



Family adaptation to cerebral palsy in adolescents: A European multicenter study



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ABSTRACT

Background and aim: Factors promoting family adaptation to child's disability are poorly studied together. The aim of the study was to describe the family adaptation to disability and to identify determinants associated with using a global theoretical model.

Materials and methods: 286 families of teenagers [13–17 years] with cerebral palsy (CP) from 4 European disability registers were included and visited at home. Face to face interviews were performed in order to measure parental distress, perceived impact in various dimensions of family life, family resources and stressors. Relationships were modelled with structural equations.

Results: 31.8% of parents living with an adolescent with CP showed clinically significant high stress requiring professional assistance. The main stressors were the level of motor impairment and behavioural disorders in adolescent. A good family functioning was the best protective factor. Respite in care and a parents' positive attitude were significantly related to less parental distress. Material support, socioeconomic level, marital status or parental qualifications did not appear to be significant protector factors.

Conclusions: Particular attention must be paid not only on physical condition but also on adolescent psychological problems to improve family adaptation. Families at risk of experiencing severe distress should be targeted early and proactive caregiver interventions on the whole family should be performed.

What this paper adds: Family is a dynamic system: facing disability, it tries to recover its balance with available resources and its perception of the situation. Literature highlights potential stressors and protecting factors that could affect the disabled child's family adaptation but few papers study a global model including most of these factors. This study validated a global theoretical model of family adaptation to disability at adolescence. It identified behaviour disorders and motor impairment level as main stressors, family functioning as the largest protecting factors, and equipment and financial support as non significant protective factors.

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Abbreviations: CFI, Comparative Fit Index; CP, Cerebral palsy; FICD+4, Revised Family Impact of Childhood Disability; PSI, Parenting Stress Index; RMSEA, Root Mean Square Error of Approximation; SD, Standard Deviation; SEM, Structural Equation Modelling; TLI, Tucker-Lewis Fit Index; WLSMV, Weight least-squares means and variance adjusted.

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Introduction

The birth of a disabled child has a long-term family impact, which has already been described (Guyard, Fauconnier, Mermet, & Cans, 2010; Rentinck, Ketelaar, Jongmans, & Gorter, 2007; Skinner & Weisner, 2007; Vermaes, Gerris, & Janssens, 2007). Seven dimensions of family life that may be influenced have been identified: constraints on time (Curran, Sharples, White, & Knapp, 2001; Saigal, Burrows, Stoskopf, Rosenbaum, & Streiner, 2000; Sen & Yurtsever, 2007), employment (Brehaut et al., 2004; Porterfield, 2002; Wallander & Venters, 1995), financial consequences (Brehaut et al., 2004; Curran et al., 2001; McCubbin, 1989), positive modifications of social relationships (Brehaut et al., 2004; Florian & Findler, 2001; Saigal et al., 2000) or negative modifications of social relationships (Green, 2003; Mugno, Ruta, D'Arrigo, & Mazzone, 2007; Sen & Yurtsever, 2007) or intrafamilial relationships (Florian & Findler, 2001; Joesch & Smith, 1997; Pirila et al., 2005; Weisner, Matheson, Coots, & Bernheimer, 2004), physical impact on caregivers (Brehaut et al., 2004; Eker & Tuzun, 2004; Lach et al., 2009) and caregiver's psychological wellbeing, with more depressive symptoms and anxiousness in parents of children with disabilities (Brehaut et al., 2004; Manuel, Naughton, Balkrishnan, Paterson Smith, & Koman, 2003; Wallander & Venters, 1995) than in other parents.

Some families adjust better than others, but factors promoting this adaptation are poorly known and seldom studied together. In this complex situation, some outcomes can explain others and may be mediated by external factors (Guyard et al., 2010; Rentinck et al., 2007; Skinner & Weisner, 2007; Summers et al., 2005). Family stress theories provide a dynamic view in which stress is explained as a process of adaptation (Lazarus & Folkman, 1984). The ABCX model of adaption from McCubbin (H. McCubbin & Patterson, 1983) offers an easy-to-understand concept of this process of adaptation: families dealing with a stressful situation experience a pile-up of stressors (A) across time. Two mediating blocks of factors influence the level of family adaptation (X): the resources available and used by the family (B) and the family's perception of the situation (C). Both of these mediators are influenced by primary stressors (A) and contribute to adaptation (X). Adaptive resources (B) can be internal or external to the family system. The perceived impact (C) refers to the family's appraisal of how consistently resources (B) match needs (A).

Here we suggest a model of family adaptation, establishing our hypotheses (Fig. 1), using factors identified in literature and based on the ABCX model.

The present study aimed to describe the family adaptation to disability based on a conceptual model related to the ABCX model in order to identify determinants associated with the level of family adaptation in families living with an adolescent with cerebral palsy (CP).

2. Materials and methods

2.1. Participants and procedure

The population targeted was families of adolescents with CP, ranging from 13 to 17 years of age at the time of the interview, living in the counties of Cork (Ireland), Haute-Garonne and Isere (France), and in Eastern Denmark. Families were recruited from a population-based registry. This research is a part of the Study of Participation of Children with Cerebral Palsy Living in Europe (SPARCLE) including nine areas in Europe (further details on the sample procedure are available elsewhere (Colver, 2006)). This sample contained 286 families, visited at home between January 2009 and April 2010.

We obtained ethical approval, or a statement that only registration was required, as appropriate to each country. We obtained signed consent from all parents and from young people in the cases where it was possible

2.2. Measures

All the questionnaires used in SPARCLE were validated. Questionnaires used for studying the family adaptation to disability are described in Table 1.

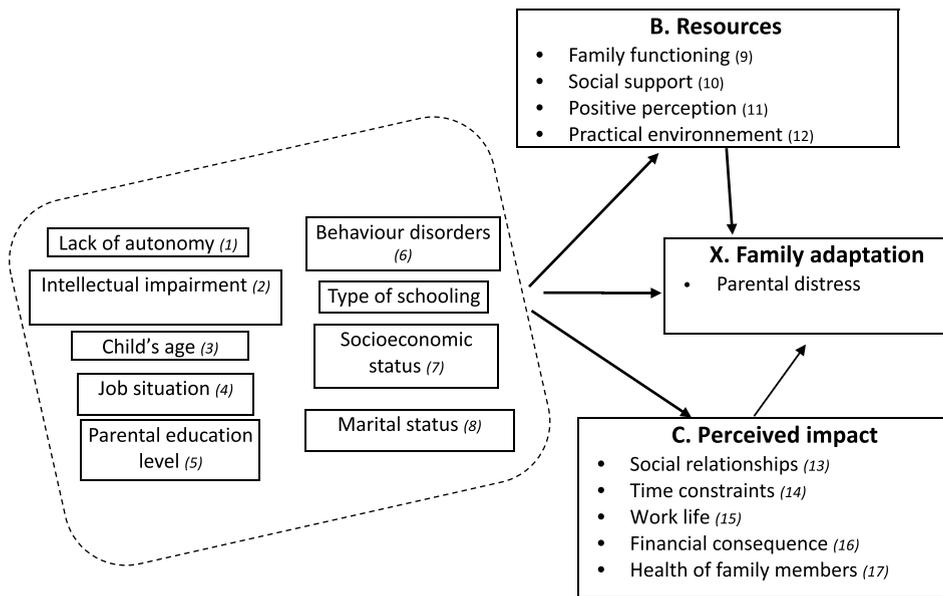
2.3. Statistical method

The data were analyzed using the structural equation method. The main outcome was the score of parental distress from the subscale of the Parenting Stress Index (PSI) (Abidin, 1995). Three measurement models (Fig. 2) were defined to assess latent concepts.

- Among stressors (A), the level of impairment was described through two latent variables: motor impairment and associated impairments. The first had three reflective indicators: walking ability, bimanual skills and difficulty feeding. The second impairment latent variable grouped three other items: cognitive impairment, communication disorders and epileptic seizures.
- Among resources (B), the positive attitude of parents coping with the disability was assessed through five items from the Family Impact of Childhood Disability – revised (FICD+4) identified previously (Guyard et al., 2012).
- The negatively perceived impact (C) was described through five latent constructs expressing the negative perception on social relationships, constraints on time, work life, financial consequences, and healthcare costs for family members. Ten

Table 1
Questionnaires and measures.

Questionnaires	Scale and description	Nber items	Range score	High score signification	Reference
Stressors (A)					
Walking ability	Gross Motor Function Classification System (GMFCS): <i>From walking without limitation to not walking</i>	5	[1;5]	Severe impairment	(Palisano et al., 1997)
Manual skills	Bimanual Fine Motor Function Classification (BFMF): <i>From at least one hand manipulation without limitation to both hands can only hold</i>	5	[1;5]	Severe impairment	(Beckung & Hagberg, 2002)
Feeding difficulties	<i>From feeding by mouth with no problem to feeding by a tube</i>	5	[1;5]	Severe impairment	
Communication difficulties	<i>From normal communication to no formal communication</i>	3	[1;3]	Severe impairment	
Cognitive impairment	<i>The child's understanding, learning and friendships. From none to severe</i>	4	[1;4]	Severe impairment	
Severity of epileptic seizure	<i>Existing seizures, medication and frequency during last year</i>	6	[1;6]	More than 1 seizure by week	
Behavioral disorders	Strengths and Difficulties Questionnaire (SDQ) <i>Total score based on emotional disorders, behavioral disorders, hyperactivity, peers difficulties.</i>	25	[0;40]	Abnormal behaviour	(Goodman, 1997)
Schooling	<i>Schooling attainment from normal school, to specialized or no school</i>	10	[1;3]	No school	
Parental work	<i>For each parent, full time work, partial or not working</i>	7	[1;3]	Both parents are not working	
Parental qualification	<i>Median qualification in parents from thesis to primary level of education</i>	6	[1;3]	Low education level	
Marital status	<i>Living with partner versus single (living alone or with parents)</i>	2	[1;2]	Single	
Socioeconomical status	Family Affluence Scale (FAS) <i>Number of cars and computers at home, number of holidays of the whole family together and having a single room for the child.</i>	4	[0;9]	High level of wealth	(Boyce, Torsheim, Currie, & Zambon, 2006)
Resources (B)					
Family functioning	Family Assessment Device <i>Agreement scale about cohesion, communication and family relationships</i>	12	[12;48]	High dysfunctioning	(Byles & Byrne, 1998)
Respite in care	European Child Environment Questionnaire (3 items: social services; child look after elsewhere for few days; assistance at home) <i>Number of kind of used formal or informal respite with child's care: from "missing" to "having", 0 meant "not needed"</i>	3	[-3;3]	High need and high access	
House adaptation	European Child Environment Questionnaire (4 items: enlargement at home; adapted toilets; modified kitchen; hoists) <i>Number of specific equipment at home: from missing to having, 0 meant "not needed"</i>	4	[-4;4]		
Financial support	European Child Environment Questionnaire (2 items: grants for special equipments; for home modifications) <i>Number of financial supports: from "missing" to "having", 0 meant "not needed"</i>	2	[-1;1]		
Positive attitude	Revised Family Impact of Childhood Disability: FICD+4 (5 items: the uniqueness of this child; special pleasures with him/her; value in life; life more meaningful and parental personal growth) <i>Agreement scale on possible consequences resulting from child disability: from "not at all" to "to a substantial degree"</i>	5	calculated latent variable [-1;1]	High positive attitude	(Guyard et al., 2012; Trute, Hiebert-Murphy, & Levine, 2007)
Negative perceptions(C)					
Finance	Revised Family Impact of Childhood Disability: FICD+4 (3 items: postponement purchases; financial cost; stress occurred) <i>Agreement scale on possible consequences resulting from child disability: from "not at all" to "to a substantial degree"</i>	3	calculated latent variable [-1;1]	High negative attitude	(Guyard et al., 2012; Trute et al., 2007)
Social	Revised Family Impact of Childhood Disability: FICD+4 (2 items: social contact; time with friends) <i>Agreement scale on possible consequences resulting from child disability: from "not at all" to "to a substantial degree"</i>	2	calculated latent variable [-1;1]	High negative attitude	(Guyard et al., 2012; Trute et al., 2007)
Time	Revised Family Impact of Childhood Disability: FICD+4 (2 items: time demands; disruption in habits) <i>Agreement scale on possible consequences resulting from child disability: from "not at all" to "to a substantial degree"</i>	2	calculated latent variable [-1;1]	High negative attitude	(Guyard et al., 2012; Trute et al., 2007)
Work	Revised Family Impact of Childhood Disability: FICD+4 (2 items: professional situation; professional ambition) <i>Agreement scale on possible consequences resulting from child disability: from "not at all" to "to a substantial degree"</i>	2	calculated latent variable [-1;1]	High negative attitude	(Guyard et al., 2012; Trute et al., 2007)
Health	Revised Family Impact of Childhood Disability: FICD+4 (1 item: health of family members) <i>Agreement scale on possible consequences resulting from child disability: from "not at all" to "to a substantial degree"</i>	1	calculated latent variable [-1;1]	High negative attitude	(Guyard et al., 2012; Trute et al., 2007)
Family adaptation (X)					
Parental distress	Parental Distress (PSI-PD) which is a subscale of the Parenting Stress Index (PSI-SF) (12 items): <i>from "strongly agree" to "strongly disagree"</i>	12	12–48	High parental stress	(Abidin, 1995; Reitman, Currier, & Stickle, 2002; Sidor, Kunz, Schweyer, Eickhorst, & Cierpka, 2011)



A : Main stressors were supposed to interact in each other. Marital status and qualification could explain the socioeconomic level and employment situation (Guyard et al., 2013). The disabled child's age influenced the parental difficulties to face disability. Schooling is supposed to depend on the child's age and its disability level (Skinner & Weisner, 2007).

B : Stressors influence resources factors: family functioning was explained by the child's behavioural disorders (Herring et al., 2006) and child's health status (Joesch & Smith, 1997). Socioeconomic status and family composition influenced the level of family functioning (King et al., 1999). Social and practical supports were assumed to be determined by the level of impairment.

C : The parental appraisal of the stressors consequences is multidimensional. Negative appraisal could exist in various dimensions of family life: social, time, work, finances, and physical health, due to stressors (A) and limited by resources (B). Social relationships were assumed to be explained by the child's autonomy (Mugno et al., 2007; Ryan et al., 2009; Thyen et al., 2003).

REFERENCES

- (1) (Ketelaar et al., 2008; Laurvick et al., 2006; Lin, 2000; Ong et al., 1998; Raina et al., 2005; Ryan et al., 2009; Skinner & Weisner, 2007; Thyen et al., 2003; Vermaes et al., 2005)
- (2) (Emerson, 2003; Laurvick et al., 2006; Lin, 2000; Majnemer et al., 2012; Raina et al., 2005; Thyen et al., 2003)
- (3) (Skinner & Weisner, 2007)
- (4) (Guyard et al., 2013; Porterfield, 2002; Thyen, Kuhlthau, & Perrin, 1999)
- (5) (Skinner & Weisner, 2007)
- (6) (Blacher & Baker, 2007; Indredavik et al., 2005; Ketelaar et al., 2008; King et al., 1999; Lach et al., 2009)
- (7) (Emerson & Hatton, 2005; Emerson, Hatton, Llewellyn, Blacher, & Graham, 2006)
- (8) (Dziak, Janzen, & Muhajarine, 2010; Florian & Findler, 2001; Joesch & Smith, 1997; M. A. McCubbin, 1989)
- (9) (Lach et al., 2009; Lin, 2000; Raina et al., 2005; Skinner & Weisner, 2007; Summers et al., 2005)
- (10) (Eker & Tuzun, 2004; Horton & J.L., 2001; King et al., 1999; Lin, 2000; Skok et al., 2006; Wallander & Venters, 1995; Wanamaker & Glenwick, 1998; Wiegner & Donders, 2000)
- (11) (Eker & Tuzun, 2004; Gupta & Singhal, 2004; Noojin & Wallander, 1997; Trute, Benzies, Worthington, Reddon, & Moore, 2010; Wanamaker & Glenwick, 1998)
- (12) (Rigby, Reid, Schoger, & Ryan, 2001; van der Burg, Jongerius, van Limbeek, van Hulst, & Rotteveel, 2006)
- (13) (Eker & Tuzun, 2004; Horton & J.L., 2001; King et al., 1999; Lin, 2000; Skok et al., 2006; Wallander & Venters, 1995; Wanamaker & Glenwick, 1998; Wiegner & Donders, 2000)
- (14) (Curran et al., 2001; Ebersold, 2005; King et al., 1999; Ong et al., 1998; Raina et al., 2005; Saigal et al., 2000; Sen & Yurtsever, 2007; Skinner & Weisner, 2007)
- (15) (Ebersold, 2005; Porterfield, 2002; Summers et al., 2005)
- (16) (Curran et al., 2001; King et al., 1999; M. A. McCubbin, 1989; Sen & Yurtsever, 2007; Thyen et al., 2003; Wang et al., 2008)
- (17) (Brehaut et al., 2004; Raina et al., 2005; Tong et al., 2003)

Fig. 1. Model of family adaptation using factors identified in literature and based on the ABCX model (Dziak, Janzen, & Muhajarine, 2010; Ebersold, 2005; Emerson & Hatton, 2005; Emerson, Hatton, Llewellyn, Blacher, & Graham, 2006; Guyard et al., 2013; Herring et al., 2006; Horton & Wallander, 2001; Rigby, Reid, Schoger, & Ryan, 2001; Thyen, Kuhlthau, & Perrin, 1999; Tong et al., 2003; Trute, Benzies, Worthington, Reddon, & Moore, 2010; van der Burg, Jongerius, van Limbeek, van Hulst, & Rotteveel, 2006; Wang et al., 2008; Wiegner & Donders, 2000).

FICD+4 items resulting from an exploratory factorial analysis (Guyard et al., 2012) were used to estimate these latent variables. The validity of these three measurement models was checked using fit indices.

The correlations assumed in the conceptual model between observed and latent variables were tested knowing covariance of the whole data structure. An iterative procedure was used to simultaneously estimate weight in the three measurement models and regression relations. These parameters were free even if the model was identified. The weighted least-squares means and variance adjusted (WLSMV) estimation method was used because of the ordinal data and the sample size (Flora & Curran, 2004; Muthén, du Toit, & Spisic, 1997). To build the best fitted structural model, we proceeded step by step. The description of the methods for intermediate steps and the main results are presented in Appendix. Only significant

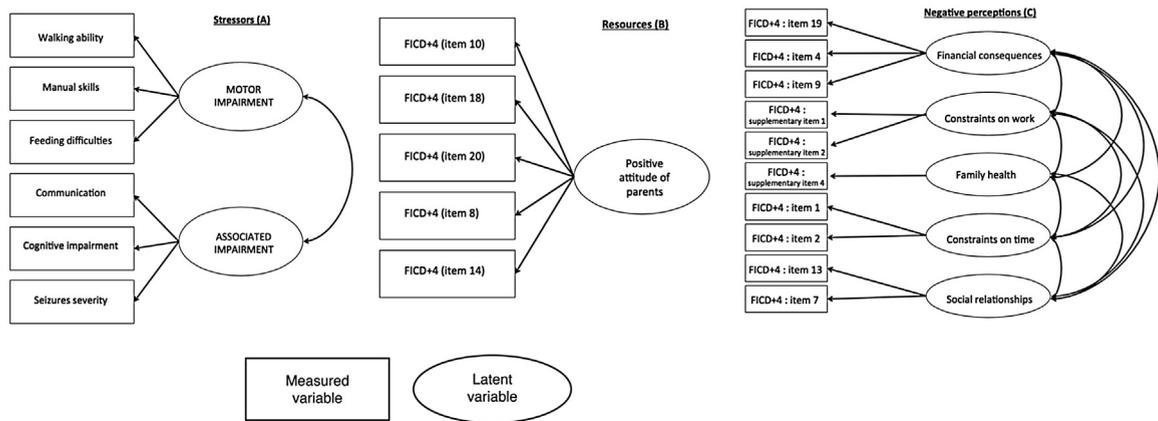


Fig. 2. The three measurement models used to calculate latent variables.

paths ($p < 0.05$) were retained. After validating the final model, the total effects of factors (direct plus indirect via mediating relationships) were calculated from standardized regression coefficients. Structural analysis was restricted to the population with no missing data. Differences between the two populations, retained versus excluded samples, were tested using the chi-squared test and the Fisher test, with significant difference defined as $p < 0.05$.

3. Theory

In the conceptual model related to the ABCX model, the psychological wellbeing of parents was the main outcome, assuming their psychological state expresses part of the family adaptation: the more parental distress decreases, the more the wellbeing of other family members increases, and vice-versa. McCubbin explained family adaptation as a continuum from “maladaptation” to optimal “bonadaptation.” The latter is defined as “meeting the needs of the individual family member to enable them to achieve their maximum potential and also the functioning of the family system and its transactions with the community (work place, school, health care system)” (McCubbin, 1993).

Parental distress is demonstrated as an outcome of the child’s intellectual impairment (Emerson, 2003; Majnemer, Shevell, Law, Poulin, & Rosenbaum, 2012). No consensus is found on the effect of the level of motor impairment (Ketelaar, Volman, Gorter, & Vermeer, 2008; Manuel et al., 2003; Ong, Afifah, Sofiah, & Lye, 1998; Raina et al., 2005; Skok, Harvey, & Reddihough, 2006). The main stressor on parental psychological wellbeing is the child’s behavioural disorders (Blacher & Baker, 2007; Indredavik, Vik, Heyerdahl, Romundstad, & Brubakk, 2005; Ketelaar et al., 2008; King, King, Rosenbaum, & Goffin, 1999; Lach et al., 2009). The parents’ ability to cope with difficult situations could protect from stressors (Eker and Tuzun, 2004; Noojin & Wallander, 1997; Wanamaker and Glenwick, 1998); this ability is explained by the family’s sociocultural characteristics (Skinner & Weisner, 2007). Social and family support goes hand in hand with good protective function (Eker & Tuzun, 2004; King et al., 1999; Skok et al., 2006; Wallander & Venters, 1995; Wanamaker & Glenwick, 1998).

4. Results

4.1. Participants

From the targeted population, 286 families were enrolled corresponding to a 53.4% response rate. A total of 220 observations (40.2% of the targeted population) were usable for the Structural Equation Modelling (SEM) analysis with no missing data. Fig. 3 describes the exclusion process for missing data and Table 2 presents characteristics of respondents and partial respondents. Even it were face-to-face interviews; some items were blank because few parents did not succeed in deciding any answer. As the whole interview took long time, we did not constraint parents in filling questionnaires.

The mean of the total PSI score was 77.80 (SD 21.92). Using a clinical cutoff of score over 90, as suggested by Abidin (1995), 31.8% of parents experienced high stress. The mean of the parental distress PSI score was 26.38 (SD 8.85).

4.2. Measurement models

Three measurement models were built. All parameters were left free except for three latent variables described below.

The impairment measurement model composed of two intercorrelated latent variables – severity of motor impairment and severity of associated impairments – was acceptable with Root Mean Square Error of Approximation (RMSEA) = 0.075, ($p(\text{RMSEA} < 5\%) = 0.163$), Comparative Fit Index (CFI) = 0.996 and Tucker-Lewis Fit Index (TLI) = 0.992. Walking ability, bimanual skills and feeding ability were largely explained by the motor latent variable (R-squared was respectively 0.749, 0.780 and 0.961). Communication and cognitive ability were strongly explained by the ‘Associated impairment’ latent variable, with,

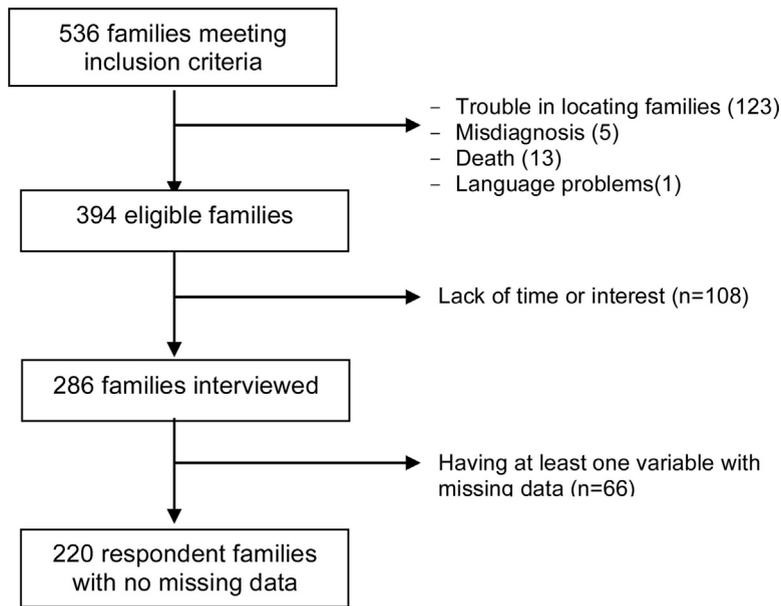


Fig. 3. Inclusion flow chart.

Table 2

Characteristics of respondents and partial respondents.

	Respondents (n = 220)		Excluded because missing value (n = 67)		
	Frequency	%	Frequency	%	p
Mother	198	90.0	62	92.5	NS
Living alone	26	11.8	13	20.0	NS
3 children and more	87	39.6	26	38.8	NS
Before university (Median qualification of parents)	174	79.1	50	79.4	NS
Parents work full-time	190	86.4	42	76.4	*
High family wealth (FAS \geq 6)	119	54.6	30	46.9	NS
Children:					
Male	122	55.5	41	61.2	NS
Mean age (SD)	15.16 (1.58)		15.20 (1.58)		NS
GMFCS IV-V	71	32.3	19	28.4	NS
BFMF IV-V	46	20.9	10	15.0	NS
Unable to communicate with speech	49	22.3	13	20.0	NS
Severe cognitive impairment	57	25.9	10	15.2	NS
Active epileptic seizures ¹	36	16.4	7	10.8	NS
Special or no school	106	58.2	30	47.6	NS
Mean SDQ score (SD)	11.59 (5.57)				NS

* $p < 0.001$.¹ At least 1 seizure during the last year.

respectively, R-squared reaching 0.993 and 0.726. The number of epileptic seizures was weakly described with R-squared at 0.475.

The parental positive attitude measurement model was acceptable with (RMSEA) = 0.041 ($p(\text{RMSEA} < 5\%) = 0.505$), CFI = 0.998 and TLI = 0.996. This measurement model was composed of five positive items from the FICD+4, explained by the positive attitude latent variable from 0.459 to 0.775 on their total variance.

The single item about health of the family's members expressed partially the latent variable, negative perceived impact on health. Thus the parameter linking this item and negative perceived impact on health was set at 95% of the item variance. An equality constraint of parameters was necessary on negative perceived impact on time and on negative perceived impact on work, unless the measurement model was not identified. The constrained model on overall negative perceived impact showed an acceptable but weak fit with RMSEA = 0.091, ($p(\text{RMSEA} < 5\%) = 0.003$), CFI = 0.983 and TLI = 0.972.

Finally, the global measurement model composed of the three measurement models showed a good fit according to the RMSEA criterion (RMSEA = 0.054, ($p(\text{RMSEA} < 5\%) = 0.274$), CFI = 0.973 and TLI = 0.973).

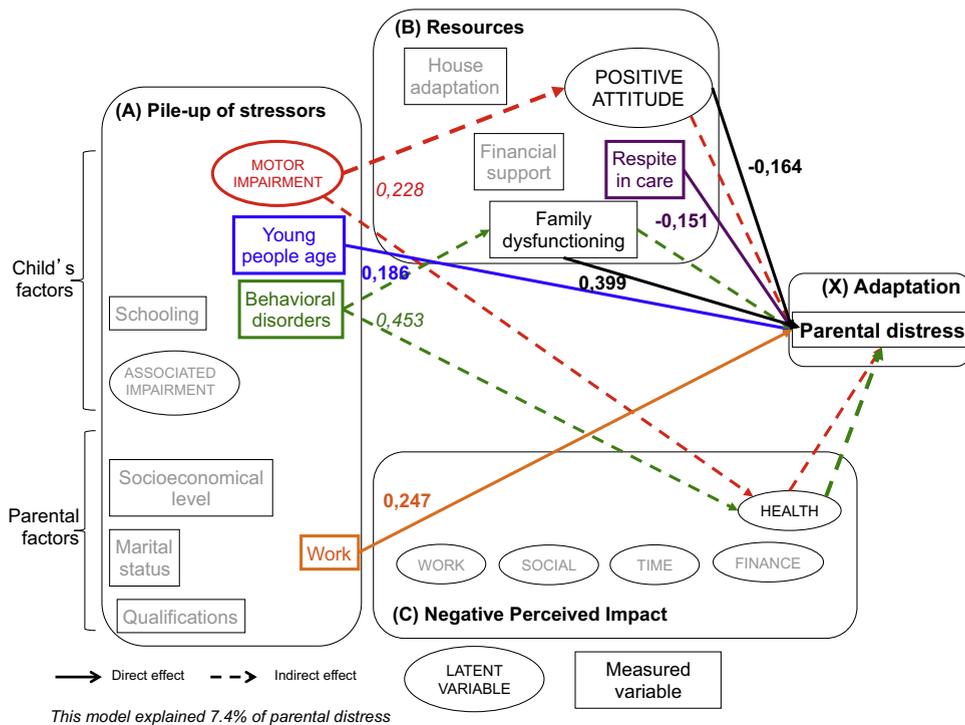


Fig. 4. Structural model of family adaptation to living with an adolescent with CP. Direct and indirect effects.

Table 3
Direct and indirect effects of factors explaining the level of parental distress.

		Negative perceived impact on					Parental Distress
		finance	work	health	time	social	
(A)	Motor impairment	0.067*	0.499	0.644	0.662	0.644	0.228
	Associated impairment	0.367					
	Behavioural disorders	0.263	0.261	0.428	0.283	0.236	0.453
	Young people age						0.186
	Parental unemployment						0.247
	Parental qualifications		0.196				-0.092*
(B)	Respite in care						-0.151
	General dysfunctioning scale	0.402			0.433	0.361	0.399
	Parental positive attitude	0.180*					-0.164
(C)	Negative perceived impact on health of the family members						0.450

This result means that an increase of one standard deviation of negative perceived impact on health score led to an increase of 0.450 unit of parental distress score.

* p < 0.05 (otherwise p < 0.01); *italic font*: indirect effect.

4.3. Structural model of family adaptation to living with an adolescent with CP

The final model is represented in Fig. 4 and the direct and indirect effects of factors explaining the level of parental distress are presented in Table 3. It validated existing relationships from the conceptual model showing acceptable fit with RMSEA = 0.044 (p(RMSEA < 5%) = 0.889), CFI = 0.966 and TLI = 0.961. The stressors factors without any effect on parental distress were the associated impairments, including intellectual impairment, the socioeconomical level, the marital status, the parental qualifications and the type of schooling. The resources showing no effect were the house adaptation and the financial support. And finally among the negative perceived impact, only health showed a significant relationship with the parental distress.

5. Discussion

Many parents having a child with a disability show a high level of stress (Parkes, Caravale, Marcelli, Franco, & Colver, 2011). Here about a third of parents living with an adolescent with CP showed clinically significant high stress requiring professional assistance, according to the cut-off score of 90 (Abidin, 1995) on the PSI total score. Based on a theoretical model

of adaptation, we identified factors aggravating parental distress and others protecting against it. Even though these results are close to previous from literature, we must notice the readers that this study is limited by the population size relatively to the number of estimated parameters.

5.1. Behavioural disorders are more disabling than motor impairment

Here, taking care of an adolescent showing abnormal behaviour, such as hyperactivity or behavioural disorders, had the worst aggravating effect on parental distress among the disability stressors. Thus autism spectrum disorders are conditions that could increase the risk of distress in parents. These behavioural disorders also significantly increased the risk of a negative parental perception of the situation. In the literature, the reverse consequence was highlighted: growing up in a stressful family environment led to behavioural disorders in children (Bakoula, Kolaitis, Veltsista, Gika, & Chrousos, 2009; Sipal, Schuengel, Voorman, Van Eck, & Becher, 2009). We could not test for this hypothesis because the present study was cross-sectional. Nonetheless, we believe this vicious circle could be turned into a virtuous circle: treating behavioural disorders in the child could contribute to reducing parental stress, and thus behavioural disorders should be reduced as well.

The second key condition was the severity of motor impairment. This showed a significant effect on both parental distress and negative perceived impacts. In previous studies, the severity of motor impairment is not unanimously identified as an aggravating factor: some validate this hypothesis (Ketelaar et al., 2008; Ong et al., 1998), while others find no significant explanation (Manuel et al., 2003; Skok et al., 2006). In three studies (Laurvick et al., 2006; Raina et al., 2005; Thyen, Sperner, Morfeld, Meyer, & Ravens-Sieberer, 2003), motor impairment is not predictive of a negative impact, but the caregiving demands and the presence of a cognitive impairment are found to significantly decrease the parents' psychological and physical wellbeing. Other literature reviews support the assumption of a negative impact of the level of dependency on the family's adaptation (Skinner & Weisner, 2007; Vermaes, Janssens, Bosman, & Gerris, 2005). Concerning social relationships, Mugno et al. (2007) find no worsening effect of a low level of autonomy, while Ryan et al. (2009) find the use of equipment fostered less dependency on the part of the child and could contribute to reducing the perceived negative social impact. Lin (2000) identifies the severity of the motor impairment plus the cognitive impairment as partially explaining the level of parental distress. These variations in results could come from the measurement used by the authors. Using SEM allowed us to estimate the concept of "motor impairment," which cannot be reduced only to the ability to walk. Pediatricians have reported high stress experienced by parents with a child requiring tube feeding. This particular consequence of motor impairment is not often taken into account in previous studies, but in our statistical results this item reflected motor impairment as strongly as walking ability (respectively, 0.966, Standard Deviation (SD) 3.762, and 0.864, SD 1.715) and discussions with parents confirm this is a significant factor of stress.

5.2. Unemployment and parental distress

The family context is also a strong predictor of distress. The parents' professional situation showed a significant effect on distress: if neither parent works, the risk of distress is greater than if at least one parent works, as in the general population (Paul & Moser, 2006). According to Murphy (Murphy & Athanasou, 1999), various benefits come from employment: a financial benefit as well as a social network, physical activity, occupational satisfaction and a feeling of contributing to society. Parents with a disabled child cope with a number of difficulties, which can result in unemployment and living alone. This combined situation carries high risk for distress in parents. Therefore, professionals caring for the child should be aware of the parental situation, including occupational and marital status. This information could be used by social institutions to target populations to monitor so as to prevent depressive symptoms in parents.

5.3. Family functioning

The strongest significant factor preventing parental distress in this model was the quality of family functioning. This result was expected because other studies find that family dysfunction is a predominant factor in limiting the family's adaptation to disability (Emerson, 2003; Khanna et al., 2011; Raina et al., 2005). In France, in a literature review on the consequences of having a disabled child, Zucman (1982) underlines the importance of the relationships within the family. To our knowledge, no interventional study has tested any kind of therapy to develop family functioning. Nonetheless, in order to promote communication and cohesion, building a family project with all the family members could be expected to show positive results on family functioning. Again, a child growing up in a healthy and stable family will show fewer behavioural disorders (Schor and American Academy of Pediatrics Task Force on the, 2003).

5.4. Positive attitude as a coping mechanism

In our results, a parental positive attitude showed a significant protective effect on parental distress. This positive attitude reflected the ability to be conscious of the gain resulting from a difficult situation and this was very helpful to protect against a deteriorating situation. It is useful to note that the latent positive attitude in the model was explained significantly and positively by the severity of motor impairment: the greater the child's motor impairment was, the more parents described strong positive consequences. In difficult times, a generally positive attitude helps parents remain positive, whereas parents

even in less stressful situations have greater difficulties perceiving positive consequences. According to the present results, the severity of motor impairment explained a small share of the positive attitude (14.1% of the variance). This attitude is therefore explained by other factors. According to Lazarus (Lazarus & Folkman, 1984), the ability to be positive in a stressful situation comes from the ability to accept the situation as irreversible: as long as a difficult situation can be modified, its consequences cannot be viewed as positive. Gupta (Gupta & Singhal, 2004) reviews all positive attitude domains. A positive attitude is related to the level of self-esteem, self-confidence and personal and family growth. So professionals having positive communication with parents, valuing their new skills developed through taking care of their child, could contribute to a positive attitude in parents and thus limit the risk of distress.

Our results showed that the more parents have a positive attitude, the more they perceived a negative financial impact. For these parents, this is related to higher wishes to promote their child's participation in leisure or social activities. These adapted activities are often rare and expensive, explaining a greater level of financial stress.

5.5. *Relieving caregivers: a possible intervention*

Using this model we tested the protective effect of three environmental factors on which a policy intervention could be possible. Parents being able to leave their child for few hours sometimes have less risk of parental distress. This type of relief is often requested by parents, and many European countries suffer from a lack of services. It should be noted that the need for relief from caregiving did not depend on the degree of impairment. According to the present results, occasionally replacing parents in the caregiving tasks could lead to better adaptation.

Financial support and how well the home is adapted to the child's disability showed no significant relation with the level of parental distress, although this does not mean that equipment or financial support is not needed. We tested an adaptation of the Double ABCX model in a cross-sectional survey, whereas the Double ABCX is a longitudinal model. This study is limited in that it only investigated the adolescent period. As a result, we did not observe the consequences of the house adaptation even on negative perception on family finances. Moreover, a large number of parents indicated that they did not need equipment, and therefore they did not need financial support (from 49.6% expressing no need for home extension to 70.9% saying they did not need a kitchen specifically modified for the child).

5.6. *The use of SEM: advantages and limits*

SEM offers numerous advantages in the research on disability, which often mixes medical criteria, social criteria and in the last decade a view on wellbeing. As explained above, it can estimate factors that are not directly measurable, including error measurements. Moreover, increasing methodological research on path estimates and statistical methods provides a high level of adaptability to specific surveys. In the present study, the WLSMV estimation method allowed us to rescale ordinal data, within item response theory, which seems to be closer to the actual complexity of responses.

Although SEM has a number of advantages in this study, it also limited certain results. Many options are therefore possible and each has their own limit. For example, we built a latent variable expressing the severity of all other impairments combined except motor impairment. This probably explained why the cognitive disorder, identified as an aggravating factor in other studies (Laurvick et al., 2006; Lin, 2000; Raina et al., 2005; Thyen et al., 2003), was not significantly related to parental distress. We wished to view the complexity of the family situation exhaustively, including many factors, but this led to less precise estimations of the number of parameters and the sample size. Nonetheless, our findings are supported by previous literature and we are confident that the results reflect half of the target population.

This model is global, allowing for simultaneously studying more relevant factors and subsequent relationships identified in literature.

5.7. *Other limits*

The importance of family functioning as a protective factor of parental distress should perhaps be mitigated; actually, it is family dysfunction that was introduced into the model. It appears that studying a negative factor is not the direct opposite of its positive reverse factor. Negative perceptions have stronger effects than their positive reverses (Ito, Larsen, Smith, & Cacioppo, 1998). This negativity bias could explain why the family functioning concept was the strongest protective factor. Nonetheless, many other studies have shown the great protective effect of family functioning.

6. Conclusions

Understanding the family adaptation to disability is central to promoting well-being in disabled children. According to Bronfenbrenner's ecological human model (Bronfenbrenner, 1986), the child's development is influenced by the environment, from immediate to societal. To enhance the well-being of disabled children, a focus on their physical condition is not enough; there is a need to understand the broad context of the disability situation. Here, the most significant protective factors appear to be the quality of family functioning, the parents' positive attitude, and the possibility of taking time off from

caregiving. These findings can be used to inform the design of proactive caregiver interventions. These interventions could first target families at risk of experiencing severe distress particularly those with children having psychological problems.

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Appendix A.

1- Steps description of model testing and intermediate results

- We searched separately relationships between variables contained in (A) and contained in (B). Then we tested assumed relationships between stressors (A) and resources (B). In a next step, we introduced in this first intermediate model the negative perceived impact (C) in each dimension and the level of parental distress (X) successively as an outcome.
- After checking for robustness of observed links between stressors and resources, a new intermediate model was built introducing simultaneously the five negative perception measurements (C).
- Finally the level of parental distress (X) was introduced as a final outcome depending on stressors (A), resources (B) and negative perceived impact (C).

Modification indices were calculated and intermediate models were revised until having a theoretically meaningful and statistically acceptable model. In order to increase the fit, not significant paths ($p > 0.05$) were excluded. In the case of two concurrent intermediate models, we chose the one explaining the largest part of variance observed in the outcome (R-squared).

An intermediate model was retained if it met at least an acceptable fit, described by the fit indexes above (Hooper, 2008):

- the root mean square error of approximation (RMSEA): value close to 0.05 indicates an acceptable fit (MacCallum, Browne et al., 1996); we considered $p(\text{RMSEA} = 0.05) = 50\%$ as showing very good fit;
- Bentler's Comparative Fit Index (CFI): value close to 0.90 and above indicates an acceptable fit (Bentler 1990);
- Tucker-Lewis Index (TLI): value close to 0.95 and above indicates an acceptable fit (Hu and Bentler 1999).

Intermediate model results

Assumptions between stressors (A) and resources to face these stressors (B) were tested. Except the chi-squared measure (Chi-squared = 250.434, $ddl = 198$, $p < 0.05$), all the goodness of fit indices indicated this model (A-B) fit well with the data (RMSEA = 0.035 [CI 90: 0.019 – 0.047], $p(=0.05) = 0.980$; CFI = 0.986; TLI = 0.984). Among stressors (A), the severity of motor impairment was strongly correlated with the severity of associated impairments. And this latter latent variable was strongly correlated with the SDQ score. The marital status and the parental median qualification explained the variance of the FAS score (14.8%) and the parental employment situation (28.5%). The school attainment was explained moderately by the adolescent's age and the severity of associated impairments. Resources (B) described in this model were explained by some stressors (A). RC was only explained weakly and negatively by the severity of associated impairments ($p = 0.042$); the more the associated impairments were severe, the less RC was available. The HA depended on the severity of motor impairment and the FS score. Moreover the FS was explained strongly by the severity of motor impairment and weakly by the FAS score. The PA was explained strongly by the severity of motor impairment; as the adolescent's motor impairment was severe, as the PA was high. The FAD was strongly explained by the SDQ score and weakly by the marital status of the respondent.

Each dimension of negative perceived impact was introduced as the only outcome of this model. This led to confirm some assumed links and to reject other hypotheses. NPFI was strongly predicted by the severity of associated impairments, moderately by FAD and weakly explained by HA and PA. negative perceived impact on time was explained strongly by the severity of motor impairment and weakly by the SDQ total score and the level of qualification of parents. The negative perceived impact on time was stronger with higher parental education level. No resource was identified as a significant predictor of the negative perceived impact on time. We found two concurrent fitting models explaining the NPIH. The negative perceived impact on health could be explained strongly by the severity of motor impairment and weakly the SDQ

total score. Or it could be explained strongly by the HA and RC. As described in methodology, the relationships described in the latter model were kept because of the largest part of variance explained by the second one (29.2% vs 43.1% of variance explained, respectively). Thus the negative perceived impact on health was found to increase with a high level of HA, reflecting the child's needs, and also to decrease with a high level of RC. The negative perceived impact on time was explained strongly by the severity of motor impairment, moderately the SDQ total score and weakly by the FAD. The NPSI was explained strongly by the severity of motor impairment, moderately by the FAD and weakly by the adolescent's age; as the adolescent was old, as the NPSI was high. Each intermediate model explained a significant part of the negative perceived impact in different dimension of family life, from 27.5% (negative perceived impact on time) to 48.0% (NPSI).

Robustness of links (and lack of robustness)

After gathering all these significant relationships in a single model, weak paths became non-significant. This concerns the marital status excluded from the factors explaining the FAD, the adolescent's age in the explanation of the NPSI and the HA in the negative perceived impact on time. This model including paths between stressors (A), resources (B) and negative perception of the situation (C) fitted well the data (Chi-squared = 604.588, ddl = 434, $p < 0.05$; RMSEA = 0.042 [0.034 – 0.050], $p(=0.05) = 0.948$; CFI = 0.971; TLI = 0.967).

Explaining the family adaptation

Before gathering all relationships verified previously and adding the final outcome, the PSI-PD, we tested effects of stressors (A) and resources (B) on the PSI-PD. This led to explain significantly the PSI-PD by the severity of motor impairment ($p < 0.001$), the SDQ score ($p < 0.001$), the adolescent's age ($p = 0.010$), the marital status of the parents ($p = 0.022$), the parental employment situation ($p = 0.004$), the PA ($p = 0.008$) and the FAD ($p < 0.001$). This model fitting well with data (Chi-squared = 263.129, ddl = 210, $p < 0.05$; RMSEA = 0.034 [0.018–0.046], $p(=0.05) = 0.987$; CFI = 0.986; TLI = 0.983) explained 47.8% of the variance of the PSI-PD. Previous significant relationships were maintained. Introducing the negative perceived impacts in the model led to increase the explained variance of PSI-PD of 10%. But only negative perceived impact on health explained significantly the PSI-PD score, with a direct effect of 0.450.

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