

# Problem Behavior in Children of Chronically Ill Parents: A Meta-Analysis

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**Abstract** The aim of this meta-analysis is to examine whether children of chronically ill parents differ from norm groups in problem behavior. We report moderator effects and overall effect sizes for internalizing, externalizing and total problem behavior assessed by children and parents. In fixed effect models, we found a significant overall effect size for internalizing problem behavior (number of studies  $k = 19$ , total sample size  $N = 1,858$ , Cohen's  $d = .23$ ,  $p < .01$ ) and externalizing problem behavior ( $k = 13$ ,  $N = 1,525$ ,  $d = .09$ ,  $p < .01$ ) but not for total problem behavior ( $k = 7$ ;  $N = 896$ ). Effects for internalizing and externalizing problem behavior were larger in non-cancer studies, in samples including younger children and younger ill parents, in samples defined by low average SES and in studies including parents with longer illness duration. In addition, effects for externalizing problem behavior were larger in studies characterized by a higher percentage of ill mothers and single parents. With exclusive self-report, effect sizes were significant for all problem behaviors. Based on these results, a family-centered approach in health care is recommended.

**Keywords** Chronic parental illness · Children · Adolescents · Problem behavior · Meta-analysis

## Introduction

Parents with a chronic medical condition (CMC) compose a significant proportion of the world's population, with prevalence ranging between 4 and 12% (Barkmann et al. 2007; Worsham et al. 1997). CMC is defined as a syndrome involving one or more organs and impairing health and psychological functioning during at least 3 months (Brown 2006). Health care professionals mainly pay attention to ill parents and their spouses, leaving children in the background even though a proportion of them become lifetime caregivers (Visser-Meily et al. 2006). In fact, children often feel overwhelmed by the demand or responsibility to care for an ill person and lack important information about the condition (Schrag et al. 2004). Research also shows that they perceive their own risk for developing illness higher than controls (Harris and Zakowski 2003). Conjointly, the offspring's functioning seems to be determined by negative circumstances of parental illness although positive aspects have also been associated with the illness such as gaining a sense of fulfillment by caring for their parent and building up a cohesive support system (Johnston et al. 1992; Newman 2002). Ill parents' offspring predominantly feel restricted in daily activities, isolate themselves from peer groups and develop health problems (Earley and Cushway 2002). It is, therefore, important to investigate the impact of parental CMC on problem behavior in children.

Children's emotional problems due to endangered parental health have been reported in many qualitative and quantitative studies (Korneluk and Lee 1998; Visser et al. 2004). Acute emotional problems among children with parental CMC, in this paper referred to as target group, are shown to be as high as 55% and frequently persist into adulthood (van de Port et al. 2007; Wong et al. 2009).

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Numerous studies conclude that children's problem behavior is determined by the amount of daily hassles and the perception of stressfulness rather than by the severity of illness (Dufour et al. 2006; Korneluk and Lee 1998; Verhaeghe et al. 2005). Children may react to the imposed stressor by isolating themselves, feeling guilty and worrying about changes in parental health. Fearing negative health outcomes or death of parents may result in psychosomatic complaints, such as headaches, cramps and weakened immune responses in the target group (Pakenham and Bursnall 2006). These symptoms are described as depressive symptoms, anxiety, withdrawn behavior and physical complaints, composing internalizing problem behavior. Children in the target group may also act out showing externalizing problems through aggressive and delinquent behavior (e.g., Diareme et al. 2006). Similarly, there may be an increased level of total problem behavior in the target group referring to a combination of externalizing, internalizing, social, identity and thought problems (e.g., Rodrigue and Houck 2001). In contrast, some studies did not find a marked effect of parental CMC on child functioning (Annunziato et al. 2007; Houck et al. 2007). The differential influence of moderators might explain this discrepancy.

According to the stress and coping theory, the threat of worsening parental health is defined as a continuous stressor exceeding children's coping resources and increasing the probability of problem behavior (Forehand et al. 1998; Pakenham and Bursnall 2006; Pedersen and Revenson 2005). Parental disease can be perceived as stressful depending on factors directly relating to children and factors associated with the situation (Lazarus 1974). Moderators related to children (e.g., age and gender) and moderators related to the situation (e.g., illness type, parental functioning, SES) may explain variations in effect sizes or differences in problem behavior between the target group and children from norm groups (Nelson and While 2002).

First, children's age seems to moderate effect sizes, but research is inconclusive whether latency-aged children are more prone to clinical problem behavior than adolescents (Visser et al. 2005). However, as adolescents are physically and cognitively more advanced than latency-aged children, they may face more caregiving tasks while struggling with identity formation and developmental tasks during puberty (Kraaij et al. 2003). The age of ill parents may also play a moderating role but has largely been ignored as predictor of problem behavior in the target group (Visser et al. 2004). Second, the parents' and children's gender may influence the size of potential effects for problem behavior in children with parental CMC. Girls especially adopt caregiving tasks and are found to be generally at higher risk for stress, depressive symptoms and other internalizing problem

behaviors than boys (Korneluk and Lee 1998). While research on children of the general population suggests that boys display more externalizing problem behavior than girls (Bongers et al. 2003; Vaalamo et al. 2002), research on children with a chronically ill parent has shown the opposite (e.g., Visser et al. 2005). In addition, girls confronted with an ill mother may display more problems than facing an ill father because they tend to identify with the parent of the same gender. Similarly, boys of chronically ill fathers may suffer more than boys of chronically ill mothers (Barkmann et al. 2007). Third, children of low socio-economic status (SES) are shown to have fewer resources to deal with parental CMC than children of high socio-economic status, which may explain increased child adjustment difficulties (Forehand et al. 1998). Low SES involves reduced social and financial resources to cope with the stressful life event of having an ill parent (Folkman et al. 1987). Fourth, ethnicity is a possible moderator although its influence on effect sizes is unclear because no relevant cross-cultural literature has been found. Fifth, the percentage of single parenthood proves to be a potential moderator: children of single parents with CMC may have more difficulties adapting to parental illness because they lack the spouse's support (Taaniila et al. 2002).

Furthermore, illness characteristics, such as illness duration, may moderate effect sizes. Medical conditions of extended duration can lead to a depletion of children's resources and initiation of activities associated with long-term difficulties, for example, frequent contact with deviant peers. This would be in accordance with the delayed-effect hypothesis (Forehand et al. 1998). Cancer studies suggest that most affected children do not display problem behavior shortly after their parent's diagnosis (Visser et al. 2007), while a longitudinal study on parental stroke shows the opposite (van de Port et al. 2007). In addition, CMC's can be categorized into three dimensions of illness according to Rolland's typology: type of illness, illness stadium and components of family functioning (Rolland 1987). CMC's differ depending on illness onset (acute or gradual), illness course (progressive, constant or episodic), illness outcome (fatal, possibly fatal, reduced longevity, non-fatal) and the degree of impairment (impairing vs. non-impairing) (Schepers et al. 2007). Further, studies can be classified as illness specific and illness non-specific (i.e., studies focusing on one specific diagnosis vs. studies with variability in diagnosis). Aside from this, we will use cancer study type as moderator considering the high amount of cancer studies (i.e.; cancer studies vs. non-cancer studies). Effect sizes may vary due to assessment type as well. Evidence suggests that parents report fewer problems than children themselves (Watson et al. 2006).

The theoretical and empirical underpinnings lead to the following hypotheses: (1) effect sizes for internalizing

problem behavior will be higher in studies with a higher percentage of girls than boys; (2) effect sizes will be larger for studies with a higher mean age than studies with a lower mean age; (3) studies including families of lower SES will manifest larger effect sizes than studies of high SES families; (4) studies including more single parent families versus two-parent families will show larger effect sizes; (5) the longer the illness persists, the larger effect sizes will be; (6) effect sizes will be smaller according to parent reports than self-reports.

Significantly, few quantitative studies involve a control group to investigate whether emotional problems are more pronounced in the target group than in children with healthy parents. Reviews to date have failed to provide an overall effect size for internalizing, externalizing and total problem behavior and have thus been inconclusive about whether having a chronically ill parent affects children's problem behavior. While some studies found evidence that these children experience more internalizing than externalizing problems (Steck et al. 2007), others found the opposite (Hough et al. 2003), so no conclusion can be drawn about how pronounced this effect is. The primary aim of this review, therefore, is to investigate whether children of chronically ill parents are at increased risk for developing these problems compared to children among the general population. This is achieved by quantitatively comparing problem behavior scores of the target group with scores of control groups (i.e., children with healthy parents) or non-clinical norm samples when studies omitted controls. We further aim to evaluate if there are variations in effect sizes for internalizing, externalizing and total problem behavior.

In summary, this review focuses on comparing problem behavior in children with a chronically ill parent with control/norm groups and examines whether child characteristics (gender, age), parent characteristics (gender, age, SES, single parenthood, ethnicity), illness characteristics (duration, variability in diagnosis, typology) and assessment type moderate group differences. By this means, we examine how children with chronically ill parents function in relation to other children. Moreover, we address moderators of problem behavior in the target group to identify risk factors for developmental problems.

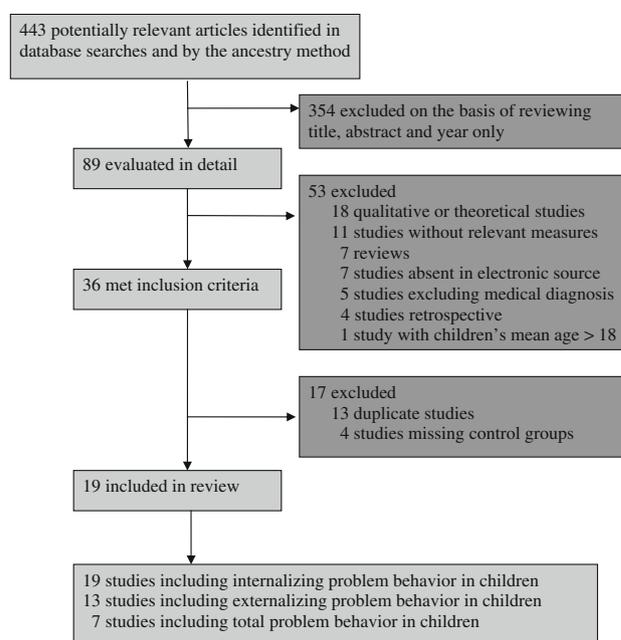
## Method

### Research Procedure

We used the search engines *Medline 1993–1996*, *Medline 1997–present*, *PsycInfo*, *PubMed* and *Web of Science* of the digital library of the University of Amsterdam. Search terms were *parent* and *illness*, *disease*, *physical* and

*chronic*, combined with *adolescent*, *child*, *family*, *internalizing*, *externalizing*, *problem*, *adjustment* and *well-being*. We additionally used terms of prevalent CMC's and illnesses associated with global burden of disease in combination with the words *parent*, *adolescent* and *child* (i.e., asthma, brain damage/contusion, diabetes, cancer, epilepsy, heart disease, HIV/AIDS, multiple sclerosis, Parkinson disease, respiratory disease, rheumatoid arthritis, spinal cord injury, stroke/cardiovascular disease) (World Health Organization 2010). Besides, we used the ancestry method to find more studies concerning children with chronically ill parents in reviews and articles reporting on empirical studies (i.e., reference sections of articles were inspected for relevant studies that had not yet been detected). When there was doubt about the relevance of these articles, they were visually inspected. The flowchart in Fig. 1 illustrates the inclusion procedure.

After exploring the search output, studies were controlled for compatibility with five criteria: (a) at least one parent was diagnosed with CMC; (b) studies included quantitative measures of internalizing, externalizing, or total problem behavior in children; (c) children's mean age in the target and control group <18 years; (d) studies were published between January 1990 and June 2010; (e) effect sizes were calculable by comparing scores of the target group with scores of controls or given norms (e.g., community samples of the Child Depression Inventory). When studies failed to provide national norm or control group scores, we used American T-score norms for the CBCL (Achenbach 1991). This is no methodological concern because research finds minor differences in CBCL norm



**Fig. 1** Flowchart of the inclusion procedure

group scores of countries whose norms were integrated in this meta-analysis (Chang et al. 1995; Verhulst et al. 2003). Studies of qualitative nature and those lacking control groups or omitting information necessary to calculate effect sizes were excluded. Studies not meeting the criteria for our definition of internalizing, externalizing and total problem behavior and CMC were also excluded. Although infection with HIV is not an illness per definition, it falls under the category of CMC, which occasionally is also referred to as illness in this article. Finally, we excluded duplicate studies and dissertation abstracts. After all, we contacted the corresponding authors of retrieved articles and considered articles suggested for inclusion.

### Dependent Measures

Our outcome variable was the effect size Cohen's *d* and was calculated for (a subscale of) internalizing, externalizing, total problem behavior in children. Subscales of the internalizing spectrum included depression, anxiety, somatic complaints and withdrawn behavior. Aggressive and delinquent behavior composed subscales of the externalizing spectrum. Total problem behavior consisted of externalizing, internalizing, social, identity, thought and sexual problems.

### Moderators

Moderators were coded by two independent researchers applying our coding criteria and were ideally verified by the corresponding author. When authors did not reply, the coding was the result of agreement between coders after consensus.

*Age.* Mean age of children and ill parents was registered in years.

*Gender.* The percentage of girls and ill mothers was calculated.

*Ethnicity.* Ethnicity was provisionally coded as *only Caucasian* (1), *mostly Caucasian* (2), *mostly African-American* (3), *only African-American* (4) and *mixed* (5). For moderator analyses, ethnicity was finally coded as *mostly Caucasian* (1) and *mostly non-Caucasian* (2).

*SES.* We estimated average SES as either *low* (1), *medium* (2) or *high* (3) depending on the percentage of parents with low education, yearly family income, mean time of education of parents and other information such as percentage of participants living in a low-income neighborhood, percentage of parental employment and indexes like the Hollingshead socioeconomic factor score (Brown et al. 2007). When the mean income of parents was less than 30 000 US dollars on a yearly basis, the mean SES of a study was coded as being low. When the study indicated that the majority of parents did not graduate from high

school or the percentage of low education (elementary school until college without graduating) and low vocational degree among parents was over 25%, SES was judged to be low unless the family income was higher than 30 000 dollars on average. An example of high SES was that the majority of parents were employed, had an income higher than 30 000 US dollars and were mostly moderately to highly educated.

*Illness type.* Studies were dichotomized into *illness specific* (1) when studies were based on one specific diagnosis (e.g., multiple sclerosis) or *illness non-specific* (2) when several illnesses were included in one study which is also called variability in diagnosis. Besides, we classified studies as *cancer non-specific* (1) and *cancer specific* (2), taking into account a possible moderator effect of study characteristics inherent to cancer. Lastly, studies were coded based on how the illness started (acute or gradual onset), its course (progressive, constant or episodic), the outcome (fatal, possibly fatal, reduced longevity or non-fatal) and the degree of impairment (impairing vs. non-impairing).

*Illness duration.* As an estimate of illness duration, time since diagnosis was defined by the duration in months since a professional had diagnosed the parent with a medical condition.

*Living condition.* Raters registered whether children were *living mostly at home with the ill parent* (1) or *living away from home* (2).

*Percentage of single parents.* Raters coded the percentage of single parents with CMC sharing a household with their children as only adult. When a parent lived with another adult such as mother, sister or partner, the definition of single parent household was not applicable.

*Assessment type.* We dichotomized studies into *self-report* (1) and *parent report* (2).

### Data Analysis

On the basis of the presentation of results, we calculated the effect size Cohen's *d* as a function of means and standard deviations of children's problem behavior scores. When scores were unavailable, we estimated the effect size based on odds ratios, *p*-values or *t*-tests. In studies using other norms than those for the CBCL and stating that the difference between the target group and the normative or control sample was not significant without providing scores, we set the effect size to be zero. The effect size in a given study was calculated by multiplying effect sizes per subgroup (e.g., girls aged 4–11) with the number of that subgroup. The resulting sum scores of subgroups were divided by the total number of participants, meaning that we took the subpopulation weight into account. In studies reporting T-scores of the CBCL, we chose normative

T-scores matched on gender and age (i.e., boys aged 4–11, girls aged 4–11, boys aged 12–18 and girls aged 12–18) (Achenbach 1991). Raw scores were compared to scores from national norms. We calculated means and standard deviations as overall mean for parent reports, which depended on the number of fathers and mothers (i.e., scores of mothers and fathers were multiplied with the number of mothers and fathers, respectively, and then divided by the total number of parents). When both parent and child reports were available, we based the effect size on parent reports to avoid moderator analyses based on duplicate data. When several measures were available to measure a spectrum of problem behavior, we averaged effect sizes by dividing the effect size for each instrument by the total number of instruments. Effect sizes of  $d \leq .20$ ,  $d = .50$  and  $d \geq .80$  were considered as indices of small, medium and large effects, respectively (Cohen 1992).

Overall effect sizes were estimated in random as well as fixed effect models with SPSS macros (Lipsey and Wilson 2001). In fixed effect models, all studies are considered equivalent, and the residual variance is based on the total number of participants, yielding higher statistical power but limiting generalizability. In random effect models, studies are not considered identical, and the residual variance takes differences between studies into account. Random effect models may be preferable because results are more generalizable than results from fixed effects modeling. Both fixed and random effects are provided for the sake of a complete picture of the effect sizes. Homogeneity of studies was tested with  $Q$  statistics at a significance level of  $p = .05$ . In a heterogeneous set of studies, differences in effect sizes were assumed to stem from study characteristics rather than subject-level sampling error. In view of the small number of studies, moderator effects were tested separately. In order to investigate possible communality of moderator effects, correlations between moderators were tested with Pearson correlations.

## Results

### Description of Studies

The smallest sample consisted of 23 children from the target group and the largest sample included 336 children. Children's age ranged between 3 and 25 years (mean age = 11.85), Table 1. Illness duration or mean time since diagnosis ranged between 2 months and 9.5 years and was 3.7 years on average. Studies included slightly more girls than boys. Over two-thirds of the ill parents were female. The average age of the ill parent was 42 years. One in 4 of the parents with CMC was estimated to be single. Almost two-thirds of all studies (63%) were executed in the United

States. The remainder was conducted in the Netherlands (16%), Australia, Germany and the United Kingdom. One study was cross-cultural involving samples from three countries in Europe. Three studies had a longitudinal design in which the first assessment of children's problem behavior was chosen, while the vast majority of studies had a cross-sectional design. Thirty-seven percent of the studies included a control group; the remainder were compared to norms from Achenbach (1991) or, in case of raw scores, to given norms.

Fourteen studies (74%) used the CBCL internalizing problem scale, while five studies used other measures, totaling 19 studies. Most studies used the CBCL to measure externalizing problem behavior (89%) and the effect size for total problem completely relied on the CBCL. Seven studies used the youth self-report (YSR) in addition to the CBCL but these were not included in the analyses due to simultaneous parent report and the emerging dependency between studies. The Teacher Report Form of the CBCL was only used in two studies and was therefore not integrated in this meta-analysis. In the majority of studies, the CBCL was filled in by both father and mother. In five cases, especially single parent studies, only one parent filled in the CBCL. The psychometric properties of instruments were generally found to be good to excellent. However, less than half of the studies evaluated the reliability and validity. Seven studies entirely focused on cancer (37%). Five studies included various CMC's two of which included parents with either HIV or hemophilia or both. Three studies included highly varying CMC's (e.g.; asthma, diabetes, heart disease, liver disease, lung disease and stroke). In one sample, CMC's were not specified but it was clear that they varied. Three studies focused on HIV/AIDS and two studies investigated MS. One study included stroke. After inspecting studies and contacting the authors, there were too many missing values to include the children's living condition as moderator. In view of insignificant variance in ethnicity across studies (79% of studies were composed of only or mostly Caucasian), we excluded ethnicity as moderator. For this reason, assessment type was also excluded. However, all 11 studies including self-report measures will be analyzed and described separately and compared to effects for exclusive parent report (15 studies). Finally, *illness typology* was discarded since a majority of studies included various CMC's to which the coding could not be applied simultaneously.

### Effect Sizes for Internalizing Problem Behavior

Nineteen independent studies were included to calculate the effect size for internalizing problem behavior ( $N = 1,858$ ), Table 1. The meta-analysis yielded a small overall effect size ( $d = .23$ ;  $p < .01$  (95% CI [.19; .28]),

fixed model;  $d = .24$ ;  $p < .01$  (95% CI [.11; .37]), random model), indicating that children in the target group displayed more internalizing problem behavior than other children. The homogeneity analysis revealed that effect sizes varied significantly between studies and moderator analyses were appropriate ( $Q(18) = 132.29$ ,  $p < .01$ ). In fixed effect models, mean age of children and ill parents were significant moderators, indicating that larger effects were found for studies including younger children and younger ill parents, Table 2. Studies including more families with low SES had significantly larger effect sizes for internalizing problem behavior than studies including more families with high SES. Effects for internalizing problem behavior were less pronounced in cancer studies than in non-cancer studies. Finally, effects were larger in studies characterized by longer illness duration. Moderation in effect sizes for internalizing problem behavior was not explained by gender of children and ill parents, the percentage of single parents and variability in diagnosis. Heterogeneity within studies was present in all fixed effect models and heterogeneity between studies applied to studies including children's and ill parents' age, SES, cancer study type and illness duration. When random effect models were computed, solely SES reached significance.

#### Effect Sizes for Externalizing Problem Behavior

Thirteen studies were used to calculate the effect size for externalizing problem behavior, which can be considered small ( $N = 1,525$ ;  $d = .09$ ;  $p < .01$  (95% CI [.04; .14]), fixed model;  $d = .15$ ;  $p = .16$  (95% CI [−.06; .37]), random model), indicating that children with parental CMC displayed slightly more externalizing problem behavior than the comparison group. The homogeneity analysis revealed that effect sizes varied significantly between studies, and moderator analyses were appropriate ( $Q(12) = 203.23$ ,  $p < .01$ ). In fixed effect models, gender of the ill parent was a significant moderator revealing that studies with a higher percentage of ill mothers showed larger effect sizes for externalizing behavior than studies with a lower percentage of ill mothers. Children's and ill parents' mean age were significant moderators, indicating that larger effects were found in studies including younger children and younger ill parents than in studies including older children and older ill parents. Studies including more families with low SES showed larger effect sizes for externalizing problem behavior than studies including families characterized by high SES. Further, studies including more single parents showed larger effects than studies with less single parents. Effects for externalizing problem behavior were smaller in cancer studies than in non-cancer studies. Finally, effect sizes were larger in studies including parents with longer illness duration than

in studies with shorter mean time since diagnosis. Heterogeneity was present in most fixed effect models. When random effect models were computed, only mean age of the ill parent reached significance.

#### Effect Sizes for Total Problem Behavior

The effect size for total problem behavior was based on 7 studies ( $N = 896$ ). The meta-analysis yielded a non-significant overall effect size ( $d = -.03$ ;  $p = .43$  (95% CI [−.09; .04]), fixed model;  $d = .02$ ;  $p = .82$  (95% CI [−.13; .16]), random model), indicating that children in the target group did not manifest more total problem behavior than children with healthy parents. The homogeneity analysis revealed that effect sizes significantly varied between studies and that moderator analyses were justifiable ( $Q(6) = 23.07$ ,  $p < .01$ ). In both fixed and random effect models, ill parents' age, illness type and cancer study type were significant moderators, revealing that effects for total problem behavior were larger in cancer studies, in samples including younger ill parents and in studies with more variability in diagnosis. Heterogeneity was present in the minority of fixed and random effect models.

#### Effect Sizes for Self-Reported Problem Behavior

First, the meta-analysis of 10 self-report studies yielded a small overall effect size for internalizing problem behavior in adolescents (mean age = 14.17;  $N = 679$ ;  $d = .25$ ;  $p < .01$  (95% CI [.18; .31]), fixed model;  $d = .27$ ;  $p < .01$  (95% CI [.14; .40]), random model), indicating that adolescents in the target group reported more internalizing problem behavior than adolescents with healthy parents, Table 3. To compare, the overall effect size for exclusive parent-reports of internalizing problem behavior was similar to effects for self-report ( $k = 15$ ;  $N = 1,628$ ;  $d = .25$ ;  $p < .01$  (95% CI [.20; .30]), fixed model;  $d = .24$ ;  $p < .01$  (95% CI [.08; .39]), random model). Second, the effect size for self-reported externalizing problem behavior in adolescents (mean age = 14.70) resulted to be significant ( $k = 6$ ;  $N = 449$ ;  $d = .22$ ;  $p < .01$  (95% CI [.14; .29]), fixed model;  $d = .28$ ;  $p = .12$  (95% CI [.07; .49]), random model), meaning that adolescents with parental CMC reported comparatively more externalizing problem behavior. The effect size for self-reported externalizing problem behavior in adolescents (mean age = 14.69) was larger than parent-reported externalizing problem behavior ( $d = .09$ ;  $p < .01$ , fixed model;  $d = .15$ ;  $p = .16$ , random model). Third, the meta-analysis yielded a small overall effect size for self-reported total problem behavior according to fixed effect models but not random effect models ( $k = 5$ ;  $N = 400$ ;  $d = .13$ ;  $p < .01$  (95% CI [.05; .20]), fixed model;  $d = .19$ ;  $p = .06$  (95% CI [.00; .38]),

**Table 1** Moderators and effect sizes in studies assessing internalizing, externalizing and total problem behavior in children with parental CMC

Authors	N	Parental CMC's	% girls	% ill mothers	Mean age child	Mean age ill parent	Illness duration	Mean SES	% single parent	Instruments	d int.	d ext.	d tot.
Annunziato et al. (2007)	211	Mixed	46.00	100.00	12.06	40.95		High	99.00	CDI-S/BPI	.00	.00	
Barkmann et al. (2007)	79	Mixed	53.24	55.70						CBCL	.61	-.13	.41
Biggar and Forehand (1998)	85	HIV/AIDS	59.00	100.00	8.68	31.25		Low	50.00	CDI	.39		
Brown et al. (2007)	40	Cancer	52.50	100.00	14.40	45.90	4.80	Medium	17.50	CBCL	.37	.25	
Harris and Zakowski (2003)	27	Cancer	66.70	81.80	15.40	48.40	2.50	Medium		CDI/ RCMAS	-.16		
Houck et al. (2007)	38	Mixed	66.78	34.22	14.92	44.70		High	.00	CBCL	.00		-.12
Hough et al. (2003)	147	HIV/AIDS	46.90	100.00	9.90	36.10	5.23	Low	78.00	CBCL	.74	1.08	
Kotchick et al. (1996)	75	Mixed	55.00	.00	12.88	38.70		Medium	.00	CBCL	.28	.02	
Pakenham and Bursnall (2006)	48	MS	56.00	95.83	15.60		9.15	Medium		BSI-18	.13		
Rodrigue and Houck (2001)	33	Mixed	45.45	63.64	12.60	38.70		Medium		CBCL/YSR	.64	.92	.35
Siegel et al. (1996)	70	Cancer	57.00	46.00	10.80			High	.00	CDI/STAI	.53		
Steck et al. (2007)	192	MS	48.44	71.53	9.80	40.80	7.36	Low		CBCL/YSR	.32	.00	.00
Steele et al. (1997)	65	Mixed	56.90	.00	13.27	39.50		Medium	.00	CBCL	.35		
Tompkins and Wyatt (2008)	23	HIV/AIDS	61.00	100.00	12.59	38.33		Medium	52.00	CBCL/YSR	-.28	.56	
Visser et al. (2007)	123	Cancer	56.14	52.38	8.00	43.00		High	8.70	CBCL/YSR	-.03	-.23	-.16
Visser et al. (2005)	336	Cancer	51.19	81.11	11.40	44.30	2.70	Medium	7.00	CBCL/YSR	.05	-.05	-.09
Visser-Meily et al. (2005b)	82	Stroke	51.00	58.26	13.20	44.40	.17	Medium	.00	CBCL	.37	.07	
Watson et al. (2006)	95	Cancer	58.19	100.00	12.00	45.00	.91	High	26.17	CBCL/YSR	.14	-.16	-.12
Welch et al. (1996)	89	Cancer	56.18	75.00	11.98	39.90	.19	Medium	6.58	CBCL	-.22	-.23	

N Sample size, CMC chronic medical condition, CDI-S children's depression inventory-short form, BPI behavior problem index, CBCL child behavior checklist, CDI child depression inventory, RCMAS revised children's manifest anxiety scale, YSR youth self-report, BSI-18 brief symptom inventory-18, STAI state-trait anxiety inventory, d Cohen's d (effect size), int. internalizing problem behavior, ext. externalizing problem behavior, tot. total problem behavior

**Table 2** Moderators of effect sizes for problem behavior in children with parental CMC

Moderator	Internalizing problem behavior				Externalizing problem behavior				Total problem behavior						
	<i>k</i>	<i>d</i>	$\beta$	<i>Q<sub>b</sub></i>	<i>Q<sub>w</sub></i>	<i>k</i>	<i>d</i>	$\beta$	<i>Q<sub>b</sub></i>	<i>Q<sub>w</sub></i>	<i>k</i>	<i>d</i>	$\beta$	<i>Q<sub>b</sub></i>	<i>Q<sub>w</sub></i>
% girls	19					13					7				
Fixed		.23	.12	1.99	130.47**		.09	.08	1.32	202.61**		-.03	.26	1.53	21.54**
Random		.24	-.02	.00	17.19		.16	.10	.12	12.04		.02	.23	.39	6.75
% ill mothers	19					13					7				
Fixed		.23	-.10	1.33	131.13**		.09	.29**	16.79**	187.14**		-.03	-.28	1.78	21.29**
Random		.26	-.14	.32	16.97		.15	.26	.89	12.40		.02	-.20	.30	7.02
Children's age	18					12					6				
Fixed		.22	-.45**	24.39**	96.84**		.10	-.32**	20.73**	179.41**		-.07	-.21	.34	7.14
Random		.22	-.41	3.26	16.44		.18	-.14	.25	11.94		-.06	-.10	.06	5.42
Ill parents' age	16					12					6				
Fixed		.21	-.49**	26.88**	87.40**		.10	-.68**	92.53**	107.61**		-.07	-.76*	4.28*	3.21
Random		.21	-.39	2.71	15.34		.17	-.60*	7.03**	12.38		-.07	-.76*	4.28*	3.21
SES	18					12					6				
Fixed		.22	-.60**	45.00**	77.23**		.10	-.51**	52.51**	147.62**		-.07	-.53	2.09	5.40
Random		.22	-.46**	4.98*	18.56		.18	-.51	3.66	10.56		-.06	-.49	1.53	4.75
% single parents	14					10					4				
Fixed		.18	.15	2.29	96.79**		.10	.47**	39.37**	138.35**		-.11	-.15	.01	.41
Random		.20	.08	.07	11.32		.13	.54	2.46	6.06		-.11	-.15	.01	.41
Variability CMC	19					13					7				
Fixed		.23	-.08	.92	131.54**		.09	-.04	.41	203.52**		-.03	.78**	14.09**	8.98
Random		.24	.10	.18	16.55		.16	.11	.15	11.56		-.002	.76**	8.65**	6.32
Cancer study type	19					13					7				
Fixed		.23	-.50**	33.07**	99.38**		.09	-.44**	38.88**	165.05**		-.03	-.66**	10.20**	12.87*
Random		.24	-.39	3.20	18.30		.15	-.45	2.91	11.20		.01	-.66*	4.58*	8.85
Illness duration	9					8					3				
Fixed		.24	.57**	27.08**	55.85**		.13	.39**	24.60**	140.08**		-.07	1.00	1.26	.01
Random		.22	.43	1.72	7.51		.14	.48	1.85	4.05		-.07	1.00	1.26	.01

CMC Chronic medical condition (positive scores mean high variability in diagnosis). Cancer study type is coded as non-cancer studies (1) and cancer studies (2), meaning that negative beta coefficients indicate smaller effects for cancer studies. *k* = number of studies; *d* = Cohen's *d* (effect size); *Q<sub>b</sub>* = *Q* statistic between studies (index of variability between the group means); *Q<sub>w</sub>* = statistic within studies (index of variability within the groups)

\* *p* < .05; \*\* *p* < .01

**Table 3** Effect sizes in studies assessing self-reported internalizing, externalizing and total problem behavior in children with parental CMC

Author	<i>N</i>	Parental CMC	Instrument	% girls	Mean age child	<i>d</i> int.	<i>d</i> ext.	<i>d</i> tot.
Biggar and Forehand (1998)	85	HIV/AIDS	CDI	59.00	8.68	.39		
Diareme et al. (2006)	27	MS	YSR	37.04	13.00	.59	.62	.59
Harris and Zakowski (2003)	27	Cancer	CDI/RCMAS	66.70	15.40	-.16		
Pakenham and Bursnall (2006)	48	MS	BSI-18	56.00	15.60	.13		
Rodrigue and Houck (2001)	29	Mixed	YSR	48.00	12.60			.40
Siegel et al. (1996)	70	Cancer	CDI/STAI	57.00	10.80	.53		
Tompkins and Wyatt (2008)	23	HIV/AIDS	YSR	61.00	12.59	.50	.80	
Visser et al. (2007)	66	Cancer	YSR	52.70	15.00	-.04	-.15	-.08
Visser et al. (2005)	222	Cancer	YSR	53.03	15.00	.21	.26	.11
Watson et al. (2006)	56	Cancer	YSR	69.64	15.00	.25	.14	.09
Welch et al. (1996)	55	Cancer	YSR	60.00	14.50	.36	.24	

*N* sample size, *CMC* chronic medical condition, *MS* multiple sclerosis, *CDI* child depression inventory, *RCMAS* revised children's manifest anxiety scale, *YSR* youth self-report, *CBCL* child behavior checklist, *BSI-18* brief symptom inventory-18, *STAI* state-trait anxiety inventory, *d* Cohen's *d* (effect size), *int.* internalizing problem behavior, *ext.* externalizing problem behavior, *tot.* total problem behavior

random model), indicating that adolescents in the target group reported slightly more total problem behavior than children with healthy parents. In contrast, the meta-analysis on parent-reported problem behavior yielded a non-significant effect size for total problem behavior, meaning that children themselves reported more total problem behavior than their parents.

#### Correlations Between Moderators

The Pearson correlations (Pearson's *r*) between moderators resulted to be positive between mean age of children and mean age of ill parents ( $r = .72$ ;  $p < .01$ ) and between percentage of single parents and percentage of ill mothers ( $r = .67$ ;  $p < .01$ ). In addition, mean age of ill parents positively correlated with cancer study type ( $r = .57$ ;  $p < .05$ ) and with average SES ( $r = .58$ ;  $p < .05$ ), indicating that older ill parents were likely to have high SES and to be represented in cancer studies. These correlations should be considered when interpreting moderator effects.

#### Publication Bias

We calculated rank order correlation (Spearman's rho) between the effect sizes and the overall sample size. Correlations for internalizing problem behavior ( $r_s = .14$ ;  $p = .58$ ), externalizing problem behavior ( $r_s = -.39$ ;  $p = .18$ ) and total problem behavior ( $r_s = -.21$ ;  $p = .65$ ) did not prove to be significant, which supports the conclusion that publication bias is not present in this meta-analysis. For self-reported problem behavior, the correlations between the total sample size of adolescents and internalizing problem behavior ( $r_s = -.07$ ;  $p = .84$ ), externalizing problem behavior ( $r_s = -.66$ ;  $p = .16$ ) and

total problem behavior ( $r_s = -.70$ ;  $p = .19$ ) were not significant either. However, the correlations between the total sample of adolescents and externalizing and total problem behavior are negative and can be considered small to moderate, meaning that studies with small sample sizes tend to have larger effect sizes, which is consistent with publication bias. In conclusion, question remains about the plausibility that effect sizes are biased due to sampling error. Therefore, we additionally estimated the number of unpublished studies reporting null effects necessary to reduce significant mean effect sizes to zero, which is referred to as fail-safe *N* (Lipsey and Wilson 2001). To yield a null effect for internalizing problem behavior, 456 additional studies with an effect size of zero would be needed, while only 34 additional studies would be necessary to reduce the mean effect size for externalizing problem behavior to zero. To create a negligible effect for self-reported internalizing problem behavior, 136 additional studies with an effect size of zero would be needed. Thirty and 6 additional studies would be needed to yield a null effect for self-reported externalizing and total problem behavior, respectively. The fail-safe results indicate that more studies should be included to be certain that the overall effect size for self-reported externalizing and total problem behavior is significant.

#### Discussion

This meta-analysis revealed that according to self-report and parent report, children with a chronically ill parent display significantly more internalizing problem behavior than children with healthy parents. To a small extent, having a parent with CMC has an effect on externalizing

problem behavior. In terms of parent-reported total problem behavior in children, there seems to be no difference between children of the target group and other children. While fixed effect models indicate a negative overall effect for total problem behavior, in random effect models, the overall effect size proves to be positive, producing a null effect. However, self-reported total problem behavior in the target group seems to be increased in comparison with controls.

The negligible effect for parent-reported total problem behavior which makes the moderator analyses difficult to interpret may stem from only few studies being involved, leading to lower power for this finding. It should also be noted that this effect size was entirely based on the CBCL, further limiting the external validity. Another explanation for this null effect is that total problem behavior of the CBCL contains six items about bowel and sexual problems and other items about gender identity and thought disorders that may rarely apply to any child. Effectively, in self-reported total problem, there is a small positive overall effect size, meaning that adolescents with parental CMC report more total problem behavior than norm groups. This result, however, may be confounded because studies using self-report were composed by adolescents rather than latency-aged children. Possibly, adolescents report more problems than their parents. This may especially be true for externalizing and total problem behavior because adolescents hardly disclose sexual problems and delinquent behaviors (Watson et al. 2006). In addition, total problem behavior may not constitute typical behaviors of children with chronically ill parents, meaning that the target group is not sensitive to these measures and hence measures for total problem behavior seem to be unspecific to the target group (Pakenham et al. 2006). With regard to influential factors, effect sizes for internalizing and externalizing problem behavior appear to be positively influenced by young ages of ill parents and children. Younger families tend to be distinguished by low SES and may benefit from fewer financial resources and education to deal with the impact of parental CMC. This was confirmed in our study showing high positive correlations between children's average age and SES and between ill parents' mean age and SES. As expected, larger effect sizes for both internalizing and externalizing problem behavior were also found in studies including more families with low SES. These findings are in agreement with the stress and coping theory stating that children with little financial support and low education may lack resources to cope with parental CMC, and therefore, experience more stress which successively results in increased problem behavior (Lazarus 1974). Last but not least, studies including parents with longer illness duration displayed larger effect sizes for internalizing and externalizing problem behavior. This

supports the delayed-effect hypothesis affirming that long-term stressors lead to depletion of resources, which may result in clinical problem behavior (Forehand et al. 1998).

Moreover, effects for all problem behaviors were smaller in studies focusing on cancer, suggesting that cancer belongs to a different category of CMC's. According to Rolland's illness typology, cancer may physically be less impairing in comparison with other CMC's. This chronic illness may also differ from other diseases because there is a chance of complete rehabilitation, especially in breast cancer which is the most represented cancer type in our study sample. While most CMC's in our sample are defined by a progressive and/or episodic course meaning unpredictability and worsening of parental condition, cancer is relatively predictable, unrelated to personality and behavioral changes and not per definition lethal. In the study population, ill parents are relatively young in comparison with cancer patients, which may implicate that they are likely to overcome their disease. Results also show that cancer study type was positively related to age and SES, indicating that older age and high SES may have contributed to the finding that cancer studies report low effects. Notably, one exceptionally high effect size was found in a cancer study focusing on the terminal phase of the parent's illness (Siegel et al. 1996). This demonstrates that reviews may benefit from taking demographics and the stage of the illness into account (Rolland 1999).

Surprisingly, effect sizes for internalizing problem behavior did not prove to be larger in studies characterized by a higher percentage of girls. In contrast to numerous studies concluding that girls manifest more internalizing problem behavior than boys among the general population (e.g., Bongers et al. 2003), this review finds no additional support for this observation among children in the target group. To summarize, it can be hypothesized that boys and girls with parental CMC struggle with similar adjustment difficulties, in particular internalizing problems, and therefore, score similarly on the internalizing spectrum (Pakenham et al. 2006). Nonetheless, an underlying interaction effect between child and parent gender might moderate effect sizes, meaning that studies focusing on boys of ill fathers and girls of ill mothers may show larger effects (Barkmann et al. 2007). Further, we found that studies including more single parents showed larger effects for externalizing problem behavior than studies including more two-parent families. The percentage of single parents was highly correlated with the percentage of ill mothers, indicating that in most cases, single parents were mothers. This indicates that children of single mothers with CMC might be at increased risk for aggression and delinquency. In the same way, research on children with single mothers versus single fathers delivers evidence that the former display more problem behavior (Hoffmann 2006). It has

also been found that specifically delinquent behavior is higher in children of single parent families than two-parent families (Eitle 2006).

The findings of the moderator effects for total problem behavior were unexpected and very different from the results of internalizing and externalizing problem behavior. Although the overall effect size for total problem behavior proves to be negligible, the effect of moderators such as age of ill parents can still be considerable. With regard to the small number of studies reporting non-significant and inconsistent effects, the moderator effects of total problem should be interpreted with care. The overall effect size for total problem behavior in children was negative and seemed to be higher in studies including older parents. This reflects the findings for internalizing and externalizing problem behaviors, providing evidence that studies including younger parents show larger effects for all problem behaviors than studies with older parents. On the one hand, moderator effects and overall effect sizes for problem behavior in the target group might generally be smaller than our results indicate because many studies did not take the between-subject dependence of children within the same families into account. This means that potential effects could also have been explained by the fact that children within the same family share a similar environment (Snijders and Bosker 1999). On the other hand, children and parents of the target group may be accustomed to their situation. Major events appearing ordinary to them may not have the same meaning for children with healthy parents, leading to believe that overall effect sizes may in fact be larger.

This review also has some limitations. Regarding total problem behavior and self-reported problem behavior in children, the number of studies is small. Accordingly, more studies are needed to rule out that these effect sizes are non-significant. Likewise, studies neglected to provide information on potential influential factors necessary for a thorough investigation into moderator effects (e.g., children's living condition, family cohesion and communication, parental functioning and quality of parent-child relationship) (Kotchick et al. 1996; Pakenham et al. 2006; Steele et al. 1997; Tompkins and Wyatt 2008; Watson et al. 2006). Studies including larger samples are required to test these moderators whose omission may explain why the selected studies were heterogeneous even after including moderators. Except for a few cases, moderator effects were not significant in the random effect models because of the small number of studies, resulting in lower power or generalizability of moderator effects. Consequently, conclusions about influential factors cannot be generalized to other studies in this field. A major limitation refers to the predominant use of the CBCL in the study sample, which limits generalizations about problem behavior and

psychological adjustment in the target group. The results should be interpreted cautiously keeping in mind that effect sizes may be marked by properties of the CBCL (i.e., reduced specificity and sensitivity for the target group). A more sensitive instrument for children with parental CMC has already been developed and may increase the validity of a meta-analysis including specific instruments only (Pakenham et al. 2006). It should also be noted that parents, teachers and children have shown to differ greatly in their perception and report of problem behavior in children (Achenbach et al. 2002). This discrepancy, however, is being kept in mind by simultaneously presenting self- and parent reports in this review. Apart from this, some studies even find little difference between self- and parent reports of children's problem behavior (e.g., Welch et al. 1996). Lastly, our results are less generalizable to the target group because the vast majority of studies included mostly Caucasian individuals.

As recommendation, future researchers should recruit culturally more representative samples. Routinely, multi-level analyses should be considered, as brothers and sisters within the same family are statistically dependent on each other, meaning that effects of parental CMC on problem behavior in offspring could be explained by clustering within families (Snijders and Bosker 1999). An improvement of reviews in this field would also imply the application of Rolland's illness differentiation into illness type, illness stadium and components of family functioning (Rolland 1987). For reviews applying Rolland's typology, studies focusing on one specific diagnosis are advantageous. Another major contribution to the knowledge of parental disease would be to investigate which characteristics are related to the finding that children whose parent has cancer are less affected than children whose parent has another CMC. Children's problem behavior may be explained by moderators omitted in this review, which requires explorative and longitudinal studies investigating the influence of family characteristics. For instance, children's living condition needs to be explored in more detail as research suggests that living with an ill parent can be seen as a continuous stressor, although living apart from an ill parent might even more be harmful because children, whose parents are unavailable, may excessively worry and lose their parental reassurance and feeling of control. Similarly, living separately from the ill parent may be an indication of the severity of illness (i.e., parents who are hospitalized might be fighting for their lives or be severely impaired) (Bakas and Burgener 2002). In addition, a suggestion for future research is to examine whether instrumental versus emotional caregiving tasks relate to increased problem behavior in children. A study found that both frequency and children's perception of caregiving tasks predicted problem behavior in children but it is

unknown if this is also true for emotional support provided by children (Meijer et al. 2008). Especially parental psychological functioning requires additional consideration as moderator since parental depression and parents' emotional well-being consistently seem to moderate children's problem behavior (Biggar and Forehand 1998; Diareme et al. 2006; Hough et al. 2003; Rodrigue and Houck 2001; Visser-Meily et al. 2005a, b). Finally, interaction effects between moderators should be investigated. The qualities of this review, however, outweigh the limitations. First, this review provides overall effect sizes for both parent and self-reports of internalizing, externalizing and total problem behavior in children with parental CMC. Second, this quantitative approach sends a clear signal of the need to consider children's psychological functioning under the impact of parental CMC. Third, we detect potential risk factors for internalizing and externalizing problem behavior in the target group. To elaborate, young age of children, young age of ill parents, low SES of families and long illness duration seem to be risk factors for these problem behaviors. Single parenthood and high percentage of ill mothers both prove to be specific risk factors for children's externalizing problems. Hence, we identified additional moderators for specific problem behavior, knowledge of which may help to develop a screening instrument for children who are at increased risk for problem behavior and need professional assistance.

In conclusion, children confronted with parental CMC appear to be at increased risk for internalizing and externalizing problem behavior. In view of the high prevalence of parental CMC in the population, the number of children at risk for depressive, anxious and somatic symptoms may be large. Health care practitioners should be aware of this and refer children with clinical levels of problem behavior to professionals offering interventions (e.g., support groups, psychological counseling, psycho-education and family therapy). Medical doctors are recommended to receive education about how illness can impact on families and how to treat undesirable behaviors and emotions of family members (Gorter et al. 2010). Counselors should also take notice of therapeutic landscapes specific to the treatment of a certain diagnosis and illness stages, for example, the need for psycho-education at the onset of muscle disease (Sperry 2009). Specific treatments for clinically elevated levels of problem behavior in children with parental CMC are required and should be evaluated in randomized control studies. Most importantly, internalizing problems are prevalent among children with parental CMC. Young families, those dealing with an ill parent with long illness duration and families with few financial resources may have an increased need for support. Standard screening of children in the target group may consist of assessing demographic risk factors and paying special attention to

risks for specific problem behaviors, for instance, single parenthood seems to pose a risk for externalizing problem behavior. Screening children soon after the parent has been diagnosed may be an important step for the prevention of persistent developmental problems. This may be achieved if professionals in contact with the target group (e.g., general practitioners, teachers, school doctors and counselors) are alert for potential problem behavior. Asking a few questions about children's adjustment during a consultation can be an important step to initiate help. The fact that parental CMC significantly affects offspring supports the idea that health care professionals should adopt a family-centered approach instead of focusing exclusively on parents (Visser-Meily et al. 2005a). We recommend integrating children during the rehabilitation of the ill parent by informing them about the disease and assisting them in their needs. Paying attention to children's adjustment to parental disease can certainly enhance their quality of life and developmental prospect.

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