The psychological defensive profile of primary Sjögren's syndrome patients and its relationship to health-related quality of life

T. Hyphantis[1], D. Mantis[1], P.V. Voulgari[2], N. Tsifetaki[2], A.A. Drosos[2]

[1]Department of Psychiatry and [2]Rheumatology Clinic, Department of Internal Medicine, Medical School, University of Ioannina, Ioannina, Greece.

Abstract

Objective

We aimed to assess the defensive profile of primary Sjögren's syndrome (SS) patients and to investigate the independent associations of psychological distress and personality variables with health-related quality of life (HRQOL).

Methods

In 40 primary SS patients we assessed psychological distress (SCL-90-R), ego defense mechanisms (Defense Style Questionnaire), hostility features (HDHQ) and HRQOL (WHOQOL-BREF). Fifty-six patients with systemic lupus erythematosous (SLE) and 80 healthy participants matched for age and sex served as controls.

Results

Primary SS patients presented higher rates of general psychological distress compared to SLE and healthy participants. Symptoms of somatisation were more prominent in SS than SLE or healthy controls. SS patients presented less use of humour defense and more help-rejecting complains and delusional guilt hostility, compared to controls. Primary SS patients' HRQOL was more impaired than healthy participants and comparable to SLE. Psychological distress was a constant independent correlate of SS patients' HRQOL, while less use of humour ($p<0.001$) and higher rates of delusional guilt ($p=0.032$) were also significantly associated with Physical HRQOL independently of psychological distress; more use of schizoid fantasy was also independently associated with impaired Environment HRQOL ($p=0.005$).

Conclusion

Primary SS patients exhibit several specific psychological difficulties in adaptation to life stressors, and clinicians and consultation-liaison psychiatrists, apart from the early assessment and treatment of psychological distress and somatisation symptoms, should consider the patients' underlying defensive profile and coping capacities, since such personality traits, although usually underestimated, are also independently associated with the disease outcome.

Key words

primary Sjögren's syndrome, quality of life, psychological distress, personality, defense style questionnaire, ego defense mechanisms, hostility

Thomas Hyphantis, MD
Dimitrios Mantis, MSc
Paraskevi V. Voulgari, MD
Niki Tsifetaki, MD
Alexandros A. Drosos, MD, FACR

Please address correspondence and reprint requests to:
Alexandros A. Drosos, MD, FACR,
Professor of Medicine/Rheumatology,
Rheumatology Clinic,
Department of Internal Medicine,
Medical School,
University of Ioannina,
45110 Ioannina, Greece.
E-mail:
adrosos@cc.uoi.gr; www.rheumatology.gr

Received on October 22, 2010; accepted in revised form on December 20, 2010.

Competing interests: none declared.

Introduction

Primary Sjögren's syndrome (SS) is a chronic, progressive, systemic autoimmune disease characterised by inflammation of the exocrine glands and functional impairment of the salivary and lachrymal glands, with dry mouth and dry eyes being the most prominent complaints (1, 2). Extraglandular involvement including arthritis, pulmonary disease, renal disease, skin vasculitis, peripheral neuropathy or glomerulonephritis may also occur (1-4), and patients belonging to this category have worse prognosis with higher mortality rates (4). Neuropsychiatric dysfunction has been also reported (5-7), mainly in the form of mild cognitive impairment with attention and concentration deficits (5), diminished cognitive capacity (6) or headache (7); symptoms of psychological distress are also common (8), and patients with primary SS have shown increased rates of clinically significant anxiety (9) and depressive symptoms (9-11), with chronic malaise being a well-recognised manifestation with a major impact on daily life (12, 13). The syndrome affects 0.09% of the adult population (14), with a male/female ratio of 9/1 (1, 15). Based on the American-European Consensus Group (AECG) criteria, prevalence among women ranges between 0.1% and 0.6% in USA, UK, and Greek cohorts (14, 16).

Evidence suggests that HRQOL is reduced in primary SS patients compared to healthy population (15, 17-20) and it is comparable to other rheumatic diseases, including rheumatoid arthritis (RA) and systemic lupus erythematosous (SLE) (21, 22). Studies investigating variables associated with primary SS patients' HRQOL showed that, among the clinical variables studied, fatigue was the dominant predictor of health status (12, 17, 23), while sicca severity did not uniquely contribute to HRQOL (17). On the other hand, although symptoms of psychological distress are well-known predictors of HRQOL in a number of medical illnesses including rheumatic diseases (24), their contribution to the primary SS patients' HRQOL has not been extensively studied. Only one recent study investigated the relationship of psychological distress with primary SS patients' HRQOL (20). In addition, to our best of knowledge, no study has focused on the relationship between the SS patients' psychological profile and their health status. Nonetheless, several personality variables and their complex interaction with psychological distress seem to play a significant role in a disease process and HRQOL. For example, our previous research has shown that disease activity was connected with the defensive personality profile in RA and inflammatory bowel disease (25, 26), while others have found that the cancer patients' underlying defensive organisation was indicator even of lowered survival (27), indicating that the study of specific personality traits such as the defensive organisation and coping capacities in relation to disease outcome may help to better identify on whom patient a psychological intervention will have a major impact. Our previous studies in RA showed that maladaptive defenses were associated with physical HRQOL, but this relationship was mediated by psychological distress (28), while in scleroderma we found that maladaptive defenses were strongly independently associated with HRQOL (29). Thus, we examine here whether the same applies to primary SS.

The aim of the present study was to assess the defensive profile of patients with primary SS and whether it is different from that of patients with another rheumatologic disorder, namely SLE, or healthy participants. We also aimed to investigate the independent associations of psychological distress symptoms and personality variables, namely ego defense mechanisms and hostility features, with primary SS patients' HRQOL.

Methods

Participants and procedures

In this cross-sectional study the sample comprised consecutive patients with primary SS attending a follow-up clinic during a one-year period at the Rheumatology Department of the University Hospital of Ioannina, which provides secondary and tertiary care for a population of 350,000 in north-western Greece. Diagnosis of primary SS was confirmed based on the respectively recommended criteria (30). Exclusion

criteria were inability to read and write Greek, history of psychotic illness, current alcohol and/or drug abuse or dementia. After complete description of the study to the 76 invited patients, 40 out of the 59 eligible patients agreed to participate (response rate: 67.8%) and signed informed consent was obtained. No statistically significant differences were found in major demographic characteristics between the participants' and non-participants' groups.

To test whether the SS patients' psychological profile differed from that of healthy participants as well as from patients with another rheumatologic disease, 56 patients with SLE who attended the same Rheumatology Department and 80 healthy participants matched for age and sex were also recruited. SLE was chosen because it shares similarities with SS and has been used for comparison in other studies evaluating outcomes in SS (7, 12, 22, 31). Diagnosis of SLE was confirmed using the American College of Rheumatology criteria (32). The healthy control group derived from a larger group of 984 subjects who participated in our study on the standardisation of Defense Style Questionnaire for the Greek population (33). This group was randomly selected from the hospital's staff-list and the hospital's visitors. Since we considered a 1:2 matching, each SS patient was matched for both sex and age with two control participants from the standardisation study. For each SS patient, the automated matching protocol selected all subjects who matched the SS patient exactly on sex and age category. If more than two participants from the standardisation sample were found to match with an SS patient, a computer generated random number selected the control subjects that were used in the study. If no exact matches were available, alternative matches were sought by allowing difference in one category. Table I presents the patients' and healthy participants' demographic profile. All the procedures followed were in accordance with the ethical standards on human experimentation (World Medical Association Helsinki Declaration) and with the local hospital's ethics committee (no. 20/14.01.2004).

Measures

Patients were examined by experienced rheumatologists (NT-PVV) and medical data were collected, including clinical features and laboratory data. The patients' medical records were reviewed for coexisting medical diseases and these were scored using the general comorbidity scale developed by Charlson and colleagues (34). The current use of any category of anti-inflammatory, anti-rheumatic or antidepressant agent was recorded from the patients' records. Each type of medication was examined separately for its effect on HRQOL. The psychological data collection was via a semi-structured interview performed by the same interviewer (DM). The following self-reported questionnaires were administered:

The *Symptom Distress Checklist (SCL-90-R)* which was used to assess patients' psychological distress, is a 90-item multidimensional self-report symptom inventory which measures a wide range of psychopathological symptoms in psychiatric and medical patients (35). It also provides the Global Symptom Index (GSI), a measurement of overall psychological distress. The utility of SCL-90-R as a psychological screening instrument in rheumatic disease patients has been well documented (36); also, it has been standardised for the Greek population (37).

Defensive profile and hostility variables

Defense mechanisms: Ego defense mechanisms are defined as "automatic psychological processes that protect the individual against anxiety and from the awareness of internal or external dangers and stressors, mediating the individual's reactions to emotional conflicts and to internal or external stressors" (38). To measure the participants' defensive profile we used the Defense Style Questionnaire (DSQ) (39), a rating scale designed to estimate behaviour indicative of 25 ego defense mechanisms. DSQ is the most widely used self-report method assessing defense mechanisms (40). Comparison of ratings using both DSQ and the observer-rated Defense Mechanism Rating Scales (DMRS) (41) showed significant correlations between measurements of DSQ and defences as assessed by DMRS (42). We used the standardised Greek version of DSQ which consists of 88-items on a 9-point Likert-type. It showed adequate internal consistency, test-retest reliability, and construct validity (33), and it has been widely used with Greek medical patients (25, 26, 28, 29).

Hostility: The Hostility and Direction of Hostility Questionnaire (HDHQ) (43) was used. It provides a measure of hostility that reflects an attitudinal personality trait and shows the participant's reaction to frustrating occurrences. The HDHQ has been used in the Greek population and with medical patients (26, 28, 29, 44).We have found that hostility features as measured by HDHQ were strongly negatively associated with physical HRQOL in systemic sclerosis (28).

Outcome (Health-Related Quality of Life)

Health-Related Quality of Life was assessed using the World Health Organisation Quality of Life Instrument, Short-Form (WHOQOL-BREF) (45,46). It assesses 4 domains: *Physical, Mental health, Social Relationships* and *Environment HRQOL*. Each item is rated on a 5-point Likert interval scale and the scores are transformed on a scale from 0 to 100. A higher score indicates better HRQOL. Data obtained from a survey of adults carried out in 23 countries including Greece showed that WHOQOL-BREF is a cross-culturally valid assessment of HRQOL (46). The WHOQOL-BREF was found to have adequate test-retest reliability, internal consistency and factor structure in people with rheumatological diseases (47). The Greek version of the WHOQOL-BREF (46,48) has been previously used as an outcome measure of HRQOL in Greek patients with rheumatic diseases (28, 29).

Statistical analyses

All the statistical analyses were performed using the Statistical Package for the Social Sciences (SPSS) 15.0 (SPSS Inc., Chicago, IL, USA) for Windows. Summary statistics for all variables were calculated. Normality was tested

Table I. Demographic characteristics, patients' disease duration and symptoms of psychological distress in patients with primary SS, SLE and healthy controls (HC).

	SS (n=40)	SLE (n=56)	HC (n=80)	*p*-value (SS *vs*. SLE)	*p*-value (SLE *vs*. HC)	*p*-value (SS *vs*. HC)
Age (years) (mean ± SD)	55.8 ± 11.1	43.1 ± 12.1	56.2 ± 11.5	<0.001*	<0.001*	0.861*
Years of education (mean±SD)	6.9 ± 3.1	10.5 ± 3.6	11.1 ± 4.0	<0.001*	0.377	<0.001*
Female gender, n. (%)	38 (95)	46 (82.1)	76 (95)	0.060¥	0.015¥	1.000¥
Marital status, n. (%)						
Single	1 (2.5)	11 (19.6)	3 (3.8)			
Married	35 (87.5)	44 (78.6)	53 (67.9)			
Divorced/Widowed/Separated	4 (10.0)	1 (1.8)	22 (28.2)	0.013¥	<0.001¥	0.065¥
Disease duration (years) (mean±SD)	9.2 ± 5.7	12.0 ± 8.1	–	0.059*	–	–
Psychological distress (mean±SD)						
Somatisation	1.65 ± 0.76	0.91 ± 0.77	0.93 ± 0.62	0.009†	0.461?	<0.001‡
Obsessive-compulsive	1.14 ± 0.67	0.78 ± 0.77	1.11 ± 0.62	0.128†	0.054?	0.891‡
Interpersonal sensitivity	1.17 ± 0.71	0.79 ± 0.74	0.89 ± 0.54	0.030†	0.273?	0.128‡
Hostility	0.79 ± 0.64	0.58 ± 0.72	0.74 ± 0.75	0.216†	0.198?	0.641‡
Anxiety	1.05 ± 0.80	0.63 ± 0.81	0.73 ± 0.67	0.145†	0.907?	0.170‡
Depression	1.13 ± 0.60	0.86 ± 0.80	1.06 ± 0.74	0.411†	0.405?	0.889‡
Phobic anxiety	0.45 ± 0.48	0.34 ± 0.56	0.43 ± 0.54	0.913†	0.517?	0.391‡
Paranoid ideation	1.33 ± 0.92	0.91 ± 0.80	1.22 ± 0.72	0.107†	0.088?	0.865‡
Psychoticism	0.65 ± 0.52	0.51 ± 0.61	0.48 ± 0.38	0.390†	0.963?	0.044‡
Global Symptom Index (GSI)	1.09 ± 0.55	0.73 ± 0.65	0.86 ± 0.49	0.039†	0.416?	0.048‡

*two-tailed *t*-tests; ¥chi-square tests; †One-way ANCOVA adjusted for age and education; ?One-way ANCOVA adjusted for age and sex; ‡One-way ANCOVA adjusted for education.

by the Kolmogorov-Smirnov test (49). Chi-square analyses for categorical data (*e.g*. sex) and two-tailed t-tests for continuous data (*e.g*. age) were carried out to assess the differences between primary SS patients and controls in major demographic, clinical and psychological variables. Since we found that primary SS patients differed from SLE patients in age and education, comparisons of psychological parameters and HRQOL between the two groups were made using one-way analyses of covariance (ANCOVA) adjusted for age and education. For similar reasons, comparison of psychological parameters and HRQOL between primary SS patients and healthy participants were carried out using ANCOVA adjusted for education (49).

To assess the relationship of primary SS patients' psychological defensive profile with HRQOL adjusting for demographic variables, disease duration and psychological distress, univariate comparisons were first conducted. Two-tailed *t*-tests were performed for dichotomous variables, and Pearson's or Spearman's correlations were calculated for continuous variables, as appropriate (49). To assess the variables most closely associated with SS patients' HRQOL, four independently produced multiple regression analyses were next performed, with dependent variables the four WHOQOL-BREF components. Selection of independent variables was based on the results of the previous univariate analyses and the most statistically significant variables were entered into the regression equations, taking into consideration the SS patient sample size. Colinearity between independent variables was tested based on variance inflation factors (VIF) and tolerances for individual variables (50).

Results

Sample characteristics and psychological distress symptoms

Table I presents the participants' demographic profile, disease duration and psychological distress symptoms. The female/male ratio of primary SS patients was 0.95. Thirty-five out of 40 of our patients complained of dry eyes, 30/40 presented dry mouth, while only 5 patients had recurrent parotid gland enlargement. None of our patients presented signs or symptoms of extraglandular or systemic manifestations. In addition, SS patients' complains for malaises were moderate and none patient had a diagnosis of concomitant fibromyalgia. SS patients had received less education than both SLE patients ($p<0.001$) and healthy participants ($p<0.001$) and were older that SLE patients ($p<0.001$). In view of these differences, subsequent comparisons were adjusted for education or for both education and age, as appropriate.

Patients with primary SS presented higher rates of symptoms of general psychological distress as measured by the SCL-90R Global Symptom Index, compared to both SLE ($p=0.039$) and healthy participants ($p=0.048$), even after adjustment for confounders. Inspection of SCL-90R subscales revealed that SS patients presented more symptoms of somatisation than both SLE patients and healthy participants ($p=0.009$ and $p<0.001$, respectively), while symptoms of interpersonal sensitivity were more prominent in SS than in SLE patients ($p=0.030$), and psychoticism symptoms were also higher in primary SS patients compared to healthy participants ($p=0.044$). Although the mean scores of SLE patients on Global Symptom Index and on most SCL-90R subscales were lower than that of healthy participants, the differences failed to reach statistical significance after controlling for age and sex.

HRQOL measurements

As shown in Table II, primary SS patients presented significantly more

Table II. Health-related quality of life among patients with primary SS (n=40), SLE (n=56), and healthy participants (n=80).

						Adjusted for age and education			
		Mean	SE	F	*p*-value	Adj. mean	SE	F	*p*-value
Physical HRQOL	SS	54.6 *	3.1	15.1	<0.0005	60.0	2.9	10.9	<0.0005
	SLE	65.8 *	2.3			63.8	2.5		
	Healthy participants	72.9 *	1.8			73.0	2.0		
Mental HRQOL	SS	58.1 *	3.1	19.1	<0.0005	59.0	2.8	14.8	<0.0005
	SLE	63.5 *	2.4			63.0	2.4		
	Healthy participants	76.5 *	1.6			76.3	2.0		
Social relations HRQOL	SS	51.9 *	3.1	16.7	<0.0005	53.3	3.3	12.3	<0.0005
	SLE	62.6 *	1.9			62.2	2.8		
	Healthy participants	73.4 *	1.8			73.0	2.3		
Environment HRQOL	SS	54.2 *	3.1	5.8	0.004	56.1	2.5	3.4	0.035
	SLE	58.1 **	1.9			57.4	2.2		
	Healthy participants	63.8 *	1.5			63.3	1.8		

*Statistically significant differences at $p<0.01$ level among subgroups; Bonferroni post-hoc tests.

Table III. Defense mechanisms of patients with primary SS, SLE and healthy controls (HC).

	SS (n=40)	SLE (n=56)	HC (n=80)	SS *vs*. SLE*	SS *vs*. HC¥
Passive aggression	2.25 ± 1.79	2.84 ± 1.33	3.11 ± 1.53	*p*=0.073	*p*=0.108
Projection	3.75 ± 1.80	3.06 ± 1.31	3.27 ± 1.37	*p*=0.227	*p*=0.336
Regression	4.58 ± 2.44	3.97 ± 2.61	4.84 ± 2.38	*p*=0.115	*p*=0.622
Inhibition	4.76 ± 2.23	4.38 ± 1.73	4.53 ± 1.54	*p*=0.973	*p*=0.494
Projective identification	2.50 ± 2.92	2.01 ± 1.96	2.47 ± 2.35	*p*=0.648	*p*=0.533
Acting out	3.96 ± 2.42	3.75 ± 2.04	4.58 ± 1.94	*p*=0.352	*p*=0.478
Withdrawal	5.25 ± 2.78	4.85 ± 2.69	5.22 ± 2.35	*p*=0.222	*p*=0.838
Schizoid fantasy	3.70 ± 3.43	2.67 ± 2.73	3.72 ± 2.95	*p*=0.145	*p*=0.506
Help rejecting complains	4.37 ± 2.83	3.10 ± 1.94	3.41 ± 1.82	*p*=0.019	*p*=0.044
Undoing	4.48 ± 2.17	3.98 ± 2.14	4.59 ± 2.14	*p*=0.230	*p*=0.987
Omnipotence	3.17 ± 1.86	3.09 ± 1.72	3.32 ± 1.82	*p*=0.844	*p*=0.325
Denial	4.37 ± 2.11	4.10 ± 1.76	4.07 ± 1.94	*p*=0.666	*p*=0.296
Splitting	3.85 ± 2.04	4.08 ± 2.17	4.25 ± 2.02	*p*=0.283	*p*=0.681
Primitive idealisation	4.37 ± 2.63	3.77 ± 2.11	4.00 ± 2.58	*p*=0.393	*p*=0.265
Isolation	3.23 ± 1.98	3.62 ± 1.59	3.98 ± 1.63	*p*=0.732	*p*=0.159
Pseudoatruism	7.82 ± 2.25	7.67 ± 1.81	7.47 ± 2.16	*p*=0.762	*p*=0.407
Reaction formation	4.87 ± 1.97	4.93 ± 1.56	5.57 ± 1.66	*p*=0.644	*p*=0.363
Humour	2.35 ± 1.51	3.26 ± 1.51	4.01 ± 1.68	*p*=0.048	*p*=0.001
Affiliation	5.60 ± 3.09	4.85 ± 2.88	5.52 ± 2.47	*p*=0.379	*p*=0.964
Sublimation	5.77 ± 3.59	4.64 ± 3.48	5.07 ± 3.19	*p*=0.717	*p*=0.106
Suppression	5.01 ± 2.66	5.27 ± 2.18	5.32 ± 1.69	*p*=0.548	*p*=0.873
Task orientation	5.77 ± 2.59	5.41 ± 2.30	6.42 ± 2.24	*p*=0.298	*p*=0.679
Anticipation	6.03 ± 2.92	6.29 ± 2.42	6.92 ± 1.90	*p*=0.356	*p*=0.244

*One-way ANCOVA adjusted for age and education; ¥One-way ANCOVA adjusted for education.

impaired HRQOL than healthy participants in all four WHOQOL-BREF components, even after controlling for confounders. Comparisons with the SLE sample showed that SS patients presented also more impaired *Physical* and *Social Relations* HRQOL, but these differences seem to be weakened after controlling for age and education, especially with respect to physical HRQOL.

Defensive profile and hostility as response to frustrating occurrences

As shown in Table III, primary SS patients presented less use of *humour defense* (p=0.048 and p<0.001) and more *help-rejecting complains* as assessed by the Defense Style Questionnaire, compared to both SLE and healthy participants (p=0.019 and p=0.044, respectively). Also, primary SS patients presented higher rates on *delusional guilt hostility* as assessed by the Hostility and Direction of Hostility Questionnaire compared to both SLE and healthy participants (p=0.049 and p=0.003, respectively), and higher rates on *delusional hostility* than healthy controls (p=0.007) (Table IV).

Psychological factors associated with primary SS patients' HRQOL

Tables V and VI present the univariate and multivariate associations of psychological variables studied with the four components of WHOQOL-BREF in the primary SS patients. As shown in Table V, among the variables associated with *Physical* HRQOL, less use of *humor* defense (p<0.001), higher rates on *delusional guilt hostility* (p=0.032) and higher psychological distress (p=0.049) were the variables most closely independently associated with

Table IV. Hostility features as response to frustrating occurrences in patients with primary SS, SLE and "healthy" controls (HC).

	SS (n=40)	SLE (n=56)	HC (n=80)	SS *vs*. SLE*	SS *vs*. HC¥
Acting-out hostility	4.10 ± 1.39	3.82 ± 1.90	3.56 ± 1.62	*p*=0.136	*p*=0.532
Criticism of others	5.81 ± 2.01	5.75 ± 2.18	6.19 ± 2.20	*p*=0.990	*p*=0.359
Delusional hostility	3.35 ± 1.71	2.69 ± 2.13	1.88 ± 1.60	*p*=0.645	*p*=0.007
Self-criticism	4.21 ± 2.33	3.98 ± 2.27	3.92 ± 1.95	*p*=0.447	*p*=0.938
Delusional guilt hostility	3.18 ± 1.63	2.42 ± 1.73	2.00 ± 1.37	*p*=0.049	*p*=0.003
Extraverted hostility	13.27 ± 3.76	12.26 ± 5.11	11.64 ± 4.15	*p*=0.458	*p*=0.379
Introverted hostility	11.62 ± 5.87	10.39 ± 5.71	9.84 ± 4.85	*p*=0.252	*p*=0.425
Total hostility	20.67 ± 6.45	18.67 ± 8.00	17.56 ± 6.00	*p*=0.255	*p*=0.201

*One-way ANCOVA adjusted for age and education; ¥One-way ANCOVA adjusted for education.

impaired *Physical* HRQOL. Similarly, impaired *Mental* HRQOL was closely associated only with higher rates of psychological distress, as measured by the SCL-90-R Global Symptom Index (*p*=0.024). General psychological distress was also the major independent correlate of *Social Relations* HRQOL (*p*=0.05), while lower education level (*p*=0.021) and more use of the *schizoid fantasy* defense (*p*=0.005) were the variables most closely associated with the *Environment* HRQOL (Table VI).

Table V. Demographic, clinical and psychological parameters associated with physical and mental HRQOL in patients with primary SS (n=40).

	Physical HRQOL			Mental HRQOL		
Independent	Univariate Analyses*	Multiple Regression¥		Univariate Analyses*	Multiple Regression¥	
Variables	*p*-value	beta†	*p*-value	*p*-value	beta†	*p*-value
Age	0.169			0.735		
Sex	0.259			0.192		
Educational level	0.203			0.154		
Divorced/Widowed/Sep.	0.206			0.123		
Disease duration	0.516			0.856		
SCL-90-R GSI‡	0.002	-0.365	0.049	<0.0005	-0.429	0.024
Defense mechanisms						
Passive aggression	0.114			0.166		
Projection	0.086			0.078		
Regression	0.489			0.932		
Acting out	0.049	-0.025	0.861	0.415		
Withdrawal	0.445			0.315		
Schizoid fantasy	0.061			0.036	-0.069	0.608
Help-rejecting complains	0.606			0.777		
Omnipotence	0.512			0.092		
Denial	0.697			0.983		
Splitting	0.399			0.451		
Pseudoatruism	0.185			0.668		
Reaction formation	0.308			0.515		
Humour	0.033	0.597	<0.001	0.162		
Suppression	0.325			0.418		
Hostility features						
Acting out	0.592			0.228		
Criticism of others	0.795			0.978		
Delusional hostility	0.072			0.003	-0.061	0.672
Self-criticism	0.122			0.013		
Delusional guilt	0.003	-0.395	0.032	<0.0005	-0.294	0.141
Cumulative R^2 Adj.		0.523			0.545	
ANOVA		$F_{(4,35)} = 10.5$, $p<0.0005$			$F_{(4,35)} = 9.3$, $p<0.0005$	

*Pearson or Spearman correlations and two-tailed *t*-tests, as appropriate; ¥Two independently produced multiple regression analyses with dependent variables the "physical HRQOL" and "mental HRQOL". Due to the small number of patients, the most significant defenses are presented and independent variables were the most significant variables resulted from univariate analyses; †Standardised beta coefficients; ‡Global Sympom Index.

Discussion

The results of the present study showed that primary SS patients presented elevated symptoms of psychological distress as measured by the SCL-90R Global Symptom Index compared to both SLE and healthy participants, in accordance to previous findings (8-11). SS patients presented also more symptoms of interpersonal sensitivity than SLE patients, less use of *humour* defense and more *help-rejecting complains* and *delusional guilt* compared to both SLE and healthy participants, indicating that SS patients may exhibit some difficulties in adaptation to life stressors and in interpersonal relationships. In line with previous findings (15, 17-22), SS patients' HRQOL was more impaired than healthy participants and comparable to SLE patients' HRQOL. General psychological distress was a constant independent correlate of most aspects of SS patients' health status, confirming a recent report (20), while less use of *humour* and higher rates of *delusional guilt* were significantly associated with impaired *Physical* HRQOL independently of psychological distress. To the best of our knowledge, this is the first study reporting that aspects of the SS patients' underlying personality structure are independently associated with *Physical* HRQOL.

A number of studies have shown that SS patients often present elevated rates of psychological distress, compared to healthy controls or patients with other rheumatic diseases (8-11), in line with our findings, and Stevenson and col-

Table VI. Demographic, clinical and psychological parameters associated with social relations and environment HRQOL in patients with primary Sjögren's syndrome (n=40).

	Social Relations HRQOL			Environment HRQOL		
Independent	Univariate Analyses*	Multiple Regression¥		Univariate Analyses*	Multiple Regression¥	
Variables	*p*-value	beta†	*p*-value	*p*-value	beta†	*p*-value
Age	0.339			0.483		
Sex	0.439			0.191		
Educational level	0.204			0.018	0.324	0.021
Divorced/Widowed/Sep.	0.496			0.099		
Disease duration	0.716			0.785		
SCL-90-R GSI‡	<0.0005	-0.410	0.050	0.004	-0.169	0.396
Defense mechanisms						
Passive aggression	0.506			0.409		
Projection	0.262			0.103		
Regression	0.952			0.721		
Acting out	0.384			0.316		
Withdrawal	0.519			0.338		
Schizoid fantasy	0.154			<0.0005	-0.437	0.005
Help-rejecting complains	0.238			0.887		
Omnipotence	0.373			0.333		
Denial	0.430			0.788		
Splitting	0.716			0.290		
Pseudoatruism	0.694			0.280		
Reaction formation	0.230			0.951		
Humour	0.287			0.109		
Suppression	0.649			0.317		
Hostility features						
Acting-out	0.452			0.800		
Criticism of others	0.499			0.692		
Delusional hostility	0.009	-0.184	0.273	0.068		
Self-criticism	0.047	0.199	0.332	0.078		
Delusional guilt	0.001	-0.275	0.254	0.010	-0.086	0.664
Cumulative R^2 Adj.	0.312			0.407		
ANOVA	$F_{(4,35)} = 4.9$, p=0.003			$F_{(4,34)} = 7.1$, p<0.0005		

*Pearson or Spearman correlations and two-tailed *t*-tests were, as appropriate; ¥Two independently produced multiple regression analyses with dependent variables the "physical HRQOL" and "mental HRQOL". Due to the small number of patients, the most significant defenses are presented and independent variables were the most significant variables resulted from univariate analyses. †Standardised beta coefficients; ‡Global Symptom Index.

leagues showed that patients with SS are at increased risk for developing depression (11). Since psychological distress is a strong independent correlate of HRQOL, as the present results showed, early recognition and treatment of psychological distress is important in order to reduce the negative impact of psychological distress on HRQOL and to prevent further deterioration.

An additional finding of the present study is that SLE patients reported fewer symptoms of psychological distress than healthy participants, a finding that contradicts the results of most studies on depression and SLE (51, 52). However, since we matched our healthy-control sample with SS patients in mind, the SLE patient sample included fewer females and comprised younger participants than the healthy control sample. Therefore, when we adjusted for age and sex, the previously observed differences failed to reach statistical significance. Even so, our finding that SLE patients did not report elevated rates of psychological distress indicates that further research is needed to better clarify the prevalence of psychological distress in SLE in relation to the general population.

Among the various subscales of the SCL-90R assessing specific aspects of psychological distress, symptoms of somatisation are particularly important, being more prominent in SS than in SLE or healthy participants. Several assumptions could be drawn regarding the reason of SS patients presenting with more bodily symptoms: these symptoms might be primarily possible indicators of the disease activity. Indeed, several somatisation symptoms as assessed by the SCL-90-R somatisation subscale resemble true physical symptoms of the SS process (*e.g.* soreness of the muscles, faintness, heavy feelings in the arms or legs etc.). Alternatively, somatisation symptoms have been considered as manifestations of anxiety and depression, as antidepressant treatment led to a reduction of scores of all these dimensions (53). Another possible explanation is that some patients with physical illness also have a high number of bodily symptoms probably unrelated to the underlying disorder, and it is these additional symptoms that are associated with impaired HRQOL (54, 55). Probably, more important is the combined effect of the physical effects of the disease process and the psychological reaction that this invokes in the individual. Therefore, apart from early addressing general psychological distress, rheumatologists should consider the somatisation process when evaluating SS patients' physical status, since early attention to this process could result in a more proper treatment and may help avoid unnecessary investigations, or inefficient and expensive interventions.

Our new findings that ego defense mechanisms (*i.e.* less use of *humour* and more use of *schizoid fantasy*) and hostility features (*i.e.* higher rates of *introverted hostility* and *delusional guilt*) were independently associated with impaired health status, underline the role of aspects of personality in SS patients' well being. *Humour* constitutes one of the cornerstone defenses comprising an adaptive defensive profile (39). It reflects a capacity to accept a conflictual and stressful situation while taking the edge off its painful aspects (39), allowing one to bear and yet focus on what is too stressful or too terrible to emerge (56), such as, for instance, the stressful feelings provoked by a severe medical illness and its impact on patient's everyday life. *Humour* also often involves an

element of self-observation or truth, and tends to relieve tension in a way that allows everyone to share in it, rather than being at one person's expense (57). On the contrary, in *schizoid fantasy*, the individual deals with stressors by excessive daydreaming as a substitute for human relationships or problem solving (57), while delusional guilt is a measure of the individual's intropunitiveness, *i.e.* a measure of the hostility directed inward on the self, connected with melancholic personality (58, 59). Delusional guilt is associated with shyness and the degree of distress felt in a variety of social situations (60), and it has been associated with poor adherence to treatment (61, 62). Taken together, it seems that SS patients with low capacity to deal with inner or external stressors by adopting a more adaptive defensive style and those who hold an intropunitive attitude towards a frustrating occurrence, such as the disease, might present higher risk for distraction from the issue in hand, *i.e.* adaptation to the disease, or they may even present difficulties in adherence to treatment. In this ways the patient's underlying personality organisation might be linked with *physical* HRQOL.

The main limitations of our study are the sample size of SS patients and the cross-sectional design. The findings need to be replicated with a larger sample in a prospective study. Moreover, the drawback of using only self-report measures means that we cannot refute the criticism that an underlying response style might have led to our results. Additionally, the inventory used to assess defensive profile (DSQ) is an attempt to describe an inferred intrapsychic phenomenon that may be out of a subject's awareness, an attempt that is fraught with difficulty (39). Studies on the validity of DSQ, though, showed that defenses assessed by DSQ were significantly correlated with defenses assessed by observer-rated scales (43), while a review of published studies indicated strong evidence that adaptiveness of defense style, as measured by the DSQ, correlates with mental health and change (40). It is also possible that other factors not included in the present study, such as social support, may also have an impact on HRQOL.

Strengths of our study include the reasonably high response rate (67.8%), and that there were no statistically significant differences between responders and non-responders; also we did use well-recognised instruments for all our measures, while our secondary and tertiary hospital provides care to the majority of the SS patients of the catchment area, suggesting that we recruited a representative sample of primary SS patients.

The main clinical implication of our study is that, apart from the early assessment and treatment of psychological distress symptoms, especially somatisation symptoms, clinicians and consultation-liaison psychiatrists should bear in mind the patients' psychological resources and coping capacities to deal with the stress of the disease, since such personality traits, although usually underestimated, are independently associated with the disease's outcome. Future longitudinal studies with larger samples are needed to confirm these associations and to investigate the specific paths that form the patients' HRQOL, especially with respect to those patients who are coping poorly with the illness because they lack the psychological resources to do so, in order to schedule proper psychotherapeutic interventions aiming to improve patients' HRQOL.

References

1. DANIELS TE, FOX PC: Salivary and oral components of Sjögren's syndrome. *Rheum Dis Clin North Am* 1992; 18: 572-89.
2. FOX RI: Sjögren's syndrome. *Lancet* 2005; 366: 321-31.
3. TZIOUFAS AG, MITSIAS ID, MOUTSOPOULOS HM: Sjögren's syndrome. *In*: HOCHBERG MC, SILMAN AJ, SMOLEN JS, WEINBLATT ME, WEISMAN MH (Eds.) Rheumatology, vol. II. 4th ed. Mosby: Elsevier; 2008; 1341-52.
4. VOULGARELIS M, TZIOUFAS AG, MOUTSOPOULOS HM: Mortality in Sjögren's syndrome. *Clin Exp Rheumatol* 2008; 26: S66-71.
5. MALINOW KL, MOLINA R, GORDON B, SELNES OA, PROVOST TT, ALEXANDER EL: Neuropsychiatric dysfunction in primary Sjögren's. *Ann Intern Med* 1985; 103: 344-50.
6. MAUCH E, VÖLK C, KRATZSCH G *et al*.: Neurological and neuropsychiatric dysfunction in primary Sjögren's syndrome. *Acta Neurol Scand* 1994; 89: 31-5.
7. HARBOE E, TJENSVOLL AB, MARONI S *et al*.: Neuropsychiatric syndromes in patients with systemic lupus erythematosus and primary Sjögren syndrome: a comparative population-based study. *Ann Rheum Dis* 2009; 68: 1541-6.
8. DROSOS AA, ANDONOPOULOS AP, LAGOS G, ANGELOPOULOS NV, MOUTSOPOULOS HM: Neuropsychiatric abnormalities in primary Sjögren's. *Clin Exp Rheumatol* 1989; 7: 207-9.
9. VALTÝSDÓTTIR ST, GUDBJÖRNSSON B, LINDQVIST U, HÄLLGREN R, HETTA J: Anxiety and depression in patients with primary Sjögren's syndrome. *J Rheumatol* 2000, 27: 165-9.
10. VALTÝSDÓTTIR ST, GUDBJÖRNSSON B, HÄLLGREN R, HETTA J: Psychological well-being in patients with primary Sjögren's syndrome. *Clin Exp Rheumatol* 2000; 18: 597-600.
11. STEVENSON HA, JONES ME, ROSTRON JL, LONGMAN LP, FIELD EA: UK patients with primary Sjögren's syndrome are at increased risk from clinical depression. *Gerodontology* 2004; 21: 141-5.
12. GODAERT GL, HARTKAMP A, GEENEN R *et al*.: Fatigue in daily life in patients with primary Sjögren's syndrome and systemic lupus erythematosus. *Ann N Y Acad Sci* 2002; 966: 320-6.
13. VAN OERS ML, BOSSEMA ER, THOOLEN BJ *et al*.: Variability of fatigue during the day in patients with primary Sjögren's syndrome, systemic lupus erythematosus, and rheumatoid arthritis. *Clin Exp Rheumatol* 2010; 28: 715-21.
14. HELMICK CG, FELSON DT, LAWRENCE RC *et al*.: Estimates of the prevalence of arthritis and other rheumatic conditions in the United States. Part I. *Arthritis Rheum* 2008; 58: 15-25.
15. THOMAS E, HAY EM, HAJEER A, SILMAN AJ: Sjögren's syndrome: a community-based study of prevalence and impact. *Br J Rheumatol* 1998; 37: 1069-76.
16. ALAMANOS Y, TSIFETAKI N, VOULGARI PV, VENETSANOPOULOU AI, SIOZOS C, DROSOS AA: Epidemiology of primary Sjögren's syndrome in north-west Greece, 1982-2003. *Rheumatology* 2006; 45: 187-91.
17. SEGAL B, BOWMAN SJ, FOX PC *et al*.: Primary Sjögren's Syndrome: health experiences and predictors of health quality among patients in the United States. *Health Qual Life Outcomes* 2009; 7: 46.
18. STEWART AL, GREENFIELD S, HAYS RD *et al*.: Functional status and well-being of patients with chronic conditions. Results from the Medical Outcomes Study. *JAMA* 1989; 262: 907-13.
19. MEIJER JM, MEINERS PM, HUDDLESTON SLATER JJ *et al*.: Health-related quality of life, employment and disability in patients with Sjögren's syndrome. *Rheumatology* (Oxford) 2009; 48: 1077-82.
20. INAL V, KITAPCIOGLU G, KARABULUT G, KESER G, KABASAKAL Y: Evaluation of quality of life in relation to anxiety and depression in primary Sjögren's syndrome. *Mod Rheumatol* 2010. DOI 10.1007/s10165-010-0329-z.
21. STRÖMBECK B, EKDAHL C, MANTHORPE R, WIKSTRÖM I, JACOBSSON L: Health-related quality of life in primary Sjögren's syndrome, rheumatoid arthritis and fibromyalgia

compared to normal population data using the SF-36. *Scand J Rheumatology* 2000; 29: 20-8.
22. SUTCLIFFE N, STOLL T, PYKE S, ISENBERG DA: Functional disability and end organ damage in patients with systemic lupus erythematosus (SLE), SLE and Sjögren's syndrome (SS), and primary SS. *J Rheumatol* 1998; 25: 63-8.
23. NG WF, BOWMAN SJ: Primary Sjögren's syndrome: too dry and too tired. *Rheumatology* (Oxford) 2010; 49: 844-53.
24. NICHOL MB, ZHANG L: Depression and health-related quality of life in patients with rheumatoid arthritis. *Expert Rev Pharmacoecon Outcomes Res* 2005; 5: 645-53.
25. HYPHANTIS TN, BAI M, SIAFAKA V *et al*.: Psychological distress and personality traits in early rheumatoid arthritis: A preliminary survey. *Rheumatol Int* 2006; 26: 828-36.
26. HYPHANTIS TN, TRIANTAFILLIDIS JK, PAPPA S *et al*.: Defense mechanisms in inflammatory bowel disease. *J Gastroenterol* 2005; 40: 24-30.
27. BERESFORD TP, ALFERS J, MANGUM L, CLAPP L, MARTIN B: Cancer survival probability as a function of ego defense (adaptive) mechanisms versus depressive symptoms. *Psychosomatics* 2006; 47: 247-53.
28. BAI M, TOMENSON B, CREED F *et al*.: The role of psychological distress and personality variables in the disablement process in rheumatoid arthritis. *Scand J Rheumatol* 2009; 38: 419-30.
29. HYPHANTIS TN, TSIFETAKI N, SIAFAKA V *et al*.: The impact of psychological functioning upon systemic sclerosis patients' quality of life. *Semin Arthritis Rheum* 2007; 37: 81-92.
30. VITALI C, BOMBARDIERI S, JONSSON R *et al*.: European Study Group on Classification Criteria for Sjögren's Syndrome. Classification criteria for Sjögren's syndrome: a revised version of the European criteria proposed by the American-European Consensus Group. *Ann Rheum Dis* 2002; 61: 554-8.
31. STRÖMBECK B, JACOBSSON LT: the role of exercise in the rehabilitation of patients with systemic lupus erythematosus and patients with primary Sjogran's syndrome. *Curr Opin Rheumatol* 2007; 19: 197-203.
32. HOCHBERG MC: Updating the American College of Rheumatology revised criteria for the classification of systemic lupus erythematosus. *Arthritis Rheum* 1997; 40: 1725.
33. HYPHANTIS T: The Greek version of the Defense Style Questionnaire: Psychometric properties in three different samples. *Comprehensive Psychiatry* 2010; 51: 618-29.
34. CHARLSON ME, POMPEI P, ALES KL, MACKENZIE CR: A new method of classifying prognostic comorbidity in longitudinal studies: development and validation. *J Chronic Dis* 1987; 40: 373-83.
35. DEROGATIS LR: SCL-90-R: Administration, scoring, and procedure manual, 3rd Ed. National Computer Systems: Minneapolis, MN, 1994.
36. PARKER JC, BUCKELEW SP, SMARR KL *et al*.: Psychological screening in rheumatoid arthritis. *J Rheumatol* 1990; 17: 1016-21.
37. DONIAS S, KARASTERGIOU A, MANOS N: Standardization of the symptom checklist-90-R rating scale in a Greek population (in Greek with English abstract). *Psychiatriki* 1991; 2: 42-8.
38. AMERICAN PSYCHIATRIC ASSOCIATION: Diagnostic and Statistical Manual of Mental Disorders. 4th ed.Washington, DC: American Psychiatric Press, 1994.
39. BOND M: An empirical study of defense styles: the Defense Style Questionnaire. *In*: VAILLANT GE, eds. *Ego mechanisms of defense: a guide for clinicians and researchers*. Washington DC, American Psychiatric Press, 1992.
40. BOND M: Empirical studies of defense style: relationships with psychopathology and change. *Harv Rev Psychiatry* 2004; 12: 263-78.
41. PERRY JC: Defense Mechanism Rating Scales (DMRS). Ed. 5. Cambridge, MA: J.C. Perry, 1990.
42. BOND M, PERRY JC, GAUTIER M, GOLDBERG M, OPPENHEIMER J, SIMAND J: Validating the self-report of defense styles. *J Pers Dis* 1989; 3: 101-12.
43. CAINE TM, FOULDS GA, HOPE K: Manual of Hostility and Direction of Hostility Questionnaire. London, University of London Press, 1967.
44. DROSOS AA, ANGELOPOULOS NV, LIAKOS A, MOUTSOPOULOS HM: Personality structure disturbances and psychiatric manifestations in primary Sjögren's syndrome. *J Autoimmun* 1989; 2: 489-93.
45. The WHOQOL Group: Development of the World Health Organization WHOQOL-BREF quality of life assessment. *Psychol Med* 1998; 28: 551-8.
46. SKEVINGTON SM, LOTFY M, O'CONNELL KA, WHOQOL GROUP: The World Health Organization's WHOQOL-BREF quality of life assessment: psychometric properties and results of the international field trial. A report from the WHOQOL group. *Qual Life Res* 2004; 13: 299-310.
47. TAYLOR WJ, MYERS J, SIMPSON RT, MCPHERSON KM, WEATHERALL M: Quality of life of people with rheumatoid arthritis as measured by the World Health Organization Quality of Life Instrument, short form (WHOQOL-BREF): score distributions and psychometric properties. *Arthritis Rheum* 2004; 51: 350-7.
48. GINIERI-COCCOSSIS M, TRIANTAFILLOU E, ANTONOPOULOU V, TOMARAS B, CHRISTODOULOU G: Quality of life in relation to WHOQOL-100 questionnaire (in Greek). Beta Medical Arts Athens, 2001.
49. ALTMAN DG: Practical statistics for medical research. London, Chapman and Hall, 1991.
50. MILES J, SHEVLIN M: Applying regression and correlation. Sage, London, 2003, pp 165-91.
51. BARBOSA F, MOTA C, ALVES M *et al*.: Alexithymia in systemic lupus erythematosus patients. *Ann N Y Acad Sci* 2009; 1173: 227-34.
52. NERY FG, BORBA EF, HATCH JP *et al*.: Major depressive disorder and disease activity in systemic lupus erythematosus. *Compr Psychiatry* 2007; 48:14-9.
53. CHATURVEDI SK, MAGUIRE GP: Persistent somatization in cancer: a controlled follow-up study. *J Psychosom Res* 1998; 45: 249-56.
54. KROENKE K, SPITZER RL, DEGRUY FV 3RD *et al*.: Multisomatoform disorder. An alternative to undifferentiated somatoform disorder for the somatizing patient in primary care. *Arch Gen Psychiatry* 1997; 54: 352-8.
55. FEDER A, OLFSON M, GAMEROFF M *et al*.: Medically unexplained symptoms in an urban general medicine practice. *Psychosomatics* 2001; 42: 261-8.
56. MEISSNER WW: Meissner's glossary of defenses. *In*: VAILLANT GE, eds. Ego mechanisms of defense: a guide for clinicians and researchers. Washington DC, American Psychiatric Press, 1992: pp. 239-51.
57. PERRY JC: Parry's Defense Mechanism Rating Scale (March 1990). *In*: VAILLANT GE, eds. Ego mechanisms of defense: a guide for clinicians and researchers. Washington DC, American Psychiatric Press, 1992: pp. 253-9.
58. FOULDS GA: Personality and Personal Illness, Tavistock Press, London (1965).
59. PHILIP AE: The development and use of the hostility and direction of hostility questionnaire. *J Psychosom Res* 1969; 13: 283-7.
60. ARRINDELL WA, HAFKENSCHEID AJP, EMMELKAM PM: The Hostility and Direction of Hostility Questionnaire (HDHQ): a psychometric evaluation in psychiatric outpatients. *Person Individ Diff* 1984; 5: 221-31.
61. PUGH R: An association between hostility and poor adherence to treatment in patients suffering from depression. *Br J Med Psychol* 1983; 56: 205-8.
62. HYPHANTIS T, KALTSOUDA A, TRIANTAFILLIDIS J *et al*: Personality correlates of adherence to type 2 diabetes regimens. *Int J Psychiatry Med* 2005; 3: 103-7.