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The association between adverse childhood experiences, self-silencing behaviours and symptoms in women with fibromyalgia

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ABSTRACT

Poor adult health has consistently been associated with Adverse Childhood Experiences (ACEs). The process is not fully understood but is likely to involve a complex interaction of biological and psychosocial factors. Early life stress can affect the developing brain resulting in long-term hyper-responses to stress and raised inflammatory biomarkers. Women with fibromyalgia syndrome (FMS) frequently report ACEs and also self-sacrificial behaviours whereby they repress their own needs and emotions to preserve personal relationships. This behavioural profile (termed self-silencing) may develop following ACEs but has not previously been considered in the context of FMS. This study examined whether self-silencing mediates the link between ACEs and FMS symptoms. Women with (N = 539) and without (N = 184) an FMS diagnosis completed measures of Silencing-the-Self, health symptoms and ACEs. Number of ACEs and symptom levels were significantly associated in both groups. One aspect of self-silencing, care and self-sacrifice (putting others needs before your own) mediated the association between ACEs and symptoms in the FMS group only. Externalised self-perception (judging oneself according to perceived external standards) was negatively associated with symptoms but presented no mediating effects. Results present preliminary new information to explain the association between ACEs and FMS. Recognition of the factors which underlie symptomology is important in understanding the condition and supporting patients.

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Silencing the Self; fibromyalgia; Adverse childhood experiences (ACEs); women's health, stress

Introduction

Fibromyalgia syndrome (FMS) is a condition typified by chronic, debilitating musculoskeletal pain together with a range of somatic, cognitive and psychological symptoms idiosyncratic to the individual (Galves-Sanchez et al., 2019; Sluka & Clauw, 2016). FMS can result in considerable physical and psychosocial impairments that impact significantly on wellbeing, and on health and social care services (Bateman et al., 2016 Creed et al, 2012). There is currently no cure for FMS, although symptoms can be managed. Increasing knowledge of the factors which may trigger and maintain symptoms is useful in understanding the condition and supporting patients.

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Adverse Childhood Experiences (ACEs) such as abuse, neglect or witnessing domestic violence have long-term negative consequences on cognitive, emotional, and behavioural processes (Cicchetti, 2016; Smith & Pollak, 2020). Chronic and severe early life stress can impact the developing brain repressing the body's ability to regulate stress at critical maturational stages, and subsequently into adulthood (Danese & McEwen, 2012). Such stress can also affect the functioning of critical neurological systems which facilitate psychological and behavioural responses to the environment, particularly to perceived threats (Danese & Lewis, 2017). People who have experienced sustained adversity in childhood show compromised immunity and raised inflammatory biomarkers when exposed to everyday stressors (Danese & Baldwin, 2016; Smith & Pollak, 2020 for reviews) and there is substantial evidence linking ACEs with a range of adult health conditions, including FMS (Groenewald et al., 2020; Hughes et al., 2017; Olivieri et al., 2012; Varinen et al., 2017). A significant mechanism in the aetiology of FMS is thought to be the development of Central Sensitivity (Burke et al., 2016; Galves-Sanchez et al., 2019; Kaleycheva et al., 2021; Nelson et al., 2020; Van Houdenhove & Luyten, 2007), whereby chronic stress leads to the amplification of sensory input to the central nervous system resulting in a perception of pain from stimuli which are usually non-painful, such as clothing against the skin (allodynia), and greater pain than would be expected from painful stimuli (hyperalgesia; Sluka & Clauw, 2016; Turk & Monarch, 2018).

Emotionally invalidating childhood experiences (e.g. psychological abuse, punitive punishments, and negative responses to displays of emotion) are associated with emotional inhibition in adulthood, including difficulties in self-expression, avoidant stress responses, depression and anxiety. Silencing-the-Self theory (Jack, 1991) explains the development and consequences of these behaviours and describes how women particularly learn to censor themselves, accommodate, and to devalue their own experiences, repressing their feelings and needs in the hope of maintaining relationships. Jack (1991) suggested that this behaviour profile stems from early experiences, and reflects a childhood coping mechanism developed for safety and to meet social expectations (Jack, 1991, 1999). As such, self-silencing is not a heritable and stable personality trait, but a cognitive schema developed as a result of social experiences, and therefore malleable and amenable to intervention (Azari et al., 2020). Silencing-the-self (STS) remains a useful psychological framework for understanding women's health outcomes (Maji & Dixit, 2019) and has been implicated in irritable bowel syndrome (IBS; Ali et al., 2000) and chronic fatigue syndrome (CFS; Hambrook et al., 2011), both chronic inflammatory conditions with uncertain aetiology and often co-morbid with FMS. In both cases, patients reported higher levels of self-silencing compared to healthy controls, with Hambrook et al. (2011) reporting care and self-sacrifice specifically significant.

There is limited extant evidence to suggest STS and fibromyalgia may be related. Wentz et al. (2004) described a pattern of behaviour they termed 'hypermanic helpfulness', whereby women's personal needs were always secondary to the needs of others. Wentz et al. reported that women with FMS displaying this behavioural profile had frequently experienced adversity and associated psychological burden in childhood. Other research has shown that women with FMS can present with a lifestyle characterised by extreme altruism and self-sacrificial tendencies linked to harm-avoidance (Van Houdenhove & Luyten, 2007).

Overall, we propose that the relational behaviours typified within STS offer a psychosocial explanation of the pathway linking ACEs and symptoms associated with fibromyalgia. As ACEs are associated with poorer health generally, we expected to observe a positive association between number of ACEs and symptoms in women with, and without, an FMS diagnosis. Our aim is to investigate how this relationship differs as a function of STS. Although we examine the effects of all four dimensions of STS, given the findings of Hambrook et al. (2011), we specifically predict that scores on the care and self-sacrifice dimension will mediate the association between ACEs and symptoms in FMS participants only.

Methods

Participants

We advertised for participants with FMS in online support groups and 540 women responded and self-declared a clinical diagnosis of FMS. One did not complete all the measures and was removed from the dataset, leaving 539 for analysis ($M_{age} = 42.30$, $SD = 12.64$). Two hundred and ninety-one (54%) declared a co-morbid diagnosis of chronic fatigue syndrome (CFS), irritable bowel syndrome (IBS; common co-morbidities which have also been linked to stress), or other chronic condition. A further sample of 184 women from the UK public ($M_{age} = 36.94$, $SD = 9.23$) took part through www.Proflic.ac.uk, an online research recruitment platform, and declared no diagnosis of FMS, CFS, IBS or any other long-term health condition. In both groups, the other inclusion criteria were that participants needed to be at least 18 years of age, have been born female and still identify as female. The groups differed in age, $t(718) = 5.30$, $p < .001$, and as this may potentially influence symptom burden we controlled for age in our analyses.

Materials and Procedure

The study was approved by the authors' university Ethics Committee. Data was collected online through a questionnaire hosted on the Qualtrics platform. Participants were given full details, and checked a box to give informed consent, before completing the following measures:

General Symptom Questionnaire-65 (GSQ-65; Hyland et al., 2019)

Participants rate how frequently they experience 65 symptoms on a 5-point scale, where 1 = 'never/almost never' and 5 = 'every day'. Responses are averaged to give a total severity score. The GSQ-65 includes a wide range of symptoms typical of FMS, and many of which are also common everyday occurrences in healthy individuals, e.g. headaches. The GSQ is therefore suitable for use with both FMS and healthy participants with expectation of lower scores in the latter group, as in previous research (Bacon et al., 2021). The GSQ showed good reliability with the present samples (FMS $\alpha = .95$, non-FMS $\alpha = .98$).

Four GSQ items relate to depression and anxiety. These are prevalent in women who self-silence (Jack, 1991) and also in FMS where they can be exacerbated by, and help to maintain, symptom burden (Thieme et al., 2004). We therefore wished to control for

these factors in our analysis. We therefore calculated the mean response to these four items separately and used the resulting score (which we refer to as depression/anxiety) as a covariate, rather than administering further measures ($\alpha = .87$ in both groups).

Silencing the Self questionnaire (Jack & Dill, 1992)

Participants respond on a 5-point Likert scale ranging from 1 (strongly disagree) to 5 (strongly agree) to items on four subscales of STS: Externalised self-perception (ESP, judging oneself by external standards; FMS $\alpha = .84$, non-FMS $\alpha = .85$), Care as self-sacrifice (CASS, putting the needs of others before one's own; FMS $\alpha = .79$, non-FMS $\alpha = .75$), Silencing the Self (STS, inhibiting one's self-expression to avoid conflict and possible loss of relationships; FMS $\alpha = .68$, non-FMS $\alpha = .82$), and Divided Self (DS, presenting compliance while inwardly angry and hostile; FMS $\alpha = .73$, non-FMS $\alpha = .79$).

Adverse Childhood Experiences scale (Felitti et al., 1998)

This widely used measure presents ten examples of adverse experiences and participants indicate whether or not they experienced each of these before the age of 18. Items reflect physical, sexual and emotional abuse as well as having parents/carers who were addicts or divorced. Yes responses are summed to give an overall ACE score (FMS $\alpha = .77$; non-FMS $\alpha = .78$).

Data analysis

Analysis was conducted using SPSS v. 25. We computed descriptive statistics and compared group means using independent samples t-tests. One-tailed Pearson product moment correlations examined associated between the key variables in each group. In linear regression analysis on reported symptoms, we first entered age, depression/anxiety as covariates, together with diagnosis (coded whereby FMS = 1 and no-FMS = 0) and number of ACEs. Then, at Stage 2 we added the four silencing-the-self subscale scores. As STS subscale scores were intercorrelated, we examined Variance Inflation Factors. These fell in the range 1.04–2.45, well below the suggested threshold of 5, therefore suggesting no problems with multicollinearity (Hair et al., 2010). Moderated mediation analysis was conducted using the PROCESS macro for SPSS, model 7 (Hayes, 2018) and included age, depression/anxiety and STS scores as covariates.

Results

Table 1 presents descriptive statistics. The FMS group scored significantly higher on all measures.

In line with our first prediction, ACEs were positively associated with symptoms in both FMS ($r = .25$, $p < .001$) and non-FMS ($r = .30$, $p < .001$) participants and these associations remained significant after controlling for depression/anxiety ($r = .18$ and $.19$, respectively, $p < .001$, one-tailed).

Results of linear regression analyses on symptoms are shown in Table 2.

At Stage 1, diagnosis, depression/anxiety and ACEs explained highly significant levels of variance as expected and these effects remained at Stage 2, though adding STS made a significant improvement to the model, $\Delta R^2 = 0.02$, $F(4, 710) = 2.88$, $p = .02$. Significant

Table 1. Descriptive statistics and comparison between groups.

Variable	FMS		No-FMS		Comparison (df = 721)
	Mean	SD	Mean	SD	
Symptoms	4.17	0.78	3.09	1.15	t = 14.26, p < .001, d = 1.11
Depression/Anxiety	4.56	1.33	3.72	1.45	t = 7.21, p < .001, d = 0.60
ACEs	3.04	2.49	2.37	2.43	t = 3.20, p = .001, d = 0.27
ESP	21.51	5.77	20.02	5.84	F = 9.18, p = .003, $\eta^2 = .01$
CASS	28.70	7.05	27.26	6.08	F = 6.04, p = .01, $\eta^2 = .01$
STS	25.72	6.04	24.18	6.72	F = 8.33, p = .004, $\eta^2 = .01$
DS	20.05	5.71	18.80	6.04	F = 6.29, p = .01, $\eta^2 = .01$

Table 2. Results of regression analyses on symptoms.

		St. β	p	95% CI		Adj. R ²
				Lower	Upper	
1.	Age	0.04	.11	-0.001	0.01	
	Depression/anxiety	0.52	<.001	0.33	0.41	
	Diagnosis	0.31	<.001	0.59	0.84	
	ACEs	0.12	<.001	0.03	0.07	
2.	Age	0.03	.22	-0.002	0.01	.50
	Depression/anxiety	0.52	<.001	0.33	0.42	
	Diagnosis	0.31	<.001	0.60	0.85	
	ACEs	0.11	<.001	0.02	0.07	
	ESP	-0.09	.02	-0.03	-0.003	
	CASS	0.11	.004	0.01	0.03	
	STS	-0.01	.85	-0.01	0.01	
	DS	0.03	.43	-0.01	0.02	
					.52	

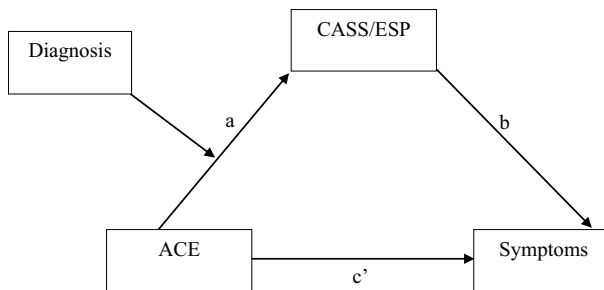


Figure 1. Hypothesised moderated mediation model.

independent effects of CASS (positive) and ESP (negative) were observed. Finally, we tested the hypothesised moderated mediation model shown in Figure 1. This allowed us to examine the conditional moderating effect of diagnosis (i.e. FMS or no-FMS) on the indirect relationship between ACEs and symptoms via silencing-the-self (path a in Figure 1), and thus to test Prediction 2.

The direct effect (path c' in Figure 1) was significant, illustrating that ACEs and symptoms were positively associated overall, $\beta = 0.05$, $SE = .01$, $p < .001$, 95%CI [0.03 0.08]. Following the regression results above, we tested mediating effects of both CASS and ESP. Diagnosis significantly moderated the effect of ACEs on CASS (path a in Figure 1), Interaction $\beta = 0.42$, $SE = .17$, $p = .01$, 95% CI [0.09, 0.76], and higher levels of CASS were associated with higher levels of symptoms (path b), $\beta = 0.02$, $SE = 0.01$, p

= .02, 95%CI [0.003, 0.03]. A significant index of moderated mediation, $\beta = 0.01$, $SE = 0.003$, 95%CI [0.001, 0.01] indicated a significant moderating effect of diagnosis in the overall indirect effect via CASS (path a + path b). The conditional indirect effect was significant in those with an FMS diagnosis, $\beta = .01$, $SE = 0.004$, 95%CI [0.001, 0.01], but not in participants with no diagnosis, $\beta = -0.002$, $SE = 0.003$, 95%CI [-0.01, 0.003].

With ESP as mediator, we observed no significant moderating effect of diagnosis (interaction $p = .17$), no significant index of moderated mediation, $\beta = 0.003$, $SE = 0.003$, 95% CI [-0.002, 0.01], and no significant indirect effect for either group of participants; FMS $\beta = 0.002$, $SE = 0.001$, 95%CI [-0.001, 0.005]; no-FMS $\beta = -0.002$, $SE = 0.002$, 95%CI [-0.01, 0.003]. A post-hoc simple main effects analysis confirmed that a significant direct effect of ESP on symptoms was only observed in the FMS group, $\beta = -0.25$, $p < .001$, 95% CI [-0.05, -0.02].

Discussion

Previous evidence suggests an association between ACEs and FMS symptoms and the present study aimed to examine whether this could be partly accounted for by behaviours encapsulated within Silencing-the-Self theory (STS; Jack, 1991, 1999). As predicted, ACEs and symptoms were significantly associated in FMS, and in a general public sample of women with no FMS diagnosis. This adds further support to studies which have suggested that adversity in childhood can be predictive of adult health status (Afifi et al., 2016; Baiden et al., 2021; Boullier & Blair, 2018; Nelson et al., 2020; Solís et al., 2015; Wade et al., 2015) and specifically in terms of FMS (Groenewald et al., 2020; Olivieri et al., 2012; Varinen et al., 2017). In addition, we predicted, and observed, a moderating effect of diagnosis (FMS vs. no FMS) on the hypothesised indirect path from ACEs to symptomology via STS. As predicted, the indirect effect was only significant in the women with a diagnosis of FMS, and only for one STS factor, care and self-sacrifice (CASS).

According to Jack (1991; 1999), CASS reflects the extent to which relationships are secured by putting the needs of others ahead of the needs of the self and such behaviours are reported in women with FMS (Wentz et al., 2004). The ongoing influence of ACEs on neurobiological stress responses is known to result in hyper-responsiveness to threat stimuli into adulthood (Danese & Lewis, 2017). Jack (1991; 1999) reports how women who self-silence grow increasingly inwardly angry and resentful as they attempt to sustain their role as a 'good' partner, mother, or daughter (Brody et al., 2014), perpetuating stress responses and coping strategies which are incompatible with self-care. We propose that self-silencing, symptom burden and heightened stress responses combine to exacerbate and maintain FMS symptoms. Further research is now needed to examine in more detail how these factors interact, both psychologically and physiologically. Future studies might also consider more specifically the role of central sensitivity by incorporating physiological markers and/or scores on self-report scales such as the Central Sensitivity inventory (Mayer et al., 2011). In addition, it is important to acknowledge that the mechanisms at work are complex, encompassing factors such as genetic predisposition, and in-vitro experiences which may influence stress reactivity and are difficult to control for.

Finally, we also observed a negative association between STS factor Externalised self-perception (ESP) and FMS symptoms. ESP has been implicated in eating disorders (Buchholz et al., 2007; Frank & Thomas, 2003) where the tendency to judge oneself by perceived external standards of appearance contributed to psychopathology and body dissatisfaction (Maji & Dixit, 2019). We know of no precedent in the literature for a negative association between self-silencing generally, or ESP in particular, and health symptoms per se. However, there is evidence to suggest that women AIDS patients with higher ESP are less likely to share their health status with others because of concerns around stigma and negative social attitudes (DeMarco et al., 1998). Our findings suggest that women with FMS who have a tendency to ESP have fewer symptoms, but it may be that they downplay the severity of their condition because of concerns about how they are perceived. Many FMS patients report negative attitudes from health professionals (Bennett et al., 2018) and this can affect treatment adherence (Rowe et al., 2019). The possibility of more generalised maladaptive beliefs about social attitudes to health in FMS requires further investigation.

The present findings may also be considered in the context of pain behaviours. Behaviours, such as limping, avoidance of certain activities, or complaining about pain, have been conceptualised as overt expressions of pain which have a communicative social function (Fordyce, 1976). Research has associated pain behaviours with the nature and quality of spousal/partner relationships, particularly in terms of how perceived negative responses can be associated with heightened pain perception (Forsythe et al., 2012). Furthermore, pain behaviours have been linked to an insecure attachment style which often develops in the context of ACEs (e.g. Murphy et al., 2014). The interesting aspect in the present context is that STS is intrinsically intertwined with spousal support seeking and maladaptive ways of soliciting this. Future research might usefully examine the relationship between these factors and highlight how interventions which focus on STS within the environment in which a woman lives may be helpful in addressing pain conditions (Forsythe et al., 2012).

These findings also have implications for practice. FMS interventions tend to focus on symptom management and supporting patients with the psychological toll of living with their condition. Treatment is typically through non-pharmacological treatments such as Mindfulness and/or Cognitive Behavioural Therapy where emotional understanding and authentic self-expression are vital for effective outcomes (Lumley & Harkness, 2007). An understanding of the role that social-behavioural factors such as self-silencing can play may support and enrich such therapies for FMS.

Limitations

We used self-report measures; however, false-positive reports of ACEs are likely to be rare and bias not sufficiently great to invalidate retrospective reports (Hardt & Rutter, 2004). Indeed, subjective retrospective reports are more predictive of psychopathology than objective court-documented evidence (Danese & Widom, 2020). Additionally, although we employed the most widely used ACE questionnaire (Felitti et al., 1998), it represents only ten potential experiences and excludes others known to influence adult outcomes, e.g. bullying and exposure to community violence (Lacey & Minnis, 2020). Our participants were self-selecting. Although online participants tend to be more

representative of the population at large than those recruited for lab-based research (Woods et al., 2015), some individuals with FMS expressed interest in the research initially, but subsequently reported being too unwell to take part. It is possible that more severely affected women may have excluded themselves from participation. Similarly, our sample may be representative of a sub-group of women with a propensity to participate in research and to express themselves. Finally, we collected self-reports of diagnosis (or lack of in the healthy group) without the benefit of corroborating evidence from a clinician.

Conclusions

This study presents some preliminary new information which contributes to explaining the previously documented association between ACEs and FMS symptoms. Behaviours encapsulated within silencing-the-self theory were significantly associated with symptom levels and mediated the association between symptoms and ACEs only in participants with a diagnosis of FMS. This relationship was observed after controlling for anxiety and depression which are pervasive in women with FMS and in those who self-silence (Jack, 1991). A biopsychosocial perspective is advocated for a comprehensive understanding of chronic pain conditions such as FMS (Turk & Adams, 2016) and our results suggest that socially mediated behavioural factors can influence and maintain symptoms. These results emphasise the importance of a holistic perspective, taking into account biological and psychosocial factors in understanding, treating and researching FMS.

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