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Stressful life event appraisal and coping in patients with psychogenic seizures and those with epilepsy

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ABSTRACT

Understanding stress and coping among individuals with psychogenic nonepileptic seizures (PNES) may have important treatment implications. 40 patients with PNES, 20 with epilepsy (EPIL), and 40 healthy control (HC) participants reported the frequency of various stressful life events (both positive and negative) and appraised the distress these events induced. They also described their habitual coping behaviors. PNES patients reported no more frequent stressful life events than EPIL patients or HC. In addition, the stressors they experienced are not objectively more severe. However, they reported more severe distress due to negative life events, especially in the domains of work, social functioning, legal matters, and health. PNES patients also engaged in less planning and active coping than HC. Neither of these two coping behaviors was associated with distress ratings. The PNES group did not engage in more denial than either group. However, greater denial among PNES patients was associated with greater perceived distress. Coping in PNES is characterized by elevated levels of perceived distress and fewer action strategies than are normally employed to reduce the impact of a stressor. These findings may inform cognitive behavioral therapy of PNES patients.

1. Introduction

Psychogenic non-epileptic seizures (PNES) are episodes of altered movement, sensation, or experience that mimic epileptic seizures, but are not caused by abnormal brain activity and are assumed to reflect an emotional disorder. Patients with PNES often have problematic behavior and unstable relationships, are occupationally disabled, and have high healthcare expenditures, even years after the non-epileptic nature of their events is identified.[1–4] Effective treatments of PNES are sorely needed,[5–7] and cognitive behavioral therapy and antidepressant medications have recently shown some promise.[8,9] Nonetheless, a better understanding of how individuals with PNES appraise, manage, and cope with stressful life events could be essential in developing and refining interventions and matching them appropriately to individual patients.

Reports of psychologically traumatic events, such as physical or sexual abuse, are commonly cited as integral to the genesis of PNES behavior.[10–18] However, the validity of such claims is often difficult to determine. Moreover, a wider variety of stressful life events may precipitate, perpetuate or be associated with PNES.[19–21] Bowman and Markand[22] found that 92% of their PNES patients reported at least one recent stressful life event, but many reported multiple events (e.g., accidents, life-role changes, personal illness, relationship conflict, or job loss). Although this study did not include a comparison group, later studies did and have often found that PNES patients report more frequent stressful life events and have higher perceived stress than patients with epilepsy (EPIL).[23,24]

PNES behavior is often conceptualized as a way for patients to cope with situations perceived to be stressful and beyond their control. Patients with PNES may be seen as having adopted the "sick role"; a status that relinquishes them of responsibility for meeting stressful life demands.[25] Indeed, some PNES patients may deny the influence of stressful life circumstances and attribute all their problems in life to the effects of seizures.[26] Consistent with this formulation are findings from Frances et al.[23] and Goldstein et al.[27] of more escape and avoidance behavior and less planful problem-solving among patients with PNES than among healthy adults.

It is widely believed that stressful life events and maladaptive coping are integral to the onset and maintenance of PNES,[28] and may therefore be relevant to its treatment. Although studies have demonstrated that PNES patients appraise stress and cope differently from patients with epilepsy,[24] none have examined

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the relationship between stress appraisal and coping. Moreover, there have been no attempts to identify subgroups of PNES patients who habitually engage in particular coping behaviors.

The goal of the current study is to determine whether there are systematic differences in the frequency and appraisal of stressful life events – be they positive stressors or negative stressors–as well as preferred methods of coping, among persons with PNES, patients with EPIL, and neurologically normal individuals. We hypothesize that patients with PNES will report more frequent and severe stressful life events, rate these as more distressing, and engage in less problem-focused coping and emotional restraint and more denial, suppression, and behavioral and mental disengagement than participants in the other two groups. Further, to explore the possibility of subgroups, we predict that PNES patients with lower IQ estimates will show exaggerated deficits in problem-focused coping (i.e., less active coping, planning, and more denial and repression).

2. Methods

2.1. Participants

Forty adults with PNES and 20 with confirmed epilepsy (EPIL) evaluated in the Johns Hopkins Hospital Epilepsy Monitoring Unit (EMU) participated in this study. All consecutive patients meeting criteria for the study were approached for participation shortly after their hospital admission. Full-time faculty epileptologists made the final diagnoses of seizure type. All patients were given definitive diagnoses based on video-EEG recording and other supporting evidence (e.g., neuroimaging). Only patients who had one or more of their typical seizure events recorded during their EMU hospitalization were included in the study. The diagnosis of definite PNES was made when one or more typical events were recorded on video which: (1) had a semiology inconsistent with epilepsy, and (2) were accompanied by EEG with no epileptiform discharge or ictal slowing. Definite epilepsy was diagnoses when video-EEG recorded the simultaneous occurrence of a stereotypic event and an epileptiform discharge.

Participants were excluded from the current analyses if they had both EPIL and PNES ($N = 4$), physiologic non-epileptic seizures (i.e., sleep myoclonus or syncope; $N = 4$), or unclear or inconclusive video/EEG findings ($N = 16$). Others were excluded because they were discharged from the hospital before completing the study procedures ($N = 12$). Forty adults who reported no history of neurologic or psychiatric disorder or chronic medical condition served as a healthy control (HC) group ($N = 40$). They were recruited from the Johns Hopkins Hospital community by posted and online advertisement.

All patients and control participants were required to be at least 18 years old and to have IQ estimates of at least 70 based on the Wonderlic Personnel Test.[29,30] Potential participants were excluded if they had been diagnosed previously with any other neurologic disease or a severe psychiatric illness (e.g., schizophrenia or bipolar disorder) or had current drug or alcohol abuse. Demographic information and medical history were collected through review of the medical chart and interview with the participant.

The research protocol was reviewed and approved by the Johns Hopkins Medicine Institutional Review Board. All participants gave informed consent after the goals and methods of the study, and the risks and benefits of participating, were explained fully.

2.2. Procedure

Participants completed a modification of the Psychiatric Epidemiology Research Interview (PERI) Life Events Scale,[31] a self-report checklist on which subjects indicated how many times they experienced specific stressful events (positive or negative). A total of 102 events from the life domains of school, work, love and marriage, having children, family, residence, crime and legal matters, finances, social activities, and health were included. Study participants provided frequency ratings for two time periods: past 12 months and one to five years ago. We also calculated severity scores by multiplying each item's empirically derived weight by how many times it was experienced. This accounts for the greater severity inherent in some life events than others (e.g., the death of one's spouse is a normatively more severe stressor than is moving one's residence). Finally, each life event that was experienced was rated by the participant for how much distress it caused (scale = 0–100).

Participants also completed the COPE,[32] a 60-item, multidimensional inventory that assesses the different ways in which people respond to stress. Fifteen coping styles are assessed: positive reinterpretation and growth, mental disengagement, focus on and venting of emotions, use of instrumental social support, active coping, denial, religious coping, humor, behavioral disengagement, restraint, use of emotional social support, substance use, acceptance, suppression of competing activities, and planning. The questions are formatted to ask about "dispositional" or trait-like coping. Respondents report the extent to which they typically engage in the behavior or cognition when they are under stress (e.g., "not at all," "a little bit," "a medium amount," and "a lot"). The COPE has been found to have good construct and external validity.[33]

The Personality Assessment Inventory (PAI)[34] was used to assess aspects of psychopathology and included in this study for descriptive purposes only. We considered scores only of the Somatic Complaints (SOM), Anxiety (ANX), and Depression (DEP) since these PAI domains are conceptually and empirically most relevant in PNES.[35–37]

2.3. Data analyses

2.3.1. Group comparisons

Three separate 3-group MANCOVAs, controlling for sex, education, and estimated IQ, were performed to determine whether there are reliable between-group differences on the PERI in (1) the frequency and (2) severity of positive, negative, recent or remote stressful life events and (3) distress ratings of positive and negative stressful life events.[1] Post hoc analyses of PERI data entailed one-way ANOVAs for each life domain. We conducted bivariate correlations between coping styles and distress ratings that differed among the groups. An additional 3-group MANCOVA was conducted on all scales of the COPE. While all univariate ANOVAS of the estimated marginal means were performed, post hoc comparisons were limited to those scales that had effect size values (η_p^2) greater than 0.10. Finally, we performed a post hoc analysis to determine the relationship between IQ and coping in PNES, we divided the PNES sample into high and low IQ groups based on their median IQ value and analyzed coping profiles by IQ.

3. Results

The groups differed on several demographic characteristics (see Table 1). The HC and EPIL group was more highly educated than the PNES group. The HC group had higher mean estimated IQ than both patient groups. The PNES and HC groups had more women than the EPIL group. The PNES group reported more somatic complaints than ES and HC, and both patient groups reported more symptoms of depression and anxiety than HC. To control for group differences

[1] A Group × Valence × Epoch mixed-model ANOVA was also conducted, but no main effects or interactions were significant.

Table 1
Description of study sample (means ± SDs).

	HC	EPIL	PNES	*p*	Post hoc
N	40	20	40		
Age, years	39.65 (11.32)	36.60 (12.52)	36.67 (11.17)	.445	NA
Sex (M:F)	7:33	9:11	3:37	.002	EPIL < HC = PNES
Education, highest grade	15.31 (2.22)	15.40 (2.54)	13.7 (1.89)	.002	PNES < HC = EPIL
Wonderlic Personnel Test:	109.08 (11.81)	99.40 (12.88)	100.77 (12.92)	.004	HC > PNES = EPIL
Age at onset of seizures	–	18.47 (17.97)	32.48 (11.47)	<.01	NA
Duration	–	17.42 (14.73)	4.20 (5.12)	<.001	NA
Number of antiepileptics	–	1.84 (0.69)	1.17 (0.98)	.01	EPIL > PNES
Seizure frequency[a]				.004[b]	
Daily	–	26.3%	35.0%	NA	
Weekly	–	15.8%	35.0%	NA	
Monthly	–	21.1%	27.5%	NA	
Yearly	–	36.8%	5.0%	NA	
PAI					
Somatic Complaints	48.13 (8.88)	65.7 (12.89)	72.8 (14.49)	<.001	PNES > ES > HC
Anxiety	49.9 (10.97)	57.75 (13.05)	59.98 (15.59)	<.001	PNES&ES > HC
Depression	49.05 (11.16)	58.65 (12.18)	63.98 (16.97)	<.001	PNES&ES > HC

NA: Not Applicable

[a] Seizure frequency is defined as: daily = one or more events each day, weekly = one or more events each week, but not daily, monthly = several each month, and yearly = several each year.
[b] Based on Chi-square.

Table 2
Stressful life events.

	HC		EPIL		PNES		*p*	η_p^2	Post hoc
	M	SE	*M*	SE	*M*	SE			
Recent positive life events (sum)	13.08	7.12	15.16	10.45	17.66	7.42	.91	0.001	
Recent negative life events (sum)	3.23	6.32	30.75	9.28	10.74	6.59	.06	0.06	
Remote positive life events (sum)	38.11	14.60	34.58	21.43	43.21	15.21	.95	0.001	
Remote negative life events (sum)	16.40	18.48	29.05	27.14	46.90	19.26	.55	0.01	
Recent positive (Freq. × weight) lg_{10}	2.49	0.11	2.52	0.15	2.22	0.09	.11	0.12	
Recent negative (Freq. × weight) lg_{10}	2.20	0.11	2.53	0.16	2.47	0.10	.46	0.06	
Remote positive (Freq. × weight) lg_{10}	2.85	0.10	2.81	0.14	2.80	0.09	.65	0.05	
Remote negative (Freq. × weight) lg_{10}	2.62	0.11	2.78	0.16	2.84	0.10	.17	0.10	
Distress positive life events (mean)	30.47	3.20	28.50	4.81	37.50	3.30	.414	0.058	
Distress negative life events (mean)	56.53	3.05	66.38	4.59	72.06	3.14	.009	0.167	PNES > HC

in sex, education and estimated IQ, all analyses were conducted with these variables entered as covariates; adjusted means are reported. Although using these covariates precludes identification of covariate-specific subgroups, none of these covariates was related systematically to stress or coping variables in the current study.

3.1. Stressful life events comparisons

The differences among groups for the overall frequency of positive or negative, recent or remote stressful life events were not significant (Hotelling's T (8, 178) = 1.03, $\eta_p^2 = 0.04$, $p = .41$; see Table 2). Although the EPIL group appeared to have more negative than positive recent life events, a pattern that is opposite from the other two groups, the interaction between group and valence of life event was not significant (Hotelling's T (2, 93) = 1.20, $\eta_p^2 = 0.03$, $p = .31$). Similarly, the frequency of negative and positive remote life events did not vary by group (Hotelling's T (2, 93) = 0.27, $\eta_p^2 = 0.01$, $p = .76$).

Severity scores similarly did not result in any differences among groups (Hotelling's T (8, 132) = 1.39, $\eta_p^2 = 0.08$, $p = .21$). However, the difference among groups in distress ratings was significant (Hotelling's $T(4, 162) = 3.54$, $\eta_p^2 = 0.08$, $p = .008$), due mainly to the higher ratings of distress in PNES. Univariate ANCOVAs revealed group differences on distress ratings of negative life events (see Table 2), with patients with PNES scoring higher than HC. Closer

Table 3
Distress ratings by life domain (means ± SE).

	HC		EPIL		PNES		*p*	η_p^2	Post hoc
	M	SE	*M*	SE	*M*	SE			
School	7.82	2.56	9.75	3.76	14.34	2.67	.234	0.031	
Work	6.07	1.77	9.83	2.60	12.99	1.85	.036	0.069	PNES > HC
Love and marriage	13.13	2.31	6.68	3.40	12.61	2.41	.275	0.027	
Children	2.82	1.05	.27	1.54	3.83	1.09	.196	0.034	
Family	9.94	2.02	9.72	2.97	15.35	2.11	.156	0.039	
Residence	8.44	1.65	5.20	2.42	9.56	1.72	.367	0.021	
Crime and legal	1.54	1.14	2.95	1.67	7.78	1.18	.001	0.131	PNES > HC&EPIL
Finances	6.26	1.45	5.24	2.13	8.30	1.51	.478	0.016	
Social	7.84	1.75	10.52	2.58	16.35	1.83	.006	0.103	PNES > HC
Health	6.83	2.63	18.97	3.87	31.91	2.74	<.0001	0.305	PNES > EPIL > HC

Table 4
Coping strategies of subject groups. *T* scores on scales of the COPE (means ± SE).

	HC		EPIL		PNES		*p*	η_p^2	Post hoc
Positive growth	52.25	1.82	47.78	2.68	48.03	1.90	.212	0.033	
Mental disengagement	46.73	1.70	49.74	2.49	48.35	1.77	.587	0.011	
Emotional venting	50.29	1.56	51.55	2.30	51.22	1.63	.875	0.003	
Social support	51.04	1.47	55.24	2.16	49.86	1.53	.144	0.041	
Active coping	54.58	1.53	48.66	2.24	44.89	1.59	.0002	0.166	HC > EPIL&PNES
Denial	47.01	1.67	54.29	2.45	50.22	1.74	.051	0.062	EPIL > HC
Religion	54.71	1.73	56.79	2.54	56.89	1.80	.650	0.009	
Behavioral disengagement	51.25	1.73	53.70	2.54	52.03	1.80	.733	0.007	
Restraint	49.44	1.51	46.37	2.22	48.87	1.57	.519	0.014	
Emotional suppression	51.11	1.56	51.05	2.28	49.36	1.62	.729	0.007	
Substance abuse	46.16	1.09	49.30	1.60	48.10	1.13	.223	0.032	
Acceptance	48.64	1.76	47.43	2.59	47.74	1.84	.907	0.002	
Suppression	52.92	1.59	48.05	2.33	50.54	1.66	.218	0.032	
Planning	53.71	1.56	49.07	2.29	45.86	1.63	.004	0.111	HC > PNES

inspection of distress ratings across various life domains (see Table 3) reveals that patients with PNES report more distress than HC related to work, social functioning, legal difficulties, and health than HC. PNES patients also report more distress than EPIL patients related to legal difficulties and health.

There were no associations between the frequency, severity, and distress associated with stressful life event ratings and seizure frequency, age at onset of seizures, or number of antiepileptic medications prescribed. However, duration of seizure disorder was correlated with more recent negative life events ($r = 0.27$, $p = .04$). In addition, negative life events are more distressing among those with more recent onset of seizures ($r = -0.40$, $p = .002$). This latter finding is driven primarily by the EPIL group ($r = -0.50$, $p = .04$). Neither the EPIL or PNES group show any relationship between duration of illness and frequency of recent negative life events.

3.2. Coping scale comparisons

The overall difference among groups in coping was significant (Hotelling's T (28, 158) = 1.68, $\eta_p^2 = 0.23$, $p = .03$). Univariate ANCOVAs revealed group differences in active coping, denial, and planning (see Table 4). More specifically, the PNES group engages in less active coping and planning than HC, and the EPIL group engages in more denial than HC. Significant associations did not emerge between coping and seizure frequency, duration of seizures, age at onset of seizures, number of antiepileptic drugs, or IQ in the overall sample. Among the PNES group, more substance abuse was associated with lower IQ scores ($r = -0.36$, $p = .03$).

3.3. Stress appraisal and coping

Bivariate correlations between distress associated with negative life events and active coping or planning were not significant at the overall sample or group level. There was a positive relationship between distress associated with negative life events and the use of denial at the overall sample ($r = 0.36$, $p < .001$). However, at a group level, this relationship was only present in the PNES group ($r = 0.47$, $p = .002$). Thus, higher distress was associated with greater levels of denial.

To explore the relationship between coping and negative life event distress further, we used a median split to divide the entire study sample by their negative life event distress ratings. In the HC and EPIL groups, coping did not differ by distress level. However, in the PNES group (see Fig. 1), the high negative distress subgroup

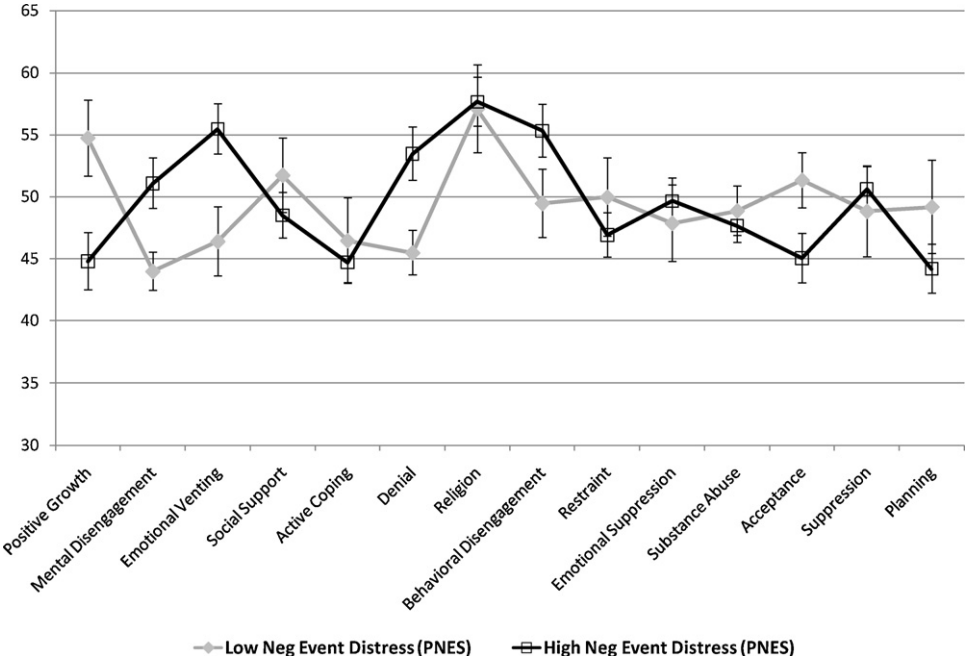

Fig. 1. Coping profiles of PNES patients reporting high or low distress related to negative life events.

($n = 28$) reported less positive growth ($p = .02$, $\eta_p^2 = 0.133$), and more mental disengagement ($p = .04$, $\eta_p^2 = 0.11$), focus on and venting of emotions ($p = .02$, $\eta_p^2 = 0.141$), and denial ($p = .03$, $\eta_p^2 = 0.12$) than the low negative distress subgroup ($n = 11$). Finally, to determine how coping varies by IQ in the PNES group, we divided the PNES group by their median IQ value (Mdn = 100). Coping scores in the PNES group did not differ between the high and low IQ groups.

4. Discussion

Contrary to our prediction, our patients with PNES did not experience more frequent recent or remote, positive or negative stressful life events than patients with epilepsy or healthy persons. In addition, the stressors they experienced are not objectively more severe. However, they subjectively experienced greater distress caused by these events. Our PNES group experienced greater distress related to work and social functioning, health status, and legal system involvement. Finally, while coping in PNES is characterized by diminished active coping and planning, we also found that higher levels of negative life event distress, especially in PNES, are associated with increased levels of denial, mental disengagement, focus on and venting of emotions, and diminished positive growth.

Our results are slightly different than those reported by Tojek et al.[24] who found a higher prevalence of stressful life events in PNES than in EPIL. Methodological differences may explain the discrepant findings, as the participants in the Tojek et al.[24] study rated the frequency of 32 rather than 102 life events. As in Tojek et al., however, our PNES group appraised events as more distressing than did the EPIL group. While Tojek et al. did not include a healthy comparison group, Frances et al.,[23] did and, as in the present study, found greater perceived stress in PNES than healthy participants. Thus, our findings and those of Tojek et al. and Frances et al. support the notion that patients with PNES perceive their lives as more stressful and chaotic than do healthy subjects or patients with EPIL.[23,24]

Neither Tojek[24] nor Frances[23] examined stress appraisal across various life domains. We found specific distress appraisal differences related to work, social, legal, and health events. What appears to be most prominent in the PNES group is the distress associated with health difficulties. The current findings suggest that the distress associated with health problems is severe for the PNES group and greater than in the EPIL group. This health-related distress may reflect somatization behavior.[38]

The coping strategies of the PNES group suggest diminished "problem-focused" coping. Thus, when faced with a stressor, PNES individuals may not formulate a plan or strategy to "handle the problem" or take direct action "one step at a time" to eliminate or neutralize the problem. The coping profile in our PNES sample suggests that the planning and action phases of dealing with problematic situations are not employed as often as they are among neurologically healthy individuals, despite their being of normal intellect. Our results are similar to those of Frances et al., but unlike that study we did not find PNES to engage in more escape–avoidance or denial than healthy controls. In fact, in our study, patients with epilepsy reported more denial than did healthy persons. We did find that PNES patients with high distress ratings, engaged in more denial than those with lower distress levels.

The relationship between coping and perceived distress suggests that greater distress related to negative life events is associated with increased use of denial in PNES patients. Though our results are cross-sectional, they are consistent with the longitudinal findings of Bodde et al.[3] who found that reductions in psychological distress of PNES patients (on the Symptom Check List; SCL-90-R) were associated with diminished dissociative symptoms (on the Dissociation Questionnaire). They also found that a reduction in seizure frequency over five years was associated with diminished distress and changes in coping. Thus, there is some empirical indication that high levels of psychological distress in PNES may be associated with the use of denial as a defense or coping mechanism. Regardless, our data appear to suggest that therapeutic approaches designed to reduce distress related to negative life events (especially health-related events) may be appropriate.

Though we attempted to measure the frequency and severity of recent and remote stressful life events, we did not survey patients about stressful life events that occurred more than five years ago. Thus, our methodology does not allow us to understand how traumatic childhood events may have influenced the development of PNES. However, there are methodological limitations inherent in relying on historical report. Moreover, approximately 80% of our PNES group had seizure onset in the last five years. Although psychodynamic theory would attempt to link the onset of PNES to early childhood psychosexual trauma, empirical support for this notion is lacking.

Contrary to our hypothesis, we did not find any relationship between IQ and coping. The lack of a relationship between IQ and coping in our sample, mirrors findings from studies of IQ and coping in other psychogenic disorders.[39] Although the range of IQ scores in our PNES sample was not overly restricted (IQ ranged from 73 to 126), we excluded study participants with IQs lower than 70 and cannot draw any conclusions about the coping habits of PNES patients with extremely low IQ scores.

In conclusion, patients with PNES report more distress associated with negative life events than do neurologically normal individuals and patients with epilepsy. Active and problem focused coping appears diminished in PNES, but unlike denial, are not associated with greater distress levels. A larger sample would provide the statistical power to examine the relationship between stress and coping at the subgroup level and provide treatment-relevant information about how personality and psychopathology might mediate perceptions of distress.

Conflict of interest

None of the authors has any conflict of interest to disclose.

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We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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