

A New Tool in the Development of Celiac Disease: Sense of Coherence Score

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Received March 02, 2020; Revised April 16, 2020; Accepted April 27, 2020

Abstract Introduction: Sense of Coherence (SOC) is a theoretical construct that was developed to explain why some people become ill and others do not, regardless of stressful events. Low SOC was reported to correlate with the development of several multifactorial, chronic diseases such as diabetes mellitus and systemic lupus erythematosus. **Aims:** To compare the SOC scores of patients diagnosed with celiac disease (CD) and matched healthy controls to investigate possible correlations between low SOC scores and CD. **Patients and Methods:** Patients provided demographic data and completed the 13-item SOC questionnaire. Patients in the study cohort were matched to healthy controls according to age, gender, education, income, employment status and marital status. **Results:** A total of 239 CD patients and 124 healthy controls answered the SOC questionnaire. Among them, 124 patients were matched to the healthy cohort according to propensity score. CD patients had a median SOC score of 57 (IQR 52.25-62), and controls 65.5 (IQR 57-75) $p < 0.001$. **Conclusions:** SOC reflects a person's resources and orientation which enable individuals to cope with stressors in a way that promotes health. Lower SOC score is correlated with the development of CD.

Keywords: celiac disease, orientation to life, sense of coherence

Cite This Article: Adi Eindor-Abarbanel, Dana ZelnikYovel, Timna Naftali, Nahum Ruhimovich, Tzipora Shalem, Shay Matalon, Haim Shirin, Tomer Ziv-Baran, and Efrat Broide, "A New Tool in the Development of Celiac Disease: Sense of Coherence Score." *International Journal of Celiac Disease*, vol. 8, no. 1 (2020): 1-4. doi: 10.12691/ijcd-8-1-1.

1. Introduction

Celiac disease (CD) is an immune-mediated disorder triggered by exposure to gluten in genetically susceptible individuals [1]. Although HLA DQ2/DQ8 predisposes for CD [2], other factors are likely to contribute to the development of the disease [3].

Similar to other chronic diseases, CD is a life-long medical condition that affects an individual's well-being and quality of life [4]. The pathogenesis behind the development of chronic diseases is multifactorial and includes genetic, environmental and emotional factors [5]. Several other common chronic diseases, such as type 2 diabetes [6,7,8] and inflammatory bowel diseases [9,10] are associated with higher rates of concurrent psychopathologies. Patients suffering from psychological distress are less adherent to recommendations for behavioral and lifestyle modifications and therefore, are

prone to more severe disease with poorer control¹¹. CD patients have been reported to be at greater risk for anxiety and depression [12]. The influence of emotional stressors on the development of CD has received little attention [15]. Research regarding the influence of the emotional distress on adherence to a gluten-free diet is contradictory [13,14].

Aaron Antonovsky, a medical sociologist, introduced the concept of Sense of Coherence (SOC) in his theory about salutogenesis. [16,17] SOC is a theoretical construct that is intended to explain why some people become ill and others do not, regardless of stressful events. [18] The theory focuses on the personal resources needed to maintain health. SOC consists of the core elements of comprehensibility, manageability and meaningfulness. Comprehensibility means that the individual considers structured environmental demands. Manageability refers to the resources that an individual has to meet these demands. Meaningfulness is the understanding that these demands are worth the resources that must be invested to

deal with them. These three components are important parts of an individual's coping strategy. The stronger one's SOC, the more likely a person is to be able to adopt appropriate coping strategies. Therefore, SOC is considered a key tool in enabling a person to deal with stressors. According to salutogenesis theory, SOC is stable and enduring and develops mainly during the first three decades of life. [8-19]

SOC was shown to correlate with the development of several multifactorial, chronic diseases, such as diabetes mellitus [20] and systemic lupus erythematosus (SLE). [21] Feritaset al. [22] investigated the relationship between SOC, anxiety and depression among patients with inflammatory bowel disease (IBD). They found that lower SOC was independently associated with higher levels of depression and anxiety. However, our previous study did not find significant differences in SOC between healthy individuals and IBD patients [23]. To our knowledge, the relationship between SOC and CD was not yet been investigated.

SOC is strongly related to perceived health. Data relating low SOC with emotional distress among patients with chronic diseases are increasing. This study explored possible associations between low SOC scores and the development of CD.

2. Methods

An anonymous, online questionnaire for celiac patients ≥ 18 years-of-age was distributed nationally through the Celiac Society in Israel and social networks. Healthy controls were also recruited online via social media advertisements. Potential participants answered an anonymous, electronic questionnaire that included demographic data and the SOC questionnaire. The healthy control group excluded individuals who took chronic medications and those who had a chronic disease.

The study was approved by the Ethics Committee of Shamir Medical Center on July 3rd 2018. The study did not require written informed consent since the patients and controls agreed to participate in the study by filling the electronic questionnaire. The study protocol conforms to the ethical guidelines of the 1975 Declaration of Helsinki as reflected prior approval by the institution's human research committee.

Sense of coherence was assessed based on the SOC orientation to life score. [24] The SOC-13 scale measures the degree to which an individual views the world as comprehensible (5 items), manageable (4 items), and meaningful (4 items) using a 7-point Likert scale. The total SOC-13 score is the sum of the items, ranging from 13 to 91, with higher scores reflecting better SOC. (The author kindly granted permission to use the scale in this study.)

2.1. Statistical Analysis

Categorical variables were described using frequency and percentage. Continuous variables were evaluated for normal distribution using histograms and Q-Q plots. Normally-distributed continuous variables were described as mean and standard deviation and non-normally

distributed data were expressed as median and interquartile range. Independent samples T-test and Mann-Whitney test were used to compare continuous variables between groups. Chi-squared test was used to compare categorical variables between the groups. Spearman correlation coefficient was applied to evaluate the association between SOC and continuous variables. Association between SOC and categorical variables was performed by using independent samples T-test or analyses of variance. Multivariate regression was used to identify independent predictors of SOC. Age, gender, education, employment status, income level, marital status and celiac status, were included in the regression analysis.

Backward selection method was used and $p < 0.1$ was chosen as a criterion for removal. The linear regression was evaluated to meet the assumptions. Patients in the CD cohort were matched to the healthy cohort using a propensity score. Logistic regression was used to calculate the propensity score, as the probability of a patient to be in the celiac group. Age, gender, education, employment status, income level and marital status were included in the regression. An absolute difference of up to 5% was considered an acceptable difference for matching. Categorical variables were compared between the matched participants using the McNemartest and continuous variables were compared using the Wilcoxon Signed-Ranks test or paired sample T-test.

All statistical tests were two-tailed. $P < 0.05$ was considered statistically significant.

Statistical analyses were performed using SPSS (IBM Corp. Released 2017. IBM SPSS Statistics for Windows, Version 25.0. Armonk, NY: IBM Corp.).

3. Results

Among 362 adults who completed the questionnaire, 238 were celiac patients with a median age of 36 years (IQR 26-46) and 189 were women (79.1%). There were 124 healthy adult controls, with a median age of 40 years (IQR 33-50) and 87 females (70.2%). Demographic data of the two groups are listed in Table 1. According to the univariate analysis, there were significant differences in age, gender, education, employment status, level of income, marital status and SOC between CD patients and healthy controls (Table 2).

Table 1. Demographic characteristics of study participants

Variable	Study Participants n=363
Age median (IQR)	38 (28-47)
Female n (%)	276 (76)
Working hours median (IQR)	40 (19-45)
Education >12 years, n (%)	275 (75.8)
Marital status, n (%)	
Single	91 (25.1)
In a relationship	248 (68.3)
Past relationship	23 (6.3)
Employed	306 (84.3)
Income level, n (%)	
Low	143 (39.4)
Medium	72 (19.8)
High	130 (35.8)

Table 2. Univariate analysis of median Sense of Coherence scores and demographic parameters in patients with celiac disease compared with healthy controls

Variable	Celiac Disease n=239	Control group n=124	p-value
Age median (IQR)	36 (26-46)	40 (33-50)	0.005
Female	189 (79.1)	87 (70.2)	0.059
Working hours	40 (10-45)	40 (28.5-45)	0.003
Education > 12 years n (%)	168 (70.3)	107 (86.3)	0.001
Marital status n (%)			<0.001
Single	75 (31.5)	16 (12.9)	
In a relationship	147 (61.8)	101 (81.5)	
Past relationship	16 (6.7)	7 (5.6)	
Employed	195 (81.6)	111 (89.5)	0.049
Income level, n (%)			<0.001
Low	110 (49.8)	33 (26.6)	
Medium	45 (20.4)	27 (21.8)	
High	66 (29.9)	64 (51.6)	
SOC median (IQR)	56 (51-61)	65.6 (57-75)	<0.001

The CD group participants were less educated with 70.3% of participants completed more than 12 years of education, compares to 86.3% in the healthy controls group ($p=0.001$), and they also had lower income levels ($p=0.001$).

In the Univariate analysis, CD patients had a SOC score of 56 (IQR 51-61) and the controls had 65.5 (IQR 57-75; $p<0.001$).

According to the propensity score, 124 healthy participants were matched to 124 CD patients. When comparing the two matched groups, CD patients had a median SOC score of 57 (IQR 52.25-62), whereas the healthy controls had a score of 65.5 (IQR 57-75; $p<0.001$).

4. Discussion

This study compared SOC between patients with CD and healthy controls. We found that SOC in CD patients is much lower than it is in the general population.

These results are similar to those documented among patients with other multifactorial, chronic diseases [13,14,22-25]. An observational study that compared SOC between patients with diabetes and healthy controls [20] found that the control population had 2.4 times higher odds of having a high SOC score as compared to patients with type 2 diabetes. On the other hand, reports on the correlation between SOC and disease control had conflicting results. Two studies [26,27] showed that SOC did not correlate with metabolic control among patients with insulin-dependent diabetes, while another study [28] evaluating patients with type 1 or 2 diabetes, reported that higher SOC scores were correlated with better glycemic control.

A study that investigated the role of SOC in the development of SLE in women [21] found that those with SLE had significantly lower SOC scores as compared to healthy controls, without correlation with disease activity.

In a previous study, comparing SOC scores between patients with a different chronic gastrointestinal disease, inflammatory bowel disease (IBD) and healthy controls, we found no difference between the two groups. [23] Furthermore, although intuitively, we would expect SOC

to be related to disease severity, the results of the diabetes [26] and SLE [21] studies indicate otherwise. This is likely related to the multifactorial etiologies of these diseases.

CD is an autoimmune enteropathy characterized by intolerance of dietary gluten. The clinical spectrum of CD, in non-adherent patients, is very wide. It includes gastrointestinal manifestations such as diarrhea and abdominal discomfort and extraintestinal manifestations such as fatigue and anemia. Even though a gluten-free diet induces mucosal healing [3] and is supposed to diminish symptoms, there are several reports that CD patients still suffer from fatigue [29] and emotional distress [30], indicating the chronic nature of the disease. Our results are in concordance with Antonovsky's theory of Salutogenesis [17], which states that individuals with lower SOC are more prone to develop chronic diseases. Furthermore, a previous study that investigated the relationship between stressful events and the development of CD, demonstrated that adults with CD reported the occurrence of serious life events in the years before the diagnosis more often than did control patients [15].

The current study had some limitations. The study population consisted of patients recruited via online anonymous questionnaires. This might influence the credibility of demographic data and disease status reported. Also bias due to the type of individuals who were motivated to respond. Additionally, the strict matching used in the study design resulted in a smaller study sample. Thus, the statistical power was also decreased. Furthermore, the meticulous matching between the 2 groups, might bring parameters that are independently influenced by SOC, such as employment and marital status, into the equation. Additionally, since HLA DQ2/DQ8 predisposes for CD [2], lack of data regarding HLA status could affect the interpretation of the results.

In conclusion, SOC consists of the core elements comprehensibility, manageability and meaningfulness, which reflect the resources that enable individuals to cope with stressors in a way that promotes health. The results of this study indicate that SOC is correlated with the development of CD. Future studies that include genetic data would add useful information to this conclusion.

Financial Support

No specific funding was received for this study

Conflict of Interest

The authors declare no conflict of interest.

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