005, p. 582). In the meantime, Freud directed his research interests to psychoanalysis, and CP ended up scarcely studied during the first half of the 20th century, also due to the fact that more common causes of disability, such as poliomyelitis and tuberculosis, were requiring greater attention (Morris, 2007). Nevertheless, given the distinctive pertinence of their contributions, both Little's etiological approach and Freud's conceptual approach still represent great milestones in the study of pediatric CP (Kavcic & Vodusek, 2005).

The issue of defining CP regained a critical focus in Minear's work on a classification system for CP (Minear, 1956). In his paper, Minear wrote eloquently about a number of definitions, previously suggested by different authors (including Winthrop Phelps, Myer Perlstein, John Pohl, C. Balf and T. Ingram), and succinctly defined CP as any symptom complex "caused by a non-progressive brain lesion (or lesions)" (Minear, 1956, p. 842). During the next year, a group of scholars from the "Oxford Study Group on Child Neurology and Cerebral Palsy", called "The Little Club", discussed a memorandum on the terminology and classification of CP. In the published form of that memorandum, CP was defined as a permanent, thought not unchanging disorder of movement and posture, emerging in the early years of life and caused by a non-progressive disorder of the brain, which resulted from interference during its development (MacKeith, MacKenzie, & Polani, 1959). This definition was then further refined, stating CP as

"a disorder of movement and posture due to a defect or lesion of the immature brain" (Bax, 1964, p. 295), and became a classic and widely cited definition for CP (Bax, Goldstein, Rosenbaum, Leviton, & Paneth, 2005).

As a result of a series of scientific meetings held in Europe and America during the late 1980s, a subsequent definition further highlighted the heterogeneity of the condition, stating CP as an "umbrella term covering a group of non-progressive, but often changing, motor impairment syndromes secondary to lesions or anomalies of the brain arising in the early stages of development" (Mutch, Alberman, Hagberg, Kodama, & Perat, 1992, p. 549). In the beginning of the 21st century, the group for the Surveillance of Cerebral Palsy in Europe (SCPE) had the merits of harmonizing the common aspects brought by the definitions of Bax (1964) and Mutch and colleagues (1992), with the statement of five key elements that should be included in any definition of CP. These **five key elements** were: CP as an umbrella term; its permanent but not unchanging nature; its relation with a disorder of movement and/or posture and of motor function; its origin in a non-progressive interference/lesion/abnormality, and finally, the occurrence of this interference/lesion/abnormality in the developing/immature brain (SCPE, 2000).

The increased understanding about antecedents and correlates of CP, along with significant changes in the care provided for individuals with disabilities, called for a reassessment of the definition of CP (Bax et al., 2005). While underlining "CP" as a clinical descriptive term, and not an etiologic diagnosis, CP was described as "a group of 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, cognition, communication, perception, and/or behavior, and/or by a seizure disorder" (Rosenbaum et al., 2005, p. 572). When planning a study on CP, its definition has to be described precisely, so that the study can be firmly based (Kavcic & Vodusek, 2005). Although more complex, this latest comprehensive definition was preferred and adopted within the present work, for two main reasons: first, it emphasized the developmental nature of CP and its potential impact on the individual's developmental trajectories; and second, it highlighted the occurrence of common comorbidities, including the behavioral disturbances (e.g. attention-deficit hyperactivity disorder; mood and anxiety disorders) (Rosenbaum et al., 2005).

The definition of the term "CP" has been debated for more than 150 years (Morris, 2007), yet it is still easier to state what CP is not than to define it precisely (Kavcic & Vodusek,

2005). Although definitions are of utter importance for ensuring accuracy and consistency in theory and research development, their pragmatic sense should not be overlooked; or as Martin Bax sharply put it: "for parents, policy makers, and the public, the label of CP defines groupings of children who are desperately in need of a service, and this seems an adequate ground (for the moment), for continuing with the unsatisfactory academically grouping of these children under the umbrella term «cerebral palsy»" (Bax, 2001, p. 75).

1.2. Diagnosis and Classification of CP

As for its definition, discussions on the best classification of CP persist to nowadays (Morris, 2007). This is in part due to the fact that variability is one of the hallmarks of CP (Liptak & Accardo, 2004), but also to the occurrence of nonlinear and variable changes in the development of children/adolescents with different CP subtypes (Vargus-Adams & Martin, 2009). In general, the distinction of CP subtypes is based on the topographic distribution of limb involvement and the quality of the movement disorder. However, during child's development, a given neurologic picture may change dramatically, and hence elicit inconsistencies between examiners or by the same examiner at different times (Murphy & Such-Neibar, 2003). The term "CP" is mostly descriptive rather than informative about the condition's etiologic factors, pathology, or prognosis (Blair & Stanley, 1997), and therefore, its diagnosis may be best approached as a dynamic process.

Although the diagnosis of CP during the first years of life is often unreliable, early risk signs may include delay in achieving motor developmental milestones, toe walking, persistent fisting, impaired head circumference growth, seizures, irritability, poor suck, handedness before 2 years old, and scissoring of the lower limbs (McMurray, Jones, & Khan, 2002). During the diagnosis process, a comprehensive neurologic, developmental and functional assessment must be developed in order to obtain the most complete history of risk factors and genetic background; for high-risk preterm infants, the best available predictor of CP is the presence of echodensities and cysts in the periventricular white matter regions of the brain, as observed from neuroimaging studies. The conduction of the diagnosis process in the earliest times of life must continually look for evidence of disease progression, and if present, exclude the diagnosis of CP (Murphy & Such-Neibar, 2003). For a more detailed description of the diagnosis process of CP, a clinical decision tree has been delineated in the scope of epidemiological research (see SCPE, 2000, p. 820). For the purpose of illustrating a provisional agreement on **diagnostic criteria for**

CP, Kavcic and Vodusek (2005) listed mandatory inclusion/exclusion criteria for the diagnosis of CP, within a continuum of clinical judgment (presented in Table 1). In the authors' own words, such agreement is "incomplete as is incomplete our knowledge of CP" (p. 586), but within the present work, it hopefully serves the intent of synthesizing the foremost diagnostic criteria of CP.

Table 1. A synthesis of diagnostic criteria for CP (reprinted with permission from: Kavcic and Vodusek, 2005, p. 586)

Possible CP *Mandatory inclusion criteria*: disorder of movement and posture manifesting as spastic diplegia, spastic hemiplegia, spastic tetraplegia, ataxia, dystonia, choreo-athetosis - alone or in any combination; onset early in life; no evidence of progression. *Mandatory exclusion criteria*: active disease that could explain the foregoing features; chromosomal disorders. *Supportive features*: other signs of brain dysfunction that could be caused by the same pathological process as the foregoing disorders of movement and posture (epilepsy, learning disorders, disorders of speech, vision or hearing); born after multiple pregnancy; vanishing twin syndrome; intrauterine growth retardation; major antenatal placental abruption; preterm birth; acute intrapartum hypoxia; reduced fetal heart rate variability from the onset of labour; extensive chorioamnionitis; congenital coagulation disorders; autoimmune disease of the mother; no child with the same/similar clinical picture in a family.
Probable CP *Mandatory inclusion criteria*: disorder of movement and posture as in possible CP; onset early in life; no evidence of progression or other disease that could explain the foregoing features at school age.
Definite CP *Mandatory inclusion criteria*: disorder of movement and posture as in probable CP, plus still no evidence of progression unrelated to aging or other disease that could explain the foregoing features at age 18 or older.

Just like its definition, the **classification of CP** has undergone constant revisions and debate for over one century. After the pioneer proposal of CP classification according to the distribution of the "paralysis" (Osler, 1889), and the aforementioned research hiatus in the study of CP, Wyllie (1951) combined a variety of etiological and neurological criteria to establish four categories: congenital symmetrical diplegia, congenital paraplegia, quadriplegia or bilateral hemiplegia, and hemiplegia. However, a much more complex framework for the classification of CP was developed few years after: Minear (1956) organized a vast number of categories to distinguish CP subtypes along physiological/motor, topographical, etiological, supplemental, neuroanatomical, functional and therapeutic dimensions. Despite its significant improvement of classification guidelines for CP, and its likely clinical relevance, Minear's classification was too

overarching to adequately serve more practical purposes in research. Notably, it was only three decades after that a classification for CP, anchored to an epidemiological perspective, did emerge. The classification system known as "Evans form" recorded motor dysfunction in terms of hypotonia, hypertonia (including stiffness, spasticity and rigidity), dyskinesia and ataxia, and enabled the documentation of functional mobility and manual dexterity in ordinal levels (Evans, Alberman, Johnson, & Mutch, 1987). Notwithstanding its apparent adequacy and utility, data on this classification system's validity and reliability was not documented (Morris, 2007).

In the same line of the harmonizing approach applied to the definition of CP, the SCPE research group further extended it to its classification, with the development of a hierarchical classification tree for CP subtypes (cf. SCPE, 2000, p. 821). Given its integrative concern and recent development within a **European epidemiological perspective**, this classification was adopted throughout the present research work. In Table 2, the definitions regarding this European classification of CP are summarized.

Table 2. European classification of CP (reprinted with permission from: SCPE, 2000, p. 821)

Spastic CP is characterized by at least two of:
- Abnormal pattern of posture and/or movement
- Increased tone (not necessarily constant)
- Pathological reflexes (increased reflexes: hyperreflexia and/or pyramidal signs, e.g. Babinski response)

Spastic CP may be either bilateral or unilateral
- Spastic bilateral CP is diagnosed if:
  - Limbs on both sides of the body are involved
- Spastic unilateral CP is diagnosed if:
  - Limbs on one side of the body are involved

Ataxic CP is characterized by both:
- Abnormal pattern of posture and/or movement
- Loss of orderly muscular coordination so that movements are performed with abnormal force, rhythm, and accuracy

Dyskinetic CP is dominated by both:
- Abnormal pattern of posture and/or movement
- Involuntary, uncontrolled, recurring, occasionally stereotyped movements

Dyskinetic CP may be either dystonic or choreo-athetotic
- Dystonic CP is dominated by both:
  - Hypokinesia (reduced activity, i.e. stiff movement)
  - Hypertonia (tone usually increased)
- Choreo-athetotic CP is dominated by both:
  - Hyperkinesia (increased activity, i.e. stormy movement)
  - Hypotonia (tone usually decreased)

1.3. Etiology and Epidemiology of CP

Although most parents of a child with CP are anxiously willing to know the origin of their child's disorder, the underlying cause remains uncertain in more than 50% of cases, thus supporting the hypothesis that CP may be most likely multi-determined (Murphy & Such-Neibar, 2003). In a recent review, Reddihough and Collins (2003) clarified the distinction between **"known causes"** and **"risk factors or associations"** in the study of CP etiology, and commented on the available evidence for each of those factors included in both etiological categories. In Table 3, an updated and evidence-based list of "known causes" and "risk factors" for CP is presented, in agreement with those authors' review.

Table 3. Causes and risk factors in the etiology of CP (according to: Reddihough & Collins, 2003)

Known causes of CP

Antenatal causes

- Congenital brain malformations (including malformations of cortical development)
- Vascular accidents (e.g. middle cerebral artery occlusion)
- Maternal infections during the 1st and 2nd trimesters of pregnancy (e.g. rubella, cytomegalovirus, toxoplasmosis)
- Metabolic disorders, maternal ingestion of toxins and rare genetic syndromes (less common)

Perinatal causes

- Problems during labour and delivery (e.g. obstructed labor, antepartum hemorrhage or cord prolapsed leading to hypoxia)
- Neonatal problems (e.g. severe hypoglycemia, untreated jaundice and severe neonatal)

Post-neonatal causes

- Meningitis neurological sequelae
- Accidental injuries (e.g. motor vehicle accidents and near-drowning episodes)
- Cerebrovascular accidents and following surgery for congenital malformations
- Septicemia and other conditions such as malaria (mostly in developing countries)

Risk factors

Risk factors before pregnancy

- Maternal factors (i.e. delayed onset of menstruation, irregular menstruation or long intermenstrual intervals; an unusually short or long interval between pregnancies; low social class; parity of three or more; previous fetal deaths; maternal medical conditions, such as intellectual disability, seizures and thyroid disease);
- Paternal and sibling factors (i.e. advanced paternal age, motor deficit in a sibling)

Risk factors during pregnancy

- Pre-eclampsia (in situations of term infants)
- Administration of thyroid hormone or estrogen in pregnancy
- Antepartum hemorrhage (in situations of preterm infants)
- Specific genetic mutations, predisposing to venous thrombosis (e.g. factor V Leiden mutation, gene for prothrombin)
- Multiple pregnancy
- Death of one twin (in monochorionic twin pregnancies)

Risk factors during labor

- Major events leading to perinatal asphyxia (including prolapsed cord, massive intrapartum hemorrhage, prolonged or traumatic delivery due to cephalopelvic disproportion or abnormal presentation, a large baby with shoulder dystocia and maternal shock from a variety of causes)
- Prolonged second stage of labour
- Emergency cesarean section
- Premature separation of the placenta
- Abnormal fetal position
- Intrauterine exposure to infection (particularly chorioamnionitis)
- Prolonged rupture of the membranes
- Presence of meconium stained fluid and tight nuchal cord (in preterm babies)

Risk factors at birth

- Decreased birth weight
- Length of the gestation
- Poor intrauterine growth (particularly in the moderately preterms)
- Low placental weight and low Apgar scores

Risk factors in the newborn period

- Neonatal seizures
- Sepsis
- Respiratory disease

As regards the **epidemiology of CP**, it has been repeatedly stated that CP is the most common physical disability in childhood (Bax, 1964; Moreno-De-Luca, Ledbetter, & Martin, 2012; Stanley et al., 2000). Similar claims have been affirmed for the number of CP cases in developed countries (Viehweger et al., 2008), and particularly in Europe (Johnson, 2002). In general, there is an estimated prevalence for CP of 2.0 to 2.5/1000 children, with little tendency to change over recent decades (Majnemer & Mazer, 2004). However, an increased prevalence of children with CP was observed during previous decades, eventually resulting from significant medical advances that occurred during the 1960s, the 1970s and the 1980s, which enabled the survival of very low birth weight infants and/or children with technology dependency (Azaula et al., 2000; McDermott et al., 1996). In fact, the prevalence of CP seriously magnifies in situations of extreme prematurity (1 in 20) and low birth weight (the occurrence rate is less than 1 in 1000 in infants with a birth weight of >2500g) (Murphy & Such-Neibar, 2003). In the USA, approximately 500.000 individuals have CP, turning this condition to be the most common of all congenital disorders (Wiley & Renk, 2007). In Portugal, a recent surveillance study revealed that the prevalence of CP at 5 years old was of 1.78/1000 children (95% CI 1.56 ‰ - 2.06‰) (Andrada, Folha, Calado, Gouveia, & Virella, 2009)

In clinical terms, it is important to note that spastic forms are the most common subtypes of CP, accounting for 70% to 85% of all cases (Chen et al., 2010). Moreover, between 1/3 and half of the total cases may present some form of intellectual disability (Kirby et al., 2011), and nearly 20% have severe intellectual deficits and are unable to walk (Johnson, 2002). Complementarily, from a healthcare perspective, the increase in the life expectancy of individuals with CP became salient: in contrast to the mid-20th century, when few people with CP survived to adulthood, nowadays, between 60% and 90% of children do survive till adult age (Zaffuto-Sforza, 2005). For this particular reason, it has been recently argued that a **life span approach** should be incorporated in the provision of healthcare for individuals with CP (Roebroeck et al., 2009).

2. A Theoretical Framework on the Adaptation to Chronic Physical Conditions in Childhood and Adolescence

2.1. Pediatric Psychology: A Developmental Perspective on Chronic Physical Conditions

Individual and family adaptation to chronic physical conditions during childhood and adolescence is a foremost object of study in pediatric psychology. Kagan (1965) first described pediatric psychology as the accomplishment of a "new marriage" between pediatrics and psychology. A few years later, Wright (1967) listed a professional profile for the "pediatric psychologist", which notably emphasized the positive feature of the profession, in comparison to the established work developed by clinical child psychologists. In so doing, Wright commented that pediatric psychologists were to deal more with child-rearing questions, positive mental health and personality development, and in contrast, to deal less with severe psychopathology.

Those multiple and distinctive facets of pediatric psychology were then conciliated in a definition that would become widely reported; in that definition, pediatric psychology is approached as "an interdisciplinary field addressing the full range of physical and mental development, health, and illness issues affecting children, adolescents, and families" and encompasses a variety of topics such as "understanding, assessment and intervention with developmental disorders; evaluation and treatment of behavioral and emotional problems and concomitants of disease and illness; the role of psychology in pediatric medicine; the promotion of health and development; and the prevention of illness and injury among children and youth (Roberts et al., 1988, p. 2). From an epistemological perspective, pediatric psychology is best understood as incorporating the interface between pediatrics and health psychology (in a more general theoretical-scientific level), and systematically developing and applying knowledge from the fields of clinical and developmental psychology (in a more specific theoretical-scientific level) (Menezes, Moré, & Barros, 2008).

As an independent interdisciplinary field, pediatric psychology is embedded in a "macro theoretical framework" where, amidst a variety of psychological, social and health disciplines, developmental psychopathology stands with particular prominence (Menezes et al., 2008). Developmental psychopathology can be viewed as a "macroparadigm" for the understanding of development and adaptation, thus integrating theoretical and empirical contributions derived from other paradigms, such as biomedical, behavioral, sociological, cognitive and family systems, which in turn develop, examine and apply a number of more specific theories (Achenbach, 1990). Interestingly enough, the fact that Spirito and colleagues (2003) highlighted "life span developmental psychopathology" as a major domain of training for pediatric psychologists,

practically illustrates the importance of liaising the practice of pediatric psychology to an overarching knowledge on development and adaptation.

For the purpose of the present work, however, a deeper analysis of some of the links between this **developmental perspective and pediatric psychology** was considered worthwhile. First, the importance of "context" (Boyce et al., 1998) and the ecological-transactional notion of development as "the ongoing interplay between an active, changing organism in a dynamic, changing context" (Cummings, Davies, & Campbell, 2000, p. 24), represent core tenets in developmental psychopathology, which have been thoroughly endorsed in pediatric psychology (Christakis et al., 2001; Fiese & Sameroff, 1989). Second, the assumption that "risk factors do not function in a static manner" (Cicchetti, 2006, p. 10), and the understanding of resilience as "a dynamic process encompassing positive adaptation within the context of significant adversity" (Luthar, Cicchetti, & Becker, 2000, p. 543) in this developmental perspective, are both in line with the dispute of a direct relationship between chronic disability and psychosocial functioning (Harper, 1991), and the pertinence of studying resource and risk factors, along with resilience outcomes, in pediatric psychology (Rose, Holmbeck, Coakley, & Franks, 2004). Third and last, given the fact that children and adolescents with chronic health conditions and their families represent a group with increased risk for psychosocial maladjustment (CCD & CPACFH, 1993; Eiser, 1997), their study is likely to improve our understanding of normative developmental processes and elucidate decisive components of adaptation that may not be typically evident (Cicchetti, 2006). Following these three considerations, and to put it succinctly, the field of developmental psychopathology has provided a conceptual and terminological framework (e.g., developmental trajectories, resilience, risk and protective processes, continuity/discontinuity of adaptive and maladaptive processes, multifinality, equifinality) that assists clinical and research efforts to explain the phenomena of interest in pediatric psychology (Holmbeck, 2002a).

2.2. Stress and Coping: The Disability-Stress-Coping Model

In the recent past, it has been commented that research on childhood and adolescent chronic physical conditions lacked firmly defined theoretical and conceptual frameworks, which had resulted in a proliferation of fragmented findings (Drotar, 1981). If indeed "the way we see the problem (childhood disability/illness) is the problem" (Harper, 1991, p. 534), the absence of a clear common rationale to guide research and practice was no minor issue. Within that context, the methodical work developed by Daniels, Moos, Billings and Miller III (1987) and Wallander and colleagues (1989a) provided a solid ground for organizing research and guiding practice. In

both cases, authors were interested in formulating and examining individual and family adaptation variability from a **"risk and resistance" perspective**, which was after all rooted in the central tenet of stress and coping theory that perceptions of stressors and resources interact to determine a given adaptation outcome (Lazarus & Folkman, 1984).

While Daniels and her colleagues (1987) were interested in generally studying the influence of a number of psychosocial risk (e.g. parent dysfunction, increased family stressors) and resistance factors (e.g. increased family resources) on children's adjustment, Wallander and his collaborators (1989a) went further and delineated a model aimed at mapping the complex relationships between risk and resistance factors and the differential psychosocial adjustment of chronically ill and handicapped children. Both groups of authors attempted to explain the mechanisms underlying the psychosocial adaptation process of children and adolescents with chronic physical conditions and their families, by refining previous general models of stress and coping (e.g. Lazarus & Folkman, 1984) with the consideration of specific illness/disability-related variables and parameters. There were, in fact, a number of models on childhood adjustment to chronic illness that were proposed several years before Wallander and his colleagues' disability-stress-coping formulation (e.g., Lipowski, 1970; Pless & Pinkerton, 1975; Moos & Schaefer, 1984), and which included overall formulations of stress and coping processes. Nevertheless, Wallander and colleagues' model had the merits of critically integrating the contributions derived from those previous conceptualizations.

The **disability-stress-coping model** (Wallander et al., 1989a; Wallander & Varni, 1992) was based on three core assumptions: first, the presence of a chronic physical condition did not necessarily represent individual or family maladjustment; second, "stressors" were defined in general terms as "problematic situations requiring a solution or some decision-making process for appropriate action" (Varni & Wallander, 1988, p. 215); and third, the notion of "competence", a core construct in developmental psychopathology (Masten, Burt, & Coatsworth, 2006), was emphasized as "the effectiveness of the coping responses emitted when an individual is confronted with problematic situations" (Varni & Wallander, 1988, p. 215). In the same line of Daniels and colleagues (1987), who previously suggested that children with additional stressors and demands to those directly related to their condition, were at increased risk for psychosocial dysfunction, Wallander and his collaborators (Wallander et al., 1989a) understood adaptation outcome as a function of the stress experienced, the nature of the problems encountered and the individual's ability to successfully cope with them. Moreover, the model further asserted that adaptation outcomes were influenced by risk and resistance factors. As graphically depicted in

Figure 1, three levels of variables were interactively outlined, namely: risk factors, resistance factors and adaptation outcomes.

At the time the disability-stress-coping model was developed, the distinction between risk and protective factors in pediatric psychology was not as clear as it turned out to be (e.g., Rose et al., 2004). For that reason, risk and resistance factors (according to Bradford, 1997, these latest were assumed as "resilience factors") were merely defined as variables that increased the likelihood of poor or positive adaptation, respectively. **Risk factors** were then grouped into three subcategories: factors related to the individual's illness and disability (e.g. severity, visibility); the individual's level of independence, and the psychosocial stressors implied by (e.g., disability and caregiving burdens) or co-existing with the condition (e.g., life events and daily micro-stressors). Within the disability-stress-coping model, the impact of those risk factors was theorized to be moderated by three groups of **resistance factors**, namely: intrapersonal factors (e.g., the individual's problem-solving or social skills); social-ecological factors (e.g., social support, family members' adaptation), and coping resources (including cognitive appraisals and coping strategies). Following this integration of risk and resistance factors into a unified theoretical framework, a central tenet of the model was that those factors could influence adaptation outcomes in both direct and indirect ways (Wallander et al., 1989a). For instance, greater handicap severity could impair the individual or his/her family caregiver's adaptation outcomes directly, or through the increase of handicap-related problems or caregiving burden. Complementarily, the impact of disability and caregiving burdens on the individual or his/her family caregiver's outcomes could be "buffered" by the amount and/or quality of their coping resources.

Despite the fact that different models have been formulated for the understanding of psychosocial adaptation processes to chronic health conditions and disabilities, such as the integrated task-based model (Samson, Siam, & Lavigne, 2007; Samson & Siam, 2008) or the perspectives from chaos and complexity theory on psychosocial adaptation to disability (Livneh & Parker, 2005), processes of stress and coping were invariantly approached in any case. The disability-stress-coping model (Wallander et al., 1989a; Wallander & Varni, 1992), however, achieved a distinct prominence in pediatric psychology, once being considered "the most sophisticated and coherent, theoretical framework to emerge so far" (Bradford, 1997, p. 146) and having inspired, till recently, the development of specific models to certain chronic physical conditions, such as CP (e.g., Raina et al., 2004). On the positive critique of the model, one must acknowledge the utility of its "general tailoring", which enables its application to any pediatric disorder within a non-categorical approach (Wallander et al., 1989a), and its simultaneous

adequacy to map individual and other family members' adaptation mechanisms to chronic physical conditions (Wallander et al., 1989a; Wallander, Pitt, & Mellins, 1990). This considerable advantage may, on the other hand, imply the difficulty of operationalizing such an overarching model into research designs. Nevertheless, it is worth noting that the authors themselves have argued that their model could not be validated as a whole, but rather more plausibly, it would have to be examined through the conduction of analyses of small clusters of variables, in order to assess if they operated in the conjectured directions (Wallander et al., 1990).

Having acknowledged the existence of varied theoretical models on individual and family adaptation to chronic conditions in pediatric psychology, it remains particularly important to justify the election of the disability-stress-coping model as the main theoretical framework to guide our studies and their discussion within the present work. The disability-stress-coping model was thought to suitably portray a "risk-resilience" framework (Raina et al., 2004) for the examination of adaptation outcomes and mechanisms in pediatric populations. Complementarily, in terms of data analyses organization, the model offered the possibility of conjecturing on both direct and indirect effects of "risk" and "resistance" factors (i.e. predictors) on adaptation outcomes (i.e. criterion variables), which have been commented to be of uttermost relevance in pediatric psychology research (Holmbeck, 2002b; Rose et al., 2004). The disability-stress-coping model also had the flexibility of being applicable to the examination of both individual and family adaptation mechanisms; more specifically, its authors asserted that a chronic physical disorder could be characterized as implying a chronic level of strain inherent to the care of a child with a chronic condition or disability (Wallander et al., 1989a). Given the fact that caregiving burden was a core variable in some of the studies conducted within the current research work, such assertion further suggested the adequacy of the model for the study of parents as primary family caregivers. In addition, despite the model's general feature, its contemplation of some variables of unique importance for the understanding of psychosocial adaptation processes in the context of CP, such as the individual's level of functional independence or cognitive functioning, was most useful to plan sample collection (e.g., definition of inclusion/exclusion criteria) and specific statistical analysis procedures (e.g., selection and inclusion of covariates in regression analyses). Interestingly enough, the disability-stress-coping model had the notable merit of clearly stating "family environment" and "family members' adaptation" as examples of social-ecological determinants of adaptation outcomes, and hence following the premises on the importance of family context on child/adolescent's adaptation and development (Bronfenbrenner, 1986), and somehow pioneering their extension to pediatric psychology settings (cf. Kazak, 1986), which were to gain a greater focus in the years to come (e.g., Drotar, 1997; Kazak, 1997). The last

comment for justifying the election of the disability-stress-coping model as the more specific theoretical framework within the present research work, relates to its consideration of a **multidimensional approach to adaptation outcomes** assessment (including the dimensions of mental health, social functioning and physical health). This multidimensional definition of outcomes underlay the selection of outcome variables in the studies here developed, along two main assumptions: first, the pertinence of assessing positive dimensions of adaptation to chronic health conditions, in addition to the more classic measures of negative outcomes (e.g., psychological maladjustment) (Barlow & Ellard, 2006; Ridder, Geenen, Kuijer, & Middendorp, 2008); second, the alignment of such multidimensionality with the concept and construct of QL adopted (Ravens-Sieberer et al., 2007; The WHOQOL Group, 1994), which was after all assumed in the present work as the "ultimate outcome" (Livneh & Antonak, 2005).

Despite these strengths of the disability-stress-coping model for guiding research within the current series of studies, some model limitations should be also acknowledged within that same context. If on the one hand, the model pertinently includes "family environment" and "family member' adaptation" as social-ecological determinants of adaptation outcomes, on the other hand, it lacks the clear representation of a possible interrelation between the adjustment of a child/adolescent with a chronic physical condition and his/her parent/caregiver's. Although none of the studies here presented were transactional in nature (cf. Fiese & Sameroff, 1989; Sameroff, 2009), it should be commented that the transactional stress and coping model for chronic childhood illness (Thompson, Gustafson, Hamlett, & Spock, 1992) complementarily portrays dyadic adaptation processes in greater detail. In addition, despite the "risk-resilience" framework of the disability-stress-coping model, the possible co-occurrence of positive and negative psychological states and emotions during the stress-coping processes, elicited by disability or caregiving-related stressors, is not clearly described. For this reason, in one of the studies here presented (specifically dealing with positive and negative dimensions of family caregiving), the contributions from a revised model of the coping process (Folkman, 1997), which accounts for the role of positive emotions and meaning-based coping on adaptation processes, were regarded as complementary to the disability-stress-coping model. An important final remark relates to the fact that the disability-stress-coping model did not specifically include any set of socio-demographic variables, such as age and gender, in its original depiction (Wallander et al., 1989a). However, different authors have highlighted the pertinence of assessing these variables when studying adaptation processes and outcomes of pediatric populations (Eiser, Havermans, Pancer, & Eiser, 1992; Gerharz et al., 2003; Holmbeck, 2002a). For this reason, and given the interest of the present research work in mapping age and gender differences (or

similarities) in adaptation outcomes and mechanisms, these variables were, as long as plausible, systematically included in the series of analyses conducted for the studies here presented.

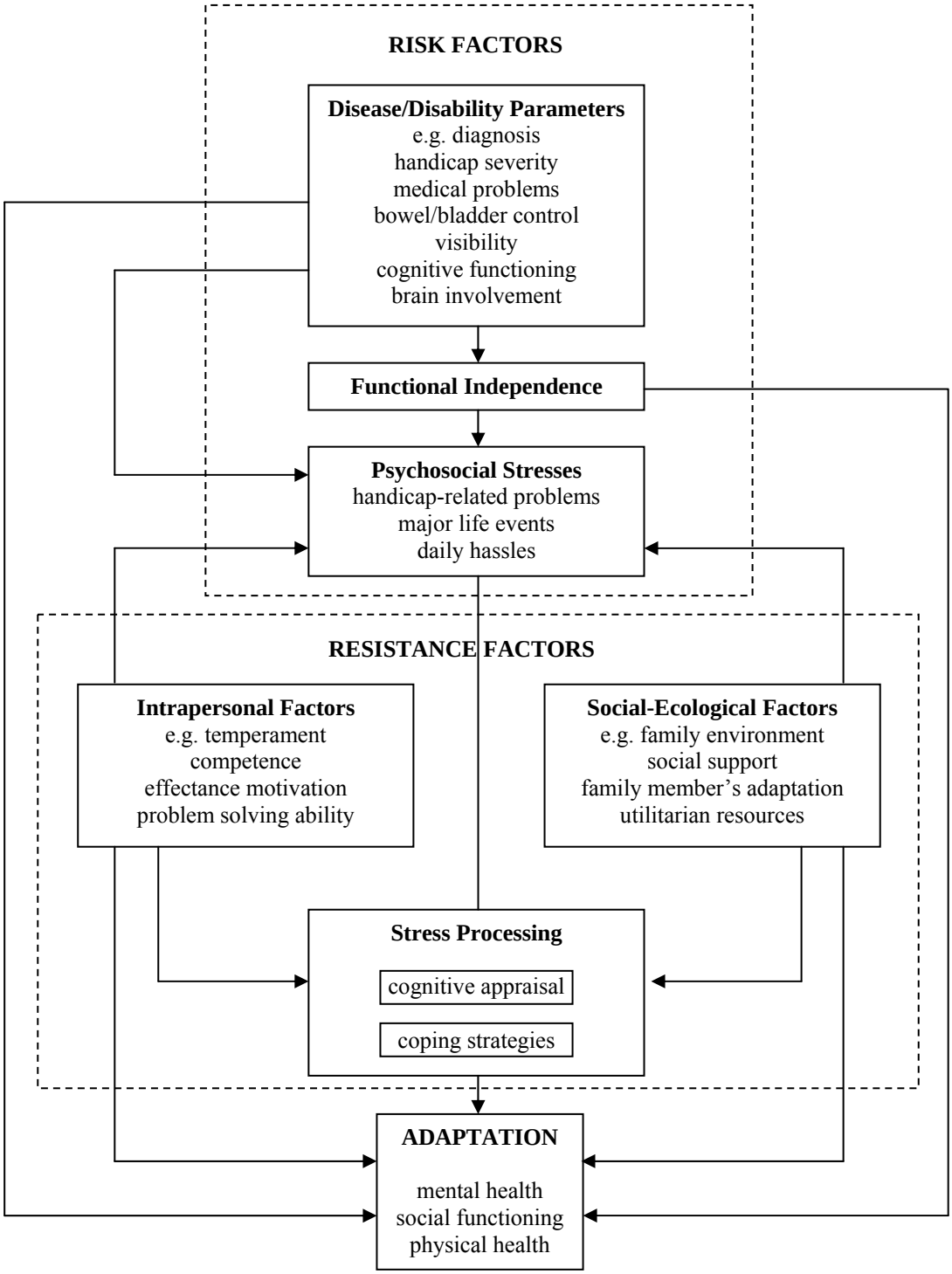

Figure 1. The Disability-Stress-Coping Model (reprinted with permission from: Wallander et al., 1989a, p. 171)

2.3. Stress, Social Support and Adaptation Outcomes

The consideration of social contexts is crucial for the understanding of human development and adaptation processes (Bronfenbrenner, 1977). A social context is operationally defined as "a set of interpersonal conditions, relevant to a particular behavior or disorder and external to, but shaped and interpreted by, the individual child" (Boyce et al., 1998, p. 143). To a considerable extent, the importance of social context in human development and individual well-being and distress has been illustrated by research on the rubric of social support (Vaux & Harrison, 1985). In fact, social support has been commented as a reflection of the individual's social environment (Schwarzer & Leppin, 1991) and as a mean of operationalizing a social-ecological model of chronically ill children and their families (Kazak, 1989). More specifically, the role of social support in understanding individual and family successful adaptation to childhood disability has been underlined (Kazak, 1987).

Social support may be defined as consisting of "social relationships that provide (or can potentially provide) material and interpersonal resources that are of value to the recipient, such as counseling, access to information and services, sharing of tasks and responsibilities, and skill acquisition" (Thompson, 1995, p. 43). Drawn from this definition, two specific features are noteworthy: first, social support operates in the context of relationships, which are psychologically complex; and second, social support is multifaceted, encompassing a variety of sources and types of support (Thompson, Flood, & Goodvin, 2006).

There are essentially three approaches to social support operationalization: as social support networks, as supportive behaviors, and as subjective appraisals of support, such as perceptions and satisfaction (Vaux & Harrison, 1985). In research aimed at predicting well-being and related outcomes, it has been recommended that the measurement of subjective aspects of social support (e.g., perceived support, satisfaction with support) should be preferred (*idem*, *ibidem*), since "health and well-being are dependent on what the person sees and believes, be it accurate or not" (Schwarzer & Leppin, 1991, p. 102).

The pertinence of studying social support in developmental psychopathology is substantiated by a number of practical applications: social support can be used to monitor the well-being of at-risk children, to improve parental conduct, and to remediate developmental outcomes. As regards this latest application, it should be emphasized that some social support key elements, such as counseling and guidance, emotional nurturance, information and skill acquisition, are indeed common components of successful psychotherapeutic interventions. Moreover, interventions targeting social support (and especially those dimensions directly linked to a child's problems) may result in the remediation of clinical symptomatology (Thompson et al.,

2006). Within the context of parent-child relationships, it has been argued that social support influences parents' well-being, improves parenting and enhances children's psychological well-being (Cochran, 1990); complementarily, the important role of parents in arranging, monitoring and facilitating their children's social experiences has been underscored (Ladd & LeSieur, 1995). Despite the fact that adolescents rely less exclusively on their parents for their emotional well-being than children, it should be noted that parents remain important, though differentiated, sources of adolescent's social support (Thompson et al., 2006).

The relevance of social support in the context of pediatric psychology is straightforwardly illustrated with the clear statement of this variable as a "resistance social-ecological factor" within the disability-stress-coping model (Wallander et al., 1989a). In addition, different authors have stressed the importance of examining social support in the adaptation processes of both youth with pediatric conditions and their families (Eiser, 1990; LaGreca, Bearman, & Moore, 2002). Two pioneer studies, based on the disability-stress-coping model, pertinently demonstrated those theoretically-established premises. In the first study, different dimensions of social support were found to contribute negative and independently to the variance of internalizing and externalizing problems in children with chronic illness and disabilities (Wallander & Varni, 1989). In the second study, significant proportions of variance in the mental and social functioning of mothers of children with chronic physical conditions were explained by specific features of their social environment (Wallander et al., 1989b).

On the topic of pediatric CP, in particular, it has been commented that social variables, such as school environment, family dynamics and peer relationships, can be potent determinants of children/adolescents' adaptation outcomes (Liptak & Accardo, 2004; Majnemer & Mazer, 2004), and that the lack of involvement in social relationships may result in poor development of social skills and social isolation (Majnemer & Mazer, 2004). Though not specifically assessing the variable of "social support", but instead other related constructs, these studies' results are indicative of the relevance of examining the role of social support in the adaptation processes of this group.

There has been a relative consensus in describing chronic health conditions or disabilities as individual and family stressful events (CCD & CPACFH, 1993; Eiser, 1997; Tijhuis, Flap, Foets, & Groenewegen, 1995). For parents who have a child with a chronic condition, the fact that caregiving demands tend to qualitative and/or quantitatively exceed the caregiving responsibilities of normative parenting, may represent a specific source of stress (Raina et al., 2004; Turner-Henson, Holaday, & Swan, 1992). This observation led some authors to suggest that caregiving burden, and not as much disability-related variables, should be studied as main

sources of stress in these parents (Horton & Wallander, 2001). Interestingly enough, parental child care has been alternatively conceptualized as a primary context of social support provision (Thompson et al., 2006). As regards the experience of increased stress in families of children with chronic physical conditions, Kazak (1987) has advanced two important questions to be answered in subsequent research: first, "who" within the family experiences the effects of stress, and second, "how" the stress presents itself. Kazak also commented on the social difficulties experienced by some of these families, thus delineating an arena for future research, namely on the links between family (members') stress and social support.

In point of fact, examining the relationships between stress, social support and adaptation outcomes turns out a complex task (Thompson et al., 2006). Since there is evidence for social support as a causal contributor of health and well-being outcomes (Cohen & Wills, 1985; Schwarzer & Leppin, 1991), two main hypotheses regarding the pathways of social support influence have been established in literature. The first one is termed the **"main effect model"**, and posits that social support is likely to elicit beneficial effects on adaptation outcomes, irrespectively of the fact that the person is under increased stress or not. The alternative hypothesis, the so-called **"buffering model"**, suggests that social support protects persons from the deleterious effects of stressful events. In the first model, social support is assumed to exert its influence via the provision of positive affect, predictability, recognition of one's self-worth, and through the reduction of the likelihood of experiencing negative events. For this reason, the main effect model has been also labeled as "stress-preventive" (Thompson et al., 2006). In the second model, social support is conjectured to either intervene between the stressful event and the individual's reaction by attenuating or preventing a stress appraisal response, or on the other hand, to intervene between the experience of stress and negative outcomes, by regulating the stress reaction or directly influencing physiological processes (Cohen & Wills, 1985). Although distinct in their formulation of operating mechanisms, both conceptualizations of social support are to be taken as complementary and not as mutually exclusive theorizations (Cohen & Wills, 1985; Thompson et al., 2006).

Albeit the sound proposition that relationships between social support and adaptation outcomes are most likely bidirectional (Tijhuis et al., 1995), social support has been mostly studied in scientific research as a determinant or antecedent of health and adaptation outcomes (Cohen, 1992). Therefore, more recently, researchers have stressed the need of exploring the mechanisms by which social support might influence outcomes, namely QL (Helgeson, 2003). This concern was also shared by Schwarzer and Leppin (1991), when they stated that "social support operates partly through other variables and exerts indirect effects on health that may

even exceed the straightforward direct effect" (p. 122/3). Implicit in these recommendations, there was the orientation for analyzing mediation effects (Baron & Kenny, 1986; Holmbeck, 1997) of certain variables on the hypothesized links between social support and adaptation outcomes, thus substantiating an **"indirect effect model"** (Bovier, Chamot, & Perneger, 2004; Ensel & Lin, 1991).

In the disability-stress-coping model (Wallander et al., 1989a), the notion of "resistance factors" and the proposition of their "buffering effect" between disability-related stressors and adaptation outcomes, call for a primary examination of social support within a statistical moderation model (Holmbeck, 1997). However, in situations of chronic stress (e.g. disability or caregiving burden), a mediating (and not a moderating) effect of social support has been reported to occur more frequently (Armstrong, Birnie-Lefcovitch, & Ungar, 2005). In short, this phenomenon could be due to the possibility of chronic stress impairing both help-seeking behavior and support provision (Gottlieb, 1992). More specifically, the study developed by Quittner, Glueckauf and Jackson (1990) stands as a major illustration of the pertinence of assessing the mediating effect of social support in situations of chronic parenting stress: in this study, conducted with mothers of children with a hearing impairment, perceived social support was found to mediate the links between child and maternal stressors, and mothers' psychological distress. This study was based on two major assumptions implied by the social **"support deterioration model"** (Lin & Ensel, 1984): first, some stressful events may elicit disparate or avoidance reactions in members of the social network, leading to more negative perceptions of support that in turn increase psychological symptoms; and second, some stressful events may demand frequent or complex needs of support, which may in the end exhaust resources or lead to negative perceptions of their usefulness and adequacy (Hobfoll & Lerman, 1988). According to Quittner and her colleagues (1990), these claims made particular sense in the context of parenting a child with a chronic condition or disability: in this case, the chronicity of the stressor could entail more pervasive deleterious effects, including in parents' social support perceptions (e.g., some support efforts could be seen as intrusive or incompetent; the exhaustion of resources could diminish help-seeking or help-offering behaviors). As regards the present research work, the interest of revisiting this mediation model of social support relied on three specific research gaps: first, there was no prior examination of this model in a sample of pediatric CP; second, the original validated model did not account for positive outcomes, such as QL, in addition to the more traditional negative dimensions of adjustment; and third, the possibility that the model could be applied to the association between caregiving burden and children/adolescents' adaptation outcomes, had not been yet explored.

2.4. A Developmental Approach to Pediatric Outcomes Assessment – The Disabkids Project

Over the last decades, the changing epidemiology and clinical understanding of childhood health and disease has been based on four distinctive considerations: first, the number of children and adolescents with a chronic health condition has increased; second, the treatment and survival of children/adolescents with more serious conditions has improved dramatically (and the same has happened for prematurely-born children); third, the potential negative impact of illness/disability in developmental processes since childhood till the transition to adulthood, has been widely acknowledged; and fourth, QL and HRQL have been assumed as major outcomes in the treatment of individuals suffering from nonlife-threatening conditions (Bruil & Detmar, 2005). Within this pediatric healthcare context, it has been argued that the traditional clinical endpoints, such as mortality and morbidity, should be complemented with more meaningful outcomes, encompassing developmental progress, educational achievement and psychosocial adjustment (Christakis et al., 2001). Therefore, given their multidimensional nature, QL and HRQL have been adopted as preferred pediatric outcome measures (Koot, 2001).

QL was defined as the "individuals' perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns" (The WHOQOL Group, 1995, p. 1405). QL was thus described as an overarching concept, accounting for individuals' physical and psychological health, level of independence, social relationships, personal beliefs and their relationships to valued features of the environment (The WHOQOL Group, 1994, 1995). HRQL, on the other hand, was defined as a multidimensional construct covering physical, emotional, mental, social and behavioral components of well-being and function as perceived by patients or proxies (Bullinger, 1991; Bullinger & Mackensen, 2004), and hence a component of the more general construct of QL (The Disabkids Group, 2006), which is thought to comprise additional themes such as political participation and economic issues (e.g., Felce & Perry, 1995). Although some criticisms have been made on the notion of HRQL, alternatively described as a marker of disease impact (Wallander, Schmidt, & Koot, 2001), the terminological distinction here provided was aimed at assuming an intermediate conceptual position between those specific disease-related markers and the more general, overarching QL concept.

In the same line of recent approaches to outcome assessment in pediatric settings in general, there has been a tendency of moving from an exclusive (or excessive) focus on impairment and function, to QL and HRQL outcomes in the context of pediatric CP (Schneider et al., 2001). While the importance of not equating disability or function to QL outcomes has

been stressed (Edwards, Huebner, Connell, & Patrick, 2002), the overall notion of QL has emerged as a foremost outcome in pediatric CP (Majnemer & Mazer, 2004; Viehweger et al., 2008). In fact, QL markers have been evaluated as the most important intervention outcomes by youths with CP, their parents and medical professionals, and thus, the overall notion of QL has been labeled as "the holy grail of outcomes" (Vargus-Adams & Martin, 2009). Since the aim of intervention approaches to pediatric CP has been moving from eliminating deficits to enhancing function, QL and HRQL measures have been adopted for a number of clinical (e.g., enhancing clinical decision making, identification of positive changes to motivate the child and family), institutional (e.g., documenting changes related to service provision) and political applications (e.g., planning of appropriate health services and allocation of resources) (Majnemer & Mazer, 2004).

During the most recent decades, although there was an increase in the scientific publications on the topics of QL and HRQL of adults with or without chronic conditions, it became evident that QL assessment in children and adolescents has been a comparatively neglected topic during that same period of time (The Disabkids Group, 2006). This might have been due to a number of methodological and conceptual questions, such as the approach of idiosyncrasies related to those developmental groups, the consideration of children's cognitive abilities and the debate surrounding the selection of respondents (self or proxy sources of information) (Drotar, 1998). In order to foster research on the topic of childhood QL and pediatric HRQL, the World Health Organization (WHO, 1993) presented general guidelines for the development of QL instruments for children, stating that these should be age-related or at least developmentally appropriate. This laudable initiative, pioneered by the WHO, denoted a concern for developing an adequate assessment of pediatric health outcomes, which was to be consistent with their classic, overarching health definition (Prince et al., 2007; WHO, 1948). It is curious to note, however, that the underlined distinction between what is considered to be an "age-related" or a "developmentally appropriate" instrument, somehow implied the difference between a measuring methodology accounting for the content specificities of a given age group, and another one focusing on format, wording and common issues along two different age groups (not to say "a larger age group"). For the purpose of illustrating the pertinence of that distinction, it is worth mentioning that examples of age-related contents include, for adolescents, the themes of body image, autonomy and planning for adult life, and for younger children, attachment to family, development of cognitive and social competence, and play with peers (Gerharz et al., 2003).

The central tenet of **a developmental approach to QL and HRQL assessment** is that adult measures are inappropriate for use with children, because of the level of abstraction required for decision making, the lack of developmental considerations, and the inclusion of certain areas that may be irrelevant, or exclusion of other areas which may be greatly valued (Spieth & Harris, 1996). Nevertheless, similar questions may be pertinently raised when adopting the same model or administering the same instrument to both children and adolescents. In these cases, and since age has been described as a primary developmental variable (Holmbeck, 2002a), stratification of instrument reliability and validation results may well represent an adequate strategy to endorse a developmental approach to QL and HRQL assessment (Gerharz et al., 2003).

As regards the operationalization of HRQL, while a definition of this concept as encompassing the physical, psychological and social domains of functioning has been stated as suitable for children and adolescents (Leidy, Rich, & Geneste, 1999; Ravens-Sieberer et al., 2006), the specific aspects that integrate those three domains may differ to some extent, and thus the item selection for instrument development should be sensitive to the experiences, activities and contexts that are relevant to the age of the sample (Matza, Swensen, Flood, Secnik, & Leidy, 2004). This is to say that although physical, psychological and social aspects of health are important for both children and adolescents, there may be substantial variation in the content of their operationalization (Rajmil et al., 2004). For this reason, items for an instrument aimed at covering a wide age range (e.g., from 8 to 18 years old) are expected to target important commonalities in terms of children and adolescents' developmental contexts and experiences. In fact, the importance of social context for the improvement of pediatric health outcomes assessment has been highlighted extensively (Barros, Matos, & Batista-Foguet, 2008; Bullinger, Schmidt, Petersen, & Ravens-Sieberer, 2006; Christakis et al., 2001; Schmidt, Petersen, & Bullinger, 2003). Moreover, since children have less opportunities and abilities to make changes to their environments, contextual factors may primarily influence the child's long-term adaptation, acting for instance as mediators between disease/treatment variables and adaptation outcomes (Matza et al., 2004). Taken altogether, these considerations account for a need to include age-relevant contexts, such as family, friends and peers, school environment, neighborhood, local health clinics and community, in pediatric QL and HRQL assessments.

The present research work endorsed **a European perspective on QL and HRQL assessment**, which was systematically developed in two different, but interrelated projects: the KIDSCREEN project ("Screening for and Promotion of Health Related Quality of Life in

Children and Adolescents - a European Public Health Perspective") (Ravens-Sieberer et al., 2001), and the DISABKIDS project ("Assessment of Health-related Quality of Life in Children and Adolescents with Chronic Health Conditions and Disabilities") (Bullinger, Schmidt, Petersen, & The Disabkids Group, 2002). Given the fact that the Portuguese versions of the generic Kidscreen instruments had been already developed (Gaspar & Matos, 2008), a substantial component of the current research project was aimed at developing the Portuguese versions of the generic module of Disabkids questionnaires (Carona, Bullinger, & Canavarro, 2011; Carona et al., 2012). For this reason, a critical appreciation of Disabkids instruments as a mean of operationalizing a developmental approach to HRQL assessment is now briefly described.

The DISABKIDS project was originally funded by the program "Quality of Life and Management of Living Resources" of the Fifth Framework of the European Union. The main objective of the project was to develop and promote the use of standardized HRQL instruments in children and adolescents with chronic health conditions (Bullinger et al., 2002). Therefore, the project was substantiated in developing and examining a battery of instruments, which has come to be known as **"The DISABKIDS Questionnaires"**. All these instruments had self and proxy-report forms, and included: a chronic generic module (with a long and a short version); seven condition specific modules (for arthritis, asthma, dermatitis, diabetes, cerebral palsy, cystic fibrosis and epilepsy), and a measure of Smileys (for younger children aged between 4 and 7 years old). For the purpose of the present research work, the agreed priority was to develop the Portuguese versions of the DISABKIDS Chronic Generic Module (known as DISABKIDS-37), and hence the need of a more detailed review on this particular instrument.

The DISABKIDS-37 consists of 37 Likert-scaled items assigned to six dimensions: "Independence" (living without impairments, confidence about future); "Emotion" (emotional problems because of the condition); "Social Inclusion" (positive social relationships); "Social Exclusion" (stigma, feeling left out); "Limitation" (functional limitations), and "Treatment" (perceived impact of taking medication). These six sub-scales are associated with three broader domains, as conceptualized in the WHO classical definition of health (WHO, 1948): "Mental" (Independence and Emotion), "Social" (Inclusion and Exclusion), and "Physical" (Limitation and Treatment). The DISABKIDS-37 questionnaires were originally developed from a simultaneous approach (i.e. different countries participating, at the same time, in the construction of a new instrument) (Simeoni et al., 2007), and their English versions revealed sound psychometric properties in terms of reliability, concordance between parent and child, and factorial,

convergent, divergent and discriminant validities (Petersen, Schmidt, Power, Bullinger, & The Disabkids Group, 2005; Simeoni et al., 2007; The Disabkids Group, 2006).

A plausible method for analyzing the extent of a developmental approach to HRQL assessment in DISABKIDS model and instruments is to critically review them, in the light of a framework of criteria to assess instrument developmental adequacy. If on the one hand, DISABKIDS-37 questionnaires clearly accomplished all the general requirements to QL assessment in children, as established by the WHO (WHO, 1993), on the other hand, different sets of criteria to assess the developmental adequacy of QL and HRQL measures for children and adolescents, have been made available in literature (Bullinger & Ravens-Sieberer, 1995; Spieth & Harris, 1996; Wallander et al., 2001). Nevertheless, it is worth mentioning that the DISABKIDS-37 questionnaires comply with all the requirements established by Bruil and Detmar (2005) for a HRQL instrument for children and adolescents. In sum, DISABKIDS-37 questionnaires seem to have successfully objectified most of the developmental considerations summarized by Wallander and colleagues (2001), namely in terms of competence in verbal comprehension; adequacy of time frames; inclusion of common markers for allowing comparisons between groups; and the selection of items based on children and adolescent's views on their values, issues and ideals (Petersen et al., 2005; The European DISABKIDS Group, 2006).

2.5. Terminological Issues in the Context of the Current Research

Language and terminology have never been minor issues in psychological science. When describing a number of terminological problems in psychological theory at the time, Thouless (1949) stated that a theoretical system was to be classified as good or bad, depending on its ability accomplish two essential requirements: first, its language should be related to a variety of empirical results, and second, that same language should facilitate the development of expectations that could be confirmed or infirmed in later experiments. As for the scientific form of human knowledge in general, and of psychological science in particular, the quintessential assumption that **"the map is not the territory"** (Korzybski, 1933, p. 750) encapsulated the premise that knowledge on the world is to be limited by the structure of human language.

Some basic problems in social science terminology have been described by Riggs (1993), and may be easily recognized in most contemporary psychological concepts and constructs: for instance, for the concept and construct of QL, there have been problems of polysemy (i.e., a variety of possible meanings imputed to the key word) and synonymy (i.e., a set of different terms

applicable to a given concept). Complementarily, Wallander and colleagues (2001) commented on the figurative distance that exists between what we are actually interested in (e.g., QL) and its respective measurements (i.e., responses to a set of items), with the latest representing imperfect indices of the former. Moreover, psychological terminology is amenable to paradigmatic changes, as the one that has recently emerged in the context of disability and rehabilitation: in this case, transition from a pathological to an integrative, social model of disability, was necessarily accompanied by the parallel movement from a deficit-focused, to a more overarching discourse that accounted for the dynamic intersections between the individual and the environment (Pledger, 2003). In sum, the complex dynamics of psychological discourse, language and terminology are noteworthy and should be approached as a crucial methodological component in psychological studies.

As regards the present research work, the inclusion of a section on terminological issues was aimed at clarifying current debates and quandaries, and mostly at justifying specific terminological options. For this reason, the terminological critical reviews here presented were not intended to be exhaustive nor conclusive. To put it briefly, the main purpose of such terminological section was to, within a plausible extent, differentiate overlapping concepts and constructs, and whenever possible, to determine the preferred, the admitted (i.e., permitted when the preferred term was not suitable for a specific context) and the deprecated terms (cf. Hirs, 1993).

Adjustment vs. Adaptation

The concepts of adjustment and adaptation have been used indistinctively, perhaps more common than not (e.g., Bradford, 1997; Davis, Brown, Bakeman, & Campbell, 1998; Yau & Li-Tsang, 1999). Although dictionary definitions tend to emphasize a minor-major change for "adjustment" and "adaptation" terms respectively, the most theoretically sophisticated distinction between the two terms has been provided by "The Resiliency Model of Family Stress, Adjustment, and Adaptation": during the "Adjustment" phase, in face of increasing demands related to a given stressor, families strive to maintain the established patterns of interaction, roles and rules, by activating resources such as family rituals and routines, family hardiness and strong social support; during the "Adaptation" phase, the family develops new coping strategies, capabilities and strengths (e.g., role flexibility, gaining information and knowledge, using humor and laughter) that often have long-term consequences between the individual and the family, and the family and the community levels (McCubbin & McCubbin, 1989; McCubbin, McCubbin, Thompson, Han, & Allen, 1997).

A historical differentiation between the two concepts has been elaborated by Harper (1991), who commented the "adjustment" term as related to a former psychopathological model on childhood illness and disability, and applied the term "adaptation" within the more positive and recent theoretical approaches that accounted for resiliency processes and variability in adaptation outcomes. In fact, the utilization of both "adjustment" and "maladjustment" terms, preceded in most instances by the adjectival forms "psychological" or "psychosocial", was relatively common in a number of studies in pediatric psychology which were developed during the 1980s and the 1990s (e.g., Drotar, 1997; Lavigne & Faier-Routman, 1991; Thompson et al., 1992; Wallander et al., 1989a).

"Adaptation" has been broadly defined as "any process whereby behavior or subjective experience alters to fit in with a changed environment or circumstance" (Colman, 2009, p. 11). This overall notion of "adaptation as a process" was preferred in the context of this research, in agreement with the description of the concept in recent studies on pediatric CP (Rentinck, Ketelaar, Jongmans, & Gorter, 2006). Complementarily, the broad expression "adaptation outcomes" was sometimes adopted to encompass different outcome measures (e.g., QL and psychological maladjustment), in the same line of the original terminology of the disability-stress-coping model (Wallander et al., 1989a). Finally, a further differentiation was endorsed whenever possible: following Thompson and his colleagues' (1994) subtle distinction, the term **"adaptation"** was mostly used to describe the overall process(es), and the term **"adjustment"** was preferably used in relation to specific outcomes (e.g., psychological adjustment or maladjustment). Even so, despite these considerations, the interchangeable use of both terms may have been sporadically admitted, in conformity with a given author's original discourse.

Quality of Life vs. Health-related Quality of Life

The scientific object of QL has been studied by different disciplines and thus approached with a variety of meanings and levels of analysis (Canavarro, 2010). In fact, one of the problematic questions in QL research has been straightforwardly put by Lawton (1997, p. 91): "quality of life is defined in so many ways by so many people and, regrettably, often is not defined". As previously mentioned, the **WHOQOL approach** was adopted in the current research work as the more general framework for a core construct in the studies developed, namely QL. The definition provided in that context highlighted that QL was subjective, multidimensional, and included both positive and negative facets (The WHOQOL Group, 1995). In addition, the related construct of HRQL was defined as a component of the more general construct of QL (Bullinger, 1997; The Disabkids Group, 2006). It has been also commented that,

in addition to the WHOQOL approach (primarily developed for adults), a European perspective to childhood and adolescent QL and HRQL assessment underlay the present research work. In fact, both the Kidscreen and the Disabkids projects clearly cited the WHOQOL approach within their theoretical and methodological foundations (Bullinger et al., 2002; Ravens-Sieberer et al., 2001).

Although the Kidscreen project was concerned with the QL of children and adolescents in general, the authors adopted the term HRQL, in a clear reference to the notion of perceived health, as defined by the WHO (WHO, 1948). In order to avoid conceptual and terminological misunderstandings, Kidscreen and Disabkids instruments were described in this research work as QL and HRQL measures, respectively. Moreover, the following HRQL definition should complement the aforementioned one (Bullinger, 1997): "[HRQL is] a multidimensional concept that includes the broad areas of functional status, psychological and social well-being, health perceptions, and disease- and treatment-related symptoms" (Aaronson et al., 1991, p. 840). In this sense, the intended meaning of HRQL in the current research implied a medical and healthcare perspective on QL (Eiser & Morse, 2001; Koot, 2001). This same distinction between QL and HRQL was also endorsed by Wallander and colleagues (2001), who went even further while criticizing the measures of HRQL as being more disease impact than QL markers.

In sum, since QL and HRQL measures were included in the studies developed within this research work, the following considerations were taken into account: first, HRQL was assumed as a distinct component of the more general QL construct, thus accounting for illness/disability-related specificities (Rosenbaum, Livingston, Palisano, Galuppi, & Russell, 2007); second, when commenting on specific results, and in strict respect with an author's original terminology, the utilization of the "HRQL" term may have been sometimes admitted in stances where the most accurate label for the construct in question would be "QL" (in those situations, additional information such as the comparison of scores with normative data was provided to enlighten the intended meaning of the construct); and third, although QL and HRQL terms were not used interchangeably, an overall expression (e.g., "QL outcomes) was sometimes used for the practical purpose of relating to both QL and HRQL constructs.

Caregiving Burden: An Aversive Label?

Variables related to caregiving stress have been incorporated under the label of "Psychosocial Stresses" in the disability-stress-coping model (Wallander et al., 1989a), and assessed in earlier studies based on that same model (e.g., Wallander et al., 1990; Wallander & Marullo, 1997). Different terms and constructs have been applied in literature to describe the

specific stress(ors) related to family caregiving (i.e., a terminological question of synonymy), and these included: "chronic parenting stress" (Quittner et al., 1990); "role strain" (Quittner, Opipari, Regoli, Jacobsen, & Eigen, 1992); "handicap-related problems" (Wallander & Marullo, 1997); "family burden" (Sales, 2003); "caregiver strain/stress" (Raina et al., 2004), and "caregiving demands" (Klassen et al., 2010). In general, stressors in the context of family caregiving have been defined as "the problematic conditions and difficult circumstances experienced by caregivers (i.e., the demands and obstacles that exceed or push to the limit one's capacity to adapt)" (Aneshensel, Pearlin, Mullan, Zarit, & Whitlatch, 1995, p. 34).

Since two of the studies developed within this research work examined differences in burden (and its relationships with other variables) between parents who had children with CP and parents of typically developing children, **caregiving burden** was broadly defined as "a multidimensional construct, addressing tension and anxiety (stress burden), changes in dyadic relationships (relationship burden), and time infringements (objective burden) resulting from caregiving" (Savundranayagam, Montgomery, & Kosloski, 2011, p. 231). A preliminary conjecture to assess caregiving burden in those studies was that differences in caregiving stress and demands could be related to differences in parents' adjustment (Wallander et al., 1990). In fact, even though caregiving burden had been studied as both a predictor and an outcome variable (Savundranayagam et al., 2011), it was conceptualized here as a determinant of adaptation outcomes, in agreement with the theoretical propositions implied by the disability-stress-coping model (Wallander et al., 1989a) and the more recent models developed in the specific context of pediatric CP (Raina et al., 2004, 2005).

Although the pertinence of studying caregiving burden is substantiated in the amount of literature that has emerged on the topic (Chou, Chu, Tseng, & Lu, 2003), there were recently some negative critiques on the term "burden", which was thought to imply "something unwanted, unrelentingly negative, imposed rather than chosen, and something a person would desire to shed" (Sales, 2003, p. 39). Moreover, not only "burden" was commented as a negative term by families undergoing the experience of continuing family caregiving, but it has been also replaced in some existing measures with the more neutral term of "strain". Finally, some authors claimed that research focusing on burden ignored positive dimensions of the caregiving experience, such as rewards and satisfaction of caregiving, and thus emphasized only "half of the equation" (Sales, 2003). In order to avoid similar criticisms, in one of the studies here compiled, the assessment of negative dimensions of caregiving (i.e., types of burden) was complemented with its positive dimensions (i.e., caregiving uplifts), in concordance with the burden measure adopted (Montgomery et al., 2006).

Positive Perceptions Related to Caregiving: Perceived Uplifts, Benefits, or Growth?

There is growing recognition that **family caregiving is a complex experience**, involving both negative and positive dimensions, such as burdens and benefits, respectively (Green, 2007; Sales, 2003). The occurrence of positive experiences related to family caregiving has been termed in literature as "uplifts of caregiving" (Pinquart & Sörensen, 2003), "positive perceptions" (Gupta & Singhal, 2004), "posttraumatic growth" or "perceived benefits" (Tedeschi & Calhoun, 2004), "growth following adversity" (Joseph & Linley, 2006), and "stress-related growth" (Finzi-Dottan, Triwitz, & Golubchik, 2011).

In face of such terminological variety, the expression **"caregiving uplifts"** was preferred in the present work, as implied by the original terminology of the instrument elected to assess caregiving burden, "The Revised Burden Measure" (Montgomery et al., 2006), which included complementary scales on burden dimensions and caregiving uplifts. In fact, a measure on perceived uplifts was preferred because the notion of "caregiving uplifts" itself was less controversial than other related concepts, such as "psychological growth" (e.g., Wortman, 2004). Moreover, items that integrated the uplifts subscale in the aforementioned instrument pertinently targeted the possibilities of a mindful approach to caregiving (Larson, 2010) and the development of meaning in that context (Folkman, 1997) (e.g., "Have your caregiving responsibilities made you cherish your time with your relative?", "Have your caregiving responsibilities given your life more meaning?"). Therefore, caregiving uplifts were broadly defined in this work as positive psychological states - such as direct enjoyment from tasks, improved relationship with the care receiver and positive affect - derived from caregiving responsibilities (Montgomery et al., 2006).

The selection of a single measure to assess both caregiving burdens and uplifts is noteworthy for two reasons: first, it has been suggested that caregiving uplifts may reduce the impact of burden, although they are independent from burden dimensions (Pinquart & Sörensen, 2003); and second, positive perceptions, such as uplifts, benefits and growth, may co-occur with substantial distress and suffering (Folkman, 1997; Tedeschi & Calhoun, 2004). It is also important to underline that although posttraumatic growth has been approached in literature as both outcome and predictor (Tedeschi & Calhoun, 2004), caregiving uplifts have been mainly studied as outcomes' determinants (Pinquart & Sörensen, 2003). Interestingly enough, in their earlier study, Wallander and colleagues (1990) used a measure of daily hassles and uplifts to operationalize some particular aspects of the overall variable of "maternal psychosocial stress", which was in turn assessed as determinant of mothers' adaptation outcomes.

Cerebral Palsy: Chronic Illness, Developmental Disability, or Chronic Condition?

CP has been classified in literature as a "symptom complex" (Minear, 1956), a "disorder" (Bax, 1964), a "group of motor impairment syndromes" (Mutch et al., 1992), a "developmental disability" (Wang & Jong, 2004), and as a "group of permanent disorders" and a "heterogeneous condition" (Rosenbaum et al., 2005). Just like in the case of its definition and classification, there is no consensual answer for the question of how to refer to CP in general terms. For the purpose of consolidating the terminological option that underlay the current research work, a series of definitions on common labels applied to CP is now presented, namely "chronic illness", "disability", "developmental disability" and "chronic condition".

According to Pless and Douglas (1971), a "chronic illness" was defined as a physical, usually nonfatal condition that lasted longer than three months in year (or required hospitalization longer than one month), and significantly interfered with the individual's regular activities to some degree. Although applicable to most CP cases in general terms, this classical definition relates to "illness", which is a term that has been seldom applied to CP in recent research and practice, and does not accurately suit the mildest forms of CP, where impairment may be almost imperceptible.

On the contrary, the term "disability" has been extensively applied to CP (e.g., Shields, Murdoch, Loy, Dodd, & Taylor, 2006). Within the context of the WHO's international classification of functioning (ICF), "disability" has been defined as "a difficulty in functioning at the body, person, or societal levels, in one or more life domains, as experienced by an individual with a health condition in interaction with contextual factors" (Leonardi, Bickenbach, Ustun, Kostanjsek, & Chatterji, 2006, p. 1220). This is indeed an accommodating concept, appropriate for any health condition, which nevertheless calls for a very specific theoretical framework, namely the ICF (WHO, 2001). In addition, the term "disability" is prone to negative societal views (Larson, 1998) and is particularly open to a number of cultural criticisms (e.g., Breckenridge & Vogler, 2001) and healthcare delimitations (Fried, Ferruci, Darer, Williamson, & Anderson, 2004). Subsequently, the expression of "developmental disability" constrained its merits to the underlining of a continuing impact of disability on the individual's developmental progress (Wang & Jong, 2004).

For the purposes of the present research work, the expression "chronic condition" (interchangeably used with "chronic health conditions" or "chronic physical conditions") was preferred in the discourse related to CP. A "chronic (health) condition" was defined within a set of criteria: conditions should have a biological, psychological or cognitive basis; have lasted or expected to last for at least one year; and produce one or more significant sequelae. These

sequelae could include: (1) limitations of function, activities or social role in comparison with healthy age peers; (2) dependency on some assistance or treatment (i.e., medications, special diet, medical technology, assistive device or personal assistance) to minimize or compensate limitations of function; and (3) a need for medical care or related services, psychological services, or educational services above the usual for the child's age or for special ongoing treatments, interventions, or accommodations at home or in school (Stein, Bauman, Westbrook, Coupey, & Ireys, 1993). This definition, though exhaustive, portrayed chronic conditions as complex, varied phenomena, which usually require the consideration of different contexts and dimensions of function assessment and intervention. Moreover, this definition was specifically developed from pediatric research (Stein et al., 1993), and the expression "chronic condition" had been utilized by the Disabkids project (The Disabkids Group, 2006).

Despite the fact that we had approached **CP as "a complex and multidimensional condition"** (Murphy, 2008, p. 160), it is important to note that the expressions of "chronic condition", "chronic health condition" and "chronic physical condition" were preferably (not exclusively) used: in different stances, in agreement with a given author's discourse or as a matter of avoiding redundant vocabulary, terms such as "disability" or "developmental disability" were also admitted. As a final remark on the evolution of scientific discourse on the matters of disability, it is interesting to observe that previous expressions, such as "disabled children" (Kazak, 1987) or "chronically ill and handicapped children" (1989), which had been progressively replaced with the standards of a "nonhandicapping language" in research (American Psychological Association, 1992), are now gaining a renewed interest as means of emphasizing a social model of disability (Colver, 2005, 2006).

3. Psychosocial Adaptation of Children and Adolescents with CP and their Parents: State of the Art and Current Challenges

3.1. Adaptation Process and Outcomes of Children and Adolescents with CP

QL and HRQL Outcomes and Related Factors

During the last couple of decades, research on the assessment procedures underlying diagnosis and intervention outcomes evaluations for children and adolescents with CP has moved from an exclusive focus on disability, impairment and functioning, to the incorporation of global, multidimensional measures on the variables of QL and HRQL (Davis et al., 2009; Schneider et al., 2001; Vargus-Adams, 2005). If on the one hand, progress in the research of pediatric QL outcomes has been slow due to a number of conceptual and operational difficulties (Drotar, 1998), on the other hand, that progress may have been even slower for pediatric CP. In fact, given the extreme variability of CP clinical manifestations (Liptak & Accardo, 2004), it is likely to conjecture that operational difficulties, such as age particularities or the debate surrounding self versus proxy reports, may have been amplified in the context of this particular condition. Specifically, the existence of communicative barriers, the limited number of validated instruments and the diversity of impairments associated with CP, represented additional challenges for the measurement of QL and HRQL in children and adolescents with CP (Livingston, Rosenbaum, Russell, & Palisano, 2007). For example, in a relatively recent literature review, it was commented that there were very few quantitative studies on the impact of pediatric CP on QL (The Disabkids Group, 2006); moreover, it was not until recently that Varni and colleagues (2005) claimed to have conducted the first study on self-reported HRQL of children and adolescents with CP.

Although general guidelines for pediatric QL assessment have stressed the need to "employ subjective self-report wherever possible" (WHO, 1993, p. 3), **a complementary approach** accounting for self and proxy sources of report has been preferred in the research of QL and HRQL of children/adolescents with CP (White-Koning et al., 2007). This preference has been justified because both parents and children are assumed to provide additional insights (Gates, Otsuka, Sanders, & Mcgee-Brown, 2010) and also because such procedure may be useful to map differences in perceptions between sources of information (Varni et al., 2005). When adopting this differential assessment procedure, a widely reported tendency of discrepancy has been verified for pediatric CP: while parents of healthy children/adolescents tend to overrate their children's QL, parents who have children with chronic conditions are more likely to underestimate their children's QL (White-Koning et al., 2007). Since this discrepancy tended to

increase as greater was the child's level of physical impairment, Oeffinger and colleagues (2007) proposed that such divergence could be explained by the following argument: "The child's perception is one of *ability* as the impairment was not acquired after a period of normal development. Children tend to score themselves at the highest level and emphasize what they *can* do. Parents have the expectation that their child should be able to do everything able-bodied children can do. Therefore, the parent's perspective is more likely one of *disability* and emphasizes what the child *cannot* do" (p. 178). In fact, in a number of studies, children and/or adolescents with CP rated their QL/HRQL higher than their parents did (Gates et al., 2010; Majnemer, Shevell, Rosenbaum, Law, & Poulin, 2007; White-Koning et al., 2007), which led some authors to conclude that "children with CP are more resilient and positive about their HRQL than their parents think they are" (Janssen, Voorman, Becher, Dallmeijer, & Schuengel, 2010, p. 344).

In sum, parent and child-reports are more complementary than mutually exclusive (White-Koning et al., 2007), and both should be combined wherever possible in pediatric CP research: if on the one hand, children's reports are a valuable mean of identifying "hidden morbidities" (Varni et al., 2005), and thus improve intervention processes and outcomes, on the other hand, parents' perception of their child's well-being is a foremost determinant for the utilization of healthcare services (Upton, Lawford, & Eiser, 2008). Moreover, it is important to bear in mind that between one third (Arnaud et al., 2008) to half (Varni et al., 2005) of pediatric CP cases have been observed to be incapable of providing self-reports, thus highlighting the pertinent need of allowing proxy-reports for those cases where QL/HRQL assessment is equally useful and required.

Children and adolescents with CP have been systematically reported to have a significantly impaired QL (or HRQL), when compared to normative samples or data (Maher, Olds, Williams, & Lane, 2008; Vargus-Adams, 2005; Varni et al., 2005) and to groups of children with other chronic health conditions (Schmidt et al., 2006; Varni et al., 2005). Nevertheless, it has been also commented that half of the children with CP may experience a QL that is similar to their typically developing peers', thus underlining the existence of a high variability of QL outcomes in this group (Majnemer et al., 2007). Contrasting with most findings reported in the literature, in a recent study, children with CP reported a similar QL to children in the general population in all domains, except schooling (where findings were inconclusive) and physical well-being (where comparisons were not performed) (Dickinson et al., 2007). These recent and unexpected findings eventually led the authors to further emphasize the role of contextual factors as important determinants of the QL/HRQL experienced by children and adolescents with CP (Dickinson et al., 2007; Majnemer et al., 2007). For the purpose of the present review, three

groups of determinants were especially considered, namely: socio-demographic factors (i.e., age, gender and socioeconomic status [SES]); clinical variables (i.e., level of physical impairment); and social factors (i.e., social support and family context).

As regards the influence of **socio-demographic factors** on QL/HRQL outcomes, the available findings are inconsistent and thus inconclusive. Some studies found no correlation between age and QL/HRQL in children and adolescents with CP (Maher et al., 2008; Majnemer et al., 2007; Vargus-Adams, 2005); however, each one of these studies focused on children or adolescents separately and hence potential differences between age-groups could not be accurately ascertained. Gender differences, on the other hand, have been scarcely studied, and apart from the verified relationship between adolescent's male gender and parent-child disagreement in HRQL assessment (Gates et al., 2010), research questions regarding the potential role of gender on QL/HRQL outcomes in pediatric CP remain understudied. Nevertheless, one study reported no differences on QL scores between girls and boys (Majnemer et al., 2007). Finally, despite the fact that family income was not related to children's QL in one study (Majnemer et al., 2007), in another two studies, SES was weakly though significantly associated with parent-reported child QL (Arnaud et al., 2008) and with self-reported QL in adolescents with CP (Maher et al., 2008).

The **level of physical impairment** (also termed as "functional dependence", "motor functioning" or "severity of impairment"), usually assessed with Gross Motor Function Classification System for CP (Palisano et al., 1997), stands as one of the clinical variables more widely studied in the context of pediatric CP. As theoretically hypothesized in the disability-stress-coping model (Wallander et al., 1989a), the association between the level of physical impairment of individuals with CP and their QL/HRQL outcomes is far from a deterministic relationship. The observed correlations between level of motor impairment and QL outcomes have been classified as variable and weak (Rosenbaum et al., 2007), moderate (Maher et al., 2008; Schneider et al., 2001) and strong (Vargus-Adams, 2005). In general terms, some evidence suggested a more impaired HRQL in cases of quadriplegia than in CP milder forms, such as diplegia or hemiplegia (Varni et al., 2005). More specifically, functioning level has been verified to be more closely related to physical than to psychosocial QL domains (Shelly et al., 2008), or even to physical well-being only (Majnemer et al., 2007). Apparently contradictory, greater severity of impairment has been demonstrated not to equate to poorer QL; in fact, poorer QL has been reported for children with milder forms of CP, thus suggesting the existence of specific factors (other than the child's severity of impairment) in determining lower QL for children with CP (Arnaud et al., 2008). In face of such incongruent findings, some authors suggested that "gross

motor function is related to HRQL only to a certain point, after which it is not a good predictor" (Maher et al., 2008, p. 54).

Although contextual factors, such as **social support** and **family members' adaptation**, have been acknowledged as important determinants of QL outcomes for children and adolescents with CP (Dickinson et al., 2007; Majnemer et al., 2007), research on these psychosocial topics is definitely lacking. Important variables of a family context, such as parenting stress (Arnaud et al., 2008) and parental well-being (White-Koning et al., 2007; White-Koning, Grandjean, Colver, & Arnaud, 2008) have been mostly studied in relation to parent-child agreement in QL assessments, and to a much lesser extent, as determinants of the child/adolescent's QL outcomes. Nevertheless, some evidence has been reported for the determinant role of family variables on children/adolescents' QL: parenting style (Aran, Shalev, Biran, & Gross-Tsur, 2007) and parenting stress and parents' depressive symptoms (Wiley & Renk, 2007) were found to be positively and negatively correlated with the proxy-reported QL of children and adolescents with CP. Complementarily, despite the fact that social support has been stated as a foremost environmental factor within the social model of disability (acting as either a facilitator or a barrier) (Mihaylov, Jarvis, Colver, & Beresford, 2004), it has been scarcely studied in children and adolescents with CP. Social support has been suggested to play a significant role in the adaptation of adults with CP (Horsman, Suto, Dudgeon, & Harris, 2010), but apart from some anecdotal evidence, the quantitative examination of such role clearly remains an understudied topic in pediatric CP.

Psychological (Mal)Adjustment Outcomes and Related Factors

Throughout the history of pediatric psychology research, there was a general tendency of moving from an eminently negative, **deficit-based and psychopathological perspective**, to a more complex, **positive and resilience-based approach** to the understanding of pediatric adaptation processes and outcomes (Bradford, 1997; Eiser, 1997). However, for the case of pediatric CP in particular, the direction or at least the intensity of such scientific (and therefore clinical) movement does not seem so marked. In fact, in contrast to the research conducted on the topics of QL and HRQL during the last two decades, studies on the behavioral difficulties and psychological (mal)adjustment of children/adolescents with CP represent a most recent tendency of psychosocial research in pediatric CP (e.g., Brossard-Racine et al., 2012a; Brossard-Racine et al., 2012b; Vles et al., 2012). Two main reasons may have accounted for this renewed interest: first, the consideration that a focus on either the positive or negative adaptation dimensions is most likely to provide an incomplete view of adaptation processes and outcomes

(Ridder et al., 2008); and second, a need to explore the nature and dimension of significant (psychological) hidden morbidities that have been reported in QL research (Varni, Burwinkle, & Lane, 2005; Varni et al., 2005).

Although psychological adjustment or difficulties have been theorized and examined as ultimate adaptation outcomes during an earlier phase of pediatric psychology research (e.g., Wallander & Varni, 1989; Wallander et al., 1989a), recent frameworks have defined them as more specific outcomes and hence as foremost QL or HRQL determinants (Bovier et al., 2004). A notable exception to the research scenario described above was a population-based study on behavior problems of children with CP that was conducted in the 1990s (McDermott et al., 1996). In that study, the authors highlighted the underestimation (and understudy) of behavior problems in pediatric CP and stated such behavioral and emotional problems as preventable psychosocial morbidities. Following their own observations, the authors concluded that children with CP were five times more likely to present behavior problems than healthy children, and that those problems mostly included difficulties related to dependent, headstrong and hyperactive behavior (McDermott et al., 1996). These findings were in line with previous reports that called the attention to the fact that children suffering from chronic conditions affecting the central nervous system were in greater risk to develop psychiatric and psychological morbidity, in comparison to children with other health conditions or no health problem (CCD & CPACFH, 1993).

In general, the most common psychological difficulties reported to date for children/adolescents with CP were interpersonal difficulties, attention-deficit and hyperactivity symptoms, reduced social skills, emotional problems, and increased dependency and withdrawal (Brossard-Racine et al., 2012a). Quite pertinently, a recent research revealed that more than one third of children with CP had psychological difficulties within the borderline to clinically abnormal range, and that peer problems were among the most common difficulties (Brossard-Racine et al., 2012a). As regards children with hemiplegia in particular, increased rates of psychological maladjustment have been reported (mostly including hyperactivity and peer problems), with boys presenting greater risk for conduct problems and hyperactivity (Parkes, White-Koning, McCullough, & Colver, 2009). In a recent European population-based study, clinical levels of psychological symptoms (i.e., requiring specialized services) were found for nearly a quarter of children with CP; in addition, better gross motor function was associated with increased psychological maladjustment (Parkes et al., 2008). This latest result was somehow concordant with the previous observation that less severely impaired children with CP were more likely to have poor QL (Arnaud et al., 2008), and could be hypothetically attributed to the fact

that children with severe CP lacked the (physical) capacity to exhibit misbehavior (Vargus-Adams, 2005). However, in a recent study where increased psychosocial maladjustment was verified for children and adolescents with CP, higher restriction in gross motor function significantly predicted worse psychosocial adjustment (Vles et al., 2012), thus suggesting a need for considering additional variables in the examination of such relationships.

It is also very important to note that relationships between psychological (mal)adjustment in children/adolescents with CP and intrapersonal or social-ecological factors (cf. Wallander et al., 1989a) are just now beginning to be explored: for example, better social skills and lower parental stress have been recently observed to correlate with positive children's behavioral adjustment (Brossard-Racine et al., 2012a). Finally, on the topic of the associations between psychological adjustment and QL/HRQL in pediatric CP, internalizing behavior problems have been identified as significant predictors of children's QL (Wiley & Renk, 2007). Depending on the source of report, HRQL has been related to both parent and child-reported internalizing problems, but only to parent-reported externalizing problems (Janssen et al., 2010).

Adaptation Process and Outcomes for Children and Adolescents with CP: Current Challenges and Questions

In the sequence of the previous sections, an immediate critical question that arises from the current body of research is the relatively narrowed focus on variables for outcome assessment: if on the one hand research on the topics of QL and HRQL for children and adolescents with CP has flourished, psychological and psychosocial adjustment in this pediatric population "remains underrepresented in current literature" (Vles et al., 2012, p. 365), and its "relationships with intrinsic or extrinsic factors are just beginning to be explored" (Brossard-Racine et al., 2012a, p. 35). This research scenario implies a need for greater investment in the study of psychological (mal)adjustment of these children/adolescents, thus broadening the scope of outcomes research, which is to encompass higher-order (e.g., QL and HRQL) and more specific health domains (Christakis et al., 2001).

Notwithstanding its pertinent development and contributions, it should nevertheless be noted that QL research in pediatric CP sometimes lacked conceptual and methodological clarity: for example, some studies reported on the concepts of QL or HRQL, but utilized measures on related though conceptually distinct variables, such as general health perception (Aran et al., 2007; Schneider et al., 2001; Vargus-Adams, 2005) or lifestyle and perceived developmental progress (Wiley & Renk, 2007). Even if some of these criticisms could be understood as methodological attempts to initiate and foster topics of research that were considerably understudied, current

methodological resources and conceptual frameworks provide a variety of instruments to reliably assess QL and HRQL in children and adolescents with CP (Viehweger et al., 2008; Waters et al., 2009), as well as theoretical and terminological distinctions that not only assume QL and HRQL as ultimate outcomes (Livneh & Antonak, 2005), but also as different (though related) constructs that need to be considered separately (Rosenbaum et al., 2007).

Regarding the more specific methodological approaches to adaptation outcomes assessment in children and adolescents with CP, two questions are particularly noteworthy: there has been an excessive reliance in comparing outcomes of this population with normative sample or data, as much as in obtaining child/adolescent-related information from proxies, namely parents. The conduction of comparison analyses between groups of healthy or typically developing individuals and those with chronic conditions has been criticized in literature as a "two-group" mentality, which does not fully account for the complex individual variability in adapting to disabling conditions (Harper, 1991); even so, a rationale for the conduction of such analyses has been argued for the study of QL in pediatric CP: for instance, as a mean of identifying major areas of impairment and assisting diagnosis and intervention processes (Viehweger et al., 2008). In order to best achieve the intended purpose of such analyses, it has been suggested that comparisons of pediatric samples with their peers (i.e., homologous samples) should be preferred to those performed with random norms (Gerharz et al., 2003). In fact, this procedure should be carefully taken into account, since results tend to be disparate when utilizing different methodologies in such comparison analyses (Lavigne & Faier-Routman, 1992). However, in the studies reviewed here, the conduction of comparison analyses with homologous control samples was far more the exception than the rule.

Complementarily, given the available evidence that at least half of children with CP can reliably self-report on their subjective states (Varni et al., 2005), aside with the evidence that parents' emotional states may influence the perception of their children's well-being (Arnaud et al., 2008), the fact that a great proportion of the studies exclusively relied on proxies to obtain child/adolescent-related information, seems regrettable. Actually, most of the recent studies on psychological adjustment of children/adolescents with CP were based on parent-reports (e.g., Brossard-Racine et al., 2012a; Brossard-Racine et al., 2012b; Vles et al., 2012; Wiley & Renk, 2007), and even in those QL studies where self and proxy-reports where obtained, these data were mainly used to assess parent-child levels of agreement (e.g., Gates et al., 2010; White-Koning et al., 2007). Given the fact that child/adolescent self-reports should be obtained wherever possible, in order to prevent the underestimation of emotional and psychological morbidity (Livingston et al., 2007; Varni et al., 2005), future studies on the psychological

(mal)adjustment of children and adolescents with CP and/or its associations with QL/HRQL should preferably incorporate self-reported measures in those constructs of interest.

Despite the increasing number of studies addressing the characterization of adaptation outcomes in children and adolescents with CP - such as QL, HRQL and psychological (mal)adjustment – research on their correlates and determinants is definitely lacking (Brossard-Racine et al., 2012a; Livingston et al., 2007; Maher et al., 2008; Majnemer & Mazer, 2004). The overcoming of this research gap is perhaps the most prominent challenge for psychosocial research in pediatric CP. For instance, QL in young people without disabilities has been related to a number of developmental variables, such as age (QL decreases from childhood to adolescence), gender (girls score lower in physical and psychological QL domains than boys), and SES (higher SES is associated with higher QL) (The European Kidscreen Group, 2006); however, there is a paucity of research examining the extent and direction of the influence of these variables in the adaptation outcomes of children and adolescents with CP. Subsequently, research on (potential) adaptation mechanisms and processes is even scarcer, which reveals an underutilization of mediation and moderation analyses that have been described as utterly important for child-clinical and pediatric psychology research (Holmbeck, 1997; Rose et al., 2004).

Age and gender have been widely studied in relation to developmental psychopathology processes (Hudziak, Achenbach, Althoff, & Pine, 2007) and pediatric adjustment to chronic diseases (Eiser et al., 1992). In addition, the influence of **SES** on children/adolescents' physical and psychosocial health has been fairly established in literature (Chen, 2004; Chen, Martin, & Matthews, 2006). However, none of these variables has been systematically addressed in the study of adaptation outcomes and determinants in pediatric CP. Age, for instance, has been considered a primary developmental variables in health and illness research (Holmbeck, 2002a), and although a functional decline from childhood to adolescence has been documented in CP (Krakovsky, Huth, Lin, & Levin, 2006; Stevenson et al., 2006), age-group differences have been rarely addressed in the study of QL and HRQL outcomes in children and adolescents with CP (Livingston et al., 2007). Similarly, although a decrease in behavior difficulties has been related to ageing (McCullough, Parkes, Kerr, & McDowell, 2011), recent research studies have failed to incorporate the examination of age-group developmental differences in their designs. In sum, the exclusion of these variables from research designs for pediatric CP is no minor issue, since "aggregating results across ages and sex may obscure true differences" (Shields et al., 2006).

Finally, for the purpose of the present research work, two psychosocial determinants of adaptation outcomes in children and adolescents with CP warrant a specific comment, namely **social support** and **parental adjustment**. The importance of environmental factors, such as

social support and family context, has been stressed in relation to health and social outcomes of children/adolescents with CP (Liptak & Accardo, 2004). Despite the fact that social support has been classically examined as a predictor of the adaptation outcomes in children with chronic conditions and disabilities (cf. Wallander & Varni, 1989), only anecdotal evidence has been gathered for the importance of social support processes on the adaptation of individuals with chronic disabilities, including CP (King, Willoughby, Specht, & Brown, 2006). In addition, even if youngsters with chronic conditions have been reported to have more peer problems than other youths, children/adolescents with chronic conditions involving the central nervous system have been commented to encounter additional peer difficulties (LaGreca et al., 2002). Interestingly enough, in a European study of parent-reported QL of children with CP, the most impaired QL domain was the one relating to social support (Arnaud et al., 2008). Nevertheless, apart from a recent study, where moderate correlations were observed between social activities/support, psychopathological symptoms and HRQL (Frontini, Crespo, Carona, & Canavarro, 2012), the quantitative examination of social support in children/adolescents with CP, as well as its relation to their adaptation outcomes, remain considerably understudied in pediatric psychosocial research. As commented above, the influence of family context and parental adjustment on the adaptation of children and adolescents with CP has been an equally neglected research topic. Previous research agendas for pediatric psychology emphasized the need to conduct studies relating parent and family functioning to children/adolescents' adaptation processes and outcomes (Drotar, 1997; Harper, 1991), however, such recommendations have not been properly addressed in the context of pediatric CP: few research has been conducted on the determinant role of parental variables, and fewer still (if any) on the potential mechanisms via which parental adjustment may influence children's adaptation outcomes. Apart from studies primarily aimed at assessing parent-child concordance in QL assessments (e.g., White-Koning et al., 2007), and those that were based in proxy-reports only (e.g. Wiley & Renk, 2007), research conducted so far has notably failed to substantiate **a parent-child perspective** on the study of adaptation mechanisms and outcomes of children and adolescents with CP.

3.2. Adaptation Process and Outcomes of Parents of Children/Adolescents with CP

Adaptation Outcomes and Related Factors

Although caregiving is a normative component of parenting in general, the amount and/or quality of parental care required by a child with a chronic physical condition often exceed the expected level of care that regularly characterizes parenting (Krulik et al., 1999; Raina et al.,

2004; Turner-Henson et al., 1992). The role of primary family caregiver, which is of paramount relevance in the present work, has been traditionally assumed by or attributed to mothers, thus it is not surprising that a focus on the maternal figure has been the dominating tendency in most of the research conducted on parental adaptation in rearing a child with a disability (Yau & Li-Tsang, 1999). There are indeed a variety of reasons accounting for the importance of studying the adaptation process and outcomes of family caregivers of children with chronic conditions and/or disabilities: first, the role of environmental factors, such as parents' health and well-being, on child's health, has been systematically emphasized; second, contemporary movements in health services delivery implied family members to be more actively involved in patient's care; and third, the economic sense underlying the prevention of costs derived from the caregivers' health protection and promotion, has been gradually acknowledged (Brehaut et al., 2004).

As it was the case for pediatric psychology in general, and for children with chronic physical conditions in particular, research on the adaptation process and outcomes of these parents has progressively moved from a traditional approach of examining the caregiving stressors and their impact on parents' stress and depression, to a more holistic and contemporary approach where variability in adaptation and multidimensional outcomes, such as QL, are greatly valued (Davis et al., 2009). Despite the consensual tenet that parents who have children with disabling chronic conditions face considerable stress, the simultaneous observation of significant variability in the ways they cope and adapt, has led researchers to embrace the transition from deficit, dysfunction-based models, to a risk-resilience framework that enables a more comprehensive understanding of the diversity of trajectories related to stress, coping and adaptation processes in those situations (Beresford, 1994; Rentinck et al., 2006; Florian & Findler, 2001).

For parents of children with CP, increased levels of parenting stress (Wang & Jong, 2004) and augmented risk for psychiatric morbidity (Mobarak, Khan, Munir, Zaman, & McConachie, 2000) have been reported. In general, the existence of worse physical and psychological health in these parents, in comparison to other parents or caregivers, has been broadly commented in literature (Brehaut et al., 2004; Raina et al., 2005). More recently, results from qualitative research suggested that caring for a child with CP affected parents' QL in a most pervasive way, including on their physical and social well-being, freedom and independence, family well-being and financial stability (Davis et al., 2010). In addition, latest findings verified that parents of school-aged children with CP were likely to experience high stress and psychological burden, associated with a negative impact of the child's health on their time, emotional status and family activities (Majnemer, Shevell, Law, Poulin, & Rosenbaum, 2012). Nevertheless, two recent studies using

the same measure to assess these parents' QL (i.e., WHOQOL-BREF; The WHOQOL Group, 1995), provided results that somehow illustrate the variability of adaptation outcomes in this population: in the first study, although more than half of the parents rated their QL as "good", QL scores were lower in all domains (i.e., Physical, Psychological, Social and Environmental), when compared to those reported by parents of healthy children (Okurowska–Zawada, Kułak, Wojtkowski, Sienkiewicz, & Paszko-Patej, 2011); in the second study, parents of children with CP showed lower scores in the Physical and Psychological QL domains, in comparison to a control group of parents of healthy children (Romeo et al., 2010).

The study of biopsychosocial issues in families of children with chronic health conditions implies a broadened focus on different dimensions of adjustment, and in a variety of factors such as SES, social support and family functioning (Lewis & Vitulano, 2003). For the present work, the following variables and their relationships to the adaptation outcomes of parents of children/adolescents with CP were reviewed in greater detail: the child's age and level of motor impairment, and parent's caregiving burden and social support.

The study of **child's age and severity of impairment** in relation to family adaptation outcomes has been early commented as potentially complex as well as a promising research direction to be pursued in pediatric psychology (Kazak, 1987). Since the **child's age** is related to a family's development phase to a greater extent than parents' age, the importance of incorporating the study of age group differences between childhood and adolescence in outcomes research for CP has been highlighted by different authors (Lin, 2000; Rentinck et al., 2006; Wang & Jong, 2004). In addition, most studies on family adaptation to CP have been conducted in families of children, and to a much lesser extent in families of adolescents or young adults (Magill-Evans et al., 2001). Therefore, the examination of adaptation differences between the developmental periods of childhood and adolescence has been studied infrequently. Although sparse qualitative evidence suggested that most central issues related to family caregiving in pediatric CP remained stable throughout childhood and adolescence (Davis et al., 2010), some quantitative evidence suggested otherwise. Older child's age (and not mother's age) along with reduced socioeconomic resources have been reported as main predictors of stress in mothers of children with CP (Mobarak et al., 2000). Moreover, families with school-aged children with CP have shown more positive coping appraisals and better social interactions than families of young adults with the same physical condition (Lin, 2000). Taken altogether, in comparison to childhood, adolescence and transition to adulthood seem to be particularly challenging periods for families of individuals with a disability, though the available empirical evidence is clearly insufficient to establish such claim.

Child characteristics, such as the **level of a child's physical impairment**, have been systematically noted as key contributors to maternal stress (Button, Pianta, & Marvin, 2001). In general, the severity of the child's impairments (and mostly when communication is also impaired) has been found to be positively correlated to parental stress (Yau & Li-Tsang, 1999). Concordantly, child's level of impairment and factors related to SES have been verified as strong predictors of maternal well-being related to caring for a child with a disability (Trivette & Dunst, 1992). As regards CP in particular, although a qualitative study has reported a positive correlation between the severity of the condition and the degree of caregiver's QL impairment (Lim & Wong, 2009), there seems to exist considerable variability in the way those two groups of variables relate to each other. In another (quantitative) study, disability severity and child's functional status did not predict maternal depression, and hence, a lower level of child's impairment was not related to a better maternal adaptation (Manuel, Naughton, Balkrishnan, Smith, & Koman, 2003). Two hypotheses were then advanced by the authors in order to explain such counterintuitive result: on the one hand, in comparison to higher functioning children (usually with less visible problems), lower functioning children could be exempt from certain expectations; on the other hand, parents of higher functioning children could experience more psychological burden than one would tentatively expect. In addition, an alternative (or at least complementary) explanation could be formulated as follows: in the sequence of a child's clinical diagnosis, parental resolution processes (cf. Marvin & Pianta, 1996) could be firmly oriented by the acceptance of aversive labels such as "disability" or "chronic condition", which could be promptly applied to most severe CP, but nevertheless trigger psychological ambivalence or resistance in parents of children with milder forms of CP, where physical impairment is likely to be small or almost imperceptible.

Given the inconsistent findings reported for the associations between child's impairment variables and parental adaptation outcomes, some authors suggested that perceived **caregiving burden and strains** should be studied in association with parents' adjustment, instead of objective disability-related parameters, such as condition severity or functional independence level (Horton & Wallander, 2001). Variables related to caregiving burden have been theoretically stated as primary risk factors in general models of family adaptation to pediatric conditions (Wallander & Varni, 1992), as well as in more specific models of caregiving process and caregiver burden that were recently developed in the context of CP (Raina et al., 2004). Complementarily, evidence has been gathered for the determinant role of caregiving burden on the psychological adjustment of family caregivers of children with chronic medical conditions (Canning, Harris, & Kelleher, 1996), and also on the physical and psychological health of family caregivers of children

with CP (Raina et al., 2005). Despite the available evidence on the determinant role of burden on parents' adaptation outcomes, a multidimensional approach (Savundranayagam et al., 2011), targeting both negative and positive dimensions of the caregiving experience (Green, 2007; Sales, 2003), has not been yet applied to the psychosocial outcomes research with parents of children/adolescents with CP.

Research on the topic of **family's social contexts and support** has been described as imperative in the context of childhood chronic illness/disability (Kazak, 1989). Indeed some early pediatric psychology research was aimed at understanding the relationships between the social environment and the adaptation of mothers of children with chronic conditions and disabilities (Wallander et al., 1989b). More recently, the relevance of environmental factors, such as social support, has been stressed in relation to the QL of parents and families of children with CP (Davis et al., 2010; Lim & Wong, 2009). For parents of children/adolescents with CP, the existence of little social support has been occasionally commented (Davis et al., 2010); however, quantitative studies have found no differences between the need for social support in these parents and those of typically developing children (Britner, Morog, Pianta, & Marvin, 2003), nor in their social support and that reported by other family caregivers (Brehaut et al., 2004). For the present review of studies on social support and parents' adaptation outcomes, three analytical approaches to social support were considered, namely its main, moderating (i.e., buffering) and mediating effects. The presence of social-ecological factors, such as greater social support, has been linked to lower burden and better emotional well-being in parents of children with neurodevelopmental disabilities, including CP (King, King, Rosenbaum, & Goffin, 1999), and with the mental health of mothers of children with chronic physical conditions (Horton & Wallander, 2001). In apparent contrast, one study with mothers of children with CP found no significant association between mothers' perceived social support and stress (Mobarak et al., 2000), although the authors have stated the low reliability of the social support measure used in the study as a significant limitation. The pertinence of analyzing the moderating, buffering role of social support against adverse events and negative outcomes has been noted for families of children with CP (Britner et al., 2003), and in one study, social support was found to moderate the association between the child's functional level and mothers' depressive symptoms (Manuel et al., 2003). Finally, social support was also found to mediate the links between child/maternal stressors and mothers' psychological distress (Quittner et al., 1990); in fact, further evidence has been recently gathered for the mediating role of social support in the adaptation of parents of children with chronic health conditions (Hatzmann, Maurice-Stam, Heymans, & Grootenhuis,

2009). Despite the clinical relevance and empirical support for this functional hypothesis of social support, it remains to be examined in parents of children/adolescents with CP.

Adaptation Process and Outcomes of Parents of Children/Adolescents with CP: Current Challenges and Questions

The following methodological limitations have been described as the most common in research on psychosocial adaptation of parents of children/adolescents with chronic physical conditions: small sample sizes; use of a single indicator of adaptation; and lack of a control group (Florian & Findler, 2001). In addition, the fact that most of research on parental adaptation to pediatric chronic conditions has been conducted in samples that mostly or exclusively included mothers, could be commented as another specific limitation. In the context of pediatric CP, some evidence has suggested that mothers and fathers may differ in the ways they perceive and cope with stressors (Yau & Li-Tsang, 1999), namely in terms of perceived social support (Magill-Evans et al., 2001). For this reason, the inclusion of fathers in studies with primary family caregivers of children/adolescents with CP has been highly recommended (Lin, 2000; Rentinck et al., 2006). Notwithstanding the pertinence of such recommendation, it should be noted that the simultaneous study of mothers and fathers as "parents" (without the conduction of separate analyzes for gender effects, often because of statistical limitations implied by small-sized subsamples) has been the main methodological trend in pediatric psychology research. However, even if there are more similarities than differences between fathers and mothers of children with chronic conditions, the consideration of methodological guidelines that may foster the examination of the abovementioned specificities should by no means be discouraged (Phares, Lopez, Fields, Kamboukos, & Duhig, 2005).

While most research conducted in families of children with chronic conditions, namely CP, has focused on the description of negative impacts and outcomes, the alternative examination of coping and adaptation processes within a risk-resilience framework has been notably emphasized (Horton & Wallander, 2001; Lin, 2000). Common criticisms reported for psychosocial research on parental adaptation to pediatric conditions, include the lack of clear theoretical frameworks and the disregard of positive dimensions related to adaptation processes (Barlow & Ellard, 2006; Lin, 2000). A **strength-based approach**, accounting for strengths and positive perceptions as means of family coping, is highly desirable in the context of parental adaptation research (and its clinical implications), since "one goal of family-centered early intervention is to identify existing family strengths and capabilities so that interventions are built on things a particular family already does well" (Judge, 1998, p. 263). The fact that positive and

negative psychological states may co-occur during caregiving processes (Folkman, 1997), has led some researchers to argue that perceived benefits and positive perceptions should be incorporated in the literature and research on burden related to parenting children with disabilities (Green, 2007; Larson, 1998). Actually, a number of positive variables has been studied, or at least commented, in relation to caregiving processes, and these included the following: "meaning-making" (Larson, 2010); "positive perceptions" (Gupta & Singhal, 2004); "stress-related growth" (Finzi-Dottan et al., 2011); "benefit finding" (Kim, Schulz, & Carver, 2007) and caregiving "uplifts" (Pinquart & Sörensen, 2003). For the purpose of the present work, the importance of negative and positive dimensions of adaptation processes related to family caregiving was assumed as "complementary", in order to substantiate a comprehensive and integrative research perspective that was endorsed as preferable. Partially given the recency of such research trend, no studies were found in the published literature neither on the assessment of caregiving uplifts, nor on the examination of their occurrence with caregiving burdens in parents of children/adolescents with CP.

Family caregiving may be described as a main developmental context for both parents and their child with a chronic physical condition (Barakat & Linley, 1992; Carter, Briggs-Gowan, & Davis, 2004). The study of parent-child adaptation outcomes and process is likely to turn out complex, and hence the relevance of studying patterns and linkages among variables has been stressed (Britner et al., 2003) as a mean of overcoming the inadequacy of main effects models to examine adaptation outcomes and mechanisms in those contexts (Button et al., 2001; Drotar, 1997). Nevertheless, the examination of such parental adaptation mechanisms has been seldom addressed in psychosocial research for pediatric CP.

In the context of parenting children with chronic physical conditions, parents' adjustment and parenting behaviors have been suggested to influence their child's developmental outcomes (Barakat & Linley, 1992; Garner et al., 2011; Wang & Jong, 2004). Complementarily, it has been reported that the adjustment of parents of children with CP may be significantly influenced by their child's behavioral adjustment (Majnemer et al., 2012; Raina et al., 2005; Romeo et al., 2010). Taken altogether, the available evidence calls for the integration of a transactional perspective to pediatric psychology research (Fiese & Sameroff, 1989), where both parents and their children are active agents in the establishment of mutual dynamics through which development occurs (Sameroff, 2009). In order to successfully apply a transactional perspective to the examination of parent-child adaptation processes, longitudinal research designs are imperative. In fact, the conduction of longitudinal research has been a common recommendation for future research with parents and families of children/adolescents with CP (Magill-Evans et al., 2001), with some

authors underlining the central tenet that "adaptation is not a single event but a multifactorial determined process over time" (Rentinck et al., 2006, p. 168).

Finally, the question of whether there are distinct patterns of adaptation for parents and families of children/adolescents with and without CP, remains to be ascertained (Britner et al., 2003). This question goes far beyond the aforementioned "two-way mentality", in the sense that contrasting adaptation mechanisms in both clinical and healthy controls samples enables the detection of possible specificities but also, and perhaps more importantly, the acknowledgement of common adaptation and development dynamics that may be essential to target in psychosocial intervention. Despite the fact that some "classic" pediatric psychology research studied the (in)variance of family adaptation mechanisms between clinical and control samples (e.g., Daniels et al., 1987; Quittner et al., 1990), this specific research topic remains to be further explored in the context of pediatric CP.

2 RESEARCH AIMS AND METHODOLOGY

Research aims and rationale

Research design

Participants

Instruments

Ethical considerations

Statistical analysis

A subsequent part of the present dissertation integrates four empirical studies, which represent the main outcomes from the research project that was developed during the period of nearly five years. This research project was aimed at deepening the existing knowledge on the psychosocial adaptation of children and adolescents with CP and their parents, while taking into account a developmental approach to pediatric HRQL assessment. The project was carried out in the context of the research work that is undertaken in the Institute of Cognitive Psychology, Vocational and Social Development (within the specific research line of "Relationships, Development, & Health"), at the Faculty of Psychology and Education Sciences of Coimbra University (*vide*: www.gaius.fpce.uc.pt/saude).

Along with a preliminary study on the cross-cultural adaptation of pediatric HRQL instruments (Carona et al., 2011), which has been published elsewhere and is here presented as a complementary attachment to the current dissertation, four empirical studies, written in article format, substantiate the empirical component of the dissertation (cf. "Introduction" section). Notwithstanding the thematic coherence that brings the different articles together under the same overarching topic, these empirical studies were submitted to different international journals and thus written and formatted accordingly. Moreover, sampling frames and analytic procedures were sometimes rather distinct from one study to another.

For these reasons, a brief introduction on the overall research aims and methodology was considered worthwhile, in order to acquaint the reader with the broader methodological framework that sustained all the research conducted. More specifically, this section was by no means intended to merely replicate information from the aforementioned articles, but instead to provide additional information on methodological choices and linkages within the sequence of different empirical studies.

1. Research Aims and Rationale

In the first part of the dissertation, a succinct literature review provided a depiction of the scientific state-of-the-art on the topic of psychosocial adaptation of children/adolescents with CP and their parents. From the literature review performed, two general remarks plainly emerged as noteworthy: first, research on psychosocial themes related to pediatric CP is rather recent and their study is still in its "infancy"; and second, there is a paucity of data on the assessment of a

broader range of individual and family adaptation outcomes, as well as on the exploration of determinants and their potential mechanisms of influence. Therefore, a number of **critical research questions and gaps** were identified as follows:

(1) The Disabkids questionnaires (The Disabkids Group, 2006) were world-renowned instruments for pediatric HRQL assessment, but their European Portuguese versions had not been yet developed;

(2) As many others pediatric HRQL questionnaires, despite the fact that Disabkids Generic Module (i.e. "Disabkids-37") was suitable for children and adolescents, the study of its psychometric properties by age-stratified groups had been reported infrequently;

(3) The psychological (mal)adjustment of children and adolescents with CP has been a neglected topic in pediatric psychology research, and most of the (few) available evidence on the topic came from studies that were conducted during the last couple of years;

(4) Regarding outcomes assessment in pediatric CP, most studies heavily relied on proxies (mostly parents) to obtain child-related information. In addition, age group differences (children vs. adolescents) were rarely examined, and the use of norms was far more the rule than the exception for the comparison and analysis of those scores;

(5) The assessment of social support, along with the examination of its determinant role on adaptation outcomes, had not been adequately addressed for children and adolescents with CP;

(6) Apart from anecdotal evidence, the nature and the influence of positive caregiving dimensions on adaptation outcomes remained unexamined for parents who have children/adolescents with CP; moreover, a multidimensional assessment of caregiving burden had been hardly ever adopted in previous studies with this population;

(7) Potential mechanisms via which caregiving variables could be linked to these parents' adaptation outcomes had not been explored; complementarily, age groups differences (childhood vs. adolescence) in parental adaptation outcomes and mechanisms had been studied infrequently;

(8) A parent-child dyadic perspective was seldom applied to pediatric psychology research conducted for CP, and thus, the examination of parent-child adaptation mechanisms, based on the integration of child and parent-reported data, was notably rare;

(9) Despite the suggestion that more similarities than differences may exist between families with and without individuals with CP, the assessment of the (in)variance of adaptation mechanisms between CP and non-CP samples has been a neglected topic in research.

Given the recency and paucity of research on the topic of psychosocial adaptation of children/adolescents with CP and their parents, the original research to be conducted was best understood as a pertinent and updated contribution, which was aimed at increasing knowledge on the research questions and gaps just outlined. In order to clarify the correspondence between those gaps and the research work performed, a brief comment on the **contributions** brought by the empirical studies developed is now presented:

- The development and validation of the European Portuguese versions of DISABKIDS-37 questionnaires was described in the first two empirical studies: the issues of instrument cross-cultural adaptation and preliminary psychometric assessment were explored in a first paper that is now presented as an attachment to the dissertation; the in-depth examination of DISABKIDS-37 psychometric properties using age group stratifications was accomplished in the empirical Study I;

- The need for broadening the scope of outcomes assessment in children and adolescents with CP was met in Study II, where specific negative outcomes (i.e., dimensions of psychological maladjustment) were analyzed in association with general HRQL outcomes, and in Study IV, where the mediating role of social support between parents' caregiving burden and their children's adaptation outcomes was examined in complementary models for psychological maladjustment and QL outcomes;

- The associations between social support and adaptation outcomes in children/adolescents with CP were explored in Study I, where specific psychopathological dimensions (i.e., internalizing and externalizing problems) were tested as potential mediators between these children/adolescents' social support and their HRQL outcomes;

- As regards the adaptation of parents of children/adolescents with CP, the adoption of a multidimensional assessment of caregiving burden and the integration of positive

dimensions (i.e., caregiving uplifts) in the examination of their caregiving experience were substantiated in Study III;

- In Study IV, the examination of a "social support deterioration model" (i.e., a mediating effect of social support between parents' caregiving burden and parent-child adaptation outcomes) was conducted within a parent-child dyadic perspective. In addition, the invariance of such potential adaptation mechanisms was ascertained between CP and non-CP samples;

- In those studies that targeted children and adolescents with CP (Study II and Study IV), there was a genuine concern in "hearing their voices" through the inclusion of their self-reports; notwithstanding, a dyadic approach to pediatric outcomes assessment (including both parent and child-reported outcomes) was complementarily adopted in Study II;

- In order to characterize a series of adaptation variables in children and adolescents with CP and their parents (Study II and Study III, respectively), comparisons of those markers were systematically performed with control samples and not with norms;

- Developmental differences and specificities between childhood and adolescence age groups were methodically addressed in all the empirical studies developed: in Study I, psychometric results were presented using age-group stratifications; in Study II and Study IV, age group was tested as a moderator within the examined models; and in Study III, age group was entered as covariate in the analyses performed.

Taken altogether, the outcomes from those empirical studies were expected to provide innovative insights as well as additional research questions to be explored in future investigations. In order to do so, general and specific aims were thoroughly outlined during the initial phase of the research process, as stated in Table 4.

Table 4. Main research aims

Empirical Studies	Aims
Preliminary Study	A. To develop the Portuguese versions of Disabkids-37 questionnaires and to ensure their cross-cultural comparability: a. To validate semantically the Portuguese versions of Disabkids-37; b. To explore the psychometric performance of Disabkids-37 in a pilot study.
I	B. To evaluate the psychometric properties of the Portuguese versions of Disabkids-37 within a developmental perspective: a. To assess the reliability and the convergent, divergent and discriminant validities of Disabkids-37 in a Portuguese sample; b. To ascertain the psychometric adequacy of Disabkids-37 for HRQL assessment in children, adolescents and mixed samples.
II	C. To examine the links between social support and adaptation outcomes in children and adolescents with CP: a. To characterize the social support and psychological maladjustment of children/adolescents with CP; b. To test the mediation effect of psychological maladjustment (i.e. internalizing and externalizing problems) on the link between social support and HRQL, and to examine the moderating role of age and gender within the hypothesized model.
III	D. To understand the nature and impact of the caregiving experience in parents of children/adolescents with CP: a. To characterize the QL and the caregiving burden and uplifts experienced by parents of children/adolescents with CP; b. To analyze the associations between caregiving burden and uplifts and the QL of these parents; c. To examine the moderating role of caregiving uplifts in the associations between burden dimensions and QL domains.
IV	E. To determine the (in)variance of hypothesized parent-child adaptation mechanisms in clinical and healthy samples. a. To examine the direct and indirect effects, via social support, of caregiving burden on the adaptation outcomes (i.e. psychological maladjustment and QL) of children/adolescents with CP and their parents; b. To assess the invariance of such models in clinical vs. healthy subsamples.

2. Research Design

All the studies integrated in the present dissertation were **cross-sectional** in nature. This design option was firmed on a two-folded methodological reasoning: first, two studies were primarily aimed at analyzing the psychometric properties of a pediatric HRQL instrument, and this could be accurately (though not exhaustively) achieved using a cross-sectional design (cf.

Petersen et al., 2005; Simeoni et al., 2007); and second, given the considerable recency of research, aside with the scarcity of available data on the core research topic, a cross-sectional design was considered a cost-effective methodological option to examine the prevalence of certain phenomena and generate critical evidence to be further explored in future studies (Ebrahim & Sullivan, 1995).

3. Participants

The empirical studies for the present dissertation were sequentially developed along two main phases: the first phase occurred between 2008 and 2010 and was aimed at the development and validation of the Portuguese versions of Disabkids (generic module) questionnaires; the second phase, mostly executed during the years of 2010 and 2011, was intended at the conduction of a study on the psychosocial adaptation of children and adolescents with CP and their parents. Altogether, five articles resulted from the research performed: the first two articles were related to the first phase of the research project, and dealt with the issues of cross-cultural instrument adaptation and the examination of the developmental approach to HRQL assessment, as proposed by the Disabkids European project; the remaining three articles were based on the work accomplished during the second phase of the research project, and specifically approached the themes of child/adolescent, parent and parent-child adaptation outcomes and potential mechanisms in the context of pediatric CP.

Despite the fact that sampling procedures were logically described in the methodological sections of the aforementioned research articles, the precise samples for each one of those studies varied significantly. During the first research phase, pilot and field validation studies of Disabkids questionnaires required distinct sampling frames; additionally, final sample sizes slightly differed between the three studies that were based on the second phase of the research project, as implied by random variations in the completion of the intended measures for each one of those empirical studies. Taken this sampling variability into account, an overall comment on the global sampling process was considered worthwhile.

The Disabkids Project in Portugal – Sampling Frames

The Disabkids European project was initially aimed at the development of standardized measures to assess HRQL in pediatric populations. The Disabkids instruments were originally

developed within a simultaneous approach (i.e., different countries participating simultaneously in the development of an instrument) (The Disabkids Group, 2006), which was described as a pioneer methodological initiative in pediatric settings (Petersen et al., 2005). Subsequently, the need for developing additional language versions (i.e., within a sequential approach of adapting and validating an existing measure from one language/cultural context to another), such as the Mexican (Medina-Castro, 2007), the Brazilian (Fegadolli, Reis, Martins, Bullinger, & Santos, 2010), the Swedish (Chaplin, Hallman, Nilsson, & Lindblad, 2011) and the Portuguese versions (Carona et al., 2011), led the coordination of the European Disabkids Group to establish guidelines for instrument translation and validation in those situations (The Disabkids Group, 2004). As a matter of fact, these procedures were in agreement with well-established guidelines for **instrument cross-cultural adaptation**, namely HRQL questionnaires (Guillemin, Bombardier, & Beaton, 1993; Hambleton, 2005; Schmidt & Bullinger, 2003).

According to those guidelines, translation procedures should conform to a sequence of (1) obtaining two independent (forward) translations, (2) conciliating both translations into a single (forward) translation, (3) providing a backward translation (performed by a third translator), (4) confronting forward and backward translations, (5) and conducting a first harmonization on problematic items. Following the translation procedures, the process of psychometric validation was organized along three additional phases: first, the conduction of a semantic and pilot validation study (in order to ensure cross-cultural and conceptual equivalence); second, the participation in a subsequent international harmonization of items; and third and last, the development of a field study to comprehensively assess the instruments' psychometric properties (The Disabkids Group, 2004).

As regards the sampling frames for the phases of semantic/pilot validation and field validation studies, two general requirements were established by the European Disabkids Group: first, a minimum of 36, and of 200 children/adolescents (and their parents) were to be included in the samples for the semantic/pilot and the field validation studies, respectively; and second, half of the sample should include cases of asthma, since this was the pediatric condition to be commonly examined by all the participating countries (The Disabkids Group, 2004). As for the case of Portugal, epilepsy was elected as the second pediatric condition to be included in the samples for validation studies, due to three main reasons: first, children/adolescents with chronic conditions affecting the central nervous system, such as epilepsy, had been reported to present an increased risk for psychological maladjustment, in comparison to children/adolescents with other chronic health conditions (CCD & CPACFH, 1993); second, the conditions of asthma and

epilepsy had been commented to share important clinical commonalities, such as the occurrence of unpredictable crises and the regular medication intake and visits to a physician; third and last, neither asthma nor epilepsy had any outwardly perceivable physical deformity (Austin, Smith, Risinger, & McNelis, 1994).

For the **semantic and pilot validation study**, a convenience sample of 36 children (8-12 years) and adolescents (13-18 years) was collected between December 2008 and March 2009, at the outpatient services of Immunoallergology and Neurology/Neuropediatrics of Coimbra University Hospitals and Pediatric Hospital of Coimbra Central Hospital, in compliance with three inclusion criteria: (1) chronological age between 8 and 18 years; (2) a clinical diagnosis of asthma or epilepsy (according to the ICD-10); and (3) illness duration of at least one year. Complementarily, subjects were to be excluded if they met any of the following criteria: (1) presence of developmental delay (including inability to understand questions, assess thoughts and emotions), as indicated by their physician; (2) severe psychiatric comorbid disorder, as indicated by their physicians, who knew the patients' clinical history; (3) clinical comorbidity of asthma and epilepsy (for the purpose of allowing reliable assessment of instrument discriminant validity, based on diagnosis). For parents, no specific inclusion/exclusion criteria were outlined in addition to the fact that the parent who accompanied the child/adolescent at the time of the consultations, was systematically included as proxy. For each chronic condition (asthma and epilepsy), a group of 9 children and a group of 9 adolescents (and their parents) was enlisted, thus achieving a total sample size of 72 participants (36 children/adolescents with chronic conditions and their parents). It is interesting to note that, in the case of the Portuguese Disabkids Group, an additional sample of 18 teachers with experience in teaching youths with chronic physical conditions was collected for semantic validation studies, as recommended by the national expert who participated in the revision of forward and backward translations. This particular sample was collected at Coimbra Cerebral Palsy Association, between October and November 2008.

For the **field validation study**, a larger convenience sample of 349 children/adolescents with chronic conditions (and their parents) was collected at Immunoallergology and Neurology/Neuropediatric outpatient services of Coimbra University Hospitals, Pediatric Hospital of Coimbra Central Hospital, Garcia de Orta Hospital (Almada) and Leiria Santo Andre Hospital, between March 2009 and December 2011. Inclusion and exclusion criteria were quite similar to the ones outlined for the phase of semantic validation, apart from the fact that regular medication intake was specifically added as inclusion criterion, for the sake of allowing the systematic conduction of analyses with the "Treatment" facet contained in Disabkids-37 questionnaires. In agreement with the requisites posed by the European Disabkids Group, a final

sample of 266 children/adolescents with asthma (and their parents) and 83 children/adolescents with epilepsy (and their parents) was attained.

Pediatric CP Sample

The clinical sample utilized for the development of three empirical studies on the core topic of parent-child psychosocial adaptation in the context of pediatric CP, was collected in different Portuguese Cerebral Palsy Associations (non-profit social organizations for tertiary healthcare), between July 2010 and July 2011. Although most cases were recruited at Coimbra Cerebral Palsy Association (n = 62), the remaining cases were collected in different locations throughout the national territory, namely in Vila Real (n = 6), Viana do Castelo (n = 4), Oporto (n = 8), Guimarães (n = 4), Leiria (n = 4), Viseu (n = 6), Almada/Seixal (n = 4), Beja (n = 2), and Faro (n = 5).

Children/adolescents were enlisted in the clinical sample if they met the following criteria: (1) diagnosis of CP, established by a physician (according to the ICD-10); (2) chronological age between 8 and 18 years old; and (3) absence of mental retardation, as indicated by a minimum intelligence quotient (IQ) of 70. Cases where results from formal IQ assessments were not available (n = 13), were still included if they were assessed as having no significant developmental delay, as suggested by gross evaluation of their cognitive abilities and the absence of previous adaptations to school curricula. The consideration of an inclusion criterion based on children/adolescents' intellectual functioning was a methodological option (or precaution) justified by the study's research design on the one hand, which emphasized the analysis of children/adolescents' self-reports, and by recent findings in literature on the other hand, which had reported a significant proportion of pediatric CP cases where the attainment of self-reports was not a feasible option (Arnaud et al., 2008; Varni et al., 2005).

Given the interest of this research project in analyzing parent-child dyads, and following the primary inclusion criteria defined for children/adolescents, parents were enrolled in this clinical sample if they met a single criterion: being the primary family caregiver of the child/adolescent with CP. The conformity to this criterion was subjectively assessed by parents themselves, who were asked to indicate who the primary caregiver was, as suggested by the attribution of the largest amount of daily time dedicated to child's health issues and care. In those cases where child informal healthcare was perceived as equally distributed between both parents,

the one who accompanied the child at the time of the assessment protocol administration was directly included.

According to the abovementioned inclusion criteria, 161 parent-child dyads were initially assigned to participate in the study. Subsequently, 56 of those cases were eliminated for a variety of reasons: seven refused to participate; forty-seven cases did not visit the institution during the established period for assessment protocol administration; and two cases related to children living in foster care placement. As a result, a final sample of 105 dyads of children/adolescents with CP and their parents was obtained.

Sample of Controls

Given the fact that many research objectives were delineated for comparing and contrasting adaptation outcomes and potential mechanisms between clinical and non-clinical groups, a sample of controls was composed. This convenience sample of controls was collected in two public schools of Coimbra district, between January and June 2010. Children and adolescents were enlisted for the study if they fulfilled two criteria: aged between 8 and 18 years old, and reporting no chronic physical condition. For their parents, a single inclusion criterion was required: to be the parent who spent more daily time with the child/adolescent. As for the clinical sample, in those situations where parents spent equal amounts of daily time with their children, the selection of an information source was indiscriminate.

In order to achieve the intended sample size, a total of 124 parent-child dyads that complied with the aforementioned criteria, were assigned to participate in the research project. Subsequently, six cases were excluded: two parents refused to participate in the study and four adolescents did not return their parents' questionnaires. Therefore, the final sample of controls was composed by 118 parent-child dyads.

As a complementary remark, it should be acknowledged that the labeling of this control group was not straightforward or definite throughout the work. This vacillation was most certainly influenced by the continuing debate on the terminological labels that may be applied to describe CP (as it was discussed in the first part of the dissertation). Nevertheless, as suggested by a number of different authors and previous research works, different expressions were reiterated throughout the dissertation and these included: "typically developing" (cf. Crnic, Hoffman, Gaze,

& Edelbrock, 2004; Russo et al., 2008), "able-bodied" (cf. Parkes et al., 2008), or (physically) "healthy" (Silver, Westbrook, & Stein, 1998) children/adolescents.

As commented earlier, this section was intended to provide a general overview of the more global sampling frames underlying the different empirical studies integrated in the present dissertation. Complementarily to this overview, detailed sample characterizations and sampling procedures were described within each one of the empirical studies presented. Nevertheless, Table 5 is included here as a concluding illustration for the overall dimension of the final sample obtained.

Table 5. Description and dimension of the samples used in the empirical studies

Research Project Phases	Empirical Studies	Sample Size Description	*N*
Disabkids Semantic Validation Study	*Preliminary Study*	18 children/adolescents with asthma 18 children/adolescents with epilepsy 36 parents 18 teachers	90
Disabkids Field Validation Study	*Study I*	266 children/adolescents with asthma 83 children/adolescents with epilepsy 349 parents	698
Study on Parent-Child Psychosocial Adaptation	*Studies II, III, IV*	105 children/adolescents with CP 118 healthy children/adolescents 223 parents	446
			1234

4. Instruments

In order to assess the different variables that were targeted in the sequence of empirical studies, a variety of measures was compiled in a general assessment protocol. Since the

delineation of more specific assessment protocols was aligned with each study's aims, and was thus detailed in the respective section of the empirical studies, the purpose of the current section was thought to provide a rationale on the selection of those instruments, which were all contained in the definition of the overall assessment protocol utilized for the development of the research project.

The use of questionnaires was clearly preferred as a cheap and practical method to obtain general pictures on different variables and dimensions of parent-child adaptation. This methodological option was nevertheless informed about its inherent problems and limitations, namely: (1) the risk for memory or response-bias errors; (2) the restrictions imposed by the specificity of scaling dimensions; (3) the requirement of certain language aptitudes; and not least important, (4) the fact that the completion of questionnaires could be regarded as tedious by some respondents (Cummings et al., 2000). Bearing these limitations in mind, the process of instrument selection was based on four methodological assumptions. First, a complementary assessment of negative and positive dimensions of adaptation was systematically adopted to broaden the scope of outcomes and determinants evaluation, as recommended in the recent literature (e.g., Barlow & Ellard, 2006; Vargus-Adams & Martin, 2009). Second, the valorization of a developmental perspective to outcomes and potential mechanisms examination, implied the incorporation of instruments that should be easy and brief to administer, allow self and proxy-reports whenever possible, and enable the conduction of comparisons between distinct age-groups (children vs. adolescents). Third, the emphasis placed on the mutual relationships between context and adaptation outcomes (Boyce et al., 1998; Kazak, 1997; Liptak & Accardo, 2004), called for the inclusion of measures on specific aspects of parent and child's social developmental contexts. Fourth and last, the research objective of examining potential parent-child adaptation mechanisms required the inclusion of measures that could promote coherence in the operationalization of child and parent's adaptation levels within a dyadic perspective.

In the next subsections, the constitution of the assessment protocol is commented with the description of the adopted measures and the rationale for their inclusion in the research project.

Socio-demographic and Clinical Variables

Socio-demographic and clinical datasheets were used in all the empirical studies conducted, but some of their items were logically adjusted to the objectives of different research

phases. For children and adolescents, simple questions addressing age, gender and diagnosis were included in the introductory section of the Disabkids questionnaires. In addition, parents were asked about their child's comorbidities, psychiatric history, recent hospitalizations, and school absenteeism related to their condition. During the phase of Disabkids validation studies, parents were further asked about their child's symptom severity, with the inclusion of three questions adapted from the respective Disabkids condition-specific modules (cf. The Disabkids Group, 2006). For the study on pediatric CP, in particular, the child/adolescent's motor impairment level was determined by experienced physical therapists, who based those clinical judgments on the expanded and revised version of the gross motor function classification system (Palisano, Rosenbaum, Bartlett, & Livingston, 2007). Generally, parents were also asked about their own socio-demographic data, including their age, gender, marital status, family composition, job and educational level. In fact, following the classification system proposed by Simões (1994), these two latest variables were combined to determine parents' SES along three levels (Low, Medium, and High). Moreover, parents were briefly asked about their own psychiatric history, chronic health problems, multiple family caregiving, and satisfaction with healthcare providers. The selection of these socio-demographic and clinical variables was drawn from previous literature review in similar research topics.

QL and HRQL Outcomes

The **chronic generic module of Disabkids questionnaires** (**Disabkids-37**) was of foremost importance throughout the research project, and especially during its initial phases. Given our interest in assessing HRQL outcomes in an understudied population, the selection of a reliable and valid measure was crucial. At that time, the worldwide dissemination of the instruments developed by Kidscreen and Disabkids projects attested their success in substantiating a European perspective to children/adolescents QL and HRQL assessment. Moreover, both projects had been developed within a theoretical and methodological matrix that was closely related to the paradigmatic WHOQOL initiative (Bullinger et al., 2002; Ravens-Sieberer et al., 2001). Finally, while the Kidscreen instruments had been already meritoriously adapted to Portuguese language and culture (cf. Gaspar & Matos, 2008), the equivalent task had not been conducted for the Disabkids questionnaires. Notwithstanding some theoretical criticisms directed at the concept of HRQL (Wallander et al., 2001), Disabkids-37 questionnaires were valued as fundamental specific assessments on those aspects of well-being directly related to limitations and treatment imposed by a chronic physical condition. This is to say that, in the

context of the present work, the importance of QL and HRQL outcomes and measures were systematically assumed as complementary to each other. Disabkids-37 assesses pediatric HRQL, as perceived by children/adolescents and/or their parents (or other proxies), through 37 items that are to be answered within a 5 point Likert scale ranging from 1 (*Never*) to 5 (*Always*). The statistical syntax for the instrument enables the computation of an overall standardized score (0-100), where lower values are indicative of a more impaired HRQL. However, despite the fact that the computation of a Treatment/Medication facet was available for Disabkids-37, given the relatively few medicated cases in our CP sample, Disabkids-37 also enabled the computation of a global score without the inclusion of that specific facet. In addition, Disabkids-37 questionnaires had incorporated a developmental approach to HRQL assessment and exhibited sound psychometric properties in validation studies that were based on heterogeneous pediatric samples, including CP cases (Petersen et al., 2005; Simeoni et al., 2007; The Disabkids Group, 2006). More recently and more specifically, some authors have commented the psychometric adequacy of Disabkids questionnaires for the assessment of QL outcomes in children/adolescents with CP (Viehweger et al., 2008).

For children and adolescents with CP, HRQL was described as "the subset of QL directly related to an individual's health" (Bjornson & McLaughlin, 2001, p. 183). The selection of a general QL instrument for children and adolescents was needed because the research project was not only interested in examining the convergent validity of the Disabkids-37 questionnaires, but also in contrasting QL outcomes between pediatric and healthy samples. As regards the validation of Disabkids-37 questionnaires, one should note that the assessment protocol designed for that phase of the research project was agreed with and approved by the coordination of the European Disabkids Group. In fact, since Disabkids and Kidscreen projects shared the so-called European perspective in children/adolescents' QL assessment, there was a pertinent interest in combining them in research. Despite the fact that QL was thoroughly defined as a multidimensional concept and construct, two main advantages underlay the adoption of **Kidscreen-10** as a unidimensional measure: first, there was a great concern in avoiding extensive assessment protocols for children, in order to avoid respondent fatigue bias, and second, the determination of a single unified score enabled the simplification of results' description and discussion. Moreover, Kidscreen-10 integrated selected items on a variety of dimensions, including the individual's physical activity, energy and fitness; depressive moods and stressful feelings; opportunities to participate in leisure time and social activities; feelings toward parents; relationships with other children; and cognitive ability and satisfaction with school achievement (Ravens-Sieberer et al., 2010). Kidscreen-10 questionnaires are available in self and proxy-report

forms and include 10 items, which are to be completed within a 5-point response scale, ranging from 1 (*Not at all/Never*) to 5 (*Extremely/Always*). The instrument then allows the calculation of a standardized score (0-100), with higher values indicating more positive QL perceptions. As regards its psychometrics, there were favorable data suggesting and demonstrating the psychometric quality of Kidscreen instruments: in Portugal, good indexes of reliability and construct validity had been reported for the longest version of Kidscreen questionnaires (Gaspar & Matos, 2008), and in an European study, those same psychometric properties were confirmed for Kidscreen-10 questionnaires (Ravens-Sieberer et al., 2010). Finally, the appropriateness of Kidscreen-10 for QL assessment in children and adolescents with neurodisabilities, namely CP, has been commented in recent literature (Davis, Shelly, Waters, & Davern, 2010; Waters et al., 2009).

The WHO approach to QL assessment was described in the introductory section as a general methodological and conceptual framework. The work developed by the WHO initiatives has established the multidimensionality and subjectivity of QL definition and operationalization, as well as the general guidelines for QL assessment in children (The WHOQOL Group, 1995; WHO, 1993). Therefore, the election of **WHOQOL-BREF** for parents' QL evaluation was rather immediate. The WHOQOL-BREF questionnaire was regarded as a sound operationalization of a multidimensional approach to adaptation outcomes assessment, because it addressed the physical, psychological, social and environmental domains related to health and well-being. The WHOQOL-BREF contains 26 items, which are to be answered within a 5-point scale ranging from 1 (*very poor/very dissatisfied/not at all/never*) to 5 (*very good/very satisfied/extremely/completely*); standardized scores (0–100) for each domain may then be computed, with the lowest scores depicting the most impaired QL. The European Portuguese version of WHOQOL-BREF had demonstrated good psychometric properties (Vaz-Serra et al., 2006), and quite recently, the instrument was used in at least two studies on the QL of parents of children with CP (Okurowska–Zawada et al., 2011; Romeo et al., 2010).

Nevertheless, in one particular empirical study, there was an interest in obtaining a single overall score for children and their parents' QL. Since the WHOQOL-BREF could not provide an overall score derived from its domains, the WHOQOL 8-item index – **EUROHIS-QOL** – was preferred at that point. This was a screening measure derived from the WHOQOL-100 and the WHOQOL-BREF instruments, which included two items to assess each of four QL domains (i.e., Physical, Psychological, Social, and Environmental) (Power, 2003). During instrument administration, those items are answered on a 5-point response format ranging from 1 (*Very poor/Very dissatisfied/Not at all/Never*) to 5 (*Very good/Very satisfied/Extremely/Completely*), thus

permitting the subsequent computation of an overall standardized score (0-100), with higher scores representing more positive QL perceptions. The reliability and validity of EUROHIS-QOL has been demonstrated in a cross-cultural field study (Schmidt, Mühlan, & Power, 2005), and in a psychometric study for the European Portuguese version (Pereira, Melo, Gameiro, & Canavarro, 2011).

Psychological (Mal)Adjustment

Contrasting with the relative recency of research on the psychological (mal)adjustment of children and adolescents with CP, an established measure was used to target that variable in our studies. The **Strengths and Difficulties Questionnaire** (**SDQ**) was adapted to the Portuguese language and population (Fleitlich, Loureiro, Fonseca, & Gaspar, 2005) and has been described as a brief, valid and reliable measure to assess prosocial behavior and psychopathology in children and adolescents (Goodman, 2001). This instrument was included in different phases of the research project, both as a mean of examining the divergent validity of Disabkids-37 questionnaires, as well as a measure on specific adaptation outcomes, namely psychopathology or psychological (mal)adjustment. In fact, the assessment of psychopathology in individuals with developmental disabilities has been strongly encouraged, because of its higher prevalence and its underdiagnosis in this population (Rush, Bowman, Eidman, Toole, & Mortenson, 2004). As for all measures on children and adolescents' adaptation outcomes (i.e., QL and HRQL outcomes), the SDQ permitted the administration of both self and proxy-report forms, and this was aligned with the recommendation of adopting multi-informant methods in pediatric psychology research (Holmbeck et al., 2008). Apart from the prosocial behavior subscale (which was not used in this research project), SDQ provides specific scores (that can be computed into a single, overall score) on the subscales of emotional symptoms, peers problems, conduct problems, and hyperactivity-inattention symptoms. SDQ items on those subscales were answered by participants within a 3-point Likert scale: 0 (*Not true*); 1 (*Somewhat true*) and 2 (*Certainly true*). Then, overall sum or mean scores were computed, and higher scores interpreted as indicative of greater psychological maladjustment. In addition to the overall score, an alternative scaling was utilized in one of the empirical studies developed. This scaling has been recommended for low-risk samples (in contrast with screening for psychological disorders) and essentially integrated the subscales of emotional symptoms and peers problems, on the one hand, and the subscales of conduct problems and hyperactivity-inattention symptoms, on the other hand, into broader subscales of internalizing and externalizing problems, respectively (Goodman, Lamping, & Ploubidis, 2010).

This alternative scaling enabled the simultaneous assessment of both psychopathological dimensions, thus substantiating the methodological guideline of looking beyond the internalizing dimension, to encompass the externalizing dimension, which has been hypothesized to play a distinct and important role in pediatric populations (Holmbeck et al., 2008). The inclusion of SDQ in the assessment protocol for this research project has been additionally validated by its inclusion in a number of recent studies describing the psychological problems of children and adolescents with CP (Brossard-Racine et al., 2012a; Brossard-Racine et al., 2012b; Parkes et al., 2008).

The brief 5-item form of the **Mental Health Inventory** (**MHI-5**) (Ware, Snow, Kosinski, & Gandek, 1993, as cited in Pais-Ribeiro, 2001, p. 86) was used in one empirical study to assess parents' psychological maladjustment. This screening measure had been adapted to the Portuguese population and commented as a valid and reliable substitute of its longer form in research settings (Pais-Ribeiro, 2001). Although the instrument also contained a subscale on positive well-being, the psychological distress subscale (encompassing items on depressive and anxious symptomatology) was utilized to concisely measure parents' psychological maladjustment. The MHI-5 items are to be completed within a 6 point response scale, ranging from 1 (*Never*) to 6 (*Always*), in order to subsequently allow the calculation of a generic score on adults' psychological maladjustment. Given its original applications in community population-based samples, the instrument content was plainly adequate for use in clinical and non-clinical samples.

Caregiving Burden and Uplifts

At the time of planning the assessment of caregiving-related variables in this research, two requirements became evident: first, the measure for parents' caregiving burden should be multidimensional, and second, that measurement should be complemented with the simultaneous or parallel assessment of positive caregiving dimensions. Given the fact that most burden literature had been developed from geriatric contexts (cf. Chou et al., 2003; Sales, 2003), a measure with those required characteristics was eventually found in that same context. The **Revised Burden Measure** (Montgomery et al., 2006) not only performed a multidimensional assessment on the objective, subjective and interpersonal aspects of caregiving burden, but also provided a subscale on caregiving uplifts. The instrument items are to be responded within a 5 point scale (1 = *Not at all*; 5 = *A great deal*); items pertaining to the objective, subjective and relationship burden dimensions may then be combined to provide an overall sum or mean score

on caregiving burden. During the process of instrument selection, a detailed revision of the items' content was carried out, in order to ensure its overall adequacy for administration in clinical pediatric and normative samples. Although the extensive validation of the Portuguese version of this instrument was not aimed for this research project, its translation adhered to very similar procedures to the ones outlined for Disabkids-37 cross-cultural adaptation. In addition, findings from preliminary psychometric studies (C. Carona, N. Silva, M. C. Canavarro, personal communication, July 27, 2011) revealed that the Portuguese version of the "Revised Burden Measure" was a valid and reliable instrument for use in pediatric settings.

Social Support

As commented earlier, this research project endorsed the recommendation of examining the subjective appraisals of social support (not social support networks, not supportive behaviors) in relation to well-being (Vaux & Harrison, 1985). For that purpose, the **Satisfaction with Social Support Scale for Children and Adolescents** (Gaspar, Pais-Ribeiro, Matos, Leal, & Ferreira, 2009) was straightforwardly selected for three essential reasons: first, it had adequate wording and enabled the simultaneous assessment of social support satisfaction in children and adolescents; second, it represented a reliable adaptation of the equivalent adult measure (i.e., "Satisfaction with Social Support Scale", described later), thus adding consistency to the measurement methodology used in the last empirical study with parent-child dyads; and third, evidence on the instrument validity had been documented (Gaspar et al., 2009). This social support measure includes 12 items, for which the child/adolescent indicates his degree of agreement along a five point Likert scale, ranging from 1 (*Totally disagree*) to 5 (*Totally agree*). Despite the fact that the instrument included two subscales, namely "Satisfaction with Social Support" and "Involvement in Social Activities", an overall score was computed in the studies that used this measure.

The selection of the **Satisfaction with Social Support Scale** (Pais-Ribeiro, 1999) to assess parents' social support was logically paired with the election of its child/adolescent version, which was commented before. This instrument comprises 15 items, which are to be answered within a 5-point scale (1 = *Totally disagree*; 5 = *Totally agree*). Although this questionnaire incorporated subscales on satisfaction with friendships, intimacy, satisfaction with family, and social activities, an overall mean score was used in the empirical study that employed this measure. In addition to the rationale described for the child/adolescent version of the

instrument, it should be noted that good validity and reliability indexes had been reported for the overall scale (Pais-Ribeiro, 1999).

Having described the constitution of the assessment protocol for research, a summary of studied variables, adopted instruments and their relation to the empirical studies, is presented in Table 6.

Table 6. Studied variables, adopted instruments and empirical studies integrated in the research project.

Variables	Instruments		Empirical Studies			
			I	II	III	IV
QL and HRQL Outcomes	KIDSCREEN-10 (Ravens-Sieberer et al., 2010; Gaspar & Matos, 2008)	*Self*	✓			✓
		Proxy	✓			
	WHOQOL-BREF (The WHOQOL Group, 1998; Vaz-Serra et al., 2006)				✓	
	EUROHIS-QOL (Schmidt et al., 2005; Pereira et al., 2011)					✓
	DISABKIDS-37 (The Disabkids Group, 2006; Carona et al., 2011)	*Self*	✓	✓		
		Proxy	✓	✓		
Psychological (Mal)Adjustment	SDQ (Goodman, 2001; Fleitlich et al., 2005)	*Self*	✓	✓		✓
		Proxy	✓	✓		
	MHI-5 (Ware et al., 1993; Pais-Ribeiro, 2001)					✓
Caregiving Burden and Uplifts	Revised Burden Measure (Montgomery et al., 2006; Carona et al., 2011)				✓	✓
Social Support	Satisfaction with Social Support Scale (Pais-Ribeiro, 1999)					✓
	Satisfaction with Social Support Scale for Children and Adolescents (Gaspar et al., 2009)			✓		✓

5. Ethical Considerations

The preparation and implementation of this research project continuously took into account the ethical guidelines and requisites established by The Declaration of Helsinki (World Medical Association, 2000) and the European Comission (Pauwels & European Comission, 2007) for the conduction of scientific research in human beings. The Declaration of Helsinki acknowledged the pertinence of developing scientific research with human beings, but also stressed that participants' well-being should prevail over any other interest. Notwithstanding the fact that The Declaration of Helsinki was first redacted for the regulation of scientific research in medical settings, it nevertheless suggested that research with human beings in any other areas of knowledge should meet the same ethical principles. In fact, the fundamental premises of The Declaration of Helsinki were integrated in the regulating guidelines published by the American Psychological Association (2010) and the Order of Portuguese Psychologists (Regulation number 258/2011, 20th April 2011).

Given the fact that this research project was directed at pediatric populations, additional ethical concerns were contemplated. In research contexts, children are assumed as a group of increased vulnerability, together with other clinical groups such as individuals with intellectual disability or severe psychiatric disorder. This position is logically implied by the inherent definition of **"vulnerability"** as the substantial inability to protect one's own interests (Vale, s.d.). The central tenet in communicating with children in research settings is that a child should never be treated as an adult, and thus his/her age, level of cognitive and moral development, and family, social and cultural contexts, should be genuinely taken into account (Vale & Oliveira, s.d.). Although any individual under the age of 18 is usually considered a minor in legal terms, the Portuguese Law determines that "consent" may be performed by any individual aged more than 14 years old and with the necessary discernment to assess the meaning and dimension of the consent provided. However, for children younger than 14 and older than 7 years, in addition to parents' consent, the child's assent should be complementarily obtained (Vale, s.d.). The consideration of the child's assent is no minor issue, since any refusal is to prevail in the assessment of discrepancies between consents and assents (Vale & Oliveira, s.d.).

As recommended by Jonas (1995), this research project, which was mainly developed with pediatric populations, sought to integrate reflective insights over the following issues: the balance between risks and benefits for participants; the simultaneous consideration of parental

informed consent and children's assent, where the latest was to prevail; and the development of research aims for increasing relevant knowledge on child's health. In sum, for the purpose of the present project, the researchers' nuclear concern was to listen to the children enrolled in the empirical studies, in a context of respect for a joint decision making between the child, his/her family, the health professionals, and the research team.

In the next paragraphs, several diligences related to the accomplishment of ethical requisites are commented within a research process perspective.

- During the preparation phase:

• *Approval of the research project by Ethics Committees or other competent organisms*: The research project and its associated assessment protocol were submitted to the appreciation, validation and approval by the Ethics Committees of all the hospital institutions that participated in the study. During the second phase of the research, the project was submitted to the Direction Board of the Portuguese Federation of Cerebral Palsy Associations and, afterwards, to the Direction Board of each Association selected to participate in the study. Authorizations for collecting the pediatric CP sample were obtained from all these institutions. As regards the collection of the sample of healthy controls, the research project was evaluated by the responsible committee at the participating public schools, and approved by the respective Direction Boards.

• *Assessment of potential risks and benefits related to the participation in the study*: This assessment was performed through literature revision and discussion with health professionals working in the pediatric field, as well as with families who had children with chronic physical conditions. During the definition of the assessment protocol, special caution was taken to avoid extensive, burdensome compilations of questionnaires, as well as the use of excessive negative wording, which may end up representing a potential threat to participants' self-esteem (cf. Waters et al., 2009).

- During the empirical research phase:

• *Protection of participants*: The protection of the individuals' life, health, dignity, self-determination and privacy was continuously advocated and ensured.

· *Guarantee of confidentiality*: This was best achieved through the attribution of a numeric code to the questionnaires collected, in order to prevent the identification of participants by third parties at any time. In those few cases where questionnaires were returned by mail, stamped envelopes with no sender's information were provided.

· *Assurance of voluntary participation*: Participants were informed that their participation was to be absolutely voluntary, and were reassured that their eventual decision on refusing or quitting the study would not, by any means, interfere with the healthcare provided to them.

· *Pertinence of the research objectives*: Research aims were defined with the main intention of gaining significant knowledge on neglected topics related to the pediatric populations under study. Subsequently, the means utilized to gather the necessary data were closely linked to those primary aims, thus avoiding the collection of redundant, superfluous data.

· *Avoidance of unnecessary constraints*: The research team placed a genuine effort in creating a cordial, supportive and friendly atmosphere during the administration of questionnaires. Although in some occasions the provision of a quiet and private place to questionnaire administration was not viable (these particular situations mostly occurred in some services of public hospital institutions), a great concern was shared by the research team to create the best possible conditions for the performance of that task. During the collection of the CP sample, assessment protocols were administered by health professionals (mostly psychologists and social workers acquainted with the research project), who usually worked with the participating children/adolescents and their parents, under the supervision of the research coordinator.

· *Informed consent:* All the participants were included in the research project following their respective informed consent. As pointed earlier, in young children, parents' informed consent was complemented with the child's assent; for adolescents older than 14, their own informed consent was always considered with their parents' assent (even if sometimes not written, nor legally required). The document for informed consent stated the research's aims and methodology and included the research coordinator's contact and institutional affiliation. This document stated the team's commitment to protect the confidentiality of participants' data, as well as their right to refuse or quit the participation in the study, without any implication on the healthcare provided to them. The information included in the informed consents was usually presented orally by the researcher, in order to ensure its full understanding by the subjects.

· *Information about the study's relevance and expected contributions*: Invitations to participate in the study were always accompanied by a simple statement on the importance of the individuals'

participation as well as on the expected impact of the research outcomes in the healthcare and lives of people facing similar situations.

- During the dissemination of results phase:

• *Objective and honest publication of results*: The publication of results was aimed at disseminating the study's contributions in a precise and detailed manner, and thus did not hinder the acknowledgement of limitations or the presentation of unexpected or inconsistent results. Funding sources and declarations of interest were systematically stated, and co-authorships were defined by a researcher's significant contribution during any phase of the project development.

• *Presentation and discussion of results with professionals:* Brief informative or training sessions were developed, mainly by the research coordinator, for the professional teams of all hospital institutions. During the first phase of the research project, for instance, those sessions were aimed at describing pediatric HRQL assessment, presenting psychometric data for the Portuguese versions of Disabkids-37 questionnaires, and reflecting on the clinical implications of such assessment procedures. Results from the second phase of the research project (i.e. study on pediatric CP) have been presented and discussed in national conferences for health and education professionals working in the area, as well as in brief training initiatives that have been mainly developed at Coimbra Cerebral Palsy Association.

6. Statistical Analysis

Statistical analysis procedures utilized throughout the research project were carefully detailed in the methodological sections of the empirical studies conducted. Nonetheless, the purpose of the present section was to briefly comment the selection of specific statistical approaches or techniques in relation to the broad scientific domain of pediatric psychology research. Specifically, three groups of general statistical procedures were commented in that regard, namely: (1) magnitude of effects; (2) mediation, moderation and moderated mediation; and (3) structural equation modeling (SEM).

The publication of **effect sizes** related to the discussion of main findings in psychological research has been strongly recommended (Wilkinson & Task Force on Statistical Inference of APA Board of Scientific Affairs, 1999). In fact, the calculation of such effect sizes is crucial to

determine the scope of core findings and the discussion of their applicability. Since psychological researchers are usually concerned about the assessment of between-groups differences or associations between variables, effect sizes are valuable indicators of "how much" difference may exist or "how strong" a given relationship may be (Durlak, 2009). In practical terms, the statistical significance (i.e., the *p*-value) of a difference or a correlation does not inform, *per se*, about the magnitude of such effects, for the reason that a small *p*-value may be related to a low, medium or high effect size (McCartney & Rosenthal, 2003; Vaughan, 2007; Volker, 2006). The report of effect sizes has been specifically required by prominent publications in the field of pediatric psychology research, where the utilization of small sample sizes is frequent and likely to affect the computation of such indices (Durlak, 2009). For the empirical studies developed in this research project, three effect size indices were mainly reported: Pearson's correlation coefficient (*r*); coefficient of determination (R^2); and partial Eta squared (λ_p^2) (i.e., the proportion of unexplained variance, after excluding the variance explained by other predictors, that is attributable to a given effect/predictor.).

Mediation analyses have been gaining increased attention in psychological research, because they are useful means of examining possible causal mechanisms in the decomposition of interesting associations, as well as of testing and developing psychological theory from a merely descriptive to a more functional understanding of the relationships between variables (Preacher & Hayes, 2004; Shrout & Bolger, 2002). **Moderation analyses**, on the other hand, are complementarily important, since they specify the factors that can affect the direction or strength of association between an independent or predictor variable and a dependent or criterion variable (Baron & Kenny, 1986). The classical approach to assess mediation effects with the Sobel test became widespread in psychological literature (Baron & Kenny, 1986), but was open to two main criticisms: first, it required very large samples, and second, it had low statistical power (MacKinnon, Lockwood, Hoffman, West, & Sheets, 2002). More recently, **bootstrapping procedures** have been approached as a preferred method to examine indirect effects. Bootstrapping is a nonparametric resampling procedure that does not impose the assumption of normality of the sampling distribution, and that essentially involves repeated sampling from the data set and estimating the indirect effect in each resampled data set (Preacher & Hayes, 2004). Through the repetition of this process thousands of time (e.g., 5000 bootstrapped samples were generally used in this dissertation), bootstrapping procedures have higher power and maintain reasonable control over Type-I error, through the appropriate construction of confidence intervals for the indirect effect(s) (Briggs, 2006). As regards mediation in particular, two recent methodological improvements are noteworthy: multiple mediation and moderated mediation.

Multiple mediation is particularly valuable to psychological research, which tends to offer more than one theoretical explanation for a given psychological phenomenon, and is also interested in examining competing theories within a single model (Preacher & Hayes, 2008). To put it concretely, multiple mediation enables the determination of the relative magnitudes of specific indirect effects associated with a number of different mediators entered in a single model. **Moderated mediation** (the so-called "conditional indirect effect"), on the other hand, aims at explaining how and when a given effect occurs, thus assessing the strength of a indirect effect as dependent on the levels of a fourth variable (i.e., the moderator) (Preacher, Rucker, & Hayes, 2007). The importance of examining mediation and moderation effects in child-clinical and pediatric psychology is well-established in the literature (Holmbeck, 1997, 2002b). To illustrate such importance, for example, it is worthy pointing out that the conduction of moderation analyses in pediatric psychology has led to a remarkable conceptual distinction between resource vs. protective factors, and vulnerability vs. risk factors to guide research in this field (Rose et al., 2004).

Finally, **SEM** has been described as a refinement of general linear modeling procedures, such as ANOVA and multiple regression analysis, which can be used to analyze the links between latent constructs that are specified by multiple measures, in both cross-sectional and longitudinal data (Lei & Wu, 2007). One of the advantages of SEM procedures is that this estimation of latent variables from indicators permits the examination of reasonably "error-free" constructs (Nelson, Aylward, & Steele, 2008). In SEM, the pattern of inter-relations between variables is specified a priori, and the subsequent goal is to ascertain whether such hypothesized model is consistent with the data collected to examine the theoretical proposal. That level of consistency is determined through "model-data fit", which can be deduced by a number of goodness-of-fit indicators that include the Normed Fit Index (NFI), Tucker-Lewis Index (TLI), Incremental Fit Index (IFI), Comparative Fit Index (CFI), and root mean square error of approximation (RMSEA) (MacCallum & Austin, 2000; Schreiber, Stage, King, Nora, & Barlow, 2006). SEM has been commented as a preferable approach to path analysis, but although the required sample size has been suggested to vary between a minimum of 100 and 200 individuals, it has also been acknowledged to depend on the model's complexity (Kline, 2005). When adopting SEM procedures within the present research work, the following recommendations were taken into account: (a) research questions suggested the use of SEM; (b) the models to be examined were theoretically grounded; and (c) graphic displays of the final models were provided (Schreiber et al., 2006). The issue of developing sound a theoretical framework and rationale was especially considered in the research conducted, thus endorsing the need to develop theory-driven research

in pediatric psychology (Kazak, 2002). For the context of the present dissertation, it is important to retain that SEM techniques have been described as innovative and sophisticated (though underutilized) statistical methodologies, which may be particularly useful in examining socio-ecological systems models in pediatric psychology (Nelson et al., 2008).

3 EMPIRICAL STUDIES

Empirical Study I
Examining a Developmental Approach to Health-related Quality of Life Assessment: Psychometric Analysis of DISABKIDS Generic Module in a Portuguese Sample

Empirical Study II
Social Support and Adaptation Outcomes in Children and Adolescents with Cerebral Palsy

Empirical Study III
The Disability Paradox Revisited: Quality of Life and Family Caregiving in Pediatric Cerebral Palsy

Empirical Study IV
Similarities Amid the Difference: Caregiving Burden and Adaptation Outcomes in Dyads of Parents and their Children with and without Cerebral Palsy

Empirical Study I |

Examining a Developmental Approach to Health-related Quality of Life Assessment: Psychometric Analysis of DISABKIDS Generic Module in a Portuguese Sample

Vulnerable Children & Youth Studies (2012), *iFirst Article*, 1-15

Examining a Developmental Approach to Health-related Quality of Life Assessment: Psychometric Analysis of DISABKIDS Generic Module in a Portuguese Sample

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Abstract

The aim of this study was to assess the properties of a generic instrument for pediatric health-related quality of life (HRQL) assessment – DISABKIDS-37 – in a sample of Portuguese children and adolescents with chronic health conditions. Participants were 349 children/adolescents with asthma or epilepsy and their parents/caregivers, who reported on children/adolescents' HRQL, along with generic quality of life and psychological adjustment measures in self and proxy-report formats. Using classical statistic validation procedures, reliability, scale inter-correlations, and convergent, divergent and discriminant validities were analyzed. Results were reported using age group stratifications, attesting the developmental appropriateness of DISABKIDS-37 questionnaire to assess HRQL in children, adolescents and mixed samples.

Keywords: health-related quality of life; children and adolescents; Disabkids; psychometrics.

Introduction

The changing epidemiology and clinical understanding of childhood health and disease, currently demands a continuous improvement on the conceptual and measurement issues of quality of life (QL) assessment (Eiser & Morse, 2001). HRQL is defined as a multidimensional construct covering physical, emotional, mental, social and behavioral components of well-being and function as perceived by patients or proxies (Bullinger, 1997), and is considered a component of the more general construct of QL (The WHOQOL Group, 1998). Despite the increasing number of QL and HRQL measures for pediatric populations, children and adolescents are usually taken as a single, unified developmental group. However, a substantial body of knowledge from interrelated disciplines, such as developmental psychopathology, developmental and pediatric psychology, assert that children and adolescents face specific developmental tasks and maturation issues, have distinct cognitive abilities, and use different coping strategies (Achenbach & Rescorla, 2006; Lerner, 1982; Spirito, Stark, Grace, & Stamoulis, 1991). Thus, an important research question in this field is to examine whether HRQL instruments that cover a wide age range, adequately and reliably assess the same construct in those two developmental groups.

The Need of Refining a Developmental Approach to HRQL Assessment

Due to a number of conceptual and methodological questions, QL assessment in children and adolescents has been a neglected topic for decades, comparatively to the amount of research published on the same issue for adults (Drotar, 1998). Following the work with the International Association for Child and Adolescent Psychiatry and Allied Professions, the Division of Mental Health of the World Health Organization (WHO, 1993) presented general guidelines to foster a consistent development of QL assessment instruments for children, stating that these should be child centered; employ subjective self-reports whenever possible; be age-related (or at least developmentally appropriate); enable cross-cultural comparisons; include a generic core and specific modules; and put an emphasis on health promotion aspects of QL, rather than solely on its negative aspects.

Within a developmental approach to QL and HRQL, adult measures are inappropriate for use with children because of the level of abstraction required for decision making, the lack of developmental considerations, and the inclusion of certain areas that may be irrelevant, or

exclusion of other areas which may be greatly valued (Spieth & Harris, 1996). However, these same considerations may be pertinently raised when administering the same instrument to children and adolescents. In fact, the lack of attention to these developmental issues may raise the question if children are sometimes considered to be "small adolescents" or if, on the other hand, adolescents tend to be merely seen as "grown up children". A way to endorse this developmental approach is to systematically test the psychometric performance of the existing measures for children and adolescents, in joint and separate samples, as many studies conducted to date covered a wide range of ages without presenting a stratification of the results by age group (Gerharz, Eiser, & Woodhouse, 2003).

There is not yet a consensual answer for what is age appropriate in QL and HRQL assessment. As regards instrument development and psychometric testing, different approaches have been adopted: some authors proposed to include specific dimensions for adolescents in a common questionnaire (Eiser & Morse, 2001); others developed a specific QL conceptual and measurement model for adolescents (Edwards, Huebner, Connell, & Patrick, 2002; Patrick, Edwards, & Topolski, 2002); a third group still, designed different age versions of the same instrument, such as the QL questionnaire KINDL ("KINDer Lebensqualitätsfragebogen", German QL questionnaire for children) and its three forms: KINDL-Kiddy (4-7 years); KINDL-Kid (8-12 years) and KINDL-Kiddo (13-18 years) (Ravens-Sieberer & Bullinger, 1998); finally, these last authors also successfully developed and tested the same HRQL instruments for both children and adolescents (e.g. Ravens-Sieberer et al., 2007). In the next section, we specifically describe and reflect on the contributions made by the DISABKIDS project, in applying a developmental approach to HRQL assessment.

The Contribution of DISABKIDS Project

The DISABKIDS project was originally funded by the Fifth Framework of the European Union, with its main goal being the development and promotion of the use of standardized instruments to assess HRQL in children and adolescents with chronic conditions (Bullinger, Schmidt, Petersen, & The Disabkids Group, 2002). This project enabled the construction and testing of a battery of instruments, which is now generally called "The DISABKIDS Questionnaires". All these questionnaires have self and proxy-report forms, and include: a chronic generic module (long and short versions); seven condition specific modules, and a measure of Smileys (for younger children aged between 4 and 7 years old). Since its original

implementation, the main distinctive features of this project remain its cross-cultural perspective; the modular system for combining generic and condition-specific aspects; the inclusion of a wide age range, and the assessment of both parents and their children's views (The European DISABKIDS Group, 2006). For the purpose of the present study, the DISABKIDS Chronic Generic Module (known as DISABKIDS-37) is described next.

The DISABKIDS-37 was originally developed from a simultaneous approach (i.e. different countries participating at the same time in the construction of a new instrument) (Simeoni et al., 2007), and revealed sound psychometric properties (Petersen, Schmidt, Power, Bullinger, & The Disabkids Group, 2005; Simeoni et al., 2007; The European Disabkids Group, 2006). The instrument clearly accomplished all the requirements to pediatric QL measures proposed by the WHO (1993), and complied with most suggested criteria to assess instrument developmental adequacy (Bruil & Detmar, 2005; Bullinger & Ravens-Sieberer, 1995; Wallander, Schmidt, & Koot, 2001).

The DISABKIDS-37 questionnaires were designed to be administered to both children and adolescents, thus adopting a conceptual and methodological perspective based on age universal markers, which is open to criticism regarding the exclusion of important age specific information (Wallander et al., 2001). Nevertheless, the DISABKIDS project adopted a number of methodological procedures which, in the light of the main cluster of research conducted so far, may be seen as relevant contributions to the refinement of a developmental approach to HRQL assessment. First, it emphasized the perceptions of children and adolescents themselves for conceptualizing HRQL and defining items accordingly, in a way that has been described as the most desirable approach (Petersen-Ewert, Erhart, & Ravens-Sieberer, 2011); second, several items addressed the notion of age-relevant contexts, such as family (e.g. "Are you able to do things without your parents?"), friends (e.g. "Do you go out with your friends?"), leisure activities (e.g. "Are you able to play or do things with other children/adolescents (like sports)?", and school (e.g. "Do you have problems concentrating at school because of your condition?"); third, the authors pursued a valuable and consistent strategy for psychometric data analysis, by systematically reporting a stratification of results by age groups (Petersen et al., 2005; Schmidt et al., 2006; The European Disabkids Group, 2006).

The need of developing new language versions led the research group to standardize additional translation and validation procedures to ensure the cross-cultural adequacy of those new versions to be developed along a sequential approach, such as the Mexican (Medina-Castro, 2007), the Brazilian (Fegadolli, Reis, Martins, Bullinger, & Santos, 2010), the Swedish (Chaplin,

Hallman, Nilsson, & Lindblad, 2011) and the Portuguese (Carona, Canavarro, & Bullinger, 2011) ones. Those procedures were based on updated guidelines for cross-cultural instrument adaptation (Schmidt & Bullinger, 2003), and included the phases of (1) translation (with the assessment of conceptual equivalence by international harmonization of items); (2) semantic validation; (3) pilot study, and (4) field study (The DISABKIDS Group, 2004). The results from semantic validation and pilot study of the Portuguese version of DISABKIDS-37 have been published elsewhere (Carona et al., 2011), and attested the comprehensibility, relevance and adequacy of items and response scales for both children and adolescents. As regards the cross-cultural adaptations of DISABKIDS-37 conducted so far, the abovementioned research gap remains pertinent: although the psychometric properties of these latest versions have been assessed in samples covering a wide age range (from 8 to 18 years old), those studies considered the differentiation of age subgroups only for the analysis of test-retest reliability (Chaplin et al., 2011) or did not consider it at all (Fegadolli et al., 2010; Medina-Castro, 2007). Thus, the main research goal for the present study was to examine the psychometric performance of the Portuguese (self and proxy-report) versions of DISABKIDS-37 in a global sample and age-stratified sub-samples, in order to verify the instrument adequacy for both children (8-12 years old) and adolescents (13-18 years old). Accordingly, the study aimed at assessing instrument reliability (internal consistency and parent-child agreement) and different types of validity (convergent, divergent and discriminant), using age group stratifications.

Method

Participants

Participants for this cross-sectional psychometric study were recruited at the Immunoallergology and Neurology/Neuropediatric outpatient services of Coimbra University Hospitals, Pediatric Hospital of Coimbra Central Hospital, Garcia de Orta Hospital (Almada) and Leiria Santo Andre Hospital, between March 2009 and December 2011. The convenience sample included subjects who met the following criteria: (1) age between 8 and 18 years old; (2) clinical diagnosis of asthma and epilepsy according to ICD-10, established by a physician; (3) minimum disease duration of 12 months; (4) regular medication intake. Subjects who presented comorbidity of asthma and epilepsy, or had major difficulties in understanding and answering questions (as indicated by doctors, following gross assessment of their cognitive abilities during

clinical interviews), were excluded from the study. Parents or caregivers (adults accompanying the child/adolescent to the hospital) who consented their child's participation, were also asked to participate in the study as proxies, with no additional inclusion criteria required for their participation. The diagnoses of asthma and epilepsy were chosen as inclusion criteria, in agreement with the previous work of semantic validation and pilot study of the Portuguese versions of DISABKIDS-37 (Carona et al., 2011). Besides, asthma was required as the common condition to be tested across all countries participating in the original DISABKIDS project (Simeoni et al., 2007) and subsequent instrument cross-cultural adaptations.

Measures

A similar assessment protocol was administered to children/adolescents and their parents, in self and proxy-report versions, which included the measures described next.

DISABKIDS-37. The Disabkids Chronic Generic Module (Carona et al., 2011; The European Disabkids Group, 2006) assesses HRQL in children (8-12 years old) and adolescents (13-18 years old) with any chronic health condition, and is available in self and proxy report forms. The instrument consists of 37 questions comprised along the following facets: Independence; Emotion; Social Inclusion; Social Exclusion; Physical Limitation and Treatment. DISABKIDS-37 is a Likert-scaled (1-5) questionnaire that provides standardized values (0-100) for each one of the facets and total score, with lower values indicating a more impaired HRQL. The standardized scale results from the calculation of the scoring algorithms of the instrument, with missing values being substituted if all but one of the items within a facet was responded to (Sandeberg, Johansson, Hagell, & Wettergren, 2010; The European Disabkids Group, 2006). In order to assess symptom severity and thus improve sample characterization, three questions from the asthma and epilepsy DISABKIDS specific modules ("When was the last time your child had an asthma attack/a seizure?"; "How many asthma attacks/seizures did your child have during the last year?"; "How severe was your child's condition during the last year?") were added to DISABKIDS-37 proxy-report questionnaire. Finally, the Portuguese versions of DISABKIDS-37 also included several questions on basic socio-demographic data; parents/caregivers' job and educational level were used to determine the socioeconomic status (SES), according to the classification system developed for the Portuguese context (Simões, 1994).

KIDSCREEN-10. The shortest version of Kidscreen questionnaires (Gaspar & Matos, 2008; Ravens-Sieberer et al., 2010), is a unidimensional measure composed of 10 questions regarding physical, psychological and social aspects of children and adolescents' QL. Kidscreen-10 was designed for individuals aged between 8 and 18, and includes both child and parent proxy reports. Each item is answered on a 5-point Likert scale, and the instrument provides an overall score (ranging between 5 and 50), where the lowest values reflect feelings of unhappiness, dissatisfaction and inadequacy towards different contexts of children and adolescents' lives (i.e. family, peers and school). Adequate Cronbach's internal consistency values were observed within our sample, for both self (a = .77) and proxy-report (a = .79) versions.

Strengths and Difficulties Questionnaire (SDQ). The SDQ (Fleitlich, Loureiro, Fonseca, & Gaspar, 2005; Goodman, 1997) assesses adjustment difficulties in children and adolescents, along four dimensions: emotional symptoms, conduct problems, hyperactivity/inattention and peer relationship problems. SDQ is available in self and proxy report forms, with 3-point Likert response scales: 0 ("not true"); 1 ("somewhat true") and 2 ("certainly true"). The overall score originated by the sum of the aforementioned sub-scales ranges between 0 and 40, with higher scores implying the existence of more psychological adjustment difficulties. Good internal consistency coefficients were obtained in this study for self (a = .77) and proxy (a = .83) versions of the instrument.

Procedure

Formal authorizations were obtained from the Ethical Committees of the aforementioned four public hospitals in Portugal. A brief description of the project's aims, methods and expected results was presented to the coordinators of the medical teams working in the departments where the sample was to be collected. Clinical cases who met the sampling criteria were identified by the responsible physician. A trained research assistant, acquainted to the project development and methodology, approached the children/adolescents, as well as their parents/caregivers, for briefly outlining the details of participation in the study. Signed informed consents were obtained from parents regarding their own and their child's participation when under 14 years old; these young children were not to be included even if the parents had previously allowed their participation, but they refused it themselves afterwards. In case of individuals aged 14 or older, informed consents were obtained from the adolescents and informed assents from their parents. Children/adolescents and their parents filled in the questionnaires in a room available at the

outpatient services in the presence of a research assistant, who answered the questions posed by the participants regarding the clarification of item content, assisted children/adolescents or parents with reading difficulties while filling in the questionnaires, and prevented information exchange between child and parent, so that the concordance between raters could be accurately assessed. In the few cases when the parent was unable to finish the questionnaire a stamped envelope was provided so that the parent could return it to the research team.

Data Analysis

Data were analyzed with SPSS 20.0 for Windows. Internal reliability was determined by calculating Cronbach's coefficient *a.* Pearson coefficients were computed to evaluate intercorrelation between facets and convergent and divergent validity. Following the suggestions of Nunnally & Bernstein (1994), alpha values ≥.70 were considered acceptable, and ≥.80 optimal; correlation coefficients between 0.1 and 0.3, 0.31 and 0.5, and those superior to 0.5, were classified as indicators of weak, moderate and strong associations, respectively. In addition, if the alpha value of a facet was higher than its correlation to the other facets, it was assumed that facet scores represented distinct aspects of HRQL (Sandeberg et al., 2010). This analysis was performed as a preliminary assessment of construct validity, because when the correlation between two subscales is less than their reliability coefficients, there is some evidence of a distinctive reliable variance measured by each subscale (Ware & Gandek, 1998). Discriminant validity was assessed through one-way between-groups multivariate analyses of covariance (MANCOVAs), examining diagnosis, age and gender differences in HRQL, separately for each factor and controlling for the remaining two factors by their inclusion as covariates. When the multivariate effect was significant we used univariate analyses (ANCOVAs) to further explore which facets of HRQL significantly differed across groups. Effect-size measures (partial Eta squared) are presented for the comparison analyses, considering $\eta_p^2 \geq .01$ as a small effect, $\eta_p^2 \geq .06$ as a medium effect and $\eta_p^2 \geq .14$ as a large effect (Cohen, 1988). Intraclass correlation coefficients (ICC) and Pearson coefficients were computed to assess the level of concordance between self and proxy-reports. All results were considered to be significant for a minimum confidence interval of 95%.

Results

Sample Characteristics

Participants were 349 children/adolescents with chronic conditions and their parents/caregivers, with a balanced distribution of the target group across age categories: children (group between 8 and 12 years old) were 56.2%. Frequencies in socio-demographic and clinical characteristics are shown in Table 1.

Table 1. Socio-demographic and clinical characteristics of the sample

	Children/Adolescents (*N* = 349)	**Parents/Caregivers** (*N* = 349)
Age (*M*/*SD*)	12.2 (2.6)	41.4 (6.2)
Age Group (*n*/%)		
Children (8-12)	196 (56.2)	
Adolescents (13-18)	153 (43.8)	
Gender (n/%)		
Male	206 (59.0)	49 (14.0)
Female	143 (41.0)	300 (86.0)
SES (*n*/%)		
Low	201 (57.6)	
Medium	102 (29.2)	
High	25 (7.2)	
Missing	21 (6.0)	
Marital status: married **(*n*/%)**		267 (76.5)
Diagnosis (*n*/%)		
Asthma	266 (76.2)	
Epilepsy	83 (23.8)	
Severity (*n*/%)		
Mild	83 (23.8)	
Moderate	163 (46.7)	
Severe	101 (28.9)	
Missing	2 (0.6)	
Comorbidity (*n*/%)	124 (35.5)	

Reliability

Acceptable and optimal internal consistency values were observed for the Disabkids' total score and each facet separately, for both age groups, with the exception of Independence (*a* =

.68), Inclusion (a = .66) and Limitation (a = .68) facets in children's self-reports, where Cronbach's alphas were slightly below the threshold for the acceptable values (see Table 2).

Inter-correlations between Facets and Total Score

Moderate to strong positive associations were verified for the correlations among facets and between facets and total HRQL scores, except for the Treatment sub-scale which correlated weakly, but still significantly, with the remaining facets (see Table 3). Across all samples, the correlation of a given facet with the other facets was always lower than the alpha value obtained for that same facet.

Convergent and Divergent Validities

Table 4 shows the results for the analyses of convergent and divergent validities. Moderate to strong associations with the expected direction were observed for the correlations between DISABKIDS-37 facets and total score, and QL (Kidscreen-10) and psychological adjustment difficulties (SDQ).

Discriminant Validity

For self-reported version of DISABKIDS-37, results presented statistically significant multivariate effects between chronic conditions (asthma vs. epilepsy), controlling for gender and age, and between age groups (children vs. adolescents), controlling for diagnosis and gender. While controlling for diagnosis and age, no multivariate effects were found for gender. Multivariate effects of diagnosis, after controlling for children's gender and age, were also statistically significant for the proxy-report version measure of pediatric HRQL. The proxy-report version of DISABKIDS-37 also demonstrated discriminant validity between children's gender, controlling for age and diagnosis, but not between age groups when diagnosis and gender effects were controlled. For both self and proxy report versions, univariate effects for each facet and global score of HRQL are presented on Table 5.

Table 2. Internal consistency reliability scores for the global sample and for separate age groups (self/proxy versions)

	Children *(self/proxy)*	**Adolescents** *(self/proxy)*	**Global Sample** *(self/proxy)*
Independence (6 items)	.68/.76	.75/.82	.70/.79
Emotion (7 items)	.85/.92	.84/.91	.84/.92
Inclusion (6 items)	.66/.72	.70/.76	.68/.74
Exclusion (6 items)	.78/.84	.75/.83	.77/.83
Limitation (6 items)	.68/.82	.70/.74	.70/.79
Treatment (6 items)	.77/.86	.84/.83	.80/.85
37 Questions	.91/.93	.92/.94	.91/.94

Table 3. Matrix of correlations between HRQL (DISABKIDS-37) total score and separate facets for self and proxy reports (self-report/proxy-report).

Children	Independence	Emotion	Inclusion	Exclusion	Limitation	Treatment
Emotion	.46/.48					
Inclusion	.55/.52	.40/.41				
Exclusion	.47/.50	.60/.67	.42/.51			
Limitation	.37/.39	.55/.66	.44/.27	.34/.42		
Treatment	.30/.30	.63/.54	.30/.26	.46/.49	.36/.40	
HRQL Total	.69/.68	.85/.87	.68/.63	.74/.80	.69/.72	.72/.71
Adolescents	Independence	Emotion	Inclusion	Exclusion	Limitation	Treatment
Emotion	.64/.63					
Inclusion	.58/.67	.43/.57				
Exclusion	.50/.62	.53/.70	.51/.62			
Limitation	.48/.47	.72/.62	.40/.43	.37/.42		
Treatment	.39/.36	.58/.57	.24/.34	.45/.50	.41/.32	
HRQL Total	.77/.79	.88/.90	.66/.77	.72/.82	.75/.69	.73/.68
Global Sample	Independence	Emotion	Inclusion	Exclusion	Limitation	Treatment
Emotion	.52/.55					
Inclusion	.57/.60	.40/.49				
Exclusion	.49/.55	.57/.68	.46/.55			
Limitation	.41/.42	.62/.64	.42/.34	.36/.42		
Treatment	.31/.33	.60/.55	.24/.30	.44/.49	.37/.36	
HRQL Total	.72/.73	.86/.88	.67/.70	.73/.81	.72/.71	.71/.70

Note. All correlations are significant, $p < .01$.

Table 5. Discriminant validity by diagnosis, age and gender for DISABKIDS-37 (self/proxy)

	Diagnosis					Age group				
Self-report	Asthma (n = 266)	Epilepsy (n = 83)	$F_{(1,345)}$	p	η_p^2	Children (n = 196)	Adolescents (n = 153)	$F_{(1,345)}$	p	η_p^2
	M (*SD*)	*M* (*SD*)				*M* (*SD*)	*M* (*SD*)			
Independence	80.72 (14.27)	76.81 (15.90)	4.06	.05	.01	77.87 (15.60)	82.24 (13.22)	8.51	<.01	.02
Emotion	81.07 (17.77)	80.29 (18.64)	.00	.97	.00	81.29 (19.00)	80.37 (16.56)	.06	.81	.00
Inclusion	80.92 (14.88)	72.49 (18.18)	18.76	<.01	.05	76.66 (16.84)	81.81 (14.67)	9.84	<.01	.03
Exclusion	91.10 (12.14)	81.33 (20.19)	27.01	<.01	.07	88.01 (16.32)	89.76 (13.16)	1.87	.17	.01
Limitation	71.74 (16.11)	79.82 (16.66)	16.78	<.01	.05	73.02 (17.24)	74.48 (15.72)	.74	.39	.00
Treatment	78.38 (20.10)	73.04 (25.44)	3.39	.07	.01	79.83 (20.74)	73.64 (22.17)	6.45	.01	.02
Global HRQL	80.67 (12.07)	77.38 (13.87)	3.54	.06	.01	79.50 (13.01)	80.38 (12.04)	.73	.39	.00
	Diagnosis					**Gender**				
Proxy-report	Asthma (n = 266)	Epilepsy (n = 83)	$F_{(1,345)}$	p	η_p^2	Boys (n = 206)	Girls (n = 143)	$F_{(1,345)}$	p	η_p^2
	M (*SD*)	*M* (*SD*)				*M* (*SD*)	*M* (*SD*)			
Independence	82.22 (14.56)	76.31 (17.10)	8.69	<.01	.03	81.82 (14.92)	79.37 (15.98)	1.33	.25	.00
Emotion	79.32 (19.60)	76.08 (19.96)	1.40	.24	.00	79.66 (18.57)	76.95 (21.20)	1.24	.27	.00
Inclusion	79.98 (15.41)	72.74 (17.82)	12.20	<.01	.03	79.07 (16.04)	77.10 (16.63)	.50	.48	.00
Exclusion	89.32 (14.07)	78.21 (19.92)	31.51	<.01	.08	86.97 (16.46)	86.25 (16.18)	.02	.90	.00
Limitation	66.76 (17.15)	78.61 (16.92)	34.80	<.01	.09	71.36 (16.06)	67.02 (19.83)	9.47	<.01	.03
Treatment	78.23 (20.31)	73.69 (22.04)	2.36	.13	.01	79.00 (20.54)	74.48 (20.94)	3.28	.07	.01
Global HRQL	79.31 (13.20)	75.94 (14.14)	3.21	.07	.01	79.65 (13.06)	76.86 (13.94)	2.95	.09	.01

Table 4. Pearson correlations coefficients between HRQL (DISABKIDS-37) and general QL (Kidscreen-10) and psychological adjustment (SDQ) measures (self/proxy versions).

		Independence	Emotion	Inclusion	Exclusion	Limitation	Treatment	HRQL Total
Kidscreen	*Children*	.57/.55	.42/.50	.56/.56	.48/.54	.44/.30	.25/.33	.60/.61
	Adolescents	.60/.63	.49/.56	.49/.59	.59/.54	.40/.44	.34/.36	.62/.67
	Global	.55/.59	.45/.53	.50/.57	.50/.53	.41/.36	.30/.34	.60/.63
SDQ	*Children*	-.41/-.50	-.47/-.49	-.40/-.49	-.53/-.64	-.36/-.34	-.32/-.42	-.56/-.64
	Adolescents	-.54/-.53	-.48/-.59	-.52/-.56	-.60/-.63	-.38/-.39	-.30/-.35	-.60/-.65
	Global	-.46/-.51	-.48/-.53	-.45/-.51	-.56/-.64	-.37/-.36	-.30/-.39	-.58/-.64

Note. All correlations are significant, p <.01.

Parent-child Agreement

Moderate levels of agreement between child/adolescent and parent-proxy reports did not differ across the total and separate age-group samples (Table 6).

Table 6. Intraclass and Pearson correlation coefficients for DISABKIDS-37 facets and total score between self (children/adolescents) and proxy (parents) reports

	Children		**Adolescents**		**Global Sample**	
	ICC	*r*	*ICC*	*r*	*ICC*	*r*
Independence	.34	.34	.44	.44	.37	.37
Emotion	.44	.44	.39	.39	.42	.42
Inclusion	.36	.36	.45	.45	.38	.38
Exclusion	.49	.49	.41	.41	.46	.46
Limitation	.44	.44	.46	.46	.45	.45
Treatment	.46	.46	.34	.34	.40	.40
HRQL Total	.50	.50	.48	.48	.49	.49

Discussion

The present paper is, to our knowledge, the first to report comprehensive results from a validation field study of DISABKIDS-37 according to the procedures outlined by the original Disabkids European project (The DISABKIDS Group, 2004), while differentiating its psychometric analyses for children and adolescents age groups. Following the development of the Portuguese versions of DISABKIDS-37 according to the latest guidelines in cross-cultural instrument adaptation (Carona et al., 2011), the main aim of this study was to assess the developmental adequacy of DISABKIDS-37 for children and adolescents with chronic health conditions, by systematically testing its psychometric properties in age-stratified samples. Key findings from this study indicate that the Portuguese versions of DISABKIDS-37 are reliable and valid measures for the assessment of HRQL in children, adolescents and mixed pediatric samples, and highlight the ability of these questionnaires for mapping differences in the HRQL of children and adolescents with chronic health conditions.

The observed results in terms of internal consistency, parent-child agreement and construct validity are similar to the originally published by the European research group (The European DISABKIDS Group, 2006), thus highlighting the importance of adopting structured instrument adaptation protocols, in order to ensure its cross-cultural validity and quality of psychometric performance. Reliability values for DISABKIDS-37 items and facets were generally very good, although scores in children-reported Independence, Inclusion and Limitation facets, and self-reported Inclusion facet, were below the commonly established value of .70.

The fact that every facet's internal consistency was always higher than its correlation to the other facets suggests that facet scores depict distinct aspects of pediatric HRQL; however, the suggested factorial structure is to be tested in further studies since the ones conducted so far reported inconsistent findings (Sandeberg et al., 2010; Schmidt et al., 2006). Regarding convergent validity, the fact that moderate to strong associations were observed between generic QL and HRQL instruments underlines the pertinence of assuming those two concepts as complementary in their applications, but somehow distinct in nature (Wallander et al., 2001). The same tenet is valid for understanding the similar strength of associations between HRQL and psychological difficulties, since mental health status has been described as a foremost

determinant of QL outcomes (Bovier, Chamot, & Perneger, 2004). These psychometric properties were systematically tested and observed in this study for children and adolescents mixed and separate samples, thus reaffirming the developmental adequacy and reliability of DISABKIDS-37 for both age groups. Despite the fact the instrument discriminated between gender (proxy version) and age (self version) groups, effect sizes were larger for discrimination between conditions. These findings have been observed in previous studies (Sandeberg et al., 2010), and emphasize the adequacy of DISABKIDS-37 to discriminate between diagnoses (the original main purpose of the instrument), without rejecting its sensitivity to developmental specificities.

Levels of agreement between parent/caregiver and child/adolescent reports were only moderate in our study, besides the fact that evidence on the discriminant validity of the questionnaires was differently observed in each report form. These results indicate that child/adolescent and parent/caregiver reports are valid and complementary to each other, and support the recommendation for "hearing the voices" of both information sources (and not just substituting one for another), depending on the specific aims of a given HRQL assessment (Theunissen et al., 1998).

Limitations and Strengths

The interpretation of results from the present study must take into account its major limitations: first, the obtained convenience clinical sample mainly included individuals with moderate to severe health conditions and from lower economic backgrounds, demonstrating discrepancies in frequency distribution for clinical (severity) and socio-demographic (SES) variables, which have been extensively reported to influence pediatric adaptation outcomes (Bullinger et al., 2002); second, interaction effects between clinical and demographic variables were not explored, even if that analysis was not among this study aims; third, the stratification of analyses for two age groups suited the sample size, but it could be further refined by testing, for instance, three groups of children, preadolescents and adolescents.

Despite these limitations, this study validates the pertinence of using DISABKIDS-37 as a single measure to assess pediatric HRQL in different age groups: if on the one hand some age-relevant information is likely to be missed, on the other hand, depending on the specific purpose of a given assessment, that restraint might be a relatively small cost for the sake of allowing between- and within-group comparisons. Besides the quality of psychometric performance observed in our study, DISABKIDS questionnaires operationalize the unique importance of

context for the refinement of pediatric health outcomes assessment (Christakis, Johnston, & Connell, 2001), by including different items with a clear reference to common age relevant contexts such as family, friends and school environment.

Conclusion

Overall, results from the present study support the use of DISABKIDS as a reliable and useful tool for assessing, in a developmentally appropriate way, the HRQL of children and adolescents with chronic conditions. Future research aiming at a comprehensive and contextual pediatric HRQL assessment could benefit from the use of both quantitative and qualitative methods, as it has been suggested for other constructs in pediatric psychology (Spirito, 1996). The combination of the use of DISABKIDS-37 with qualitative methodologies, the analysis of its performance in relatively understudied samples (e.g. cerebral palsy, obesity), and the examination of its factorial structure with exploratory and/or confirmatory factor analyses, are promising venues for future research in this field.

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Empirical Study II |

Social Support and Adaptation Outcomes in Children and Adolescents with Cerebral Palsy

Submitted article, under review

Social Support and Adaptation Outcomes in Children and Adolescents with Cerebral Palsy

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Abstract

Objectives. This study had two main objectives: first, to describe the social support and psychological maladjustment of children and adolescents with cerebral palsy (CP); and second, to test a mediation model where psychological maladjustment was hypothesized to mediate the link between social support and health-related quality of life (HRQL). In addition, the moderating role of gender and age was examined for this mediation model. **Methods.** Self and proxy-report questionnaires on the aforementioned variables were administered to a sample of 96 children/adolescents with CP and 118 healthy controls, as well as one of their parents. Univariate and multivariate analyses of covariance were conducted to examine differences in social support and psychological maladjustment, respectively. PROCESS computational tool was used for path analysis-based mediation, moderation and moderated mediation analyses. **Results.** Children/adolescents with CP reported lower levels of social support than their healthy peers, but no significant differences emerged in terms of their psychological maladjustment. For children/adolescents with CP, internalizing and externalizing problems were found to mediate the link between social support and HRQL, and these indirect effects were not conditional upon age or gender. **Discussion.** Children and adolescents with CP are likely have more negative perceptions of social support, but not necessarily more psychological adjustment problems than their healthy, able-bodied peers. Results further suggest that interventions targeting social support perceptions may positively affect HRQL outcomes in children/adolescents with CP, through the improvement of internalizing and externalizing dimensions of their psychological adjustment.

Keywords: social support; adaptation; health-related quality of life; psychological adjustment; cerebral palsy.

Introduction

Cerebral palsy (CP) was recently described as an "umbrella term" for a group of disorders of movement and posture, attributed to non-progressive disturbances that occurred in the developing fetal or infant brain (Rosenbaum et al., 2005). With a prevalence of 1.5-3.0/1000 live births (SCPE, 2000), CP is the most common physical disability in childhood (Moreno-De-Luca, Ledbetter, & Martin, 2012).

Although there is growing evidence for impaired health-related quality of life (HRQL) (Rosenbaum et al., 2007; Varni et al., 2005) and increased psychological maladjustment in children/adolescents with CP (Brossard-Racine et al., 2012a, 2012b), research on their psychosocial adjustment remains underrepresented in current literature (Vles, Hendriksen, Vles, Kessels, & Hendriksen, 2012), and little is known about the association of specific factors, such as social support, with their HRQL outcomes (Livingston, Rosenbaum, Russell, & Palisano, 2007). Given the fact that such contextual factors are important determinants of HRQL in individuals with disabilities (Majnemer, Shevell, Rosenbaum, Law, & Poulin, 2007), more research is needed to examine the circumstances under which social support influences specific outcomes, as well as the potential mechanisms via which it may operate. A deeper understanding on these matters may improve the effectiveness of current psychosocial interventions for children/adolescents with CP, by ascertaining the importance of social support perceptions in their associations with psychological adjustment and HRQL outcomes.

Adaptation Outcomes in Pediatric Cerebral Palsy: Moving From Outcomes Description to Outcomes Prediction

"Adaptation" is broadly defined as "any process whereby behavior or subjective experience alters to fit in with a changed environment or circumstance" (Colman, 2009, p. 11). In pediatric settings, adaptation outcomes have been operationalized in terms of child/adolescent's mental health, social functioning and physical health (Wallander, Varni, Babani, Banis, & Wilcox, 1989). Therefore, the multidimensionality of these outcomes is likely to encompass the constructs of psychological (mal)adjustment and HRQL. As regards the adaptation of individuals with CP, research has recently moved from an exclusive focus on

impairment and function, to a broader framework where quality of life (QL) and HRQL measures are complementary to traditional functional and medical assessments (Schneider, Gurucharri, Gutierrez, & Spira, 2001).

Although sometimes used interchangeably with the notion of HRQL, QL has been adopted as one of the most important goals of current research in CP (Bjornson & McLaughlin, 2001), and is perhaps "the holy grail of [intervention] outcomes", as perceived by youths with CP, their parents and medical professionals (Vargus-Adams & Martin, 2009). HRQL may be seen as a component of the holistic concept of quality of life (QL), because it encompasses physical, social and mental dimensions of functioning, along with condition/treatment facets, but excludes a broader range of aspects such as political freedom and economical issues (The European Disabkids Group, 2006). For the purpose of the present review, comments on previous research were based on the theoretical assumption that QL and HRQL are somehow overlapping, but nevertheless, distinct concepts.

Psychological (mal)adjustment in children and adolescents, on the other hand, is generally associated with two broadband dimensions: internalizing and externalizing problems (Bornstein, Hahn, & Haynes, 2010). Internalizing problems essentially affects the child's internal psychological states, rather than the external world, and include withdrawn, anxious and depressive behavior; externalizing problems, in contrast, relate to children's outward behavior as negatively acting on the external environment, and include disruptive, aggressive and hyperactive behavior (Liu, 2004). Although the predominance of internalizing over externalizing problems has been a consistent finding for children with chronic medical conditions (Thompson, Gustafson, Hamlett, & Spock, 1992), the distinctive importance of examining externalizing problems in pediatric populations has been emphasized (Holmbeck et al., 2008).

On a theoretical level, psychological (mal)adjustment may be regarded as a specific adaptation outcome (Wallander et al., 1989), and as a QL determinant (Bovier, Chamot, & Perneger, 2004; Janssen, Voorman, Becher, Dallmeijer, & Schuengel, 2010). In fact, QL has been described as "the ultimate outcome" in psychosocial rehabilitation practice (Livneh & Antonak, 2005, p. 13). Moreover, the simultaneous assessment of positive and negative dimensions has been recommended as a mean of providing a more complete picture on the individual's adaptation outcomes (Ridder, Geenen, Kuijer, & Middendorp, 2008). Psychological maladjustment is likely to imply increased burden of disease and deteriorate internal resources (e.g., mastery, self-esteem), and thus impair an individual's QL (Bovier et al., 2004). Therefore, from a conceptual and methodological point of view, psychological (mal)adjustment may be

assumed as both a QL determinant and a "first-order outcome" (more specific), and QL and HRQL as "second-order outcomes" (more general).

Children and adolescents with CP have been reported to experience a markedly impaired HRQL, in comparison to children/adolescents with other chronic health conditions (Schmidt et al., 2006) and to their healthy/able-bodied peers (Varni et al., 2005). As a notable exception in challenging these widespread findings, self-reported QL of children with CP has been found to be mostly similar to the QL perceived by children in the general population (Dickinson et al., 2007). Notwithstanding the contributions of such studies, QL research in pediatric CP has been characterized by a number of criticisms: QL and HRQL have been often used interchangeably or inadequately assessed; children and adolescents have been typically studied as one single group; the adoption of children/adolescents' self-reports has been rare, in contrast to an excessive reliance on parent/proxy-reports; scores have been usually compared to norms and not to homologous peer samples; and determinants of QL and HRQL outcomes have been scarcely studied (Davis et al., 2009; Livingston et al., 2007; Rosenbaum et al., 2007).

On the topic of psychological adaptation outcomes, children/adolescents with chronic conditions are at higher risk for psychological maladjustment (Stawski, Auerbach, Barasch, Lerner, & Zimin, 1997). Moreover, the study of developmental specificities in pediatric populations has suggested that internalizing problems are more common in older children, and that gender differences in externalizing problems tend to emerge earlier than gender differences in internalizing problems (Pinquart & Shen, 2011). As regards pediatric CP in particular, psychological problems seem frequent and include peer difficulties, inattention-hyperactivity, emotional symptoms, increased dependence, withdrawal, obstinacy and antisocial characteristics (Brossard-Racine et al., 2012a; Parkes et al., 2008). In fact, children and adolescents with CP have been reported to achieve less psychosocial adjustment (Vles et al., 2012), besides being five times more likely to present parent-reported behavior problems than their healthy peers (McDermott et al., 1996). Complementarily, some age and gender specificities are noteworthy for CP: a decrease in the frequency of behavior problems has been related to ageing (McCullough, Parkes, Kerr, & McDowell, 2011), and an increased risk for conduct and hyperactivity problems has been observed for boys (Parkes, White-Koning, McCullough, & Colver, 2009). In addition, even if some evidence has been gathered for the role of psychological maladjustment as a QL predictor in children/adolescents with CP (Majnemer et al., 2007; Wiley & Renk, 2007), the selection of informants on children's outcomes seems influential: while internalizing problems have been related to child and parent-reported HRQL, externalizing problems were related with

parents' reports only (Janssen et al., 2010). Regrettably, research on the psychological adjustment of children/adolescents with CP is open to some of the abovementioned criticisms to QL research, namely in terms of the excessive reliance on proxy-reports, the tendency to perform comparisons with norms, and the scarcity of data on psychological adjustment determinants.

Compared to the amount of literature on the adaptation outcomes for other chronic health conditions, studies on pediatric CP are remarkably few (The European Disabkids Group, 2006). A considerable amount of research in this area has been devoted to the description of differences in the adaptation outcomes experienced by clinical and non-clinical populations. Those studies are important because they promote insights on the differentiation of groups, which may be useful for mapping needs and/or allocating resources, but they are still of limited heuristic value to ascertain potentially modifiable associations between determinants, such as social support, and adaptation outcomes (Livingston et al., 2007; Majnemer & Mazer, 2004).

Social Support and Adaptation Outcomes in Children and Adolescents with Cerebral Palsy

Social support was defined here as "social relationships that provide (or can potentially provide) material and interpersonal resources that are of value to the recipient, such as counseling, access to information and services, sharing of tasks and responsibilities, and skill acquisition" (Thompson, 1995, p. 43). This definition was preferred because it implicitly links the individual's context of social relationships with his/her behavioral development. Social support has been commented as an important factor in developmental psychopathology (Thompson, Flood, & Goodvin, 2006), and stated as a determinant social-ecological factor of adaptation outcomes in the so-called "disability-stress-coping" model of individual and family adaptation to chronic physical conditions (Wallander et al., 1989). In literature, the influence of social support on adaptation outcomes has been hypothesized in two distinct, but complementary models: on the one hand, social support may generally improve adaptation outcomes, whether the person is under increased stress or not (i.e., "main effect model"); on the other hand, social support is likely to impede, reduce or control the detrimental effects of stressful situations (i.e., "buffering model") (Cohen & Wills, 1985; Thompson et al., 2006). The refinement and examination of a main effect model (i.e. "an indirect effect model", Bovier et al., 2004) in children and adolescents with CP was selected as a general framework for the present research work.

Social support has been described as a potential causal determinant of psychological symptomatology and other health-related variables (Schwarzer & Leppin, 1991), but the understanding of the mechanisms via which social support is likely to determine such outcomes, still needs to be addressed in research (Helgeson, 2003). In fact, social support may exert indirect effects (i.e. through other variables) on adaptation outcomes, which may even surpass its straightforward direct effects (Schwarzer & Leppin, 1991). Moreover, in research aimed at predicting well-being outcomes, the measurement of subjective aspects of social support, such as perceived support or satisfaction with support, has been recommended (Vaux & Harrison, 1985). Social support has been reported to increase children and adolescents' QL, with girls perceiving better support than boys, and children better than adolescents (Malkowska, Mazur, & Woynarowska, 2004). As commented earlier for the definition adopted, social support provides a context for the development of social competence in children, which is a good predictor of later externalizing and internalizing problems in preadolescence, and externalizing problems in adolescence (Bornstein et al., 2010). Social support has been also hypothesized to promote QL outcomes through the preservation of feelings of connectedness and a sense of belonging (Helgeson, 2003). On the topic of other age and gender differences, it should be noted that the amount of social support sources tends to increase in adolescence, with multiple resources being related to better adjustment (Levitt et al., 2005), and that girls seem more likely to perceive higher levels of social support than boys (Bokhorst, Sumter, & Westenberg, 2010).

In pediatric populations, increased social support has been related to improved psychological adjustment, and identified as a significant predictor of internalizing and externalizing problems in children/adolescents with chronic physical conditions (Wallander & Varni, 1989). In that study, no interaction effects between social support and age or gender were verified. As regards the determinant role of social support in children and adolescents with chronic conditions, it is important to note that internalizing and/or externalizing problems may be a response to stressful social situations, such as peer rejection (Pinquart & Shen, 2011). Additionally, decreased HRQL due to impaired social functioning has been reported for adolescents with physical disabilities (Stevens et al., 1996). In fact, it has been suggested that children with conditions that involve the central nervous system (such as CP) may face additional social difficulties (LaGreca, Bearman, & Moore, 2002). Nevertheless, for pediatric populations in general, and for pediatric CP in particular, the examination of direct and indirect effects (via the internalizing/externalizing dimensions of psychological adjustment) of social support on HRQL has not been addressed in research. In the same way, gender and age specificities have not been thoroughly examined in models that hypothesize the links between social support and adaptation

outcomes. This rationale calls for the conduction of mediation and moderated mediation analyses, which became popular in developmental and behavioral pediatric research (Rose, Holmbeck, & Franks, 2004). After all, these analyses allow theory development and testing, as well as the identification of potentially modifiable links between variables of interest (Preacher & Hayes, 2004).

The Current Study

The present study adopted a developmental dyadic approach to adaptation outcomes assessment, by examining two complementary models: in the first model, social support, psychological maladjustment and HRQL were exclusively self-reported; in the second model, parent-reported psychological maladjustment and HRQL were combined with child-reported social support. Although more complex, such methodological procedure was thought to improve the clinical validity of our study (cf. Smith, 2007).

The aims for the present study were defined as follows: first, to assess the satisfaction with social support and the psychological maladjustment (i.e. internalizing and externalizing problems) of children/adolescents with CP, in comparison to typically developing children/adolescents; and subsequently, to examine a potential process through which social support may influence HRQL, by testing two dimensions of psychological maladjustment (i.e. internalizing and externalizing problems) as mediators of that relationship. The study further explored age and gender differences in social support and psychological maladjustment, as well as the possibility that the indirect effects of social support on HRQL differed between gender and age groups, and more specifically, whether gender and age group moderated the path from social support to internalizing/externalizing problems and/or the path from these clusters of psychological symptoms to HRQL. Figure 1 graphically depicts the hypothesized moderated mediation model, as described and adapted from Hayes (2012a).

According to these objectives, three theoretically-driven hypotheses were outlined: (1) children/adolescents with CP would report decreased levels of social support in comparison to their healthy, able-bodied peers; (2) higher levels of self and proxy-reported internalizing and externalizing problems would be observed for children/adolescents with CP, than for healthy, able-bodied children/adolescents; (3) boys would report more externalizing problems than girls, and adolescents would report more internalizing problems than children. Although internalizing and externalizing problems were tested as mediators in the relationship between social support

and HRQL, and age and gender moderation effects were examined within that mediation model, we made no specific predictions for those analyses.

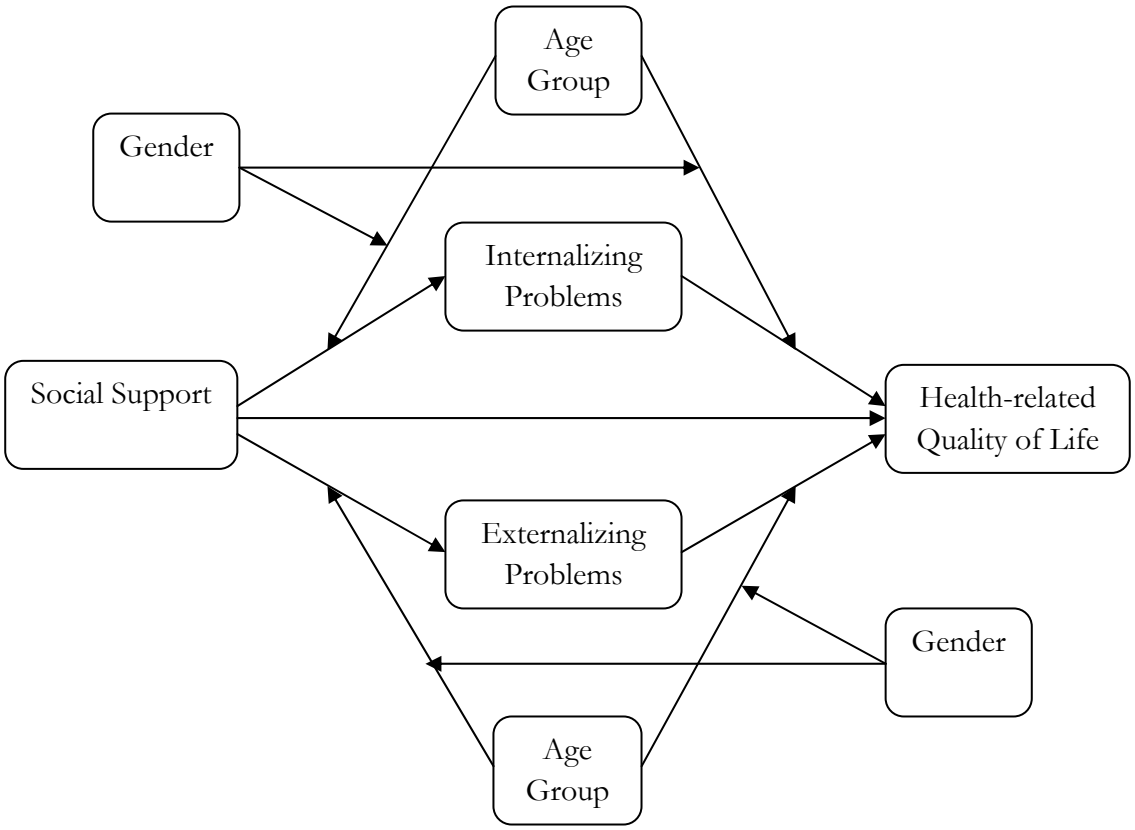

Figure 1. Gender and age group as moderators of multiple mediated pathways from social support to HRQL

Methods

Participants

The sample for this study (*N* = 96 children/adolescents with CP and one of their parents) was recruited in ten Portuguese Cerebral Palsy Associations between July 2010 and July 2011. Pediatric subjects were assigned to the study if they met the following criteria: (1) a clinical diagnosis of CP; (2) age between 8 and 18 years old; (3) minimum intelligence quotient (IQ) of 70. Cases where results from previous formal assessments of IQ were not available (*n*=13) were still included in the study, if they didn't present significant cognitive delay, as indicated through gross evaluation of their cognitive abilities, and simultaneous absence of any previous adaptation in their school curricula. For parents, a single inclusion criterion was considered: being the primary family caregiver of the child/adolescent with CP, as suggested by the largest amount of

time dedicated to child's health issues and care. One hundred and sixty one children/adolescents with CP and their parents were assigned to the study, out of which 65 were progressively excluded because of the following reasons: 7 cases refused to participate; 47 did not visit the institutions during the established period for sample collection; 2 cases were only able to provide self-reports, since children were living in foster care placement, and 9 cases did not complete all the questionnaires.

Complementarily, a convenience sample of controls was collected in two public schools of Coimbra district, between January and June 2010. Children and adolescents included in this sample were to fulfill two criteria: aged between 8 and 18 years old, and reporting no diagnosed chronic health condition. For their parents, a single inclusion criterion was considered: being the parent who spent more daily time with the child/adolescent. A total of 124 children/adolescents were assigned to participate in the study. Since two parents did not allow their children's participation, and four adolescents did not return their parents' questionnaires, a total sample of 118 healthy controls was obtained.

Measures

Satisfaction with Social Support Scale for Children and Adolescents (SSSS). This scale assesses satisfaction with social support, as perceived by children and adolescents (Gaspar et al., 2009). The instrument includes 12 items, for which the individual indicates his degree of agreement along a five point Likert scale from 1 (*totally disagree*) to 5 (*totally agree*). The items may be grouped in two factors: "Satisfaction with Social Support" (e.g. "I am satisfied with the activities and things I do with my group of friends"), and "Activities connected to Social Support" (e.g. "I would like to participate more in organized activities, such as sport clubs, scouts"). The overall score resulting from the sum of both factors varies between 12 and 60, with higher values indicating more satisfaction with social support. Adequate internal consistency values were found for our samples of healthy controls (a = .80) and pediatric CP (a = .76).

. ***Strengths and Difficulties Questionnaire (SDQ).*** SDQ is a brief measure of psychological adjustment for children and adolescents, allowing both self and proxy-reports (Goodman, 2001). SDQ comprises 20 items assessing psychopathological symptoms, and 5 items targeting prosocial behavior, for which the respondent indicates his perception within a 3 point Likert response scale: 0 (*not true*); 1 (*somewhat true*) and 2 (*certainly true*). Apart from the prosocial factor, an alternative scaling for SDQ has been recently proposed: emotional

symptoms (e.g. "I am often unhappy, down-hearted or tearful") and peer problems (e.g. "Other children or young people pick on me or bully me") were integrated into a broader scale of internalizing problems; conduct problems (e.g. "I get very angry and often lose my temper") and hyperactivity-inattention symptoms (e.g. "I am constantly fidgeting or squirming") were combined into a scale of externalizing problems (Goodman, Lamping, & Ploubidis, 2010). Scores for each one of these broad subscales vary between 0 and 20, with higher values indicating increased psychological maladjustment. Acceptable internal consistency values were observed for internalizing and externalizing subscales within our global sample, for both self and proxy-reports, with Cronbach's alphas ranging between .67 (CP self-reported internalizing problems) and .82 (controls proxy-reported externalizing problems).

DISABKIDS-37. The generic module (long version) of Disabkids questionnaires is available in self and proxy formats and assesses HRQL of children and adolescents with chronic health conditions (Carona, Bullinger, & Canavarro, 2011; The European Disabkids Group, 2006). Disabkids-37 items are to be answered within a 5 point Likert scale, and may be grouped into six facets: Independence (e.g. "Are you able to do things without your parents?"), Emotion (e.g. "Are you unhappy because of your condition?"), Inclusion (e.g. "Do your friends enjoy being with you?"), Exclusion (e.g. "Do you feel different from other children/adolescents?"), Physical Limitation (e.g. "Is your life ruled by your condition?") and Treatment (e.g. "Does taking medication bother you?"). Given the fact that most CP cases in our sample were not medicated (n = 58), a syntax for a standardized global score (0-100) based on the remaining 31 items was preferred, with lower scores indicating the most impaired HRQL. Good internal consistency values were found in our sample for self (a = .88) and proxy reports (a = .95).

Procedure

Authorizations were obtained from the Direction Boards of Portuguese Cerebral Palsy Associations participating in this study. Informed consents were obtained from parents regarding their own and their child's participation (when under 14 years old); these young children were also to assent their own participation, and not to be included even if the parents had previously allowed their participation, but they refused it themselves afterwards. For individuals aged 14 or older, informed consents were obtained from adolescents.

Authorizations for the collection of the control sample were given by the Direction Boards of both public schools involved in this research study. After the selection of a number of

classes to fairly achieve the intended sample size, questionnaires were administered to children/adolescents in the classroom. Parents completed their questionnaires at home and returned them through their children. General procedures for obtaining informed consents/assents were similar to the ones described for the clinical sample.

Data Analysis

Internal consistency of questionnaires integrating the assessment protocol was measured through the calculation of their Cronbach's alphas, which were then classified as minimally acceptable (≥.65), acceptable (≥.70) and optimal (≥.80) (DeVellis, 1991; Nunally & Bernstein, 1994). Descriptive statistics were obtained for clinical and socio-demographic variables, and the homogeneity between clinical and control samples was tested through mean differences tests (Student's *t* tests) or frequency differences for categorical variables (chi-square tests). In order to compare psychological adjustment outcomes between groups, multivariate analysis of covariance (MANCOVA) was performed, examining condition (CP vs. healthy controls), age (children vs. adolescents) and gender (boys vs. girls) group differences in self and proxy-reported internalizing and externalizing problems. When multivariate effects were significant, univariate analyses were used to further explore which dimensions of psychological adjustment significantly differed across groups. Differences in social support between condition, age and gender groups were examined through univariate analysis of covariance (ANCOVA). Given the observed discrepancy in SES frequencies between clinical and control samples ($\chi 2 = 29.38$; $p = .00$), SES was dichotomized (0 = *low*; 1 = *medium/high*) and entered as covariate in univariate and multivariate analyses. Effect-size measures (partial Eta squared) were presented for the comparison analyses, considering $\eta_p^2 \geq .01$ as a small effect, $\eta_p^2 \geq .06$ as a medium effect, and $\eta_p^2 \geq .14$ as a large effect (Cohen, 1988). No effect sizes were calculated for multiple mediation models because of the inclusion of covariates. Pearson's bivariate correlation coefficients were computed to assess associations between variables, while adopting the following guidelines to classify their strength: ±.10 - ±.29 (weak); ±.30 - ±.49 (moderate); ±.50 - ±1.0 (strong) (Cohen, 1988).

As conceptually depicted in Figure 1, two moderators (age group and gender) were hypothesized to influence the mediator effects of internalizing and externalizing problems on the links between social support and HRQL. Hence, multiple moderated mediation analyses were elected because they permitted the assessment of both "how" and "when" an indirect effect

would occur in models where more than one mediator and one moderator were included. Moderated mediation is said to exist when the mediating effect of a given variable in the relationship between a predictor and outcome depends on a level of a moderator. To test for multiple moderated mediation, PROCESS was used as a computational tool for path analysis-based moderation and mediation analyses, as well as their combination in the so-called "conditional process model" (Hayes, 2012b). Bootstrapping procedures have been reported to be superior to other traditional methods of studying mediation, since they do not require the assumption of a normal distribution to be met, and demonstrate higher power with reasonable control over the Type-I error rate, through appropriate control of confidence intervals (Mackinnon, Lockwood, Hoffman, West, & Sheets, 2002; Preacher & Hayes, 2008). In bootstrapping procedures, cases from the original dataset are randomly re-sampled with replacement to re-estimate the sampling distribution, and from this new sampling distribution, bias-corrected and accelerated confidence intervals (BCa CIs) are then created, with an indirect effect being significant if zero is not contained within the lower and upper CIs (Shrout & Bolger, 2002). In moderated mediation analyses, indirect effects are thus to be separately computed across the levels of a moderator. PROCESS computational tool provides a command guide where the expansion of the number and complexity of models combining moderation and mediation is clearly portrayed (Hayes, 2012a). In this command guide, for instance, the hypothesized model for the present study (Figure 1) is graphically depicted in conceptual and statistical terms as "model 72", where "*X*" would stand for SS, "*Y*" for HRQL, "M_j" for internalizing and externalizing problems, "*W*" for age group and "*Z*" for gender. This multiple moderated mediation model allows the simultaneous testing of single and combined moderator effects (e.g. *X*W*, *X*Z*, *W*Z*, *X*W*Z*). Other models that were used in the present study, for examining single moderation (model 1), multiple moderation (model 3) and multiple mediation (model 4), are also included in the aforementioned command guide. All PROCESS analyses were run through a SPSS macro, with 5000 bootstrap samples being systematically drawn. In addition, mean centered products computed for moderation analyses, and gross motor function level was entered as covariate (0 = *no mobility limitations*, 1 = *with mobility limitations*), since it was significantly correlated with both self and proxy-reported HRQL (as recommended by Tabachnik & Fidell, 2007). All analyses were conducted for a 95% confidence interval, even though some marginally significant results (i.e. $p \leq .09$) were presented and/or commented for the purpose of clarifying a given sequence of analyses.

Results

Sample characteristics

With the exception of SES, homologous age and gender distributions were observed for children/adolescents in both samples (see Table 1). The majority of proxy respondents were mothers (>80%), and most cases were classified as pertaining to low/medium socioeconomic backgrounds (possibly due to the fact that school and healthcare contexts elected for sample collection were respectively public and semi-private institutions). As regards the clinical sample, more than half of the cases corresponded to milder forms of CP, including spastic subtypes (88.5%) with no limitations in walking (62.5%).

Differences in Social Support and Psychological Maladjustment and Inter-correlations between Variables

Regarding social support, when controlling for SES, significant differences were found between children and adolescents with CP and healthy controls, $F(1, 202) = 4.96$, $p = .03$, $\eta_p^2 = .02$ (see Table 2), but not between age, $F(1, 202) < .01$, $p = .97$, $\eta_p^2 = .00$, or gender groups, $F(1, 202) = 1.06$, $p = .30$, $\eta_p^2 = .01$.

Results on psychological maladjustment indicated the absence of statistically significant multivariate differences between children and adolescents with CP and healthy controls, controlling for SES, $F(4, 199) = 1.55$, $p = .19$, Wilks' Lambda = .97, $\eta_p^2 = .03$. Univariated effects for each dimension of psychological maladjustment were presented in Table 2. Multivariated effects of age, $F(4, 199) = 2.78$, $p = .03$, Wilks' Lambda = .95, $\eta_p^2 = .05$, and gender, $F(4, 199) = 3.53$, $p = .01$, Wilks' Lambda = .93, $\eta_p^2 = .07$, were found. Univariate analyses for age groups showed, however, only a marginally significant difference on parent-reported internalizing problems, $F(1, 202) = 2.90$, $p = .09$, $\eta_p^2 = .01$, with parents of adolescents ($M = 5.57$, $SD = 3.94$) reporting higher levels of internalizing symptoms than parents of children ($M = 4.55$, $SD = 3.40$). Regarding gender, univariate analyses indicated higher prevalence of externalizing problems, both self, $F(1, 202) = 10.81$, $p < .01$, $\eta_p^2 = .05$, and parent-reported, $F(1, 202) = 10.31$, $p < .01$, $\eta_p^2 = .05$, for boys ($M = 5.96$, $SD = 3.29$ for self-reports; $M = 6.80$, $SD = 3.88$ for proxy-reports), when compared to girls ($M = 4.42$, $SD = 3.07$ for self-reports; $M = 5.02$, $SD = 3.55$ for proxy-reports).

As presented on the right side of Table 2, moderate correlations were generally observed between social support, dimensions of psychological maladjustment, and HRQL.

Table 1. Socio-demographic and clinical characteristics of CP and control samples

	CP Clinical Sample		**Healthy Controls Sample**		
	Children/ Adolescents (*N* = 96)	**Parents** (*N* = 96)	**Children/Adolescents** (*N* = 118)	**Parents** (*N* = 118)	*Differences between Samples*[5]
Age (*M/SD*)	12.3 (2.8)	41.8 (6.7)	12.3 (3.0)	42.7 (5.3)	t = -.09; p = .93
Age Group (*n*/%)					
Children (8-12)	48 (50.0)	-	62 (52.5)	-	$\chi 2$ = .05; p = .82
Adolescents (13-18)	48 (50.0)		56 (47.5)		
Gender (n/%)					
Male	56 (58.3)	12 (12.5)	59 (50.0)	22 (18.6)	$\chi 2$ = 1.16; p = .28
Female	40 (41.7)	84 (87.5)	59 (50.0)	96 (81.4)	
Marital status: married **(*n*/%)**	-	72 (75.0)	-	99 (83.9)	-
SES[1] **(*n*/%)**					
Low	59 (61.5)		31 (26.3)		$\chi 2$ = 29.88; p = .00
Medium	23 (24.0)		65 (55.1)		
High	11 (11.5)		22 (18.6)		
Missing	3 (3.0)		-		
CP Type[2] **(*n*/%)**					
Spastic unilateral	48 (50.0)				
Spastic bilateral	37 (38.5)				
Dyskinetic	4 (4.2)				
Ataxic	2 (2.1)				
Missing	5 (5.2)				
GMFCS[4] **(*n*/%)**					
I	60 (62.5)				
II	13 (13.5)				
III	11 (11.5)				
IV	7 (7.3)				
V	3 (3.1)				
Missing	2 (2.1)				
Epilepsy (*n*/%)	12 (12.5)				
IQ (*M/SD*)	92.9 (17.8)				
Cognitive level (*n*/%)	31 (32.3)				
Borderline[3] (71-84)	13 (13.5)				
Missing					

Notes.
[1] Socio-economic status (SES) was determined using a classification system based on parents' job and educational level (Simões, 1994).
[2] Classification of CP subtypes according to the Surveillance of CP in Europe project (SCPE, 2000).
[3] Borderline Intellectual Functioning [V62.89], as defined in DSM-IV (APA, 1994).
[4] Levels of function according to the Gross Motor Function Classification System (GMFCS) – Expanded and Revised (Palisano, Rosenbaum, Bartlett, & Livingston, 2007).
[5]Results of comparison tests for children/adolescents' variables.

Table 2. Differences in internalizing/externalizing problems and social support and matrix of inter-correlations among variables (correlations for CP sample only)

	CP *(N = 93)*	**Healthy controls** *(N = 118)*									
	M (SD)	*M (SD)*	$F_{(1,202)}$	*p*	η_p^2	1	2	3	4	5	6
1. Self-reported internalizing problems	5.99 (3.50)	5.13 (3.06)	1.29	.26	.01						
2. Self-reported externalizing problems	5.47 (3.25)	5.07 (3.30)	.34	.56	.00	.45**					
3. Proxy-reported internalizing problems	6.23 (3.64)	4.12 (3.49)	4.79	.03	.02	.48**	.29**				
4. Proxy-reported externalizing problems	6.81 (3.83)	5.31 (3.71)	3.10	.08	.02	.20*	.53**	.30**			
5. Self-reported social support	42.89 (8.08)	46.63 (7.76)	4.96	.03	.02	-.39**	-.33**	-.30**	-.34**		
6. Self-reported HRQL	-	-	-	-	-	-.59**	-.38**	-.35**	-.35**	.50**	
7. Proxy-reported HRQL	-	-	-	-	-	-.37**	-.20*	-.58**	-.29**	.35**	.49**

Note. * $p \leq .05$, ** $p \leq .01$.

Multiple Moderated Mediation Models

Following the examination of a multiple moderated mediation model (see Figure 1), no significant conditional indirect effects were found for the relationships between social support, dimensions of psychological maladjustment and HRQL; however, when considering parents' reports, an interaction effect between Social Support*Age Group*Gender was found for the link between social support and externalizing problems (b = .42, p <.05). Given this fact, the moderating role of age group and gender in the relationship between social support and proxy-reported externalizing problems was subsequently explored (testing for model 3 in PROCESS),

with the significance of the conditional effect of Social Support*Age Group interaction observed only in girls (b = .37, p = .01). After restricting analyses for the sample of girls, age group was further examined as a single moderator in the same relationship (testing for model 1 in PROCESS), with a significant conditional effect of social support on externalizing problems observed only in female children (b = -.31, p <.001). Finally, PROCESS-generated data were used to graphically depict this conditional effect: as shown in Figure 2, the conditional effect of social support on externalizing problems was only significant for children, i.e., the externalizing problems decreased as social support increased only in female children (b = -0.311, t = -3.65, p < .001); for female adolescents, the slope did not differ significantly from zero, i.e., the conditional effect was not significant (b = 0.05, t = 0.58, p =.57).

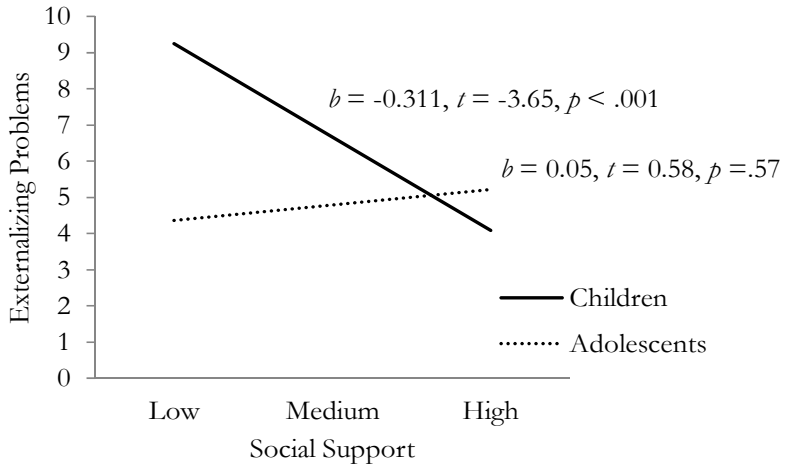

Figure 2. The moderating effect of age group in the association between social support and proxy-reported externalizing problems of child and adolescent girls with CP

Multiple Mediation Models

Since no conditional indirect effects were found, a simplified version of the initial model was tested. This latest version represented a multiple mediation model with no moderating variables (testing for model 4 in PROCESS). As presented in Table 3, both internalizing and externalizing problems were found to mediate the association between social support and HRQL. Moreover, this mediation effect was significant for the model based on self-reports only, as well as for the model combining self and proxy-reports.

Table 3. Summary of multiple mediation analyses for models including self and proxy-reported psychological maladjustment and HRQL (5000 bootstraps).

	Independent variable	Mediating variable	Dependent variable	Effect of IV on M	Effect of M on DV	Direct effect	Indirect effect		Total effect
				b (SE)	*b* (SE)	*b* (SE)	*b* (boot SE)		*b* (SE)
	(IV)	(M)	(DV)	(a)	(b)	(c')	(a*b)	95% CI	(c)
Self-reported psychological maladjustment and HRQL	Social Support	Internalizing problems	HRQL	-.15 (.04)**	-1.43 (.34)**	.46 (.14)**	.22 (.08)	(.09-.41)	.76 (.15)**
		Externalizing problems		-.12 (.04)**	-.63 (.36)[a]		.08 (.05)	(.01-.23)	
Proxy-reported psychological maladjustment and HRQL	Social Support	Internalizing problems	HRQL	-.11 (.04)*	-2.57 (.40)**	.31 (.18)[a]	.28 (.12)	(.05-.55)	.71 (.20)**
		Externalizing problems		-.16 (.05)**	-.76 (.37)*		.12 (.08)	(.01-.33)	

Notes. * $p \leq .05$, ** $p \leq .01$. [a] marginally significant ($p = .09$)

Discussion

Main findings of this study may be summarized as follows: first, children/adolescents with CP reported lower levels of social support in comparison to healthy, able-bodied controls; second, there were no significant differences in the levels of psychological maladjustment between those two groups; third, no conditional indirect effects were observed in the examination of a moderated mediation model; and finally, both dimensions of psychological maladjustment (i.e., internalizing and externalizing problems) mediated the association between social support and HRQL.

When compared to their healthy peers, children and adolescents with CP reported lower levels of social support. This difference was small, yet confirmed our first hypothesis. This finding is consistent with previous assertions of impaired social relationships in pediatric chronic conditions, and particularly in those affecting the central nervous system, such as CP (LaGreca et al., 2002). Given the fact that social support was assessed in terms of the individual's subjective satisfaction, the observation of lower levels of social support may reflect the existence of adverse social conditions (e.g. isolation) and/or negative subjective appraisals on the received social support. For this reason, interventions aimed at improving satisfaction with social support in children/adolescents with CP, may target their perceptions of adequacy and availability of support sources and/or the objective number of social contacts and activities in their lives.

Contrary to previous reports on the psychological adjustment of children/adolescents with CP (Brossard-Racine et al., 2012a, 2012b; Vles et al., 2012), our results infirmed the study's second hypothesis: there were no significant differences in self and parent-reported psychological maladjustment between children/adolescents with CP and their healthy peers. Interestingly, a similar unexpected finding was recently reported for the QL of children with CP, which was observed to be mostly analogous to the QL reported by children in the general population (Dickinson et al., 2007). We believe this unexpected finding may derive from the use of appropriate controls and the inclusion of self-reports (as recommended by Wallander, Schmitt, & Koot, 2001), rather than from the higher frequency of milder forms of CP in our sample, which are not necessarily related to better adjustment (Arnaud et al., 2008). Another plausible explanation is that these results may well depict what has been coined in literature as the "disability paradox" (Albrecht & Devlieger, 1999) or "response shift" (Sprangers & Schwartz, 1999). These notions account for those clinical cases where maladjustment would be

greatly expected by external observers, but it is not verified (or is even contradicted) through patients' reports. Response shift, for instance, has been described as an adaptation process to health stressors, and its further examination in the context of pediatric CP could greatly expand our current knowledge on the diversity of trajectories that may emerge during these children and adolescents' development.

Regarding the dimensions of internalizing and externalizing problems, results were in fair agreement with previous research and partially confirmed our third hypothesis: boys presented more externalizing problems than girls, but adolescents did not positively present more internalizing symptoms than children (Gortmaker, Walker, Weitzman, & Sobol, 1990; Yang, Li, Zhang, Tein, & Liu, 2008). The fact that gender differences in externalizing problems were evident through parents and their children's reports, somehow challenges the general assumption that parents are more reliable raters of their children's externalizing problems than children themselves (Youngstrom, Loeber, & Stouthamer-Loeber, 2000).

As regards the hypothesized moderated mediation model, the fact that no conditional indirect effects were observed, may well attest the model's adequacy in portraying a potential adaptation mechanism in children and adolescents with CP. Nevertheless, as suggested by our literature review, age and gender differences are likely to occur in the comparison of isolated variables or specific links between them. After all, this plausible claim was to be verified in our study, where age and gender were found to moderate the link between social support and parent-reported externalizing problems. This particular moderation effect may reflect the suggestion that social support is more influential on the level of externalizing problems in girls (Bender & Lösell, 1997), in addition to the observations that such problems are more common in children than in adolescents with CP (McCullough et al., 2011), and that statistical correlations with externalizing problems may only occur in proxy-reports (Janssen et al., 2010). However, given the small sample size in which these effects were detected, their interpretation should be merely assumed as exploratory.

Finally, we found support for a multiple mediation model in which the link between social support and HRQL was mediated by two dimensions of psychological maladjustment, namely internalizing and externalizing problems. The need to identify variables that may mediate the path from social support to QL outcomes has been stressed in literature (Helgeson, 2003) and, in general terms, the understanding of such mechanisms is potentially enriching for both theory and practice (Shrout & Bolger, 2002). Our results indicate that one of the ways by which social support may be linked to the HRQL of children/adolescents with CP, is through its

negative associations with psychological maladjustment. In terms of model reliability and clinical validity, it is noteworthy that such pattern of associations was consistent in both models accounting for self-reports only, and for concomitant child and parent-reports. Nevertheless, these findings do not exclude the existence of other mediating variables in the relationship between social support and HRQL (e.g. coping, health-related behaviors), and even more precisely, in the associations between social support and psychological maladjustment (e.g. social skills), and between psychological maladjustment and HRQL (e.g. psychosomatic reactions, stigma). Definitely, more research is needed to disentangle these varied patterns of associations between psychosocial determinants and HRQL outcomes.

Limitations and Strengths

The cross-sectional design of this study remains its major limitation. Although the direction of the relationship between variables was hypothesized, the study was based on a clear theoretical rationale and implemented reliable statistical procedures that allowed confidence in results for answering the research questions. Given the scarce literature on the theme, this cross-sectional study offers promising insights to be further examined in future research. Sampling frames may also stand as a relevant limitation: heterogeneous distribution in a number of clinical variables, including a higher frequency of milder CP forms, argues for additional caution in generalizing the observed findings. Besides, our clinical sample was recruited in tertiary healthcare institutions, which may be prone to some form of selection bias (McDermott et al., 1996).

Albeit these limitations, three distinctive features of our study are to be acknowledged as considerable strengths: first, it "gave voice" to children/adolescents with CP (Varni et al., 2005), while simultaneously including parents' reports in a dyadic perspective to outcome assessment (White-Koning et al., 2007), which has been more suggested than examined in research; second, it sought to integrate negative (i.e. psychological maladjustment) and positive (i.e. HRQL) dimensions in the assessment of adaptation outcomes; and last, it applied bootstrapping statistical procedures as a mean of model development for a pediatric group where psychosocial research is sparse, in order to clarify some of the mechanisms through which social support is likely to influence HRQL.

Clinical Implications and Future Directions

Findings from this study showed that social support was linked to HRQL both directly and indirectly, via internalizing and externalizing problems. In terms of clinical formulation, these results suggest that negative social support perceptions may impair the HRQL of children/adolescents with CP, through the deterioration of their psychological adjustment. In clinical practice, interventions targeting satisfaction with social support may assume a variety of forms (e.g. increasing participation, training social skills, enhancing positive family relationships), and may positively affect HRQL through the improvement of these child/adolescent's psychological adjustment (i.e., prevention or reduction of internalizing and externalizing symptoms). There is some evidence, for instance, on the efficacy of cognitive-behavioral interventions in reducing isolation and increasing social competence for certain pediatric populations (Barlow & Ellard, 2004), but the effectiveness of such interventions remains to be ascertained in children and adolescents with CP.

Longitudinal research is needed to clarify the directionality of the associations observed in this and other cross-sectional studies. Moreover, it would be important to further examine the occurrence of the "disability paradox" in pediatric CP, along with the identification of determinants (such as social support) related to that counterintuitive phenomenon. Finally, the comparison of adaptation patterns between children/adolescents with and without CP would be interesting to differentiate commonalities and specificities that may exist in the adaptation patterns of clinical and normative populations.

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Empirical Study III |

The Disability Paradox Revisited:
Quality of Life and Family Caregiving in Pediatric Cerebral Palsy

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The Disability Paradox Revisited: Quality of Life and Family Caregiving in Pediatric Cerebral Palsy

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Abstract

Parents who have children with cerebral palsy (CP) have been reported to have a more impaired QL and higher levels of burden than parents of typically developing children; however, little is known about the positive dimensions of their caregiving experience. In this study, WHOQOL-Bref and The Revised Burden Measure were administered to a sample of 105 parents of children/adolescents with CP (clinical group) and 117 parents of children/adolescents with no disabilities (control group). Despite the fact that parents of children with CP reported more Subjective Burden and less caregiving Uplifts, there were more similarities than differences in the variables compared between clinical and control groups. For parents of children with CP, the associations between Burden dimensions and QL, and between caregiving Uplifts and QL, were respectively moderate and weak. Caregiving Uplifts were found to moderate the links between Objective Burden and Psychological QL, and between Relationship Burden and Social QL. In addition, differential main effects of Burden dimensions and caregiving Uplifts were verified for Physical, Psychological and Social QL domains. These results highlight the adaptation variability of parents who have children with CP, as well as the importance of acknowledging caregiving uplifts as a resource that may attenuate the impact of burden on their QL.

Keywords: family caregiving; quality of life; cerebral palsy; burden; uplifts.

Introduction

The "disability paradox" was defined as the discrepancy between the objective limitations and suffering posed by certain disabilities, and the reasonable or excellent quality of life (QL) reported by some individuals living with them (Albrecht & Devlieger, 1999). In their attempt to explain why some individuals adapt well despite adverse health conditions, Albrecht and Devlieger suggested that, amidst a variety of factors related to positive outcomes, psychological growth and inner strength could provide a "balanced perspective on life" (p. 983), which in turn could maintain or improve QL for those individuals. Since considerable variability has been reported for adaptation outcomes of parents who have children with chronic health conditions or disabilities (King, King, Rosenbaum, & Goffin, 1999; Raina et al., 2004), the present study reveals a renewed interest in determining the extent of such variability, as well as in moving away from the simplistic cause-effect relationship between caregiving burden and parental maladjustment (Jones & Passey, 2004). A focus on parents as primary family caregivers of children with chronic conditions and disabilities, and the assumption of parental burdens as stressful caregiving circumstances, enables the redefinition of the so-called "disability paradox" as follows: "why do some of these parents report increased levels of caregiving burden, and still perceive a similar or superior QL, in comparison to those parents who care for healthy/able-bodied children?"

For the purpose of illustrating the assessment of this "disability paradox", cerebral palsy (CP) was specifically elected for this study because of the heterogeneity of forms it may assume, along its significant prevalence and related caregiving burden, thus maintaining the assumption of CP as an interesting prototype of childhood disability (Raina et al., 2004). The core question implied by the redefined "disability paradox" has been put forward by different authors, who were interested in explaining why (or how) some parents adapt better than others to specific demands imposed by the responsibility of caring for a child with a chronic condition or disability. As regards pediatric CP in particular, it has been suggested that the way parents cope with increased caregiving demands, may have implications on their physical and psychological health (Brehaut et al., 2004). In this line of thought, coping is assumed here as a process of cognitive and behavioral efforts to manage demands related to stress (Lazarus, 1993), which may well encompass positive reappraisals as means for reframing a stressful situation in order to acknowledge its positive features (Folkman, 1997).

The reiterated recommendation for placing an emphasis on the understanding the ways that these parents cope, with varying adaptation outcomes, with the care of their child (Beresford, 1994; Eiser, 1990), is particularly important within the pediatric context for two main reasons: first, little is known about the factors that may lead some parents to exhibit a pattern of resiliency (i.e. positive adaptation following or coexisting with adverse circumstances) (Ha, Hong, Seltzer, & Greenberg, 2008); second, such emphasis could improve the adequacy and specificity of current intervention practices, for which a scarceness of evidence-based literature has been acknowledged (Anderson & Davis, 2011). Additionally, research on this topic is urgently needed because the quality and amount of care provided by these parents to their children represents a significant reduction of public healthcare costs; however, deterioration of their QL may have serious negative consequences such as decreased work productivity, increased healthcare costs for the caregiver, and impairment of their child's adjustment (Davis et al., 2010; Hatzmann, Maurice-Stam, Heymans, & Grootenhuis, 2009).

There were several reasons for our interest in revisiting the notion of "disability paradox": first, the idea that a pediatric chronic condition not only affects the child/adolescent who has it, but also other family members, particularly their parents (Kazak, 1989); second, the well-established occurrence of considerable heterogeneity in parental adaptation, which challenges the potential of approaching parental stress in those situations from an exclusive deficit model perspective (Yau & Li-Tsang, 1999); third, the possibility of different types of burden and benefit finding coexisting in the subjective experience of raising a child with a chronic condition or disability (Green, 2007); finally, notwithstanding the acknowledged value of a non-categorical approach to illness/disability adaptation processes (Wallander, Pitt, & Mellins, 1990), differences in parental stress and the corresponding professional help are likely to exist across a variety of medical diagnoses, including CP (Wang & Jong, 2004).

In their original article, besides presenting a sharp description of the "disability paradox", Albrecht and Devlieger (1999) further explained the existence of such paradox in two forms: on the one hand, the discrepancy between serious self-reported limitations and daily adverse experiences, and the good or excellent QL perceived by people with disabilities or chronic health conditions; on the other hand, the divergence between the negativistic views hold by general public and health professionals about those peoples' daily existence, and the fact they report a good or excellent QL. These two forms may be delineated within the present notion of the "disability paradox revisited": indeed many parents cope and adapt successfully to stressful pediatric caregiving demands (Yau & Li-Tsang, 1999), and health professionals may portray the

same prejudices about disability/disease that are prevalent in society, thus labeling some families' positive discourse as unrealistic or confusing, and underscoring their adaptation levels (Green, 2007; Larson, 1998; Yau & Li-Tsang, 1999).

One of the possible mechanisms underlying the "disability paradox" has been termed in literature as "response shift", describing "a change in the meaning of one's self-evaluation of QL as result of changes in internal standards, values and the conceptualization of QL" (Sprangers & Schwartz, 1999, p.1509); "response shift" was described as a mediator of an individual adaptation process, while explaining the paradox of a stable self-reported QL in face of life-threatening diseases or disabilities. In order to further conceptually map our study on the "disability paradox revisited", models deriving from stress-coping theory were preferred because of their central tenet stating that the ways people perceive, input meaning and cope with stress and adversity, differentially influence adaptation (Folkman, 1997). The "disability-stress-coping model" is one of such examples, and was developed to encompass the adjustment continuum experienced by families of chronically ill children (Wallander & Varni, 1998). In this model, risk factors (i.e. disease/disability parameters, functional dependence and psychosocial stressors) and resistance factors (i.e. intrapersonal and socio-ecological factors) are hypothesized to act in complex interplays to determine adaptation outcomes. Within this broad theoretical framework, positive meanings, benefits or uplifts attributed to caregiving process have been pointed out as personal resources linked to successful adaptation (Larson, 2010); complementarily, the emotions elicited by positive meaning finding have been suggested not only to result from certain coping strategies, but also to sustain coping process itself (Folkman, 1997). In fact, even if it seems rather consensual that caring for a child with a chronic condition or disability may become burdensome, high well-being, personal growth and uplifts may coexist and/or derive from challenging caregiving demands (Gupta & Singhal, 2004; Larson, 2010). A similar claim was underlined by Tedeschi and Calhoun (2004), when exploring a model for posttraumatic growth, defined as the occurrence of positive change resulting from struggling with challenging life crises; these authors sustained that such growth could coexist with significant psychological distress, thus suggesting the existence of a ramifying meaning attributed to stressful events.

Such "ramifying meanings" and "outcomes variability" have not been properly addressed, or at least demonstrated, in the research literature on pediatric family caregiving. The adoption of sensitive measurements of the whole range of adaptation (i.e. encompassing physical, mental and social functioning) (Wallander et al., 1990), led to a recent emergence of studies on the QL of parents who have children with chronic conditions or disabilities. These

parents have been reported to be at risk for an impaired QL (Hatzmann et al., 2009), and to present a marginally poorer psychological well-being, compared to parents with healthy/non-disabled children (Ha et al., 2008). Furthermore, socioeconomic status (SES) has been observed to influence these parents' coping and adaptation (Raina et al., 2004; Yau & Li-Tsang, 1999).

Pediatric family caregiving burden or stress has been typically studied as a risk factor within the "disability-stress-coping model", even if its sources have rarely been described (Wallander & Marullo, 1997). Given the assumption that certain types of burden may differentially affect key outcomes, a multidimensional approach to burden has been argued (Savundranayagam, Montgomery, & Kosloski, 2011). Objective and emotional burdens of care have been commented to be particularly increased in these parents, with greater levels of stress related to child's older age (Ha et al., 2008) and severity of impairments (Yau & Li-Tsang, 1999). In fact, it has been suggested that increased objective burden may be determined by socio-structural constraints related to caregiving (Green, 2007). Some authors found evidence for a negative association of burden (and not disability objective parameters) with psychological adjustment of mothers who cared for children with chronic physical conditions (Horton & Wallander, 2001); moreover, maternal stress was shown to be uniquely associated with maternal mental health, but not physical or social functioning, even when controlling for demographic and clinical variables (Wallander et al., 1990). Burden and caregiving stress have been hypothesized to influence parental QL via intrapsychic and coping factors (Raina et al., 2004), but research on the role of potential buffers or protective factors that may alleviate their impact is definitely lacking.

Although not abundant, research on burden has been conducted to a much larger extent than the one on positive dimensions of parents' caregiving experience (such as personal growth, benefit finding and uplifts). Current studies indicate the possibility of growth for families of children with disabilities (Yau & Li-Tsang, 1999), and draw attention to mothers' common perception of valuable benefits in having a child with a disability, despite of the strain imposed by objective burdens (Green, 2007). Indeed Gupta and Singhal (2004) commented that positive perceptions were frequent in parents of children with disabilities, and included a variety of themes such as the child as a source of happiness; the child providing a challenge or opportunity to learn and develop; strengthened intimate relationships; increased personal strength and spirituality, and a changed, balanced perspective in life. Curious enough, most of these positive perceptions were coincident to the five domains of posttraumatic growth proposed by Tedeschi and Calhoun (2004). It has been suggested that finding benefits in the caregiving experience may have a positive impact on the caregiver's emotional and physical health (Green, 2007), eventually

through the promotion of psychological flexibility or the regulation of the deleterious physiological effects of stress (Gupta & Singhal, 2004), but most of these assertions remain untested for pediatric populations. However, although significant subjective burden tends to decrease the perceived benefits of caring (Green, 2007), pediatric family caregivers with high well-being have been reported to generate positive emotions by using meaning-making reappraisals, such as the experience of personal growth, caregiving uplifts and benefit finding (Larson, 1998; 2010).

In order to improve the study of negative and positive dimensions of pediatric family caregiving, we considered worthwhile the integrated analysis of both variables within a risk-resilience framework (Wallander & Varni, 1998). In the scope of the broader concept of "adaptation", resilience has been defined as a "dynamic process encompassing positive adaptation within the context of significant adversity" (Luthar, Cicchetti, & Becker, 2000). At this point, it is also noteworthy that despite the frequent interchangeable use of "adaptation" and "adjustment" terms, we endorse the distinction between "adaptation" as an overall process, and "adjustment" as a specific outcome (Thompson et al., 1994). From this standpoint, our literature review suggested that, in addition to burden, caregiving uplifts should be studied as QL predictors (Larson, 2010), and particularly as resources or protective factors (Gupta & Singhal, 2004). According to these perspectives, the role of caregiving uplifts could be examined in two analytical levels: first, as a single determinant of parents' QL (i.e. a main effect), and second, as a moderator in the association between burden and QL (i.e. an interaction effect). Moderator variables affect the strength and/or direction of the relation between a predictor and an outcome, and their analysis became popular in the study of resilience in pediatric contexts (Rose, Holmbeck, Coakley, & Franks, 2004). If one assumes the experience of caregiving uplifts as a protective or resource factor, one will expect it to modify, ameliorate, or alter the impact of burden on parents' QL (Armstrong, Birnie-Lefcovitch, & Ungar, 2005). For the present purpose, a further distinction is worthy to note: while a "protective factor" serves its protective role only in the context of adversity, a "resource factor" has a positive impact on the outcome regardless of the presence or absence of adverse conditions (Rose et al., 2004).

Research addressing the QL and caregiving experience of parents who have children with CP is scarce and mostly based on heterogeneous samples, which limits the conclusions that may be drawn about CP specifically (Britner et al., 2003). Moreover, the few studies directly addressing CP are characterized by negative approaches to family adaptation (Magill-Evans, Darrah, Pain, Adkins, & Kratochvil, 2001), unclear conceptual frameworks with no regard for

resiliency (Lin, 2000), and an excessive focus on families of young children that largely ignores the adolescence period (Magill-Evans et al., 2001).

Results from studies on the adaptation outcomes of these parents are diverse: some studies found few differences in adaptation patterns (Britner, Morog, Pianta, & Marvin, 2003) and a similar life satisfaction (Magill-Evans et al., 2001), in comparison to parents of typically developing children; other studies observed poorer mental health (Florian & Findler, 2001), and more psychological and physical health problems (Brehaut et al., 2004), in comparison to parents of children without physical disabilities and other family caregivers, respectively. Complementarily, a qualitative study adopting a grounded theory framework verified an impaired QL in all domains (i.e. physical, psychological and social) for parents caring for a child with CP (Davis et al., 2010).

Regarding the research on burden, it has been argued that higher levels of parental stress in CP do not necessarily equate to lower levels of adaptation (Rentinck, Ketelaar, Jongmans, & Gorter, 2006); nevertheless, caregiving demands were shown to strongly influence physical and psychological health of caregivers of children with CP (Raina et al., 2005). Increased stress and psychological risk have been reported for mothers of children with CP (Florian & Findler, 2001), and issues of relationship burden (i.e. child-related demands and behavior) have been suggested to significantly affect parental stress and well-being (Raina et al., 2005; Wang & Jong, 2004). The need for examining parental adaptation differences between child's age groups has been acknowledged (Florian & Findler, 2001), with the existing literature suggesting better adaptation in families with younger school-aged children, than in families with adolescents (Lin, 2000). For mothers of children with CP, child's older age and family economic conditions were related to increased maternal stress (Mobarak, Khan, Munir, Zaman, & McConachie, 2000). Although disability severity may be an important predictor of parental stress and mental health (Rentinck et al., 2006), it has been suggested that parents of higher functioning children may present higher levels of psychological burden, possibly derived from conflicting expectations (Manuel, Naughton, Balkrishnan, Smith, & Koman, 2003); in another study, the interaction between child's level of impairment and partner support significantly predicted maternal stress (Button, Pianta, & Marvin, 2001). Finally, demonstrating a considerable research gap, we found no references on the study of personal growth, caregiving benefits or uplifts in parents of children with CP.

Our study adopted a balanced and multidimensional approach to the adaptation of parents who have children with CP, through the integration of both positive and negative

dimensions of family caregiving, and the selection of multidimensional burden and QL measures. In order to succinctly illustrate the notion of the "disability paradox revisited", the objectives of our study were: (1) to characterize the QL and the caregiving Burden and Uplifts experienced by parents of children with CP, in comparison to parents of healthy/able-bodied children/adolescents; (2) to analyze the associations between caregiving Burden and Uplifts and the QL of those parents, and (3) to assess the moderating role of caregiving Uplifts in the relationship between Burden dimensions and QL domains.

Accordingly, the following theoretically-driven hypotheses were outlined: first, parents of children with CP would report poorer physical, psychological and social QL, in comparison to parents of children with no chronic physical condition; second, parents of children with CP would present higher levels of Objective, Subjective and Relationship Burdens, and lower levels of caregiving Uplifts, in comparison to parents of children with no chronic physical condition; third, Burden dimensions and caregiving Uplifts would present moderate (negative and positive, respectively) correlations with all QL domains; fourth and last, caregiving Uplifts would moderate the associations between Burden dimensions and Psychological QL (i.e. parents with higher levels of Uplifts would report a better QL across different burden conditions, than those experiencing less caregiving Uplifts).

Method

Participants

The clinical group for this study (N = 105 parents of children/adolescents with CP) was collected in ten Portuguese Cerebral Palsy Associations (social institutions of tertiary health care) between July 2010 and July 2011. These parents were assigned to the study if they met the following criteria: (1) having a child aged between 8 and 18 years old, with a diagnosis of CP established by a physician, and a minimum intelligence quotient (IQ) of 70; (2) being the primary family caregiver of the child/adolescent with CP, as suggested by the largest amount of time dedicated to child's health issues and care. In those situations where informal health care was perceived as equally distributed between parents, the one who accompanied the child at the time of assessment protocol administration was included. Cases where results from formal assessments of child's IQ were not available (n = 13) were still included in the study, if their child did not present significant cognitive delay, as indicated by gross evaluation of the child's cognitive abilities, and the simultaneous absence of adaptations to school curricula. The inclusion criterion based on child's IQ was considered, because children/adolescents were to simultaneously participate in a parallel study where their self-reports were required. According to the aforementioned criteria, 161 parents of children/adolescents with CP were assigned to participate in the study; subsequently, the further exclusion of 56 parents was based on the following reasons: seven refused to participate; forty-seven did not visit the institutions during the established period for data collection, and two cases were related to children living in foster care placement.

Parents for a group of controls (N = 117) were recruited in two public schools of Coimbra district, between January and June 2010, considering two inclusion criteria: (1) having a child aged between 8 and 18 years old with no chronic health condition or disability; (2) being the parent who spent more daily time with the child/adolescent. In order to achieve the intended sample size, a total of 124 parents were assigned to participate in the study (with no matching procedures), but seven were excluded afterwards: two parents refused to participate; four did not return the questionnaires, and one parent did not report on all the measures.

Measures

The World Health Organization Quality of Life Assessment – Brief Version (WHOQOL-BREF). WHOQOL-BREF questionnaire comprises 26 items addressing four QL domains: Physical, Psychological, Social Relationships (henceforth: "Social" domain), and Environmental (Vaz-Serra et al., 2006). The Physical domain (e.g. "Do you have enough energy for everyday life?") integrates the facets of pain and discomfort, energy and fatigue, sleep and rest, dependence on medication, mobility, activities of daily living, and working capacity; the Psychological domain (e.g. "How satisfied are you with yourself?) assesses the facets of positive and negative feelings, self-esteem, thinking, learning, memory and concentration, body image, and spirituality, religion and personal beliefs; and lastly, the Social domain (e.g. "How satisfied are you with your personal relationships?") comprises the facets of personal relationships, sex and social support. The subscale assessing the Environmental domain was not used in this work, because it was not aligned with our study's specific aims; we also did not use the general QL facet, provided by two questions, because of its unacceptable internal consistency in the overall sample (a = .50). WHOQOL-BREF items are to be answered within a 5-point scale ranging from 1 (*very poor/very dissatisfied/not at all/never*) to 5 (*very good/very satisfied/extremely/completely*); standardized scores (0-100) for each domain are then computed, with the lowest scores portraying the most impaired QL. Adequate internal consistency values were observed for our general and clinical samples: Physical (a = .82/.84); Psychological (a = .81/.80), and Social QL domain (a = .76/76).

The Revised Burden Measure. This self-report questionnaire includes distinct, but complementary burden and uplifts measures (Montgomery et al., 2006). Although originally developed for geriatric caregivers, this measure has been successfully applied to pediatric populations (Crespo, Carona, Silva, Canavarro, & Dattilio, 2011). The instrument includes three burden subscales: Objective Burden (e.g. "Have your caregiving responsibilities left you with almost no time to relax?"); Subjective Burden (e.g. "Have your caregiving responsibilities created a feeling of hopelessness?"), and Relationship Burden (e.g. "Have your caregiving responsibilities caused conflicts with your relative?"). Additionally, a measure of caregiving Uplifts is contained within the instrument (e.g. "Have your caregiving responsibilities given your life more meaning?"). Caregiving Uplifts represent a positive psychological state related with caregiving, and include such things as the direct enjoyment from caregiving tasks, an improved relationship with the child, and a generalized positive affect. Responses for all items are to be provided within a 5-point scale (1 = *Not at all*; 5 = *A great deal*), with scores ranging between 6 and 30 for Objective Burden and caregiving Uplifts, and between 5 and 25 for Subjective and Relationships

burdens. Adequate internal consistency values were obtained in our general and clinical groups, for all subscales: Objective Burden (*a* = .82/.84); Subjective Burden (*a* = .81/.80), Relationship Burden (*a* = .76/76) and caregiving Uplifts (*a* = .76/76).

Procedure

After getting authorizations from the Direction Boards of participating Portuguese Cerebral Palsy Associations, cases that met the inclusion criteria were identified. Informed consents were then obtained from parents who agreed to participate in the study. During their visit to the institution, parents completed the questionnaires in a room provided for the purpose, with the permanent assistance of a psychologist or social worker acquainted with the research project.

Regarding the collection of control group, authorizations were obtained from the Direction Boards of participating schools. A sufficient number of classes were selected to reasonably achieve the intended sample size (nearly 100 participants). These classes were then visited by a researcher who delivered informed consents and assessment protocols to students, who in turn were expected to return them completed by their parents, nearly one week after.

Statistical Analysis

Statistical analyses were conducted with Statistical Package for the Social Sciences (SPSS, v.20). Missing data, as they were random and low level, were handled by individual mean score substitution, except for socio-demographic and clinical data. The clinical and control groups were characterized with descriptive statistics for socio-demographic and clinical variables, and the homogeneity of characteristics between clinical and control groups was examined with comparison tests (independent samples *t*-tests and chi-squared tests, for continuous and categorical variables respectively). Reliability of the measures used in this study was assessed through the calculation of their Cronbach's alphas, which were then interpreted as indicators of acceptable (≥.70) or optimal (≥.80) internal consistency (Nunally & Bernstein, 1994).

Differences between conditions (parents of children/adolescents with CP vs. parents of children/adolescents without disabilities) and between age groups (parents of children vs. parents of adolescents) were tested with two multivariate analyses of covariance (two-way MANCOVA), one with the three QL domains and another one with the dimensions of caregiving Burden and Uplifts as dependent variables. These analyses were controlled for SES,

by including it as covariate, since there was a significant discrepancy in this variable distribution between clinical and control groups. When multivariate effects were significant, univariate analyses were performed to examine which dimensions of QL and caregiving Burden or Uplifts significantly differed between groups. Effect-size measures (partial Eta squared) were presented for the comparison analyses, considering $\eta_p^2 \geq .01$; $\eta_p^2 \geq .06$; and $\eta_p^2 \geq .14$ as small, medium and large effects, respectively (Cohen, 1988).

In order to assess associations between variables, Pearson's correlation coefficients were computed, and their strength of association interpreted according to the following classification parameters: ±.10 - ±.29 (weak); ±.30 - ±.49 (moderate); ±.50 - ±1.0 (strong) (Cohen, 1988).

Moderation effects were examined with multiple regression analyses performed in SPSS. Prior to the conduction of moderation analyses, independent and moderator variables were mean centered for products calculation, and afterwards, covariates were entered in the first block, and interaction product terms in the last block of regression analyses. Following the identification of significant interaction effects, the simple slope procedure (i.e. creating three groups based on the mean levels of the moderator variable) was selected for probing possible moderator effects (Aiken & West, 1991). Post-hoc probing of interaction effects between two variables (one independent and one moderator) was necessary for specifying the conditions under which a predictor was significantly related to the outcome (i.e. whether either of the simple slopes was significantly different from zero) (Holmbeck, 2002). This procedure was optimized with the utilization of PROCESS (Hayes, 2012a), a computational tool that provides a SPSS macro for the examination of diverse statistical models that are numbered and analytically described in a command guide supplied for the effect (e.g. simple moderation is represented as "model 1"). These statistical procedures were sequentially used because we were interested in testing both main and interaction effects, and also in obtaining the sophisticated outputs delivered by PROCESS, which facilitated the graphical depictions of significant interaction effects. For all the examined models, different covariates were introduced to statistically account for shared associations between variables (Hayes, 2012b) and thus explain additional variability in the outcome variables (MacKinnon & Luecken, 2008). These covariates were clinical and socio-demographic variables that were entered because of their significant associations with the outcome variables (Tabachnik & Fidell, 2007): child's age group and function level (0 = *no mobility limitations*, 1 = *with mobility limitations*) were entered as covariates for analyses with the outcome variable of Physical QL; SES and child's age for Psychological QL, and child's age for Social QL. Effect sizes of main and interaction (moderating) effects derived from the regression

analyses were based on the values of R^2, which were then classified as small ($R^2 \geq .02$), medium ($R^2 \geq .13$) and large ($R^2 \geq .26$) (Cohen, 1992). A minimum confidence interval of 95% was considered for all the analyses performed in this study.

Results

Sample Characteristics

As presented in Table 1, the collected sample mainly included mothers (more than 80% of the cases), who were married (nearly 80% of the cases). Except for SES, no significant differences were observed for the socio-demographic variables between clinical and control groups. Data on children's variables, such as age group and gender, were homogenously distributed across both samples. Regarding the clinical group, the majority of CP cases implied, were spastic forms (≈ 89%), with no limitations in walking (63.8%).

Table 1. Socio-demographic and clinical characterization of clinical and control samples

	Parents of Children with CP (*n* = 105)	Parents of Children without Disabilities (*n* = 117)	Differences between Samples[1]
Parents' Variables			
Age (*M*/*SD*)	41.5 (6.5)	42.8 (5.2)	$t = 1.56$; $p = .12$
Gender (n/%)			
Male	12 (11.5)	22 (18.8)	$\chi^2 = 2.23$; $p = .14$
Female	93 (88.5)	95 (81.2)	
Marital status:			
Married **(*n*/%)**	79 (76.0)	99 (84.6)	$\chi^2 = 2.63$; $p = .11$
SES[2] (*n*/%)			
Low	67 (63.8)	31 (26.5)	$\chi^2 = 34.77$; $p < .01$
Medium-High	34 (32.4)	86 (73.5)	
Missing	4 (3.8)	-	
Children's Variables			
Age (*M*/*SD*)	12.0 (2.9)	12.3 (3.0)	$t = .83$; $p = .41$
Age Group (*n*/%)			
Children (8-12)	59 (56.2)	61 (52.1)	$\chi^2 = .47$; $p = .49$
Adolescents (13-18)	46 (43.8)	56 (47.9)	
Gender (n/%)			
Male	63 (60.0)	59 (50.4)	$\chi^2 = 2.05$; $p = .15$
Female	42 (40.0)	58 (49.6)	
CP Type[3] (*n*/%)			
Spastic unilateral	53 (50.5)		
Spastic bilateral	40 (38.1)		
Dyskinetic	4 (3.8)		
Ataxic	3 (2.9)		
Missing	5 (4.8)		
GMFCS[4] (*n*/%)			
I	67 (63.8)		
II	15 (14.3)		
III	12 (11.4)		
IV	6 (5.7)		
V	3 (2.9)		
Missing	2 (1.9)		

[1] Results of homogeneity testing between clinical and control samples.

[2] SES levels were determined using a classification system based on parents' job and educational level (Simões, 1994), followed by variable dichotomization.

[3] According to the classification proposed by the Surveillance of CP in Europe project (SCPE, 2000).

[4] Levels of function according to the Gross Motor Function Classification System (GMFCS) – Expanded and Revised (Palisano, Rosenbaum, Bartlett, & Livingston, 2007).

Differences in QL, Burden Dimensions and Caregiving Uplifts

Regarding QL, the two-way MANCOVA presented no significant multivariate effect of condition, indicating that there were no differences in QL between parents of children/adolescents with CP and the control group, Wilks' Lambda = .98, $F_{(3, 210)} = 1.75$, $p = .16$, $\eta_p^2 = .02$. Children's age had a significant multivariated effect on parents' QL, Wilks' Lambda = .93, $F(3, 210) = 4.92$, $p < .01$, $\eta_p^2 = .07$, specifically on the Psychological domain, where parents of younger children presented better Psychological QL than parents of adolescents (see Table 2). No multivariated interaction effects of condition and age group were found on parents' QL, Wilks' Lambda = .97, $F_{(3, 210)} = 1.87$, $p = .14$, $\eta_p^2 = .03$. The two-way MANCOVA for the dimensions of Burden and caregiving Uplifts, indicated significant multivariate effects of condition, Wilks' Lambda = .80, $F_{(4, 209)} = 12.96$, $p < .01$, $\eta_p^2 = .20$, and age group, Wilks' Lambda = .93, $F_{(4, 209)} = 3.67$, $p = .01$, $\eta_p^2 = .07$, as well as of the interaction between the two factors, Wilks' Lambda = .94, $F_{(4, 209)} = 3.36$, $p = .01$, $\eta_p^2 = .06$. The univariate analyses, presented in Table 2, showed that parents of children/adolescents with CP reported more Subjective Burden and less caregiving Uplifts than parents of children/adolescents without disabilities, and parents of younger children experienced more caregiving Uplifts than parents of adolescents. Univariate analyses for the interaction effects indicated that parents of adolescents with CP had more Objective Burden than parents of young children with CP, whereas for the control sample, parents of young children reported more Objective Burden than parents of adolescents.

Correlations between Burden Dimensions, Caregiving Uplifts and QL Domains

Subjective Burden was observed to be moderately correlated with Physical and Social QL domains, and strongly correlated with Psychological QL. Weak to moderate associations were found between Relationship and Objective Burdens, and QL domains. Caregiving Uplifts were weakly correlated with QL domains, and had no significant associations with Burden dimensions (see Table 3). Given the fact that QL domains targeted different dimensions of the same construct, their inter-correlations were accordingly strong.

Table 2. Differences in QL, Burden dimensions and caregiving Uplifts between clinical and control samples

	Parents of Children with CP		**Parents of Children without Disabilities**										
	Children (n = 56)	**Adolescents** (n = 44)	**Children** (n = 61)	**Adolescents** (n = 56)	Condition effects (CP vs. control sample)			Age group effects (children 8-12 vs. adolescents 13-18)			Interaction effects (condition X age group)		
	M (SD)	M (SD)	M (SD)	M (SD)	$F_{(1,212)}$	p	η_p^2	$F_{(1,212)}$	p	η_p^2	$F_{(1,212)}$	p	η_p^2
Quality of Life													
Physical QL	77.36 (13.05)	70.13 (19.13)	74.53 (13.90)	76.40 (13.44)	0.03	.86	.00	1.73	.19	.01	4.32	.04	.02
Psychological QL	73.36 (11.64)	65.15 (16.96)	76.23 (11.03)	71.50 (13.92)	1.94	.17	.01	**12.73**	**< .01**	**.06**	.61	.44	.00
Social QL	71.88 (14.78)	65.34 (19.60)	72.27 (15.12)	71.73 (15.54)	2.34	.13	.01	2.56	.11	.01	1.89	.17	.01
Caregiving Burden and Uplifts													
Relationship Burden	9.05 (3.88)	7.70 (3.76)	8.80 (3.47)	8.43 (3.74)	0.07	.79	.00	2.87	.09	.01	.86	.36	.00
Objective Burden	13.16 (4.92)	13.32 (5.13)	14.28 (5.09)	12.05 (4.89)	1.14	.29	.01	2.27	.13	.01	**3.78**	**.05**	**.02**
Subjective Burden	11.66 (4.62)	12.82 (4.78)	10.10 (3.66)	9.13 (4.45)	**17.22**	**< .01**	**.08**	0.03	.88	.00	3.24	.07	.02
Caregiving uplifts	21.07 (4.02)	20.00 (5.12)	24.26 (3.84)	22.30 (4.92)	**20.11**	**< .01**	**.09**	**6.21**	**.01**	**.03**	.43	.51	.00

Table 3. Matrix of inter-correlations among variables for parents of children with CP

	Physical QL	Psychological QL	Social QL	Relationship Burden	Objective Burden	Subjective Burden
Psychological QL	.71**					
Social QL	.54**	.61**				
Relationship Burden	-.26**	-.35**	-.26**			
Objective Burden	-.42**	-.31**	-.29**	.49**		
Subjective Burden	-.46**	-.56**	-.35**	.56**	.62**	
Caregiving Uplifts	.13**	.29**	.26**	-.09	-.02	-.14

* $p \leq .05$, ** $p \leq .01$

Main and Interaction Effects of Burden Dimensions and Caregiving Uplifts on Parents' QL

Results from regression analyses examining main and interaction (moderating) effects of caregiving Burden and Uplifts on parents' QL are detailed in Table 4. No main or interaction effects were found for caregiving Uplifts on Physical QL, but Relationship (b = -1.07, $p < .01$), Objective (b = -1.26, $p < .001$) and Subjective (b = -1.34, $p < .001$) Burdens respectively explained 6.4%, 12.5% and 15% of the variance in this QL domain.

As graphically depicted in Figure 1, caregiving Uplifts were found to moderate the negative association between Objective Burden and Psychological QL, $F(5, 96) = 8.15$, $p < .001$, $R^2 = .30$, with those parents who acknowledged medium (b = -0.82, t = -3.17, $p < .01$) to high (b = -1.34, t = -3.71, $p < .001$) levels of Uplifts, reporting a less impaired QL than those experiencing low levels of Uplifts (b = -0.29, t = -.90, p = .37). This moderating effect of caregiving Uplifts was far more evident under low to medium Burden conditions, while tending to decrease in situations of high Objective Burden. Caregiving Uplifts were also found to have a significant main effect on Psychological QL, along with Relationship Burden, $F(5, 96) = 8.36$, $p < .001$, $R^2 = .30$], and Subjective Burden, $F(5, 96) = 12.68$, $p < .001$, $R^2 = .40$.

As illustrated in Figure 2, a moderating effect of caregiving Uplifts was observed in the negative association between Relationship Burden and Social QL, $F(4, 101) = 5.91$, $p < .001$, $R^2 = .19$: parents who experienced medium levels of Uplifts, reported a less impaired QL under conditions of increased Relationship Burden (b = -1.10, t = -2.74, p <.01), when compared to those who experienced low levels of caregiving Uplifts (b = -2.15, t = -3.32, p

=.001). Moreover, parents reporting high levels of caregiving Uplifts seemed to benefit from a relative stability in their Social QL across different levels of Relationship Burden (*b* = -.05, *t* = -.08, *p* = .93), when compared to parents reporting low to medium levels of Uplifts. In other words, the association between Relationship Burden and Social QL was significant only for individuals with low to medium levels of caregiving Uplifts. Finally, caregiving Uplifts were positively related to Social QL, along with Objective Burden, $F(4, 101) = 4.78$, $p < .001$, $R^2 = .16$, and Subjective Burden, $F(4, 101) = 5.12$, $p < .001$, $R^2 = .17$.

Table 4. Regression analyses (main and interaction/moderating effects) for parents of children with CP

	Dependent variables					
	Physical QL		Psychological QL		Social QL	
	B (SE)	ΔR^2	*B* (SE)	ΔR^2	*B* (SE)	ΔR^2
Relationship Burden						
Child's age[a]	-1.27 (.54)*/ -4.82 (3.21)	.076*	-1.42(.44)**/ 5.53 (2.68)*	.117**	-1.20 (.54)*	.042*
Relationship Burden	-1.07 (.40)**	.064**	-1.19 (.33)***	.102**	-1.11 (.40)**	.063**
Uplifts	.41 (.34)	.010	.91 (.28)**	.066**	.95 (.34)**	.047*
Relationship B. x Uplifts	.07 (.11)	.004	.14 (.09)	.018	.23 (.11)*	.038*
Total R^2	.16		.30		.19	
Adjusted R^2	.11		.27		.16	
F (final model)	3.59**		8.36***		5.91***	
Objective Burden						
Child's age[a]	-1.08 (.51)*/ -2.81 (3.13)	.076*	-1.14 (.44)*/ 7.72 (2.70)**	.117**	-1.00 (.55)	.042*
Objective Burden	-1.26 (.30)***	.125***	-.80 (.26)**	.060**	-.85 (.32)**	.060*
Uplifts	.30 (.33)	.013	.78 (.29)**	.083**	.84 (.36)*	.055*
Objective B. x Uplifts	-.08 (.06)	.015	-.12 (.05)*	.038*	-.03 (.06)	.002
Total R^2	.23		.30		.16	
Adjusted R^2	.19		.26		.13	
F (final model)	5.85***		8.15***		4.78**	
Subjective Burden						
Child's age[a]	-.85 (.52)/ -5.08 (3.07)	.076*	-.90 (.42)*/ 6.45 (2.48)*	.117**	-.87 (.55)	.042*
Subjective Burden	-1.34 (.31)***	.15***	-1.43 (.25)***	.218***	-.95 (.33)**	.082**
Uplifts	.29 (.32)	.006	.81 (.26)**	.060**	.77 (.34)*	.042*
Subjective B. x Uplifts	-.02 (.05)	.001	.03 (.04)	.002	.03 (.06)	.002
Total R^2	.23		.40		.17	
Adjusted R^2	.20		.37		.14	
F (final model)	5.97***		12.68***		5.12**	

Note. The unstandardized regression coefficients (*B*) concern the analyses in which all main and interaction effects were entered (last step).

[a] Child´s age was entered as covariate in all regression analyses performed, along with function level (for Physical QL) and SES (for Psychological QL).

* $p < .05$; ** $p < .01$; *** $p < .001$

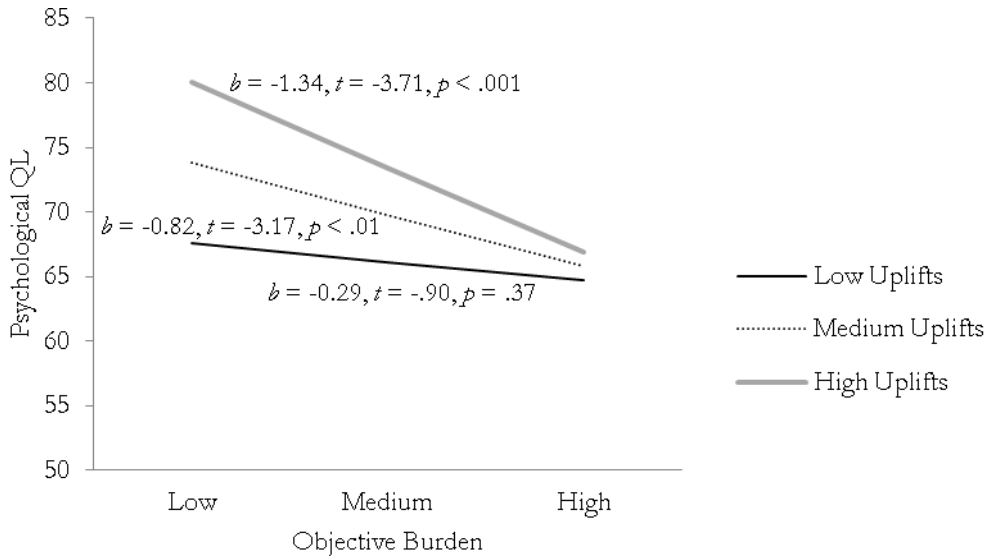

Figure 1. The moderating effect of caregiving Uplifts on the association between Objective Burden and Psychological QL of parents of children with CP

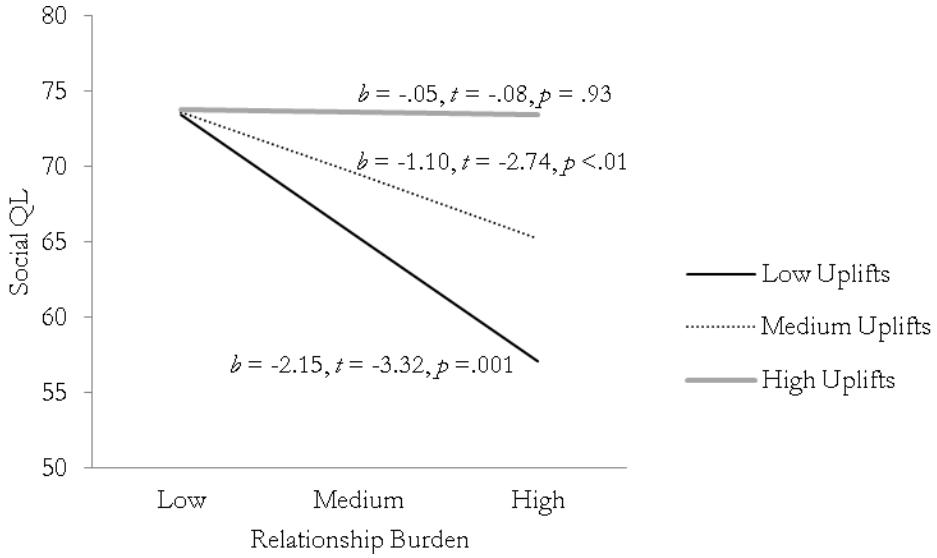

Figure 2. The moderating effect of caregiving Uplifts on the association between Relationship Burden and Social QL of parents who have children with CP

Discussion

The notion of the "disability paradox" (Albrecht & Devlieger, 1999) was revisited in this work within the context of pediatric family caregiving. This "disability paradox revisited" was then defined as the discrepancy between the burdensome caregiving experienced by parents who have children with chronic health conditions or disabilities, and the similar or superior QL levels reported by them, when compared with parents of healthy/able-bodied children. For the purpose of illustrating the "disability paradox revisited", a study on the QL and the positive and negative caregiving dimensions was conducted within a sample of parents who had children with CP. The obtained results confirm the variability of adaptation outcomes in these parents, and highlight the importance of targeting positive and negative dimensions of family caregiving in psychotherapeutic or psychosocial interventions aimed at improving their QL.

Main findings of our study may be summarized as follows: first, QL differences emerged between age groups (i.e. parents of children vs. parents of adolescents), and not between health/function conditions (i.e. children with CP vs. typically developing children); second, parents of children with CP reported more Subjective Burden and less caregiving Uplifts than parents of children without disabilities; third, the QL of parents of children with CP was, in general, moderately associated with Burden dimensions, and weakly related to caregiving Uplifts; fourth, caregiving Uplifts moderated the associations between Objective Burden and Psychological QL, and between Relationship Burden and Social QL of those parents; additionally, there were significant main effects of different Burden dimensions on parents' QL, with main effects also observed for caregiving Uplifts, but only in relation to Psychological and Social QL.

In contrast with the majority studies included in our literature review, and contradicting our initial hypothesis, parents of children with CP in our study reported a similar QL to those parents of children with no disabilities. This contrasting result highlights the importance of challenging professional and societal pessimistic perceptions, which tend to undervalue the adaptation potential of these parents and families (Yau & Li-Tsang, 1999). Furthermore, differences with medium magnitude in parental QL emerged between age groups for both parents caring for children with CP or typically developing

children, with parents of adolescents reporting lower Psychological QL than parents of children. In fact, adolescence is a developmental period marked by certain tensions for parents and their children, which may increase child-rearing stress and negatively interfere with parents' well-being (Seginer, Vermulst, & Gerris, 2002). The most striking and straightforward insight from these results is the possibility of more similarities than differences existing between parents of children with CP and those of children without a physical disability (Magill-Evans et al., 2001).

In the same line of thought, our second hypothesis was partially confirmed: parents of children with CP reported increased Subjective Burden and decreased caregiving Uplifts. Although levels of Relationship and Objective Burden did not differ between clinical and control groups, apparently disputing previous findings (Raina et al., 2005; Wang & Jong, 2004), considerable prudence is to be taken in generalizing such results. Our clinical group mainly included cases related to high-functioning forms of CP, and excluded those cases with comorbid intellectual disability, though severity of child's impairments and communication competence have been linked to increased parental stress (Yau & Li-Tsang, 1999). Nevertheless, it is worthy to note that parents of higher functioning children with CP may indeed report higher levels of psychological burden than one would expect (Manuel et al., 2003). Those differences observed in our study for Subjective Burden depict a medium effect and reiterate a need for caution in adopting simplistic "normalizing" attitudes in working with parents of children with CP, because in so doing, important intervention needs may be not properly screened and targeted. In our total sample and somehow consistent with the aforementioned results for Psychological QL, caregiving Uplifts were significantly lower in parents of adolescents than in parents of children, although such difference between age groups was smaller than the one between physical health conditions. Complementarily, while parents of children in control sample reported increased Objective burden than parents of adolescents, the opposite tendency was observed in our clinical sample, where parents of adolescents with CP reported higher Objective burden than parents of children with CP. This is to say that, despite most parents acknowledge childhood parenting as more enjoyable (even if more physically demanding), and adolescence parenting as more stressful (Seginer et al., 2002), such differences may assume distinctive features in the context of CP. During the adolescence period, the performance or achievement of certain developmental tasks related to family relationships, peers and autonomy may be more complicated for youths with CP, and his parents may gravely realize the stability of their child's impairments, along with the probable occurrence

of life-long challenges and the need for respective adjustments and caregiving (Lin, 2000; Magill-Evans et al., 2001). The fact of having a child with a disability may drive some parents to seek alternative meanings for their caregiving daily experiences, through positive reappraisals and benefit finding (Larson, 2010), but those positive appraisals tend to diminish during adolescence and the transition to adulthood (Lin, 2000). Additionally, cultural beliefs and prejudices about disability may be fostered within this context of seemingly increased vulnerability, and withdraw parents of children with CP from experiencing positive perceptions on their lives and parenting.

Despite the fact that moderate associations between caregiving variables and QL were conjectured in our third hypothesis, the obtained results were not that linear and depict a more complex and varied frame of correlations. Most of the associations between Burden types and QL domains were moderate, but Subjective Burden was strongly related to Psychological QL, and weak to moderate correlations were observed between Relationship Burden and QL domains. Complementarily, the strength of the association between caregiving Uplifts and parents' QL was weak, and no significant association was verified between Burden dimensions and caregiving Uplifts. This latest result is particularly noteworthy since it suggests that relatively opposite, contradictory aspects of family caregiving do not necessarily correlate as negative or as stronger as one would intuitively predict. In fact, it has been commented that distress and psychological dysfunction may occur with positive experiences of personal growth (Joseph & Linley, 2006). An immediate implication of such assertion is that, even if burden dimensions and caregiving Uplifts do not portray different facets of the same construct, they may indeed be assumed as plausible (and often simultaneous) reactions to the complex experience of caring for a child with a disability. Interestingly enough, the successful effort of these parents on integrating and finding a balance between positive and negative facets of their exceptional caregiving experience has been labeled as "the embrace of paradox" (Larson, 1998). The experience of such "paradox" is further discernible in our results: moderate correlations between Burden and QL constitute additional evidence for the risk of equaling caregiving stress to parents' adaptation (Beresford, 1994; Rentinck et al., 2006). In addition, although significant and slightly in line with previous statements (Larson, 2010), associations between caregiving Uplifts and QL domains were weak. Thus, despite the influence caregiving Uplifts may have on these parents' well-being, the experience of such positive perceptions should not be addressed as exclusive factors for the improvement of their QL. Finally, given the fact that the adopted measure for the assessment of Subjective Burden mainly included items

on the experience of emotional stress, tension and anxiety, the stronger association observed between this Burden dimension and Psychological QL seems straightforwardly explicable.

Since moderation effects of caregiving Uplifts were found not only for Psychological QL, and not for all Burden dimensions, our fourth and last hypothesis was not confirmed. However, thought-provoking results did emerge: caregiving Uplifts were found to moderate the relationship between Objective Burden and Psychological QL, and between Relationship Burden and Social QL. It has been suggested that it is not caregiving workload (i.e. Objective Burden) per se that causes psychological distress, but rather the interpretation that caregivers attach to the caregiving activities (Savundranayagam et al., 2011). This claim partially explains our first moderation, where parents with medium and high levels of caregiving Uplifts reported a better Psychological QL than those with low caregiving Uplifts; nonetheless, such effect was most visible under low to medium Burden conditions, and notably tended to vanish in the condition of high Objective Burden. This is to say that although positive caregiving perceptions may buffer the impact of Objective Burden on psychological well-being (Gupta & Singhal, 2004), they are not a sufficient mean to prevent Psychological QL deterioration when parents are facing increased Objective Burden; in those situations, parents would benefit more from interventions targeting effective task sharing and time management than, for instance, from cognitive reframing techniques. In those situations where Objective Burden is low to medium, adjunctive interventions seeking to improve the experience of caregiving Uplifts may be valued to promote the best Psychological QL possible. In the second interaction effect observed in our study, caregiving Uplifts were found to moderate the association between Relationship Burden and Social QL, thus adding some evidence for their effects on this particular domain, besides on the physical and psychological ones (Green, 2007). In this moderation effect, parents of children with CP who experienced high levels of caregiving Uplifts reported a better Social QL than parents experiencing low or medium levels of Uplifts, across all conditions of Burden intensity. Parents who experienced high levels of caregiving Uplifts seemed to benefit from a relative stability in their Social QL across different Burden conditions, in comparison to the other groups of parents, who experienced a stronger association between Burden increase and Social QL impairment. In practical terms, one may say that motivating, teaching and fostering the ability of parents of children with CP, to acknowledge and experience uplifts from their caregiving activity, may prevent them from the deleterious effects of Relationship Burden in their Social QL. In both

moderations observed in our study, caregiving Uplifts influenced the strength (and not the direction) of the association between Burden and QL; moreover, caregiving Uplifts positively influenced QL outcomes, regardless of the presence of adversity (i.e. across all Burden conditions). According to Rose et al. (2004), caregiving Uplifts were then to be regarded as "resource factors", and not as "protective factors", which would otherwise decrease the likelihood of a negative outcome, but only under adverse conditions (i.e. high Burden condition). A final remark on the observed interaction effects relates to the correlational matrix verified for the associations between Burden dimensions, caregiving Uplifts and QL domains. Although moderation analysis typically requires fewer assumptions on the associations between variables than mediation, it has been suggested that having a moderator variable that is uncorrelated with both the predictor and the criterion, increases the likelihood of obtaining clearly interpretable interaction terms (Baron & Kenny, 1986). The fact that in our study, Uplifts were uncorrelated with Burden dimensions, but still weakly associated with QL domains, might have influenced the consistency of results to some degree.

In addition to the aforementioned moderation effects, we also found evidence for some main effects of caregiving Burden and Uplifts on the QL of parents of children with CP. Relationship Burden had a small effect on Physical QL and Objective and Subjective Burden, medium ones, whilst no significant effect was detected for caregiving Uplifts on that same QL domain. These results confirm the significant impact of Burden (mostly Objective and Subjective types) on these parents' physical well-being (Raina et al., 2005), but do not support the hypothesis of caregiving Uplifts influencing their Physical QL (Green, 2007). As regards Psychological QL, Relationship and Subjective Burdens, along with caregiving Uplifts, displayed medium and large main effects, respectively. Concordantly, some authors have previously commented the significant impact of issues related to relationship (Raina et al., 2005) and Subjective Burden (Ha et al., 2008) on the well-being of parents of children with disabilities (or specifically with CP). As expected, caregiving Uplifts presented the highest main effects for the Psychological QL domain, since positive caregiving perceptions have been related to increased subjective well-being (Larson, 2010), and more specifically, to greater psychological flexibility and improved self-esteem (Gupta & Singhal, 2004). At last, small main effects were observed for the links between Objective and Subjective Burdens, together with caregiving Uplifts, and Social QL. This result adds support to previous qualitative findings, where parents of children with CP reported impairments in their social well-being due to caregiving responsibilities,

which included poorer social support and difficulty in maintaining social relationships (Davis et al., 2010). In agreement with our findings on mean differences between age groups, this variable (along with SES for Psychological domain) was a significant predictor of QL outcomes variability. This result was rather consistent across different QL domains, but despite its concordance with previous reports (Lin, 2000), it challenges the hypothesis of age-related attenuation of the consequences of having a child with a disability, due to parents 'adaptation to stress over time (Ha et al., 2008). Our results on the significant (although small) effect of SES on Psychological QL also lead us to conjecture that the financial burden that has been observed in parents of children with CP (Florian & Findler, 2001; Mobarak et al., 2000), may play an influential role on their psychological well-being.

The cross-sectional design of the present study represents its major limitation: even with careful selection of statistical procedures tailored to answer our research questions, causal relations between variables cannot be drawn from correlational research. As a matter of fact, we have no way of ascertaining if the observed differences between age groups, for instance, are developmental in nature (Magill-Evans et al., 2001). In addition, despite the fact that WHOQOL-BREF questionnaire discriminated parents' QL between age groups, we only had previous evidence of its discriminant validity between clinical and healthy populations (Vaz-Serra et al., 2006). This research work sought to offer and discuss innovative insights into adaptation variables and mechanisms that may underlie the adaptation of parents who have children with CP; nevertheless, we entirely subscribe the idea that "adaptation is not a single event but a multi-factorial determined process over time" (Rentinck et al., 2006, p. 168). Moreover, despite the fact that the comparison of adaptation patterns (e.g. main and interaction effects) between families of children with and without CP remains an understudied topic (Britner et al., 2003), we do acknowledge that such analyses were beyond the aims of this study, for they should be conducted in future research. Another major limitation of our study regards its sampling frames: despite the fact that our sample included cases from the three main regions of national territory, and that some of those cases were visiting the institution only once or twice a year, tertiary health care institutions have been commented to represent a biased context for sample collection (Brehaut et al., 2004). Furthermore, the obtained sample for our study mainly included mothers caring for children with milder forms of CP, thus lacking a wider range of functional ability levels, which could portray a more accurate depiction of the variety of CP forms. Since gender differences have been reported for the adaptation of parents of children with disabilities (Ha et al., 2008), these two sample characteristics (i.e. function and

gender) call for particular caution in generalizing the results here verified. Another potential limitation of our study relates to the risk of a social desirability bias in the participants' response style, since such bias is likely to occur, to some extent, in situations where people are asked about positive emotions or outcomes derived from stressful events they have experienced (Tomich & Helgeson, 2004). Finally, given the fact that our study was centered on the topic of pediatric family caregiving, our assessment protocol solely relied on a single informant (i.e. the primary family caregiver), as well as on the level of individual members, rather than on the family as a whole (Magill-Evans et al., 2001).

Despite bearing in mind the limitations just discussed, we acknowledge the innovative features and promising insights derived from this study. Very little attention has been given to the study of positive dimensions of family caregiving (Green, 2007), and to the best of our knowledge, our study was the first to quantitatively analyze the experience of caregiving Uplifts in the context of pediatric CP, and its interactions with parents' Burden and QL. With this research, we sought to move from an excessive focus on negative outcomes to the study of resiliency, within a clear conceptual framework, namely the stress-coping models. Besides, we conducted an assessment of parents' adjustment that was not restricted to pathological terms (Wallander et al., 1990) or to psychological functioning (Brehaut et al., 2004), and that further included understudied variables such as burden (Horton & Wallander, 2001), here approached from a multidimensional perspective that has been rarely adopted in past research (Savundranayagam et al., 2011). Other strengths of our study corresponded to the overcoming of two important gaps in previous research: one was the inclusion of an adequate control group, and the other was the comparison of different age points (Florian & Findler, 2001). Also in terms of statistical analyses, we examined interactions effects, because the exclusive analysis of main effects could be insufficient for understanding the different conditions under which a variety of determinants operate (Button et al., 2001).

A straightforward implication of our study reflects the need of changing professional attitudes regarding parents of children with disabilities in general, and with CP in particular. Parents may feel more motivated to acknowledge positive aspects of their caregiving, if they are embedded in a social context that facilitates personal and comprehensive meaning making of their parenting experience (Gupta & Singhal, 2004). In fact, health professionals working closer with these parents benefit from a privileged opportunity to offer a more realistic and positive regard on their experience, which may

then counteract some of the prejudices hold by society (Larson, 1998). Far more different than adopting a "normalizing" attitude, health professionals should acknowledge variability in the adaptation of parents of children with CP and assume themselves as positive sources of social support that may actually make a difference. Within such context, parents could openly develop their search for meaning, thus increasing their ability to experience positive caregiving perceptions (Gupta & Singhal, 2004). In other words, health professionals could help these parents "embracing the paradox" of their caregiving experience (Larson, 1998), by genuinely "embracing the paradox" of their clinical challenges themselves.

Another general clinical implication from the present study is the need to incorporate a multidimensional approach to parents' QL and pediatric family caregiving. Our study demonstrated that the relationships between caregiving variables are not necessarily linear, and their impact is quite differential. A multidimensional assessment of burden may increase intervention effectiveness, through an appropriate allocation of resources (Savundranayagam et al., 2011). Sharing caregiving responsibilities with other sources of support, learning to manage emotional stress, and implementing child behavior modification techniques, for instance, are distinct intervention strategies that may follow a multidimensional assessment to reduce Objective, Subjective and Relationship Burdens respectively. Nevertheless, any caregiving assessment exclusively focusing on negative dimensions may only provide an incomplete picture. It stands clear from our work that considerable levels of caregiving burden and uplifts may indeed coexist, so that despite a component of the intervention may be designed to decrease burden, other may be implemented to foster caregiving uplifts or utilize them as a therapeutic resource.

Given the clinical group that served the basis for our study, some additional clinical implications may be specifically drawn for parents of children with CP: first, interventions targeting distinct burden dimensions may differentially improve these parents' QL; second, caregiving Uplifts seem to be particularly relevant for the promotion of parents' Psychological and Social QL, and third, increased levels of caregiving Uplifts may alleviate the impact of Objective and Relationship Burden on parents' Psychological and Social QL, respectively. Moreover, our results highlight the need of adopting a developmental perspective in working with these parents: the adolescence period may represent a developmental context of increased risk for the reduction of psychological well-being and positive caregiving perceptions. For this reason, greater attention should be directed to these parents' emotional needs during the transition period from childhood to adolescence.

As it has been stated for interventions facilitating personal growth following adversity (Joseph & Linley, 2006), the development of caregiving Uplifts is to be encouraged, not imposed. In this sense, parents who engage in a mindful experience of their caregiving, may benefit from a broadened attention to different (and often conflicting) aspects of their parenting, and thus mitigate the effects of a narrowed focus on its burdensome aspects (Larson, 2010). For that same purpose, in the psychotherapeutic work with these parents, one should bear in mind that if we do not ask positive questions, we will hardly get a positive answer (Gupta & Singhal, 2004). Furthermore, if psychological interventions often seek to change rigid meanings attached to the individual's experience, we would also suggest that these parents are to be encouraged to value the ramified meaning of their parenting and, after all, of their "caregiving paradox". This clinical implication makes particular sense if one assumes coping as a process where searching and finding positive meanings may elicit positive emotions, which then sustain adaptive coping processes themselves (Folkman, 1997).

Future directions for the research of adaptation processes of parents who have children with CP were sharply synthesized by Britner and colleagues (2003), who argued for longitudinal, multi-measure and multi-respondent designs. Longitudinal designs are needed to determine causal links between variables and enlighten the dynamic interplay between negative and positive dimensions of adaptation across time. Age differences observed in literature and in our study underline the need of researching adaptation change and/or stability from childhood to adolescence, and from adolescence into adulthood. Moreover, there is a considerable research gap on the nature, extent and impact of personal growth and perceived benefits experienced by parents of children with CP. This research gap calls for the incorporation of qualitative methods in mixed designs that also include quantitative measures, in order to comprehensively capture the complexity of that phenomenon in this group. Finally, for the purpose of exploring mutual interplays between child and parents' adaptation, the applicability of transactional models to pediatric CP remains to be examined, preferably in multi-respondent research designs.

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Empirical Study IV |

Similarities Amid the Difference:
Caregiving Burden and Adaptation Outcomes in
Dyads of Parents and their Children with and without Cerebral Palsy

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Similarities Amid the Difference: Caregiving Burden and Adaptation Outcomes in Dyads of Parents and their Children with and without Cerebral Palsy

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Abstract

Objective. This study had two main objectives: first, to examine the direct and indirect effects, via social support, of caregiving burden on the adaptation outcomes of children/adolescents with cerebral palsy and their parents; and second, to assess the invariance of such models in clinical vs. healthy subsamples. **Methods.** Participants were 210 dyads of children/adolescents and one of their parents (Total *N*= 420), divided in 93 dyads of children/adolescents with cerebral palsy and 117 dyads of children/adolescents with no medical diagnosis. Data on caregiving burden, social support and adaptation outcomes were obtained through self-report questionnaires. **Results.** Caregiving burden was linked to parents and their children's psychological maladjustment and quality of life both directly (except for children's quality of life) and indirectly through social support. Findings were invariant across clinical and healthy subsamples. **Conclusion.** Caregiving burden may influence adaptation outcomes of children/adolescents with CP and their parents both directly and via their social support perceptions. These patterns are similar to those observed in typically developing children/adolescents.

Keywords: caregiving burden; cerebral palsy; social support; adaptation.

Introduction

Cerebral palsy (CP) is a chronic condition of movement and posture due to non-progressive disturbances that occurred in the developing fetal or infant brain (Rosenbaum et al., 2005). Given its clinical variability and elevated prevalence, CP may be regarded as an interesting prototype of developmental disabilities (Raina et al., 2004).

Research conducted so far has shown that children and adolescents with CP and their parents are at increased risk for impaired quality of life (QL) and psychological maladjustment (Brehaut et al., 2004; Brossard-Racine et al., 2012; Varni et al., 2005). However, there is a paucity of data on the psychosocial factors influencing those outcomes (Livingston, Rosenbaum, Russell, & Palisano, 2007; Rentinck, Ketelaar, Jongmans, & Gorter, 2006), as well as on the comparability of adaptation patterns exhibited by these families and those with typically developing children/adolescents (Magill-Evans, Darrah, Pain, Adkins, & Kratochvil, 2001). Furthermore, the recommended assessment of both child and parent adaptation levels (Barlow & Ellard, 2006) has been rarely adopted, even if such contextual factors have been underlined as important determinants for the QL of individuals with CP (Majnemer, Shevell, Rosenbaum, Law, & Poulin, 2007). The identification of potentially modifiable factors within a parent-child perspective is important to effectively promote more positive psychosocial outcomes and thus reduce the costs related to individual and family burden of disability and care. Complementarily, the examination of adaptation patterns in families with and without children with CP will improve the clinical understanding on commonalities and specificities underlying the psychosocial interventions to be developed. Therefore, the general aim of the present study was twofold: firstly, to examine the direct and indirect effects, via social support, of caregiving burden on the adaptation outcomes of children/adolescents with CP and their parents, and subsequently, to assess the (in)variance of the hypothesized parent-child adaptation mechanisms in clinical versus healthy subsamples.

Pediatric Family Caregiving as a Developmental Context

Family is the primary social context in which human development takes place (Bronfenbrenner, 1986). In pediatric psychology, the role of the family as a context for the understanding and treatment of chronic health conditions is well-established (Fiese & Sameroff, 1989). Specifically, the social-ecological model of adaptation and challenge in families of chronically ill children has argued for research and intervention practices based on the assessment of the child, parents and their social support networks (Kazak, 1989). The caregiving context, in particular, is crucial when examining childhood behavioral development (Carter, Briggs-Gowan, & Davis, 2004), since the most influential aspects of social context are those directly related to children's core developmental needs (Boyce et al., 1998). In fact, a considerable amount of research has demonstrated a significant relationship between the quality of caregiving and a child's ability to adapt to adversity (Armstrong, Birnie-Lefcovitch, & Ungar, 2005).

Although caregiving is a normative component of parenting children and adolescents, the nature and amount of care required by a child with chronic limitations and possible long-term dependence, such as several cases of CP, are distinct (Raina et al., 2005). For some parents, the continuous provision of such care may become burdensome and have deleterious effects on their physical and psychological well-being (Raina et al., 2004). Within a parent-child perspective, it has been claimed that parents' psychological distress significantly contributes to behavioral and emotional disturbance of chronically ill children (Canning, Harris, & Kelleher, 1996); moreover, parents' successful management of illness-related stressors has been linked to better social functioning and less distress in their children (Moos, 2002). The examination of models that describe how family context may influence the psychological adjustment of children with chronic health conditions, has been stated as a research priority for pediatric psychology in general (Drotar, 1997), and for CP in particular (McDermott et al., 1996). In addition, more recently, the assessment of models that account for positive dimensions of adaptation, such as QL outcomes, has been equally recommended (Barlow & Ellard, 2006).

Caregiving Burden, Social Support and Parent-Child Adaptation Outcomes

The influence of family environment, social support and parents' adjustment on the adaptation of children with chronic conditions has long been theoretically established in the disability-stress-coping model (Wallander, Varni, Babani, Banis, & Wilcox, 1989). These core premises were further developed in the transactional stress and coping model for chronic childhood illness, to encompass the mutual interplay between parental and child adaptation (Thompson, Gustafson, Hamlett, & Spock, 1992). In both theoretical formulations, caregiving context was defined by the inclusion of variables regarding illness stressors, social support and family functioning.

For the purpose of the present study, caregiving context was primarily operationalized through caregiving burden. This is a multidimensional construct integrating negative mood alterations, changes in dyadic caregiver-care recipient relationships, and time infringements resulting from caregiving (Montgomery et al., 2006). Caregiving burden has been found to be a foremost predictor of the psychological maladjustment experienced by caregivers of children with chronic medical conditions (Canning, Harris, & Kelleher, 1996), and of the well-being of caregivers of children with CP in particular (Raina et al., 2005). Three pediatric studies, which were conducted in the context of pediatric CP, observed significant associations between parental stress and their children's behavioral adjustment (Brossard-Racine et al., 2012) and QL (Majnemer et al., 2007; Wiley & Renk, 2007). Interestingly, these studies mostly relied on parents' report on their children's behavior and well-being, so it remains to be ascertained if these associations will be verified when examining more complex models accounting for both parents and child's reports on different adaptation variables.

Social support, defined here as the existence or availability of significant others to provide adequate help, care or company (Sarason, Levine, Basham, & Sarason, 1983), has been studied as a major determinant of adjustment in children with chronic physical conditions and their parents (Wallander & Varni, 1989, 1998). Within the social-ecological model, it has been commented that mothers' positive perceptions of social support are related to more positive attitudes towards themselves and their children, as well as to the provision of more adequate caregiving (Bronfenbrenner, 1986). Accordingly, for parents of children with disabilities, increased social support was found to be associated with better

individual well-being, more positive attitudes and more positive influences in parent-child interactions (Dunst, Trivette, & Cross, 1986). For parents of children with CP, in particular, social support has been found to be positively related to parents' mental health (Rentinck et al., 2006).

An alternative and specific way of examining social support as a mediator between parenting stressors and outcomes in pediatric populations has been described by Quittner, Glueckauf, and Jackson (1990). The rationale for the mediator hypothesis was that the chronicity of parenting stress in pediatric health conditions could elicit more negative perceptions of support which, in turn, could increase psychological symptoms. In their study of mothers of children with a disability, the authors found evidence for this mediator effect of social support on the links between child/maternal stressors and mothers' psychological distress (Quittner et al., 1990). Therefore, their study gathered additional evidence for a "social support deterioration model" (Lin & Ensel, 1984), which posits that stigmatizing or chronic stressful events may exhaust social resources or elicit avoidant or inadequate responses from network members.

When studying adaptation patterns across different populations (e.g. pediatric vs. healthy), it is important to bear in mind that specific family factors may be of differential importance in various conditions (Daniels, Moos, Billings, & Miller III, 1987). In fact, the invariance of adaptation patterns between families of children with and without CP remains an understudied topic (Britner, Morog, Pianta, & Marvin, 2003). In their study on the mediating role of social support between caregiver stressors and psychological distress, Quitner and colleagues (1990) verified that, although between-groups differences existed at the level of means comparison, the mediation model was valid for both clinical and control samples. A similar stability of associations between variables was reported in other pediatric studies: in one study, mother's higher adjustment and social support were related to better child adjustment in families of children with or without handicaps (Barakat & Linney, 1992); in another study, behavior problems and parenting stress significantly covaried across time in both families of typically developing and developmentally delayed children (Neece, Green, & Baker, 2012). As regards children and adolescents in particular, Moos (2002) suggested that associations between life stressors, social resources and adaptation might be similar among ill, distressed and healthy youths. Nevertheless, despite the evidence for a general association between risk and resistance factors and childhood adaptation, Daniels and colleagues (1987) noted that certain variables, such as burden of

illness in the family, were stronger predictors of adaptation for pediatric patients than for healthy individuals.

The Current Study

The present study was conducted to examine how caregiving burden is associated with parents and children's adaptation outcomes in normative and clinical parent-child samples. Three specific objectives were defined: first, to assess the associations between caregiving burden and parents and children's psychological maladjustment and QL; second, to examine the mediating effect of parents and children's social support on the links between caregiving burden and psychological maladjustment and QL; third, to ascertain if the mediation model was moderated by condition (CP vs. typically developing children), gender (boys vs. girls), and age group (children vs. adolescents).

Accordingly, four hypotheses were devised for our study:

Hypothesis 1: Caregiving burden would be positively related to parents and children's psychological maladjustment and negatively related to their QL;

Hypothesis 2: Caregiving burden would be negatively associated with parents and children's social support;

Hypothesis 3: Social support would mediate the links between caregiving burden and the adaptation outcomes of both parents and their children;

Hypothesis 4: Direct and indirect effects between caregiving burden and parent and child adaptation outcomes would be equivalent in clinical and community subsamples. Finally, we also examined such model invariance for age and gender groups, but no specific predictions were made in that regard.

Method

Participants

Participants were 210 dyads of children/adolescents and one of their parents (Total *N*= 420), divided in 93 dyads with children/adolescents with CP and 117 dyads with healthy, able-bodied children/adolescents.

The clinical sample for the present study was recruited in ten Portuguese Cerebral Palsy Associations (social and tertiary healthcare institutions) between July 2010 and July 2011. Pediatric subjects were assigned to the study if they met the following criteria: (1) diagnosis of CP established by a physician; (2) age between 8 and 18 years old; (3) minimum intelligence quotient (IQ) of 70. For their parents, a single inclusion criterion was considered: being a primary family caregiver of the child/adolescent with CP. Cases where results from formal IQ assessments were not available (n = 13), were still included if they were assessed as having no significant developmental delay (as indicated by gross evaluation of their cognitive abilities and the absence of previous adaptations to regular school curricula). The consideration of an inclusion criterion based on children/adolescents' intellectual functioning was implied by the methodological design of the study, which relied on children/adolescents' self-reports (in fact, there were no proxy-reports in this study).

The control sample (i.e. typically developing children) was collected in two Portuguese public schools, between January and June 2010. Children/adolescents were included in this sample if they fulfilled two criteria: aged between 8 and 18 years old, and reporting no diagnosed chronic health condition. For their parents, a single inclusion criterion was considered: to be the parent who spent more daily time with the child/adolescent.

Children /adolescents (53.8% boys) were between 8 and 18 years old (M = 12.34; SD = 2.91). Parents, mostly mothers (83.8%) and married (81%), were between 23 and 58 years old (M = 42.34; SD = 5.72). Descriptive results for both samples, group differences in socio-demographic characteristics, and clinical characteristics for CP sample are depicted in Table 1. Participants in CP and healthy samples only differed significantly in their socioeconomic status (SES): there was a higher percentage of dyads from high and medium

SES in the healthy sample, and a higher percentage of dyads from low SES in the CP sample. Regarding the clinical sample, it is worth mentioning that more than half of the cases were related to milder forms of CP, including spastic subtypes (88.1%) with no limitations in walking (62.4%).

Table 1. Socio-demographic and clinical characteristics of the sample.

	CP Sample		**Healthy Controls Sample**		
	Children/Adolescents (*N* = 93)	**Parents** (*N* = 93)	**Children/Adolescents** (*N* = 117)	**Parents** (*N* = 117)	*Differences between Samples*[d]
Age (*M*/*SD*)	12.39 (2.83)	41.79 (6.32)	12.31 (2.97)	42.7 (5.18)	*C/A:* $t = -.20$; $p > .05$ *P*: $t = 1.21$; $p > .05$
Age Group (*n*/%) Children (8-12) Adolescents (13-18)	46 (49.5) 47 (50.5)		61 (52.1) 56 (48.9)		$\chi^2_{(1)} = .15$; $p > .05$
Gender (n/%)					*C/A:*
Male	54 (58.1)	12 (12.5)	59 (50.4)	22 (18.8)	$\chi^2_{(1)} = 1.22$; $p > .05$
Female	39 (41.9)	84 (87.5)	58 (49.6)	95 (81.2)	*P*: $\chi^2_{(1)} = 1.82$; $p > .05$
Marital status: married **(*n*/%)**	-	71 (76.3)	-	99 (84.6)	$\chi^2_{(1)} = 1.88$; $p > .05$
SES[a] **(*n*/%)**					
Low	56 (60.2)		31 (26.5)		$\chi^2_{(2)} = 27.12$;
Medium	23 (24.7)		64 (54.7)		$p < .001$
High	11 (11.8)		22 (18.8)		
Missing	3 (3.2)		-		
CP Type[b] **(*n*/%)**	47 (50.5)				
Spastic unilateral	35 (37.6)				
Spastic bilateral	4 (4.4)				
Dyskinetic	2 (2.2)				
Ataxic	5 (5.3)				
Missing					
GMFCS[c] **(*n*/%)**					
I	58 (62.4)				
II	13 (14.0)				
III	10 (10.8)				
IV	7 (7.5)				
V	3 (3.2)				
Missing	2 (2.2)				

Notes.

[a] Socioeconomic status (SES) was determined using a classification system based on parents' job and educational level (Simões, 1994).

[b] Classification of CP subtypes according to the Surveillance of CP in Europe project (SCPE, 2000).

[c] Levels of function according to the Gross Motor Function Classification System (GMFCS) – Expanded and Revised (Palisano, Rosenbaum, Bartlett, & Livingston, 2007).

[d] Results of comparison tests for socio-demographic variables.

Measures

Caregiving burden.

The Revised Burden Measure. This self-report questionnaire included three subscales for different types of burden, namely: objective burden (e.g. "Have your caregiving responsibilities changed your routine?"), subjective burden (e.g. "Have your caregiving responsibilities created a feeling of hopelessness?") and relationship burden (e.g. "Have your caregiving responsibilities caused conflicts with your relative?") (Montgomery et al., 2006). Participants answered these questions on a 5-point scale (1 = *Not at all*; 5 = *A great deal*). Those subscales were then combined into an overall mean score of caregiving burden.

Social Support.

Satisfaction with Social Support Scale. This instrument assesses adults' subjective appraisals on their satisfaction with social support obtained from significant others and activities (Pais-Ribeiro, 1999). The questionnaire comprises 15 items, which target four dimensions of satisfaction with SS: satisfaction with friends (e.g. "I am satisfied with the kind of friends I have"), intimacy (e.g. "When I need to let off steam, I can easily find someone to support me"), satisfaction with family (e.g. "I am satisfied about the way I get along with my family") and social activities ("I lack social activities that satisfy me"). A mean score of social support was computed, based on the responses provided within a 5-point scale (1 = *Totally disagree*; 5 = *Totally agree*).

***Satisfaction with Social Support Scale for Children and Adolesc*ents.** This scale assesses satisfaction with social support based on children and adolescents' perceptions on their social experiences with parents, friends and social organizations (Gaspar et al., 2009). The instrument comprises two subscales: satisfaction with social support (e.g. "I am satisfied with the activities and things I do with my group of friends"), and activities connected to social support (e.g. "I would like to participate more in organised activities, such as sport clubs, scouts"). An overall mean score was calculated from the answers provided for each item within a five point Likert scale ranging from 1 (*Totally disagree*) to 5 (*Totally agree*).

Adaptation Outcomes.

Psychological maladjustment.

Mental Health Inventory – short form (MHI-5). The MHI-5 is a screening instrument aimed at the assessment of two general dimensions of adult mental health: psychological distress and psychological well-being (Ware, Snow, Kosinski, & Gandek, 1993, as cited in Pais-Ribeiro, 2001). The 3-item psychological distress subscale was used in this study to assess the frequency of anxiety and depressive symptoms (e.g. "How much of the time, during the past month, have you felt downhearted and blue?"; "How much of the time, during the past month, have you been a very nervous person?"), within a 6 point response scale ranging from 1 (*Never*) to 6 (*Always*). Responses were then computed into global mean scores.

Strengths and Difficulties Questionnaire (SDQ). The SDQ is a measure of psychological adjustment for children and adolescents (Goodman, 2001). The self-report version of SDQ was used in this study to assess psychological difficulties related to four main factors: emotional symptoms (e.g. "I worry a lot"), peer problems (e.g. "I get on better with adults than with people my own age"), conduct problems (e.g. "I get very angry and often lose my temper") and hyperactivity-inattention (e.g. "I am constantly fidgeting or squirming"). For each one of the SDQ items, the respondent states his/her perception within a 3-point Likert scale: 0 (*Not true*); 1 (*Somewhat true*) and 2 (*Certainly true*). The computation of an overall mean score was performed in order to assess children/adolescents' psychological maladjustment.

Quality of life.

The World Health Organization Quality of Life Assessment (WHOQOL) – 8-item index (EUROHIS-QOL). EUROHIS-QOL is a screening measure derived from the WHOQOL-100 and the WHOQOL-BREF instruments (Pereira, Melo, Gameiro, & Canavarro, 2011; Schmidt, Mühlan, & Power, 2005). This measure includes two items to assess each of four QL domains: physical (e.g. "Do you have enough energy for everyday life?"), psychological (e.g. "How satisfied are you with yourself?"), social (e.g. "How satisfied are you with your personal relationships?") and environmental (e.g. "How satisfied are you with the conditions of your living place?"). Participants answered items on a 5-

point response format ranging from 1 (*Very poor/Very dissatisfied/Not at all/Never*) to 5 (*Very good/Very satisfied/Extremely/Completely*). The overall score was then obtained from the mean of those item scores.

KIDSCREEN-10. The shortest version of Kidscreen questionnaires is a unidimensional measure of 10 items on physical (e.g. "Have you felt full of energy?"), psychological (e.g. "Have you felt sad?") and social (e.g. "Have you had fun with your friends?") aspects of children and adolescents' QL (Gaspar & Matos, 2008; Ravens-Sieberer et al., 2010). The self-report form was used in the present study. Items of KIDSCREEN-10 were completed in a 5-point Likert scale, ranging from 1 (*Not at all/Never*) to 5 (*Extremely/Always*). An overall QL score was then derived from the mean of those item scores.

Procedures

The authorizations for sample collection were obtained from the Direction Boards of the participating institutions, namely: the Portuguese Federation of Cerebral Palsy Associations, ten Portuguese Cerebral Palsy Associations and two public schools enrolled in the research project. The Direction Boards of these institutions were responsible for the evaluation and approval of research projects, in a similar way to regular institutional review boards.

According to the inclusion criteria defined for the clinical sample, 161 parent-child dyads were assigned to participate in the study. Afterwards, 68 of those cases were excluded for a variety of reasons: seven cases refused to participate; forty-seven cases did not visit the institutions during the established period for sample collection; two cases related to children living in foster care institutions; and 12 cases did not report in all the intended measures. Therefore, a final clinical sample of 93 dyads of children/adolescents with CP and their parents was attained. These parent-child dyads were administered the assessment protocol during their regular visits to the institution, in a room provided for the purpose and under the supervision of a professional acquainted with the research project.

In order to achieve the projected size for a sample of controls, 124 parent-child dyads that complied with the aforementioned criteria were enrolled to participate in the research project. Subsequently, seven cases were excluded: two parents refused to

participate in the study; four adolescents did not return their parents' questionnaires; and one case did not complete all the required measures. As a result, the final sample of healthy controls was composed by 117 parent-child dyads. Children and adolescents completed their questionnaires in the classroom, under the supervision of a researcher, and were asked to deliver and return their parents' questionnaires, which were to be completed at home.

All subjects participated voluntarily in the study. In strict adherence to legal and ethical requirements, informed consent forms were obtained from all parents and from children older than 13 years; informal assents were obtained from younger children. Children who refused to participate were not included in the study, even if their parents had previously authorized their participation.

Results

Descriptive and Zero-order Correlations

Descriptive statistics and correlations for all of the measures for both samples are presented in Table 2. Hypothesis 1 and 2 were partially supported. Caregiving burden was positively related to parents' psychological maladjustment and negatively related to their QL and social support. For children, parents' caregiving burden was associated with psychological maladjustment in the expected positive direction; however, there were no significant associations with children's QL and social support, except a marginally significant correlation (p = .06) between caregiving burden and social support for the CP sample.

Path Models: Testing Direct and Indirect Links between Caregiver Burden and Adaptation Outcomes

Two SEM path models were run with the whole sample testing the direct and indirect links between caregiving burden and adaptation outcomes via social support.

For Model 1, the specified outcomes were parents and children's psychological maladjustment, whereas for Model 2, the outcomes were parents and children's QL. Analysis of raw data with the maximum likelihood estimation method was used. After obtaining the results for the predicted models, we trimmed these models by removing non-significant paths, endorsing a model-generation application of SEM (Jöreskog, 1993, described in Kline, 2005).

In Model 1 (Figure 1 and Table 3), results demonstrated direct and indirect links, through social support, between caregiving burden and parents and children's psychological maladjustment. Results for Model 2 (Figure 2 and Table 4) showed that caregiving burden was associated with parents' QL directly and indirectly, also via social support. In this model, caregiving burden was associated with children's QL only indirectly via children's social support. Results from both models supported this study's third hypothesis.

Subsequently, we ran several multi-group analyses for both models according to condition, gender and age group, in order to test for model invariance (Hypothesis 4). Firstly, with regard to condition, we found that the differences between the unconstrained models and the structural weights models were non-significant for Model 1 ($\Delta\chi^2(6) = 7.97$, $p > .05$) and for Model 2 ($\Delta\chi^2(5) = 3.76$, $p > .05$), confirming, as expected, that both models were valid for the healthy as well as for the CP samples. Secondly, regarding gender, the difference between the unconstrained and the structural weights model was also non-significant for Model 1 ($\Delta\chi^2(6) = 5.50$, $p > .05$) and for Model 2 ($\Delta\chi^2(5) = 2.64$, $p > .05$), as predicted. Lastly, with regard to age group, the difference between the unconstrained and the structural weights model was non-significant for Model 1 ($\Delta\chi^2(6) = 11.91$, $p > .05$). A non-expected significant difference ($\Delta\chi^2(5) = 11.37$, $p = .05$) was found between the unconstrained and the structural weights model for Model 2. We then performed separate equality constraints for each of paths in the model and verified that the

significant difference was located in the path linking caregiving burden to parents' QL: this specific standardized coefficient was only significant for the parents-adolescents' dyads (β = -.25, p < .001) and not for the parents-children's (β = .01, p> .05). Finally, given the fact that the CP sample had more subjects from lower socioeconomic backgrounds than the non-CP sample, we re-ran both models separately for each of the samples, controlling for SES, and results did not change significantly for the psychological maladjustment or the QL models.

Table 2. Descriptive statistics and matrix of inter-correlations among study variables for parents and children/adolescents in CP (figures in bold font) and healthy samples (figures in non-bold font).

	Parents				**Children/Adolescents**			
	1	2	3	4	5	6	7	8
Parents								
1. Caregiving burden								
2. Social support	**-.35****/-.45**							
3. QL	**-.39****/-.25**	**.47****/.48**						
4. Psychological maladjustment	**.51****/.31**	**-.45****/-.45**	**-.66****/-.61**					
Children/Adolescents								
5. Social support	**-.20~**/-.13	**.26*/**.14	**.16**/.09	**-.45****/-10				
6. QL	**-.15**/-.11	**.17**/.24**	**.21*/**.06	**-.23***/-.17~	**.54****/.51**			
7. Psychological maladjustment	**.24***/.30**	**-.22*/ -.25****	**-.19~/**-.18~	**.25*/**.17~	**-.42****/-.53**	**-.47**/**-.46**		
8. Age	**.04**/-.12	**-.17**/-.03	**-.22*/**-.03	**.20~/**.22*	**.09**/-.04	**-.12/**-.35**	**.07**/-.09	
9. Gender	**-.31****/-.12	**.19**~/.08	**.21*/**.03	**-.13**/-.07	**.00**/.15	**.01/**.03	**-.18~**/-.18~	**-.16**/.05
Mean	**2.18**/.1.97	**3.61**/3.67	**3.63**/3.78	**2.79**/2.33	**3.56**/3.89	**4.02**/4.09	**.58**/.51	
SD	**.72**/.72	**.67**/.67	**.52**/51	**1.12**/.94	**.67**/.65	**.57**/.55	**.29**/.25	
Cronbach's alpha	**.90**/.94	**.86**/.88	**.79**/.82	**.86**/.89	**.76**/.81	**.75**/.75	**.77**/.75	

Note. ** $p < .01$; * $p < .05$; ~ $p \leq .08$

Table 3. Unstandardized coefficients and standard errors (SE) for all parameters and bias-corrected (BC) bootstrap confidence intervals (CI) for indirect effects in Model 1.

Parameters	Unstandardized coefficients (SE)	*p*	BC Bootstrap 90% CI for Indirect effects
Direct effects			
Within-participants			
Parents			
Caregiving burden→ Social Support (P)	-.38 (.06)	<.001	
Caregiving burden→ Psych. Maladjustment (P)	.42 (.09)	<.001	
Social Support (P) → Psych. Maladjustment (P)	-.51 (.10)	<.001	
Children			
Social Support (C)→ Psych. Maladjustment (C)	-.18 (.02)	<.001	
Across-participants			
Caregiving burden→ Social Support (C)	-.18 (.06)	.01	
Caregiving burden→ Psych. Maladjustment (C)	.08 (.02)	<.001	
Indirect effects			
Within-participants			
Caregiving burden→ Psych. Maladjustment (P)	.19 (.06)	<.001	[.11, .30]
Across-participants			
Caregiving burden→ Psych. Maladjustment (C)	.03 (.01)	.01	[.02, .05]

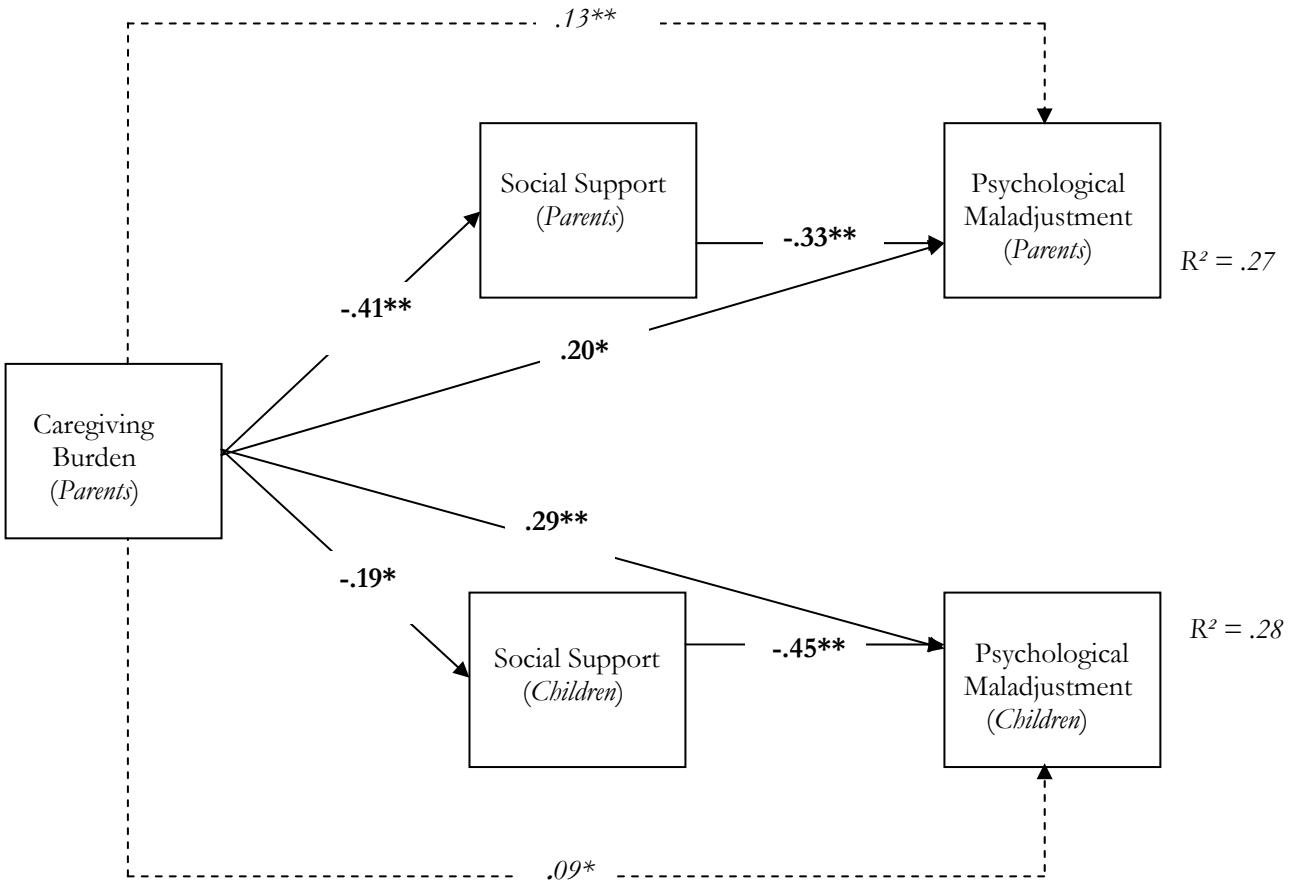

Figure 1. Model 1: Structural equation model testing the direct and indirect effects of caregiver burden on parents and children's psychological maladjustment via social support.

Note. Non-italic bold figures represent standardized coefficients for direct paths; italic figures represent standardized coefficients for indirect paths. Fit indices for the model were: $\chi^2(2, N = 210) = 3.74$; $p > .05$; CFI = .99; RMSEA = .07. For simplicity, error terms are not shown; ** $p \leq .001$; *$p \leq .01$.

Table 4. Unstandardized coefficients and standard errors (SE) for all parameters and bias-corrected (BC) bootstrap confidence intervals (CI) for indirect effects in Model 2.

Parameters	Unstandardized coefficients (SE)	*p*	BC Bootstrap 90% CI for Indirect effects
Direct effects			
Within-participants			
Parents			
Caregiving burden→ Social Support (P)	-.38 (.06)	<.001	
Caregiving burden→ Quality of life (P)	-12 (.05)	≤.01	
Social Support (P) → Quality of life (P)	.32 (.05)	<.001	
Children			
Social Support (C)→ Quality of life (C)	.43 (.05)	<.001	
Across-participants			
Caregiving burden→ Social Support (C)	-.18 (.06)	≤.01	
Indirect effects			
Within-participants			
Caregiving burden→ Quality of life (P)	-.12 (.06)	.03	[-.24, -.09]
Across-participants			
Caregiving burden→ Quality of life (C)	-.08 (.01)	.03	[-.16, -.04]

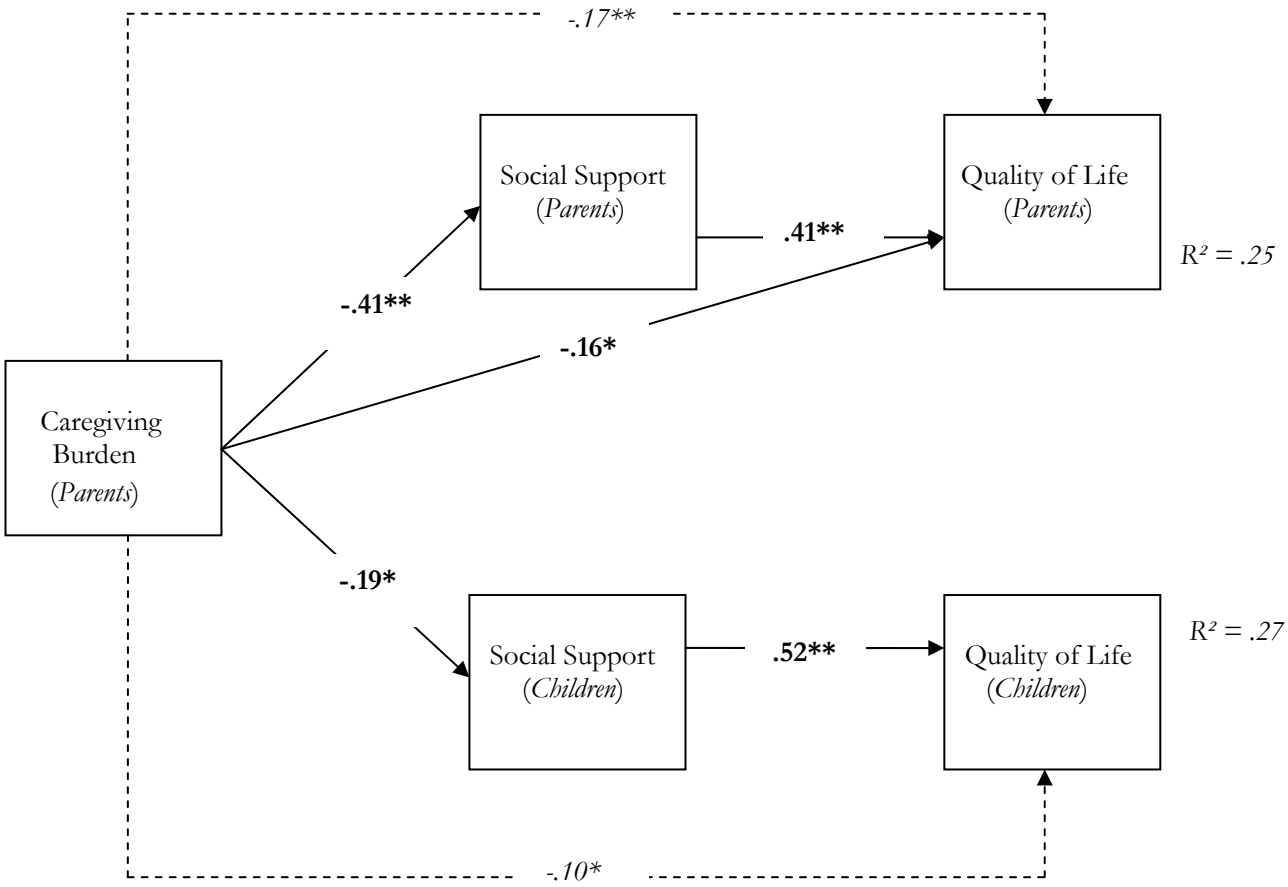

Figure 2. Model 2: Structural equation model testing the direct and indirect effects of caregiving burden and on parents and children's QL, via social support.

Note. Bold non-italic figures represent standardized coefficients for direct paths; figures in italic represent standardized coefficients for indirect paths. Fit indices for the model were: $\chi^2(3, N = 210) = 3.97$; $p > .05$; CFI = .99; RMSEA = .04. For simplicity, error terms are not shown; ** $p \leq .001$; *$p \leq .01$.

Discussion

Our main findings verified that parents' caregiving burden was associated with parents and children's adaptation outcomes through their perceptions of social support. Furthermore, a similar pattern of associations was observed for parent-child dyads of both children/adolescents with CP and healthy, able-bodied children/adolescents. Since Drotar (1997) established a research agenda for the study of parent-child relationships in pediatric contexts, few studies addressed those questions for children/adolescents with CP and their parents. In addition, it was only recently that some authors claimed to have conducted the first assessment of health-related QL from the perspective of children with CP (Varni et al., 2005). To the best of our knowledge, in the area of pediatric CP, our study was the first to examine potential mechanisms through which caregiving burden may affect parent/child adaptation outcomes, while considering children's self-reports and a healthy sample to explore the invariance of those adaptation mechanisms.

Partially confirming this study's first two hypotheses, caregiving burden was significantly related to parents' psychological maladjustment, QL and social support and to children's psychological maladjustment only. These results are aligned with previous research reports (Brossard-Racine et al., 2012; Canning, Harris & Kelleher, 1996; Raina et al., 2005). The absence of a significant relationship between burden and children's QL somehow contradicts previous findings (Majnemer et al., 2007; Wiley & Renk, 2007), a result that might be due to the reduced size of the subsamples in our study. Our findings suggest that caregiving burden is an important determinant of adaptation outcomes for parents and their children with CP, though it may affect children/adolescents in a less pervasive way. This implies that burden assessment in future research should be conducted in relation to family member's specific outcomes, and that interventions targeting caregiver's burden may positively influence parents and their children's psychological adjustment and parents' QL.

Our third hypothesis aimed at testing the indirect effects of caregiving burden on parents and children's outcomes, through their perceptions of social support. This hypothesis was fully corroborated by our findings: social support perceived by

children/adolescents and their parents mediated the links between caregiving burden and their psychological maladjustment and QL. These results add evidence to the mediating effect of social support on the links between chronic caregiving stressors and parental adjustment (Quittner et al., 1990). Furthermore, the present study's results extend the relevance of such mediation model in that it may be applied, in addition to parents, to children's adaptation outcomes. The main implication of this finding is that interventions targeting caregiving burden in CP may possibly exert its influence on parent-child improved outcomes, via enhanced parent and child's perceptions of social support. Thus, in order to capture the effects of such interventions in the more global social context of children/adolescents with CP and their parents, the assessment of social support perceptions should be taken into account. In fact, caregiving burden was only indirectly linked to children's QL through their associations with social support. This particular finding suggests that, for children and adolescents, parents' caregiving burden may only influence specific outcomes when they are related to children/adolescents' perceptions of social support. More precisely, increased parental caregiving burden may elicit negative perceptions of support from parents, friends and social organizations in children/adolescents with CP, and thus impair their psychological adjustment and their QL.

Finally, and quite remarkably, our last research hypothesis was confirmed, in that no differences emerged in the mediation model for the clinical and the healthy samples. This result substantiates the existence of a general association between risk and resistance factors and childhood adaptation (Daniels et al., 1987), and further extends the assertion that more similarities than differences may exist between families of children/adolescents with CP and families with typically developing children/adolescents (Magill-Evans et al., 2001). Although such evidence is important to deconstruct negative expectations hold by society and health professionals towards families of people with disabilities (Green, 2007), in clinical practice, it should be borne in mind that important differences between adaptation variables may exist (Quittner et al., 1990) and that certain associations between them may matter distinctively for different groups (Daniels et al., 1987). Moreover, differences in these patterns of relationships seem most likely to emerge during critical developmental transitions (Quittner et al., 1990), which were not considered in our study. Nevertheless, our findings indicate that the increase in parents' caregiving burden may operate through similar mechanisms and eventually lead to similar outcomes in both dyads of parents and their children with or without CP. The outcomes desired by parents of

children with disabilities include the achievement of social inclusion and an "ordinary life", the experience of a life that is not confined to their role as parents/caregivers, and the enjoyment of quality time with their children, which is to be over and above caregiving activities (Arksey, Beresford, Glendinning, Greco, & Sloper, 2007). These desired outcomes are certainly shared by many other parents, and in this sense, our findings further posit that the social deterioration model (Lin & Ensel, 1984) may be useful in understanding caregiving stress processes in dyads of parents and children with or without CP.

In this study, the analysis of the invariance of effects between groups was also performed based on gender and age subsamples. Since no gender differences emerged, this was indicative of the models' adequacy for both boys and girls. Regarding age groups (children vs. adolescents), the direct effect of burden on parents' QL was only significant for parent-adolescent dyads. It would be tentative to conjecture that such direct effect could only emerge in later stages of child's development, when an extension of burden over time would have a direct impact on the most global adaptation outcomes; alternatively, the demands of family reorganization during the transition to and the period of adolescence might explain why burden affects parents of teenagers in a significant direct way. However, to fully examine such hypotheses, longitudinal study designs would be required.

Limitations, strengths, and future directions

As recommended in a recent agenda for pediatric psychosocial research (Barlow & Ellard, 2006), this study had the merits of "hearing the voices of children" and including a parent-child perspective in the research approach to a pediatric group that has been notably understudied. Nevertheless, its cross-sectional design stands as its major limitation: even with SEM techniques, which have been underutilized in pediatric psychology research (Nelson, Aylward, & Steele, 2008), a significant path coefficient remains a necessary but not a sufficient criterion to establish causality (King, King, Rosenbaum, & Goffin, 1999). Despite this major limitation, we endorse the importance of such cross-sectional studies in identifying promising relationships, which may be then further examined in longitudinal designs (Quittner et al., 1990). Our clinical sample was collected in tertiary healthcare centers, which may be related to some selection bias (Brehaut et al., 2004; McDermott et al., 1996); furthermore, this sample mainly included mothers (as primary caregivers) and milder forms of CP. Given the adoption of self-reports in our research design, children/adolescents with an intellectual disability (i.e., IQ < 70) were excluded from the

study, in order to safeguard the reliability of those reports. Although severity of impairment (especially when communication is also impaired) has been positively related to parental stress in the context of developmental disabilities in general (Yau & Li-Tsang, 1999), one study on pediatric CP revealed that a lower level of child's impairment was not associated to a better maternal adaptation (Manuel, Naughton, Balkrishnan, Smith, & Koman, 2003). Therefore, additional caution must be taken in generalizing the results here discussed. Finally, this study was conducted in a Portuguese context. The scales used in this study were all Portuguese validated versions of English original measures, except for the scales of social support, which were first developed in Portugal. Although we expect that similar results would be obtained in other Western countries, future research in other cultural contexts is warranted. The CP sample in our study mainly came from a low-medium socioeconomic background. Results indicated that, controlling for this variable, the overall mediation results remained similar; however, further research should address this issue with more diverse samples in terms of socioeconomic backgrounds.

Future research should longitudinally examine the patterns of relationships that have gained some support from previous cross-sectional studies; it would be important to assess differences in the adaption patterns exhibited by families with children with CP versus families with typically developing children, during periods of critical developmental transitions, such as the child entering school or the transition to adolescence. In addition, although the role of a primary family caregiver is crucial, there are other relevant influences inside and outside the family (Armstrong et al., 2005), and thus the role of fathers, siblings and peers on children/adolescents' outcomes should be studied in greater depth.

Conclusion

These findings call for special consideration of a parent-child perspective when developing psychosocial interventions in the context of pediatric CP. The observed results add evidence for a potential mechanism via which caregiving burden may influence parent-child adaptation outcomes, namely through the deterioration of social support perceptions. Moreover, our findings reveal important similarities that have been notably understated in literature, as the invariance of the hypothesized adaptation mechanisms between dyads of parents and children with and without CP.

In general, these results emphasize the relevance of assessing and targeting core dimensions of an individual's context, as an effective clinical guideline for understanding

and improving individual's adaptation outcomes. The observed results further highlight the importance of applying a more comprehensive approach to pediatric family caregiving context, thus encompassing child and parents' social support perceptions in assessment and intervention routines. Complementarily, the reduction of family caregiving burden in pediatric CP could be regarded as a strategic intervention target, since it may elicit beneficial effects on both parental and child levels. Finally, and perhaps most importantly, this study's findings add support for the clinical guideline that psychosocial interventions with these families should acknowledge general adaptation processes in the specific context of CP. Although parents and their children with CP may face specific challenges and difficulties, which have been fairly documented in literature, the clinical approach to this population is likely to benefit from the consideration of normative developmental issues and adaptation mechanisms as well. Therefore, psychosocial interventions with these families should be more a matter of finding "similarities amid the difference", rather than assuming the fact of having a child with CP as an all-determining difference.

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4 DISCUSSION

Summary and integration of main findings

Methodological considerations: Strengths and limitations

Scientific implications and future directions in research

Clinical implications

The research work presented throughout this dissertation was broadly aimed at improving current understanding on how children and adolescents with CP and their parents (primary family caregivers) adapt to the challenges associated with that chronic physical condition. In this last chapter of the dissertation, that general research aim is revisited by synthesizing main findings from the research project developed and discussing their meaning and their scientific and conceptual integration. Accordingly, the theoretical framework that underlay the process of research development will be used at this stance to guide the description and critical comment of those main findings.

The macro conceptual framework adopted in this research project incorporated a **developmental and ecological perspective** on human development and adaptation (Achenbach, 1990; Cicchetti, 2006), in conjunction with a stress-coping formulation of individual and family adaptation to pediatric conditions - the **disability-stress-coping model** (Wallander et al., 1989a). Even so, despite its intended comprehensiveness, such conceptual framework was further enriched, or at least detailed, with the occasional consideration of more specific theoretical models at particular points of the research development. These specific contributions were seen as refinements or elaborations of the most general assumptions established by the aforementioned perspectives, and included, for instance, the formulations of stress-coping processes accounting for the role of positive emotions (Folkman, 1997; Folkman & Moskowitz, 2000) or the social support deterioration model (Lin & Ensel, 1984; Quittner et al., 1990).

This final discussion begins with a **summary of main findings and their integrated discussion**, which is followed by a reflection on core methodological issues, namely the **research strengths and limitations**. The discussion then draws attention to a number of **scientific implications** raised by the empirical studies conducted, along with the delineation of **future directions** in the research theme of individual and family psychosocial adaptation in the context of pediatric conditions in general, and CP in particular. Finally, the **clinical implications** of the observed main findings are discussed in terms of evidence-based guidelines for clinical and psychosocial rehabilitation practice.

1. Summary and integration of main findings

During the research process underlying the present dissertation, four empirical studies were conducted to develop the Portuguese versions of Disabkids-37 HRQL questionnaires (Study I), to describe psychosocial determinants and adaptation outcomes of children/adolescents with CP (Study II) and their parents (Study III), and to examine a mediation model of social support within a parent-child perspective in the context of pediatric CP (Study IV). More specifically, we sought to determine the psychometric adequacy of the Portuguese versions of Disabkids-37 questionnaires to assess HRQL outcomes in two distinct age groups, namely children and adolescents (Study I). Subsequently, another study (Study II) examined a potential adaptation mechanism in which social support was hypothesized to determine HRQL outcomes in children/adolescents with CP, via its influence on their levels of internalizing and externalizing psychological problems. A multidimensional assessment of the caregiving experience of parents of children/adolescents with CP was conducted in Study III, where the associations between caregiving burdens and uplifts were examined in relation to those parents' QL outcomes. Finally, a last study assessed the (in)variance of a parent-child social support deterioration model in CP (clinical) and non-CP (healthy controls) samples (Study IV). Having comprehensively described the respective results in each one of the aforementioned studies, we now highlight the most important findings brought by the research developed:

1. Despite the fact that Disabkids-37 questionnaires cover a wide age range, the developmental and psychometric adequacy of the Portuguese versions for children (8-12 years old) and adolescents (13-18 years old) was confirmed throughout the phases of instrument cross-cultural adaptation, namely semantic validation and field psychometric examination.

2. Children and adolescents with CP reported more negative perceptions of social support than their typically developing peers, but no significant differences emerged in terms of their psychological adjustment.

3. Perceptions of social support were moderately associated with the HRQL of children and adolescents with CP. Internalizing and externalizing problems were found to mediate the link between social support and HRQL outcomes in this pediatric group, and those indirect effects of social support were not conditional upon age group or gender.

4. Parents of children/adolescents with CP reported levels of QL that were similar to those observed in parents of typically developing children/adolescents. Nevertheless, the former group of parents reported the experience of more subjective burden and less caregiving uplifts than the latter.

5. Subjective burden had small to medium main effects on all QL domains of parents of children/adolescents with CP. Caregiving uplifts, on the other hand, had small main effects on these parents' psychological and social QL. Caregiving uplifts were found to moderate the associations between objective burden and psychological QL, and between relationship burden and social QL.

6. Caregiving burden was found to be linked to parents and their children's psychological maladjustment and QL both directly (except for children's QL) and indirectly, through their subjective appraisals of social support. These patterns of associations were invariant in dyads of parents and their children with and without CP.

The Disabkids project in Portugal: The developmental and psychometric adequacy of the Portuguese versions of Disabkids-37 to assess HRQL outcomes in children and adolescents

Standing as a major outcome from this research project, the European Portuguese versions of Disabkids-37 (generic module, long version) were made available and have been requested for a number of clinical and scientific utilizations in Portugal ever since. Results from **the process of semantic validation confirmed the importance, comprehensibility and suitability of DISABKIDS-37 questionnaires** in general, and their items in particular, for children and adolescents with chronic conditions and their parents. The methodological procedures adopted for the development of the Portuguese versions of Disabkids-37 were in agreement with updated guidelines for the translation and cross-cultural adaptation of HRQL instruments (Hambleton, 2005; Schmidt & Bullinger, 2003; The Disabkids Group, 2004). In fact, the results observed in the preliminary empirical study (regarding semantic and pilot validation analyses) were remarkably similar to those reported for the same phases of Disabkids-37 cross-cultural adaptation in Mexico (Medina-Castro, 2007) and Brazil (Fegadolli et al., 2010). This similarity in results between countries is thus likely to reflect the consistency of cross-cultural adaptation procedures that were implemented throughout different adaptation processes and systematically supervised by the original European coordination center. The first phases of the

development of the Portuguese versions of Disabkids-37 were conducted in tandem with the Brazilian Disabkids Group, and during that time a considerable attention was devoted to translation equivalence as a mean of achieving the desired conceptual equivalence (Skevington, 2002). This "conceptual equivalence" was essentially concerned with the establishment of commonalities in the ways different populations may conceptualize health and QL, thus endorsing a universalist approach to HRQL instrument development (Herdman, Fox-Rushby, & Badia, 1998).

Since a "test adaptation guideline" has been defined as a relevant practice for performing the adaptation of psychometric tests (Hambleton, 2005), the quality and comparability of semantic and pilot validation results, for the Portuguese versions of Disabkids-37, attest the pertinence of assuming cross-cultural adaptation procedures proposed by The Disabkids Group (2004), as effective guidelines for pediatric HRQL instrument adaptation. As a matter of fact, it is worth noting several advantages in adapting preexisting HRQL measures to another cultural context of a target population, such as: the provision of a common instrument to assess HRQL in different cultural contexts; the availability of a standard measure for use in international, multicentric studies; the facilitation of including immigrants in national studies, thus avoiding the frequent bias of exclusively depicting the dominant culture of the country; and the reduction of financial and time costs commonly related to the generation of a new instrument (Guillemin et al., 1993).

The fact that the Disabkids-37 questionnaires cover a wide age range (from 8 to 18 years old) may be regarded as both the instruments' greatest advantage and greatest risk. If on the one hand age universal markers enable consistent sets of items that can be straightforwardly compared across age groups, on the other hand, important age-specific information is likely to be missed (Wallander et al., 2001). Given the interest of Disabkids project in substantiating a developmental approach to HRQL assessment, the construction of a single instrument, which could be used longitudinally from childhood to adolescence, made good sense in terms of its clinical and research applications. One way of examining the developmental and psychometric adequacy of a broadband instrument like Disabkids-37, is to conduct separate analyses for different age groups (Gerharz et al., 2003; Matza et al., 2004). In the preliminary cross-cultural adaptation study, **age-appropriate formatting, wording, design and content was acknowledged for Disabkids-37** by children and adolescents with chronic health conditions, as well as their parents. In addition, that same generic validation was obtained from teachers with experience in teaching pediatric populations and from experts in the areas of child psychological

assessment and pediatric psychology. Subsequently, Study I further analyzed the psychometric properties of Disabkids-37 in age-stratified and mixed samples, and verified a highly satisfactory psychometric performance for both children and adolescents with chronic conditions. In addition, Disabkids-37 also discriminated between age-groups (self-reports) and gender (parent-reports), thus suggesting its ability to map significant developmental differences in HRQL assessments.

Despite the fact that basic domains of HRQL may be equally relevant for different age groups (Bruil & Detmar, 2005), their particular operationalization in a given assessment procedure is likely to elicit additional challenges. Moreover, even if Disabkids project has directly involved children and adolescents since the earlier phases of project development (i.e., participation in focus groups underlying item development) (The Disabkids Group, 2006), the examination of QL assessments that cover a wide range of ages, with stratification of results, has been performed infrequently (Gerharz et al., 2003). Therefore, our results are especially important because they highlight **the psychometric quality and the developmental adequacy and sensibility of Disabkids-37 for use in children and adolescents with chronic medical conditions**. Notwithstanding these general observations, Study I also reported slight discrepancies in results between children and adolescents age-groups, mostly in terms of the instrument's reliability. Even if those inconsistencies did not significantly affect the study's main findings and conclusions, our understanding is that they may be indicative of a need for conducting further psychometric analyses using modern statistical techniques, such as confirmatory factor analysis or Rasch analysis, in addition to those based on traditional or classical test theory.

As a final remark on this topic, one should remind that the pertinence of pediatric HRQL assessment is not consensual: HRQL has been commented as narrow concept (almost resembling the notion of "disease impact"), and its utilization has been criticized as a mean of differentiating QL outcomes that should be common for "those who have specific health conditions, any health conditions, and no health condition" (Wallander et al., 2001, p. 573). While we endorse a definition of QL that is closely linked to universal standards of human rights (cf. Wallander et al., 2001), our preferred approach to QL and HRQL assessment in children and adolescents is best described as **"differential"**. First, QL and HRQL measures are best understood within a "pyramid model" with different levels, where generic QL instruments (e.g., Kidscreen Questionnaires) stand on the basis, generic HRQL instruments (e.g., Disabkids-37) occupy an intermediate level, and condition-specific instruments (e.g., Disabkids specific module

for CP) are placed on the top (the direction of the "pyramid" follows the dimension of the target populations) (cf. Baars et al., 2005). Second, in agreement with the finality of a given assessment, the administration of different measures in the study of a clinical case or a research group is likely to improve assessment outcomes, by increasing their depth; nevertheless, the election of a particular QL or HRQL measure should follow the differentiation of the primary aims underlying the need of assessment. For example, a Kidscreen questionnaire may be particularly useful in an epidemiological study aimed at detecting discrepancies in health care needs between children/adolescents with CP and their healthy peers (cf. Rajmil et al., 2006); in another stance, a clinician may find the administration of Disabkids-37 most pertinent to monitor the impact of an intervention targeting an adolescent' medical compliance; still at a different point, a research team may be interested in assessing the impact of a communication device on the daily well-being of a child with CP, and perhaps this could be best achieved through the consideration of Disabkids CP specific module. Third and last, we reiterate that QL standards must be universal and thus advocate the universality of human rights (Wallander et al., 2001); nevertheless, for the moment, we endorse the incorporation of HRQL measures in pediatric settings as a mean of refining a developmental approach to QL assessment: health and treatment contexts may assume a developmental prominence for children and adolescents with chronic health conditions, which is over and above the normative level of health/treatment experiences of their healthy peers. Therefore, the consideration of those contexts in HRQL assessment is likely to prevent the disregard of crucial developmental differences and elucidate about health-related specificities in the well-being experienced by pediatric populations.

Social support and psychological adjustment in children and adolescents with and without CP: The importance of (no) differences

Children and adolescents with CP reported lower levels of social support than their typically developing peers. Although this difference was statistically small, this result is indicative of some impairment in the subjective appraisals of social support (i.e., satisfaction with social support) of children and adolescents with CP. This finding represents a first empirical evidence for the levels of social support perceived by this pediatric population, and emphasizes social support perceptions as a mean of indirectly targeting contextual factors in intervention. Even if contextual factors in general have been commented as foremost QL determinants for children and adolescents with CP (Dickinson et al., 2007; Majnemer et al., 2007), our finding highlights the specific need for improving their social support appraisals. In pediatric settings,

physical restrictions have been related to restricted social activities (Meijer, Sinnema, Bijstra, Mellenbergh, & Wolters, 2000); in addition, though youngsters with chronic conditions do not have more peer problems than their healthy peers, children with conditions that are stigmatizing or that affect the central nervous system (like CP), have been reported to encounter significant peer difficulties (LaGreca et al., 2002). Therefore, our finding substantiates these former generic reports for the context of pediatric CP, while taking into account these children and adolescent's perceptions.

Social support was stated as a social-ecological factor in the disability-stress-coping model (Wallander et al., 1989a), but it was measured in our study through individuals' subjective appraisals (perceptions, satisfaction). Although such assessment has been recommended for studies examining relations between social support and well-being (Vaux & Harrison, 1985), as it was the case, the study of child and adolescent's developmental contexts is likely to increase accuracy (and complexity) with the inclusion of multiple informants (Cummings et al., 2000). Moreover, our results are solely based on an overall score of social support that was computed through a variety of items on different facets of social support, such as support from parents, support from friends, and involvement in social activities (Gaspar et al., 2009). Given the fact that effects of support sources or providers may be quite differential in children and adolescents (Bokhorst, Sumter, & Westenberg, 2010; Colarossi & Eccles, 2003), our finding is merely indicative of a general tendency in the reported perceptions of social support by children and adolescents with CP. Nevertheless, our study has notably given voice to a pediatric group whose social support perceptions had been characterized infrequently; in addition, it provided a first glance on the impairment of those perceptions, which reflect in any case these children/adolescents' satisfaction with parental support, relationships with peers/friends and/or social involvement.

On the other hand, since no significant differences emerged between CP and non-CP samples, **our findings contrast with previous reports of (greatly) increased psychological maladjustment in children and adolescents with CP** (Brossard-Racine et al., 2012a, 2012b; McDermott et al., 1996). Possible explanations for this contrast certainly include methodological options, since previous studies heavily (if not exclusively) relied on parents' reports and on comparisons with norms, which have been commented to increase risk detection (Lavigne & Faier-Routman, 1992). On the other hand, sampling issues in our study could be raised as an alternative reason for the aforementioned results, since most participants included in the CP sample represented milder forms of CP, with few limitations in walking. Nonetheless, it is worth

noting that better gross motor function has been associated with increased psychological maladjustment (Parkes et al., 2008) and poorer QL (Arnaud et al., 2008), and for this reason, we do not simply attribute the nature and dimension of our findings to the sampling characteristics of our study. In any case, some of those specific findings support the relevance of assuming parent and their children's reports as complementary to each other, and not as mutually exclusive. The slight discrepancies between parents and their children's reports were relatively unsurprising, since scores on psychosocial adjustment tend to differ by respondent, with parents generally providing the most negative ratings of their children's psychological adjustment (Barlow & Ellard, 2006). As regards such discrepancy, children/adolescents with CP might have underreported their psychological problems either because they wanted to portray themselves as healthy functioning individuals, or because their parents underestimated the ability of children to adapt toward their condition (Pinquart & Shen, 2011).

Social support and HRQL outcomes in pediatric CP: The mediating role of internalizing and externalizing problems

Our empirical research gathered preliminary evidence for a potential adaptation mechanism of children and adolescents with CP, in which **social support was linked to HRQL outcomes via its associations with internalizing and externalizing problems**. In addition, the observed results confirmed that this mediation model was not conditional upon age group or gender. This is to say that our findings corroborated an original indirect effect model of social support for children and adolescents with CP (cf. Bovier et al., 2004; Ensel & Lin, 1991). In fact, the originality of this mediation model is best understood as an integration of two series of previous findings: first, social support had been studied as a significant determinant of psychological adjustment and other health-related variables, including QL (Colarossi & Eccles, 2003; Helgeson, 2003; Malkowska, Mazur, & Woynarowska, 2004); and second, psychological adjustment had been described as a foremost determinant of QL outcomes (Bovier et al., 2004; Janssen et al., 2010).

In the context of pediatric psychology, the contribution of our findings may be regarded as an extension of the evidence gathered for the disability-stress-coping model (Wallander et al., 1989a), now in the context of pediatric CP. Firstly, Wallander and Varni (1989) had demonstrated that social support predicted internalizing and externalizing problems in children with chronic physical conditions, in a study that was exclusively based on mothers' reports on

their children adjustment. Secondly, social support had been stated as a determinant and resistance factor in the disability-stress-coping model (Wallander et al., 1989a), which nevertheless did not distinguish between specific and general levels of the so-called "adaptation outcomes". Given the fact that mediation analyses allow theory development and testing (Preacher & Hayes, 2004), our results underlined **social support as an important predictor of HRQL outcomes in children and adolescents with CP**, and clarified a possible mechanism via which social support may exert its effects on their HRQL, namely through both dimensions of psychological (mal)adjustment (i.e., internalizing and externalizing problems).

For the purpose of interpreting the corroborated model, it is worth recalling that stressful social circumstances, such as being restricted from positive activities and being teased by peers, may elevate internalizing and externalizing problems, respectively (Pinquart & Shen, 2011). In fact, perceived social support may affect psychological adjustment by increasing beliefs and skills that are negatively related to those two psychopathological dimensions (Bornstein, Hahn, & Haynes, 2010; Colarossi & Eccles, 2003).Complementarily, increased psychological difficulties may deteriorate an individual's resources and thus impair his/her QL (Bovier et al., 2004). Taken altogether, the applicability of the model stands clear for the purposes of guiding preventive and psychosocial rehabilitation interventions directed at children and adolescents with CP. Furthermore, the developmental adequacy of the overall model was validated in our study, since those indirect effects of social support were not conditional upon gender or age group (even if gender and/or age differences may exist at the level of single variables or in specific associations between two variables). In order to ascertain its consistency and clinical validity, the mediation model just described was observed to be valid in a research design exclusively relying on children/adolescents' self-reports, and in another one crossing children/adolescents' self-reported social support with parent-reported psychological maladjustment and HRQL.

The caregiving experience of parents of children/adolescents with CP: How does it compare to that of parents with typically developing children/adolescents?

For the purpose of the present discussion, the expression "caregiving experience" relates to parents' QL and caregiving burden and uplifts. In our empirical research, **parents of children/adolescents with CP presented a similar QL to that reported by parents of typically developing children**. Nevertheless, the former group reported **more subjective burden and less caregiving uplifts** than the latter, and those differences were medium-sized. In general, parents of adolescents experienced a worse psychological QL and less caregiving uplifts than parents of children, though these differences had a small magnitude effect.

The fact that **no differences emerged between the QL of parents with and without a child with CP** represents an innovative finding, since it challenges a number of previous assertions and research reports that widely underscored the impairment of their health, well-being and QL (e.g., Brehaut et al., 2004; Davis et al., 2010; Hatzmann et al., 2009; Okurowska-Zawada et al., 2011; Romeo et al., 2010). Although most cases in our sample were milder forms of CP, the observed lack of differences may be hardly attributed to that fact, since a lower level of child's impairment is not necessary related to parental adaptation (Manuel et al., 2000). On the other hand, the verification of **increased levels of subjective burden** (i.e., psychological burden) in these parents confirmed some previous findings (Majnemer et al., 2012; Wang & Jong, 2004). Subjective burden has been defined as the mental pain related to caregiving, and encompasses negative feelings associated with caregiving and the child's disability, such as tension, hopelessness, guilt, shame, resentment and entrapment (Green, 2007; Schwartz, 2003). Unlike perceived objective and relationship burdens, which are largely implied by the occurrence of certain stressful events, such as time constraints or parent-child conflicts, subjective burden denotes caregivers' attitudes towards caregiving experience, and is thus the product of a distinctive, interpretative process (Chou, 2000). Interestingly, this notion of subjective burden appears conceptually related to the classical concept of "chronic sorrow"; this term has been coined and applied to describe the grief experienced by parents of mentally or physically disabled children in their struggle to cope with the loss of a "perfect child" (Olshansky, 1962; Burke, Hainsworth, Eakes, & Lindgren, 1992). Similarly to the notion of subjective burden, chronic sorrow is commonly experienced not only with sadness and sorrow, but also with fear, helplessness, anger, frustration and other feelings typical of grieving states (Eakes, Burke, & Hainsworth, 1998). In a recent study, many parents of children with CP were observed to experience increased grief following a triggering event, even several years after their child's diagnosis (Whittingham, Wee, Sanders, & Boyd, 2012a). In this sense, our findings add evidence for an intensification of negative feelings related to caregiving in parents of children and adolescents with CP, which may be linked to an underlying chronic sorrow over their child's diagnosis.

As illustrated by our results, the circumstance of caring for a child with a chronic physical condition, though challenging and potentially stressful, does not necessarily equate to negative adaptation outcomes. This observation specifically underlines the pertinence of adopting a **risk-resilience and strength-based approach** (Beresford, 1994; Rentinck et al., 2006) in the research work and clinical intervention with parents who have children with CP. A central tenet of such strength-based approach is that family adaptation to the adversity of chronic physical

conditions is best achieved and understood through the identification and development of families' strengths and capabilities (Judge, 1998). In this context, positive reappraisals on caregiving demands that seem unmanageable have been stated as effective coping mechanisms to sustain the well-being of parents who have children with disabilities (Judge, 1998, Larson, 2010). In our study, the construct of "caregiving uplifts" was preferred to assess positive emotional states related to caregiving and the extent to which parents perceived their own caregiving as emotionally and mentally gratifying.

The fact that **parents of children/adolescents with CP reported fewer caregiving uplifts than parents with typically developing children** reveals a decrease in the experience of positive emotions, which are assumed as important facilitators of adaptive coping and adjustment to the chronic stress of caregiving (Folkman, 1997). Complementarily, the experience of positive emotions related to caregiving (i.e., caregiving uplifts) may be indicative of appraising stressful situations more as a challenge than as threat (Folkman & Moskowitz, 2000). Therefore, the experience of caregiving uplifts may be more common for parents of children with disabilities that approach their caregiving as a "calling" or a "commitment", than for those who perceive it as a "tragedy" or a "punishment" (Gupta & Singhal, 2004; Schwartz, 2003). The combination of our findings on increased subjective burden and decreased caregiving uplifts is particularly dramatic, because parents of children with disabilities are more likely to experience gratification from caregiving when they perceive low subjective burden (Schwartz, 2003). In addition, positive reappraisals to caregiving have been conjectured as effective internal resources to manage negative feelings associated with chronic sorrow (Eakes et al., 1998). Our findings further suggest that despite the fact that parents in general experience intense negative and positive affect during parenting (Duncan, Coatsworth, & Greenberg, 2009), the experience of those emotions may be markedly unbalanced in parents who have children with CP. Furthermore, in our study, caregiving uplifts were observed to be fewer in parents of adolescents than in parents of children, and this may be particularly damaging for parents of adolescents with CP, who may be facing increased stress, paired with decreased positive coping appraisals, during a particularly challenging period of development (Ha, Hong, Seltzer, & Greenberg, 2008; Lin, 2000). In short, our findings seem to depict the difficulty of these parents in "embracing the paradox" of caring for their children with CP (Larson, 1998), since they end up experiencing more subjective burden and fewer caregiving uplifts than parents of typically developing children. As regards this particular phenomenon, Green (2007) observed that perceived stigma could impact on subjective burden and hence decrease the perceived benefits of caring for a child with a disability; however, such claim has not been specifically addressed for pediatric CP

and thus remains as an interesting, unexplored research question. Since the "embrace of the paradox" is much about acknowledging the coexistence of positive and negative aspects in caregiving, the experience of increased uplifts could well denote "a stage beyond acceptance, which involves appreciation of a positive aspect of life with a child with a chronic disability, such as the parent's personal growth" (Schwartz, 2003, p. 583).

Quality of life and family caregiving in pediatric CP: Main and interaction effects of caregiving burden and uplifts

Our empirical research implemented a multidimensional approach to caregiving burden, which encompassed anxious and depressive feelings (subjective burden), disruptions in dyadic parent-child relationships (relationship burden), and time infringements (objective burden) resulting from caregiving. This multidimensionality, though enabling greater specificity in the analysis of the obtained results and their implications, logically increased their complexity. Having described and commented in greater detail the differential effects of each burden dimension on the physical, psychological and social QL domains (*vide* Study III), for the purpose of the present discussion, it is worth highlighting that subjective burden was the only burden dimension that displayed small to medium direct effects on the three QL domains. These results demonstrate the pervasive negative impact that subjective burden is likely to have on the QL of parents who have children with CP, indicating that the increase in negative feelings related to caregiving may elicit extensive detrimental effects on their physical, psychological and social function and well-being. The **differential impact of burden dimensions** on these parents' QL is in agreement the idea that those dimensions represent relatively independent constructs, which go beyond the mere assumption of workload per se as the major predictor of caregiver outcomes (Savundranayagam et al., 2011). Even if most researchers tend to use a global burden measure resulting from the integration of different burden dimensions (Pinquart & Sörensen, 2003), our multidimensional assessment allowed a specification of previous research findings (Canning et al., 1996; Ha et al., 2008; Raina et al., 2005; Wang & Jong, 2004) in the sense that subjective burden in particular, emerged as a foremost predictor for the QL outcomes of parents of children and adolescents with CP. Therefore, these results also suggest that this specific type of burden is probably more associated with parents' adjustment than the child's disability defined by objective parameters (Horton & Wallander, 2001).

While incorporating the assessment of negative and positive dimensions of caregiving (Green, 2007; Sales, 2003), our empirical research also revealed small **direct effects of caregiving uplifts on the psychological and social QL of parents of children/adolescents with CP**. Although positive perceptions related to caregiving have been commented to have a positive impact on parents' physical health (Green, 2007), we did not find support for the applicability of such claim in the context of pediatric CP. In our study, the operationalization of caregiving uplifts was implicitly linked to two classes of coping mechanisms, namely positive reappraisal (i.e., focus on the good in what is happening or what has happened) and creation of positive events (i.e., creating a positive psychological time-out by imputing positive meaning to ordinary events) (cf. Folkman & Moskowitz, 2000). Complementarily, caregiving uplifts were defined as positive psychological states derived from caregiving responsibilities. The role of such positive emotions on human adaptation has been formulated in the "broaden-and-build model of positive emotions" (Frederickson, 1998). According to this model, positive emotions not only "broaden" the scope of attention (e.g., noticing achievements or positive events), cognition (e.g., integrating opposites and thinking creatively) and action (e.g., involving oneself in more varied activities), but also "build" physical (e.g., muscle growth from joyful physical exercise), intellectual (e.g. psychological flexibility) and social (e.g. pro-social behavior) resources. Therefore, the experience of caregiving uplifts in parents of children/adolescents with CP may support well-being (namely psychological QL) by providing a sense of stability, facilitating personal agency and persistence, and redefining daily priorities (Larson, 2010). On the other hand, given the significance of positive emotions in establishing and maintaining social relationships (Frederickson, 1998), such caregiving uplifts may well have beneficial effects on these parents' social QL.

Positive and negative emotions tend to co-occur in chronically stressful situations, and those positive emotions may serve the functions of sustaining coping efforts, providing a "breather" and restoring exhausted resources (Folkman, 1997; Folkman & Moskowitz, 2000). These premises served as the basic rationale for our examination of **caregiving uplifts as moderators** of the associations between caregiving burden and QL outcomes of parents of children/adolescents with CP. Consequently, we found that caregiving uplifts moderated the associations between objective burden and psychological QL, and between relationship burden and social QL. In the first situation, caregiving uplifts were observed to attenuate the negative impact of objective burden on parents' psychological QL, even if that mitigation effect was larger in low-medium than in high objective burden conditions. This result confirmed the previous suggestion that caregiving uplifts could reduce the negative effects of objective burden on

parents' psychological outcomes (Gupta & Singhal, 2004; Pinquart & Sörensen, 2003); nevertheless, it also indicated that caregiving uplifts are expected to weaken (or even loose) that attenuating effect in conditions of considerable objective burden. In the second situation, parents experiencing high levels of caregiving uplifts reported a better social QL than those experiencing low to medium levels of uplifts, across all conditions of burden intensity; moreover, parents reporting the highest frequency of caregiving uplifts exhibited a relative stability in their Social QL across all levels of relationship burden. This particular result suggests that a greater frequency of uplifts may prevent or reduce the deleterious effects of the burden arising from parent-child caregiving relationship, on the social QL of parents who have children with CP. Following a conceptual distinction proposed in the context of pediatric psychology (cf. Rose et al., 2004), in both situations of moderating effects, caregiving uplifts functioned as resource factors (and not as protective factors), since they positively influenced QL outcomes across all conditions of burden intensity (i.e., regardless of the presence of adversity). Taken altogether, our findings on the associations between caregiving burden and uplifts and the QL of parents of children/adolescents with CP draw attention to the pertinence of refining more classical theoretical formulations on family adaptation to pediatric conditions, such as the disability-stress-coping model (Wallander et al., 1989a), through the integration of two specific considerations: first, different types of caregiving burden may influence parents' adaptation outcomes quite distinctively, and second, the role of positive reappraisals in the context of stress processing mechanisms is to be highlighted.

Caregiving burden and parent-child adaptation outcomes: The extensive validity of a social support deterioration model

In a last empirical study conducted for the present dissertation, caregiving burden was found to be linked to the adaptation outcomes of children/adolescents with CP and their parents, through their perceptions of social support. In addition, the validity of such potential adaptation mechanism was observed for dyads of parents and their children with and without CP. The theoretical rationale for the conduction of this study was essentially based on the **"social support deterioration model"**, which stated that chronic, traumatizing or stigmatizing events might either elicit avoidant and inadequate responses by members of the social network, or exhaust their supportiveness through frequent help-seeking behaviors, thus leading to more negative perceptions of support, which could in turn impair adaptation outcomes (Barrera, 1986; Lin & Ensel, 1984). This conceptual model had been previously examined in mothers of children

with a disability (Quittner et al., 1990), but nevertheless, our empirical research represented a significant expansion of previous findings and formulations by providing three innovative contributions: first, adaptation outcomes were not limited to psychological distress, but also encompassed the more positive and overarching concept of QL; second, the model's adequacy was examined for parent and child adaptation levels; and third, the applicability of the model was established for dyads of parents and their children with and without CP. Taken altogether, our findings gathered specific evidence for some general claims that have been commented in literature, namely that caregiving burden may have negative consequences for parents (e.g., deterioration of health status, occurrence of psychological problems, restrictions in social activities) and their children as care-recipients (e.g., increased psychological distress) (Chou, 2000).

On the level of parental adaptation, **caregiving burden was linked to the psychological maladjustment and the QL of parents of children/adolescents with CP, via their perceived social support**. This is to say that increased burden may deteriorate these parents' satisfaction with social support by diverse means (the dimensions stated between brackets follow the scaling of the social support measures used in our study): it may reduce time and opportunities for meeting up with friends (satisfaction with friendships); it may elicit perceptions of the difficulty in finding someone who truly understands one's situation, someone to help "getting it off one's chest" (intimacy); it may also require the dedication of an excessive amount of family time to caregiving activities, or in turn impair the quality of family time spent together (satisfaction with family); and it may decrease parents' participation in social activities that promote one's feelings of belonging and connectedness (social activities). These negative perceptions of social support are then likely to impair these parents' psychological adjustment and QL through the emergence, maintenance and/or intensification of feelings of isolation, inadequacy, exhaustion, and helplessness.

On the level of child adaptation, **parents' caregiving burden was linked to the psychological maladjustment and QL of children and adolescents with CP, via their perceptions of social support**. The interpretation of this finding should bear in mind that parents are primary sources of social support for their children and act as important mediators of their children's access to other formal and informal sources of social support (Thompson et al., 2006). Consequently, increased parents' caregiving burden may reduce the amount and/or quality of family time, as well as restrict the opportunities to fully participate in age-appropriate social activities, such as meeting up with friends or attend social organizations or initiatives for

youths, and thus elicit negative perceptions of social support in children and adolescents with CP. Complementarily, this deterioration in perceived social support is likely to develop feelings of emotional deprivation, defectiveness/shame, social isolation or alienation in these children and adolescents, and thus impair their psychological adjustment and QL. On the whole, and revisiting the disability-stress-coping model (Wallander et al., 1989a), our results offer additional insights on the understanding of individual and family adaptation to pediatric conditions. In the first place, although social support is stated in the disability-stress-coping model as a "resistance factor", thus suggesting its examination as a moderator variable (cf. Holmbeck, 1997), our findings are indicative of the pertinence of (also) examining it as a mediator variable between stressors and outcomes. Furthermore, our results underline the significance of incorporating a dyadic perspective in the study and promotion of parents and their children's adaptation to chronic physical conditions.

Finally, our empirical research observed that the aforementioned **social support deterioration model was valid for both dyads of parents and their children with and without CP**. This result indicates that increased caregiving burden, either in the context of parenting a typically developing child or a child with CP, may exert detrimental effects on parents and children's adaptation outcomes through the deterioration of their perceived social support. Given the fact that caregiving is an inherent component of parenting in general (Raina et al., 2004), our understanding is that the concept of "caregiving burden" emphasizes similar aspects of parenting stress (or child-rearing stress) in the specific context of caring for a child with a chronic condition (cf. Quittner et al., 1992; Seginer, Vermulst, & Gerris, 2002; Wang & Jong, 2004). Typically developing children and adolescents (i.e. able-bodied, physically healthy) experience periods of emotional turmoil and may face significant behavioral and academic difficulties, which were not controlled within the sampling frames for our study, but may actually increase their parents' stress, or more specifically, their caregiving demands (Seginer et al., 2002). Therefore, our results on the invariance of a psychosocial adaptation mechanism, namely a social support deterioration model, between CP and non-CP samples, reveal important similarities that are usually understated in literature, and thus add evidence for the existence of general mechanisms of childhood adaptation (Daniels et al., 1987).

The psychosocial adaptation of children and adolescents with CP and their parents: A different matter or the matter of a difference?

Having commented the core results from our empirical research, this last section intends to provide a final, conciliatory remark on the essential meaning of those findings. For that purpose, the title of the dissertation is now revisited in order to propose an answer for the question posed: is the psychosocial adaptation of these parents and their children a different matter, or is it a matter of a difference? This question sharply confronts two opposite appraisals on the lives, development and adaptation of individuals with chronic physical conditions and their families. On the one hand, the question may sound rhetorical in scientific terms, since the assumption of this scientific object as "a different matter" could be partly verified in the detection of major differences in adaptation mechanisms, or would ultimately not permit the conduction of between-group comparison analyses or the straightforward consideration of universal models on human development and adaptation to adversity for its study, as it happened in our research work. On the other hand, that same question portrays the constructed, negativistic views commonly held by society as regards the lives of people with chronic physical conditions (or "disabilities", to emphasize the visibility of that physical difference in the case of CP) and the families caring for them. In this case, the existence of a chronic condition or disability, such as CP, tends to be regarded as a tragedy, as a living condition that basically deserves pity and sorrow, and perhaps most gravely, as a deterministic factor to infer a negative life quality and substantially different needs throughout development (Dickinson et al., 2007; Green, 2007; Gupta & Singhal, 2004; Larson, 1998).

Therefore, the theoretical framework (e.g., risk and resilience models) and methodological design (e.g., comparative analysis between CP and non-CP samples) underlying the present dissertation, immediately depict our understanding on the question posed in its title: **the psychosocial adaptation of these parents and their children is not an inherently different matter, but rather a constructive (not to say constructed) "matter of a difference"**. Despite the (unfortunate) fact that a physical difference, such as CP, may pose specific challenges and eventually originate negative physical, psychological and social outcomes under certain circumstances, our approach to those additional differences is that they are important signs and markers on what still needs to be done for the improvement of those lives. In clinical and social interventions aimed at that improvement, it is then particularly important to bear in mind that sometimes "the many individual differences between people are more important than their similarities" (Skevington, 2002, p. 136). To put it briefly, the working

question in those contexts should not be the difference per se (i.e., "a different matter"), but rather the issue of how people perceive that difference (i.e., "the matter of a difference"), and ultimately, how they adapt (to) it. In this sense, the psychosocial adaptation of children/adolescents with CP and their parents is best understood within the contemporary social model of disability, which describes "disability" as "resulting from the interaction between individuals and their respective environments rather than as something within the individual" (Colver, 2006, p. 502). Actually, the summary of our main findings in comparative analyses is reasonably aligned with those premises: the observed differences indicated lower levels of social support for children and adolescents with CP, and increased subjective burden and decreased caregiving uplifts for their parents, in comparison to other parents and their typically developing children. Complementarily, no significant differences emerged in the psychological adjustment of children/adolescents with CP or in their parents' QL and specific types of caregiving burden (i.e. objective and relational), and the similarity of specific adaptation mechanisms in CP and non-CP parent-child dyads was supported. In sum, one may say that differences and similarities coexisted in our results.

Besides our methodological design and research findings, the adopted theoretical framework provides the primary rationale for approaching the psychosocial adaptation of this pediatric population as "a matter of a difference": firstly, the investigation of non-normative conditions can expand our knowledge on normal developmental processes (Cicchetti, 2006); and secondly, despite the fact that specific theoretical models (e.g. the disability-stress-coping model) may increase effectiveness in researching and working with specific populations (e.g., pediatric populations, such as children/adolescents with CP and their parents), they do not discard the pertinence and applicability of universal theoretical models on the understanding of human development and adaptation to adversity (e.g., Bronfenbrenner, 1986; Lazarus & Folkman, 1984). Taken altogether, the observed findings in our comparative analyses highlight those same tenets and point to the need of targeting "differences" (i.e. needs and specificities) in research and clinical contexts of pediatric CP, to a parallel extent of acknowledging and promoting "similarities" with the general population. In fact, a second cluster of our research findings was aimed at increasing the "know-how" of psychosocial interventions with these parents and their children; specifically, the observation of how social support and parents' caregiving burden may influence the adaptation outcomes of children/adolescents with CP, and the ascertainment of how caregiving burden and uplifts may determine their parents' adaptation outcomes, offered critical insights on what can be done to ensure that these families experience most of life as do other families in general.

2. Research strengths and limitations

The acknowledgement of distinctive strengths in the empirical studies that were carried-out for this dissertation, confirms the validity and pertinence of the contributions brought by our research. Firstly, the underlying research design integrated a **multidimensional and comprehensive approach to adaptation** (Barlow & Ellard, 2006; Cummings et al., 2000), thus emphasizing the assessment of positive and negative outcomes (i.e., psychological maladjustment and QL outcomes) and including measures on a number of understudied variables in children/adolescents with CP (e.g., social support) and their parents (e.g., caregiving burden and uplifts). This methodological option enabled a broadening of outcomes assessment and a clearer depiction of adaptation variables and mechanisms for this population. Secondly, the development of the Portuguese versions of Disabkids-37 questionnaires adhered to **updated and methodical test adaptation procedures** that ensured the cross-cultural and developmental adequacy of a widespread instrument to assess pediatric HRQL, which has now become available for use in Portugal. Thirdly, we sought to expand the available knowledge on the core research topic not only by the assessment of specific outcomes that were underrepresented in literature, such as the psychological adjustment of children/adolescents with CP, but also by **moving from outcomes description to the examination of potential mechanisms** linked to the promotion of those outcomes. This movement was thought to develop both theory and practice of psychosocial adaptation of children/adolescents with CP and their parents, for which evidence-based formulations and guidelines are definitely lacking. Fourthly, self and proxy-reports were obtained for the operationalization of a **complementary approach to the assessment of outcomes in children/adolescents with CP**, which has been infrequently adopted in pediatric psychology research. In addition, for the purpose of examining a **dyadic parent-child perspective** on the understanding of psychosocial adaptation in the context of pediatric CP, two-leveled analyses substantiated a distinctive procedure of "giving parents and their children a voice". Fifthly, the **utilization of a sample of controls**, which was specifically collected for the purpose of enabling reliable comparative analyses, challenged a tendency of comparing scores from CP or other pediatric samples, to norms derived from the general population. Finally, the assessment and consideration of age group differences and specificities, namely between children and adolescents, were systematically performed throughout the research project, thus endorsing previous recommendations of not straightforwardly assuming children and adolescents as a single, unified group (Livingston et al., 2007; Magill-Evans et al., 2001; Shields et al., 2006).

Notwithstanding the abovementioned strengths, some important limitations should be complementarily acknowledged and considered in the interpretation of findings. The first and the main limitation of our studies is their **cross-sectional design**. Although cross-sectional design stood as an adequate methodology for examining the prevalence of certain phenomena, as well as for identifying relationships between variables and testing hypothesized links between them, the lack of measurement over time limited interpretations from the positive establishment of causal links and directionality. This limitation was discussed throughout the series of empirical studies, but even with the commitment to a theoretically-driven research and the adoption of a number of methodological diligences to ensure the studies' validity and reliability, their cross-sectional design remains a most important limitation of our research work. In essence, we endorse the criticism that unidirectional patterns "may not fully describe what is most likely a reciprocal process" (Quittner et al., 1990, p. 1277). A second limitation relates to the **sampling frames** adopted for empirical research: all our studies were based on convenience samples, but though this limitation might be less salient in the psychometric studies, it becomes prominent for putting in perspective the results observed in the studies on CP. The clinical sample used in those studies was exclusively collected in social healthcare and rehabilitation institutions and it mainly included "milder" forms of CP with no intellectual impairment; in addition, most parents were mothers and came from low-medium socioeconomic backgrounds. Although many of these sampling limitations are common in similar pediatric psychology research (Brehaut et al., 2004; McDermott et al., 1996; Phares et al., 2005), some of them call for considerable caution in generalizing our findings to a population that is inherently characterized by marked variability (Liptak & Accardo, 2004). The **adoption of one-dimensional measures or the consideration of overall scores** for variables that are multidimensional in nature represents a third limitation in our research work. The option of using overall scores for parents' caregiving burden or their children's social support, for instance, enabled a viable operationalization of conceptual models, but it does not permit an in-depth interpretation of findings and it is still not aligned with the theoretical assumption that caregiving dimensions may impact differently on health (Savundranayagam et al., 2011), or that specific social support sources may matter distinctively for children and adolescents (Bokhorst et al., 2010). Also the utilization of a QL index is somewhat problematic, since it considerably limits interpretation of findings for a construct that is essentially described as multidimensional (The WHOQOL Group, 1995). Finally, the assessment of children and adolescents' socioeconomic background, through parent-based income and occupation measurement, may generate inconsistent findings (Boyce, Torsheim, Currie, & Zambon, 2006). Although such measurement might have been adequate for parents, it

should have been complemented with an **age-appropriate measure of socioeconomic conditions** for children and adolescents, such as the Family Affluence Scale (Boyce et al., 2006).

3. Scientific implications and future directions in research

Given the dialectical nature of knowledge development, any piece of research work is expected to offer new questions and additional insights to be explored subsequently (Connell, 1985). Following our own research work, it stands clear that there is a pertinent need of **conducting more studies that seek to translate the complexity and variability of psychosocial adaptation processes in children/adolescents with CP and their parents**. The development of future studies in this area could be significantly enriched by the incorporation of the following methodological guidelines: first, prospective and longitudinal research is needed to clarify the directionality of associations between variables and to assess the occurrence of developmental differences; second, the generalizability of results would benefit from the study of more varied samples, which are to include a wider range of CP forms, a balanced proportion of mothers and fathers, and an ample variety of cases in terms of socioeconomic backgrounds and accessibility to/utilization of healthcare infrastructures; third, quantitative data obtained from questionnaires may increase its meaningfulness with the future consideration of qualitative methods, such as structured interviews on the targeted variables; and lastly, measures on children/adolescents' QL outcomes should be preferably available in self and proxy-report formats, include generic and specific modules, and adhere to a cross-cultural perspective. As regards this latest guideline, our research work specifically underlines the utility and meaningfulness of using generic, though developmentally appropriate measures that target childhood and adolescence age groups, for detecting age-related differences or developmental (dis)continuity in QL outcomes.

Additionally, the discussion of the results observed in our empirical studies permits the identification of promising research venues on the topics of individual and family psychosocial adaptation in pediatric settings in general, as well as in the specific context of pediatric CP. The development of the Portuguese versions of Disabkids-37 questionnaires, described in two empirical studies, is to be understood as an initial milestone of the **continuing implementation of "The Disabkids Project" in Portugal**, which is expected to proceed with the following initiatives: conduction of additional psychometric studies on Disabkids-37, including the confirmatory analysis of its factorial structure; examination of the psychometric performance of

Disabkids-37 in understudied pediatric samples; development of the Portuguese versions of the brief version of Disabkids generic module (known as "Disabkids-12"); and the adaptation of Disabkids Smiley Questionnaire (HRQL measure for 4 to 7-year-olds) and condition-specific modules for use in Portugal (including Disabkids CP Module).

As regards the study of psychosocial adaptation in the context of pediatric CP, two particular recommendations follow our own study's limitations: first, the investigation of parents should be expanded to incorporate the **study of other family members**, such as siblings, and the **examination of family as a unit** (Barlow & Ellard, 2006); and second, the (in)variance of the adaptation mechanisms examined in our Studies II and III (and logically of other models to be developed) should be ascertained in samples with typically developing children/adolescents and their parents. Other promising venues for psychosocial research in pediatric CP may be summarized in the need for increasing the "breadth" (i.e., developmental contexts and social ecology) and "depth" (i.e., intrapsychic and meaning-making processes) of the studied adaptation variables and mechanisms. As regards the increase of "breadth" in research, the investigation of **social attitudes and cultural beliefs** as contextual precursors for meaning-making processes (cf. Gupta & Singhal, 2004; Lim & Wong, 2009) seems crucial in this particular context, since some of our findings portray a negative emotional pattern of experiencing the parenting of a child with a disability, or indicate negative perceptions of social support in children/adolescents with CP. Furthermore, in agreement with Bronfenbrenner (1977), the examination of contextual influences at the microsystem level (i.e., parents, peers and siblings) should be complemented with the consideration of **potential sources of influence at the exosystem level**, such as the child/adolescent's school environment (cf. Barros et al., 2008) or the availability of family-centered caregiving in healthcare institutions (cf. King et al., 1999. As to the need of increasing the "depth" of research, some of our findings fostered the elaboration of hypotheses regarding the role of cognitions on parents and their children's adaptation. On this topic, there is an interest to investigate meaning-making processes and their relation to beliefs on illness and disability. For parents, the study of their **resolution of the diagnosis of CP** (Marvin & Pianta, 1996) should be conducted in later phases of their child's development (i.e. childhood and adolescence), since there is some recent evidence suggesting that parents of children with CP may experience significant chronic sorrow symptoms even many years after the diagnosis (Whittingham et al., 2012a). This research could benefit greatly from the contributions arising from the so-called "third wave of behavioral and cognitive therapies" (cf. Hayes, 2004; Hayes, Luoma, Bond, Masuda, & Lillis, 2006), which offer a most interesting theoretical framework to explore **parental acceptance processes** in the context of pediatric chronic conditions. In fact,

experiential avoidance, for instance, has been recently observed to be a foremost predictor of chronic sorrow symptoms in parents of children with CP (Whittingham, Wee, Sanders, & Boyd, 2012b). For children and adolescents with CP, life satisfaction has been found to be strongly associated with the individuals' perceptions of their CP but not with their functional walking ability (Chong, Mackey, Broadbent, & Stott, 2012). This particular finding draws attention to the vital importance of assessing **cognitive representations of CP** and to further examine their developmental origins, stability and relationships with coping mechanisms. Ideally, in the context of pediatric CP, a transactional research design would be desired to clarify those dyadic meaning-making processes within parent-child developmental dynamics.

4. Clinical implications

The last section of the discussion, and hence of the dissertation, is aimed at describing the implications for psychotherapeutic and psychosocial practice with children/adolescents with CP and their parents. These implications were derived from the observed findings in our own research work and their integration in the available body of knowledge of the interrelated scientific domains of pediatric psychology, clinical psychology and developmental psychopathology. For this reason, the implications here discussed are best understood as evidence-based guidelines for clinical and psychosocial rehabilitation practice in the context of pediatric CP. Despite the fact that most of these guidelines are primarily related to a specific pediatric population, at least some of them may be applicable to the general practice of pediatric psychology or even adapted for guiding psychotherapeutic and psychosocial interventions with children and adolescents with other chronic health conditions and their parents. In general, the formulation of these clinical implications was based on three main sources of knowledge: the evidence provided by the empirical studies conducted within the present research work; our own clinical experience in the professional fields of clinical and pediatric psychology; and the general intervention principles and strategies currently established in the mainstream literature from the scientific domains of pediatric psychology (Drotar, 2006; Spirito & Kazak, 2006), cognitive-behavioral psychotherapy (Reinecke, Dattilio, & Freeman, 2003; Hayes, Strosahl, & Wilson, 1999) and health psychology (Camic & Knight, 2004).

In agreement with the theoretical framework initially outlined, the clinical implications presented in this final section are best understood within **a developmental perspective to pediatric psychology practice**. This developmental perspective essentially asserts that

assessment and intervention procedures in pediatric psychology should be framed by the context of child/adolescent developmental processes, and may be applicable at two levels of intervention: first, this developmental perspective emphasizes the understanding of the differential impact that health conditions may assume during childhood and adolescence, in relation to the characteristics that define each of those developmental periods; and second, it stresses the need of selecting and implementing therapeutic interventions in agreement with the child or adolescent's cognitive and social developmental level (Barros, 1999). On the level of family or parental intervention in pediatric psychology, it is worth noting that family has a differential impact and importance throughout the process of development, and that an emphasis on family issues in pediatric psychology does not necessarily imply the involvement of the whole family or the achievement of profound changes in intervention (Barros, 2010). The consideration of these general assumptions and principles is decisive to promote the best application of the following guidelines in clinical practice.

Incorporating generic HRQL instruments in pediatric routine assessment

Within a multi-level approach, the assessment of QL outcomes may be used to describe the function and well-being of populations with or without chronic medical conditions (epidemiological perspective), as criterion measures for intervention results and impact (clinical perspective), and as a basis for decision making in the healthcare field (political perspective) (Bullinger, 1997). Disakids-37 questionnaires are renowned pediatric HRQL instruments that may be applied by clinicians, researchers, decision-makers and healthcare providers to: document the HRQL of children and adolescents; characterize the impact of a health condition or treatment on the child/adolescent's well-being; evaluate pediatric health outcomes in health economic research, and perhaps most importantly, give parents and their children a voice in healthcare (The Disabkids Group, 2006). These questionnaires target children and adolescent's HRQL as an intermediate outcome between the outcome levels of generic and condition-specific QL measures: if on the one hand, generic QL measures may be useful to compare the levels of QL between children/adolescents with and without medical conditions, and to provide a broader picture on their subjective well-being, they are also likely to miss important health-related information; on the other hand, condition-specific measures may be more sensitive for detecting the most specific changes in HRQL, but they are of limited valued in clinical situations of comorbidity or less prevalent chronic conditions (Eiser & Morse, 2001; Spieth & Harris, 1996).

Therefore, the Disabkids-37 questionnaires are capable of providing profile and overall scores on HRQL outcomes, which may play a particularly relevant role in guiding interventions directed at children/adolescents with chronic physical conditions, for whom health-related contexts and experiences may assume a distinctive importance in their lives and development. The effects of chronic illness or disability and child/adolescent development have been described as reciprocal: for example, a chronic physical condition may impair the child/adolescent's psychosocial development (e.g., infantilization, social isolation), to a parallel extent that developmental issues may affect chronic health conditions (e.g., consequences of exploratory risk-taking behaviors, poor developed abstract thinking/planning and difficulty in imaging the future) (Suris et al., 2004).

Given their developmental adequacy for both children and adolescents, Disakids-37 questionnaires may be used as means of **implementing a developmental approach to HRQL assessment in pediatric settings**, while serving a number of clinical applications: identification of psychosocial "hidden morbidities" (Varni et al., 2005), possibly even before they assume increased clinical proportions (for example, the occasional occurrence of bullying experiences may signalize the need for developing coping strategies in interventions, and thus prevent the increasing proportions of the problem and its deleterious effects); differentiation of HRQL outcomes by age, gender and condition groups, in order to define intervention guidelines in agreement with a group's specificities or vulnerabilities; evaluation of change over time, following the child/adolescent's developmental trajectories, the natural evolution of his/her health condition or disability, or his/her response to therapeutic interventions; and the assistance for the formulation of prognoses regarding the child/adolescent's development in articulation with his/her chronic condition and related factors (Spieth & Harris, 1996; Viehweger et al., 2008).

Assessing and promoting the psychological adjustment of children and adolescents with CP

Although we have no systematic information on the importance that has been given to the psychological (mal)adjustment of children and adolescents with CP in clinical settings, recent research has been commenting that their psychosocial adjustment "remains underrepresented in current literature" (Vles et al., 2012, p. 1), and that "few studies have described behavior problems in children with CP" (Brossard-Racine et al., 2012a). These observations suggest that

there might be a possibility for psychological maladjustment standing as a neglected "hidden morbidity" in these children and adolescents.

Even though our results contrasted with the available literature on the topic, indicating that differences in psychological adjustment between CP and non-CP samples may be smaller, or even insignificant, than the reports from previous research would otherwise suggest, they do not positively discard the idea that there may be an increased risk for psychological maladjustment in children and adolescents with CP. In addition, the slight discrepancies between parents and their children's reports are clinically relevant, because parents are usually the ones who monitor their children's well-being more closely and signalize their needs for referral. Moreover, our results underlined the determinant role of psychological adjustment on the HRQL outcomes of this population (Janssen et al., 2010). Taken altogether, these results enable the proposition of two interrelated recommendations for clinicians assessing these children and adolescents' psychological adjustment: one, "do not be narrow-minded" by equating disability or complex health conditions to maladjustment, and thus admit the possibility of resilience; and two, "do not be naïve" by assuming child/adolescent's reports as the definite evidence for their level of psychological adjustment, since such positive reports may in fact entail compensatory mechanisms aimed at presenting a healthier image of oneself or a self concept as being "bullet proof".

During assessment procedures, if a child/adolescent is observed to have no clinically significant psychological difficulties, such information should be shared with parents, because it may sound surprising "from the perspective of a non-disabled adult imagining what it would be like to be disabled, but probably not from the perspective of a child whose sense of self from birth incorporates their impairment and who embraces growth, development, and living with the same excitement as most children" (Dickinson et al., 2007, p. 2177). In the clinical work with CP, especially during times of increased distress for families, **sharing positive information** may be especially important: the information on a child's "normal" psychological adjustment may be reassuring for parents and broaden their focus to positive dimensions of their child's development, and ultimately, of their role as parents. Complementarily, for clinicians interested in primarily **preventing psychological difficulties** in children and adolescents with CP, the promotion of their psychological well-being may be achieved through the facilitation of positive social relationships with parents, peers and siblings and the encouragement for effective involvement in health-related decision making (Edwards & Titman, 2010). For those children and adolescents who present increased levels of psychological maladjustment, there is currently an array of **cognitive-behavioral techniques that may be adapted and applied to the**

treatment of internalizing and externalizing problems in the context of pediatric CP (Jongsma, Peterson, McInnis, & Bruce, 2006; Reinecke et al., 2006). Interestingly, a substantial component of those interventions strategies directly or indirectly seek to improve the amount and/or quality of the individual's social relationships and support (e.g. scheduling social activities, building social skills, managing peer conflict, improving parent-child communication). Despite the fact that internalizing problems seem more prevalent than externalizing problems in children/adolescents with chronic health conditions, we suggest that this broadband classification of childhood and adolescent psychopathology should be adopted in pediatric psychology assessment and intervention processes, not only because internalizing and externalizing psychopathological dimensions tend to be related to different stressors (Pinquart & Shen, 2011), but also because they may exert differential effects in the child/adolescent's adaptation to his/her condition (Holmbeck et al., 2008).

Targeting social support perceptions of children and adolescents with CP as a means of improving their adaptation outcomes

As suggested by our findings, interventions targeting social support perceptions may display positive effects in the HRQL of children and adolescents with CP, through the improvement or promotion of their psychological adjustment. In addition, our results generally suggest that such effect may be applicable to boys and girls, and to children and adolescents, thus highlighting the overall adequacy of such social support interventions in the context of pediatric CP. Also following our own observations, it should be noted that the assessment of perceived social support may constitute a pertinent baseline to identify overall needs or at-risk groups, but it has to be further elaborated with complementary assessment procedures (e.g., measures on social embeddedness, semi-structured clinical interview), in order to determine the more specific social support needs (e.g., social skills deficit, social isolation, social anxiety), which in turn call for the delineation of specific intervention strategies. Given the fact that social support was decreased in children/adolescents with CP and linked to both their psychological adjustment and HRQL, in clinical settings, it could be targeted as a strategic variable for assessment and intervention. As for **planning social support interventions** in children and adolescents with CP, two general recommendations are noteworthy: first, as implied by these age groups, particular attention should be directed to the needs of their parents and families, which may be indicative of a need for two-generation interventions (Thompson et al., 2006); and second, social sources of internalizing problems (e.g. restriction in positive activities, peer rejection) may be

more common than social sources for externalizing problems (e.g., abusive or neglecting parents, negative peer comparisons at school) (Pinquart & Shen, 2011), and thus the implementation of generic social support interventions should be regarded as impacting differently each of those dimensions of psychological adjustment.

Interventions aimed at promoting the wellness of children and adolescents generally seek to enhance competency and positive mental health by activating social support, enriching existing social ties, modifying dysfunctional social networks or dysfunctional beliefs, and introducing new network members, such as support group participants (Barrera & Prelow, 2000). This is in fact a pertinent clinical guideline that is most aligned with our own research results. According to Gottlieb (2000), there are essentially two types of social support interventions: introduction of new ties and intervention within the natural network. The **introduction of new ties** would be most adequate for those conditions when specialized knowledge and expert opinion are needed, or when the existing social network is either impoverished/drained/conflicted, or reinforces undesirable behaviors, or lacks experiential knowledge. This type of social support intervention could be particularly indicated for those clinical situations of children/adolescents with CP that experience problems of social isolation, for instance. **Interventions within the natural network**, on the other hand, are warranted in the following conditions: when the attainment of health goals strongly depends on the behavior of one or more network members; when the current network needs strengthening to meet long-term, continuing support needs; when the presenting problem or outside intervention is highly stigmatizing, or when there is a gap between the support recipient and the external providers. This type of social support intervention would be primarily recommended for those clinical situations where the child/adolescent with CP experiences peer rejection or parental unavailability due to increased caregiving burden. At this point it is worth recalling that despite the specificity of these types of intervention, they are both assumed to eventually contribute for the improvement of children/adolescents' social support perceptions; therefore, the administration of perceived social support measures throughout the process of intervention would be recommended to monitor its effective impact on those perceptions of support.

The delineation of **social support interventions for children and adolescents with CP** may be based on five major pathways to well-being, which were identified by Cowen (1994), as follows: (1) forming wholesome attachments; (2) acquire age- and ability-appropriate competencies; (3) engineering settings that promote adaptive outcomes; (4) acquiring skills needed to cope effectively with life stressors, and (5) promoting empowerment. In the clinical

practice of pediatric psychology with this group, the following intervention strategies (mentioned as examples) could respectively substantiate the development of the aforementioned pathways: (1) promoting positive parent-child interactions and child-rearing approaches; (2) teaching social skills and scheduling the involvement in age-relevant social activities; (3) modifying parents or family's negative discourse on the child/adolescents' (dis)abilities; (4) developing the child/adolescent's coping repertoire for increasing his/her sense of mastery in managing age and condition-related stressors (e.g. being teased by peers, being unable to perform certain physical exercises in the gymnastics classes); (5) involving the child/adolescent in decision-making processes related to important aspects of his/her life and fostering his/her participation in mastery activities.

Adopting a multidimensional approach to caregiving assessment and intervention

The notion of a multidimensional approach to family caregiving measurement is two-folded: first, there is a need to assess multiple dimensions of burden because they relate differently to both key predictors and outcomes of caregiving (Chou et al., 2003; Savundranayagam et al., 2011); and second, positive dimensions of caregiving, such as caregiver's gratification and growth, should be complementarily assessed in order to provide a most complete depiction of the caregiving experience (Sales, 2003). As regards this latest aspect, it is worth noting that social, clinical and scientific discourses have repeatedly discouraged parents of children with chronic conditions from finding and acknowledging positive aspects of their caregiving (Green, 2007; Larson, 1998). In fact, a number of widespread measures to assess parents' caregiving experience in pediatric settings tend to exclusively focus on the negative dimensions of the caregiving experience, such as physical and psychosocial exhaustion associated with burden (cf. Angold et al., 1998; Brannan, Heflinger, & Bickman, 1997; England & Roberts, 1996). Although the utilization of these measures may be harmonized with the parallel administration of other instruments on positive variables, **the administration of a single measure that simultaneously encompasses positive and negative dimensions of caregiving is to be preferred**, since such procedure reduces the workload related to pediatric assessment routines (both parental and professional), and infuses that same assessment with a sense of encouraging parents to be mindful of their caregiving experience. Results from our research represented additional support for the reliability, validity and applicability of "The Revised Burden Measure" (Montgomery et al., 2006) in pediatric settings in general (cf. C. Carona, N. Silva, M. C. Canavarro, personal communication, July 27, 2011; Crespo, Carona,

Silva, Canavarro, & Dattilio, 2011), and in the context of pediatric CP in particular. Therefore, the utilization of "The Revised Burden Measure" in parents who have children with chronic conditions is recommended, because: it provides a comprehensive assessment on positive and negative dimensions of family caregiving; it is easy to administer, score and interpret; and given the overall adequacy of its items, it may be used in parents of children from different age groups and eventually throughout different phases of family development.

In clinical practice, the implementation of the aforementioned **multidimensional assessment of family caregiving may be especially useful to operationalize a strength-based approach in promoting positive family adaptation** (Beresford, 1994; Judge, 1998). To put it briefly, the core tenet of such approach is stated in the clinical pertinence of not only describing "what is wrong and how to correct it", but also to focus on "what is right and how to use it". In fact, from our own clinical experience, parents will more frequently express their interventions needs in terms of "increased caregiving exhaustion" than in terms of "decreased caregiving gratification". Therefore, it is crucial to bear in mind that parents of children/adolescents with chronic conditions or disabilities tend to experience more prolonged periods of caregiving stress than parents with typically developing children (Gupta & Singhal, 2004), and thus the assessment of their caregiving experience should not be limited to its positive dimensions. This recommendation is after all aligned with the theoretical assumption that positive and negative emotions may coexist during stress and coping processes (Folkman, 1997; Folkman & Moskowitz, 2000).

In the development of clinical interventions in pediatric settings, a multidimensional assessment of parents' caregiving burden and uplifts has a number of advantages and applications, which include the following: balancing the extent of negative and positive assessments on parents' caregiving experience; facilitating specialized referral to effectively target the most affected burden dimensions (e.g., increased levels of objective burden primarily call for distinct intervention strategies from those applied to the management of subjective or relational burdens); and determining the extent of caregiving gratification and uplifts, which may then be targeted for improvement or activation as supportive resources in clinical intervention processes.

Facilitating the development of caregiving gratification in parents who have children with CP

Our research findings illustrated how caregiving uplifts can positively affect the adaptation outcomes of parents of children/adolescents with CP, either by displaying direct

beneficial effects or by alleviating the negative impact of burden in their psychosocial QL. For this reason, in clinical practice, the promotion of positive perceptions related to caregiving may be aimed at directly improving parents' QL or at attenuating the deleterious effects of specific types of burden on their psychosocial QL. In this section, the discussion of clinical implications from those findings is generally drawn from the available literature on the topics of growth following adversity (Joseph & Linley, 2006; Tedeschi & Calhoun, 2004); stress, coping and positive emotion (Folkman, 1997; Folkman & Moskowitz, 2000); and meaning-making or mindfulness-based interventions (Larson, 2010; Shapiro, Carlson, Astin, & Freedman, 2006). In the next paragraphs, general clinical guidelines are firstly commented, and then followed by the description of more specific intervention strategies.

Different authors have commented that social context does not typically encourage parents who have children with chronic conditions in the acknowledgement and enjoyment of gratifications implied by their caregiving (Green, 2007; Gupta & Singhal, 2004). The immediate implication of such statement is that clinical context for psychotherapeutic change is expected to counteract that general tendency by broadening the scope of clinical attention to encompass both positive and negative dimensions of family caregiving. Given the fact that parents of children/adolescents with CP are most likely to experience mixed, contradictory emotions of sadness and joy, hope and fear, the clinician's role is perhaps best described as **facilitating the "embrace of caregiving paradox"** (cf. Larson, 1998).

In order to facilitate such process, it is worth noting that the alleviation of distress does not straightforwardly imply the facilitation of caregiving-related growth, gratification or perceived benefits. Instead, this (re)redefinition of the clinician's role has to be accompanied by a change in the therapeutic goals, which then incorporate the achievement of caregiving gratification (e.g., caregiving uplifts), in addition to the alleviation of distress (e.g., caregiving burden) (Joseph & Linley, 2006). In this context, clinicians should be mindful that adversity does not necessarily lead to maladjustment or positive change, and recall that **caregiving-related growth, perceived benefits or gratifications are to be encouraged, not imposed**, in the same way that they can't be created, but rather facilitated. Another general recommendation is that the facilitation of gratification should be conducted in the context of the parents' struggle with the stressful event (i.e., caring for a child with CP), and not in the event itself (i.e., having had a child with CP); this is most important not to imply that there is something inherently positive in the event (*idem*, *ibidem*).

In our own clinical work, we find most valuable the adoption of a compassionate attitude towards parents' caregiving experience (cf. Halifax, 2011), and the presentation of an intervention rationale that heavily relies on the theoretical premises proposed by the "broaden-and-build" model (Frederickson, 1998), and on the importance of acceptance, willingness and commitment, to dialectically generate change (Hayes, 2004). This crucial therapeutic strategy simply denotes the importance of counteracting widespread discourse tendencies: indeed if we don't ask positive questions to these parents, we will hardly get any positive answers (Gupta & Singhal, 2004; Schwartz, 2003).

According to Frankl (1994), there are essentially three ways of finding meaning in life: by creating a work or a deed; by experiencing something or encountering someone; and by the attitude we take toward unavoidable suffering. It is this latest pathway to meaningfulness that has to be emphasized in the facilitation of caregiving gratification in parents of children/adolescents with CP. In the light of stress and coping models, the most general goal of such therapeutic intervention is **to promote the appraisal of stressful events (i.e. continuous caregiving) "more as a challenge than as a threat"** (Folkman & Moskowitz, 2000, p. 115). In the initial phases of meaning-making interventions, a key component in the Socratic dialogue constructed with parents is a non-judgmental examination of some commonly held beliefs, such as the child's disability as "a God's punishment" or as an "ultimate tragedy", which may be related to maladaptive coping strategies, such as behavioral disinvestment, blaming, isolation or denial.

Following that motivational change, the facilitation of caregiving gratification may be mapped by the promotion of three coping mechanisms: positive reappraisal (e.g., seeing how one's efforts can benefit other people; imputing a meaning of "pride" for not leaving difficult situations); problem-focused coping (to help combat feelings of hopelessness that may characterize many aspects of the situation); and creation of positive events (e.g., scheduling positive events, noting positive events when they occur spontaneously, or instilling common events with positive meaning) (Folkman, 1997; Folkman & Moskowitz, 2000). These coping mechanisms have been described as "meaning-based" and operate at a "situational level", as opposed (but still in relation) to the most "global meaning-making" that deals with the order and predictability of life and goal-striving (Larson, 2010). This classification is aligned with the distinction between "meaning as comprehensibility" (i.e., understanding the event and why it happened) and "meaning as significance" (i.e., understanding the philosophical, world view implications of the event) (Janoff-Bulman & Frantz, 1997). In the clinical work of facilitating

caregiving-related growth and gratification, the achievement of "meaning as significance" in parents is a necessary ground for further therapeutic developments (Joseph & Linley, 2006).

The use of meaning-making reappraisals that foster a sense of control and identity has been linked to the well-being of parents caring for children with disabilities; moreover, intentional mindfulness has been suggested to generate positive emotions in those parents, which then "broaden and build" their coping repertoires and resources (Larson, 2010). Mindfulness is a core concept in the "third wave of cognitive-behavioral therapies" and is defined as "the awareness that emerges through paying attention, on purpose, in the present moment, and nonjudgementally to the unfolding of experience moment by moment" (Kabat-Zinn, 2003, p. 145). Mindfulness approaches have been applied to general parenting (Duncan et al., 2009; Hughes et al., 2009), but have also gathered some support for their effectiveness in reducing stress and increasing self-compassion and personal growth in parents of children with chronic conditions and special needs (Benn, Akiva, Arel & Roeser, 2012; Minor, Carlson, Mackenzie, Zernicke, & Jones, 2006). In the face of such promising evidence and in agreement with our own clinical observations, we suggest that **mindful-based cognitive therapy techniques** (Baer, 2006; Wells, 2006) **may be effective in facilitating caregiving-related growth, gratification and benefits** (and in reducing the psychological distress) of parents who have children with CP. The practice of mindfulness involves learning a "reperceiving" attitude towards one's feelings, thoughts and emotions, and may lead to the aforementioned positive outcomes through a series of mechanisms that include improved self-regulation, greater clarification of values, increased psychological flexibility and greater exposure (Shapiro et al., 2006). Nevertheless, the establishment of the effectiveness and the examination of the mechanisms of mindfulness-based interventions, in the adaptation of parents who have children with CP, remain to be ascertained in empirical research.

Electing caregiving burden as a strategic target for interventions, while acknowledging the similarity of specific adaptation mechanisms in families with and without a child/adolescent with CP

Main findings from our research attest the pertinence and applicability of a social support deterioration model (Lin & Ensel, 1984; Quittner et al., 1990) for understanding the effects of caregiving burden in parents-child adaptation outcomes in the context of pediatric CP. These findings suggest a number of **general guidelines for clinical assessment**: first, caregiving burden could be routinely assessed to identify parent-child dyads in greater need for caregiver

support interventions; second, burden assessment in the context of pediatric CP could be improved with the simultaneous administration of social support measures that reliably indicate the occurrence of extended detrimental effects of burden (i.e., social support deterioration); and third, the assessment of parents' burden should be conducted in relation to their own and their children's adjustment, while including measures on their specific (i.e., psychological maladjustment) and general (i.e., QL and HRQL) adaptation outcomes. Additionally, despite the fact that our findings were based on overall burden scores, we highly recommend the implementation of multidimensional assessments in practice, in order to effectively plan interventions that are most likely to meet the specific needs implied by different types of caregiving burden (Chou, 2000; Savundranayagam et al., 2011).

As regards clinical intervention, our results suggest that **the election of primary family caregivers' burden as a preferred target may well represent a strategic, cost-effective option for improving psychological and QL outcomes**, not only for parents, but also for their children with CP. This suggestion highlights the idea that family intervention in the context of pediatric CP does not have to necessarily imply the involvement of the whole family (Barros, 2010). In fact, our results indicate that parents' greater involvement in family caregiving activities may eventually impair their children's adaptation outcomes, and thus, they somehow depict what is likely to be a specific pattern of "miscarried helping" in the context of pediatric CP. "Miscarried helping" has been defined in pediatric psychology as "well-intentioned support attempts that fail because they are excessive, untimely, or inappropriate" (Anderson & Coyne, 1991, p. 167), which sometimes occurs between parents and their children with chronic health conditions. Therefore, clinical interventions should prevent parents from (physical, emotional, relational) over-involvement in family caregiving, which may ultimately exert undesirable effects in both the parent as care-provider, and the child/adolescent as care-recipient. The costs related to continuous caregiving (e.g., caregiver burnout) may be better understood as manifestations of "compassion fatigue" (Figley, 2002). Compassion is generally defined as the desire to alleviate suffering (Lazarus & Lazarus, 1994), and its application to family caregiving in the context of pediatric CP could be particularly valuable for developing parents' awareness that their child's well-being (i.e., compassion towards the others) is (also) a matter of their own well-being (i.e., self-compassion). This is to say that an essential therapeutic goal in the clinical work with these parents would be "to keep the balance right" in meeting their own and their children's needs. In psychotherapeutic interventions, the achievement of such goal could be promoted through the development of compassionate attributes and skills directed at the self and the child with CP, for which a number of clinical strategies are now available in literature (Gilbert, 2009).

The election of caregiving burden as a strategic target for intervention is also an interesting contribution for operationalizing the process of family-centered caregiving: family-centered care involves treating parents supportively and meeting family-reported needs, and it has been observed to influence the levels of caregiving burden experienced by parents of children with developmental disabilities (King et al., 1999). Following the recommended multidimensional assessment, **parents with increased levels of certain burden types may be referred to specialized intervention**: for instance, parents reporting increased objective burden may mostly benefit from sharing the tasks of caregiving, scheduling meaningful activities for rest and pleasure, or developing effective time management; other parents with marked subjective burden could be referred to psychological interventions aimed at developing emotional self-regulation, and increasing acceptance and values-committed action; other parents still, reporting elevated levels of relational burden, would be most amenable to those interventions focusing the management of behavioral difficulties or the development of specific parenting skills in the context of disability.

In general, interventions targeting parental caregiving burden may integrate any of the following dimensions: strengthening caregiver's competence (and thus developing his/her sense of mastery); developing task-specific or problem-solving skills; providing information and emotional counseling; and linking caregivers to resources (Reinhard, Given, Petlick, & Bemis, 2008). Although the effectiveness of psycho-educational interventions for parents who have children with chronic conditions is not well-established (Barlow & Ellard, 2004), it has been recently commented that cognitive-behavioral and problem-solving therapies are effective interventions for improving these parents' mental health and parenting behavior as well as their children's health (Eccleston, Palermo, Fisher, & Law, 2012). Moreover, sensible adaptations of general positive parenting programs, to the specific context of behavioral family intervention for children with developmental disabilities, have been proved to be effective in improving child's behavior and parents' stress and parenting style (Roberts, Mazzucchelli, Studman, & Sanders, 2006).

In addition to the election of caregiving burden as a strategic target in intervention, the clinical implications of its relationships with parents and their children's perceived social support are noteworthy. As suggested by Barrera (1986), in those times where caregiving burden cannot be prevented or readily changed, parents and their children might be protected from its deleterious impact (or at least its extended impact) on their perceptions of social support, by preparing parents' significant others for increased strains and to arrange for positive social

interactions. The same author actually suggests that, in such situations, **cognitive-behavioral interventions could be especially useful in preventing distorted/negative perceptions of support**. Many of the clinical guidelines for social support interventions in the context of pediatric CP, have been described in a previous section of this discussion; nevertheless, in agreement with the adopted measure for adults' social support (Pais-Ribeiro, 1999), we recommend the following guidelines for targeting these parents' perceptions of support: referring the parent for a parent support group (this may reduce the feelings of not being genuinely understood); fostering marital intimacy; arranging for participation in social or other meaningful activities; and increasing quality time with family and friends (i.e., not restricted to caregiving activities or concerns).

The last implication of our research results is perhaps the one that most strikingly goes beyond the clinical arena: our understanding is that a sizeable amount of literature from the field of pediatric psychology tends to emphasize differences, to a much larger extent than similarities, between the adaptation of pediatric and healthy, normative populations. The fact that the abovementioned social support deterioration model was valid for both dyads of parents and their children with and without CP represents additional evidence (which tends to be infrequently reported) for "a general association between certain risk and resistance factors and childhood adaptation" (Daniels et al., 1987, p. 295). The main implication of this assertion is that families of children/adolescents with CP are likely to cope with and adapt to adversity in similar ways to those observed in families with typical developing children; more specifically, our findings suggest that the deterioration of perceived support (and thus of adaptation outcomes) is related to increased caregiving burden in both CP and non-CP families. This is to say that in face of increased caregiving stress, both groups of families may display similar adaptation outcomes, through similar adaptation mechanisms. This remark does not dispute or diminishes the existence of specific challenges and prolonged periods of increased stress in families of children/adolescents with CP, but rather calls the attention for completing the characterization of such scenario with the consideration of commonalities in parent-child adaptation mechanisms and outcomes. In clinical practice, this means that **parents and their children with CP should be treated differentially** (i.e., in regard to their specificities) **and not differently** (i.e., as needing something rather different from others families facing distress and adversity). Actually, the acknowledgement of common adaptation and development mechanisms in the clinical practice with pediatric CP may be as important for families and health professionals, as their knowledge on condition-related specificities. Given the observation that health professionals may convey similar disabling attitudes to those held by society in general (Larson, 1998), **a**

"depathologizing" and empowering professional discourse may shape the families' own discourse, while ensuring that normative developmental and adaptation needs are not being neglected in the implementation of specialized interventions that are equally needed.

This clinical recommendation is then to be extended to society in general: the issues of stigma and socio-structural constraints (and their relations to burden) have been highlighted in the context of parenting a child with a disability (Green, 2007), and in this sense, our key assertion (though it may sound a truism) is that families of children/adolescents with CP need essentially the same things that any other family would need if facing a similar situation. In fact, no adversity changes our common human condition. Therefore, as a concluding remark, it is worth recalling an updated and positive discourse on disability (Dickinson et al., 2007), but now rephrased to the specific ambit of our discussion: in order to change maladaptive attitudes, we need to support social policies that acknowledge the similarity between the lives of parents and their children with and without CP, and that promote their rights as citizens, rather than as "families with a disabled child", to fully participate in society and enjoy life and fulfillment.

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ATTACHMENTS |

Preliminary Study |

Assessing Paediatric Health-related Quality of Life within a Cross-cultural Perspective: Semantic and Pilot Validation Study of the Portuguese Versions of DISABKIDS-37

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Assessing Paediatric Health-related Quality of Life
within a Cross-cultural Perspective:
Semantic and Pilot Validation Study of the Portuguese Versions of DISABKIDS-37

C. Carona, M. Bullinger, & M.C. Canavarro

Abstract

The aims of this study were to validate semantically the Portuguese versions of DISABKIDS-37 (quality of life questionnaires for children and adolescents with chronic conditions) and to explore their psychometric performance in a pilot study. For each health condition (asthma and epilepsy), groups of children (aged 8–12 years) and adolescents (aged 13–18 years) were formed, including nine individuals and their parents (total = 72 subjects), to whom the Portuguese versions of DISABKIDS-37 and the semantic validation questionnaires were administered. Data on symptom severity were obtained from parents' reports. The study also included a sample of 18 teachers with experience in teaching youths with chronic conditions. Results of semantic validation supported the relevance, comprehensibility and adequacy of the Portuguese versions. Internal consistency ranged between 0.84 and 0.92 for the self-reported version, and between 0.78 and 0.92 for the proxy-reported version. Correlations between facets and domains were mainly strong (0.61–0.77) and very strong (0.82–0.87). Self- and proxy-reported health-related quality of life (HRQoL) was lower for the epilepsy group (social domain), and proxy-reported HRQoL (physical limitation facet) was poorer for the asthma group. Adolescents reported higher levels of HRQoL than children, and parent–child concordance was weak. These results reaffirm that semantic and exploratory validation procedures are important to achieve adequate levels of equivalence between different versions of HRQoL questionnaires.

Keywords: children and adolescents; health-related quality of life; instrument adaptation.

Introduction

In order to tailor treatment approaches to patients' needs, as well as to assess their effectiveness and monitor disease trajectories, a greater emphasis on patient-reported outcomes has been proposed (Patrick, 2003). Among these, health-related quality of life (HRQoL) has become particularly salient as a multidimensional construct covering physical, emotional, mental, social and behavioural components of well-being and functioning as perceived by patients or families (Bullinger, 1997). As a result of the international interest in the multiple applications of HRQoL, the translation of many existing measures and the assessment of equivalence between source and target questionnaires have received increased attention (Herdman, Fox-Rushby, & Badia, 1997).

In contrast to the proficient progress in adult HRQoL research over the last decades, paediatric HRQoL research has developed more slowly due to conceptual and operational difficulties, such as age particularities, the selection of a source of report and the child's cognitive ability to perform that assessment (Drotar, 1998). However, the Division of Mental Health of the World Health Organization stated that measures to assess quality of life (QoL) in children should be child-centered, self-reported, age-dependent and cross-culturally comparable (Gerharz, Ravens-Sieberer, & Eiser, 1997). Integrated within a developmental framework for the study of QoL, the DISABKIDS project proved to be an innovative project integrating a variety of distinctive features, such as the cross-cultural perspective modular system with a combination of specific and generic QoL aspects, the inclusion of a wide age range and the representation of parents and children's views (The European DISABKIDS Group, 2006).

The DISABKIDS Project (Quality of life in children and adolescents with disabilities and their families – assessing patients' view and patients' needs for comprehensive care) was related closely to the KIDSCREEN Project, which was concerned with the development of a generic QoL questionnaire for children and adolescents in the general population. The instruments devised by these two projects form a three-level modular structure: generic, chronic generic and condition-specific (Baars et al., 2005; The European DISABKIDS Group, 2006; Petersen, Schmidt, Power, Bullinger, & The DISABKIDS Group, 2005). By the end of the project, an English version of DISABKIDS instrument tool set for children and for proxies included the following modules: DISABKIDS Chronic Generic Measure – long form (DCGM-37); DISABKIDS Chronic Generic Measure – short form (DCGM-12); DISABKIDS Smiley Measure; and seven condition-specific modules (Baars et al., 2005; The European DISABKIDS

Group, 2006). The focus of the present study is on the long form of the Chronic Generic Module, known as DISABKIDS-37.

According to the DISABKIDS validation procedures, the selection of two chronic conditions is required to proceed with the instrument adaptation process: one condition is asthma, common to all countries who participated in the original study or who wish to develop a new version of the questionnaires; the other condition is selected according to scientific, clinical or public health reasons with particular interest to a given country or culture. As well as asthma having been acknowledged recently as the most prevalent chronic health condition in Portuguese children up to 15 years old (INE & INSA, 2006), epilepsy was selected as the second condition for the Portuguese validation study, supported by the following reasons: (1) children and adolescents with conditions that affect the central nervous system may be at a higher risk for psychological problems than children with other chronic conditions (CCD & CPACFH, 1993); (2) epilepsy and asthma are both chronic health conditions characterized by the occurrence of unpredictable episodes that generally require regular medication intake and regular visits to a physician; and (3) neither clinical group has any outwardly observable physical deformity (Austin, Smith, Risinger, & McNelis, 1994).

Although semantic validation studies for Portuguese versions of DISABKIDS-37 have been performed previously in Brazil (Fegadolli, Reis, Martins, Bullinger, & Santos, 2010), this was the first initiative to conduct that same study in Portugal, where language differences and specificities are substantial in comparison to Brazilian Portuguese. While encouraging the development of new DISABKIDS modules and the adoption of a cross-cultural perspective in paediatric HRQoL instrument development, the objectives of our work were to validate semantically the experimental Portuguese versions of DISABKIDS-37 and to explore their psychometric performance in a pilot study.

Methods

The DISABKIDS methodology

One of the most distinctive features of the DISABKIDS project was its emphasis on a simultaneous approach to international instrument development: in contrast to the traditional sequential approach (instruments are translated and adapted from one language/culture to another), the construction of DISABKIDS measures relied on a joint effort between experts from different countries, which enabled the inclusion of cultural specificities from the beginning

of the instrument development (Schmidt et al., 2006; Simeoni et al., 2007). However, upon completion of the original DISABKIDS project, a sequential approach was adopted in the development of new versions of the instruments, such as Portuguese. The European DISABKIDS Group prepared a formal document where specific translation and validation procedures for the development of new versions were outlined extensively. The steps described in this protocol were as follows: (1) translation of questionnaires; (2) semantic validation; (3) pilot study; (4) field study; and (5) norming (if possible). The translation methodology, for instance, was organized according to the latest guidelines in cross-cultural instrument adaptation (Schmidt & Bullinger, 2003) and included the following specific procedures: (1) production of two independent forward translations; (2) reconciliation of items into a single forward version; (3) back-translation; (4) review of forward- and back-translation; (5) assessment of conceptual equivalence/first harmonization of problematic items; (6) semantic validation (cognitive interviews); and (7) international harmonization. It is important to note that for each phase of instrument adaptation process, its aims, agents, methods, instruments and expected outcomes were defined clearly (The DISABKIDS Group, 2004). Moreover, (re)formulation of items during harmonization phases was conducted under the supervision of the European DISABKIDS Group coordination.

Participants

Following the sampling frames indicated by the European DISABKIDS Group (The DISABKIDS Group, 2004), for each condition (asthma and epilepsy) a group of children (aged 8–12 years) and a group of adolescents (aged 13–18 years) were recruited, including each of nine individuals and their parents, thus achieving a total sample of 72 subjects (children and parents only). Sample collection was carried out between December 2008 and March 2009 at the outpatient services of Allergology and Neurology of Coimbra University Hospitals and Coimbra Paediatric Hospital. Children and adolescents were included if they met the following inclusion criteria: (1) age between 8 and 18 years; (2) clinical diagnosis of asthma or epilepsy according to the international classification systems (ICD-10) by a physician; (3) illness duration of at least one year; and (4) a consent form from the patients and/or their parents. Individuals were excluded from this study according to these criteria: (1) children (less than 14 years old) who refused to participate, disregarding their parents' previous consent; (2) significant developmental delay, interfering with the ability to understand questions, assess thoughts and emotions and maintain an adequate conversation with an adult (as grossly assessed by their physician); (3) severe psychiatric comorbid disorder, currently receiving psychiatric/psychotherapeutic care (as

indicated by the doctors, according to the patient's clinical history); and (4) existence of asthma and epilepsy comorbidity (in order to assess instrument discriminant validity). Additionally to the DISABKIDS translation and validation protocol, a group of teachers was also included in our sample, enabling a complementary assessment of the quality and adequacy of the Portuguese versions of DISABKIDS-37. This was thought to be a valuable contribution to the semantic validation phase, because teachers have increased knowledge of the language abilities and vocabulary of children and adolescents and could easily suggest eventual item reformulations while ensuring the maintenance or improvement of their adequacy and comprehensibility. All the teachers integrating our sample had extensive experience in teaching children and adolescents with chronic health conditions, in both community and rehabilitation settings. For each group of academic levels (primary/elementary and secondary) nine teachers were included, thus completing a total of 18 teachers. This part of the sample was collected at Coimbra Cerebral Palsy Association between October and November 2008.

Procedure

After the presentation of a comprehensive description of the research project for the validation studies of the Portuguese versions of DISABKIDS-37, formal authorizations were obtained from the Ethics Committees of Coimbra University Hospitals and Coimbra Paediatric Hospital, as well as from the Direction Board of Coimbra Cerebral Palsy Association. Informed consent forms were signed by the adolescent and his/her parent when he/she was more than 14 years old, and only by parents when younger than 14 years. A specific consent form was also available for the teachers' group. Children and their parents were conveniently recruited by the research team at the moment of admission to medical consultations at the aforementioned services, according to a previous case selection (made by the responsible physicians) of those who met the criteria to participate in the study. After completing the DISABKIDS questionnaires, children and their parents were asked to rate the instrument in general ("general impression" phase) and a given set of items in particular ("cognitive debriefing" phase). During this process, teachers did not complete the questionnaires but were asked only to assess the instruments in general and the items in particular, through their perceptions of the children's cognitive and linguistic abilities at a given academic level. Given the exhaustion associated with the task required for the semantic validation cognitive debriefing, each subject was assigned to only assess a group of items of the 37 DISABKIDS questions, which were clustered into three subsets: subset A (items 1–12); subset B (13–25); and subset C (26–37). Throughout the sample collection, subjects were assigned sequentially to one of these clusters, as the example given:

subject 1 to subset A, subject 2 to subset B, subject 3 to subset C, subject 4 to subset A and so on. During the administration of the questionnaires, a research assistant was present in order to support children and parents when necessary and to prevent information exchange regarding self and proxy perceptions. For the same reason, children of four parents who opted to answer the questionnaires at home and return them later by mail were asked to complete the questionnaires at the hospital services.

Measures

The following instruments were administered to both the child and his/her parent/family caregiver, or the parents only. The teachers' group was administered only the last two of the listed instruments.

- DISABKIDS-37 (QoL questionnaire for children and adolescents with chronic conditions – DISABKIDS chronic generic measure) (The DISABKIDS Group, 2006) – the generic DISABKIDS module is designed to assess HRQoL in children and adolescents, aged between eight and 18 years, with any chronic condition. The questionnaire includes 37 items, which are similar in both self and proxy versions, relating to the child's global functioning and well-being during the last four weeks. A five-point Likert response scale is adopted in both versions of the instrument (1 = *never*; 2 = *seldom*; 3 = *quite often*; 4 = *very often*; 5 = *always*), although negative items (8–25 and 32–37) need to be recoded inversely. The 37 questions are grouped into six facets: independence; emotion; social inclusion; social exclusion; physical limitation; and treatment. The global raw score (minimum = 37, maximum = 215) represents the computation of these six facets, thus considering HRQoL as a second-order construct. For the purpose of this study, all raw scores were standardized within a percentage scale (minimum = 0, maximum = 100). These questionnaires also include specific fields regarding basic socio-demographic data of the children, such as age and gender.

- DISABKIDS severity assessment scales for asthma and epilepsy (The DISABKIDS Group, 2006) – the severity assessment scales for asthma and epilepsy symptoms originally integrated the specific HRQoL modules for these two conditions and were utilized here as a brief measure of symptom severity assessment. The proxy-report scales used in our study are adequate to be administered to parents and other family/informal caregivers of children and adolescents with chronic conditions, and their questions are related to how much trouble a child/adolescent has had with their asthma/epilepsy during the last year. Global raw scores for

each one of these symptom severity assessment scales range between a minimum of 3 and a maximum of 15. For the purpose of the present study, and given the relatively small size of our sample, we considered a unified global score between the two conditions and two severity categories: mild-moderate (global score between 3 and 8) and moderate–severe (global score between 9 and 15). In our sample, adequate levels of internal consistency were observed for these scales independently ($a = 0.77$ for asthma; $a = 0.81$ for epilepsy) and jointly ($a = 0.78$).

- DISABKIDS general impression sheet (DISABKIDS group document, s/d) – the general impression sheet aims at an overall assessment of the DISABKIDS questionnaires by children and adolescents with chronic conditions and their parents. The general impression sheet includes seven questions covering a variety of general features on questionnaires quality and applicability, such as: (1) global qualitative evaluation of the questionnaire; (2) item understandability; (3) straightforward use of the response scales in relation to the questions; (4) relevance of the questions for the child's condition; (5) willingness to change something in the questionnaire; (6) willingness to add something to the questionnaire; and (7) indication of items the child/parent might not want to answer.

- DISABKIDS cognitive debriefing sheet (DISABKIDS group document, s/d) – this cognitive debriefing sheet is the core instrument for the semantic validation process of DISABKIDS instruments. For each item of the DISABKIDS-37 questionnaires, the child/adolescent and their parents are asked to indicate whether that particular item (1) is relevant for the child/adolescent situation; (2) is difficult to understand; and whether (3) the response scales are simple and in agreement with the question posed.

Analysis

Data analyses were performed using the Statistical Package for Social Sciences (SPSS, version 15.0). Item frequencies were calculated to describe the pattern of responses obtained with the semantic validation sheets (general impression and cognitive debriefing) while considering a minimum frequency of three subjects for the cognitive debriefing in a given negative response category for an item to be assumed as problematic, and thus possibly requiring additional revision. Internal consistency was assessed with Cronbach's *a* and Guttman's split-half reliability coefficient. Pearson's correlation coefficients were computed in order to assess scale inter-correlations, and the non-parametric Mann–Whitney *U*-test was used to compare means between small independent groups. Parent–child concordance was assessed with intraclass

correlations for average measurements within a two-way mixed model (absolute agreement type). Most statistical analyses were conducted for a confidence interval of 95%, but given the exploratory intend of the pilot study, a confidence interval of 90% was accepted for some of the analyses performed.

Results

Sample characteristics

Children and adolescents of our sample were generally distributed homogeneously along the demographic and clinical categories assessed across both age groups (Table 1). Despite its classification as a convenience sample, homogeneity in children and adolescent age and gender variables across both chronic health conditions was also achieved: for the asthma group, 11 boys and 7 girls were assessed, with a mean age of 12.44 years [standard deviation (SD)=3.18]; for the epilepsy group, 9 boys and 9 girls were assessed, with a mean age of 12.44 years (SD=3.34). Also, most of the children and adolescents assessed had no comorbidity with other medical condition (94.4% children and 88.9% adolescents) or developmental delay (83.3% children and 100% adolescents).

Table 1. Demographic and clinical characteristics of children and adolescents

Characteristics		**Children (8-12 years)** (*n* = 18)	**Adolescents (13-18 years)** (*n* = 18)
Demographic			
Age (years)	(M/SD)	9,6 / 1,3	15,3 / 1,6
Gender	Male (*n*/%)	13 / 72,2	7 / 38,9
	Female (*n*/%)	5 / 27,8	11 / 61,1
Clinical	(*n*/%)		
Main diagnosis (clinician info)	*Asthma*	9 / 50,0	9 / 50,0
	Epilepsy	9 / 50,0	9 / 50,0
Medication	*Yes*	16 / 88,9	18 / 100
Severity (parent rating)	*Mild-Moderate*	10 / 55,6	11 / 61,1
	Moderate-Severe	8 / 44,4	7 / 38,9
Comorbidity (parent info)	*Yes*	1 / 5,6	2 / 11,1
Developmental delay (clinician info)	*Yes*	3 / 16,7	0 / 0

Note. SD, Standard Deviation.

Semantic validation – general impression phase

Children and adolescents with chronic health conditions, their parents and teachers reported an overall positive impression on the Portuguese versions of DISABKIDS-37. All parents and teachers and almost all children/adolescents rated their general impression on the questionnaires as "good" or "very good". The large majority of the subjects from these three groups found no difficulty in understanding the items of the Portuguese DISABKIDS-37 or in using the response scales. Interestingly enough, while most parents and teachers considered the questions as "very relevant", children and adolescents tended to assess their relevance more partially, but mainly as "relevant" and "sometimes relevant". With the exception of the teachers' group, where nearly one-third of the subjects expressed their wish to change something in the questionnaire and 16.7% suggested the need for additional information to be asked, almost all subjects reported no need to add or change something in the questionnaires. In all three groups, 94.4% of subjects regarded the questions as adequate and non-intrusive. Results from the general impression phase of the semantic validation are described in greater detail in Table 2.

Table 2. Portuguese DISABKIDS General Impression ratings by children/adolescents, their parents and teachers.

Questions	Answer categories	*Frequency (%)* Children/Adolescents (*n* = 36)	*Frequency (%)* Parents (*n* = 36)	*Frequency (%)* Teachers (*n* = 18)
(1) What do you think about our questionnaire in general?	*Very good*	58,3	58,3	72,2
	Good	38,9	41,7	27,8
	Not good	2,8	-	-
(2) Are the questions understandable?	*Easy to understand*	80,6	94,4	77,8
	Sometimes difficult	19,4	5,6	22,2
	Not understandable	-	-	-
(3) What about the answer categories? Did you have any difficulties to use them?	*No difficulties*	80,6	91,7	88,9
	Some difficulties	19,4	8,3	11,1
	A lot of difficulties	-	-	-
(4) Are the questions relevant for the health condition/disease of you/your child?	*Very relevant*	61,1	80,6	83,3
	Sometimes relevant	36,1	16,7	16,7
	Not relevant at all	2,8	-	-
(5) Would you like to change something in the questionnaire?	*No*	97,2	94,4	72,2
	Yes	2,8	5,6	27,8
(6) Would you like to add something in the questionnaire?	*No*	94,4	91,7	83,3
	Yes	5,6	8,3	16,7
(7) Were there any questions you did not want to answer?	*No*	94,4	94,4	94,4
	Yes	5,6	5,6	5,6

Semantic validation – cognitive debriefing phase

Because cognitive debriefing was conducted for three different subsets clustering the 37 items of DISABKIDS questionnaires, we will approach the results obtained for each one, then separately. For subset A (items 1–12), three children/adolescents considered questions nine ("Is your life ruled by your condition?") and 12 ("Does your condition bother you when you play or do other things?") as not relevant for their condition, the same occurring within the parents' group for question nine. This same question was indicated by three teachers as eventually raising difficulties of understanding by younger children. None the less, items from this subset were largely evaluated as relevant and understandable by the three groups of subjects, and the

response scales were seen as appropriate for the questions asked. For subset B (items 13–25), all the groups mainly reported these questions as greatly relevant and found no difficulty in understanding them nor in using their respective answer scales. Finally, regarding subset C (items 26–37), items 33 ("Is it annoying for you to have to remember your medication?") and 34 ("Are you worried about your medication?") were rated by three children/adolescents as not relevant for their health condition, but this was not observed for the parents and the teachers' item relevance assessment. Children/adolescents, their parents and their teachers indicated no difficulty in understanding the questions from this subset nor in using the response scales provided.

Reliability

Internal consistency reliability alpha and Guttman's split-half coefficients for the self and proxy versions of Portuguese DISABKIDS-37 are presented in Table 3. Both versions of the instrument exceeded the reliability standard of 0.70, which has been recommended previously by Nunnally and Bernstein (1994, cited in Varni, Burwinkle, Rapoff, Kamps, & Olson, 2004).

Table 3. Internal consistency values for the Portuguese self-report version (child version) and the proxy version of DISABKIDS-37.

	α	*Split-half*	Number of cases	Number of items
Questions	0.92/0.92	0.90/0.91	34/36	37
Facets	0.84/0.78	-	34/34	6

Scale inter-correlations

Table 4 shows Pearson's coefficients obtained for the correlations between subscales and total score of the Portuguese child and proxy versions of DISABKIDS-37 questionnaires. Apart from the treatment facet, which displayed only moderate associations with the emotion and limitation facets, most of the other inter-facet correlations are moderate or strong. The emotion facet was the only one to present moderate to strong associations with all the other facets of the instrument in both self and proxy versions. With the exception of emotion (self- and proxy-reported) and social inclusion (self-reported) facets, which achieved very strong correlations, all

the remaining facets were found to be correlated strongly with the total score of DISABKIDS-37. Such correlations between facets, and between them and the total score, are suggestive of a higher-order QoL factor.

Table 4. Inter-correlations between total score and subscales for the child and proxy versions of Portuguese DISABKIDS-37

	Independence	**Emotion**	**Social Inclusion**	**Social Exclusion**	**Limitation**	**Treatment**
Emotion	0.34*/0.47**					
Social Inclusion	0.70**/0.71**	0.66**/0.41*				
Social Exclusion	0.34*/0.66**	0.69**/0.57**	0.67**/0.59**			
Limitation	0.28/0.36*	0.77**/0.68**	0.57**/0.35*	0.61**/0.39*		
Treatment	0.17/0.14	0.61**/0.57**	0.28/0.06	0.32/0.14	0.41*/0.32	
Total score	0.57**/0.67**	0.87**/0.87**	0.82**/0.65**	0.75**/0.69**	0.76**/0.70**	0.67**/0.61**

Note. *$p < .05$ **$p < .01$

Discrimination between clinical and socio-demographic groups

The Portuguese DISABKIDS-37 questionnaires distinguished between differences on the impairment of self and proxy-reported HRQoL of children and adolescents with chronic health conditions (Table 5). In terms of symptom severity, the DISABKIDS-37 proxy version distinguished HRQoL outcomes in the independence facet between parent-reported severity levels (Mann–Whitney *U*-test = 97.5, $n_1 = 21$, $n_2 = 15$, $p = 0.05$). The DISABKIDS child version also differentiated between age groups, with adolescents reporting a better HRQoL in the independence (Mann–Whitney *U*-test=79, $n_1 = n_2 = 18$, $p = 0.01$) and the social inclusion (Mann–

Whitney *U*-test = 99, $n_1 = n_2 = 18$, $p = 0.05$) facets than children. Finally, DISABKIDS-37 self and proxy versions did not discriminate between gender categories.

Table 5. Differences in self and proxy-reported HRQoL between asthma and epilepsy groups.

	Asthma (*n* = 18) *M* (*SD*) Self/Proxy	**Epilepsy** (*n* = 18) *M* (*SD*) Self/Proxy	***U*** Self/Proxy	***p*** Self/Proxy
Independence	78.9 (18.1) / 81.4 (16.0)	80.1 (20.1) / 77.1 (17.3)	149.5/125.0	NS/NS
Emotion	76.4 (24.1) / 80.5 (16.8)	71.4 (19.3) / 73.1 (24.0)	135.0/130.5	NS/NS
Social inclusion	82.9 (17.6) / 81.6 (15.5)	72.5 (21.4) / 79.2 (16.4)	103.5/139.5	0.06/NS
Social exclusion	91.4 (13.1) / 90.4 (13.2)	82.1 (18.4) / 82.4 (13.6)	113.5/88.5	NS/0.02
Limitation	68.3 (18.3) / 65.7 (14.7)	73.8 (20.3) / 80.1 (16.2)	136.0/104.5	NS/0.07
Treatment	68.1 (28.8) / 77.2 (19.7)	65.1 (27.1) / 70.3 (27.2)	131.0/131.0	NS/NS
Total score	79.5 (15.7) / 79.5 (12.1)	74.1 (15.3) / 76.8 (13.4)	134.5/130.0	NS/NS

Notes: NS, Not significant; SD, Standard deviation.

Parent-child concordance

Intraclass correlation coefficients (ICC) that were computed in order to assess parent–child concordance (i.e. inter-rater agreement) are listed in Table 6. These coefficients indicate a moderate convergence between child/adolescent and parent reports for the facets of emotion, social inclusion, social exclusion and treatment, and a non-convergence of reports for the remaining facets and total score.

Table 6. Intraclass correlation coefficients (ICC) for total score and sub-scales of the self-report version (child version) and the proxy version of Portuguese DISABKIDS-37 questionnaires

Domain	Facet	ICC	*p*
Mental	Independence	0.210	NS
	Emotion	0.476	0.05
Social	Social inclusion	0.411	0.06
	Social exclusion	0.413	0.06
Physical	Limitation	0.353	NS
	Treatment	0.402	0.07
Total score		0.250	NS

Note. NS, Not significant.

Discussion

The objective of this study was to validate semantically the Portuguese versions of DISABKIDS-37, ensuring their adequacy for paediatric HRQoL assessment in Portugal within a cross-cultural perspective. Additionally, some preliminary results on basic psychometric properties of those versions were obtained with exploratory pilot analyses. The main finding of our study was the evidence on the quality, relevance and adequacy of the Portuguese versions of DISABKIDS-37 for both children and adolescents with chronic health conditions and their parents. The preliminary psychometric data gathered additionally may also be suggestive of interesting outcomes to be verified in future research, using larger samples to assess accurately the reliability and the validity of the Portuguese versions of these questionnaires.

Similar to the results obtained from the semantic validation studies of DISABKIDS-37, which were carried out in Mexico and Brazil (Fegadolli et al., 2010; Medina-Castro, 2006), the Portuguese versions of the instruments were generally rated as important, understandable and adequate by children and adolescents with chronic conditions and their parents. Although children and adolescents tended to slightly judge the DISABKIDS-37 questions as less relevant than their parents and teachers, the overall impression was very positive and highlights the instrument appropriateness to assess paediatric HRQoL in Portuguese samples. The Portuguese semantic validation study of DISABKIDS questionnaires was the first to include a group of teachers with experience in working with children and adolescents with chronic conditions. This

inclusion represented a significant contribution, as their reports identified at least one critical item which might need additional revision, so that the Portuguese questionnaires may achieve the intended level of immediate item understanding by children and youths. This semantic validation study is an example of best practices for the future development of new versions of paediatric HRQoL measures which have been constructed originally in a different country or culture.

In terms of the preliminary psychometric properties assessed, results need to be taken cautiously, given the exploratory nature of the pilot study performed. Nevertheless, internal consistency and facet inter-correlation coefficients were within the desirable psychometric range and in notable agreement with the results observed in the original DISABKIDS and the Mexican and Brazilian studies. This agreement may be viewed as a significant outcome drawn from the accuracy devoted to the previous phases of translation of the DISABKIDS-37 adaptation process. Regarding the comparisons that were conducted between clinical and socio-demographic subgroups in our sample, the results from our pilot study are limited in terms of statistical significance, but interesting enough at an exploratory level of analysis. Self- and proxy-reported HRQoL for children and adolescents with epilepsy was more impaired in facets belonging to the social domain, while proxy-reported limitation (one of the facets of the physical domain) was greater for the asthma group. It is curious to note that, despite the different scope of these studies, our preliminary results tend to coincide with those that were previously observed in a comparative study including children with asthma and epilepsy (Austin et al., 1994). The Portuguese versions of DISABKIDS-37 also discriminated HRQoL outcomes between chronically ill children and adolescents, with the latter group reporting higher scores in the facets of independence and social inclusion than the former. Although we have not controlled the analyses for illness duration, it is tempting to hypothesize its resilient contribution on health-related coping development and individual adaptation to chronic conditions during childhood and adolescence, as has been suggested for asthma (Erickson et al., 2002), for instance. Finally, for parent–child concordance in paediatric HRQoL assessment, agreement was generally weak. This low concordance between informants, which has not been observed to this degree in previous DISABKIDS-37 validation studies, may represent an actual divergence between self and proxy HRQoL assessment, or be due simply to the reduced clinical sample on which we based our analyses. Hopefully, the next steps for the validation process of DISABKIDS questionnaires in Portugal will enable us to clarify this topic.

Although we have stressed the pilot nature of this study and the exploratory level of the statistical analyses performed, we acknowledge the small sample size as its major limitation.

Moreover, our assessment protocol included a minimum of clinical and socio-demographic variables which served the main purpose of the study, but prevented us from additional analyses and interpretations of results. Optimally, in the context of semantic validation, we could also have tested the response scales by using virtual analogue scales in order to assess the conceptual equivalence of response scales, as suggested by Schmidt and Bullinger (2003).

Despite these limitations, this study represents an important contribution to paediatric HRQoL assessment in Portugal, as well as an application of an instrument validation framework based on a cross-cultural perspective. By making the DISABKIDS-37 questionnaires available in Portugal, we are proposing an innovative instrument which may serve as a basis for important decision-making at clinical, institutional and political levels in this country, and broadening the possibilities for the conduction of future cross-cultural HRQoL studies with paediatric populations. Following the DISABKIDS instrument adaptation protocol, we are currently collecting a larger sample of children and adolescents with chronic conditions (field study), with the administration of a more comprehensive instrument tool set, which will enable us to assess with greater accuracy the psychometric properties of the Portuguese versions of DISABKIDS-37 questionnaires. In addition to the conduction of cross-cultural HRQoL studies, future directions in paediatric HRQoL research should include the simultaneous assessment of the child/adolescent and his/her parents/caregivers' health outcomes, while placing greater emphasis on positive dimensions of individual and family adaptation to childhood chronic health conditions.

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