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UNIVERSITY OF COPENHAGEN



PHD THESIS · 2014  
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# Towards a Better Understanding of Caregiver Distress in First-Episode Psychosis

A Study of Psychological Factors

# Towards a Better Understanding of Caregiver Distress in First-Episode Psychosis: A Study of Psychological Factors



REGION SJÆLLAND  
PSYKIATRIEN  
*-vi er til for dig*

PhD thesis by Jens Einar Jansen

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**Towards a better understanding of caregiver distress in first-episode  
Psychosis: A study of psychological factors**

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“Schizophrenia cannot be understood without understanding despair.”

– R.D. Laing

“Nobody realizes that some people expend tremendous energy merely to be normal.”

– Albert Camus



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## **Preface**

This PhD-thesis was carried out at the Early Psychosis Intervention Center, Region Zealand, Denmark. The study described here is part of a larger research project seeking to compare single-family treatment with multi-family treatment for caregivers of persons with first-episode psychosis. The Region Zealand Health Scientific Research Foundation provided funding for this research. The research was carried out in close collaboration with colleagues in the Early Psychosis Intervention Center as well as the clinical staff working in Opus, Region Zealand.

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The thesis is dedicated to my father, Jens Arne Jansen.

Jens Einar Jansen  
Copenhagen, June 2014

## Guide to reading the thesis

This PhD study comprises three empirical papers and a systematic review that together examine how various psychological variables are associated with caregiver distress in first-episode psychosis. The specific objectives are described in the next section.

The first chapter contextualises the current study by describing key concepts and theories. First, a contemporary understanding of schizophrenia and psychosis is presented, which is in stark contrast to the traditional, more pessimistic view of the illness. This highlights the early intervention movement, to which Early Psychosis Intervention Center and Region Zealand Psychiatry has been an important contributor, especially through the TIPS (Early Treatment and Intervention in psychosis) project. Second, the literature on family involvement and its influence on the course of illness is briefly reviewed. The concept of 'expressed emotion', which has been a cornerstone of family research, is being critically evaluated, followed by a presentation of psychological variables found to relate to caregiver distress. Finally, the concept of metacognition is defined and discussed, with the work of Adrian Wells and Paul Lysaker as central points of reference. As these ideas have not been examined in relation to caregiver distress before, the aim of this section is to describe their relevance to the current study.

The second chapter describes the design and methodological considerations for the study, including inclusion and exclusion criteria, procedure, assessment instruments and statistical analyses. The third chapter presents the results from the three empirical studies and the systematic review. The fourth chapter discusses the main findings and is divided thematically according to the four papers, namely the systematic review, subjective appraisal, expressed emotion, metacognitive capacity and maladaptive metacognitions. Clinical implications are discussed in chapter five, strengths and limitations of the study in chapter six, and implications for future research in chapter seven. Finally, chapter eight summarise the study in English and Danish, followed by the conclusion in chapter nine.

## Objectives

The overall goal of this PhD was to get a better understanding of what and how psychological factors relate to caregiver distress in first-episode psychosis. With this in mind, the specific aims were to: 1) systematically review the current literature that has examined psychological factors accounting for caregiver distress; 2) examine subjective appraisals and emotional over-involvement as predictors of caregiver distress, while addressing some of the methodological limitations in earlier studies; and 3) examine the concept of metacognition from two different theoretical perspectives as predictors of caregiver distress. The specific study hypotheses are presented in the method section.

## List of papers

### Paper 1

Jansen, J. E., Gleeson, J., Cotton, S. (2014). Towards a better understanding of caregiver distress in first-episode psychosis: A systematic review of the psychological factors involved. In review

### Paper 2

Jansen, J. E., Haahr, U. H., Harder, S., Trauelsen, A. M., Lyse, H. G., Pedersen, M. B., Simonsen, E. (2014). Caregiver distress in first-episode psychosis: the role of subjective appraisal, over-involvement and symptomatology. *Social Psychiatry and Psychiatric Epidemiology*.

### Paper 3

Jansen, J. E., Lysaker, P. H., Harder, S., Haahr, U. H., Lyse, H. G., Pedersen, M. B., Trauelsen, A. M., Simonsen, E. (2013). Positive and negative caregiver experiences in first-episode psychosis: Emotional overinvolvement, wellbeing and metacognition. *Psychology and Psychotherapy. Theory, Research and Practice*.

### Paper 4

Jansen, J. E., Harder, S., Haahr, U. H., Lyse, H. G., Pedersen, M. B., Trauelsen, A. M., Simonsen, E. (2014). The role of metacognitions in expressed emotion and distress: a study on caregivers of persons with first-episode psychosis. *Clinical Psychology and Psychotherapy*.

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## List of abbreviations

ACT	Acceptance and Commitment Therapy
CAS	Cognitive-Attentional Syndrome
CBT	Cognitive Behaviour Therapy
CC	Critical comments
DBT	Dialectic Behaviour Therapy
DUP	Duration of Untreated Psychosis
ECI	Experience of Caregiving Inventory
EOI	Emotional over-involvement
EE	Expressed emotion
FQ	Family Questionnaire
FEP	First-episode psychosis
GAF	Global Assessment of Functioning scale
GHQ-30	General Health Questionnaire, 30 item version

ICC	Intra Class Correlation
ICD-10	International Classification of Disease, 10 <sup>th</sup> ed.
IPII	Indiana Psychiatric Illness Interview
MBCT	Mindfulness-based Cognitive Therapy
MAS-A	Metacognitive Assessment Scale – Abbreviated
MCQ-30	Metacognitions Questionnaire
MCT	Metacognitive Therapy
MERIT	Metacognitive Reflective Insight Therapy
PANSS	Positive and Negative Syndrome Scale for Schizophrenia
PTSD	Post-traumatic stress disorder
S-REF	Self-Regulatory Executive Function theory
SPSS	Statistical Package for the Social Sciences
TOM	Theory of Mind

# 1. Background

## 1.1. Introduction

Nonprofessional caregivers, usually family members, often play an important role in the recovery of persons with first-episode psychosis (FEP). They often provide care, act as advocates, collaborate with treatment team members and generally attend to many needs that are unmet by the psychiatric services (Glynn, 2012; Onwumere et al., 2011a; Shor and Birnbaum, 2012). Supportive families have been found to enhance recovery, and seem to reduce both duration of untreated psychosis and compulsive treatment, possibly by ensuring timely access to relevant services (Kuipers and Bebbington, 1985; Morgan et al., 2006, 2005; O'Brien et al., 2006). For this reason, family- involvement and collaboration is increasingly recognised as a central aspect of treatment of persons with FEP (Agius et al., 2010).

Caregivers are most often parents or partners, and although there are various degrees of acceptance of the role, caring for someone with psychosis is generally not a matter of choice – it is often impossible just to walk away from (Kuipers et al., 2010). However, this is not to say that the relationship can not be characterised as reciprocal and inter-dependent, as shown by Horwitz (1996). The relationship with close relatives or partners can influence the course of illness in both positive and negative ways. The course of illness can in turn influence caregivers, who often report high levels of distress and burden. This compromises their ability to support the person with psychosis, sometimes even making the illness worse. For this reason service systems must aim at improving recovery in the whole family, not just in the service user. There has also been an increased recognition of caregivers' own needs, leading experts in the field to raise the question as to whether it is "time for a separate psychosis caregiver service." (Kuipers, 2010 p. 401).

However, most studies have examined distress and burden in more chronic forms of illness and many of the interventions have been developed within this context. For this reason, a number of authors have addressed the need for developing interventions more specifically tailored to FEP families (Gleeson et al., 1999; Linszen et al., 1996). One challenge has been the limited understanding of caregiver distress, especially its psychological underpinnings, from which to build a broader base of interventions.

With this in mind, the first part of the thesis will discuss the current view of psychosis and schizophrenia, focusing on the course of illness and its treatment. This is followed by a brief review of the research on caregiver distress and family work. Finally, the concept of metacognition will be presented as one potential area for developing new interventions suitable for families with a member suffering from FEP.

## 1.2. Schizophrenia: from pessimism to a more optimistic outlook

Contrary to traditional beliefs about persons with schizophrenia, many can achieve recovery or significant improvement over time (Lysaker and Buck, 2008). The rather pessimistic outlook is often attributed to Emil Kraepelin (1919) who saw it as an inherent degenerative disorder starting in adolescence or young adulthood. For this reason Kraepelin coined the term “dementia praecox” or “premature dementia”. However, progressive deterioration may be a hallmark for some, but it is more of an exception than a rule (Harding et al., 1992).

Recently, a more positive view has been adopted by clinicians and researchers, a view which is based on a range of evidence. First, there has been a series of long-term studies demonstrating a very good outcome in a large number of individuals diagnosed with schizophrenia (Jobe and Harrow, 2005). Second, a growing consumer movement among people with schizophrenia have revised our understanding of what recovery might look like. According to this view, *subjective* areas of recovery such as ratings of meaningfulness, self-esteem and quality of life, are just as important as, and not necessarily correlated with, *objective* domains of recovery such as symptom remission, employment and leisure time activities (Bellack, 2006; Deegan, 2001; Liberman and Kopelowicz, 2002; Lysaker and Buck, 2008). Complete recovery is possible for a number of people and the course of the disorder is more fluid and malleable than depicted earlier (Davidson et al., 2008; Lysaker and Buck, 2008). Finally, within the Early Intervention paradigm starting in the 1990s, new specialised clinical services have been established, first in Melbourne (McGorry et al., 1996) and soon after in many key locations in the UK, Europe, North America and Asia (McGorry et al., 2008). This has resulted in a resurgence of research interest, new evidence and widespread international efforts to reform services and treatment approaches (Killackey and Yung, 2007).

Some of the central aspects of the movement have been early detection of persons with psychosis and implementation of evidence-based treatment (Jeppesen et al., 2005a; McGorry et al., 1996; Petersen et al., 2005a). An important finding has been that reducing the period of untreated psychosis is possible through information campaigns and specialised services, and that this reduction improves long-term outcome for persons diagnosed with schizophrenia (Friis et al., 2005; Hegelstad et al., 2012; Marshall et al., 2005). Schizophrenia typically emerges during the sensitive developmental period of adolescence and emerging adulthood, and it complicates developmental conditions such as identity, independence, sexuality, intimate relationships, study and career plans (McGorry et al., 2006). This highlights the importance of an understanding family, as well as the added challenges caregivers experience in supporting their young adult through this transitional phase.

After the first onset of psychosis, there are often a number of social risk factors such as disruption of peer and family networks and vocational discontinuation, and psychological risk factors such as comorbid depression and anxiety, substance use, personality dysfunction and post-traumatic stress disorder (PTSD) (McGorry et al., 2008; Salokangas and McGlashan, 2008). These risk factors can have a detrimental influence on the development of the personality of a young person and they probably constitute the main reasons for the deterioration seen in schizophrenia. However, as pointed out by Birchwood and Fiorillo (2000), the first five years after onset of psychosis can be characterised as a 'critical period' in which the disorder is more malleable by psychosocial interventions to improve recovery than in later chronic states. In other words, the critical period is a 'window of opportunity' to preventing avoidable deroute and promoting recovery.

### *1.3. Family involvement in treatment*

Engaging and supporting the family is a central part of the early intervention service for psychosis and the active participation of the family is an invaluable resource for clinicians (Jeppesen et al., 2005b). Studies looking at family environments have found that positive family interactions can improve the course of illness (González-Pinto et al., 2011; López et al., 2004; O'Brien et al., 2006). According to Addington et al. (2007), family members of persons with psychosis also are taking an increasingly active and participating role in the care of their relative. In fact, there seems to be a development over the last forty years, of nonprofessional caregivers assuming steadily greater responsibility for their adult family member (Dyck et al., 1999).

However, there has also been a long tradition in psychosocial research to examine how family members and particularly the emotional environment in the family can negatively affect the course of illness in persons with schizophrenia. The concept of expressed emotion (EE) has been a cornerstone in family research since it was first formulated by Brown and colleagues (1972, 1962). High EE refers to relatives being critical (making negative comments about the behaviour or characteristics of the patients), hostile (making very general criticism or reject the patient) or emotionally over-involved (showing an exaggerated emotional response, over-intrusive or self-sacrificing behaviour, and/or over-identify with the patient). More recently, these three categories are often reduced, and EE commonly refers to relatives making excessive critical comments (CC) and/or being emotionally over-involved (EOI).

The historical backdrop was some surprising findings that, when the big mental institutions closed in the 1960s, patients that went to live with their parental or matrimonial home relapsed significantly more than the patients who went on



to live either in lodgings or with siblings. A recurring finding was that persons diagnosed with schizophrenia living in families in which there were high levels of expressed emotion, had significantly more relapses compared to persons with schizophrenia living in families with low levels of expressed emotion (Vaughn and Leff, 1976). These findings have been replicated in later studies and supported by meta-analyses (Bebbington and Kuipers, 1994; Butzlaff and Hooley, 1998; Kavanagh, 1992; Kuipers et al., 2006; Nuechterlein et al., 1992).

While it is not known how EE leads to relapse, most theories are based on the stress-vulnerability model (Zubin and Spring, 1977). From this view interactions with high-EE individuals are stressful for patients, rendering them vulnerable to relapse. According to Kuipers et al. (2006), family environments increase relapse rates via affect, which was supported by their finding that patients living with high EE parents had higher levels of anxiety and depression and lower self-esteem compared to patients living with carers that showed low EE. Related to this, Tarrier et al. (1988a) used electrodermal measures and found that subjects showed significantly higher frequencies of nonspecific skin-conductance response with a high-EE relative present than subjects with a low-EE relative present.

According to Breitborde et al. (2009b), the conceptual foundation of EE is underdeveloped. The model of EE described in the early writings (e.g., Leff & Vaughn 1980) tended to view EE in a “trait-like manner” (Barrowclough and Hooley, 2003, p. 850). However, the level of EE has also been found to change during the course of illness (Patterson et al., 2000), leading some authors to call for a developmental perspective towards EE that goes beyond simple cause-effect dichotomies (Smith et al., 1993a). McFarlane and Cook (2007) found EE to be significantly higher in a first-episode sample compared to an at-risk group, suggesting EE to be a reaction to deterioration of function in the patient rather than a trait. It has also been argued that the two components of EE are related to different factors. Schreiber et al. (1995) found EOI and warmth to be more strongly related to the state of the patient, and expression of CC to be the same for both ill and well siblings, thus suggesting that it represents a parental trait. Alvarez-Jiménez et al. (2010) found that EOI was more related to caregiver distress, while CC was more related to duration of untreated illness. One way to look at EE is as a ‘snapshot’ of the state of on-going transaction in which “stress, burden, coping, negative attributions of behaviour and unresolved grief are the key elements” (Smith et al., 1993b, p. 15). In summary, while there are a number of factors found to predict EE, there is, as pointed out by Breitborde et al. (2009b), a lack of an overarching psychological conceptualisation.

#### *1.4. Family work and relapse prevention*

The attention to family factors in EE research has been closely linked to the development of family interventions (Dixon et al., 2001) and a number of studies and meta-analyses have found family involvement to be effective in preventing relapse (Falloon, 1985; Hogarty, 1986; McFarlane et al., 1995; Pfammatter et al., 2006; Pharoah et al., 2001; Pilling et al., 2002; Pitschel-Walz et al., 2001).

There is a range of structured family intervention programmes, but most of them are based on broad psycho-educational and cognitive behavioural therapy approaches (Askey et al., 2007). The interventions typically consist of the following components in various combinations: improving communication, information about psychosis, problem-solving, reducing criticism and conflict, improving activity, and (sometimes) the emotional processing of grief, loss and anger (Kuipers et al., 2002; McFarlane and Anderson, 2011; Tarrier and Barrowclough, 1990).

However, while the association between EE and relapse is considered to be robust in more chronic forms of illness, the findings are less conclusive in the early course (Bird et al., 2010). In fact, a number of studies have not found this association in FEP samples (MacMillan et al., 1986; Stirling et al., 1991). Some authors argue that too much focus on EE might even cause harm in FEP families, and that some of the family approaches developed for long-term service users are not congruent with the hope and recovery advocated by the early intervention paradigm (Birchwood and Fiorillo, 2000; Gleeson et al., 1999; Linszen et al., 1996). Some of the arguments have been that EE interventions may interfere with the grief process, that “adverse” labelling effects, e.g. addressing communications skills may be stigmatising, and finally that high EE is being used as a criterion for receiving family intervention rather than the support being “needs led” (Linszen and Birchwood, 2000).

#### *1.5. Caregiver distress*

Caring for someone with a mental illness often involves both positive and negative feelings (Szmukler, 1996). Many caregivers report positive personal transformations and describe rewarding experiences such as clarity about their priorities in life, ‘inner strength’, greater closeness in the family, and being more sensitive to other people (Chen and Greenberg, 2004; Greenberg, 1995; Kulhara et al., 2012). However, many caregivers also experience high levels of burden and distress, including anxiety, depression, stigma, social isolation, increased family conflict and economic strain (Awad and Voruganti, 2008; Barrowclough et al., 1996; Sczufca and Kuipers, 1996; Schene et al., 1998; Treasure et al., 2001). Some studies have found that as many as a third of caregivers meet criteria for post-traumatic stress disorder (Barton and Jackson, 2008). Martens and Adding-

ton (2001) also found that relatives of persons with first-episode psychosis were more distressed than relatives of persons with a more chronic course of illness.

While earlier studies of caregiver distress examined various 'objective' causes of distress such as patients' symptomatology, reduced personal freedom and financial concerns, most recent studies have adopted a stress-appraisal-coping framework (Joyce et al., 2003; Raune et al., 2004; Szumukler et al., 1996). Based on Lazarus and Folkman's (1984) model, the illness and its impact are stressors appraised by caregivers. Primary appraisal involves an assessment of the event's relevance or impact and secondary appraisal involves an evaluation of coping options. Lazarus and Folkman describe two types of coping, namely "problem-focused" and "emotion-focused". Problem-focused coping involves facing the problem and attempts to deal with it through planning, problem-solving and seeking advice and support. Emotion-focused coping involves trying to diminish the negative emotional impact by seeking emotional support or by behavioural or cognitive avoidance (e.g. pretending it doesn't exist, substance abuse, or not wanting to talk about the problem). Emotion-focused strategies tend to be used when stressor are appraised as exceeding one's coping resources. This increases negative emotional states such as distress, depression and anxiety.

Within the stress-appraisal-coping-framework, a number of factors relating to caregiver distress have been examined. First, caregivers' appraisal of the impact of illness has been found to be related to the level of distress in caregivers, and symptomatology is often of less importance to the level of caregiver distress in the early stages of illness (Addington et al., 2005). Second, avoidant coping has been associated with higher levels of caregiver distress in psychosis (Cotton et al., 2013; Onwumere et al., 2011b). Third, studies have examined how distress is related to caregivers' attributions, or appraisals of whether patients can control their symptoms and behaviour (Barrowclough and Hooley, 2003; Kuipers et al., 2010; Lobban et al., 2006; Peterson et al., 2004). Hooley et al. (2002) found that relatives with high EE attribute more personal control over the illness to the patient, and are themselves more behaviourally controlling than relatives with low EE. Finally, while EE has been shown to have less value in predicting relapse in FEP, it has been found to be relevant in predicting caregiver distress. A number of studies have shown that EE, especially EOI, is linked to caregiver burden and distress, (Boye et al., 1998; Patterson et al., 2005). In a prospective study, Breitborde et al. (2009a) also found baseline EOI to predict long-term distress in caregivers. In Boye and colleagues' (1998) view, emotional over-involvement can be seen as an analogue to a 'stress syndrome' (p. 493), arguing that EOI might be a manifestation of unhealthy ways of coping with trauma.

Taken together, recent models of caregiving in psychosis and schizophrenia, are framed within a stress-appraisal-coping model, in which caregivers' appraisals of the impact of illness and their coping seem to be of central concern

to their level of distress. Furthermore, the concept of EE, particularly EOI, seems associated with distress. However, few studies have examined families of persons with FEP and little is known about the psychological underpinnings of caregiver distress in FEP (Addington et al., 2005; Gleeson et al., 2010). Moreover, while appraisal and coping seem important in caregivers' overall experience of the caregiving experience, little is known about what factors within caregivers are involved in appraisal and caregiver distress. According to the stress-appraisal-coping model, personality characteristics and style influence appraisal and coping and may constitute a mediating factor of distress (Folkman and Moskowitz, 2000; Folkman, 1997; Szmukler et al., 1996). Few studies have examined the psychological factors within caregivers in FEP and the question as to why some caregivers are more distressed than others remains largely unanswered. In the following, the concept of metacognition will be described as one potential new approach to further our understanding.

### *1.6. Thinking about thinking*

One way to encompass the psychological factors associated with caregiver distress and the subjective view of caregiver experiences is through the concept of metacognition. Metacognition, which can be described as thinking about thinking, originally evolved within the tradition of cognitive psychology. The first appearance of the concept is often identified as Flavell's (1979) work on education, referring to the awareness of which conditions best enabled learning. In psychotherapy literature, there are various ways in which the concept has been used, and two of the most extensively researched approaches will be examined in this thesis. The first is Lysaker et al.'s (2013) and Semerari et al.'s (2003) use of the concept, which views metacognition as a broad cognitive process in which information is integrated into representations of self and others in various degrees of complexity and integration. The second is Wells et al. (2008) that describes a set of maladaptive metacognitive beliefs involved in the control, monitoring and regulation of attention. These metacognitions activate or maintain repetitive thought patterns (rumination and worrying), threat monitoring (hypervigilance) and unhelpful coping, which have been found important to a number of psychological problems. In the following these two approaches will be described in more detail.

In contrast to the cognitive model of caregiving, by which much family work is inspired, these metacognitive models are less focused on the *content* of cognition – that is, the appraisal of the challenging situation. Rather, they understand distress as influenced by the way thinking processes are controlled and the style they take (Wells, 2008) or by the capacity to make a comprehensive understanding of the situation.

### *1.7. Metacognitive capacity*

This concept of metacognition has been defined as “a spectrum of activities which involve thinking about thinking, ranging from the consideration of discrete thoughts and feelings to the synthesis of discrete perceptions into an integrated representation of self and others” (Lysaker et al., 2014, p. 2). Rooted in developmental literature and literature on inter-subjectivity, it has an interest in how humans form coherent ideas of self and others in close relationships with other people (Lysaker et al., 2012a; Meins et al., 2002). Metacognition parallels a number of related concepts such as Theory Of Mind (Brüne, 2005), mentalisation (Fonagy et al., 2011) and some aspects of social cognition (Penn et al., 2008).

The capacity to reflect upon one’s thinking has been found relevant to a range of disorders such as personality disorders (Bateman and Fonagy, 2004; Dimaggio et al., 2007), eating disorders (Skårderud, 2007) and psychosomatic conditions (Vanheule et al., 2011). Thus metacognition can be used as a conceptual framework for developing therapeutic interventions in a number of disorders and can therefore be viewed as a trans-diagnostic concept (Allen and Fonagy, 2006; Fonagy et al., 2011; Lysaker and Dimaggio, 2014). Psychotherapy is a setting, in which metacognitive abilities such as self-reflectivity, understanding of others and emotional problem solving can be developed through interventions adjusted to the persons current level of these skills (Carcione et al., 2008; Harder and Folke, 2012; Lysaker and Dimaggio, 2014). Thus, treatment may help a person construct more sophisticated explanations for their psychological distress and therefore better be able to deal with emotional difficulties.

A number of studies have suggested that many with FEP or schizophrenia experience diminished metacognitive capacity, including Self-reflectivity; Awareness of others’ thoughts and feelings; Decentration, or the ability to understand others as independent agents; and Mastery, or the ability to use metacognitive knowledge to handle psychological challenges (Lysaker et al., 2013). According to Lysaker and colleagues, this can have important clinical implications. First, at the most elemental level of self-awareness, studies have found persons with schizophrenia to have difficulties in fully experiencing themselves as agents who are the cause of their own behaviour (Mishara et al., 2013). With this limited ability to experience ownership of one’s actions and experiences, the likelihood of having delusions is greater – i.e. there may be a tendency to interpret these as emanating from another source than oneself. Second, a reduced capacity for metacognition may also complicate the process of constructing a meaningful and coherent narrative in which past and present are integrated. This further complicates the process of making realistic plans for the future, including adjusting one’s behaviour to the constraints of current difficulties (Lysaker et al., 2005). Third, a larger sense of personal identity and the ability to solve problems related to work and personal relationships may be compromised,

which has implication for the prospects of personal recovery (Lysaker et al., 2013). Finally, associations have been found between metacognition and a number of important outcomes such as work performance (Lysaker et al., 2010a), social functioning (Lysaker et al., 2010b), current and prospective symptom levels (Hamm et al., 2012) and self reported therapy alliance in Cognitive Behaviour Therapy (CBT) (Davis et al., 2011). These difficulties are related to, but are not synonymous with, neurocognitive deficits, and they can be considered as semi-independent phenomena (Lysaker et al., 2005). Taken together, the ability to form complex accounts of self and others, and to use this to solve social and emotional problems, seems to be related to distress and well-being in a number of emotional disorders.

However, little is known about how the concept of metacognition relates to distress and well-being in a non-clinical population, that is, in a population in which one would not expect to find metacognitive deficits, but rather variations within a 'normal range'. The closest is a study by Lysaker and colleagues, assessing the level of metacognition in patients with HIV (Lysaker et al., 2012b). But as metacognition represents a trans-diagnostic framework, incorporating concepts like self-reflection, empathy and emotional understanding, which have been found relevant to a range of psychological difficulties, the idea being that it may also further the understanding of caregiver distress. Also, as metacognitive capacity is at the same time both relatively stable and also dynamic and malleable (Lysaker et al., 2007), it may especially inform a psychological understanding of how caregivers form subjective appraisals. To the best of this author's knowledge, the current study is the first attempt to use a metacognitive framework in the context of caregiver distress.

### *1.8. Maladaptive metacognitions*

In contrast to the concept of metacognition above, Wells' (2008) metacognitive model is not concerned with how people form broad representations of self and others, but rather with the way people assess, monitor and control their thinking and consciousness. Drawing on the seminal work of Wegner and colleagues (2000), a central concern is our general unwillingness to accept intrusive, unwanted or uncomfortable thoughts and emotions. This often leads to attempts to suppress or avoid them, which according to Wegner et al., leads to the opposite – namely increasing them. Another central concern is rumination (Nolen-Hoeksema et al., 2008) and worry (Borkovec et al., 1998) which are forms of recurrent negative thinking about past or future events that are implicated in the maintenance of depression and anxiety. As with suppression, rumination and worry are considered as an understandable although maladaptive coping strate-

gy, which often backfires and cause yet more psychological distress (Wells and Matthews, 1996; Wells, 2010).

Wells' theoretical foundation is the Self-Regulatory Executive Function theory (S-REF; Wells and Matthews, 1996). Within the S-REF model, psychological distress is maintained and increased by the cognitive-attentional syndrome (CAS; Wells, 2000), consisting of a range of unhelpful coping strategies such as perseverative thinking (e.g. rumination and worrying), threat monitoring, avoidance and thought suppression. The CAS makes persons less able to modify maladaptive self-beliefs and increases the accessibility of negative information about the self. According to Wells' model, a specific set of metacognitions underlies the CAS and can be described as follows: (1) positive beliefs about worry, or the degree to which a person finds perseverative thinking useful; (2) negative beliefs about worry concerning uncontrollability and danger, or the degree to which a person finds perseverative thinking overwhelming and dangerous; (3) cognitive confidence, or a person's reliance on their attention and memory; (4) beliefs about the need to control thoughts; and (5) cognitive self-consciousness, or the tendency to monitor one's thoughts and focus one's attention inwards. Beliefs of a metacognitive nature are of central concern as they guide the cognitive system and leads to styles of thinking that facilitate or impede emotional processing (Wells and Sembi, 2004a).

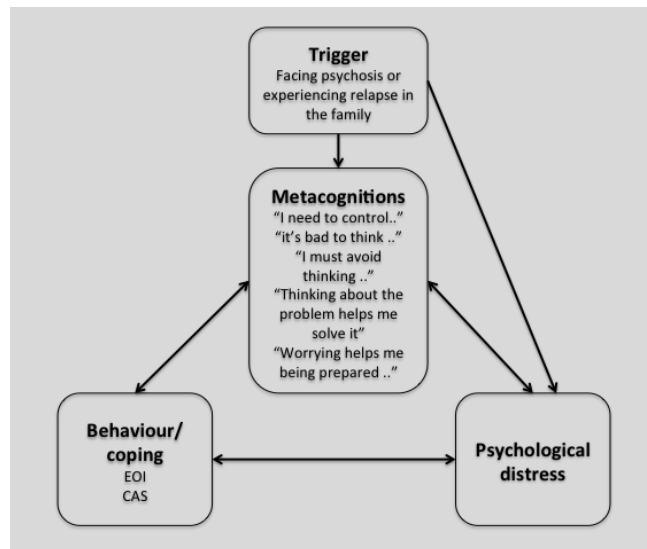
This trans-diagnostic theory has influenced an array of more disorder-specific models and treatment protocols and metacognitions have been found to predict distress in a variety of disorders, including depression (Papageorgiou and Wells, 2001), general anxiety disorder (Wells, 2010), psychosis (Morrison and Wells, 2003), posttraumatic stress-disorder (Wells and Sembi, 2004b) and addictive behaviours (Spada and Wells, 2005; Spada et al., 2008b). Recently, two studies have been particularly interesting to the current study. First, McNicol and colleagues (McNicol et al., 2012) found metacognitive beliefs to be influencing the level of distress in cancer patients. Distress is a normal and expected reaction to experiencing a close one having a psychotic episode, as well as to receiving treatment for a potentially life threatening disease such as cancer. But people differ in their level of distress and coping, and in the degree to which the worry and rumination continues after the initial crisis is 'settled', thus prolonging the suffering. Second, Spada and colleagues (2012) found parental over-protection to predict worry and anxiety in their children, an association which was partly mediated by the development of maladaptive metacognitions.

The metacognitive model has some interesting overlap with EOI, which in many ways also involves increased rumination, worrying, attempts at control, and possibly, suppressing of uncomfortable thoughts. In a prospective study of EOI, Boye et al. (1998) found that caregivers who remained high EOI from baseline to follow-up, scored higher on measures of 'intrusion' and 'psychophysiology

ical activation', or being in a state of alertness. The authors speculated that this was associated with extensive worrying and possibly suppression of thoughts in caregivers due to a 'stress condition' from being traumatised. To the best of this author's knowledge, the current study is the first attempt to use this metacognitive framework to examine caregiver distress.

Figure 1 illustrate how maladaptive metacognitions may influence, and be influenced by, caregiver distress. Caregivers respond with a range of feelings such as anxiety, sadness, grief and shock to the crisis of having a close one going through a psychosis. They also try to handle this situation the best they can, often in ways that are functional in the short term but detrimental to their well-being in the long run, e.g. too closely monitoring their ill family member, and being too protective and self-sacrificing. According to the S-REF model (Wells and Matthews, 1996; Wells, 2000) the prolonged and exacerbated distress is maintained by the CAS (worrying/ruminations, dysfunctional coping that backfires) which has overlaps with the behavioural component of EOI. These processes are, as argued by Wells and colleagues, partly fuelled by dysfunctional metacognitions. Interventions such as detached mindfulness, modification of maladaptive metacognitions and less dysfunctional coping strategies will limit the vicious circle of metacognitions, rumination/worrying and negative emotions, thus reducing the overall caregiver distress.

**Figure 1.** Maladaptive metacognitions and caregiver distress



*Note:* EOI = Emotional Over-Involvement: Over-protective, over-intrusive, self-sacrificing and over-identifying; CAS = Cognitive-Attentional System: Thought control/suppression, avoidance, rumination, worrying and hyper-vigilance. Based on Metacognitive Therapy (MCT; Wells et al., 2000).



## 2. Methods

### 2.1. Aim and Design

The study had a cross-sectional design and participants comprised the baseline of an intervention study seeking to compare multi-family interventions with individual-family interventions. The participants were consecutively included.

The overall goal of the present study has been to get a better understanding of what and how psychological factors relate to caregiver distress in first-episode psychosis. With this in mind, the specific aims were to: 1) systematically review the current literature that has examined psychological factors accounting for caregiver distress; 2) examine subjective appraisals and emotional over-involvement as predictors of caregiver distress, while addressing some of the methodological limitations in earlier studies; and 3) examine the concept of metacognition from two different theoretical perspectives as predictors of caregiver distress.

### 2.2. Research questions and hypotheses

The systematic review addressed the following specific research questions: What psychological processes, resources or competencies within caregivers have been empirically tested in studies to account for variations in caregiver distress? What were the findings and what was the methodological quality of these studies (Paper 1)?

To further address the aim of this study, a number of specific hypotheses were proposed. First, caregivers would experience high levels of distress already in the early phase of illness. This would be associated with more negative appraisal, and higher levels of EOI, but not with the level of symptoms and overall functioning in the patients (paper 2). Second, greater capacity for metacognition in caregivers would be associated with less negative and more positive experience of caregiving (paper 3). Finally, caregivers with more maladaptive metacognitions would report higher levels of distress. We also expected EOI to be associated with distress and that EOI would function as a mediator between metacognitions and distress (paper 4).

### 2.3. Participants and setting

Every person recently diagnosed between April 2011 and April 2013 with a first-episode non-affective psychotic disorder and currently enrolled in treatment within Region Zealand in Denmark (population: 816,670) was invited to participate.

The total numbers of eligible individuals were 99 patients enrolled in the out-patient treatment service based on assertive community treatment principles (OPUS) (Petersen et al., 2005b) together with 154 caregivers. Diagnoses were confirmed using Operational Criteria Checklist (OPCRIT; McGuffin, 1991) by a clinical psychologist, MD or psychiatrist. Of note, the different sub-studies represented by the different papers in this thesis had different number of participants, which is detailed in the papers. There are various reasons for this. First, the transcription and rating of the MAS-A was very time-consuming, and therefore only a consecutively and randomly chosen sample ( $n = 40$ ) could be analysed within the limited time-frame of this PhD project. Second, the MCQ-30 was added to the interview material after the commencement, leaving a sample of hundred and twenty-seven caregivers.

The median number of weeks between enrolling in treatment and participating in interviews was 4.5 weeks for caregivers (range 0.5 to 19.5) and 3.9 weeks for patients (range 0 to 16.5). At the time of interview twenty-four per cent ( $n=24$ ) of the patients were in symptom remission according to the criteria proposed by the Remission in Schizophrenia Working Group (Andreasen et al., 2005).

#### *2.4. Inclusion and exclusion criteria*

The inclusion criteria were: 1) ICD-10 criteria for a first-episode non-affective psychotic disorder (F20-29, except F21), 2) first-ever psychiatric treatment because of this disorder, 3) age 18-35. The only exclusion criterion, was insufficient Danish language skills for the interview to be completed, which none of the participants met.

#### *2.5. Materials*

##### *2.5.1. Caregiver measures*

*General Health Questionnaire* (GHQ-30; Goldberg and Williams, 1988). The level of distress and general well-being was measured using the GHQ-30, which is a 30 item self-report questionnaire. The GHQ was chosen as it has been used in numerous studies on caregivers of persons with psychosis and schizophrenia and is probably the most popular screening instrument for detecting psychiatric disorder in patient and community samples (Whittington and Huppert, 1998). For each item, the caregivers must rate the occurrence of a particular symptom on a 4-point Likert scale ranging from 0 to 3. The total score could thus vary from 0 to 90. The current severity of symptoms is compared to the habitual state of the person. There are four response categories to the positively worded items, which are labelled 'better than usual/more so than usual', 'same as usual', 'less than

usual' and 'much less than usual'. The response categories for the negatively worded items are 'not at all', 'no more than usual', 'more than usual', and 'much more than usual'. Based on Huppert et al. (1989), the GHQ was divided into subscales for anxiety, depression, wellbeing, social dysfunction and coping failures. GHQ-case score was also calculated, each item scored as 'present' or 'absent' (0-0-1-1), where total score could vary from 0 to 30. A case score above five is usually considered to be high, and suggesting clinically significant distress (Goldberg and Williams, 1988). For exploratory post-hoc testing, two subscales based on Boye et al. (1998) were computed, namely "intrusion" and "psychophysiological activation". This was in order to examine whether EOI was related to intrusive thoughts and "alertness" as suggested by the metacognitive theory of Wells (2000) and whether EOI could be considered a stress- or trauma reaction as suggested by Boye et al. (1998). The GHQ-30 has been shown to have high internal consistency and good test-retest reliability, as well as concurrent validity with a number of other clinical assessments (Goldberg and Williams, 1988).

*Experience of Caregiving Inventory* (ECI; Szmukler et al., 1996). The ECI is 66-item self-report questionnaire, designed to measure the subjective experience of providing care for a person with a serious mental illness such as schizophrenia. The questionnaire consists of 10 subscales: eight negative areas of caregiving (difficult behaviours, negative symptoms, stigma, problems with services, effects on the family, need to provide back-up, dependency and loss) and two positive (positive personal experiences and positive aspects of the relationship). The ECI measures how often caregivers have thought about each issue during the preceding month. The items are scored on a 5-point Likert scale with the response categories 'never', 'rarely', 'sometimes', 'often' and 'nearly always'. The maximum score for the negative subscale is 208 and 56 for the positive subscale. The ECI has been widely used for caregivers of persons with more chronic courses of psychotic illness and for persons at the early stage of illness, and has been shown to be a useful measure of the perception and appraisal of caregiving (Addington et al., 2005; Joyce et al., 2003). The internal consistency and construct validity of the scale have been found to be high (Joyce et al., 2000; Szmukler et al., 1996).

*Indiana Psychiatric Illness Interview* (IPII; Lysaker et al., 2002). The IPII is a semi-structured interview developed to assess illness narratives in patients with schizophrenia. This was modified for caregivers of patients with first-episode psychosis in collaboration with its first author. The interview typically lasts for 30-60 minutes, and is audiotaped and transcribed. The interview is divided into five sections. First, rapport is established and the caregivers are asked to tell the story of their family in as much detail as they can, starting from around the birth of the child that currently receives treatment. Second, they are asked if they

think their son or daughter has a mental illness, and how they understand it. Third, they are asked to consider whether this has had any influence on their occupational, social or emotional life, or on their personality. Fourth, they are asked how being a caregiver controls their lives, and how they cope with being a caregiver. This is followed by questions about how caring for the patient affects others and how others influence their way of being caregivers. Finally, they are asked how they think the patient's need for care will develop in the future. The tone is conversational. There are no direct questions of specific symptoms and the interviewer only asks for clarification when unclear. The result is a narrative of self and challenges related to being a caregiver, from which metacognitive capacities can be analyzed.

*The Metacognitive Assessment Scale abbreviated* (MAS-A; Lysaker et al., 2005). The MAS is a rating scale to assess metacognitive abilities and was originally designed to be used on psychotherapy transcripts in personality disorders (Semerari et al., 2003). This study used a modified version, the MAS-A adapted to analyzing IPII transcripts. This abbreviated version contains four scales: 'Understanding of one's own mind', 'understanding of others' minds', 'decentration' and 'mastery'. Decentration refers to the ability to see that others have independent motives and unique perspectives on life-events. Mastery refers to the ability to conceptualize one's difficulties and implement effective strategies to cope with problematic mental states and emotions. For each scale higher ratings reflect greater capacities to engage in more complex metacognitive acts. The maximum total score is 28. The MAS-A has been widely used with schizophrenia patients where it has acceptable reliability and validity, and it has been able to predict various psychosocial outcomes independently of symptoms and neurocognition (Lysaker et al., 2011b, 2010a, 2010b). It has also been used to study metacognition among persons with prolonged non-psychiatric medical conditions (Lysaker et al., 2014b, 2012b). To the author's knowledge, this is the first study using the MAS-A to assess metacognition in caregivers of persons with first-episode psychosis.

*Metacognitions Questionnaire 30* (MCQ-30; Wells & Cartwright-Hatton 2004). Individual differences in metacognitions were measured by the MCQ-30, which is a 30-item self-report questionnaire with five subscales: (a) positive beliefs about worrying (e.g. "Worrying helps me avoid problems in the future"); (b) negative beliefs about worry, concerning uncontrollability and danger (e.g. "When I start worrying I can't stop"); (c) cognitive confidence (e.g. "I do not trust my memory"); (d) beliefs about the need to control one's thoughts (e.g. "If I did not control a worrying thought and then it happened it would be my fault"); and (e) cognitive consciousness (e.g. "I constantly observe my thoughts"). Each state-

ment is rated on a 4-point Likert-scale as “Agree very much”/“Agree moderately”/“Agree slightly”/“Do not agree” thus yielding a score ranging from 30 to 120. The MCQ-30 has been widely used for a number of psychological difficulties ranging from depression, anxiety and addictive behaviors to psychosis. The MCQ-30 has good internal consistency and convergent validity, as well as acceptable test-retest reliability (Spada et al., 2008a; Wells and Cartwright-Hatton, 2004). The MCQ-30 was developed and validated on a normal sample. To the author’s knowledge this is the first study using the MCQ-30 to assess metacognitions in caregivers of persons with first-episode psychosis.

*Family Questionnaire* (FQ; Wiedemann et al., 2002). Caregivers’ levels of expressed emotion were assessed by the FQ. This is a 20-item self-report questionnaire examining how the family deals with everyday challenges, especially focusing on negative comments, criticism and emotional over-involvement. Questions like ‘I have a tendency to neglect myself because of her/him’ and ‘she/he irritates me’ are rated as ‘never/very seldom’, ‘seldom’, ‘often’ or ‘very often’. The measure consists of two subscales with 10 items each: Emotional over-involvement (EOI) and Critical comments (CC). The EOI includes over-intrusive, self-sacrificing, overprotective behavior, or exaggerated emotional response, and over-identification with the patient; CC is defined as unfavorable comments on the behavior or the personality of the patient (Vaughn and Leff, 1976). The items are scored from 1 to 4 yielding a maximum score of 40 in each of the two subscales. Caregivers are classified as high EE if they score 23 or greater on the CC subscale or 27 or greater on the EOI subscale. The FQ has good psychometric properties including a clear factor structure, high internal consistency of subscales and good concurrent validity in relation to the widely used Camberwell Family Interview (Vaughn and Leff, 1976). In addition, the FQ has displayed a similar level of accuracy and substantially higher sensitivity as compared to the Five Minute Speech Sample, which is another widely used measure to assess EE (Magaña et al., 1986).

### 2.5.2. Patient measures

*Positive and Negative Syndrome Scale for Schizophrenia* (PANSS; Kay et al., 1987). This is a 30-item instrument (each rated on a scale from 1 to 7) for the assessment of positive and negative symptoms of psychosis. Symptoms during the past week are rated, and higher scores indicate more severe symptoms. The positive symptoms sub-scale and negative symptoms sub-scale each consist of seven items and there is also a general pathology sub-scale consisting of 16 items. The PANSS is the most widely used instrument for the assessment of schizophrenia symptoms in clinical trials and its psychometric properties have been found to

be adequate (Peralta and Cuesta, 1994; Santor et al., 2007). For this study, the PANSS was divided into Positive, Negative, Cognitive, Depressive and Excitatory Component scores based on Bentsen et al. (1996b). Symptom remission was defined according to Andreasen et al.'s (2005) recommendation, namely scores of 3 or less on each of eight core symptoms: Delusions, Unusual Thought Content, Hallucinatory Behavior, Conceptual Disorganization, Mannerisms and Posturing, Blunted Affect, Social Withdrawal, and Lack of Spontaneity. Global functioning was measured using the DSM-IV's *Global Assessment of Functioning scale* (GAF) (APA, 1987).

### 2.5.3. Notes on outcome measures

The project began with using both the GHQ and the ECI as main outcomes since these have been among the most widely used for similar studies and populations. Some of the studies that we used as inspiration for the current design, included the ECI as a dependent measure (Alvarez-Jiménez et al., 2010; Cotton et al., 2013; Patterson et al., 2005; Raune et al., 2004). Later it was discovered that the ECI was better used as an independent measure of subjective appraisal, as used in other studies (Addington et al., 2003; Gleeson et al., 2010; Tomlinson et al., 2013). The field of metacognition has not been examined in relation to caregiver distress before this study, and investigating how it might be related has, to a certain extent, been explorative. For this reason, one paper has used the ECI as a dependent variable (Jansen et al., 2013) while another has used it as an independent variable, measuring the subjective appraisal within the stress-appraisal-coping model (Jansen et al. in press).

## 2.6. Statistics

Statistical analyses were carried out using SPSS version 20 (IBM Inc., Chicago, IL, USA), including Mean and Standard Deviations, Pearson's correlations, t-tests, Linear mixed model analysis and mediation analyses. Multilevel modeling was used to accommodate the challenge of non-independence resulting from more than one caregiver per participant (Tabachnick and Fidell, 2014).

For the mediation analysis used in paper two the recommended criteria set out by Preacher & Hayes (2008a, 2004) were followed and we used the authors' macros designed for SPSS which can be downloaded from the internet (<http://afhayes.com/spss-sas-and-mplus-macros-and-code.html>). The statistical significance of the indirect coefficients ( $a*b$ ) was tested using a nonparametric bootstrapping method. Randomly drawing participants from the original dataset and replacing each value as it was sampled created 5,000 identically sized datasets. Confidence intervals for the 5,000  $a*b$  values were then derived using Z-score biased corrections. In this approach mediation is significant if the upper

and lower bounds of confidence intervals do not contain zero (Efron and Tibshirani, 1993).

There are many approaches to statistical inference for indirect effects and the most cited in psychological research has been that of Baron and Kenny (1986). In recent years, the relative performances of these various approaches have been studied extensively (Hayes, 2013), and the bias-corrected bootstrapping confidence interval used in this study currently seems to be the preferred approach (Breitborde et al., 2010; Mackinnon et al., 2004; Zhao et al., 2010). Although the use of mediation analysis on cross-sectional data has been questioned (Cole and Maxwell, 2003; Maxwell and Cole, 2007), the current study uses mediation analysis as a first approximation in exploring the hypothesis of the EOI being a mediator, following the suggestions by Hayes (2013) and Mackinnon (2008).

## 2.7. Inter-rater reliability

Inter-rater agreement on the measures included are shown in Table 4. Of particular note, most ICC values can be considered to represent “excellent reliability (>.75) except PANSS Excitative component, GAF symptom and GAF function which are within the category “fair to good reliability” (.04 to .75) (Fleiss, 1986).

**Table 1.** Inter-rater agreement on the measures included

Scale	ICC (2,k)	95% CI	Sig.
PANSS Positive component	.81	.60 to .92	.000
PANSS Negative component	.90	.77 to .96	.000
PANSS Depressive component	.86	.71 to .94	.000
PANSS Excitative component	.64	.29 to .85	.001
PANSS Cognitive component	.81	.61 to .92	.000
GAF symptom	.73	.44 to .89	.000
GAF function	.62	.24 to .84	.003
MAS-A	.85	.66 to .92	.000

*Note:* ICC = Intra Class Correlation; CI = Confidence Interval; PANSS = Positive and Negative Syndrome Scale for Schizophrenia; GAF = Global Assessment of Functioning scale; MAS-A = Metacognitive Assessment Scale - Abbreviated

## 2.8. Ethical considerations

All participants gave written informed consent and the study was approved by the Regional Committee for Research Ethics. All participants were debriefed following the interview and completion of questionnaires, to give an opportunity to discuss eventual aversive effects caused by talking about emotionally difficult material. None of the participants reported any aversive effects.

### **3. Results**

#### *3.1. A systematic review of psychological factors (Paper 1)*

The aim of the first paper was to address the questions as to what psychological processes, resources or competencies within caregivers have been empirically tested to account for caregiver distress, and moreover, to highlight the findings and the methodological quality.

The main findings from the review are presented in Table 2. The search identified 14 papers describing 12 studies together comprising 977 caregivers of persons with first-episode psychosis. The mean age of caregivers was 47.3 years (SD = 10.8), of whom 70.6 % were female and 74.2% were parents. Nine different psychological variables were examined in these studies, which were categorized in the following non-mutually exclusive groups: coping, appraisal/attribution and interpersonal response. There were moderate to high effect sizes supporting the link between distress and such psychological factors as avoidant coping, appraisal and EOI. However, the possibilities of drawing conclusions were limited by a number of methodological limitations, including cross-sectional data, small sample-sizes, confounding variables not being accounted for, and a wide variation in outcome measures. The various definitions of caregiver distress and outcome measures used in the included studies are presented in Table 3. As can be seen, the most widely used distress measures were the GHQ (50%, n=6) and ECI (67%, n=8) making direct comparison possible for a number of scores and subscores.



**Table 2.** Studies investigating psychological factors in caregiver distress

Study Location	N Caregivers	Age (SD)	% female	Design	Distress measures	Psychological constructs	Measure of psychol. Con-structs	Key findings
Addington et al. 2003 Calgary, Canada	238	-	-	Cross-sectional	PGWB	Appraisal	ECI	The family's appraisal of the impact of the illness was associated with their psychological well-being.
Alvarez-Jimenez et al. 2010 Melbourne, Australia	63 (48)	44.4 (11.1)	-	Prospective, 7 month follow-up	GHQ-28 ECI	Coping model Attachment model	FQ	EOI predicted family distress and burden at baseline and follow-up.
Cotton et al. 2013 Melbourne, Australia	124	47.2 (8.3)	82.3	Cross-sectional	ECI K10	Coping style	WOC FQ	Avoidance coping strategies were related to psychological distress, EOI and burden.
Gleeson et al. 2010 Melbourne, Australia	63 (21)	45.5 (9.5)	-	RCT	GHQ-28	Appraisal	ECI	There were significant group effects for aspects of the appraisal of caregiving (RPT vs. TAU). There were no significant effects for caregiver distress.
Hinrichsen et al. 1999 NY, USA	63	-	-	Cross-sectional	PRS BI SCL-90	Attribution Coping	PDAQ HDLFC DMSS	Attributions and reported ways of coping were linked to indices of caregivers' emotional adjustment.
Jansen et al. 2013 Roskilde, Denmark	40	-	60	Cross-sectional	ECI GHQ-30	Level of metacognition in caregivers	MAS-R FQ	Greater levels of general distress and EOI were related to more negative caregiver experience, while greater metacognitive capacity was related to more positive experiences of caregiving.
McNab et al. 2007 Melbourne, Australia	53	49.1 (5.3)	66	Cross-sectional	GHQ-28	Attribution Utility beliefs	IPQ (note: only four items) UB FQ	Distress was associated with utility beliefs (beliefs about the utility of self-sacrifice and person-focused criticism) and EOI.
Moller-Leimkuhler 2005	83 (35 F20)	46.8 (11.5)	44.4	Cross-sectional	FBQ SCL-90 LQLP Bf-S	Coping "Negative stress response" (cop-	SVF FKK NEO-FFI FMSS	The most relevant predictors of burden were EE, emotion-focused coping strategies and generalised

Munich, Germany						ing) Personality dimensions	FQ	negative stress response. Illness characteristics were not predictive of distress.
Moller-Leimkuhler 2006	70 (34 F20)	49.2 (10.9)	48.6	Prospective, 1 year follow-up	FBQ SCL-90 LQLP Bf-S	Coping "Negative stress response" (coping) Personality dimensions	SVF FKK NEO-FFI FMSS FQ	Caregiver burden was predicted by EE, neuroticism, generalised negative stress response (negative coping) and life stressors.
Munich, Germany								
Moller-Leimkuhler et al. 2008	63 (34 F20)	48.3 (17.9)	47.6	Prospective, 2 year follow-up	FBQ SCL-90 LQLP Bf-S	Coping "Negative stress response" (coping) Personality dimensions	SVF FKK NEO-FFI FMSS FQ	Caregiver burden was predicted by EE, neuroticism, generalised negative stress response (negative coping) and life stressors.
Munich, Germany								
Onwumere et al. 2011	141	50.2 (11.8)	79.3	Cross-sectional	GHQ-28	Coping	COPE	Caregiver distress was associated with avoidant coping strategies but not with duration of illness.
London and East Anglia, UK								
Patterson et al. 2005	50 (39)	43.9 (11.2)	-	Prospective, 9 month follow-up	CDSS ECI	Loss Attachment model  (Appraisal/attributions)	TRIG CFI	Subjective burden was linked to loss and not EE status. The appraisal of loss was linked to EOI but not CC at baseline. Loss was reduced in those who changed EE status from high EOI to either high CC or low EE.
Birmingham, UK								
Raune et al. 2004	46	47.2 (14.4)	72	Cross-sectional	GHQ-28 ECI SFS BDI	Appraisal Coping	The cope instrument (COPE) CFI	High EE was associated with avoidant coping, subjective burden and lower perceived patient interpersonal functioning. High EE might be a way of coping with the burden.
London, UK								
Tomlinson et al. 2013	24	51.7 (5.8)	83.3	Cross-sectional	HADS	Social cognition	Hinting task FEEST FAS ECI	Negative experience of caregiving was related to high EE, anxiety and depression. Both patients and carers showed impaired social cognition, but social cognition was not related to burden or EE.
London, UK								

**Table 3.** The various ways of describing and measuring caregiver experience

Description in paper	Measurement	Authors
“Appraisal of impact of illness/stressors/caregiving experience”	ECI	Alvarez-J. et al., 2010 Cotton et al., 2013 Gleeson et al., 2010 Addington et al., 2003 Tomlinson et al., 2013
“Subjective experience of caregiving”	ECI	Raune et al., 2004
“Psychological distress”	K10	Cotton et al., 2013
“Burden of care/subjective burden”	ECI	Patterson et al., 2005 Raune et al., 2004
	FBQ	Moller-L. et al., 2005, 06, 08
	LQLP	Moller-L. et al., 2005, 06, 08
“Caregiver distress”	GHQ-30 GHQ-28	Jansen et al., 2013 McNab et al., 2007 Onwumere et al., 2011 Raune et al., 2004
“Level of stress”	PGWB	Addington et al., 2003
“General well-being”	GHQ-30 PGWB Bf-s SCL-90	Jansen et al., 2013 Addington et al., 2013 Moller-L. et al., 2005, 06, 08 Moller-L. et al., 2005, 06, 08
“Emotional adjustment to the care of the patient”	PRS BI SCL-90	Hindrichsen et al., 1999 Hindrichsen et al., 1999 Hindrichsen et al., 1999
“Carer symptoms”	GHQ-28	Alvarez-Jimenez et al., 2010
“Depression” or “anxiety”	CDSS BDI HADS	Patterson et al., 2005 Raune et al., 2004 Tomlinson et al., 2013

*Note:* PGWB = Psychological General Wellbeing Scale; GHQ-28 = General Health Questionnaire, 28-item version; ECI = Experience of Caregiving Inventory; K10 = Kessler Psychological Distress Scale; PRS = Patient Rejection Scale; BI = The Burden Interview; SCL-90 = Symptom Checklist-90; DMSS = Dementia Management Strategies Scale; GHQ-30 = General Health Questionnaire, 30-item version; FBQ = Family Burden Questionnaire; LQPL = Lancashire Quality of Life Profile ;Bf-S = Befindlichkeitsskala; CDSS = Calgary Depression Scale for Schizophrenia; BDI = Beck Depression Inventory; HADS = Hospital Anxiety and Depression Scale.

### 3.2. Subjective appraisal and expressed emotion (paper 2)

The aim of paper two was to examine subjective appraisal and EOI as predictors of caregiver distress, while addressing some of the methodological challenges found in earlier studies such as sample size, controlling for patient symptoms and level of functioning, and caregiver demographics.

Characteristics of caregivers and patients are presented in Table 4 and caregiver measures are presented in Table 5. As can be seen, most caregivers are fe-

male (62%), parents (81%), living with the patient (57%) and employed (71%). While there are no norms available for the PANSS, the mean score of 57.14 (SD = 12.94) would suggest generally low levels of psychotic symptoms in the sample (Leucht et al., 2005). At the time of interview 24% (n = 24) of the patients were in symptom remission according to the criteria proposed by The Remission in Schizophrenia Working Group (Andreasen et al., 2005). Caregivers presented with high levels of distress, as evidenced by 37 per cent (n = 57) meeting criteria for clinical significant distress, or caseness (> 5 on GHQ-30).

**Table 4.** Characteristics of caregivers and persons with FEP

	<b>M (SD)</b>	<b>% (n)</b>
Gender female	-	62.3 (96)
Age	49.09 (9.17)	-
Relationship		
Parent	-	80.5 (124)
Stepparent	-	5.2 (8)
Sibling	-	5.8 (4)
Spouse	-	5.8 (9)
Other	-	5.8 (9)
Cohabitation	-	57.1 (88)
Face-to-face contact > 1 x week	-	83.8 (129)
Employment	-	71.4 (110)
Characteristics of persons with FEP		
Gender female	-	37.4 (37)
Age	21.09 (3.44)	-
PANSS total score	57.14 (12.94)	-
Positive	11.22 (4.57)	-
Negative	20.31 (7.31)	-
Emotional distress	9.88 (3.23)	-
Cognitive disorganisation	5.35 (1.86)	-
Excitement	6.60 (1.63)	-
Global functioning, GAFs	40.21 (12.14)	-
Global functioning, GAFF	40.80 (12.34)	-

*Note:* N = 154 caregivers and 99 service users with FEP. M = Mean; SD = Standard Deviation; PANSS = Positive and Negative Syndrome Scale for Schizophrenia; GAF = Global Assessment of Functioning Scale; GAFs = Symptom score; GAFF = Function score.

**Table 5.** Caregiver clinical measures

Measures	M (SD)	% (n)
GHQ	29.85 (14.6)	-
GHQ-caseness	-	37 (57)
ECI – Negative subscale	73.95 (31.79)	-
ECI – Positive subscale	27.03 (8.65)	-
FQ – EOI	23.34 (5.1)	-
FQ – CC	18.33 (5.45)	-
FQ – High EOI	-	28 (43)
FQ – High CC	-	21 (32)
FQ – High EE on one subscale	-	36 (55)
FQ – High EE on both subscales	-	12 (18)

*Note:* N = 154. M = Mean; SD = Standard Deviation; GHQ = General Health Questionnaire; ECI = Experience of Caregiving Inventory; FQ = Family Questionnaire; EOI = Emotional Over-Involvement; CC = Critical Comments

The results of the linear mixed model analysis with distress (GHQ-30) as the main outcome are presented in Table 6. High scores on EOI and Negative Appraisal were significantly related to increased caregiver distress ( $\beta = 1.13$ , 95% CI = .48 to 1.78,  $p = 0.001$  and  $\beta = 0.16$ , 95% CI = .04 to .29,  $p = 0.011$  resp.) after controlling for caregivers' gender and cohabitation, and the level of symptoms and overall functioning in patients. Caregivers' positive appraisals related significantly to decreased caregiver distress ( $\beta = -.49$ , 95% CI = -.76 to -.21,  $p = 0.001$ ) while criticism did not significantly contribute to the model ( $\beta = -.31$ , 95% CI = -1.04 to .42,  $p = 0.403$ ).

**Table 6.** Linear mixed model analysis with distress as dependent variable

Predictors	$\beta$	95% CI	p
Expressed emotion			
FQ – Emotional Over-Involvement	1.13	.48 to 1.78	<b>.001</b>
FQ – Critical Comments	-.31	-1.04 to .42	.403
Subjective appraisal			
ECI – Negative subscale	.16	.04 to .29	<b>.011</b>
ECI – Positive subscales	-.49	-.76 to -.21	<b>.001</b>
Caregiver demographics			
Gender	-.28	-4.87 to 4.32	.905
Cohabitation	-2.59	-7.53 to 2.34	.298
PANSS			
Positive component score	.15	-.42 to .72	.592
Negative component score	.04	-.37 to .45	.853
Cognitive component score	.23	-1.25 to 1.70	.759
Depressive component score	.06	-.77 to .88	.893
Excitatory component score	.11	-1.42 to 1.63	.887
Global functioning			
GAFs	.21	-.27 to .33	.836
GAFf	1.37	-.10 to .51	.179

Note: N=154. CI = Confidence Intervals; FQ = Family Questionnaire; ECI = Experience of Caregiving Inventory; PANSS = Positive and Negative Syndrome Scale for Schizophrenia; GAF = Global Assessment of Functioning Scale; GAFs = Symptom score; GAFf = Function score.

The relationships between distress and the two subscales of EE – EOI and CC – are presented in Table 7. There was a consistent pattern that caregivers with higher levels of either EOI or CC had higher levels of overall distress and higher score on the particular subscales than caregivers with lower levels of EOI and CC. of particular note, high EOI had consistently higher levels of distress than high CC, and high CC did not have significantly higher scores on the subscales of low self-esteem and depression compared with low CC.

**Table 7.** The relationship between EE and subscales of caregiver distress

	EOI		CC	
	Low	High	Low	High
GHQ-total	25.3 (11.4)	42.0 (15.2)**	28.5 (14.5)	35.7 (13.8)*
Anxiety	6.4 (4.6)	13.0 (4.9)**	7.6 (5.5)	10.7 (4.9)**
Low self-esteem	4.1 (1.8)	5.4 (1.8)**	4.4(1.9)	5.0 (1.6)
Depression	2.1 (2.3)	4.8 (3.6)**	2.8 (2.9)	3.4 (3.1)
Coping difficulties	4.4 (1.7)	7.4 (2.9)**	5.0 (2.5)	6.0 (2.5)*
Social dysfunction	3.2 (1.3)	4.4 (2.0)**	3.4 (1.6)	4.0 (1.5)*

Note: N = 154. Mean (Standard Deviation). Statistical analyses by the independent sample Student's t-test. EE = Expressed Emotion; GHQ = General Health Questionnaire; EOI = Emotional Over-Involvement; CC = Critical Comments  
\*P < 0.05, \*\*P < 0.0005

### 3.3. Metacognitive capacity (paper 3)

The aim of the third paper was to examine whether the capacity for metacognition – or forming complex accounts of own and others' thoughts and feelings, including using this understanding to solve emotional and social problems – were associated with less negative and more positive experiences of caregiving.

Means and Standard Deviations for the study on metacognitive capacity are presented in Table 8 and Pearson correlations are presented in Table 9. Greater levels of distress and EOI were associated with more negative experiences of caregiving while greater metacognitive capacity was associated with more positive experiences of caregiving. There were also significant associations between all the subscales of metacognition and positive experiences: Self-reflectivity (.46  $p < .01$ ), Understanding of others (.46  $p < .01$ ), Decentration (.60  $p < .01$ ) and Mastery (.47  $p < .01$ ). No significant association was found between capacity for metacognition and distress (-.03  $p = .88$ ), or between metacognition and caregiver criticism (-.03  $p = .83$ ). There was no significant correlation between positive and negative caregiver experiences (.107,  $p = .51$ ).

**Table 8.** Descriptive statistics for EOI, distress, positive/negative caregiver experience and capacity for metacognition

Measures	M	SD
EOI	23.83	5.21
Caregiver distress (GHQ-30)	32.60	16.75
Positive caregiving experiences (ECI - Positive)	25.90	9.16
Negative caregiving experiences (ECI - Negative)	74.73	34.10
Capacity for metacognition (MAS-A)	19.83	4.99

Note: N = 40. M = Mean; SD = Standard Deviation; ECI = Experience of caregiving Inventory; GHQ-30 = General Health Questionnaire 30 item version; MAS-A = Metacognitive Assessment Scale Abbreviated

**Table 9.** Pearson correlations for EOI, distress, positive/negative caregiver experience and capacity for metacognition

	<u>Positive Experiences</u>	<u>Negative Experiences</u>
Emotional Over-involvement (EOI)	.19	.67*
Distress (GHQ-30)	.02	.62*
Capacity for metacognition (MAS-A)	.52*	.03

Note: N = 40. EOI = Emotional Over-Involvement; GHQ-30 = General Health Questionnaire 30 item version; MAS-A = Metacognitive Assessment Scale Abbreviated.

\*p < 0.01

### 3.4. Metacognitions (paper 4)

The aim of paper four was to examine caregiver distress from the metacognitive model of Wells et al. (Wells and Matthews, 1996; Wells, 2000), namely testing whether the S-REF model could accommodate the concepts of EOI and caregivers' specific dysfunctional metacognitions in understanding distress. The hypothesis was that EOI would mediate the effect of metacognition on caregiver distress, that is, EOI would function as a coping mechanism driven by metacognitions.

Descriptive statistics and Pearson correlations of variables in the study on metacognitions are presented in Table 10. In the univariate analyses, there were significant associations between distress and three of the metacognitions subscales (negative beliefs about worry concerning uncontrollability and danger,



cognitive confidence and beliefs about need to control thoughts). When controlling for EE, the linear mixed model analysis found that negative beliefs about worry concerning uncontrollability remained as significant predictors of distress. This variable was carried forward to the mediation analysis.

**Table 10.** Descriptive statistics and Pearson correlations for EE, distress and the subscales of metacognitions

Study variables	M	SD	1	2	3	4	5	6	7	8
1. GHQ-30	29.83	14.29	-	.58**	.30*	.13	.42**	.33**	.26**	.09
2. FQ-EOI	23.36	4.89		-	.51**	.22	.39**	.22*	.21**	.09
3. FQ-CC	18.13	5.23			-	.14	.23*	.09	.23**	.12
4. MCQ-30-1	9.66	3.90				-	.36**	.14	.41**	.57**
5. MCQ-30-2	11.48	4.17					-	.59**	.59**	.45**
6. MCQ-30-3	10.68	4.57						-	.44**	.29**
7. MCQ-30-4	9.22	3.38							-	.52**
8. MCQ-30-5	11.59	4.44								-

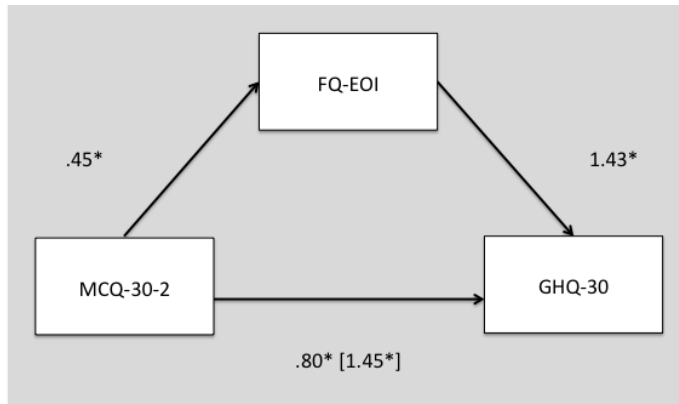
Note: N=127. M = Mean; SD = Standard Deviation; GHQ-30 = General Health Questionnaire-30; FQ = Family Questionnaire; EOI = Emotional; CC = Critical Comments; MCQ-30 = Metacognitions Questionnaire-30; MCQ-1 = positive beliefs about worry; MCQ-2 = negative beliefs about worry concerning uncontrollability and danger; MCQ-3 = beliefs about cognitive confidence; MCQ-4 = beliefs about the need to control thoughts; MCQ-5 = cognitive self-consciousness.

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

In order to explore the hypothesis that emotional over-involvement mediates the relationship between metacognitions and distress, a mediation analysis was carried out. Because both the a-path ( $B = .45$ ,  $se = .10$ ,  $p < .001$ ) and the b-path ( $B = 1.43$ ,  $se = .22$ ,  $p < .001$ ) were significant, premises for the mediation analysis using bootstrapping with bias-corrected confidence estimates were met (Mackinnon et al., 2004; Preacher and Hayes, 2008b, 2004). The results of the mediation analysis were consistent with a possible mediational role of emotional over-involvement in the relation between negative beliefs about worry concerning uncontrollability and danger and distress in caregivers ( $B = 1.45$ ;  $CI = .37$  to  $1.06$ ). The results indicated that the direct effect of negative beliefs about worry concerning uncontrollability and danger on distress was reduced, but remained significant ( $B = .80$ ,  $se = .26$ ,  $p < .001$ ) when controlling for emotional over-involvement, thus suggesting only partial mediation. This suggests that negative beliefs about worry concerning uncontrollability and danger influence caregiv-

ers' distress directly as well as indirectly through emotional-over-involvement ( $B = 1.45$ ,  $se = .28$ ,  $p < .001$ ). Figure 2 displays the results.

**Figure 2.** Mediation model. Metacognitions, EOI and distress



*Note:* N=124. FQ-EOI = Family Questionnaire – Emotional Over-involvement subscale; GHQ-30 = General Health Questionnaire; MCQ-30 = Metacognitions Questionnaire-30; MCQ-2 refers to negative beliefs about worry concerning uncontrollability and danger. \* $p < .001$ .

Finally, exploratory post-hoc analyses for this thesis were carried out to examine whether EOI were related to ‘psychophysiologic activation’, or ‘alertness’, and ‘intrusion’ based on the GHQ-30 subscales suggested by Boye et al. (1998). Compared with caregivers characterised as low EOI, caregivers characterised as high EOI had significantly higher mean score of ‘intrusion’ ( $t(124) = 6.43$ ,  $p < .001$ ) and ‘psychophysiologic activation’, or level of alertness ( $t(124) = 6.03$ ,  $P < .001$ ).

## 4. Discussion

The overall goal of this PhD project was to get a better understanding of what and how psychological factors relate to caregiver distress in first-episode psychosis. With this in mind, the specific aims were to: 1) systematically review the current literature that has examined psychological factors accounting for caregiver distress; 2) examine subjective appraisals and emotional over-involvement as predictors of caregiver distress, while addressing some of the methodological limitations in earlier studies; and 3) examine the concept of metacognition from two different theoretical perspectives as predictors of caregiver distress.

In general, the current study revealed high levels of distress, reflected in the finding that more than one-third of the caregivers were suffering from clinical significant distress. These results concur with the general finding in the literature that caregivers already in the early phase of illness experience high levels of distress and report many of the same sorts of problems as long-term caregivers (Kuipers and Raune, 2002). The percentage of case scores on the GHQ was considerably higher than what was found by Tennakoon et al. (2000) (12%), but slightly lower than what was found by Alvarez-Jiménez et al. (2010) (55.5%) and by Barrowclough and Parle (1997) (57%).

#### *4.1. Psychological factors involved in caregiver distress*

The systematic review found that nine different psychological variables were studied in the included studies, which were here categorised in the following non-mutually exclusive groups: coping, appraisal/attribution and interpersonal response. There were moderate to large effect sizes supporting the link between distress and psychological factors such as avoidant coping, appraisal and EOI. However, many of the studies included had some methodological limitations in terms of cross-sectional data, small sample-sizes, confounding variables not being accounted for, and a wide variation in outcome measures. Thus, the conclusions must have these in mind. However, this review highlighted the relevance of psychological factors within caregivers to the understanding of caregiver distress. Moreover, these various predictors also seem to fit within the stress-appraisal-coping model as well as the cognitive model of caregiving in FEP (Kuipers et al., 2010; Szmukler et al., 1996).

#### *4.2. Subjective appraisal and caregiver distress*

Caregiver distress was found to be more related to the caregivers' subjective appraisal and EOI, than to variations in symptoms and global functioning of the person diagnosed with FEP. This is in line with other studies in FEP (Addington et al., 2003; Patterson et al., 2005) and with studies of more chronic forms of illness (Barrowclough et al., 2001; Boye et al., 2001; Harvey et al., 2001; Szmukler et al., 1996). However, while subjective appraisal is more associated with distress than symptomatology in the early phase of illness, prospective studies have shown that symptoms may be more predictive of caregiver distress at later stages of illness (Dyck et al., 1999). While there is limited data on this, a plausible explanation would be that burden and distress increase as the illness and disturbing symptoms persist, with disappointment and stress associated with recurrent relapse or periods with symptom exacerbation. The findings lend further support to the stress-appraisal coping model (Joyce et al., 2000; Szmukler et al.,

1996) and the cognitive model (Barrowclough and Tarrrier, 1992; Kuipers et al., 2010; Tarrrier et al., 1988b) of caregiver distress in FEP. They also highlight supportive interventions aimed at changing or modifying unhelpful cognitions to improve distress, as suggested by the cognitive therapy model (Beck, 1979). However, as pointed out by Harvey (2001) further research is needed to determine predictors of caregivers' appraisals. There are at least two ways of working with appraisals within the cognitive behaviour tradition. The first is the classic approach, in which the content of thoughts are the focus, and unhelpful appraisals are modified or "restructured" into more supportive and helpful thoughts. The second, is the third-wave or contextual behaviour therapy approach, in which the focus is how one relates to the unhelpful thought or appraisal, rather than trying to change the content. The idea is to see "thoughts as just thoughts", practice "letting go" without either getting caught up in it or trying to suppress it or making it go away (Hayes et al., 2011; Segal et al., 2002; Wells, 2000).

#### *4.3. Expressed emotion and caregiver distress*

The finding that EOI predicts caregiver distress corresponds to studies in FEP (Alvarez-Jiménez et al., 2010; Patterson et al., 2005) as well as to studies of more chronic forms of psychosis (Boye et al., 1998; Breitborde et al., 2009a). From the current study, a number of suggestions as to a psychological understanding of the concept of EE can be made. First, as suggested by Alvarez-Jiménez et al. (2010), the two subscales of EE seem to be associated with different factors, where EOI seems to be more closely related to caregiver distress than CC. Studies have found CC to be more of a 'trait' in the caregivers, but also to be associated with Duration of Untreated Psychosis (DUP), illness length and frequent psychotic episodes (Alvarez-Jiménez et al., 2010; Schreiber et al., 1995; van Os et al., 2001). Second, EOI may be viewed as a coping mechanism adopted by caregivers as an attempt to reduce the impact of illness and their own distress (van Os et al., 2001).

The above is also in line with Patterson et al. (2000), who found EOI to be associated with perceived loss in their sample of FEP. Loss and grief is according to Patterson et al., a central feature of caregiver experience in FEP but is often not attended to enough in family work. too much emphasis on EE, communication and problem-solving might not help caregivers "integrate" their traumatic experience, but rather support a coping characterised by "sealing over" (McGlashan, 1975). The latter coping style has been found to result in a more distant relationship with the patient (Patterson et al., 2000). This is an important issue as many caregivers become entrenched in long-term grief (Patterson et al., 2000). Davis and Schultz (1998) found that 43% of caregivers reported high levels of grief more than ten years after their children were

diagnosed with schizophrenia and Eakes (1995) found that eight out of ten caregivers were experiencing chronic sorrow.

However, the relationship between distress, burden, grief and EE is complex and the explanation above do not preempt the discussion. Some authors have argued that EE is better viewed as a “thermometer” of the family climate (Patterson et al., 2000, p. 192), others have argued that it is nothing more than a proxy or an epiphenomenon of other variables, e.g. appraisal (Breitborde et al., 2009b, p.45). While the theoretical understanding of EE is underdeveloped, as pointed out by Breitborde et al. (2009b), there seems to be agreement that it is not a one way causality, but rather a vicious circle, in which illness and caregiver distress reciprocally influence each other in a negative way (Kuipers et al., 2002). While distress seems less influenced by illness characteristics compared to subjective appraisals in the early phase, with time it might possibly increase when the illness progresses in a negative way but then decrease when things go well.

#### *4.4. Metacognitive capacity*

While interpersonal skills, such as the capacity to form complex representations of oneself and others and constructing meaningful explanations for psychological distress, are central to family functioning (Lysaker et al., 2011b), the current study has been the first attempt to examine these phenomena in relation to caregiver distress and caregiver experiences. The findings in this study suggest that positive and negative aspects of caregiving are associated with different variables. Negative experiences were associated with EOI and general well-being, while positive experiences were associated with the capacity for metacognition. Contrary to expectation, metacognition did not relate to negative experiences. Greater metacognitive capacity does not seem to reduce suffering and distress in the early phase of illness, which is a healthy and normal reaction to having a close one suffering from psychosis. On the other hand, it might help broaden the perspective, allowing for both negative and positive experiences. In the only related study examining the capacity for understanding others’ thoughts and feelings, Tomlinson et al. (2013) found that the performance on social cognition tasks was impaired in both caregivers and patients compared with expected performances for the measure. Taken together, these results suggest that abilities to reflect upon and make sense of other peoples’ cognitive and emotional states can be reduced in families with FEP, as shown by Tomlinson et al., but is of importance to the overall caregiving experience, as found in the current study. As in the current study, Tomlinson did not find social cognition to be predictive of caregiver distress or burden in caregivers. More studies are needed to ascertain

knowledge in this field and future studies should examine whether these factors may predict long-term distress and burden.

As pointed out by Patterson et al. (2005), the cognitive literature places great emphasis on the importance of subjective appraisal when trying to understand how the illness impacts on caregivers, but takes little account of how these appraisals develop. Metacognitive capacity may be one such determinant. As pointed out earlier, the early stage of adaption to psychosis often involves a process akin to grieving in caregivers. However, certain aspects of the traditional family work models, e.g. problem-solving, communication training and conflict management, may disturb the emotional processing of loss and grief. As noted by Patterson et al. (2005), “Anger, denial, frustration, ‘search for what has been lost’ and later ‘acceptance’ of the new situation are all recognisable as part of the response to loss” (p. 63). Metacognitively oriented therapy or Metacognitive Reflective Insight Therapy (MERIT) (Lysaker et al., 2014a, 2011a) could assist this process by providing a setting in which caregivers can get the opportunity to tell their story with the aim of creating a more complex narrative, process grief and loss and expand their capacity to form complex accounts of own and others’ thoughts and feelings, i.e. their metacognitive capacity. In Mentalisation-Based Therapy (MBT), this is referred to as ‘minding’ self and others (Allen and Fonagy, 2006). In the words of Skårderud (2007), “mentalising can be seen as “simplifying the basic steps in psychotherapeutic encounters, either in individual, group or marital and family treatment contexts” (p. 326). According to Skårderud, “a mentalising attitude means an inquisitive, playful, curious and open-minded style in dialogues, with a focus on minding the mind” (p.326). In many ways, this is a different approach to families than the structured psycho-educational intervention, and might serve as an important supplement. While the data supporting the efficacy of family psycho-education is compelling, it does not meet the needs of all families, as noted by Dixon (2000).

#### *4.5. Maladaptive metacognitions*

With regard to specific metacognitions as discussed by Wells’ (2000), the current study found evidence to support their relevance in caregiver distress. The positive associations between distress and the three dimensions of metacognitions (negative beliefs about worry concerning uncontrollability and danger; cognitive confidence; and beliefs about the need to control thoughts) are in keeping with earlier studies and these particular three subscales have been found to be implicated across many domains of emotional difficulties (Spada et al., 2010). The mediation analysis also found that EOI could be seen as mediating the effects of metacognitions on caregiver distress. Finally, the exploratory post-hoc analyses

revealed that caregivers with high EOI experienced higher levels of ‘intrusion’ and ‘alertness’, compared to caregivers with low EOI.

One way to interpret these findings in relation to Metacognitive Therapy (MCT) and the S-REF model (Wells and Matthews, 1996; Wells, 2000), is that caregivers respond with anxiety and distress to the crisis of having a close one being diagnosed with schizophrenia, which is a normal and healthy reaction. However, some caregivers tend to respond with more unhelpful coping such as over-intrusiveness, self-sacrificing, over-protective behavior, exaggerated emotional response or over-identification with the patient. According to Wells (2000), metacognitions such as beliefs about worry concerning uncontrollability and danger may lead to a perseveration of unhelpful coping in terms of rumination and worrying, suppression of threatening thoughts and hyper-vigilant monitoring of potential threats. This form of ‘coping’ tends to backfire and cause more emotional suffering and distress. The coping strategies often work in the short-term, by giving some sense of control over the situation, but become detrimental in the long-term causing higher levels of emotional distress in the caregivers, and possibly also the persons with psychosis.

The view of EOI as a coping mechanism, that is the caregiver’s attempt to bring the illness under control by being over-protective, over-intrusive or self-sacrificing, corresponds with other findings (van Os et al., 2001). According to McNab et al. (2007), relatives who engage in these behaviors also hold beliefs in the usefulness of them in terms of dealing with the illness. McNab et al. refer to these as ‘utility beliefs’. From a CBT and MCT perspective, these behaviors would be referred to as ‘safety behaviors’ (Beck, 1979; Wells, 2000). While these reactions are perfectly understandable when faced with a traumatic experience, they are associated with higher levels of distress in caregivers and are to be considered as ‘unhelpful coping’ from Wells’ metacognitive model (Wells and Matthews, 1996; Wells, 2000). However, as Boye et al. (1998) pointed out, while most caregivers tend to fluctuate and be less over-involved with time, some caregivers present with continuously high EOI. Boye et al.’s study found that the relatives who remained high EOI at follow-up had higher scores on measures of ‘intrusion’ and ‘psychophysiologic activation’, which according to the authors, could be associated with excessive worrying and suppression of uncomfortable thoughts and emotions. These results correspond to the exploratory post-hoc analyses in this study, in which caregivers with high EOI also had significantly higher mean scores of both ‘intrusion’ and ‘psychophysiologic activation’ than caregivers with low EOI.

Taken together, these findings lend preliminary support for using Wells’ metacognitive model in family work for persons with psychosis. One promising research avenue could be to examine the more specific mechanisms in which metacognitions control attention through the activation of the CAS, as found in many

other psychological difficulties such as anxiety, depression and PTSD (Papageorgiou and Wells, 2001; Wells and Sembi, 2004b; Wells, 2010). Caregiver distress is a natural reaction to being in a difficult situation and often involves experiences such as loss, grief, anxiety and shock. However, high levels of caregiver distress also seem to be related to excessive worry and (unhelpful) attempts to control thinking and/or the illness. This is predicted by the CAS and found to be a trans-diagnostic feature of mental illness, in fact of all human suffering. However, EOI has also been found to be helpful for some persons with psychosis in the early phase of illness, as they may be in need of someone being over-protective and self-sacrificing (Bentsen et al., 1996a; van Os et al., 2001). The crucial thing for caregivers then, is to step back after the initial stages of illness in order to allow for independence in their relative and to avoid detrimental consequences to their own mental well-being.

The current study is a first step toward understanding the role of metacognitions in caregiver distress, thus opening up for the possibility of using interventions from the so-called 'third-wave' cognitive behavioural therapy (Hayes, 2004), or more recently referred to as 'contextual behavior therapies' (Hayes et al., 2011). Contextual behavior therapies include therapies such as Acceptance and Commitment Therapy (ACT), Metacognitive Therapy (MCT), Dialectic Behavior Therapy (DBT) and Mindfulness-based Cognitive Therapy (MBCT), and refer to their mutual focus on changing the context from which one relates to one's thoughts instead of trying to change the content of thoughts. Some examples of interventions from MCT are detached mindfulness, attention training and modification of dysfunctional metacognitions (Wells and Matthews, 1996; Wells, 2000).

## **5. Clinical implications**

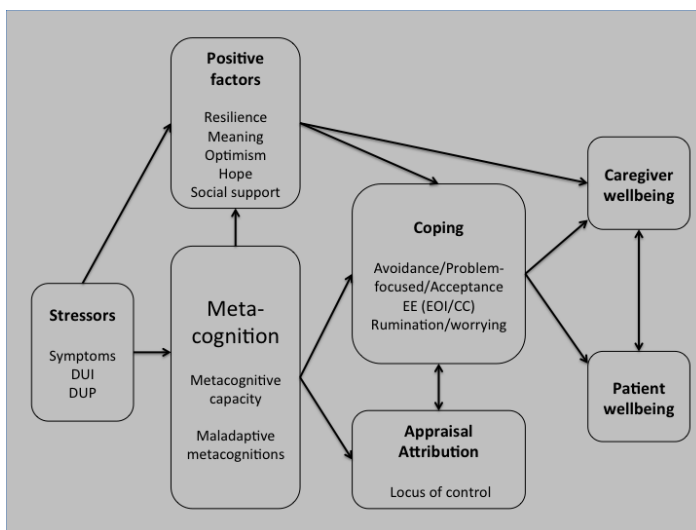
There are a number of clinical implications from this study. First, while EOI seems to be associated strongly with distress, CC does so only to a limited extent and the effect seems to disappear when controlling statistically for other variables. This corresponds to other studies, and highlights the need to address this early in treatment to prevent entrenchment of EOI and long-term grief. Not only in order to reduce the risk of relapse in persons with psychosis, as earlier suggested, but also with the aim of reducing the impact of caregiving and distress in caregivers. In addition, it seems important from the perspective of staying close to the ill family member, as unresolved loss and grief have been associated with a more distant relationship (Patterson et al., 2000). Second, caregivers appraisal of the situation seem to be more important than actual symptomatology and level of global functioning in the early phase, which highlights the need to address



these appraisals. Third, caregivers should get the opportunity to tell their story in order to help broaden the perspective of the caregiving experience, and developing their ability to form complex accounts of own and others' minds, thus allowing for both positive and negative experiences. Finally, metacognitions seem to play a role in caregiver distress, and may activate worry, rumination and dysfunctional coping, as they have been found to do in a range of psychological disorders. With replication and a broader examination of the metacognitive model, interesting new avenues of contextual interventions may open, including detached mindfulness, attention training and addressing of metacognitive beliefs in family work.

Based on the findings in this study as well as findings in the well-researched field, a preliminary model of all the psychologically variables involved in caregiver distress and well-being is proposed in Figure 3. As can be seen in the model, metacognitive capacity and specific metacognitions are considered as parallel processes, both influencing appraisal and coping, including EOI, avoidance and repetitive thinking such as excessive worrying and ruminations.

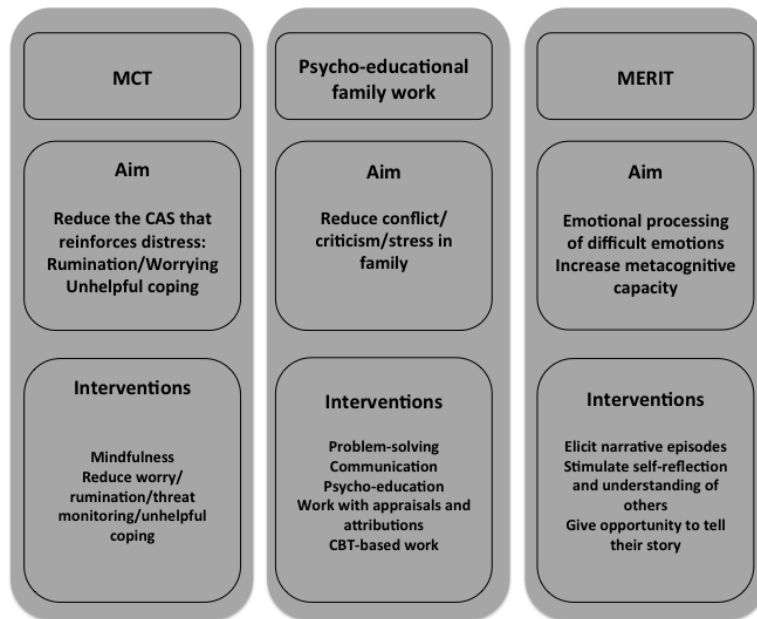
**Figure 3.** Psychological factors involved in caregiver distress



*Note:* DUI = Duration of Illness; DUP = Duration of Untreated Psychosis; EE = Expressed Emotion; EOI = Emotional Over-Involvement; CC = Critical Comments.

Figure 4 presents an overview of the aims and interventions suggested by the three 'therapeutic schools' involved in the current study, well knowing that only fractions of the theories have been tested out directly with regards to caregiver distress.

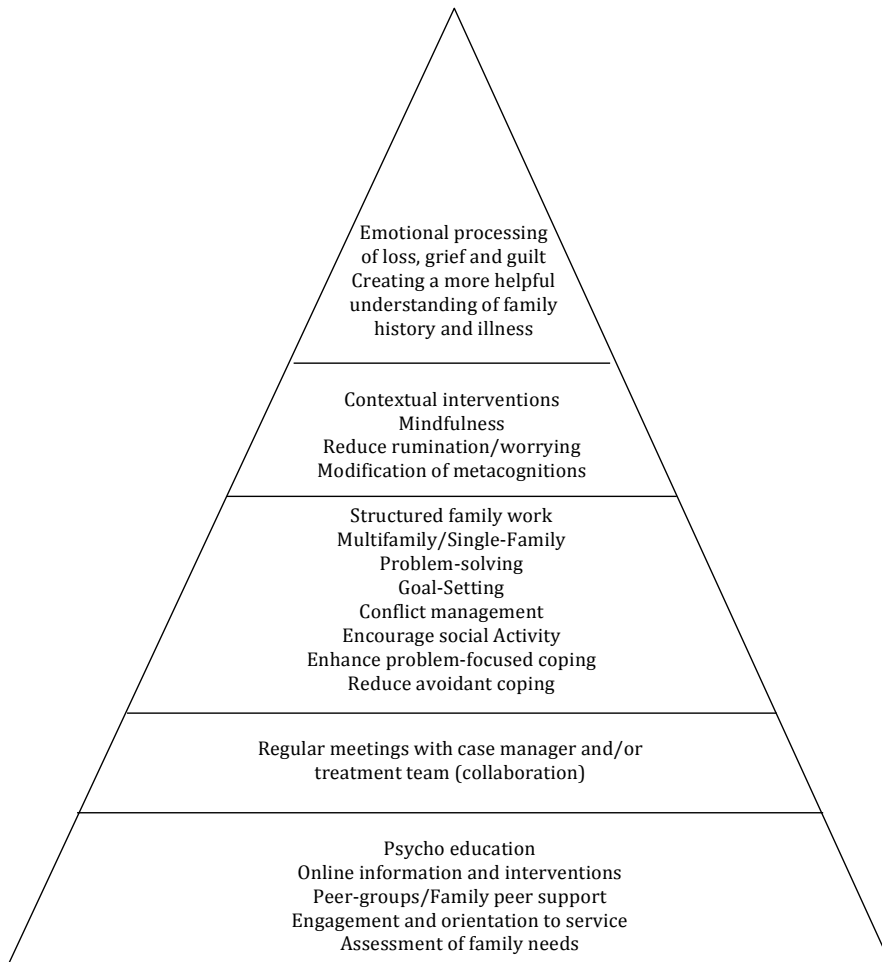
**Figure 4.** Model of three approaches to family support



*Note:* MCT = Metacognitive Therapy (Wells, 2000); MERIT = Metacognitive and Reflective Insight Therapy (Lysaker et al., 2014); CAS = Cognitive Attentional System; CBT = Cognitive Behavior Therapy.

Finally, a need-based model of family work for caregivers of persons with first-episode psychosis is proposed in Figure 5. The obvious starting point is to acknowledge, that families vary in terms of how much help they need and/or want. For some, psycho-education, either accessed from the Internet or delivered by clinicians individually or in groups, is sufficient. Others feel they are best supported in peer-groups with other caregivers. And some caregivers would benefit from more intensive support ranging from more regular meetings with the case manager, through structured family work, to more comprehensive interventions such as family therapy or individual psychotherapy. At the apex of the pyramid, there are interventions based on the metacognitive framework described in this thesis. Again, caregivers are different and have different needs and preferences with regards to therapeutic interventions. Some may benefit from getting an opportunity to tell their family story in detail, developing a more complex narrative account of self and others, and thus developing their metacognitive capacity. Others may wish for more concrete coping skills in terms of mindfulness to reduce excessive worrying and rumination.

**Figure 5.** A need-based model of family work in FEP



*Note:* Level 1-2: Normal crisis reactions; relatively good improvement of symptoms/remission; Level 3-4: Patient's problems are more complex and symptoms are more persistent; Comorbidity; multiple relapses; Caregivers are presenting with higher levels of Emotional Over-Involvement; Level 5: Long-term or high levels of distress; High levels of trauma or PTSD in caregivers; Entrenched in grief, loss and/or Expressed Emotion.

Two inter-dependent and equally important aspects seem relevant in therapy. The first is the capacity to form complex accounts of own and others' thoughts and feelings, which may be of particular importance to the emotional processing of grief, loss and guilt. The second is the ability to step back from one's thoughts and observe them as 'just thoughts' without being either caught up in them or avoiding them. This may be particularly important in EOI relationships with a lot of worrying, overprotection and attempts to control. The first is associated with

metacognitive focused therapy and mentalisation-based therapy (Allen and Fonagy, 2006; Dimaggio et al., 2007; Lysaker et al., 2011a). The latter might be informed by the so-called 'third-wave' cognitive behavioural therapy (Hayes, 2004), or more recently referred to as 'contextual psychotherapy' (Hayes et al., 2011), and includes interventions aimed at directing attention, and at reducing worrying and rumination, hypervigilance and avoidant coping. However, while the model has a hierarchical structure, in practice the need for and use of interventions go beyond this structure. As an example, many would benefit from interventions such as mindfulness, which is a general skill to handle any psychological challenge one is faced with, and not exclusively focused on high levels of caregiver distress. The same can be said about metacognitively focused therapy to help process loss and grief, as well as developing a more comprehensive account of the family and the caregiving situation.

## **6. Limitations and strengths**

There are some overall limitations to this study. First, self-report biases and social desirability may have contributed to errors in the measures we used. Second, the cross-sectional design does not allow for firm causal inferences. It is not possible to rule out for instance, that distress was increasing the report of negative appraisals or over-involvement. While the application of mediation analysis has been justified by experts in the field (Hayes, 2013), future studies should also address causality in longitudinal and experimental designs. Finally, caregiver inclusion was based on consent by the patients, which represents a selection bias. The unwell patients may be more inclined to refuse participation, as noted by Friis et al (2004). There is also the possibility that patients who had a good relationship with their caregivers were inclined to consent, which also may be reflected in the low representation of caregivers with high CC.

The overall strength of the study is the relatively large sample drawn from a specialist early intervention service within a defined catchment area, which increases the likelihood of a sample that is representative of a specifiable target population. In addition, the study presented in paper three, adjusted for caregiver- and patient characteristics and included two caregivers per patient in a multilevel analysis. Most studies use only one caregiver (Patterson et al., 2005), selected by asking the person with psychosis to choose the significant other with whom they are closest, or choosing the higher of two scores to determine the EE level in the household (López et al., 2004). The inclusion of two caregivers possibly explains why we have more male participants compared with other studies on caregiver distress. Most studies include only one significant other which typically is the mother (Cotton et al., 2013; Onwumere et al., 2011b).

## 7. Implications for future research

This study has examined some novel ideas in the field of family work and caregiver distress, and the findings lend support to understanding some of the mechanisms involved from a metacognitive framework. There are a number of possible areas for future studies. First, from the systematic review it is clear that prospective designs and larger samples are needed to capture and model changes in trajectories of caregiver distress. It also highlighted the need to develop and test interventions found to be associated with distress such as avoidant coping and appraisal. Second, future studies should examine long-term effects of baseline levels of metacognitive capacity, which may be important both in terms of how recurring challenges with a psychotic illness are dealt with as well as how this relates to individual differences in appraisal and EE. One hypothesis would be that EOI is lower at follow-up in persons with high capacity for metacognition at baseline, that is, they may be better able to step back from over-involvement. Third, future studies should test the S-REF model more comprehensively, by including measures of rumination, worrying, thought suppression and coping. One interesting avenue, related to this, would be examining caregivers level of acceptance coping and 'experiential avoidance' (Hayes et al., 1996). Interventions based on so-called third-wave CBT or contextual behaviour therapies, including mindfulness-based interventions, should be modelled and tested for efficacy in family work. Finally, while caregiver distress warrants focus in itself, future studies should also model the reciprocal influence of EE, metacognition and distress and patient characteristics including symptoms, level of functioning, quality of life and recovery.

## 8. Conclusions

The overall goal of the current study was to get a better understanding of what and how psychological factors relate to caregiver distress in first-episode psychosis, thus getting closer to answering the questions as what makes some caregivers more distressed than others.

A systematic review was carried out which found twelve empirical studies investigating the relationship between distress and psychological factors within caregivers. Moderate to large effect sizes supported the importance of subjective appraisal, coping and emotional over-involvement. Moreover, the effect of subjective appraisal and emotional over-involvement was tested and these were found to predict caregiver distress, after controlling for patient symptomatology and global functioning. From this, it may be asserted that caregivers' subjective experience and appraisal, together with an over-involved behaviour style, seem

more important than illness characteristics in the early phase of a psychotic disorder. Finally, the concept of metacognition, as approached from two different perspectives, was found to be related to caregiver distress and caregiver experience. First, the capacity to form complex representations of self and others, that is, understanding their thoughts and emotions, were related to more reports of positive caregiver experiences. Second, specific metacognitions, which are involved in the control and monitoring of attention, were also found to be associated with caregiver distress. From the metacognitive theory, these metacognitions are considered to activate repetitive thinking such as worrying and rumination, as well as unhelpful coping strategies, which together increase psychological distress. Emotional over-involvement, including being too protective, intrusive and self-sacrificing may represent one such coping strategy that increases the level of distress in caregivers.

With replication there might be important clinical implications in terms of a broadening of the interventions offered in treatment services to reduce caregiver distress and to increase a focus on caregivers' need for support independently of their ill family member. There is substantial data supporting the need for clinicians to address negative subjective appraisals, avoidant coping and over-involved behaviour. There also seems to be preliminary basis for using interventions from contextual behaviour science, such as mindfulness-based interventions, to reduce excessive worry and rumination. Finally, helping caregivers to develop complex accounts of the caregiving experience, including both positive and negative experiences, and by creating a space to tell their story seem important in addition to standard family work models such as psycho-education and problem-solving. However, more studies are needed to replicate, expand on and test interventions based on the findings.

## **9. Summaries**

### *9.1. Summary in English*

#### **Background**

Contrary to traditional pessimistic beliefs about persons with psychosis and schizophrenia, many can achieve recovery or significant improvement over time, especially if they receive adequate support at the time of the first-episode of psychosis (FEP). Nonprofessional caregivers, usually family members, often play an important role in the recovery process and a supportive family has been found to enhance recovery. Family involvement thus, is recognised as a central aspect of the treatment of persons with FEP. While caring for someone with a mental disorder involves both positive and negative feelings, studies have shown that care-

givers often experience high levels of burden and distress, including anxiety, depression, stigma, social isolation, increased family conflict and economic strain. However, most studies have examined distress and burden in more chronic forms of illness and many of the interventions have been developed within this context. For this reason, a number of authors have addressed the need for developing interventions more specifically tailored to FEP families. One challenge has been the limited understanding of caregiver distress, especially its psychological underpinnings, from which to build a broader base of interventions.

## Aim

The overall goal of this PhD was to get a better understanding of what and how psychological factors relate to caregiver distress in first-episode psychosis. With this in mind, the specific aims were to: 1) systematically review the current literature that has examined psychological factors related to caregiver distress; 2) examine subjective appraisals and emotional over-involvement as predictors of caregiver distress, while addressing some of the methodological limitations in earlier studies; 3) examine the concept of metacognition from two different theoretical perspectives as predictors of caregiver distress.

## Design

Within a cross-sectional design, a total number of 99 patients and 154 caregivers enrolled in Opus, participated between April 2011 and April 2013. The following semi-structured interviews, rating scales and questionnaires were used: GHQ-30, ECI, IPII, MAS-A, MCQ-30, FQ, PANSS and GAF. The PhD study comprises three empirical papers and a systematic review that together examine how various psychological variables are associated with caregiver distress in first-episode psychosis. Two papers have been published in international peer-reviewed journals and two have been submitted and are under review.

## Results

The systematic review of psychological factors related to caregiver distress identified nine different psychological variables that have been empirically examined, which have here been categorized in the following non-mutually exclusive groups: coping, appraisal/attribution and interpersonal response. These were based on 14 papers describing 12 studies together comprising 977 caregivers of persons with first-episode psychosis. Moderate to large effect sizes supported the importance of appraisal, coping and emotional over-involvement in predicting caregiver distress. However, many of the studies included had some methodological limitations in terms of cross-sectional data, small sample-sizes, confounding variables not being accounted for, and a wide variation in outcome measures. Thus, the conclusions must keep these limitations in mind.

The data from the interviews, revealed high levels of caregiver distress, reflected in the finding that thirty-seven per cent of the caregivers were suffering from clinically significant distress. Moreover, when testing the effect of subjective appraisal and emotional over-involvement, these were found to predict caregiver distress, after controlling for patient symptomatology and global functioning. From this, it appears that caregivers' subjective experience and appraisal, together with an over-involved behaviour style, seem more important than illness characteristics in the early phase of a psychotic disorder. Finally, the concept of metacognition, as approached from two different perspectives, was found to be related to caregiver distress and caregiver experience. First, the capacity to form complex representations of self and others, that is, understanding their thoughts and emotions, is related to more reports of positive caregiver experiences. Second, specific metacognitions, which are involved in the control and monitoring of attention, were also found to be associated with caregiver distress. According to the metacognitive theory, these metacognitions activate repetitive thinking such as worrying and rumination, as well as unhelpful coping strategies, that together increase psychological distress. Emotional over-involvement, including being too protective, intrusive and self-sacrificing may represent one such coping strategy that increases the level of distress in caregivers.

### Clinical implications

There might be important clinical implications in terms of a broadening of the interventions offered in treatment services to reduce caregiver distress and a higher focus on caregivers' need for support independent of their ill family member. There is substantial data supporting the need for clinicians to address negative subjective appraisals, avoidant coping and over-involved behaviour. There also seems to be preliminary basis for using interventions from contextual behaviour science, such as mindfulness-based interventions, to reduce excessive worry, rumination and distress. Finally, helping caregivers to develop complex accounts of the caregiving experience, including both positive and negative experiences, by creating a space to tell their story seem important in addition to standard family work models such as psycho-education and problem-solving.

## *9.2. Summary in Danish*

### Baggrund

Behandling af psykose og skizofreni har traditionelt set været forbundet med stor grad af pessimisme. Nyere forskning viser imidlertid at de fleste opnår betydelig forbedring over tid og mange kommer sig helt, især hvis de modtager adækvat støtte på et tidligt stadium af forløbet. Samarbejde med familien er en



vigtig del af den tidlige indsats, og forskning har vist at en støttende familie øger sandsynligheden for at komme sig. At være pårørende til mennesker med psykose er forbundet med mange følelser, positive såvel som negative. Mange beskriver dog stor grad af belastning såsom ængstelse, tristhed, stigmatisering og social isolation.

De fleste studier har imidlertid undersøgt pårørende til personer med mere kroniske former for psykose, og den familiebehandling der generelt er til rådighed er blevet udviklet til denne målgruppe. Det er derfor udtrykt behov for at udvikle familieinterventioner der mere specifikt er rettet imod de særlige udfordringer i den tidlige fase af psykosen. En af udfordringerne har imidlertid været en begrænset forståelse af hvilke psykologiske faktorer der er involveret i pårørendebelastning, hvilket kan hæmme udviklingen af nye interventioner.

### Formål

Det overordnede mål med dette studie var derfor at få en større forståelse for hvilke psykologiske faktorer, der er involveret i pårørendebelastning ved førstegangs psykose. Med udgangspunkt i dette, var de mere specifikke mål: 1) At lave et systematisk review over psykologiske faktorer relateret til pårørendebelastning; 2) at undersøge betydningen af pårørendes subjektive vurdering og emotionelle overinvolvering for pårørendebelastning; og 3) at undersøge hvordan begrebet metakognition er relateret til pårørendebelastning og hvordan familiearbejde kan forstås ud fra en metakognitiv forklaringsmodel.

### Design

Undersøgelsen er et tværsnitstudie af 99 patienter og 154 pårørende, der fik behandling i Opus mellem april 2011 og april 2013. Følgende semi-strukturerede interviews, rating scales og spørgeskemaer blev anvendt: GHQ-30, ECI, IPII, MAS-A, MCQ-30, PANSS og GAF. Afhandlingen består af tre empiriske artikler samt et systematisk review. To af artiklerne er publiceret i internationale tidsskrifter og to er indsendt og under vurdering.

### Resultater

I studiets første del, fandt det systematiske review at ni forskellige psykologiske variable er blevet empirisk undersøgt. De er i denne afhandling blevet kategoriseret i følgende grupper: coping, subjektiv vurdering/attribution og interpersonlig respons. Der var moderate til store effektstørrelser for sammenhængen mellem henholdsvis subjektiv vurdering, coping og emotionel overinvolvering, og grad af pårørendebelastning. Resultaterne er baserede på 12 studier beskrevet i 14 artikler, som tilsammen omfattede 977 pårørende til personer med førstegangs psykose. Reviewet peger også på en række metodiske be-

grænsninger ved de inkluderede studier, såsom tværsnitsdata, små samples, confounders, samt anvendelsen af mange forskellige mål for pårørendebelastning.

Den anden del studiet, der omhandlede subjektiv vurdering og emotionel overinvolvering viste for det første at de pårørende rapporterede en høj grad af belastning; 37 procent havde en score svarende til 'klinisk signifikant belastning'. Pårørendes grad af belastning var relateret til deres subjektive vurdering og grad af emotionel overinvolvering, også efter at der blev taget højde for patienternes symptom- og funktionsniveau. Med andre ord, kan det se ud til at pårørendes belastning i den tidlige fase af psykoseforløbet er mere påvirket af deres subjektive vurdering af situationen, samt grad af emotionel involvering, end af hvordan den unge mere 'objektivt' set har det.

Den tredje del af studiet viste at metakognition havde betydning for pårørendes subjektive oplevelse og grad af belastning. For det første, var pårørendes evne til at danne komplekse repræsentationer af egne og andres tanker og følelser, relateret til mere positive beskrivelser af det at være omsorgsgiver. For det andet, var specifikke metakognitioner, der er involveret i kontrol og monitorering af opmærksomheden, relateret til pårørendes grad af belastning. Ifølge den metakognitive teori, aktiverer og vedligeholder disse metakognitioner en tænkning præget af bekymring og ruminering, i tillæg til dysfunktionelle copingstrategier, der tilsammen øger psykologisk belastning. Emotionel overinvolvering, hvor den pårørende for eksempel er overbeskyttende og tilsidesætter sig selv og egne behov, kan anskues som dysfunktionel coping og forbundet med forøget belastning indenfor den metakognitive model.

### Kliniske implikationer

Samlet set peger dette studiet på mulighederne for en udvidelse af det interventionsrepertoire, der i dag tilbydes familier med førstegangs psykose. Undersøgelsen understøtter vigtigheden af at reducere pårørendebelastning i den tidlige fase, samt at sætte større fokus på pårørendes selvstændige behov for støtte, uafhængigt af familiemedlemmet med en psykoselidelse. Der synes at være tilstrækkelig evidens for, at behandlere med fordel kunne arbejde med de pårørendes subjektive vurdering af omsorgssituationen. Her med særligt fokus på at mindske en alt for negativ vurdering af psykosen og dens påvirkning, samt arbejde med at reducere undvigende coping og emotionel overinvolvering. Der synes også at være basis for at anvende interventioner fra kontekstuel adfærdspsykologi, såsom mindfulness-baserede interventioner for at reducere overdreven bekymring og ruminering. Endelig kan pårørende hjælpes til at udvikle mere komplekse og nuancerede repræsentationer af sig selv, patienten og pårørenderollen, der inkluderer både positive og negative oplevelser. Dette kan gøres ved at tilbyde dem mere plads til at fortælle familiens historie samt arbejde med konkrete narrative episoder. Disse tilgange kan således understøtte og supplere

de mere almindelige modeller for familiearbejde såsom psykoedukation, problemløsning og konflikthåndtering.

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## Papers

### Paper 1

Jansen, J. E., Gleeson, J., Cotton, S. (2014). Towards a better understanding of caregiver distress in first-episode psychosis: A systematic review of the psychological factors involved. In review

### Paper 2

Jansen, J. E., Haahr, U. H., Harder, S., Trauelsen, A. M., Lyse, H. G., Pedersen, M. B., Simonsen, E. (2014). Caregiver distress in first-episode psychosis: the role of subjective appraisal, over-involvement and symptomatology. *Social Psychiatry and Psychiatric Epidemiology*.

### Paper 3

Jansen, J. E., Lysaker, P. H., Harder, S., Haahr, U. H., Lyse, H. G., Pedersen, M. B., Trauelsen, A. M., Simonsen, E. (2013). Positive and negative caregiver experiences in first-episode psychosis: Emotional overinvolvement, wellbeing and metacognition. *Psychology and Psychotherapy. Theory, Research and Practice*.

### Paper 4

Jansen, J. E., Harder, S., Haahr, U. H., Lyse, H. G., Pedersen, M. B., Trauelsen, A. M., Simonsen, E. (2014). The role of metacognitions in expressed emotion and distress: a study on caregivers of persons with first-episode psychosis. *Clinical Psychology and Psychotherapy*.

# Paper 1



## **Towards a better understanding of caregiver distress in first-episode psychosis: A systematic review of the psychological factors involved**

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## 1. Introduction

Caregivers of persons with first-episode psychosis (FEP) or schizophrenia play an important role in the process of recovery but often experience high levels of distress, including depression, anxiety and subjective experience of burden (Martens and Addington, 2001). An early understanding and assessment of this, followed by appropriate support, is an important part of a comprehensive early intervention programme to prevent chronic problems and long-term distress in the whole family following the onset of psychosis (McGorry et al., 2008).

While early studies often described the caregiver situation in terms of “objective” burden (Baronet, 1999; Yarrow et al., 1955), later studies seem to agree on a stress-appraisal-coping framework in understanding how subjective experience of distress develops (Lazarus and Folkman, 1984; Szmukler et al., 1996). In other words, the extent to which these stressors are related to a caregiver’s negative mental health is dependent on the caregiver’s appraisal, or subjective evaluation, of the stressors and his/her ability to cope with them. A number of illness-characteristics have been found to be associated with caregiver distress, including negative symptoms, reduced levels of functioning and ‘disruptive behaviour’ (Addington et al., 2003; Boye et al., 2001; Tennakoon et al., 2000; Wolthaus et al., 2002). However, the findings are inconclusive and others have not found these to be related (Carrà et al., 2012; Heikkilä et al., 2002). Studies have also looked at caregiver demographics such as age, gender and ethnicity, but the findings here are also inconclusive (Baronet, 1999; Boydell et al., 2013).

While there are a limited number of studies looking at FEP, data suggest that caregivers at this stage report a higher levels of distress compared to family members of individuals who have a more chronic course of illness (Martens and Addington, 2001). In the early phase of psychosis, caregivers face a number of issues, such as shock, grief and poor understanding of illness and the psychiatric system (Addington and Burnett, 2004). Also, incidences such violence, disruptive behaviour and

forced admission can be traumatizing, sometimes warranting a diagnosis of posttraumatic stress disorder in caregivers themselves (Onwumere et al., 2011a).

In relation to reducing the distress, and helping caregivers to help their psychotic relatives, one of the most influential concepts in psychosocial research on psychosis has been Expressed Emotion (EE). EE refers to interactions between persons with psychosis and their caregivers that are characterised by either critical comments (CC)/hostility or emotional over-involvement (EOI) (Brown et al., 1962). EE is a robust predictor of relapse and symptom exacerbation in schizophrenia and other mental illnesses (Butzlaff and Hooley, 1998; Wearden et al., 2000). The concept has been of considerable importance in the development of family intervention and family support programmes in psychosis (Glynn, 2012; Pfammatter et al., 2006). However, the relevance of the concept in the early phase of psychosis has been questioned. Findings relating to the association between EE and relapse are mixed in FEP (Bird et al., 2010; Linszen et al., 1996) and certain aspects of EE, especially EOI, might be useful for persons with psychosis in the early phase of illness (Bentsen et al., 1996). As an example, EOI has been associated with greater participation in care, including supporting adherence to treatment and attention to needs that are unmet by the treatment system (van Os et al., 2001). For this reason some researchers caution against relying too much on EE-focused interventions in FEP, and argue that, if EE is too crudely targeted in this population, it may have detrimental effects (Gleeson et al., 2010; Jansen et al., 2013; Linszen et al., 1996).

In the current research literature, the experience of caregiving is conceptualised as a complex phenomenon, influenced by characteristics of the person with psychosis as well as the caregiver, and comprising both negative and positive experiences (Jansen et al., 2013; Joyce et al., 2003). In addition, while earlier studies focused on caregiver distress in terms of its relationship to EE and relapse, recently there has been an increased recognition of the needs of caregivers independent of the course of illness in their ill relative (Kuipers, 2010; Lobban et al., 2013).



However, less is known about the psychological underpinnings of caregiver distress in first-episode psychosis, and there seems to be a limited set of psychological models accounting for variations in caregiver distress. An improved understanding of this would assist in developing need-based support to caregivers of persons with FEP. Furthermore, by studying caregiver distress early in the course of illness, findings are less likely to be confounded by duration of the illness and factors associated with more chronic forms of psychosis.

### 1.1. Aims of the review

Through this current review we sought to identify empirical studies that have investigated psychological factors accounting for caregiver distress in FEP, with the aim of summarising key findings and critically evaluating the methodology used.

This review addresses the following questions:

- 1) What psychological processes, resources or competences within caregivers have been empirically tested in studies to account for variations in caregiver distress and what are the findings?
- 2) What is the methodological quality of these studies?

## **2. Material and method**

### 2.1 Search strategy

Systematic searches using Cochrane methodology were performed to find relevant trials and, observational studies from the following databases: PsycINFO, EMBASE and Cochrane Central Register of Controlled Trials (CENTRAL). Databases were searched up to March 2014. The computerized search used the following strategy: (\*psychosis/ or exp schizophrenia/) AND (caregiver\* OR carer\* OR

parent\* OR famil\*) AND (distress\* OR burden\* OR wellbeing OR well being OR stress\* OR depress\* OR anxiety OR loss OR grief) AND (attribution\* OR coping OR attachment OR metacognition OR “metacognitive belief\*” OR “expressed emotion\*”).

Online titles and abstracts were reviewed after duplicates were removed. Papers that did not meet the inclusion criteria were discarded, and full text papers were retrieved for potentially eligible studies. Electronic searches were supplemented by hand-searching reference lists of retrieved papers, previous reviews and book chapters. We also manually searched relevant journals such as JAMA Psychiatry, Psychiatry Research, Journal of Nervous and Mental Disease, Schizophrenia Bulletin, Schizophrenia Research, Psychology and Psychotherapy: Theory, Research and Practice, Clinical Psychology Review, Early Intervention in Psychiatry, Acta Psychiatrica Scandinavica, Psychological Medicine and British Journal of Psychiatry. Finally, several experts were consulted about potential additional studies.

## 2.2. Inclusion and exclusion criteria

Inclusion criteria were that studies: 1) reported on caregiver distress or wellbeing in caregivers of persons with FEP and schizophrenia spectrum disorders; 2) empirically tested hypotheses regarding psychological factors within caregivers accounting for caregiver distress and 3) were published in peer-reviewed journals in English. Excluded were: 1) studies with a primary aim of investigating predictors of EE or studies without a hypothesis on how psychological factors within caregivers explain variance in caregiver distress, 2) secondary publications (reviews, commentaries, editorials and letters), (3) theses, dissertations and conference papers, 4) qualitative studies and case reports. No date restrictions were applied to the selection of studies. Two reviewers (JEJ, JG) independently assessed relevant articles for inclusion, based on a template developed for this purpose. Any disagreements were resolved through discussion.

## 2.3 Data extraction and tabulation of data

A data extraction template was developed based on the Cochrane Consumers and Communication Review Group's data extraction template (2011) and the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) checklist (Moher et al., 2009). Two reviewers (JEJ, JG) tabulated key information independently and discrepancies were reconciled by consensus.

#### 2.4. Quality criteria

All selected papers were evaluated in terms of methodological quality to assess risk of bias and generalizability. This was based on the STROBE (STrengthening the Reporting of Observational studies in Epidemiology) guidelines (von Elm et al., 2007) and PRISMA checklist (Moher et al., 2009), of which central aspects were implemented in the data extraction template. As suggested by Beller (2013), we specified a set of methodological components for description, assessment and evaluation. These were: a) confounding factors including duration of untreated psychosis, duration of illness, time of caregiver assessment and symptomatology b) independence of data collection; c) definition of outcome variables; and d) verification of diagnosis.

### 3. Results

The search and exclusion process is summarized in Figure 1. The initial data base search produced 870 abstracts. A total number of 14 papers describing 12 studies, published between 1999 and 2013, met the inclusion criteria. These are summarized in Table 1.

#### 3.1. Study characteristics

Most of the studies used consecutive sampling from a defined catchment area (91.7%, n = 11), except for one, which recruited a convenience sample (Möller-Leimkühler and Obermeier, 2008; Möller-Leimkühler, 2006, 2005). Most studies recruited from a specialist early psychosis service setting

(66.7%, n=8) and one study (8.3%) did not specify the recruitment site. The characteristics of the participants who chose not to participate were not reported in the studies included.

#### a) Persons with psychosis

There were a total of 1002 persons with FEP in the included studies. Their mean age was 24.7 years (SD = 6.6), based on data from eight studies (70.3%, n = 705). Of the participants with psychosis included, 65.3% (n = 430) were male and 34.7% (n = 229) were female. Five studies reported no data regarding gender (343 participants). Six of the studies (54.1%, n = 542) reported verification of the psychiatric diagnoses using standardized instruments such as the Schedule for Clinical Assessment in Neuropsychiatry (SCAN; WHO, 1992), Operational Criteria Checklist (OPCRIT; McGuffin, 1991) or the Structured Clinical Interview (SCID-I; Spitzer, 1992).

#### b) Caregivers

There was data on a total of 977 caregivers in the studies included. Based on data from nine studies the mean age was 47.32 (SD = 10.79). No data regarding age were reported on 341 caregivers (34.9%). Based on the data from seven studies, 70.6% (n = 361) were female and 29.4 % (n = 150) were male. Five studies reported no data regarding gender (47.7%, n = 466). Most of the caregivers were parents (74.2%, n = 725) and 81.8% (n = 742) were living with the person with FEP. Two studies did not report on cohabitation (n = 70). Education level of caregivers was reported in only three studies, which precludes any meaningful description. Based on the data from six studies, 61.6% (n = 307) of caregivers were employed either part- or full time. No data regarding employment status were provided in six studies (68.6%, n = 670)

### 3.4. Study quality

There were a number of strengths in the studies included, such as reliable and valid distress measures,

recruitment from FEP specialist service, which increased the likelihood of a representative sampling, and hypotheses that were derived from a stress-appraisal-coping framework. However, there were some considerable methodological issues with regard to the variables we set out to evaluate. There were few studies on caregiver distress in FEP and only six of the studies included had caregiver distress as a main variable of interest. Additionally, most studies used cross-sectional data, had relatively small samples and lacked a comparison group to enable distinguishing the effect of being close to a person with psychosis from that of being close to a person with other mental health disorders. Finally, none of the studies specified the time of family assessment and only four reported on the duration of illness or duration of untreated psychosis. With few exceptions, these variables were not accounted for in most statistical analyses of distress (Addington et al., 2003; Onwumere et al., 2011b).

### 3.5. Measurement of caregiver distress

Only one of the studies included had an explicit definition of distress (Möller-Leimkühler, 2005), and the studies used a variety of overlapping concepts and descriptions of caregiver experiences (see Table 3 for an overview). Across the 12 studies, 15 different measures of caregiver distress or psychological well-being were employed. The number of distress measures included within each individual study ranged from one to four and three studies used more than two different measures. None of the studies included used measures of physical illness or visits to the general practitioner as indicators of distress. Caregivers' distress was the main variable of interest in six (50%) of the studies, while three (25%) defined EE as the main focus. In the remaining studies it was not made clear whether distress was to be considered as a dependent or an independent variable.

Of the 15 measures, the *Experience of Caregiving Inventory (ECI)* (Szmukler et al., 1996) and the *General Health Questionnaire (GHQ)* (Goldberg and Williams, 1988) were the most frequently used and will be described below.

The *ECI* is a measure of caregivers' appraisal of the impact of the illness and comprises ten subscales: eight negative (difficult behaviors, negative symptoms, stigma, problems with service, effects on the family, the need to provide back-up, dependency and loss) and two positive (rewarding personal experiences and good aspects of the relationship). The *ECI* has been shown to have excellent psychometric properties (Szmukler et al., 1996). Eight of the included studies used the *ECI* (66.7%), five as a dependent measure (Alvarez-Jiménez et al., 2010; Cotton et al., 2013; Jansen et al., 2013; Patterson et al., 2005; Raune et al., 2004) and three as an independent measure (Addington et al., 2003; Gleeson et al., 2010; Tomlinson et al., 2013) predicting depression, scores on *GHQ* and impact of illness. Only five reported on the positive subscales (Addington et al., 2003; Cotton et al., 2013; Gleeson et al., 2010; Jansen et al., 2013; Patterson et al., 2005).

The *GHQ* is a screening instrument covering a range of psychiatric symptoms. It was originally developed as a 60-item questionnaire, and was later adapted into 30-, 28- and 12-item versions. The *GHQ* can be scored as a continuous measure with higher scores indicating increased levels of mental distress. It can also be scored as a 'case-score', indicating the presence or absence of clinically significant distress. The *GHQ* has also been shown to have excellent psychometric properties (Goldberg and Williams, 1988). Six of the included studies used the *GHQ* (50%). Five studies used the *GHQ-28* (Alvarez-Jiménez et al., 2010; Gleeson et al., 2010; McNab et al., 2007; Onwumere et al., 2011b; Raune et al., 2004) and one study used the *GHQ-30* (Jansen et al., 2013).

The following distress measures were used in one or two studies: *The Beck Depression Inventory* (BDI; Beck & Steer, 1987); *The Psychological General Well-Being Scale* (PGWB; Beck, 1993); *The Kessler Psychological Distress Scale* (K10; Andrews & Slade, 2001); *Patient rejection scale* (Addington et al., 1992); *The Family Burden Questionnaire* (FBQ; Pai & Kapur, 1981); *The Symptom Checklist-90* (SCL-90; Derogatis, n.d.) (used in two studies); *The Burden Interview* (Zarit and Zarit, 1983); *The Lancashire Quality of Life Profile* (LQLP; Oliver, Huxley, Priebe, & Kaiser, 1997); *Befindlichkeitsskala* (Bf-S; von Zerssen, 1976); *The Calgary Depression Scale for Schizophrenia* (CDSS;

Addington et al., 1992); *Texas Revised Inventory of Grief* (TRIG; Miller, Dworkin, Ward, & Barone, 1990); *The Hospital Anxiety and Depression Scale* (HADS; Zigmond & Snaith, 1983); and *The Social Functioning Scale* (SFS; Birchwood, Smith, Cochrane, Wetton, & Copestake, 1990).

### 3.6. Psychological models and constructs examined and their measurement

Across the twelve studies, nine different psychological processes, resources or competences relating to caregivers were examined and empirically tested in terms of their association with caregiver distress. These were: appraisal, coping, attribution, loss, expressed emotion, metacognition, attachment, social cognition and utility beliefs which for the purpose of overview and synthesis, we have organized into three non-mutually exclusive groups below, namely (a) coping, (b) appraisal/attribution and (c) interpersonal response. The various psychological factors were assessed by the following rating scales: The Family Questionnaire (FQ; Wiedemann, Rayki, Feinstein, & Hahlweg, 2002); The Experience of Caregiving Questionnaire (ECI; Szmukler, Burgess, Herrman, Bloch, Benson, & Colusa, 1996); The Ways of Coping Checklist (WOC; MacCarthy, Kuipers, Hurry, Harper, & LeSage, 1989); The Psychiatric Disabilities Attribution Questionnaire (PDAQ; Krauss, 1989); The Health and Daily Living Form Indices of Coping (HDLFC; Moos, Cronkite, Billings, & Finey, 1984); The Dementia Management Strategies Scale (DMSS; Hinrichsen & Niederehe, 1994); The Metacognitions Assessment Scale-Abbreviated (MAS-R; Lysaker et al., 2005); The Illness Perception Questionnaire (IPQ; Barrowclough, Lobban, Hatton, & Quinn, 2001); 'Utility Beliefs' (McNab et al., 2007); The Stressverarbeitungsbogen (SVF; Janke & Erdmann, 1997); The Fragebogen zu Kompetenz- und Kontrollüberzeugungen (FKK; Krampen, 1991); The Neo Five-Factor Inventory (NEO-FFI; Borkenau & Ostendorf, 1993); The Five-Minute Speech Sample (FMSS; Magaña et al., 1986); The Cope instrument (COPE; Carver, Scheier, & Weintraub, 1989); The Texas Revised Inventory of Grief (TRIG; Miller et al., 1990); The Camberwell Family Interview (CFI; Vaughn & Leff, 1976); The Hinting Task (Corcoran et al., 1995); The Facial Expression of Emotion Stimuli (FEEST; Young, Perrett, Calder, Sprengelmeyer, & Ekman, 2002); The

Family Attitude Scale (FAS; Kavanagh et al., 1997).

(a) Coping

Five studies (Cotton et al., 2013; Hinrichsen and Lieberman, 1999; Möller-Leimkühler and Obermeier, 2008; Möller-Leimkühler, 2006, 2005; Onwumere et al., 2011b; Raune et al., 2004) explored associations between coping and distress. In a study that recruited two relatively large samples (in total  $n = 141$ ) (of different illness durations), Onwumere et al. (2011b) found that distress was associated with avoidant coping in caregivers ( $r = .51, p < .01$ ). There were no associations between caregivers' coping strategies and demographic variables (caregiver's sex, age and cohabitation with the patient), and avoidant coping remained associated with distress after controlling for illness length (partial  $r = .51, p < .01$ ). An association between caregiver burden and avoidant coping was also found by Hinrichsen and Lieberman (1999) even though the participants had a reasonably good emotional adjustment, with an average burden score between 'mild to moderate'. Multivariate analyses found avoidant coping to be associated with all three indices used by Hinrichsen et al. to measure emotional adjustment in caregivers – patient rejection ( $\beta = .37, p < .001$ ), burden ( $\beta = .39, p < .001$ ) and SCL-90 ( $\beta = .45, p < .001$ ) – while neither family nor patient characteristics were related to caregiver burden. Moreover, Cotton et al. (2013) found avoidant coping to be associated with psychological distress ( $\beta = .58, p < .001$ ), EOI ( $\beta = .51, p < .001$ ), CC ( $\beta = .29, p < .001$ ) and burden ( $\beta = .41, p < .001$ ). In their study, tension reduction, cognitive escape and optimistic coping together explained 50% of the variance in caregiver distress. In one of the few prospective studies, Möller-Leimkühler and colleagues (2008; 2006, 2005) found that emotion-focused coping predicted caregiver burden as measured by SCL-90 in a logistic regression (Wald  $\chi^2 = 7.39, p = .007$ ) and that general negative stress response predicted burden as measured by BF-S (Wald  $\chi^2 = 16.52, p \leq .001$ ), SCL-90 (Wald  $\chi^2 = 7.53, p = .006$ ) and LQ (Wald  $\chi^2 = 18.96, p \leq .001$ ) at two years follow-up. Of particular note, severity of illness, the type of symptoms, and psychosocial functioning at first admission were not associated with caregivers' ratings of objective burden or other stress indicators. Möller-Leimkühler (2005) concluded that both



EE and burden were “significantly associated with several psychosocial resources and dispositions of the relatives” (p. 223).

In summary, avoidance coping has consistently been shown to be associated with distress and burden even in the early stages of the illness, a finding that is consistent with studies of more long-term or chronic patients (Fadden et al., 2000; Kuipers et al., 2010, 2006) and qualitative studies (Gerson et al., 2011; Huang et al., 2008). There are few prospective studies and therefore it is difficult to assess the direction of these effects. Other studies have found a number of factors associated with avoidant coping, including internalised stigma (Yanos et al., 2008), insight (Cooke et al., 2007), shorter duration of untreated psychosis (Skeate et al., 2002) and high EE (Kuipers et al., 2006). To the best of our knowledge, no study has examined whether interventions aimed at reducing avoidant, or emotional coping, influence the level of caregiver distress. Coping based on acceptance, e.g., through mindfulness-based approaches, has also not been examined. However, this has been shown to reduce distress in a number of other studies, including anxiety (Vøllestad et al., 2012), depression (Hofmann et al., 2010) and psychosis (Khoury et al., 2013).

#### (b) Appraisal and attribution

Six studies (Addington et al., 2003; Gleeson et al., 2010; Hinrichsen and Lieberman, 1999; McNab et al., 2007; Patterson et al., 2005; Raune et al., 2004) explored how caregiver distress was related to one of the following: the way caregivers appraised the impact of illness, the extent to which they attributed control of illness to the patients, or the views they held regarding the effectiveness of their EE behaviour. Addington et al. (2003) found that caregivers’ appraisal of the impact and consequences of the illness, as measured by the ECI negative subscale, had the greatest association with their psychological well-being ( $\beta = .52, p < .05$ ) and that appraisal together with the patient’s age and age of onset explained 36% of the variance in caregivers’ psychological well-being. The authors also concluded that, while “the difficult experiences of being a caregiver are associated with patient variables such as negative and positive symptoms and poor social functioning ... at this early stage of

the illness specific patient characteristics seem to play a minor role in families' appraisal of their situation" (p. 288).

In the only randomised controlled study on caregiver intervention in FEP to be included, Gleeson et al. (2010) examined the effect of an intervention based on psychoeducation and relapse prevention strategies on caregiver appraisal. Their intervention showed a specific advantage in caregiver appraisals in relation to greater positive personal experiences of caring ( $F(1,62.17) = 4.19, p = .045$ ) and greater overall positive scores ( $F(1,63.58) = 4.39, p = .040$ ). However, this did not influence caregivers' level of distress as measured by GHQ-28. There were no significant association on the overall negative subscale. This finding casts doubt on whether the manipulation of positive appraisals alone is sufficient to improve distress, and suggests that negative appraisals instead need to be addressed. Another interpretation might be that distress is an unavoidable aspect of being close to someone with a psychotic disorder in the early phase of illness, and that other measures such as quality of life, social functioning and meaningfulness might be equally important to overall well-being. However, this obviously does not mean that reducing prolonged and unnecessarily high levels of distress is not an important objective of family intervention.

In a prospective study, Patterson et al. (2005) found that caregivers' appraisal of loss was related to their level of distress as measured by depression ( $r = .44, p < .01$ ) and burden ( $r = .43, p < .01$ ). Moreover, their appraisal of loss was associated with EOI but not CC at baseline. Interestingly, the appraisal of loss was reduced at nine months follow-up in those changing from high EOI to either high CC or low EE, suggesting that EOI might evolve into criticism as a way of easing the unresolvable loss. Hinrichsen and Lieberman (1999) found poorer emotional adjustment as measured by the Burden Interview in caregivers who attributed patients' psychiatric problems to moral failings ( $\beta = .32, p < .05$ ) or to psychological problems from earlier life ( $\beta = .28, p < .05$ ). With the Burden Interview as outcome measure, attribution was found to explain 45% of the variance in caregiver distress. Finally, in an attempt to understand reasons for engaging in EE-behaviour, McNab et al. (2007) assessed

caregivers' 'utility beliefs', i.e. their beliefs in the utility of criticism and self-sacrifice when interacting with the person with psychosis. They found that some caregivers often thought self-sacrifice or coercive behaviour would be helpful in changing patients' behaviour for the better. As hypothesised, they found that caregiver distress was associated with the endorsement of utility beliefs concerning person-focused criticism ( $r = .24, p < .05$ ) and self-sacrifice ( $r = .27, p < .05$ ).

In summary, there is some consistency across findings that caregivers' appraisal of various illness related factors – ranging from control, symptoms, loss, interpersonal functioning and utility of EE behaviour – are predictive of the level of distress and burden that caregivers experience. The findings correspond with those of studies of more chronic forms of psychosis (Barrowclough et al., 2001; Szmukler et al., 1996). They are also in agreement with the stress-appraisal-coping model (Lazarus and Folkman, 1984) of distress and with a cognitive model of caregiving (Kuipers et al., 2010). From this perspective, the appraisals and related behaviour are seen as understandable responses made by caregivers to the demands of caring for their psychiatrically impaired relative. However, while these appraisals are often unhelpful and based on fear and lack of knowledge about psychosis, they are open for modification in a supportive collaborative relationship (Barrowclough et al., 1996). With regard to our understanding of appraisal and attribution, prospective studies with measures of distress as outcome variables are needed in order to determine direction of effects and potential moderators and mediators. Furthermore, the question of why some caregivers engage in more negative and less positive appraisals and attributions remains largely unanswered, as well as the question as to whether interventions aimed at reducing these appraisals are effective.

### (c) Interpersonal response in caregivers

Six studies (Alvarez-Jiménez et al., 2010; Jansen et al., 2013; Möller-Leimkühler and Obermeier, 2008; Möller-Leimkühler, 2006, 2005; Patterson et al., 2005; Raune et al., 2004; Tomlinson et al., 2013) examined the association between EE and distress. In cross-sectional studies, Jansen et al. (2013) found EOI to be associated with distress ( $r = .67, p < .01$ ), which corresponds to McNab et al.'s (2007)

findings (high vs. low EOI:  $t(47) = 2.22, p < .05$ ). In a prospective study, Alvarez-Jimenez et al. (2010) found that baseline EOI predicted caregiver distress ( $F(1) = 5.76, p < .05$ ) and burden ( $F(1) = 8.53, p < .01$ ) at seven months follow-up. In another prospective study, Möller-Leimkühler (2006) found EE to predict caregiver burden as measured by SCL-90 ( $\beta = -.34, p < .001$ ) as well as in interaction with other variables for other measures of distress at one year follow-up. They also found EE to predict distress at two-year follow-up when measured by FBQ in a logistic regression (Wald  $\chi^2 = 28.01, p < .001$ ), FBO (Wald  $\chi^2 = 28.21, p < .001$ ) and SCL-90 (Wald  $\chi^2 = 20.04, p < .001$ ). Finally, Raune et al. (2004) found higher burden in caregivers high in EE ( $t(44) = -2.4, p = .02$ ) and Tomlinson et al. (2013) found distress to be associated with high EE ( $r = .65, p < .01$ ). Patterson et al. (2005) on the other hand, did not find a significant association between burden and EE, although high EOI relatives reported higher burden scores ( $M = 84.5, SD = 28$ ) than low EOI ( $M = 66.4, SD = 17.9$ ),  $p < .01$ .

While interpersonal skills, including emotional regulation and psychological problem solving are central and inter-related to family functioning (Lysaker et al., 2011b), only two studies have examined caregivers' abilities to reflect upon and make sense of other peoples' cognitive and emotional states. Jansen et al. (2013) studied caregivers' capacity for metacognition, which is defined as the understanding of one's own- and others' thoughts and emotions, as well as the ability to solve emotional problems. They found that, while higher levels of metacognition in caregivers were not associated with less negative caregiver experiences, they were associated with reports of more positive caregiver experiences ( $r = .52, p < .01$ ). One possibility is that greater capacity to form complex ideas about one self and others does not necessarily reduce distress and burden, which is a natural and understandable reaction when faced with psychosis. However, it seems to allow for a broader perspective on caregiving, allowing for both positive and negative experiences.

In a related study on social cognition, Tomlinson et al. (2013) found that performance on social cognition tasks was impaired in both caregivers and patients compared with the expected means for the measure ( $t(40) = 4.75, p < .001$ ). This is in line with previous studies that have shown reduced

social cognition capabilities in first-degree relatives of individuals with psychosis (Janssen et al., 2003). However, in Tomlinson et al.'s study, social cognition did not significantly predict their level of distress or burden. Finally, in the only study investigating personality dimensions included, Möller-Leimkühler and colleagues (2008; 2006, 2005) found that neuroticism was predictive of caregiver distress measured by FBQ in a logistic regression (Wald  $\chi^2 = 18.51$ ,  $p < .001$ ), FBO (Wald  $\chi^2 = 28.48$ ,  $p = .001$ ) and LQLP (Wald  $\chi^2 = 7.80$ ,  $p = .005$ ) both at one- and two-year follow-ups. Möller-Leimkühler and colleagues (2008) argue, that neuroticism may operate as a vulnerability factor in caregivers, rendering them more susceptible to critical events such as mental illness in the family. Referring to a meta-analysis by Connor-Smith et al. (2007), showing that that neuroticism strongly relates to engaging in more avoidant or emotional coping styles, Möller-Leimkühler et al. (2008) argue for including this as a vulnerability factor within the stress-appraisal-coping framework of caregiver distress.

In summary, there seem to be a robust association between EE, especially EOI, and distress in caregivers of FEP, a finding that corresponds with those from studies of more chronic forms of psychosis (Boye et al., 1998; Breitborde et al., 2009). Only a few studies have examined caregivers' capacity to understand their own and others' thoughts and feelings, which taps into a range of inter-related concepts such as mindfulness (Baer, 2006), metacognition (Lysaker et al., 2008), mentalization (Fonagy et al., 2011), social cognition (Penn et al., 2008) and theory of mind (Brüne, 2005). However, while metacognitive and social cognitive abilities were found to be compromised in caregivers of persons with FEP in the included studies, the findings are preliminary and the correlational designs preclude any firm conclusions with regard to whether they are a reaction to a difficult situation (state) or some characteristics inherent in the caregivers (trait). In general, studies on caregivers' capacities and personality characteristics and their relation to distress are sparse. An exception however, was the studies of Möller-Leimkühler and colleagues (2008; 2006) who found neuroticism in caregivers to be predictive of distress, which is in line with findings from studies on more long-term or chronic patients (Möller-Leimkühler and Wiesheu, 2012).

## 4. Discussion

We sought to identify empirical studies that have investigated psychological factors accounting for caregiver distress in FEP, with the aim of summarising key findings and critically evaluating the methodologies used.

### 4.1. Psychological factors in caregiver distress

Within a stress-appraisal-coping framework, psychological factors seem to account for a great deal of variation in caregiver distress although there are still a limited number of studies on FEP samples. For the purpose of overview and synthesis, we organized the encountered psychological variables into the three non-mutually exclusive groups of coping, appraisal/attribution and interpersonal response. There were moderate to high effect sizes supporting the link between distress and coping as well as distress and appraisal of various illness related factors – ranging across control, symptoms, loss, interpersonal functioning and utility of EE behaviour. Moreover, there was a strong association between EE, especially EOI, and caregiver distress. EE has received much attention in the published research on psychotic disorders, although not so much as a predictor of caregiver distress as a risk factor for relapse (Butzlaff and Hooley, 1998; Hooley, 2007; Kavanagh, 1992). Finally, there seems to be less knowledge as to whether some caregivers are more vulnerable to appraising the caregiving experience as more negative and to having more negative interpersonal responses faced with a difficult caregiving situation. One way of understanding this might be in terms of caregivers' abilities to form complex accounts of their own and other people's mental states and utilizing this knowledge to regulate emotions and to solve interpersonal problems with the person with a psychotic disorder. In the included studies, these abilities were examined in terms of social cognition, metacognition and neuroticism, which might operate as vulnerability factors within the stress-appraisal-coping model. However, while studies on individual characteristics are promising, more studies are needed before firm clinical implications can be drawn.

#### 4.2. Methodological limitations and future research

We identified a number of important methodological issues in the literature. First, the overwhelming majority of the studies were cross-sectional and the consideration of potential confounding factors such as time of assessment, length of illness or duration of untreated psychosis was limited. Second, only one of the studies made an explicit definition of distress and an extensive list of overlapping terms and measures for distress were used across and within studies. This makes comparison between studies difficult. Moreover, only half of the studies had caregiver distress as a main variable of interest, which might be a reflection of the fact that caregiver distress has, until recently, been primarily studied as a predictor of EE and relapse. An interesting observation was also that none of the studies reported on physical indicators of distress, such as physical illness or visits to a general practitioner, which are otherwise important indicators of general well-being. Finally, there was a predominance of observational studies as opposed to trials, which further limits the ability to test the direction of effects.

Future studies would benefit from using prospective designs and larger samples to model and capture changes in trajectories of caregiver distress. In addition, controlled studies, including the use of comparison groups, and controlling for confounding variables, would further the field. Moreover, statistical analyses incorporating mediation and moderation effects of variables such as social support, fear of stigma, self-efficacy and expectancy of recovery should be employed (Breitborde et al., 2010). Finally, interventions for psychological factors mentioned in this review, including avoidant coping and subjective appraisal, should be further developed and then tested in randomized controlled trials

#### 4.3. Strengths and limitations of the review

The strengths of the current review include the systematic research strategy based on the PRISMA guidelines, the exclusion of non-empirical papers and the focus on psychological hypotheses and

models behind the studies. All of the potentially eligible studies were first assessed for inclusion and then assessed for methodological quality by two independent raters. There were also some limitations of the review. First, potential papers written in languages other than English were excluded, which may have resulted in language- or cultural bias by omitting relevant studies. Second, while based on validated measures and standards (Beller et al., 2013; Cochrane Consumer and Communication Review Group, 2011; von Elm et al., 2007), the templates for data extraction and methodology assessment was developed for this review and validity has not been established. Finally, there might have been a publication bias favoring reports of significant findings and working against failures to replicate.

#### 4.4. Clinical implications.

The overall findings of this review have a number of therapeutic implications for supporting caregivers, mostly in terms of a broadening of the repertoire of interventions. First, it might be important to focus more on other aspects than solely reducing EE in FEP. Clinicians would benefit from adopting the view of EOI as an understandable reaction to a crisis and also a sign of care and willingness to be engaged in treatment and support (van Os et al., 2001). In addition, having someone 'taking over responsibilities' might actually be of benefit to the person with psychosis in the early phase of illness. Moreover, caregivers' coping style such as emotional or avoidant coping and their appraisal of the impact of the illness seem to be associated with distress already in the early phase of illness and need to be addressed. Second, in addition to psychoeducation and problem-solving caregivers may benefit from having a better opportunity to tell their story. Good rapport with a therapist may also constitute a setting in which caregivers' metacognitive abilities are expanded and developed (Lysaker et al., 2011a). In addition to the non-specific therapeutic factors such as being heard and understood, a more comprehensive and rich narrative of illness can be developed, encompassing caregivers' grief and loss, as well as coping efforts, appraisal and understanding of illness. Third, interventions would benefit from targeting caregivers' negative appraisals of the



caregiving situation. This could involve helping caregivers to more accurately perceive the situation they are in, including increasing knowledge about the patient's illness and the possibility of recovery, as well as assisting in handling difficult thoughts, appraisals and emotions. This, of course, is more than just thinking differently or positively, but rather helping 'being with', 'making room for' and 'accepting' difficult and understandable emotions (Hayes et al., 2011; Morris et al., 2013).

## 5. Conclusion

In conclusion, this review has considered how psychological factors within caregivers of FEP have been investigated empirically to explain variations in caregiver distress. We organized the psychological variables found through the search into the three non-mutually exclusive groups of coping, appraisal/attribution and interpersonal response. A number of methodological issues were noted in the studies included and caution must be taken when generalizing to clinical practice. Within the limitations of the studies, the reviewed evidence suggests moderate to high effect sizes supporting the continued focus on psychological factors such as avoidant coping, negative appraisal and EOI when working with caregivers early in the course of illness. Future studies should aim at: 1) replicating findings with larger, prospective samples which controls for confounders such as time of assessment and duration of illness; 2) expanding the intervention repertoire within family work for psychosis, beyond EE-based interventions; and finally 3) testing these psychological factors in controlled clinical trials.

### **Declaration of interest**

none

### **Author contributions:**

Analyzed the data: JE JG. Wrote the first draft of the manuscript: JE. Contributed to the writing of the manuscript: JE JG SC. ICMJE criteria for authorship read and met: JE JG SC. Agree with manuscript results and conclusions: JE JG SG.

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Table 1. Summary of studies investigating psychological factors accounting for caregiver distress in first-episode psychosis

Study	Location	N Care-givers	Age (SD)	% fe-male	Design	Distress measures	Psychological constructs	Measure of psychol. Constructs	Key findings
Addington et al., 2003	Calgary, Canada	238	-	-	Cross-sectional	PGWB	Appraisal	ECI	The family's appraisal of the impact of the illness was associated with their psychological wellbeing.
Alvarez-Jimenez et al., 2010	Melbourne, Australia	63 (48)	44.4 (11.1)	-	Prospective, 7 month follow-up	GHQ-28 ECI	Coping model Attachment model	FQ	EOI predicted family distress and burden at baseline and follow-up.
Cotton et al., 2013	Melbourne, Australia	124	47.2 (8.3)	82.3	Cross-sectional	ECI K10	Coping style	WOC FQ	Avoidance coping strategies were related to psychological distress, EOI and burden.
Gleeson et al., 2010	Melbourne, Australia	63 (21)	45.5 (9.5)	-	RCT	GHQ-28	Appraisal	ECI	There were significant group effects for aspects of the appraisal of caregiving (RPT vs. TAU). There were no significant effects for caregiver distress.
Hinrichsen et al., 1999	NY, USA	63	-	-	Cross-sectional	PRS BI SCL-90	Attribution Coping	PDAQ HDLFC DMSS	Attributions and reported ways of coping were linked to indices of caregivers' emotional adjustment.
Jansen et al., 2013	Roskilde, Denmark	40	-	60	Cross-sectional	ECI GHQ-30	Level of metacognition in caregivers	MAS-R FQ	Greater levels of general distress and EOI were related to more negative caregiver experience, while greater metacognitive capacity was related to more positive experiences of caregiving.
McNab et al., 2007	Melbourne, Australia	53	49.1 (5.3)	66	Cross-sectional	GHQ-28	Attribution Utility beliefs	IPQ (note: only four items) UB FQ	Distress was associated with utility beliefs (beliefs about the utility of self-sacrifice and person-focused criticism) and EOI.
Moller-Leimkuhler, 2005	Munich, Germany	83 (35 F20)	46.8 (11.5)	44.4	Cross-sectional	FBQ SCL-90 LQLP Bf-S	Coping "Negative stress response" (coping) Personality dimensions	SVF FKK NEO-FFI FMSS FQ	The most relevant predictors of burden were EE, emotion-focused coping strategies and generalised negative stress response. Illness characteristics were not predictive of distress.
Moller-Leimkuhler, 2006	Munich, Germany	70 (34 F20)	49.2 (10.9)	48.6	Prospective, 1 year follow-up	FBQ SCL-90 LQLP Bf-S	Coping "Negative stress response" (coping) Personality dimensions	SVF FKK NEO-FFI FMSS FQ	Caregiver burden was predicted by EE, neuroticism, generalised negative stress response (negative coping) and life stressors.
Moller-Leimkuhler et al., 2008	Munich, Germany	63 (34 F20)	48.3 (17.9)	47.6	Prospective, 2 year follow-up	FBQ SCL-90 LQLP Bf-S	Coping "Negative stress response" (coping) Personality dimensions	SVF FKK NEO-FFI FMSS FQ	Caregiver burden was predicted by EE, neuroticism, generalised negative stress response (negative coping) and life stressors.
Onwumere et al., 2011	London and East Anglia, UK	141	50.2 (11.8)	79.3	Cross-sectional	GHQ-28	Coping	COPE	Caregiver distress was associated with avoidant coping strategies but not with duration of illness.

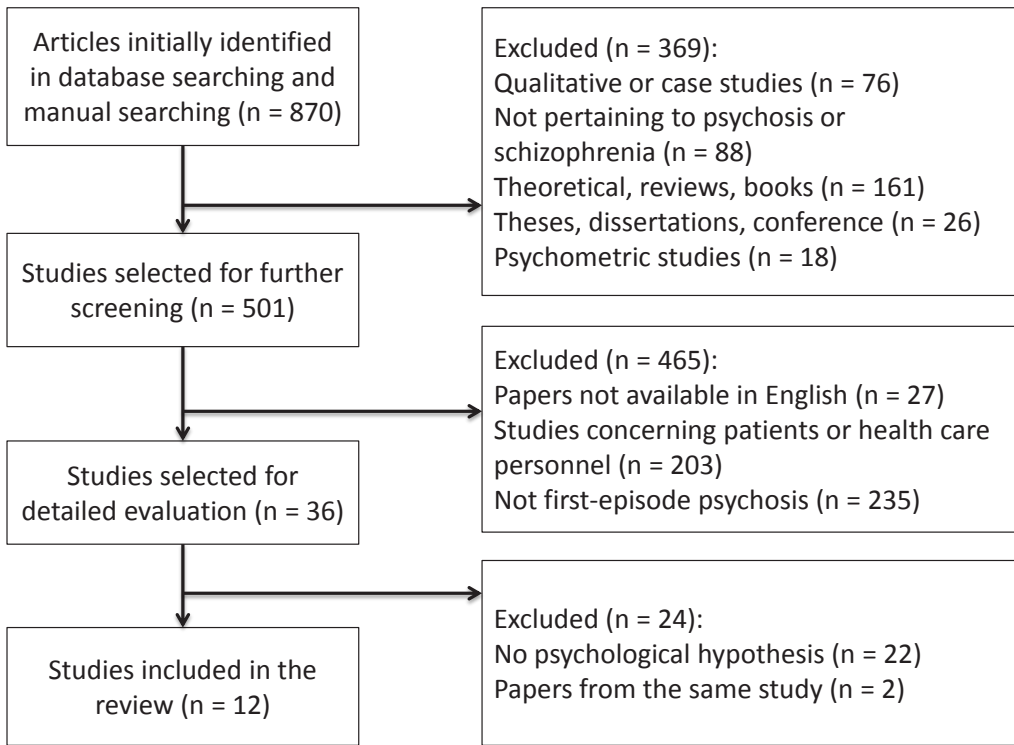
Patterson et al., 2005	Birmingham, UK	50 (39)	43.9 (11.2)	-	Prospective, 9 month follow-up	CDSS ECI	Loss Attachment model  (Appraisal/attribut ion)	TRIG CFI	Subjective burden was linked to loss and not EE status. The appraisal of loss was linked to EOI but not CC at baseline. Loss was reduced in those who changed EE status from high EOI to either high CC or low EE.
Raune et al., 2004	London, UK	46	47.2 (14.4)	72	Cross-sectional	GHQ-28 ECI SFS BDI	Appraisal Coping	The cope instrument (COPE) CFI	High EE was associated with avoidant coping, subjective burden and lower perceived patient interpersonal functioning. High EE might be a way of coping with the burden.
Tomlinson et al., 2013	London, UK	24	51.7 (5.8)	83.3	Cross-sectional	HADS	Social cognition	Hinting task FEEST FAS ECI	Negative experience of caregiving was related to high EE, anxiety and depression. Both patients and carers showed impaired social cognition, but social cognition was not related to burden or EE.

Note. N = number; "-" = not reported; PGWB = Psychological General Wellbeing Scale; GHQ-28 = General Health Questionnaire, 28-item version; ECI = Experience of Caregiving Inventory; FQ = Family Questionnaire; EOI = Emotional over-involvement; CC = Critical comments; EE = Expressed emotion; K10 = Kessler Psychological Distress Scale; WOC = Ways of Coping Checklist; TAU = Treatment As Usual; RPT = Relapse Prevention Therapy; RCT = Randomised controlled trial; PRS = Patient Rejection Scale; BI = The Burden Interview; SCL-90 = Symptom Checklist-90; PDAQ = Psychiatric Disabilities Attribution Questionnaire; HDLFC = Health and Daily Living Form Indices of Coping; DMSS = Dementia Management Strategies Scale; GHQ-30 = General Health Questionnaire, 30-item version; MAS-R = Metacognition Assessment Scale-Revised; IPQ-SCV = Illness Perception Questionnaire; UB = 'Utility beliefs'; FBQ = Family Burden Questionnaire; LQLP = Lancashire Quality of Life Profile; Bf-S = Befindlichkeitsskala; SVF = Stressverarbeitungsbogen; FKK = Fragebogen zu Kompetenz- und Kontrollüberzeugungen; NEO-FFI = Neo Five-Factor Inventory; FMSS = Five-Minute Speech Sample; COPE = The Cope Instrument; CDSS = Calgary Depression Scale for Schizophrenia; TRIG = Texas Revised Inventory of Grief; CFI = Camberwell Family Interview; SFS = Social Functioning Scale; BDI = Beck Depression Inventory; HADS = Hospital Anxiety and Depression Scale; FEEST = Facial Expression of Emotion Stimuli Test; FAS = Family Attitude Scale.

Table 3. The various ways of describing and measuring caregiver experience in the studies included

Description in paper	Measurement	Authors
“Appraisal of impact of illness/stressors/caregiving experience”	ECI	Alvarez-J. et al., 2010 Cotton et al., 2013 Gleeson et al., 2010 Addington et al., 2003 Tomlinson et al., 2013
“Subjective experience of caregiving”	ECI	Raune et al., 2004
“Psychological distress”	K10	Cotton et al., 2013
“Burden of care/subjective burden”	ECI FBQ LQLP	Patterson et al., 2005 Raune et al., 2004 Moller-L. et al., 2005, 06, 08 Moller-L. et al., 2005, 06, 08
“Caregiver distress”	GHQ-30 GHQ-28	Jansen et al., 2013 McNab et al., 2007 Onwumere et al., 2011 Raune et al., 2004
“Level of stress”	PGWB	Addington et al., 2003
“General well-being”	GHQ-30 PGWB Bf-s SCL-90	Jansen et al., 2013 Addington et al., 2013 Moller-L. et al., 2005, 06, 08 Moller-L. et al., 2005, 06, 08
“Emotional adjustment to the care of the patient”	PRS BI SCL-90	Hindrichsen et al., 1999 Hindrichsen et al., 1999 Hindrichsen et al., 1999
“Carer symptoms”	GHQ-28	Alvarez-Jimenez et al., 2010
“Depression” or “anxiety”	CDSS BDI HADS	Patterson et al., 2005 Raune et al., 2004 Tomlinson et al., 2013

Note. PGWB = Psychological General Wellbeing Scale; GHQ-28 = General Health Questionnaire, 28-item version; ECI = Experience of Caregiving Inventory; K10 = Kessler Psychological Distress Scale; PRS = Patient Rejection Scale; BI = The Burden Interview; SCL-90 = Symptom Checklist-90; DMSS = Dementia Management Strategies Scale; GHQ-30 = General Health Questionnaire, 30-item version; FBQ = Family Burden Questionnaire; LQLP = Lancashire Quality of Life Profile ;Bf-S = Befindlichkeitsskala; CDSS = Calgary Depression Scale for Schizophrenia; BDI = Beck Depression Inventory; HADS = Hospital Anxiety and Depression Scale.



**Figure 1.** Flow diagram of the study selection process

# Paper 2



# Caregiver distress in first-episode psychosis: the role of subjective appraisal, over-involvement and symptomatology

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## Abstract

**Background** Caregivers of persons with first-episode psychosis (FEP) often report high levels of distress. Preventing long-term or chronic distress within the whole family is an important focus of early intervention for psychosis. However, a more comprehensive understanding of the psychological factors involved is needed.

**Aims** To examine the impact of subjective appraisals and expressed emotion on caregiver distress in FEP.

**Method** Within a cross-sectional design, 154 caregivers of 99 persons with FEP in a clinical epidemiological sample completed a series of questionnaires to examine potential predictors of caregiver distress.

**Results** Thirty-seven percent of caregivers were suffering from clinically significant distress. A linear mixed model analysis found that, after controlling for caregiver socio-demographic factors, service-user symptoms and global

functioning, emotional over-involvement and subjective appraisal of caregiving were significant predictors of caregiver distress.

**Conclusion** Caregiver distress is significant in the early phase of illness, and this seems to be more related to their subjective appraisal and over-involvement, than to variations in symptoms and global functioning of the person diagnosed with FEP. This lends further support to the stress-appraisal coping model and the cognitive model of caregiving in FEP, and highlights supportive interventions aimed at handling unhelpful cognitions and behaviors.

**Keywords** Caregivers · Distress · Emotional over-involvement · First-episode psychosis

## Introduction

Informal caregivers often constitute an invaluable source of support in the early phase of a psychotic illness, and they can attend to a number of needs that are unmet by the established treatment system [1]. Unfortunately, this is often accompanied by a great deal of burden and distress including anxiety, depression and economic strain [2–5]. Clinicians have traditionally focused on family environments from the vulnerability-stress framework [6], aiming to reduce stress, symptoms exacerbation and relapse [7]. Recently, attention has broadened to encompass caregivers' need for support in their own right to prevent long-term or chronic distress [8].

However, there is still a challenge to get a more comprehensive account of the various psychological factors involved in caregiver distress in first-episode psychosis (FEP). Two parallel lines of research have been central to the field. The first is the stress-appraisal coping model [9] and the cognitive model of caregiving [10], suggesting that

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caregivers' appraisal of the stressors associated with the illness, inform what type of coping they will engage in. Higher levels of distress are associated with more unhelpful appraisals or cognitive representations of the illness and more dysfunctional coping behavior [4, 11]. In the only study in FEP, Addington and colleagues [3] found that caregivers' distress was associated with their subjective appraisals of the impact of illness, but not with patients' characteristics such as positive and negative symptoms.

The second line of research is the expressed emotion (EE) model, which has been a cornerstone in family research for the last 50 years [12, 13]. High EE is defined as interpersonal interaction characterized by critical comments (CC), hostility or emotional over-involvement (EOI) and has been shown to have strong associations with relapse and symptoms exacerbation in schizophrenia [13, 14]. While the predictive validity is less conclusive in FEP [15–17], there has been an increased interest in the impact that expressed emotion—especially emotional over-involvement—has on caregivers' own mental well-being [18, 19]. Converging evidence suggests that EOI is more related to distress than CC [20, 21].

However, while a few studies have examined appraisal and distress in FEP these have reported mixed findings [3, 15, 22]. Furthermore, there have been methodological limitations such as small sample sizes, distress not being the main outcome and a lack of adjustment for variations in illness characteristics. To the best of our knowledge no study has examined both emotional over-involvement and subjective appraisal within a model adjusting for illness characteristics and with caregiver distress as the main outcome in FEP.

The aims of the current study were to determine the level of distress experienced by caregivers in a FEP sample and examine whether this was predicted by: (1) the level of expressed emotion in the family, (2) caregivers' subjective appraisal of the impact of caregiving and illness, when (3) controlling for patient characteristics such as symptoms and global functioning.

Based on the studies presented above, the following hypotheses were proposed: (1) Caregivers of persons with FEP will experience high levels of distress and this will be associated with (2) more negative appraisals and (3) higher levels of emotional over-involvement, but not criticism. Finally, (4) caregiver distress will not be associated with patient characteristics such as symptoms and overall functioning.

## Method

### Design

The study had a cross-sectional design and participants comprised the baseline of an intervention study seeking

to compare multi-family interventions with individual-family interventions. The participants were consecutively included.

### Participants and settings

Every person recently diagnosed with a schizophrenia spectrum disorder and currently enrolled in treatment within Region Zealand in Denmark (population: 816,670) was approached for participation between April 2011 and April 2013. The inclusion criteria were: (1) meeting ICD-10 criteria for schizophrenia spectrum disorders (F20–29, except F21), (2) first-ever psychiatric treatment because of this disorder, (3) age 18–35. The only exclusion criterion was insufficient Danish skills for the interview to be completed. The numbers of eligible individuals were 99 patients enrolled in the outpatient treatment service based on assertive community treatment principles (OPUS) [23] together with 154 caregivers. Diagnoses were confirmed using operational criteria checklist (OPCRIT) [24] by a clinical psychologist, MD or psychiatrist.

## Measures

### Demographic data

A standardized form developed by the research group was employed to collect demographic data such as caregivers' age, gender, relationship status, cohabitation and employment status.

### Caregiver distress

Caregiver distress was assessed using General Health Questionnaire (GHQ-30) [25]. The GHQ-30 is a 30-item self-report questionnaire. For each item a rating must be given for the occurrence of a particular symptom, on a 4-point Likert scale ranging from 0 to 3. The total score can thus vary from 0 to 90. The four response categories to the positively worded items are labeled 'better than usual/more so than usual', 'same as usual', 'less than usual' and 'much less than usual'. The response categories for the negatively worded items are 'not at all', 'no more than usual', 'more than usual', and 'much more than usual'. The GHQ was divided into five subscales for anxiety, depression, well-being, social dysfunction and coping failures based on factor analysis by Huppert et al. [26]. GHQ-case scores were also calculated, in which each item was scored as 'present' or 'absent' (0–0–1–1), where total score could vary from 0 to 30. A case score above five is normally considered to be high, suggesting clinically significant distress [25]. The scale has shown high internal consistency

and good retest reliability, as well as concurrent validity with a number of other clinical assessments [25].

#### Subjective appraisal of caregiving

Subjective appraisal of caregiving was assessed by the experience of caregiving inventory (ECI) [4]. The ECI is a 66-item self-report questionnaire designed to measure the subjective experience of caregiving for a person with serious mental illnesses. The questionnaire consists of 10 subscales: eight negative areas of caregiving (difficult behaviors, negative symptoms, stigma, problems with services, effects on the family, need to provide back-up, dependency and loss) and two positive (positive personal experiences and positive aspects of the relationship). The ECI measures how often caregivers have thought about each issue during the last month. The items are scored on a 5-point Likert scale with the response categories 'never', 'rarely', 'sometimes', 'often' and 'nearly always'. The maximum score for the negative subscale is 208 and 56 for the positive subscale. The internal consistency and construct validity of the ECI have been found to be high [4, 27].

#### Expressed emotion

The caregivers' level of expressed emotion was assessed by the Family Questionnaire (FQ) [28]. The FQ comprises 20 items measured on a 4-point Likert scale with the response categories 'never/very seldom', 'seldom', 'often' and 'very often'. Participants were asked about their way of handling everyday challenges with statements like 'I have a tendency to neglect myself because of her/him' and 'she/he irritates me'. The measure consists of two subscales: emotional over-involvement (EOI) and critical comments (CC). EOI includes over-intrusive, self-sacrificing, over-protective behavior, or exaggerated emotional responses, and over-identification with the patient; CC is defined as unfavorable comments on the behavior or the personality of the patient [29]. The items are scored from 1 to 4 yielding a maximum score of 40 in each subgroup. Caregivers are classified as high EE if they score 23 or greater on the CC subscale or 27 or greater on the EOI subscale [28]. The FQ has good psychometric properties including a clear factor structure, good internal consistency of subscales and good concurrent validity in relation to the widely used Camberwell family interview (CFI) [29]. The FQ has also displayed similar level of accuracy and higher sensitivity compared to the five minute speech sample (FMSS) [30], another popular measure to capture EE [28].

#### Illness characteristics and global functioning

Symptoms were assessed with Positive and Negative Syndrome Scale for Schizophrenia (PANSS) [31]. This is a

30-item instrument (each rated on a scale from 1 to 7) for the assessment of positive and negative symptoms of psychosis. Symptoms during the past week are rated, and higher scores indicate more severe symptoms. The positive symptoms subscale and negative symptoms subscale each consist of seven items and there is also a general pathology subscale consisting of 16 items. PANSS is the most widely used instrument for the assessment of schizophrenia symptoms in clinical trials and its psychometric properties have been found to be adequate [32, 33]. For this study, PANSS were divided into positive, negative, cognitive, depressive and excitative component scores based on Bentsen et al. [34]. Global functioning was measured using the DSM-IV's Global Assessment of Functioning scale (GAF) [35].

#### Procedure

Once the persons with psychosis were enrolled in treatment, they were approached for participation in the study. Following oral and written information about the study and then written consent, caregivers were asked to fill out the three questionnaires and the patients were asked to participate in a semi-structured diagnostic interview. All participants were debriefed following the completion of the questionnaires and the PANSS, but none of the respondents reported any adverse experiences in answering the questions. The Regional Committee for Research Ethics approved the study (reg.nr. 2008580020).

#### Statistical analyses

Statistical analyses were carried out using SPSS version 20 for Mac (IBM Inc., Chicago, IL, USA). First, descriptive statistics of socio-demographic and clinical variables were calculated using means and standard deviations for quantitative variables and frequencies with percentages for categorical variables. Second, as some of the patients had two caregivers participating, a linear mixed model regression analysis was carried out to account for the possible correlation between the pairs of caregivers. Finally, the Student's *t* test was used to compare levels of distress on the various subscales of GHQ-30 between caregivers rated as high and low in expressed emotion.

## Results

#### Sample characteristics

The characteristics of the sample are presented in Table 1. There were 154 caregivers between 21 and 87 years (median 50) of 99 patients between 18 and 33 (Median 20).

**Table 1** Characteristics of caregivers and persons with FEP

	M (SD)	% (n)
Gender female	–	62.3 (96)
Age	49.09 (9.17)	–
Relationship		
Parent	–	80.5 (124)
Stepparent	–	5.2 (8)
Sibling	–	5.8 (4)
Spouse	–	5.8 (9)
Other	–	5.8 (9)
Cohabitation	–	57.1 (88)
Face-to-face contact >1 × week	–	83.8 (129)
Employment	–	71.4 (110)
Characteristics of persons with FEP		
Gender female	–	37.4 (37)
Age	21.09 (3.44)	–
PANSS total score	57.14 (12.94)	–
Positive	11.22 (4.57)	–
Negative	20.31 (7.31)	–
Emotional distress	9.88 (3.23)	–
Cognitive disorganisation	5.35 (1.86)	–
Excitement	6.60 (1.63)	–
Global functioning, GAFs	40.21 (12.14)	–
Global functioning, GAFf	40.80 (12.34)	–

*n* = 154 caregivers and 99 service users

FEP first-episode psychosis, *M* mean, *SD* standard deviation, PANSS Positive and Negative Syndrome Scale for Schizophrenia, GAF Global Assessment of Functioning Scale, GAFs Symptom score, GAFf Function score

A further 69 eligible patients included in the treatment service did not participate, while 20 of these accepted for the caregivers to be involved. For 16 persons we could not arrange for an interview within time limits set by protocol, one was too unwell for interview and the rest gave no reason. Only 25 caregivers declined participation of particular note, the majority of caregivers were female, living with the patient and employed. Both higher and lower socio-economic classes were well represented.

#### Questionnaires and clinical measures

In this sample 43 (28.3 %) caregivers could be characterized as emotionally over-involved (*M* 23.34, *SD* 5.01) while 32 (21.3 %) could be characterized as critical (*M* 18.33, *SD* 5.45). A total number of 55 (35.7 %) caregivers could be characterized as high EE on at least one of the subscales and 18 (11.7 %) were high EE on both subscales. Fifty-seven (37 %) caregivers had a GHQ-caseness score above the threshold for clinical significant distress and the mean continuous score was 29.85 (*SD* 14.60). The mean

score for ECI negative subscale was 73.95 (*SD* 31.79) and ECI positive subscale was 27.03 (*SD* 8.65).

The mean total scores on the PANSS and GAF are presented in Table 1. While there are no norms available for the PANSS, the mean score of 57.14 (*SD* 12.94, range 30–89) would suggest low levels of psychotic symptoms [36].

#### Predictors of caregiver distress

The results of the linear mixed model analysis with distress (GHQ-30) as the main outcome are presented in Table 2. High scores on EOI and Negative Appraisal were significantly related to increased caregiver distress after controlling for caregivers' gender and cohabitation, and the level of symptoms and overall functioning in patients. Caregivers' positive appraisals also related significantly to decreased caregiver distress while criticism did not significantly contribute to the model.

As demonstrated in Table 3, when compared to caregivers with low EOI, caregivers with high EOI had a significantly higher level of total distress and a higher score on all of the five subscales, namely anxiety, low self-esteem,

**Table 2** Linear mixed model analysis with distress as dependent variable

Predictors	$\beta$	95 % CI	<i>p</i>
Expressed emotion			
FQ—emotional over-involvement	1.13	0.48 to 1.78	<b>0.001</b>
FQ—critical comments	−0.31	−1.04 to 0.42	0.403
Subjective appraisal			
ECI—negative subscale	0.16	0.04 to 0.29	<b>0.011</b>
ECI—positive subscales	−0.49	−0.76 to −0.21	<b>0.001</b>
Caregiver demographics			
Gender	−0.28	−4.87 to 4.32	0.905
Cohabitation	−2.59	−7.53 to 2.34	0.298
PANSS			
Positive component score	0.15	−0.42 to 0.72	0.592
Negative component score	0.04	−0.37 to 0.45	0.853
Cognitive component score	0.23	−1.25 to 1.70	0.759
Depressive component score	0.06	−0.77 to 0.88	0.893
Excitatory component score	0.11	−1.42 to 1.63	0.887
Global functioning			
GAFs	0.21	−0.27 to 0.33	0.836
GAFf	1.37	−0.10 to 0.51	0.179

Bold values indicate statistical significance (*p* > 0.05)

*n* = 154

CI confidence intervals, FQ Family Questionnaire, ECI Experience of Caregiving Inventory, PANSS Positive and Negative Syndrome Scale for Schizophrenia, GAF Global Assessment of Functioning Scale, GAFs Symptom score, GAFf Function score

**Table 3** The relationship between the two components of expressed emotion and subscales of caregiver distress

	EOI		CC	
	Low	High	Low	High
GHQ-total	25.3 (11.4)	42.0 (15.2)**	28.5 (14.5)	35.7 (13.8)*
Anxiety	6.4 (4.6)	13.0 (4.9)**	7.6 (5.5)	10.7 (4.9)**
Low self-esteem	4.1 (1.8)	5.4 (1.8)**	4.4(1.9)	5.0 (1.6)
Depression	2.1 (2.3)	4.8 (3.6)**	2.8 (2.9)	3.4 (3.1)
Coping difficulties	4.4 (1.7)	7.4 (2.9)**	5.0 (2.5)	6.0 (2.5)*
Social dysfunction	3.2 (1.3)	4.4 (2.0)**	3.4 (1.6)	4.0 (1.5)*

*n* = 154. Mean (standard deviation). Statistical analyses by the independent sample Student's *t* test

GHQ General Health Questionnaire, EOI emotional over-involvement, CC critical comments

\* *p* < 0.05, \*\* *p* < 0.0005

depression, coping difficulties and social dysfunction. In addition, compared to caregivers with low CC, caregivers with high CC had a significantly higher mean level of total distress and a higher score on the subscales anxiety, coping difficulty and social dysfunction, but not on self-esteem and depression.

## Discussion

The current study examined predictors of caregiver distress in first-episode psychosis, focusing on the subjective appraisal of the impact of caregiving and expressed emotion. Consistent with our hypotheses, caregivers had high levels of distress, and more than one-third (37 %) of them reached a level of clinically significant distress. Also, consistent with our hypotheses, subjective appraisal and emotional over-involvement were significant predictors of caregiver distress. These results remained significant after controlling for caregiver demographic characteristic, such as gender and cohabitation, and patients' symptoms and overall functioning.

### Caregiver distress

In accordance with earlier studies [5, 37, 38], we also found that many of the caregivers had high levels of distress. The percentage of case scores on the GHQ was considerably higher in the present study than was found by Tennakoon [39] (12 %), but slightly lower than was found by Alvarez-Jiménez et al. [20] (55.5 %) and by Barrowclough and Parle [40] (57 %).

### Appraisal and distress

The positive association between distress and increased caregivers' subjective appraisal is also congruent with earlier studies [3, 39]. The findings are consistent with the

stress-appraisal coping model [9, 27] and the cognitive model of caregiving [10]. One way to interpret these findings is that while caregivers might be negatively influenced by the impact of illness, variations in caregiver distress seem less influenced by the patients' symptomatology and overall functioning, than by their own subjective appraisals or cognitive formulations. Furthermore, this implies that the distress experienced by caregivers is not necessarily alleviated or even reduced when their son, daughter or partner's symptoms improve, but they may sometimes need individually tailored interventions addressing 'unhelpful' cognitions and behaviors.

While there is still limited knowledge about what causes negative and positive appraisals in caregivers, factors such as control attribution [41], negative caregiver self-concept [42] and metacognition [43] have been suggested. Jansen and colleagues [43] found that EOI and general well-being were more closely related to negative appraisals of caregiving, while higher levels of metacognition—or the capacity to construct complex and integrated representations of self and others—were associated with more positive appraisals.

The question as to whether interventions aimed at reducing negative appraisals also reduce general distress, as suggested by the stress-appraisal and cognitive model, remains to be empirically resolved. In a randomized controlled study of family intervention, Gleeson et al. [15] found that although they were able to increase positive appraisal of the impact of caregiving, this did not significantly reduce the level of distress as measured by GHQ. According to the authors, more interventions directly aimed at reducing negative appraisals, in addition to the positive, are warranted.

### Expressed emotion and distress

This study found that 37.7 % of caregivers could be characterized as having high expressed emotion, which is

slightly less than that reported in some other FEP samples [20, 44, 45] but considerably less than found by McNab et al. [46] (73.5 %). While EOI was a strong predictor of caregiver distress in the linear mixed model analysis, criticism did not significantly contribute to the model that was fitted. These findings are in line with recent evidence suggesting that criticism and emotional over-involvement are related to different factors, and that the latter is more closely related to distress in caregivers [18, 20]. It is notable, however, that this was a low CC sample, which may also explain the weak influence of CC on caregiver distress. The findings are also in line with the view that emotional over-involvement can be seen as an attempt to reduce the perceived stressfulness of the caring role [21, 47]. It has also been argued that this way of coping might benefit some patients in the early stages of illness when they are less able to take care of themselves [1, 48]. However, emotional over-involvement seems to have a detrimental effect on the caregivers' own mental well-being [18], and possibly on the long-term well-being of the patients [14]. Distress and over-involvement is thus a natural and understandable reaction to the difficult and painful experience of having someone close to you going through a psychosis, but early intervention aimed at preventing long-term over-involvement is important to promote recovery in both patients and their caregivers.

#### Limitations, strengths and clinical implications

There are some limitations to this study. First, self-report biases and social desirability may have contributed to errors in the self-report measures we used. Second, the cross-sectional design does not allow for firm causal inferences. It is not possible to rule out for instance, that distress was increasing the report of negative appraisals or over-involvement. Finally, caregiver inclusion was based on consent by the patients, which represents a potential selection bias. It could be that patients who had a good relationship with their caregivers were more inclined to consent, and that this was reflected in the lower ratings of CC. The strength of the study is the relatively large sample drawn from a specialist early intervention service within a defined catchment area, which increases the likelihood of a sample that is representative of the target population. In addition, the study adjusted for caregiver and patient characteristics and included two caregivers per patient in a multilevel analysis. The inclusion of two caregivers possibly explains why we have more male participants compared with other studies on caregiver distress as most studies include only one significant other which typically is the mother [49, 50].

With these limitations and strengths in mind, the current study provides further support for the stress-appraisal coping theory and the cognitive model of caregiving in

families with first-episode psychosis. First, in the early stages of illness, caregivers are clearly distressed and have numerous concerns that need to be attended to by early intervention services. Second, it adds to the existing literature of cognitive appraisals and attributions in understanding caregiver distress. Third, it adds to the literature regarding the understanding of expressed emotion and its different correlates in first-episode psychosis. Future studies should examine the potential positive side of emotional over-involvement in the early stages of illness and the modification of appraisals in controlled studies. In addition, further studies should address the long-term effects of EOI and negative appraisals on caregiver distress in longitudinal designs to better assess the direction of the effects.

#### Conclusion

Distress in caregivers of persons with FEP seems to be more related to their subjective appraisal and over-involvement, than to variations in the patients' symptomatology and overall functioning in the early phase of illness. This lends further support to the stress-appraisal coping model and the cognitive model of caregiver distress in FEP, and highlights the need for supportive interventions aimed at reducing unhelpful cognitions and behaviors.

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**Conflict of interest** None.

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# Paper 3







## Positive and negative caregiver experiences in first-episode psychosis: Emotional overinvolvement, wellbeing and metacognition

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**Objectives.** While caregivers of persons with first-episode psychosis often report a range of negative experiences, little is known about what psychological factors are involved. The aim of this study was to examine how caregivers' general wellbeing, emotional overinvolvement and metacognition influenced their reports of both positive and negative caregiving experiences.

**Design.** A prospective consecutive cross-sectional study.

**Methods.** Forty caregivers of patients with first-episode psychosis were interviewed using semi-structured interview and questionnaires.

**Results.** Greater levels of distress and overinvolvement were associated with more negative experiences of caregiving while greater metacognitive capacity was associated with more positive experiences of caregiving.

**Conclusions.** The experience of positive and negative aspects of caregiving seems to be associated with different variables. Greater metacognitive capacity does not necessarily alleviate the suffering and distress, which is a healthy and normal reaction to having a close one suffering from psychosis. But it might help broaden the perspective, allowing for both negative and positive experiences. Clinical implications in terms of expanding the range of therapeutic interventions are discussed.

### Practitioner points

- Assessing the capacity for metacognition in a non-clinical population.
- Understanding what factors are involved in positive and negative caregiver experiences in first-episode psychosis.
- A broadening of family interventions by encompassing the concept of metacognition.

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Recent research suggests that contrary to long-standing view, most persons diagnosed with schizophrenia experience some significant degree of recovery over their life span (Lysaker & Buck, 2008). Recovery can involve a range of factors including symptom remission, but also a subjective sense of wellbeing (Davidson, Schmutte, Dinzeo, & Andres-Hyman, 2008). Subjective wellbeing within the patient may interact with and be influenced by the experiences of wellbeing within the family. Thus, models of recovery are increasingly focused on family systems and not just the person diagnosed (Martens & Addington, 2001).

Many relatives assume great responsibility for the care of persons with schizophrenia but often feel overwhelmed by the task (Glynn, 2012). Studies reveal high levels of psychological distress, including anxiety and depression, as well as economic strain, reduced quality of life and stigmatization (Martens & Addington, 2001; Scazufca, Kuipers, & Menezes, 2001; Tennakoon *et al.*, 2000; Treasure *et al.*, 2001). Caregiver burden has been related to higher levels of expressed emotion (EE), which has been shown to predict symptom exacerbation and relapse (Brown, Monck, Carstairs, & Wing, 1962; Butzlaff & Hooley, 1998; Vaughn & Leff, 1981). While EE, defined as a relationship between patient and caregivers characterized by criticism and/or emotional overinvolvement (EOI) (Barrowclough & Parle, 1997), is one of the most studied psychosocial predictor of psychosis, the concept is far from fully understood (Kanter, Lamb, & Loeper, 1987; Van Os, Marcelis, Germeys, Graven, & Delespaul, 2001). There seems to be some agreement though that EE is an attempt to guide, influence or 'control' the behaviour of the patient – often with the best intention to help, but at the cost of great distress in the caregivers (Bentsen, Boye, & Munkvold, 1996; Hooley, 1985; Van Os *et al.*, 2001). There also seems to be agreement that the level of burden and EE are best understood from a stress-coping perspective (Lazarus & Folkman, 1984), focusing on relatives' appraisal of circumstances rather than on patients' actual deficits and 'objective' measures of caregiving experiences (Joyce *et al.*, 2003; Kuipers *et al.*, 2007; Scazufca & Kuipers, 1996). Finally, from an attachment perspective, Patterson, Birchwood, and Cochrane (2005) have recently found EE to be adaptive reactions to perceived loss in caregivers.

However, any comprehensive consideration of family experiences of caregiving should take into account that they involve a range of both negative and positive experiences (Diener, Suh, Lucas, & Smith, 1999; Martens & Addington, 2001; Szmukler *et al.*, 1996). These may be related to one another but not necessarily so and caregivers may experience a predominance of one or experience both. As an illustration, anecdotally many caregivers describe feelings of deep sadness and worry, and yet experience a deeper connection with the ill family member together with feelings of stronger bonds within the family. This might provide some perspective and work against negative experiences that result in lack of hope for the future. Little is known though about what psychological factors within the family members contribute to negative and positive experiences of caregiving either separately or combined (Martens & Addington, 2001).

One way to encompass these psychological factors is by the concept of metacognition, which has been found underlying many difficulties adapting to psychological challenges for persons with or without mental illness (Dimaggio, Semerari, Carcione, Nicolò, & Procacci, 2007; Semerari *et al.*, 2003). Metacognition refers to a spectrum of activities, which call for thinking about thinking. These include more discrete activities, which include noticing a specific thought and more synthetic activities, which call for integrating discrete information into complex representations of the self and others (Lysaker *et al.*,

2005). Metacognition includes among other skills, the ability to understand emotional reactions in self and others, the ability to consider one's thoughts as subjective, and to understand that the point of view of others may be different from one's own (Carcione *et al.*, 2011; Lysaker *et al.*, 2013). Metacognition might affect caregiver experience for several reasons. Firstly, making sense of a loved one's illness, in a way that allows for both positive and negative experiences, may require the creation of a complex and integrated account of life changing events and therefore the changes experienced by a range of persons. To be able to create such narratized accounts of oneself and one's loved ones would intuitively seem to require greater metacognitive capacities. Family members with lower metacognitive capacities might be less able to form complex accounts, leading to a poorer shared understanding between caregivers and patients, thus leading to fewer positive experiences. Also with greater metacognitive capacity it is likely that family members would be better able to manage their own distress and again see events in a perspective that allow for both good and painful elements to be present. In support of this at least one study found that higher levels of metacognition in patients diagnosed with schizophrenia were linked to more effective coping in the face of identified psychological problems (Lysaker, Erickson, *et al.*, 2011).

To study this issue we gathered reports of positive and negative experiences of caregiving from family members of patients with first-episode psychosis. We then sought to determine the extent to which both were (1) related to one another and (2) related to three different predictors: Overinvolvement, general wellbeing and metacognitive capacity within family members. We chose to focus on only one component of EE, EOI, because this has been found more strongly related to caregiver distress and burden (Alvarez-Jiménez *et al.*, 2010). Also there is a growing recognition that the two components of EE are uncorrelated, related to different variables and best studied separately (Scazufca & Kuipers, 1998; Van Os *et al.*, 2001).

We predicted greater levels of distress would be associated with greater levels of reported negative experiences in light of previous findings linking reports of negative experiences to caregiver distress (Joyce, Leese, & Szmulker, 2000; Martens & Addington, 2001). We also predicted greater levels of overinvolvement in relatives reporting greater levels of negative experiences in light of research understanding EOI as an attempt to bring things to a 'status quo' (Bentsen *et al.*, 1996) and findings on the relation between subjective burden and EOI (Raune, Kuipers, & Bebbington, 2004). Finally, we predicted that the caregivers' capacity for metacognition would be associated with reports of more positive and less negative caregiver experiences.

## Method

### *Design and participants*

The study had a prospective consecutive cross-sectional design and participants were 40 relatives of persons with first-episode psychosis (age 18–35). The sample is drawn from a larger intervention study seeking to compare multi-family interventions with individual-family interventions. Participants were interviewed between April 2011 and June 2012. All patients met ICD-10 criteria for schizophrenia and were included in an outpatient treatment service based on assertive community treatment principles (Petersen *et al.*, 2005). Patients were asked to choose one or two relatives 'or significant others' to participate in family-work and/or research. Excluded from the study were relatives who did not speak Danish (N).

Of the 95 patients enrolled in treatment during this period, 71 agreed to participate while 24 declined. Of the 71 patients, six were not relevant due to change of diagnosis and four later withdrew from the study (because of leaving treatment). From the remaining 61 patients, relatives of 50 patients were randomly included in the study leaving us with a sample of 40 relatives.

The Regional Committee for Research Ethics approved the study. All patients and relatives entering the study gave written informed consent.

## **Measures**

### *Indiana psychiatric illness interview (IPII)*

The IPII is a semi-structured interview developed to assess illness narratives in patients with schizophrenia (Lysaker, Clements, Plascak-Hallberg, Knipscheer, & Wright, 2002). This was modified for caregivers of patients with first-episode psychosis in collaboration with its first author. The interview typically lasts for 30–60 min, and is audiotaped and transcribed. The interview is divided into five sections. First, rapport is established and the caregivers are asked to tell the story of their family in as much detail as they can, starting from around the birth of the child that currently receives treatment. Second, they are asked if they think their son or daughter has a mental illness, and how they understand it. Third, they are asked to consider whether this has had any influence on their occupational, social or emotional life, or on their personality. Fourth, they are asked how being a caregiver controls their lives, and how they cope with being a caregiver. This is followed with questions about how caring for the patient affects others and how others influence their way of being caregivers. Finally, they are asked how they think the patient's need for care will develop in the future. The tone is conversational. There are no direct questions of specific symptoms and the interviewer only asks for clarification when confused. The result is a narrative of self and challenges related to being a caregiver, from which metacognitive capacities can be analysed.

### *The metacognition assessment scale abbreviated (MAS-A)*

The MAS is a rating scale to assess metacognitive abilities and was originally designed to be used on psychotherapy transcripts in personality disorders (Semerari *et al.*, 2003). This study used a modified version, MAS-A adapted to analysing IPII transcripts (Lysaker *et al.*, 2005). This abbreviated version contains four scales: 'Understanding of one's own mind', 'understanding of others' minds', 'decentration' and 'mastery'. Decentration refers to the ability to see that others have independent motives and unique perspectives on life-events. Mastery refers to the ability to conceptualize one's difficulties and implement effective strategies to cope with problematic mental states and emotions. For each scale higher ratings reflects the capacity to engage in more complex metacognitive acts and the maximum total score is 28. MAS-A has been widely used with schizophrenia patients with acceptable reliability and validity and has been able to predict various psychosocial outcomes independent of symptoms and neurocognition (Lysaker, Dimaggio, *et al.*, 2010; Lysaker, Erickson, *et al.*, 2011; Lysaker, Shea, *et al.*, 2010). It has also been used to study metacognition among persons with prolonged non-psychiatric medical conditions (Lysaker *et al.*, 2012). To our knowledge, this is the first study using the MAS-A to assess metacognition in caregivers of persons with first-episode psychosis.

*Family questionnaire (FQ)*

Caregivers' level of EE were assessed by the FQ (Wiedemann, Rayki, Feinstein, & Hahlweg, 2002). In a 20-item self-report questionnaire they are asked about how the family deals with everyday challenges, especially focusing on negative comments, critique and emotional over-involvement. Questions like 'I have a tendency to neglect myself because of her/him' and 'she/he irritates me' are rated as 'never/very seldom', 'seldom', 'often' or 'very often'. The measure consists of two subscales: EOI and critical comments (CC). EOI includes over-intrusive, self-sacrificing, overprotective behaviour, or exaggerated emotional response, and over-identification with the patient; CC is defined as unfavourable comments on the behaviour or the personality of the patient (Vaughn & Leff, 1976b). The items are scored from 1 to 4 yielding a maximum score of 40 in each subgroup. Caregivers are classified as high EE if they score 23 or greater on the CC subscale or 27 or greater on the EOI subscale. The FQ has good psychometric properties including a clear factor structure, good internal consistency of subscales and good concurrent validity in relation to the widely used Camberwell Family Interview (Vaughn & Leff, 1976a).

*General health questionnaire (GHQ-30)*

The level of distress and general wellbeing was measured using the GHQ-30 (Goldberg & Williams, 1988), a 30 item self-report questionnaire. For each item, the caregivers must rate the occurrence of a particular symptom on a four-point Likert scale ranging from 0 to 3. The total score could vary from 0 to 90. The four response categories to the positively worded items are labeled 'better than usual/more so than usual', 'same as usual', 'less than usual' and 'much less than usual'. The response categories for the negatively worded items are 'not at all', 'no more than usual', 'more than usual', and 'much more than usual'. GHQ-case score was also calculated, each item scored as 'present' or 'absent' (0-0-1-1), where total score could vary from 0 to 30. A case score above five is normally considered to be a high score, suggesting clinical significant distress.

*Experience of caregiving inventory (ECI)*

The ECI is 66-item self-report questionnaire, designed to measure the subjective experience of caregiving for a person with serious mental illnesses such as schizophrenia (Szmukler *et al.*, 1996). The questionnaire consists of 10 subscales: eight negative areas of caregiving (difficult behaviours, negative symptoms, stigma, problems with services, effects on the family, need to provide back-up, dependency and loss) and two positive (positive personal experiences and positive aspects of the relationship). The ECI measures how often caregivers have thought about each issue during the last month. The items are scored on a 5-point Likert scale with the response categories 'never', 'rarely', 'sometimes', 'often' and 'nearly always'. Maximum score for the negative subscale is 208 and for the positive subscale 56.

**Procedures and statistics**

Diagnoses were confirmed using OPCRIT (McGuffin, Farmer, & Harvey, 1991) by a clinical psychologist or psychiatrist. As soon as a patient enrolled in treatment, they were asked permission to being informed about the research project by one of the research staff.

Upon agreement, oral and written information about the study were given to patient and relatives. Following consent, participants were interviewed with the IPII, followed by questionnaires in the following order: ECI, FQ, and GHQ-30. Duration of interview was approximately 1 hr per participant. The IPII interview was audiotaped and later transcribed. The ratings of the transcripts were done before typing the data from EE, ECI and GHQ-30 to prevent bias from previous knowledge of the participants. There were two separate raters. The first author was trained in Indianapolis by the MAS-A developer while the second rater was trained by the first. Inter-rater reliability was assessed for 12 transcripts (30%), and good reliability was found with an intraclass correlation of .85 ( $p < .01$ ). Mean, Standard deviation and Pearson correlations were calculated by SPSS version 20 (IBM Inc., Chicago, IL, USA).

## Results

Participant characteristics are presented in Table 1. Twenty-eight (70%) of the patients were living with their parents, and having daily face-to-face contact. Thirteen (32.5%) caregivers could be characterized as critical (scoring  $\geq 23$  on FQ, CC subscale), while twelve (30%) caregivers could be characterized as emotionally overinvolved (scoring  $\geq 27$  on FQ, EOI subscale). The mean and standard deviation of EOI, caregiver distress, positive and negative caregiver experience, and metacognitive capacity are presented in Table 2. Of note, twenty (50%) of caregivers had a GHQ-case score of  $>5$  suggesting clinical significant distress. The Pearson correlations between EOI and caregiver experiences, distress and caregiver experiences, and capacity for metacognition and caregiver experiences are presented in Table 3. There were significant associations between EOI and negative experiences of caregiving (.66,  $p < .01$ ), between distress (GHQ-30) and negative experiences (.62,  $p < .01$ ), and between capacity for metacognition and positive

**Table 1.** Characteristics of the participants ( $n = 40$ )

Parents (%)	35 (87.5)
Spouses (%)	3 (7.5)
Stepparents (%)	2 (5)
Face-to-face contact $\geq 1$ pr. day (%)	28 (70)
Living with the patient (%)	28 (70)
Gender female (%)	24 (60)

**Table 2.** Mean and standard deviations. EOI, distress, positive/negative caregiver experience and capacity for metacognition ( $n = 40$ )

EOI, $M$ ( $SD$ )	23.83 (5.21)
Caregiver distress (GHQ-30), $M$ ( $SD$ )	32.60 (16.75)
Positive caregiving experiences (ECI – Positive), $M$ ( $SD$ )	25.90 (9.16)
Negative caregiving experiences (ECI – Negative), $M$ ( $SD$ )	74.73 (34.10)
Capacity for metacognition (MAS-A), $M$ ( $SD$ )	19.83 (4.99)

ECI = experience of caregiving inventory; GHQ-30 = General Health Questionnaire 30 item version; MAS-A = Metacognitive Assessment Scale Abbreviated; EOI = emotional overinvolvement;  $M$  = mean;  $SD$  = standard deviation.

**Table 3.** Pearson correlations. Positive and negative caregiver experience, EOI, wellbeing and capacity for metacognition ( $n = 40$ )

	Positive experiences	Negative experiences
EOI	0.191	0.665*
Distress (GHQ-30)	0.024	0.621*
Capacity for metacognition (MAS-A)	0.518*	0.030

EOI = emotional overinvolvement; GHQ-30 = General Health Questionnaire 30 item version; MAS-A = Metacognitive Assessment Scale Abbreviated.

\* $p < 0.01$ .

experiences of caregiving ( $.51, p < .01$ ). There were also significant associations between all the subscales of metacognition and positive experiences: Self-reflectivity ( $.46, p < .01$ ), Understanding of others ( $.46, p < .01$ ), Decentration ( $.60, p < .01$ ) and Mastery ( $.47, p < .01$ ). No significant association was found between capacity for metacognition and distress ( $-.03, p = .88$ ), or between metacognition and caregiver criticism ( $-.03, p = .83$ ). There was no significant correlation between positive and negative caregiver experiences ( $.107, p = .51$ ) Tables 1–3.

## Discussion

This study examined how the experience of caregivers of persons with first-episode psychosis was associated with three sets of psychological factors: the caregivers' levels of EOI, general wellbeing and metacognition. First, consistent with our predictions ratings of positive and negative caregiving experiences were not associated with the same variables, supporting the view that that these are independent constructs (Diener, Oishi, & Lucas, 2009; Diener *et al.*, 1999). Second, consistent with our predictions caregivers who reported greater levels of distress and overinvolvement reported having more negative experiences of caregiving while caregivers who had greater levels of metacognition reported having more positive experiences of caregiving.

While the cross-sectional design does not allow us to directly address the question of causality, the finding that greater levels of metacognition were related to more positive reports of caregiving, gives a number of hypotheses to explore further. One possibility is that, with a greater capacity to form complex ideas about one self and others it may be possible to have a more balanced experience of caretaking in which there are both positive and negative experiences. For instance, caregivers with poorer metacognition might not fully appreciate distress and its impact on them, and it might reduce the ability to see their problems in perspective and so manage their distress. Poorer metacognition, including lack of critical distancing or understanding the ill one, might also lead to misunderstandings between caregivers and patients, thus leading to fewer positive experiences. There was a somewhat unexpected finding that while metacognitive capacity was linked to positive experiences it was not associated with negative experiences. One possible explanation for this is that being able to think in more complex ways about oneself and others does not necessarily decrease distress or perception of the negative. It merely helps persons perceive positive aspects of the relationships and accurately perceiving real loss and suffering. This resonates with ideas within the recovery movement, which holds that you can live satisfying, meaningful lives, despite of experiencing distressing symptoms (Davidson, O'Connell, Tondora, Lawless, &



Evans, 2005). It also resonates with ideas from the so-called third-wave cognitive therapies, e.g. Acceptance and Commitment Therapy (ACT), focusing on “making space” for painful emotions, while moving towards positive/meaningful areas of living (Bach, Gaudiano, Pankey, Herbert, & Hayes, 2006; Hayes, Luoma, Bond, Masuda, & Lillis, 2006; Morris, Johns, & Oliver, 2013). From studies on well-being in general, it has also been noted that pleasant and unpleasant affect seem to be independent and having different correlates – they are not simply opposites of one another (Diener *et al.*, 1999; Gilbert *et al.*, 2012; Seligman, Rashid, & Parks, 2006). In other words, when working towards creating hope and meaningfulness, it might not be enough only to focus on reducing clinical symptoms and distress. Of equal importance is focusing on an accurate perception of distress as well as developing positive affects, resilience and meaningfulness.

The finding that negative experiences were predicted by EOI and distress is also in keeping with previous research (Barrowclough & Parle, 1997; Martens & Addington, 2001; Tennakoon *et al.*, 2000). Mental illness such as psychosis and schizophrenia is mostly experienced as a major crisis by relatives, friends and partners. They sometimes describe it in terms of loss – loss of ‘control’, loss of their personal freedom, and loss of the relationship they used to have with the patient – and they are faced with the challenge of coping with this loss (Patterson *et al.*, 2005). According to Van Os *et al.* (2001), EOI can be seen as the relatives’ attempt to take an active partnership in the care for the patients and should not be considered as a sign of a ‘dysfunctional family’. It is known that relatives play a crucial part in the recovery process, often attending to unmet needs on the part of the established treatment system (Glynn, 2012). EOI is rather a sign of a caring family in Van Os *et al.* (2001) view, something that was reflected in their finding that caregivers who actively participated in treatment were the ones that also scored high on measures of EOI. Bentsen *et al.* (1996) also found EOI to be associated with reduced drug abuse and less aggressive behaviour. From this perspective, EOI, might actually be beneficial to the patient in the early stages of the illness: someone taking care of them at a time, when they are vulnerable and lack the energy, self-esteem and motivation to take care of themselves. As the cost often is higher burden, distress and financial strain, EOI might be more detrimental to caregivers than to patients. This was also noted by Breitborde, López, Chang, Kopelowicz, and Zarate (2009) who found baseline overinvolvement to be a marker both for poor current health in caregivers and for poor health at follow up.

There are some limitations of this study to be considered. First, the cross-sectional design does not allow us to directly address the question of causality. Rival hypotheses cannot be ruled out including the possibility that more positive experiences enhance metacognition or that negative experiences create overinvolvement. Second, there were two different interviewers, and various degrees of rapport might have influenced the narratives obtained. Third, there were a considerable number of patients who, for various reasons, did not want to participate. This might have influenced the generalizability of the sample.

### **Clinical implications and future studies**

With replication results may have clinical implications. First, targeting EOI as most family interventions do may reduce negative experiences of caregiving though not necessarily lead to more positive ones. The two affects might best be studied separately to gain the complete picture of caregivers’ wellbeing. Maybe the focus on decreasing EOI in early-phases of the illness is less relevant than focusing on increasing positive experiences? Moreover, if we consider EOI as potentially providing some positive

contributions to development in the early phase of treatment, then typically EE-based interventions developed for a more chronic population, may have unintended negative consequences. As an example, the focus on reducing EOI may potentially discourage caregivers' natural helping behaviour instead of praising their efforts, supporting their self-esteem and giving them a sense of hope. Of great importance in this regard, might be the study of concepts like hope, optimism and meaning as emphasized by the recovery movement (Snyder & Lopez, 2005). Many authors have called for EE based interventions to be more 'need-based' and 'refined' for them to be useful in first-episode psychosis (Gleeson *et al.*, 2010). Offering metacognitively oriented therapy (Lysaker, Buck, *et al.*, 2011) to relevant families could provide opportunities for caregivers to develop a broader, more complex and coherent understanding, or narrative of the illness – including their suffering, coping effort and rewarding experiences. Future studies should examine long-term effects of baseline levels of metacognition on EOI, caregiver distress and patient outcome. The level of overinvolvement is not a stable characteristic, but tends to fluctuate during the illness period (Bentsen *et al.*, 1996; Patterson, Birchwood, & Cochrane, 2000). An interesting question in this regard is whether overinvolvement is lower at follow-up in relatives with greater metacognitive capacity, and whether this affects the experience of distress and wellbeing, both within the patient and within the family. It may be that relatives with greater metacognitive capacity might regain a broader perspective and overview earlier than relatives with lower metacognitive capacity, thus 'stepping back' from overinvolvement as the patient develops/regains skills? Also, metacognition is a complex phenomenon, involving a poorer shared understanding between two persons, leading to fewer positive experiences in caregivers, but also potentially to difficulties in the patients. As an example, Meins *et al.* (2002), found 'parental mind-mindedness', defined as their proclivity to comment appropriately on their infants' mental states, to predict their children's later theory of mind understanding. Future studies thus should explore the interaction of metacognition in both the patient and the caregiver, including a detailed analysis of how the various metacognitive domains can explain variations in caregiver experience.

### **Conclusion**

The experience of positive and negative aspects of the relationship and caregiver burden seems to be associated with different variables. Negative experiences are more closely related to EOI and distress, as measured by GHQ, while the experience of positive and rewarding experiences of caregiving is related to the level of metacognition. The level of metacognition may be a relevant factor in understanding the relatives' ability for, and approach to, caregiving in first-episode psychosis. Greater levels of metacognition do not alleviate suffering and distress, which is a healthy and normal reaction to having a relative with serious mental illness. However, greater capacity to form complex ideas about oneself and others seems to allow for a more balanced perspective of caregiving, in which there is room for both positive and negative experiences.

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# Paper 4





# The Role of Metacognitions in Expressed Emotion and Distress: A Study on Caregivers of Persons with First-Episode Psychosis

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**Background:** In first-episode psychosis, the family is considered an important part in the recovery process. This is often accompanied by significant distress, which is acknowledged in numerous studies. However, little is known about the psychological factors involved.

**Method:** One hundred and twenty-seven caregivers of persons with first-episode psychosis completed a series of questionnaires aimed at investigating the contribution of expressed emotion and metacognitions to caregiver distress.

**Results:** Linear mixed model analysis found that emotional over-involvement and metacognitions independently predicted caregiver distress. Mediation analysis using bootstrapping showed that emotional over-involvement could be seen as mediating the effect of metacognitions on distress.

**Conclusion:** The current study is a first step towards understanding the role of metacognitions in caregiver distress, thus opening up for the possibility of using interventions from 'contextual behaviour therapies'. Implications and future studies are discussed. Copyright © 2014 John Wiley & Sons, Ltd.

## Key Practitioner Message:

- This study is the first attempt to address caregiver concerns from the perspective of contextual cognitive behavioural therapy.
- The metacognitive framework for caregiver distress in first-episode psychosis may help develop new therapeutic interventions to better support families.
- The study proposes a psychological understanding of emotional over-involvement and caregiver distress.

**Keywords:** Caregivers, Distress, Emotional Over-involvement, Metacognition, First-episode Psychosis, Contextual Behaviour Therapies

## INTRODUCTION

In first-episode psychosis, the family is considered an important part in the recovery process, and many relatives go to great lengths in supporting their ill family member (Glynn, 2012; Szmukler *et al.*, 2003). Unfortunately, this often involves a great deal of distress including anxiety, depression and economic strain (Addington, Coldham, Jones, Ko & Addington, 2003; Kuipers, Bebbington & Barrowclough, 2005; Szmukler, Burgess *et al.*, 1996). Traditionally, studies on caregivers

have been concerned with the relationship between caregiver behaviour and relapse (Pilling *et al.*, 2002), but recently, there has been an increased focus on the psychological well-being of caregivers themselves (Breitborde, López & Kopelowicz, 2010; Kuipers *et al.*, 2005).

The concept of expressed emotion (EE)—defined as caregiver criticism, hostility and over-involvement—has been the cornerstone of research on caregivers and is considered as a robust predictor of relapse in schizophrenia (Brown, Monck, Carstairs & Wing, 1962; Kuipers *et al.*, 2006; Pilling *et al.*, 2002). Recently, however, the concept has been questioned from a number of sources. First, it has been argued that the components of EE are related to different variables and are best studied separately. Alvarez-Jimenez and colleagues (Alvarez-Jimenez *et al.*, 2012; Alvarez-Jimenez *et al.*, 2008) found that only

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criticism seemed to predict relapse, while emotional over-involvement was more strongly related to caregiver distress. Second, the view of EE as a marker for a 'dysfunctional family' has been criticized (Jansen *et al.*, 2013; Van Os, Marcelis, Germeys, Graven & Delespaul, 2001), suggesting that especially over-involvement is a natural and understandable reaction to a crisis and is associated with caregivers who are involved in treatment and care. Finally, it has been argued that while family interventions aimed at reducing EE have reduced relapse in persons with chronic schizophrenia, the results are somewhat mixed in studies of first-episode psychosis (Bird *et al.*, 2010; Linszen *et al.*, 1996). While the association between emotional over-involvement and relapse in first-episode psychosis is being questioned, there has been an increased interest in the impact that it has on caregivers' own mental well-being (Breitborde, López, Chang, Kopelowicz & Zarate, 2009).

Increasingly, caregiver experience is recognized as a complex phenomenon involving both internal and external factors on the part of the caregivers (Jansen *et al.*, 2013; Joyce *et al.*, 2003). While earlier studies focused on 'objective burden' (e.g., Baronet, 1999), more recent studies have focused on distress as a subjective experience that is best understood within a stress-coping framework (Lazarus & Folkman, 1984) (Kuipers, Onwumere & Bebbington, 2010; Szmukler, Herrman, Bloch, Colusa & Benson, 1996). Studies into caregivers' appraisals and attributions have led the way to a cognitively based family approach, viewing emotional over-involvement partly as a coping strategy (Kuipers *et al.*, 2010; Raune, Kuipers & Bebbington, 2004). Moreover, McNab and colleagues (McNab, Haslam & Burnett, 2007) found that caregivers who were more distressed tended to endorse beliefs in the efficacy of more problematic kinds of interpersonal behaviour, particularly in the importance of self-sacrifice.

However, there is still limited knowledge about the psychological factors underlying emotional over-involvement and caregiver distress, and the question as to why some caregivers engage in more emotional over-involved behaviour and become more distressed than others remains largely unsolved. One way to further our understanding of the psychological factors underlying emotional over-involvement may be provided by the account of 'unhelpful' coping of Wells and Matthews (1996) in their Self-Regulatory Executive Function (S-REF) model. In the S-REF model, it is argued that people have a set of specific metacognitions that guide, influence and control their way of responding to their own cognitive experiences. Examples of metacognitions include 'I need to control my thoughts at all times', 'If I worry I will be prepared', and 'I cannot stop thinking bad thoughts'. Metacognitions are believed to be involved in the activation and perseveration of the 'cognitive

attention syndrome' (CAS). The CAS is characterized by perseverative thinking (rumination and worry), threat monitoring, attentional inflexibility and dysfunctional coping. The activation of CAS under stress causes a reduction in the cognitive resources needed to control attention, leading to coping behaviours that maintain, exacerbate or prolong distress.

Studies have found metacognitions to be relevant to a range of disorders including addictive behaviours (Spada, Nikčević, Moneta & Wells, 2008; Spada & Wells, 2005), depression (Papageorgiou & Wells, 2001), general anxiety disorder (Wells, 2010), psychosis (Morrison & Wells, 2003) and post-traumatic stress disorder (Wells & Sembi, 2004). Finally, and particularly relevant to the present study, Spada and colleagues (2012) found over-protection in parents to be associated with worry and anxiety in their children, and they suggested that this was partly mediated by the development of maladaptive metacognitions.

To address these questions, we gathered reports on metacognitions, EE and distress in caregivers of persons with first-episode psychosis. We then sought to examine the relationship between caregivers' specific metacognitions, EE and their level of distress. First, we expected that caregivers with higher levels of metacognitions would report higher levels of distress. Second, we expected that emotional over-involvement would be associated with higher levels of distress. Finally, we explored whether caregivers with greater levels of metacognitions would engage in more over-involved behaviour, leading to more distress. That is, whether emotional over-involvement would function as a mediator between metacognitions and distress.

## METHOD

### *Participants and Procedure*

The study had a cross-sectional design, and participants were 127 relatives of persons with first-episode psychosis (age 18–35 years old). The sample is drawn from a larger intervention study seeking to compare multi-family interventions with individual-family interventions and was included consecutively. Eligible individuals were caregivers of patients who (a) met International Classification of Diseases 10 criteria for schizophrenia, (b) previously had not received any treatment for psychosis, (c) had not received anti-psychotic medication for more than 6 months and (d) were currently enrolled in an outpatient treatment service based on assertive community treatment principles (Petersen *et al.*, 2005). All patients in Region Zealand, Denmark, were approached for participation between April 2011 and April 2013. The only exclusion criterion was an inability to speak Danish, either on the part of the patient

or the caregiver. In the event, no participants were excluded on this criterion.

Of the 147 patients enrolled in treatment during this period, 116 (78.9%) gave consent. Three (2%) were not relevant due to change of diagnosis, and six (4.1%) later withdrew from the study (because of leaving treatment). From the remaining 107 patients, 127 relatives were included in the study. The age of the caregivers ranged from 21 to 87 years old (mean = 49.5, standard deviation [SD] = 9.2, median = 50). Seventy-nine (62.2%) of the participants were female. Seventy-two of the participants (56.7%) lived together with the patients, and 107 (84.3%) had weekly contact. They included 102 (80.3%) parents, 5 (3.9%) partners, 7 (5.5%) step-parents, 3 (2.3%) grandparents and 4 (3.1%) siblings.

Diagnoses were confirmed using operational criteria (McGuffin, 1991) by a clinical psychologist or psychiatrist. Once patients enrolled in the treatment, they were asked permission to being informed about the research project by one of the research staff. Upon agreement, oral and written information about the study was given to both patient and relatives. Following written consent, caregivers were given questionnaires in the order that they are presented below. The duration was approximately 20 min per participant. All participants were debriefed following the completion of the questionnaires. The Regional Committee for Research Ethics approved the study.

### *Self-Report Measures*

Family Questionnaire (FQ; Wiedemann *et al.*, 2002). Caregivers' level of EE was assessed by the FQ, which comprises 20 items measured on a 4-point Likert scale rated as 'never/very seldom', 'seldom', 'often' or 'very often'. They were asked about their way of handling everyday challenges using questions like 'I have a tendency to neglect myself because of her/him' and 'she/he irritates me'. The measure consists of two subscales: emotional over-involvement (EOI) and critical comments (CCs). EOI includes over-intrusive, self-sacrificing, over-protective behaviour or exaggerated emotional response and over-identification with the patient; CC is defined as unfavourable comments on the behaviour or the personality of the patient (Vaughn & Leff, 1976). A third variable—hostility—is normally associated with high levels of CC. The items are scored from 1 to 4 yielding a maximum score of 40 in each subgroup. Caregivers are classified as high EE if they score 23 or greater on the CC subscale or 27 or greater on the EOI subscale. Wiedemann *et al.* (2002) found the FQ to have good psychometric properties including a clear factor structure, good internal consistency of subscales and good concurrent validity in relation to the widely used Camberwell Family Interview (Vaughn & Leff, 1976). They also found that the FQ displayed

similar level of accuracy and higher sensitivity compared with the Five Minute Speech Sample (Magaña *et al.*, 1986).

General Health Questionnaire 30 (GHQ-30; Goldberg & Williams, 1988). The level of distress was measured using the GHQ-30, which is a 30-item self-report questionnaire. For each item, the caregivers must rate the occurrence of a particular symptom on a 4-point Likert scale ranging from 0 to 3. The total score could thus vary from 0 to 90. The four response categories to the positively worded items are labelled 'better than usual/more so than usual', 'same as usual', 'less than usual' and 'much less than usual'. The response categories for the negatively worded items are 'not at all', 'no more than usual', 'more than usual' and 'much more than usual'. A GHQ-case score was also calculated, each item scored as 'present' or 'absent' (0–0–1–1), where the total score could vary from 0 to 30. A case score above 5 is usually considered to be high and suggesting clinically significant distress (Goldberg & Williams, 1988).

Metacognitions Questionnaire 30 (MCQ-30; Wells & Cartwright-Hatton, 2004). Individual differences in metacognitions were measured by the MCQ-30, which is a 30-item self-report questionnaire with five subscales: (a) positive beliefs about worrying (e.g., 'Worrying helps me avoid problems in the future'); (b) negative beliefs about worry concerning uncontrollability and danger (e.g., 'When I start worrying I can't stop'); (c) cognitive confidence (e.g., 'I do not trust my memory'); (d) beliefs about need to control thoughts (e.g., 'If I did not control a worrying thought and then it happened it would be my fault'); and (e) cognitive consciousness (e.g., 'I constantly observe my thoughts'). Each statement is rated on a 4-point Likert scale as 'agree very much', 'agree moderately', 'agree slightly' and 'do not agree' with a score ranging from 30 to 120. The MCQ-30 has good internal consistency and convergent validity, as well as acceptable test-retest reliability (Spada, Mohiyeddini & Wells, 2008; Wells & Cartwright-Hatton, 2004). The MCQ-30 was developed and validated on a normal sample. To our knowledge, this is the first study using the MCQ-30 to assess metacognitions in caregivers of persons with first-episode psychosis.

### *Statistical analyses*

Statistical analyses were carried out using SPSS version 20 for Mac (IBM Inc., Chicago, IL, USA). Since we intended to test the relationship between metacognitions, EOI and distress, the first part of the analysis involved computing Pearson's correlations. As some of the patients had two caregivers participating, a linear mixed model analysis was carried out to account for the possible correlation between two caregivers (Gueorguieva & Krystal, 2004; O'Connor, 2004). In order to test the prediction that EOI

mediates the relationship between metacognitions and distress, we performed a mediation analyses following the recommended criteria set out by Preacher and Hayes (2004, 2008b). The statistical significance of the indirect coefficients ( $a*b$ ) was tested using a non-parametric bootstrapping method. Randomly drawing participants from the original data set and replacing each value as it was sampled created 5000 identically sized data sets. Confidence intervals for the 5000  $a*b$  values were then derived using  $z$ -score biased corrections. In this approach, mediation is significant if the upper and lower bounds of confidence intervals do not contain 0 (Efron & Tibshirani, 1993).

While there are many approaches to statistical inference for indirect effects and their relative performance has been studied extensively (Hayes, 2013), the bias-corrected bootstrapping confidence interval currently seems to be the preferred approach (Breitborde, Srihari, Pollard, Addington & Woods, 2010; Mackinnon, Lockwood & Williams, 2004; Zhao, Lynch & Chen, 2010). Although the use of mediation analysis on cross-sectional data has been questioned (Cole & Maxwell, 2003; Maxwell & Cole, 2007), the current study uses mediation analysis as a first attempt to explore the hypothesis of EOI being a mediator, following the suggestions by Hayes (2013) and Mackinnon (2008).

## RESULTS

Descriptive statistics and Pearson's product-moment correlations for distress, EE (EOI and criticism) and the five metacognitions subscales are presented in Table 1. Data were examined for skewness, kurtosis and outliers by visual inspection of histograms and scatterplots, and they were found symmetrically distributed and without outliers. There were positive and significant associations between distress and three of the metacognition subscales (negative beliefs about worry concerning uncontrollability

and danger, cognitive confidence and beliefs about need to control thoughts). There were also positive and significant associations between distress and the two subscales of EE (over-involvement and criticism). In this sample, 44 (28.2%) caregivers could be characterized as emotionally over-involved (scoring  $\geq 27$  on FQ, EOI subscale), while 33 (21.2%) could be characterized as critical (scoring  $\geq 23$  on FQ, CC subscale). It is noteworthy that 47 (37%) of the caregivers had a GHQ-case score of  $>5$  suggesting clinical significant distress.

### Linear Mixed Model Analysis

To evaluate whether metacognitions and EE (EOI and criticism) predicted distress, a linear mixed model analysis was run with distress (GHQ-30) as an outcome (Table 2). The two subscales of EE (EOI and CC) and the three metacognitions that were found to be significant in the correlation analysis (5, 6 and 7 in Table 1) were included in the model. The mixed model analysis showed that negative beliefs about worry concerning uncontrollability and danger and EOI were significant predictors of distress.

### Mediation Analysis

In order to explore the hypothesis that EOI mediates the relationship between metacognitions and distress, a mediation analysis was carried out. Because both the  $a$ -path ( $B = .45$ ,  $SE = 0.10$ ,  $p < 0.001$ ) and the  $b$ -path ( $B = 1.43$ ,  $SE = 0.22$ ,  $p < 0.001$ ) were significant, premises for the mediation analysis using bootstrapping with bias-corrected confidence estimates were met (Mackinnon *et al.*, 2004; Preacher & Hayes, 2004, 2008a). The results of the mediation analysis were consistent with a possible mediational role of EOI in the relation between negative beliefs about worry concerning uncontrollability and danger and distress in caregivers ( $B = 1.45$ ;  $CI = 0.37$  to  $1.06$ ). The results indicated that the direct effect of negative beliefs about worry

Table 1. Descriptive statistics and Pearson product-moment correlations of study variables

Study variables	M	SD	1	2	3	4	5	6	7	8
1. GHQ-30—caregiver distress	29.83	14.29	—	0.58**	0.30*	0.13	0.42**	0.33**	0.26**	0.09
2. FQ-EOI—emotional over-involvement	23.36	4.89	—	0.51**	0.22	0.39**	0.22*	0.21**	0.09	
3. FQ-CC—critical comments	18.13	5.23	—	—	0.14	0.23*	0.09	0.23**	0.12	
4. MCQ-30-1—positive beliefs about worry	9.66	3.90	—	—	—	0.36**	0.14	0.41**	0.57**	
5. MCQ-30-2—negative beliefs about worry concerning uncontrollability and danger	11.48	4.17	—	—	—	—	0.59**	0.59**	0.45**	
6. MCQ-30-3—beliefs about cognitive confidence	10.68	4.57	—	—	—	—	—	—	0.44**	0.29**
7. MCQ-30-4—beliefs about the need to control thoughts	9.22	3.38	—	—	—	—	—	—	—	0.52**
8. MCQ-30-5—cognitive self-consciousness	11.59	4.44	—	—	—	—	—	—	—	—

Note:  $n = 127$ . GHQ-30 = General Health Questionnaire-30. FQ-EOI = Family Questionnaire-emotional over-involvement subscale. FQ-CC = Family Questionnaire-critical comment subscale. MCQ-30 = Metacognitions Questionnaire-30.

\* $p < 0.05$ . \*\* $p < 0.01$ .

Table 2. Linear mixed model analysis with distress as dependent variable and dimensions of expressed emotion and metacognitions as predictor variables

Predictors	B	95% CI	p
FQ-emotional over-involvement	1.30	0.76 to 1.82	<0.001
FQ-critical comments	0.03	-0.45 to 0.52	0.89
MCQ-2 uncontrollability and danger	0.73	0.04 to 1.43	<0.05
MCQ-3 cognitive confidence	0.36	-0.21 to 0.92	0.21
MCQ-4 need to control thoughts	-0.02	-0.77 to 0.73	0.96

Note:  $n = 124$ . FQ = Family Questionnaire. MCQ-30 = Metacognitions Questionnaire-30. MCQ-2 refers to negative beliefs about worry concerning uncontrollability and danger, MCQ-3 refers to cognitive confidence, and MCQ-4 refers to and beliefs about need to control thoughts.

concerning uncontrollability and danger on distress was reduced but remained significant ( $B = 0.80$ ,  $SE = 0.26$ ,  $p < 0.001$ ) when controlling for emotional over-involvement, thus suggesting only partial mediation. This suggests that negative beliefs about worry concerning uncontrollability and danger influence caregivers' distress directly as well as indirectly through emotional over-involvement ( $B = 1.45$ ,  $SE = 0.28$ ,  $p < 0.001$ ). Figure 1 displays the results.

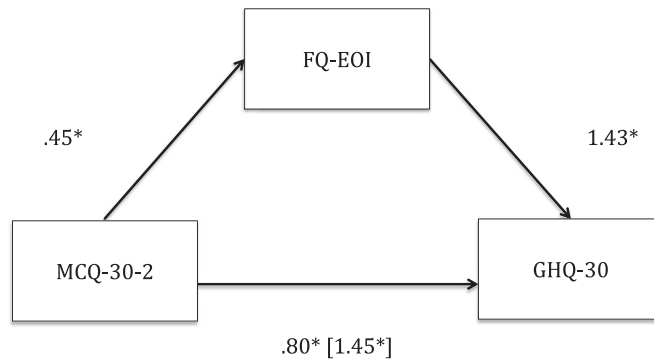
DISCUSSION

The current study examined the relationship between metacognitions, EE and caregiver distress. First, consistent with our prediction, there was a positive and significant

association between metacognitions and distress. Caregivers with higher levels of dysfunctional metacognitions reported higher levels of distress. Second, also consistent with our prediction, there was a positive and significant association between EOI and distress. A linear mixed model analysis showed that negative beliefs about worry concerning uncontrollability and danger, and EOI, were significant predictors of distress. Finally, we explored a mediation model according to which caregivers with greater levels of negative beliefs about worry concerning uncontrollability and danger would engage in more over-involved behaviour, leading to increased distress. Overall, our findings lend support to the hypotheses proposed and are consistent with the metacognitive model of psychological dysfunction. According to this model, metacognitions contribute to distress by inducing persons to engage in coping strategies that are somewhat unhelpful and counterproductive.

The positive associations between distress and the three dimensions of metacognitions (negative beliefs about worry concerning uncontrollability and danger, cognitive confidence and beliefs about the need to control thoughts) are in keeping with earlier studies. According to Spada, Georgiou and Wells (2010), these three subscales have consistently been found to be implicated across many domains of emotional difficulties.

In terms of the theoretical understanding of EE, the findings are also in line with the view that the two dimensions are related to different factors and that EOI is more closely related to distress in the caregivers than criticism (Álvarez-Jiménez *et al.*, 2008; Breitborde *et al.*, 2009). When



Note:  $N = 124$ . FQ-EOI = Family Questionnaire – Emotional Over-involvement subscale; GHQ-30 = General Health Questionnaire; MCQ-30 = Metacognitions Questionnaire-30; MCQ-2 refers to negative beliefs about worry concerning uncontrollability and danger. \* $p < .001$ .

Figure 1. Mediation model.  $n = 124$ . FQ-EOI = Family Questionnaire-emotional over-involvement subscale. GHQ-30 = General Health Questionnaire. MCQ-30 = Metacognitions Questionnaire-30. MCQ-2 refers to negative beliefs about worry concerning uncontrollability and danger. \* $p < 0.001$

distress was regressed onto EE, only EOI remained as a significant predictor. The findings also lend some support to the view that EOI can be seen as a coping mechanism (Kuipers *et al.*, 2006; Raune *et al.*, 2004).

One way to interpret these findings in relation to the S-REF model (Wells, 2000; Wells & Matthews, 1996) is that caregivers respond with worry and distress to the crisis of having a close one being diagnosed with schizophrenia, this being a normal and healthy reaction. However, some caregivers tend to engage in more unhelpful coping (EOI) such as over-intrusiveness, self-sacrificing, over-protective behaviour, exaggerated emotional response or over-identification with the patient. These coping strategies often work in the short term, by giving some sense of control over the situation, but become detrimental in the long term causing higher levels of emotional distress in the caregivers and possibly also the persons with psychosis. Most caregivers would periodically be emotionally over-involved and distressed, and this often fluctuates during the course of the patient's illness (Patterson, Birchwood & Cochrane, 2000), but for some, the distress is exacerbated and prolonged. According to Wells (2000), metacognitions such as beliefs about worry concerning uncontrollability and danger may lead to dysfunctional coping in terms of rumination and worrying, suppression of threatening thoughts and hyper-vigilant monitoring of potential threats. This form of 'coping' tends to backfire and cause more emotional suffering and distress.

With replication and confirmation through longitudinal designed studies, these preliminary findings can have a number of therapeutic implications for supporting caregivers. First, considering people's metacognitions as an important driving mechanism for unhelpful coping adds to the existing literature of cognitive appraisals and attributions in understanding caregiver distress. Second, understanding EOI as a coping mechanism within a metacognitive model can broaden the intervention repertoire to encompass interventions from 'contextual behaviour therapies' (Hayes, Villatte, Levin & Hildebrandt, 2011), including detached mindfulness, attention training and modification of dysfunctional metacognitions (Wells, 2000; Wells & Matthews, 1994).

It is recognized that there are several limitations to this study, and the results must be considered with these in mind. First, self-report biases and social desirability may have contributed to errors in the self-report measures that we used. Second, the cross-sectional design does not allow for causal inferences. It is not possible to rule out, for instance, that distress was increasing the report of metacognitions or EOI. While the application of mediation analyses to cross-sectional designs has been justified by experts in the field (Hayes, 2013), future studies should also address causality in longitudinal and experimental designs. However, the relationship between EE, coping

and distress probably includes many mediators and moderators that need to be studied further in terms of their effect on both caregivers and persons with psychosis (Breitborde, Srihari *et al.*, 2010). Third, it could be that the EOI subscale of FQ actually is tapping into worrying, and it is still somewhat unclear what aspects of EOI are causing distress. Also, in terms of the metacognitive model, future studies should expand on the measurement of coping strategies used in this study to test the S-REF model more comprehensively. As an example, avoidant coping, thought suppression and rumination could be studied as possible mediators and social support and positive caregiver experiences as possible moderators of caregiver distress. Longitudinal studies could also explore whether people with high levels of metacognitions have prolonged periods of EOI compared with persons with low levels of metacognitions and how this relationship influences the level of distress.

Despite these limitations, the current study provides preliminary support for a metacognitive theory of distress in caregivers, which incorporate earlier theories of EOI as an understandable, but to an extent also unhelpful, coping effort to deal with distress. With replication, this may expand the intervention repertoire used in family support programmes to better tailor the needs of the family.

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