

Spasmodic torticollis – a multicentre study on behavioural aspects IV: psychopathology

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The GSI (General Symptom Index) of the Symptom Checklist 90 R (SCL 90 R) (as a global indicator of the severity of psychiatric symptoms) of 27% of the spasmodic torticollis (ST) sample fell outside the 95% range of the normal control group (two standard deviations). Patients with a higher GSI were younger, more functionally disabled and subject to higher psychosocial stress due to the illness. The highest scores were reached on the subscales of somatization, interpersonal sensitivity and depression. On the depression scale, 23% of the patients' scores were abnormal. This scale correlated significantly with the neurological signs, particularly the TSUI-index and laterocollis. A statistically significant correlation also existed between psychiatric morbidity and a family history of mental disorder. More than 50% of the patients reported that stressful life events had triggered their illness. In order of frequency, a death came first, followed by marital strife, changes in employment and family arguments. The data suggest that psychopathology in ST should generally be considered as a result of a variety of interacting factors, biological, psychological and social.

Keywords: Spasmodic torticollis – Psychosocial adjustment – Life events psychopathology – Coping

INTRODUCTION

In this paper we report on psychopathological symptoms and their interrelations with the neurological signs, the neurological symptoms and psychosocial changes in spasmodic torticollis (ST). In particular one of the questions outlined in the first paper in this series (Scheidt *et al.*, 1996) will be addressed, namely how do psychopathological symptoms in ST compare to a normal control group or to other clinical samples taken from the literature, and how is psychopathology related to the neurological syndrome and to psychosocial disability?

In a series of thorough studies Jahanshahi and Marsden (1988a,b; 1990a,b) and Jahanshahi (1991) compared psychopathology in ST and cervical spondylosis. The prevalence of psychiatric disorders in both groups was relatively high: 27.1% of the torticollis group and 26.5% of the cervical spondylosis group had either a premorbid psychiatric history or concurrent psychiatric symptomatology (see Scheidt *et al.*, 1996). Compared with each other the two groups did not differ with regard to psychiatric

morbidity, but the ST patients had significantly higher mean scores on the Beck Depression Inventory (Jahanshahi and Marsden, 1988a). Self-referent negative cognitions such as self-blame, self-accusation, self-punitive thoughts and a negative body image emerged as the prominent components of depression (Jahanshahi, 1991). These results suggest an interplay of the neurological disorder with self-perception and coping as an important determinant of psychopathology (see Nickel *et al.*, 1996). Close associations between signs, symptoms, psychosocial changes and psychopathology can be expected according to this hypothesis.

The prevalence rates of psychopathology reported in the present paper refer to a patient population in which 70% of the patients were undergoing treatment with botulinum toxin (see Heinen *et al.*, 1996). Treatment with botulinum toxin improves the neurological condition in a substantial number of patients (Blackie and Lees, 1990; Lorentz *et al.*, 1991; Moore and Blumhardt, 1991). Follow-up studies of treatment

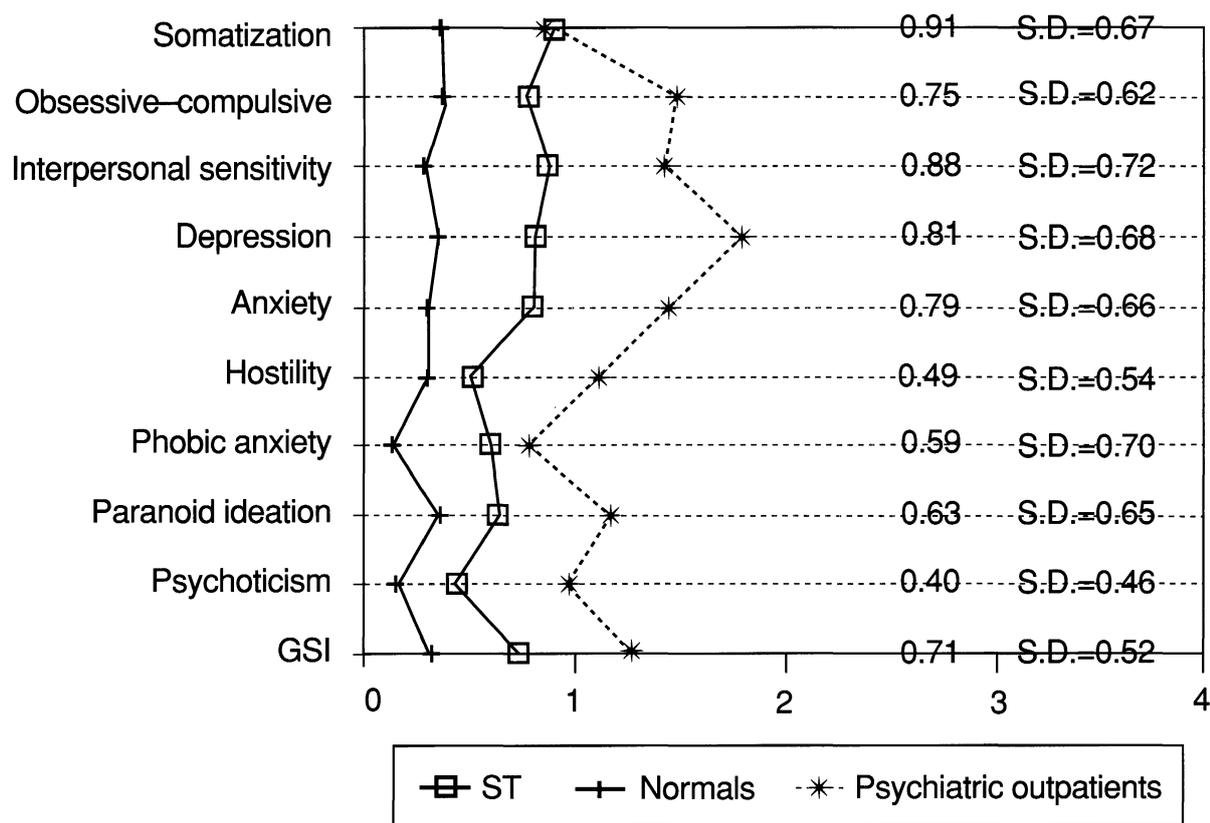


FIG. 1. SCL 90 R mean scores and standard deviations of ST patients compared to two reference groups.

programmes with botulinum toxin comparing pre-treatment and post-treatment levels of psychopathology are rare. The few studies available show that botulinum toxin improves not only the neurological condition but also the degree of psychological distress (Knoll *et al.*, 1992; Jahanshahi and Marsden, 1992). However the results of the study presented illustrate that, in about a third of the patients, considerable psychological distress persists even under treatment with botulinum toxin.

RESULTS

Prevalence of psychopathological symptoms

On all but one subscale of the Symptom Checklist 90 R (SCL 90 R), ST patients scored in the range between the two reference groups, normals and psychiatric outpatients (Fig. 1).

The mean general symptom index (GSI) of the ST patients was 0.72 (SD = 0.52) compared to 0.31 (SD = 0.31) in the normal controls and 1.26 (SD = 0.68) in the psychiatric outpatient group taken from the literature (Derogatis, 1986). Of the

ST patients 27% had a GSI higher than two standard deviations above the normal group. Fifteen per cent of the ST patients scored more highly than the average of the psychiatric outpatients.

The mean scores on the subscales of the SCL 90 R are detailed below.

The highest mean scores were reached on scale 1, *somatization*. This scale consists of items referring to dysfunctions of the body such as headache, dizziness, heart and chest pain and backpain. The mean score of the ST sample on this scale was very similar to that of the psychiatric reference group (0.91, SD = 0.67 in ST versus 0.87, SD = 0.75 in the psychiatric group). Fifty-seven per cent of the ST patients scored higher than the average of the psychiatric group. The scores of the ST group on the somatization scale did not seem to be contaminated by the neurological signs. Pearson correlation coefficients between the TSUI-index, rotation, laterocollis or antero-retrocollis and the somatization scale of the SCL 90 R were not significant.

The mean score on the scale *interpersonal sensitivity* ranked second behind somatization. Items of this scale refer to feelings of social insecurity such as

TABLE I. Correlations between psychopathology (SCL 90 R), signs (neurological assessment), symptoms (self-reported) and coping (Freiburger Questionnaire for Coping; FKV)

	Somatization	Obsessive-compulsive	Interpersonal sensitivity	Depression	Anxiety	GSI
Age	0.03	0.00	-0.16**	-0.06	-0.10	-0.10
Age at onset	0.07	-0.04	-0.21**	-0.05	-0.14*	-0.10(*)
Duration	-0.02	0.10	0.10	0.01	0.08	0.06
Neurological signs						
TSUI-index	0.13(*)	0.16*	0.04	0.19**	0.05	0.13*
Rotation	0.02	-0.01	0.00	-0.02	-0.01	-0.01
Laterocollis	0.06	0.17*	0.05	0.14*	-0.03	0.11
Antero-retrocollis	0.07	0.03	0.10	0.01	0.01	0.04
Neurological symptoms						
Deviation head	0.36***	0.27***	0.30***	0.30***	0.24***	0.32***
Muscle tension	0.44***	0.30***	0.29***	0.39***	0.31***	0.39***
Tremor of head	0.31***	0.18**	0.29***	0.26***	0.44***	0.33***
Pain	0.46***	0.20***	0.20***	0.34***	0.29***	0.34***
Coping						
Depressive	0.38***	0.61***	0.64***	0.68***	0.59***	0.68***
Active problem focused	0.26***	0.17**	0.13*	0.13*	0.16**	0.20**
Wishful thinking	0.22***	0.37***	0.31***	0.34***	0.20***	0.34***

* $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$. High correlations were found between the subscales of the SCL 90 R and the neurological symptoms and coping, whereas the correlations with the neurological signs are weak.

shyness towards the other sex, not feeling understood or appreciated by others and feeling self-conscious in relation to others. The mean score of the ST patients on this scale was 0.90 (SD = 0.72) compared to 0.29 (SD = 0.39) in the normal control and to 1.41 (SD = 0.89) in the psychiatric group. Thirty-three per cent of the ST patients scored higher than two standard deviations above the normal mean. For this scale no significant correlations emerged with the neurological signs (TSUI-index and the deviation of the head in different planes) (Table I). However the scale correlated negatively with age and age at onset ($r = -0.16$, $r = -0.21$, $p < 0.01$).

On the *depression scale*, 23% of the ST patients scored more than two standard deviations above the normal mean. The mean score was 0.82 (SD = 0.68) compared to 0.36 (SD = 0.44) in the normal control group and 1.79 (SD = 0.94) in the psychiatric group. The depression scale correlated significantly with the neurological signs, particularly the TSUI-index and laterocollis (Table I). Although the correlations are weak, there is a discernible trend pointing to a *relationship between depression and salient rotation*.

The mean value on scale 5, *anxiety*, ranked fourth. Items of this scale describe symptoms of manifest anxiety including somatic equivalents such as tension and restlessness, and cognitive components of anxiety such as worry and anxious anticipations. The mean score of the ST patients on this scale was 0.79 (SD =

0.66) compared to 0.30 (SD = 0.37) in the normal control group and 1.47 (SD = 0.88) in the psychiatric group. Twenty-eight per cent of the ST patients scored outside the normal range (mean \pm 2 SD). Fifteen per cent exceeded the mean score of the psychiatric group. No correlations emerged with the neurological signs.

In summary, the GSI of the SCL 90 R indicates that barely one-third (27%) of the torticollis patients exhibit more obvious psychological symptoms. For about a quarter to a third, values were slightly elevated on the subscales of somatization, interpersonal sensitivity, depression and anxiety.

In the following sections we report on some aspects of psychopathology in detail and discuss the correlations with other clinical features.

ST patients with a high GSI

To test whether or not ST patients with an abnormal GSI differ from the rest of the group in other features, chi-square-tests were used (see Scheidt *et al.*, 1996).

These analyses yielded statistically significant differences for the following variables: age ($p < 0.05$), psychosocial distress (factor 1) ($p < 0.001$), functional disability (factor 2) ($p < 0.01$), illness triggered by stressful life events ($p < 0.01$) and presence of other dystonic symptoms ($p < 0.05$).

The direction of the differences indicated that patients with a high GSI are younger (on the average 4.3 years), are subject to greater psychosocial stress

TABLE II. Correlations between psychosocial changes (factor 1 illness-related psychological distress and factor 2 functional disability), signs (neurological assessment), symptoms (self-reported), coping (FKV) and psychopathology (SCL 90 R)

	Illness-related psychological distress	Functional disability
Age	-0.13*	0.08
Age at onset	-0.18**	0.17**
Duration	0.09	-0.10
Neurological signs		
TSUI-index	0.03	0.21**
Rotation	-0.04	-0.12*
Laterocollis	0.01	0.22**
Antero-retrocollis	-0.04	0.28***
Neurological symptoms		
Deviation head	0.34***	0.54***
Muscle tension	0.36***	0.55***
Tremor of head	0.38***	0.06
Pain	0.26***	0.48***
Coping		
Depressive	0.64***	0.12*
Active problem focused	0.07	0.21**
Wishful thinking	0.30***	0.02
Self-distraction	-0.05	0.13*
Religiosity	0.04	0.20**
Psychopathology		
Somatization	0.34***	0.31***
Obsessive-compulsive	0.57***	0.13*
Interpersonal sensitivity	0.66***	0.11*
Depression	0.63***	0.26***
Anxiety	0.53***	0.16**
Hostility	0.49***	0.11(*)
Phobic anxiety	0.48***	0.26***
Paranoid ideation	0.49***	0.02
Psychoticism	0.55***	0.10
GSI	0.65***	0.23***

* $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$. Functional disability correlated more highly with the neurological signs and the neurological symptoms, whereas illness-related psychological distress correlated more highly with psychopathology and coping (depressive coping and wishful thinking).

due to the illness and are more limited functionally. They more frequently listed stressful life events as triggers of the illness, suffered more frequently from additional dystonic symptoms and more often had a history of mental disorder in their family.

No significant differences were noted for sex, social status (retirement), length and severity (TSUI-index) of the illness or course (symptom-free intervals).

Family history of mental disorder

Of the total sample, 13.1% reported first degree relatives (parents or siblings) with a severe mental

disorder. As mentioned above, a statistically significant correlation existed between psychiatric morbidity and a family history of mental disorder.

However, the remaining variables (age, sex, TSUI-index, social status) did not differ significantly between those with a history of mental disorder in the family and the rest of the group.

Life events as trigger

Of the patients, 53.5% ($n = 137$) reported that stressful life events had triggered their illness. In order of frequency, a death came first ($n = 40$, 15.6% of the total sample), followed by marital strife, changes in employment and family arguments. Women outnumbered men in attributing their illness to psychosocial stress ($p < 0.01$). The group whose illness was preceded by a stressful life event demonstrated significantly higher measures of psychological symptoms ($p = 0.05$).

Interrelations of psychopathology and psychosocial changes

Psychosocial changes have been reported in detail in the accompanying paper (Nickel *et al.*, 1996). Here we will discuss interrelations between psychopathology and psychosocial changes referring to the two factors, psychological distress due to the illness (factor 1) and functional impairment (factor 2) (for method see Scheidt *et al.*, 1996).

Table II shows the correlations of the two factors with age, age at onset, duration, neurological signs, neurological symptoms, coping and psychopathology. Factor 1 correlates consistently with all subscales of the SCL 90 R. Factor 2 correlates with the subscales somatization, depression, phobic anxiety and the GSI. Correlations of the various subscales of the SCL 90 R with factor 1 are consistently higher than with factor 2. Factor 2, however, in contrast to factor 1, correlates significantly with the neurological signs.

Psychopathology and coping

The two coping styles which are highly interrelated with psychopathology are depressive coping ($r = 0.64$, $p < 0.001$) and wishful thinking ($r = 0.30$, $p < 0.001$). Depressive coping refers to a set of cognitive and emotional attitudes towards the illness such as blaming oneself, feeling guilty, etc. Wishful thinking describes a tendency towards denial of the illness and escapism into fantasy.

As can be seen from Table I, depressive coping is highly interrelated with almost all subscales of the SCL 90 R and with the GSI (Pearson correlation coefficients ranging from $r = 0.38$, $p < 0.001$ to $r =$

0.68, $p < 0.001$). Interrelations between wishful thinking and the subscales of the SCL 90 R tended to be lower (ranging from $r = 0.20$, $p < 0.01$ to $r = 0.37$, $p < 0.001$).

Loss of professional activity

Among the severe social consequences of ST is the loss of working capacity and with it the loss of an important social role. Twenty-three per cent of the patients in our sample had to retire because of their illness. The group of retirees differed from the rest of the sample in three variables. These were *age* ($p = 0.01$), *functional disability* ($p < 0.01$) and the *duration of the illness* ($p < 0.05$). The correlations indicated that the retired patients were on average 5.5 years older, perceived themselves as more functionally disabled (factor 2) and had been ill approximately 2.5 years longer than their counterparts.

No differences emerged with regard to sex, the TSUI-index or the psychological stress resulting from the illness (factor 1). Neither did the groups differ with regard to the GSI or either dysfunctional style of coping. This means that neither severity of the signs, nor mental stress due to the illness (factor 1), nor the psychiatric symptomatology (SCL 90 R) had any direct influence on retirement.

Psychopathology, signs and symptoms

The *neurological symptoms* correlated significantly with all subscales of the SCL 90 R included in Table I. In contrast the *neurological signs* did not correlate with the subscales of the SCL 90 R except for weak associations of TSUI-index and laterocollis with obsessive-compulsive symptoms and depression.

Additional dystonic symptoms

Thirty-four per cent of the patients had *additional dystonic symptoms* in other areas of the body. These patients are of particular interest as the prevalence of multiple dystonic symptoms in ST may confirm the concept of focal dystonia as a "forme fruste" of "dystonia musculorum deformans" (Marsden, 1976).

Patients with multiple dystonic symptoms in our study differed from the rest of the group in the following variables: *duration of illness* ($p < 0.05$), *psychological stress* due to the illness ($p < 0.05$), *functional disability* ($p = 0.001$) and *GSI* ($p = 0.001$). The variables age, sex, retirement, TSUI-index and the occurrence of remissions in the course of the illness did not differ significantly. The direction of the differences indicates that patients with multiple extrapyramidal motor symptoms had a longer duration of their illness (approximately 3 years), were

more functionally disabled (factor 2) (while just failing to reach the level of statistically significant difference for the TSUI-index), and reported more psychiatric symptoms.

The subscales of the SCL 90 R yielded significant differences between the two groups for the scales *somatization* ($p < 0.001$), *obsessive-compulsiveness* ($p < 0.05$), *interpersonal sensitivity* ($p < 0.05$), *depression* ($p < 0.05$), *anxiety* ($p < 0.01$) and *psychoticism* ($p < 0.001$). On all of these scales, patients with multiple dystonic symptoms demonstrated values higher than those of the rest of the group.

Psychopathology and course

Of the patients, 19.5% reported symptom-free intervals during the course of ST. It may be worth considering whether these patients can be distinguished from the rest of the group on any other features of their illness.

The patients with symptom-free intervals had higher scores ($p < 0.05$) on the *GSI* than the rest of the group. Other differences were found for the subscales of *psychoticism* ($p < 0.05$) and *paranoid ideation* ($p < 0.01$) indicating higher scores for the group with symptom-free intervals than the rest of the sample on these two scales. No statistically significant differences were found for the following variables: age, sex, social status (retirement), psychological stress (factor 1), functional disability (factor 2) or TSUI-index. Neither did the frequency of life-event triggers differ between the two groups.

Predictors of psychopathology

In order to investigate the predictors of psychopathology a stepwise multiple regression analysis was calculated.

All variables which were interval-scaled and had a Pearson correlation coefficient higher than 0.32 (at least 10% of shared variance) with the GSI of the SCL 90 R were included. These were: the *neurological symptoms* (deviation of the head, muscle tension, tremor of the head and pain), *coping* (depressive coping, wishful thinking and total score of coping) and *illness-related psychological distress* (factor 1). *Functional disability* (factor 2), which correlated with some of the subscales of the SCL 90 R more than with the GSI, was also included. Missing data were pair-wise excluded. The results are presented in Table III. *Depressive coping* was the most significant single predictor of psychopathology and accounted for 46.4% of the variance. Illness-related psychological distress (factor 1) and functional disability (factor 2) also entered the regression analysis and accounted for 7.5% and 3.2% of the variance respectively.

TABLE III. Multiple regression analysis with GSI as dependent variable

Step	Variable	Betaweight	r ²
1	Depressive coping	0.32	0.46
2	Factor 1	0.54	0.39
3	Factor 2	0.57	0.15
4	Coping total score	0.59	0.16

Depressive coping is the most powerful predictor of psychopathology.

However the neurological symptoms did not enter into the equation.

DISCUSSION

The mean scores of the SCL 90 R in our sample on all but one subscale ranged approximately midway between the two comparison groups. In 27% of the ST patients, the GSI lay beyond two standard deviations of the mean of the normal control group and was therefore considered to fall into the pathological range. Increased values were found particularly on the subscale *somatization*, on which the score of 57% of the patients exceeded the mean score of the psychiatric outpatients. *Interpersonal sensitivity* ranked second behind somatization. Thirty-three per cent of the ST patients on this subscale scored abnormally. On the depression scale, 23% of the ST patients scored in the pathological range. This scale correlated significantly with the neurological signs, particularly with the TSUI-index and the lateral inclination of the head. Although the correlations are weak, there is a discernible trend pointing to a relationship between depression and salient rotation (laterocollis).

Summarizing these frequencies, about a quarter to a third of the ST patients, according to their self-report of psychopathological symptoms in the SCL 90 R, fall into a range of clinical significance. This prevalence is in agreement with other studies on psychopathology in ST, which reported depressive symptoms in about a third of the patients (Jahan-shahi, 1988a, 1990a; Schulze and Stephan, 1987).

As outlined in the first paper in this series (Scheidt *et al.*, 1996), the factors to which psychopathology in ST may be attributed have been a matter of debate. On the basis of the data presented here, the following conclusions are proposed:

1. The *neurological signs* in our study correlated only weakly with psychopathology. This is true for the total sample as well as for the subgroup of patients with "severe" psychopathology (a GSI in the

pathological range). By contrast, the correlations between psychopathology and the *neurological symptoms* were statistically significant throughout.

2. According to the results of the regression analysis with psychopathology (GSI) as dependent variable, *depressive coping* and the two factors *functional disability* and *psychological distress*, accounted for 57% of variance. The neurological symptoms did not enter into the equation. This indicates that the neurological symptoms are less predictive of psychopathology in ST than coping and psychosocial changes. (Because of their low Pearson correlation coefficients with the GSI neurological signs were not included.)
3. *Depressive coping* has already been discussed in the accompanying paper (Nickel *et al.*, 1996) as a predictor of psychosocial changes. Its relevance is confirmed as a predictor of psychopathology.
4. Various other factors are also likely to contribute to psychopathology in ST, but have not been included in the multiple regression analysis because of their lack of interval scaling. Such factors are a *family history of mental disorder*, *multiple dystonic symptoms* and *life events* as a trigger of the illness. All of these variables were significantly associated with psychopathology. This suggests that psychopathology in ST must generally be considered as a result of a variety of interacting factors, biological, psychological and social.

We will comment briefly on some of these aspects.

Somatization

Somatization has been considered an *expression of a negative basic affect* that is represented at the level of non-specific physical complaints. The correlations between the somatization scale of the SCL 90 R and the burnout scales of Jones (1980) and Maslach and Jackson (1981), and particularly with the subscales *distress* and *emotional fatigue* (Franke, 1995) support this line of thought. The high scores of the ST patients on the somatization scale of the SCL 90 R may indicate an emotional state of distress and fatigue, which is expressed at a physical level. This implies that physical illness and somatization are phenomena which do not exclude but rather may facilitate each other.

Interpersonal sensitivity

The values beyond the normal range on the scale of *interpersonal sensitivity* correspond to the fact that a substantial number of patients (53.7%) felt themselves observed and critically looked at by the public and hence withdrew from social activities. Similar

clinical observations have been reported in the literature (Duane, 1988). Jahanshahi and Marsden (1988a) have emphasized the negative impact of the external disfigurement on the self-image in ST. The increased values on the scale *interpersonal sensitivity* can be linked to social insecurity due to an impairment of the body-image. Age seems a relevant factor modulating vulnerability in this area. The negative correlation between interpersonal sensitivity and age speaks for the fact that external disfigurement is more poorly tolerated by younger than by older patients.

Life events as trigger

More than 50% of the patients reported that their illness was triggered by stressful life events. This corresponds to the general findings in the literature (Scheidt *et al.*, 1996). The patients, who attributed their illness onset to stressful life events in our study, showed increased values on the SCL 90 R. They seemed to experience more psychological distress than the rest of the group. The high scores on psychopathology in the triggered-onset group may be interpreted as evidence of a high premorbid psychological vulnerability. However, patients who attribute their illness to life events may generally report more psychopathological symptoms.

Treatment

As has been pointed out the results presented are likely to differ from those obtained in a sample of untreated patients. The presence of psychopathology in about a third of cases on treatment gives rise to the question whether additional psychiatric and psychotherapeutic support should be offered to these patients. Identification of those in need of psychotherapy requires a comprehensive assessment, including the biological, psychological and social aspects of the condition. For clinical as well as methodological reasons, however, future studies should take into account that signs and symptoms in ST represent relatively independent dimensions which need to be evaluated separately.

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