Caregiver Coping, Mental Health and Child Problem Behaviours in Cystic Fibrosis: A Cross-Sectional Study

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Abstract

Background In children with cystic fibrosis (CF) sleep, eating/mealtime, physiotherapy adherence and internalising problems are common. Caregivers also often report elevated depression, anxiety and stress symptoms.
Purpose To identify, through principal components analysis (PCA), coping strategies used by Australian caregivers of children with CF and to assess the relationship between the derived coping components, caregiver mental health symptoms and child treatment related and non-treatment related problem behaviours.

Method One hundred and two caregivers of children aged 3 to 8 years from three CF clinic sites in Australia, completed self-report questionnaires about their coping and mental health and reported on their child's sleep, eating/mealtime, treatment adherence and internalising and externalising behaviours.
Results Two caregiver coping components were derived from the PCA: labelled 'proactive' and 'avoidant' coping. 'Avoidant' coping correlated moderately with caregiver depression (0.52), anxiety (0.57) and stress (0.55). For each unit increase in caregiver use of avoidant coping strategies, the odds of frequent child eating/mealtime behaviour problems increased by 1.3 (adjusted 95 % CI 1.0 to 1.6, *p*=.03) as did the odds of children experiencing borderline/clinical internalising behaviour problems (adjusted 95 % CI 1.1 to 1.7, *p*=.01). Proactive coping strategies were not associated with reduced odds of any child problem behaviours.
Conclusions Avoidant coping strategies correlated with caregiver mental health and child problem behaviours. Intervening with caregiver coping may be a way to improve both caregiver mental health and child problem behaviours in pre-school and early school age children with CF.

Keywords Cystic fibrosis · Child behaviour · Caregiver coping · Mental health · Principal components analysis

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Introduction

Cystic fibrosis (CF) is a chronic, life-shortening illness which affects about 1 in 2,874 children born in Australia [1], and is the most common inherited disease in Caucasian children [2]. Raising a child with a chronic health condition, such as CF, is associated with additional burdens for

caregivers, children and their families, including higher rates of mental health difficulties, less cohesive family functioning and greater demands on both caregiver and family time [3]. The management of CF is considered particularly burdensome given that much of the daily management is performed in the home by the primary caregiver. Caregivers need to ensure a high-fat, high-calorie diet, pancreatic enzyme replacement with every meal and snack, vitamin supplementation, regular antibiotics (oral and often inhaled) and airway clearance, such as chest physiotherapy, with time-consuming nebuliser therapy also often required [2, 4]. In addition, children require regular attendance at hospital clinic appointments. Caregivers can encounter problems in any of these areas in addition to other behaviour problems that are not specific to CF but nonetheless common in children.

Our previous work highlighted a high prevalence and persistence of CF-specific problems (i.e. physiotherapy adherence) as well as non-specific problems including behavioural sleep difficulties (e.g. refusing to go to bed, difficulty falling asleep), eating/mealtime problems and internalising behaviours (e.g. anxiety, depression, withdrawal) [5, 6]. How a family manages such problems may depend on a number of factors including the severity of the child's illness, the child's temperament and caregiver coping style and mental health.

The Role of Caregiver Coping In CF

Coping strategies are considered to be volitional behaviours, thoughts and feelings a person uses or experiences in relation to stressors [7, 8]. Active coping behaviours (described as problem focused, active or adaptive coping) and passive coping behaviours (described as avoidant or disengaged) have been well described in the literature, with particular coping strategies described as offering more or less adaptive ways of managing stressors [7]. Examples of active coping strategies in relation to CF include those behaviours that focus attention on the condition in an active and positive way, by being positive and optimistic, seeking information and support, monitoring child illness symptoms, cognitive restructuring of unhelpful thoughts and trust in treatment and in health professionals [3, 7, 9]. An example of adaptive coping strategies may include acceptance of the condition or related demands of the condition [10]. Examples of avoidant or passive coping strategies include those that divert attention away from the condition and related demands in an avoidant or distracting way, such as being pessimistic, denying the presence of the illness related demands or particular aspects of the illness that are too distressing, distraction, wishful thinking, self-blame, passivity or using numbing strategies, such as the use of substances [7, 9, 11].

In CF, regular use of active or adaptive coping strategies may allow caregivers to more consistently follow through with the required treatment regimen, thereby potentially avoiding problem behaviours related to CF management [4]. However, no studies have examined the relationship between caregiver coping strategies and CF-specific child problem behaviours. Rather they have focused on the relationship between caregiver coping and child and caregiver mental health, quality of life (QoL) and adjustment to the illness. For example, in a sample of children aged 5 to 12 years, positive coping strategies (as assessed by the Brief COPE) [12], such as seeking emotional support from family and friends, reduced the overall emotional impact of CF on the parent and were associated with a better overall sense of parental adjustment to the child's illness [11]. On the other hand, parent use of avoidant/ passive coping strategies such as 'self blame' for their child's CF was associated with higher reported parent and child mental health difficulties as well as higher emotional impact experienced by parents resulting from their child having CF. Frequent use of parental 'disengaging' strategies (i.e. giving up attempts to manage/deal with the stressors associated with CF) was associated with higher levels of parental symptoms of anxiety [11].

Over a broader age range, caregivers of children with CF aged one to 22 years who used passive coping strategies (as assessed by the Freiburg Questionnaire of Coping with Disease, see Staab et al.) [9], such as 'depressive coping', 'control of emotions' and 'retreat from social relations' reported their own health related quality of life (HRQoL) as poorer. Compliance, trust in treatment and self-encouragement strategies were positively related to caregiver HRQoL. Furthermore, caregiver coping explained more of the variance in HRQoL scores than their child's illness severity when measured both objectively and subjectively [9]. Other studies have found that maternal coping strategies may impact on child physical health status, such as lung function and body mass index (BMI) [4, 13]. In a group of 7 to 12-year-old children with mild pulmonary disease, maternal coping (as assessed by the Coping Health Inventory for Parents, CHIP) [14], predicted better child growth outcomes (BMI z scores) but not the child's CF related dietary adherence [15]. In longitudinal studies, infrequent caregiver use of avoidant/passive coping strategies has been associated with better adjustment to the child's illness [16], while more frequent use of active coping strategies has been associated with a positive trend in lung function over a 10-year period [4].

Given these studies, it is evident that aspects of active caregiver coping behaviours are associated with better parent and child adjustment to CF and caregiver HRQoL, and avoidant or passive coping strategies are associated with higher reported parent and child mental health difficulties. However, it remains unknown if caregiver coping strategies are associated with CF-specific and non-specific child

problem behaviours, such as behavioural sleep difficulties, eating/mealtime problems and physiotherapy adherence problems. It is possible that caregivers who engage in active coping may protect the health of their child by better managing the demands associated with treatment adherence, seeking extra support for care when needed and by responding faster to CF exacerbations rather than avoiding them. Alternatively, caregivers who engage in avoidant or externalised ways of coping, such as substance use, distraction , behavioural disengagement or internalising coping behaviours such as blaming themselves or wishful thinking, may feel overwhelmed managing the demands of their child's CF and may therefore be less effective in managing their child's treatment and non-treatment related problem behaviours. If there is a relationship between caregiver coping and child problem behaviours, then developing and implementing an intervention to improve caregiver coping may lead to better health outcomes for the child and their caregivers.

Measuring Coping

A number of measures of caregiver coping are available. We chose the Brief COPE [12] to assess caregiver coping in relation to the presence of child problem behaviours because of its brevity and previous use in the CF population. Additionally, principal components analysis can be performed on the Brief COPE which can: (a) reduce the number of coping strategies from the original 14 subscales into fewer dimensions and therefore reduce type I and II errors during analysis, and (b) assist in identifying clusters of coping behaviours to be targeted in an intervention. This approach to measuring coping has not been conducted in Australian caregivers of children with CF.

The aims of this study therefore were to: (a) investigate caregiver coping strategies using the Brief COPE [12] derived from the current multi-centre sample; (b) report the relationship between symptoms of caregiver mental health difficulties and the derived caregiver coping strategies; and (c) report the associations between caregiver coping strategies and child problem behaviours, specifically sleep, eating/mealtime behaviour, adherence with treatment (physiotherapy and electrolyte/enzyme supplements) and externalising and internalising behaviour problems, adjusting for potential confounders including caregiver mental health.

Methods

Participants

Between May and October 2010, Australian families from a multi-centre cohort, originally recruited in 2007 [6], were

surveyed. The original cohort included 117 caregivers of children with CF, attending outpatient clinics at either The Royal Children's Hospital, Melbourne (RCH), Victoria, Monash Medical Centre (MMC), Victoria (RCH and MMC provide care for all children with CF within Victoria), or Sydney Children's Hospital (SCH), New South Wales (NSW; SCH provides care for approximately 40 % of children with CF in NSW).

Recruitment and Consent

Caregivers were mailed an information statement and survey. Caregiver consent was implied upon return of the completed survey. This study was approved by the Human Research and Ethics Committee at each site: RCH (HREC29105A), MMC (09289B) and SCH (09/168).

Measures

Caregiver Coping

Caregiver coping strategies were measured using the 28 item Brief COPE [12]. Caregivers were asked to consider the extent to which they used each coping strategy to deal with the difficulties associated with raising their child with CF. Items were rated on a four-point Likert scale from 1 = 'I haven't been doing this at all' to 4 = 'I've been doing this a lot'. The Brief COPE derives 14 subscales, consisting of two items each, which were described as representing both potentially positive and negative strategies. The subscales are; self-distraction, active coping, denial, substance use, use of emotional support, use of instrumental support, behavioural disengagement, venting, positive reframing, planning, humour, acceptance, religion, and self-blame. Cronbach's alpha reliability coefficients of the Brief COPE subscales ranged from 0.50 to 0.90 [12]. Within the current sample the Cronbach's alpha reliability coefficients of the 14 Brief COPE subscales ranged from 0.14 to 0.93 (see Table 1).

Caregiver Mental Health

Caregiver mental health was measured using the 21-item Depression, Anxiety and Stress Scales (DASS) [17], a self-report questionnaire measuring the presence of emotional disturbance, relating to symptoms of depression, anxiety and stress. Higher scores indicate more severe symptoms of depression, anxiety or stress. The distributions of these continuous variables were positively skewed and Spearman's correlation coefficients were therefore used. For regression analysis, cut-points to indicate risk of depression (score > 9), anxiety (score >7) and stress (score > 14) were calculated (see Sheehan et al. [5] for prevalence data).

Table 1 Internal consistency coefficients and factorability of the 14 subscales of the Brief COPE within the current sample

Subscale of the Brief COPE	Sample size of respondents per subscale	Cronbach's alpha	Kaiser–Meyer–Olkin measure of sampling adequacy
1. Self-distraction (1) Turning to work or other activities to take my mind off things (19) Doing something to think about it less, such as going to movies, watching tv, reading, daydreaming, sleeping, or shopping	101	0.72	0.71
2. Active coping (2) Concentrating my efforts on doing something about the situation I'm in (7) Taking action to try to make the situation better	99	0.77	0.71
3. Denial (3) Saying to myself 'this isn't real' (8) Refusing to believe that it has happened	100	0.27	0.48
4. Substance use (4) Using alcohol or other drugs to make myself feel better (11) Using alcohol or other drugs to help me get through it	101	0.93	0.67
5. Use of emotional support (5) Getting emotional support from others. (15) Getting comfort and understanding from someone	96	0.79	0.71
6. Use of instrumental support (10) I've been getting help and advice from other people. (23) I've been trying to get advice or help from other people about what to do	101	0.74	0.72
7. Behavioural disengagement (6) Giving up trying to deal with it. (16) Giving up the attempt to cope	101	0.14	0.67
8. Venting (9) I've been saying things to let my unpleasant feelings escape (21) I've been expressing my negative feelings	101	0.58	0.78
9. Positive reframing (12) Trying to see it in a different light, to make it seem more positive (17) Looking for something good in what is happening	101	0.70	0.84
10. Planning (14) Trying to come up with a strategy about what to do (25) Thinking hard about what steps to take	100	0.77	0.72
11. Humour (18) I've been making jokes about it. (28) I've been making fun of the situation	101	0.70	0.49
12. Acceptance (20) Accepting the reality of the fact that it has happened (24) Learning to live with it	101	0.65	0.79
13. Religion (22) I've been trying to find comfort in my religion or spiritual beliefs. (27) I've been praying or meditating.	101	0.83	0.79
14. Self-blame (13) Criticizing myself (26) Blaming myself for things that happened	100	0.72	0.68
Overall values		0.79	0.72

Caregiver mental health problems were then dichotomised as '0' or '1', where '1' identified caregivers who scored above the cut point on any one of the depression, anxiety or stress subscales.

Child Behaviour

Primary caregivers rated several areas of child behaviour. Child's current sleep problems were rated as either 'no',

'mild', 'moderate' or 'severe' problem. Ratings were then dichotomised into no/mild or moderate/severe sleep problem [18]. Eating/mealtime problems were assessed using the Behavioral Pediatrics Feeding Assessment Scale (BPFAS) [19], with the frequency of problem eating/mealtime behaviours rated on a five-point scale from 1 = 'never' to 5 = 'always'. Caregivers also indicated if the behaviour was a problem (yes or no). Item ratings were summed to obtain the frequency score. The total number of problem behaviours was the problem score. The clinically significant frequency score and clinically significant problem score were defined as a score > 1.5 standard deviations (SD) above the normative mean. Problems with adherence to treatment regimens were assessed using three items that required caregivers to rate whether physiotherapy, electrolyte or enzyme treatment adherence was a problem on a seven-point scale, from 0 = 'no problem' to 6 = 'severe problem'. The scores for enzyme and electrolyte adherence were summed and reported together, as per the baseline reporting [6]. Results for each of physiotherapy and combined electrolyte/enzyme adherence were dichotomised into a positive score for a problem (≥ 2 and ≥ 4, respectively). Externalising and internalising behaviour problems were measured using the Child Behavior Checklist (CBCL; 1.5–5 years or 6–18 years depending on child age) [20, 21], a 99- or 112-item parent-report scale, respectively, that generates externalising and internalising behaviour problem subscale scores, based on the age form and gender (from 6 to 18 years only). The CBCL requires caregivers to rate the frequency of child behaviours over the previous two months (from 1.5 to 5 years) or 6 months (from 6 to 18 years) from 0 = 'not true' to 2 = 'very true or often true'. Summed scores were converted to T scores (mean = 50, SD = 10). Scores in the borderline/clinical range (T score ≥ 60) for externalising and internalising scales are reported. Scores within the borderline/clinical range may reflect behaviours that require clinical intervention/assistance, with children in borderline/clinical internalising and externalising ranges in the standardisation sample five to six times more likely, respectively, to be clinically referred than children with lower scores [21]. All prevalence data for child problem behaviours are reported elsewhere (see Sheehan et al.) [5].

Demographic Information

Caregivers reported child age and number of admissions to hospital in the past 12 months, primary caregiver age, marital status, education and employment status and partner age, education and employment status.

Statistical Analysis

For aim a, we used a principal components analysis to determine the coping components derived from the current

sample. For aim b, we used Spearman correlation coefficients to report correlations between symptoms of caregiver mental health difficulties and the derived coping components. For aim c, we conducted unadjusted and adjusted (child age, sex, number of admissions to hospital in the last 12 months, primary caregiver education level and mental health difficulties) logistic regression analysis with the derived caregiver coping components and each of the child problem behaviours. Odds ratios and 95 % confidence intervals are presented with p-values only for statistically significant results ($p \leq 0.05$). Analyses were conducted using Stata release 11.0 [22].

Results

Descriptive Statistics

Of the original 117 families, 1 child was excluded (no longer considered to have diagnosis of CF) and 102/116 (88 %) caregivers returned the survey. One caregiver did not complete the Brief COPE items; this family's data were excluded from the analysis. Responders did not differ from non-responders on parent education status, child age or child gender. All data reported in this paper are cross-sectional. Sample characteristics are presented in Table 2.

Factor Structure of the Coping Strategies

The factorability of the 28 Brief COPE [12] items within this study was assessed using the Kaiser–Meyer–Olkin measure of sampling adequacy (value=0.68), which indicated appropriate sampling adequacy (values of ≥ 0.6 are required for factor analysis) [23].

The factor structure of the Brief COPE [12] was identified by replicating Carver's [12] Principal Components Analysis (PCA), with oblique rotation on the 28 items. Ten components with eigenvalues greater than one were generated, which generally represented seven of Carver's 14 subscales (active coping, one item from planning, use of emotional support, one item from use of instrumental support, self-distraction, one item from venting and substance use). A scree plot, used to identify the point at which the amount of variance accounted for by each additional factor is relatively low (i.e. the elbow) [23], indicated a four-component solution. Based on the limited number of items (11) that would be retained using these two criteria, and the consideration that only seven of the original 14 subscales were replicated, we performed a second PCA on the Brief COPE [12] subscales, which is similar to the protocol performed by Benson [24].

Three subscales were not supported for inclusion in the PCA (denial, poor alpha reliability coefficient and poor

216

Int. J. Behav. Med. (2014) 21:211–220

Table 2 Demographic information

Variable	Sample size	Value
Child	101	
Male (n (%))		54 (53)
Age in years (mean (SD); (range))		5.5 (1.7); 3.1, 8.5
Hospital admissions in the previous year (n (%))		
0		66 (65)
1		23 (23)
2 or more		12 (12)
Primary caregiver	101	
Mother (n (%))		97 (96)
Father (n (%))		3 (3)
Grandparent (n (%))		1 (1)
Age in years (mean (SD); (range))		37.6 (5.3); 26.6, 61.6
Education status (n (%))		
Did not complete high school		26 (26)
Completed high school only		15 (15)
Completed tertiary studies		60 (59)
Employed (n (%))		58 (57)
Partner	94	
Age in years (mean (SD); (range))		38.9 (5.4); 30.2, 64.6
Education status (n (%))		
Did not complete high school		28 (30)
Completed high school only		16 (17)
Completed tertiary studies		50 (53)
Employed (n (%))		88 (94)

sampling adequacy; behavioural disengagement, poor alpha reliability coefficient and humour poor sampling adequacy). Removal of these subscales increased the Kaiser–Meyer–Olkin sampling adequacy from 0.72 to 0.76 and increased the overall reliability of the scale from 0.79 to 0.80 (individual subscale sampling adequacy values are reported in Table 1). Bartlett's test of sphericity for the remaining 11 subscales was significant (x^2 (65)=320.05, $p<0.0001$). As a result the three subscales were removed from the PCA, leaving 11 subscales for analysis.

The correlation matrix for the 11 subscales revealed numerous correlations in excess of 0.30 and several that were considerably higher, indicating that patterns in responses to the items were present. PCA with oblique rotation on the 11 subscales of the Brief COPE [12] generated three components with eigenvalues greater than one, based on item coefficients of ≥0.4 on one component and ≤0.4 on all other components. The third component, however, contained only two items and was therefore considered unstable [23]. Two components were supported by the scree plot further indicating the rejection of the third component. A forced two component PCA was then performed. The component loading coefficients of the final two derived components with items details are shown in Table 3.

The two retained components explained 49 % of the total variance. Component one (labelled 'proactive coping' strategies) contained four Brief COPE subscales (planning, active coping, positive reframing and acceptance), all of which reflect caregivers efforts to focus their attention on and address stressors associated with their child's CF in a proactive way. Component two (labelled 'avoidant coping' strategies), contained three Brief COPE subscales (self-distraction, substance use and self-blame), reflecting caregivers efforts to avoid, distract or focus their attention away from the CF related stressors, including using externalised coping strategies and internal focused strategies. A score for each of the coping components was then calculated by summing the scores for each subscale contributing to the component, for use in analysis.

Correlations between Caregiver Coping Strategies and Caregiver Mental Health Symptoms

Avoidant coping strategies were moderately correlated with caregiver depression, anxiety and stress (see Table 4). That is, parents who reported using more avoidant coping strategies were more likely to report symptoms of depression, anxiety and stress. Proactive coping strategies were weakly

Table 3 Forced two component PCA with oblique rotation on 11 subscales of the Brief COPE[a]

Variable	Component 1 Proactive coping	Component 2 Avoidant coping
Proportion of variance explained	26.9 %	21.8 %
Cronbach's alpha	0.83	0.71
Brief COPE subscale with items (item number in parenthesis)		
Planning	**0.47**	0.03
(14) Trying to come up with a strategy about what to do		
(25) Thinking hard about what steps to take		
Active coping	**0.44**	0.01
(2) Concentrating my efforts on doing something about the situation I'm in		
(7) Taking action to try to make the situation better		
Positive reframing	**0.40**	−0.00
(12) Trying to see it in a different light, to make it seem more positive		
(17) Looking for something good in what is happening		
12. Acceptance	**0.40**	−0.16
(20) Accepting the reality of the fact that it has happened		
(24) Learning to live with it		
Self-distraction	0.03	**0.48**
(1) Turning to work or other activities to take my mind off things		
(19) Doing something to think about it less, such as going to movies, watching TV, reading, daydreaming, sleeping, or shopping		
Substance use	−0.19	**0.46**
(4) Using alcohol or other drugs to make myself feel better		
(11) Using alcohol or other drugs to help me get through it		
Self-blame	−0.12	**0.42**
(13) Criticizing myself		
(26) Blaming myself for things that happened		

[a] Component loading coefficients are in bold

associated with caregiver stress. Given this, caregiver mental health was considered a potential confounder in the regression analyses.

Associations Between Caregiver Coping Strategies and Child Problem Behaviours

Unadjusted and adjusted logistic regressions for the two derived caregiver coping components and child behaviours are presented in Table 5. For each unit increase in caregiver

Table 4 Spearman's rank correlation coefficients between caregiver symptoms of mental health difficulties and the derived coping components[a]

Derived coping components	Caregiver mental health symptoms		
	Depression	Anxiety	Stress
Proactive coping	0.03	0.15	0.20*
Avoidant coping	0.52**	0.57**	0.55**

*p≤.05; **p<.001

[a] Sample size ranged from 98 to 100

use of avoidant coping strategies, the odds of children experiencing frequent eating/mealtime problem behaviours increased by 1.3 (adjusted 95 % CI 1.0 to 1.6, p=.03), and the odds of children experiencing borderline/clinical internalising behaviour problems increased by 1.3 (adjusted 95 % CI 1.1 to 1.7, p=.01). Caregiver use of proactive coping strategies was not associated with reduced odds of experiencing any of the child problem behaviours.

Discussion

This is the first study to investigate the factor structure of caregiver coping strategies, and the relationship between the derived coping strategies, caregiver mental health and the presence of child problem behaviours within a multicentre sample of Australian children with CF. Two main coping components were derived, which were labelled 'proactive' and 'avoidant' coping. Caregiver 'proactive' coping (planning, being actively involved, trying to see things differently and attempts at acceptance) was not associated with the child problem behaviours. 'Avoidant' coping (self-

Table 5 Unadjusted and adjusted logistic regression analyses of the relationship between child problem behaviours and the derived caregiver coping components[ab]

Child problem behaviour	Caregiver coping components based on the forced 2 factor solution of the Brief COPE subscales			
	Proactive coping		Avoidant coping	
	Unadjusted OR (95 % CI)	Adjusted OR (95 % CI)	Unadjusted OR (95 % CI)	Adjusted OR (95 % CI)
Moderate/severe sleep problem	1.0 (0.9, 1.1)	1.0 (0.9, 1.1)	1.0 (0.8, 1.2)	0.9 (0.7, 1.1)
Eating/mealtime problems (BPFAS)				
Significant frequency score	1.0 (0.9, 1.1)	1.0 (0.9, 1.1)	1.3** (1.1, 1.5)	1.3* (1.0, 1.6)
Significant problem score	1.0 (0.9, 1.1)	1.0 (0.9, 1.1)	1.1 (1.0, 1.3)	1.2 (1.0, 1.4)
Adherence				
Physiotherapy	1.0 (0.9, 1.1)	1.0 (0.9, 1.1)	1.1 (1.0, 1.3)	1.0 (0.9, 1.2)
Electrolytes/enzymes	1.0 (0.9, 1.2)	1.0 (0.8, 1.2)	1.1 (0.9, 1.4)	1.0 (0.7, 1.4)
CBCL				
Externalising				
Borderline/clinical (T score ≥ 60)	1.1 (0.9, 1.2)	1.1 (1.0, 1.2)	1.1 (0.9, 1.3)	1.0 (0.8, 1.3)
Internalising				
Borderline/clinical (T score ≥ 60)	1.1 (1.0, 1.2)	1.0 (0.9, 1.2)	1.3** (1.1, 1.5)	1.3* (1.1, 1.7)

BPFAS Behavioral Pediatrics Feeding Assessment Scale [19], *CBCL* Child Behavior Checklist [20, 21]

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

[a] Analysis adjusted for child age, sex, number of admissions to hospital in the last 12 months, primary caregiver education level and caregiver mental health difficulties

[b] Sample size ranged from 92–100 for unadjusted analysis and 91–99 for adjusted analysis

distraction, use of substances and self-blame) was associated with more frequent child eating/mealtime problem behaviours and borderline/clinical level internalising behaviours. Caregivers who used 'avoidant' coping strategies were more likely to report symptoms of depression, anxiety and stress.

The two coping components derived within the current study are similar to broader coping dimensions reported within the literature. For example the 'proactive' coping component reflected an active, monitoring, optimistic approach, which is considered within the coping literature to focus attention on the stressors (task/problem focused), and is likely to reflect a successful way of managing stressors [7]. The 'avoidant' coping component reflected an avoidant, passive-repressive approach, as well as encompassing a pattern of utilising external resources to deal with stressors. This approach is therefore less problem-focused and has been described in the literature to divert attention away from stressors in an unhelpful way [7].

Finding from the current study that avoidant coping is associated with child internalising problems in CF has also been observed by Wong and Heriot [11] who found that caregiver ratings on the behavioural disengagement and self-blame subscales of the Brief COPE [12] were significantly moderately correlated (−.48 and −.47, respectively) with child symptoms of anxiety and depression. Our findings that caregiver avoidant coping was associated with poorer caregiver mental health also echo those of Benson [24] who found that in parents of children with autism, higher use of maternal avoidant and distraction coping strategies was associated with increased caregiver distress.

The relationships between avoidant caregiver coping strategies and child problem behaviours in the current study were weaker than those between coping and caregiver mental health. This suggests that for caregivers of younger children, intervening in caregiver coping in order to improve child problem behaviours may best be targeted to parents

reporting avoidant coping. The relationship between avoidant coping strategies and poorer caregiver mental health was stronger, suggesting a role for intervention, focusing on both caregiver coping and mental health, in order to improve outcomes for caregivers. However, there is little literature to support intervening with caregiver coping in a whole of population approach, with intervention studies promoting strengthening of coping strategies unable to show improvements in caregiver coping, child outcomes or parental health beliefs [25, 26]. Additionally, developing an intervention for this population is not as simple as offering specific adaptive coping strategies as evidenced by Hodgkinson and Lester [3] where mothers who were instructed to try to compartmentalise their child's CF (appraisal-focused coping strategy) from other aspects of their life and functioning, feeling guilty and a sense of failure when they could not effectively use this strategy. Conversely, interventions targeting child behaviour directly have shown secondary gains in areas such as caregiver mental health in healthy populations [27], but it remains to be seen whether the same is true of the CF population.

This study has several strengths. First, we assessed a multi-centre sample of caregivers of children with CF, suggesting our findings may generalise to the wider population of children with CF in Australia in this age group. Second, we utilised a coping measure that is validated and has been used within the CF population. Third, by performing PCA and reporting coping in the context of the two derived coping components, we have reduced difficulties associated with power that performing multiple calculations using the original 14 subscales of the Brief COPE [12] would present.

Our study has some limitations. It was cross-sectional so we cannot infer causality. Data were collected by parent report which may impact both the reporting of child problem behaviours and the types of coping strategies endorsed. Diagnostic interviews to control for reporting of socially desirable responses could be considered in future studies. However, a number of caregivers reported 'negative' coping strategies and mental health symptoms, suggesting response bias for this outcome was low in our study. Additionally, in the absence of validated measures of adherence, we used study designed questions (e.g. for physiotherapy adherence). We cannot tell if parents who reported adherence to be a problem were reflecting overall family disorganisation or specific child opposition to therapy. However, the latter appears more likely as over 70 % of parents who reported a problem with physiotherapy cited lack of child compliance.

There are several implications for both clinical practice and research. For the multi-disciplinary teams who work with caregivers of children with CF, it is important to consider caregiver coping and mental health in the presence of child problem behaviours. Caregivers who report more frequent use of avoidant coping strategies may have children at risk of eating/mealtime or internalising behaviour problems. Supporting adaptive coping strategies that are consistent with the parent's beliefs or past positive practices may offer assistance for these caregivers [11]. Future research should include longitudinal studies to clarify the role of coping strategies in the development and persistence of child problem behaviours. Investigating the relationship between caregiver coping strategies and caregiver persistent mental health problems within this population would also be valuable. Finally, rigorous evaluation of interventions targeting only child problem behaviours versus interventions targeting problem behaviours and caregiver coping and mental health are required. Outcomes of such interventions will determine the best approach to managing child problem behaviours in CF.

In conclusion, there is evidence to support the implementation of an intervention targeting avoidant caregiver coping in order to improve some child problem behaviours in this age group. Intervention focusing on both caregiver coping and mental health may also improve outcomes for caregivers.

Conflict of interest The authors declare they have no competing interests in this research or its outcomes, or any relevant affiliations.

Author contributions The project was conceived, initiated and supervised by JS, MH and HH. JS led the project and the paper, supervised by MH and HH, with all authors providing critical contributions to reviewing, editing and approving its final version. JS conducted the analyses with the supervision of MH and HH. JS is the guarantor and accepts full responsibility for the conduct of the study, had access to the data, and controlled the decision to publish.

References

1. Massie RJ, Olsen M, Glazner J, Robertson CF, Francis I. Newborn screening for cystic fibrosis in Victoria: 10 years' experience (1989–1998). Med J Aust. 2000;172(12):584–7.
2. O'Sullivan BP, Freedman SD. Cystic fibrosis. Lancet. 2009;373 (9678):1891–904.
3. Hodgkinson R, Lester H. Stresses and coping strategies of mothers living with a child with cystic fibrosis: implications for nursing professionals. J Adv Nurs. 2002;39(4):377–83.

4. Patterson JM, Budd J, Goetz D, Warwick WJ. Family correlates of a 10-year pulmonary health trend in cystic fibrosis. Pediatrics. 1993;91(2):383–9.

5. Sheehan J, Massie J, Hay M, Jaffe A, Glazner J, Armstrong D, et al. The natural history and predictors of persistent problem behaviours in cystic fibrosis: A multicentre, prospective study. Archives of Disease in Childhood. 2012; doi:10.1136/archdischild-2011-301527.

6. Ward C, Massie J, Glazner J, Sheehan J, Canterford L, Armstrong D, et al. Problem behaviours and parenting in preschool children with cystic fibrosis. Arch Dis Child. 2009;94(5):341–7.

7. Abbott J. Coping with cystic fibrosis. J R Soc Med. 2003;96 Suppl 43:42–50.

8. Lazarus RS, Folkman S. Stress, appraisal and coping. New York, USA: Springer; 1984.

9. Staab D, Wenninger K, Gebert N, Rupprath K, Bisson S, Trettin M, et al. Quality of life in patients with cystic fibrosis and their parents: what is important besides disease severity? Thorax. 1998;53(9):727–31.

10. Compas BE, Jaser SS, Dunn MJ, Rodriguez EM. Coping with chronic illness in childhood and adolescence. Annu Rev Clin Psychol. 2012;8:455–80.

11. Wong MG, Heriot SA. Parents of children with cystic fibrosis: how they hope, cope and despair. Child Care Health Dev. 2008;34 (3):344–54.

12. Carver CS. You want to measure coping but your protocol's too long: consider the brief COPE. Int J Behav Med. 1997;4:92–100.

13. Patterson JM, McCubbin HI, Warwick WJ. The impact of family functioning on health changes in children with cystic fibrosis. Soc Sci Med. 1990;31(2):159–64.

14. McCubbin HI. CHIP–coping health inventory for parents: an assessment of parental coping patterns in the care of the chronically ill child. J Marriage Fam. 1983;45(2):359–70.

15. Anthony H, Paxton S, Bines J, Phelan P. Psychosocial predictors of adherence to nutritional recommendations and growth outcomes in children with cystic fibrosis. J Psychosom Res. 1999;47(6):623–34.

16. Thompson RJJ, Gustafson KE, Gil KM, Kinney TR, Spock A. Change in the psychological adjustment of children with cystic fibrosis or sickle cell disease and their mothers. J Clin Psychol Med S. 1999;6(4):373–92.

17. Lovibond SH, Lovibond PF. Manual for the depression anxiety stress scales. 2nd ed. Sydney: Psychology Foundation; 1995.

18. Martin J, Hiscock H, Hardy P, Davey B, Wake M. Adverse associations of infant and child sleep problems and parent health: an australian population study. Pediatrics. 2007;119(5):947–55.

19. Crist W, McDonnell P, Beck M, Gillespie CT. Behavior at mealtimes and the young child with cystic fibrosis. J Dev Behavl Pediatr. 1994;15(3):157–61.

20. Achenbach TM, Rescorla LA. Manual for the ASEBA preschool forms & profiles. Burlington VT: University of Vermont, Research Center for Children, Youth & Families; 2000.

21. Achenbach TM, Rescorla LA. Manual for the ASEBA school-age forms & profiles. Burlington, VT: University of Vermont Research Center for Children, Youth, & Families; 2001.

22. StataCorp. Stata statistical software: Release 11. College Station, TX: StataCorp LP. 2009.

23. Tabachnick BG, Fidell LS. Using multivariate statistics (Fifth Edition). United States of America: Pearson Education; 2007.

24. Benson PR. Coping, distress, and well-being in mothers of children with autism. Res Autism Spect Dis. 2010;4(2):217–28.

25. Goldbeck L, Babka C. Development and evaluation of a multifamily psychoeducational program for cystic fibrosis. Patient Educ Couns. 2001;44(2):187–92.

26. Klebanov PK, BrooksGunn J, McCormick MC. Maternal coping strategies and emotional distress: results of an early intervention program for low birth weight young children. Dev Psychol. 2001;37(5):654–67.

27. Hiscock H, Bayer J, Gold L, Hampton A, Ukoumunne OC, Wake M. Improving infant sleep and maternal mental health: a cluster randomised trial. Arch Dis Child. 2007;92(11):952–8.