Illness Identity in Adults with a Chronic Illness

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Abstract

Objective. The present study examines the concept of illness identity, the degree to which a chronic illness is integrated into one's identity, in adults with a chronic illness by validating a new self-report questionnaire, the Illness Identity Questionnaire (IIQ). **Methods.** Self-report questionnaires on illness identity, psychological and physical functioning were assessed in two samples: adults with congenital heart disease (22-78 year old; *n*=276) and with multisystem connective tissue disorders (systemic lupus erythematosus or systemic sclerosis; 17-81 year old; *n*=241). **Results**. The IIQ could differentiate four illness identity states (i.e., engulfment, rejection, acceptance, and enrichment) in both samples, based on exploratory and confirmatory factor analysis. All four subscales proved to be reliable. Rejection and engulfment were related to maladaptive psychological and physical functioning, whereas acceptance and enrichment were related to adaptive psychological and physical functioning. **Conclusion**. The present findings underscore the importance of the concept of illness identity. The IIQ, a self-report questionnaire, is introduced to measure four different illness identity states in adults with a chronic illness.

KEYWORDS: chronic illness, congenital heart defects, multisystem connective tissue disorders, illness identity, psychological functioning

Introduction

Having a chronic illness can pose major challenges to a person's life due to many lifestyle changes, such as adhering to a daily treatment regimen. Although most patients succeed in adjusting to their illness, some patients experience substantial difficulties, which can negatively affect their physical and psychosocial functioning (Morea, Friend, & Bennett, 2008). To understand why some patients succeed in managing these challenges, whereas others experience more difficulties, the present study examined the concept of illness identity from an integrative point of view. In line with Charmaz (1995), we define illness identity as the degree to which a chronic illness becomes integrated into one's identity (Oris et al., 2016). Four different illness identity states were distinguished (i.e., rejection, engulfment, acceptance, and enrichment) and their relation to psychological and physical functioning was examined.

Illness Identity in Chronic Illness

Inspired by Erikson's (1968) seminal work on lifespan ego-development, identity is viewed as the degree to which (i) an individual (manages to) integrates different self-assets into a coherent sense of self, and (ii) such a coherent sense of self translates itself into daily life and guides choices and values. This gives rise to a feeling of continuity and sameness and has been demonstrated to contribute to psychological well-being (Campbell, Assanand, & Paula, 2003; Erikson, 1968; Schwartz, 2001). When confronted with a chronic illness, individuals need to understand what this means to their identity and try to create or regain a coherent sense of self (Leventhal, Idler, & Leventhal, 1999). In other words, they need to integrate their chronic illness into their identity, a process originally conceptualized as illness identity in the sociological literature (Charmaz, 1995). Over the years, many related constructs from different theoretical backgrounds have been forwarded related to the topic of illness identity. Only recently, to bridge these traditions, Oris et al. (2016) have introduced and validated a new questionnaire, the Illness Identity Questionnaire (IIQ), in youth (ages 14-25) with type 1 diabetes (T1D). The IIQ extends the Illness Self-Concept Scale, which assesses engulfment and acceptance on one dimension (Morea et al., 2008), by explicitly distinguishing among these states and by adding two

additional constructs that have been suggested by Charmaz (1999) and have demonstrated to be also important (Senol-Durak, 2014; Tilden, Charman, Sharples, & Fosbury, 2005): rejection and enrichment. Hence, The IIQ focuses on four different illness identity states: rejection, engulfment, acceptance, and enrichment. The first two identity states assessed by the IIQ, *engulfment* and *rejection,* capture a lack of illness integration (Oris et al., 2016). *Engulfment* refers to the degree to which chronic illness dominates a person's identity and daily life. Individuals completely define themselves in terms of their illness, which invades all domains of life, at the expense of other important self-assets (Morea et al., 2008). Next, the state of *rejection* is mainly based on qualitative studies that tried to understand poor treatment adherence in patients with T1D and asthma (Adams, Pill, & Jones, 1997; Tilden et al., 2005). These studies concluded that some patients tend to neglect their illness, resulting in suboptimal treatment adherence (Oris et al., 2016). These individuals also try to avoid thinking and talking with others about their illness (Tilden et al., 2005). Hence, *rejection* refers to the degree to which the chronic illness is rejected as part of one's identity and is viewed as a threat or as being unacceptable to the self.

In contrast to these two illness identity states, two other states capture ways of adaptive illness integration: *acceptance* and *enrichment* (Oris et al., 2016). *Acceptance* captures the degree to which individuals accept the illness as part of their identity without being overwhelmed. Chronic illness plays a peripheral role in one's identity, besides other personal, relational, and social self-assets, and does not pervade all life domains (Morea et al., 2008). Patients try to lead as normal a life as possible, whereas, at the same time, they do not deny having a chronic illness (Adams et al., 1997). Finally, with respect to the fourth illness identity resolution (*enrichment*), positive changes as a result of negative life events, such as chronic illness, have been referred to as benefit finding or stress-related growth (Helgeson, Reynolds, & Tomich, 2006; Senol-Durak, 2014). Such positive changes manifest themselves in different ways, including an increased appreciation for life, changed life priorities, increased personal strength, and more positive interpersonal relationships (Tedeschi & Calhoun, 2004). In contrast to the broader concepts of benefit finding or post-traumatic growth, *enrichment* specifically

refers to positive changes related to one's identity. Hence, it refers to the degree to which chronic illness enriches one's sense of self, and enables one to grow as a person.

Although these four concepts have been examined previously (e.g., Evers et al., 2001; Helgeson et al., 2006; Morea et al., 2008; Tilden et al., 2005), no study or existing questionnaire assessed these four illness identity states simultaneously and/or forwarded an integrative framework. For example, the Illness Cognition Questionnaire (Evers et al., 2001) focused on helplessness (which is somewhat similar to engulfment), acceptance, and perceived benefits, but did not assess rejection. Although previous measures have substantially improved our understanding of illness identity, the IIQ, which taps into these four illness identity states, allows fine-tuning the assessment of illness identity. Hence, the concept of illness identity could provide an integrative framework, potentially guiding both research and clinical practice.

Psychological and Physical functioning

Attaining an identity structure in which different self-assets are integrated into a coherent whole has been found to contribute to psychological well-being (Campbell et al., 2003). Hence, the degree to which individuals achieve to attain such a coherent identity in the context of chronic illness may influence psychological functioning as well (Morea et al., 2008). As such, rejection may give rise to suboptimal functioning, as a potentially important self-asset is being ignored (Baumeister, 1999). Also, a chronic illness that intrudes upon all life domains (cf. engulfment) has demonstrated to be related to maladaptive functioning (Luyckx, Rassart, & Weets, 2015; Oris et al., 2016). In contrast, acceptance and enrichment have been related to adaptive psychological functioning (Helgeson et al., 2006; Oris et al., 2016).

In addition, the degree to which individuals integrate a chronic illness into one's identity might relate to physical functioning as well (Leventhal et al., 1999). As physical symptoms may disrupt everyday functioning, they may interfere with identity roles and instigate individuals to rethink one's identity (Leventhal et al., 1999). In addition, illness identity may also influence physical functioning and symptom experience (Leventhal et al., 1999; Luyckx, Vanhalst, Seiffge-Krenke, & Weets, 2010). For

example, acceptance of the illness as part of one's identity might lead to more adaptive better coping and self-care (Richardson, Adner, & Nordstrom, 2001) and, hence, better (perceived) physical health (Karademas, Tsagaraki, & Lambrou, 2009). Research has indeed demonstrated that acceptance was related to less illness symptoms, whereas concepts related to engulfment were related to more symptoms (Evers et al., 2001; Morea et al., 2008).

Research Objectives and Hypotheses

The present study aims to provide evidence for the concept of illness identity in adults with chronic illness by assessing the validity of these four states and by examining associations with psychological and physical functioning. In examining our research objectives, two patient samples were used: congenital heart disease (CHD) and multisystem connective tissue disorders (MSDs). First, CHD is the most frequent birth defect (9:1,000 births; van der Linde et al., 2011) and comprises a wide spectrum of structural heart lesions, varying from simple to complex severity lesions (Vander Velde et al., 2005). Because almost 90% of children with CHD survive into adulthood (Moons, Bovijn, Budts, Belmans, & Gewillig, 2010), a long-term follow-up throughout the lifespan is needed to decrease rates of morbidity and mortality (Warnes et al., 2008). Hence, although adults with CHD generally manage to successfully engage in different adult life responsibilities and roles, they are also confronted with various medical, psychosocial, and behavioral challenges, such as restricted employment opportunities because of physical limitations (Kovacs, Sears, & Saidi, 2005).

Second, MSDs are chronic auto-immune conditions characterized by a complex pathogenesis and inflammation of multiple organ systems (Medsger, 2003; Simard & Costenbader, 2007). The present study focuses on two such MSDs: Systemic lupus erythematosus (SLE) and Systemic sclerosis (SSc). SLE is a systemic auto-immune disease which has a highly variable course and prognosis. It is characterized by, for instance, organ involvement, but also by joint and muscle pains, skin rashes, and fatigue (Simard & Costenbader, 2007). SSc is characterized by three cardinal pathogenic features: activation of the immune system, fibrosis of the skin and internal organs, and microvascular involvement (Medsger, 2003). Prevalence rates of both diseases vary greatly geographically, with

ranges from 1.4-21.9/100,000 inhabitants for SLE and from 7-700/1,000,000 for SSc. Both diseases occur primarily in women (male-to-female ratio of about 9:1 in SLE and 3:1 to 8:1 in SSc), with usual disease onset between ages 15 and 40 in SLE and between ages 35 and 55 in SSc (Lisnevskaia, Murphy, & Isenberg; Simard & Costenbader, 2007; Valentini & Black, 2002). Given the heterogeneous and unpredictable disease course, with high morbidity rates and high mortality rates in the case of SSc, both disorders have a substantial impact on daily life (Dobkin, Da Costa, & Dritsa, 1999; Haythornthwaite, Heinberg, & McGuire, 2003). Although CHD and MSDs are different medical conditions, both groups of patients are confronted with common challenges, such as lifestyle changes and recognizing symptoms related to their condition. Research has indeed demonstrated that chronic illnesses have general stressors and tasks in common, although differences in the degree and type of stressors do exist (Heijmans et al., 2004). Hence, in the present study, integration of chronic illness into one's identity is viewed as a common task across diagnostic categories (Schulman-Green et al., 2012).

Objective 1: Factorial Validity and Reliability of the IIQ

Given that subscale scores on the IIQ have only been validated in youth with T1D (Oris et al., 2016) and illness integration is a lifelong challenge and process (Leventhal et al., 1999; Schulman-Green et al., 2012), our first objective was to validate subscale scores on the IIQ in adults with CHD and MSDs. Furthermore, internal consistencies of the four illness identity states were examined.

Objective 2: Associations with Demographic and Clinical Parameters

The present study explored mean differences in illness identity states based on demographic and clinical variables. First, based on a recent study on illness identity (Oris et al., 2016), no sex and age differences in illness identity were expected (Oris et al., 2016). Second, in patients with MSDs, disease duration was expected to be unrelated to illness identity (Oris et al., 2016). As the study by Oris et al. (2016) focused on youth (ages 14-25), we aimed to explore if consistency of the results for sex, age, and disease duration could be demonstrated in adults. Third, we explored mean differences in illness identity states between patients with CHD and MSDs. As these patient groups have not been directly compared before, we did not have specific hypotheses. Finally, we compared differences

within conditions. Complex heart defects and SSc could lead to greater disruptions in a person's life as compared to simple/moderate heart defects and SLE, respectively (Haythornthwaite et al., 2003; Kovacs et al., 2005), possibly leading to engulfment (Beanlands et al., 2003). Such disruptions could also increase the odds that the chronic illness would be rejected as part of one's identity, as confrontation would be too overwhelming (Mozzetta et al., 2008). However, as self-growth is more likely to occur with more severe stressors (Helgeson et al., 2006), complex heart defects and SSc could lead to feelings of enrichment as well. Consequently, patients with a more complex heart defect and with SSc were expected to score higher on engulfment, rejection, and enrichment than patients with a simple defect and SLE, respectively.

Objective 3: Associations with Psychological and Physical Functioning

Depressive and anxiety symptoms were used as an indicator of psychological functioning in both patient groups. Perceived illness symptoms and pain were used as an indicator of perceived physical functioning in patients with CHD and MSDs, respectively. We expected rejection and engulfment to be positively related to depressive and anxiety symptoms (Oris et al., 2016), and, for engulfment, also to illness symptoms and pain (Morea et al., 2008). Acceptance and enrichment would be negatively related to depressive and anxiety symptoms (Oris et al., 2016), and for acceptance, also to illness symptoms, and pain (Evers et al., 2001).

Methods

Participants and Procedure

Sample 1. As part of the Belgian branch of APPROACH-IS (Assessment of Patient-Reported Outcomes in Adults with Congenital Heart Disease – International Study; Apers et al., 2015, 2016), patients were selected from the database of congenital cardiology of the University Hospitals Leuven (Belgium) using the following criteria: (1) diagnosis of CHD, defined as a structural abnormality of the heart and/or intra-thoracic great vessels present at birth and actually or potentially functionally significant (including mild, moderate, and severe heart defects; Mitchell, Korones, & Berendes, 1971); (2) born before 1991; (3) diagnosis established before the age of 10 (i.e., before adolescence to warrant

198 sufficient experience of living with CHD); (4) continued follow-up at our center; and (5) physical,
199 cognitive, and language capabilities required to complete the self-report questionnaires. Patients are
200 excluded from study participation if they (1) underwent prior heart transplantation; (2) have primary
201 pulmonary hypertension; or (3) have impaired cognitive abilities. A total of 400 patients who fulfilled
202 these criteria were randomly selected to participate, of which 377 (94%) were retained after a final
203 check of the criteria. All participants received a postal study package including: (1) a study information
204 letter; (2) a copy of the survey package; (3) the informed consent form; and (4) an addressed, pre-
205 stamped return envelope. A total of 276 patients (54.3% men; response rate: 73.2%) returned
206 completed questionnaires. Demographic characteristics are presented in Table 1. Age ranged from 22
207 to 78 years (*M*=36.8, *SD*=11.4). The complexity of heart defects was determined based on Task Force
208 1 of the 32nd Bethesda conference as simple (33.7% of the sample), moderate (54.3%), or complex
209 (12%) (Warnes et al., 2001).

210 *Sample 2.* Patients were selected from the database of rheumatology of the University
211 Hospitals Leuven using the following criteria: (1) diagnosis of SLE or SSc, (2) Dutch-speaking, (3) the
212 patient is able to fill in an informed consent form, (4) the patients' cognitive or medical condition allows
213 for filling out the questionnaire, and (5) absence of a severe psychiatric disorder. A total of 285 patients
214 fulfilled these criteria, who all received a postal study package including: (1) a study information letter;
215 (2) a copy of the survey package; (3) the informed consent form; and (4) an addressed, pre-stamped
216 return envelope. A total of 241 patients (17.4% men; response rate: 85%) returned completed
217 questionnaires (53.1% patients with SLE). Demographic characteristics are presented in Table 1. Age
218 ranged from 17 to 81 years (*M*=52.8, *SD*=14.9). Mean disease duration was 11.36 years (*SD* = 9.60).
219 Informed consent was obtained from all individual participants included in both studies. All procedures
220 performed in both studies involving human participants were in accordance with the ethical standards
221 of the institutional and/or national research committee and with the 1964 Helsinki declaration and its
222 later amendments or comparable ethical standards.

223 ***Measures***

Illness identity. The Illness Identity Questionnaire (IIQ) was used to assess the four illness identity states (Oris et al., 2016). Eight items were initially formulated for each of the illness identity states: rejection, engulfment, acceptance, and enrichment. This item pool was generated based on a broad literature search into existing measures focusing on illness identity or related constructs (e.g., Illness Cognition Questionnaire; Evers et al., 2001). Further, newly generated items, which were semantically based on these measures, were also included in this initial item pool. Patients were asked to indicate how much they agreed with each statement on a 5-point Likert scale ranging from 1 (*strongly disagree*) to 5 (s*trongly agree*).

Depressive and anxiety symptoms. Depressive and anxiety symptoms were measured with the depression and anxiety subscales of the Hospital Anxiety and Depression Scale (HADS; Spinhoven et al., 1997; Zigmond & Snaith, 1983), which consists of seven items for each subscale with a 4-point scale ranging from 0 to 3. Scores can range from 0 to 21, with high scores indicating more depressive and anxiety symptoms. Sample items are "I still enjoy the things I used to enjoy" (depressive symptoms) and "I get a sudden feeling of panic" (anxiety symptoms). Cronbach's alpha's for depressive and anxiety symptoms, were .83 and .87 in patients with CHD and .84 and .85 in patients with MSDs, respectively.

Physical functioning. Physical functioning, as subjectively experienced by the patient, was assessed with a single item. Patients with CHD responded to the item "How much do you experience symptoms from your illness" (illness symptoms) of the Brief Illness Perception Questionnaire, on a 0-10 response scale (Broadbent, Petrie, Main, & Weinman, 2006; de Raaij, Schroder, Maissan, Pool, & Wittink, 2012). Patients with MSDs responded to the pain/discomfort item of the EQ-5D-5L on a 5-point scale from "I have no pain or discomfort" to "I have extreme pain or discomfort" (Herdman et al., 2011).

Statistical Analysis

Analyses were conducted in three steps, according to the three main objectives. First, we conducted principal axis factoring with promax rotation in Sample 1 on the 32 items of the IIQ using SPSS 23

(Brown, 2015). Both the scree test (Cattell, 1966) and parallel analysis (using the 95th percentiles of the distributions of eigenvalues; Buja & Eyuboglu, 1992; Horn, 1965) were used to select the appropriate number of factors (Brown, 2015; Worthington & Whittaker, 2006). All items with loadings less than .40 on their intended factor and/or with cross-loadings exceeding |.32| were deleted in a stepwise approach, because such items could be poor indicators of the intended factor (Brown, 2015; Worthington & Whittaker, 2006). Next, to evaluate the model fit of the factor solution based on EFA, we conducted Confirmatory Factor Analysis (CFA) using Mplus 7 in Sample 2. To deal with non-normal data distributions, Maximum Likelihood Mean Variance (MLMV) was used as a robust estimation method (Kline, 2005). To evaluate model fit, we used the χ^2-index, which should be as small as possible. Given that χ^2- index is sensitive to sample size (i.e., it becomes significant with large sample size; Hu & Bentler, 1999), we additionally used the normed χ^2 (χ^2/df), which should be less than 2 (Ulman, 2013), and used alternative fit indices (Brown, 2015; Kline, 2005): the Root Mean Square Error of Approximation (RMSEA), which should be less than .08; the Comparative Fit Index (CFI), which should exceed .90; and the Standardized Root Mean Square Residual (SRMR), which should be less than .09 (Kline, 2005). Second, multivariate analyses of variance (MANOVA), using Wilks' Lambda, were used to test for mean differences in illness identity (as dependent variable) based on sex, condition (CHD or MSDs), disease complexity in CHD, and diagnosis (SSc or SLE) in MSDs. For age and disease duration, Pearson correlation coefficients were calculated with the four illness identity states. Third, to examine the associations linking illness identity to psychological and physical functioning, Pearson correlation coefficients were calculated (if age and gender correlate significantly with illness identity, partial correlations would be calculated).

Results

Objective 1: Factorial Validity and Reliability of the IIQ

Exploratory Factor Analysis in Sample 1

Factor retention. Based on the scree test four factors were retained. Based on parallel analysis, using the 95th percentiles of the distributions of eigenvalues (Buja & Eyuboglu, 1992; Horn, 1965), seven

factors needed to be retained. However, based on recommendations of Brown (2015), a number of interrelated reasons seemed to indicate that seven would be too many factors to use in our case. First, and foremost, on three of the seven factors only two items had salient loadings, which means these factors are poorly defined and can be eliminated. Second, a limitation of Exploratory Factor Analysis (EFA) is that correlated indicator errors cannot be included. Hence, EFA may suggest more factors while the relationships between some items (indicators) may be better explained by correlated errors signalling method effects rather than additional latent factors. Third, some authors argue that parallel analysis using principal axis factoring tends to select too many factors (Buja & Eyuboglu, 1992). As such, combining these three considerations, a more parsimonious four-factor solution is preferred. Hence, we could conclude that four factors needed to be retained based on exploratory factor analysis.

Item retention or deletion. A total of five items had to be deleted, two items because of no loading above .40 and three items because of a negative cross-loading. Hence, the item pool was reduced to 27 items. Two additional items with relatively low loadings were deleted based on conceptual grounds as well. The final four-factor solution, which explained 60.81% of the variance, consisted of a 7-item enrichment scale, an 8-item engulfment scale, a 5-item acceptance scale, and a 5-item rejection scale. Factor loadings are given in Table 2.

Confirmatory Factor Analysis in Sample 2

After including three error correlations (based on the highest modification indices) between items that are somewhat similarly worded (i.e., method effect for items 6 – 7, items 17 – 18, and items 24 – 25; Brown, 2015), the four-factor model provided an adequate fit to the data of patients with MSDs (df=266; χ^2=382.82, p<.001, χ^2/df=1.44; RMSEA=.046; CFI=.909; SRMR=.067)[7]. Further, all three-factor models demonstrated poor fit to the data and, based on Bayesian Information Criteria and χ^2

[7] Additionally, CFA was conducted in sample 1 (CHD) and full measurement invariance (i.e., configural, metric, and scalar invariance) could be established across both patient samples. This means that the IIQ measures the same concept(s) across both samples (Vandenberg & Lance, 2000), which is necessary to compare mean scores and correlations with psychological and physical functioning across groups. For more information about the analyses of the measurement invariance, readers can contact the corresponding author.

298 difference testing (Brown, 2015), had significantly worse fit than the four-factor model, further
299 testifying to the distinctiveness of the illness identity states. Table 2 presents all standardized factor
300 loadings of the final four-factor solution.

301 *Reliability, correlations, and means of the IIQ*

302 Cronbach's alphas for CHD and MSDs, respectively, were .75/.75 for rejection, .83/.85 for
303 acceptance, .92/.91 for engulfment, and .95/.90 for enrichment. Acceptance correlated positively with
304 enrichment, but negatively with rejection and engulfment in both patient groups. Engulfment
305 correlated positively with rejection and enrichment in patients with CHD, and negatively with
306 acceptance in patients with MSDs (Table 3). All factor correlations were below .80, which points to
307 discriminant validity (Brown, 2015). Ancillary analyses demonstrated that these correlations did not
308 significantly differ across both samples.

309 ***Objective 2: Associations with Demographic and Clinical Parameters***

310 First, age correlated positively with engulfment (r=.18; p=.003), rejection (r=.20; p=.001), and
311 enrichment (r=.15; p=.018) in patients with CHD, and with rejection (r=.23; p<.001) in patients with
312 MSDs. Second, we found no significant multivariate sex effects for illness identity in both patients with
313 CHD (F(1,268)=0.35, p=.847, η^2=.01) and MSDs (F(1,230)=1.93, p=.106, η^2=.03). Third, in patients with
314 MSDs, disease duration correlated positively with acceptance (r=.24, p<.001). Fourth, we found
315 significant multivariate effects of condition for illness identity (F(1,500)=20.02, p<.001, η^2=.14).
316 Patients with MSDs scored higher on engulfment and rejection and lower on acceptance than patients
317 with CHD (See Table 4). Fifth, in patients with CHD, significant multivariate effects of disease
318 complexity were found for illness identity (F(2,267)=2.73, p=.006, η^2=.04). Patients with a complex
319 heart defect scored higher on engulfment and enrichment than patients with a simple heart defect.
320 Patients with a moderate heart defect scored higher on enrichment than patients with a simple heart
321 defect (See Table 5). Finally, in patients with MSDs, we found significant multivariate effects of
322 diagnosis for illness identity (F(1,230)=4.62, p=.001, η^2=.08). Patients with SSc scored higher on
323 engulfment and rejection, and lower on acceptance than patients with SLE (Table 5).

Objective 3: Associations with Psychological Functioning and Physical Functioning

Given that age was significantly correlated with illness identity, correlations were calculated controlling for age. These partial correlations are displayed in Table 3. In both patients with CHD and MSDs, engulfment correlated positively with depressive and anxiety symptoms, and acceptance correlated negatively with depressive and anxiety symptoms. In patients with CHD, engulfment, rejection, and enrichment correlated positively with illness symptoms, but acceptance correlated negatively with illness symptoms. In patients with MSDs, engulfment correlated positively with pain, whereas acceptance correlated negatively with pain.

Discussion

The present study provides initial evidence that subscale scores on the Illness Identity Questionnaire (IIQ; Oris et al., 2016) seem to represent the four intended illness identity states (i.e., engulfment, rejection, acceptance, and enrichment) in patients with CHD and MSDs. Patients' responses to the IIQ were reliable in both samples and were related to psychological and physical functioning as hypothesized (indicative of concurrent criterion validity).

Objective 1: Factorial validity and reliability of the IIQ

In line with a recent study in youth with T1D (Oris et al., 2016), the four illness identity states assessed in the IIQ could be differentiated, and scores on all four subscales proved to be reliable. Hence, the present study demonstrated that subscale scores of the IIQ are valid indicators of illness identity in adults with CHD and MSDs. The correlational pattern was similar across patient samples and indicated that they were distinct but interrelated states (Brown, 2015). Engulfment and rejection were positively interrelated, and were both negatively related to acceptance, as they both were hypothesized to capture a lack of illness integration. Next, acceptance and enrichment were positively interrelated, as they both capture instances of adaptive illness integration. In addition, enrichment and engulfment were positively related, which might be explained by impact of the illness. When individuals experience a substantial impact of chronic illness on their daily life, they may feel engulfed by the illness (Beanlands et al., 2003). However, such an illness impact may also enable people to grow

as a person (Helgeson et al., 2006). To enhance the self in the context of a stressor, individuals indeed initiate cognitive efforts, such as construing benefits, which tend to be greater when stressors are (perceived as) more severe (Taylor & Brown, 1988).

Objective 2: Associations with Demographic and Clinical Parameters

No differences were found between men and women in the way they (fail to) integrate their illness into their identity. Older patients scored higher on rejection in both patients samples, and on engulfment and enrichment in patients with CHD. Older people tend to experience more disability, combined with lower feelings of control as compared to younger people (Heijmans et al., 2004), which makes the illness a potentially greater identity threat (Leventhal et al., 1999). Hence, one can respond by rejecting the illness, because confrontation would be too overwhelming (Beanlands et al., 2003), or one can feel engulfed by the illness, as if the illness takes over control of one's life. On the other hand, this increased threat might also give rise to more enrichment, because the construction of benefits might be greater when stressors are more severe (Helgeson et al., 2006). With regard to disease duration, patients with MSDs were able to accept their illness more when they lived longer with the disease, which is in contrast to the study in youth with T1D (Oris et al., 2016). However, other studies have suggested that patients who lived longer with the illness were able to accept their illness more, because they learned to cope with the illness challenges over time (Sparud-Lundin, Öhrn, & Danielson, 2010). Hence, these inconsistent results suggest that clinicians should realize that a longer disease duration is not necessarily accompanied with more acceptance.

Further, mean differences in illness identity were found across patient samples. Patients with MSDs scored higher on engulfment and rejection, and lower on acceptance than patients with CHD. MSDs might pose larger identity threats than CHD as, for example, unemployment is more often the case in MSDs than CHD (Haythornthwaite et al., 2003; Kovacs et al., 2005). Also, mean differences in illness identity were found within patient samples. First, with respect to CHD, patients with simple, moderate, and complex heart defects showed equal levels of rejection and acceptance, potentially because most patients can successfully engage in adult responsibilities and roles (Kovacs et al., 2005).

This might limit the threat to one's identity, even in the case of moderate or complex heart defects, which might make it less necessary to reject the illness and facilitate acceptance. However, complex heart defects were related to more engulfment and enrichment, and moderate heart defects were related to more enrichment as compared to simple heart defects. Patients with simple heart defects experience few, if any, physical limitations, compared to moderate and complex defects (Kovacs et al., 2005). These aspects of the illness experience may play into experiencing engulfment and enrichment (Beanlands et al., 2003; Helgeson et al., 2006). In sum, patients with simple, moderate, and complex heart defects showed more similarities than dissimilarities on illness identity, but future research should demonstrate which illness aspects play into these dissimilarities.

With respect to MSDs, patients with SSc scored higher on engulfment and rejection, and lower on acceptance than patients with SLE. This is in line with research suggesting that the degree of physical disability is greater in SSc than in other chronic rheumatic diseases, partly because skin thickening and tightening limits hand and limb functioning (Haythornthwaite et al., 2003). This disability limits patients in performing different daily activities, which might interfere with their identity roles, such as work (Haythornthwaite et al., 2003). Hence, it may be more difficult to integrate SSc as part of one's identity.

Objective 3: Associations with Psychological and Physical Functioning

By relating illness identity to psychological and physical functioning, we obtained a more clinically relevant account of the four illness identity states in adults with a chronic illness. As expected, engulfment was related to maladaptive psychological and physical functioning, that is, more depressive and anxiety symptoms and more illness symptoms and pain (Oris et al., 2016). This might be because the chronic illness interferes with other valued self-assets (e.g., social relationships and work) (Luyckx, Goossens, Van Damme, & Moons, 2011). In the other direction, experiencing symptoms and pain may interfere with everyday functioning and behaviors (Leventhal et al., 1999), which might lead to feelings of engulfment.

For patients with CHD, rejection was related to more illness symptoms Individuals might reject their illness as part of their identity in order to avoid that the illness threatens their identity (Leventhal

et al., 1999). Hence, when the threat is appraised as more severe, for example when patients experience more symptoms, they might reject the illness more, as a defense mechanism (Mozzetta et al., 2008). In line with results in adolescents and emerging adults with type 1 diabetes, rejection was unrelated to depressive (and anxiety) symptoms (Oris et al., 2016). By avoiding confrontation with a chronic illness, rejection might limit the emotional impact of the illness.

In line with previous research (Oris et al., 2016), acceptance was related to less depressive and anxiety symptoms, and less illness symptoms and pain. Hence, the present findings testify to the importance of integrating an illness in one's identity and retaining a coherent identity, as acceptance was strongly related to adaptive functioning. Acceptance might enable patients to better cope with illness challenges and might also lead to better self-care behaviors (Luyckx et al., 2010; Richardson et al., 2001), which might lead to better psychological and physical functioning (Karademas et al., 2009). However, in the other direction, experiencing few symptoms and pain might enhance acceptance, because the illness does not interfere much with everyday functioning and behaviors (Leventhal et al., 1999).

Finally, enrichment was related to more illness symptoms in patients with CHD, as individuals have to experience a substantial impact of their illness in order to be able to grow as a person (Helgeson et al., 2006).

Clinical Implications

Given the cross-sectional nature of our study, only some preliminary clinical implications can be formulated. The concept of illness identity may provide a valuable integrative framework in clinical practice to understand and recognize how patients integrate (or fail to do so) a chronic illness into their identity (Zangi, Hauge, Steen, Finset, & Hagen, 2011).. Hence, acknowledging patients' core identity issues might be an important first step in clinical practice, in order to help patients become more aware of how their illness impacts their daily life and how they perceive themselves. To that extent,the IIQ could be used to assess illness identity in clinical care. However, longitudinal studies that provide information on antecedents, mechanisms, and outcomes of illness identity, are necessary to

understand how illness identity is related to psychological and physical functioning. Based on such longitudinal studies, more profound clinical implications may be formulated

Limitations and Suggestions for Future Research

The present study is characterized by some limitations. First, the most important limitation is the cross-sectional study design, which does not allow drawing conclusions on the directions of effects linking illness identity and functioning. Hence, in the current study, the evidence for validity of subscale scores on the IIQ is limited to concurrent criterion validity, which is a weaker form of validity than predictive validity. More specifically, based on our cross-sectional data, we cannot conclude whether illness identity is an antecedent or consequence of psychological and physical functioning. In other words, illness identity might predict psychological and physical functioning, but psychological and physical functioning might also predict the way in which individuals integrate their illness into their identity (i.e., illness identity). These mutual relations need to be investigated in future longitudinal research. Future research should also investigate how illness identity emerges and develops over time, by focusing on younger age groups such as adolescents. Second, all measures were self-report questionnaires. Although these are the most appropriate method to gather information regarding internal processes such as identity, other methods (e.g., interviews or objective physical functioning) should be used in future research. This will allow a more in-depth understanding of illness identity. Third, future research should examine the associations between illness identity and illness perceptions from the Common Sense Model (CSM; Leventhal, Meyer, & Nerenz, 1980), which are different but related constructs (Benyamini, 2011; Leventhal et al., 1999). Indeed, illness perceptions are part of the broader self-concept (Benyamini, 2011), but they do not explicitly capture the degree to which individuals manage to integrate their illness into their identity (i.e., illness identity; Oris et al., 2016). However, in order to know how a person understands his (or her) chronic illness, we do not only need to understand how a person perceives the illness and its treatment (i.e., illness perceptions), but also how a person views him- or herself as a person *with* an illness (i.e., illness identity; Kihlstrom & Kihlstrom, 1999). In the present manuscript, we only used one illness perception, illness symptoms, as

a measure of subjective physical functioning in patients with CHD (Leventhal et al., 1980), but future research should examine the relationship between illness perceptions and illness identity more in depth. Fourth, our sample consisted solely of Caucasian European patients from a single-center setting in Belgium. Although University Hospitals Leuven are the largest in Belgium, this might reduce the generalizability of our findings. Fifth, we do not claim that the four illness identity states are exhaustive. Other states might fit into our framework on illness identity as well and might be added based on future research. However, this is the first time that four states are investigated together within an integrative identity framework. Finally, because of different study designs, different measures of physical functioning were used in both patient samples. Preferably, similar measures should be used in future research to increase comparability of the results.

Conclusion

In sum, the scores of the IIQ are valid and reliable to capture four different ways of integrating an illness into one's identity in adults with a chronic illness. Further, as expected, engulfment and rejection capture rather maladaptive illness identities, whereas acceptance and enrichment are more adaptive ways of illness integration. Hence, these findings demonstrate the need of differentiating among these four illness identity states in adults with a chronic illness.

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Table 1

Demographic and Clinical Characteristics of Patients with CHD and MSDs

Variables	CHD	MSDs
Educational level (*n*=274/*n*=240)		
University degree	52 (19.0)	25 (10.4)
University college degree	80 (29.2)	58 (24.2)
High school degree	131 (47.8)	126 (52.5)
Less than high school degree	11 (4.0)	31 (12.9)
Work situation (*n*=275/*n*=241)		
Working fulltime	174 (63.3)	47 (19.5)
Working part-time	47 (17.1)	39 (16.2)
Disability/government financial assistance	24 (8.7)	55 (22.7)
Retired	10 (3.6)	69 (28.6)
Homemaker	8 (2.9)	/
Seeking for a job or unemployed	7 (2.5)	6 (2.5)
Other	5 (1.8)	25 (10.4)
Relationship (*n*=275/*n*=241)		
Married/remarried	127 (46.2)	161 (66.8)
Unmarried/never married	77 (28.0)	19 (7.9)
Living with a partner	58 (21.1)	26 (10.8)
Seperated/divorced	12 (4.4)	21 (8.7)
Widowed	1 (0.4)	12 (5.00)
Other	/	2 (0.8)

Note. Data are presented as numbers and percentages (within parentheses).

Table 2

Standardized factor loadings of the Illness Identity Questionnaire

	EFA Sample 1				CFA Sample 2
	1	2	3	4	
Engulfment items					
18. My illness limits me in many things that are important to me.	.86				.71
17. My illness prevents me from doing what I would really like to do.	.84				.64
15. My illness completely consumes me.	.82				.79
16. It seems as if everything I do, is influenced by my illness.	.79				.82
14. My illness influences all my thoughts and feelings.	.75				.80
11. My illness dominates my life.	.75				.77
13. I am preoccupied with my illness.	.72				.67
12. My illness has a strong impact on how I see myself.	.70				.72
Rejection items					
5. I just avoid thinking about my illness.		.74			.66
4. I never talk to others about my illness.		.68			.53
3. I hate being talked to about my illness.		.68			.73
2. I'd rather not think of my illness.		.53			.57
1. I refuse to see my illness as part of myself.		.41			.57
Acceptance items					
6. My illness simply belongs to me as a person.			.88		.54
7. My illness is part of who I am.			.79		.57
9. I am able to place my illness in my life.			.72		.89
8. I accept being a person with a illness.			.61		.78
10. I have learned to accept the limitations imposed by my illness.			.47		.68
Enrichment items					
23. Because of my illness, I have learned a lot about myself.				.90	.83
20. Because of my illness, I know what I want out of life.				.89	.68
21. Because of my illness, I have become a stronger person.				.87	.80
22. Because of my illness, I realize what is really important in life.				.85	.81
19. Because of my illness, I have grown as a person.				.83	.69
25. Because of my illness, I have learned to enjoy the moment more.				.81	.76
24. Because of my illness, I have learned to work through problems and not just give up.				.80	.65

Note. Only factor loadings exceeding |.32| are presented. For CFA, all factor loadings are significant at $p<.001$. Illness was formulated as "heart defect" and "lupus/scleroderma" in Sample 1 and Sample 2, respectively.

Table 3

(Partial) Correlations Among Illness Identity and Psychological Functioning

	1.	2.	3.	4.	5.	6.	7.
Illness identity							
1. Engulfment	1	.21***	-.43***	.22***	.55***	.45***	.61***
2. Rejection	.11	1	-.35***	-.01	.10	.02	.16**
3. Acceptance	-.39***	-.23**	1	.20**	-.22**	-.21***	-.26***
4. Enrichment	.11	.06	.20**	1	-.00	-.01	.22***
Functioning							
5. Depressive symptoms	.58***	.11	-.33***	-.10	1	.62***	.44***
6. Anxiety symptoms	.45***	.11	-.32***	-.12	.66***	1	.36***
7. Illness symptoms[a]/ pain[b]	.40***	.07	-.17**	-.05	.30***	.43***	1

*p<.05. **p<.01. ***p<.001. Correlations of patients with CHD are presented above the diagonal, correlations of patients with MSDs below the diagonal. Superscripts a and b refer to patients with CHD and MSDs, respectively. Correlations between illness identity and functioning were controlled for age.

Table 4

Univariate ANOVAs, Means, and F-values for Patient Sample

Variables	Total	Patient Sample		*F*-value (η^2)
		CHD	MSDs	
Illness identity				
Engulfment	2.11 (0.96)	1.84 (0.88)	2.43 (0.95)	50.74*** (.02)
Rejection	2.74 (0.95)	2.63 (0.95)	2.88 (0.93)	8.88** (.11)
Acceptance	3.86 (0.93)	4.14 (0.81)	3.53 (0.95)	62.20*** (.09)
Enrichment	3.01 (1.08)	3.04 (1.15)	3.04 (1.16)	0.56 (.001)

Note. SD's are given within parentheses.

*p<.05; **p<.01; and ***p<.001.

Table 5

Univariate ANOVAs, Means, and F-values for Disease Complexity (CHD) and Diagnosis (MSDs)

Variables	Total CHD	Disease Complexity (CHD)			*F*-value (η^2)	Total MSD	Diagnosis		*F*-value (η^2)
		Simple	Moderate	Complex			SLE	SSc	
Illness identity									
Engulfment	1.85 (0.88)	1.70 (0.83)[a]	1.86 (0.89)[a, b]	2.19 (0.92)[b]	3.61* (.03)	2.43 (0.95)	2.25 (0.90)	2.63 (0.98)	9.57** (.04)
Rejection	2.63 (0.95)	2.75 (0.96)	2.54 (0.94)	2.69 (0.94)	1.53 (.01)	2.88 (0.93)	2.73 (0.99)	3.05 (0.83)	6.88** (.03)
Acceptance	4.15 (0.81)	4.23 (0.67)	4.13 (0.86)	4.00 (0.94)	1.11 (.01)	3.53 (0.95)	3.67 (0.98)	3.37 (0.88)	5.63* (.02)
Enrichment	3.04 (1.16)	2.76 (1.17)[a]	3.14 (1.13)[b]	3.38 (1.12)[b]	4.75** (.03)	2.97 (0.97)	3.05 (1.03)	2.89 (0.90)	1.49 (.01)

Note. SD's are given within parentheses. For disease complexity, means sharing a common superscript are not statistically different at $p<.05$ according to the Tukey HSD procedure.

*$p<.05$; **$p<.01$; and ***$p<.001$.