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High Psychiatric Comorbidity in Spasmodic Torticollis: A Controlled Study

HARALD GÜNDEL, M.D.,[1] ANJA WOLF, M.A.,[1] VASSILIKI XIDARA, M.D.,[1] RAYMONDE BUSCH, M.S.,[2] KARL-HEINZ LADWIG, PH.D.,[1,3] FRANK JACOBI, PH.D.,[4] MICHAEL VON RAD, M.D.,[1] and ANDRES O. CEBALLOS-BAUMANN, M.D.[5]

Disturbed body image and negative self-referent cognitions caused by the postural disfigurement of the head are regarded as the main reason for elevated depression scores in spasmodic torticollis (ST), but this factor was never controlled for. We therefore compared 48 patients with ST and 48 patients with alopecia areata (AA) who were matched for age, sex, and body image dissatisfaction. Psychiatric diagnoses were based on a structured psychiatric interview (SCID-I). Results of patients with ST and AA were compared with a matched sample of the representative German population. Odds ratios to develop psychiatric comorbidity for patients with ST compared with patients with AA were significantly increased throughout nearly all assessed DSM-IV categories. Logistic regression analysis showed that (1) depressive coping and (2) belonging to the group of patients with ST correlated with a significantly higher rate of current psychiatric diagnosis. We conclude that high psychiatric comorbidity in ST is unlikely to be a mere consequence of chronic disease and disfigurement.

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Spasmodic torticollis (ST), also called *cervical dystonia*, is a form of focal dystonia in which tonic and phasic involuntary movements of the cervical musculature cause intermittent or sustained deviations of the head and neck. ST is the most common form of adult-onset focal dystonia, with an incidence of 1.2 per 100,000 (Claypool et al., 1995) to 5.4 per 100,000 (Castelon Konkiewitz et al., 2002). Mean age of onset is 41.6 years (SD 14.1), and the male to female sex ratio is 1:1.3 (Castelon Konkiewitz et al., 2002). Clinical severity of ST may be measured by the Tsui index (Tsui et al., 1986). Individual scores reported by Tsui et al. (1986) range between 6 and 16; Kutvonen et al. (1997) report a mean Tsui score of 13.2 in 39 patients with ST. Although 10% to 20% of patients may experience remission, nearly all patients relapse within 5 years and are left with persistent disease (Dauer et al., 1998).

Spasmodic torticollis is now regarded as a focal dystonia. On the basis of electrophysiological (for review, Berardelli et al., 1998) and functional imaging studies (for review, Ceballos-Baumann and Brooks, 1998), it has been concluded that primary dystonias result from a functional disturbance of the basal ganglia causing altered thalamic control of cortical motor planning and executive areas. It has been shown that direct affective input from the amygdala and orbitofrontal cortex is provided to the caudate nucleus and at the thalamic level, thus helping to elicit or suppress specific patterns of motor behavior in response to emotional states (Mogenson et al., 1980; Rolls, 1995). Interestingly, it is in the models of frontal-subcortical connectivity that the dystonias and hysteria have recently been brought together (Ron, 2001). Before the advent of sophisticated electrophysiology, functional imaging, and genetics, the dystonias were regarded as unexplained

[1] Institut und Poliklinik für Psychosomatische Medizin, Psychotherapie und Medizinische Psychologie, TU München, Klinikum rechts der Isar, Langerstr. 3, D-81675 München, Germany. Send reprint requests to Dr. Gündel.
[2] Institute for Medical Statistics and Epidemiology, Technische Universität München, München, Germany.
[3] GSF—National Research Center for Environment and Health, Institute of Epidemiology, Neuherberg, Germany.
[4] Klinische Psychologie und Psychotherapie, AG Epidemiologie und Versorgungsforschung Technische Universität Dresden, Dresden, Germany.
[5] Department of Neurology, TU München, Klinikum rechts der Isar, München, Germany.

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neurological symptoms; in fact, many patients with dystonia were diagnosed as hysterical.

Abnormalities in the generation of motor programs and readiness to move at the level of striatothalamocortical circuits were suggested as neuroanatomical correlates of hysterical sensorimotor loss in patients using functional imaging (Vuilleumier et al., 2001), similar to results of studies in dystonia (for review, Ceballos-Baumann and Brooks, 1998). Indeed, an interaction of motor and affective control at a subcortical level could explain subtle primary psychiatric abnormalities in ST within the concept of dystonia as a brain disease. In the only population-based study on the natural history of ST, 6 of the 11 new patients identified with ST had a psychiatric diagnosis. In five, the psychiatric diagnosis preceded the onset of dystonia (Claypool et al., 1995).

Psychological factors have been implicated in the cause of ST in the past. However, chronic disease is commonly complicated by psychiatric comorbidity (Cohen-Cole et al., 1993). For instance, social phobia as significant psychiatric comorbidity coexists with stuttering, essential tremor, and ST (George and Lydiard, 1994; Gündel et al., 2001; Oberlander et al., 1994; Stein et al., 1996). There has been ample work on the extent of psychosocial distress and on the interaction between primarily somatic and psychological factors in ST. Stressful life events before onset of the illness have been found repeatedly in 30% to 50% of the patients studied (Jahanshahi and Marsden, 1988a; Matthews et al., 1978; Scheidt et al., 1996). In addition, an increased number of psychodynamic personality abnormalities have been described in a subgroup of patients (Cleveland, 1959; Mitscherlich, 1979; Scheidt, 1995).

The problem when addressing psychiatric issues in ST has been the lack of an appropriate control group. In previous studies, psychiatric aspects in ST have been investigated mainly with the aid of standardized questionnaires (Cleveland, 1959; Jahanshahi and Marsden, 1988a; Jahanshahi and Marsden, 1988b; Scheidt et al., 1996). Five of these previous studies used control groups, on conversion hysteria (Cleveland, 1959), surgical patients (Cockburn, 1971), operated herniated disc (Choppy-Jacolin et al., 1977), Parkinson disease (PD) (Naber et al., 1988), and cervical spondylosis (Jahanshahi and Marsden, 1988a; Jahanshahi and Marsden, 1988b). Two of these studies used nonstandardized testing (Cleveland, 1959; Cockburn, 1971), two studies used self-rating questionnaires (Choppy-Jacolin et al., 1977; Jahanshahi and Marsden, 1988a; Jahanshahi and Marsden, 1988b), and none of these studies controlled for the disfigurement. So far there has been no controlled study about the prevalence of psychiatric comorbidity in ST as defined by DSM-IV criteria (American Psychiatric Association, 1994).

A recent study showed that patients with ST experience considerable stigma resulting in avoidance of others; feelings of being avoided by others, of being different, of unattractiveness, and of self-consciousness; and being apologetic (Papathanasiou et al., 2001). These authors concluded that self-consciousness about the altered physical appearance combined with perceived stigma could lay the grounds for the development of depression in ST, leaving the person feeling devalued and subject to negative stereotyping. Indeed, Jahanshahi and Marsden (1990) found that negative self-referent cognitions such as self-blame, self-accusation, and negative body image were the prominent components of depression in ST, and we found that more than 50% of ST patients met DSM-IV clinical criteria A to G for current social phobia (Gündel et al., 2001). Jahanshahi and Marsden (1988b) proposed that future studies should investigate the contribution of self-perceived disfigurement to the experience of depression in ST. An appropriate control group for the patients with ST is therefore a crucial precondition to address this issue further.

We screened different populations of patients with disfigurement in the craniocervical region. Recruitment of age-matched adults with cleft palate who underwent operations during early childhood was not practical, and this cohort is probably less disfigured than patients with ST. We investigated patients with facial burns from a national burns center and found that isolated facial burns are extremely rare. Patients with a history of facial trauma treated at a maxillofacial surgery unit often had the confound of coincident brain injury, and as in patients with facial burns, most cases showed disfigurement out of proportion with ST. Eventually, we chose patients with alopecia areata (AA) as control group.

Alopecia areata is a form of hair loss that may be limited to patches on the scalp but may also involve the entire scalp and other hair-bearing regions of the body. AA affects men and women equally. Sixty percent of patients seek treatment for their first patch before 20 years of age, but patients usually seek treatment for several episodes of hair loss and hair regrowth during their lifetime. Its clinical presentation can vary from a single patch of hair loss to multiple patches or total hair loss. Loss of all scalp hair (alopecia totalis) is seen in 5% to 10% of cases. Sites other than the scalp (eyelashes, beard, general body hair) are affected in 10% of patients. Alopecia universalis involves the loss of all scalp and body hair. Complete recovery for 10 to 15 years is seen in one third of cases (Colon et al., 1991; Madani and

Shapiro, 2000). It is a nonmalignant disease that does not affect multiple organ systems. It is suspected to be an autoimmune disease that is hitherto not known to interfere with basal ganglia–thalamocortical circuits (Madani and Shapiro, 2000), but, like ST, is especially disfiguring in the craniocervical region (Garcia-Hernandez et al., 1999). Similar to ST, it is always visible and obvious to others, and it may fluctuate in extent of disease, thus making patients feel different (Gupta and Gupta, 1998). Individual embarrassment because of the stigmatized physical appearance is present both in ST and AA and often leads to growing social isolation and eventually to a high amount of psychiatric comorbidity (Garcia-Hernandez et al., 1999; Gündel et al., 2001; Koo et al., 1994). Here, we report the psychiatric comorbidity in ST compared with AA and with a large representative German population sample.

Methods

Patients

Inclusion criterion for patients with ST was a diagnosis compatible with primary cervical dystonia amenable to botulinum toxin treatment (*i.e.*, no other known cause for the dystonia, no tardive dystonia). Inclusion criterion for AA patients was bald patches on the head. Additional inclusion criteria for both groups were age older than 18 years with no history or present use of neuroleptic medication and the ability and willingness to participate in the study. Patients were recruited from dermatological and neurological outpatients at the university hospitals of Munich and Erlangen. Patients with ST were referred for botulinum toxin injections. Patients with AA came for diphenylcyclopropenon treatment.

Disturbed body image in ST may be the main reason for depression in this group (Jahanshahi and Marsden, 1988b), and additionally, it has been shown that the prevalence of psychiatric comorbidity correlates with older age and female gender (*e.g.*, Carter et al., 2001). Therefore, from the studied cohort of 116 ST and 76 AA patients, 48 ST and 48 AA patients (28 women, 20 men) were matched for age, sex, and body image dissatisfaction according the guidelines for 1:1 case-control studies (Woodward, 1999). Informed consent was obtained from all subjects, and the study was approved by the local ethics committee.

Measures

Patient Evaluation. Patients with ST were examined with a validated functional scale that included amplitude and duration of sustained movement (rotation, tilt, anteversion/retroversion), shoulder elevation, and tremor (Tsui et al., 1986) by a neurologist blinded to the results of the psychiatric evaluation who specialized in the diagnosis and treatment of dystonia. A structured interview was developed for the purpose of the study. Items referred to demographic variables, past medical history, stressful life events within the year before disease onset, the course of the disease, previous treatments, and current neurological and dermatological status.

To our knowledge, there is no validated instrument to measure subjective stigma caused by disfigurement; therefore, patients assessed their body image dissatisfaction on a visual analogue scale (VAS). Ratings were 0, normal, *i.e.*, no image dissatisfaction at all, to 10, maximal dissatisfaction. Participants also completed self-report scales designed to provide information regarding general psychopathology and coping style.

SCL-90-R. The SCL-90-R (Derogatis, 1986) is a 90-item questionnaire assessing general psychiatric symptomatology; it yields an overall index of psychiatric distress, the General Symptomatic Index. Research has documented the reliability and validity of this original scale (Derogatis, 1977) and its version in the German language (Franke, 1995).

Freiburger Fragebogen zur Krankheitsverarbeitung. The Freiburger Fragebogen zur Krankheitsverarbeitung (FKV-LIS; Muthny, 1989) takes into account a wide spectrum of cognitive, emotional, and activity-related coping strategies. It consists of 35 items, each rated on a 5-point scale. The authors describe five factor-analytical scales. Scores indicate the extent of each coping strategy individually used. The optimal cutoff score was determined for each scale to differentiate high versus low scorers using the receiver operating characteristic (ROC) method.

Social Phobia Scale. The Social Phobia Scale (SPS; Heimberg et al., 1992) was used in its validated German version (Stangier et al, 1999) to measure social anxiety. The 20 items of each questionnaire are rated on a 5-point scale. Higher scores are associated with greater pathology.

Psychiatric Evaluation

The occurrence of psychiatric disorders was assessed during a structured psychiatric interview (Structured Clinical Interview for DSM-IV, axis I, German version, Wittchen et al., 1997a) according to

DSM-IV criteria (American Psychiatric Association, 1994). The SCID assesses current (last 4 weeks before interview) and lifetime psychiatric status for major axis I psychiatric disorders using criteria in accordance with the DSM-IV. DSM-IV excludes people with social anxiety secondary to disfiguring or disabling physical conditions from the diagnostic category of social phobia. To qualify for the diagnosis of social phobia in DSM-IV, subjects must meet criteria A to H. DSM-IV criterion H specifies that social anxiety may not be in association with a "medical illness factor." The resulting formal assignment of patients with ST meeting DSM-IV criteria A to G, but not H, to the category of anxiety disorder not otherwise specified (300.00) still describes a psychiatric disorder, but it completely blurs the specific clinical situation. As proposed by other authors (George and Lydiard, 1994; Oberlander et al., 1994; Stein et al., 1996), we therefore modified DSM-IV criteria to permit a diagnosis of social phobia if only the clinical criteria A to G were met. Our previous work supports the construct validity of this modified definition of social phobia (Gündel et al., 2001). According to guidelines concerning interviewer qualifications and training (Spitzer et al., 1992), the clinically experienced interviewing authors attended a training course for the SCID-I interview technique run by a certified SCID trainer (H. U. Wittchen, Munich, Germany) or performed 10 supervised SCID interviews, or both. Because of the clinical context of the interview, interviewers could not be blinded to patient groups.

The data on psychiatric morbidity in the German general population come from the German National Health Interview and Examination Survey and its Mental Health Supplement (GHS-MHS), the first nationwide, epidemiological study of mental health in Germany (Jacobi et al., 2002; Wittchen et al., 1998a, 1998b). The psychopathological and diagnostic assessments were based on the computer-assisted version of the Munich Composite International Diagnostic Interview (Wittchen and Pfister, 1997b), a fully structured interview that allows the diagnosis of DSM-IV mental disorders. Test–retest reliability and procedural validity were found to be acceptable (Wittchen et al., 1998b). The GHS-MHS sample consisted of a stratified, cross-sectional, representative sample from 130 sites ($N = 4181$ adult respondents) throughout Germany. Details are described elsewhere (Jacobi et al., 2002).

Statistical Analyses

Between-group differences in SCID data were assessed by unpaired *t*-test, Mann-Whitney, and chi-square statistics (when appropriate) with the software package SPSS (Chicago, IL). Alpha was set to .05 (two-tailed). In order to investigate the predictors of current psychiatric comorbidity, a stepwise multiple logistic regression was calculated. All main clinical variables were included. Continuous variables were dichotomized by ROC analyses. Objective variables were education, marital status, severity as functional score (Tsui et al., 1986) and extent (isolated pure rotational torticollis versus complex torticollis with adjacent dystonic involvement) of ST, duration of the illness (<5 or ≥ 5 years), and belonging to the ST or AA group. Subjective variables were pain (VAS 0 to 10); extent of psychopathology (SCL-90-R mean general symptom index $<.5$ or $\geq .5$); incriminating life event within the year before onset of disease, yes/no; depressive coping (FKV subscore <22 or ≥ 22); and extent of body image dissatisfaction (VAS <5 or ≥ 5). Odds ratios (Ors) with 95% confidence intervals (CIs) were calculated to estimate increased risks in psychiatric comorbidity in patients with ST and AA compared with the general population (controlling for age and gender) and for comparisons between the ST and AA groups (additionally controlled for body image dissatisfaction).

Results

Baseline Sociodemographic and Illness Characteristics of ST and AA Groups

The basic characteristics of the two samples are summarized in Table 1. Other demographic data (marital status, educational level, socioeconomic level) did not differ significantly.

Psychiatric Comorbidity in ST and AA Groups

Analyses revealed that psychiatric comorbidity was more common in the ST than in the AA group (Table 2). There was a higher rate of current psychiatric comorbidity in the ST (77.1%) than in the AA group (41.7%; $p = .01$, Fisher exact test) with a different pattern of psychological distress: of the 48 patients with ST, 33 (68.8%) fulfilled DSM-IV criteria for current (4-week) anxiety disorders and 9 (18.8%) for mood disorders. In the group of 48 patients with AA, only 16 (33.3%) fulfilled DSM-IV criteria for current anxiety disorders and 6 (12.5%) for mood disorders. Lifetime prevalence of psychiatric comorbidity was 91.7% in the patients with ST *vs.* 60.4% in the patients with AA ($p \leq .01$, chi-square).

TABLE 1
Baseline Sociodemographic and Illness Characteristics of the ST and AA Groups

	ST Mean (SD)	AA Mean (SD)
Mean age	47.5 (11.0)	47.5 (11.0)
Age at onset	34.9 (13.3)	38.8 (14.0)
Duration of illness, years	12.5 (11.4)	8.7 (10.2)*
Body image dissatisfaction (VAS 0–10)	4.7 (3.3)	5.2 (3.0)
	N (%)	*N* (%)
Sex ratio (female:male)	29:19 (60.4:39.6)	29:19 (60.4:39.6)
Severity of ST[a] and AA	Tsui score ≤6: 21 (43.8)	Alopecia vulgaris: 13 (27.1)
	Tsui score 7–9: 14 (29.2)	Alopecia liminaris: 8 (16.7)
	Tsui score ≥10: 13 (27.1)	A. totalis/universalis: 27 (56.3)
Clinical course since onset		
No change	10 (20.8)	1 (2.1)
Deterioration	29 (60.4)	23 (47.9)
Improvement	2 (4.2)	8 (16.7)
Permanent fluctuations	7 (14.6)	16 (33)

[a] Overall range of clinical impairment in ST and AA as measured with the Tsui index (ST) or subcategorized according to pattern or extent of the hair loss (AA) was not subject to further statistical analysis.
* $p < .05$.

TABLE 2
Frequency of Psychiatric Disorders (DSM-IV, Axis I) in a Group of 48 Patients With ST (N = 48) and AA (N = 48) Compared with the General Population (GHS-MHS; N = 4181)

	Current ST Patients		Current AA Patients		Current General Population	Lifetime ST Patients		Lifetime AA Patients		Lifetime General Population
Diagnosis (DSM-IV)	*N*	%	*N*	%	%	*N*	%	*N*	%	
No diagnosis	11	22.9	28	58.3	80.2	4	8.3	19	39.6	57.2
	$p = .01$ OR = 4.7 95% CI: 1.95–11.40					$p = .001$ OR = 7.20 95% CI: 2.22–23.4				
Mood disorders	9	18.8	6	12.5	6.3	19	39.7	15	31.3	17.3
Major depressive disorder—single episode	6	12.5	5	10.4	2.1	16	33.3	13	27.1	9.3
Major depressive disorder—recurrent	3	6.3	1	2.1	1.4	3	6.3	2	4.2	5.5
Dysthymia	1	2.1	0	0	3.6		*a*		*a*	4.5
Anxiety disorders[b]	33	68.8	16	33.3	8.7	40	83.3	22	45.8	>15.4[d]
Social phobia[c]	26	54.2	14	29.2	1.2	34	70.8	19	39.6	*d*
Panic disorder with or without agoraphobia	3	6.3	0	0	1.1	4	8.3	3	6.3	3.9
Other anxiety disorders	13	27.1	3	6.3	6.9	14	29.2	4	8.3	*d*
Adjustment disorders	4	8.3	5	10.4	*e*	10[e]	20.8	8	16.7	*e*
Other psychiatric disorders (without presence of a mood or anxiety disorder)	7	14.6	1	2.1	7.2	8	16.7	2	4.2	16.3

[a] A SCID diagnosis was given only if criteria for this disorder were fulfilled at the time of interview.
[b] Without obsessive-compulsive disorder, posttraumatic stress disorder.
[c] using DSM-IV criteria A–G.
[d] 12-Month prevalence; lifetime prevalence in anxiety disorders only assessed in panic disorders.
[e] Not assessed in the GHS-MHS; in the present study, lifetime diagnosis of adjustment disorder was assessed in order to elucidate how many patients developed the diagnosis after manifestation of ST or AA but managed to cope with this chronic disease in its further course.

TABLE 3
Comparison of the ORs to Develop Psychiatric Comorbidity Between ST or AA Patients and a Representative Sample of the German General Population (GHS-MHS)

	ST *vs.* GP[a] OR (95% CI)	Significance of Difference ST *vs.* GP	AA *vs.* GP[a] OR (95% CI)	Significance of Difference AA *vs.* GP	ST *vs.* AA[b] OR (95% CI)	Significance of Difference ST *vs.* AA
Any depressive disorder—current diagnosis	3.91 (1.89–8.08)	<.000	2.04 (0.84–4.96)	NS	2.01 (0.65–6.23)	NS
Any depressive disorder—lifetime diagnosis	5.15 (2.75–9.65)	<.000	2.39 (1.26–4.53)	<.008	2.39 (0.99–5.79)	<.053
Any anxiety disorder[c]—current diagnosis	21.30 (11.04–41.10)	<.000	5.09 (2.72–9.54)	<.000	4.61 (1.85–11.47)	<.001
Any anxiety disorder[c]—lifetime diagnosis	28.23 (12.32–64.66)	<.000	4.10 (2.25–7.45)	<.000	9.50 (3.42–26.36)	<.000
Any disorder—current diagnosis	13.12 (6.61–26.04)	<.000	3.60 (2.03–6.38)	<.000	4.76 (1.72–13.17)	<.003
Any disorder—lifetime diagnosis	29.39 (7.04–122.64)	<.000	2.81 (1.51–5.25)	<.001	34.51 (4.01–297.38)	<.002

[a] Odds ratios from logistic regression, controlled for age and sex.
[b] Odds ratios from logistic regression, controlled for age, sex, and body dissatisfaction.
[c] Without obsessive-compulsive disorder, posttraumatic stress disorder.

Comparison With Epidemiological Findings on Psychiatric Comorbidity According to DSM-IV Criteria in the German Representative Population

There is a significantly higher amount of psychiatric comorbidity in patients with ST and AA within the categories of anxiety and depressive disorders according to DSM-IV criteria than in the representative German population (Table 3). Psychiatric comorbidity is significantly higher in patients with ST and AA compared with the general population (OR = 2.39 to 29.39, controlled for age and gender), with the exception of patients with AA in the category current depressive disorders. With the same exception, we found significant ORs (additionally controlled for body image dissatisfaction) for patients with ST compared with patients with AA throughout all assessed DSM-IV categories (Table 3).

Self-Report Questionnaires: General Psychopathologic Symptoms (SCL-90-R) and SPS

Patients with ST or AA showed a significant increase in general psychopathology (SCL-90-R—global score) and in all but one SCL-90-R subscale (SCL6: hostility) compared with the original sample of normal subjects described by Derogatis (1977) (*t*-test, $p < .01$). No significant difference in self-rated psychopathology (SCL-90-R subscales and global score) was found between patients with ST and AA.

The self-rating scale SPS measuring social phobia showed a higher score ($p = .008$) in the patients with ST (median, 15.5) than in the patients with AA (median, 8.0), thereby confirming the findings of the SCID interview.

Predictors of Current Psychiatric Comorbidity

Only two parameters emerged as independent significant predictors of current psychiatric comorbidity in patients with ST and AA: (1) depressive coping ($p < .001$; OR = 5.3; 95% CI, 2.0 to 14.3) and (2) belonging to the ST group compared with the AA group ($p < .01$; OR = 3.7; 95% CI, 1.4 to 9.7). Thus, belonging to the group of patients with ST was associated with a ×3.7 increased risk for current psychiatric diagnoses compared with the group of patients with AA when controlling for the factor depressive coping.

Discussion

This study indicates that in patients with ST, a high prevalence of current and lifetime psychiatric comorbidity may not be explained as secondary to altered physical appearance or chronic disease alone. Psychiatric comorbidity in ST and in the control group was mainly caused by anxiety and depressive disorders. ST was significantly associated with a ×3.7 increased risk for current psychiatric comorbidity when compared with a matched group with altered physical appearance in the head region and similar body image dissatisfaction. Compared with the age-matched and gender-matched representative German population, the risk for psychiatric comorbidity in patients with ST was markedly increased for current psychiatric comorbidity (OR = 13.12; 95% CI, 6.61 to 26.04) and for lifetime psychiatric comorbidity (OR = 29.39, 95% CI, 7.04 to 122.64). As could be expected in chronic disease with altered physical appearance, the AA group also had a signif-

icantly higher current and lifetime psychiatric comorbidity compared with the representative German population (current: OR = 3.60, 95% CI, 2.03 to 6.38; lifetime: OR = 2.81, 95% CI, 1.51 to 5.25).

We found only five previous studies on the psychological aspects of ST that included a control group. Cleveland et al. (1959), using projective tests (*e.g.*, Rorschach), found in patients with ST significantly more "fantasies featuring guilt and shame" compared with "conversion hysterics." Cockburn (1971), using nonstandardized interviews and a retrospectively answered personality inventory, compared 46 patients with ST with matched surgical patients to assess the premorbid personality. It was concluded without statistical tests that there were no differences between the two groups. Choppy-Jacolin et al. (1977) compared the performance of 34 patients with ST with operated herniated disc on a number of tests, including the MMPI and the Rorschach. These authors concluded that the patients with ST were "more neurotic." It has to be mentioned that these older studies did not rely on reliable diagnostic criteria that were established with the third edition of the DSM.

Naber et al. (1988) investigated 32 patients with ST, but not the control group with PD, with a structured psychiatric interview. There was a low current psychiatric comorbidity in the ST group (2/32) according the DSM-III interview. However, there was a considerable bias because patients with ST were recruited by advertisement or came from a patient association. A number of questionnaires were completed by the patients with ST and PD, and no significant differences were found between groups. It is questionable whether PD as a progressive neurodegenerative disease is an appropriate control group for ST. Jahanshahi and Marsden (1988a, 1988b) compared 85 patients with ST to a control group of 49 patients with cervical spondylosis to establish the effect of torticollis on the prevalence of psychiatric disorder and on self-reports of depression, hopelessness, and neurotic traits. The response rate to questionnaires was 68% for patients with ST and 40.2% for patients with spondylosis. They reported increased mean scores on the Beck depression inventory (Jahanshahi and Marsden, 1988b) and no difference in terms of any of the personality dimensions evaluated (Jahanshahi and Marsden, 1988a). The authors considered this extra degree of depression found among patients with ST a consequence of the postural abnormality of the head (Jahanshahi and Marsden, 1988b). However, the results of the study by Jahanshahi and Marsden (1988b) are limited by the assessment with mailed self-rating questionnaires, in contrast with our study, which applied a structured psychiatric interview according to DSM-IV (SCID-I) and validated questionnaires.

Our approach using the SCID interview disclosed a clear statistical difference between the two clinical groups with altered physical appearance in the prevalence of current and lifetime psychiatric comorbidity.

The discrepancy between comparable results in the self-rated questionnaire SCL-90-R on the one hand and significant differences in psychiatric comorbidity between the ST and AA groups using a personal structured interview (SCID) on the other hand supports the assumption that self-ratings alone may not reflect true psychiatric comorbidity (Scheidt, 1995; Scheidt et al., 1999). A comparison of the screening properties of the SCL-90-R and the SCID in a primary care setting showed that there are indeed some patients with psychiatric comorbidity according to the SCID interview who score normally on the SCL-90-R (Schmitz et al., 1999). Additionally, the validity of our results concerning psychiatric comorbidity was supported by the finding that symptoms of social phobia as measured with the self-rating scale SPS were significantly worse among patients with ST than patients with AA.

Could the increased psychiatric comorbidity in ST compared with AA be a result of factors other than disease-related factors? Disfigurement as a main feature of both diseases was comparable between both groups according to VAS scores. Duration of disease was 8.7 years in the AA group and 12.5 years in the ST group, but duration of illness has been shown to have no significant impact on psychiatric comorbidity in ST or other neurological disorders (*e.g.*, Cleveland, 1959; Fruehwald et al., 2001; Gündel et al., 2001). In addition, no significant relationship between duration of illness and stigma scores (Papathanasiou et al., 2001) or self-rated psychopathology as measured with the SCL-90-R (Scheidt et al., 1996) has been observed in patients with ST. Likewise, we found no significant relationship between duration of AA and prevalence of current psychiatric comorbidity in our AA sample. Thus, our results should not be influenced by different duration of disease in both groups.

Patients with AA did not report pain, in contrast with the patients with ST, who scored a median of 4.0 on the VAS. However, our patients with ST reported pain at a time when they came for a repeat botulinum toxin treatment, which efficiently relieves pain in ST. Jahanshahi and Marsden (1992) reported that 22 of 26 patients with ST considered their torticollis to be better as a result of the botulinum toxin injection. Straightening of the head and relief of neck pain was rank-ordered as the first or second most important benefit by 11 patients. Therefore,

pain levels reported by our patients at the time of interview should not be considered a lasting phenomenon.

Furthermore, one third of patients with ST are reported to be completely pain-free, and if affected by pain, it is moderate in most instances (Kutvonen et al., 1997). In a community-based population survey, ST was associated with a low probability of pain, also (Claypool et al., 1995). Likewise, it is unlikely that an interviewer bias may have confounded the results, because SCID-I follows strict manualized diagnostic rules and was applied by trained raters.

Conclusions

High psychiatric comorbidity, mainly depressive and anxiety disorders, occurs in ST and is unlikely to be a mere consequence of chronic disease and disfigurement. Therefore, other factors must be sought to explain the striking difference in psychiatric comorbidity in ST compared with the control group. The current concept on the pathophysiology of ST is that as a primary dystonia, it results from a functional disturbance of the basal ganglia causing altered thalamic control of cortical motor planning and executive areas (Berardelli et al., 1998). Current conceptions of the neurophysiology of the basal ganglia emphasize an arrangement involving at least five separable cortico-striato-pallido-thalamo-cortical loops. The motor loop links the basal ganglia via the ventral and dorsomedial thalamus to frontal cortex. Other loops are thought to mediate attentional, cognitive, and limbic functions (Alexander et al., 1990). Their degree of segregation and the extent of interaction are still a matter of debate (Bergman et al., 1998).

However, why should the pathology of primary dystonia affect the motor circuit exclusively? Clinically, interactions between different basal ganglia-thalamocortical loops in degenerative diseases like PD or Huntington disease are well accepted (Cummings, 1998). In these disorders, mood and motivational disturbances were related to dysfunction in the limbic loop (Austin et al., 1995). Obviously, in primary dystonia, such nonmotor dysfunction should be more subtle and confounded by effects of chronic disease. However, a predominant dysfunction in one loop (here, motor loop) could also affect another loop (*i.e.*, limbic loop) through crosstalk and reflect a more generalized dysfunction of basal ganglia-thalamo-cortical connectivity in primary dystonia or ST (Bergman et al., 1998). Our theory may explain the notion of many clinicians during the last century that ST is more than just a motor disorder, because the primary pathology affects not only motor behavior but also affective processing. In a time when the mental–motor functional duality of the brain appears progressively unified, this theory may help bridge the fruitless debate about psychogenicity and organicity of ST.

In summary, our results together with models of basal ganglia thalamocortical connectivity could imply that the high psychiatric comorbidity in ST relative to AA should not be interpreted merely as a consequence of the disfigurement. A hypothesis to be explored further is that the dysfunction of basal ganglia-thalamo-cortical connectivity extends beyond motor control circuitries, at least in a subgroup of patients with ST, and (subtle) affective dysfunction may represent an intrinsic part of the disease, at least in a subgroup of patients with ST.

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