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Original Research Article

Psychometric Evaluation of the Caregiver Burden Inventory in Children and Adolescents With PANS

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Abstract

Objectives To establish the psychometric properties of the Caregiver Burden Inventory (CBI) in patients with Pediatric Acute-onset Neuropsychiatric Syndrome (PANS), which is characterized by the abrupt onset of obsessive-compulsive disorder and/or restricted eating and at least two additional psychiatric symptoms. Parents of patients with PANS have reported high caregiver burden. However, no validated instrument of burden exists for this population. **Methods** Study took place at a community-based PANS clinic where the CBI is administered as part of routine clinical care. The first CBI available during an active disease flare was analyzed ($N = 104$). Construct validity was evaluated within a confirmatory factor analytic framework. Associations between the CBI and patient/family characteristics were explored, and preliminary normative data for this population are presented. **Results** Item-factor loadings were strong, and the overall fit of the model was good (root mean square error of approximation = .061). Strict/metric measurement invariance was demonstrated across age. The mean Total Score in this sample was 36.72 ± 19.84 (interquartile range 19–53). Total Scores on the CBI were significantly elevated for parents of children who switched schools because of their illness (Cohen's $d = 0.75$, 95% confidence interval [CI] 0.28–1.22) and for those who had reduced work hours to accommodate the child's illness (Cohen's $d = 0.65$, 95% CI 0.10–1.20). However, in this relatively high-status sample, socioeconomic variables did not predict Total Scores. **Conclusions** Parents of patients with PANS experience high caregiver burden. The CBI may be confidently used to assess caregiver burden in this population.

Key words: neuropsychology; parent stress; research design and methods.

Introduction

Caregiver burden has been defined as the perceived negative effects of caring for a family member (Zarit, Todd, & Zarit, 1986). Caregivers' emotional, physical, and social well-being are often overlooked, leading some to call caregivers "the neglected patient" (Adelman, Tmanova, Delgado, Dion, & Lachs, 2014). Researchers use self-report scales to measure caregiver burden. Some scales were originally validated in caregivers of older adults before being applied to pediatric populations (Dada, Okewole, Ogun, & Bello-Mojeed, 2011; Lane et al., 2017; Novak & Guest, 1989;

Poulshock & Deimling, 1984; Steele, Long, Reddy, Luhr, & Phipps, 2003; Zarit et al., 1986). Other scales of caregiver burden and related concepts have been developed specifically for pediatric populations (Aitken et al., 2009; Brannan, Heflinger, & Bickman, 1997; Camfield, Breau, & Camfield, 2001; Everhart, Fiese, & Smyth, 2008; Khanna et al., 2012; Molteni et al., 2017; Streisand, Braniecki, Tercyak, & Kazak, 2001).

In this study, we evaluated the psychometric properties of a scale used to assess caregiver burden in a population of patients with Pediatric Acute-onset Neuropsychiatric Syndrome (PANS). PANS is defined by the abrupt onset of obsessive-compulsive disorder (OCD) and/or severely restricted food intake with at least two comorbid neuropsychiatric symptoms from the following categories: anxiety, mood dysregulation, irritability/aggression/oppositionality, behavioral regression, cognitive deterioration, sensory or motor abnormalities, and somatic symptoms (Chang, Frankovich, et al., 2015; Swedo, Leckman, & Rose, 2012). A distinct subcategory of PANS is Pediatric Autoimmune Neuropsychiatric Disorder Associated with Streptococcal infections (PANDAS), in which a group A streptococcal infection is coincident with the neuropsychiatric deterioration (Swedo et al., 2012). The broader category of PANS, on the other hand, is agnostic to trigger. Patients with both PANS and PANDAS present most commonly with a relapsing and remitting illness course, in which patients oscillate between disease flares and periods at baseline functioning (Frankovich et al., 2015).

Caregivers of patients with PANS consistently report distress; however, no validated burden scale exists in this population (Thienemann et al., 2017). Anecdotally, caregivers devote significant time and energy to caregiving, which can affect emotional and physical health as well as social interactions. To study caregiver burden in PANS, we wanted a scale that captured the multiple dimensions of burden, especially burden because of time dependency, on which the aforementioned scales did not focus sufficiently (Aitken et al., 2009; Brannan et al., 1997; Camfield et al., 2001; Everhart et al., 2008; Khanna et al., 2012; Molteni et al., 2017; Poulshock & Deimling, 1984; Streisand et al., 2001; Zarit et al., 1986).

We chose to collect the Caregiver Burden Inventory (CBI) in our clinic. The CBI conceptualizes burden in terms of five categories (time dependency, emotional health, physical health, development, and social relationships) (Novak & Guest, 1989). This multidimensional view of caregiver burden allows clinicians to better target support strategies for caregivers. If the CBI is valid in PANS, research studies could also assess the longitudinal relationship between disease severity and the dimensions of caregiver burden, possibly improving the precision with which caregiver support is offered.

In this study, to evaluate the psychometric properties of the CBI in children and adolescents with PANS, we (1) establish internal factor validity, (2) evaluate measurement invariance across younger and older children, and (3) assess external validity by correlating CBI with disease and demographic variables. We also provide some preliminary normative data for this population, which will provide clinicians and researchers with context for interpreting CBI scores. We hypothesized, a priori, that variables corresponding to greater psychosocial stress and burden for families and caretakers (e.g., socioeconomic status, school/work disruption) would be associated with higher scores on the CBI. We also hypothesized that the existing five-factor structure would be supported.

Methods

Patients and Procedures

This study was approved by the Stanford Panel on Human Subjects Institutional Review Board. Participants were parents/caregivers of child and adolescent patients of the Stanford PANS clinic between September 1, 2012 and October 17, 2016. Generally, patients at the Stanford PANS clinic live within 90 miles of clinic, have access to a primary care provider, and are of high socioeconomic status. Entry into clinic requires referral from an established primary care provider. Participants gave informed consent before data collection. Participants were selected from 256 consecutively evaluated patients. Patients who refused research consent were excluded ($n = 5$). Inclusion criteria were (a) child meets criteria for PANS/PANDAS ($n = 147$), (b) CBI was administered during active disease flare ($n = 114$), (c) the child is >4 years and <18 years old at the time of the CBI ($n = 107$), and (d) the CBI had <30% of items missing ($n = 104$). The first CBI to meet these criteria was selected for each child (usually from the initial presentation to the clinic), for a final sample of 104 CBIs. The CBI is a survey composed of 24 questions on five subscales: time-dependence, developmental, physical, social, and emotional burden (see Figure 1 for item names) (Novak & Guest, 1989). Each item is rated on a five-point scale (0—*Never*, 1—*Rarely*, 2—*Sometimes*, 3—*Quite Frequently*, 4—*Nearly Always*) with a higher score suggesting higher burden. The CBI is valid and reliable in caregivers of Alzheimer's patients (Novak & Guest, 1989). It also has adequate validity and reliability in caregivers of Rett Syndrome, a pediatric condition (Lane et al., 2017). A score >36 is used conventionally in adult populations to indicate the need for respite services (Lund, Wright, Caserta, & Utz, 2006). We chose to study the CBI, as opposed to another caregiver burden scale, because it measures multiple dimensions of caregiver burden.

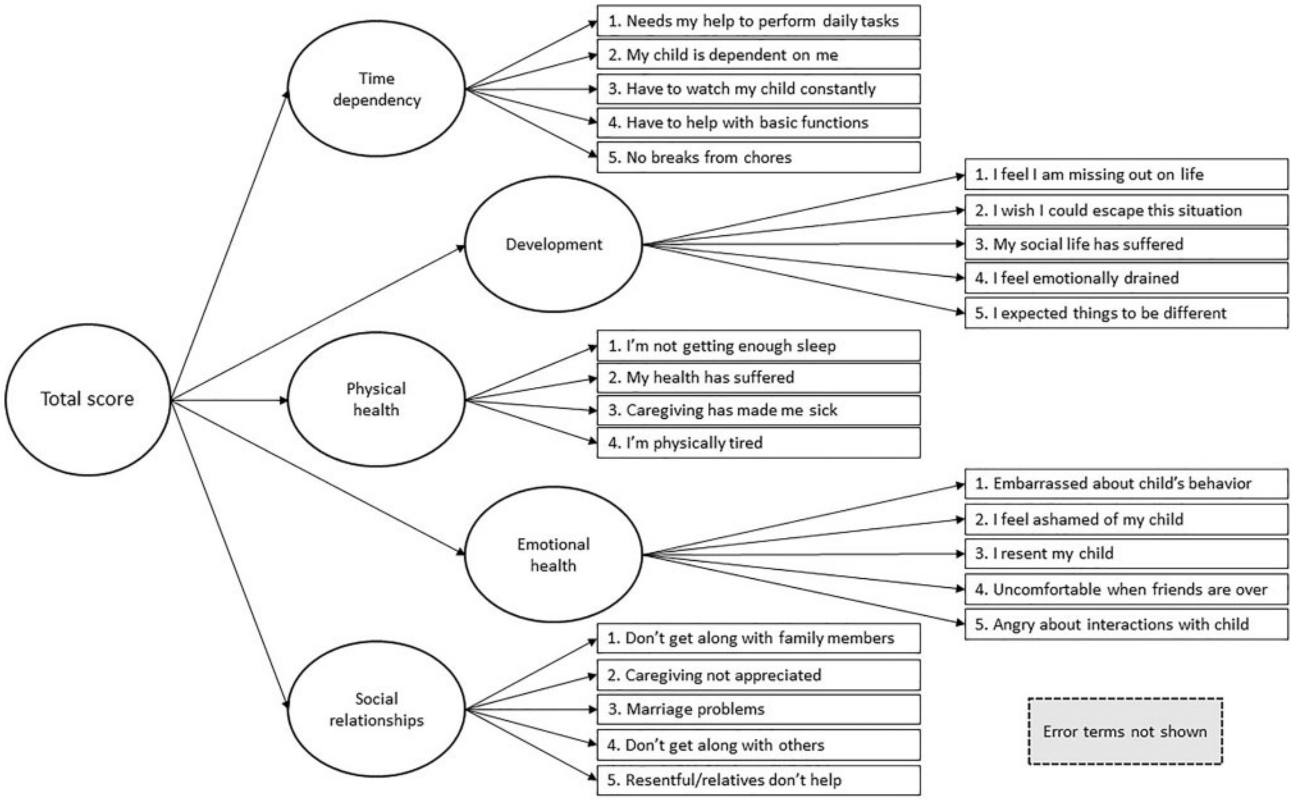

Figure 1. Factor structure of the CBI in the Stanford PANS clinic.

Note. CBI = Caregiver Burden Inventory; PANS = Pediatric Acute-onset Neuropsychiatric Syndrome.

We collected CBIs as part of an extensive Patient Questionnaire that caregivers fill out online before each clinic visit. The CBI is only collected in this questionnaire every 3 months or if the patient is in a new disease flare. If caregivers forget to fill out the form before the visit, we allow them to fill out the survey on an iPad in the waiting room. Caregivers have filled out a questionnaire for 83.8% of clinic visits; however, this value is an underestimate of the actual response rate because we do not send questionnaires for some clinic visits (e.g., visits spaced within a week of the previous visit with no indication that disease severity has changed).

For this study, we also sent out a one-time demographic survey that queried families on socioeconomic status (parental education and annual income), home environment (child's living arrangement, number of care contributors, at-home caregiving services), and school/work changes due to PANS. These variables were selected based on the a priori hypothesis that they would be associated with caregiver burden. Specifically, we included socioeconomic status because past research found a relationship between socioeconomic status and burden (Motenko, 1989). We included home environment variables because we hypothesized that caregiver burden would be lower in families with more care contributors and at-home caregiving services (although we did think it possible that families who hire at-home caregiving services could have children with higher disease severity, which would confound the association with burden). Finally, we hypothesized that school/work disruption would be associated with greater burden.

Statistical Analysis

Confirmatory factor analytic procedures were performed in MPlus Version 7 software (Muthén & Muthén, 2012). Data preparation and all other analyses were completed in SAS/STAT Version 9.3. The factor structure of the CBI is shown in Figure 1. First, data were prepared by collapsing response categories with low response rates (i.e., if fewer than nine respondents selected a response of '4' for a given item, '4' responses were combined with '3' responses for that item). Second-order confirmatory factor analysis was performed using the 24 items as ordered categorical indicators of the five factors, which were themselves indicators of a latent total score variable. Model estimation was performed using the mean and variance-adjusted weighted least-squares method, which tolerates missing data and makes no distributional assumptions about the observed variables (Li, 2016). Thus, missing data were not imputed for the factor analysis.

Model fit was assessed using a combination approach (Hu & Bentler, 1999). The χ^2 test of model fit is reported, but as it is sensitive to sample size and magnitude of correlation in the data, it often is statistically significant (thereby rejecting the null hypothesis of acceptable model fit) and is not a specific indicator of model fit. The root mean square of approximation (RMSEA), the standardized root mean squared residual (SRMR), and weighted root mean square residual (WRMR) are goodness-of-fit statistics that indicate better fit by lower values. RMSEA values <.06, SRMR values <.08, and WRMR values <1.0 are considered "good." The comparative fit index (CFI) and Tucker–Lewis index (TLI) indicate better fit with values closer to 1.0, where "good" is usually defined as >.97.

Given the wide age range of participants, preliminary measurement invariance analyses were performed for the first-order factor model. The grouping variable was age, split at 10 years. A sequential series of tests of increasingly restricted parameters was performed, including weak/configural invariance, strong/metric invariance, and strict/scalar invariance. Change in CFI was used to assess invariance; the simulation-based threshold for an unacceptable decrease in fit is .002 for ΔCFI (Meade, Johnson, & Braddy, 2008).

Given that this was a retrospective study, a priori power analysis was not completed. There is no agreed-upon adequate sample size for factor analysis. Although many authors settle on a rule-of-thumb of 10 observations per item, simulations suggest that adequate sample size depends on several conditions, including the strength of loadings and quality of the factor solution (Wolf, Harrington, Clark, & Miller, 2013). For this reason, we proceeded with analyses with the understanding that measures of fit may be adversely affected by the relatively small sample size.

The remaining analyses were performed with the CBI total score (i.e., items equally weighted and summed). For these analyses, missing item-level data were replaced with the sample mean; missing data were uncommon, and this was necessary for no more than four observations per item. As an initial demonstration of external validity, we present the association between CBI total score and demographic variables. These analyses were performed in a subset of 79 patients with a completed demographics form. We used analysis of variance (ANOVA) to test whether CBI total scores differed between levels of each demographic variable; differences are presented as Cohen's *d* effect sizes with 95% confidence intervals (CIs). Finally, we performed an exploratory, post hoc analysis comparing CBI total scores in patients diagnosed with PANDAS with CBI total scores in patients diagnosed with PANS only.

Results

In the factor analysis sample ($N = 104$), ages ranged from 4 to 17 years ($M = 10.9$, $SD = 3.5$). Most patients were male (62 of 104, 60%). All patients met criteria for PANS; 55 of 104 patients (53%) additionally met criteria for PANDAS. CBI was filled out by mother of patient in 79 of 104 instances (76%), by father of patient in 5 of 104 instances (5%), by both parents jointly in 13 of 104 instances (13%), and by another caregiver in 7 of 104 instances (7%). In total, 12 of 104 CBIs (12%) had at least one missing response item. The overall rate of missingness was 21 of 2,496 responses (0.008%). In total, 13 of the 24 response items had categories with a low response rate, and these categories were collapsed for factor analysis. As shown in Table I, all indicators loaded strongly and significantly onto the prescribed factors. The chi-square test of model fit was significant [$\chi^2(247) = 341.05$, $p = .0001$], rejecting the null hypothesis of model fit, but all other fit indices were in the good or adequate range. The RMSEA point estimate was .061 (95% CI, 0.044–0.076), the WRMR was .855, and the SRMR was .078. The CFI and TLI were .983 and .981, respectively.

For the measurement invariance analyses, the sample was split into children <10 years old ($n = 43$) and those ≥10 years old ($n = 61$). Configural, metric, and scalar invariance were not rejected. The configural (RMSEA = .063, CFI = .984), metric (RMSEA = .062, CFI = .984), and scalar (RMSEA = .057, CFI = .985) models all had good fit. The $\Delta\chi^2$ and ΔCFI both indicated that the metric invariance restrictions did not worsen model fit relative to the configural model [$\Delta\chi^2(19) = 25.67$, $p = .14$; $\Delta\text{CFI} = 0$] nor did the scalar model worsen fit relative to the metric model [$\Delta\chi^2(51) = 44.22$, $p = .74$; $\Delta\text{CFI} = +.001$].

Preliminary normative subscale and total score data for PANS patients are shown in Table II. The mean total score in the sample was 36.7 ± 19.8, and the subscale with the highest score was Time Dependency (mean score of 10.1 ± 4.6).

Demographic data were missing for 25 of 104 participants (24%); external validity analyses were performed in the remaining 79 participants (see Table III). The subset with a completed demographic form did not differ from those without the full demographics form on age [$F(1, 102) = 0.29$, $p = .59$], but they were more likely to be male (66 vs. 40%, $\chi^2 = 5.26$, $p = .02$). For several demographic variables, there was not enough variability in the sample to assess its relationship with the CBI. Only 7 of 79 patients (9%) identified as non-White. Regarding family variables, 10 of 79 patients (13%) had caregivers who were not married and 10 of 79 patients (13%) had no siblings. Thus, the final set of external validity variables included those relevant to school/work and socioeconomic status.

Generally, the school/work variables were positively related to CBI total score. Parents of children

Table I. *Standardized Factor Loadings (With SE) of Second-Order Confirmatory Factor Analysis (N = 104)*

Item	Time dependency	Development	Physical health	Emotional health	Social relationships
Help to perform daily tasks	0.89 (0.04)				
(S)he is dependent on me	0.83 (0.04)				
Watch constantly	0.78 (0.05)				
Needs help with basic functions	0.81 (0.04)				
No break from chores	0.96 (0.04)				
I am missing out on life		0.93 (0.02)			
I wish I could escape		0.87 (0.03)			
Social life has suffered		0.94 (0.02)			
I feel emotionally drained		0.91 (0.02)			
I expected different		0.92 (0.02)			
Not getting enough sleep			0.85 (0.04)		
My health has suffered			0.97 (0.02)		
Caregiving has made me sick			0.90 (0.03)		
I am physically tired			0.90 (0.04)		
I feel embarrassed				0.89 (0.03)	
I feel ashamed of my child				0.85 (0.05)	
I resent my child				0.84 (0.06)	
Uncomfortable when friends over				0.87 (0.04)	
Angry about child interactions				0.89 (0.04)	
Do not get along/family members					0.92 (0.03)
Caregiving not appreciated					0.84 (0.04)
Marriage problems					0.91 (0.03)
Do not get along/others					0.80 (0.05)
Resentful relatives do not help					0.95 (0.03)
Loading on second-order total score factor	0.64 (0.06)	0.97 (0.02)	0.76 (0.05)	0.77 (0.05)	0.84 (0.04)

Note. Loadings are standardized, meaning that they may be interpreted similarly to a correlation coefficient. All loadings, $p < .0001$.

Table II. *Preliminary Normative Data for the CBI in PANS Patients, Drawn From the Stanford PANS Clinic (N = 104)*

Subscale or total score	Possible score	*M* ± *SD*	Median	Interquartile range
Time dependency	0–20	10.1 ± 4.6	11	7–13
Development	0–20	9.7 ± 6.1	10	4–14
Physical health	0–16	6.6 ± 4.5	6	3–10
Emotional health	0–20	5.1 ± 4.4	5	1–8
Social relationships	0–20	5.1 ± 5.1	5	0–8
Total score	0–96	36.7 ± 19.8	37	19–53

Note. CBI = Caregiver Burden Inventory; PANS = Pediatric Acute-onset Neuropsychiatric Syndrome.

who had been forced to switch schools because of their illness reported significantly higher scores than those who did not (Cohen's $d = 0.75$, 95% CI 0.28–1.22), as did parents who reported child missing 1+ days of school per week, on average, because of PANS symptoms ($d = 0.53$, 95% CI 0.07–0.99). Parents who reported that the mother reduced her work hours because of the child's illness reported significantly higher scores than those who did not ($d = 0.65$, 95% CI 0.10–1.20). While the mean score among parents of children who had been hospitalized because of PANS was slightly higher than those who had not been, this difference was not statistically significant ($d = 0.09$, 95% CI −0.53 to 0.71). As shown in Table III, the sample had relatively high socioeconomic status (SES). No parental SES variable was associated with CBI total score.

The total score of patients diagnosed with PANS only ($M = 39.4$, $SD = 19.6$) was slightly higher than the total score of patients also diagnosed with PANDAS ($M = 34.4$, $SD = 20.0$); however, the difference was not significant ($d = 0.23$, 95% CI −0.18 to 0.65, $p = .2$).

Discussion

Caregiver burden is clinically relevant in pediatric disease because burden can cause negative health outcomes for both caregiver and care receiver (Cousino & Hazen, 2013). The CBI is well validated in adult populations, but it had not been validated in a pediatric condition until a recent study in Rett Syndrome (Caserta, Lund, & Wright, 1996; Lane et al., 2017; Marvardi et al., 2005; McCleery, Addington, & Addington, 2007).

This study sought to validate the CBI in a PANS population. In assessing this work, Holmbeck and Devine's five-item checklist for development of a new

Table III. *External Validity Analyses (n = 79)*

Variable		*n*	%	CBI total score($M \pm SD$)	ANOVA
Changed schooling arrangement[a]	No	31	39	29.8 ± 19.9	$F(1, 76) = 10.18, p = .002$
	Yes	47	59	43.1 ± 16.7	
School disruption[b]	No	32	41	32.2 ± 20.4	$F(1, 75) = 5.19, p = .03$
	Yes	45	57	42.0 ± 17.4	
Psychiatric hospitalization	No	64	81	37.6 ± 19.3	$F(1, 74) = 0.09, p = .8$
	Yes	12	15	39.3 ± 15.0	
Mother reduced work hours	No	17	22	28.4 ± 21.1	$F(1, 76) = 5.52, p = .02$
	Yes	61	77	40.4 ± 17.8	
Primary caregiver	Mother	34	43	39.3 ± 18.4	$F(1, 77) = 0.44, p = .5$
	Mother and father	45	57	36.4 ± 19.6	
Childcare services[c]	No	41	52	37.7 ± 18.5	$F(1, 77) = 0, p = 1.0$
	Yes	38	48	37.6 ± 19.9	
Maternal Education	Up to college grad.	39	49	37.2 ± 20.1	$F(1, 76) = 0.07, p = .8$
	Advanced grad./prof.	39	49	38.4 ± 18.4	
Annual Income	$200K+	38	48	40.9 ± 17.4	$F(1, 70) = 0.39, p = .5$
	<$200K	34	43	38.4 ± 17.4	

[a] A common example of changing schooling arrangement is switching from traditional school to homeschool. Other examples include switching classes within a school or developing an Individualized Education Program.

[b] Defined as missing 1+ days of school per week, on average, because of PANS.

[c] Defined as parents using childcare services, including nannies, after school programs, extended family, and formal respite care.

Note. Some missing data, % do not all sum to 100. ANOVA = analysis of variance; CBI = Caregiver Burden Inventory; PANS = Pediatric Acute-onset Neuropsychiatric Syndrome.

measure is useful (Holmbeck & Devine, 2009). Although we did not develop the CBI, this checklist allows us to assess the evidence base for using the measure in a population of patients diagnosed with PANS/PANDAS.

The first item of the checklist is establishing a scientific need for the measure, which is especially important for the present study, in which an existing measure is applied to a new population. Caregiver burden is clinically relevant in PANS/PANDAS, an acute-onset neuropsychiatric disease. Previous research has found that brain-related conditions are more highly associated with greater familial distress than are other chronic conditions (Holmes & Deb, 2003). Specifically, OCD tends to lead to high caregiver burden (Storch et al., 2009). PANS is a brain-related condition with severe OCD symptoms. OCD and other neuropsychiatric symptoms cause suffering and interfere with a child's ability to complete tasks of daily living, requiring the caregiver to devote more resources to maintain family function. Past research also shows that families of patients with unstable courses of disease are more likely than those with stable courses to report that they needed respite services in the past 12 months (Nageswaran, 2009). PANS typically follows an unstable course (Frankovich et al., 2015). Clinical observations consistently reveal that while symptoms improve or remit with treatment, parents' relief is tempered by fear and hypervigilance that the symptoms will recur. Further, the disease is rare and controversial, which may make caregivers feel more isolated (Chang, Koplewicz, & Steingard, 2015). Families have reported, as well, that their social support systems and schools may be unaware of or disbelieving about the nature, impact, and useful treatment strategies for the relatively newly described illness, making caregivers feel doubted and thwarted, even abandoned.

While we could not "build in" content validity to the measure development process, as suggested by the second item of the checklist, we did assess the CBI for content validity in this new pediatric care receiver population. There are obvious differences between the adult care receiver population and the pediatric care receiver population; for example, the caregiver–care receiver relationship may be fundamentally different, so the caregiver may perform different tasks. Caregiver demographics may differ. However, based on our initial evaluation of the scale, we felt that it had sufficient content validity for application in the pediatric care population. In this study, we focused on the third checklist item, which involves testing the hypothesis of content validity. Our confirmatory factor analysis provided evidence that the original five-factor solution (Novak & Guest, 1989) was valid for use in this pediatric population. Further, measurement invariance analyses demonstrated that the psychometric properties of the scale do not depend on the age of the child; it is valid for use in younger and older children. As described above, we felt that the face validity of the CBI was sufficient for use in this population without modification; however, other investigators have come to different conclusions. In a recent study of the CBI in children with Rett syndrome, the investigators felt that the "Developmental Burden" items were inappropriate and excluded them from the outset (Lane et al., 2017). So, while they did not empirically test the question of whether the five-factor solution was valid for use in Rett syndrome, at least one study provides an alternative formulation in another pediatric sample.

Finally, in this study, we began to address the fourth checklist item, which is establishing external validity. We found that greater caregiver burden was associated with several variables, including variables measuring disruptions in work and school. These data support the convergent validity of the instrument. We did not find significant associations with some SES variables, including family income and maternal education, as we hypothesized. However, the overall SES of this sample was homogenous and high, and not reflective of the broader pediatric population. The impact of SES on caregiver burden may be diminished at higher levels. However, our ability to properly evaluate the external validity of the CBI in this sample was limited by our ability to include only 76% of participants in these analyses. The design of this study was such that we were unable to address the final item of Holmbeck and Devine's checklist—assessing responsiveness to treatment or diagnostic utility—but future work in this population should address these important questions.

The results of this study demonstrate the excellent factorial validity of the CBI when used in patients with PANS/PANDAS, and the invariance of these properties across the age range. Our findings may be generalizable to other populations of patients with PANS/PANDAS, especially as our population is similar in age and gender distribution to past reports of patients with PANS/PANDAS, which report an early age of onset and a disproportionate number of male patients (Swedo et al., 1998). Given this evidence, we propose that the CBI can be used confidently as a measure of caregiver burden in patients with PANS/PANDAS between ages 4 and 18 years.

This research also supports the hypothesis that PANS caregivers experience high burden. A common threshold for defining high burden in adult care receiver populations is a CBI score of 36 (Lund et al., 2006); although further work is needed to understand whether this cutoff is valid for use in a pediatric care receiver population, comparison is still useful. The median score in this sample was 37, indicating that the level of burden in these families is relatively high. However, only two families reported hiring formal respite care (not including nannies, after school programs, and family members who provide respite care).

Currently, this population's respite need is not met. We did not ask families why they did not use respite care, but insurance coverage may play a role. The percentage of uninsured families who need respite but do not have it is higher than for insured families (Nageswaran, 2009). However, having insurance does not guarantee respite care. In fact, insurance coverage of respite care is low in the United States. Some insurance policies, including Medicare, pay for limited respite care for caregivers of patients who qualify for hospice. Caregivers of patients with Alzheimer's Disease would qualify (Centers for Medicare and Medicaid Services, n.d.). To our knowledge, no PANS families qualify for respite care covered by insurance. However, recent action by the Center for Medicare and Medicaid Innovation (CMMI) is encouraging. CMMI is experimenting with new payment structures for pediatric care to incentivize services like respite care (Center for Medicare and Medicaid Innovation, 2017). Other funding opportunities may exist, depending on state law. For example, in some California counties, funding from the state's Mental Health Services Act is used to support caregivers of pediatric patients with mental health conditions (Southard, 2015).

These data will affect our research and clinical practice going forward. In terms of research, we plan to study longitudinal trends of CBI in our population. Longitudinal studies of caregiver burden are especially warranted both to further validate the scale and to better target interventions for decreasing caregiver burden (Adelman et al., 2014). We hypothesize that disease severity correlates highly with CBI, that CBI fluctuates as patients experience disease flares and quiescence, but that CBI tends to decrease throughout the course of a patient's time in our clinic. In clinic, we will try to find ways to decrease caregiver burden to reduce suffering in parents while improving outcomes in patients (Adelman et al., 2014). Data do suggest that family-based psychological interventions can improve outcomes for parents (Law, Fisher, Fales, Noel, & Eccleston, 2014).

Conclusions

We established the factor validity of the CBI in a PANS population, and showed that high levels of caregiver burden are reported in the Stanford PANS clinic. Interventions for PANS/PANDAS may be enhanced by including the CBI as part of routine clinical assessment and by providing targeted resources to parents where appropriate.

Supplementary Data

Supplementary data can be found at: http://www.jpepsy.oxfordjournals.org/.

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