

Patient-reported outcome measures on mental health and psychosocial factors in patients with Brugada syndrome

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Aims

Brugada syndrome (BrS) is a hereditary arrhythmic disease, associated with sudden cardiac death. To date, little is known about the psychosocial correlates and impacts associated with this disease. The aim of this study was to assess a set of patient-reported psychosocial outcomes, to better profile these patients, and to propose a tailored psychosocial care.

Methods and results

Patients were recruited at the European reference Centre for BrS at Universitair Ziekenhuis Brussel, Belgium. Recruitment was undertaken in two phases: phase 1 (retrospective), patients with confirmed BrS, and phase 2 (prospective), patients referred for ajmaline testing who had an either positive or negative diagnosis. BrS patients were compared to controls from the general population. Two hundred and nine questionnaires were analysed (144 retrospective and 65 prospective). Collected patient-reported outcomes were on mental health (12 item General Health Questionnaire; GHQ-12), social support (Oslo Social Support Scale), health-related quality of life, presence of Type-D personality (Type-D Scale; DS14), coping styles (Brief-COPE), and personality dimensions (Ten Item Personality Inventory). Results showed higher mental distress (GHQ-12) in BrS patients (2.53 ± 3.03) than in the general population ($P < 0.001$) and higher prevalence (32.7%) of Type D personality ($P < 0.001$) in patients with confirmed Brugada syndrome (BrS +). A strong correlation was found in the BrS + group (0.611 , $P < 0.001$) between DS14 negative affectivity subscale and mental distress (GHQ-12).

Conclusion

Mental distress and type D personality are significantly more common in BrS patients compared to the general population. This clearly illustrates the necessity to include mental health screening and care as standard for BrS.

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Graphical Abstract

Patient reported outcome measures (PROMs) on mental health and psychosocial factors in patients with Brugada Syndrome

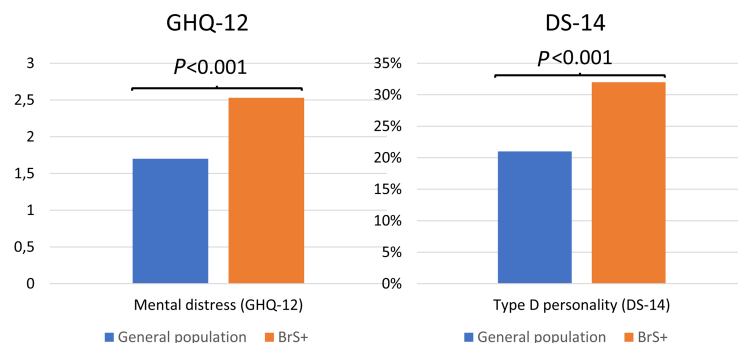
CONSORT diagram for retrospective and prospective phase

Retrospective phase (BrS+)
n = 144
40% female

Prospective phase (BrS+ or BrS-)
n = 65
18 BrS+
47 BrS-
43% female

Total study sample
n = 209
BrS+ = 162
BrS- = 47
41% female

Higher mental distress (GHQ-12) and higher prevalence of type D personality in BrS patients compared with the general population



Keywords

Brugada syndrome • patient-reported outcome measures • mental health • Type D personality

What's new?

- Patients with Brugada syndrome experience higher mental distress than the general population.
- Type D personality (distressed, with a strong tendency to have negative emotions across many situations but avoiding to express these) is significantly more common in patients with Brugada syndrome.
- Results from this study highlight the need for routine assessment and support for mental health in this patient population.

Introduction

Brugada syndrome (BrS) is a genetically inherited arrhythmogenic disease that poses an elevated risk for sudden cardiac death. Diagnosis of BrS often occurs following family screening after the unexpected death of a relative.¹

Cardiac ion channel dysfunction plays an important role in the pathogenesis of BrS in 20% of BrS patients.² The most commonly associated mutated (causal) gene in BrS is *SCN5A*, which accounts for 20% of molecularly confirmed cases of BrS.^{3–5} Common variants in other genes presumably underly the BrS phenotype in the 80% of the patients, and these may also lead to some form of 'cardiac ion channel dysfunction'. Interestingly, research has suggested various potential connections between psychological factors and BrS. There is an overlap between genes implicated in both BrS and a subset of patients with recent onset schizophrenia spectrum disorders,^{4,6} and there is increasing evidence that central pathways that convert stress into autonomic effects on the heart may be considered therapeutic targets.⁷ On the other hand, BrS diagnosis itself might prompt or unmask pathological psychological mechanisms.

To gain a better understanding of the relationship between psychological factors and BrS, further research is needed.

Patient-reported outcome measures (PROMs) are emerging tools in evaluating psychological and emotional impact of heart diseases.⁸ Addressing psychosocial aspects such as mental distress and social

support can improve the overall quality of life for patients affected by the BrS, impacting treatment adherence, health outcomes, and ability to cope with diagnosis.^{9–12} Differences in personality traits or the prevalence of a certain personality type, such as Type D personality (the combination of a tendency towards negative affectivity and social inhibition), may inform about the risk of mental health problems and the associated need for mental health screening. Several studies looking into inherited cardiac conditions have shown that including PROMs is useful to provide whole-patient care.^{9,13,14} However, the value of PROMs to guide a tailored care and psychosocial support in BrS is unknown.^{11,15}

Therefore, the aim of this study is to examine PROMs on mental health, personality type, coping style, psychological functioning, quality of life, and perceived social support in BrS. Moreover, the BrS cohort is compared with a sample of the general population (non-BrS).

Methods

Design and study population

All consecutive patients diagnosed with BrS following current guidelines and included in the UZ Brussel monocentric BrS registry (NCT05283759) were screened.¹⁶ They were included in two stages of the diagnostic process: retrospective and prospective. In the retrospective stage, patients with confirmed BrS diagnosis were selected from the UZ Brussel monocentric BrS registry. They were contacted by email and invited to fill out an online questionnaire. Patients aged under 18 and those without valid e-mail address were excluded. In the prospective stage, all patients (aged 18 or older) who were planned for an ajmaline test were asked to complete the questionnaire before the test (either online or on paper in-hospital).¹⁷ The prospective stage resulted in a group with BrS [ajmaline positive, people who tested positive for BrS (BrS+)] and a group without BrS [ajmaline negative, people who tested negative for BrS (BrS-), control group]. Through the implementation of both a retrospective phase involving confirmed diagnosis (BrS+) and a prospective phase preceding potential diagnosis, we were able to examine the impact of BrS diagnosis on patients' self-reported outcome measures in comparison to a control group that underwent identical procedures (i.e. ajmaline testing). Ethical approval for this study was granted by the UZ Brussels Committee for Medical Ethics (approval number B.U.N.

143201732280). All participants gave informed consent, and data were stored on a secured server within UZ Brussel.

Measures

The following sociodemographic variables were collected: gender, age, level of education, employment status, marital status, religious affiliation, number of children, alcohol use, drug use, smoking behaviour, and sports activity.

Symptomatic BrS patients were defined as patients with at least one of the following: previous aborted sudden death, syncope, sinus node dysfunction, or ICD shock.

Mental health was assessed with the 12 item General Health Questionnaire (GHQ-12) which is a screening tool for current mental disturbances and disorders that can be completed without supervision. The GHQ-12 is especially suitable for use in clinical practice, due to its brevity.¹⁸ It has been adopted as a screening tool by the World Health Organization to assess psychological disorders in primary health care, and it is considered the most extensively validated among similar screening tools. The instrument contains 12 symptom questions, with a recall period of a couple of weeks, which are scored on a four-point scale regarding intensity, ranging from much-less-than-usual to much-more-than-usual (e.g. *Have you been able to concentrate on all your activities in the past few weeks?*). As recommended by the designer of the GHQ-12, results were scored bimodally, resulting in a dichotomous scale (0-0-1-1 instead of 0-1-2-3). Total score ranges from 0 to 12, where higher scores indicate more severe symptoms of psychological distress. A score of 2 or higher indicates that further screening by a mental health professional is needed, while a score of 4 or more indicates a possible mental disorder.¹⁸

Social support was measured by the three item Oslo Social Support Scale (OSSS-3), a three-item scale that inquires on the number of close confidants, the sense of perceived concern from other people, and the relationship with neighbours, so focusing on the accessibility of practical help. The OSSS-3 has good predictive as well as construct validity, which is supported by its significant associations (in the predicted directions) with measures of positive mental health, mental health problems, psychological distress and depressive symptoms, depression, and satisfaction with life.¹⁹ The summated scores for the three items are interpreted as follows: 3–8 indicate poor social support, 9–11 denote moderate social support, and 12–14 represent strong social support.

Health-related quality of life (HRQL) was measured by the heart-related quality of life (HeartQoL) questionnaire which consists of a physical (10 items) and an emotional (4 items) subscale making up a 14 item global scale, where higher values representing better HRQL. All items on the physical (e.g. 'In the last 4 weeks, have you been bothered by having to lift or move heavy objects?') and the emotional subscale (e.g. 'In the last 4 weeks, have you been bothered by being worried?') are answered on a four point scale ranging from 'bothered a lot' (=0) to 'not bothered' (=3). A global HeartQoL score is calculated as the mean score of the 14 items. The HeartQoL questionnaire has been extensively validated for use in ischaemic heart disease patients.²⁰

Type D personality (which refers to a Distressed Personality) was assessed with the Type D Scale (DS14), which is a brief, psychometrically sound measure of negative affectivity (7 item subscale) and social inhibition (7 item subscale) that can readily be used in epidemiologic and clinical research.²¹ Subjects rate their personality on a five point scale ranging from 0 (=false) to 4 (=true). Summated scores equal to 10 or higher on both subscales indicate a Type D personality. The combination of a tendency towards negative affectivity and social inhibition often coincides with poor cardiac prognosis.²¹ Individuals with Type D personality exhibit heightened sensitivity to distress compared to the general population, often experiencing feelings of worry, sadness, and reduced confidence. They tend to expend considerable effort to conceal their emotions, which may lead to adverse health outcomes, including an increased risk for coronary artery disease, compromised immune function, and chronic inflammation.²²

Personality dimensions were assessed with the Ten Item Personality Inventory (TIPI), a brief assessment of the big five personality dimensions: (i) Extraversion, (ii) Agreeableness, (iii) Conscientiousness, (iv) Emotional Stability, and (v) Openness to Experience. All 10 items are rated on a scale from 1 (=disagree strongly) to 7 (=agree strongly). Although the TIPI provides a brief measurement of personality traits, the instrument has reasonable psychometric properties and is widely used in studies where brevity is important and where the focus is on an overall description of personality

traits.²³ Scoring is done in two steps: first, reverse-scored items are recoded (e.g. a 7 is recoded as a 1, a 6 as a 2, etc.). Second, the average of the two items that make up each scale (one standard item and one recoded, reverse-scored item) is computed. The five personality dimensions are scored as follows: Extraversion: items 1 and 6R (with 'R' denoting a reverse-scored item); Agreeableness: items 2R and 7; Conscientiousness: items 3 and 8R; Emotional Stability: items 4R and 9; and Openness to Experiences: items 5 and 10R.

Coping style was assessed using Brief-COPE, a 28 item self-report questionnaire designed to measure effective and ineffective ways to cope with a stressful life event. Coping is defined broadly as an effort done to minimize distress associated with negative life experiences. The scale is typically used in healthcare settings to ascertain how patients are emotionally responding to a serious health threat and determines a person's primary coping style and provides scores on three subscales: problem-focused coping (directed at reducing the threats and losses of the illness), emotion-focused coping (EFC) (directed at reducing the negative emotional consequences of the illness), and avoidant coping (AC) (trying to avoid certain situations, thoughts, or feelings rather than dealing with them). In addition, the following facets of coping are reported: self-distraction, denial, substance use, behavioural disengagement, emotional support, venting, humour, acceptance, self-blame, religion, active coping, use of instrumental support, positive reframing, and planning. The 28 items of the Brief-COPE are rated on a four point scale: ranging from 1='not at all', over 2='a little bit', and 3='medium', to 4='a lot'. Average scores are computed for three overarching coping styles (i.e. considering items that constitute the three respective scales), reflecting to which extent the respondent has been engaged in that coping style. The Brief-COPE has been thoroughly evaluated and was demonstrated to have strong psychometric properties.²⁴

Data collection

Data were collected using an online survey, on a secured server, in the retrospective phase of the study from August 2020 to July 2021 (up to two reminders were sent in case of no reaction to the invitation to participate). Patients in the prospective phase were asked to fill out the questionnaire (on paper) during their hospital visit, while waiting for ajmaline testing, between September 2021 and October 2022. Data were compared between BrS+ and BrS- prospective cohort and between BrS+ and standard Belgium population reference data.

Statistical analysis

All variables were tested for normality with Shapiro-Wilk test. Descriptive statistics are presented as means and standard deviations for quantitative variables and absolute and relative frequencies for categorical variables. Pearson correlation coefficient and test was used to assess for linear association. *t*-test was performed to evaluate the differences in mean scores obtained in the retrospective and prospective patients.²⁵ Furthermore, scores obtained in our sample were compared to general population norms, to account for the fact that patients who scored negatively on the ajmaline test could still consider themselves at risk, which may affect their responses. Proportion sizes were compared with *z*-tests. Significance tests were performed with $\alpha = 0.05$. Stepwise linear multiple regression was performed to evaluate predictors of mental distress. All statistical analyses were performed with SPSS (version 23) and R software version 3.6.2 (R Foundation for Statistical Computing, Vienna, Austria).

Results

In the retrospective phase, 403 email invitations with a link to the online questionnaires were sent. After maximum two reminders, a total of 144 completed questionnaires (response rate 35.7%) were received and analysed. Of these BrS+ patients, 87 (60%) were male and 57 (40%) female (Figure 1). In the prospective phase of the study, 65 individuals undergoing ajmaline test were included: 37 (57%) males and 28 (43%) females. Of these, 47 (72%) were BrS- and 18 (28%) BrS+. No patients had a previous diagnosis of any psychiatric disorder.

A total number of 209 questionnaires were completed and included in the analyses: 162 BrS+ and 47 BrS- (Table 1). The mean age was

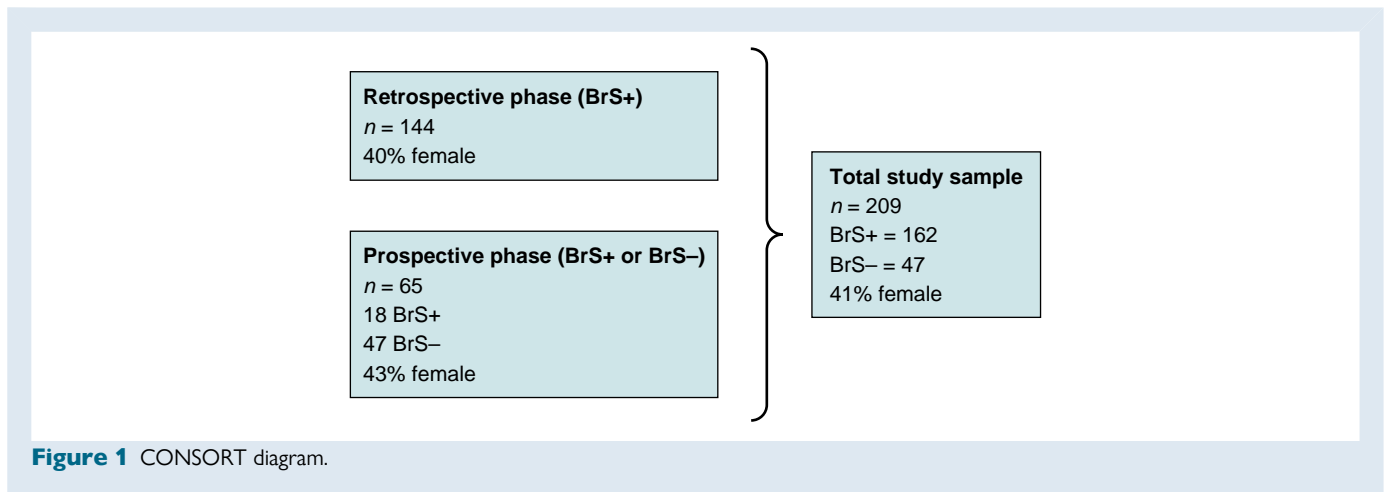


Figure 1 CONSORT diagram.

47.5 ± 15.0 years. Most of the individuals in our sample had earned a high school degree or higher (85.7%), had a partner (80.4%), and were professionally active (68.9%). An overview of all sociodemographic characteristics of the participants is shown in *Table 1*. The BrS+ and BrS- groups were roughly comparable, except for age (49.6 vs. 40.5, $P < 0.001$) and the amount of time (minutes per week) during which they engaged in sports (185.8 vs. 343.9, $P < 0.001$).

Responses to the 12 item General Health Questionnaire

Patients diagnosed with BrS (BrS+) reported higher levels of mental distress compared to general Belgium population norms as reported in the Belgian Health Interview Survey of 2018. Specifically, a one sample *t*-test showed that the GHQ-12 score for the BrS+ group (2.53 ± 3.03) was significantly higher than that of the general population (1.7, $P < 0.001$).²⁶

Additionally, 48.8% of the BrS+ group had a GHQ-12 score of at least 2, indicating mental distress and the need for further screening by a healthcare worker. Moreover, 34.0% of the BrS+ group had a score of at least 3, and 28.4% had a score of at least four symptoms, indicating a high likelihood of mental disorder. This was significantly higher than the 17.7% that reported at least four symptoms in the general Belgian population in 2018 ($P < 0.001$).²⁶ A significant difference in self-reported mental distress was observed between men and women in the BrS+ group; women (3.41 ± 3.38) had significantly higher scores on the GHQ-12 than men (1.93 ± 2.65, $P = 0.002$).

Comparing results in the BrS+ group from the retrospective phase with those from the prospective phase, BrS patients in the prospective phase showed significantly higher mental distress scores (2.28 ± 2.87 vs. 4.5 ± 3.6, $P = 0.003$).

When comparing the BrS+ group (2.53 ± 3.02) with the BrS- group (2.15 ± 3.16) overall, there was no significant difference in self-reported mental distress ($P = 0.451$); however when comparing the BrS+ group in the prospective phase with the BrS- group in the prospective phase, there was a significant difference (4.5 ± 3.60 vs. 2.15 ± 3.16, $P = 0.012$).

Responses to the three item Oslo Social Support Scale

Differences in mean social support scores between the BrS+ group and the BrS- group were not significant (10.14 ± 2.03 vs. 10.74 ± 1.80, $P = 0.065$). Both groups show a mean score that reflects moderate social support.

As no OSSS-3 norms are available for Belgium, we compared the mean score from participants in the BrS+ condition (10.14 ± 2.03) with the mean score in the general German population (10.16 ± 2.07) and found no significant difference ($P = 0.90$).¹⁹

Responses to the 14 item Health-related Quality of Life Questionnaire

While the HeartQoL emotional subscale did not differ between the BrS+ and the BrS- group, there was a tendency towards lower physical and global HeartQoL in the BrS+ group (*Table 2*).

We further compared Global HeartQoL scores in patients with BrS with scores found in ischaemic heart diseases as reported by Oldridge *et al.* (2014) and found that the BrS+ group had better global quality of life.²⁰ HeartQoL physical subscale scores in BrS+ were higher than in ischaemic heart disease, while HeartQoL emotional subscale scores tended to be lower in the BrS+ group.

Responses to the 14 item Type D personality questionnaire

In the BrS+ population ($n = 162$), there were 53 (32.7%) individuals with Type D personality. In the BrS- group ($n = 47$), there were 14 (29.8%), $P = 0.35$. Compared with the general population norm of 21% in Belgium, Type D personality seemed significantly more prevalent in our BrS+ sample ($P < 0.001$).²¹

Furthermore, we found a moderate correlation (0.463, $P < 0.001$) between the DS14 total score and mental distress (GHQ-12) in the BrS+ group and a strong correlation (0.611, $P < 0.001$) between the DS14 negative affectivity subscale and mental distress (GHQ-12).

Responses to the Ten Items Personality Inventory

Two hundred and eight questionnaires were completed: 143 in the retrospective phase and 65 in the prospective phase. In the BrS+ population ($n = 161$), we compared our results with known normative data for men and women (*Table 3*).²³ Compared with the population norms, BrS+ men scored lower on extraversion, agreeableness, emotional stability and openness to new experiences, and higher on conscientiousness. BrS+ women tended to have a slightly lower score on extraversion, scored lower on agreeableness, higher on conscientiousness, lower on emotional stability, and lower on openness to new experiences.

Table 1 Sociodemographic characteristics of the participants

Characteristics	Total sample (n = 209)	BrS+ (n = 162)	BrS- (n = 47)	P
Gender				
Male	124 (59.3%)	96 (59.3%)	28 (59.6%)	0.97
Age (years) (mean, SD)	47.5 ± 15.0	49.6 ± 14.5	40.5 ± 14.8	<0.001
Proband	79 (37.8%)	79 (48.8%)	NA	NA
Spontaneous BrS I pattern	25 (11.9%)	25 (15.4%)	0 (0.0%)	<0.001
Symptoms				<0.001
Syncope	44 (21.1%)	44 (27.2%)	0 (0.0%)	
Sinus node dysfunction	5 (2.4%)	5 (3.1%)	0 (0.0%)	
Previous aborted sudden death	7 (3.3%)	7 (4.3%)	0 (0.0%)	
ICD	50 (23.9%)	50 (30.1%)	0 (0.0%)	<0.001
ICD shock	6 (2.9%)	6 (3.7%)	0 (0.0%)	
SCN5A+	33 (15.8%)	33 (20.4%)	NA	NA
Education				
Primary education	8 (3.8%)	5 (3.1%)	3 (6.4%)	
Lower secondary education	22 (10.5%)	13 (8.0%)	9 (19.1%)	
Higher secondary education	74 (35.4%)	57 (35.2%)	17 (36.2%)	
Bachelor	53 (25.4%)	43 (26.5%)	10 (21.3%)	
Master	48 (23.0%)	41 (25.3%)	7 (14.9%)	
PhD	4 (1.9%)	3 (1.9%)	1 (2.1%)	
Employment				
Employee 1 job	115 (55.0%)	87 (53.7%)	28 (59.6%)	
Employee multiple jobs	6 (2.9%)	3 (1.9%)	3 (6.4%)	
Entrepreneur	23 (11.0%)	18 (11.1%)	5 (10.6%)	
Student	9 (4.3%)	3 (1.9%)	6 (12.8%)	
Unemployed	4 (1.9%)	1 (0.6%)	2 (4.3%)	
Other	53 (25.3%)	50 (30.9%)	3 (6.4%)	
Marital status				
Married	116 (55.5%)	96 (59.3%)	20 (42.6%)	
Partner, not living together	22 (10.5%)	17 (10.5%)	5 (10.6%)	
Partner, living together	30 (14.4%)	24 (14.8%)	6 (12.8%)	
Divorced	12 (5.7%)	9 (5.6%)	3 (6.4%)	
Single	28 (13.4%)	15 (9.3%)	13 (27.7%)	
Widowed	1 (0.5%)	1 (0.6%)	0 (0.0%)	
Number of children				
Has 1 child	22 (10.5%)	14 (8.6%)	8 (17.1%)	
Living in house	59.1% ^a	50.0% ^a	75.0% ^a	
Has 2 children	89 (42.6%)	74 (45.6%)	15 (31.9%)	
Living in house	64.0% ^a	64.2% ^a	63.3% ^a	
Has 3 children	27 (12.9%)	25 (14.2%)	4 (8.5%)	
Living in house	38.3% ^a	37.3% ^a	25.0% ^a	
Religion				
None (atheist)	65 (31.1%)	46 (28.4%)	19 (40.4%)	
Roman Catholic	118 (56.5%)	97 (59.9%)	21 (44.7%)	
Liberal	9 (4.3%)	5 (3.1%)	4 (8.5%)	
Muslim	5 (2.4%)	4 (2.5%)	1 (2.1%)	
Greek orthodox	2 (1.0%)	2 (1.2%)	0 (0.0%)	
Protestant	3 (1.4%)	2 (1.2%)	1 (2.1%)	

Continued

Table 1 Continued

Characteristics	Total sample (n = 209)	BrS+ (n = 162)	BrS- (n = 47)	P
Other	7 (3.3%)	6 (3.7%)	1 (2.1%)	
Alcohol use				
No	47 (22.5%)	37 (22.8%)	10 (21.3%)	
Yes	162 (77.5%)	125 (77.2%)	37 (78.7%)	
Average glasses per day:				
<1	99 (47.4%)	79 (48.8%)	20 (42.6%)	
1	39 (18.7%)	27 (16.7%)	12 (25.5%)	
2	19 (9.1%)	14 (8.6%)	5 (10.6%)	
3	2 (1.0%)	2 (1.2%)	0 (0.0%)	
4	3 (1.4%)	3 (1.9%)	0 (0.0%)	
Smoking				
No	189 (90.4%)	147 (90.7%)	42 (89.4%)	
Yes	20 (9.6%)	15 (9.3%)	5 (10.6%)	
Cigarettes per day:				
<10	11 (5.3%)	8 (4.9%)	3 (6.4%)	
11–20	8 (3.8%)	6 (3.7%)	2 (4.3%)	
>30	1 (0.5%)	1 (0.6%)	0 (0.0%)	
Sport				
No	96 (45.9%)	77 (47.5%)	19 (40.4%)	
Yes	113 (54.1%)	85 (52.5%)	28 (59.6%)	
Minutes per week	225 (± 174.9)	185.8 (± 143.4)	343.9 (± 208.4)	<0.001

BrS, Brugada syndrome; BrS+, patients with confirmed BrS; BrS-, people who tested negative for BrS; ICD = implantable cardioverter defibrillator.

^aAverage percentage of children living in house relative to number of children.

Responses to the 28 item Coping Style Questionnaire

We found no statistically significant differences in the three major coping styles between BrS+ and BrS- groups. We also compared the different facets that constitute these three coping styles (Table 4) and found self-distraction to be lower in the BrS+ group than in the BrS- group (2.90 ± 0.69 vs. 3.14 ± 0.68 , $P = 0.04$).

We further compared the retrospective results with the prospective results within the BrS+ population to determine if receiving BrS+ diagnosis may affect coping style. Results show that the use of EFC was significantly higher in the prospective BrS+ group (2.69 ± 0.28) than in the retrospective BrS+ group (2.45 ± 0.37), $P = 0.01$. Similarly AC was used more in the prospective BrS+ group (2.15 ± 0.43) than in the retrospective BrS+ group (1.85 ± 0.41), $P < 0.01$.

Finally, we compared our results from the BrS+ group with already published standards for healthy adults from the study by Poulus et al. (2020).²⁷ Results are presented in Table 5 and show that all facets that are part of the three major coping styles in BrS+ patients differ significantly from known general population norms.

Regression model

A stepwise linear multiple regression analysis was performed on the BrS+ data, to determine which variables significantly predicted the mental distress score (GHQ-12). Four explanatory variables (HeartQoL emotional subscale, DS14 negative affectivity subscale, symptomatic BrS patient, and age) contribute significantly to the model with standardized β coefficients respectively equal to -0.442 ($P <$

0.001), 0.304 ($P < 0.001$), 0.356 ($P < 0.001$), and -0.110 ($P = 0.048$). Higher mental distress as measured with GHQ-12 coincides with lower scores on the HeartQoL emotional subscale, higher negative affect scores on the DS14, symptoms, and younger age.

Discussion

Patients with BrS showed significantly more symptoms of mental distress than the general Belgian population (as measured by the GHQ-12), with 48.8% of the BrS+ group reporting at least two symptoms on the GHQ-12, indicating mental distress and the need for further screening by a professional. Additionally, a significant difference in self-reported mental distress was observed between men and women in the BrS+ group; women scored higher on the GHQ-12 than men, which is consistent with other studies.^{28,29} When comparing the BrS+ group with the BrS- group overall, there was no difference in self-reported mental distress; however, there was a significant difference between diagnostic phases. Indeed, BrS patients in the prospective phase showed significantly higher mental distress scores than BrS patients in the retrospective phase. This suggests that BrS+ patients report more symptoms of current mental distress before knowing their diagnosis, than their BrS- counterparts. It is possible that COVID-19 restrictions could have affected the mental health of the study participants in the prospective phase, but this effect would be expected to apply to both BrS+ and BrS- patients and may have less impact on the observed differences between these groups. Still, our data found significantly higher GHQ-12 scores in the prospective part of the study compared with the retrospective part, indicating that uncertainty about the Brugada diagnosis outcome might coincide with higher levels

Table 2 Comparison of mean HeartQoL scores

Instrument	BrS+	BrS–	Oldridge	df	t	P
HeartQoL (14 items)	2.44 (±0.59)	2.58 (±0.52)	2.2 (±0.5)	206	1.51	0.132
				6543	5.13	<0.001
HeartQoL physical (10 items)	2.48 (±0.60)	2.66 (±0.53)	2.2 (±0.7)	206	1.81	0.072
				6543	5.85	<0.001
HeartQoL emotional (4 items)	2.32 (±0.73)	2.39 (±0.66)	2.4 (±0.6)	206	0.55	0.582
				6543	1.38	0.17

HeartQoL, health-related quality of life; BrS+, patients with confirmed Brugada syndrome; BrS–, people who tested negative for Brugada syndrome; note that BrS + is compared with both BrS– and with values found in ischaemic heart disease (Oldridge); hence the two lines in the last three columns

Table 3 Big five personality traits in BrS +

Personality trait	BrS + men (n = 95)	Norm data men (n = 366)	t (df = 459)	P
Extraversion	3.82 (±1.57)	4.30 (±1.15)	2.79	0.005
Agreeableness	4.53 (±1.06)	5.05 (±1.11)	4.22	<0.001
Conscientiousness	5.62 (±1.10)	5.27 (±1.17)	2.73	0.007
Emotional stability	4.71 (±1.37)	5.27 (±1.32)	3.58	<0.001
Openness to experiences	4.81 (±1.20)	5.39 (±1.09)	4.28	<0.001
Personality trait	BrS + women (n = 66)	Norm data women (n = 760)	t (df = 824)	P
Extraversion	4.43 (±1.33)	4.68 (±1.47)	1.45	0.15
Agreeableness	4.57 (±0.87)	5.36 (±1.12)	6.90	<0.001
Conscientiousness	5.77 (±0.86)	5.56 (±1.09)	1.86	0.06
Emotional stability	4.33 (±1.46)	4.65 (±1.46)	1.71	0.09
Openness to Experiences	5.00 (±1.02)	5.45 (±1.04)	3.41	<0.001

BrS+, patients with confirmed Brugada syndrome.

of mental distress and significantly more in BrS + patients. Future longitudinal research should reveal whether levels of mental distress change over time in relation to diagnosis and whether there is, for example, a habituation effect.

Regarding health-related quality of life, BrS patients scored significantly better than patients with ischaemic heart disease, with higher scores in the physical subscale. This might be the result of the relatively high prevalence of asymptomatic BrS patients and highlights the usefulness of the HeartQoL for distinguishing the quality of life in BrS patients from other groups with cardiac diseases.³⁰

Our study also identified a higher proportion of individuals with Type D personality among BrS + patients compared with the general population.²¹ Furthermore, there was a moderate to strong correlation between Type D personality and mental distress in the BrS + group. These findings emphasize the importance of routine assessment and support for mental health in the BrS + population, given the link between Type D personality, psychological distress, cardiovascular disease, and mortality.^{31,32}

Regarding personality differences, men with BrS tended to be less extraverted, agreeable, emotionally stable, and open to new experiences than the general population norm for men. However, they scored higher on conscientiousness. Women with BrS also tended to be less agreeable and open to new experiences, with slightly lower

emotional stability than the general population norm for women. These personality differences can lead to typical behaviours, which should be considered in clinical practice to provide more individualized care.

Examples of behaviours driven by such personality traits in men are the following: missing appointments, having reduced therapeutic adherence, challenging healthcare professionals' opinions, non-compliance with hospital protocols, and unusual emotional reactions. In women, examples can be: expressing their opinions even if they conflict with healthcare professionals, be hesitant to try new medications, and requiring additional information or education to consider alternative treatment options. In severe cases, they may become overwhelmed by medical information and require additional support to manage their emotions effectively.

It is important to note, however, that statistical averages cannot be directly applied to each individual patient, and the clinical interpretation of personality differences should be made with caution.

The present study found a significant difference in coping styles between BrS + participants in the prospective and retrospective phases. Problem-focused coping, EFC, and AC are three main coping strategies that individuals use to deal with stress.³³ Problem-oriented coping involves taking immediate action to solve the problem causing stress, while EFC involves managing one's emotional response to stress, and

Table 4 Coping styles

	BrS + (mean ± SD; n = 162)	BrS- (mean ± SD; n = 47)	t (df = 207)	P
PFC	3.14 (±0.55)	3.27 (±0.52)	1.44	0.15
Active coping	3.39 (±0.67)	3.57 (±0.54)	1.69	0.09
Use of instrumental support	2.75 (±0.75)	2.85 (±0.87)	0.78	0.44
Positive reframing	3.11 (±0.69)	3.20 (±0.73)	0.78	0.44
Planning	3.31 (±0.72)	3.46 (±0.67)	1.28	0.20
EFC	2.48 (±0.37)	2.51 (±0.44)	0.47	0.64
Use of emotional support	2.64 (±0.81)	2.84 (±0.90)	1.45	0.15
Venting	2.52 (±0.70)	2.46 (±0.78)	0.50	0.61
Humour	2.28 (±0.73)	2.31 (±0.76)	0.25	0.81
Acceptance	3.23 (±0.65)	3.37 (±0.70)	1.28	0.20
Self-blame	2.57 (±0.65)	2.50 (±0.70)	0.64	0.52
Religion	1.62 (±0.87)	1.59 (±0.98)	0.20	0.84
AC	1.88 (±0.42)	1.91 (±0.38)	0.44	0.66
Self-distraction	2.90 (±0.69)	3.14 (±0.68)	2.11	0.04
Denial	1.66 (±0.70)	1.80 (±0.88)	1.14	0.26
Substance use	1.29 (±0.58)	1.18 (±0.44)	1.20	0.23
Behavioural disengagement	1.69 (±0.69)	1.52 (±0.71)	1.48	0.14
	BrS + retrospective (mean ± SD; n = 144)	BrS + prospective (mean ± SD; n = 18)	t (df = 160)	P
PFC	3.14 (±0.56)	3.15 (±0.42)	0.07	0.94
Active coping	3.41 (±0.67)	3.28 (±0.62)	0.78	0.44
Use of instrumental support	2.71 (±0.74)	3.06 (±0.78)	1.88	0.06
Positive reframing	3.13 (±0.69)	2.92 (±0.71)	1.21	0.23
Planning	3.31 (±0.71)	3.33 (±0.80)	0.11	0.91
EFC	2.45 (±0.37)	2.69 (±0.28)	2.65	0.009
Use of emotional support	2.60 (±0.81)	3.00 (±0.69)	2.00	0.047
Venting	2.50 (±0.69)	2.75 (±0.75)	1.44	0.15
Humour	2.27 (±0.71)	2.42 (±0.90)	0.82	0.41
Acceptance	3.23 (±0.64)	3.22 (±0.75)	0.06	0.95
Self-blame	2.52 (±0.64)	2.97 (±0.65)	2.81	0.006
Religion	1.60 (±0.88)	1.78 (±0.81)	0.82	0.41
AC	1.85 (±0.41)	2.15 (±0.43)	2.91	0.004
Self-distraction	2.86 (±0.71)	3.14 (±0.48)	1.63	0.11
Denial	1.61 (±0.68)	2.06 (±0.68)	2.65	0.009
Substance use	1.25 (±0.55)	1.61 (±0.74)	2.51	0.013
Behavioural disengagement	1.67 (±0.67)	1.81 (±0.82)	0.81	0.42

BrS+, patients with confirmed Brugada syndrome; BrS-, people who tested negative for Brugada syndrome; PFC, problem-focused coping; EFC, emotion-focused coping; AC, avoidance coping.

AC involves avoiding or escaping the source of stress. The effectiveness of each coping style varies depending on the stressor.³⁴

EFC and AC were found to be more prominent in BrS + participants in the prospective phase than in those in the retrospective phase, suggesting that waiting for a diagnosis may contribute to increased mental distress. Previous research has linked these coping styles to worse mental health outcomes.³⁵ Furthermore, the potential dangers of EFC for BrS + patients are significant due to the link between the development of arrhythmias and hypervagal stimulation. Therefore, future research

should focus on developing stress-reducing approaches that minimize this risk. Such intervention should at least consist of stress reduction techniques that do not increase the risk of vagal stimulation (i.e. no meditation or deep relaxation techniques) as well as cognitive behavioural therapy techniques that increase resilience and prevent the patient from resorting to dysfunctional coping mechanisms, which are known to lead to undesirable mental health outcomes.

Our findings indicate that mental health aspects should receive attention during the BrS diagnosis process since coping styles and mental

Table 5 BrS + coping styles compared with normative data

	BrS +, n = 162 mean (\pm SD)	General population (norms) n = 316 mean (\pm SD)	t (df = 476)	P
PFC	3.14 (\pm0.55)	2.47 (\pm0.63)	11.99	<0.001
Active coping	3.39 (\pm 0.67)	2.79 (\pm 0.79)	8.71	<0.001
Use of instrumental support	2.75 (\pm 0.75)	2.03 (\pm 0.88)	9.36	<0.001
Positive reframing	3.11 (\pm 0.69)	2.42 (\pm 0.92)	9.21	<0.001
Planning	3.31 (\pm 0.72)	2.65 (\pm 0.83)	9.00	<0.001
EFC	2.48 (\pm0.37)	2.23 (\pm0.49)	6.24	<0.001
Use of emotional support	2.64 (\pm 0.81)	1.99 (\pm 0.89)	8.03	<0.001
Venting	2.52 (\pm 0.70)	2.06 (\pm 0.81)	6.44	<0.001
Humour	2.28 (\pm 0.73)	2.67 (\pm 0.83)	5.27	<0.001
Acceptance	3.23 (\pm 0.65)	2.99 (\pm 0.78)	3.56	<0.001
Self-blame	2.57 (\pm 0.65)	2.39 (\pm 0.90)	2.50	0.013
Religion	1.62 (\pm 0.87)	1.29 (\pm 0.63)	4.29	<0.001
AC	1.88 (\pm0.42)	1.64 (\pm0.45)	5.77	<0.001
Self-distraction	2.90 (\pm 0.69)	2.47 (\pm 0.89)	5.83	<0.001
Denial	1.66 (\pm 0.70)	1.37 (\pm 0.61)	4.47	<0.001
Substance use	1.29 (\pm 0.58)	1.19 (\pm 0.47)	1.90	0.058
Behavioural disengagement	1.69 (\pm 0.69)	1.53 (\pm 0.71)	2.38	0.018

BrS+, patients with confirmed Brugada syndrome; PFC, problem-focused coping; EFC, emotion-focused coping; AC, avoidance coping.

distress scores may impact patient well-being. Moreover, the prevalence of Type D personality suggests the importance of a systematic screening and assessment of possible mental health problems.

Limitations

First, we acknowledge that scores of mental well-being (GHQ-12) may have been influenced by the restrictions associated with the COVID-19 pandemic; this effect however would be expected to apply to both BrS + and BrS- patients. Second, we state that all reported values were self-reported by study participants; it is thus possible that socially desirable answers were given to some specific questions (e.g. alcohol consumption). Third, these are results from a single-centre registry; it is therefore possible that the psychological impact of BrS may have been influenced by specific management approaches that are not transferable to other settings or applicable in other health care models. Fourth, all patients within the retrospective registry were invited to complete the questionnaire, resulting in a response rate of 35.7%. It is possible that personality type and levels of mental distress influenced the decision to respond to the questionnaire, implying that the sample is not representative of the overall cohort. Finally, a correlation between BrS and mental stress was shown in this study; further research needs to be conducted to evaluate possible causality.

Conclusion

The current study highlights the need for routine assessment and support for mental health in BrS + patients, especially those with Type D personality traits. Interventions aiming at reducing psychological distress in individuals with BrS could improve quality of life and potentially reduce the risk of adverse outcomes. Such interventions should focus on stress reduction techniques that do not increase vagal tone as well as cognitive behavioural therapy techniques to avoid dysfunctional coping mechanisms.

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Data availability

The data underlying this article cannot be shared publicly due to the privacy of individuals who participated in the study. The data will be shared on reasonable request to the corresponding author.

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