

Short communication

Alexithymia in adolescents with chronic fatigue syndrome

Elise M. van de Putte^{a,*}, Raoul H.H. Engelbert^b, Wietse Kuis^a,
Jan L.L. Kimpfen^a, Cuno S.P.M. Uiterwaal^c

^aDepartment of Pediatrics, Wilhelmina Children's Hospital, University Medical Center Utrecht; The Netherlands

^bDepartment of Pediatric Physical Therapy and Pediatric Exercise Physiology, Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands

^cJulius Center for Health Sciences and Primary Care, University Medical Center Utrecht, The Netherlands

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Abstract

Background: Alexithymia is postulated as an important factor in the development of medically unexplained physical symptoms. Chronic fatigue syndrome (CFS) is presently medically unexplained. The aim of this study was to investigate whether the prevalence of alexithymia was higher in adolescents with CFS compared to healthy adolescents. Comorbidity such as anxiety and depression were analyzed as possible confounding factors. Secondly, alexithymia was investigated as a prognostic factor for the recovery of CFS. **Methods:** A cross-sectional study was performed among 40 adolescent outpatients diagnosed with CFS and 36 healthy controls. The 20-item Toronto Alexithymia Scale was used to assess all participants for alexithymia. Additionally, all participants completed a number of questionnaires regarding fatigue (Checklist Individual Strength), somatic complaints (Checklist Somatization Inventory), depression (Children's

Depression Inventory), and trait anxiety (Spielberger State Trait Anxiety Questionnaire). A follow-up study was performed among the CFS adolescents 1 1/2 years after the initial assessment. **Results:** CFS adolescents scored higher only on the subscale identifying feelings of the TAS-20 [mean difference after adjustment for depression and anxiety 2.8 (95% CI: 0.6; 4.9)]. Twelve CFS adolescents (30%) fulfilled criteria for alexithymia. This subgroup was characterized by higher scores for depression and anxiety and equal scores for fatigue and somatic complaints. At follow-up, no differences in recovery were established between the alexithymic and nonalexithymic CFS adolescents. **Conclusions:** Alexithymia neither appears to be a unique correlate of CFS nor to be a prognostic factor for recovery of the CFS illness.

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Introduction

Alexithymia has been defined as a deficit in cognitive processing and regulation of emotions, characterized by difficulties in describing and differentiating emotions and a cognitive style focused on external events instead of inner experience [1]. Alexithymia is considered an important risk factor for somatization [2]. Supposedly, emotions of highly

alexithymic individuals are not well represented mentally, with an ensuing tendency to focus on somatic sensations that accompany emotional arousal and to misinterpret these as signs of illness [3]. Alexithymic individuals are considered vulnerable to incorrectly attributing innocent bodily sensations to physical disease and to seeking medical care for their symptoms, for which subsequently no medical explanations can be found [4]. Chronic fatigue syndrome (CFS) is one of the medically unexplained illnesses, also designated as one of the functional somatic syndromes [5]. However, little is known about the role of alexithymia in the pathogenesis of CFS, and studies concerning alexithymia in adolescents with CFS are lacking.

* Corresponding author. KE04.133.1, University Medical Center Utrecht, PO Box 85090, 3508 AB Utrecht, The Netherlands. Tel.: +31 30 2504001; fax: +31 30 2505349.

E-mail address: e.vandeputte@umcutrecht.nl (E.M. van de Putte).

A study on alexithymia should simultaneously measure other constructs that might mediate alexithymia such as depression and anxiety [6–8].

Alexithymia was shown to be a predictor of an unfavorable outcome of adult patients with somatoform disorders, independent of other psychopathology or illness severity [9], although not confirmed in a recent study [4]. We hypothesized that adolescents with CFS are more alexithymic than healthy adolescents after adjustment for depression and anxiety. We further expected that alexithymic CFS adolescents would have a worse prognosis.

Methods

Population

A total of 70 adolescents (12–18 years old), referred with severe fatigue to the University Medical Center Utrecht in 2003 and 2004, were all examined by a paediatrician. A final diagnosis of CFS was established in 47 adolescents after medical and psychological examinations. In addition to the Centers for Disease Control and Prevention (CDC) exclusion criteria [10], patients with somatic comorbidity interfering with fatigue ($n=4$) and one patient because of severe primary depression were excluded. Two adolescents refused to participate. Of the remaining 40, 36 fulfilled all criteria for CFS of the CDC [10]. Four patients had less than 4 additional symptoms at the moment of the research examinations but were nevertheless included.

A follow-up study of 36 of these 40 CFS patients was performed 1 1/2 years after the initial study assessments (2006).

As a reference group, 102 adolescents aged 12–18 years from a secondary school were invited to participate with their parents. Families with an adoptive child or a child with a chronic illness were excluded ($n=3$). From the remaining 99 adolescents, 36 (37%) agreed to participate, including four pairs of siblings.

Instruments (questionnaires)

Alexithymia was assessed with a validated Dutch translation of the 20-item Toronto Alexithymia Scale (TAS-20), comprising three factors: difficulties identifying feelings, difficulties expressing feelings, and externally orientated thinking [11–13]. Items consist of statements presented in a five-point Likert scale (score, 1–5) along a “strongly disagree” to “strongly agree” continuum, with higher scores indicating more alexithymia. TAS-20 scores can be used dimensionally (score range, 20–100) and categorically, indicating yes or no alexithymia (score, ≥ 60 and < 60 , respectively) [14]. TAS-20 is validated for young adults (mean age, 21.5 years) and is not adapted for children.

Fatigue was assessed dimensionally with the Checklist Individual Strength (CIS-20) on fatigue in the preceding

2 weeks. There are four subscales: subjective experience of fatigue, concentration, motivation and physical activity, each item scored on a Likert scale (score, 1–7). Internal consistency is high, as is the discriminative validity for CFS [15].

Depression was measured with a validated Dutch translation of the Children’s Depression Inventory (CDI) [16,17]. CDI quantifies depressive symptoms in the past 2 weeks using 27 items rated on a three-point scale (range, 0–2).

Trait anxiety was assessed with a Dutch translation of the Spielberger State-Trait Anxiety Inventory for Children (STAIC) [18,19], consisting of 20 statements on a three-point scale that assess the level of anxiety a person reports as generally characteristic of himself.

Somatic complaints were assessed with a validated Dutch translation of the Children’s Somatization Inventory (CSI), rating the presence of each of 35 somatic symptoms in the preceding 2 weeks using a five-point Likert scale ranging from “not at all” to “a whole lot” (range, 0–4) [20,21].

All questionnaires were completed individually in, on average, 30 min and in separate rooms in a university building in May to September 2004.

The medical ethics committee of the University Medical Center Utrecht approved the study. Written informed consent was obtained from both adolescents and parents.

Outcome measures in the follow-up study

Questionnaires (CIS-20 and Child Health Questionnaire) were filled out at home in January 2006. The score on the subscale subjective fatigue of the CIS-20 was chosen as the major outcome variable in the follow-up of CFS cases. A cutoff score was set, (mean plus 2 S.D. of subjective fatigue distribution in healthy adolescents), to dichotomize outcome as recovery (< 40) or nonrecovery (≥ 40). This outcome measure was combined with the health perception question from the Child Health Questionnaire [22]. “My health is ‘excellent,’ ‘very good,’ or ‘good’” were all classified as “recovery” (in good health). My health is “moderate” or “bad,” were both classified as “nonrecovery” (in poor health). Final recovery classification required a positive score in both categories.

Analysis

Of the relevant variables, group-specific means and standard deviations were calculated.

We used linear regression with the variable of interest as dependent and a group indicator as independent variable to explore group differences. Results are presented as coefficients representing mean differences between CFS adolescents and healthy controls with 95% confidence intervals. The same models were used to adjust for possible confounding factors as age, gender, anxiety, and depression.

CFS adolescents were classified as alexithymia (yes/no) by the TAS-20 score (≥ 60 =alexithymic). These groups were described by mean values of depression, anxiety, somatic

Table 1
Characteristics of adolescents with CFS and healthy controls

	CFS (<i>n</i> =40) [mean (S.D.)]	Healthy (<i>n</i> =36) [mean (S.D.)]	Difference (95% CI)	<i>P</i> value
Mean symptom duration (months)	23.4 (11.3)	NA		
Age (years)	16.0 (1.5)	16.8 (1.4)	−0.8 (−1.5 to −0.1)	.019
Gender (% girls)	78	67	11 (−10 to 31)	.298
Intact families (%)	82	93	−9 (−27 to 5)	.185
Fatigue assessment				
Total score CIS-20 (20 items; 20–140)	101.8 (17.8)	48.0 (18.8)	53.9 (45.5–62.2)	<.001
Score subjective fatigue subscale (8 items; 8–56)	46.9 (7.4)	19.4 (10.0)	27.5 (23.5–31.5)	<.001
Somatic complaints (CSI 35 items; 0–140)	35.6 (20.1)	13.0 (9.3)	22.6 (15.5–29.7)	<.001
Health concerns (CHQ) (11 items; 11–55)	39.8 (5.7)	22.6 (7.3)	17.2 (14.2–20.2)	<.001
Psychological adjustment				
Anxiety disposition (STAIC; 20 items; 20–60)	36.9 (7.8)	30.3 (6.4)	6.6 (3.3–9.9)	<.001
Depression disposition (CDI; 27 items; 0–54)	11.7 (6.1)	5.6 (4.4)	6.1 (3.7–8.5)	<.001

CHQ, Child Health Questionnaire.

complaints, and outcome, and differences were analysed with independent-samples *t* tests.

The association between alexithymia and outcome at follow-up in the CFS adolescents was quantified through odds ratios using logistic regression, with outcome (yes/no) as dependent and the TAS-20 score as independent variable.

Internal consistency of the TAS-20 was determined with the intraclass correlation coefficient.

In all analyses, the significance level was set at $P < .05$ (two-tailed tests).

Results

Table 1 shows a similar gender distribution in both adolescent groups, but the CFS adolescents were significantly younger (16 vs. 16.8 years), for which was accounted by adjustment for age.

Evidently, the CFS patients showed a higher score on the CIS-20 than the healthy adolescents. Somatic complaints, anxiety, and depression were more prominent in the CFS group. The mean CDI score in the CFS adolescents (11.7) was below that for a depressive disorder, which is 22.8 in a reference sample [23].

Table 2 shows the alexithymic features of both groups of adolescents. The CFS adolescents scored higher on the TAS-20, in particular, the subscales identifying feelings and expressing feelings. Adjusting for depression and anxiety

attenuated the differences, and only the subscale identifying feelings remained statistically significant. The internal consistency of the total TAS-20 was good (intraclass coefficient, 0.79). A proportion of 30% ($n=12$) fulfilled criteria for alexithymia (TAS-20 score ≥ 60). One healthy adolescent was alexithymic.

The 12 CFS adolescents with alexithymia were compared with the 24 CFS adolescents without alexithymia. The scores on the CIS-20 (subscales subjective fatigue and somatic complaints) were identical for both groups. However, anxiety and depression were more prevalent in the CFS adolescents with alexithymia.

At follow-up, 17 (47%) of the 36 CFS adolescents fulfilled criteria for full recovery. The presence of alexithymia was not a risk factor for non-recovery (odds ratio, 1.027; 95% CI, 0.96–1.10; $P=.465$).

Discussion

CFS adolescents exhibit more alexithymic features than healthy adolescents; however, this finding is mediated by depression and anxiety. The 30% who fulfilled criteria for alexithymia are characterized by higher anxiety and depression scores. Alexithymic features seemed associated with the cluster of fatigue, somatic symptoms, anxiety, and depression both in healthy adolescents and adolescents with CFS, rather than with the main symptom of CFS—fatigue.

Table 2
Alexithymic features of adolescents with CFS and healthy controls

	CFS (<i>n</i> =40) [mean (S.D.)]	Healthy (<i>n</i> =36) [mean (S.D.)]	Difference (95% CI)	<i>P</i> value	Adjusted difference ^a (95% CI)	<i>P</i> value
Alexithymia						
Sum score TAS-20 (20 items; 20–100); RC = 0.79	54.1 (9.9)	44.3 (7.8)	9.8 (5.7; 13.9)	<.001	3.8 (−0.5; 8.1)	.081
Subscale identifying feelings (7 items; 7–35); RC = 0.83	19.2 (5.8)	12.7 (4.1)	6.5 (4.2; 8.7)	<.001	2.8 (0.6; 4.9)	.012
Subscale expressing feelings (5 items; 5–25); RC = 0.77	13.7 (4.5)	10.9 (3.3)	2.8 (0.9; 4.6)	.003	1.4 (−0.8; 3.5)	.205
Subscale externally oriented thinking (8 items; 8–40); RC = 0.54	21.2 (4.4)	20.6 (3.9)	0.5 (−1.4; 2.5)	.576	−0.3 (−2.3; 1.7)	.763

RC, reliability coefficient, Cronbach's α .

^a Adjusted for age, gender, anxiety, and depression.

The association between alexithymic features and this cluster of symptoms might be due to an overlap between the measurements of alexithymia, anxiety, and depression, leading to an imperfect differentiation between the three phenomena [24]. Alexithymia does not appear to be a unique correlate of CFS. Depression and anxiety partly explained and mediated its relationship with CFS. Neither is alexithymia a predictor for recovery of CFS.

Limitations and strengths of this study

Our study is, to our knowledge, the first on the meaning of alexithymia in CFS in adolescence. We could explicitly control for confounding factors such as depression and anxiety. Some aspects need to be considered in further interpretation. Having recruited CFS adolescents from a tertiary facility may hamper generalization to all adolescents with CFS. There may have been selection bias in the control group, in particular, in the light of the low response rate, but we consider it unlikely that the association between fatigue and alexithymia differs between responders and non-responders. The TAS-20 total score of our control group was similar to previously published Dutch data from 519 slightly older adolescents [13]. Alexithymia was assessed with a self-report TAS-20 questionnaire, which requires at least some ability to identify feelings.

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References

- [1] Taylor GJ. Alexithymia: concept, measurement, and implications for treatment. *Am J Psychiatry* 1984;141:725–32.
- [2] Grabe HJ, Spitzer C, Freyberger HJ. Alexithymia and personality in relation to dimensions of psychopathology. *Am J Psychiatry* 2004;161:1299–301.
- [3] Taylor GJ, Bagby RM. New trends in alexithymia research. *Psychother Psychosom* 2004;73:68–77.
- [4] Kooiman CG, Bolk JH, Rooijmans HG, Trijsburg RW. Alexithymia does not predict the persistence of medically unexplained physical symptoms. *Psychosom Med* 2004;66:224–32.
- [5] Wessely S, Hotopf M, Sharpe M. *Chronic fatigue and its syndromes*. New York: Oxford University Press, 1999.
- [6] Honkalampi K, Koivumaa-Honkanen H, Tanskanen A, Hintikka J, Lehtonen J, Viinamaki H. Why do alexithymic features appear to be stable? A 12-month follow-up study of a general population. *Psychother Psychosom* 2001;70:247–53.
- [7] Hintikka J, Honkalampi K, Lehtonen J, Viinamaki H. Are alexithymia and depression distinct or overlapping constructs?: a study in a general population. *Compr Psychiatry* 2001;42:234–9.
- [8] Saarijarvi S, Salminen JK, Toikka T. Temporal stability of alexithymia over a five-year period in outpatients with major depression. *Psychother Psychosom* 2006;75:107–12.
- [9] Bach M, Bach D. Predictive value of alexithymia: a prospective study in somatizing patients. *Psychother Psychosom* 1995;64:43–8.
- [10] Fukuda K, Straus SE, Hickie I, Sharpe MC, Dobbins JG, Komaroff A. The chronic fatigue syndrome: a comprehensive approach to its definition and study. International Chronic Fatigue Syndrome Study Group. *Ann Intern Med* 1994;121:953–9.
- [11] Bagby RM, Parker JD, Taylor GJ. The twenty-item Toronto Alexithymia I. Scale-item selection and cross-validation of the factor structure. *J Psychosom Res* 1994;38:23–32.
- [12] Bagby RM, Taylor GJ, Parker JD. The Twenty-Item Toronto Alexithymia Scale-II. Convergent, discriminant, and concurrent validity. *J Psychosom Res* 1994;38:33–40.
- [13] Kooiman CG, Spinhoven P, Trijsburg RW. The assessment of alexithymia: a critical review of the literature and a psychometric study of the Toronto Alexithymia Scale-20. *J Psychosom Res* 2002;53:1083–90.
- [14] Kooiman CG, Bolk JH, Brand R, Trijsburg RW, Rooijmans HG. Is alexithymia a risk factor for unexplained physical symptoms in general medical outpatients? *Psychosom Med* 2000;62:768–78.
- [15] Vercoulen JH, Swanink CM, Fennis JF, Galama JM, van der Meer JW, Bleijenberg G. Dimensional assessment of chronic fatigue syndrome. *J Psychosom Res* 1994;38:383–92.
- [16] Kovacs M. The Children's Depression Inventory (CDI). *Psychopharmacol Bull* 1985;21:995–8.
- [17] Timbremont B, Braet C. Psychometrische evaluatie van de Nederlandstalige Children's Depression Inventory. *Gedragstherapie* 2001;34:229–42.
- [18] Papay JP, Spielberger CD. Assessment of anxiety and achievement in kindergarten and first- and second-grade children. *J Abnorm Child Psychol* 1986;14:279–86.
- [19] Houtman IL, Bakker FC. The anxiety thermometer: a validation study. *J Pers Assess* 1989;53:575–82.
- [20] Walker LS, Garber J, Greene JW. Somatization symptoms in pediatric abdominal pain patients: relation to chronicity of abdominal pain and parent somatization. *J Abnorm Child Psychol* 1991;19:379–94.
- [21] Meesters C, Muris P, Ghys A, Reumerman T, Rooijmans M. The Children's Somatization Inventory: further evidence for its reliability and validity in a pediatric and a community sample of Dutch children and adolescents. *J Pediatr Psychol* 2003;28:413–22.
- [22] Raat H, Landgraf JM, Bonsel GJ, Gemke RJ, Essink-Bot ML. Reliability and validity of the child health questionnaire-child form (CHQ-CF87) in a Dutch adolescent population. *Qual Life Res* 2002;11:575–81.
- [23] Timbremont B, Braet C, Dreessen L. Assessing depression in youth: relation between the Children's Depression Inventory and a structured interview. *J Clin Child Adolesc Psychol* 2004;33:149–57.
- [24] Marchesi C, Fonto S, Balista C, Cimmino C, Maggini C. Relationship between alexithymia and panic disorder: a longitudinal study to answer an open question. *Psychother Psychosom* 2005;74:56–60.