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REVIEW ARTICLE



Investigating the relationship between specific negative symptoms and metacognitive functioning in psychosis: A systematic review

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Abstract

Background: Disrupted metacognition is implicated in development and maintenance of negative symptoms, but more fine-grained analyses would inform precise treatment targeting for individual negative symptoms.

Aims: This systematic review identifies and examines datasets that test whether specific metacognitive capacities distinctly influence negative symptoms.

Materials & Methods: PsycINFO, EMBASE, Medline and Cochrane Library databases plus hand searching of relevant articles, journals and grey literature identified quantitative research investigating negative symptoms and metacognition in adults aged 16+ with psychosis. Authors of included articles were contacted to identify unique datasets and missing information. Data were

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For Affiliation refer page on 19

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extracted for a risk of bias assessment using the Quality in Prognostic Studies tool.

Results: 85 published reports met criteria and are estimated to reflect 32 distinct datasets and 1623 unique participants. The data indicated uncertainty about the relationship between summed scores of negative symptoms and domains of metacognition, with significant findings indicating correlation coefficients from 0.88 to -0.23. Only eight studies investigated the relationship between metacognition and individual negative symptoms, with mixed findings. Studies were mostly moderate-to-low risk of bias.

Discussion: The relationship between negative symptoms and metacognition is rarely the focus of studies reviewed here, and negative symptom scores are often summed. This approach may obscure relationships between metacognitive domains and individual negative symptoms which may be important for understanding how negative symptoms are developed and maintained.

Conlclusion: Methodological challenges around overlapping participants, variation in aggregation of negative symptom items and types of analyses used, make a strong case for use of Individual Participant Data Meta-Analysis to further elucidate these relationships.

KEYWORDS

anhedonia, apathy, metacognition, psychosis, schizophrenia

Practitioner Points

- Higher levels of negative symptoms are associated with greater metacognitive deficits.
- Negative symptoms and metacognition are multifactorial constructs that require granular assessment and evaluation.
- Most studies do not include people with severe negative symptoms and so our current treatment formulation models may be under-developed for that subgroup.

INTRODUCTION

Negative symptoms can be profoundly disabling (Strassnig et al., 2018). They are often persistent and associated with lower rates of recovery in psychosis (Strauss et al., 2010). Factor analyses of negative symptom scales suggest two main sub-domains: Anhedonia, amotivation and asociality represent deficits of *experience*, and alogia and affective blunting represent *expressive* deficits (Messinger et al., 2011). These symptom clusters have been associated with different underlying factors (Marder & Galderisi, 2017), and arguably, exist transdiagnostically, or within several diagnostic categories.

Several treatments have focused on changing cognitive distortions (e.g. about the value of expending effort to pursue goals, and the likelihood of success) and impaired neurocognitive processing (such as working memory, effort allocation) as a mechanism for improvement in negative symptoms, with

3

moderate impact compared with treatment as usual (Lutgens et al., 2017). However, the results suggest that additional mechanisms are needed to explain negative symptom formation and maintenance. Also, given that some treatments may improve recovery from expressive deficits more than experiential deficits (Lutgens et al., 2019; Sevy et al., 2020), a greater understanding of individual negative symptoms and the mechanisms that lead to their amelioration is required to improve treatment precision, similar to other areas of psychosis (Lincoln & Peters, 2019).

Metacognition, broadly conceptualised as 'thinking about thinking', refers to a spectrum of activities. At the lower end of this spectrum, *discrete* metacognitive capacity refers to the ability to make accurate inferences about another's perspective, and generally involves lower level neurocognitive skills such as being able to recognise what information an individual is presented with, or interpreting others' facial expressions. These discrete skills overlap with elements of social cognition, which is a separate, and also multifaceted construct, incorporating additional knowledge and skills related to social rank, competition and competitive behaviours (Frith & Frith, 2012). At the higher end of the metacognitive spectrum, metacognitive capacity is less focused on the accuracy of social inferences, but rather encapsulates an individuals' capacity to create an *integrated* understanding of how individuals, including the self, make sense of the social world and act on this understanding. This more synthetic ability relies on higher level cognitive processing such as building a coherent narrative and developing meaning from experiential learning (Lysaker, Olesek, et al., 2011; Moritz & Lysaker, 2018). Metacognitive capacity has been related to processes involved in the development of positive symptoms (such as the jumping to conclusions bias [Buck et al., 2012]), and enhanced metacognitive capacity may be protective for people with psychosis, for example, in reference to social functioning (Fischer et al., 2020).

This review focuses on disrupted metacognition (at this higher level) as a mechanism involved in the development and maintenance of negative symptoms. For example, if individuals have difficulty labelling and giving meaning to the thoughts of themselves and others, they may find it more difficult to identify goals and initiate action towards them (van Donkersgoed et al., 2016) and may avoid social interactions where uncertainty over the intentions of others may cause discomfort (Salvatore et al., 2007). Individuals experiencing negative symptoms have been shown to have difficulties generating self- and other- reflective narratives, and have lower levels of intrinsic motivation (Tas et al., 2012) and these difficulties appear to persist over time (McLeod et al., 2014). This focus may enhance our understanding of mechanisms of negative symptoms and add to the existing literature focusing on discrete neurocognitive capacities and rule-governing beliefs (i.e. if there's a low chance of success there is no point in trying; Faith et al., 2020).

Like negative symptoms, metacognitive capacity, when considered in this way, can also be separated into sub-components: understanding of oneself (self-reflectivity, SR); understanding others (understanding other's minds, UOM); the capacity to understand the social world as separate from oneself (decentration, D); and the ability to utilise this information to respond to psychological problems (mastery, M [Lysaker et al., 2005]). A previous meta-analysis explored those outcomes which were most strongly associated with metacognition in individual studies, including negative symptoms. Results showed a relatively strong relationship between metacognitive capacities and a summed score of negative symptoms, but the analyses did not compare negative symptoms with all metacognitive subdomains or include all statistical results available (including weaker correlations expressed in some studies; Arnon-Ribenfeld et al., 2017).

At present, is it unclear whether metacognition and negative symptoms are consistently related across studies. Also, given that both metacognition and negative symptoms are multidimensional, it is important to determine whether subdomains of these constructs have different relationships. Few studies have investigated this, and those which cannot identify the unique influence of each metacognitive domain because participants are grouped based on metacognitive scores across multiple domains (Lysaker, Gumley, et al., 2011). Further analysis of this literature is also warranted to identify and understand the evidence comparing individual negative symptoms to metacognitive capacity.

This systematic review aimed to bring greater clarity to this literature by identifying all the unique data sets exploring the relationship between metacognition and negative symptoms. Then, we aimed to characterise the samples included and type of research conducted, focusing on how the relationship between metacognition and negative symptoms have been conceptualised and measured. The relevant findings across studies are summarised to demonstrate the degree to which metacognition and negative symptoms appear related, and to what extent this literature considers the subdomains of each construct separately. We also set out to critically assess the literature and risk of bias. Compiling this literature is a critical first step in identifying uncertainties in the current understanding of how negative symptoms and metacognition are related. These data provide essential information for future quantitative research approaches such as Individual Participant Data Meta-Analysis.

METHOD

Protocol and registration

Methods were developed according to a protocol, available on PROSPERO (registration number CRD42019130678). This protocol was intended for the dual purpose of describing both the systematic review and a subsequent IPDMA; however, the methods and criteria shown here pertain to the systematic review only.

Eligibility criteria

Participants and exposures

Studies including participants aged 16+ with any level of negative symptoms, as indicated by any clinical symptom measure, were eligible for inclusion. Comparison groups were not required.

Outcomes

The main outcome of interest were the characteristics of reported relationships between metacognition and negative symptoms reported across studies, including how frequently metacognition and negative symptoms were treated as a superordinate construct or examined by symptom subtypes. To examine specific sub-profile effects, studies needed to have measured metacognition and negative symptoms using reliable and validated measures. There was no cut-off score for studies to be included in the review. It was anticipated that missing data for these scales might not be reported, particularly for older papers; therefore, no specifications were made around measure completeness. These characteristics of the studies were instead summarised. Measures of social and occupational functioning were included as secondary outcomes to be presented descriptively. Inherent to exploration of these outcomes, a narrative description of the included studies and their characteristics was required, including considering the possible reporting of singular datasets across multiple study reports.

Study design

Only quantitative, English language publications before the last search date (30th April 2019) were included in the review. Case studies were excluded.

Search strategy

Broad search terms around experiences of psychosis, negative symptoms and metacognition were used to maximise the identification of relevant papers. Adhering to Cochrane Collaboration Guidelines (Higgins & Green, 2011), the search string for each information source was devised to ensure a comprehensive search using free-text keywords and database-specific index terms where possible. The following databases were searched:

- PsycINFO (1887- April 2019, updated weekly) via EBSCOhost.
- MEDLINE (Ovid MEDLINE® Epub Ahead of Print, In-Process & Other Non-Indexed Citations, Ovid MEDLINE® Daily, Ovid MEDLINE and Versions® – 1946 – April 2019, updated daily and weekly) via Ovid.
- EMBASE (1947 April 2019, updated daily) via Ovid.
- The Cochrane Database of Systematic Reviews (2007 April 2019) via Wiley Online Library.
- The Cochrane Central Register of Controlled Trials via Wiley Online Library (1966 April 2019) via Wiley Online Library.

Appendix S1: Item 1 includes the search string for PsycINFO. An update of the first search (May 2018) was conducted in April 2019. The reference lists of included reports and relevant reviews were hand-searched along with forward citation searching of included reports. Grey's literature including Google Scholar and Open Grey, and the Directory of Open Access Journals were searched using the free-text keywords. Authors from eligible studies were contacted to identify other data sources or reports.

Study selection

Search results were de-duplicated in Endnote (version x9) and screened for eligibility. Title and abstract screening were followed by full-text review of records that were identified as definitely or potentially meeting inclusion criteria. Eligibility assessment for 100% of the records was completed by the primary reviewer, and a second reviewer (W.A.) independently reviewed a randomly selected 10% of the records to check whether the screening process was replicable with substantial reliability (Cohen's Kappa = .74). Where there was insufficient information to judge whether a study met inclusion criteria and that work could not be accessed in full, reports citing the work in question were consulted alongside original authors where possible, and where reports were indicated to not meet inclusion criteria they were excluded.

Data extraction

Relevant meta-data and report details were extracted to an excel spreadsheet recording information, including the study aims, methods, participant characteristics and results. Authors of the original reports were contacted to confirm details where there was insufficient clarity in reporting, including whether multiple reports represented the same research data. One reviewer extracted 100% of the data from included reports and a second reviewer independently extracted 10% of report data blind to the primary reviewer's output (percentage agreement 85% following calibration).

Analysis

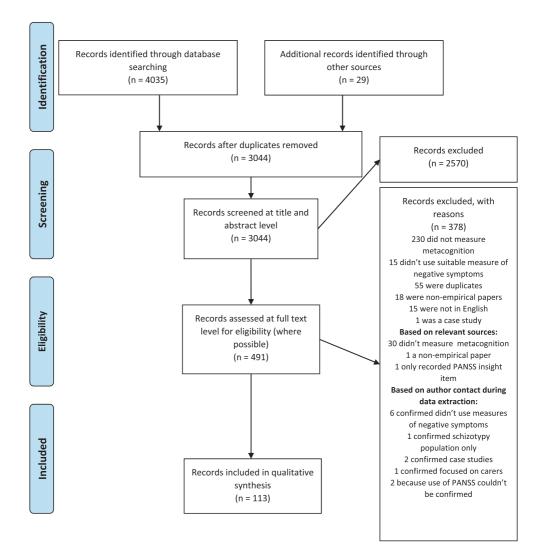
Included studies were described using narrative synthesis based on previous guidance (Popay et al., 2006) to explore reporting of relationships between metacognition and negative symptoms, and to ascertain the clarity of reporting and risk of bias in the data. The Quality in Prognosis Studies (QUIPs) tool (Hayden et al., 2013) was used by two reviewers (N.M. and S.A.), independently, to assess risk of bias in

reporting (Cohen's Kappa = .77 following calibration). It assesses all relevant domains to understanding the representativeness of the included population in each study, the measurement methods used, attrition, confounding, statistical analysis and reporting.

RESULTS

Search results

As illustrated in the PRISMA flow diagram in Figure 1, there were 4064 total returns from all included academic databases and hand searching. After de-duplicating, 3044 unique records were screened at title and abstract level, with the main reason for exclusion being the identification of further non-exact duplicate records. In all, 491 records were screened at full-text level. Of those included in data extraction, contact with study authors resulted in the exclusion of 12 reports because it was confirmed they did not meet inclusion criteria, or in 2 cases, ability to meet inclusion criteria



could not be confirmed. This resulted in a final 113 final reports – comprising theses, conference abstracts, letters to the editor and journal articles – being compared to determine whether they utilised the same datasets.

Identifying multiple study reports

Reports were identified as corresponding to the same dataset if they provided matching information across records, including measures and interventions used, and recruitment and data collection procedures. Where conference abstracts or theses and final published reports were identified to reflect the same research study they were combined for this review. Only one dataset was reported as a conference abstract only and not in any journal article, as results were still being analysed. Ten other conference abstracts were unattached to other study reports of the same dataset, as their aims or methods varied slightly but as they are not substantially different, they are not discussed further in this review.

Overall, 85 unique reports were identified and reviewed, which were confirmed to refer to 32 unique datasets measuring metacognition and negative symptoms (summarised in Appendix S1: Item 2). We estimate that there were 1623 unique participants across studies based upon the maximum sample size reported for each dataset across any report (see Appendix S1: Item 3). The second England sample were recruited and reported on concurrently with participants completing follow-up from England sample 1, following author confirmation, we describe their sample using the total minus those participants from England sample 1.

Study authors confirmed identification of multiple study reports, and this would have been impossible without author input. Although most of the 68 secondary data analyses papers reported that this was an analysis of existing data, only 29 reported where the data originated from. Furthermore, seven articles which analysed secondary data did not specify this or reporting was unclear (Abu-Akel et al., 2015; Bo et al., 2013, 2014; Lysaker et al., 2008, 2012, 2014; Popolo et al., 2017; Vohs et al., 2015). It was also unclear where one thesis and one published article were of the same data (Mitchell et al., 2012; Reilly, 2011). Additionally, most reports did not specify where results were a new analysis of existing data, or new data collected from individuals already contributing to previous studies. Multiple reports of the same data also varied in sample size due to the data completeness for variables of interest resulting in participants being dropped from some analyses and not others.

Characteristics of the reports

The included reports were largely developed using data from intervention studies (although these reports did not explore the interventions themselves) and predominantly came from USA sites. The comparisons are largely cross-sectional and investigate the ability of specific variables to predict metacognition. Related to our primary outcome, how the relationship between metacognition and negative symptoms was conceptualised across studies, some studies directly compared this relationship, and some studies analysed the relationship between metacognition and other variables with negative symptoms as a covariate. Thirty-four studies highlighted negative symptoms specifically as an outcome of interest in relationship with metacognition. Only nine studies highlighted an interest in individual negative symptoms, including anhedonia, or more specifically consummatory/anticipatory deficits (Buck et al., 2014; Luther, Firmin, Minor, et al., 2016); blunted affect and emotional withdrawal (Bo et al., 2015); intrinsic motivation (Luther et al., 2020; Luther, Firmin, Minor, et al., 2016; Tas et al., 2012; Vohs & Lysaker, 2014); and deficits in 'specific negative symptoms' (Austin et al., 2019; Nicolò et al., 2012).

Descriptive summaries of negative symptoms and metacognition across samples

Sixty-six reports provided negative symptom data (35 more papers than expected given the number of papers expressing hypotheses about negative symptoms). Two studies reported no negative symptom subscale data but did include negative symptoms in covariate analyses (Kukla et al., 2013; Rabin et al., 2014). Four studies reported Brief Psychiatric Rating Scale (BPRS), BPRS-Extended (BRPS-E) and Intrinsic Motivation measures. It is not possible to derive a single quantitative summary of the range of negative symptom scores present in the remaining 62 reports using the Positive and Negative Syndrome Scale (PANSS; Kay et al., 1987) because of the various factor structures used. Three studies also reported individual PANSS items and a further three reported negative symptom data per group of participants who were clustered together based on certain additional characteristics (e.g. intervention group at baseline, metacognition levels), making it impossible to extract negative symptom severity scores independent of these additional constructs. Table 1 gives the range of *average* negative symptom scores reported across studies.

The PANSS was the most common measure of negative symptoms. Only four reports used variants of the BPRS (Bargenquast & Schweitzer, 2014; Massé & Lecomte, 2015; Popolo et al., 2017; Schweitzer et al., 2017). The studies specifying an interest in individual negative symptoms either used specific measures (e.g. of intrinsic motivation or anhedonia) or reported on individual PANSS (Kay et al., 1987) items (e.g. blunted affect, emotional withdrawal). Of the 80 reports using the PANSS, 63 used an alternative to the original PANSS factor structure to analyse symptom data, for example (Bell et al., 1994; van der Gaag et al., 2006), Thirteen reports used the original negative symptoms subscale. The remaining studies assessed individual items, for example (Buck et al., 2012; Minor et al., 2015), or used overall PANSS scores as a cut-off to determine if individuals had eligible levels of symptom severity (Davis et al., 2011; van Kleef et al., 2015). The different combinations of items contributing to negative symptoms analyses (summarised in Figure 2) create different possible total scores making it problematic for aggregating analyses using these measures. Additionally, 17 reports failed to specify which factor structure they used to measure negative symptoms.

It is also crucial to understand how metacognition was conceptualised and measured across studies. Only eight reports used a measure other than the Metacognition Assessment Scale (MAS-A) developed by Lysaker et al. (2005); including 4 reports using the Metacognition Assessment Interview (MAI; Davies et al., 2017; Wright et al., 2019a, 2019b, 2020), and 4 using the Revised version of the MAS (MAS-R; MacBeth et al., 2014, 2016; Mitchell et al., 2012; Reilly, 2011).

The range of MAS-A scores reported across subdomains and the total score are highlighted in Table 2, based on 71 studies which gave descriptive data for at least some elements of the MAS-A. Seven studies with interest in particular subscales of the MAS-A did not descriptively summarise these, and instead some grouped participants by high, intermediate and low scores in these domains (Bonfils et al., 2018; Davis et al., 2011). Seven studies excluded the decentration subscale from their description of the MAS-A; with only three studies giving a rationale for this. This complicates interpretations of

TABLE 1	A quantitative summary of PA	ANSS Negative Symptom score	s reported across studies.

PANSS factor structure	Bell et al. (<mark>1994</mark> ; PTSR 8-56)	Original (PTSR 7-49)	van der Gaag et al. (2006; PTSR 2-62)
Total number of reports using PANSS factor structure specified	48	13	12
Number of studies reporting data	39	11	12
Reported symptom range of mean scores for negative subscale	13.9–24.5	10.97–22.2	12.54–19.41

Abbreviation: PTSR, Possible Total Score Range

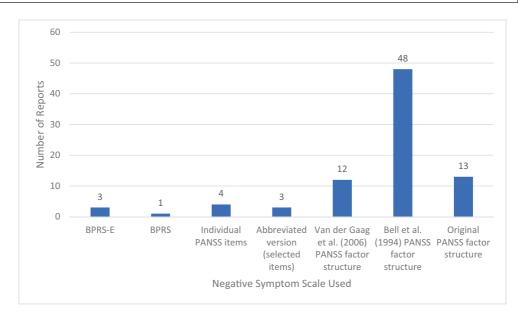


FIGURE 2 Negative symptom structures measured across reports: *Note*: The use of PANSS in one dataset (Vernal et al., 2018) does not contribute to this figure as results were not analysed at the time of writing.

TABLE 2	MAS-A characterist	ics across reports	and ranges reported.
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	Self- reflectivity (range 0–9)	Understanding others' minds (range 0–9)	Decentration (range 0–3)	Mastery (range 0–9)	Total MAS-A score (range 0–28)
Number of studies with metacognition- specific hypotheses	27	20	16	25	25
Number of studies reporting MAS-A item scores	47	41	34	46	48
Reported range (means across studies)	3.375-5.51	2.27-4.43	0.36-1.69	1.77-4.75	8.48-14.6

total metacognition levels because it is then unclear whether these scores also account for decentration. Similar to negative symptoms, some studies also summarised MAS-A scores by subgroup leading to more extreme scores (i.e. high achievers on a learning task in one study (Tas et al., 2012) scored higher total metacognition than is reported in any other study: 16.55).

Analyses reported across studies

Sixty-two studies reported analyses of negative symptoms and metacognition. Tables 3 and 4 summarises the study characteristics and findings of research making a direct comparison between negative symptoms and metacognition. Across analyses, only 35 analyses reported a direct correlation coefficient between negative symptoms and metacognition. Remaining analyses including secondary outcomes and their relationship to metacognition and negative symptoms are summarised in Appendix S1: Item 5.

Table 5 describes the range of correlations (including Pearsons and Spearmans coefficients) between subtypes of metacognition and total negative symptom measures and their statistical significance. However, given the different factor structures used there are different items contributing to analyses across studies. As only one study (Reilly, 2011) used Kendall's Tau as a measure of association, we refer interested readers to this paper for comparison. A clearer indication of the studies contributing to the

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Major study, country	Sub/secondary analyses	Does study hypothesis specify interest in symptoms?	Metacognition measures	Negative symptom measure(s)	Sample size (of schizophrenia sample)	Type of sample	Findings
Massé and Lecomte (2015), Canada		No	MAS-A (excl. Decentration)	BPRS-E	50	First Episode Psychosis (FEP)	No significant difference across participants grouped by metacognitive profile on BRPS negative scores
Lysaker et al. (2018), Chile		Negative symptoms	MAS-A	PANSS-Bell et al. (1994) factor structure (PANSS-B)	26	People with schizophrenia	Significant correlation between MAS-A Other, Mastery and Total score and PANSS Negative Symptoms (NS) total score, no other significant relationships
Abu-Akel and Bo (2013), Denmark		No	MAS-A (Total (T), SR and UOM only, alternative scoring)	PANSS – abbreviated	42	Patients with schizophrenia	No significant correlations between PANSS NS abbreviated version total score and MAS-A
	Abu-Akel et al. (2015)	°Z	MAS-A (excl. decentration, alternative scoring)	PANSS – abbreviated	- 20	Forensic patients with schizophrenia	No significant correlations between PANSS NS abbreviated version total score and MAS-A. Significant correlation between MAS-A subscales and total scores
	Bo et al. (2015)	Individual negative symptoms	MAS-A (alternative scoring)	PANSS (blunted affect, emotional withdrawal)	62	Primarily criminal and violent patients with schizophrenia	Significant correlation between decentration and emotional withdrawal – no other significant correlations between PANSS N1 and N4 items and MAS-A subscales or total score
Traucisen et al. (2016), Denmark		Negative symptoms	MAS-A	PANSS van der Gaag et al. (2006) factor structure (PANSS- VDG)	97	ΗEP	PANSS NS, subscale total score significantly correlated with MAS-A total score and all subscales
	Trauelsen et al. (2019)	Negative symptoms	A-SAM	PANSS-VDG	92	Non-affective FEP	MAS-A Total and subscale scores all significantly correlated with PANSS NS subscale total score and with each other
Bröcker et al. (2017), Germany		No	MAS-A	PANSS-VDG	22	Individuals with Schizophrenia Spectrum Disorders	No significant correlations between MAS-A subscales and total score and PANSS NS subscale

TABLE 3 Cross-sectional associations between negative symptoms and metacognition.

Rabin et al. (2014),	Sub/secondary analyses	hypothesis specify interest in symptoms?	Metacognition measures	Negative symptom measure(s)	Sample size (of schizophrenia sample)	Type of sample	Findings
Israel		Negative symptoms	MAS-A (SR and UOM)	D-SSNA9	39	Persons with schizophrenia	SR and UOM subscales of the MAS-A are significantly correlated with the PANSS NS total score and with each other
Nicolò et al. (2012), Italy		Individual negative symptoms	MAS-A (excl. decentration)	PANSS (blunted affect, emotional withdrawal, and disturbance of volition)	45	Outpatients with schizophrenia	Controlling for age and education, blunted affect significantly correlated with SR and UOM subscales, emotional withdrawal significantly correlated with SR subscale, and disturbance of volition significantly correlated with SR subscale and total score. No other significant correlations found
Po	Popolo et al. (2017)	Negative symptoms	MAS-A	BPRS	26	Patients with schizophrenia	BRPS Withdrawal/retardation subscale score correlated significantly with MAS-A SR and UOM subscales and total score. No significant correlations with D and M subscales
van Kleef et al. (2015), Netherlands		Symptoms	MAS-A	PANSS Original factor structure (Kay et al., 1987; PANSS-O)	52	People with schizophrenia	MAS-A UOM subscale and total score correlated significantly with PANSS NS subscale total score, no other subscales significantly correlated
Mitchell et al. (2012), Scotland		No	MAS-R	O-SSN Vd	29	People with schizophrenia with/without a history of interpersonal violence	PANSS NS subscale total score was significantly correlated with the MAS-R UOM and M subscales
MacBeth et al. (2014), Scotland		Negative symptoms	MAS-R	PANSS-VDG	34	FEP	Significant relationship between MAS-R Understanding Other's Minds subscale and PANSS NS total score
Tas et al. (2014), Turkey		No	MAS-A	PANSS-O	30	People with schizophrenia	No significant correlations between PANSS NS and MAS-A subscales (Continues)

TABLE 3 (Continued)

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Major study,	Sub/secondary	Does study hypothesis specify interest in	Metacognition	Negative symptom	Sample size (of schizophrenia	alma of camp	Hinditores
Lysaker et al. (2005), USA	、	Negative symptoms	MAS-A (excl. decentration)	PANSS (blunted affect, emotional withdrawal, disturbance of volition)	19	People with schizophrenia	Significant relationship between solf-reflectivity, understanding the mind of the other and mastery with emotional withdrawal, but not with blunted affect or disturbance of volition
Buck et al. (2012), USA		Symptoms	MAS-A (Mastery)	PANSS-B	40	People with schizophrenia	Found individuals grouped by their level of mastery had significantly different PANSS NS total scores
	Fridberg et al. (2010)	Symptoms	A-SAM	PANSS-B	62	People with schizophrenia	Significant correlation between the self-reflectivity subscale of the MAS-A and the PANSS NS subscale total score
	Lysaker et al. (2007)	Symptoms	MAS-A (SR and D)	PANSS-VDG	69	People with schizophrenia	Significant relationship between MAS-A self- reflectivity and PANSS NS total score, but not decentration
	Lysaker, Dimaggio, et al. (2010)	Symptoms	MAS-A (excl. Decentration)	PANSS-B	102	People with schizophrenia	No significant relationship between MAS-A Total score and SR, UOM & M subscales with PANSS NS total score
	Lysaker, Shea, et al. (2010)	No	MAS-A (M)	PANSS-B	102	Persons with schizophrenia	No significant relationship between MAS-A Mastery and PANSS NS total score
	Lysaker et al. (2012)	Negative symptoms	A-SAM	PANSS-B	95	People with schizophrenia	Factor analytical structure of metacognitive constructs (Beck Cognitive Insight Scale and MAS-A total) not significantly related to negative symptoms scores
Minor and Lysaker (2014), USA		Symptoms	A-SAM	PANSS-B	68	People with schizophrenia	Significant negative correlation between MAS-A Total score and SR and M subscales, and PANSS NS Total score, but not UOM and D subscales
Vohs et al. (2014), USA		Symptoms	MAS-A	PANSS-B	98	FEP/Multiple Episode Psychosis sample	PANSS NS subscale total score significantly correlated with MAS-A total score and all subscales except D
	Vohs et al. (2015)	Symptoms	A-SAM	PANSS-B	40	FEP	MAS-A UOM and M subscales and total score significantly correlated with PANSS NS total score. SR subscale appears not significantly correlated as not reported

TABLE 3 (Continued)

Major study, country	Sub/secondary analyses	Does study hypothesis specify interest in symptoms?	Metacognition measures	Negative symptom measure(s)	Sample size (of schizophrenia sample)	Type of sample	Findings
Bonfils et al. (2018), USA		No	A-SAM	PANSS-B	56	People with schizophrenia	Significant correlation between MAS-A Self-reflectivity subscale and PANSS NS subscale total score
Bonfils (2017), USA		No	MAS-A (total)	PANSS-B	58	People with schizophrenia	No significant correlation between MAS-A total score and PANSS NS subscale
	Bonfils et al. (2019)	°Z	MAS-A	PANSS-B	58	People with schizophrenia or schizoaffective disorder	PANSS NS subscale total score significantly correlated with MAS-A total score and SR subscale, no other significant correlations identified
Luther et al. (2020), USA		Individual negative symptoms	MAS-A	Quality of Life Scale (QLS) motivation index MAP-SR CAINS	56	People with schizophrenia spectrum disorders	No measure of motivation was significantly associated with MAS-A total score
MacBeth et al. (2016)*, Scotland		Negative symptoms	MAS-R	PANSS-VDG	34	FEP	No significant correlations between MAS-A items and negative symptoms
Snethen et al. (2014)*, USA		Negative symptoms	MAS-A	PANSS-B	44	People with schizophrenia	No significant correlations between MAS-A subscales and PANSS NS subscale total score
Luther, Firmin, Vohs, et al. (2016)*, USA		Individual negative symptoms	A-SAM	PANSS-B QLS motivation index	175	People with schizophrenia	Higher metacognition significantly predicted higher levels of intrinsic motivation
Gagen et al. (2019)*, USA		Negative symptoms	A-SAM	PANSS-B	334	People with schizophrenia	PANSS NS total subscale score significantly negatively correlated with MAS-A total score
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(Continued)

TABLE 3

Major study, country	Does study hypothesis specify interest in symptoms?	Metacognition measures	Negative symptom measure(s)	Sample size (of schizophrenia sample)	Type of sample	Findings
Austin et al. (2019), Denmark	Individual negative symptoms	A-2AM	PANSS- VDG (and Harvey et al., 2017 two factor model; and individual items blunted affect, emotional withdrawal, poor rapport, passivity and alogia)	50	FEP	Expressive negative symptom domain (composed of PANSS NS items N1, N3 and N6) significantly correlated with S and M MAS-A subscales, no other significant correlations, and experiential domain (N1 and N4) was not significantly correlated with any MAS-A subscales. PANSS N1, N2, N3, N4 and N6, expressive and experiential components, and total score, all significantly correlated with MAS-A Total score at baseline, N1, N3 and N6, total score, and expressive component all significantly correlated at follow-up. Significant relationship retained for N1 and N3 when controlling for baseline negative symptoms
Wright et al. (2019b), England	Negative symptoms	MAI (total)	PANSS-O	FEP	26	MAI baseline and follow-up total composite scores significantly correlated with PANSS NS subscale baseline and follow-up scores
McLeod et al. (2014), Scotland	Negative symptoms	MAS-A	DANSS-O	45	People with early psychosis	MAS-A SR, UOM, D and M subscales significantly negatively correlated with negative symptoms at 6 months, no significant correlations at 12 months (1 trend, decentration). Addition of MAS-A scores to predictive models of negative symptoms explained 62% of variance at 6 months and same model explained 38% of variance at 12 months
Breustedt (2017), Scotland	No	MAS-A	D-SSNA9	12	Individuals experiencing acute psychosis	Significant correlations between MAS-A decentration and total score with PANSS NS total score
Tas et al. (2012), Turkey	Individual negative symptoms	MAS-A	PANSS-O IMI-SC	30	Patients with symptomatically remitted schizophrenia	All subdomains of metacognition significantly correlated with all subdomains of intrinsic motivation, except interest and enjoyment
Davis et al. (2011), USA	No	MAS-A (M)	PANSS-B	63	People with schizophrenia	Found individuals grouped by their level of mastery had significantly different PANSS NS total scores
Hamm et al. (2012), USA	Negative symptoms	MAS-A (Total)	PANSS-B	49	People with schizophrenia	Significant correlation between MAS-A Total scores and PANSS NS subscale total scores at baseline and 6 months

TABLE 4 Longitudinal associations between negative symptoms and metacognition.

Findings	Reduced baseline motivations significantly related to increased baseline negative symptoms and lower metacognition. Decreased motivation at 6-month follow-up also associated with decreased baseline motivation, anticipatory pleasure and metacognition amongst other factors. Metacognition was a significant contributor to a model predicting prospective motivation	When participants grouped by metacognition level (low, moderate, high), the low metacognition group had significantly higher overall negative symptoms. Participants with low MAS-A scores at baseline had a trajectory of worsening negative symptoms over time. Results were consistent across all treatment groups	No significant differences on PANSS NS subscale total scores for participants grouped by levels of low, intermediate and high mastery. MAS-A M subscale significantly correlated with intrinsic motivation over time, and there were significant differences across mastery groups on intrinsic motivation at all timepoints
Type of sample	Individuals with schizophrenia spectrum disorder	People with schizophrenia	Individuals with prolonged schizophrenia
Sample size (of schizophrenia sample)	21	53	75
Negative symptom measure(s)	PANSS-B QLS motivation index TEPS	PANSS-B	PANSS-B QLS motivation index
Metacognition measures	A-SAM	MAS-A (Total)	A-SAM
Does study hypothesis specify interest in symptoms?	Individual negative symptoms	Negative symptoms	Individual negative symptoms
Major study, country	Luther, Firmin, Minor, et al. (2016), USA	Lysaker et al. (2015), USA	Vohs and Lysaker (2014), USA

TABLE 4 (Continued)

	Self-reflectivity, N (% significant)	Understanding others' minds, N (% significant)	Decentration, N (% significant)	Mastery, N (% significant)	Total metacognition, <i>N</i> (% significant)
Total Negative symptom Comparisons	25 (44%)	24 (50%)	17 (41.18%)	23 (39.13%)	24 (66.67%)
Range of coefficients (for st	atistically significant re	lationships only)			
Min.	-0.23	-0.29	-0.422	-0.286	-0.28
Max.	-0.54	-0.60	0.88	-0.70	-0.636

TABLE 5 Summary of correlation comparisons between negative symptoms and domains of metacognition.

ranges listed in Table 5, plus further information about the reported relationships between metacognitive domains and individual negative symptom items, and other measures of negative symptoms, are listed in Appendix S1: item 4.

In summary, significant findings of a relationship between subscales of metacognition and summed measures of negative symptoms were not consistently observed, and when they were, the strength of association ranged from small to large. Total metacognition was significantly associated with negative symptoms more than any singular metacognitive domain, and the correlation coefficients were not consistent with summation of coefficients observed for individual domains. No one metacognitive domain emerged as consistently and significantly related to total negative symptoms. Regression and covariate analyses were similarly mixed, but mirrored similar results such as metacognition predicting levels of intrinsic motivation (Luther, Firmin, Vohs, et al., 2016) and total negative symptoms (Hamm et al., 2012), and the relationship between negative symptoms and metacognition appeared to be independent of demographics (Nicolò et al., 2012). Similarly, relationships between negative symptoms and MAS-R subscales also showed a range of significant and non-significant relationships (MacBeth et al., 2016; Mitchell et al., 2012). The only study with relevant analyses using the MAI also showed a significant relationship between PANSS total negative symptoms and metacognition over time (Wright et al., 2019b).

Only five reports give comparisons of domains of metacognition and specific negative symptoms. One study suggested individual metacognitive domains were related to different elements of intrinsic motivation, but like other studies, no single metacognitive domain emerges as more consistently and strongly related to negative symptom items than any other (Tas et al., 2012). As detailed further in the Supporting Information, individual PANSS negative symptom items were often not significantly correlated with MAS-A subscale scores. Only one study (Austin et al., 2019) summarised relationships between individual items clustered by expressive and experiential deficits (as suggested by Harvey et al. [2017]) as well as individual negative symptom relationships. Expressive negative symptoms when treated individually and when grouped together appeared more consistently associated with MAS-A scores at baseline and follow-up than experiential negative symptoms. There is limited evidence to draw a clear conclusion about the relationship between metacognitive domains and specific negative symptoms.

Several studies grouped their participants by a range of variables including metacognitive levels and composite scores of various symptom domains. Studies clustering participants into low, medium and high levels of mastery found a range of significant and non-significant relationships between metacognition and negative symptoms (Davis et al., 2011; Vohs & Lysaker, 2014). Similarly, participants grouped into low, high and medium levels of total metacognition also showed significant differences on negative symptom scores (Lysaker et al., 2015). Of the two studies grouping participants by metacognitive profiles (high, mixed or low metacognitive abilities, and composite self-reflectivity/decentration scores respectively), both significant and non-significant associations were found (Lysaker et al., 2007; Massé & Lecomte, 2015). Given that clustering likely differs based on sample size, and the sample present, it is unclear whether clustering approaches have contributing to the inconsistency of results and so it is difficult to draw comparisons across studies investigating the relationship between metacognition and negative symptoms.

Risk of bias assessment

The risk of bias summary is included as a separate item in the Supplementary documents (Appendix S1: Item 6). The conference abstract (Vernal et al., 2018) of the dataset still being analysed and otherwise not reported was not included in the risk of bias assessment as it was not possible to assess the methodology adequately. Studies were mostly rated as moderate or low risk of bias, with the main sources of bias being unclear reporting around whether the samples included in secondary data analyses were different to the original sample, and insufficient information about the use of measures and analyses procedures. There were also few reports which explicitly specified whether data were missing, and it was hard to identify the impact of refusal to participate on sample size. There seemed to be few identifiable errors in reporting (e.g. scores reported which were greater than the maximum possible score for a specific measure). No major inconsistencies were found between reports of the same dataset, and there was no individual study at such a high risk of bias as to warrant exclusion from this review.

DISCUSSION

Negative symptoms are often unresponsive to treatment, and their relationship to psychological treatment targets such as metacognition is poorly understood, despite studies showing metacognition is a protective factor for psychosis symptoms more generally. This systematic review aimed to summarise the relationship between metacognition and negative symptoms across studies, with reference to the conceptualisation and measurement of each construct, and types of analyses employed. Our review demonstrates that a substantial number of studies of metacognition in psychosis have measured negative symptoms, often as a covariate, but not usually as the main focus of research. The range of analyses were vast, with the relationship between the total constructs as opposed to sub-domains being explored, and the majority of findings were mixed in directionality, effect size, and statistical significance. In the case of the most comparable analyses across studies (direct correlations comparing total negative symptoms and metacognitive domains) only half of the included studies report statistically significant results. The evidence presented does not suggest a clear relationship between metacognition and negative symptoms, although there is positive evidence of a strong relationship between the two constructs.

This review's aims were similar to that of Arnon-Ribenfeld et al. (2017), who summarised the direction and magnitude of relationship between metacognition and measures of symptoms and psychosocial functioning in people with psychosis, although with a more specific focus on negative symptoms. Their findings, that symptoms were negatively associated with metacognitive abilities is clearer than the results related to negative symptoms in the current review; however, the amalgamation of distinct psychosis symptoms, and use of different effect size for analyses (Cohen's *d*), might be responsible for the differences across studies. Additionally, both studies show that findings for studies investigating the relationship between metacognition and negative symptoms are highly heterogeneous, in the case of the earlier review, this heterogeneity is higher than that in other psychosis symptom subtypes. Factors, such as participant's levels of insight into their own pain or degree to which internal experiences affect their behaviour are cited as possible confounding factors that might have influenced these results (Arnon-Ribenfeld et al., 2017).

There are several methodological factors which might caveat the current review findings. There are only 32 unique datasets attributed to the 85 included reports, and only 12 papers written for publication which do not appear to include data which has been published elsewhere (including in studies out with the scope of this review). Reassuringly, the largest proportion of reports (by the research team who originally developed the MAS-A) showed mixed results and reflected similar patterns to other research groups' findings not directly involved in the development of this measure, lessening the risk of publication bias. Nonetheless, the issues of multiple comparisons across reports of the same datasets may mean that the estimates reported across studies, when not considered in aggregate, are at risk of higher imprecision than currently estimated.

Studies were also generally a low-to-moderate risk of bias across all QUIPs domains. One particular area of risk of bias (discussed in the Supplement), lack of inter-rater reliability computations in the measurement of negative symptoms and metacognition within studies, indicates that measuring consistency in rating across researchers may be a key area for improvement in the field. Indeed, this alongside heterogeneity in the negative symptom measurements selected for analyses, and a lack of systematic comparison between negative symptoms and all metacognitive domains across studies, may have contributed to the lack of a clear relationship being observed. Furthermore, we notice that the range of mean scores reported for both negative symptoms and metacognition are skewed towards low to median possible scores on each of these scales. This could perhaps indicate sampling bias where individuals with more severe negative symptoms or metacognitive difficulties are not adequately represented within studies.

Finally, there are significant limitations to the generalisability of these findings due to methodological heterogeneity across studies. Exact determination of the unique number of participants was not possible due to sample overlap across publications, and traditional meta-analyses would have resulted in over 50% of data loss given the range of statistical relationships reported and heterogeneity in items contributing to analyses. This influenced the decision not to conduct an aggregate data meta-analysis and means no summary estimate of effect is available.

Theoretical and clinical implications

Relationships with negative symptoms were around or below 50% significant across studies for all metacognition subdomains. Interestingly, of the few studies investigating subtypes of negative symptoms, differential relationships with metacognitive domains were identified. For example, one study (Tas et al., 2012) found that Understanding Others' Minds and Decentration were related to perceived effort, an experiential negative symptom construct, but not Self-reflectivity or Mastery. In comparison, both these latter constructs were significantly related to expressive negative symptoms in another study (Austin et al., 2019). This calls into question whether the tendency to treat negative symptoms as monolithic might obscure any relationship between these symptoms and metacognitive subtypes. However, while we see some signals of potential interesting relationship between these constructs, we are also not able to rule out the possibility that no true relationship between metacognition and negative symptoms exists, as quantified using these measures.

Strengths and limitations

The reduction of reports from 4061 to the final 85 records reviewed perhaps indicates that the search was initially relatively broad; however, this does indicate that the results were likely to be inclusive of all studies. The choice to include conference abstracts identified an additional dataset, but most of these reports did not provide any usable data so the benefit of including them is questionable. Author contact also allowed the reviewers to identify publication of overlapping datasets which would not have been possible through examination of the published records alone.

While this search was relatively comprehensive, our focus on 'negative symptoms' as search terms may have excluded papers which examined phenomenologically similar experiences in wider diagnostic groups, such as people experiencing depressive symptoms (Moritz et al., 2019). Given our focus on negative symptoms as they present within psychotic disorders, we argue that our narrower focus is justified. Also, the heterogeneity of measurement tools, analyses, and risk of bias around reporting of these studies may point to the need for cautious interpretation of any of the findings. While the overall constructs assessed possess similarities, heterogeneity in outcomes consistently reported in studies examining the relationship between negative symptoms and metacognition restricted focus on any singular outcome.

CONCLUSION

This review has identified a previously under-acknowledged gap in understanding of how the relationship between metacognition and negative symptoms is conceptualised and measured, and comprehensively estimates the evidence base available. The high heterogeneity in the literature was attributed to methodological differences in the use of negative symptom and metacognition measures and lack of clarity in reporting around overlapping participants in datasets. This makes it unclear as to whether the lack of consistency in relationships between negative symptoms and metacognition are due to measurement error or sampling bias (either in the variables selected or range of participants included). The findings also raise questions around whether homogenising negative symptoms and metacognition may obscure potential significant relationships between individual negative symptoms and metacognitive subtypes. This makes a strong case for future research which investigates negative symptoms and metacognition at the item level, and ultimately, research designed specifically to investigate the relationship between metacognition and negative symptoms.

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CONFLICT OF INTEREST STATEMENT

The authors attributed to this manuscript declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that supports the findings of this paper are provided by the co-authors listed here. Most data pertains to documents reviewed in the public domain. Restrictions apply to the availability and retention of raw data processed for this paper, which were used under license for these purposes. Data are available from the authors upon reasonable request at the discretion of co-authors and subject to retention agreements.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

Appendix S1.

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