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COMPREHENSIVE REVIEW

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Family environmental factors in at-risk mental states for psychosis

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Abstract

The family environment represents an important psychosocial factor that impacts psychosis prognosis, but little is known about its effect on the at-risk stages of psychosis. This study presents a comprehensive review and summarizes the state of the art of study on the wide range of family factors related to family functioning in the At-Risk Mental State (ARMS) for psychosis, as well as family interventions in ARMS individuals. Publications were retrieved by an extensive search on MEDLINE, PsycINFO and SCOPUS (1990–2020). Expressed Emotion is the most studied variable in ARMS literature, but there is scarce evidence of the role of other significant family factors at the ARMS stage. Overall, high Expressed Emotion did not appear to be reactive to ARMS patients' poor clinical status. However, initial evidence has suggested that relatives' beliefs about the disorder may play a significant role, either as mediators of these relationships or as predictors of Expressed Emotion. Available literature yet to yield a consistent pattern of findings on the association between Expressed Emotion or other family functioning indicators and negative outcomes, but some longitudinal studies highlight the greater potential for the protective effects of positive family environments at the ARMS stage. Family-based interventions have demonstrated benefits for both ARMS individuals and family dynamics. An increased focus on the impact of the at-risk stage of illness on relatives' mental well-being is required to provide family support based on their needs and to clarify the mechanisms leading to dysfunctional family dynamics during the critical ARMS period.

KEYWORDS
caregivers, Expressed Emotion, family functioning, family interventions, high risk for psychosis, review

1 | INTRODUCTION

The family environment has been extensively studied as an influential psychosocial factor that impacts psychosis prognosis (Butzlaff & Hooley, 1998; Kavanagh, 1992; Schlosser et al., 2012). In this context, family members' emotional attitudes towards the patient, as measured by the level of Expressed Emotion (EE; Brown et al., 1972), have received most of the attention in psychosocial research. High EE (characterized by the presence of elevated levels of criticism, hostility and/or Emotional Over-Involvement [EOI]) has consistently shown to be a robust predictor of relapse in Schizophrenia (Cechnicki et al., 2013; Marom et al., 2005; O'Driscoll et al., 2019; Wearden et al., 2000).

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Within the literature on caregivers of individuals with Schizophrenia, the theoretical attributional model (Barrowclough et al., 1994; Barrowclough & Hooley, 2003) was developed for elucidating the relationships between relatives' illness attributions about psychosis and EE. The attributional model postulates that relatives' beliefs about the causes of the patients' illness are linked to relatives' emotional attitudes towards patients. In this regard, critical relatives tend to blame patients for their behaviours because they believe that the patient can control their symptoms, whereas relatives who feel excessively blameworthy regarding the patient's illness may resort to over-involvement or self-sacrificing attitudes (Bentsen et al., 1998).

A parallel line of research focused on the impact of the psychotic disorder on the family well-being has demonstrated that the responsibility of caring for a family member with a psychotic disorder can lead to show elevated levels of distress, anxiety and depression in caregivers (Barrowclough et al., 1996; Jansen, Gleeson, & Cotton, 2015; Jungbauer & Angermeyer, 2002). To date, most research on family factors has mainly been carried out on patients with chronic forms of the psychosis phenotype (for a review, see Butzlaff & Hooley, 1998; Caqueo-Urízar et al., 2014; Hooley, 2007; Miklowitz, 2004) or individuals with recent onset of psychosis (i.e., first episode of psychosis [FEP] patients) (for a review, see Jansen, Gleeson, & Cotton, 2015; Koutra et al., 2014). Conversely, the family environment has been less studied in the phases preceding the onset of illness (i.e., high risk for psychosis or At-Risk Mental State [ARMS] stage).

The ARMS stage comprises a heterogeneous group of symptoms traditionally described during the prodromal phase of psychosis. Current standard definitions and operationalization yield three clusters of individuals: (1) people with attenuated positive psychotic symptoms; (2) people who have experienced brief intermittent episodes of frank psychotic symptoms lasting no more than a week with spontaneous full recovery; and (3) individuals with either genetic risk (having a first-degree relative diagnosed with a psychotic disorder) or meeting the criteria for Schizotypal Personality Disorder in addition to a significant decrease in social functioning in both cases (McGlashan et al., 2001; Miller et al., 2003; Yung et al., 2004).

The study of family factors at the ARMS stage has the advantage of not being biased by the plethora of confounding factors associated with chronic and/or more advanced stages of the illness (e.g., the onset of marked clinical symptoms, impairment, major co-morbidity and severe medication side effects) and could ultimately improve understanding of the early development of high-EE attitudes for prevention purposes. Furthermore, since family factors may play a critical role in the management and outcome of psychosis, developing an understanding of the family functioning at the very early stages of psychosis can help develop early interventions designed to prevent negative dynamics in the family environment.

Although the study of EE has played a central role in family environment research, different theoretical frameworks of family functioning have emphasized other-dimensional indicators of family welfare, such as levels of cohesion and adaptability within the family, quality of parent–child communication and/or problem-solving skills. Importantly, findings from previous research show that relatives caring for an individual with psychosis are at-risk for difficulties in family functioning related to cohesion, flexibility and/or poor coping styles (Friedmann et al., 1997; Gupta & Bowie, 2018; Phillips et al., 1998; Raune et al., 2004). Furthermore, it appears that these overall family functioning indicators may have an impact on psychiatric outcomes (Gurak & Weisman de Mamani, 2016; Weisman, 2005). This parallel yet less explored line of research considers other family-related constructs, which are of great relevance and warrant further discussion, especially in the earlier stages of psychosis.

Key Practitioner Message

- Relatives' psychological needs in the at-risk for psychosis period have been overlooked, and there is an urgent need for more research to better understand the impact of the pre-psychotic stage on relatives' mental well-being.
- Longitudinal studies highlight the significant role of positive family aspects as predictors of at-risk for psychosis patients' clinical and/or functional improvement over time.
- The effect of criticism on ARMS patients' clinical course is inconsistent across studies, while emotional over-involvement appears to act as a protective factor and could have some positive effects on patients' outcomes at the ARMS stage.
- Relatives' illness attributions seem to be an important predictor factor and mediator in the relationship between expressed emotion and ARMS clinical/functional features.
- Family-based interventions have demonstrated benefits for both ARMS individuals and family dynamics.

To date, only one systematic review has so far attempted to describe the effects of the EE construct during the at-risk for psychosis stage (Izon et al., 2018), but it is limited only to EE, without analysing its relationships with other significant family factors involved in family functioning that could explain the potential mechanisms underlying the manifestation of EE in at-risk stages. Therefore, the present study review seeks to expand Izon et al. (2018) by providing the state of the art of study on the wide range of family environmental factors related to family functioning at the pre-psychotic stage and discuss the evidence they provide in light of existing theories on EE development (Barrowclough et al., 1994; Barrowclough & Hooley, 2003; Hooley, 2007; Kavanagh, 1992; Kuipers et al., 2006; Miklowitz, 2004; Patterson, 2013; Patterson et al., 2000, 2005; Raune et al., 2004). Moreover, given the central role of the EE construct in this field, the examination of findings is followed by an in-depth analysis of the possible explanatory models that could account for the emergence of EE in the earliest stage of psychosis.

The research reviewed is divided into four different sections:

1. studies investigating the association of EE and other family environment constructs with ARMS symptoms/functioning and/or relatives' psychological variables;

TABLE 1 Summary of studies researching family environment variables and family interventions in the At-Risk Mental Stage (ARMS) of psychosis

Study	Location	Design and participants	Family variables	Family measures	Key findings
Studies investigating the association of EE and other family environment constructs with ARMS symptoms and/or functioning and/or relatives' psychological variables					
McFarlane and Cook (2007)	Portland, New Rochelle and Kingston (USA)	Cross-sectional study 69 subjects with established psychotic disorders and their parents 50 subjects with prodromal symptoms and their parents	EE	SAS-III (relatives' self-reports)	- Criticism and EOI were higher in parents of patients with established psychotic disorders than in parents of ARMS individuals. - Warmth was significantly higher in ARMS families than in parents of patients with established psychotic disorders. - In ARMS families, maternal criticism and EOI increased over time, whereas maternal and parental warmth decreased over time after the first signs of illness began to appear.
Meneghelli et al. (2011)	Milan (Italy)	Cross-sectional study 77 FEP and their relatives 66 ARMS and their relatives	EE	CFI (interview based on relatives' reports)	FEP families: - High EE was related to longer DUI. - High paternal EOI related to DUP. - Patients' severity of symptoms/functioning not related to EE. ARMS families: - High EE not related to DUI. - The severity of illness/functioning not related to EE.
Smith et al. (2018)	London (UK)	Cross-sectional study 80 FEP relatives and ARMS relatives	EE Subjective appraisals of caregiving Perceived mental well-being	CFI (interview based on relatives' reports) ECI (relatives' self-reports) SF-36 (relatives' self-reports)	- One third of the sample reported at least one incident of patient-initiated violence. - Reports of violence were associated with poorer mental well-being scores among caregivers and more negative appraisals of caregiving. - Patient-initiated violence also correlated with greater criticism and hostility expressed towards patients.

(Continues)

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
Hamaie et al. (2016)	Sendai (Japan)	Cross-sectional study 43 FEP and their relatives 56 ARMS and their relatives	EE	FAS (relatives' self-reports)	FEP relatives: - Criticism was associated with relatives' higher depression symptoms and with FEP patients' negative and general symptoms. - Criticism was explained by relatives' depression symptoms and FEP patients' general symptoms in the regression analyses. ARMS relatives: - Criticism was unrelated to ARMS patients' clinical status. Criticism was explained by relatives' higher educational levels in the regression analyses.
Hinojosa-Marqués et al. (2020)	Barcelona (Spain)	Prospective study, 6-month follow-up 55 ARMS and 36 FEP patients and their relatives at baseline 33 relatives of ARMS and 13 relatives of FEP patients at follow-up	EE Perceptions of illness Distress	FQ (relatives' self-reports) IPQS-R (relatives' self-reports) SCL-90-R (relatives' self-reports)	- Relatives' psychological distress and illness attributions predicted criticism and EOI over and above patients' illness characteristics at baseline and 6-month follow-up. - Relatives' increased levels of anxiety, attributions of blame towards the patients, emotional negative representation about the disorder and decreased levels of self-blame attributions predicted EE criticism at baseline. - Relatives' anxiety and negative emotional representation of the disorder (i.e., negative emotional response to the patient's mental health problems) were the only

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
					significant predictors of EE criticism at follow-up, whereas anxiety, attributions of control by the relative and an emotional negative representation about the disorder predicted EE-EOI both at baseline and in 6-month follow-up assessments.
Domínguez-Martínez et al. (2017)	Barcelona (Spain)	Cross-sectional study 37 FEP relatives 41 ARMS relatives	EE Perceptions of illness Distress	FQ (relatives' self-reports) IPQS-R (relatives' self-reports) SCL-90-R (relatives' self-reports)	- Anxiety, depression and attributions of blame towards the patient predicted relatives' criticism in both ARMS and FEP relatives' groups. - Attributions of control by the patient and emotional negative representation of the disorder predicted relatives' EOI. - Anxiety more strongly associated with relatives' criticism in ARMS relatives than in FEP relatives. - Anxiety associated with relatives' EOI in ARMS but not in FEP relatives.
Hinojosa-Marqués, Domínguez-Martínez, Cristóbal-Narváez, et al. (2019)	Barcelona (Spain)	Cross-sectional study 34 ARMS relatives 21 FEP relatives	EE Domains of ESM questionnaire[a]	FQ (relatives' self-reports) ESM questionnaire (relatives' self-reports)	- Momentary criticism and EOI were significantly associated with the two FQ-EE dimensions, respectively, supporting the criterion validity of real-world assessed EE dimensions. - Momentary and FQ-EE dimensions were associated with decreased positive affect, as well as with appraisals of less effective coping in daily life. - Only momentary EE dimensions were

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TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
					associated with increased momentary negative affect. - Momentary criticism and FQ-criticism were more consistently related to situational stress and burden than momentary EOI and FQ-EOI. - Neither momentary nor FQ-EE dimensions showed distinct patterns of associations with illness attributions. - Findings partly support the construct validity of momentary criticism and EOI as well as the construct and ecological validity of the FQ as a sensitive measure of EE.
Hinojosa-Marqués, Domínguez-Martínez, Sheinbaum, et al. (2019)	Barcelona (Spain)	Cross-sectional study 30 FEP relatives 48 ARMS relatives	EE Perceived loss Attachment	FQ (relatives' self-reports) MIV-TIG (relatives' self-reports) PAM (relatives' self-reports)	- Relatives' perceived loss was associated with EE dimensions. - Relatives' attachment anxiety, but not avoidance, mediated the relationship of perceived loss with both criticism and EOI.
Tsai et al. (2015)	Los Angeles (USA)	Cross-sectional study 49 ARMS Non-Latino White: 38 Latino: 11	EE	PC and PW (patients' self-reports)	- Analyses examining the entire sample showed that patients' perceived levels of criticism were negatively associated with ARMS negative symptomatology. - Race/ethnicity moderated the relationship between perceived criticism/warmth and CHR clinical symptomatology.
Carol and Mittal (2015)	Boulder (USA)	Cross-sectional study 37 ARMS 42 control group	EE	FMSS (interview based on relatives' reports)	- Relatives of ARMS provided significantly fewer positive comments about patients than relatives of the control group.

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
					- No significant differences emerged between relatives of ARMS and the control group in criticism or EOI. - Relatives' criticism was related at the trend level to ARMS patients' positive symptoms. - Relatives' EOI, warmth and positive comments were unrelated to ARMS patients' clinical status.
Domínguez-Martínez et al. (2014)	Barcelona (Spain)	Cross-sectional study 24 FEP and their relatives 20 ARMS and their relatives	EE Perceptions of illness	FQ (relatives' self-reports) IPQS-R (relatives' self-reports)	- Relatives' EOI was associated with patients' higher negative and general symptoms, and worse social and role functioning. - Relatives' criticism was associated with patients' higher positive, negative and general symptoms, depressive symptoms and worse social and role functioning. - The association between EE and patients' symptoms/functioning did not differ between FEP and ARMS groups. - Attributions of blame towards the patient mediated most of the relationships between EE and illness severity.
Welsh and Tiffin (2015)	Northeast England (UK)	Cross-sectional study 26 FEP 44 ARMS 140 control group	Family perceptions	FPS (patients' self-reports)	- ARMS and FEP showed more maladaptive family perceptions than the control group. - ARMS showed poorer perceived problem-solving and nurturing behaviour than the control group. - Family perceptions not related to symptom severity in

(Continues)

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
					either ARMS and FEP groups; however, higher scores on the EE subscale of the FPS were related to manic symptomatology in both ARMS and FEP groups.
Bentley et al. (2016)	Baltimore (USA)	Cross-sectional study 36 ARMS 60 help-seeking control group	Quality of parent–child relationship	BASC-2 (patients' self-reports)	- ARMS reported less positive parent–child relationships (P-CH-R) and a higher perception of social stress than the control group. - P-CH-R moderated the relationship between ARMS diagnosis and perception of social stress. - ARMS informing poorer P-CH-R reported increased perceptions of social stress. - ARMS informing positive P-CH-R reported decreased perceptions of social stress.
Thompson et al. (2019)	Maryland (USA)	Cross-sectional study 52 ARMS	Family perceptions	FAD (ARMS' self-reports)	- Perceived family functioning moderated the effect of ARMS patients' symptoms on social/role functioning. - For individuals who perceived lower levels of family functioning, symptoms were moderately associated with social and role functioning. - For individuals who perceived higher levels of family functioning, symptoms were not significantly associated with social/role functioning. - Positive symptoms and perceived family

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
					functioning were not associated, suggesting that perceived family functioning did not directly impact symptom severity and vice versa.
Peh et al. (2020)	Singapore (Singapore)	Cross-sectional study 164 ARMS 510 controls	Parental bonding	PBI (patients' self-reports)	- ARMS individuals were more likely to report having affectionless-controlling mothers, significantly lower maternal and paternal care and higher maternal and paternal overprotection than controls. - Higher paternal overprotection was significantly associated with a range of worse clinical and functioning outcomes among ARMS individuals.
Predictive value of EE and other family environment constructs on symptoms and/or functional ARMS outcomes					
O'Brien et al. (2006)	Los Angeles (USA)	Prospective study, 3-month follow-up 26 ARMS and their primary caregivers at baseline and follow-up	EE	CFI (interview based on relatives' reports)	- Relatives' criticism at baseline was not associated with changes in symptoms or social functioning at follow-up. - Relatives' EOI at baseline was associated with improvement in negative symptoms and social functioning at follow-up. - Relatives' positive remarks at baseline were associated with improvements in negative and disorganized symptoms at follow-up. - Relatives' warmth at baseline was associated with improvements in social functioning at follow-up.

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TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
Schlosser et al. (2010)	Los Angeles (USA)	Prospective study, 6-month follow-up 63 ARMS and their relatives at baseline and follow-up	EE	CFI (interview based on relatives' reports) PC and PW (patients' self-reports) FMPC and FMPW (relatives' self-reports)	- Patients' perceived criticism at baseline significantly predicted worsening of attenuated positive symptoms at follow-up. - Relatives' criticism and hostility at baseline (as measured by CFI) significantly predicted worsening of attenuated positive symptoms at follow-up. - Relatives' EOI (moderate levels) and relatives' warmth (as measured by CFI) interacted such that they jointly predicted improved functioning at follow-up.
O'Brien et al. (2008)	Los Angeles (USA)	Prospective study, 4-month follow-up 40 ARMS and their primary caregivers at baseline and follow-up	EE Supportive and conflict-engaging behaviour	CFI (interview based on relatives' reports) FIT (interactional patterns)	- Relatives' positive remarks at baseline were associated with a decrease in negative symptoms at follow-up. - Relatives' warmth at baseline predicted an improvement in social functioning at follow-up. - Relatives' behaviours exhibited during the interactional task were not predictive of ARMS patients' symptoms/functioning at follow-up.
O'Brien et al. (2009)	Los Angeles (USA)	Prospective study, 6-month follow-up 33 ARMS and their primary caregivers at baseline 27 ARMS and their primary caregivers at follow-up	Social problem-solving behaviour	Problem-solving discussion (interactional patterns) FIT (interactional patterns) CFI (interview based on relatives' reports)	- Baseline ARMS' skilful problem-solving, constructive communication and relatives' constructive communication were associated with ARMS' enhanced social functioning at follow-up. - Baseline ARMS' conflictual communication was

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
					associated with increased positive symptoms at follow-up.
Wang et al. (2015)	Shanghai (China)	Prospective study, 6-month follow-up 32 ARMS from the general population at baseline 25 ARMS from the general population at follow-up 256 control group	Perceptions of family functioning Family cohesion Family adaptability	FAD (ARMS' self-reports) FACES-II (ARMS' self-reports)	- ARMS reported worse family functioning than the control group. - Positive perception of problem-solving and affective responsiveness from parents at baseline predicted less severe positive and negative symptoms at follow-up. - Better family cohesion and adaptability were associated with decreased general symptoms at baseline. - Better family cohesion and adaptability at baseline were associated with decreased disorganized and general symptoms at follow-up.
Haidl et al. (2018)	Multicentric European study	Prospective study, 18-month follow-up 235 ARMS at baseline 205 ARMS at follow-up	EE	LEE (patients' self-reports)	- Patients' perceived irritability was found to be a predictor of conversion for ARMS into FEP. - The importance of this family environmental risk factor was further demonstrated by an improvement of risk estimation in the original European Prediction of Psychosis Study (EPOS) predictor model.
Family environmental factors as the main outcome explored in ARMS families					
Wong et al. (2008)	New York (USA)	Cross-sectional study 12 FEP and their relatives 11 ARMS and their relatives	Burden	FEIS (interview based on relatives' reports)	- Levels of both subjective and objective burden were comparable between ARMS and FEP relatives.
Gerson et al. (2011)	New York (USA)	Cross-sectional study 12 FEP relatives	Coping	CCOPE (relatives' self-reports)	- Families reported moderate use of

(Continues)

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
		11 ARMS relatives			'approach' coping and rare use of 'avoidant' coping strategies.
Yee et al. (2020)		Cross-sectional study 75 ARMS 79 healthy controls	Coping with family stress	RSQ (patients' self-report)	- ARMS and control groups reported similar levels of engagement strategies. - ARMS were more likely to use disengagement strategies when coping with family stress than the healthy control group. - Engagement strategies predicted greater perceptions of the availability of advice support. - Disengagement strategies were associated with anxiety and depression symptoms and predicted lower perceived social support from the family and greater family strain.
Santesteban-Echarri et al. (2018)	Toronto (Canada)	Cross-sectional study 52 ARMS with early mood or anxiety symptoms 108 ARMS with subthreshold psychotic symptoms 41 non-help-seeking youth with risk factors 42 control group	Family functioning	FACES-IV (patients' self-reports)	- Families in all groups fell within ranges that reflected healthy family functioning. - Family satisfaction is lower in youth at risk for severe mental illness who present with early signs of mood, anxiety or subthreshold psychotic symptoms.
Salinger et al. (2018)	8 research centres of the NAPLS-2 consortium	Cross-sectional study 58 ARMS and their respective relatives 58 at risk for bipolar disorder patients and their respective relatives	Family communication	Problem-solving discussion (interactional patterns)	- Mothers of youth at clinical high risk for psychosis showed significantly more conflictual and less constructive communication during problem-solving discussions than did mothers of youth at clinical high risk for bipolar disorder.

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
Baron et al. (2019)	San Francisco (USA)	Qualitative cross-sectional study 12 ARMS relatives	Associative stigma	Semi-structured interviews	- Perceptions of stigma and coping with stigma emerged as main domains according to parents' experiences. - Parents expressed the increased experience of the stigma associated with psychotic-like symptoms as compared to other psychiatric symptoms. - Parents attempted to combating stigma through public disclosure with adolescents' privacy and autonomy needs. - Parents generally found that participation in research is less stigmatizing than non-speciality care. - Stigma impacted treatment-seeking and participation in family groups.
Izon et al. (2019)	Manchester (UK)	Qualitative cross-sectional study 14 ARMS relatives	Barriers and facilitators when providing support to ARMS patients	Semi-structured interviews	- Key barriers included a lack of understanding of individual's symptoms/ behaviour and how to support them, as well as unproductive coping strategies impacting on their health or/and well-being. - Key facilitators included early access and involvement of services, access to social support and open communication between family/ carers and the individual.
Izon et al. (2020)	Manchester (UK)	Qualitative prospective study 10 ARMS relatives	Family/carers' experiences supporting ARMS	Semi-structured interviews	- Over 12 months, four factors were important for family/carers to

(Continues)

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
			individuals over 12 months		facilitate their caring role: (a) looking after their well-being; (b) accessing additional support from family intervention; (c) communicating openly with the individual; and (d) engaging with services for the individual. - All these aspects were important for improving family communication, meeting family/carers' unmet needs and helping them feel more confident and less isolated in their role as carers.
Family intervention studies					
O'Brien et al. (2014)	Eight research centres in the NAPLS-2 consortium	Randomized control trial Prospective study, 6-month follow up Family Focused Therapy for individuals at clinical high risk (FFT-CHR) with 18 sessions over 6 months vs. three-session family psychoeducational intervention 51 ARMS and their relatives at baseline 38 ARMS and their relatives at follow-up	Family communication	Problem-solving discussion (interactional patterns)	- ARMS and their respective families assigned to FFT-CHR showed greater improvement from baseline to follow-up in constructive communication and greater decreases in conflictual behaviours than those who participated in a brief psychoeducational intervention. - FFT-CHR was found to be effective in improving active listening and calm communication and decreasing irritability, anger, complaints and criticism in both patients and family caregivers.
Miklowitz et al. (2014)	Eight research centres in the NAPLS-2 consortium	Randomized control trial Prospective study, 6-month follow up. Family Focused Therapy for individuals at clinical high risk (FFT-CHR) with 18 sessions	-	-	- ARMS participants assigned to FFT-CHR showed greater improvement in positive symptoms from baseline to follow-up than participants

TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
		over 6 months vs. three family psychoeducation sessions FFT-CHR was administered to patients and their parents 129 ARMS at baseline 102 ARMS at follow-up			assigned to a brief family psychoeducational intervention (three sessions of family psychoeducation). - Negative symptoms improved independently of psychosocial treatments. - Changes in psychosocial functioning over time were age dependent: patients over 19 showed greater role improvement in FFT-CHR, whereas participants aged between 16 and 19 showed more role improvement in the brief psychoeducational intervention.
O'Brien et al. (2015)	Eight research centres in the NAPLS-2 consortium	Randomized control trial Prospective study, 6-month follow up Family Focused Therapy for individuals at clinical high risk (FFT-CHR) with 18 sessions over 6 months vs. three-session family psychoeducational intervention FFT-CHR was administered to patients and their parents 90 ARMS and their mothers at baseline 41 ARMS and their mothers at follow-up	EE	PC (patients' and mothers' self-reports)	- Perceived maternal criticism decreased from pre- (baseline) to post-treatment (6 months) for both treatment groups (FFT-CHR and brief psychoeducational intervention), and these changes in criticism predicted decreases in attenuated positive symptoms at 12-month follow-up. - Decreases in mothers' reports of criticism were marginally significant predictors ($p = .06$, two-tailed).
McFarlane et al. (2015)	Six mental health agencies and settings belonging to the Early Detection and Intervention for the Prevention of Psychosis Program (EDIPPP) (United States)	Quasi-experimental multisite trial Prospective study, 6-, 12- and 24-month follow-up Family-Aided Assertive Community Treatment (FACT) with a minimum duration of 1 year vs. community care	Only patient's outcome data were reported	-	- No differences in transition between groups were found. - The FACT showed some beneficial effects over community care in reduction of symptoms and improved global functioning.

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TABLE 1 (Continued)

Study	Location	Design and participants	Family variables	Family measures	Key findings
		Interventions were administered to young people at risk for psychosis and their families 337 ARMS and their relatives at baseline 222 ARMS and their relatives at 24-month follow-up			- Over the 24-month follow-up period, the FACT group increased its level of participation in work and school. - Effects for symptoms, functioning and global outcomes proved to be larger for the group having very early psychosis than for the clinical high-risk group.
Landa et al., 2016		Open uncontrolled pilot trial Prospective study, 3-month follow-up Group- and Family-based Cognitive Behavioural Therapy (GF-CBT) for youth at risk for psychosis for 15 weeks 6 ARMS individuals and family members at baseline 5 ARMS individuals and 4 family members at 3-month follow-up			GF-CBT showed significant decreases in symptoms and improvement in the functioning of ARMS individuals. Family members showed significant improvements in the use of CBT skills, enhanced communication with their offspring and greater confidence in their ability to help.
Law et al. (2021)	Two mental health trusts in Northwest England (UK)	Single (rater), blinded, randomized feasibility study Cross-sectional study Combined Individual and Family Cognitive Behavioural Therapy (IFCBT) plus enhanced treatment as usual (ETAU) vs. ETAU alone for ARMS and their family/carers 70 ARMS and their family/carers		FMSS (interview based on relatives' reports) FQ (relatives' self-reports) FMPC and FMPW (relatives' self-reports) BDI (relatives' self-reports) SIAS (relatives' self-reports) ECR-RS (relatives' self-reports)	- The study recruited 92% of the target, showing that it is feasible to identify and recruit participants. - ARMS individuals presented with high levels of co-morbid emotional disorders such as moderate to severe depression and high levels of social anxiety. - CAARMS scores revealed high levels of distress in relation to symptoms, particularly on the non-bizarre ideas' subscale. - Family members/carers also presented with moderate depression, anxiety and physical health problems.

Note: The order of appearance of the studies follows the comments on the results in the body text to facilitate reading.
Abbreviations: ARMS, At-Risk Mental State; BASC-2, Behaviour Assessment System for Children, Second Edition; BDI, Beck Depression Inventory; CCOPE, Carver's Coping Orientations to Problems Experienced Questionnaire; CFI, Camberwell Family Interview; DUI, duration of untreated illness; DUP, duration of untreated psychosis; ECI, Experience of Caregiving Inventory; ECR-RS, Relationship Structure Questionnaire; EE, Expressed Emotion; EOI, Emotional Over-Involvement; FACES-II, Family Adaptability and Cohesion Evaluation Scale II; FACES-IV, Family Adaptability and Cohesion Evaluation Scale IV; FAD, Family Assessment Device; FAS, Family Attitude Scale; FEIS, Family Experiences Interview Schedule; FEP, first episode of psychosis; FIT, The Family Interaction Task; FMPC, family member's perceived criticism; FMPW, family member's perceived warmth; FMSS, The Five Minute Speech Sample; FPS, Family Perception Scale; FQ, Family Questionnaire; IPQS-R, Illness Perception Questionnaire for Schizophrenia Relatives' version; LEE, Level of Expressed Emotion Scale; MIV-TIG, Mental Illness Version of the Texas Inventory of Grief; NAPLS-2 consortium, Emory University, Harvard University, University of Calgary, University of California Los Angeles, University of California San Diego, University of North Carolina, Yale University and Zucker Hillside Hospital; NAPLS-2, North American Prodrome Longitudinal Study; PAM, Psychosis Attachment Measure; PBI, Parental Bonding Instrument; PC, Perceived Criticism Scale; PW, Perceived Warmth Scale; RSQ, Responses to Stress Questionnaire; SAS-III, Social Adjustment Scale-III; SCL-90-R, Symptom Checklist-90-Revised; SF-36, The Short Form-36 Health Survey; SIAS, Social Interaction Anxiety Scale.

[a] Momentary EE, affect at the moment, appraisals of effective coping, appraisals of the current situation, appraisals of burden, illness attributions, appraisals related to the self, positive appraisals of the patient and appraisals of the patient when relatives were in direct contact and/or had had recent contact with the patient.

2. the predictive value of EE and other family environment constructs on ARMS symptoms and/or functional outcomes;
3. family environmental factors (other than EE) explored in ARMS families; and
4. the effect of family interventions on symptomatic and/or functional ARMS outcomes.

2 | METHOD

2.1 | Search strategy

Publications were retrieved through an extensive search of three electronic databases: MEDLINE, PsycINFO and SCOPUS (1990–2020). The search was conducted using the terms 'expressed emotion', 'family expressed emotion', 'emotional over-involvement', 'criticism', 'hostility', 'warmth', 'family environment', 'family functioning', 'family cohesion', 'family adaptability', 'family burden', 'family coping', 'family distress', 'relatives' illness attributions', 'caregivers', 'carers', 'relatives', 'at-risk mental states', 'prodrome', 'ultra-high risk', 'clinical high-risk for psychosis' and 'at-risk for psychosis'.

2.2 | Inclusion/exclusion criteria

Inclusion criteria: (1) published in peer-reviewed journals in English; (2) date of publication from 1990 to 2020 (December); (3) samples of ARMS patients and/or their relatives; and (4) research focused on (a) the impact of illness on family members' attitudes or psychological health; (b) the influence of family environment variables on ARMS' clinical and/or functional outcomes; (c) relatives' psychological factors accounting for EE; (d) the effects of family interventions on both ARMS' clinical and/or functional outcomes; and (e) studies with at least one family measurement and/or relevant constructs to understand the early emergence of EE.

Exclusion criteria: (1) secondary publications (commentaries, editorials and letters); (2) theses, dissertations and conference papers; and (3) case reports. We did not limit the search to studies that only included biological mothers and fathers but also included studies that recruited other relatives, such as stepparents, siblings and partners. Additionally, we also included studies assessing family factors in mixed samples of ARMS patients and/or their family members with healthy controls, FEP and/or clinical psychosis individuals given that mixed samples are common in the early psychosis literature.

3 | RESULTS

A total of 35 studies met the inclusion criteria. Table 1 summarizes the main characteristics of the studies included.

3.1 | Study aims

The aims and scope of the reviewed studies varied greatly. Regarding the family environment variables, 15 studies (42.9%) explored EE (Carol & Mittal, 2015; Domínguez-Martínez et al., 2014, 2017; Haidl et al., 2018; Hamaie et al., 2016; Hinojosa-Marqués et al., 2020; Hinojosa-Marqués, Domínguez-Martínez, Cristóbal-Narváez, et al., 2019; Hinojosa-Marqués, Domínguez-Martínez, Sheinbaum, et al., 2019; McFarlane & Cook, 2007; Meneghelli et al., 2011; O'Brien et al., 2006, 2008; Schlosser et al., 2010; Smith et al., 2018; Tsai et al., 2015) and 14 (40%) explored other relevant family environment constructs as the main outcome (such as burden, problem-solving skills, communication, cohesion, adaptability, perception of the quality of relationships within the family, family functioning, coping, barriers and facilitators in the caring role) (Baron et al., 2019; Bentley et al., 2016; Gerson et al., 2011; Izon et al., 2019, 2020; O'Brien et al., 2009; Peh et al., 2020; Salinger et al., 2018; Santesteban-Echarri et al., 2018; Thompson et al., 2019; Wang et al., 2015; Welsh & Tiffin, 2015; Wong et al., 2008; Yee et al., 2020). Finally, six studies (17.1%) focused on family-based interventions and their effects on ARMS clinical outcomes, high-EE attitudes and/or family communication (Landa et al., 2016; Law et al., 2021; McFarlane et al., 2015; Miklowitz et al., 2014; O'Brien et al., 2014, 2015). Reviewed studies also varied in their assessment methods. While some of them assessed the family's emotional climate through relatives' self-reports and/or interviews based on relatives' reports (Carol & Mittal, 2015;

Domínguez-Martínez et al., 2014, 2017; Gerson et al., 2011; Hamaie et al., 2016; Hinojosa-Marqués et al., 2020; Hinojosa-Marqués, Domínguez-Martínez, Cristóbal-Narváez, et al., 2019; Hinojosa-Marqués, Domínguez-Martínez, Sheinbaum, et al., 2019; McFarlane & Cook, 2007; Meneghelli et al., 2011; O'Brien et al., 2006, 2008, 2009; Smith et al., 2018; Wong et al., 2008), other studies assessed family variables from the patient's viewpoint (Bentley et al., 2016; Haidl et al., 2018; Peh et al., 2020; Santesteban-Echarri et al., 2018; Thompson et al., 2019; Tsai et al., 2015; Wang et al., 2015; Welsh & Tiffin, 2015; Yee et al., 2020), only two studies considered both patients' and relatives' perspectives (O'Brien et al., 2015; Schlosser et al., 2010) and three very recent studies used a qualitative methodology approach (Baron et al., 2019; Izon et al., 2019, 2020). Family interaction tasks were also used to assess family-related variables (O'Brien et al., 2008, 2009, 2014; Salinger et al., 2018).

3.2 | Studies investigating the association of EE and other family environment constructs with ARMS symptoms/functioning and/or relatives' psychological variables

The scant existing literature on the relationship between EE and specific symptoms and functioning in ARMS patients contains a substantial level of contradictory findings, much more so than the research conducted on patients with chronic Schizophrenia.

Some cross-sectional studies have aimed to explore the assumption that EE develops as a reaction to patients' symptom severity by comparing the prevalence of EE at different stages of illness severity and/or by using retrospective reports on the duration of patients' symptoms to examine their impact on EE (McFarlane & Cook, 2007; Meneghelli et al., 2011). McFarlane and Cook (2007) did not use patients' measures to test their association with EE and instead compared EE levels between families of patients with clinical psychosis and ARMS individuals. Furthermore, they analysed the impact of relatives' perceived duration of prodromal symptoms on EE. They found that parents of individuals with established psychotic disorders reported higher levels of criticism and EOI and lower levels of warmth than parents of ARMS individuals. Further analyses conducted only within the ARMS sample revealed that levels of maternal criticism and EOI increased once the first signs of illness began to appear, whereas maternal and paternal warmth decreased over the pre-psychotic development. The latter analyses were conducted using retrospective reports from parents regarding the duration of the subpsychotic syndrome. This in turn provided a measure of parents' perceived duration of the prodromal symptoms, which was analysed in association with the current EE values. The authors suggested that EE is a reaction to patients' deterioration, that is, increasing symptom severity and disability across the ARMS stage. Conversely, Meneghelli et al. (2011) stated that the duration of untreated illness was not related to EE in relatives of ARMS patients. Both duration of untreated illness and duration of untreated psychosis were measured from the relatives' perspective. Results also showed that families of FEP and ARMS patients have the same prevalence of high EE mostly due to EOI and that EE was not related to the severity of symptoms or psychosocial functioning in either group. The authors concluded that results partially support the assumption that high-EE attitudes develop as a reaction to patients' clinical/functional status. This led them to speculate the existence of intrinsic components regarding family functioning which might also be involved in the genesis of EE (e.g., relatives' beliefs about the illness). In a slightly different way, Smith et al. (2018) set out to examine how other patient-related variables (not strictly clinical), such as patient-initiated violence, impacted the manifestation of EE, as well as on relatives' negative appraisals of caregiving and perceived mental well-being. They found that relatives' reports of violence in patients were associated with poorer mental well-being, more negative appraisals of caregiving and greater criticism and hostility expressed towards patients.

In a recent Japanese study, Hamaie et al. (2016) assessed the impact of patients' clinical characteristics on relatives' criticism by also considering the contribution of relatives' psychological factors, in a sample of 56 ARMS and 43 FEP patients with their caregivers. Findings showed that ARMS caregivers' criticism was not related to patients' clinical status or caregivers' depressive symptomatology but rather to caregivers' higher educational levels. Authors suggested that high levels of education may contribute to developing higher expectations regarding the capacity for control of ARMS individuals over their illness-related behaviours, which in turn would be expressed in critical attitudes. In a more comprehensive predictive model of EE, Hinojosa-Marqués et al. (2020) also examined the contribution of early psychosis clinical and functional status to EE along with relatives' psychological factors. They explored the association of relatives' EE dimensions (criticism and EOI) with relatives' psychological distress and illness attributions and with patients' clinical/functioning features at baseline and 6-month follow-up in 91 dyads of early psychosis patients and their relatives (comprising 55 ARMS and 36 FEP patients). Furthermore, they explored whether relatives' psychological factors predicted EE dimensions over and above patients' baseline clinical and functional status at both time points. The most relevant finding of this study revealed that relatives' psychological distress and subjective appraisals of the illness accounted for significant variance over and above patients' clinical and functional status in the prediction of criticism and EOI both at baseline and at 6-month follow-up (please see Table 1 for more details of specific results).

Other studies have placed greater emphasis on the relevance of examining relatives' psychological variables to identify the mechanisms underlying EE. Specifically, Domínguez-Martínez et al. (2017) highlighted the role of relatives' distress and cognitive representations of illness. Results from this study showed that relatives' distress and attributions of blaming patients for their illness predicted criticism in both ARMS and FEP caregivers, whereas beliefs that symptoms are within patients' control and an emotional negative representation about the illness predicted EOI in both groups. On a correlational level, relatives' EE dimensions (i.e., criticism and EOI) were highly associated with relatives' distress and several types of illness attributions. Also, comparisons between groups revealed that ARMS relatives scored higher on criticism than FEP relatives. Moreover, anxiety was more strongly related to criticism in ARMS than FEP relatives and associated with EOI in ARMS but not FEP relatives.

Findings from Hinojosa-Marqués, Domínguez-Martínez, Cristóbal-Narváez, et al. (2019) also highlighted the fact that EE dimensions (criticism and EOI) were significantly associated with relatives' negative affective states and negative illness attributions in the realm of daily life by using the Experience Sampling Method (ESM) in relatives of ARMS and FEP patients. The authors examined the way EE dimensions, as measured by momentary and psychometric self-reports (Family Questionnaire [FQ]; Wiedemann et al., 2002), were expressed in daily life and related to a wide variety of real-world experiences. Overall, these findings provide a valid ecological insight of EE correlates, thus pointing to the role of negative affective states and negative illness attributions as potential mechanisms underlying the expression of EE (please see Table 1 for more details of specific findings). Further research (Hinojosa-Marqués, Domínguez-Martínez, Sheinbaum, et al., 2019) underscored the importance of investigating the role of relatives' perceived loss in the development of EE in early psychosis caregivers (i.e., ARMS and FEP relatives). No differences emerged between ARMS and FEP relatives in terms of perceived loss. Findings indicated that relatives' perceived loss was associated with both EE dimensions and that relatives' attachment anxiety, but not attachment avoidance, mediated the relationship between perceived loss and EE components. These findings therefore also highlighted the importance of examining the role of relatives' attachment characteristics for understanding how perceptions of loss could impact the manifestation of EE attitudes at the early stages of psychosis.

Alternative cross-sectional studies focused mainly on examining the influence of EE on ARMS patients' symptoms and/or functioning (Domínguez-Martínez et al., 2014; Tsai et al., 2015). Carol and Mittal (2015) have a comparable goal but also explored the relationship between EE and ARMS endocrine activity and self-concept. Findings showed that relatives of ARMS reported similar levels of criticism and EOI but significantly lower frequency of positive comments about patients in comparison with relatives of healthy individuals. Analyses of the ARMS group suggested that relatives' criticism (but not EOI) was related at the trend level to ARMS patients' positive symptoms. Neither relatives' warmth nor positive comments were related to patients' symptomatology. Results also revealed a moderate correlation at the trend level between relatives' criticism and ARMS' heightened levels of resting cortisol. Moreover, relatives' criticism was related to ARMS increased negative self-concept while relatives' warmth was associated with ARMS increased positive belief about the self.

Tsai et al. (2015) found that higher levels of patients' perceived maternal criticism were associated with lower levels of negative symptoms across the entire sample of ARMS young adults. Authors speculated that criticism could also reflect mothers' involvement or engagement in their offspring's care, thus serving as a protective factor in adolescent populations. Further analysis indicated that race/ethnicity moderated the relationship between patients' perceived levels of parental warmth/criticism and patients' symptom expression (please see Table 1 for more details of specific findings). Although the results showed cross-cultural differences among ARMS patients, the study has several limitations that do not allow to generalization of the results.

Domínguez-Martínez et al. (2014) showed that relatives' criticism and EOI were related to increased several symptom dimensions and functional impairment at both the ARMS and FEP stages of psychosis. No differences emerged between ARMS and FEP relatives in terms of EE or its relationship with patients' symptoms/functioning, suggesting that both patients' samples were similarly influenced by family environment variables. Going a step further, this study analysed the potential role of relatives' attributions in the association between EE and patients' symptoms. It was found that relatives' attributions of blaming patients for their illness-related difficulties mediated most of the relationships between EE and illness severity.

Some studies have examined how other family variables, as rated by ARMS patients' perceptions, are related to patients' symptom expression in the subclinical stages of psychosis. Welsh and Tiffin (2015) showed that ARMS and FEP patients scored significantly higher on EE in comparison with a community control sample. Interestingly, ARMS patients were likely to report poorer perceived problem-solving and lower levels of nurturing behaviour in their families compared with the community control group. Self-reported family perceptions were not related to symptom severity in either ARMS or FEP groups, although higher scores on the EE subscale of the self-reported family perceptions measure were significantly related to manic symptomatology in both ARMS and FEP groups. Bentley et al. (2016) found that ARMS individuals reported less positive parent–child relationships and higher levels of social stress than those who did not meet ARMS criteria but receive mental health services. Parent–child relationships moderated the relationship between ARMS diagnosis and social stress. The severity of social stress was only dependent on the quality of family relationships in the ARMS group. ARMS individuals who reported poorer parent–child relationships tended to report greater perceptions of social stress, whereas those who described positive parent–child relationships tended to report lower perceptions of social stress. Similarly, Thompson et al. (2019) showed that ARMS individuals' perceived family functioning moderated the effect of symptoms on social/role functioning. Specifically, in patients who perceived lower levels of family functioning, psychosis-risk symptoms were moderately associated with social and role functioning. However, this effect was not present in patients reporting higher levels of family functioning. A recent study (Peh et al., 2020) explored parent–child relationships in ARMS individuals ($n = 164$), compared to controls ($n = 510$), in an Asian setting by exploring the association between parental bonding, symptom severity and functioning. Findings showed that, compared with controls, ARMS individuals were more likely to report having affectionless, controlling mothers, significantly lower maternal and paternal care and higher maternal and paternal overprotection. Moreover, higher paternal overprotection was significantly associated with a range of worse clinical and functioning outcomes among ARMS individuals.

3.3 | Predictive value of EE and other family environment constructs on ARMS symptoms and/or functional outcomes

Preliminary research examining family environment as a predictor of outcome in ARMS patients underlined the important role of a positive family environment. In a longitudinal study of 26 ARMS adolescents and their primary caregivers, O'Brien et al. (2006) indicated that caregivers' criticism (assessed by the Camberwell Family Interview [CFI; Vaughn & Leff, 1976]) at baseline was unrelated to changes in symptoms or social functioning at 3-month follow-up. However, caregivers' EOI at baseline was associated with improvements in negative symptoms and social functioning at follow-up. Likewise, caregivers' positive remarks were associated with an improvement in negative and disorganized symptoms at follow-up, and caregivers' warmth was also associated with enhanced social functioning at follow-up. Authors suggested that EOI could be developmentally appropriate in adolescence, acting as a protective factor. Along the same lines, Schlosser et al. (2010) suggested that when EOI is reported at moderate levels and in interaction with warmth can act as a protective factor and improved functioning over time. These authors also found that ARMS patients' perception of parental criticism predicted a worsening of attenuated positive symptoms at follow-up. Moreover, interview-based ratings of relatives' hostility and criticism significantly predicted a change in positive symptoms at follow-up. In another study, O'Brien et al. (2008) showed that caregivers' warmth predicted an improvement in social functioning while caregivers' positive remarks were associated with a decrease in negative symptoms 4 months later. Besides, it was observed that parents who expressed more positive remarks regarding their ill family member during the CFI tended to exhibit more constructive behaviours during an interactional task. Besides, criticism was positively related to parents' conflict engaging behaviours during the interactional task. Finally, the behaviours exhibited by relatives during the interactional task were not predictive of ARMS patients' symptoms/functioning at a 4-month follow-up. However, in a 6-month follow-up report, O'Brien et al. (2009) showed that ARMS individuals' skilful problem-solving and constructive communication as well as caregivers' constructive communication exhibited during face-to-face problem-solving discussions were associated with enhanced social functioning of patients at follow-up. Conversely, ARMS individuals' conflicting communication during the problem-solving discussion with their parents was associated with increased positive symptoms at follow-up.

In their longitudinal study of 32 ARMS from a non-clinical population, Wang et al. (2015) found that self-reported family functioning, specifically, better perceived problem-solving and affective responsiveness from their parents predicted less severe positive and negative symptoms at 6-month follow-up. Perceived family cohesion and adaptability were negatively associated with general symptoms at baseline but were also negatively associated with general and disorganized symptoms at follow-up.

As part of the European Prediction of Psychosis Study (EPOS) designed to develop a prediction model of psychosis, Haidl et al. (2018) followed up a large cohort of 235 ARMS individuals during 18 months. Results revealed that ‘perceived irritability’, a domain of the Level of Expressed Emotion Scale (LEE; Cole & Kazarian, 1988), was found to be predictive for the conversion to psychosis. That is, patients' perception of irritability (responsive to stress and less able to cope with it) by a key relative was a predictor of conversion from high-risk status to FEP. The predictive value of this family environmental risk factor was further underpinned by an improvement of risk estimation in the original EPOS psychosis prediction model (Ruhrmann et al., 2010) by improving risk prediction above *0.90*.

3.4 | Family environmental factors (other than EE) explored in ARMS families

Families' psycho-emotional and/or coping factors have scarcely been explored as the primary outcome (regardless of relatives' EE levels or the course of the illness in the affected relative) in the at-risk for psychosis stage.

In a small exploratory study aimed at examining family burden in the early stages of psychosis, Wong et al. (2008) indicated that levels of both subjective and objective burden were comparable between ARMS and FEP caregivers. Family members also report that they help patients with their everyday activities yet deny any resentment or subjective burden. They also denied the need to monitor or control behaviours in patients. Worry was a common feeling among family members, but their lives have not been altered, and they do not have much anger or resentment.

Two studies have focused on coping styles but from different perspectives. On the one hand, Gerson et al. (2011) explored self-reported coping styles in a small cohort of families of individuals with ARMS and FEP. They found that families reported moderate use of ‘approach’ coping (such as planning, social support seeking, positive reinterpretation, acceptance and turning to religion) and little use of ‘avoidant’ coping strategies (such as denial/disengagement and alcohol and drug use). On the other hand, Yee et al. (2020) focused on how ARMS individuals cope with stress with their relatives as one of the most significant stressors in the family environment. Specifically, this study examined differences in coping strategies (i.e., engagement and disengagement) in reaction to family stressors between ARMS individuals and healthy controls, as well as their association with social support and clinical symptoms. In comparison with the control group, ARMS individuals did not differ in their use of most engagement coping strategies (such as problem-solving, emotion regulation, positive thinking, cognitive change and distraction) but were more likely to use disengagement strategies (i.e., avoidance, denial and wishful thinking) when coping with family stress. Engagement strategies were linked to higher perceptions of social support (in the form of having a source to go to for advice and guidance), whereas the use of disengagement strategies was associated with anxiety and depression symptoms, as well as with lower perceptions of family support and increased family strain.

Other studies have focused on exploring aspects related to family functioning in the ARMS stage. Santesteban-Echarri et al. (2018)

examined differences in family functioning, family satisfaction and communication in a sample of participants at risk of severe mental illness across different clinical stages (i.e., non-help-seeking youth with risk factors, help-seeking youth with early mood and anxiety symptoms and distress, and youth with an attenuated psychiatric syndrome) and a control group. Findings showed that families in all groups fell within ranges that reflected healthy family functioning. Moreover, all the groups reported moderate levels of family communication, suggesting that participants generally feel good about communication patterns within the family. However, family satisfaction was lower in youth at risk for severe mental illness, who present with early signs of mood, anxiety or subthreshold psychotic symptoms, than other participants. Likewise, a recent study (Salinger et al., 2018) sought to examine whether families of youth at clinical high risk for psychosis or bipolar disorder differ in communication patterns during problem-solving discussions. Results indicated that mothers of youth at risk for psychosis displayed significantly more conflictual and less constructive communication than those of youth at risk for bipolar disorder.

The following studies used the qualitative methodology approach to explore relatives' experiences and family factors associated with caring for an ARMS family member. Baron et al. (2019) examined the lived experience of associative stigma in 12 parents of adolescents at clinical high risk for psychosis. This study aimed to empower families to tell their stories and use their narrative to make recommendations and improve families' experiences of coping with clinical high risk for psychosis identification. Perceptions of stigma and coping with stigma emerged as the main domains according to parents' experiences. The narrative of the participants showed that (a) there is an increased experience of the stigma associated with psychotic-like symptoms as compared to other psychiatric symptoms; (b) parents attempt to balance combatting stigma with the privacy needs of adolescents; and (c) stigma impacted treatment-seeking and participation in family groups that were seen as both potentially supportive and threatening. These findings indicate that parents of ARMS children are probably reconciling their vulnerability to associative stigma with their notions of mental illness, which may include stigmatizing beliefs. The other two qualitative studies (Izon et al., 2019, 2020) were conducted with the same sample of family/carers belonging to a larger Individual and Family Cognitive Behavioural Therapy trial study (Law et al., 2021). Izon et al. (2019) explored the barriers and facilitators for those supporting ARMS individuals through semi-structured interviews with 14 caregivers. Findings suggest that caring for someone with ARMS can have an emotional and psychological impact, as caregivers reported high levels of worry, uncertainty, distress, anger, feeling unsupported by health services, as well as depression symptoms and suicidal feeling, as needing the most immediate support from services. In summary, key barriers included relatives' unmet needs and limited confidence in accessing and providing support, whereas facilitating factors included open communication with the individual, flexibility, understanding employers and feeling that they were supported. A further follow-up qualitative study (Izon et al., 2020) was conducted to understand the different types of changes, similarities and reflections of those caregivers 12 months after taking part in Izon et al. (2019). Ten of the 14 caregivers completed the follow-up interview. Over 12 months, four factors were important for caregivers to facilitate their caring role: (a) looking after their well-being; (b) accessing additional support from family intervention; (c) communicating openly with the individual; and (d) engaging with services for the individual. All these aspects were important in improving family communication, meeting caregivers' unmet needs and helping them feel more confident and less isolated in their caring role.

3.5 | Effect of family interventions on symptomatic and/or functional ARMS outcomes

Several trials including family interventions for ARMS populations have been developed, most of which have been conducted within the eight-site North American Prodrome Longitudinal Study (NAPLS). The family-focused therapy, consisting of an 18-session intervention that included explicit communication and problem-solving training, was administered to CHR (FFT-CHR) relatives and patients. Results from O'Brien et al. (2014) indicated that ARMS individuals and their respective families showed greater improvement from baseline to 6-month follow-up in constructive communication (such as active listening) and greater decreases in conflicting behaviours (such as criticism) than those who participated in a brief psychoeducational intervention. Importantly, the FFT-CHR was found to be effective in reducing high-EE attitudes and improving positive communication within the family. Miklowitz et al. (2014) examined the effects of the FFT-CHR in reducing the risk for psychosis onset among 129 ARMS individuals. Patients undertaking FFT-CHR showed greater improvement in positive symptoms from baseline to 6 months than those assigned to a brief family psychoeducational intervention. Changes in psychosocial functioning over time were age dependent, such that patients over 20 demonstrated greater functional improvement in FFT-CHR, whereas those between the ages of 16 and 19 showed greater functional improvement in the brief psychoeducational intervention. O'Brien et al. (2015) explored whether FFT-CHR can reduce levels of perceived criticism and whether decreases in perceived criticism predicted an improvement in symptoms. Findings indicated that perceived criticism reduced from baseline to 6 months for both treatment groups (FFT-CHR and brief family psychoeducational intervention). A reduction in ARMS individuals' perceived criticism from baseline to 6 months predicted improvement in attenuated positive symptoms at 12 months over and above symptom improvement at 6 months.

In a pilot study with 337 youth (age 12–25) at risk of psychosis (McFarlane et al., 2015) from the multisite trial of the Early Detection and Intervention for the Prevention of Psychosis Program in the United States, participants were assigned to different treatment groups based on the severity of positive symptoms. Individuals at the highest risk of psychosis transition ($n = 250$) were assigned to a Family-Aided Assertive Community Treatment (FACT), while those with clinically lower risk ($n = 87$) were assigned to community care. Although there were no differences in transition, family treatment had certain beneficial effects over community care, as shown by the reduction of positive, negative, disorganized and general symptoms, increases in global functioning and greater overall improvement. Over the 24-month follow-up period, the FACT group increased their level of participation at work or school. Besides, effects for symptoms, functioning and global outcomes proved to be larger for the group having very early psychosis than in the clinical high-risk group (McFarlane et al., 2015).

A further small ($N = 6$), open, uncontrolled trial pilot study (Landa et al., 2016) examined a different Group- and Family-based Cognitive Behavioural Therapy (GF-CBT) intervention intended to facilitate psychosocial recovery, decrease symptoms and prevent transition to psychosis in at-risk youth. Findings showed statistically significant decreases in attenuated psychotic symptoms, negative symptoms, depression and improvements in functioning in at-risk youth, whereas family members showed significant improvements in the use of CBT skills, enhanced communication with their offspring and greater confidence in their ability to help. However, the results should be taken with caution because this study has important limitations including lack of control group, unblinded assessments and potential confounds and the short 3-month follow-up.

A recent study from a single-blind, pilot randomized controlled trial comparing a Combined Individual and Family Cognitive Behavioural Therapy intervention with treatment as usual in ARMS individuals and their key caregiver reported the results of the rationale, design and baseline characteristics of the study feasibility, but, given that it is currently underway, no data are yet available on the effectiveness of the trial (Law et al., 2021).

4 | DISCUSSION

To the best of our knowledge, this is the first review examining the wide range of factors related to family functioning in the ARMS stage. We sought to identify empirical studies that have explored (1) relationships of EE and other family environment constructs with ARMS symptoms/functioning and/or relatives' psychological variables; (2) the predictive value of EE and other family environment constructs on ARMS symptoms and/or functional outcomes; (3) family environmental factors (other than EE) explored in ARMS families; and (4) the effect of family interventions on clinical and/or functional ARMS outcomes.

This section is intended to present a theoretical discussion of the findings based on several questions addressed by the existing theoretical models of EE to analyse their usefulness in the understanding of the early emergence of EE at the risk stage for psychosis. For the remaining reviewed literature that cannot be dealt with using this theoretical perspective, a more general discussion of findings will be provided.

4.1 | Is EE a reaction to ARMS patients' clinical and functional characteristics?

Given that high-EE environments have consistently been related to poor outcomes among patients with Schizophrenia (Butzlaff & Hooley, 1998; Cechnicki et al., 2013; Marom et al., 2005; Wearden et al., 2000), some of the reviewed cross-sectional studies aimed to explore the factors contributing to the early manifestation of EE. One line of enquiry has been the examination of whether ARMS' concurrent clinical status impacted on relatives' EE (Hamaie et al., 2016; McFarlane & Cook, 2007; Meneghelli et al., 2011) since it has been suggested that patients' clinical features may predict relatives' levels of EE (for a review, see Hooley, 2007; Miklowitz, 2004). With exception of McFarlane and Cook (2007), the rest of the reviewed studies showed that relatives' high-EE status did not seem to be reactive to ARMS patients' poor clinical status (Hamaie et al., 2016; Meneghelli et al., 2011). However, as suggested by Smith et al. (2018), it seems that other patient-related variables, not restricted to clinical characteristics, such as patient-initiated violence, contribute to the expression of higher levels of criticism and hostility in relatives of ARMS patients.

It is important to note that most of the cross-sectional studies conducted in FEP samples also show inconsistent results regarding this issue. Although patients' poor clinical and functional status has been related to an increase in relatives' EE in some FEP studies (King, 2000; Koutra et al., 2016; Mo et al., 2007), other studies have suggested that patients' symptoms/functioning have limited or no impact on relatives' EE (Álvarez-Jiménez et al., 2010; Bachmann et al., 2002; Heikkilä et al., 2002; Raune et al., 2004). The differences in results among the above-mentioned studies (possibly due to the variability of the samples, measures or study design) leave unanswered the question as to what extent EE is a reaction to the severity of the relative's psychotic disorder (Heikkilä et al., 2002).

As suggested by Hooley (2007) and Miklowitz (2004), understanding EE as a unilateral reaction on the part of a relative to a patient's clinical characteristics is an incomplete approach to studying the ontogenesis of EE. In fact, developmental as well as bidirectional processes are involved in the genesis of high-EE attitudes (Hooley, 2007). There is therefore still an urgent need for prospective designs to determine whether EE levels increase over time as a consequence of continued exposure to ARMS patients' poor clinical and functional status. Moreover, given the above-mentioned results from Smith et al. (2018), it would be necessary to examine a broader range of patients' variables concerning EE, including their psychological and emotional state. However, caution must be taken to avoid an oversimplistic view of EE. It is therefore of the utmost importance to include relatives' psychological variables in the early manifestation of EE. Following this line, Hinojosa-Marqués et al. (2020) confirm that, as in Schizophrenia, relatives' emotional state and their cognitive representation of psychosis play an important role in the emergence, expression and maintenance of emotional attitudes towards the patient, over and above patients' poor clinical and functional status in the early stages of psychosis.

4.2 | Is EE related to ARMS relatives' psychological variables?

The scant evidence available suggests that a slightly different picture emerges when EE is considered as a more relational variable in which relatives' cognitive representations of the disorder or generalized stress reactions contribute to its emergence. Unfortunately, the sparse literature available on relatives' psychological factors associated with EE precludes drawing meaningful conclusions.

Some of the reviewed studies based on the attributional model in Schizophrenia (Barrowclough et al., 1994; Barrowclough & Hooley, 2003) highlight the significant role of relatives' illness attributions as mediators of the association between relatives' EE and early psychosis patients' clinical/functional features (Domínguez-Martínez et al., 2014) or as cross-sectional predictors of criticism (Domínguez-Martínez et al., 2017). To expand these preliminary cross-sectional reports by using an extended sample and a longitudinal design, Hinojosa-Marqués et al. (2020) tested a more comprehensive predictive model of EE in early psychosis (ARMS and FEP individuals) by including both patients' illness-related variables and relatives' psychological factors. Findings confirmed that relatives' psychological distress and negative illness attributions predicted EE dimensions (over and above patients' clinical and functional features) across time at both subclinical and onset stages of psychosis, lending further support to the attributional model (Barrowclough & Hooley, 2003). This result also indicates that the emergence of EE attitudes also stems from negative emotional responses and negative 'hot' cognitions in the ARMS and recent-onset stages of psychosis.

Hinojosa-Marqués, Domínguez-Martínez, Cristóbal-Narváez, et al. (2019) also found a similar pattern of associations between EE dimensions (assessed by both momentary and psychometric self-reports [FQ]) and relatives' illness attributions in daily life. Some of these associations are in line with previous early psychosis studies (e.g., Bolton et al., 2003; McNab et al., 2007; Vasconcelos e Sa et al., 2013) and support the attributional model of EE (Barrowclough & Hooley, 2003). Hinojosa-Marqués, Domínguez-Martínez, Cristóbal-Narváez, et al. (2019) argued that at the early stages of psychosis, many relatives may still exhibit low-defined illness attributions due to confusion, uncertainty and the lack of knowledge about the disorder. Thus, relatives may believe they can control the disorder by themselves and, at the same time, that patients can have significant control over their behaviour. As the disorder progresses, relatives may express more clearly defined illness attributions which, in turn, would result in more specific behavioural reactions. Presumably, the high emotional impact of early psychosis on family members may lead them to a low understanding of their feelings (i.e., diminished emotional clarity). Low levels of emotional clarity may influence the way they interpret patients' behaviours, thus causing relatives' cognitive ambivalence regarding the control of the disorder (Hinojosa-Marqués, Domínguez-Martínez, Cristóbal-Narváez, et al., 2019). According to Domínguez-Martínez et al. (2017), the attributional model of EE should be tailored to the developmental specificities of early psychosis thereby including the differences related to the various stages which families undergo across the psychosis continuum. This would require integrating the critical role of emotional factors influencing the psychological experience of relatives in at-risk stages.

To date, the attributional model of EE has received some empirical support at the recent-onset stage of psychosis. For example, families of FEP patients have shown that critical relatives tend to believe that symptoms are within the patients' control (McNab et al., 2007; Vasconcelos e Sa et al., 2013). Nevertheless, the attributional model of EE is a strictly cognitive-oriented paradigm that does not consider the pivotal role of emotional and affective factors influencing the psychological experience of relatives in at-risk stages. It should therefore be borne in mind that relatives of ARMS patients are exposed, probably for the first time, to early signs of psychosis. This potential threat may not only lead to creating cognitive appraisals about the causes of the disorder but also significant affective reactions to a recently processed situation. According to Hinojosa-Marqués et al. (2020), both cognitive and emotional factors, and even their interaction, are important predictors of the emergence and maintenance of EE in the ARMS period.

To date, relatives' emotional states (such as distressing affective states) and its relationship with EE have rarely been studied in the pre-psychotic phase, which precludes the possibility of examining whether EE arises from the perceived stress related to the caregiving role, as suggested by the 'carer appraisal model of EE' based on FEP samples (Kuipers et al., 2006; Raune et al., 2004). Some of the studies that have examined these relationships presented contradictory findings. Hamaie et al. (2016) only observed links between relatives' depression and criticism in FEP relatives but not in ARMS relatives. However, similar proportions of mild to moderate depressive symptoms were reported for both ARMS and FEP relatives. Authors suggested that the interaction between caregivers' emotional distress and criticism may emerge after the onset of psychosis but not at the at-risk stage. Conversely, Domínguez-Martínez et al. (2017) found differences between the ARMS and FEP relatives regarding the relationship between EE indices and anxiety but not with depression. Moreover, Hinojosa-Marqués et al. (2020) found strong associations between relatives' baseline levels of anxiety and depression and relatives' EE dimensions at both baseline and 6-month follow-up. These findings contradict the assumption by Hamaie et al. (2016) by showing that relatives' emotional states are strongly linked to EE attitudes at the at-risk and onset stages of psychosis, even at 6-month follow-up. Indeed, relatives' levels of anxiety and depression proved to be predictors of critical attitudes in both ARMS and FEP groups. The difference in results between these studies can be explained by cultural differences between the Japanese (Hamaie et al., 2016) and Spanish samples (Domínguez-Martínez et al., 2017; Hinojosa-Marqués et al., 2020). However, more studies are required on the association between EE and relatives' levels of anxiety and depression at the early stages of psychosis to be able to theorize about it.

Following this line, Hinojosa-Marqués, Domínguez-Martínez, Cristóbal-Narváez, et al. (2019) showed that relatives' EE attitudes are closely related to momentary negative affective experiences, increased appraisals of situational stress and feeling burdened by the patient and decreased positive affect in daily life at both the subclinical and onset stages of the illness. Overall, these results seem to be partially consistent with previous early psychosis findings indicating an association between EE and relatives' distress and/or burden (Domínguez-Martínez et al., 2017; Hinojosa-Marqués et al., 2020; Raune et al., 2004; Tomlinson et al., 2014) but do not replicate previous early psychosis research suggesting that EOI is more closely related to distress and burden than criticism (Álvarez-Jiménez et al., 2010; González-Blanch et al., 2010; Jansen, Gleeson, & Cotton, 2015).

Unfortunately, the study of relatives' distress and EE in the high-risk period is still very scarce compared with FEP literature. Converging evidence suggests that greater levels of psychological distress in FEP caregivers appear to be related to increased levels of EE (Álvarez-Jiménez et al., 2010; Jansen, Gleeson, & Cotton, 2015; Jansen, Haahr, et al., 2015; Koutra et al., 2014; Raune et al., 2004; Tomlinson et al., 2014). There is therefore an urgent need for more research aimed at examining caregivers' distress during the pre-psychotic stage to achieve a better understanding of the impact of prodromal phases of illness on relatives' mental well-being.

Only one study (Hinojosa-Marqués, Domínguez-Martínez, Sheinbaum, et al., 2019) pointed to the role of relatives' perceived loss as a major driver of EE attitudes in a mixed sample of ARMS and FEP relatives. Moreover, results from this study expanded previous findings from the FEP literature (Patterson, 2013; Patterson et al., 2000, 2005), showing that relatives' attachment anxiety may be a mediating mechanism whereby perceived loss influences the manifestation of both criticism and EOI attitudes in at-risk stages. This study constitutes a novel contribution, but further research focused on the early grief reactions of ARMS family members is required.

4.3 | Are EE or other family-related variables influencing clinical and/or functional ARMS characteristics?

Beyond studies focused on disentangling the patients and/or relatives' factors involved in the early development of EE, another parallel line of research has focused on testing the impact of EE on ARMS patients' concurrent symptoms and/or functioning (Carol & Mittal, 2015; Domínguez-Martínez et al., 2014; Tsai et al., 2015).

Although cross-sectional studies reported mixed results concerning this issue, it seems that relatives' criticism is more closely associated with patients' concurrent clinical and functional status than EOI. However, the scant research available makes it difficult to draw firm conclusions. Overall findings on the cross-sectional associations between EE and family functioning (as rated by patients or relatives) with patients' status are inconclusive in the ARMS literature. However, it should be noted that not all studies on the family environment in Schizophrenia and/or FEP samples find correlations between levels of EE and the severity of patients' concurrent clinical status (Álvarez-Jiménez et al., 2010; Bachmann et al., 2002; Cutting et al., 2006; Heikkilä et al., 2002; Vasconcelos e Sa et al., 2016). Accurate predictions of the influence of EE on ARMS patients' status are probably impossible to identify using cross-sectional designs.

Regarding the reviewed studies exploring the longitudinal impact of family environment variables on ARMS symptomatic relapse, it appears that most of the research reviewed highlighted the role of positive family aspects. Interestingly, studies revealed that (1) relatives' positive EE components (such as positive remarks and warmth) (O'Brien et al., 2006, 2008) and (2) observed positive interactional patterns within the family (O'Brien et al., 2009) and positive self-reported family functioning (Wang et al., 2015) were predictors of improvement in ARMS patients' symptoms and/or functioning over time. As suggested by a recent review (Butler et al., 2019), the protective effects of positive family attitudes on symptomatic relapse are most evident at the ARMS stage. Indeed, it was argued that the reduced chronicity of co-morbid difficulties (such as social anxiety, low mood and substance use; McGorry & Yung, 2003) could imply that there is greater potential for the protective effects of positive family environments in the early course of psychosis (Butler et al., 2019).

Another differential aspect of the ARMS stage is that relatives' EOI appeared to act more as a protective factor, given that it was found to be related to improvement in patients' clinical/functional status over time (O'Brien et al., 2006; Schlosser et al., 2010). In this line, it is important to assess the differential predictive power of the two EE components (criticism and EOI) since criticism has been found to make a greater contribution to symptomatic relapses in Schizophrenia (Cechnicki et al., 2013; Kavanagh, 1992; Marom et al., 2005) and even in the FEP period (Koutra et al., 2015). However, results regarding the predictive value of criticism in ARMS samples are somewhat mixed: while one study found that relatives' criticism predicted a worsening of attenuated psychotic symptoms over time (Schlosser et al., 2010), others did not report the same association (Haidl et al., 2018; O'Brien et al., 2006). Given the controversial relationship between criticism and patients' clinical attributes in the high-risk period, further study is required to determine whether cumulative exposure to critical attitudes can impact negatively on ARMS symptoms and/or functioning. Furthermore, Haidl et al. (2018), the first study exploring the predictive value of perceived EE in conversion into FEP, highlighted how other valuable constructs, such as patients' perceived irritability of a key relative, were predictive of ARMS conversion into FEP. These significant results should encourage future studies to adopt a more holistic approach to studying family environment variables contributing to relapse, rather than focusing solely on relatives' criticism and EOI attitudes, including positive affect that showed to be protective of relapse in FEP individuals (Lee et al., 2014).

From a diathesis-stress model perspective, stress within the family environment has been shown to contribute to the development and maintenance of symptoms in psychotic disorders (Hooley & Gotlib, 2000). Recently, research interest in family stress, coping strategies and the parent–child relationship at the ARMS stage has increased. It has been demonstrated that families reported moderate use of ‘approach’ coping and occasional use of ‘avoidant’ coping strategies (Gerson et al., 2011), whereas ARMS individuals were more likely to use disengagement strategies (such as avoidance, denial and wishful thinking) to cope with family stress that is associated with less social support and greater anxiety and depression, potentially compounding the social stressors and clinical burden these individuals are experiencing (Yee et al., 2020).

Moreover, from a parent–child relationship perspective, Peh et al. (2020) provide empirical evidence supporting the hypothesis that parent–child relationships are associated with the ARMS state. Specifically, they suggest that an affectionless-overprotective-controlling parenting style could not be beneficial for young people who are vulnerable to psychosis. Using similar measures, further cross-sectional studies showed that other family variables (such as nurturing behaviour, problem-solving and quality of parent–child relationships), as rated by patients' perceptions, were unrelated to psychotic symptom severity in both ARMS and FEP groups (Welsh & Tiffin, 2015), except for the association between the EE subscale and patients' manic symptomatology. However, a different picture emerges when considering ARMS patients' family perceptions as moderators. Bentley et al. (2016) found that in the ARMS group, the severity of social stress is dependent on the patient's perceived quality of the parent–child relationship. Likewise, Thompson et al. (2019) showed the moderating effect of ARMS patients' perceived family functioning between symptoms and social/role functioning.

4.4 | To what extent is the family emotional climate affected by the ARMS stage?

Studies exploring family factors as the main outcome are still very scarce in the ARMS literature but have been increasing in recent years. Only eight studies focused on examining family-related variables regardless of relatives' EE levels or patients' symptom severity (Baron et al., 2019; Gerson et al., 2011; Izon et al., 2019, 2020; Salinger et al., 2018; Santesteban-Echarri et al., 2018; Wong et al., 2008; Yee et al., 2020). Wong et al. (2008), the first study examining burden in the ARMS stage, showed that family burden was comparable between ARMS and FEP families. ARMS families were characterized by worry and active involvement in care, but their lives had not yet been disrupted by the disorder, which only had a limited impact on their daily routines. One possible explanation for these results could be that in the ARMS stage, characterized by less symptom severity and chronicity, relatives' active involvement or worry is not yet associated with significant emotional disturbances in their lives. Instead, findings from the FEP literature suggest that high levels of burden and psychological distress are already present after the recent onset of psychosis (Addington et al., 2003; Boydell et al., 2014). Continued exposure to psychopathology or poor functioning, as well as the caregivers' emotional shock caused by the early onset of the illness, therefore probably lead to the reinforcement of burden and/or distress responses.

Findings from Gerson et al. (2011) also supported the fact that ARMS relatives are characterized by active involvement in care, as they reported moderate use of ‘approach’ coping (such as planning, seeking social support, positive reinterpretation and acceptance) and only occasional use of ‘avoidant’ coping strategies (such as denial/disengagement and alcohol and drug use). Avoidant coping strategies have been more frequently described in relatives of patients with chronic Schizophrenia (Fortune et al., 2005; Huang et al., 2008), which may indicate that these types of coping strategies become more prevalent over time as a consequence of relatives' increased fatigue or burden.

To date, little is known about relatives' burden and/or coping reactions in the high-risk period. As suggested by the previous literature (Gerson et al., 2011; Wong et al., 2008), it would appear that relatives of ARMS patients are not yet as negatively affected by the disorder as they are in chronic stages, although they already show the emotional and psychological impact of their caring role (such as higher levels of worry, fear, distress, anger, anxiety and depressive symptoms) that are closely linked to the lack of understanding, uncertainty, feelings of lack of support from health services and unmet needs (Izon et al., 2019).

The only study focused on examining family functioning from the patients' perspective (Santesteban-Echarri et al., 2018) indicated that ARMS patients reported lower family satisfaction compared with healthy controls or non-help-seeking participants with risk factors for mental illness. However, ARMS patients reported healthy family functioning, in terms of cohesion and adaptability within the family. In contrast to these findings, another of the reviewed studies indicated that ARMS individuals scored less than the control group on perceived cohesion and adaptability within the family (Wang et al., 2015) and reported poorer perceived problem-solving and lower levels of nurturing behaviour in their families compared to controls (Welsh & Tiffin, 2015). Moreover, Salinger et al. (2018) indicated that communication patterns also appeared to be affected.

Although more research is required to determine the generalizability of the findings, dimensional approaches, which adopt a broader view of family functioning in terms of cohesion and adaptability and other global indicators of family welfare (such as problem-solving, communications patterns and nurturing behaviour), have been suggested as being more useful and comprehensive for capturing the potential disruptions of the family environment caused by the disorder (Koutra et al., 2015).

Finally, findings from Baron et al. (2019) expand the literature on the topic of caregivers of adolescents at risk for psychosis and provide a different perspective through the narrative of parents on their perceptions of stigma and how they cope with it. This is the first qualitative study to focus on this topic in the at-risk stage of psychosis. The authors pointed out that the ‘psychosis risk’ label carries more stigma than other more common mental health disorders, which may create a barrier to seeking appropriate treatment.

4.5 | Do family-based interventions improve outcomes in the ARMS stage?

In comparison with longer term illness (Barrowclough & Tarrier, 1990; Berglund et al., 2003; Chien & Norman, 2009) and recent-onset psychosis groups (for a review, see Askey et al., 2007; Bird et al., 2010; Claxton et al., 2017; Penn et al., 2005), there have been fewer family-based intervention studies at the ARMS stage and the evidence based on its efficacy during this pre-illness stage is still limited. Some early intervention programmes include multifamily psychoeducation, individual and/or group family interventions as part of the integrated treatment focused on preventing progression to FEP. However, some of them have reported the overall effects of the set of interventions on outcomes, which limits the interpretation of the specific effect of family intervention in preventing the transition to psychosis (e.g., Bechdolf et al., 2012; Nordentoft et al., 2006). The reviewed studies suggest that, as in Schizophrenia or FEP stages, family-based interventions are effective in reducing both relatives' high-EE attitudes (O'Brien et al., 2014) and patients' perceived levels of criticism (O'Brien et al., 2015) as well as in improving family communication and ARMS patients' clinical and functional outcomes over time (Landa et al., 2016; McFarlane et al., 2015; Miklowitz et al., 2014; O'Brien et al., 2015). The primary objective of the reviewed family-based interventions at the ARMS stage was to delay or prevent the transition to psychosis by reducing the negative aspects of EE. However, given the potentially protective effects of positive family attitudes (such as warmth, positive remarks and positive interactional patterns; O'Brien et al., 2006, 2008, 2009; Wang et al., 2015), it would also appear relevant to implement treatment strategies to foster the positive aspects of the family environment (Butler et al., 2019).

4.6 | Methodological limitations and future research

We identified several important methodological limitations in the reviewed literature. First, there was a high degree of heterogeneity among the studies included. There were differences regarding study designs, methods of assessment (family and patients' measures), different types of mixed samples, follow-up or other features that make the comparison between studies difficult due to the heterogeneity between them. Given the characteristics of the present theoretical and comprehensive review, we do not provide an analysis of the methodological quality of the studies, which would be important to address in further systematic reviews using quality appraisal tools.

Second, most of the studies were cross-sectional with small sample sizes. Future studies could shed light on variations in patient–family dynamics over time by using longitudinal designs and large samples of ARMS patients and their respective relatives. Third, most of the reviewed studies recruited combined samples of ARMS and FEP relatives. Although some of them reported separate results for the ARMS sample or offered comparative results between the samples, this precluded a more detailed examination of the different aspects of ARMS relatives concerning family environment variables. Another limitation is that EE and/or other family environment constructs were assessed based on either relatives' or patients' perspectives. Only two studies assessed both patients' and relatives' perceptions of EE (O'Brien et al., 2015; Schlosser et al., 2010). Given that EE reflects a transactional process between patients and relatives (Strachan et al., 1989), further studies should consider the importance of obtaining information from both relatives' and patients' perspectives. A final limitation is that only a few studies considered the role of relatives' psychological features (such as illness beliefs, distress and stress reactions) in explaining EE attitudes. Unfortunately, this precluded a detailed discussion of the usefulness of existing theoretical models for explaining EE in the at-risk stage of psychosis. There is therefore still an enormous need for research aimed at exploring the needs of caregivers regardless of their EE attitudes or the course of illness of their affected relative.

4.7 | Clinical implications

Although a great deal of research on family-based interventions with ARMS populations remains to be done, the evidence summarized in this review concerning the family treatment in ARMS stages suggests that early interventions benefit individuals at risk for psychosis as well as family dynamics. Family work with ARMS relatives is therefore essential for maximizing the adaptive functioning of the family and minimizing disruption to family life and the risk of ARMS individuals' deterioration given their high vulnerability to environmental stressors (Fusar-Poli et al., 2017).

Findings of this review showed that relatives' own needs and the emotional impact of caregiving are still a neglected intervention area in the ARMS stage. An increased focus on the impact of the at-risk psychosis stage on relatives' mental well-being is required to expand family support based on their psychological needs and to clarify the mechanisms leading to dysfunctional family dynamics during the critical ARMS period, considering other family factors beyond EE. Some of the identified barriers for family members include a lack of understanding of individuals' symptoms, limited confidence in their ability to support them, unproductive coping strategies impacting their health and/or well-being (Izon et al., 2019) and the associated stigma to 'psychosis risk' label, which may create a barrier to seeking appropriate treatment (Baron et al., 2019). Besides, it has been widely demonstrated that psychosis impacts family members' emotional state and family dynamic throughout the different stages of the disorder. Consequently, it is necessary to develop family resources and alternative intervention approaches, to educate, normalize and validate their own experiences. Izon et al. (2020) suggest some aspects to consider in clinical settings to facilitate relatives' role as caregivers, improve family communication and improve their quality of life: (a) exploring caregivers' coping strategies to manage their worries, anxiety and distress can help them prioritize their health and well-being; (b) psychoeducation and normalizing caregivers' experiences can help them feel validated and reduce their worry; (c) facilitating open communication between the patient and their family can positively impact on their relationship; (d) early engagement between services and ARMS individuals can help families feel less isolated; and (e) when feasible, services should invite/involve all the family members of ARMS individuals in sessions and explore family support strategies in managing their distress. Furthermore, Baron et al. (2019) also suggest that stigma should be included as an important topic to work on within psychoeducation and family therapeutic groups to help families cope with it and prevent a delay in adequate patient treatment. Besides, alternative interventions approach, such as online programmes and forums that families can access conveniently, asynchronously and more anonymously, may be useful in the beginning to prevent associative stigma in parents.

Furthermore, it has been highlighted the importance of offering caregivers about psychotic symptoms and tailors psychological interventions according to the stage of the disorder (considering that psychotic disorder is not necessarily the final outcome). This would help relatives to handle difficult thoughts, negative appraisals, and distress, to adequately cope and address the challenges of the disorder over time (Domínguez-Martínez et al., 2017), especially when they are dealing with the emergence of high-risk symptoms, where difficulties related to the caring role different from those with onset or well-established psychotic disorder.

5 | CONCLUSIONS

Although much research is needed to gain a better understanding of family dynamics in the pre-psychotic stage, it is important to appreciate existing research efforts to encourage the study of the family risk factors across the psychosis continuum. The recent interest in this area is beneficial in the shift to a paradigm in family environment research that will hopefully continue to attract the attention of researchers in the near future.

In conclusion, the preliminary evidence summarized in this review reflects the state of the art of the study of family environmental factors at the ARMS stage. Given the enormous heterogeneity of the studies reviewed, it is difficult to compare them all or to draw meaningful conclusions. The scant literature researching potential contributing factors to the early manifestation of EE has precluded the identification of the specific mechanisms underlying its ontogenesis. Overall, relatives' high-EE status did not appear to be reactive to ARMS patients' poor clinical status. However, relatives' cognitive representations of psychosis may have an important role to play, either as mediators of the relationship between EE and ARMS clinical/functional features or as predictors of EE attitudes. However, there is still a dearth of literature on relatives' psychological appraisals in terms of their relationship to EE. Likewise, relatives' distress and/or burden and its association with EE have been largely unexplored or even as the main variables of interest at the ARMS stage, meaning that relatives' psychological needs in the prodromal period have been overlooked. Furthermore, the available literature has failed to achieve consistent results when analysing the impact of EE on ARMS patients' concurrent symptoms and/or functioning. In the same vein, further dimensional family functioning indicators (such as cohesion, adaptability, nurturing behaviour, problem-solving, quality of parent–child relationships and communication patterns) do not appear to impact negatively on psychotic symptom severity. However, some initial evidence suggests the moderating role of these family functioning indicators in shaping the expression of ARMS patients' symptoms. Although evidence is still limited, several longitudinal studies highlight the significant role of positive family aspects as predictors of ARMS patients' clinical and/or functional improvement over time. Nevertheless, there is an urgent need for studies aimed at determining the predictive value of EE dimensions (such as criticism and EOI) on the clinical outcome of ARMS patients. There is insufficient consistency across longitudinal studies regarding the contribution of criticism to ARMS patients' clinical course. Moreover, EOI appears to act more as a protective factor and could have some positive effects on patients' outcomes at the ARMS stage. Finally, family-based interventions have demonstrated enormous potential for reducing high-EE attitudes and/or improving ARMS patients' outcomes. Determining relatives' needs and supporting family caregivers also warrant further attention.

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AUTHOR CONTRIBUTIONS

All the authors listed have made a substantial, direct and intellectual contribution to the work and approved it for publication.

CONFLICT OF INTERESTS

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

DATA AVAILABILITY STATEMENT

Data sharing does not apply to this article as no new data were created or analysed in this study.

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