

PATIENT ENGAGEMENT IN HEALTH AND WELL-BEING: THEORETICAL AND EMPIRICAL PERSPECTIVES IN PATIENT CENTERED MEDICINE

EDITED BY: Guendalina Graffigna and Elena Vegni
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PATIENT ENGAGEMENT IN HEALTH AND WELL-BEING: THEORETICAL AND EMPIRICAL PERSPECTIVES IN PATIENT CENTERED MEDICINE

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At present citizens are more aware of their health and care rights and more literate about their disease. Furthermore the continuous development of technological and bio-medical solutions are alimending the expectation for longer and better life expectancy, even despite the diagnosis. Patients require to be higher involved in the decision making about their care and are willing to deeply entangle all the possible treatment options, their advantages, and their risks. In other terms, citizens today want to be treated not only as “client” but mainly as partners of the medical action and as co-authors of the success of their healthcare pathway.

Due to this socio-psychological change in patients’ attitude, healthcare systems today are claimed to a deep revision of their practices and organizational models in order to become better responsive to patients’ expectation and more sustainable and effective in the management of their services.

Patient participation and engagement in healthcare management, indeed, is today acknowledged by policy makers and healthcare experts as a valuable option to orient changes and actions of the healthcare systems. Several empirical studies have demonstrated the positive outcomes of a participatory care approach at the clinical, psychosocial, and economic levels. Patient Engagement, thus, appears today not only an ethical but also a pragmatic imperative for the innovation and the improvement of healthcare system.

Moving from these premises, this e-book collect first research experiences, conceptual contribution and review of good practices in the area of Patient Engagement promotion. The e-book also discuss the relevance and the theoretical linkages between the concept of Patient Engagement and that one of Patient Centered Medicine.

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Editorial: Consumer Engagement in Health and Well-being: Theoretical and Empirical Perspectives in Patient Centered Medicine

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Keywords: consumer engagement, health, well-being, patient-centered medicine, health behavior

Editorial on the Research Topic

Consumer Engagement in Health and Well-being: Theoretical and Empirical Perspectives in Patient Centered Medicine

The growing understanding of the key role of people in improving healthy behaviors and clinical outcomes has led healthcare to search for innovative ways to foster individuals' roles in the care and health promotion processes. It comes as no surprise, therefore, that making consumers active agents in their health and care is up today recognized as a key priority for services' innovation. In the era of participatory health, the concept of "engagement" may be particularly promising to give consumers a starring role in managing their health and well-being.

The healthcare field has recently introduced the term "engagement" in its lexicon to call for a renewed partnership among the actors (i.e., patients, caregivers, practitioners, decision makers...) implied in the health and care management. Overall, the concept of engagement attempts to offer a compass for action in the current complex and uncertain context of healthcare design and health promotion initiatives. The main aim is giving (back) a leading role to patients and taking them on board for a more efficient and effective process of care delivery and of health promotion initiatives. Furthermore, consumer health engagement can be the key to systematically diagnose and make sense of the different organizational, relational, and psychological components in play in the dynamic exchange between "demand" and "supply" of health and care. This challenge could or even should be integrated with a complex attempt coming from the literature on medicine and regarding a new medical model that should be offered to patients/clients/consumers: that of a patient centered medicine, based on a biopsychological epistemology.

Patient engagement may be defined as an umbrella term that qualifies the systemic relation that occurs between the demand and the supply of healthcare, at different levels and in different situation. If considered according to this meaning, patient engagement overarches the other terms more traditionally used to denote the active role of patients in their care such as patient adherence, patient compliance, patient involvement, patient participation, and patient activation (Graffigna et al., 2016).

Precisely, the concept of "activation" differs from the concept of "engagement" since it is mainly referred to the knowledge, attitude, and skills of patients in their self-management; while the concept of "engagement" is also applicable to the patients' proactive role in other settings, such as health promotion and prevention. Furthermore, the motivational and emotional nature of "engagement" is crucial and differentiates this concept from activation which is rather more cognitive and behavioral in its nature.

Other concepts are strongly anchored to the area of disease management. For instance, the classical concepts of "adherence" and "compliance" express a hierarchical representation of the

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clinical consultation, where the healthcare professional is considered as “the expert” and he/she prescribes to the patient (not expert) the rules to manage his/her disease. These concepts imply the implicit evaluation of the patient behavior in self-management such as more or less good, more or less able to respond adequately to the expert’s requirements (Haynes et al., 1979; Vlasnik et al., 2005). Furthermore, these concepts evoke a medicalizing idea of the patient such as passive and needing to be correct in order to better functioning, not only at the clinical but also at the psychosocial level. It is evident how the concept of “engagement” move from a very different philosophy and representation of the patient role along the healthcare journey. From this perspective, the different actors implied in the healthcare journey are considered “experts” based on their specific subjective experience of illness and of its management. The concept of “engagement” aims at democratizing the clinical consultation and at legitimizing care receivers in a more starring role.

On the other hand, the concepts of “involvement” and “participation,” refers instead to the dyadic context of the medical consultation and the cognitive/emotional attitude of the patient to the negotiation of clinical decision making. There is an evident conceptual link between the concepts of “involvement,” “participation,” and the concept of “engagement.” All of these concepts advocate for the proactive role of patients in the decisions about care. However, these concepts relate to different levels of healthcare services demand-supply exchange (Murray et al., 2006; Thompson, 2007), since the concepts of “involvement” and “participation” are mostly limited to the dyadic context of the doctor-patient consultation in shared decision making, whereas the concept of “engagement,” refers to the role of patients and to how he/she approaches the healthcare system in its complexity (where medical consultation is one of the possible settings).

Finally, the concept of “empowerment” entertains evident areas of overlapping and potential synergies with the concept of “engagement,” although with a different breadth. The concept of “empowerment” relates to the level of patients’ power and of agency upon their healthcare condition. Particularly, “empowerment” refers to patients’ reacquisition of the subjective sense of control over their disease (Aujoulat et al., 2007). Given this definition, “empowerment” may be defined as a potential prerequisite for the process of engagement, although in turn it is fed by the good experiences that the patient makes in his evolutionary journey of exchange with the healthcare system, and thus along the “patient engagement journey” itself. Furthermore, if “empowerment” is primarily a cognitive and behavioral condition, “engagement” is nurtured by the emotional and motivational components of patients experiences along the care pathway.

Alongside with this desire to engage patients a tendency toward a more patient centered clinical intervention has developed. At the end of the 60 s, Balint introduced the term of patient centered medicine focusing the attention, during the

medical encounter, not only to the biological-technical aspects but also on the emotional and relational dimensions (Balint, 1964). The patient-centered medicine is a large concept variously described, till to the proposal of a “transformed clinical method” by the Canadian group: they suggested to involve the patient and consider his/her own perspective (Stewart et al., 2003) not only in the clinical encounter but in the process of care. The patient centered medicine is conceptualized as a clinical method of a bio-psycho-social model of care and has the aim to transform the clinical approach to patients and enhance their involvement. So far, patient engagement and the patient-centered medicine seem to have a common ground and similar aims, but their overlapping is not clear and not frequently explored in the literature. Furthermore, shared guidelines about how to translate into the clinical practice the imperative of patient engagement have still to come. In this light, this Frontiers Research Topics has been conceived as an arena to bridge research and theoretical expertise mastered in different disciplinary domains in order to set the ground for a shared definition of what consumer engagement in health and well-being is and on useful guidelines for practice a consumer engagement in a patient centered medicine.

We invited authors from different disciplinary domains to contribute original as well as review article in order to set a debate about patient engagement applications and its conceptual relationship with patient centered medicine.

In the present issue, readers will find the two concepts differently approached. The importance of engagement in health and well-being increased in the last years outlining the role of patient and legitimizing the starring role in the care receivers is described in the contribution by Bigi: in particular the issue that engagement involves a profound change in the doctor’s behavior is discussed. The increase of chronic diseases has highlighted the need for increased engagement in patients (Menichetti and Graffigna; Zhang et al.). Menichetti and Graffigna underlined the importance of intervention (PHEinAction) to support patient engagement and lifestyle change and maintenance, facilitating emotional and psychological processes. The relevance of emotion in engagement process appears also in the study conducted on patients with HIV by Leone et al. The importance of this construct in cancer studies (Villani et al.; Saita et al.), and mental health studies (Degli Stefani and Biasutti; Singh et al.; Oliveira-Maia et al.) seems to suggest the usefulness of psychosocial interventions in addressing the care, for patients and the family. Last but not least, the training for health care workers appears to be crucial in order to enable cares to engage patient, particularly for nurses to learn strategies and assessment measures in clinical practice are key points in supporting the realization of patient engagement in healthcare (Barello et al.).

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All authors listed have made a substantial, direct and intellectual contribution to the work, and approved it for publication.

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Illness Representations of HIV Positive Patients Are Associated with Virologic Success

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Introduction: It is important for HIV positive patients to be engaged in their care and be adherent to treatment in order to reduce disease progression and mortality. Studies found that illness representations influence adherence through the mediating role of coping behaviors. However, no study has ever tested if patient engagement to the visits mediate the relationship between illness perceptions and adherence. This study aimed to explore illness representations of HIV positive patients and test the hypothesis that illness representations predict adherence through the mediating role of a component of behavioral engagement.

Methods: HIV-positive patients treated with highly active antiretroviral therapy (HAART) for at least one year and presenting to a check-up visit were eligible to participate in the study. Patients completed the Illness Perception Questionnaire-Revised. Behavioral engagement was measured based on the patients' clinical attendance to the check-up visits; adherence to HAART was measured by viral load. Undetectable viral load or HIV-RNA < 40 copies/ml were considered indexes of virologic success.

Results: A total of 161 patients participated in the study. Most of them coherently attributed the experienced symptoms to HIV/HAART; perceived their condition as chronic, stable, coherent, judged the therapy as effective, and attributed their disease to the HIV virus and to their behavior or bad luck. The majority of patients (80.1%) regularly attended check-up visits and 88.5% of them reached virologic success. The mediation model did not show good fit indexes. However, a significant direct effect of two independent variables on virologic success was found. Specifically, the perception that the disease does not have serious consequences on patient's life and the prevalence of negative emotions toward HIV were associated with virologic success. On the contrary, the patient's perception that the disease has serious consequences on his/her life and the prevalence of positive emotions were associated with virologic failure. This model showed good fit indexes (CFI = 1; TLI = 1; RMSEA = 0.00; and WRMSR = 0.309).

Discussion: Results do not support the mediating role of behavioral engagement in the relationship between illness representations and adherence. As perception of serious consequences coupled with positive emotions are directly associated with virologic failure, clinicians should take them into account to promote treatment adherence.

Keywords: adherence, HAART, HIV, illness representations, IPQ-R, patient engagement, virologic success

INTRODUCTION

In the last 20 years, the care of HIV positive patients has changed dramatically. The introduction of highly active antiretroviral therapy (HAART) has allowed patients to achieve an undetectable plasma viral load (HIV – RNA level) and so a virologic success (Das et al., 2010; Gill et al., 2010), thus reducing mortality and morbidity (Mocroft et al., 2003). Due to this pharmacological success, HIV infection has moved from a mortal disease to a chronic condition. The key to virologic success seems to be the adherence to HAART (Gardner et al., 2011). Although viral suppression has become possible with moderate adherence (less than 95%) to HAART regimens, adherence is fundamental in reducing HIV related symptoms, mortality, and the side effects related to the treatments (Paterson et al., 2000; García et al., 2002; Bangsberg, 2006).

It is known that adherence is a complex behavior, influenced by the type of treatment regimen, patient–provider relationship and patient characteristics, including psychological factors (Chesney, 2003; Mills et al., 2006). In order to improve adherence and promote treatment success it is crucial to develop models that predict adherence (DiMatteo et al., 2012) and identify risk factors for non-adherence (Paterson et al., 2000; Ickovics and Meade, 2002; Wood et al., 2003). Psychological models that posit a relation between psychological factors and the management of a chronic illness can be broadly categorized (Horne and Weinman, 1998) as social cognitive models, such as the health belief model (Rosenstock, 1974), stage models, such as the precaution adoption process model (Weinstein, 1988), and the hybrid common-sense self-regulatory model (CS-SRM) of illness representations (Leventhal et al., 1980). The CS-SRM has been adopted by several researchers to predict adherence in several patient populations (e.g., Meyer et al., 1985; Weinman et al., 1996). This model suggests that patients create mental representations of their illness experience through cognitive and emotional processes in order to make sense of it (Leventhal et al., 1997; Cameron and Leventhal, 2003). Cognitive illness representations are composed of five essential dimensions: (1) identity (the nature of the illness and the symptoms the patient considers associated to the disease), (2) cause (personal beliefs about the cause of the illness), (3) timeline (the perceived chronicity of the illness), (4) consequences (perceptions about the short- and long-term effects of the illness), and (5) control (beliefs about the degree of illness and if treatment can be controlled). Beside these cognitive representations, emotions elicited by illness, such as fear, anger, or anxiety, are also integral to illness representations and develop simultaneously with the cognitive components (Leventhal et al., 1997). Based on the CS-SRM, Weinman et al. (1996) developed the Illness Perception Questionnaire (IPQ), in order to measure cognitive illness representations. The IPQ was later revised by Moss-Morris et al. (2002) into the Illness Perception Questionnaire Revised (IPQ-R) to include also the emotional components of patients' representations.

According to the CS-SRM, when faced with a health-related problem, patients undergo a process, named the self-regulation process, which encompasses three phases: (1) illness representations; (2) coping strategies; and (3) evaluation (Broadbent et al., 2006). First, the patient seeks to understand the illness. From the various internal stimuli, such as illness activity or side effects of the disease, and external information, such as relationship with healthcare providers or public opinion, the cognitive and emotional representations of the health threat are constructed. Thus, illness representations are not necessarily scientifically based as they are often formulated from personal experience, social influences, and/or interaction with healthcare providers. Second, illness representations will lead to the selection of coping strategies to eliminate or control the threat posed by the disease (Broadbent et al., 2006). There are several classifications of coping strategies. One of the most common classifications divided the coping strategies into problem-focused strategies, such as gathering information, and taking action to manage the problem, and emotion-focused strategies aimed to manage distress by minimizing, reducing, or preventing the emotional components of a stressor, such as venting emotions, distracting or using avoidance strategies (Lazarus and Folkman, 1984). Third, patients evaluate the effectiveness of the coping strategy on the outcome or goal of the disease, as, for example, virologic success.

The usefulness and validity of the CS-SRM model has been confirmed by numerous studies on patients suffering from several chronic diseases, such as diabetes, cardiovascular disease, asthma, and cancer (Petrie and Weinman, 1997; Cameron and Leventhal, 2003; French et al., 2013; Mickevičienė et al., 2013). Several studies on different chronic illnesses have confirmed the mediating role of coping strategies in the relationship between illness representations and outcomes, such as mood, quality of life, and patients' satisfaction (Diefenbach and Leventhal, 1996; Leventhal et al., 2001; Rutter and Rutter, 2002; Hagger and Orbell, 2003; Llewellyn et al., 2007; Gould et al., 2010; Catunda et al., 2016). However, some studies have suggested that illness representations are also directly associated with outcomes regardless of coping strategies, and that illness representations are more strongly associated with outcomes than coping strategies (e.g., Moss-Morris et al., 1996; Scharloo et al., 1998). In addition, some studies that did not measure coping in their research designs have shown strong direct relationships between some illness representations and outcome measures (Petrie et al., 1996; Schiaffino et al., 1998). Illness representations were found to predict patients' self-management of their disease (Leventhal et al., 1997; Petrie and Weinman, 1997; Cameron and Leventhal, 2003), decisions to seek health care and comply with medical advice (Leventhal et al., 1980), functional adaption and adherence (Moss-Morris et al., 2002).

Overall, these studies highlight that coping strategies may not be the only mediator in the relationship between illness representations and adherence. Recently, patient engagement has received increased attention in the healthcare literature as a psychological factor that may influence adherence. The increasing number of people living with a chronic condition

brings to light the importance of engaging patients in their health, helping them to integrate the disease into their identity and life, self-manage their disease and properly use the healthcare resources (Graffigna et al., 2014). A recent model developed by Graffigna et al. (2014) defined patient engagement as a dynamic process, in which patients experience four phases (blackout, arousal, adhesion, and eudaimonic project), each encompassing emotional, cognitive, and behavioral dimensions. According to this model, engagement is the final outcome of a series of emotional, cognitive, and behavioral reframing of the patient's health condition (Barello and Graffigna, 2014). More specifically, fully engaged patients are able to integrate the disease into their identity and life, manage their own care and mobilize healthcare services proactively if needed. Among the different aspects of engagement, a behavioral dimension is highlighted that relates to the patient adhesion to medical prescriptions, retention in care and visit attendance. The importance of engagement in HIV care has been acknowledged by several authors as it was found to be associated with HIV outcomes and reduced risk transmission behaviors (Marks et al., 2005; Giordano et al., 2007; Metsch et al., 2008; Yehia et al., 2014).

Illness representations and engagement seem to play an important role in understanding HIV positive patients' self-care behaviors and treatment adherence (Sacajiu et al., 2007; McGavock and Trehan, 2011). However, to our knowledge, no study has ever explored the relationship between illness representations, patient engagement and adherence.

Few studies have explored the relationship between illness representation and adherence in HIV positive patients (Cooper et al., 2009; Reynolds et al., 2009; Pala Norcini and Steca, 2015). Reynolds et al. (2009) found that cognitive illness representations were associated with self-care frequency and effectiveness in the context of HIV care. Cooper et al. (2009) found that adherence to HAART was influenced by individuals' experiences of both HIV and HAART-related symptoms. Pala Norcini and Steca (2015) identified three configurations of perceptions of illness influence (low, moderate, and high) using the Brief IPQ-R questionnaire. They found that a higher perception of illness influence on patients' lives (in terms of consequences, negative emotions and intensity of symptoms) was associated with greater viral load, with the mediating role of dysfunctional coping strategies in response to HIV-related stressors. In particular, this research found that high and moderate illness influence perception correlated with passive coping, which consisted in a lack of action in response to HIV-related stressors. Passive coping might also be seen as patients' perception of helplessness or behavioral disengagement due to their expectation of poor outcomes (Carver et al., 1989).

Based on the results of the literature presented, we hypothesized that the relationship between illness representations and adherence in HIV positive patients could be mediated by behavioral engagement. The present study has two aims: (1) to explore illness representations of HIV positive patients in HAART and (2) to test the hypothesis that illness representations predict adherence through the mediating role of

a component of behavioral engagement. Specifically, we tested if regular visit attendance mediates the relationships between illness representations and virologic success.

MATERIALS AND METHODS

Participants

Participants were recruited at the outpatient clinic of infectious diseases of a university hospital in the north of Italy. This center cares for an average of 1000 HIV positive patients per year.

Inclusion criteria were: (1) HIV patients under treatment for at least 1 year; (2) age ≥ 18 years; (3) able to understand and provide informed consent; (4) able to understand Italian according to (depending on) the physician's and/or researcher's judgment; (5) no history of psychiatric symptoms; and (6) no actual alcohol or drugs abuse.

Data Collection

Participants were recruited in the waiting room, before their check-up visit. A researcher with training in clinical psychology presented the study to the patients. Patients who accepted to participate in the study signed an informed consent and were asked to complete the IPQ-R (Giardini et al., 2007). Socio-demographic, clinical, and adherence data were collected from inspection of the medical records. The research protocol was approved by the hospital ethics committee.

Measures

Socio-Demographic and Clinical Data

Socio-demographic information that was collected included gender, age, relationship status, educational level, nationality. Clinical information included HIV mode of transmission, year of HIV diagnosis, and therapeutic regimen (fix dose, other drugs, and number of pills per day).

Illness Representations

Illness representations were measured with the Italian version of the IPQ-R (Giardini et al., 2007). The questionnaire is composed of three sections: (a) identity; (b) opinions, and (c) causes. While items on identity section have dichotomous responses (yes/no), items regarding opinions and causes are rated by patients on a 5-point Likert scale (from "1 = completely disagree" to "5 = completely agree").

(a) The *identity* section explores patients' beliefs about the disease's nature. Out of 14 symptoms, patients are asked to mark the ones they have experienced since being diagnosed with HIV and, which they believed to be linked to the disease/treatment. Two subscales were measured: *reported symptoms* and *associated symptoms*. A high score on identity scales indicates a great number of symptoms experienced and attributed to the disease.

(b) The *opinions* section is composed of 38 items exploring patients' illness representations. Items are grouped into seven subscales: (1) *timeline* (perception of the disease as chronic); (2) *cyclical symptoms* (perception of a cyclic disease); (3) *consequences* (perception that the disease has serious physical,

psychological, and social consequences on the patient's life); (4) *personal control* (perception that actions can be taken to effectively manage the disease); (5) *treatment control* (high trust in the treatment and its efficacy); (6) *coherence* (high understanding of the disease); and (7) *emotional representations* (prevalence of negative emotions related to the disease).

(c) The *causes* section lists 18 possible illness causes and patients have to rate their level of agreement with each item as a cause of their disease. Only the three most represented causes were used for statistical analysis.

Behavioral Engagement

According to Graffigna et al. (2014), engagement is a complex concept encompassing cognitive, emotional, and behavioral dimensions. In this study, a behavioral component of engagement was assessed by measuring the patients' regular attendance to the last check-up visit. Check-up visits are scheduled by the clinicians every three months, according to the internal guidelines of the clinic of infectious disease where the study was conducted.

Adherence

Based on the literature on adherence measures in HIV (Marcellin et al., 2013), an objective index measuring the viral load (level of HIV-RNA copies/ml) was adopted. The use of an objective index is a headway compared to previous studies on predictors of HIV patients' adherence that commonly used self-reported measures (Ubbiali et al., 2008). An undetectable viral load or HIV-RNA < 40 copies/ml, observed at least at the two previous visits, was considered as virologic success. Virologic success as such is a clinical outcome. However, it was considered as an index of adherence because it is reached only through a regular and correct therapy intake. Indeed, in the clinic of infectious disease where the study was conducted, patients undergo a screening for HIV/RNA baseline, CD4⁺ baseline and drug resistance before starting the HAART treatment. The optimal HAART therapy, which is appropriated also for drug resistance, is then chosen for each patients according to these screening results. Therefore, regardless of their baseline viral load, patients are expected to reach virologic success after three to six months of treatment if they adhere to prescriptions.

Data Analysis

Means, standard deviations, frequencies, percentages, skewness, and kurtosis were used to describe demographic, clinical, IPQ-R, and adherence data. Subscales with skewness and kurtosis indexes > |1| were normalized with a logarithmic transformation.

Correlations between IPQ-R dimensions (identity and opinions) and socio-demographic variables were conducted. Coefficients were calculated according to the type of socio-demographic variable.

A mediation model was tested through structural equation modeling. IPQ-R dimensions (identity and opinions) were entered as independent variables, clinical attendance as a mediator, and virologic success as dependent variable. The socio-demographic variables which significantly correlated with IPQ-R dimensions were used as control variables. All direct and indirect effects were calculated. The model was evaluated by using the

following fit indices: root mean square error of approximation (RMSEA), comparative fit index (CFI), Tucker Lewis index (TLI), and weighted root mean square residual (WRMSR). Values below 0.08 at the RMSEA (Browne and Cudeck, 1993), and values above 0.90 or higher at the CFI and TLI (Bentler, 1990) were judged as indicating an acceptable fit. Values below 0.06 at the RMSEA, and values above 0.95 at the CFI and TLI were judged as indicating a good fit (Hu and Bentler, 1999). We considered values <1 at WRMR as indicating a good fit (Yu, 2002).

The model was modified by removing the variables which did not show significant relations, until a good fit was reached. Data were analyzed using SPSS version 21.0 for Windows and Mplus version 4.0 for Windows.

RESULTS

Of 231 eligible patients, 171 (74%) participated in the study. Ten patients did not return the questionnaire or returned it incomplete, therefore 161 questionnaires were collected.

Demographic and clinical characteristics of participants are shown in Table 1.

Illness Perception and Relationship with Socio-Demographic and Clinical Variables

Identity

One hundred and forty patients (87%) reported to have suffered from at least one of the symptoms listed since being diagnosed with HIV (mean = 4.8 ± 3.5 , range 0–14). The most frequently reported symptoms were: fatigue (67.5%), loss of strength (52.2%), and sleep difficulties (41.3%); 98 patients (60.9%) deemed that at least one of the symptoms experienced was linked to HIV/HAART (mean = 2.6 ± 3.1 , range 0–13). The most frequently symptoms associated to HIV/HAART were: loss of strength (72.8%), weight loss (68.5%), and nausea (65%).

Opinions

Means illness representation of the seven opinions are reported in Table 2. Timeline, personal control, treatment control, and coherence' means are skewed to the higher values of the IPQ-R range; while cyclical symptoms' mean is skewed to the lower values of the IPQ-R range.

Causes

The three most frequently perceived reasons for becoming HIV were: a germ or virus (88.6%), my own behavior (70.4%), and chance or bad luck (54.1%) (Figure 1).

Relationship between Illness Representation, Behavioral Engagement, and Adherence

The majority of patients (80.1%) showed a regular attendance to the check-up visits, and 88.5% of the patients reached a virologic success. The correlation matrix between IPQ-R dimensions (identity and opinions) and socio-demographic variables is

TABLE 1 | Participants' socio-demographic and clinical data.

Characteristics	Value
Gender, <i>n</i> (%)	
Male	120 (74.5)
Age	
Mean (<i>SD</i>), range	45.2 (10.3), 21–78
Relationship status, <i>n</i> (%)	
Single	97 (63.8)
Cohabiting/married	39 (25.7)
Divorced/widow	16 (10.5)
Educational level, <i>n</i> (%)	
Primary	46 (29.3)
High school	83 (52.9)
Graduate	28 (17.8)
Nationality, <i>n</i> (%)	
Italian	148 (91.9)
Eastern European	1 (0.6)
South American	7 (4.3)
African	3 (1.9)
Asian	2 (1.2)
HIV mode of transmission, <i>n</i> (%)	
Homosexuals	70 (45.2)
Heterosexuals	48 (31)
Drug addiction	33 (21.3)
Vertical or transfusion transmission	4 (2.6)
Year of HIV diagnosis	
Mean (<i>SD</i>), range	2000 (8), 1984–2013
Current fix dose combination regimen, <i>n</i> (%)	
Yes	58 (36)
No	103 (64)
Other drugs therapies (comorbidities), <i>n</i> (%)	
Yes	76 (47.8)
No	83 (51.6)
Total number of pills per day	
Mean (<i>SD</i>), range	2.6 (1.8), 1–10

reported in **Table 3**. As gender and educational level correlated with at least one of the study variables, they were included as control variables in the mediation model.

The initial mediation model did not show good fit indexes (**Figure 2**). Coefficients are shown in standardized form. Specifically, IPQ-R dimensions were not significantly associated with visit attendance, and visit attendance was not significantly associated with virologic success. Significant indirect effects were not detected. Since the mediation model did not fit the data, we tested a direct model between illness representations and virologic success and we proceeded by removing the variables which did not show significant relations, until a model with good fit indexes was obtained. The final path-analysis model is reported in **Figure 3** and presented good fit indexes (CFI = 1; TLI = 1; RMSEA = 0.00; and WRMSR = 0.309). Coefficients are shown in standardized form. Only two independent variables were significantly associated with virologic success: the consequences scale, as a negative predictor, and the emotional representations scale, as a positive predictor. In other words, the perception that

the disease does not have serious consequences on the patient's life and the prevalence of negative emotions were associated with virologic success.

DISCUSSION

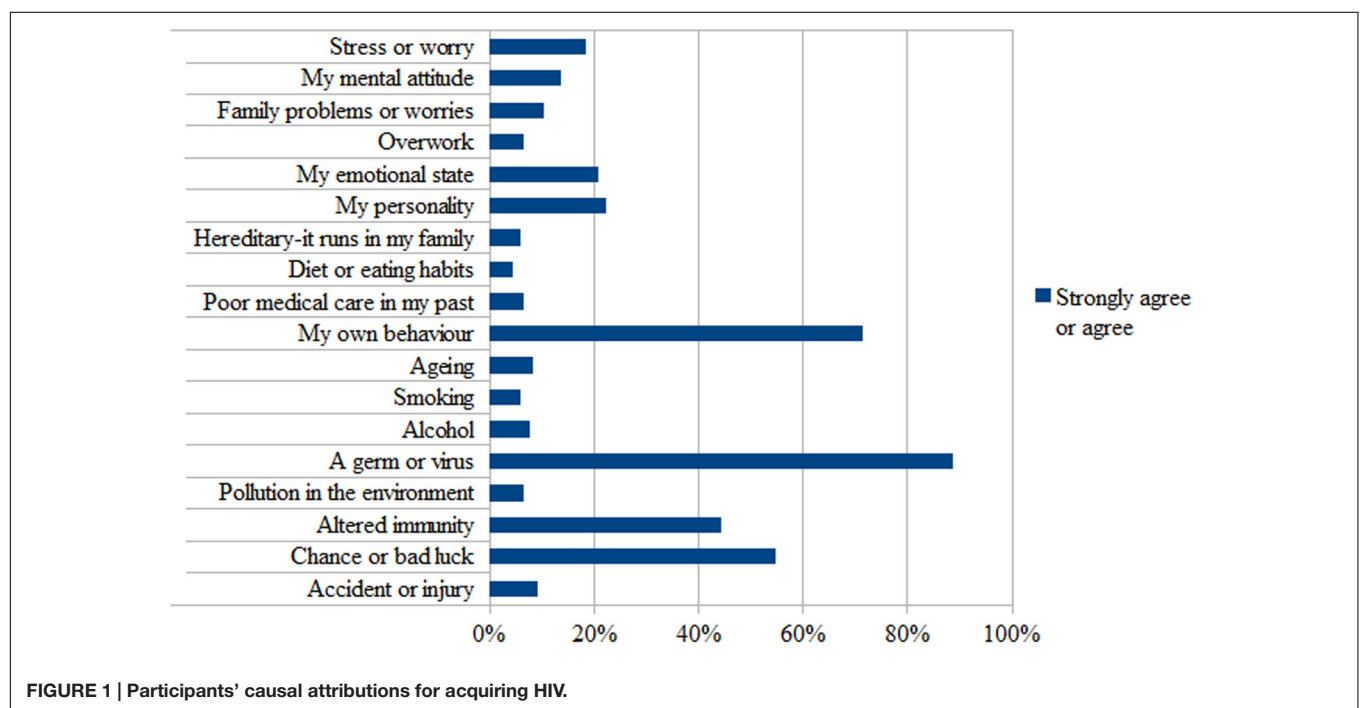
Literature on chronic disease and HIV showed that patients' illness representations affect illness experiences, management, and adherence (Moss-Morris et al., 1996; Petrie and Weinman, 1997; Spire et al., 2002; Cameron and Leventhal, 2003; Reynolds et al., 2009). The present study aimed to (1) explore illness representations of HIV positive patients in HAART and (2) test the hypothesis that illness representations predict adherence through the mediating role of a component of behavioral engagement.

For what concerns the first aim, results showed that most participants (87%) experienced various symptoms since HIV diagnosis and linked them to HIV and/or HAART coherently with their clinical condition. Overall, patients perceived HIV as a chronic and stable disease. They presented a good understanding of HIV, a general sense of personal control over the disease management and high trust in the treatment. Around 50% of patients did not perceive HIV as having serious consequences on their life and did not report negative related emotions. These results probably reflect the effectiveness of HAART treatment, which has transformed HIV from a terminal to a chronic disease, allowing patients to develop good disease control (Siegel and Lekas, 2002). With regard to the causal attribution for acquiring HIV, our findings highlighted that patients mainly attributed HIV to an "objective" cause (a germ or virus), consistently with a biomedical interpretation of their disease. The majority of patients seemed to be engaged in terms of their attendance to the check-up visits and to have a good adherence to HAART therapy.

As to the relationship between illness representations, behavioral engagement and adherence, our findings show that patients' attendance to the check-ups does not mediate the relationship between illness representations and virologic success. However, illness representations were found to be directly associated with virologic success. Our findings highlighted that virologic success is related to a specific combination of cognitive and emotional representations in HIV patients. Notably, patients who perceived HIV as not having serious consequences on their life but reported negative emotions connected to the disease were found to reach virologic success. On the contrary, patients who experienced HIV as having more serious consequences on their life, but reported positive emotions, were found to be less adherent, showing a high viral load. According to the theoretical model of patient health engagement (PHE) (Graffigna et al., 2014), which describes the patient engagement process as a transition across four phases, each encompassing emotional, cognitive and behavioral dimensions, our findings seem to describe patients who are positioned at different phases of their health journey. Patients who failed to reach virologic success are those who perceive HIV as having more serious consequences on their lives but, at the same time, present more positive emotions. These patients

TABLE 2 | Descriptive statistics of participants' illness representation (IPQ-R opinions).

	Mean (SD)	Sample range	IPQ-R opinions range and descriptions
Timeline	23.4 (5)	10–30	Range: 6–30 High: perception of chronic duration Low: perception of acute duration
Cyclical symptoms	9 (3.8)	4–20	Range: 4–20 High: perception of a cyclic disease Low: perception of a stable disease
Consequences	18.4 (3.8)	7–30	Range: 6–30 High: perception that the disease has serious consequences Low: perception that the disease has not serious consequences
Personal control	22.2 (4.2)	6–30	Range: 6–30 High: perception of high control in disease management Low: perception of low control in disease management
Treatment control	20.2 (2.9)	11–25	Range: 5–25 High: high trust in the disease treatment Low: low trust in the disease treatment
Coherence	19.1 (3.9)	5–25	Range: 5–25 High: high understanding of the disease Low: low understanding of the disease
Emotional representations	17.1 (5.5)	6–30	Range: 6–30 High: prevalence of negative emotions Low: prevalence of positive emotions



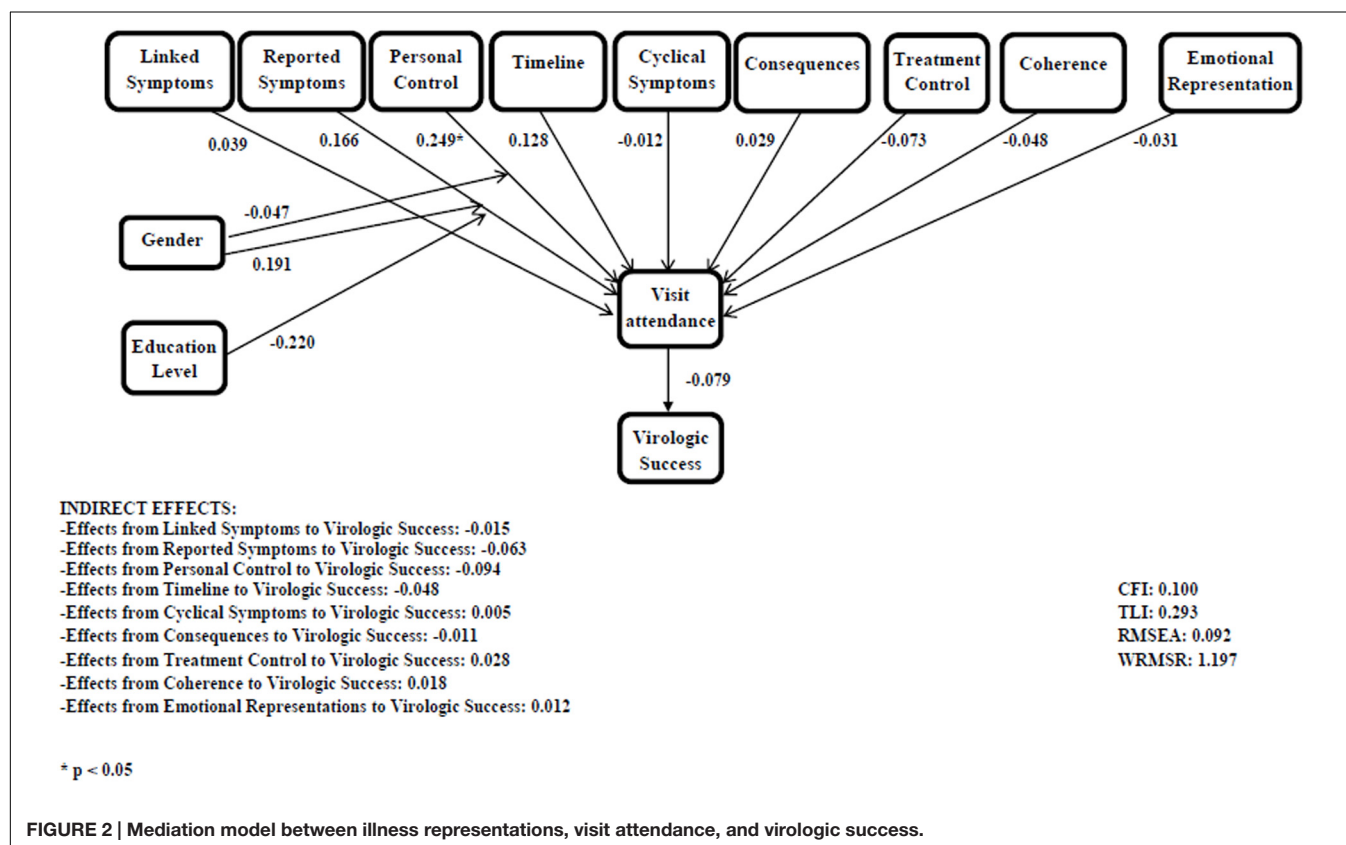
are probably in the earlier stages of the PHE model and we could hypothesize that, faced with serious illness consequences, they use maladaptive coping strategies, such as emotional detachment and denial, that lead them to refer a prevalence of positive emotions. These patients may not perceive the importance of taking medications for maintaining good health in the long term. These results are consistent with previous

studies on HIV infection and heart conditions, which found that patients perceiving their disease as having serious consequences reported higher viral load (Pala Norcini and Steca, 2015) and lower adherence to exercise therapy (Flora et al., 2015). On the contrary, patients who reached virologic success seem to be at an advanced stage of PHE process (such as adhesion and eudaimonic project) as they referred negative emotions related

TABLE 3 | Correlation matrix between IPQ-R dimensions (identity and opinions) and socio-demographic variables.

	Age ¹	Educational level ²	Gender ³	Relationship status ⁴	Year of HIV diagnosis ¹
Timeline	−0.029	0.068	0.052	0.044	−0.021
Cyclical symptoms	0.001	−0.143	0.088	0.156	0.096
Consequences	−0.109	−0.009	0.024	0.146	0.042
Personal control	−0.083	0.135	0.158*	0.168	−0.117
Treatment control	0.045	−0.035	−0.029	0.022	−0.033
Coherence	−0.096	0.071	−0.138	0.151	−0.013
Emotional representations	−0.091	−0.057	0.078	0.115	0.034
Identity reported symptoms	0.045	−0.231**	0.187*	0.093	0.131
Identity HIV/HAART associated symptoms	−0.004	−0.108	0.071	0.140	0.129

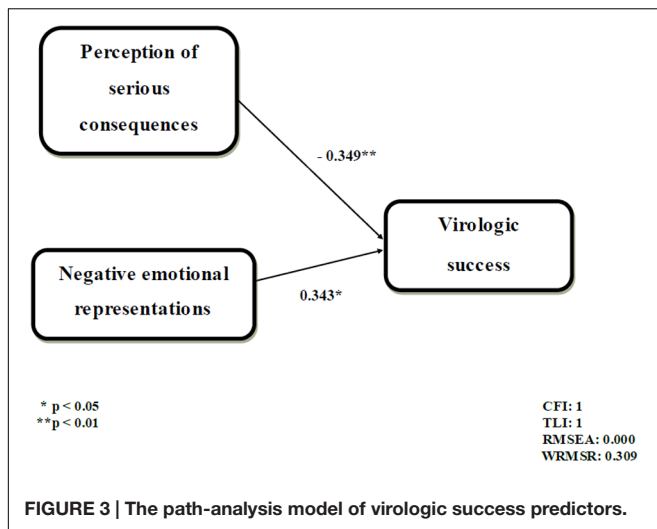
¹Pearson coefficient *r*; ²Spearman rho coefficient; ³Point-biserial coefficient; ⁴Eta coefficient. **p* < 0.05; ***p* < 0.01.

**FIGURE 2 | Mediation model between illness representations, visit attendance, and virologic success.**

to HIV instead of denying them and, at a cognitive level, they did not perceive HIV as having too many consequences on their lives. Patients in these stages may present a correct intake of the HAART therapy, and thus are more likely to reach virologic success.

The fact that negative emotions were found to be associated with virologic success appears to be in contradiction with previous literature which identified depressive symptoms as the most consistent predictor of treatment non-adherence in HIV (Paterson et al., 2000; Ickovics and Meade, 2002; Ammassari et al., 2004) and other diseases (DiMatteo et al., 2000). This discrepancy may be due to the conceptual difference between emotional representations and depression.

According to the CS-SRM, the emotional representations of illness reflect the patient's cognitive evaluation of the emotional impact of the illness (Moss-Morris et al., 2002). Emotional representations refer to a cognitive elaboration of the emotional impact of the disease and play an important role in the adaptation process. Emotional representations can indeed motivate the patient to develop an action plan, or can be so overwhelming, resulting in less or no action taken with respect to the disease (Diefenbach and Leventhal, 1996). On the contrary, according to the CS-SRM the depressive symptoms are considered an illness outcome and refer to a dysfunctional emotional reaction that is not necessarily processed by a cognitive elaboration. Future studies should explore the



mechanism by which emotional representations are translated into lived emotions, and should investigate the relationship between emotional representation and emotional outcomes (e.g., depression) in relation to adherence to HAART among HIV positive patients.

Interestingly, visit attendance did not mediate the relationship between illness representations and adherence. The lack of mediation may be differently discussed. It is possible that the use of visit attendance as an index of behavioral engagement may have reduced the complexity of the construct of patient engagement yielding non-significant results. It is also possible that attendance to the visits, as a form of problem-focused coping strategy, was not effective in order to reduce viral load among HIV patients. A recent meta-analysis (Dempster et al., 2015) highlighted that different coping strategies may work best for different conditions. It is possible that other problem-focused coping strategies should be considered or that emotional-focused strategies (e.g., acceptance/self-blame, avoidance, or social support) may be more effective to mediate or moderate the relationship between illness representations and virologic success in HIV positive patients.

Overall, our findings provided us with a more comprehensive picture of the relationships between illness representation, attendance to the visits and HAART adherence among HIV positive patients. Significant implications for theory and practice can be highlighted. At a theoretical level, our study has advanced research on adherence to HAART compared to previous literature which has mainly focused on socio-demographic, treatment-related or psychological characteristics without exploring the relationships between variables. As illness representations were not found to be associated with visit attendance, but were found to predict adherence, future research could explore the impact of other variables on visit attendance and virologic success, such as coping strategies. Self-blame or acceptance coping were found to mediate the effect of perceived consequences on outcomes among patients with irritable bowel syndrome. Patients who reported little consequences were more

likely to accept the illness, achieve a better quality of life and be more satisfied with their health (Rutter and Rutter, 2002). In addition, the role of perceived social stigma could be assessed in relationship to visit attendance and virologic success.

At a practical level, our results shed light on specific illness representations that lead to virologic failure in HIV positive patients. Perceived consequences and emotional representations are potentially modifiable factors that may be targeted in future interventions to enhance patient adherence to HAART therapy, as already done for other illness and outcomes (Petrie et al., 2002; Llewellyn et al., 2007; Phillips et al., 2012). As reported by Llewellyn et al. (2007) in a study on patients with head and neck cancer, illness representations could be targeted for intervention in the time period between diagnosis and shortly after treatment in order to maximize longitudinal outcomes. Psychological group interventions could be implemented for HIV patients presenting specific illness representations in order to help them cope with the disease and process their emotions, thus promoting illness adaptation and empowerment. Another practical implication concerns also clinician-patient communication. The importance of exploring patients' illness experience to promote adherence has been mentioned by several authors (Zolnieriek and DiMatteo, 2009) and has been embraced by a patient-centered model of care (Borghi et al., 2016). During clinical encounters, clinicians should pay particular attention to their patients' illness representations regarding HIV as a deeper understanding of the patients' perspective could promote patient adherence (Borghi et al., 2016).

Our study has several limitations that are principally due to the convenience sample recruited, which presented a high prevalence of adherent patients as well as the small number of patients. Therefore, the generalizability of the findings is limited. Moreover, as the study is cross-sectional, the relationship between illness perception, attendance to visits and viral load have to be interpreted with caution especially for what concerns the direction of causality. Finally, our study focused on illness representations and visit attendance, but did not investigate the role of other variables in affecting adherence in HIV patients.

CONCLUSION

Our findings revealed a perception of good personal control in disease management, high trust in the therapy and good adherence to treatment. Since patients' perceived consequences and emotions influence virologic success, clinicians should explore them in order to promote adherence to treatment.

ETHICS STATEMENT

The research protocol was approved by the San Paolo Hospital Ethics Committee. Patients who accepted to participate in the study signed an informed consent.

AUTHOR CONTRIBUTIONS

DL, EV, TB, and AdM contributed to the conception and design of the work; DL contributed to the acquisition of data; LiB and LuB contributed to the data analysis; all authors contributed to the interpretation of data. DL, LiB, GL, and LuB contributed to the draft of the work; EV, TB, and AdM revised the work critically. All authors gave their final approval of the version to be published and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity

of any part of the work are appropriately investigated and resolved.

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Profiles of Recovery from Mood and Anxiety Disorders: A Person-Centered Exploration of People's Engagement in Self-Management

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Context: A shift toward person-centered care has been occurring in services provided to people with mood and anxiety disorders. Recovery is recognized as encompassing personal aspects in addition to clinical ones. Guidelines now recommend supporting people's engagement in self-management as a complementary recovery avenue. Yet the literature lacks evidence on how individualized combinations of self-management strategies used by people relate to their clinical and personal recovery indicators.

Objectives: The aims of this study were to identify profiles underlying mental health recovery, describe the characteristics of participants corresponding to each profile, and examine the associations of profiles with criterion variables.

Method: 149 people recovering from anxiety, depressive, or bipolar disorders completed questionnaires on self-management, clinical recovery (symptom severity), personal recovery (positive mental health), and criterion variables (personal goal appraisal, social participation, self-care abilities, coping).

Results: Latent profile analysis (LPA) revealed three profiles. The *Floundering* profile included participants who rarely used self-management strategies and had moderately severe symptoms and the lowest positive mental health. The *Flourishing* profile was characterized by frequent use of self-empowerment strategies, the least severe symptoms, and the highest positive mental health. Participants in the *Struggling* profile engaged actively in several self-management strategies focused on symptom reduction and healthy lifestyle. They concomitantly reported high symptom severity and moderately high positive mental health. The study revealed that *Floundering* was associated with higher probabilities of being a man, being single, and having a low income. People in the *Flourishing* profile had the most favorable scores on criterion variables, supporting the profiles' construct validity.

Discussion: The mixed portrait of *Struggling* participants on recovery indicators suggests the relationship between health engagement and recovery is more intricate than anticipated. Practitioners should strive for a holistic understanding of their clients' self-management strategies and recovery indicators to provide support personalized to their profile. While people presenting risk factors would benefit from person-centered support, societal efforts are needed in the long term to reduce global health inequalities. The integration of constructs from diverse fields (patient-centered care, chronic illness, positive psychology) and the use of person-oriented analysis yielded new insights into people's engagement in their health and well-being.

Keywords: self-management, recovery, mood and anxiety disorders, person-centered approach, health engagement, positive mental health

INTRODUCTION

Contemporary mental health services are more person-centered¹ than they used to be (Mechanic, 2007). Mental health providers increasingly seek to support people's engagement in their idiosyncratic recovery process rather than prescribing a rigid treatment plan (Corrigan, 2015). As an overarching philosophy behind person-centered care (Storm and Edwards, 2013), the notion of recovery orients the services offered to people living with mental disorders in several countries, such as the US (President's New Freedom Commission on Mental Health, 2003), England (National Institute for Mental Health in England, 2005), New Zealand (Mental Health Commission, 2012), and Canada (Mental Health Commission of Canada, 2009). From a clinical approach, recovery refers to the reduction of symptoms below the clinical threshold (e.g., Frank et al., 1991). In contrast, in a person-centered approach, recovery refers to "a movement toward health and meaning rather than avoidance of symptoms" (Clarke et al., 2012, p. 303). Self-management (i.e., daily actions a person takes to manage symptoms and well-being) has been proposed as a crucial pathway to recovery from mental disorders (Slade, 2009). Building on people's engagement in their own well-being and health (Graffigna et al., 2014), supporting self-management appears to be an exemplary person-centered practice. However, the notion of self-management mainly derives from the chronic disease literature (Lorig and Holman, 2003; Sterling et al., 2010), and its application in mental health recovery research is still limited (see Mueser et al., 2002, for a review in the mental health field). The aim of this study was to examine recovery from mood and anxiety disorders by focusing on the person and his/her active role. The present study constitutes a first exploration of individual profiles underlying mental health recovery. It highlights different combinations of self-management strategies used by people in relation to recovery

indicators. To this end, innovative person-oriented analyses were conducted to discern how self-management and recovery are related at the person level, in contrast to traditional variable-oriented analyses that consider relationships between variables across whole groups of participants (Meyer et al., 2013).

Recovery from Mood and Anxiety Disorders

Mood and anxiety disorders are among the most prevalent mental disorders in the world (Kessler et al., 2005, 2007). In the US, lifetime prevalence has recently been estimated at 17.5% for any mood disorder (major depressive and bipolar disorders) and 31.6% for any anxiety disorder (panic, generalized anxiety, agoraphobia, social phobia, specific phobia, separation anxiety, post-traumatic stress, obsessive-compulsive disorders) (Kessler et al., 2012). In Canada, an estimated 11.6% (point prevalence) of the adult population reported having a mood or anxiety disorder (Public Health Agency of Canada, 2015). Mood and anxiety disorders are often recurrent. The estimated cumulative recurrence rate for major depressive disorder has been estimated at 42.0% at 20 years after remission (Hardeveld et al., 2013). Indicative of chronicity, in a study of people living with anxiety disorders, the average time spent in an illness episode represented over 70% of the 12-year study course (Bruce et al., 2005). Mood and anxiety disorders are also highly comorbid. For example, a study with a large nationally representative sample in the Netherlands estimated (12-month prevalence) that 54.3% of people with a mood disorder also had an anxiety disorder, and 33.4% of those with an anxiety disorder also had a mood disorder (de Graaf et al., 2002). Given the comorbidity and similitudes between these disorders, "it is sensible to consider them as a single group," as argued by the International Society for Affective Disorders², the leading international scientific society in that field.

Mental health recovery from mood and anxiety disorders has usually been defined using a *clinical* approach, i.e., as a reduction of clinical symptoms to below a threshold for a certain period of time, following Frank et al.'s (1991) definition (see review from Fava et al., 2007). However, this pathogenic approach is

¹As described by Davidson et al. (2015), person-centered services in the mental health field emerged from the patient-centered model of care in the medical domain, for which the 2001 Institute of Medicine Report made a strong case. The expression "person-centered" is preferred to "patient-centered" throughout this article, as it is more consistent with the aim of these services, i.e., recognizing the person and his/her active role beyond the "patient" status (Davidson et al., 2015). For the same reason, the word "person" (or "client") is preferred to "patient" in the paper.

²See <https://www.isad.org.uk>; see also the journal of the association, the Journal of Affective Disorders.

now being deemed too limited in comparison with how mental health consumers themselves define recovery (Zimmerman et al., 2006; Johnson et al., 2009; McEvoy et al., 2012). From their perspective, recovery is better defined as “a deeply personal, unique process of changing one’s attitudes, values, feelings, goals, skills, and/or roles. It is a way of living a satisfying, hopeful, and contributing life even with limitations caused by illness” (Anthony, 1993, p. 527). This *personal* approach to recovery is concordant with the recent field of positive psychology that aims to cultivate human strengths, well-being, and dimensions that make life worth living (Seligman and Csikszentmihalyi, 2000; see Provencher and Keyes, 2010, 2011, 2013).

Personal and clinical approaches to recovery have mainly been examined in distinct streams of research. However, Whitley and Drake (2010) recently proposed a theoretical conceptualization of recovery that encompasses both clinical and personal aspects. Their model postulates five recovery dimensions: clinical (e.g., reduction and control of symptoms), existential (e.g., emotional and spiritual well-being), functional (e.g., employment and education), physical (e.g., diet and exercise), and social (e.g., social support and community integration). Although Whitley and Drake (2010) suggest a list of several measurable outcomes that could be used to explore these dimensions of recovery, to our knowledge their comprehensive assessment has yet to be fully operationalized.

Provencher and Keyes (2010, 2011, 2013) also proposed a comprehensive model: the Complete Mental Health Recovery model. Based on this model, recovery should be assessed on two indicators. The first is the experience of restoration from mental illness symptoms; the second is the experience of optimization of positive mental health. The first indicator mostly pertains to the clinical recovery approach, while the second mostly relates to themes from the personal recovery approach (Slade, 2010). Positive mental health is defined as a syndrome composed of several manifestations of well-being (Keyes, 2002), at the emotional (e.g., interest, satisfaction), psychological (e.g., purpose in life, personal growth), and social levels (e.g., social contribution, social integration). Provencher and Keyes’ model is based on several psychometric studies using large non-clinical samples showing mental illness and positive mental health to be two coexistent dimensions, and not merely the two ends of a single dimension (Keyes and Lopez, 2002; Keyes, 2005; Keyes et al., 2008; Westerhof and Keyes, 2010).

Formed by the intersection of these two dimensions, Provencher and Keyes (2010, 2011, 2013) model proposes different states of recovery. In partly recovered states, the person shows low symptoms³ concomitantly with low positive mental health (state labeled as *languishing* by Keyes and Lopez, 2002)

or high symptoms and high positive mental health (labeled as *struggling* with life). In the completely recovered state, the person shows both low symptoms and high positive mental health (labeled as *flourishing*). In the opposite state, the person is non-recovered on both aspects (labeled as *floundering*). The model also proposes two more states, in which people have a moderate level of positive mental health but are either recovered or not from their symptoms. According to their situation in terms of recovery indicators (symptom severity and positive mental health), individuals are expected to fall into one of these states. However, this classification has never been explored in clinical samples of people with mood and anxiety disorders.

Self-Management in Mental Health Recovery

Exploring recovery from a person-centered perspective necessitates considering what people actually do in their pathway toward recovery. Self-management refers to actions people implement day-to-day to manage their symptoms, prevent recurrence, and optimize well-being (Lorig and Holman, 2003). Self-management harnesses people’s sense of agency, responsibility, empowerment, and motivation to get better (Barlow et al., 2005; Slade, 2009). Self-management support is now recommended in clinical guidelines for mood and anxiety disorders (Swinson et al., 2006; Patten et al., 2009; National Institute for Health and Care Excellence, 2014). Supporting self-management is intended to complement, not to replace, standard psychological, and pharmacological treatments (Fournier et al., 2012). It is a useful approach to complement such evidence-based treatments, which, although efficient, are limited by the fact that not all people respond positively to antidepressants or psychotherapy (Bystritsky, 2006; Lanouette and Stein, 2010; Berlim et al., 2015), and that several of them relapse (Boland and Keller, 2009; Boschen et al., 2009) or must deal with incapacitating residual symptoms (Fava et al., 2007; Kaya et al., 2007).

The value of self-management for coping with physical chronic illness such as diabetes and asthma has been well established (Barlow et al., 2002). This is in line with a prolific stream of theoretical and empirical work in medicine on the broader concepts of engagement and active involvement in one’s own health and care (see review from Menichetti et al., 2014). While similar to self-management, health engagement has recently been proposed as an umbrella term (Graffigna et al., 2015b) representing a multidimensional process that includes not only behaviors (Gruman et al., 2010) but also the person’s cognitions and emotions regarding his/her health (Graffigna et al., 2014). These dimensions can be considered at different levels of the person’s systemic context (e.g., individual, organizational, societal; Carman et al., 2013).

In contrast, self-management is more specific, as it focuses on strategies (behaviors) that the person enacts, considered as one positive outcome of the engagement process (see review from Graffigna et al., 2015b), while patient activation focuses on the knowledge, skills, and confidence for performing such

³In addition to a measure of symptom severity, Provencher and Keyes (2010, 2011, 2013) suggest that a measure of functional impairment should also be included to assess recovery from mental illness, based on the usual practice for schizophrenia. However, in the case of mood and anxiety disorders, the focus of the present article, inclusion of such a measure is not mandatory (e.g., Goldberg et al., 2007; Radhakrishnan et al., 2013). Furthermore, a task force (Rush et al., 2006) has specifically recommended that such a measure should not be included when evaluating clinical recovery, as observed functional impairment may be unrelated to the mental illness under consideration. For this reason, the level of functional impairment is not taken into account as a recovery indicator in the present article.

strategies (Hibbard and Mahoney, 2010). Notions of engagement and activation have received only limited attention in the mental health field or in psychology (Kukla et al., 2013; Menichetti et al., 2014; Sacks et al., 2014; Moljord et al., 2015). Similarly, research and interventions on self-management are less frequent in the context of mental illness than in medicine (Cook et al., 2009; Lorig et al., 2014). In the present article, while the center of attention is self-management, the findings also have the potential to contribute to the incipient knowledge base on the application of these related concepts to the field of mental health.

Self-management strategies implemented by people with mood and anxiety disorders have rarely been studied, with the exception of a few recent qualitative studies (Murray et al., 2011; van Grieken et al., 2014, 2015; Chambers et al., 2015; Villaggi et al., 2015). Participants have reported a wide variety of strategies focused on reducing and preventing symptoms (e.g., mood monitoring, obtaining mental health services), as well as other strategies to promote positive mental health (e.g., meditating, socializing). In the study from Villaggi et al. (2015), participants with depressive, bipolar, and anxiety disorders reported overall similar strategies, suggesting that a transdiagnostic approach to self-management is appropriate.

Based on this qualitative study (Villaggi et al., 2015), our research team developed the Mental Health Self-management Questionnaire (MHSQ), the first instrument to provide a quantitative indicator of the frequency with which people use a diversity of strategies (Coulombe et al., 2015). The validation study revealed three distinct types of self-management strategies: (a) clinical (getting help and using resources, e.g., taking medication, consulting a professional); (b) empowerment (building upon strengths and positive self-concept to gain control, e.g., acknowledging one's successes, arranging one's schedule around one's capabilities); and (c) vitality (having an active and healthy lifestyle, e.g., practicing sports, maintaining healthy eating habits).

In our cross-sectional validation study of the MHSQ (Coulombe et al., 2015), positive mental health was associated positively with empowerment and vitality strategies but unrelated to clinical ones. Depressive and anxiety symptom severity indicators were found to be negatively related to empowerment and vitality strategies. However, symptom severity was positively related to clinical self-management. This was interpreted as suggesting that participants with more severe symptoms may have focused on using clinical strategies, given their acute needs in that regard. Indeed, people with severe symptoms have been shown to be more likely to use health services (Hämäläinen et al., 2008), one of the so-called clinical strategies. In contrast, people with less severe symptoms may have been more likely to use empowerment and vitality strategies, since they probably had reached a different state of recovery and now faced the task of increasing their positive mental health (Provencher and Keyes, 2011). These interpretative hypotheses illustrate the need for further studies to disentangle the complex relationships between self-management and recovery indicators.

The Value of Person-Oriented Statistical Analysis

As people have been shown to use their personal “recipe” of self-management strategies (Chambers et al., 2015; Villaggi et al., 2015), it is important to go beyond the group level when exploring self-management and recovery. Given the variety of self-management strategies and possible situations in terms of recovery indicators (i.e., forming six different states according to Provencher and Keyes, 2010, 2011, 2013), it is pertinent to ask how these all vary together, and whether, across individuals, there are diverse profiles of interrelationships among these variables. Exploring such profiles quantitatively calls for person-oriented analyses. In contrast to the variable-oriented approach (e.g., correlational analysis), person-oriented analysis [e.g., cluster analysis, latent profile analysis (LPA)] allows variables to be related differently across the people in the sample (Meyer et al., 2013). The individual is seen as a system of variables that “can combine in various ways that have implications for how they are experienced and relate to other variables of interest” (Meyer et al., 2013, p. 191). Person-oriented analyses are intended to provide a holistic perspective, offering a richer source of information for person-centered services (Cloninger, 2013). One person-oriented analysis that is gaining in popularity is LPA, which provides a way to uncover unobserved (i.e., latent) profiles of participants showing distinctive patterns of interaction among continuous variables. In LPA, the number of profiles is selected based on the estimation and comparison of statistical models, allowing for more objectivity than other procedures, such as cluster analysis (DiStefano and Kamphaus, 2006; Pastor et al., 2007; Morin et al., 2011b).

Once these profiles underlying mental health recovery are identified, it is possible to explore the background characteristics (on clinical and sociodemographic variables) associated with each profile. Notably, although evidence is still scarce, people might show different profiles depending on their diagnoses. A recent study (Vermeulen-Smit et al., 2015) suggests that having a depressive or anxiety disorder is associated with lower probability of endorsing a healthy lifestyle (i.e., vitality self-management strategies), while this is not the case with bipolar disorder. Treatments currently in progress are another factor to consider. People with more severe symptoms could be more likely to receive mental health services (Hämäläinen et al., 2008) and concomitantly to display a profile characterized by the use of self-management strategies focused on symptoms (Coulombe et al., 2015).

Sociodemographic variables are also important. Recovery- and person-centered policies and research emphasize the importance of holistic approaches that take into account social determinants of health, such as gender, income, and marital status (Jayadevappa and Chhatre, 2011; Weisser et al., 2011; Commonwealth of Australia, 2013; Cloninger et al., 2014). Nevertheless, in their review, Weisser et al. (2011) concluded that recovery has mainly been studied as “an individual journey,” so the existent literature “falls short on an analysis of the role of

gender and other social and structural inequities in mental health problems” (p. 6). For instance, because of their endorsement of traditional masculinity norms, men would probably use fewer self-management strategies, such as seeking professional help (Möller-Leimkühler, 2002). Also, being married is associated with increased adherence to health recommendations, possibly because of the social support offered by a life partner (e.g., Trivedi et al., 2008). Finally, being from a low-income background is associated with less health engagement (Greene and Hibbard, 2012), as there are economic barriers to self-management (Henderson et al., 2014). Despite the formative evidence, background factors have never been examined specifically in relation to self-management and to clinical and personal recovery.

As stated by Morin et al. (2011b, p. 61), “the advantages of LPA do not offset the need to assess the construct validity of the classification.” Profiles are considered valid to the extent that their pattern of association with criterion variables is consistent with theoretical expectations (Bauer and Curran, 2004; Morin et al., 2011b). Thus, the associations of recovery profiles with meaningful criterion variables need to be examined. In the present study, four were selected: personal goal appraisal, social participation, self-care abilities, and coping.

Personal goals constitute a pervasive theme in the recovery literature (Andresen et al., 2003). Empirical research has consistently related mental health indicators to positive appraisal of one’s personal goals, in terms, for example, of how important they are or how effective one is at achieving them (Little, 2007). Negative goal appraisal has been related to depression, anxiety, and hypomania (Lecci et al., 1994; Meyer et al., 2004; Dickson et al., 2011). Getting and seizing opportunities for *social participation* have also been highlighted as important components in recovery (e.g., Noordsy et al., 2002; Onken et al., 2007; see Provencher and Keyes, 2011). For people in recovery, regaining some of their previous social roles and engaging in new ones can give meaning to their life (Mezzina et al., 2006). *Self-care abilities* refer to people’s knowledge and competence concerning activities they need to perform for their health (Britz and Dunn, 2010; Seed and Torkelson, 2012). These are foundational skills for effective self-management. Similarly, the way people cope with illness has been related to psychological adjustment (Roesch and Weiner, 2001). In this context, *coping* refers to people’s adaptive (e.g., planning, seeking support) and maladaptive (e.g., denial, substance use) efforts to deal with the stress associated with their disorder (Meyer, 2001; Roesch and Weiner, 2001).

Self-management and recovery indicators, being comprehensive variables, were chosen as the key parameters driving the profile exploration in the present study. In contrast, personal goal appraisal, social participation, self-care abilities, and coping are more specific notions. These are nevertheless interesting to consider as criterion variables, given their importance in recovery theories and findings. However, because the profiles have never been explored before, their precise nature is still unknown; thus it would be premature to propose specific hypotheses concerning their associations with the criterion variables (Morin et al., 2011b).

Objectives

The aim of this study was to explore person-centered recovery profiles presented by individuals who reported having received a diagnosis of mood and anxiety disorders. The first objective was to identify and draw the general portrait of the distinct profiles concerning individuals’ use of self-management strategies (clinical, empowerment, and vitality) and scores on recovery indicators (symptom severity and positive mental health). The second objective was to describe the profiles by exploring their associations with (a) the frequency of use of specific self-management strategies and (b) background characteristics (clinical and sociodemographic variables). The third objective was to verify the construct validity of the profiles by examining their pattern of association with criterion variables.

MATERIALS AND METHODS

Procedure

The present study was part of a larger research project to validate the (MHSQ, see Section Self-Management). Validation results have been published elsewhere (Coulombe et al., 2015). Using data from that study, the present paper is distinct by virtue of its different analytical strategy (person-oriented analysis) and its consideration of an array of variables (e.g., gender, low income, personal goals, etc.) that were not treated in the validation article. The study was approved by the institutional research ethics board for research involving human participants at Université du Québec à Montréal, Canada.

Recruitment

Thirteen community organizations in Quebec (Canada) and France were asked to send an email invitation to members of their mailing list and to advertise the study on their website. An invitation was also published in a Montreal (Canada) free newspaper. The invitation included a URL link for participants to complete the study online. After reading and consenting to an online consent form, participants answered self-reported preliminary questions to verify their eligibility. Participants had to be at least 18 years old; understand written French; have received a diagnosis of anxiety, depressive and/or bipolar disorder(s) at least 1 year prior to responding; and be in treatment or have been treated (with psychotherapy and/or pharmacotherapy) for the disorder(s). The “time since diagnosis” criterion was intended to ensure the person had had sufficient time to implement self-management strategies. Pregnant women or those who had given birth in the previous year were excluded, given that the recovery process is different in these situations (Hendrick et al., 2000). To prevent symptom exacerbation due to filling out the questionnaire, people scoring high on symptom measures (see Section Recovery Indicators) were excluded and presented with a list of available help resources. The same list was presented to all participants after questionnaire completion. The questionnaire was filled out on a secured online survey platform.

Participants

The final sample was composed of 149 participants. The detailed sample description has been published in the MHSQ validation

paper (Coulombe et al., 2015). The majority of participants reported having been diagnosed with a depressive disorder (55.7%), while self-reported anxiety (36.9%), and bipolar (36.2%) disorders were less prevalent. In terms of comorbidity, around one-quarter (26.8%) reported having been diagnosed with more than one of these disorders. Based on the scores on the depression severity measure (see Section Recovery Indicators; Kroenke and Spitzer, 2002), at the time of the study, 34.2% of the participants reported moderate symptoms, 30.2% mild symptoms, and 35.6% less than mild symptoms. Based on the scores on the anxiety severity measure (Spitzer et al., 2006), 26.2% reported moderate symptoms, 26.8% mild symptoms, and 47.0% less than mild symptoms. The vast majority reported they had been undergoing pharmacotherapy (85.2%) in the past month, and less than half of the sample was currently undergoing psychotherapy (40.3%). Participants were mostly female (80.1%) and were on average 41.5 years old ($SD = 12.2$; from 18 to 71). Most reported being from Canada or having immigrated there (91.9%). The sample was very educated: 60.4% had a university degree, which is much higher than the Canadian figure (30.8%, Statistics Canada, 2013). The remaining participants either had a vocational (9.0%) or a college (pre-university) degree (21.5%), or a high school diploma or less (9.0%). About half the participants were married or had a life partner (47.9%) while the other half (52.1%) were single. As explained below (Section Background Characteristics), low-income status was calculated only for those from Canada; nearly one-quarter (23.0%) were living under the low-income threshold (Statistics Canada, 2015).

Measures

The questionnaire included the validated French version of the following instruments.

Self-Management

Self-management was measured using the MHSQ developed as part of the larger study (see **Table 5** for the complete item list). Items were created on the basis of qualitative interviews (Villaggi et al., 2015), and a multidisciplinary expert team helped reduce the number of items and improve wording. As reported in the validation paper (Coulombe et al., 2015), exploratory and confirmatory analyses of data collected from the present sample indicated the presence of three distinct subscales: (a) clinical (5 items, e.g., *I look for available resources to help me with my difficulties (websites, organizations, healthcare professionals, books, etc.)*; *I participate in a support or help group to help me manage my difficulties*); (b) empowerment (9 items, e.g., *I take my capabilities into account when arranging my schedule*; *I congratulate myself for my successes, large and small*); and (c) vitality (4 items, e.g., *I do activities I like to maintain an active lifestyle*; *I engage in sport, physical activity*). For each item, participants were asked to indicate to what extent they had used the strategy during the two previous months, on a scale from 0 (*Never*) to 4 (*Very often*). Each subscale had adequate internal consistency: $\alpha = 0.69$ for clinical, $\alpha = 0.81$ for empowerment, and $\alpha = 0.75$ for vitality.

Recovery Indicators

Three recovery indicators were included, two measuring recovery from the clinical perspective (symptom severity) and one from the personal perspective (positive mental health).

The Patient Health Questionnaire 9 (PHQ-9; Kroenke and Spitzer, 2002) was used to assess severity of depressive symptoms. The PHQ-9 requires participants to rate to what extent they had experienced nine symptoms (e.g., *little interest or pleasure in doing things*) during the two previous weeks, on a 4-point frequency scale: 0 (*Not at all*), 1 (*Several days*), 2 (*More than half the days*), and 3 (*Nearly every day*). The Generalized Anxiety Disorder 7 (GAD-7; Spitzer et al., 2006) was used to assess severity of anxiety symptoms (e.g., *feeling nervous, anxious, or on edge*) on the same response scale. According to a systematic review (Kroenke et al., 2010), the PHQ-9 and GAD-7 have adequate sensitivity and specificity for detecting symptoms of depressive and anxiety disorders and monitoring their severity. Both scales had adequate internal consistency in the current study: $\alpha = 0.85$ for PHQ-9 and $\alpha = 0.86$ for GAD-7. Sums of scores for each scale were used as recovery indicators in the analyses, but also to verify eligibility, with participants presenting severe symptoms (PHQ-9 ≥ 20 ; GAD-7 ≥ 15 ; Kroenke and Spitzer, 2002; Spitzer et al., 2006) being excluded, as explained above. The Altman Self-Rating Mania Scale (ASRMS, Altman et al., 1997) was used to exclude participants who were in current mania (ASRMS ≥ 6 ; Altman et al., 1997).

The Mental Health Continuum–Short Form (MHC-SF; Keyes, 2002; Lamers et al., 2011; Salama-Younes, 2011) was used to assess the degree of participants' positive mental health in terms of their experience of 14 well-being manifestations, related to positive emotions (e.g., *feel satisfied with your life*), psychological functioning (e.g., *feel that your life has a sense of direction or meaning to it*), and social functioning (e.g., *feel that you belonged to a community*) during the past month. Participants were required to answer on a 6-point frequency scale: 0 (*Never*), 1 (*Once or twice*), 2 (*About once a week*), 3 (*About two or three times a week*), 4 (*Almost every day*), and 5 (*Every day*). The MHC-SF has been shown to be valid and reliable in diverse samples (Keyes et al., 2008; Lamers et al., 2011) and has been successfully used in national surveys (e.g., Canadian Community Health Survey—Mental Health, Statistics Canada, 2012). Results from several studies with large samples (>1000) across the world show that MHC-SF scores are not simply the inverse of mental illness symptom indicators, as they measure two distinct factors that correlate negatively but only moderately (Keyes et al., 2008; Lamers et al., 2011; Petrillo et al., 2015). Internal consistency was satisfactory ($\alpha = 0.92$) in the present study.

Background Characteristics

In terms of clinical variables, diagnosis (depressive, anxiety and/or bipolar disorders) and ongoing treatments (undergoing psychotherapy and/or pharmacotherapy) were self-reported as part of the eligibility questions. The questionnaire also included a sociodemographic form including age, gender, education level, marital status, number of people in the household, and household income. Using the last two variables, each participant's status as living or not in a low-income household was determined based

on the national cut-off depending on household size (Statistics Canada, 2015). For comparability purposes, only participants from Canada, who made up the vast majority of the sample, were included in the analysis pertaining to low income.

Criterion Variables

Assessment of participants' personal goal appraisal was based on the Personal Project System Rating Scale (PSRS; Little, 1988; Pychyl and Little, 1998; Chambers, 2007), which was translated into French and adapted for the purposes of the present study. Participants were asked to appraise their goal system (presented as their current goals, activities, commitments, and projects considered on the whole) on a scale from 1 (*Not significant for me*) to 10 (*Very significant for me*) along six dimensions: meaningfulness, manageability, progress, support, stress (reversed), and enjoyment. Cronbach's alpha of the overall scale was satisfactory ($\alpha = 0.82$).

Social participation was measured with the Social Participation Scale (Richard et al., 2009). The scale assessed to what extent participants had taken part in 10 social activities (e.g., visiting friends or family, shopping, volunteering) in the previous 6 months, on a 5-point frequency scale: 0 (*Never*), 1 (*Less than once a month*), 2 (*At least once a month*), 3 (*At least once a week*), and 4 (*Almost every day*). Internal consistency was satisfactory ($\alpha = 0.70$).

The Therapeutic Self-Care Scale (Doran et al., 2002; Paradis, 2009) was used to assess participants' perceived self-care abilities. The 12 items were developed for patients living with physical illness, but are also pertinent in a mental health context. The scale measures knowledge and competence with regard to management of the disorder, such as understanding what needs to be done to address one's symptoms, being able to take one's medication (if applicable), etc. Answers were given on a 6-point Likert scale, from 0 (*Not at all*) to 5 (*Completely*). Cronbach's alpha was high ($\alpha = 0.86$).

Use of coping strategies was measured with the Brief COPE (Carver, 1997; Muller and Spitz, 2003), in which participants indicated to what extent they had used 28 strategies to deal with the stress associated with their mental health problem, on a 4-point scale: 0 (*Not at all*), 1 (*A little bit*), 2 (*Moderately*), and 3 (*A lot*). Instead of using the instrument's 14 original subscales, four coping subscales were created to reduce the number of variables in the analysis, following the procedure used by Desbiens and Fillion (2007): emotional (venting and emotional support; $\alpha = 0.80$), behavioral (active coping, planning, and instrumental support; $\alpha = 0.85$), cognitive (acceptance, positive reframing, humor, and religion; $\alpha = 0.77$), and avoidance (substance use, denial, behavioral disengagement, and self-distraction; $\alpha = 0.67$).

Analysis

As a preliminary analysis, bivariate correlations were examined between the main study variables. To achieve the first objective, LPA was then performed using the Robust Maximum Likelihood estimator (MLR) available in the Mplus software (Muthén and Muthén, 1998–2010) to identify latent profiles of participants (Morin et al., 2011b; Morin, 2016), based on participants' scores on the three subscales of self-management strategies and on the

three recovery indicators. To ensure the analysis did not converge on a local solution, the estimation process aimed to replicate the solution, using 3000 sets of random starts and 100 iterations, and retaining the 100 best sets of starting values for final stage optimization⁴, following Morin's recommendation (2016). Models with increasing numbers of profiles were compared using a variety of statistical criteria. Lower values of the Akaike Information Criterion (AIC), Consistent AIC (CAIC), Bayesian Information Criterion (BIC) and sample-size adjusted BIC (SSA-BIC) indicated better fit. Tests comparing each model with the model having one less profile (Vuong-Lo-Mendell-Rubin Likelihood Ratio, VLMR; Adjusted Lo-Mendell-Rubin Likelihood Ratio, ALMR; and Bootstrap Likelihood Ratio Test, BLRT) were also considered: significant *p*-values for these tests indicated that the model with more profiles was more adequate. Models including profiles in which fewer than 5% of the participants are classified should be rejected (Hamza and Willoughby, 2013). Finally, although entropy (which varies from 0 to 1) cannot be used to identify the optimal number of latent profiles in the data, it provides useful information regarding the accuracy of the participants' classification into the various latent profiles, with higher levels being indicative of less classification error (Tein et al., 2013; Morin, 2016).

Once the number of profiles was selected, each profile's standardized means on the self-management subscales and recovery indicators were graphed and compared with the overall sample mean. The profiles were also compared to one another on these variables, by re-running the LPA in Mplus, but adding an auxiliary command named "auxiliary (e)," which provides equality of means tests across profiles. Introduction of variables using such a command does not have an impact on the nature of the profiles (Morin et al., 2011b). Using an auxiliary command has recently been presented as one of the best ways of studying the association between variables and latent profiles (Asparouhov and Muthén, 2014b; Feingold et al., 2014). It recognizes classification uncertainty, and thus each participant is correctly considered as having a degree of probability of being a member of every profile (Bolck et al., 2004; Morin, 2016). For pragmatic purposes, as an additional analysis that could facilitate interpretation for practitioners, we performed an analysis in which participants were classified into only one of the profiles based on their *Most Likely Latent Profile Membership*. Each self-management subscale and recovery indicator was dichotomized into high and low scores, using the documented clinical cut-off when available (for symptom severity) or, when not available, by splitting the variable at the nearest score above the overall mean. The distributions of high (vs. low) scores were then compared across profiles with chi-square using SPSS software. Despite the fact that this involves a certain loss of information compared to the auxiliary command, this supplementary analysis is particularly informative for transposing our results to clinical

⁴We freely estimated means in all profiles. We also tested models in which variance was freely estimated (Morin et al., 2011a). However, these models were not retained given that their solutions failed to be sufficiently replicated or that they converged on improper solutions (negative variance). These problems suggest that more parsimonious models (in which variance is constrained to be equal across profiles) were more appropriate (Morin et al., 2011b), as further indicated by their better fit.

settings, in which practitioners will find useful to have a clear portrait of clients that would be assigned to each profile.

To achieve the second objective (part a), scores on individual items of the self-management questionnaire were compared across profiles. To do so, the LPA was re-run in Mplus, but this time variables corresponding to the individual self-management items were integrated using another auxiliary command, named BCH, designed for such purposes (Asparouhov and Muthén, 2014a). This tested equality of means across profiles for each self-management strategy.

To achieve the second objective (part b), associations between profiles and participants' background characteristics (clinical and sociodemographic variables) were examined by introducing these characteristics using the auxiliary BCH command for continuous variables (in our case, only the age variable) and another similar command, named DCAT, designed for categorical variables (all the variables other than age) (Asparouhov and Muthén, 2014a). The DCAT command provides a between-profile comparison of the estimated probability of each characteristic.

To achieve the third objective, associations between profiles and criterion variables were examined with the BCH command (Asparouhov and Muthén, 2014a) to test equality of means across profiles on criterion variables.

Data Preparation

Table 1 shows descriptive statistics for self-management subscales and recovery indicators. As shown in this table, only a small proportion of missing values were observed for these variables (between 0.0 and 2.0%). The same was found for background characteristics (between 0.0 and 4.7%) as well as criterion variables (between 0.0 and 0.7%). For deriving the latent profiles, which was the analysis at the core of the study, models were estimated in Mplus using a full information maximum likelihood (FIML) algorithm. This estimation method does not require deletion of cases with missing data but instead uses the information available from all the participants (Schlomer et al., 2010). This algorithm has proved to be the most robust approach for dealing with missing values without deleting cases (Newman, 2014). For the analysis performed in SPSS, deletion of cases with missing values was used. This deletion should have a negligible impact, given the very low percentage (<5%) of missing values (De Vaus, 2002; Tabachnick and Fidell, 2013).

RESULTS

Exploring the Overall Bivariate Relationships of Self-Management and Recovery Indicators

As shown in **Table 1**, the three types of self-management strategies were positively related (correlations either significant or marginally significant). With regard to the recovery indicators, positive mental health had a negative relationship with both depression and anxiety symptom severity. Supporting the discriminant validity of the measures, the confidence interval of the correlation coefficients of positive mental health with depression and anxiety symptom severity did not include 1

(Cheng, 2011). The same observation applied for depression and anxiety symptom severity, which were positively related, but the confidence interval also did not include 1. As for the association between self-management strategies and recovery indicators, clinical strategies were positively related to depression and anxiety symptom severity, but not to positive mental health. Empowerment and vitality strategies were both negatively associated with depression and anxiety symptom severity and positively associated with positive mental health.

Identifying the Number of Latent Profiles and Drawing Their General Portrait

LPA was performed using clinical, empowerment, and vitality self-management strategies, as well as depression severity, anxiety severity, and positive mental health as recovery indicators. The analysis was performed multiple times, each time increasing the requested number of profiles. As shown in **Table 2**, in each case, all the profiles contained more than 5% of participants (Hamza and Willoughby, 2013). *P*-value of the BLRT test suggested that adding profiles was necessary up to seven profiles. Values for AIC and SSA-BIC were increasingly lower, suggesting better fit as the number of profiles increased. A graphical examination (elbow plot, Morin, 2016) of the evolution of these indicators showed that the slope flattened after four profiles (with only minimal decrease with more profiles subsequently). BIC and CAIC were lowest for the four-profile model. However, according to the VLMR and ALMR, models with more than three profiles were not necessary. Given this pattern of indices, the three-profile and four-profile models were both examined. Three profiles from these two models showed a very similar pattern in terms of self-management and recovery indicators. The only difference was the fourth profile of the four-profile model. This profile did not add substantive meaning (i.e., scores were moderate on all indicators, which is not particularly relevant in terms of Provencher and Keyes' theory). For the sake of parsimony and because of its greater theoretical conformity, the three-profile model was thus selected as the final one. The entropy value was high. **Table 3** shows the classification quality was satisfactory, with high probabilities of participants' belonging in the assigned profile (between 0.92 and 0.97) and low cross-probabilities (between 0.01 and 0.07).

Figure 1 shows the standardized means of participants in each profile on the variables used in the LPA. **Table 4** presents results from the equality of means and chi-square tests comparing the profiles to one another on these variables. Based on the overall pattern of these results, a summary label inspired by Keyes and Lopez's (2002) classification was assigned to each profile, which admittedly could not fully convey, in just a few words, the recovery dynamics underlying each profile.

The first profile—those who were *Floundering*, yet trying to manage their symptoms—included 52 participants (34.9%). These had moderately severe depression and anxiety symptoms, as well as the lowest level of positive mental health among the three profiles. More than half scored over the clinical cut-off for moderate depression and anxiety symptoms, and only one participant had a high level of positive mental health. Their use

TABLE 1 | Correlations between the main study variables and descriptive statistics ($N = 146\text{--}149$).

Variables	r (95% CI) ^a					
	1.	2.	3.	4.	5.	6.
1. Clinical self-management	–					
2. Empowerment self-management	0.15 [†] (–0.02, 0.31)	–				
3. Vitality self-management	0.16 [†] (–0.02, 0.31)	0.37*** (0.21, 0.52)	–			
4. Depression symptom severity	0.21** (0.08, 0.34)	–0.34*** (–0.48, –0.19)	–0.30*** (–0.45, –0.17)	–		
5. Anxiety symptom severity	0.20* (0.01, 0.35)	–0.21** (–0.38, –0.03)	–0.23** (–0.40, –0.06)	0.70*** (0.61, 0.78)	–	
6. Positive mental health	–0.03 (–0.16, 12)	0.59*** (0.46, 0.69)	0.41*** (0.24, 0.54)	–0.65*** (–0.74, –0.56)	–0.45*** (–0.58, –0.30)	–
<i>M</i>	2.32	2.39	2.10	7.70	5.64	2.65
<i>S.D.</i>	0.85	0.68	0.88	5.47	4.40	1.03
Skewness	–0.41	0.14	0.18	0.34	0.40	–0.04
Kurtosis	–0.18	–0.63	–0.68	–1.05	–1.08	–0.90
Missing	0.00%	2.01%	0.67%	0.00%	0.00%	0.00%

Total sample size varies between 146 and 149 due to missing data on some variables.

^aBias-corrected accelerated confidence intervals based on $N = 1000$ bootstrap samples.

*** $p \leq 0.001$, ** $p \leq 0.01$, * $p \leq 0.05$, [†] $p \leq 0.10$.

TABLE 2 | Fit of the compared latent profile models with increasing numbers of profiles ($N = 149$).

Number of profiles (k)	LL	FP	AIC	BIC	CAIC	SSA-BIC	P -value VLMR	P -value ALMR	P -value BLRT	Entropy	<5% of sample
1	–1638.53	12	3301.07	3337.11	3349.11	3299.14	–	–	–	–	No
2	–1532.69	19	3103.39	3160.46	3179.46	3100.33	0.000	0.000	0.000	0.87	No
3	–1507.23	26	3066.45	3144.56	3170.56	3062.27	0.020	0.022	0.000	0.90	No
4	–1484.83	33	3035.66	3134.79	3167.79	3030.35	0.176	0.186	0.000	0.85	No
5	–1472.26	40	3024.52	3144.67	3184.67	3018.08	0.424	0.433	0.010	0.86	No
6	–1457.29	47	3008.58	3149.77	3196.77	3001.03	0.382	0.387	0.000	0.87	No
7	–1445.78	54	2999.56	3161.77	3215.77	2990.88	0.463	0.469	0.030	0.88	No
8	–1433.93	61	2989.86	3173.10	3234.10	2980.05	0.305	0.309	0.070	0.89	No

LL, loglikelihood; FP, number of free parameters; AIC, Akaike Information Criteria; BIC, Bayesian Information Criteria; CAIC, Consistent AIC; SSA-BIC, Sample-Size-Adjusted BIC; VLMR, Vuong-Lo-Mendell-Rubin Likelihood Ratio Test for $k-1$ profiles vs. k profiles; ALMR, Adjusted Lo-Mendell-Rubin Likelihood Ratio Test for $k-1$ profiles vs. k profiles; BLRT, Bootstrapped Likelihood Ratio Test for $k-1$ profiles vs. k profiles.

of self-management strategies was overall low to moderate, and empowerment and vitality strategies were used significantly less often than in the other profiles. Less than 10% of participants in this profile used these strategies often or very often. The second profile—*Struggling*, but fully engaged—was comprised of 14 participants (9.4%) and included those who, overall, performed self-management strategies often, and more frequently than those in other profiles for clinical and vitality strategies. Their use of vitality strategies was more than one SD above the overall sample mean. All participants in this profile scored above the clinical cut-off for depression symptom severity. They also reported a higher level of anxiety symptoms compared to the overall sample (more than one SD above the mean). Despite this pattern of symptoms similar to the *Floundering* profile,

participants from the *Struggling* profile reported experiencing a higher positive mental health level. The last profile—those well on the way to *Flourishing*—was the most frequent ($n = 83$, 55.7%) and included participants with relatively high levels of self-management, especially empowerment. They had the least severe symptoms of depression and anxiety (<3% above the clinical-cut off) compared to other profiles, as well as a high level of positive mental health (65% had high scores).

Describing the Specific Self-Management Strategies Used in Each Profile

Profiles were compared regarding use of the 18 specific self-management strategies measured in the questionnaire. As shown in **Table 5**, only two self-management strategies were used to the

TABLE 3 | Average latent profile probabilities for most likely latent profile membership (row) by latent profile (column) (N = 149).

	Profile 1: Floundering	Profile 2: Struggling	Profile 3: Flourishing
Profile 1: Floundering	0.952	0.024	0.024
Profile 2: Struggling	0.070	0.921	0.009
Profile 3: Flourishing	0.021	0.007	0.972

**FIGURE 1 | Plot of the standardized means of the latent profiles on indicators (N = 149) compared to the overall sample mean.**

same extent by people in the different profiles: participating in a support or help group (low frequency) and taking medication for one's mental health problem (high frequency). The remaining clinical strategies (looking for available help resources, consulting a professional, and being actively involved in one's follow-up with professionals) were used more frequently, between *often* and *very often*, by people in the *Struggling* profile as compared to the two other profiles. Overall, empowerment strategies were used between *very rarely* or *sometimes* by people in the *Floundering* profile. In contrast, as a general pattern, participants in the *Struggling* and *Flourishing* profiles used these strategies between *sometimes* and *often*. These two profiles used the following empowerment strategies more frequently, compared to *Floundering* participants: trying to solve one's problem one step at a time, trying to recognize relapse signs, focusing one's attention on the present moment, learning to live with one's strengths and weaknesses, trying to love oneself, and finding comfort in people around oneself. Finally, participants in the *Struggling* profile used all the vitality strategies more frequently (overall between *often* and *very often*) than those in the other profiles: doing activities one enjoys to maintain a healthy lifestyle, engaging in sports, having healthy eating habits, and doing relaxation exercises.

Characterizing the Participants in Each Latent Profile

Table 6 presents the profiles' associations with the participants' background characteristics. Probability of self-reporting a depression diagnosis was higher for the *Floundering* or *Struggling*

profiles than for the *Flourishing* profile. Probability of self-reporting an anxiety disorder diagnosis was higher for the *Floundering* profile than for the *Flourishing* profile. Probability of self-reporting a bipolar disorder diagnosis was higher for the *Flourishing* profile than for the *Floundering* profile. Being currently involved in psychotherapy was more likely for the *Struggling* profile than for the two other profiles. Probability of being a man was higher in the *Floundering* profile than the other two profiles. Probability of living in a low-income household or probability of being single were higher for the *Floundering* profile than for the *Flourishing* profile.

Verifying the Associations of Profiles with Criterion Variables

As shown in Table 7, people in the *Struggling* and *Flourishing* profiles appraised their personal goals more positively and reported participating more frequently in society, compared to those in the *Floundering* profile. They also reported having more developed self-care abilities and using more adaptive coping (behavioral and cognitive) to deal with the stress associated with their mental health problem. This is consistent with these people's higher levels of positive mental health and engagement in self-management strategies. Also converging with the fact that the highest level of self-management was found in the *Struggling* profile, this profile had among the highest scores for all coping types. Interestingly, the *Struggling* and the *Floundering* profiles scored as high for avoidance coping. Their scores indicated a relatively low frequency of this type of coping, but nevertheless higher than in the *Flourishing* profile. This shared aspect of the *Floundering* and *Struggling* profiles, in terms of the use of this maladaptive coping style, is consistent with the fact that both profiles presented more severe symptoms.

DISCUSSION

In line with the shift of mental health services toward a person-centered approach (Corrigan, 2015), the present study explored for the first time individual recovery profiles. The results suggest three such profiles underlying the engagement of people with mental disorders in their recovery. Their pattern of associations with criterion variables (personal goal appraisal, social participation, self-care abilities, coping) was consistent with previous theoretical and empirical work on factors that form the foundation of successful self-management and mental health recovery. In keeping with the description of these profiles in terms of recovery indicators and self-management strategies, the *Floundering* profile presented the most unfavorable portrait on the criterion variables, while the *Flourishing* profile presented the most favorable portrait, and the in-between *Struggling* profile presented a mostly favorable, yet mixed portrait.

Understanding Self-Management Differently

Although traditional variable-oriented analytical strategies are useful for seeing the big picture of how specific variables relate to each other at the group level, they are insufficient to inform

TABLE 4 | Comparison of the latent profiles on the profile variables (continuous) and their dichotomized version.

Continuous indicators	Equality of means results ^a				Chi-square results ^b				
	Floundering <i>M</i> (S.E.)	Struggling <i>M</i> (S.E.)	Flourishing <i>M</i> (S.E.)	χ^2	Dichotomized indicators	Floundering <i>n</i> (%)	Struggling <i>n</i> (%)	Flourishing <i>n</i> (%)	χ^2
SELF-MANAGEMENT^c									
Clinical	2.21 _a (0.11)	3.12_b (0.19)	2.24 _a (0.10)	24.43***	Score ≥ 3 (<i>strategies used often</i>)	8 _a (15.4)	10_b (71.4)	18 _a (21.7)	19.53***
Empowerment	1.98 _a (0.08)	2.70_b (0.18)	2.59_b (0.07)	31.38***	Score ≥ 3 (<i>strategies used often</i>)	3 _a (5.9)	6_b (46.2)	26_b (31.7)	15.36***
Vitality	1.47 _a (0.10)	3.03_b (0.20)	2.33 _c (0.09)	66.84***	Score ≥ 3 (<i>strategies used often</i>)	2 _a (3.8)	9_b (64.3)	25 _c (30.5)	25.68***
RECOVERY INDICATORS									
Depression ^d	12.77_a (0.54)	11.06_a (1.19)	3.89 _b (0.35)	172.61***	Score ≥ 10 (clinical cut-off)	31 _a (59.6)	14_b (100.0)	2 _c (2.4)	81.99***
Anxiety ^e	8.86 _a (0.44)	11.89_b (0.57)	2.47 _c (0.26)	311.56***	Score ≥ 8 (clinical cut-off)	41_a (78.8)	8_a (57.1)	2 _b (2.4)	86.58***
Positive mental health ^f	1.73 _a (0.10)	2.90_b (0.22)	3.20_b (0.09)	102.92***	Score > 3 (<i>positive manifestations about 2 or 3 times/week</i>)	1 _a (1.9)	5_b (35.7)	54_b (65.1)	53.12***

Total sample size varies between 146 and 149 due to missing data on some of the variables.

^aFor each indicator, means with different subscripts are different at $p \leq 0.05$ according to equality of means results, and cells in bold highlight the profiles with the highest average scores.

^bPercentages calculated on non-missing data. For each indicator, proportions with different subscripts are different at $p \leq 0.05$ according to post-hoc tests (Bonferroni), and cells in bold highlight the profiles with the highest proportions.

^cMeasured with the Mental Health Self-management Questionnaire, scores from 0 (Never) to 4 (Very often).

^dMeasured with the Patient Health Questionnaire 9, scores from 0 (None) to 27 (Severe).

^eMeasured with the General Anxiety Disorder 7, scores from 0 (None) to 21 (Severe).

^fMeasured with the Mental Health Continuum–Short Form, scores from 0 (Never) to 5 (Every day).

*** $p \leq 0.001$.

health professionals working from a person-centered perspective (Cloninger, 2013). In contrast, there is a natural fit between the person-centered philosophy of care and person-oriented statistical analysis, because both recognize the person as more than the sum of parts (Laursen, 2015). Nevertheless, person-oriented analysis is still rarely used even to study topics closely related to person-centered care, such as people's engagement in self-management and recovery. Our study illustrates that person-oriented analysis can provide insightful results with the potential to stimulate reflection.

By definition, from a traditional variable-oriented perspective, positive associations would have been expected between self-management and recovery. By extension, it would have been expected that those who were more engaged in strategies to reduce their symptoms (clinical self-management), trying more actively to gain control by harnessing their positive sense of self (empowerment self-management), and adopting a healthier and active lifestyle (vitality self-management) would have had less severe symptoms as well as higher levels of positive mental health. Of the three identified profiles, the *Floundering* and *Flourishing* profiles were overall in line with this reasoning. Participants in the *Floundering* profile used empowerment and vitality self-management strategies less frequently than did those who were *Flourishing*. As a corollary, people in the former profile scored more negatively on recovery indicators than did those in the latter profile. However, despite their different scores on recovery indicators, people in both profiles reported using clinical strategies to the same extent (only moderately) as part of their self-management "recipe." This provides evidence that the relationship between self-management and recovery indicators is not as straightforward as might be thought, at least when studied from a cross-sectional perspective.

In that same vein, a surprising result was seen in the *Struggling* profile, where respondents reported high self-management co-existing with moderately severe symptoms. People in this profile were the most activated and were involved in a diverse combination of frequently used clinical, empowerment, and vitality self-management strategies. They were also more likely to be currently involved in psychotherapy, potentially indicating or resulting from their higher engagement (see review from Kreyenbuhl et al., 2009, on engagement and treatment). Their symptoms were among the most severe observed across the different profiles, suggesting that a high level of engagement, even in clinical strategies specifically targeting symptoms, is not necessarily associated with reduced symptomatology. Indeed, these participants had on average the most severe anxiety levels and used avoidance coping strategies (i.e., substance use, denial, behavioral disengagement) to the same extent as did *Floundering* participants. Even though *Struggling* participants' score on the use of such maladaptive coping strategies was low, it was nevertheless similar to levels observed in studies with other clinical samples (Meyer, 2001; Nazir and Mohsin, 2013). One of those studies (Meyer, 2001) suggested that the use of maladaptive coping is associated with higher symptom severity. A review of the literature supports the notion that avoidance coping could be associated with relapse, recurrence, and greater time to recovery in mood disorders (Christensen and Kessing, 2005). Over the long term, use of avoidance coping has been shown to generate stress, which can increase symptoms (Holahan et al., 2005).

It is also possible that *Struggling* participants' focus on working through their symptoms elevated their stress level. This would be consistent with literature suggesting that, as part of the health engagement process, people with chronic diseases tend to experience a phase of arousal in which they are hyper-attentive

TABLE 5 | Comparisons of latent profiles on the frequency of use of self-management strategies.

Items from the Mental Health Self-management Questionnaire	Floundering <i>M (S.E.)</i>	Struggling <i>M (S.E.)</i>	Flourishing <i>M (S.E.)</i>	χ^2
CLINICAL SELF-MANAGEMENT				
I look for available resources to help me with my difficulties (websites, organizations, healthcare professionals, books, etc.).	2.23 _a (0.16)	3.69_b (0.19)	2.42 _a (0.13)	39.23***
I consult with a professional (doctor, psychologist, social worker, etc.) concerning my mental health disorder.	2.59 _a (0.18)	3.43_b (0.30)	2.19 _a (0.16)	13.41***
I get actively involved in my follow-up with the healthcare professionals I consult (physician, psychologist, social worker, etc.).	2.22 _a (0.18)	4.00_b (0.11)	2.45 _a (0.16)	96.12***
I participate in a support or help group to help me manage my difficulties.	0.59 (0.15)	1.71 (0.51)	0.68 (0.13)	4.19 n.s.
I take medication for my mental health problem as directed by a healthcare professional.	3.12 (0.22)	3.45 (0.33)	3.50 (0.13)	2.26 n.s.
EMPOWERMENT SELF-MANAGEMENT				
I try to solve my problems one step at a time.	2.13 _a (0.14)	2.84_b (0.27)	2.52_b (0.11)	6.73*
I try to recognize the warning signs of a relapse of my mental health disorder.	2.40 _a (0.13)	3.05_b (0.26)	3.01_b (0.11)	12.77**
I learn to differentiate between my mental health problem and myself as a person.	1.83 _a (0.15)	2.01 _{a,b} (0.27)	2.50_b (0.14)	10.56**
I focus my attention on the present moment.	1.83 _a (0.14)	2.47_b (0.26)	2.72_b (0.11)	25.03***
I learn to live with my strengths and weaknesses.	2.08 _a (0.13)	3.08_b (0.21)	2.82_b (0.10)	23.18***
I congratulate myself for my successes, large and small.	1.73 _a (0.16)	2.44 _{a,b} (0.35)	2.43_b (0.13)	11.83**
I try to love myself as I am.	1.73 _a (0.13)	2.57_b (0.28)	2.65_b (0.11)	30.41***
I take my capabilities into account when arranging my schedule.	1.97 _a (0.17)	2.67 _{a,b} (0.41)	2.42_b (0.12)	5.19 ^t
I find comfort, I feel listened by people around me.	1.92 _a (0.14)	3.01_b (0.32)	2.42_b (0.12)	12.06**
VITALITY SELF-MANAGEMENT				
I do activities I like to maintain an active lifestyle.	1.36 _a (0.12)	3.31_b (0.27)	2.49 _c (0.12)	64.43***
I engage in sport, physical activity.	1.02 _a (0.15)	3.41_b (0.22)	2.19 _c (0.15)	84.06***
I have healthy eating habits.	2.06 _a (0.13)	3.54_b (0.19)	2.99 _c (0.10)	49.51***
I do exercises to relax (yoga, tai chi, breathing techniques, etc.).	1.12 _a (0.15)	2.85_b (0.34)	1.72 _c (0.14)	22.05***

Response scale: 0 (Never), 1 (Very rarely), 2 (Sometimes), 3 (Often), and 4 (Very often). Items were presented to participants in French. The English version above was produced using a back-translation approach (Vallerand, 1989).

Total sample size varies between 142 and 149 due to missing data on some of the items. In each line, means with different subscripts are different at $p \leq 0.05$, and cells in bold highlight the profiles with the highest average scores.

*** $p \leq 0.001$, ** $p \leq 0.01$, * $p \leq 0.05$, ^t $p \leq 0.10$.

to their symptoms yet are still unable to cope adequately, causing them anxiety (Barello et al., 2014; Graffigna et al., 2014). Taking part in a psychotherapeutic process can also be demanding for a person, especially when using stressful procedures such as exposure (Wills, 2008). The burden associated with self-management can also cause stress (Sav et al., 2013). Seeking to improve one's happiness has been shown to be "a delicate art" that can backfire (Catalino et al., 2014, p. 1160). Likewise, our cross-sectional results may suggest that actively seeking to get better and wanting to do the best for one's health might put additional stress on people with mood and anxiety disorder, at least temporarily or in the short term.

An alternative interpretation is that participants in the *Struggling* profile engaged in self-management to deal with their residual symptoms. The literature on depression (the diagnosis most reported in this profile) is clear on the fact that, even when responding successfully to pharmacotherapy or psychotherapy, a significant proportion of people still have to contend with incapacitating residual symptoms (see review from Fava et al.,

2007, and by Nierenberg, 2015). Anxiety is one of the most common residual symptoms in depression disorders (Fava et al., 2007; D'Avanzato et al., 2013). From that standpoint, it is possible that *Struggling* participants' symptoms (notably their relatively high anxiety) did not result from their active self-management, but rather were the very reason why they actively engaged in self-management. These participants' attempts to deal with stressful residual symptoms may also explain their involvement in a diversity of coping strategies, as shown by their elevated coping scores, even on apparently contradictory subscales (e.g., avoidance vs. behavioral coping). As put forward by Folkman and Lazarus (1991), a person may seek and try several, sometimes opposite, ways of dealing with a stressful situation. While persons in this profile may not be reaping the benefits of their coping and self-management efforts in the moment, they might experience less severe symptoms over the longer term. Longitudinal studies exploring how symptom severity and self-management relate to each other over time are needed to verify this.

TABLE 6 | Associations between participants' background characteristics and latent profiles.

Characteristics	Estimated probability of each characteristic within each profile			χ^2
	Floundering	Struggling	Flourishing	
SELF-REPORTED DIAGNOSIS				
Depressive disorder	0.69 _a	0.83 _a	0.43 _b	13.24***
Anxiety disorder	0.53 _a	0.28 _{a,b}	0.29 _b	6.61*
Bipolar disorder	0.21 _a	0.30 _{a,b}	0.47 _b	10.32**
Comorbidity between depressive, anxiety and/or bipolar disorders	0.34	0.43	0.19	4.78 ^t
SELF-REPORTED TREATMENTS				
Pharmacotherapy in the last month	0.81	0.81	0.89	1.65 n.s.
Current psychotherapy	0.43 _a	0.90 _b	0.28 _a	26.17***
SOCIODEMOGRAPHIC VARIABLES				
Age	<i>M</i> = 40.11; <i>S.E.</i> = 1.90	<i>M</i> = 44.61; <i>S.E.</i> = 3.26	<i>M</i> = 41.77; <i>S.E.</i> = 1.36	1.37 n.s.
Gender (man vs. woman)	0.31 _a	0.06 _b	0.15 _b	5.07 ^t
Education level (university vs. lower)	0.53	0.54	0.67	2.40 n.s.
Low income (yes vs. no)	0.39 _a	0.15 _{a,b}	0.14 _b	7.45*
Single (yes vs. no)	0.76 _a	0.43 _{a,b}	0.39 _b	16.04***

For the low-income variable, only participants from Canada were included, given that this variable was created only for this subgroup. Thus, probabilities were calculated on available data (total sample size varies between 135 and 149 depending on the characteristic considered). In case of a significant chi-square, for each indicator, probabilities with different subscripts are different at $p \leq 0.05$, and the cell in bold highlights the profile with the highest probability.

*** $p \leq 0.001$, ** $p \leq 0.01$, * $p \leq 0.05$, ^t $p \leq 0.10$.

TABLE 7 | Comparisons of latent profiles on criterion variables.

Criterion variables	Floundering <i>M</i> (<i>S.E.</i>)	Struggling <i>M</i> (<i>S.E.</i>)	Flourishing <i>M</i> (<i>S.E.</i>)	χ^2
Personal goal appraisal ^a	4.58 _a (0.25)	6.99_b (0.37)	7.16_b (0.14)	80.01***
Social participation ^b	1.00 _a (0.06)	1.71_b (0.18)	1.50_b (0.06)	37.16***
Self-care abilities ^c	3.50 _a (0.11)	4.17_b (0.14)	4.41_b (0.06)	55.28***
Emotional coping ^d	1.86 _a (0.10)	2.59_b (0.15)	2.13 _c (0.08)	15.66***
Behavioral coping ^d	1.47 _a (0.10)	2.32_b (0.18)	2.12_b (0.08)	29.86***
Cognitive coping ^d	0.88 _a (0.07)	1.45_b (0.07)	1.50_b (0.06)	51.12***
Avoidance coping ^d	1.02_a (0.07)	0.94_a (0.13)	0.66 _b (0.06)	16.41***

Total sample size varies between 148 and 149 due to missing data on a criterion variable. In each line, means with different subscripts are different at $p \leq 0.05$, and cells in bold highlight the profiles with the highest average scores.

^aMeasured with the Personal Project System Rating Scale, scores from 1 (Very negative) to 10 (Very positive).

^bMeasured with the Social Participation Scale, scores from 0 (Never) to 4 (Almost every day).

^cMeasured with the Therapeutic Self-Care Scale, scores from 0 (Not at all) to 5 (Completely).

^dMeasured with the Brief COPE, scores from 0 (Not at all) to 3 (A lot).

*** $p \leq 0.001$.

Supporting and Expanding the Complete Mental Health Recovery Model

Provencher and Keyes's Complete Mental Health Recovery model (2010, 2011, 2013) was developed on the idea that symptom severity and positive mental health are two distinct dimensions and that their intersections form six states of recovery. This proposition was based on studies in which participants from the general population were artificially classified into different profiles corresponding to these six states (Keyes, 2005, 2007).

Our results based on an inductive method of classification (LPA) confirm the existence of some of these profiles, thereby providing general supporting evidence for their model.

The *Flourishing* profile found in the present study resembles the state described by Provencher and Keyes (2010, 2011, 2013) in which the person is recovered in terms of symptom severity and shows a moderately high level of positive mental health. Similarly, the *Floundering* profile mirrors their description of the opposite state (non-recovered from the mental illness and low positive mental health). Finally, the *Struggling* profile echoes Provencher and Keyes' (2010, 2011, 2013) state of non-recovery from symptoms concomitant with a moderate level of positive mental health. Although our participants were not numerous in this profile, its existence is supported by the model's adequate fit and the satisfactory classification probabilities. The existence of this profile is essential because it demonstrates the foundational idea that people with important mental health symptoms can nevertheless experience frequent manifestations of well-being that help make their life worth living, as positive psychologists would say (Seligman et al., 2004). Three others states (e.g., recovered from mental illness and low positive mental health) proposed by Provencher and Keyes (2010, 2011, 2013) were not found in the present study. However, it is possible that, with a larger sample size, probabilities of observing these would have been augmented. Even in the large general population studies cited above (Keyes, 2005, 2007), such states have been shown to be among the least frequent.

Beyond providing confirmation, the present study complements the Complete Mental Health Recovery model by explicitly incorporating self-management strategies. Provencher and Keyes (2010) recognized people's active role in their recovery and gave examples of strategies that could promote the process.

The present study expands on this by providing unprecedented empirical data on the level of self-management engagement shown by people in different profiles of recovery. It also reveals specific self-management strategies that people in each profile tend to combine.

The level of engagement in almost all self-management strategies was lowest for participants in the *Floundering* profile. Although time since onset of their disorder was not collected, this profile relates to the description of people who are in the beginning of the recovery process (Provencher and Keyes, 2010). Researchers have labeled this the “moratorium” stage, characterized by hopelessness and self-protective withdrawal (Andresen et al., 2003, 2006). Taking their medication as prescribed was the only self-management strategy that participants from this profile implemented on a regular basis, which seems consistent with the dependence on external support that distinguishes this beginning stage (Andresen et al., 2006).

People in the *Struggling* profile had the highest level of self-management. Their combination of self-reported strategies was characterized by regular use of help-seeking strategies (e.g., inform oneself about resources, consult with a professional), in line with their higher probability of being involved in psychotherapy and having more severe symptoms (Hämäläinen et al., 2008). They also were keeping themselves physically active and healthy by maintaining a good diet and engaging in sports and relaxation exercises. Among other strategies, they were trying to solve their problems one step at a time and to focus on the present moment. These self-management strategies evoke lifestyles changes, behavioral activation, problem resolution, and mindfulness activities that are suggested or recommended in clinical guidelines (e.g., National Institute for Health and Care Excellence, 2009; Scottish Intercollegiate Guidelines Network, 2010). Participants in this profile may have been encouraged to use such strategies by a psychotherapist or other health professional they consulted. Given their use of potentially physically energizing strategies, it is not surprising that their level of positive mental health was relatively high, in keeping with a recent qualitative study showing a sense of energy to be a marker of positive mental health in people with mental disorders (Mjøsund et al., 2015). Although such a profile of self-management strategies has not been described explicitly in the literature before, it bears some resemblance to descriptions of recovery stages after the initial “moratorium” (Andresen et al., 2003). In those stages the individual struggles with the illness but, at some turning point, manages to move into action (Davidson and Strauss, 1992; Spaniol and Wewiorski, 2012).

As for those in the *Flourishing* profile, their moderately high self-management scores suggested that, although well on the way to full recovery, they were still very engaged in getting better. Even though taking their medication as prescribed and recognizing relapse signs were important for them, in all likelihood their main focus was not on managing the disorder for itself, but rather for the benefit of optimizing their overall well-being. Provencher and Keyes (2011, p. 64), described people at similar states of recovery: “They look for opportunities to challenge themselves and to reach a sense of serenity and peace

of mind. [...] When deficits are still present, individuals are well aware of them and know how to best use them while continuing to grow and to optimize their own potential in the pursuit of challenging goals.” Consistent with this portrayal, the strategies characteristic of the *Flourishing* profile were related to accepting, working around, and transcending difficulties, such as arranging their schedule around their capabilities and congratulating themselves on their successes. This pattern of self-management strategies is consistent with the final stages of the recovery process (“rebuilding” and “growth”), in which people forge a new positive sense of self and develop a feeling of confidence in their abilities to face challenges (Andresen et al., 2003, 2006).

Bringing Background Characteristics and Recovery Inequalities to the Foreground

Guidelines for person-centered health services emphasize the importance of culturally sensitive assessment and intervention practices (Adams et al., 2004; Porche, 2013) that are tailored or individualized to the person’s background (Lauver et al., 2002). The present study revealed several background characteristics associated with each profile. Most notably, the least favorable profile (*Floundering*) was characterized by an array of clinical (self-reported depressive or anxiety disorder) and sociodemographic variables (male gender, low income, and singlehood). In contrast, the most favorable profile (*Flourishing*) was characterized by a different clinical background (self-reported bipolar disorder), as well as the opposite sociodemographic variables (being a female, having sufficient income, and having a life partner). These variables may represent risk and protective factors for practitioners to consider in their holistic comprehension of their clients’ situation.

Consistent with a previous study (Vermeulen-Smit et al., 2015) suggesting that anxiety disorders could be associated with a form of unhealthy lifestyle, the *Floundering* profile was the profile most clearly characterized by an overrepresentation of people with a self-reported anxiety disorder, and was the least engaged in vitality self-management strategies. Also of particular interest was the association of the *Floundering* profile with social variables (gender, singlehood, low income), in line with several previous studies in the wider mental health field. For example, several studies have shown singlehood to be related to higher prevalence of depression and anxiety (see Martins et al., 2012). In a recent study of people with a depressive disorder, single marital status at baseline predicted non-recovery in terms of depressive symptoms 11 years later (Markkula et al., 2016), which is congruent with a stream of research concerning the association of marital status with health and health behaviors. This relation could be due to multiple reasons, such as the fact that economic, psychological, and social resources are less accessible to single people (see reviews from Robards et al., 2012; Robles et al., 2014). Economic disadvantage is also associated with higher prevalence of depression and anxiety disorders (see Martins et al., 2012). It has been suggested that psychosocial resources helpful for coping effectively with life stressors, such as personal control and social support, may be less available to disadvantaged people (Taylor

and Seeman, 1999). People with low incomes are also more likely to face financial barriers to obtaining mental health services (Sareen et al., 2007).

Concerning gender, although anxiety and mood disorders prevalence rates are generally higher in women than in men (Faravelli et al., 2013; see reviews from Piccinelli and Wilkinson, 2000; Bekker and van Mens-Verhulst, 2007), research has documented several health challenges faced by men, such as lower subjective well-being (Graham and Chattopadhyay, 2013) and higher suicide rates (Nock et al., 2008). Men also tend to have less healthy lifestyles (Von Bothmer and Fridlund, 2005) and to consult less than women in cases of emotional problems, due to their endorsement of traditionally masculine cultural norms (Möller-Leimkühler, 2002). Overall results from the present study expand these previous findings by pointing out potential social inequalities in terms of chances of recovery from mood and anxiety disorders.

Implications for Patient-Centered Interventions

From a person-centered care perspective, people's idiosyncratic recovery profiles (in terms of self-management strategies and recovery indicators) should be considered by professionals who intervene with them. Traditional self-management support interventions usually focus on symptom reduction (e.g., Bilsker and Patterson, 2007; Lorig et al., 2014). Our findings confirmed that people use different combinations of self-management strategies, focusing not only on symptoms, but also on promoting their overall positive mental health. Thus, health professionals should consider the whole diversity of self-management behaviors implemented by their clients. Through a comprehensive investigation, professionals can seize opportunities to build clients' confidence by offering sincere praise for their self-management actions, even small ones, in line with solution-focused principles (Winbolt, 2011).

The low frequency of self-management strategies observed in the *Floundering* profile might warrant discussions with clients in such a profile to identify potential emotional (e.g., feeling of incompetence) and cognitive (e.g., lack of knowledge) barriers to self-management. Health engagement in the context of chronic illness is intertwined with emotional and cognitive processes (Graffigna and Barelo, 2015; Graffigna et al., 2015a). If done appropriately and respecting the individual's wishes, working through these barriers together could help set the client on a path of increased engagement in self-management, and ultimately into the *Flourishing* profile. To that end, the recently validated Patient Health Engagement Scale (Graffigna et al., 2015a) is a 5-item short scale to help practitioners identify their clients' position in their engagement process, considering the emotional and cognitive components. Discussing this scale's results in the clinical encounter can be useful to stimulate person-centered communication between practitioner and client (Graffigna et al., 2015a). Such a client-practitioner partnership could facilitate engagement in self-management (Trivedi et al., 2007).

Results from the *Struggling* profile highlight a possibility that anxiety can arise, at least temporarily, from engaging deeply in self-management. Although the level of self-management was not sufficiently high to be deemed excessive in itself in the present study, the existence of this profile raises a yellow flag. In self-management, as in other domains of life, it is possible that excessiveness causes stress and leads to negative outcomes (Witkin, 1985). While being respectful of clients' engagement, professionals could personalize follow-ups to support people in achieving the delicate balance between actively managing their illness and pursuing other life activities and goals without undue stress.

Our findings suggest that additional efforts should be expended to ensure that mental health services effectively reach and support men, single persons, and those with low incomes in their self-management and recovery. Examples of interventions from the chronic illness or physical health field can be instructive for this purpose, such as self-management interventions developed for people on low income with diabetes (Eakin et al., 2002), or the Scottish Premier League football clubs, which promote weight reduction in men through a gender-sensitized context, content, and style of delivery (Hunt et al., 2014). In 2014, the Geneva Declaration on Person- and People-centered Integrated Health Care for All was adopted, which encouraged commitment to reducing health inequalities and to making person-centered care available for all (Cloninger et al., 2014). This requires not only adapting professional services to people's profiles, but also committing to social justice and participating in wider efforts aimed at "creating well-being-promoting societies as well as treating illness" (Slade, 2010, p. 9).

Limitations and Future Research

The present study is limited by its cross-sectional design. The profiles discovered represent static "snapshots" of the recovery experience taken at one moment in time. As recovery is thought to unfold across time, with "setbacks and plateaus along the way" (Farkas, 2007, p. 72), it is possible that the different profiles are experienced at different moments in the recovery process. Capturing time elapsed since the onset of the disorder would have enabled a first examination of this question, but unfortunately it was not measured in this study. Provencher and Keyes (2011) suggested that people transition from one state to another on the pathway toward complete mental health recovery. One can intuitively conceive that the *Flourishing* profile is more likely to be experienced later in the recovery process, while the *Floundering* profile is more likely to be experienced at the beginning of the process. The *Struggling* profile might represent an intermediate state in which the person becomes deeply engaged in self-management, possibly paving the way toward flourishing. It might also be an end-state for some people who need to deal with residual symptoms over the long run. Such speculations illustrate a set of research questions that have yet to be explored with longitudinal designs.

Although the current sample size appears to be sufficient to conduct LPA according to some suggested guidelines (e.g., Formann, 1984 in Tuma and Decker, 2013; Williams and Kibowski, 2016), it remains limited in terms of generalizability.

Our sample size was modest for multivariate statistics like LPA (Mueller et al., 2010), warranting further studies to replicate the findings, especially the existence of the *Struggling* profile, in which only a limited number of participants were classified. If the power was sufficient to detect meaningful differences between profiles, larger sample sizes would make it possible to verify the few associations that were only marginally significant.

Online research provides valid data (Gosling et al., 2004) and makes it possible to reach individuals who are dispersed geographically (Wright, 2006). However, future studies would benefit from using a traditional face-to-face method, allowing the use of structured clinical interviews (e.g., Structured Clinical Interview for DSM Disorders; First et al., 2002) to thoroughly measure participants' clinical symptoms. Such objective symptom assessment could help rule out alternative interpretations for the findings. In the present study, it is possible that people in the *Struggling* profile, being focused on getting better through self-management and psychotherapy, were more conscious of their symptoms and thus biased toward giving higher scores to self-reported severity measures such as the PHQ-9 and GAD-7.

Beyond background characteristics, several other variables possibly related to profiles warrant examination. Notably, while self-management refers mainly to the *actions* involved in taking care of one's health, other cognitive (e.g., knowledge about their health) and emotional variables (e.g., feelings of confidence) are also likely to be involved and should be considered as potential determinants (Graffigna and Barelllo, 2015; Graffigna et al., 2015a). Also, the study did not examine health professionals' (e.g., psychiatrists, psychologists, general practitioners) contribution to self-management and recovery. A recent measure such as the INSPIRE questionnaire (Williams et al., 2015) could be useful in this regard to assess the extent to which professionals support clients in their personal recovery.

Conclusion

Mood and anxiety disorders figure among the 20 leading causes of disability worldwide (Institute for Health Metrics and Evaluation, 2013). At the heart of person-centered approaches in mental health services (Davidson et al., 2015) lies the principle that people can play an active role in dealing with such incapacitating disorders and in promoting their complete recovery. Yet systematic research-based evidence on self-management and recovery from these disorders is scarce. The present study represents a first thorough quantitative examination of recovery, combining self-management strategies used and recovery indicators.

Although the results need to be replicated, the person-oriented analyses conducted in this study yielded insights for practitioners interested in developing services that are personalized to clients' unique profiles and backgrounds. The list of profiles identified in the study is in no way definitive. Thus, we advise practitioners not to strive to classify their clients into these exact profiles. Rather, we hope the individualized person-centered approach developed in this study can encourage them to adapt their services to their clients' own profiles.

At the theoretical levels, this study integrated notions from different domains of research and interventions, such as the chronic illness, mental health, positive psychology, and patient-centered care literature. We hope the findings will stimulate reflection on how an integrative theoretical framework and innovative methods can provide original empirical information on people's health engagement and how it supports their health and well-being.

AUTHOR CONTRIBUTIONS

SC developed the research design, coordinated data collection, performed the statistical analyses, and wrote the manuscript as part of his Ph.D. thesis. SR contributed to the design of the study and critically reviewed the paper several times. SM conducted the qualitative interviews that served as the basis for the validated self-management questionnaire, enriching the manuscript with her experience. HP took part in planning the study as a co-investigator of the larger research project. Her knowledge as a recovery expert was useful in improving the manuscript. CH is a co-investigator of the larger research project and took part in planning the study. Her expertise on self-management helped improve the manuscript. PR took part in planning the study as a co-investigator of the larger research project. As an expert in mental health services, she critically reviewed the manuscript. MP is a co-investigator in the larger research project and contributed to its planning. His contribution to the study concerned the evaluation of symptom severity. JH is the principal investigator of the larger research project of which the present study is a part. As SC's thesis advisor, JH closely supervised all research stages and critically reviewed the paper. The authors have approved the article and agree to be accountable for all aspects of the work.

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Adaptation and Validation of a Chinese Version of Patient Health Engagement Scale for Patients with Chronic Disease

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The Patient Health Engagement Scale (PHE-s) was designed to assess the emotional and psychological attitudes of patients' engagement along their healthcare management journey. The aim of this study was to validate a culturally adapted Chinese version of the PHE-s (CPHE-s). Three hundred and seventy-seven participants were recruited from eight community health centers in a sample of patients with chronic disease in Hunan Province, China. The original Italian PHE-s was translated into Mandarin Chinese using a standardized forward-backward translation. The Rasch model was utilized and presented uni-dimensionality and good items fitness of the PHE-s. The internal consistency was 0.89 and the weighted Kappa coefficients of the items (test-retest reliability) ranged from 0.52 to 0.79. Both principal component analysis and confirmatory factor analysis supported a single-factor structure of the PHE-s. In testing the external validity, the PHE-s showed a significant moderate correlation with patient activation but not with medicine adherence behavior, which requires further exploration. The result suggested that the PHE-s is a reliable and valid instrument to assess the level of patient engagement in his or her own health management among chronic patients in China. Further analysis of reliability and validity should be assessed among other patient cohorts in China, and future directions for testing changes after patient engagement interventions should be developed by exploring some clinical relevance.

Keywords: patient engagement, patient engagement measure, patient health engagement scale, patient activation, psychometric properties

INTRODUCTION

Chronic diseases are the leading health concerns of the twenty-first century. Due to longer life expectancy and an aging population, control and prevention measures are urgently needed in the healthcare system (Ali et al., 2015). Deaths from chronic diseases rose by just under 8 million between 1990 and 2010, accounting for two-thirds of global deaths, half of all disabilities, and rapidly growing costs (Lozano et al., 2012). The low- and middle-income countries are projected to experience the greatest challenge resulting from chronic disease, which makes up 80% of the causes of death among the world's population (Bloom et al., 2011) and bears

more serious burden (Boutayeb, 2006). In China, rapid transitions are also occurring on account of demography and epidemiology (Zhou et al., 2016). According to the latest data from the “2015 Report on Chinese Nutrition and Chronic Disease” released by the National and Family Planning Commission (NHFPC) (http://www.chinadaily.com.cn/m/chinahealth/2015-07/08/content_21224293.htm), the prevalences of hypertension and diabetes among Chinese adults in 2012 were 25.2 and 9.7%, respectively. The incidence rate of cancer was also on the rise, reaching 235 per 100,000 people in 2013. Moreover, 533 out of every 100,000 Chinese residents died from chronic disease in 2012, resulting in 86.6% of all deaths with cardio-cerebrovascular disease, cancer, and chronic respiratory disease as the top causes (Yang et al., 2013).

As a way of combating this growing health crisis, health systems are under a paradigm shift in the planning and delivery of healthcare from patients being viewed as passive recipients of care to being more active and accountable for their own health (Osborn and Squires, 2012). More and more theories and practices have advocated considering patients to be key resources in self-management of chronic diseases. The evidence-based Chronic Care Model illustrated the importance of productive interactions between patients and health practitioners and also highlighted the crucial connection between patient engagement and desirable health outcome (Wagner et al., 2001; Bodenheimer et al., 2002; Wasson and Coleman, 2014). The increasing evidence has also demonstrated that patients' engagement in their own health care process is a core generic components to achieving patient-centered care and successful health management (Ishikawa and Yano, 2011; Miles and Mezzich, 2011). This is particularly true in the case of chronic disease (Simmons et al., 2014). These growing findings in previous serial studies have shown that patients who are more active and engaged in healthcare frequently report improved patient satisfaction and better care experiences, more effective medication management and improved medication safety, higher quality of life, access to health behaviors, and better clinic outcomes, even likely using fewer health care services and contributing to a reduction of healthcare costs (Coulter, 2012; Osborn and Squires, 2012; Hibbard and Greene, 2013; Barello and Graffigna, 2014; Cunningham, 2014; Laurance et al., 2014).

Clearly, patient engagement is perceived to be of importance in the health care system. It is essential to have a good understanding of the level of patient engagement by assessing what works, how it works, and whether engagement efforts are improving outcomes over time (Carman et al., 2013; Wasson and Coleman, 2014). However, to the best of our knowledge, the measurement of patient engagement remains a substantial issue for policy makers and healthcare practitioners at present; few scientifically validated assessment tools exist to help identify patients' level of engaging in their care. Most generic instruments developed in the field of chronic disease—such as the Patient Enablement Instrument designed to capture patients' ability to

understand the nature of diseases and copy with their health problems (Howie et al., 1998); the Partners in Health scale, which is a generic assessment scale for patients managing their chronic medical conditions (Battersby et al., 2003); the Patient Activation Measure, which provided an evaluation of individuals' knowledge, skill, and confidence for managing their own health or healthcare (Hibbard et al., 2005); and the Self-Management Ability Scale focused on self-management abilities in relation to well-being (Cramm et al., 2012) can be grouped into one category aimed at assessing the patients' ability to self-manage disease conditions (Eikelenboom et al., 2015). In fact, apart from oriented measurements toward health literacy and behavioral components of patients' self-management, increasing evidence was also accumulating to learn from patients' life stories and call on giving birth to patients' chronic illness trajectory across time (Morales-Asencio et al., 2014), particularly on the emotive component of patient engagement. Those were conceived of as the patients' process elaborating and adjusting to the disease and often appeared to be the first movers of patients' confidence and ability to increase health literacy and reinforce self-management behaviors (Hudson et al., 2014; Graffigna and Barello, 2016). Several existing studies have demonstrated that patients with increased and positive emotion and psychology to attend their own health care are more likely to perform improved patient activation (Hibbard and Mahoney, 2010), enact specific health behavior (Graffigna et al., 2015a), and even have ameliorative health-related biological markers (Ismail et al., 2004). On the contrary, patients are often being debilitated because of emotive disorders, and which may further affect their behavioral choices (Shubin et al., 2015) and may even develop into a vicious cycle (Hibbard and Mahoney, 2010).

However, minimal attention has been given in this aspect to evaluate the emotional and psychological dynamic of patients' engagement experiences along their care management journeys using measures like the Patient Health Engagement Scale (PHE-s), which was recently developed by Graffigna et al. (2015a). Even though the psychometric properties of the PHE-s may not yet be well established, it was still able to be generalized in other countries, because it offered a measure of patient engagement for the first time that more holistically considers the psychological elaboration of the patient based on a rigorous conceptualization model—namely, PHE model. In this model, patient engagement was deemed to be a dynamic and evolutionary process that features four stages: blackout, arousal, adhesion, and eudaimonic project (Graffigna and Barello, 2014; Graffigna et al., 2014). The original Italian version of the PHE-s has only five items and is easy to answer due to its shortness. Each item has seven response options that allow patients to position themselves along a continuum of patient engagement experiences (according to the phases featured by the PHE model). The scale has an ordinal nature and is easy to apply. The instrument can be self-administered by the patient and gives interesting and pragmatic cues to the clinicians in order to best customize their communicational and relational strategies aimed at sustaining patient engagement.

At present, a Chinese version of the PHE-s remains unavailable. Therefore, in the current study, we aimed to translate

Abbreviations: PHE, patient health engagement; PHE-s, patient health engagement scale; CPHE-s, The Chinese version of patient health engagement scale.

the original Italian PHE-s into Chinese Mandarin and to evaluate its psychometric properties in a group of patients with chronic disease in China.

MATERIALS AND METHODS

Translation and Culture Adaptation of the PHE-s

In this study, the original version of the PHE-s was translated as recommended by the WHO (World Health Organization) procedures for cross-cultural validation and adaptation of a self-report instrument. The method involved the following steps: (1) forward translation by two bilingual language experts who translated the PHE-s from Italian into English; (2) forward translation by two bilingual language experts who translated the PHE-s from English to Mandarin Chinese; (3) experts' qualitative interviews in order to evaluate semantic and content equivalence, which included eight professionals with broad work experience in chronic care, health research, clinical psychology, and translating; (4) back translation by two additional bilingual experts who translated the Chinese version scale back into English; (5) back translation by two additional bilingual experts who translated the scale from English into Italian while the original author was invited to distinguish the back translation from the English and original versions; and (6) a pilot test among 27 patients with chronic diseases to check the readability and understandability of items and cognitive equivalence of the translation. The final version of the Chinese PHE-s (CPHE-s) was established by consensus and attached online as Appendix I.

Other Instruments

Patient Activation Measure—Short Form

The American short form of the Patient Activation Measure (PAM13) developed by Hibbard et al. (2005) is an interval-level, unidimensional, Guttman-like scale to provide an assessment of the potential or capacity for patients to be engaged in their health care from three aspects of disease self-management including patient knowledge, skills, and confidence. The response categories of the 13-item scale ranged from strongly disagree to strongly agree and "not applicable." The raw scores were converted from the continuous Rasch item response theory logit scale to activation scores between 0 and 100 with higher scores indicating greater patient activation. The scale has been widely used and has been shown to be reliable and validated in many different contexts (Maingal et al., 2009; Brenk-Franz et al., 2013; Brucki et al., 2014; Graffigna et al., 2015c; Moljord et al., 2015). In this study, we used the Mainland Chinese version of the PAM13 (PAM13-C) obtained by following the WHO guidelines for cross-cultural adaptation of instruments, and permission for use was obtained from Insignia Health, Inc. The Cronbach's alpha of the PAM13-C was 0.84, indicating good internal consistency among the items. A partial credit Rasch model was used to assess the item fit of the PAM13-C. The item statistics ranged from 0.81 to 1.25 for the infit mean square (MNSQ) and from 0.82 to 1.12 for the outfit MNSQ, suggesting that all the items are productive measurements (Linacre, 2011).

Morisky Medication Adherence Scale

The Morisky Medication Adherence Scale (MMAS-4) (Morisky et al., 1986) is a 4-item self-reported scale used to assess patients' medication-taking behavior. The MMAS-4 addressed the essential reasons for non-adherence including forgetting, carelessness, and stopping the drug when feeling better or worse. Response categories were yes and no for each item with a dichotomous response. Scores obtained from the MMAS-4 ranged from 0 to 4. Scores of 0, 1 to 2, and 3 to 4 were classified as high, medium, and low adherence, respectively. The Chinese version of the MMAS-4 was available, and the psychometric properties have been established, showing adequate internal consistency with a Cronbach's alpha coefficient of 0.76 for the total scale (Xu et al., 2007; Li et al., 2010).

Demographic and Clinical Variables

Participants were asked for information regarding age (continuous), gender, marital status, educational attainment, occupational status, type of diagnosis, and insurance type. Education attainment level was categorized as "primary school and below," "junior secondary school," "senior and specialized secondary school," and "college or higher."

Procedure

To enhance representativeness, a hierarchical sampling process was employed to recruit participants with chronic illnesses from community health centers in Hunan Province of China. Hunan Province was stratified to four geographic areas, and one city was randomly selected from each area. Data from the Hunan Health Ministry was used to compile a sampling frame of all community health centers in each of the four selected cities. All community health centers that agreed to participate in the study were provided with unique code numbers using SPSS 23.0, and then two community health centers were randomly selected as settings for recruiting participants. One hundred and ten participants were targeted in the two selected community centers in each of the four cities and recruited using a consecutive sampling method if they met the following selection criteria: (1) age > 18 years, (2) diagnosed with one or more chronic disease, and (3) following a chronic treatment for their diseases. Participants with cognitive impairment, uncontrolled psychiatric illness, or serious hearing impairment were excluded from the study. A total of 377 participants were recruited between November, 2015, and February, 2016, and completed the questionnaire; the sample frame and the detailed numbers of participants from each city are presented in **Figure 1**. Moreover, a sub-sample of 27 participants from one community health center in Changsha completed a test-retest evaluation of the CPHE-s through a telephone investigation after a 2-week interval.

ETHICS STATEMENT

Permissions were obtained from the authors and copyright owners of the original scale development research. The study was approved by the Ethical Committee of Xiang Ya Nursing School, Central South University in China. All participants received a short study description and were asked to give oral permission

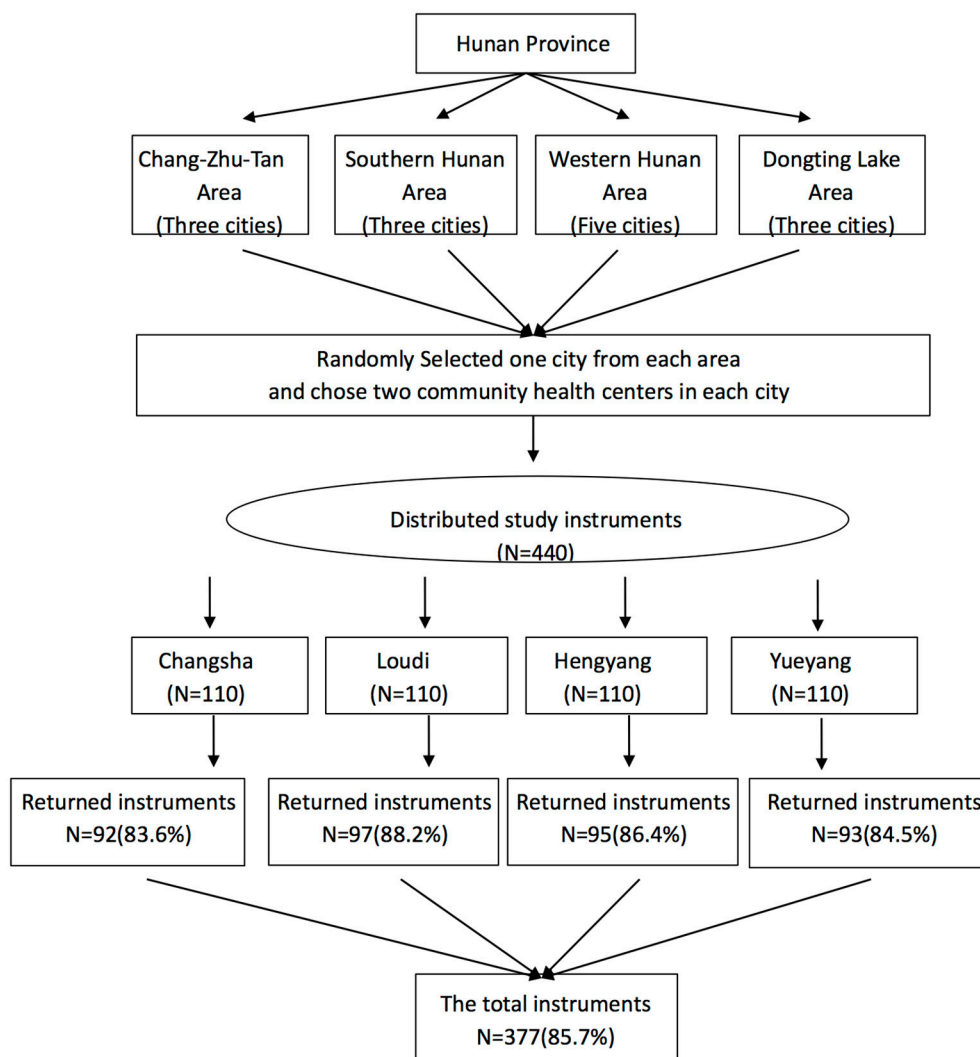


FIGURE 1 | Flow chart of sample frame and numbers of participants from each city.

to take part. Patients were also informed about anonymity and their right to withdraw from the study at any time without consequences. The results were analyzed at the group level and for scientific purposes exclusively. There are no vulnerable populations in this study.

DATA ANALYSIS

The data were entered by EpiData 3.02 and analyzed using IBM SPSS 23.0, Amos 23.0, and R 3.2.4. Appropriate descriptive statistics for ordinal data were used to summarize and present the data. The content validity was assessed through the application of the content validity index (CVI), and the item-level CVI (I-CVI) and Scale-level Index Average (S-CVI/Ave) were reported to compare the relevance of the translated Chinese version with the English version in this study (Polit and Beck, 2012). The partial credit Rasch model specifying additivity and uni-dimensionality

was employed to assess the item fit of the CPHE-s. The infit MNSQ statistic was used to assess the item fit. An infit value of an item falling between 0.5 and 1.5 indicates that the item is productive for the underlying measurement (Linacre, 2011). The internal consistency of the items of the CPHE-s was assessed using Ordinal Alpha via Empirical Copula Index (Bonanomi et al., 2012, 2015) and inter-item polychoric correlation given the ordinal nature of the items. A reliability index ≥ 0.7 , 0.8, or 0.9 can be interpreted as acceptable, good, or excellent, respectively (Gliem and Gliem, 2003). Test-retest reliability of the instrument was assessed using weighted kappa. Weighted kappa values can be interpreted as follows: ≤ 0.20 as poor, 0.21–0.40 as fair, 0.41–0.60 as moderate, 0.61–0.80 as substantial, and > 0.8 as almost perfect (Altman, 1991). Principal component analysis (CATPCA) was performed to identify the factor structure of the polytomous items of the CPHE-s. Confirmatory factor analysis (CFA) was performed to study the replicability of the factor structure obtained by CATPCA. The estimation method was

asymptotically distribution free, particularly suitable for ordinal data and not-Gaussian distributions. Measurement invariance by gender was also investigated. Finally, to assess the external validity of the CPHE-s, we evaluated the correlations between patient engagement (CPHE-s) and the PAM13 as well as medication adherence (MMAS-4) using SPSS 23.0 with Pearson's correlation coefficient. We expected high positive correlations between the CPHE-s and PAM13 as well as the MMAS-4. Significance level was set to $\alpha = 0.05$.

RESULTS

Translation and Adaptation

During the translation process, some modifications were necessary to address cultural differences between Western and Eastern countries. For example, the word “blackout” refers to a sudden status of power outages or flameout that is too abstract to understand for Chinese, especially elders with chronic disease. Therefore, we translated “I feel like I have blacked out” into “I feel my mind/brain goes blank.” Further, there is no obvious difference between “aware” and “conscious,” which both illustrate a status of attitude or cognition. “I have gained some understanding gradually” and “I know my disease/status” were used to distinguish two different intensities. In the current study, an expert panel including three clinicians experienced in chronic care or psychology from university teaching hospitals and two faculty members from university was asked to rate each item of the CPHE-s. The I-CVI was between 0.80 and 1.00, and the S-CVI/Ave was 0.92 in the final version of the CPHE-s. These results indicate acceptable content validity for the CPHE-s. More detailed information of the forward and back translations is presented in Appendix II.

Sample

Among 440 chronic patients who participated in the current study, 377 completed the survey for the psychometric analysis for an overall response rate of 85.7%. The most common reasons for refusal were lack of time and poor physical condition on the day of the survey. Demographic and clinic characteristics of the participants are summarized in **Table 1**. It was noted that, for these clinic diagnoses, the groups were not independent since most of the participants had more than one chronic disease. Additionally, 27 out of 30 participants from a randomly selected sub-sample completed the test–retest evaluation of the CPHE-s after a 2-week interval. Of those, 59.3% were women, and 40.7% were men with a mean age of 53.8 years ($SD = 11.0$) and a range of 34–78 years.

Responses to the Chinese Patient Health Engagement Scale (CPHE-s)

Since the Pearson correlation between the ordinal factorial score and median is very high (equal to 0.86), the median of the CPHE-s is considered to be a robust and efficient estimator of the real latent score of the construct—namely, the level of patient engagement. **Table 2** provides the item-level descriptive statistics for all items. Given the ordinal nature of the items, the median and the Shannon Entropy Index were calculated as tendency

TABLE 1 | Demographic and clinical characteristics of the sample ($n = 377$).

Characteristics	n (%)
AGE (YEARS)	
< 60	107 (28.4)
60–74	182 (48.3)
≥ 75	88 (23.3)
GENDER	
Male	172 (45.6)
Female	205 (54.4)
MARITAL STATUS	
Never married	1 (0.3)
Married	316 (83.8)
Divorced	6 (1.6)
Widowed	54 (14.3)
EDUCATION	
Primary school and below	122 (32.4)
Junior secondary school	124 (32.9)
Senior and specialized secondary school	80 (21.2)
College or higher	51 (10.75)
EMPLOYMENT	
Student	1 (0.3)
Unemployed	131 (34.7)
Retired	209 (55.4)
Employed	36 (9.5)
INSURANCE TYPE	
UEBMI	208 (55.2)
URBMI	122 (32.4)
NRCMS	45 (11.9)
Uninsured	2 (0.5)
CHRONIC DISEASES(%)	
Hypertension	271 (71.9)
Diabetes	110 (29.2)
Cerebrovascular disease	50 (13.3)
Cardiovascular disease	102 (27.1)
COPD	41 (10.9)
Cancer	15 (4.0)
Rheumatoid arthritis	9 (2.4)
Osteoarthritis	21 (5.6)
Osteoporosis	1 (0.3)
Thyroxine disorder	5 (1.3)
Uremia	4 (1.1)
Asthma	1 (0.3)
Hepatitis	6 (1.6)
Hypercholesterolemia	9 (2.4)
Depression	1 (0.3)

UEBMI, Urban employee basic medical insurance; URBMI, Urban residents' basic medical insurance; NRCMS, New rural cooperative medical system.

central and dispersion indices, respectively. Furthermore, there was no severe floor or ceiling effect for the summary score of the CPHE-s since only 17 (4.5) and 44 (11.7%) of the participants achieved the lowest and highest possible scores, respectively.

TABLE 2 | Item-level descriptive statistics for ranks on the CPHE-s.

CPHE-s item	Rank range	Minimum	Maximum	Median	Shannon entropy
Item 1	1–4	1	4	3	0.77
Item 2	1–4	1	4	3	0.77
Item 3	1–4	1	4	3	0.81
Item 4	1–4	1	4	3	0.84
Item 5	1–4	1	4	3	0.83

Item Analysis

A Partial Credit Rasch Model was implemented to further investigate whether the CPHE-s was uni-dimensional and whether all items fit the model well. Infit MNSQ statistic was computed to check whether the items fit the expected model. MNSQ determines how well each item contributes to defining a single underlying construct (uni-dimensionality). Infit is more sensitive to misfit responses to items closest to the person's ability level.

The logit measures and infit MNSQ statistics of the CPHE-s items are given in **Table 3**. The logit measures of the items ranged from 0.47 to 0.50, indicating that the item difficulties are not greatly varied. None of the items had infit MNSQ statistics < 0.5 or > 1.5, suggesting that all the items are productive for the underlying measurement (Linacre, 2011). The person separation index (PSI) was calculated to evaluate the reliability in the Rasch Model (PSI = 0.884).

RELIABILITY

Internal Consistency and Test-Retest Reliability

The CPHE-s had very good internal consistency since the value of the Ordinal Alpha via Empirical Copula was equal to 0.89. In **Table 4**, the Ordinal Alpha was evaluated after deleting individual items. Each item contributed significantly to the PHE-s score. The internal consistency of the CPHE-s was satisfactory. The Ordinal Alpha if item is deleted and weighted kappa coefficients (test-retest reliability) are shown in **Table 4**. The weighted Kappa coefficients of the items ranged from 0.52 to 0.79, suggesting that the strength of agreement of the test-retest values of the CPHE-s items could be interpreted as moderate to good (Altman, 1991).

Internal Structure Exploratory Analysis

The sample has been divided randomly into two sub-groups with an exclusive and exhaustive procedure: Group 1 ($n = 227$, 55.9% women, aged 36–90 years old; $M = 66.1$ years, $SD = 9.8$) for exploratory analysis and Group 2 ($n = 150$, 52% women, aged 26–87 years old; $M = 65.4$ years, $SD = 10.6$) for confirmatory analysis.

An exploratory categorical CATPCA was conducted on the final CPHE-s in Group 1 because of the ordinal nature of the items. An initial analysis was performed without any restriction on the number of metric factors to be estimated. The initial

TABLE 3 | Logit measures and mean square Infit statistics for the partial credit Rasch model of the CPHE-s.

CPHE-s	Logit measure (error)	Infit MNSQ
Item 1	0.11 (0.16)	1.43
Item 2	−0.47 (0.16)	1.22
Item 3	0.50 (0.16)	0.79
Item 4	−0.13 (0.16)	0.74
Item 5	0.00 (0.16)	0.77

TABLE 4 | Reliability indices for the CPHE-s.

CPHE-s item	Ordinal alpha if item deleted	Test-retest reliability weighted kappa
Item 1	0.88	0.74
Item 2	0.87	0.65
Item 3	0.86	0.52
Item 4	0.85	0.53
Item 5	0.85	0.79

analysis yielded one factor with eigenvalue 4.1, explaining 87.1% of the total variability. **Table 5** shows the factor loadings for the one solution of the CATPCA. All factor loadings had a very high value (>0.8).

All the conducted analyses (Rasch Model, Item fit analysis, PSI, Ordinal Alpha, eigenvalue, and explained variability of the first component of CATPCA) confirmed the uni-dimensionality of the scale. **Table 6** gives the inter-item polychoric correlation matrix of the CPHE-s. The average inter-item polychoric correlation is a subtype of internal consistency reliability. It is obtained by taking all the items on a test that probes the same construct, determining the polychoric correlation coefficient for each pair of items, and finally taking the average of all of these polychoric correlation coefficients. All inter-item polychoric correlations were higher than 0.7, indicating good inter-correlation between the items. The average inter-item polychoric correlation is equal to 0.84, indicating a high correlation between items.

Confirmatory Analysis

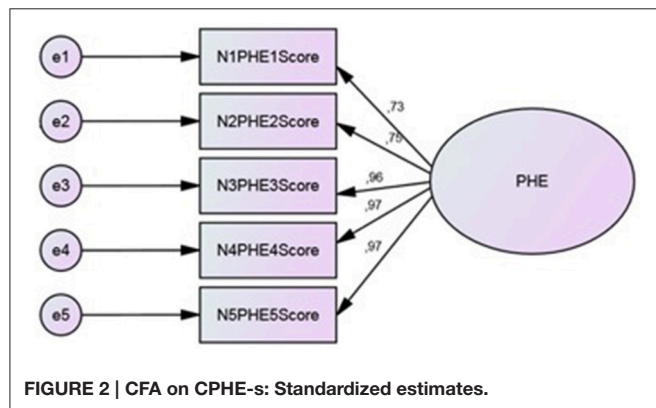
CFA (**Figure 2**) was performed on Group 2 to study the replicability of the factor structure obtained by CATPCA. The estimation method was asymptotically distribution free, particularly suitable for ordinal data and not-Gaussian distributions. To evaluate the closeness of the hypothetical model to the empirical data, multiple goodness-of-fit indexes were used, including the ratio of the chi-square to degrees of freedom (χ^2/df), the Comparative Fit Index (CFI), the Standardized Root Mean Square Residual (SRMR), the Goodness of Fit Index (GFI), and the Root Mean Square Error of Approximation (RMSEA). To test the model, each variable was allowed to load on only one factor, and one variable loading in the latent factor was fixed at 1.0. For the remaining factor loadings, residual variances were freely estimated.

TABLE 5 | Factor loadings from CATPA – One factor solution.

CPHE-s item	One factor solution
Item 1	0.74
Item 2	0.71
Item 3	0.84
Item 4	0.89
Item 5	0.88

TABLE 6 | Polychoric correlation matrix for the items of CPHE-s.

CPHE-s	Item 1	Item 2	Item 3	Item 4	Item 5
Item 1	–	0.94	0.79	0.75	0.72
Item 2		–	0.80	0.81	0.79
Item 3			–	0.93	0.90
Item 4				–	0.98
Item 5					–

**FIGURE 2 |** CFA on CPHE-s: Standardized estimates.

CFA showed reasonable goodness of fit indices. The fit indices met the criteria of fit for the hypothesized one-factor structure. Chi square ($\chi^2 = 6.65$, $df = 4$, $p = 0.156$) value and goodness of fit indices (CFI = 0.983, SRMR = 0.014, GFI = 0.979, RMSEA = 0.067) suggested that the model is coherent with the data. The analysis of modification indices did not suggest any relation between the error covariance of the items, avoiding overlapping problems.

To verify the validity and generalizability of the factor structure, a multi-group confirmatory analysis tested measurement invariance in the two subsamples divided by gender. **Table 7** shows the verified invariance hypothesis. The $\Delta\chi^2$ between the unconstrained and constrained models did not yield significant results. The factor structure was invariant by gender.

External Validity

The external validity of the CPHE-s was assessed by correlating the median score of the scale with the PAM-13 and MMAS-4 scores using Pearson's correlation coefficient. A moderate correlation was found between the CPHE-s and PAM-13 ($r =$

TABLE 7 | Multigroup CFA by gender.

Model	χ^2	Df	RMSEA	CFI	$\Delta\chi^2$ (df)	P
Unconstrained	6.6	4	0.059	0.971	–	–
Invariant factor loading	21.6	16	0.048	0.994	14.9 (12)	0.25

0.43, $p < 0.001$). However, there was no significant correlation between the CHPE-s and MMAS-4 ($r = -0.04$, $p = 0.464$).

DISCUSSION

This cross-sectional study provided the first report on the translation and validation of the PHE-s into the Chinese Mandarin language. The Rasch analysis presented a series of good infit values (ranging from 0.74 to 1.43) for each item of the CPHE-s. The data demonstrated good internal consistency of the PHE-s for patients with chronic disease through satisfactory Ordinal Alpha ($\alpha = 0.89$), which is higher than the original Italian Cronbach coefficient ($\alpha = 0.85$) (Graffigna et al., 2015a), and good test–retest analysis (the weighted Kappa coefficients of the items ranging from 0.52 to 0.79) (intra-class correlation coefficient = 0.68), which is lower than the value of the Italian measure (Graffigna et al., 2015a) but is still accepted. The exploratory categorical CATPCA and CFA suggest that the PHE-s belongs to a single-factor and uni-dimensionality scale.

To assess the external validity of the CPHE-s, the CPHE-s factor scores were first evaluated in relation to the PAM13 with strong psychometric properties as a golden standard, and the Pearson's correlation coefficient was 0.43, which is consistent with the Italian finding (Graffigna et al., 2015a). With some degree of conceptual overlapping, the terms “patient engagement” and “patient activation” are often used interchangeably. Patient activation contributed to describe the degree of patient engagement as an active agent in managing their own health, and higher levels of activation have been associated with greater patient engagement in health care (Carman et al., 2013; Hibbard and Greene, 2013; Graffigna et al., 2015a,b; Menichetti et al., 2016).

In contrast to the Italian data (Graffigna et al., 2015a), the Chinese results failed to support significant and negative associations between PHE and MMAS-4 scores. One plausible explanation for this finding stems from previous studies (Young et al., 2014; Awwad et al., 2015) that suggest that medicine adherence reports may tend to be skewed in favor of reporting higher adherence. Some responders might not disclose non-adherence as it might be deemed undesirable behavior, especially in the face-to-face investigation. Another explanation for this finding is that a four-item scale is not sufficient to represent the entire domain of the medication adherence construct (Morisky and DiMatteo, 2011). The eight items of the MMAS, which were shown to have higher reliability than the original four-item scale (Morisky et al., 2008), should be taken into consideration to further explore the correlation between patient health engagement level and medication adherence in future research.

Concerning potential shortcoming and limitations, first, this study is liable to recall bias like other studies based on self-reported measures, because it is hard to differentiate between patients who have actually addressed a high level of patient engagement and those reporting a high level of patient engagement for social desirability. Second, the relative heterogeneity of samples may be regarded as a weakness. Participants enrolled in community health centers represent a wide range of patients with chronic diseases, including patients with acute care, patients undergoing routine examinations, and patients with multi-morbidity. This might be negatively affected and lead to some errors in concurrent analysis like the relationship between patient health engagement and medicine-taking behaviors. Hence, further research is warranted to confirm the validation of the CPHE-s in a stratified manner representative of the Chinese chronic population.

In conclusion, our research adds to the accumulating evidence that the PHE-s has good validity and reliability in the context of Eastern culture. Healthcare practitioners can use it in primary care settings to better understand the patient engagement levels among patients taking part in their own health management. Differences in relevance between patient health engagement and medication-taking behavior require further investigation considering a revised MMAS-8 or other questionnaires and some objective indexes such as pharmacy refill records and pill counts (Santra, 2015).

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AUTHOR CONTRIBUTIONS

YZ was responsible for the literature search, analysis, data collection and coordination, interpretation of the data, drafting, writing, and revising the work. All authors contributed to the design (GG, SB, HF), data collection and coordination (PM, HF), analysis and interpretation of data (AB, KC), and/or writing and revising the work critically for important intellectual content (GG, AB, SB, HF). All authors read and approved the final manuscript and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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SUPPLEMENTARY MATERIAL

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The Mental Health Recovery Measure Can Be Used to Assess Aspects of Both Customer-Based and Service-Based Recovery in the Context of Severe Mental Illness

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Within clinical psychiatry, recovery from severe mental illness (SMI) has classically been defined according to symptoms and function (service-based recovery). However, service-users have argued that recovery should be defined as the process of overcoming mental illness, regaining self-control and establishing a meaningful life (customer-based recovery). Here, we aimed to compare customer-based and service-based recovery and clarify their differential relationship with other constructs, namely needs and quality of life. The study was conducted in 101 patients suffering from SMI, recruited from a rural community mental health setting in Portugal. Customer-based recovery and function-related service-based recovery were assessed, respectively, using a shortened version of the Mental Health Recovery Measure (MHRM-20) and the Global Assessment of Functioning score. The Camberwell Assessment of Need scale was used to objectively assess needs, while subjective quality of life was measured with the TL-30s scale. Using multiple linear regression models, we found that the Global Assessment of Functioning score was incrementally predictive of the MHRM-20 score, when added to a model including only clinical and demographic factors, and that this model was further incremented by the score for quality of life. However, in an alternate model using the Global Assessment of Functioning score as the dependent variable, while the MHRM-20 score contributed significantly to the model when added to clinical and demographic factors, the model was not incremented by the score for quality of life. These results suggest that, while a more global concept of recovery from SMI may be assessed using measures for service-based and customer-based recovery, the latter, namely the MHRM-20, also provides information about subjective well-being. Pending confirmation of these findings in other populations, this instrument could thus be useful for comprehensive assessment of recovery and subjective well-being in patients suffering from SMI.

Keywords: recovery, quality of life, needs assessment, schizophrenia, schizoaffective disorder

INTRODUCTION

Recovery is a concept that cuts across medicine, with particular importance in the context of chronic disease. Clinical definitions of recovery are generally related to reduction or remission of symptoms and return to pre-morbid or full levels of functioning. However, these definitions are variable according to disease or disorder and, frequently, consensus definitions are difficult to obtain. In the contexts of clinical psychiatry, mental health policy and psychiatric research, the concept of recovery from severe mental illness (SMI) has become increasingly relevant (Slade, 2010). This is particularly true for conditions such as schizophrenia, where recovery is very heterogeneous (Lieberman et al., 2008), since there are arguments that stigma and negative stereotyping are self-fulfilling attitudes directly resulting from a misconception of a very limited potential for recovery (Lieberman and Kopelowicz, 2005).

The conceptualization of recovery has been challenging for psychiatric disorders, in part because different groups use the term differently. Clinical psychiatry has traditionally defined recovery based on symptoms and several dimensions of function (service-based, objective, or clinical recovery – SBR), while consumer movements advocate for recovery to be defined as the process that involves overcoming mental illness, regaining self-control and establishing a meaningful and fulfilling life (customer-based, subjective, or personal recovery – CBR) (Schrang and Slade, 2007). In patients suffering from schizophrenia, recovery has many predicting factors, including socio-demographic variables, among others (Westermeyer and Harrow, 1984; Wieselgren et al., 1996). Specifically, age and functional status at onset, better cognitive functioning at stabilization, shorter duration of psychosis and early remission seem to best predict functional SBR (Robinson et al., 2004; Lambert et al., 2008). While there is less data for CBR, it has been proposed that subjective well-being at onset and marital satisfaction are associated with increased subjective recovery (Lambert et al., 2008; Tse et al., 2014). In any case, the concept of CBR has gained increasing relevance, given the movements toward promotion of patient-centered medicine and patient engagement in healthcare (Barello et al., 2012; Mullins et al., 2012; Domecq et al., 2014; Richards et al., 2015).

Unfortunately, lack of precision in the definition of these constructs and their dimensions has led to variable use of these terms within the literature. In fact, CBR has been directly or indirectly equated to other measures of subjective experience, such as quality-of-life (QoL) (Roe et al., 2011), and the degree to which CBR and SBR are separable constructs is not consensual (Resnick et al., 2004; Andresen et al., 2010; Lloyd et al., 2010; Roe et al., 2011, 2012; Norman et al., 2013; Stanhope et al., 2013). To address this question empirically, as we propose here, stringent conceptualizations of these recovery constructs have been considered by several authors. Specifically it has been proposed that the distinction between CBR and SBR should result from the methods according to which the two constructs and their respective measurement instruments are defined and derived (Campbell-Orde et al., 2005; Andresen et al., 2010). According to these conceptualizations, CBR is considered to be

recovery defined by users/patients and measured by instruments developed according to the accounts of users/patients (e.g., focus groups, qualitative analysis of patient interviews). SBR, on the other hand, is recovery defined by service providers and experts, and is measured using instruments developed according to the expertise of service providers and experts (Schrang and Slade, 2007). Nevertheless, this approach to distinguish recovery constructs is questionable, and there has been insufficient empirical work to support the distinction between the two.

Here, we set out to clarify the relationship between CBR and SBR constructs in a population of patients with SMI. Since there are no validated instruments for measurement of CBR in Portuguese patients with SMI, we initially translated and validated the Mental Health Recovery Measure (MHRM) (Bullock and Young, 2003; Young and Bullock, 2005) for use in this patient population. This instrument was chosen because it is one of only two self-rated measures of CBR according to the stringent definition presented above (Campbell-Orde et al., 2005; Andresen et al., 2010), i.e., it was developed according to the accounts of service-users. Furthermore, the MHRM has several versions with excellent psychometric properties (Bullock and Young, 2003; Young and Bullock, 2005; Chang et al., 2013; Armstrong et al., 2014) and has been successfully translated and validated into other languages (van Nieuwenhuizen et al., 2014). Once this instrument was validated, we proceeded to compare customer-based and service-based recovery and clarify their differential relationship with other constructs, namely needs and subjective QoL. The four constructs were assessed simultaneously using either clinician-reported (SBR and needs) and/or self-reported (CBR, needs and subjective QoL) measures.

MATERIALS AND METHODS

Participants

A sample of 101 users of a community mental health service (CMHS) at the Department of Psychiatry and Mental Health of Centro Hospitalar Cova da Beira (CHCB) in Covilhã, Portugal, was recruited. The ethics committee of CHCB approved the study and written informed consent was obtained from all participants, in accordance with the declaration of Helsinki. Individuals diagnosed with schizophrenia or schizoaffective disorder according to the 10th revision of the International Classification of Disorders (World Health Organization, 1993 – diagnostic codes F20 and F25, respectively), were eligible for enrolment and were identified by review of the institutional patient database, with diagnosis confirmed by chart review. Eligible individuals were contacted consecutively from January 2010 to December 2011, to schedule data collection. Exclusion criteria included illiteracy, presence of comorbid dementia or development disorder, acute exacerbation of positive psychotic symptoms, acute intoxication with alcohol or other substances or acute non-psychiatric disease. Patients no longer receiving regular clinical care at the CMHS were also excluded.

Mental Health Recovery Measure

To measure CBR, we used the MHRM, developed by Young and Bullock at the University of Toledo (Bullock and Young, 2003; Young and Bullock, 2005). The development of this self-report instrument was based on theoretical analysis of qualitative interview data regarding recovery-related experiences of persons with SMI, namely recurrent major depression, bipolar disorder, or schizophrenia spectrum disorders (Young and Ensing, 1999), followed by a series of revisions according to formal psychometric analyses (Young and Bullock, 2005). The current English version of the MHRM includes 30 items (MHRM-30), scored using a five point Likert scale that are added in a total score ranging from 0 (low recovery) to 120 (high recovery). The scale has eight domains: overcoming stuckness, self-empowerment, learning and self-redefinition, basic functioning, overall well being, new potentials, advocacy/enrichment and spirituality. The Flesch-Kincaid reading level is grade 7–8 and the administration time is of approximately 5 min. To develop a Portuguese version of this scale, translation, back-translation and adaptation of the MHRM-30 was performed by a team of three bilingual researchers, with expertise in clinical psychiatry, in order to obtain a final consensus version for application in a Portuguese population. Psychometric refinement of the original scale was performed according to results of an exploratory factor analysis (Young and Bullock, 2005) of MHRM-30 items with adequate item-total correlation, followed by item-reduction for domains with five or more items, following increasing order of item-domain correlations, until effects on Cronbach's α for that domain were no longer negligible or a minimum number of four items was reached. According to these methods, a smaller 20-item Portuguese version of the MHRM (MHRM-20) was obtained.

Other Evaluation Instruments

Quality-of-life was assessed using the TL-30S, a shortened version of the Lehman QoL scale which has been used extensively in individuals with SMI (Lehman, 1996). A subjective subscale score was calculated by adding scores from Likert scales for satisfaction in eight life domains (living situation, family, social relations, leisure, work, safety, finances, and physical health), as well as general life satisfaction, each rated from terrible (=1) to delighted (=7) (Dixon et al., 2007). The Camberwell Assessment of Need (CAN) assesses the clinical and social needs of people with SMI over the previous month, across 22 mental health and social domains (Phelan et al., 1995). Each domain is rated on a 3-point scale from the absence of need (=0) to the presence of an unmet need (=2), and a total score is calculated by adding the domain ratings. The CAN may be applied separately according to the perspective of the user, a clinical staff member or a caretaker (Phelan et al., 1995). The Global Assessment Functioning Score (GAF) was used as a clinician-rated measure of function-related service-based recovery. This is a global scale to measure psychological, social and occupational functioning on a hypothetical continuum ranging from 0 (maximal dysfunction from mental illness) to 100 (high functioning and health) (Jones et al., 1995), with evidence for concurrent validity to assess functioning in patients suffering

from schizophrenia (Startup et al., 2002). Portuguese translations of psychometric instruments, with extensive prior use in patients suffering from SMI (Gago, 1996; Fernandes et al., 2009; Brissos et al., 2012; Talina et al., 2013; Cardoso et al., 2016), were used.

Data Collection

A mental health nurse and a psychiatrist collected demographic, clinical and psychometric data for each patient on a single occasion. One-hundred-and-one individuals were recruited and evaluations were performed when users visited the outpatient clinic of the CMHS, or when the community mental health team visited their home. One team member oversaw the self-administration of the MHRM-30 and TL-30S scales while the alternate team member, thus blinded to the MHRM-30 and TL-30S responses, interviewed the participant to obtain clinical data, apply the CAN scale and assess the GAF score. In a convenience subsample of 40 participants, the MHRM-30 was applied again approximately 3 months later, to assess test-retest reliability. Participants in this subsample were slightly younger than the remainder of the sample (48 ± 14.9 vs. 55 ± 12.4 years, $p < 0.01$), but not significantly different regarding education or duration of disease ($p > 0.05$, t -tests, data not shown), nor regarding gender, marital status and substance abuse status ($p > 0.05$, χ^2 tests, data not shown).

Data Analysis

Data were analyzed using SAS software (version 9.3, SAS Institute, Cary, NC, USA). All continuous measurements were normally distributed according to skewness, kurtosis and comparison of mean and median. Sequential multiple linear regression models were used to test the association between MHRM-20 and GAF scores, when adjusting for other psychometric scores and for demographic variables. In these models, model assumptions were tested by analyses of the distribution of residuals and influence diagnostics were conducted using Cook's distance. Data transformations and polynomial models were used to test the better alternative to fit continuous predictors. Data for duration of disease were omitted from these models due to concerns about multicollinearity.

RESULTS

Demographic, clinical and psychometric data are summarized in **Table 1**. None of the patients invited for the study declined to participate. However, six participants did not complete the MHRM-30 scale, three of which also did not complete the TL-30S scale.

Psychometric Properties of the Portuguese Version of the MHRM

Regarding the full Portuguese translation of the MHRM (MHRM-30), while overall internal consistency was high (Cronbach's $\alpha = 0.9$), it was medium-low or low for several domains, specifically 'overcoming stuckness,' 'self-empowerment,' 'basic functioning,' and 'advocacy/enrichment' ($0.36 < \alpha < 0.65$). The remaining domains ('learning and

TABLE 1 | Description of the data collected for this study.

Variable ¹	% or Mean \pm SD	Range
Gender (% male)	76.2%	–
Marital status (% married ²)	27.7%	–
Substance abuse (% positive ³)	34.7%	–
Psychiatric home care (%)	12.9%	–
Age (years)	52.2 \pm 13.8	18–83
Education (years)	6.3 \pm 3.8	0–16
Duration of disease (years)	24.4 \pm 11.9	1–49
MHRM-30 (total score)	74.8 \pm 15.2	43–115
MHRM-20 (total score)	49.4 \pm 12.5	20–78
TL30S (subjective subscore)	45.7 \pm 7.7	27.5–60
CAN user (total score)	8 \pm 4.4	0–20
CAN staff (total score)	9.3 \pm 5.4	0–28
GAF (score)	49.6 \pm 19.4	10–90

¹Number of observations for all variables was 101, with the exception of MHRM ($n = 95$) and TL30S ($n = 98$). ²Percentage of individuals legally married and not separated, or cohabiting with a primary partner. ³Percentage of individuals that self-report abuse of alcohol or illicit substances. Range, Minimum and maximum values; mean \pm SD, Mean and standard deviation.

self-redefinition,’ ‘overall well-being,’ ‘new potentials,’ and ‘spirituality’) had adequate internal consistency ($0.70 < \alpha < 0.98$; Supplementary Table 1). Furthermore, eight items of the full scale had item-total correlations less than 0.40 (Supplementary Table 2). Thus, psychometric refinement of the full scale was performed according to an exploratory factor analysis of the 22 original MHRM items with adequate item-total correlation (Supplementary Table 3). The items were thus assigned to six novel domains, several of which corresponded, at least in part, to the domains in the original scale with higher α , and which were thus similarly named (‘empowerment,’ ‘redefinition,’ ‘identity,’ ‘social functioning,’ ‘overall well-being,’ and ‘optimism’). Item-reduction (see Materials and Methods) resulted in a 20-item revised MHRM scale (MHRM-20), consisting of six domains with adequate internal consistency ($\alpha > 0.71$; Supplementary Table 1). The revised scale had excellent internal consistency, overall ($\alpha = 0.92$) and for each of the six domains ($0.72 < \alpha < 0.83$), as well as adequate item-total correlations ($r \geq 0.40$; Supplementary Table 4). Test-retest reliability was assessed in a subsample of 40 participants, approximately 3 months later, according to Pearson’s r correlation coefficient, and found to be adequate (MHRM-20 total score: $r = 0.89$, $p < 0.0001$). Validity measures were also calculated and, as expected, were adequate (see below and Table 2). Because socio-demographic variables have been found to modulate the prognosis of schizophrenia (Westermeyer and Harrow, 1984; Wieselgren et al., 1996), discriminant validity was assessed across categories of age, education and disease duration, and found to be robust (Supplementary Table 5).

Relationship between CBR, SBR, Needs and Subjective QoL

As expected, the MHRM-20 score was positively correlated with QoL (TL30S-subjective) and GAF, and negatively correlated with

TABLE 2 | Correlations between scores on the MHRM scale and other psychometric instruments.

	MHRM-20	
	r	p
TL30S-subjective	0.49	<0.0001
CAN user	−0.58	<0.0001
CAN staff	−0.48	<0.0001
GAF	0.65	<0.0001

r , Pearson r correlation coefficient; p , p -value.

CAN scores. In absolute terms, correlations for the total MHRM-20 scores were lowest with CAN-staff and highest with GAF (Table 2). We had similar findings when using the scores for the MHRM-30 scale (data not shown).

The association between MHRM-20 and GAF scores was further assessed in sequential linear regression models (Tables 3 and 4), with either MHRM-20 (Table 3, models 1–3) or GAF (Table 4, models 4–6) as the dependent variable. Base models (models 1 and 4) included only demographic (age, gender, marital status, and education) and clinical variables (substance abuse). The base models were first incremented with either GAF (model 2) or MHRM-20 (model 5), and then with TL-30S-subjective (models 3 and 6). In base models, demographic and clinical variables explained only 15% of the variance of the MHRM-20 score (model 1, $R^2 = 0.15$), and 26% of the variance of the GAF score (model 4, $R^2 = 0.26$). When GAF score was added to model 1, the resulting model had a 26% increment in explaining variance of the MHRM-20 score (model 2, $R^2 = 0.41$). Similarly, when the MHRM-20 score was added to model 3, the resulting model had a 23% increment in explaining variance of the GAF score (model 5, $R^2 = 0.49$). However, while the explanatory potential of the MHRM-20 model (model 2) was further incremented by 10% when the TL-30S-subjective score was included in the model (model 3, $R^2 = 0.51$), when the TL-30S-subjective score was included in the GAF model (model 5), this had no impact (model 6, $R^2 = 0.49$). Indeed, while the adjusted association between the MHRM-20 and TL-30S-subjective scores was statistically significant ($\beta = 0.6 \pm 0.2$, $p < 0.0001$; model 3), that between the GAF and TL-30S-subjective score was not ($\beta = -0.06 \pm 0.2$, $p = 0.8$; model 6).

DISCUSSION

Here, we have contributed to clarify the construct of customer-based, subjective, or personal recovery (CBR), namely its relationship with another measure of recovery (service-based, objective, or clinical recovery – SBR) and an alternative measure of subjective experience (subjective QoL). Previous literature is not consensual regarding the dimensions within the complex construct of recovery. CBR is typically used to refer to personal experiential dimensions, including aspects such as empowerment and sense of hope (Schrack and Slade, 2007), and is frequently named subjective recovery. SBR is related to more clinical and social indicators, namely symptoms, functionality,

TABLE 3 | Sequential multiple linear regression models for the MHRM-20 score.

	Model 1 ($R^2 = 0.15$)		Model 2 ($R^2 = 0.41$)		Model 3 ($R^2 = 0.51$)	
	β (SE)	p	β (SE)	p	β (SE)	p
Age	−0.2 (0.1)	0.1	0.09 (0.1)	0.5	0.06 (0.1)	0.6
Male gender	5.7 (3.8)	0.1	5.3 (3.2)	0.1	4.3 (2.9)	0.1
Marital status	8.1 (3.4)	0.02	−0.3 (3.1)	0.9	−0.9 (2.9)	0.8
Education	0.96 (0.5)	0.06	0.8 (0.4)	0.06	0.7 (0.4)	0.07
Substance abuse	−3.5 (3.4)	0.3	−3.2 (2.8)	0.3	−2.5 (2.6)	0.3
GAF			0.5 (0.08)	<0.0001	0.4 (0.07)	<0.0001
TL30S-subjective					0.6 (0.2)	<0.0001

R^2 , R-squared statistic; β , Beta weight; SE, Standard error of β ; p , p -value.

TABLE 4 | Sequential multiple linear regression models for the GAF score.

	Model 4 ($R^2 = 0.26$)		Model 5 ($R^2 = 0.49$)		Model 6 ($R^2 = 0.49$)	
	β (SE)	p	β (SE)	p	β (SE)	p
Age	−0.6 (0.2)	<0.0001	−0.5 (0.1)	0.001	−0.5 (0.1)	0.001
Male gender	0.8 (4.3)	0.9	−2.8 (3.6)	0.4	−2.8 (3.6)	0.4
Marital status	16.6 (3.9)	<0.0001	11.5 (3.3)	0.001	11.5 (3.3)	0.001
Education	0.3 (0.6)	0.6	−0.3 (0.5)	0.5	−0.3 (0.5)	0.5
Substance abuse	−0.6 (3.8)	0.9	1.6 (3.2)	0.6	1.6 (3.2)	0.6
MHRM-revised			0.6 (0.1)	<0.0001	0.6 (0.1)	<0.0001
TL30S-subjective					−0.06 (0.2)	0.8

R^2 , R-squared statistic; β , Beta weight; SE, Standard error of β ; p , p -value.

participation in community and employment (Lloyd et al., 2010), and is generally equated to objective recovery. The use of such terminology has been misleading, with confusion between subjectivity and self-report resulting in constructs such as QoL to be considered as equivalent to CBR (Silverstein and Bellack, 2008; Lloyd et al., 2010; Roe et al., 2011). The results described here contribute to disambiguate these concepts. In fact, we found that the self-reported measure of CBR was correlated with two self-reported measures (subjective quality of life and a self-reported measure of needs – CAN-user), but was nevertheless better correlated to GAF, a clinician-rated measure of SBR. Linear regression models further confirmed the relationship between MHRM-20 and GAF, even when adjusting for demographic and clinical variables, while also showing that subjective QoL was related to MHRM-20 incrementally to GAF. The contrary, however, was not true, i.e., subjective QoL was not related to GAF incrementally to MHRM-20. Considered globally, these findings suggest that CBR, as evaluated by the MHRM-20 scale, could be used to measure global recovery, while also incorporating aspects of subjective QoL. Furthermore, these findings could contribute toward the reconciliation of clinical and consumer perspectives of recovery, as has been previously proposed (Davidson et al., 2005), while still demonstrating their complementary nature.

In previous empirical research, diverse findings have been reported relative to the relationship between CBR and SBR. Resnick et al. (2004) were the first to address this question. The authors found that, in a large sample of patients with

schizophrenia, severity of symptoms, as measured by a shortened version of the Symptom Check List 90, was negatively associated to several dimensions of what was described as ‘recovery as an attitude or life orientation,’ namely life satisfaction, hope and optimism, knowledge about mental illness and services, and empowerment. While this data was used to argue against the polarization of recovery perspectives, it must be noted that these authors did not use CBR measures developed strictly from the accounts of users/patients. However, subsequent research using the two measures of CBR meeting this more stringent definition, i.e., the MHRM-30 and the recovery assessment scale (RAS) (Campbell-Orde et al., 2005; Andresen et al., 2010), have mostly confirmed a negative correlation between CBR scores and symptom-based measures of SBR, namely the Kessler-10 scale (Andresen et al., 2010), the Colorado Symptom Index (Stanhope et al., 2013) and the Scale for Assessment of Negative Symptoms and Scale for Assessment of Positive Symptoms (Norman et al., 2013). Others, using the Brief Psychiatric Rating Scale as a symptom-based measures of SBR, have not replicated this association (Roe et al., 2012), or have replicated it only when restricting analyses to subgroups of patients, according to age at disease onset (Roe et al., 2011).

Research comparing CBR and function-based measures of SBR or needs has been less consensual, motivating the work performed here. Lloyd et al. (2010) found a negative correlation between scores in the RAS and the CAN Short Appraisal Schedule. The strength of this correlation ($r = -0.51$)

is similar to what, we found here between scores for the MHRM-20 and the CAN-user ($r = -0.54$) or the CAN-staff ($r = -0.45$). Andresen et al. (2010) found that MHRM-30 and GAF scores were positively correlated, but with a much weaker correlation than what we found ($r = 0.24$ vs. $r = 0.64$). Furthermore, in the latter and other studies (Andresen et al., 2010; Roe et al., 2011, 2012), an association between RAS and GAF scores was not found. However, as discussed by the respective authors (Roe et al., 2011), the patient populations where correlations between CBR and function-based SBR were weak or non-existent had particular characteristics. Specifically, patients were selected according to the presence of high support needs (Andresen et al., 2010), or were recruited at psychiatric rehabilitation residential centers (Roe et al., 2011, 2012). On the contrary, here and in the other study finding stronger correlations (Lloyd et al., 2010), patients were recruited from multiple clinical settings or a CMHS, and presumably were more representative of patients with SMI in general. Thus, we propose that our findings support prior research suggesting convergence between CBR and SBR (Andresen et al., 2010; Lloyd et al., 2010; Norman et al., 2013; Stanhope et al., 2013).

The convergence found, in this population, between CBR and function-based measures of SBR could be explained by different mechanisms. One possibility is that the distinction between CBR and function-based SBR is artificial, and that these two visions of recovery from SMI are actually reflections of a common and unitary construct. An alternate interpretation, however, is that CBR and SBR have common determinants, conditioning co-variation of their respective measures. One interesting possibility is that social factors, critical in the context of mental health (Fisher and Baum, 2010; Allen et al., 2014), determine aspects of both SBR and CBR. In fact, this possibility is partially supported by prior data, since there is evidence that social factors influence measures of CBR as well as GAF, even if to a lesser degree (Corrigan and Phelan, 2004; Hendryx et al., 2009). Several mechanisms are thought to underlie social influences on mental health, including social influence, self-esteem, sense of control, belonging, companionship, purpose and meaning, and perceived support availability (Thoits, 2011). Such proposed mechanisms are actually in line with many of the elements underlying the CBR construct, further supporting the connection between social factors and recovery. In fact, the hypothesis that common determinants, namely social factors, underlie the correlation between CBR and SBR, could explain why this finding is not common to all patient populations, as described above. In patients that have high support needs or reside at psychiatric rehabilitation centers (Andresen et al., 2010; Roe et al., 2011, 2012), it is conceivable that specificities of social characteristics, or interventions tailored to promote their modification, could modify the relationship between CBR and SBR. In any case, these hypotheses were not directly addressed with the work described here, and should be tested in the future.

The results of this study should be interpreted in the context of its cross-sectional experimental design. Thus, while the methods are adequate to query the relationship between the constructs of

interest, they do not allow for exploration of causal relationships between them. Longitudinal studies are needed to explore the role of customer-based recovery either as a prognostic factor or a measure of outcome. Furthermore, the use of the GAF score to assess functionality has been criticized in the past (Roy-Byrne et al., 1996). Nevertheless, the limitations of the GAF score are not consensual (Startup et al., 2002), and it is of frequent clinical use to assess functionality. Finally, comparisons of these results with those of previous studies are hindered by the fact that the patient sample described here has particular socio-demographic characteristics – patients are older, have low schooling and long disease duration. To the best of our knowledge, this is the first study on recovery from SMI conducted in a rural setting, which could explain such differences. However, the relationship between GAF and MHRM-20 does not seem to be dependent on the specificities of our sample, since it was mostly unchanged after adjustment for age, education level and duration of disease in linear regression models. Importantly, while the nature of the patient population in this study hinders the comparison with results from previous research, our findings also provide evidence, for the first time, that self reported measures of CBR can be used successfully in rural populations of patients with SMI, with low schooling and long disease duration.

CONCLUSION

Our results demonstrate that, in certain patients populations, constructs for recovery from SMI are convergent, suggesting that recovery can be assessed using tools developed based on the experiences of users (CBR) as well as the knowledge of mental health experts (SBR). Specifically, we found that the MHRM-20, an instrument for self-assessment of CBR, assesses SBR and QoL, in addition to CBR, in chronic and elderly patients in a rural community mental health setting. Thus, when considering the ease of application and scoring of the MHRM-20 scale, relative to measures depending on user interview, such as the CAN scale, or on clinical evaluation, such as the GAF score, this measure gains appeal as an inexpensive tool for broad use in CMHSs, and possibly even in e-health instruments (Graffigna et al., 2014). Furthermore, given its underlying customer-based philosophy and development, the use of the MHRM-20, or similar measures, as tools for evaluation or assessment of outcome, holds promise to stimulate and further develop collaborative patient-clinician environments (Barello et al., 2012; Mullins et al., 2012; Domecq et al., 2014; Richards et al., 2015), firmly rooted in a recovery-oriented model for mental health services. Under a patient engagement framework, promoting transition from a more authoritative to a more collaborative model of healthcare provision (Graffigna et al., 2015), we expect that active monitoring of clinical outcomes using self-rated and patient-developed tools, such as the MHRM, will potentiate patient participation, involvement and empowerment, hopefully leading toward strengthening of the therapeutic alliance between patients and caregivers, and ultimately improving outcomes of clinical care (Barello et al., 2014; Graffigna et al., 2015).

AUTHOR CONTRIBUTIONS

AO-M, CM, and JG conceived and designed research. CM and MP acquired data. AO-M and MC analyzed data. AO-M, CM, MC, and JG interpreted data. AO-M and MC drafted the work that was critically revised by CM, MP, and JG. All authors approved the version to be published.

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SUPPLEMENTARY MATERIAL

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Effects of Music Therapy on Drug Therapy of Adult Psychiatric Outpatients: A Pilot Randomized Controlled Study

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Objective: Framed in the patients' engagement perspective, the current study aims to determine the effects of group music therapy in addition to drug care in comparison with drug care in addition to other non-expressive group activities in the treatment of psychiatric outpatients.

Method: Participants ($n = 27$) with ICD-10 diagnoses of F20 (schizophrenia), F25 (schizoaffective disorders), F31 (bipolar affective disorder), F32 (depressive episode), and F60 (specific personality disorders) were randomized to receive group music therapy plus standard care (48 weekly sessions of 2 h) or standard care only. The clinical measures included dosages of neuroleptics, benzodiazepines, mood stabilizers, and antidepressants.

Results: The participants who received group music therapy demonstrated greater improvement in drug dosage with respect to neuroleptics than those who did not receive group music therapy. Antidepressants had an increment for both groups that was significant only for the control group. Benzodiazepines and mood stabilizers did not show any significant change in either group.

Conclusion: Group music therapy combined with standard drug care was effective for controlling neuroleptic drug dosages in adult psychiatric outpatients who received group music therapy. We discussed the likely applications of group music therapy in psychiatry and the possible contribution of music therapy in improving the psychopathological condition of adult outpatients. In addition, the implications for the patient-centered perspective were also discussed.

Keywords: music therapy, drug treatments, adult psychiatric outpatients, neuroleptics, antidepressants

INTRODUCTION

It is well known that music is connected to mood and that a certain piece of music can make people feel blessed, sad, lively, or relaxed (Biasutti, 2015a). The effects of music on a person's mindset and well-being are evident, and music has been used in various settings and conditions to control and improve health conditions (Biasutti and Concina, 2013). Music has been studied in several therapeutic settings for managing psychological conditions such as anxiety and stress, and there is strong scientific evidence supporting these positive effects. Music therapy treatments

are categorized as expressive therapies and non-verbal techniques for facilitating, expanding and shaping patients' expression and communication modes (Manarolo, 2005). The improvement of these skills provide patients with a comfort zone in which to express themselves while developing confidence and self-efficacy.

Music therapy treatment may help in shifting from a disease-centered perspective to a patient-centered perspective in the psychiatric setting and in determining new dynamics in the care relationship. Music therapy may facilitate a patient-centered perspective with an approach based on creativity and personal empowerment rather than external control of symptoms, as is the case with drug control.

A patients' engagement in healthcare is crucial and is widely recognized as a critical component of a high-quality health care system (Mead and Bower, 2000; Coulter and Ellins, 2007; Coulter, 2012; Graffigna et al., 2015). Several health care institutions at the forefront of policies support the promotion of a patient-centered perspective (Hibbard et al., 2004; Barello et al., 2012). The consumer-centered mental health framework could be used for finding alternative solutions to complement drug usage in order to improve the symptoms of patients and thus improve the participation and effective collaboration of patients in the management of their care (Lawn et al., 2007; Kreyenbuhl et al., 2009; Kukla et al., 2013).

There are several advantages for the use of music therapy in mental health care that can facilitate the shift from a disease-centered perspective to a patient-centered perspective. Compared with drug therapies, music therapy can help to improve patients' symptoms and quality of life in alternative and more holistic ways. This approach can lead to patients more effectively engaging in their care management, consequently resulting in improved health and well-being outcomes, including such effects as positive recovery attitudes (Loh et al., 2007; Patel et al., 2008; Livingston et al., 2013).

Background

The current study is focused on how a patient-centered perspective could be applied in psychiatry using music therapy as a rehabilitation technique. There are many studies in the literature that have focused on the effects of music therapy in such settings as psychiatry. Several reviews and meta-analyses have shown that music therapy, in combination with drug treatments, has significant effects on the positive and negative symptoms of psychosis, depression and the well-being of affected individuals (Maratos et al., 2008; Gold et al., 2009; Carr et al., 2013). The positive effects of music therapy have also been observed in the pathology of schizophrenia (Yang et al., 1998; Talwar et al., 2006). Jung and Newton (2009) performed a comparative analysis of the existing literature using the Cochrane database to explore the effects of 28 alternative therapies for schizophrenia, psychosis, and bipolar disorder. They noted that music therapy is one of the four most effective interventions in psychiatry. Other research has considered the effectiveness of music therapy on patients' quality of life and spirituality (Grocke et al., 2014), among schizophrenic in-patients needing acute care to reduce negative symptoms and improve interpersonal contact (Ulrich

et al., 2007), and for reducing patients' depression (Erkkilä et al., 2011).

The issue of the reduction of drug use by mental health patients as an effect of the outcomes of music therapy sessions (which is the subject of the current research) has been regarded with less interest by researchers, likely because of the difficulty of accessing such data and the complexity of data retrieval. Changes in drug treatments are often evaluated secondarily without being deemed a fundamental aspect of research. Chen et al. (2014) compared the drug dosages of patients in two groups (experimental and control) and found similarities in drug intake between the two groups. However, we argue that drug dosage should be considered a fundamental variable of research because it has a significant impact on disease management and because it involves a number of obvious advantages in an individual's treatment.

The literature also discusses how music therapy techniques can become complementary or adjuvant to drug therapy and how music therapy can provide psychological support with the consequent need to carefully consider the scientific evidence of the effectiveness of music therapy. In a meta-analysis of five studies, Maratos et al. (2008) compared the efficacy of music therapy to other therapies, such as drug and psychological support, and found that in four of the five studies analyzed, a greater reduction in symptoms of depression was reported among randomized patients after music therapy compared to those who received standard care. Similar results have also been highlighted by Erkkilä et al. (2011). Other studies have shown that in various types of patients, the therapeutic value of music can lead to a reduction of the anxiety state and a consequent decrease of drugs for the control of anxiety (Lepage et al., 2001). Lepage et al. (2001) found that the use of music resulted in decreased use of sedatives in interventions involving spinal anesthesia.

Few studies have examined changes in drug treatment as a result of music therapy interventions. Morgan et al. (2011) considered variations in drug treatment in patients between the ages of 17 and 55 years with a diagnosis of schizophrenia, schizoaffective disorder, and bipolar affective disorder with a framework of psychotic symptoms in the acute phase after participating in music therapy sessions. The patients were divided into two randomized groups: the experimental group, which participated in four sessions of individual active music therapy, and the control group, which participated in four sessions of music listening. Several scales have been proposed for patients before and after treatment, such as the Brief Psychiatric Rating Scale and the Calgary Interview Guide for Depression, and the average weekly patient dose of drugs, such as antipsychotics, mood stabilizers, benzodiazepines and antidepressants, has been monitored. There were no significant differences in drug dosage before and after music therapy treatments, whereas significant results were found using the Brief Psychiatric Rating Scale. The outcomes of this research are likely to be related to the short duration of music therapy treatment.

Literature Review Summary

The background analysis showed the effectiveness of music therapy treatments in psychiatry and suggested that music

therapy may provide a means of improving mental health among psychiatric patients (Yang et al., 1998; Talwar et al., 2006; Maratos et al., 2008; Gold et al., 2009; Jung and Newton, 2009; Carr et al., 2013). However, its effects on drug therapy have not been deeply explored, and no significant reduction in the drug treatment of patients following music therapy treatments has been shown (Morgan et al., 2011). The results of the research by Morgan et al. (2011) were likely influenced by the limited number of music therapy sessions (only four). Few studies have correlated the effects of music therapy with drug dosage in the treatment of psychiatric patients by considering these variables fundamental in the formulation of the experimental hypotheses. The need for better methodological control and conditions with a specific focus on the music therapy approach has also been noted.

Aim of the Study and Research Question

The current research aimed to fill the gaps of previous research by examining how prolonged group music therapy may affect drug assumption in adult psychiatric outpatients in a controlled trial. The research methodology adopted a quantitative approach using an experimental design that included an experimental group (MT) and a control group (CTR). The MT group participated in group music therapy sessions, whereas the CTR group experienced other non-expressive group activities (psycho-educational group activities on well-being). The research resulted in an analysis of drug dosages for all of the patients by consulting their medical records. The pharmacological treatments were classified according to the following four primary categories: neuroleptics, benzodiazepines, mood stabilizers, and antidepressant medications. The following research question was considered: does participation in group music therapy sessions affect drug dosage in adult outpatients with serious psychiatric problems?

MATERIALS AND METHODS

Participants

The sample consisted of 27 psychiatric outpatients ranging in age from 27 to 57 years (average age, 42 years) who were followed at an outpatient Mental Health Centre (MHC) in the northeast region of Italy. Participants were recruited by considering all the patients under the care of the MHC. The inclusion criteria required that the participants' primary diagnoses were F20 (schizophrenia), F25 (schizoaffective disorders), F31 (bipolar affective disorder), F32 (depressive episode), and F60 (specific personality disorders) according to the ICD-10 classification. These inclusion criteria were determined by the need to focus the research on psychiatric disease. Other types of diseases were not considered, such as drinking problems, post-traumatic stress disorder, and substance misuse disorder. Additionally, the outpatients were required to have pharmacological treatments in the following four primary drug categories: neuroleptics, benzodiazepines, mood stabilizers, and antidepressant medication. Fourteen participants were assigned to the MT group, and 13 were randomly assigned to the CTR group. Only the MT group participated in the group music

therapy activities, whereas the CTR group experienced other non-expressive group activities (psycho-educational group activities on well-being).

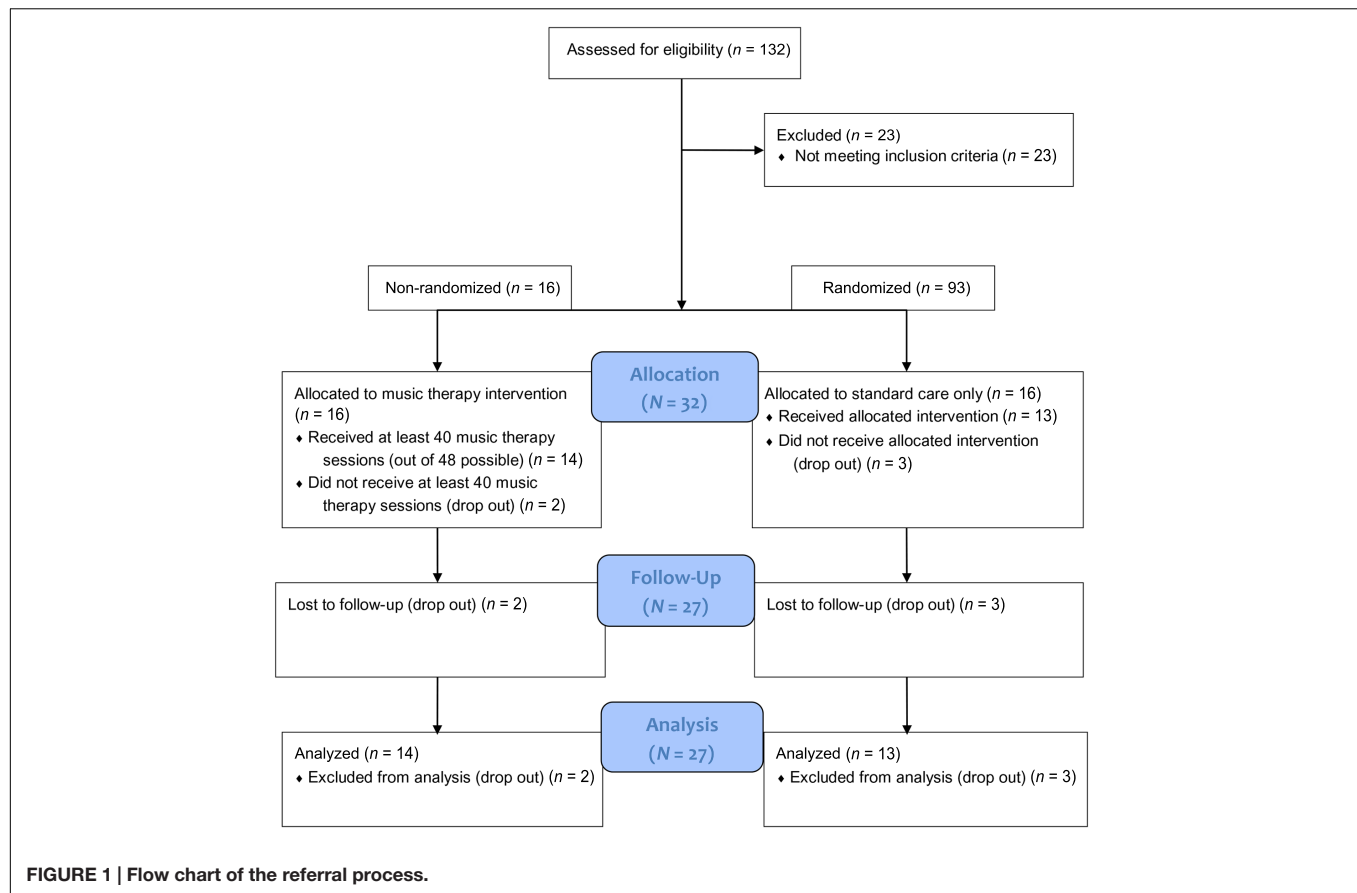
We assessed 132 participants for eligibility. Of this group, 23 were deleted because they did not meet the inclusion criteria. Of the remaining 109 participants ($132 - 23 = 109$), 16 participants were assigned to the music therapy activities. Of the remaining 93 subjects ($109 - 16 = 93$), a group of 16 participants was randomly extracted for the control group. The process of random assignment was performed by an independent collaborator who was not responsible for determining the eligibility of the patients. The random assignment was determined with a random number generator with the use of sequentially numbered, opaque, sealed envelopes (SNOSE). A flow chart of the referral process is presented in **Figure 1**.

The participants had been admitted to the MHC in a day-care community. Their medical records had been established for at least 6 years. They were not in an acute or subacute state, and they had no hospitalizations in the previous 6 months. The participants of the two groups had similar characteristics with regard to age, gender, prognostic factors, and psychiatric disease. The age of the 14 psychiatric outpatients of the MT group ranged from 29 to 57 years (average age of 43 years), and the group was composed of seven males and seven females. The age of the 13 psychiatric outpatients of the CTR group ranged from 27 to 55 years (average age of 41 years), and the group was composed of five males and eight females. A group comparison using the χ^2 test showed no significant differences between the MT group and the CTR group with regard to age, gender, and psychiatric disease.

The psychiatric participants were diagnosed with psychosis, bipolar disorder, and/or borderline personality disorder, and all displayed psychotic features. The patients involved in the study were considered serious with regard to psychosis. The participants were independent and autonomous. The majority of them lived with their family of origin. Of the participants, 63% were men, and 22% were married and living with their spouse, whereas only one was living in a community apartment. Regarding socio-cultural status, 14% had a university degree, and 37% had a high school diploma, and only 37% had a job.

Music Therapy Activities

The music therapy activities were conducted in group sessions of 2 h each on a weekly basis throughout 1 year for a total of 48 sessions. The music therapy activities were based on active techniques in which the participants were asked to play and improvise. Improvisation was based on the processes outlined by Biasutti and Frezza (2009) and Biasutti (2015b). The purpose of the group music therapy activities was to constitute a stable human therapeutic frame capable of supporting the relevant manifestations of suffering expressed by the outpatients. "The therapeutic process is based on the mutual construction of meaning of emerging thoughts, images, emotional content, and expressive qualities that often originate from the musical experience and are then conceptualized and further processed in the verbal domain" (Erkkilä et al., 2011, p. 134). The setting consisted of chairs arranged in a circle with the instruments



(e.g., djembe, darbuka, cymbals, wood blocks, rattles, bongos, and maracas) placed in the center.

The music therapy team, led by a music therapist, included a psychiatrist-psicotherapist, and a nurse. In addition to these professionals, a volunteer psychologist and a music therapist trainee attended the sessions. The large number of operators was necessary for the validity of the setting and is a constitutive feature of group music therapy activities (Manarolo, 2006). All of the members of the music therapy team participated in the group sessions and were involved in the musical productions. At the end of each session, a reflective discussion was scheduled in which the participants had the opportunity to express the feelings they experienced during the group music therapy session. Moreover, there was a monthly meeting of the music therapy team for sharing and discussing the progress of the sessions. The 14 participants in the MT group were divided into two groups of eight and six participants who had separate group sessions to ensure a lower dispersion and greater attention by the music therapy team. The two groups were conducted by the same music therapy team using the same music therapy methodology. These groups were designed for a maximum of 10 participants (Manarolo, 2006).

Procedure

The data were collected and the patients' medical records over a 1-year period were analyzed to extrapolate the historical

drug dosage for each patient. The data collection transpired over a sufficiently long period of time to appreciate the general course of drug therapy. Pre- and post-music therapy periods were distinguished. The pre-music therapy period was when the patients did not attend the music therapy sessions. This period was useful for collecting baseline data on drug dosage. The post-music therapy period was after the end of the music therapy sessions. The patients participated in an ongoing music therapy activity for a year, which is considered the minimum duration for detecting significant results of music therapy (Gold et al., 2009). Before starting and at the end of the activities, the dosages of the drugs taken by the patients on a daily basis were noted, and the total quantity of drugs consumed in a month was calculated. Medications were controlled by psychiatrists, who determined whether doses should be increased or decreased. Medications were controlled every 2 weeks, and this control was independent of the music therapy sessions offered. The psychiatrists who controlled the drug dosage of the participants were blind to the research procedures and to the participants' allocation, and they had no role in the research.

With regard to drugs, the participants were administered a variety of drugs summarized into 35 types, which are placed in the following four broad categories: neuroleptics, benzodiazepines, mood stabilizers, and antidepressants. To obtain standardization

of the data, the dosage was reported in Mg and was later converted into a percentage by dividing the daily dose into the maximum dosage per day. The mean daily dose for the long-acting depot drugs, which were taken every 2/4 weeks, was also calculated. Additionally, the adult outpatients participated in monthly individual monitoring visits in which their clinical picture was verified. The data from these visits were used to triangulate the results.

This study was conducted in accordance with the recommendations of the British Psychological Society with informed consent from all subjects. All subjects gave informed consent in accordance with the Declaration of Helsinki. For medical record consultations, the director of the MHC obtained the proper authorisation.

RESULTS

Methods for Data Analysis

The data collected included the changes over time of drug treatments for the four types of drugs during pre- and post-conditions. For data analysis, the statistical program SPSS 22 was used. The descriptive statistics were computed, and the Kolmogorov–Smirnov normality test was conducted to verify whether the distributions of the considered pharmacological groups were normal. The results showed that all of the categories met the criteria of normality, with the exception of mood stabilizers at $p < 0.05$. For this category, the non-parametric Friedman Test was performed, which allowed us to separately isolate the time effect within the groups. To calculate the other drug categories, a paired t -test was used to compare the MT and CTR groups in the pre- and post-conditions. In addition, a group comparison of the drug dosages at baseline was performed, and no group differences were found.

Drug Treatment Neuroleptics

Changes in the monthly dosage of neuroleptics for the MT group and the CTR group in the month before the commencement of music therapy and after the group music therapy intervention are shown in **Figure 2**. The two groups had different trends over time with regard to the doses of neuroleptics: the MT group decreased, whereas the CTR group increased. Paired t -tests showed that these changes were significant in both groups: the MT group with $t = 2.28$, $df = 13$, $p < 0.040$, and the CRT group with $t = -2.32$, $df = 12$, $p < 0.039$.

Benzodiazepines

Changes in the monthly dosage of benzodiazepines for the MT group and the CTR group in the month before the commencement of group music therapy and after the music therapy intervention are shown in **Figure 2**. There was no significant difference (paired t -test, $p > 0.05$) between the MT group and the CTR group. The benzodiazepines exhibited a similar trend in both groups, and the drug dosage appeared to be stable over time.

Mood stabilizers

Changes in the monthly dosage of mood stabilizers for the MT group and the CTR group in the month before the commencement of music therapy and after the group music therapy intervention are shown in **Figure 2**. The non-parametric Friedman test was computed for the mood stabilizers, which showed no significant differences in drug dosage over the two periods for the MT and CTR groups.

Antidepressants

Changes in the monthly dosage of antidepressants for the MT group and the CTR group in the month before the commencement of music therapy and after the group music

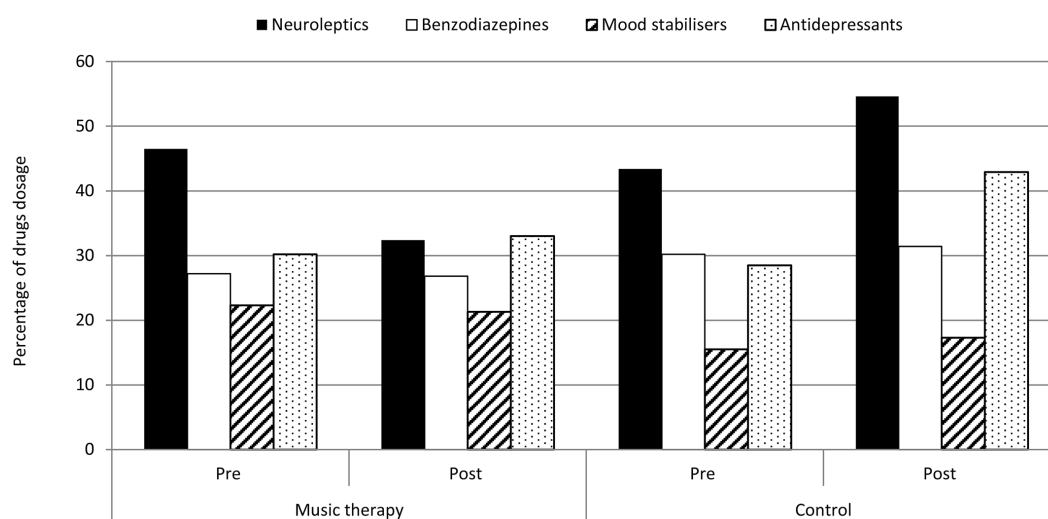


FIGURE 2 | Changes from baseline (PRE) to post-intervention (POST) in the dosage of the four drug categories for the MT group ($n = 14$) and the CTR group ($n = 13$).

therapy intervention are shown in **Figure 2**. The drug dosage displayed a similar trend in both groups, but the CTR group increased more than the MT group. This difference is evident in the paired *t*-tests, which showed a significant change only for CRT group with $t = -2.56$, $df = 12$, $p < 0.025$, whereas the MT group reported no significant difference (paired *t*-test, $p > 0.05$).

DISCUSSION

The present study evaluated the effects of a music therapy intervention on drug dosages in adult psychiatric outpatients. Doses of neuroleptics decreased significantly in the MT group and increased significantly in the CTR group. Doses of antidepressants did not change significantly in the MT group but increased significantly in the CTR group. In contrast, benzodiazepines and mood stabilizers did not demonstrate significant changes in either group. The adult outpatients participated in monthly individual monitoring visits in which their clinical picture was verified. The data from these visits showed an improvement or no worsening of their clinical picture during the group music therapy activities, confirming the results of the diminished drug dosage of neuroleptics. Neuroleptics are an essential drug therapy for psychiatric patients who display symptoms of psychosis, and a significant difference between the MT and the CTR groups emerged. The dosage in the MT group tended to decrease significantly, whereas the dosage in the CTR group increased significantly. The results provide evidence that a group music therapy treatment in combination with standard care, including pharmacological treatment, can affect the dose of neuroleptics (reducing or stabilizing their consumption) and can enhance the improvement of the clinical picture. The data in the present study are in contrast to the findings by Morgan et al. (2011), who reported no significant differences in the dosage of neuroleptics between the experimental and control groups. These differences likely relate to the different number of music therapy sessions in the study by Morgan et al. (2011) compared to the present research. A more extensive explanation is due to the nature of neuroleptics that are used in the treatment of schizophrenia and in diseases such as bipolar disorder, depressive episodes with psychotic onset, and personality disorders. These drugs act to reduce positive symptoms, such as hallucinations, deliria, aggressiveness, and excitement, and have less direct effects on negative symptoms, such as ideational impoverishment, autism, depression, and withdrawal into oneself. Active music therapy plays an important role in reducing the symptoms of psychosis, such as auditory hallucinations, deliria and psychomotor agitation, schizophrenia, and schizoaffective disorders, in patients suffering major depressive disorders. Research has analyzed patients with diagnoses similar to those who participated in this study. It could be argued that group music therapy may have induced the attenuation of positive symptoms as well as negative ones, favoring the likelihood of reduction in the daily dose of the drug.

Regarding benzodiazepines, no significant differences were found for the MT and CTR groups. This observation likely depends on the characteristics of the drug. Benzodiazepines are used as muscle relaxants and are effective for treating symptoms such as anxiety and agitation. They are used for anxiety disorders and depression as well as psychotic disorders, such as schizophrenia. The literature emphasizes that treatment using benzodiazepines should be cyclical; the dose cannot be increased for a period of time, and the drug is typically administered continuously for a relatively short period (Dell'osso and Lader, 2013).

The increase in antidepressant therapy corresponds to the reduction of secondary neuroleptic therapy and to the greater stability of the basis of the psychotic framework. This is a fact that might suggest a maturing capacity of introspection and insight in patients who are able to address greater awareness of the disease and related depressive experiences. This hypothesis could also be related to the induction of more anxious aspects and to the use of benzodiazepines.

With regard to mood stabilizers, the data did not respond to the normality criteria and were subjected to non-parametric tests that showed no significant differences for the MT and CTR groups. It is useful to remember that the changes in dosages for this type of drug are usually less "pointed" than the other categories evaluated because the mechanism of action requires a long time to achieve a therapeutic effect.

These results provide evidence that music therapy can stabilize or reduce the daily amount of drugs administered to psychiatric patients. This observation contrasts with the outcome of Morgan et al. (2011), who did not find significant differences between groups for mood stabilizers.

The other aspects that may have contributed to the positive results of this study include the number of sessions, the music therapy technique and the reflective discussion at the end of the session. The subjects attended 48 sessions within a year lasting 2 h each, which is a suitable number according to the literature that suggests that 16 to 51 sessions are adequate to obtain a positive result (Gold et al., 2009). The music therapy technique was an active approach (i.e., the patients were asked to actively participate in the music performance) and has been considered the most significant means for treating severely ill psychiatric patients (Manarolo, 2005). These methods are considered effective because patients have the opportunity to express feelings that they would not be able to explain in any other way. Another factor is the reflective discussion employed at the end of the music therapy sessions, which allows patients to think about the content that emerged during the sessions and share their thoughts with the group (Bruscia, 1998; Erkkilä et al., 2011). The discussion is a crucial aspect in which the effects of the sound-musical element allow the patient to express or verbalize latent content of which the patient is unaware. This phase, which is considered critical and therapeutic, is supported by the psychiatrist and the entire music therapy team who act as a container for the patient's emotional reactions.

CONCLUSION AND RESEARCH PERSPECTIVES

The results of this research have demonstrated the long-term effectiveness of group music therapy activities with adult outpatients with serious psychiatric issues. In this research, the most relevant analyzed drugs were neuroleptics and antidepressants. These drugs are more potent than other drugs, both in terms of their effects and their side effects. The remaining two types of drugs, mood stabilizers and benzodiazepines, have different characteristics and are not indicative of psychotic pathology. Psychotic drugs are crucial in the treatment course of a psychiatric condition; however, they have several side effects that reduce the patient's quality of life physically, such as tremors, fatigue, and drowsiness, and socially, such as flattened emotional and social spheres, decreased motivation in participating in daily activities, and a sense of isolation. Group music therapy was effective in controlling psychotic symptoms and successfully reducing the dosage of neuroleptics, thus indicating greater stability in the patient's clinical picture, which was a significant achievement. The music therapy intervention has the advantage of no side effects and may be beneficial to improving patients' quality of life (Grocke et al., 2014) by reducing the drug dosage.

The data presented in this study are encouraging considering that the patients' characteristics are similar to the clinical picture of severe psychiatric issues. For these patients, continuity in attendance at music therapy sessions is already a significant indicator of success because there was a sharp drop-out in participation for other activities proposed by the MHC of the current research. This research should be considered a pilot study with a limited number of patients. Several limitations could be discussed, such as the low sample size and the measurement of the psychophysical conditions of the outpatients. The data are significant and provide ideas for the continuation of research; however, the data should be validated with a larger group of subjects before generalization. In addition, a set of tools could be associated to measure the enhancement of the psychophysical conditions of the outpatients. The data could be verified in additional research with regard to whether these activities might induce persistent and meaningful effects in combination with drug treatments and how music therapy might limit drug usage. Other aspects that could be explored in future trials include a better understanding of the relationship between the patient and the therapeutic process in relation to individual diseases and the different contexts in which the patient is placed.

Implications for the Patient-Centered Perspective

The results of the current research could also be discussed in the framework of the patient-centered perspective considering two

main points: (1) the potential role of music therapy in providing care that is more holistic and aligned with a patient-centered perspective; and (2) the potential virtuous cycle of symptom reduction through music therapy.

Considering point one, the positive benefits of music therapy in improving symptoms and reducing drug usage in light of the introduction of alternative treatments should be considered. These treatments can better sustain the quality of life of patients and add to their resources for improving symptoms. Furthermore, the findings of the current research provided evidence of the importance of reducing drug usage through alternative methods, such as music therapy, in mental health care. This is an example of how to promote partnerships with patients as well as patient engagement in their care management (Lawn et al., 2007; Kreyenbuhl et al., 2009). In addition, the contribution of music therapy in providing a more holistic and broader approach to mental health care and consequently ensuring that mental health care supports a consumer-centered perspective could be discussed (Lawn et al., 2007; Kukla et al., 2013; Livingston et al., 2013). In this framework, mental disorders are considered not only as something isolated to treat but also as an expression of the individuality of the person (Loh et al., 2007; Patel et al., 2008). Moreover, a bio-psychosocial approach could be adopted, which proposes that symptoms should be treated considering the biological, psychological, and cultural backgrounds of each patient.

Regarding point two, the potential virtuous cycle of symptom reduction through music therapy could be highlighted. In this case, the reduction of drug use induces an improvement in patient engagement with an influence on the general well-being of the person. This cycle could induce a deeper engagement of patients in their care management and therefore exert an influence on such aspects as positive recovery attitudes, higher levels of hope, and fewer symptoms of emotional discomfort (Loh et al., 2007; Kreyenbuhl et al., 2009; Kukla et al., 2013; Livingston et al., 2013). In this framework, music therapy in addition to drug treatment can help in shifting from a disease-centered perspective to a patient-centered perspective in the psychiatric setting to establish new dynamics in the care relationship (Degli Stefani and Xodo, 2015).

AUTHOR CONTRIBUTIONS

MDS: Supervised the clinical setting, implemented the music therapy activities, and collected the data. MB: Revised critically the literature, analyzed the data, discussed the results, and wrote the whole paper.

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“PHE in Action”: Development and Modeling of an Intervention to Improve Patient Engagement among Older Adults

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The increasing prevalence of chronic conditions among older adults constitutes a major public health problem. Thus, changes in lifestyles are required to prevent secondary conditions and sustain good care practices. While patient engagement received great attention in the last years as key strategy to solve this issue, to date no interventions exist to sustain the engagement of older chronic patients toward their health management. This study describes the design, development, and optimization of *PHEinAction*, a theoretically-driven intervention program to increase patient engagement in older chronic populations and consequently to foster healthy changes that can help reduce risks of health problems. The development process followed the UK Medical Research Council’s (MRC) guidelines and involved selecting the theoretical base for the intervention, identifying the relevant evidence-based literature, and conducting exploratory research to qualitatively evaluate program’s feasibility, acceptability, and comprehension. The result was a user-endorsed intervention designed to improve older patients’ engagement in health management based on the theoretical framework of the Patient Health Engagement (PHE) model. The intervention program, which emerged from this process, consisted of 2 monthly face-to-face 1-h sessions delivered by a trained facilitator and one brief telephonic consultation, and aimed to facilitate a range of changes for patient engagement (e.g., motivation to change, health information seeking and use, emotional adjustment, health behaviors planning). *PHEinAction* is the first example of a theoretically-based patient engagement intervention designed for older chronic targets. The intervention program is based on psychological theory and evidence; it facilitates emotional, psychological, and behavioral processes to support patient engagement and lifestyle change and maintenance. It provides estimates of the extent to which it could help high-risk groups engage in effective health management and informs future trials.

Keywords: patient engagement, intervention development, older patients, chronic disease, patient activation

INTRODUCTION

Aging of the population is a major health challenge and a considerable concern for public health authorities and institutions (Ebrahim, 1997; Lutz et al., 2008). Older adults are likely to suffer from chronic diseases (Denton and Spencer, 2010), they often have multiple unmet health needs (Wolff et al., 2002; Chatterji et al., 2015), and they often have limited access to personal and contextual

resources needed to purposeful engage in accomplishing health goals (Shearer et al., 2009). It is difficult for health services to meet those needs, because of the lack of resources. Consequently, renewed models of care where patients are involved as main partners of their health management are needed (Anderson and Funnell, 2005; Thomson et al., 2005). Indeed, it is becoming ever more important for patients to be partners in care, not simply recipients of care as in older paradigms, because the actions people do—as well as do not—are critical for successful disease prevention and management (Mosen et al., 2007). The existing evidences suggest that patients who are partners in care have the potential to improve health outcomes through the adoption of health-enhancing behaviors and the reduction of health inequities (Coulter, 2005, 2012; Hibbard and Cunningham, 2008; Jordan et al., 2008; Cosgrove et al., 2013; Hibbard and Greene, 2013). This is particularly true among older patients who often suffer multiple disease conditions. Moreover, engaging patients in the health management can have a pivotal role in improving the effectiveness and efficiency of care (Holman and Lorig, 2004; Remmers et al., 2009). It can also improve client's satisfaction with the care process and the maintenance of an active role in society (Mosen et al., 2007; Kubina et al., 2013). This may not only contribute to the reduction of direct costs of the healthcare system, but also concur with the (re)orientation of economic resources in the management of healthcare systems to reduce costs (Fisher et al., 2009; Remmers et al., 2009; Greene and Hibbard, 2012; Hibbard and Greene, 2013).

Not surprisingly, the importance of engaging patients in their care has been gaining increased attention from clinicians, researchers, and policymakers alike (Simmons et al., 2014; Weil, 2016). Different labels have been adopted in the scientific literature to denote the process of making patients active stakeholders of their health management (i.e., patient activation, patient empowerment, patient engagement, and patient involvement; Barelo et al., 2014). Among these, the use of the term “patient engagement” has been showing an increasing trend, probably for its capacity to represent an “umbrella term” that encompasses different interconnected conceptualizations and labels (Barelo et al., 2016). With this term, which is taken from the marketing literature (Hardyman et al., 2015), the dynamic relationship between the patient (“the supply”) and the healthcare system (“the demand”) and its multi-level determinants (individual, relational, contextual, organizational) are highlighted (Graffigna et al., 2014a, 2015). As showed by Graffigna et al. (2014b, p. 87), the phenomenon of patient engagement is a “multi-dimensional psychosocial process resulting from the conjoint cognitive, emotional, and behavioral enactment of individuals toward their health condition and management.” According to this definition and to other authors' explanations of patient engagement (Hibbard et al., 2004; Gruman et al., 2010; Carman et al., 2013), three main dimensions featured the patient engagement process: behavioral (the concrete actions that patients do to manage their health condition), cognitive (the thoughts and information that patients have concerning their health condition), and emotional (the feelings and emotions that patients experience when adjusting to their new health condition) (Barelo et al., 2016). All these dimensions

help patients become experts in managing their health and care. According to this broad conceptualization and different qualitative studies on the care experiences of chronic patients, the Patient Health Engagement (PHE) Model has been developed (Graffigna et al., 2014a,b; Barelo and Graffigna, 2015; Barelo et al., 2015). According to this Model, after a diagnosis of chronic illness, people move through a series of phases that express different needs for engagement in care. In a blackout phase, patients feel unable to manage their health condition and are upset. Subsequently, they can experience an arousal phase in which they perceive anxiety and worry for their condition. In an adhesion phase, they learn to manage their health condition but have problems in adjusting their health habits to new life situations. Finally, in an eudemonic phase, they feel confident in autonomously managing their health conditions, they are optimistic about their future, and they perceive themselves as the main actors of their health and their life. A 5-items unidimensional validated self-report scale (PHE-Scale) has been recently validated strongly rooted in this model, showing the ability to detect these four main patient engagement phases along the care process (Graffigna et al., 2015).

However, the findings of a recent systematic review on patient engagement interventions revealed that although the link between patient engagement and improved health outcomes has been demonstrated, few interventions exist in the literature (Simmons et al., 2014). The few existing interventions target only some components and dimensions of the patient engagement process. This could limit the evidences of such interventions. Furthermore, few studies that aimed to engage patients rarely quantified and measured patient engagement (Simmons et al., 2014). Theoretical assumptions of those studies are often weak. Additionally, little research in this area has involved older people as the main target of the research (Wetzels et al., 2007). Besides the difficulties and specific needs that older patients may have to address in self-managing their health, specific solutions might be required for this population. Despite the considerable potential of patient engagement for older adults, this field remains underdeveloped (Kane and Kane, 2001; Elliott et al., 2016). To date, little is known about how to concretely engage older patients in their health management in a way that could be integrated in the clinical practice and tailored to the older patients' specific needs and goals.

This study reports on the design, development, and optimization processes of a new theory-driven intervention program aimed at improving the engagement of older chronic patients in care management. The intervention was based on the PHE Model, because of its broader view of the patient engagement process, and it proposed the operationalization of the PHE Model in practice based on three main research phases: literature review, experts and patients' opinion.

METHODS

Study Design

The Medical Research Council (MRC) Framework was used to assist with the development and optimization of an intervention program to improve the older chronic patients' engagement in

health management. This phased approach aimed to provide a robust methodological basis for the development and evaluation of complex interventions (Campbell et al., 2000, 2007; Craig et al., 2008). According to the MRC framework, prior to formal evaluation, dissemination, and monitoring of a new intervention, two main steps in the development and modeling need to be implemented to identify the theoretical base for the intervention, define contents and processes, structure the intervention, and model the procedures according to stakeholders' evaluations. In this study, this development and optimization process featured three main research phases, evidence exploration, experts' tune-up, and patients' fine-tuning. For every phase, activities, aims, and methods are described in **Table 1**. A small team of health psychologists responsible for the coordination and project implementation discussed the key findings of every research phase. The results of these discussions were used to set up and gradually refine the resulting intervention.

RESULTS

The findings from the research phases, so as details about the resulting intervention, are presented in the following sections.

Literature Review on Patient Engagement Techniques

The synthesized literature evidence demonstrated that interventions to engage chronic patients in their health and care management at the individual level (i.e., the patient) were generally scant, often poorly described in their components and delivery, and rarely of high methodological quality. Furthermore, they rarely targeted specifically older patients.

Despite these aspects, some key components and techniques adopted by the identified interventions to engage patients in their health and care management recurred. Thus, after selecting articles describing interventions for patient engagement, we analyzed and extracted the techniques used by the selected interventions. We thereafter summarized those techniques and synthesized them considering the three main domains of the patient engagement process:

- behavioral (the concrete actions that patients do to manage their health condition),
- cognitive (the thoughts and information that patients have concerning their health condition), and
- emotional (the feelings and emotions that patients experience when adjusting to their new health condition).

TABLE 1 | Methodological process for intervention development and modeling.

Phase	Activity	Aims	Methods
Phase 1: "Evidences exploration"	Systematic literature review	(i) To identify existing strategies, techniques and solutions for patient engagement (ii) To match literature findings with the PHE model components	- Scientific databases: Medline, PsychInfo, Scopus, Cochrane. - Search strategy: ("patient engagement" OR "patient activation") AND ("intervention" OR "trial" OR "program") - Inclusion criteria: (i) involving chronic patients, (ii) pre-/post-evaluations
Phase 2: "Tune-up with experts"	Experts' group discussion	(i) To revise, discuss and prioritize results of Phase 1 (ii) To optimize intervention's contents and procedures (iii) To collect experts' opinion about feasibility of the interventions' components	- Participants: 22 healthcare professionals caring for older patients trained in patient engagement theories, measures, and actions - Procedures: (i) presentation of literature results, (ii) group discussion to revise and optimize evidence-based results, (iii) <i>ad-hoc</i> questionnaire to evaluate feasibility, utility and adoption of an intervention for patient engagement - Data analysis: transcription of group discussion was thematically analyzed by members of the research team (JM, GG) to identify key issues within data (Braun and Clarke, 2006); quantitative data gathered through the evaluation sheets were synthesized with descriptive analyses by using the SPSS software 21.0.
Phase 3: "Fine tuning with patients"	Repeated qualitative semi-structured interviews	(i) To explore older patients' expectations and needs for engagement (ii) To collect patients' opinion about comprehensibility and acceptability of the intervention (iii) To collect patients' feedback to optimize the intervention	- Participants: 8 purposively selected patients >65 years-old affected by at least one chronic condition - Procedures: (i) preliminary qualitative semi-structured interview lasting about 30 minutes before the involvement in a prototype training, (ii) prototype training simulation (patients participated in a prototype version of the training), (iii) qualitative semi-structured interview lasting about 60 min just after the second session of the prototype training - Data analysis: Two researchers independently analyzed interviews by using audiotape transcripts of interviews. A thematic approach was adopted in order to synthesize main themes within data (Braun and Clarke, 2006).

Most interventions targeted the behavioral and/or cognitive domains. Consequently, positive psychology exercises (Seligman et al., 2005; Sin and Lyubomirsky, 2009; Schueller and Parks, 2012) and expressive writing tasks (Pennebaker and Beall, 1986; Rosenberg et al., 2002; Frisina et al., 2004) were also included within the emotional part of our intervention because they showed promising results in helping patients positively adjust to their illness experience. Furthermore, the main theories and models on which the programs were based and used to elucidate the role of cognitive, emotional, and social factors in health behavior were summarized. **Table 2** describes the resulting map of techniques reported in the scientific literature and used to deliver interventions for patient engagement.

Personalization of actions based on baselines assessment level was also adopted in some analyzed studies to ensure that actions were tailored to participants' experiences (Hibbard et al., 2004; Rise et al., 2016).

Finally, literature reported that even relatively short interventions could increase patient engagement, and suggested to track patient engagement and specific behavioral outcomes over time using validated measures to achieve positive health outcomes (Simmons et al., 2014).

Healthcare Professionals' Tune-Up

Overall, 22 healthcare professionals (e.g., nurses, physicians) who were experts in patient engagement theories and strategies participated in the prototype intervention's presentation and evaluation. These were community based practitioners caring for older chronic patients in a North-Italian hospital who were previously involved in a formative program on patient engagement theories, measures, and strategies. The researchers (JM, GG) recruited willing participants, most of whom were women (72%) with the mean age of 46 years old (range: 29–62), and with the mean number of years of experience of 26 years. Evidence-based literature on patient engagement strategies/techniques and suggested exercises based on summarized techniques were presented to experts. These

exercises were a prototype version of those ones described in the last section of the results and in **Table 7**. Thereafter, they were invited to discuss the presented input, to revise the proposed framework, to provide suggestions about possible ameliorations, and to identify possible advantages/barriers to the implementation of an intervention including the revised techniques. Subsequently, experts discussed a prototype of training's contents and procedures in small groups and finally evaluated the prototype intervention using a worksheet that was developed *ad-hoc* to address the following domains: easiness to use, relevance and utility, perceived competence, and willingness of adoption.

Generally, experts evaluated the proposed techniques and the framework of action positively. They particularly appreciated standardization and documentation of procedures and the possibility offered by techniques and proposed exercises to support the patient through simple and concrete steps and tasks. Thereafter, they provided suggestions and recommendations for the training's contents and procedures. The main themes that emerged from group discussion are summarized in **Table 3** with corresponding quotes.

Finally, on average, experts rated the proposed prototype intervention on a 5-point Likert scale as highly relevant and useful (ranging from moderately to extremely), and they reported high willingness to implement the training in their clinical practice (ranging from moderately to extremely). They generally evaluated the exercises as moderately easy to use (ranging from slightly to very) and felt moderately able to deliver them (ranging from slightly to very) (see **Figure 1**).

Listening Older Patients: Fine Tuning the Intervention

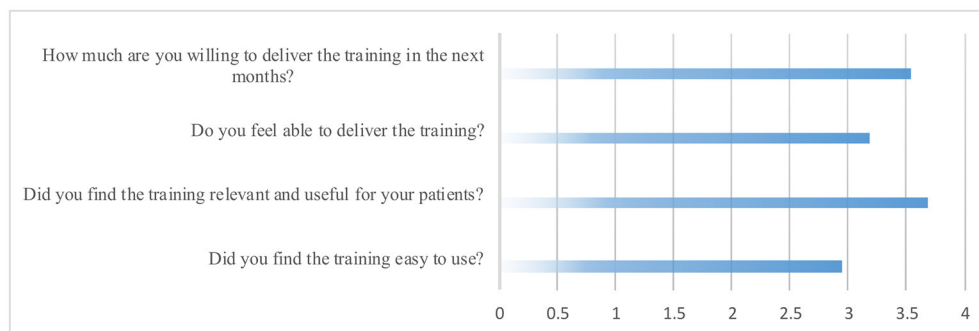
Eight participants older than 65 years accepted to participate in the "fine tuning" phase. Those patients were purposively and sequentially recruited by an external researcher through community senior centers, senior associations, or community medical centers. No exclusion criteria with the exception of age (>65 years old) or diagnosis (>1 chronic health condition) were applied. Most participants were women (72%), affected

TABLE 2 | Main techniques and theories emerged from literature considering the three PHE domains.

PHE domains	Techniques	Theories/Models
Behavioral	<ul style="list-style-type: none"> Goal setting and planning (Shively et al., 2005, 2013; Riegel et al., 2006; Kersten et al., 2015; Shah et al., 2015) Motivational interviewing (Anderson et al., 1995; Riegel et al., 2006; Linden et al., 2010; Benzo et al., 2012) 	Patient Activation Theory (Hibbard et al., 2004) Transtheoretical Model (Prochaska and DiClemente, 1986) Self-Regulation Theory (Leventhal, 1984)
Cognitive	<ul style="list-style-type: none"> Question-asking tasks (Hochhalter et al., 2010; Deen et al., 2011; Maranda et al., 2014; MacLachlan et al., 2016) Psycho-education sessions (Druss et al., 2010; Maindal et al., 2011; O'Leary et al., 2015; Krouse et al., 2016; Lara-Cabrera et al., 2016) Salutogenesis exercises to map external resources (Tan et al., 2016) Daily diaries for self-monitoring (Nagykaldi et al., 2012; Lee et al., 2013) 	Health Belief Model (Janz and Becker, 1984) Patient Activation Theory (Hibbard et al., 2004) Social Cognitive Theory (Bandura, 1986) Self-determination Theory (Deci and Ryan, 2008)
Emotional	<ul style="list-style-type: none"> Positive psychology exercises Expressive writing tasks Salutogenesis exercises to strengthen inner resources (Tan et al., 2016) Illness experience maps (Hall et al., 2015) 	Stress Coping Model (Lazarus and Folkman, 1984) Salutogenesis (Antonovsky, 1996) Positive Psychology (Seligman and Csikszentmihalyi, 2014)

TABLE 3 | Experts' recommendations main themes and quotes.

Main themes	Quotes
An evidence-based standardized guide to follow	<i>"All the proposed exercises are reported in literature and have a solid background, this helps"</i> <i>"It's easier to follow the program if exercises and procedures are well described"</i>
Working on multiple domains	<i>"I think that the most valuable aspect of the intervention could really be that it allows working on different aspects of the patient's experience"</i> <i>"I know that my patient has different need, and it's important to me to offer him actions for all his/her different needs"</i> <i>"It's true that patients need to be informed, but also to understand what is happening to them"</i>
Optimizing available resources	<i>"I usually do most of the things you described, but I do them without thinking...having a guide could help me better organizing my actions"</i> <i>"Most of the aids are already available to the patient, but he/she doesn't know that there are and how to use them"</i>
Supporting good communication exchanges	<i>"Some exercises are like a guide for our exchanges with the patient"</i> <i>"This can help having a guide for my communication with the patient...I already ask to my patient his story, but with some of these exercises I can have some practical tool"</i>
A tool to create bridges	<i>"Results of assessment and exercises can be used by other colleagues to continue the work with the patient"</i> <i>"Patients can feel accompanied by professionals also outside the hospital bridges, it can be a way to stay in touch with the patient"</i>
Motivating patients to change	<i>"the process of patient engagement could require a pre-existing degree of motivation on the individuals' part"</i> <i>"it's important to consider motivation of patients"</i>
Autonomy vs. presence	<i>"the illness experience map is useful if you use it in the first encounter with the patient"</i> <i>"I think that this intervention is easily suitable and implementable in clinical practice, but time and spaces are surely a potential barrier...I appreciate that some exercises can be autonomously managed by the patient"</i> <i>"Is useful to give to the patient some at-home exercises, it could be a way for the patient to bring patient engagement into the home walls"</i>
Supporting a patient-centered organizational culture	<i>"we are speaking about a cultural change"</i> <i>"we need an organizational structure supporting the introduction of a similar training"</i> <i>"looking only to the disease can be a barrier, professionals should be trained to support a patient-centered culture"</i>
Working in tandem with patients and caregivers	<i>"you know...most of my patients are older...some of them have a low educational level, live far from the hospital, or have impairing conditions...what about a training also for caregivers?"</i> <i>"I think it could be useful for our population of patients to train also caregivers"</i>

**FIGURE 1 | Healthcare professionals' evaluations responses.**

primarily by type 2 diabetes (57%). Most had an elementary education (57%), and indicated that they were married (71%). All of them were retired. Three participants were overweight, but none of them was a smoker. Almost all participants had multiple diseases and comorbidities, mainly with cardiovascular disorders. For further details about the characteristics of the sample, see **Table 4**.

Participants completed two face-to-face semi-structured qualitative interviews conducted in their homes just before and just after the involvement in a simulation of the prototype training (this was a second generation prototype version of the training modified based on the experts' revision of the primary generation prototype training) and participated in the presentation of the training's materials and

procedures (see **Table 5** for further details about the interviews' track).

The first interviews round aimed to elicit the needs and expectations for engagement among participants. All participants reported interest in participating in an intervention designed to engage them in their health management with different motivations (*"to become more calm, I'm too anxious when I manage my health," "to succeed in changing my lifestyle according to my health condition,"* and *"I'm really upset, I need to understand what it is happening to me and reorganize my life"*). They did not have particular expectations for a patient engagement intervention. Most of them (57%) preferred an individual intervention.

TABLE 4 | Characteristics of participants (n = 8).

	mean (SD)/ n (%)
Age	73 (4)
SEX	
Male	2 (25%)
Female	6 (75%)
Education (years)	8 (4)
DIAGNOSIS	
Type 2 diabetes	4 (50%)
Cardiovascular disease	2 (25%)
Chronic respiratory disease	1 (12,5%)
Inflammatory bowel disease	1 (12,5%)

TABLE 5 | Interviews' guide.

Area	Exemplificative questions
PRELIMINARY INTERVIEW	
Experiences and needs for engagement	<ul style="list-style-type: none"> • In your experience, what would help one in being more engaged in managing his/her health? What would help you in being more engaged? • Would you be interested in participating in initiatives aimed to foster your engagement in health management? Why would you engage in efforts to be more involved in your health management?
Expectations for a patient engagement intervention	<ul style="list-style-type: none"> • What features should have an intervention to engage you in managing your health? • Which contents and which way of delivery would you prefer?
SECONDARY INTERVIEW	
Patients' experiences about their participation in the intervention	<ul style="list-style-type: none"> • Could you describe me the main reasons which bring you in participating in the intervention? • Could you describe what happens in the sessions, in your own words? • How did you feel when you participate in the intervention? How did you feel before and after starting the sessions? • If you had to describe what the intervention means to you, what would you say? What images/metaphors come to your mind?
The intervention effects on the daily life and on health management	<ul style="list-style-type: none"> • In your opinion, how the intervention improved your engagement and attitude toward managing your health? How did it affect your daily life? • If you think about your way of managing your health, what aspects of the intervention have contributed to it? How? • If you think about your daily life and lifestyle, what aspects of the intervention have contributed to them? How?
Intervention satisfaction and feedbacks	<ul style="list-style-type: none"> • How would you rate and define your satisfaction toward the intervention? What aspects satisfied you more? What satisfied you less? Why? • What were the obstacles and difficulties? • What would you change or improve?

After this preliminary interview, patients were invited to participate in a simulation of the prototype training and to adopt the developed at-home exercises in their daily life. Patients were also asked to complete a battery of questionnaires before and after the prototype training to evaluate the potential compilation burden and the feasibility of measures. Participants were informed that the training would have been refining basing on their feedbacks and invited to point out possible difficulties with comprehension or other aspects of the program. One month after the preliminary interview and the involvement in the first prototype session, patients participated in the second prototype intervention session followed by a second round of interviews in which materials and procedures were discussed with patients.

The responses to the intervention were generally positive. Adherence to the home practice was high, as all participants used all of the provided instruments and engaged in a sustained effort to pursue their health goals. **Table 6** summarizes the main themes that emerged from this second round of interviews.

The average response for recommending the training to another person was high. In particular, participants indicated that they would suggest the intervention to just diagnosed people and to people who are less interested in managing their health, although those people were also perceived as difficult to engage in the intervention.

"PHEinAction": An Intervention for Older People Health Engagement

According to the main results of the previously described research phases, the prototype version of the training, which was

TABLE 6 | Main themes and quotes of patients' perception of the intervention.

Main themes	Quotes
A new perspective to the disease	<p><i>"I already knew all these things, but I kept them insight myself and gave no importance to them, with this intervention I eviscerate them and thus I faced them"</i> (Int. 1, F, 77 years-old)</p> <p><i>"It was important to me because I slowed down and I reflected on my situation, it was difficult but important and satisfactory"</i> (Int. 6, F, 68 years-old)</p> <p><i>"it is now for me like some light into the fog is appearing...and this light changes your perspective...it is like I'm realizing some things"</i> (Int. 2, F, 69 years-old)</p>
A stimulus to change	<p><i>"it was useful for me to manage my emotions and my anger...it helped me"</i> (Int. 5, F, 70 years-old)</p> <p><i>"I want to thank you because I never thought to be able to do something to better manage my health"</i> (Int. 2, F, 69 years-old)</p> <p><i>"it was like a flowered field with the sun...this pathway made me serene"</i> (Int. 1, F, 77 years-old)</p>
Clinicians have to play their part	<p><i>"doctors did not play their part"</i> (Int. 3, M, 76 years-old)</p> <p><i>"asking questions to my doctor is hard, I prepared myself for the doctor's visit but the encounter was brief and it ended on the hoof"</i> (Int. 2, F, 69 years-old)</p>
Improving repetitiveness and comprehensibility	<p><i>"sometimes home-works appeared repetitive and I would have benefitted by more concise exercises"</i> (Int. 6, F, 68 years-old)</p> <p><i>"I only completed the elementary degree so some words were difficult to me to understand"</i> (Int. 5, F, 70 years-old)</p>

outlined based on the PHE theory and literature analysis, was finally refined.

The resulting intervention was an individual training consisting of 2 monthly 1-h sessions, one brief telephonic consultation between the two sessions, and a set of instruments to be used by participants at home. Each encounter contributed to the promotion of manageability and meaningfulness disposition to manage health.

The first face-to-face session was used to (i) collect information on the patient's background through a patient's experience map, (ii) assess the starting phase of engagement of participants (i.e., blackout phase, arousal phase, adhesion phase, eudaimonic project phase) through a structured and validated questionnaire (The PHE Scale of Graffigna et al., 2015), (iii) define a purpose of engagement and manageable behaviors to sustain this purpose, and (iv) administer the instruments to support those purpose and behaviors based on the baseline PHE phase. Thus, for every phase of engagement, an individually tailored goal with subsequent emotional, informational, and behavioral actions was defined. Goals and behaviors defined in the individual sessions were based on the PHE phase of participants and were driven by PHE theory. The contract regarding the defined goals and behaviors was made with participants according to their particular needs and expectations for care. Furthermore, for every goal and actions, a set of instruments was developed to sustain the PHE process at home. Those instruments were developed based on the literature review of the existing strategies/techniques adopted to engage patients in their care management (see Table 2) and on experts and patients' feedbacks. Following the PHE theory, they covered three main areas of action (i.e., emotional adjustment, health information seeking and use, and health behavior change). For every area, instruments were personalized to each of the four PHE phases, yielding four packages of instruments that became increasingly challenging across phases. Indeed, as suggested by the literature and endorsed by experts, personalization of actions based on PHE phases and consequently on patients' needs and desires for care was considered a key aspect of the intervention. Four

different paths of training, with specific goals and consequent specific selection of exercises related to each area of action, were thus featured to enhance flexibility and personalization of the intervention (see Figure 2 for further details about the main goals of the four training's paths based on the baseline PHE phase of participants). During the first session, a personalized PHE plan was thus defined with home-based exercises. Participants were invited to follow their plan in the next month and to actively adopt instruments of their plan to reach their engagement goal. Table 7 provides further details about the instruments' aims and contents.

A telephonic consultation was conducted 2 weeks after the first encounter to maintain motivation of participants and discuss potential difficulties.

Finally, a second face-to-face session was used to (i) collect the experience of participants and discuss the adopted instruments, (ii) re-assess the PHE phase of participants through the PHE-Scale, (iii) provide feedbacks and reinforce improvements, and (iv) define a new engagement goal with related actions and instruments.

Figure 3 provides further details about the structure and the sessions' goals of the final intervention.

These two sessions were conceived to be the minimal unit of action for the patient engagement change, and further "units of actions" were suggested in critical points of the care process to sustain the change process.

DISCUSSION

This paper describes the process of the development and refinement of a theory-based individual training aimed to engage older chronically ill patients in their health and care management. To our knowledge, this is the first intervention that aims to sustain health engagement among older chronic population. The development process allowed to define, refine, and optimize the contents of the training. Overall, patients and healthcare professionals provided positive feedbacks for the

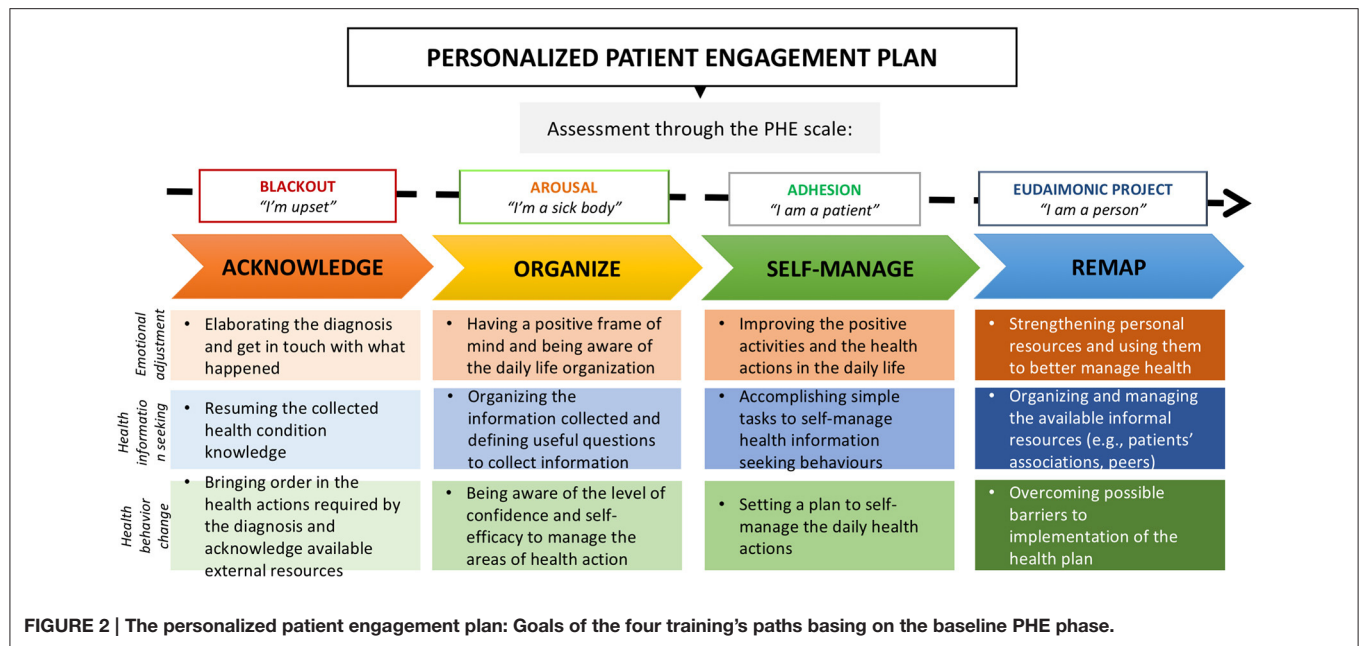


TABLE 7 | "PHEinAction" home-based instruments' key components, aims and procedures.

Instruments	Aims	Exercises
Instrument 1. Emotional adjustment	<ul style="list-style-type: none"> To foster a process of adjustment to the diagnosis and to the patient's role To activate and strengthen skills and inner resources of the patient 	1) Expressive writing exercise on the illness experience; 2) Daily diary with small positive thinking tasks; 3) Map of wellbeing/discomfort areas in the daily life and strengthening exercise of wellbeing areas; 4) Positive psychology exercise to identify 3 personal strengths and apply them to better manage health
Instrument 2. Health information seeking and use	<ul style="list-style-type: none"> To improve health information seeking/use processes To sustain the adoption of external resources 	1) Disease knowledge elicitation exercise; 2) Question-asking exercise; 3) Prompts to self-manage information-seeking behaviors; 4) Map of adopted informal information channels (e.g., internet, peers, books...)
Instrument 3. Health behavior change	<ul style="list-style-type: none"> To sustain the plan and organization of health behaviors To improve self-efficacy in managing health 	1) Map of areas of action that patient needs to manage (diet, physical activity, medications...) and of informal resources supporting the management of these areas; 2) Self-evaluation exercise concerning self-efficacy level for every area of action that patients need to manage and identification of reasons for self-evaluation; 3) Behavioral plan to activate health actions; 4) Imagination exercise of possible barriers getting in the way for the plan and of solutions to handle these barriers

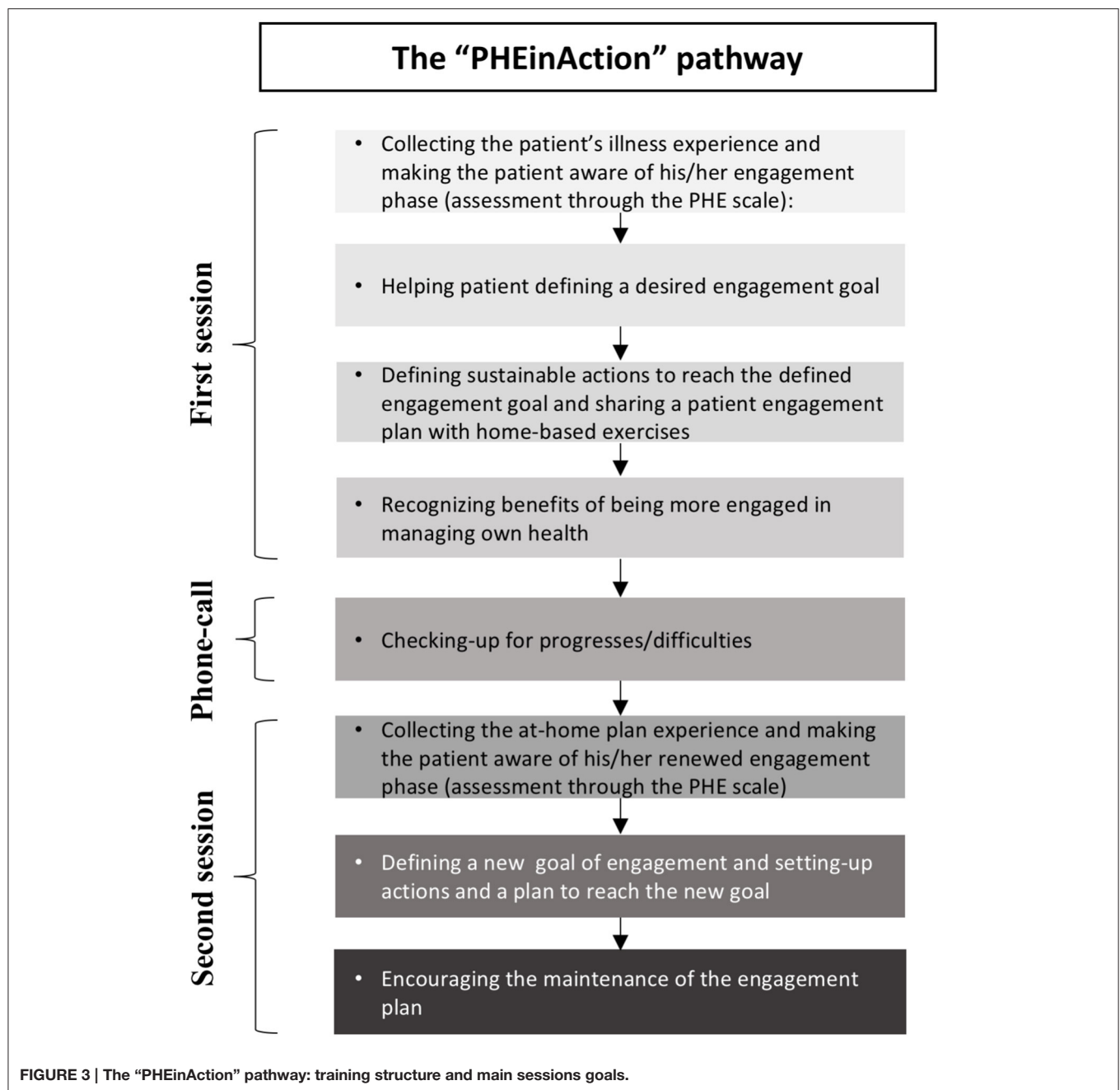
training contents and procedures. The training still requires formal evaluation.

The development process highlighted some key points that to our opinion need to be discussed.

First, although the training was initially developed for patients affected by different chronic conditions, and no differences in the fruition of the intervention among the different clinical conditions were found, it might need to be adjusted to specific chronic conditions to enhance potential benefits of the training. Even more, the results revealed that the few months following the

diagnosis can represent an optimal window of action to deliver the training. This is supported by the literature suggesting that the period just after diagnosis is particularly important to allow a process of diagnosis adjustment to be started (de Ridder et al., 2008). This should be tested by further studies and evaluations.

Second, experts envisaged families and caregivers as crucial to sustain patient engagement, especially in situations in which patients are physically compromised. Probably, a training could be specifically developed to engage also caregivers and families in the care process to better sustain patient engagement. As



highlighted in the literature, giving support, and engaging families and caregivers could help ensure high-quality care at home (Wellard and Street, 1999; Donelan et al., 2002), thus strengthening the engagement of patients and supporting them when directly engaging them is hindered by physical or contextual barriers.

Third, clinicians were described by patients as a potential barrier to change in patient engagement. Complex programs that train clinicians to embrace requests of patients for engagement and, even more, enable patients to become partners in their care management could make patient engagement more effective.

The role of clinicians in advancing the patient engagement have been increasingly emphasized, as they play a crucial role in guiding patients on their care journey (Greene and Yedidia, 2005; Killaspy et al., 2015).

Finally, it is important to consider that this study aimed to report the development and refinement process of a new intervention, rather than to quantitatively evaluate its effect on validated measures. More data are needed to demonstrate the effectiveness of the intervention, especially in the long term, and to drive conclusions. The limited sample size (and the broadness of the inclusion criteria) did not allow us to

make conclusions on the efficacy of the program. Patients' views expressed in this study were from a particular sample, mainly woman, and it was difficult to retrieve recently diagnosed participants. Furthermore, patients' feedback recorded in this study was qualitative in nature, and as such, it needs to be complemented by quantitative evaluations. The intervention needs to be further evaluated using a larger sample of males and females with different degree of engagement at baseline and including also recently diagnosed patients. It would be advisable to test the feasibility of the intervention in more homogeneous populations. Furthermore,—although this was not the primary aim of this study—preliminary results collected through the PHE Scale suggested few changes in the engagement scores of patients enrolled in the intervention. In particular, only patients with lower levels of engagement at baseline improved their scores after the training. Participants reporting high levels of engagement at baseline (equal to or greater than the adhesion phase measured through the PHE scale) generally maintained their baseline scores after the training. This is consistent with other studies showing that particularly less engaged patients might benefit from activation interventions (Deen et al., 2011). A more systematic effectiveness study is needed to explore the stability of these first preliminary results. Additionally, changes in engagement scores might need more time to be detected, and follow-up evaluations could be particularly relevant when conducting a similar training.

To conclude, this study described the development process and optimization of a new individual intervention program to engage older chronic patients in their care management. The study utilized a step-wise structured approach to develop

complex interventions (MRC) and a theoretical model based on qualitative studies and grounded on the specific needs of the target group. Indeed, grounding health interventions on qualitatively-based theories and adjusting them to the specific needs and context of final users can help deliver ecological studies. The intervention components, developed and evaluated by experts and older patients, were considered feasible and acceptable as well as useful and easily implementable in clinical practice. Some suggestions for changes in health actions and attitudes after the training were also envisaged. Further work is needed to improve and adapt the intervention components and tackle issues related to their delivery and implementation within healthcare professionals' existing clinical practice.

AUTHOR CONTRIBUTIONS

GG and JM designed the study and developed the methodology. JM collected the data, performed the analysis, and wrote the manuscript. GG supervised the data collection and analysis process, and revised the manuscript.

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Engaging Elderly Breast Cancer Patients: The Potential of eHealth Interventions

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Breast cancer is the most common cancer in women in the world and age is the strongest risk factor for breast cancer in women. Incidence of breast cancer still increases even if at a slower rate in women aged over 50 years until age of 80 years (DeSantis et al., 2011). Actually the incidence of breast cancer represents a critical health concern in the growing aging population and requires specific evidence-based recommendations (Petrakis and Paraskakis, 2010; Biganzoli et al., 2012). From a psychological point of view, some authors found that older breast cancer patients have more difficulty in adjusting to breast cancer and related treatments than younger women and these differences resulted from several age-related factors (Park et al., 2011).

Among several treatment options, chemotherapy treatment is experienced as distressing and traumatizing and frequently considered as emblematic of the treatment and of cancer itself (Richer and Ezer, 2002), also for postmenopausal women (Browall et al., 2006). One of the worst experiences associated with the chemotherapy treatment is that of losing hair. This feeling is frequently ranked among the first three important side effects for breast cancer patients, together with nausea and fatigue (Lindop and Cannon, 2001; Carelle et al., 2002). Some studies suggest that side effects are experienced as less distressing as patients can anticipate them (Golant et al., 2003; Frith et al., 2007). This preparation constitutes a form of anticipatory coping—coping which involves the preparation for managing the stressful consequences of an upcoming event, which is likely or certain to occur (Aspinwall and Taylor, 1997).

Therefore, sustaining the engagement of breast cancer patients at different ages represents an important aim. Specifically, within the whole process of patient engagement that is composed of four incremental and evolutionary phases (Graffigna et al., 2013a,b), the period before the initiation of adjuvant chemotherapy can be seen as the second phase of the breast cancer patient engagement process (*arousal phase*). On the one hand, the diagnosis is still a recent event (*blackout phase*) and the management of the emotional reactions is still difficult. On the other hand, to increase knowledge about the treatment and its related side effects and develop strategies to cope with anxiety (*adhesion phase*) represents an important challenge that contributes in maintaining future outcomes of the cancer treatment (Su et al., 2005) and in recapturing a positive life planning oriented to the future.

Age-appropriate patient education interventions might need to be designed and realized to prepare older women for the social, physical, function and treatment-related effects of breast cancer and thus reducing their anxiety and increasing their control over the situation (Treacy and Mayer, 2000; Seçkin, 2011). eHealth interventions allow to develop integrated, sustainable and patient-centered services, to promote and enhance health and to augment the efficacy and efficiency of the process of healthcare (Eysenbach, 2001; Graffigna et al., 2014; Barelo et al., 2016). As investigated by Fogel et al. (2002), age, length of time since diagnosis, and breast cancer stage are unrelated to

Internet use and an increasing number of patients of any age are accessing health information on the Internet (Seçkin, 2011).

Offering Web support as part of regular care can be a powerful tool to help breast cancer patients manage their illness (Ventura et al., 2013). Recently, several studies showed the efficacy of Web-based illness management systems, containing components for symptom monitoring, tailored information and self-management support, compared to usual care (Børøsund et al., 2014) in promoting emotional processing (Baker et al., 2011) and reducing depression and anxiety levels (Yun et al., 2012). A recent review suggests a positive relationship between the use of Internet - or interactive computer-based education program - and the knowledge of breast cancer patients; this relationship also has a positive effect on patient satisfaction (Ryhänen et al., 2010). However, Internet educational programs available for breast cancer patients are still rare and mostly focused on increasing patients' knowledge, focusing more on "basic details" related to the disease and information about procedures rather than on diagnosis, treatment, recovery, and quality of life (Warren et al., 2014).

Among the few eHealth interventions aimed to improve the women well-being, it is possible to describe different approaches. One includes the use of personal websites to improve the emotional wellbeing of breast cancer women by helping them to construct a narrative of their experience, express emotions, and receive the social support they need, particularly from friends and extended family (Harris et al., 2015). A second is aimed to enhance social support through online peer support interventions, and recently older women reported that they receive more benefits from using online support groups especially in regard to feeling in control of their health and feeling less distressed than younger women (Seçkin, 2011). A third proposes coping skills trainings aimed to take under control patients' affective state. With this aim, Owen et al. (2005) employed a randomized controlled design to pilot the efficacy of a self-guided coping skills training and support intervention provided over the Internet. Treatment participants showed a trend toward greater improvement in emotional well-being compared to control participants. With a similar approach, recently, Villani et al. (2016) developed a 2 weeks eHealth protocol based on Meichenbaum's Stress Inoculation Training

(SIT) (Meichenbaum, 1985) intervention for helping elderly women undergoing chemotherapy to cope with impeding hair loss and other treatment side effects. The protocol was composed by three phases coherent with the general SIT objectives (Serino et al., 2014): (1) increasing knowledge about the stress process, (2) developing self-regulation skills and (3) helping individual to use the acquired coping skills in real contexts. SIT has been already applied in other studies to cancer patients and appeared beneficial in altering anxiety-related behaviors (Moore and Altmaier, 1981).

To conclude, design and develop eHealth interventions for elderly breast cancer patients represents a challenge for future interventions. These could be particularly helpful for older women who for medical, geographic, and/or social reasons, find themselves isolated and could have difficulties in accessing to other psychological services. Thus, first as elderly is not an homogenous group, different sociodemographic characteristics but also individual characteristics, such as personality traits and computer self-efficacy (Rockmann and Gewald, 2015), should be assessed as these could have implications on Internet adoption. Second, by considering this specific patient engagement phase, eHealth interventions can be used to manipulate the affective state of patients and help them in recovering control over their own experience by using different approaches and strategies (Villani and Riva, 2012; Carissoli et al., 2015).

Patient engagement constitutes a new frontier for health care models where eHealth could maximize its potentialities by targeting interventions to specific diseases and different phases of the life span (Riva et al., 2016).

AUTHOR CONTRIBUTIONS

DV conceived the ideas presented in the article and took the lead role in drafting the article. CC, DT, FS, and GR assisted in drafting the article.

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Promoting Patient and Caregiver Engagement to Care in Cancer

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The positive outcomes associated with Patient Engagement (PE) have been strongly supported by the recent literature. However, this concept has been marginally addressed in the context of cancer. Limited attention has also received the role of informal caregivers in promoting physical and psychological well-being of patients, as well as the interdependence of dyads. The Cancer Dyads Group Intervention (CDGI) is a couple-based psychosocial intervention developed to promote engagement in management behaviors, positive health outcomes, and the quality of the relationship between cancer patients and their informal caregivers. The article examines the ability of the CDGI to promote adaptive coping behaviors and the perceived level of closeness by comparing cancer patients participating in the intervention and patients receiving psychosocial care at usual. Results indicate that individuals diagnosed with cancer attending the CDGI present significant increases in Fighting Spirit and Avoidance, while reporting also reduced levels of Fatalism and Anxious Preoccupation. Initial indications suggest that the intervention may contribute to strengthening the relationship with the primary support person.

Keywords: engagement, cancer, patient, caregiver, group-based intervention

INTRODUCTION

Patient engagement (PE) is defined as the cognitive, emotional, and behavioral activation of patients in their care (Graffigna et al., 2013). With increasing demands for a more active role of individuals in their healthcare (Crawford et al., 2002; Davis et al., 2005; Bellardita et al., 2012; Barelo et al., 2014a,b; Menichetti et al., 2014; Barelo and Graffigna, 2015), this concept is emerging as a key factor to promote healthy behaviors, better outcomes in the context of chronic diseases, as well as greater satisfaction with quality of care (Barelo et al., 2012; Graffigna et al., 2013). In contrast with the great attention received elsewhere, PE has been marginally addressed within the context of cancer despite the evidence collected. For example, a recent survey conducted by CancerCare (2016) on 3000 adults highlighted that physical, emotional, financial, and social costs of cancer are not currently met because of the challenges patients report in collecting information about the disease, understanding their diagnosis, and communicating with the healthcare team. Furthermore, while PE has been traditionally promoted by focusing on individual factors, there is increasing attention to the family system (Carman et al., 2013; Donato and Bertoni, 2016). As stated by Carman et al. (2013) “those who engage and are engaged include patients, families, caregivers, and other consumers and citizens” (Carman et al., 2013, p. 224). This view of engagement as inclusive of the patient’s tapestry of relationships has essential implications for cancer, since it is now well-established in the literature that cancer is a relational illness (Revenson et al., 2005; Kayser et al., 2007; Manne and Badr, 2008).

Not only the disease can negatively affect the patient's quality of life by reducing physical and psychological well-being (Epplein et al., 2011; Drake, 2012; Poghosyan et al., 2013), but the illness equally affects partners and family members, who often assume the role of informal caregivers (Meeker et al., 2011; Kim et al., 2015). While significant attention has been dedicated to cancer patients' adaptation to diagnosis and treatment, the role of caregivers has been only recently addressed by the literature (Institute of Medicine, 2008). This is in contrast with the caregiving literature, which has confirmed over the years the burden associated with this role (Given et al., 2004; Kim et al., 2006). Caregiving is often related to sleep disorders (Hearson and McClement, 2007; Fletcher et al., 2008), difficulties maintaining an occupation (Rossi Ferrario et al., 2003; Stetz and Brown, 2004; Bishop et al., 2007), emotional distress (Hagedoorn et al., 2008; Kim and Given, 2008; Kim et al., 2010), as well as high levels of anxiety and depression (Rhee et al., 2008; Cipolletta et al., 2013).

Similarly, the individual focus registered in the PE literature is antithetic to the literature on couple relationships. The extensive body of knowledge collected in the last 20 years about stress and coping has highlighted that patients' and partners' adjustment to cancer is indeed interdependent, therefore supporting the need to assume a relational perspective when working with dyads (Li and Loke, 2008; Traa et al., 2014; Vellone et al., 2014). Some authors, in fact, have identified how the couple relationship has a crucial role in promoting partners' well-being, healthy behaviors, and this datum has been confirmed across illnesses and even among healthy couples (Revenson et al., 2005; Kayser et al., 2007; Manne and Badr, 2008; Saita and Cigoli, 2009; Bertoni and Bodenmann, 2010; Bertoni et al., 2012; Carpenter et al., 2015; Donato et al., 2015; Pagani et al., 2015). The concept of dyadic coping (Bodenmann, 2005) is of particular relevance in the process to move from an individualistic to a relational view of PE. Since PE represents a dynamic and changing process, the relational context can significantly affect the individual's ability to adjust to the disease (Barello et al., 2012). It therefore follows that to support this process, the attention of the researcher should be on strengthening the coping abilities of the patient and the informal caregiver working with the dyad as a unit.

Donato and Bertoni (2016) have recently proposed a model of individual, interactive, and dyadic engagement organized on two axes: appraisal and actions of care. The authors see patient-partner healthcare patterns as a result of individual vs. shared appraisal of health management, and of an individualistic vs. relational view of the health management strategies. But how is it possible to translate this relational framework in interventions that promote patients and partners' engagement? Surprisingly, a paucity of studies have examined the association of partners' relational processes, exchanges, and engagement. Even less couple-based interventions have been recorded in the literature (Scott and Kayser, 2009; Baik and Adams, 2011; Regan et al., 2012; Badr and Krebs, 2013; see for example Badr et al., 2013). An aspect of limitation is that these experiences have focused mainly on breast, prostate, and gynecological cancers, and that only recently the literature has started to focus on other types of cancer (i.e., lung cancer). Furthermore, despite their relational focus, most contributions concentrated on patients

and caregivers' outcomes separately. Hence, a significant gap in the current literature is the limited knowledge available about best practices to promote patients as well as informal caregivers' activation and engagement (Donato and Bertoni, 2016). This evidence supports the need to develop psychosocial interventions dedicated to the patient-caregiver dyad, grounded in a theoretical model that values the role of close relationships and that can be ultimately aimed at increasing the level of patients and caregivers' engagement in behaviors that (a) contribute to more beneficial adaptation to the cancer experience, and (b) support the bond between the individual and the informal caregiver.

The present contribution examines the effectiveness¹ of the Cancer Dyads Group Intervention (CDGI); an innovative protocol developed to promote engagement in management behaviors, maximize positive health outcomes, and the quality of the relationship between cancer patients and their informal caregivers. The unique features of this approach can be identified in its theoretical framework, relational focus, and in the fact that the program can be easily translated into routine care in a variety of settings (from hospitals to community-based centers, and private practice). An overview of the program, its theoretical foundation, and techniques is available in **Table 1**. While participating in the CDGI program, patients, informal caregivers, and healthcare providers engage in an active partnership aimed at ensuring the best quality of care. The article presents the available empirical evidence about the ability of the CDGI to promote adaptive coping behaviors and the quality of the relationship by comparing participants and individuals receiving usual psychosocial care.

MATERIALS AND METHODS

Participants

Participants were 50 cancer patients recruited from two hospitals in the Northern part of Italy. Sixteen patients participated in the CDGI, while the remaining 34 were used as a control group. Cancer patients who did not participate in the group intervention were referred to usual psychosocial care available at the institutions involved in the study (psychologists and psychiatrists), where the most common type of psychosocial support is individual therapy. Institutional Review Board approval was obtained from the University IRB as coordinating center of the study and each participating institution (E. Bassini Hospital, Milano, Policlinico Hospital, Monza). The inclusion criteria for the study required that the participants were: (1) 18 years old or older, (2) free of dementia symptoms and a psychiatric diagnosis, (3), involved in a relationship with a significant other (partner, spouse, family member, friend), (4) Italian-speaking, and (5) had received a diagnosis of cancer in the last 3 months.

The average age of the participants in both groups was 62 years ($SD = 8.80$ for CDGI, $SD = 8.12$ for the Control Group). In the CDGI group, the majority of the patients were women (87.5%) diagnosed mostly with breast cancer (68.8%), where only a small number of participants were in treatment for

¹Cfr. (Haynes, 1999).

TABLE 1 | An overview of the Cancer Dyads Group Intervention.**The Cancer Dyads Group Intervention**

The Cancer Dyads Group Intervention (CDGI) is a supportive group-based intervention for cancer patient and caregiver dyads theoretically inspired by the Bio-psychosocial Model (Engel, 1977), the Symbolic Relational Model (Cigoli and Scabini, 2006; Scabini and Cigoli, 2012), and the Psycho-Educational Approach (Fawzy and Fawzy, 1998). The Bio-Psychosocial Model offered a holistic alternative to the biomedical model, therefore stating that the illness must be addressed focusing on three dimensions: the biological, the psychological, and the social domain (Engel, 1977). This perspective allows to contextualize care not only as the limited application of scientific knowledge (Saba, 2002), but as an action that occurs in the interaction between individuals where trust is essential (Saita et al., 2015b). As a consequence, it is possible to treat the illness while also validating the life experience of the single individual. In this sense, the ability of the individual to cope with cancer is influenced not only by the suffering of the body, but also the ability to sustain the emotional dimension of sorrow, loss, uncertainty, and –sometimes– helplessness (Saita, 2009).

The Symbolic Relational Model is the second theoretical foundation of the intervention. It is aimed at investigating family relations by focusing on the connection existing between individuals and family members (Cigoli and Scabini, 2000). Although the essence of health is perceived to be associated with the quality of the close relationships (Cigoli, 2002), only a limited number of studies have focused on the relational network. For this reason, the intervention enhances the caregiving relationship rather than the patient and the caregiver as individuals. While the first two theoretical frameworks have inspired the authors' attention to multiple determinants of health, and the crucial role the relationship with a significant other has during the time of illness, the Psycho-Educational Approach (Fawzy and Fawzy, 1998) assumes a significant value when considering patients and partner's engagement. Since it focuses on the relevance clear communication—of symptoms, treatment, and the implications on lifestyle—has in the context of illness, this aspect becomes essential when designing programs aimed at promoting patients' engagement. Moreover, the group setting has been proved to promote the emotional disclosure of participants and to facilitate the exchange and communication among its members, thus facilitating the engagement of both patients and caregivers (Saita et al., 2016).

Furthermore, the decision to use the group as a clinical tool is supported by the psychoanalytic concept of "group thought," which refers to the experience of thinking together (Neri, 1995, 2003). Although when referring to group-based interventions it is necessary to consider numerous issues (for example: the type of group used, the kind of intervention to be planned, the techniques to be used, the setting, the strategy to conduct the sessions, and the socio-affective dynamics), it is not our intention to address here the complexity of these dimensions. The group setting becomes relevant in the development of the CDGI because the group is a psychological entity different from the sum of single individuals (Bion, 1961; Foulkes, 1964, 1973). According to Foulkes (1973), the individual unconscious is connected with the group unconscious, which the author compares to a network where each individual is metaphorically denoted by a knot. This reflection informed our idea of "thinking about oneself and the other," which represents the basis of an intervention aimed at supporting crucial relationships during cancer, including the affective bonds the patient develops with relatives, friends, and healthcare providers.

The influence of these theoretical models has shaped and informed the techniques used in the meetings with the participants. The CDGI is organized in eight sessions and the group meets every 2 to 3 weeks for a couple of hours in a conference room of the hospital where patients are treated and where they have been recruited. Every session deals with a specific topic and begins with an exercise aimed at identifying and strengthening new coping repertoires of the dyad. The product of each exercise is later shared with the rest of the group to promote patients' and caregivers' closeness, and a sense of belonging among the participants. Two practitioners with extensive knowledge and experience in psychosocial oncology are the facilitators of the program. More practically, in collaboration with the multidisciplinary team individuals receiving care at the participating institutions are invited to participate. The group usually begins when enough dyads are recruited, from a minimum of 6 to a maximum of 10 participants. A brief overview of the CDGI is presented in the next paragraphs, while a more detailed presentation is available in a previous work (Saita et al., 2014). While the CDGI was initially developed for patients diagnosed with breast cancer, over the years the intervention has been easily adapted to meet the needs of individuals diagnosed with other types of cancer (like rare tumors; e.g., epithelial tumors of some organs or different types of sarcoma) and their caregivers. Furthermore, the CDGI has been applied not only with partners in a committed relationship, but also with other types of dyads where the role of caregiver is assumed by a member of the family system (brother, sister, daughter, or son), or a peer (for example when friends are involved).

Session 1_My coping, your coping, our coping

The first session is aimed at facilitating the identification of the individual's coping strategies and to develop bonds among the members of the group, and with the two conductors. After the participants are introduced to each other, the facilitators present and read the stories of two cancer patients presenting opposite coping styles: active versus avoidance and denial. By comparing their own experiences with the two proposed stories, participants are encouraged to explore the concept of coping with cancer and to recognize their own coping strategy. This is also the moment when the facilitators introduce the idea that the coping process involves the partner or significant others; a strategy to bring the concept of dyadic coping in the setting of the intervention (Acitelli and Badr, 2005; Bodenmann, 2005; Donato and Bertoni, 2016).

Session 2_Understanding Cancer

The session is focused on enhancing patients and caregivers' understanding of the illness. By involving a physician, it is possible for the participants to increase their knowledge about the diagnosis, treatment consequences, and overall impact on the quality of life. This is a crucial moment not only to clarify what are resources available to the patient, but also because the presence of the physician offers the opportunity to engage in an open communication which promotes the patient-provider relationship and their interaction becomes more meaningful and authentic. This meeting is divided in three main phases. In the first part, patients and caregivers can express their concerns about cancer, its treatment, and the overall cancer care continuum. The second phase involves the presence of the oncologist, who is invited to join the group to answer questions prepared by the participants or issues emerged in the first part of the meeting. This moment is particularly important to reduce the stress and uncertainty associated with cancer; especially for patients whose diagnosis is less common in the literature. Finally, dyads are invited to reflect together on the illness, to share thoughts, emotions, and concerns connected with the management of the disease. In particular, attention is given to concerns and challenges as well as to their hopes for the future.

Session 3_Before/After

Cancer requires the patient and the caregiver to assume new roles within the family and the relational system, with significant adjustments of the dynamics of giving and receiving care. Hence, the third session focuses on the change introduced by the diagnosis (participants are usually at the beginning the active treatment phase when they attend the intervention). Each dyad is invited to identify differences between the time before and after cancer, and later these topics are shared with the group. The clinical work of the two conductors is aimed at supporting the verbalization of concerns and aspects of change connected not only to the management of the illness, but more importantly to their relationship and the link with the supportive network (family members, close friends, colleagues); aspects which are often very difficult to verbalize and to process. As a consequence, therapists are attentive to feelings of uncertainty, resentment, denial, and inability to manage the demands of the illness. By offering participants a safe space to allow these feelings and concerns to emerge and to be shared with others facing the same stressor, it follows that participants become more aware of the impact of the illness on the life experiences of the patients, but also on the lives of caregivers, partners and family members.

(Continued)

TABLE 1 | Continued*Session 4_Looking for strength and resilience through the generations*

Continuing the work to highlight the dyad's ability to engage in behaviors that facilitate a more beneficial adaptation to the cancer, as well as the relevance each other has for their well-being, the fourth session use the genogram (McGoldrick et al., 1999) as a strategy to identify strength and resilience through the generations. Participants are also asked to include relationships with significant others that may not be included in the traditional family structure. When the genogram is completed, dyads are invited to present their products to the group and to describe their family history, significant events happened to family members, and/or family myths. Finally participants are asked how the illness has been or can be integrated in the broader and larger family history.

Session 5_ "Place me like a seal over your heart, like a seal on your arm": the Coat of Arms

The fifth session deepens participants' understanding of how relationships can become resources during the cancer experience by using the instrument of a family coat of arms. After providing some example, every dyad is asked to draw a coat of arms that would represent their family and its key features (some participants have even added a motto that summarized their strength and resources). The goal of the exercise is to discover positive aspects, resources and competencies already available within their close relationships, so that no resource is lost during this time of need.

Session 6_Body Image and Cancer

The core element of this meeting is the body and its transformation as a consequence of the illness, offering both patients and caregivers the opportunity to reflect about the beauty and strengths still present despite the negative impact of the treatment and its side effect on the body image of the patient. Using a photo-elicitation technique, each patient is invited to choose one image (from a set of 20) representing famous statues of female or male bodies (for examples the Donatello's David or the Venus de Milo), then each dyad is invited to write about the emotions associated to the image and to explore the meanings for his/her life experience. These products, which are then shared with the other participants, contribute to the discussion about body image and to the impact of cancer on intimate relationship and intimacy.

Session 7_Mind/Body Connection

Session seven focuses on the concept of mindfulness. It begins with a brief relaxation exercise which can be completed without any specific support (a chair is enough). Subjects are given instructions to repeat the exercise outside the setting of the intervention. The relaxation exercise introduces a reflection about the mind-body connection and the reciprocal influence, aimed at identifying strategies to handle negative emotions and the stress experienced as the end of the treatment nears. This session ends with the request to each dyad to select or create an object that symbolizes what experienced during the program and to bring it to the last session. The facilitators do the same, by selecting an object that denotes their experience as well.

Session 8_Making Meaning and Closure

The last session begins with the presentation of the objects the dyads have chosen or created, to support the dyad making meaning of the experience while also bringing closure to the intervention. Then, each participant is given the opportunity to verbalize what the group and the contents of the sessions may have done for him/her. Symbolically, the session ends with diplomas presented to every dyad and with a gift from the facilitators.

rare cancer (31.3%). Overall, participants in the intervention group were married (56.3%) and were not highly educated (62.6% did not graduate from high school). Subjects in the control group present similar socio-demographic characteristics. Most patients were women (79.4%), and individuals with rare cancer diagnoses represented one third of the group (32.4%). Similar to what reported for the intervention group, 76.5% of cancer patients were married. However, members of the control group were more highly educated, with 32.4% being high school graduates and 8.8% being college graduates. Informal caregivers of individuals in the CDGI group were mostly romantic partners (75%), with a mean age of 65 (mean 64.8, $SD = 9.1$), low level of education (52% had only completed junior high school), and currently retired (60%).

Procedure

Participants were initially screened by a psychologist to determine their eligibility. After a brief interview about the cancer experience, study participants completed a set of questionnaires measuring closeness with their informal caregiver, and coping strategies at time of recruitment (within 3 months from diagnosis). The same questionnaires were then completed within the 1 month after the end of the intervention, while for individuals in the control group the post-test data collection occurred 6 months after the initial contact.

Measures

Individual Coping. To identify the prevailing coping style used to cope with cancer, the Italian version of the Mini-Mental Adjustment to Cancer Scale (Mini-MAC) (Watson et al., 1994; Grassi et al., 2005) was selected. The instrument is a 29-item questionnaire which identifies five coping strategies: Fighting Spirit, Hopeless/Helplessness, Anxious Preoccupation, Fatalism and Avoidance. Hopelessness/Helplessness indicates a coping style characterized by the belief of low control on events, which is associated with high levels of anxiety and depression. Individuals with a fatalistic coping behavior show low sense of control, resignation and passive acceptance of fate. Anxious Preoccupation is used to describe a coping modality with high levels of anxiety and worry about the cancer diagnosis, which can impact the quality of life of the individual. The patient is either looking for constant reassurance or is distancing herself/himself from the healthcare environment. Avoidance indicates the tendency to minimize cancer and to refrain from the search of information. Fighting Spirit is characterized by an optimistic attitude toward one's ability to cope with the illness. Next to low levels of anxiety and depression, individuals presenting fighting spirit tend to perceive the illness as a challenge. They implement diverse and flexible cognitive strategies, which contribute to a positive appraisal of the experience. This coping style has been associated with better psychological morbidity, increased sense of control and better prognosis (Pettingale et al., 1985; Burgess et al., 1988; Saita et al., 2015a).

Interpersonal Closeness. Perceived level of closeness with the primary support person was measured by the Inclusion of the Other in the Self (IOS) Scale (Aron et al., 1992). The measure consists of seven pairs of overlapping circles that are drawn to show varying levels of overlap, indicating an increasing degree of closeness in the relationship. A 7-point scale is used to score the degree of closeness. In the present study, we asked the individual to indicate up to five persons who provide support to them and describe each relationship by choosing one of the seven circles. Although the scale is not formally validated in the Italian population, the very easy and intuitive nature of the questionnaire -being a single item pictorial tool- has contributed to its use with Italian subjects, as documented by earlier works of these authors (Saita et al., 2015a), other Italian researchers (ex. De Panfilis et al., 2015), or studies conducted including Italian samples (Karremans et al., 2011).

Data Analysis

Descriptive statistics of cancer patients were obtained for all the variables compiling frequency tables, histograms, and bar graphs. Differences in the coping style behavior for the intervention and control group were examined with Independent Sample *t*-test, while differences within patients were assessed calculating paired samples *t*-test. IBM SPSS Statistics 22 was used for data screening and data analysis. Changes in the perceived degree of closeness with the informal caregivers were described comparing who were the sources of support identified by the cancer patients and by calculating the mean scores originating from the position of the IOS Scale used to describe these relationships

RESULTS

Differences between Patients: Examining Changes in Coping Style between the Intervention and Control Group at Pre-test and Post-test

An Independent Samples *t*-test was used to compare the mean score of each coping strategy of individuals participating in the CDGI intervention and those of the control group at pre and post-test, in order to identify if the two groups were already different in their coping behaviors at pre-test and if a change occurred at the post-test. While results indicate that no statistically significant difference existed at pre-test, at post-test individuals who participated in the CDGI presented significantly higher Fighting Spirit [$t_{(48)} = 2.71, p < 0.01$] than cancer patients who received usual psychosocial care.

Differences within Patients: Pre-test/Post-test Comparison among the Participants of the Intervention and Control Group

The previous findings were confirmed when a pre-test/post-test comparison was conducted on each group, using Paired Samples *t*-test. Results indicate that individuals diagnosed with cancer attending the CDGI present significant increase in Fighting Spirit [$t_{(14)} = -2.31, p < 0.05$] and Avoidance [$t_{(14)} = -4.65, p <$

0.001], while reporting also reduced mean scores in Fatalism [$t_{(14)} = 3.42, p < 0.01$] and Anxious Preoccupation [$t_{(14)} = 3.40, p < 0.01$]. On the contrary, the changes registered in the control group indicate that individuals reported significantly higher scores of Hopelessness/Helplessness [$t_{(32)} = -2.41, p < 0.05$] next to reduced Fatalism [$t_{(32)} = 4.54, p < 0.001$] (Tables 2, 3).

While the results about Hopelessness/Helplessness may be considered in contrast with the overall aim of the intervention, they may be contextualized referring to the types of cancer included in the study. Given the limited number of subjects in the two groups, this analysis is only exploratory in nature. When the mean scores have been compared differentiating between breast and rare cancer patients, results indicate that at pre-test individuals with rare tumors in the CDGI presented significantly higher scores of Hopelessness/Helplessness [$t_{(14)} = -2.71, p < 0.05$] compared to women with breast cancer, while on the contrary this group scored higher on Fighting Spirit [$t_{(14)} = 2.63, p < 0.05$] and Fatalism [$t_{(14)} = 2.88, p < 0.05$]. The same differences were found also in the control group, where breast cancer patients also shown higher Avoidance [$t_{(32)} = 2.75, p < 0.05$]. At post-test, it clearly emerges how the intervention contributed to reduced Hopelessness in patients with rare tumors [$t_{(14)} = 4.19, p < 0.01$], while patients with breast cancer presented significantly higher Fighting Spirit [$t_{(14)} = 4.15, p < 0.01$] and significantly lower Fatalism [$t_{(14)} = -3.9, p < 0.01$]. In the control group, differences were registered on Fatalism [$t_{(32)} = -6.2, p < 0.001$] and Hopelessness [$t_{(32)} = 7.7, p < 0.001$], with individuals with breast cancer presenting higher Hopelessness and lower Fatalism (Tables 4, 5).

Examining Changes in the Perceived Degree of Closeness with the Primary Support Person

The relational perspective of the intervention determines the need to explore the perceived level of closeness with the informal caregiver. To examine changes in the two groups we first considered which persons were identified as the primary source of support, and then we compared the mean values obtained from the picture selected by the participants as indication of closeness. For individuals in the CDGI, the source of their support is identified in the relationship with partners, children and siblings. This indication is confirmed also at post-test, with these three categories being the most listed by the participants. One patient also indicated that the family as a whole became the source of support. For the control group, while at pre-test the most commonly identified individuals were partners, children, and siblings, 6 months after the initial contact patients started to expand their supportive network and to include parental figures and the healthcare system.

When focusing specifically on the degree of closeness with the primary support person, which are presented in Figures 1, 2, it is possible to notice how members of the CDGI reported elevated levels of closeness with the primary support person; as indicated by higher scores in the relationships with partners, children and siblings. We want also to note that while the perceived closeness with friends was very elevated at pre-test,

TABLE 2 | CDGI Pre-post test comparison.

Variable	Time	Mean	SD	t	p	95% CI	
						LL	UL
Fighting spirit	Pre-test	2.99	0.79592	-2.308	0.036	-0.84150	-0.03350
	Post-test	3.43	0.50091				
Hopelessness/Helplessness	Pre-test	1.67	0.55812	-2.844	0.012	-1.40003	-0.20059
	Post-test	2.47	0.79120				
Fatalism	Pre-test	2.96	0.64174	3.424	0.004	0.411759	1.769491
	Post-test	1.87	0.818325				
Anxious preoccupation	Pre-test	2.14	0.52599	3.402	0.004	0.077864	0.339011
	Post-test	1.93	0.648717				
Avoidance	Pre-test	2.39	0.59139	-4.652	0.000	-0.934138	-0.347112
	Post-test	3.03	0.442672				

Bold values indicates significant results.

TABLE 3 | Control Group Pre-post test comparison.

Variable	Time	Mean	SD	t	p	95% CI	
						LL	UL
Fighting spirit	Pre-test	2.77	0.63038	-1.327	0.194	-0.45831	0.09654
	Post-test	2.95	0.61335				
Hopelessness/Helplessness	Pre-test	1.86	0.60824	-2.410	0.022	-0.96772	-0.08169
	Post-test	2.38	1.01721				
Fatalism	Pre-test	2.70	0.69186	4.537	0.000	0.467034	1.226496
	Post-test	1.86	0.735365				
Anxious preoccupation	Pre-test	2.09	0.70088	0.880	0.385	-0.153928	0.388772
	Post-test	1.97	0.556457				
Avoidance	Pre-test	2.67	0.87083	-0.352	0.727	-0.478033	0.337150
	Post-test	2.74	0.715798				

Bold values indicates significant results.

this relationship was no longer indicated at post-test. On the contrary, friends were substituted by the family as a whole, indicating greater reliance of the patient on the family system.

Figure 2 illustrates the change registered among individuals in the control group. The perceived level of closeness with the partner increased, but for those patients who identified the primary support person with children or siblings, the perceived closeness was reduced at post-test. Furthermore, the relationship with peers lost relevance over time, substituted by a closer connection with the parental figures. It is also important to highlight that for this group the family as a whole did not assume a meaningful role over time, and that external sources of support were searched in the relationships established with healthcare providers, such as physicians, nurses, and mental health professionals.

DISCUSSION

In this paper we have supported the relevance close relationships have in the promotion of PE in the context of cancer. As a result of the need to assume a relational view of this concept, we have illustrated the CDGI as a way to operationalize PE as involving patients and informal caregivers. This program has been developed with the goal to increase patients' engagement in management behaviors, enhance awareness of the emotional dimension associated with the illness, and to promote the quality of the relationship between cancer patients and their informal caregivers. Furthermore, this article aspired to provide further empirical evidence about the effectiveness of the CDGI to activate patients' coping abilities and strengthening the senses of closeness with the primary support person, while previous works have been mostly focused on the

TABLE 4 | Pre-test comparison by cancer type.

Group	Variable	Cancer Type	Mean	SD	t	p	95% CI	
							LL	UL
CDGI	Fighting spirit	Breast cancer	3.29	0.70550	2.63	0.02	0.17549	1.73542
		Rare Tumor	2.34	0.58885			0.19381	1.71710
	Hopelessness/Helplessness	Breast cancer	1.46	0.40268	-2.71	0.02	-1.22514	-0.14268
		Rare tumor	2.15	0.60063			-1.41430	0.04648
	Fatalism	Breast cancer	3.21	0.59635	2.88	0.01	0.20947	1.42689
		Rare tumor	2.40	0.28284			0.34622	1.29014
	Anxious preoccupation	Breast cancer	2.12	0.61938	-0.25	0.74	-0.70127	0.55563
		Rare tumor	2.19	0.27181			-0.55076	0.40513
Control Group	Fighting spirit	Breast cancer	2.43	0.52549	0.40	0.69	-0.57228	0.83591
		Rare tumor	2.30	0.77862			-0.81509	1.07873
	Hopelessness/Helplessness	Breast cancer	3.03	0.58535	5.46	<0.001	0.42533	1.18534
		Rare tumor	2.22	0.27372			0.50522	1.10545
	Fatalism	Breast cancer	1.67	0.53597	-2.88	0.007	-0.98894	-0.16545
		Rare tumor	2.25	0.58392			-1.01474	-0.13964
	Anxious preoccupation	Breast cancer	2.91	0.68444	2.75	0.010	0.16479	1.10857
		Rare tumor	2.27	0.49736			0.21151	1.06185
Avoidance	Anxious preoccupation	Breast cancer	2.03	0.79625	-0.69	0.492	-0.70749	0.34753
		Rare tumor	2.21	0.44962			-0.61719	0.25724
	Avoidance	Breast cancer	2.89	0.93844	2.75	0.010	0.05553	1.28487
		Rare tumor	2.22	0.48047			0.17412	1.16628

Bold values indicates significant results.

TABLE 5 | Post-test comparison by cancer type.

Group	Variable	Cancer Type	Mean	SD	t	p	95% CI	
							LL	UL
CDGI	Fighting spirit	Breast cancer	3.67	0.33344	4.15	0.001	0.37573	1.17882
		Rare tumor	2.90	0.37914			0.31064	1.24390
	Hopelessness/Helplessness	Breast cancer	2.86	0.58485	4.20	0.001	0.60317	1.86411
		Rare tumor	1.63	0.42953			0.65690	1.81037
	Fatalism	Breast cancer	1.48	0.666742	-3.90	0.002	-1.912	-0.555328
		Rare tumor	2.72	0.303315			-1.753	-0.713319
	Anxious preoccupation	Breast cancer	1.88	0.744678	-0.45	0.65	-0.934735	0.607462
		Rare tumor	2.05	0.410792			-0.789682	0.462409
Control Group	Fighting spirit	Breast cancer	3.09	0.314498	0.78	0.44	-0.327733	0.709551
		Rare tumor	2.90	0.675463			-0.632810	1.014628
	Hopelessness/Helplessness	Breast cancer	3.02	0.68621	1.00	0.32	-0.23184	0.68401
		Rare tumor	2.80	0.40927			-0.15970	0.61187
	Fatalism	Breast cancer	2.90	0.79322	7.71	<0.001	1.06471	2.10328
		Rare tumor	1.31	0.40415			1.16541	2.00258
	Anxious preoccupation	Breast cancer	1.48	0.450557	-6.24	<0.001	-1.521	-0.772675
		Rare tumor	2.63	0.598787			-1.5795	-0.714950
Avoidance	Anxious preoccupation	Breast cancer	1.91	0.492031	-0.90	0.37	-0.600940	0.232482
		Rare tumor	2.09	0.681147			-0.673038	0.304580
	Avoidance	Breast cancer	2.77	0.635979	0.25	0.80	-0.476157	0.608370
		Rare tumor	2.70	0.893156			-0.573572	0.705786

Bold values indicates significant results.

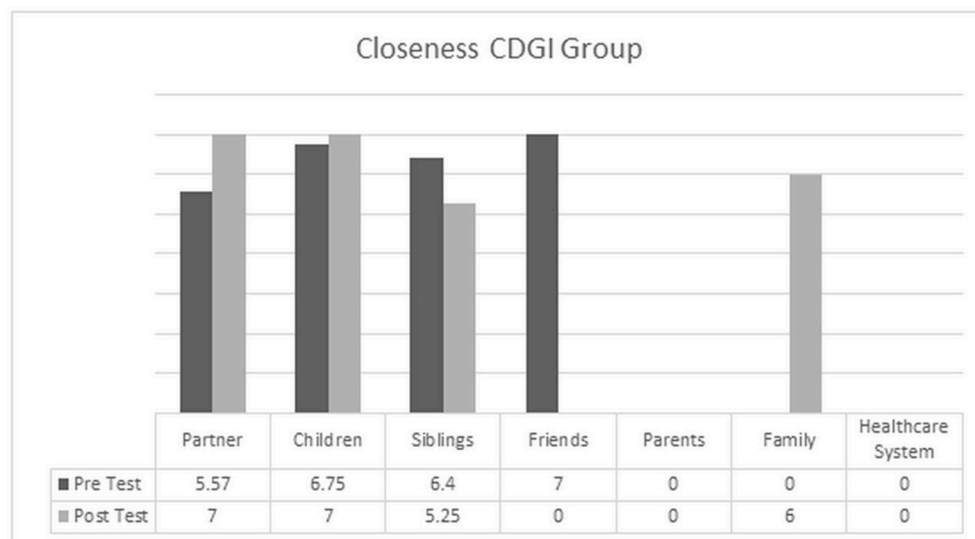


FIGURE 1 | Pretest/Posttest comparison of the mean value of closeness for individuals in the CDGI.

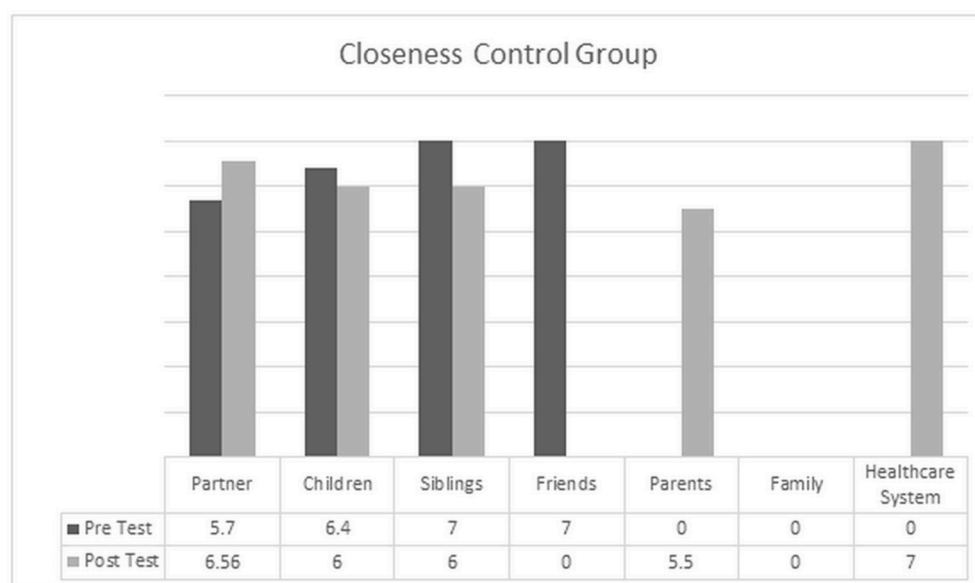


FIGURE 2 | Pretest/Posttest comparison of the mean value of closeness for the control group.

process of the intervention (Saita et al., 2014). The analysis conducted in the present contribution focused on the coping strategies and the perceived degree of closeness, by comparing pre-test and post-test scores of patients who participated in the intervention and those of were referred to usual psychosocial care. Three are the most relevant findings to discuss.

First, differences in coping strategies between patients highlight how individuals who participated in the CDGI have developed higher levels of Fighting Spirit at the end of the program, which suggests that participants were able to develop

more adaptive behavioral and emotional strategies. They seem to be better equipped to cope with the potentially stressful events and feelings associated with the cancer experience than the individuals who did not. Moreover, the enactment of coping behaviors characterized by Fighting Spirit also reveals the willingness of the individual to face the multiple stressors a cancer diagnosis originates, and the realization of mastering the abilities necessary to face the disease. This aptitude also sustains the hope to foresee a future with no cancer, despite the uncertainty associated with this illness (Coward and Kahn, 2004; Saita et al., 2015b).

This finding is also supported by the pre-test and post-test comparison of the two groups. In our study, the control group reported statistically significant increase in Hopelessness and Fatalism. Individuals receiving usual care showed over time indication of low control on events, resignation, and passive acceptance, which has been associated with negative quality of life outcomes in the literature (Meerwein, 1989; Barraclough, 2001). On the contrary, individuals in the intervention group presented aspects of change in all the coping styles evaluated. While the indication of higher Fighting Spirit and Avoidance, reduced Fatalism, and lower Anxious Preoccupation are in line with the outlined goals, the statistically significant increase in Hopelessness/Helplessness appeared in contrast with our hypotheses. However, these findings may be clarified considering how the two types of cancer in the sample (breast cancer vs. rare tumors) can affect the quality of life of the individual, given the dissimilar treatments, outcomes, and survivorship issues. Although it is necessary to be mindful of the very limited number of individuals with rare tumors included in the study, it is possible that the different level of information and knowledge available about rare types of cancer (including the preparedness of the healthcare team) can contribute to affect the outcome of the intervention. A second consideration then pertains the feelings associated with a cancer diagnosis, which are often unexpected and destabilizing for the individual, whose sense of psychological and physical integrity is suddenly threatened. Despite the differences between facing a well-known and studied pathology vs. a rare disease, the possibility to receive information related to the illness and its treatment contributes to higher adherence (Cousson-Gélie et al., 2008). In this sense, the higher level of Hopelessness/Helplessness registered among patients in the control group can be partially explained by the sense of loneliness and isolation experienced when facing cancer alone. The attendance of the CDGI can, on the contrary, alleviate the feelings of helplessness and lack of control the diagnosis has originated.

Finally, given the theoretical foundation of our work and the role close relationships have for physical and psychological well-being of patients and informal caregivers, we examined patients' closeness. These findings suggest that the CDGI experience represents a setting where the relationship with the primary support person can be nurtured and strengthened. Not only CDGI participants continued to identify as sources of support the relationships established within the family of origin or the partner, but the descriptive analysis about the mean level of closeness indicates that the program contributed to increased degree of proximity and support with the informal caregiver and the family as a whole. Differently, individuals in the control group experienced lower sense of closeness with children and siblings, and some of them showed a tendency over time to rely more on the healthcare system. This movement mirrors "the stress-coping cascade effect" (Bodenmann, 2005), which describes how individual coping strategies are substituted by dyadic approaches (involving partners, relatives, and friends), and ultimately healthcare professionals. Hence, when a coping strategy is no longer functional, the individual continues to search support until he can find an adequate response to his

needs. Results of the descriptive analysis of the IOS Scale data are of particular interest because while extensive attention has been given to the development of psychosocial interventions to promote coping, a limited number of studies have investigated the less conscious aspect of the relationship between cancer patient and his/her informal caregiver (Aron and Aron, 1986; Aron et al., 1991).

Given the important clinical implications the intervention has for the current debate about best-practices to promote patients and informal caregivers' engagement to care, it is important to describe how this contribution is affected by several limitations. First, the present work relied on a small sample size, which limits our ability to generalize these findings and also represents a limitation in the selection of the data analysis strategy. While difficulties in the recruitment of dyads in research are extensively reported, this article also represents the result of years of collaboration with hospital settings and illustrates a strategy to move from a qualitative analysis of the intervention (Saita et al., 2014) toward a quantitative approach. Moreover, in this contribution it was not possible to include and analyze data from both members of the dyad. Although this may seem in contrast with the relational perspective that has inspired our work, the need to focus only on patients' data was influenced by the fact that the questionnaires completed by the participating caregivers, although being collected as part of the research protocol, were not fully available at the time of the analysis. It will be therefore important to include data about patients and caregivers' change as part of the participation in the CDGI to provide further empirical evidence to the findings we have published so far. Third, as the intervention promotes dyadic coping strategies to strengthen adaptation and engagement in both partners, a measure of dyadic coping should be included in future works. Similarly, it will be critical to add a measure of PE and to target participants' satisfaction with care; aspects that were not investigated in the current work. Finally, from a methodological perspective, data should be analyzed using a dyadic data approach, in order to account for the interdependence of patients' and partners' scores.

Summarizing, while a relational view of the concept of PE has received increasing attention in the literature, the present work has illustrated how it is possible to develop interventions that support the bond between the individual and his/her informal caregiver. Within these experiences, the CDGI contributes to foster PE as a "process-like and multidimensional experience" (Graffigna et al., 2014, p. 1), by focusing both on health information (working together with providers) and on the affective dimension of the illness. Participants reported significant reduction in coping strategies like Anxious Preoccupation and Fatalism, and higher Fighting Spirit than controls. Furthermore, from this initial analysis it is possible to appreciate how the CDGI represents a setting where it is possible to support the relationship between patient and informal caregiver.

Not only this program is almost unique in the field of PE in the context of cancer, but while previous studies have targeted only individuals outcomes, the intervention described here helps participants to find the resources to cope with the

illness indentifying the relationship as the therapeutic tool. Since support within the context of a close relationship leads to better outcomes, we propose that forms of intervention that focus on these dyads would be appropriate and potentially effective in promoting and enhancing engagement and quality of life for both the patient and partner (Kayser and Scott, 2008; Saita et al., 2014). This brings us back to the suggestions identified by Donato and Bertoni (2016) about the development of interventions aimed at promoting patients and partners' enagement. The authors discuss the relevance of a clear theoretical framework, of addressing both patients and partner's needs, and to being able to integrate individual, interactive, and relational levels of intervention. These indications guided our work and were used to support the effectiveness of the intervention, given that the CDGI integrates the Bio-psychosocial Model (Engel, 1977), the Symbolic Relational Model (Cigoli and Scabini, 2006; Scabini and Cigoli, 2012), and the Psycho-Educational Approach (Fawzy and Fawzy, 1998). Similarly, the intervention focuses on the dyad

and the relationship between patients and caregiver, therefore integrating excercises and activities with individual, interactive and relational focus. Our study then represents a nice integration of their model, as it introduces also a third dimension to consider when developing interventions to promote PE using a relational perspective. As the appraisal of care and the actions of care define three levels of patient-partner engagement, psychosocial interventions should be placed along a continuum of settings (ranging from individual, couple, and group of patients or caregivers, and groups of dyads) with the goal to offer the most appropriate and effective setting of intervention given the unique characteristics of the participants the equipe is working with.

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Literature Review: CA and SM. Description of Intervention: ES. Data collection and Data analysis: ES and CA. Discussion: All authors.

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Effectiveness of Caregiver Training in Mindfulness-Based Positive Behavior Support (MBPBS) vs. Training-as-Usual (TAU): A Randomized Controlled Trial

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Caregivers of individuals with intellectual and developmental disabilities (IDD) often end up having their medical and psychological well-being compromised due to the stressful nature of caregiving, especially when those in their care engage in aggressive behavior. In this study, we provided caregivers with mindfulness-based training to enable them to better manage their psychological well-being and, through this, to also enhance specific indices of quality of life of the individuals in their care. Thus, the aim of the present study was to evaluate in a randomized controlled trial (RCT) the comparative effectiveness of Mindfulness-Based Positive Behavior Support (MBPBS) and Training-as-Usual (TAU) for caregivers in a congregate care facility for individuals with severe and profound IDD. The comparative effects of the two training conditions were assessed in terms of caregiver variables care recipient variable (number of aggressive events), and agency variables Results showed that MBPBS was significantly more effective than TAU in enabling the caregivers to manage their perceived psychological stress, and to reduce the use of physical restraints and stat medications for aggressive behavior of the individuals in their care. In addition, there were significant reductions in aggressive events by the individuals in their care, 1:1 staffing of individuals with aggressive behavior, and staff turnover. Furthermore, the MBPBS training was significantly more cost-effective than the TAU training. If replicated in future RCT studies, MBPBS may provide an effective means of enhancing socially acceptable bidirectional engagement of caregivers and care recipients within a person-centered context.

Keywords: Mindfulness-Based Positive Behavior Support, MBPBS program, 1:1 staffing, aggressive behavior, physical restraints, psychological stress, staff turnover, benefit-cost analysis

INTRODUCTION

Caregivers, regardless of whether they are unpaid family members or paid staff, often end up having their psychological well-being compromised due to the stressful nature of caregiving (McIntyre et al., 2002; Hastings et al., 2006; Herring et al., 2006). Many change jobs because of psychological stress (Hastings and Beck, 2004) and burnout (Chung and Harding, 2009),

or require therapy if they continue in their role of caregiving. In this context, psychological stress results from emotional and physiological reactions to job-related demands that a caregiver is unable to cope with, and burnout results when prolonged stress exhausts the physical and emotional strength of the caregiver. Recent research suggests that caregivers can enhance the self-management of their psychological stress by engaging in a disciplined practice of meditation (Singh, 2014). There are several mechanisms that may come into play when a caregiver regularly practices meditation, especially mindfulness (Anālayo, 2016). For example, enhanced mindfulness may provide caregivers better emotional self-regulation during periods of acute stress. It may also increase cognitive flexibility, when their responses are informed by awareness of what is unfolding in the present moment without the distortions of their own emotions and perceptions of the events. Every time caregivers are able to view each unfolding event with a beginner's mind, perceiving each event as if it is occurring for the first time, then "right action" emerges (Suzuki, 1970).

Caregivers of individuals with intellectual and developmental disabilities (IDD) face additional stress from the severe challenging behaviors of individuals in their care (Hensel et al., 2012; Didden et al., 2016). For example, individuals with IDD evince aggression that is often of low frequency but high intensity, and are likely to physically hurt their caregivers as well as their peers. The prevalence rate of aggression in this population varies considerably, ranging from about 7% (Emerson et al., 2001) to over 50% (Tenneij et al., 2009; Jahoda et al., 2013). Current research suggests that when caregivers are stressed due to the aggressive behavior of the individuals in their care, they tend to develop a negative attitude toward the individuals, eventually leading to negatively interacting with them or avoiding them (Jahoda et al., 2013). Indeed, caregiver stress may lead them to recommend that individuals in their care who are aggressive be treated with restrictive procedures such as psychotropic medications, emergency medications, and physical restraints (Singh et al., 2011a; Deveau and McGill, 2014).

In response, agencies have typically taken one of three general approaches to assist caregivers in delivering services to individuals with IDD. In the most widely used approach, caregivers are provided additional training in managing the challenging behaviors of individuals in their care. This could take many forms. For example, caregivers often receive new employee and in-service training in behavior management procedures, typically involving the principles and practice of positive behavior support (PBS; MacDonald, 2016; Morris and Horner, 2016). When implemented with fidelity, PBS has been shown to be immensely successful in caregiver management of the challenging behaviors of individuals with IDD. However, PBS procedures may not be used with fidelity in actual practice because of staff shortages, the inability of staff to use the procedures with more than one person at a time, caregiver stress, and the intensity of effort required (Allen et al., 2005; Didden et al., 2016). While assisting caregivers to better manage the aggressive behavior of the individuals is a viable and logical approach, it does not help the caregivers to manage their own stress and burnout.

A second approach involves providing training that enables caregivers to better manage their work-related psychological distress. For example, Noone and Hastings (2009, 2010) showed that when caregivers participate in Promotion of Acceptance in Carers and Teachers training, it enables them to significantly decrease their psychological distress even when faced with a slight increase in occupational stress. This training included three key components of Acceptance and Commitment Therapy (Hayes et al., 1999)—acceptance, cognitive mindfulness, and values clarification. In a similar approach, Brooker et al. (2013) showed that when caregivers participate in occupational mindfulness (OM) training, they are able to decrease their stress, enhance psychological well-being, and increase job satisfaction. The OM training includes mindfulness practices, aspects of positive psychology (e.g., signature strengths; Seligman, 2002), and various cognitive therapy exercises.

A third approach is to enhance the ability of the caregivers to skillfully use behavior management strategies and to learn ways of reducing their own occupational stress. For example, Singh et al. (2009) added a mindfulness-based training to the existing training in behavior management principles for caregivers, and demonstrated positive changes in the behaviors of both the caregivers and the individuals in their care. Brooker et al. (2014) essentially replicated these results using a different set of mindfulness-based training procedures. In a small multiple-baseline design study, Singh et al. (2015) evaluated the effects of an integrated mindfulness-based training with PBS training (i.e., Mindfulness-Based Positive Behavior Support [MBPBS]; Singh et al., 2016a). The results suggested that the MBPBS training enabled the caregivers to greatly reduce their psychological stress, eliminate staff turnover, and substantially reduce and then eliminate the use of physical restraints with individuals who evinced aggressive behavior. In a proof-of-concept quasi-experimental design study, Singh et al. (2016b) further evaluated the effectiveness of MBPBS training for caregivers. The results corroborated earlier findings of lowered caregiver psychological stress and staff turnover, and significantly less use of physical restraints. Furthermore, in both studies, a benefit-cost analysis showed substantial financial savings for the agency due to their staff participating in the MBPBS training (Singh et al., 2015, 2016b).

These studies strongly suggest that caregivers can learn to regulate their emotions more effectively through MBPBS training than with standard agency in-service training. Furthermore, enhanced caregiver emotional regulation appears to have benefits for the caregivers (e.g., reduced psychological stress and staff turnover), as well as for those in their care (e.g., reduced engagement in aggressive behavior). These studies focused on caregivers who provided services to individuals with IDD who functioned at mild to moderate levels but did not include those who functioned at severe and profound levels. In addition, these studies did not use control groups to compare the effects of the different training using a robust experimental design. Thus, the aim of the present study was to evaluate the comparative effectiveness of MBPBS and Training-as-Usual (TAU) for caregivers in a congregate care facility for individuals with severe and profound IDD, using a randomized controlled

trial (RCT). The comparative effects of the two training conditions were assessed in terms of caregiver variables (i.e., use of physical restraints, use of stat [emergency] medications, perceived psychological stress), a care recipient variable (i.e., number of aggressive events), and agency variables (i.e., 1:1 staffing of individuals with aggressive behavior, staff turnover, benefit-cost of the two trainings).

individuals exhibited these behaviors at a level of severity and/or frequency that required the caregivers to implement formal behavior management plans. Of the 34 with formal behavior management plans, up to 10 of them required 1:1 staffing on any given day for aggressive behavior toward staff and/or peers. A 1:1 staffing level was used for the safety of the individual, staff and peers.

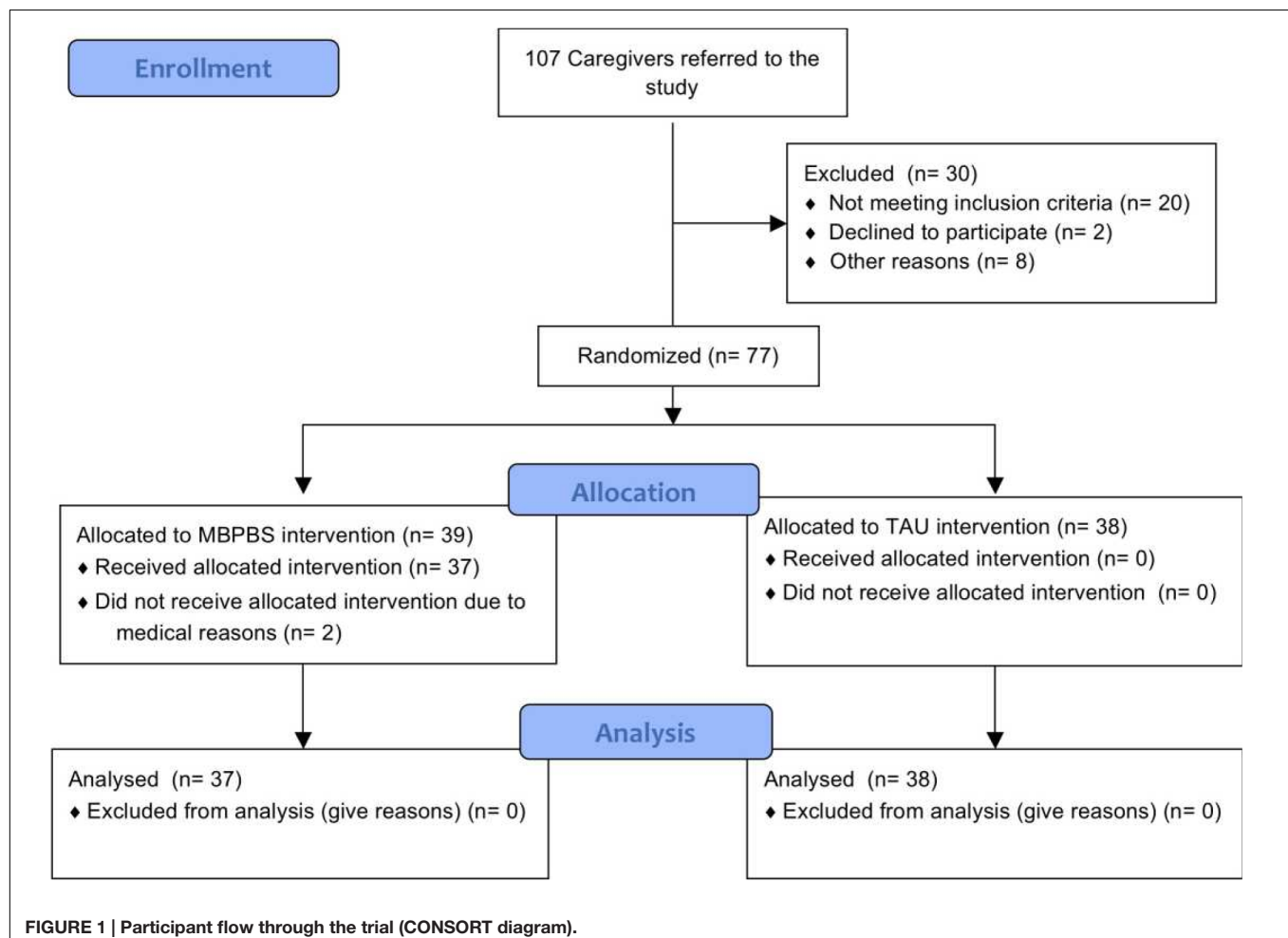
MATERIALS AND METHODS

Setting

The study was conducted at a large congregate care facility for individuals with IDD. The agency served individuals who were at the severe and profound levels of functioning and exhibited varying levels of challenging behaviors (e.g., physical aggression, property destruction, pica, rumination, stereotypy). The individuals resided in six homes, with each home accommodating between 6 and 10 individuals (mean = 8 per home). In total, there were 48 long-term beds, all of which were filled except when an individual needed short-term admission at a local hospital for acute medical care. All individuals exhibited challenging behaviors, but only 34 of the 48

Participants

As a part of the facility's consumer engagement plan for enhancing the health and wellness of individuals with IDD, the entire caregiver staff was required to receive additional in-service training. All 107 caregivers were enrolled, of which 30 did not meet the inclusion criteria (i.e., full-time employment, consent to participate in the training, and availability during the training). Using a random number generator, the remaining 77 caregivers were randomized into MBPBS or TAU conditions. Of the 39 caregivers randomized to the MBPBS group, 2 dropped out before the study due to personal reasons (i.e., one due to change of job and the other due to late stage pregnancy). None of the 38 caregivers randomized to the TAU group dropped out. **Figure 1** presents a CONSORT participant flow diagram.



The sociodemographic data for both the caregivers and the individuals with IDD in their care are presented in **Table 1**.

Ethics Statement

All training procedures in the study were in accord with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Written informed consent was obtained from all caregivers who participated in the study. The provider agency, caregivers, and facility review committee approved the MBPBS and TAU training and data collection.

Procedure

Experimental Design

A RCT was conducted to evaluate the comparative effectiveness of MBPBS vs. TAU on caregiver, individuals with IDD, and agency variables. Unless otherwise stated, standard agency data collection procedures were followed as part of the study protocol.

Interventions

Mindfulness-Based Positive Behavior Supports

The standard 7-day MBPBS protocol, as reported by Singh et al. (2015, 2016b), was used. The training was presented in three parts, spread over a 10-week period. Part I lasted one 8-h day, Part II was five 8-h days (i.e., 40 h), and Part III was one 8-h day. All 37 caregivers in this condition received all the MBPBS training in a group format. **Table 2** presents the MBPBS program and a brief outline of each day's training.

Part I was on the first day of the first week of training, during which the caregivers received instructions in and practiced three foundational meditations: Samatha, Kinhin, and Vipassanā. The caregivers received in-depth instructions and practiced the fundamentals of meditation posture. They were instructed to sit comfortably with a straight spine, without slouching or stretching the shoulders, with their head tilted slightly forward, eyes slightly open or closed, with the tip of their tongue lightly touching the upper palate, the right hand resting over the left hand on the lap, with thumbs just touching, and breathing evenly (Buksbazen, 2002). They were taught to focus on their breathing, without

deliberately changing the length of each breath. They learned to count an inhalation and exhalation as one breath until they reached 10 breaths, before restarting the counting cycle. They were taught to simply observe their discursive thoughts and emotions, without interacting with them or trying to suppress them. That is, they were required to focus their awareness on whatever took place in their mind without judgment or engagement. Samatha meditation is the foundational meditation that provides the practitioner with the stability of mind on which to build all other meditation practices. In addition, they were taught Kinhin (walking) meditation and Vipassanā (insight) meditation (McDonald, 2005). Kinhin is a walking meditation that enables a person to be in the present moment while walking slowly and mindfully. Vipassanā meditation is used to gain insight into the true nature of reality through mindfulness of breathing, thoughts, feelings, and actions (Shonin et al., 2015). Toward the end of the first day of training, all caregivers were instructed to develop a personal meditation practice, beginning with a few minutes each day and incrementally increasing it until they reached between 20 and 30 min of daily practice. Finally, they were required to log their daily meditation practice. The caregivers were required to practice the three meditations daily until training in Part II that was scheduled for the fifth week.

During Part II (days 2 to 6 of training), the caregivers received instructions on the nature of the Four Immeasurables (equanimity, lovingkindness, compassion, and empathetic joy), and meditation practices in equanimity, lovingkindness, compassion, and empathetic joy (Kyabgon, 2004). They also received instructions on the concepts and application of the beginner's mind (Suzuki, 1970), being in the present moment, the three poisons—attachment, anger and ignorance (Kyabgon, 2004), shenpa and compassionate abiding (Chödrön, 2007, 2010; Kongtrül, 2008), meditation on the Soles of the Feet (Singh et al., 2011b), and the general concept of emotion regulation and application of the various meditations in the caregivers' work and private life.

Part III was scheduled on the first day of the 10th week (i.e., seventh full day of training), for follow-up, wrap-up,

TABLE 1 | Socio-demographic characteristics of the caregivers and individuals with IDD in their care for the Mindfulness-Based Positive Behavior Supports (MBPBS) and Training-as-Usual (TAU) conditions.

	MBPBS		TAU	
	Caregivers	Individuals with IDD	Caregivers	Individuals with IDD
Number of participants	37	24	38	24
Mean age in years (SD)	43.05 (10.39)	39.21 (7.61)	45.08 (7.87)	42.33 (9.22)
Age range (years)	23–62	27–54	25–59	24–57
Gender: males	14 (37.83%)	16 (66.67%)	10 (26.32%)	16 (66.67%)
Level of functioning				
Severe	na	9 (37.5%)	na	7 (29.17%)
Profound	na	15 (62.5%)	na	17 (70.83%)
Number of individuals on psychotropic medications	na	20 (83.33%)	na	19 (79.16%)
Number of individuals with mental illness	na	20 (83.33%)	na	19 (79.16%)
Number of individuals with behavior plans for aggressive behavior	na	18 (75%)	na	16 (67%)

na, not applicable.

TABLE 2 | Outline of the 7-day MBPBS program.**PART I****Day 1**

(First 1-day training)

Samatha meditation
 Kinhin meditation
 Vipassanā meditation
 Five hindrances—sensory desire, ill will, sloth and torpor, restlessness and remorse, and doubt
 Daily logs and journaling

PART II**Day 2**

(First day of 5-day intensive training)

Review of meditation practice
 Introduction to the Four Immeasurables (*Brahmavihara*: *metta*—lovingkindness; *karuna*—compassion; *mudita*—empathetic joy; *upekkha*—equanimity)
 Equanimity meditation
 Beginner's mind
 Applications to PBS practice

Day 3

Review of day 2 instructions and practices
 Further instructions on the Four Immeasurables
 Equanimity meditation
 Lovingkindness meditation
 Being in the present moment
 Applications to PBS practice

Day 4

Review of days 2 and 3 instructions and practices
 Further instructions on the Four Immeasurables
 Equanimity meditation
 Lovingkindness meditation
 Compassion meditation
 The three poisons—attachment, anger, and ignorance
 Applications to PBS practice

Day 5

Review of days 2 to 4 instructions and practices
 Further instructions on the Four Immeasurables
 Equanimity meditation
 Lovingkindness meditation
 Compassion meditation
 Joy meditation
 Attachment and anger—shenpa and compassionate abiding meditations
 Applications to PBS practice

Day 6

Review of days 2 to 5 instructions and practices
 Review and practice Samatha, Kinhin, and Vipassanā meditations
 Review of the Four Immeasurables
 Practice equanimity, lovingkindness, compassion, and joy meditations
 Attachment and anger—meditation on the soles of the feet
 Review of applications to PBS practice
 Review of the MBPBS training program

PART III**Day 7**

(Second 1-day training)

Review of the meditation instructions and practices (daily logs)
 Review and practice Samatha, Kinhin, and Vipassanā meditations
 Review of the Four Immeasurables
 Practice equanimity, lovingkindness, compassion, and joy meditations
 Emotion regulation and anger—meditation on the soles of the feet
 Instructions for practicing three ethical precepts—refrain from (a) harming living creatures, (b) taking that which is not given, and (c) incorrect speech
 Applications to PBS practice
 Review of the 7-day MBPBS training program

and follow-through meditation practices. This involved further meditation practice, review of the caregivers meditation practices and experiences, questions and answers from the group, and how the caregivers would continue their practice till the end of week 40—when formal aspects of the study concluded—and beyond.

For the PBS component of the MBPBS training, Part I was devoted to ascertaining the current knowledge of the caregivers in the principles and practice of PBS, and collaboratively developing a training program in PBS within the context of mindfulness-based practices. The PBS training program was informed by current literature on PBS (Morris and Horner, 2016) and staff training in PBS (MacDonald, 2016), the caregivers' lived experience of working with individuals with IDD who periodically engaged in high-intensity but low frequency aggressive behavior, and the seamless interface with the caregivers' personal practice of mindfulness. During Part II (i.e., the 5-day training), the caregivers were instructed in the following five components of standard PBS plans: setting event strategies, preventive strategies, teaching strategies, consequence strategies, and quality of life outcomes (Lucyshyn et al., 2015). In terms of interfacing with their mindfulness practices, they were given instructions on mindful observation of the individual's behavior, mindful communication (with a focus on mindful prompting and feedback), mindful pause between requests and prompts, and mindful use of reinforcement contingencies that focused on the rate, quality, magnitude, delay, and specificity of the reinforcement delivered contingently and non-contingently to the individuals with IDD in their care (Singh et al., 2016a). Part III (i.e., the seventh full day of training), involved a review of the mindfulness-based PBS practice, discussion of the need for formal PBS programs, questions and answers regarding the MBPBS practice, and follow-through PBS practices.

Training-as-Usual

The TAU in-service training followed the same three-Part, 7-day training timeline as in the MBPBS training. The training staff of the provider agency provided this training. It covered the following general areas of applied behavior analysis: definitions and characteristics; principles, processes and concepts; behavioral assessment; evaluation of outcomes; development and implementation of behavior management plans; and ethical considerations in using a behavioral approach to interventions (Cooper et al., 2007; Mayer et al., 2012). The caregivers were given instructions in reading, understanding, and implementing behavior management plans, observing the implementation of behavior support plans by expert staff, role playing implementation of PBS plans, and getting feedback on their implementation efforts. Further, they discussed their current behavior support plans, their effectiveness, fidelity of implementation, data collection, graphing the data, and revisions based on the data.

Training Adherence

For both training conditions, the caregivers' attendance at the 7-day training program was documented. In addition, caregivers in both training conditions were requested to record the time they spent in daily meditation during the 40 weeks of the study.

All caregivers in both training conditions attended and fully participated in the 7 days of training. The daily logs showed that all 37 caregivers in the MBPBS training condition began their practice of meditation following Part I training (i.e., first 1-day training) and continued throughout the 40 weeks of the study. The duration of meditation gradually increased from a few minutes during Part I and averaged 15 min by the end of training in Part II (i.e., the 5-day training). There was a further increase following the 5-day training, and this averaged 33 min by the end of training in Part III. Thereafter, the caregivers averaged between 25 and 40 min of daily meditation, with occasional meditation holidays. Overall, on average, the caregivers in the MBPBS condition meditated for 89% (range: 0–96%) of the days. Two caregivers from the TAU training condition had a personal meditation practice prior to the study and both continued their meditation practice during the 40 weeks of the study. On average they meditated between 20 and 30 min daily, with occasional breaks from meditation.

Trainers

The MBPBS trainer was an experienced behavior analyst at the BCBA-D level, with over 35 years of hands-on experience in developing and implementing behavior support plans. In addition, the trainer had a 40-year personal meditation practice and experience in the mindful delivery of services in behavioral health. Segments of training in Parts I, II, and III were videotaped and 10 randomly selected segments of 10–15 min from each day of the 7-day training (i.e., 70 training segments) were rated for fidelity of training by another qualified meditation trainer who was also an expert in PBS. The fidelity of the MBPBS training was rated at 100% for both the meditation instructions and the training in PBS.

The TAU trainer was an experienced behavior analyst at the BCBA level, with over 20 years of training experience in behavior management. This trainer did not have a personal meditation practice. As with the MBPBS training, segments of training in Parts I, II, and III were videotaped and 10 randomly selected segments of 10–15 min from each day of the 7-day training (i.e., 70 training segments) were rated for fidelity of training by another qualified trainer in behavior management. The fidelity of the behavioral training was also 100%.

Measures

Aggressive Events

An aggressive event was defined as an individual hitting, biting, scratching, punching, kicking, slapping, or destroying property. Staff recorded each instance of an aggressive event on an incident reporting form at the point of occurrence and this was later entered in the facility's incident management database. By policy, each incident was double-checked by the home supervisor for occurrence and accuracy of reporting. The reliability of reporting and logging the occurrence of aggressive events was 98% (range: 94–100%).

Physical restraints

A physical restraint was defined as a brief physical hold of an aggressive individual by a caregiver when there was imminent danger of physical harm to the individual, peers or staff, and the

behavior could not be controlled with verbal redirection. Staff recorded each instance of the use of a physical restraint at the point of occurrence and this was later entered in the facility's risk management database. By policy, each use of physical restraint was double-checked by the home supervisor for occurrence and accuracy of reporting. The reliability of reporting and logging the occurrence of physical restraints was 100%.

Stat medicine

Stat medicine is prescribed during medical, psychiatric or behavioral emergencies for the immediate safety of the individual. Stat medication was defined as an emergency medication prescribed for the immediate calming of an individual who was aggressive and could not be managed by other means, including physical restraints. Each prescription was counted as one event as recorded by a registered nurse in the individual's Medication Administration Record (MAR). Only those prescriptions that were prescribed specifically as emergency medication for aggressive behavior were counted.

One-to-one staffing

A 1:1 staffing is the level of supervision used when an individual needs close attention for a specific reason and it is designed to ensure the safety of the individual, peers or staff. For the purpose of this study, 1:1 staffing was defined as the level of enhanced observation ordered by a physician or psychologist for an individual with IDD who evinces aggressive behavior. Each individual's treatment team determined the need for level of supervision, the nursing administration assigned the staff, and the home manager ensured the provision of level of supervision on a shift-by-shift basis. Level of supervision staff was recorded as being present for the assigned duties 100% of the time.

Staff stress

Caregivers in both training conditions completed the Perceived Stress Scale-10 (PSS-10; Cohen et al., 1983) as a measure of perceived stress at two time points: on the first and last day of the 40-week study. The PSS-10 provides an index of the degree to which people perceive their lives as stressful and indicates how often they have found their lives to be unpredictable, uncontrollable, and overloaded in the last month. This rating scale includes items such as, "In the last month, how often have you found that you could not cope with all the things that you had to do?" The caregivers responded to 10 questions on a five-point scale, ranging from 0 (never) to 4 (very often). Their responses were summed to create a psychological stress score, with higher scores indicating greater psychological stress. The PSS-10 has adequate psychometric characteristics (Cohen and Williamson, 1988). In the present study, Cronbach's alpha was 0.82, indicating good reliability.

Staff turnover

The facility's Human Resources Department provided the staff turnover data, which included all instances of any staff member leaving the employment of the agency due to staff injury on the worksite during the 40 weeks of the study period. Data were extracted only for the fulltime staff involved in the MBPBS and TAU training conditions.

Cost effectiveness data

The facility's Finance Department provided cost data on (1) work days lost due to staff injury, (2) instances of 1:1 staffing, (3) staff needing medical and physical rehabilitation therapy due to injury, (4) staff resigning due to staff injury who were replaced, (5) staff required for MBPBS and TAU training, and (6) temporary staff required during MBPBS or TAU training. All costs were included, regardless of whether the costs were borne by the agency or by workers' compensation.

Data Analyses

The effectiveness of the MBPBS and TAU conditions were examined in several ways. For several variables, the unit of analysis was a count variable for an entire condition, not for individuals within a condition. These examples included the number of aggressive events per week, the number of uses of physical restraints per week, the number of emergency stat medicine prescriptions per week, and the number of additional 1:1 staffing needed per week. As these variables are not at the individual level, they do not lend themselves to traditional analyses for RCTs. Therefore, we employed two strategies to examine change across time within conditions and differences across groups.

First, we were able to examine change across time within each condition by treating each group as an n of 1. In doing so, we plotted the count of each variable for each condition across all weeks of the study. Second, we averaged counts per week across the 10-week Training phase and the 30-week post-training phase, respectively, for each condition. The resulting M 's (with SD 's) could then be compared across groups by phase with independent samples t -test (and corresponding Cohen's d values as effect size measures). Using paired samples t -test, we compared change across phases within each condition. Note that alternative approaches, such as use of a mixed-model ANOVA, were not possible because the unit of analysis was not individuals.

The data on perceived stress were unique to individuals. Therefore, we used a mixed-model ANOVA to compare main effects of condition, time, and their interaction. Effect sizes reported include η^2 for an overall effect size, and Cohen's d 's for direct comparisons for a specific phase across conditions, or for a specific condition across time.

RESULTS

Demographic Variables

We ran a series of Chi-Square and Independent Samples t -test to compare demographic characteristics of participants in the MBPBS and TAU conditions. There were no statistical differences between the groups (all p 's > 0.05).

Caregiver Variables

Perceived Stress

Figure 2 shows there was a decrease in the perceived stress score from the first day of Training (Time 1) to the last day of post-training (Time 2) of 36.15% in the MBPBS condition and 9.02% in the TAU condition. Differences across time and between

conditions on PSS were examined with a 2 (condition: MBPBS vs. TAU) \times 2 (timepoint: pre vs. post) mixed model ANOVA. The results revealed a significant interaction, $F(1,73) = 73.70$, $p < 0.001$ ($\eta^2 = 0.50$). **Figure 2** shows there was no significant difference between groups on the first day of Training (pre) phase (Cohen's $d = 0.36$), but there was a significant difference, with a large effect size, on the last day of the post-training phase (Cohen's $d = 2.78$). The within-groups effect sizes from pre to post for the MBPBS and TAU conditions were as follows: Cohen's $d = 2.60$ and 0.70 , respectively.

Physical Restraints

As evident in **Figure 3**, the weekly use of physical restraints decreased substantially from the Training to the post-training phase for the MBPBS condition, while there was no such decrease in the TAU condition. On average, the use of physical restraints in the MBPBS condition was 8.00 (range = 2–13) per week during the Training phase and 0.53 (range = 0–3) in the post-training phase. Similarly, on average, the use of physical restraints in the TAU condition was 13.60 (range = 9–17) per week during the Training phase and 10.77 (range = 5–16) in the post-training phase. During the Training phase, there was a statistically significant difference in physical restraint use per week across the MBPBS ($M = 8.00$, $SD = 4.06$) and TAU conditions ($M = 13.60$, $SD = 2.72$), $t(18) = 3.62$, $p = 0.002$ (Cohen's $d = 1.62$). During the post-treatment phase, use of physical restraints was lower in the MBPBS condition ($M = 0.53$, $SD = 0.90$) than in the TAU condition ($M = 10.77$, $SD = 3.21$), $t(58) = 1.73$, $p = 0.10$ (Cohen's $d = 4.34$).

Stat Medication

As shown in **Figure 4**, the weekly use of stat medications decreased substantially from the Training to post-training phase for the MBPBS condition, while there was no such decrease in the TAU condition. On average, the use of stat medication in the MBPBS condition was 6.40 (range = 2–11) per week during the Training phase and 0.23 (range = 0–3) in the post-training phase. Similarly, on average, the use of stat medication in the TAU condition was 8.60 (range = 5–12) per week during the Training phase and 7.17 (range = 2–14) in the post-training phase. During the Training phase, there was no statistically significant difference in mean use of stat medications per week between the MBPBS ($M = 6.40$, $SD = 3.24$) and TAU conditions ($M = 8.60$, $SD = 2.37$), $t(18) = 1.73$, $p = 0.10$ (Cohen's $d = 0.77$). During the post-training phase, the use of stat medications in the MBPBS condition was significantly lower ($M = 0.23$, $SD = 0.68$) than in the TAU condition ($M = 7.17$, $SD = 3.30$), $t(58) = 11.28$, $p < 0.001$ (Cohen's $d = 2.91$).

Care Recipient Variable

Aggressive Events

Figure 5 shows the number of aggressive events by each training condition. On average, in the MBPBS condition there were 21.75 (range = 19–27) aggressive events per week during the Training phase and 5.91 (range = 0–21) in the post-training phase. Similarly, on average, in the TAU condition there were 22.25 (range = 17–26) aggressive events per week during the

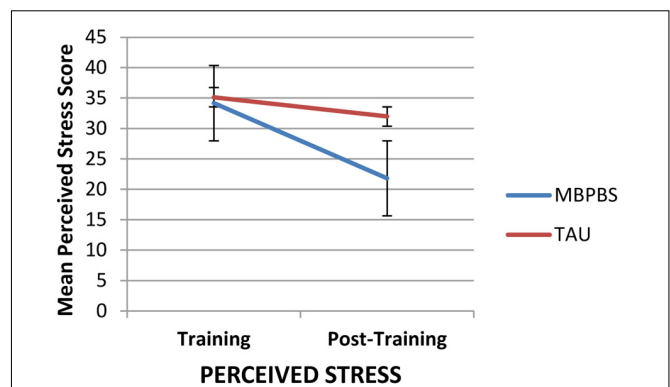


FIGURE 2 | Ratings of caregiver perceived stress on PSS-10 on the first day of training and the last day of post-training in the two conditions, Mindfulness-Based Positive Behavior Supports (MBPBS) and Training-as-Usual (TAU). Note that higher scores indicate greater psychological stress. Error bars report standard error of the mean.

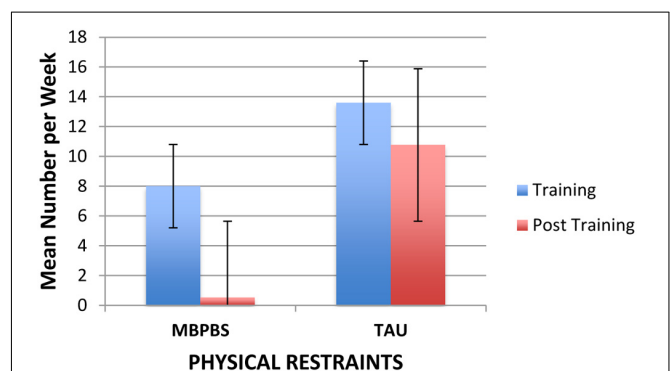


FIGURE 3 | Mean number of physical restraints per week used by caregivers contingent on aggressive behavior of the individuals in the MBPBS and TAU conditions. Error bars report standard error of the mean.

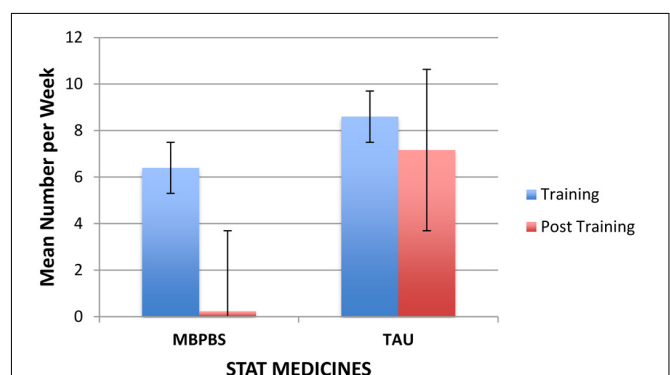
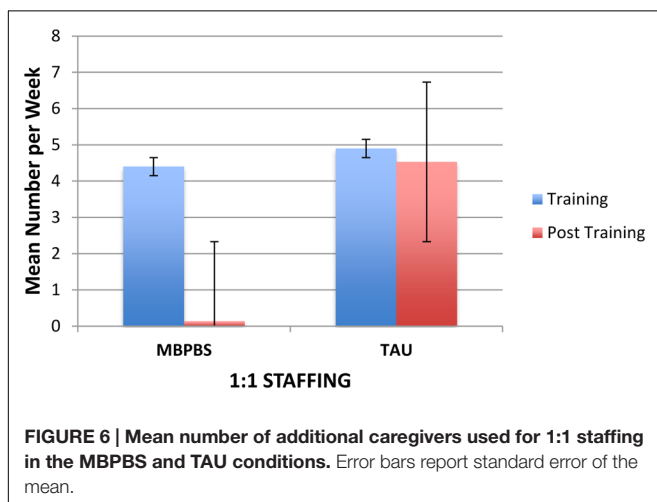
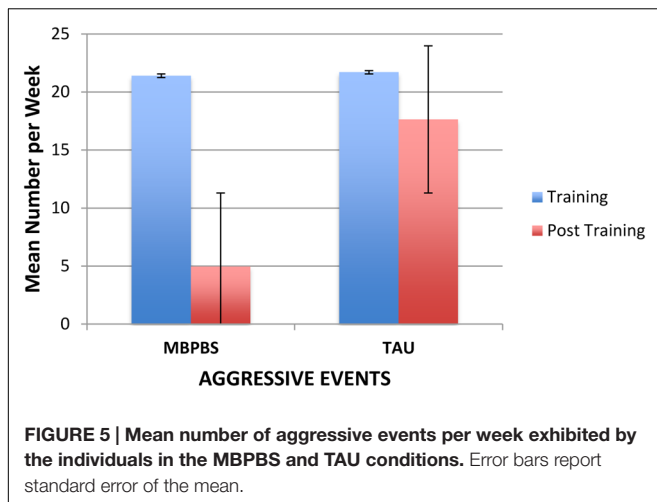


FIGURE 4 | Mean number of stat medicines per week used by caregivers contingent on aggressive behavior of the individuals in the MBPBS and TAU conditions. Error bars report standard error of the mean.

Training phase and 17.75 (range = 10–24) in the post-training phase. There was no significant difference between the MBPBS ($M = 21.40$, $SD = 2.76$) and TAU conditions ($M = 21.70$,



$SD = 3.71$), $t(18) = 0.21$, $p = 0.84$ (Cohen's $d = 0.09$) during the Training phase. However, the difference between conditions was significant in the post-training phase, $t(58) = 11.01$, $p < 0.001$ (Cohen's $d = 2.84$), with the MBPBS condition ($M = 4.97$, $SD = 4.86$) demonstrating fewer aggressive events than the TAU condition ($M = 17.63$, $SD = 4.01$).

Agency Variables

1:1 Staffing

Figure 6 shows the average 1:1 staffing (in addition to regular staffing) required for the care, safety and protection of staff and peers from individuals with IDD who were periodically aggressive. In the MBPBS condition, on average, 4.40 ($SD = 1.78$; range = 2–7) additional staff was required each week during Training and 0.13 ($SD = 0.35$; range = 0–1) during post-training. In the TAU condition, on average, 4.90 ($SD = 1.45$; range = 3–7) additional staff was required each week during Training and 4.53 ($SD = 1.59$; range = 3–9) during post-training. The difference between the two groups during the Training phase was not statistically significant, $t(18) = 0.69$, $p = 0.50$, but the difference

between MBPBS vs. TAU post-training phases was statistically significant, $t(58) = 14.80$, $p = 0.0001$.

Staff Turnover

In terms of staff turnover, no caregiver in the MBPBS condition resigned due to injury and stress during the Training or post-training phases. Three caregivers resigned (all due to injury) during the Training phase in the TAU condition and 11 caregivers resigned (seven due to injury, four due to stress and injury) during the post-training phase. The difference between the MBPBS ($M = 0.00$, $SD = 0.00$) and TAU ($M = 0.30$, $SD = 0.48$) Training phases was not statistically significant, $t(18) = 1.98$, $p = 0.06$. During the post-training phase, the difference between the MBPBS ($M = 0.00$, $SD = 0.00$) and the TAU ($M = 0.37$, $SD = 0.67$) conditions was statistically significant, $t(58) = 3.02$, $p = 0.004$.

Cost Effectiveness

Table 3 presents the cost effectiveness data for the MBPBS and TAU conditions. When compared to the TAU condition, the number of lost days of work due to staff injury was reduced by about 92% in the MBPBS condition, with a savings of \$82,992.00. Commensurate with this savings, there was additional savings of \$72,128.00 during the 40 weeks of the study in terms of additional costs of 1:1 staffing. The cost of medical and physical rehabilitation therapy services for the two injured staff during the MBPBS condition was \$39,000.00 compared to \$351,000.00 for the 18 staff injured during the TAU condition. While no additional costs were incurred in the MBPBS condition due to any staff turnover during the 40-week study period, the cost of training 14 new staff in the TAU condition was \$17,680.00. The cost of providing alternate (temporary) staff for those who were in the MBPBS training was \$41,440.00 compared to \$42,560.00 in the TAU condition. Finally, the cost of providing the MBPBS training was \$30,000.00 compared to \$2,000.00 for TAU training. Overall, when compared to the TAU condition, there was a savings of \$457,920.00 for an equivalent period during the MBPBS condition; that is, a savings of over 78%.

DISCUSSION

Caregivers in mental health provide services for individuals with diverse abilities and challenging behaviors that often sap their mental and physical resources, and lead to stress and burnout. The traditional solution has been to teach caregivers specialized techniques, such as behavior analytic skills, that will help them to be more effective in managing the challenging behaviors of the individuals in their care (MacDonald, 2016; McIntyre and Neece, 2016). While this approach has been found effective, caregivers often work in situations where such skills alone may not be enough to curb their stress and burnout, because of the intensity and frequency of the challenging behaviors they face, multiple individuals engaging in challenging behaviors, the shortage of well-trained staff when the need arises, and the emotional toll of such work (Hastings, 2002; Allen et al., 2005; Crawford et al., 2010; Deveau and McGill, 2014).

TABLE 3 | Comparative costs for 40 weeks of MBPBS training condition ($n = 37$) compared to 40 weeks of TAU training condition ($n = 38$) for the human services variables.

	Cost Variables		Cost	
	MBPBS	TAU	MBPBS	TAU
Lost days of work and cost due to staff injury	42	536	\$7,056.00	\$90,048.00
Number of staff-days and cost of 1:1 staffing	96	740	\$10,752.00	\$82,880.00
Number and cost of staff needing medical and physical rehabilitation therapy	2	18	\$39,000.00	\$351,000.00
Number of staff resigned due to staff injury and training costs for new hires	0	14	\$0.00	\$17,680.00
Number of training days and cost of training	10	10	\$30,000.00	\$2,000.00
Cost of temporary staff during training	37	38	\$41,440.00	\$42,560.00
Total additional costs for the two time periods			\$128,248.00	\$586,168.00
Total overall savings				\$457,920.00

An emerging approach involves not only enhancing the management skills of the caregivers in terms of the needs of the individuals they serve, but also teaching them self-management skills that enhance the caregivers' psychological well-being, thereby making them more resilient in their work situation (Noone and Hastings, 2009, 2010; Brooker et al., 2013, 2014). For example, a series of exploratory proof-of-concept studies reported that adding mindfulness-based skills to caregivers previously or concurrently trained in PBS reduces their stress and burnout, as well as the challenging behaviors of those in their care (Singh et al., 2009, 2015, 2016b). Related studies have supported the notion that caregivers and their clients with disabilities mutually benefit when the caregivers are given training in mindfulness-based approaches (Brooker et al., 2013, 2014).

The present study strengthens the evidence-base for this approach. In a RCT, the current study demonstrated multiple beneficial effects of training caregivers in MBPBS compared to TAU in a congregate care long-term care facility for individuals with IDD. First, with regard to caregiver variables, the effects of the MBPBS training were evident in terms of statistically significant reductions in perceived psychological stress and job resignations due to stress and work-related injury. In addition, following training in MBPBS, staff greatly reduced using physical restraints and stat medicines contingent on the individuals' aggressive behaviors. These findings confirm earlier reports of significant reductions in the use of physical restraints and emergency medications for individuals with aggressive behavior following training of caregivers in MBPBS (Singh et al., 2015, 2016b). Furthermore, Brooker et al. (2014) reported similar findings in terms of reduced staff use of PRN ("as needed") medicines for behavior control, seclusions, and emergency chemical restraints following training in a mindfulness-based training program. Any reduction or elimination of the use of restrictive procedures indicates an enlightened approach to the care of people with diverse abilities, particularly those with IDD (Singh, 2016).

Second, with regard to the care recipient variable, reductions in aggressive incidents were significantly greater with the individuals receiving care from caregivers trained in MBPBS, than with those receiving care from caregivers trained in

TAU. Caregivers in the MBPBS training condition did not receive instructions on how to manage the behavior of specific individuals in their care who engaged in aggressive behavior. In addition, existing behavior management plans for aggressive behavior were not reviewed or revised as a component of the MBPBS training condition. Given that the only difference between the MBPBS and TAU conditions was the nature of the training the caregivers received, the significantly reduced frequency of aggressive behavior evident in the MBPBS condition could be attributed to the personal change in the caregivers due to training in MBPBS. Similar reductions in incidents of verbal and physical aggression were reported in previous studies in which staff was trained in MBPBS (Singh et al., 2009, 2015, 2016b). All these findings suggest that training in MBPBS may change the very nature of the reciprocal interactions between the caregivers and the individuals in their care, moving them from a negative to a positive trajectory (Sameroff, 1995).

Third, with regard to agency variables, there was a significant reduction in the assignment of 1:1 staffing of individuals with aggressive behavior in the MBPBS condition when compared to the TAU condition. Indeed, when compared to the TAU condition, 1:1 staffing was rarely used following caregiver training in MBPBS. There was no staff turnover in the MBPBS condition compared to 14 in the TAU condition. Finally, the cost-effectiveness data showed substantial savings in the MBPBS condition when compared to the TAU condition. Costs were estimated for several standard variables (i.e., lost days of work due to staff injury, 1:1 staffing, treatment related costs for work-related staff injury, hiring and training of new staff, MBPBS and additional TAU training costs, temporary staff during training days) during the study period. There was an overall savings of 78.12% with the MBPBS training condition compared to the TAU training condition. Except for the costs of MBPBS training, which was much higher than for TAU, cost savings were realized on all other key variables in the MBPBS condition. Although the overall cost savings were somewhat less in the present study, they still aligned well with those from previous studies that reported savings 87.75% (Singh et al., 2015) and 89% (Singh et al., 2016b). Coupled with the enhanced psychological well-being of the caregivers and reduced aggressive behavior of the care recipients, these cost savings suggest that MBPBS

may be a clinically useful and financially viable training for caregivers.

The results of this RCT indicate that the traditional training provided in large facilities and community group homes for individuals with IDD may not be very effective in ameliorating the stress and burnout of the caregivers. While such training may enable caregivers to provide the required services detailed in each individual's Individual Support Plan (ISP), the training is not very effective in assisting caregivers to effectively manage the behavior of individuals who engage in severe challenging behaviors, such as aggression, property destruction, and self-injury (Didden et al., 2016; Hoch et al., 2016). Furthermore, traditional training falls short in another respect—it does not teach caregivers how to successfully respond to workplace stress, compassion fatigue, and burnout. It is inevitable that caregivers will be stressed in large congregate care facilities for people with disabilities because of inherent demands in the job. Thus training should encompass strategies that enable caregivers to change their relationship to the job demands. Fortunately, mindfulness-based training does just that by teaching caregivers how to respond differently to the same daily work stresses, because they are typically not in position to change the nature of the job requirements (Hwang and Singh, 2016).

The MBPBS training enabled staff to respond in a calm and mindful way rather than to react negatively to the challenging behaviors of the individuals (Singh et al., 2016a). We suspect that disciplined meditation practice enables the caregivers to gradually change their relationship to their perceived mental and emotional experiences that arise when providing care to the individuals. This ability to step back and observe their thoughts and emotions as they occur results in cognitive, emotional and behavioral flexibility which helps them to respond more adaptively to difficult situations, thus reducing psychological stress and burnout (Shapiro et al., 2006). Indeed, it is likely that this metacognitive awareness enables caregivers to distance themselves from their reactive thoughts and emotions, and perceive difficult situations as transient mental events (Safran and Segal, 1990). The Samatha and Kinhin meditations in the MBPBS training enable the caregivers to pay attention to the individuals with non-judgmental awareness, supporting them to work through their ISPs rather than by controlling their challenging behaviors. The meditations on the Four Immeasurables enable the caregivers to develop more equanimity in the face of daily work hassles and stresses, view the individuals and other staff with lovingkindness and compassion, and to demonstrate empathic joy as events unfold. Their training in adopting a beginner's mind enables them to see more possibilities in terms of how to provide care to the individuals with challenging behaviors, buttressed by their training in seeing each individual and each event as if for the first time, without the baggage of history and emotional biases. For example, this mindset enables them to avoid reacting to the challenging behaviors of the individuals based on their premature cognitive commitment to control aggressive behavior through physical restraints and stat medications. While the actual mechanisms of the observed change due to training in mindfulness are yet to be explicated, it is increasingly evident that training staff in

mindfulness produces behavioral changes in the staff as well as the individuals in their care (Eliassen et al., 2016).

In addition to its strengths as a RCT, this study has limitations as well. An important one is the issue of equivalence of the training in each of the two conditions of the study. It can be argued that the MBPBS condition included more training components than the TAU condition and, thus, should produce better outcomes. A standard control group design could not be used because congregate care facilities are required by policy to provide in-service training, thus restricting the comparison to the TAU condition. To address this possible limitation, future studies could evaluate the differential effects of MBPBS against standard behavioral training, the current gold standard of training in the care of individuals with challenging behaviors. Also, this study was executed in a congregate care facility for individuals who functioned at the severe to profound levels of IDD. Whether similar outcomes would be expected in other settings is open to speculation. Previous studies were executed in community group homes, and with individuals who had higher levels of intellectual functioning, factors that could likely produce similar or better results. In addition, it would be instructive to ask the caregivers in the MBPBS condition their views on what and how changes occurred as a consequence of their participation in the meditation practices. Furthermore, future research should investigate the importance of the mindfulness trainer's personal meditation practice and the authenticity of the teachings as factors that may impact outcomes (Carmody and Baer, 2009; McCown et al., 2010).

Mindfulness-Based Positive Behavior Supports provides a paradigm shift in terms of how caregivers and care recipients can be mutually engaged in enhancing the quality of their lives. This model is based on multiple theoretical underpinnings, including Sameroff's (1995) bidirectional transactions, mindfulness (Shonin et al., 2015), PBS (Morris and Horner, 2016), and patient engagement (Institute of Medicine, 2011; Barello et al., 2014; Graffigna et al., 2016), which is known as person-centered planning in the field of IDD (Ratti et al., 2016). In their lexicographic literature review, Barello et al. (2014) noted that the concept of patient engagement has changed over time, and still remains an elusive concept in clinical practice and health care policy. Coulter (2011, p. 10) has suggested that when patients and caregivers work together on patient engagement, they "promote and support active patient and public involvement in health and healthcare and... strengthen their influence on healthcare decisions, at both the individual and collective levels." The MBPBS model is grounded in mutual engagement of both the caregiver and care recipient, and by changing the behavior of one of the pair, the behavior of the other changes because of the bidirectional transactions that occur (Sameroff, 1995; Singh and Singh, 2001). Training caregivers in MBPBS not only elicits and enhances the individual's activation—his or her ability and willingness to self-manage challenging behaviors—but also promotes engagement in positive behavior, as evidenced in the present and previous studies (Singh et al., 2015, 2016b).

There are several implications of this study. First, these findings need further validation in terms of the effects of MBPBS against other proven techniques, such as behavior analytic

strategies, as well as replications by other research teams. Second, the effects of MBPBS need to be assessed in different care settings, such as institutions, group homes, family homes, and within the larger community settings. Third, the effects of MBPBS need to be further evaluated with parents and teachers of individuals with IDD, caregivers of other populations, such as the elderly and individuals with neurodevelopmental disorders. These kinds of studies will help refine the MBPBS model and delineate the boundary conditions for its effectiveness. Furthermore, future research should also focus on dissemination, especially given the finding that few evidence-based practices are actually implemented in real-world settings (Katz, 2010). The challenge is to make MBPBS not too intensive or effortful, but still produces replicable and long-lasting effects at a reasonable cost (Glasgow et al., 2003; Singh, 2016).

In sum, this RCT provides further evidence that MBPBS may be a viable approach to caregiver training to improve the psychological well-being of both caregivers and the care recipients. This study showed that, when provided training in MBPBS, caregivers could significantly reduce their own stress levels and the use of restrictive procedures (e.g., physical restraints and emergency medications) when confronted with challenging behaviors by individuals with intellectual disabilities. When caregivers changed the nature of their care, the individuals

reduced their aggressive behaviors and obviated the need for 1:1 staffing. Finally, the cost effectiveness data suggest that agencies may want to implement MBPBS training not only because of the benefits for the caregivers and care recipients, but also for financial savings.

AUTHOR CONTRIBUTIONS

NS: designed and executed the study, assisted with the data analyses, and wrote the paper. GL: collaborated with the design and writing of the study. BK: analyzed the data and wrote part of the results. JC: collaborated with the design and writing of the study. AW: collaborated in the writing and editing of the final manuscript.

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Communication Skills for Patient Engagement: Argumentation Competencies As Means to Prevent or Limit Reactance Arousal, with an Example from the Italian Healthcare System

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The paper discusses the role of argumentative competencies for the achievement of patient engagement through communication in doctor-patient consultations. The achievement of patient engagement is being proposed by recent studies as a condition that can facilitate in particular patient adherence, which involves behavior change. One obstacle to behavior change that has been observed is reactance, i.e., resistance to persuasive messages when a threat to freedom is perceived. In the medical field, reactance theory has been mostly applied in the field of mental health, less frequently to understand non-adherence in general. However, a few studies have revealed that reactance can actually explain in part the motives behind non-adherence. These studies propose that the arousal of reactance could be limited or prevented by adopting relational measures aimed at giving patients the feeling that they still hold some control over the process of care and that the “impositions” on their freedoms are acceptable because they have had the opportunity to decide about them. However, they do not discuss how these strategies should be operationalized at the dialogical level. A debated issue in the study of reactance is the role played by knowledge. It seems that pure information regarding an issue is likely to represent a threat in itself. Complementary to this is the finding that quality of argument does not impact on the degree of reactance. These findings pose a problem in view of the goal of patient education, itself considered as a necessary premise for any process of patient engagement and adherence. It seems necessary to move away from a conception of education as mere transmission of information and look for more effective ways of transferring knowledge to patients. With regard to this issue, the paper argues that useful insights can be found in studies on science education, in which it is shown experimentally that argumentative processes favor learning and understanding. Drawing on previous studies and taking an interdisciplinary perspective on the issue, the paper brings into the discussion on engagement concepts developed in the field of argumentation theory, showing how the suggestions for avoiding reactance could be realized dialogically.

Keywords: psychological reactance, deliberation dialogue, patient engagement, adherence, patient education

INTRODUCTION

The paper discusses the role of argumentative competencies for the achievement of patient engagement through communication in doctor-patient consultations.

Patient engagement “qualifies the relation that the patient [...] may establish with his/her reference healthcare system [...] in the different phases of the care process” (Graffigna et al., 2015, p. 8). It is a concept borrowed from the marketing and consumer behavior literature to describe consumers’ positive attitudes toward brands or products (Gambetti and Graffigna, 2010). In the healthcare context, it is meant to signify a proactive attitude of patients, understood as “consumers” of healthcare. Such engaged attitude, when successfully achieved, would imply that patients are proactive on two levels: (1) they are able to correctly solicit the healthcare system when in need for assistance; (2) they are able to correctly manage their health condition without improperly referring to the healthcare system (patient autonomy).

In this sense, the concept of engagement takes a step forward in comparison to the notions of compliance, adherence, self-management, patient empowerment and patient activation, as it refers to the relationship between patients and healthcare systems in their complexity. Moreover, it implies a more active view of patients, who are not simply “activated” by their providers, but independently take an active role in the management of the whole process of care that concerns them (Graffigna et al., 2015, pp. 16–20).

Full patient engagement is the result of a gradual process that allows patients to rise from a condition of blackout, to one of eudaimonic project, in which the disease is not the center of attention anymore, but is fully integrated in the patient’s life (Graffigna et al., 2014). In other words, in the eudaimonic project condition (the “perfection” of engagement) patients do not perceive themselves just as “patients,” but as individuals who also have a health condition that requires some attention. But the actions that need to be taken in order to manage this health condition are not felt as impairing as they might have been at the beginning (Graffigna et al., 2014, 2015). In the Patient Health Engagement Model (PHE), Graffigna and colleagues have detailed the phases patients go through in their progress toward the condition of eudaimonic project. The Model is represented in **Figure 1**.

The PHE Model details and relates the cognitive, emotional and behavioral conditions that characterize the pathway from the initial condition of blackout, usually following the first diagnosis, to the condition of being fully engaged and able to integrate the health condition within a life project. As full engagement is the final outcome of this progress, it appears that, especially at the beginning but also along the way, individuals need to be educated to understand their new status as patients, and motivated to take up any new behavior that their health condition requires (Graffigna et al., 2015, pp. 36–38). Thus, it appears that the condition of “being engaged” originates in the one-to-one relationship with healthcare providers (medical doctors, nurses, counselors, etc.) and is triggered by their ability to motivate patients and support their efforts toward behavior change.

Particularly in this respect, “being engaged” should include among other behaviors the ability for patients to more easily adhere to providers’ therapeutic prescriptions and suggestions for healthier lifestyles (Graffigna et al., 2014). As such, engagement is understood as a pre-requisite for adherence (Graffigna et al., 2015, p. 17).

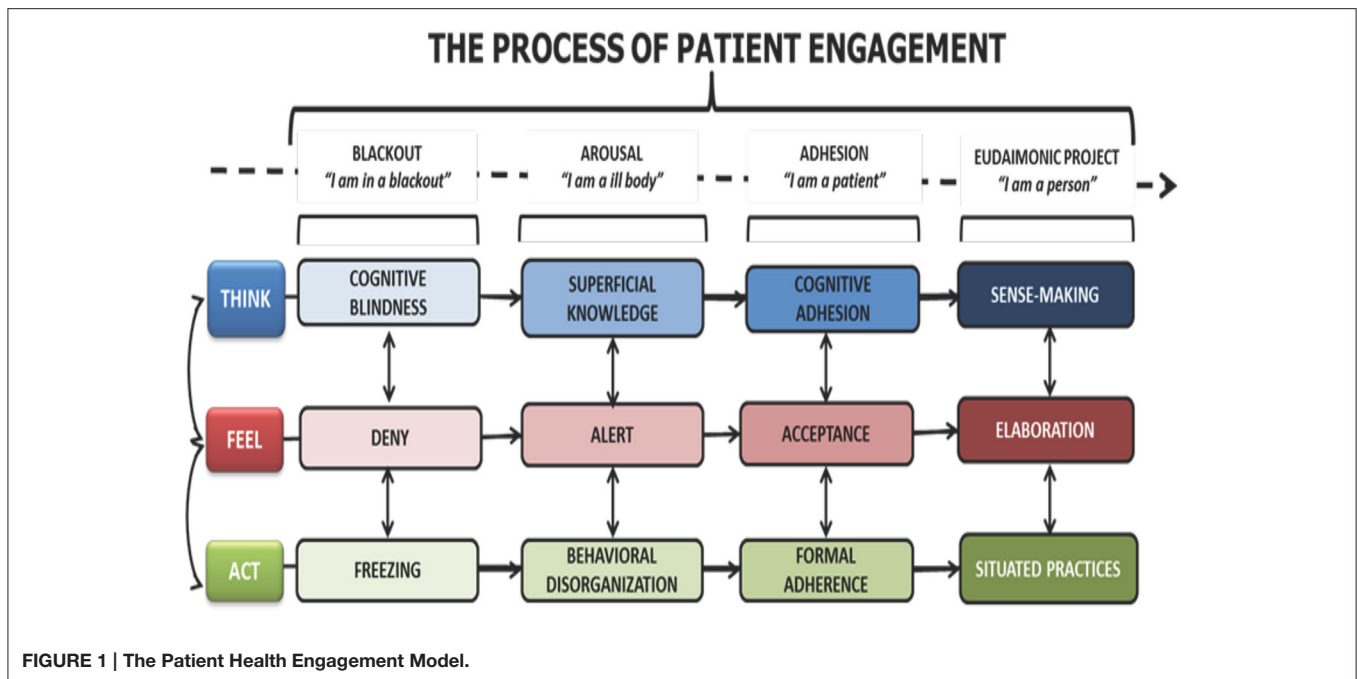
In the following discussion, attention is devoted in particular to the communicative processes that may play a role in the activation of patient engagement. The assumptions on which the discussion is based are that: (1) patient engagement originates in the relationship with one or more healthcare providers in their capacities as representatives of the healthcare system; (2) the tools that support and maintain this relationship are communicative in nature. More specifically, the paper discusses the case of psychological reactance, which can hinder motivation to behavior change and engagement. As such, reactance can be considered as one of the many judgment biases that have been identified by researchers in the behavioral sciences during the past 40 years or so (Fischhoff, 1975; Kahneman et al., 1982; Petty and Cacioppo, 1986). For many of these biases, corresponding debiasing techniques have been studied and tested (Fischhoff, 1982; Arkes, 1991; Lewandowsky et al., 2012; Croskerry et al., 2013). Also drawing on previous studies (Bigi, 2014a, 2015; Bigi and Lamiani, 2016), it is argued that reactance can be prevented or limited by an appropriate use of argumentative strategies, used within the structure of the deliberation dialogue. In this sense, argumentation strategies could be considered and should be further experimentally tested as a form of debiasing.

PSYCHOLOGICAL REACTANCE IN RELATION TO THE PROBLEM OF NON-ADHERENCE

Psychological reactance is described as resistance to persuasive messages when a threat to freedom is perceived (Brehm, 1966; Brehm and Brehm, 1981). This condition brings individuals to do the contrary of what they are asked to do or to persist in a wrong behavior even in the face of evidence. The key factors involved in the arousal of reactance are freedom, threat to freedom, reactance and restoration of freedom. Studies have shown that reactance is the result of a combination of cognitive and affective processes, in which negative cognition and anger play a major role (Dillard and Shen, 2005; Rains and Turner, 2007).

The analysis of reactance as a potential threat to the achievement of patient engagement is relevant for two orders of reasons: (1) the institutional and asymmetrical nature of interactions between patients and providers, which could imply in itself a threat to freedom; (2) the fact that little attention has been paid so far to the role reactance could play in relation to non-adherence.

In the medical field, reactance theory has been mostly applied in the field of mental health, less frequently to understand non-adherence in general (Fogarty, 1997). However, a few studies have revealed that reactance can actually explain in part the motives behind non-adherence (Fogarty, 1997; Dillard and Shen, 2005; Orbell and Hagger, 2006; Rains and Turner, 2007). These



studies propose that the arousal of reactance could be limited or prevented by adopting certain measures, e.g., spending time with patients familiarizing them with the procedures they will undergo, answering their questions and listening to their non-medical comments; simplifying the behavior changes requested of patients; reducing the magnitude of the requested tasks; when possible, offering the opportunity to choose among different and equally acceptable solutions to the problem (Fogarty, 1997). These and other strategies are aimed at giving patients the feeling that they still hold some control over the process of care and that the "impositions" on their freedoms are acceptable because they have had the opportunity to decide about them. However, these studies do not discuss how these strategies should be operationalized at the communicative level, and more specifically at the level of dialogue. In particular, it is not clear what role is played by knowledge and persuasion.

As far as knowledge is concerned, it seems that pure information regarding an issue, e.g., a disease, is likely to represent a threat in itself, as it sheds light on possible limitations to freedom for the individual, who in turn will experience reactance (Brehm and Brehm, 1981; Fogarty, 1997). In particular, experimental findings (Dillard and Shen, 2005) suggest that those who design health messages should pay attention to health topics that may interfere with strength of threat: the study reported an experiment with messages related to flossing vs. responsible drinking, targeting college students. The latter were found to arouse more reactance because they were perceived as more threatening of freedom. The problem is that responsible drinking runs counter to social norms of conduct in certain groups, therefore complying with messages suggesting different styles of drinking puts individuals at risk of being stigmatized within their social group (Dillard and Shen, 2005, pp. 163–164). These

findings suggest that the role and provision of information to patients deserves careful consideration especially at the dialogical level.

With regard to persuasion, another study found that quality of argument does not impact on the degree of reactance (Rains and Turner, 2007), because it seems that perceiving a threat to freedom is enough to cause anger and negative cognitions that actually make the quality of arguments irrelevant. In other words, once certain information has been provided and reactance has been aroused, the potential benefits of persuasion are reduced by the emotional component of reactance itself, i.e., anger and negative cognition. It also seems that reactance is strengthened by the perception of dominance, i.e., the extent to which a message reveals that the sender believes s/he can control the receiver. On the other hand, when justifications for requesting a certain behavior are provided, this softens the feeling of threat and reduces the arousal of reactance. It is possible to distinguish in these findings two different ways in which argumentation is used: with the aim of persuading (in the sense of obtaining consensus or compliance) and with the aim of providing reasons to support a claim (with the aim of finding agreement over a debated issue).

In all the studies reported, there seems to be a close connection between the arousal of reactance and the processes of providing information about and reasons for action. In particular, it seems that the quantity, quality and context of the messages that are conveyed to patients can dramatically change the way threats to freedom are perceived. It appears therefore justified to try and understand the informative and argumentative processes from a dialogical perspective, which may allow to clarify their structures and roles in relation to reactance on the one hand, and engagement on the other. To this end, in the following sections insights from the field of argumentation theory are

called into the picture and discussed in relation to the notion of reactance.

PREVENTING REACTANCE AROUSAL THROUGH EFFECTIVE PRAGMATIC ARGUMENTATION

In her discussion on reactance and patient non-adherence, Fogarty (1997, pp. 1282–1283) puts forward a series of practical suggestions that should limit patients' reactance to providers' indications.

The final goal of providers' efforts at patients' involvement is, in Fogarty's terms, fueling patients' perception that they are retaining some degree of control over the procedures they need to undergo, and that they are freely conceding something to the provider, instead of being persuaded against their will into something they did not want to do. This should decrease the perception of loss of freedom, thereby also reducing reactance. The process of eliciting patients' cooperation should also increase the likelihood of future adherence, as studies show that those who comply with small requests are more likely to comply again, even with larger requests, in the future (Freedman and Fraser, 1966; Snyder and Cunningham, 1975; Souchet and Girandola, 2013). By explicitly requesting patients' cooperation at the beginning of the encounter, Fogarty suggests that an atmosphere of "mutual interdependence" (1997, p. 1283) is created, which can be preserved by letting patients become active participants in the discussion regarding therapeutic regimens, so that the final decision will result in a "negotiated regimen" formulated through the integration of patients' perspectives. In this respect, Fogarty provides a list of practical suggestions for providers:

1. discuss with patients how to fit the prescribed regimen into their and their families' lifestyles;
2. keep treatments as simple as possible (on this concept, see also more recent experiments by Fogg, <http://tinyhabits.com>);
3. show willingness to keep therapies as short as possible and to eliminate unnecessary proposals;
4. offer more than one effective alternative, whenever possible, and let patients select the one that best fits their preferences and possibilities. This suggestion in particular is aimed at communicating the perception of the provider as someone who is willing to make concessions. A universal rule of behavior (Cialdini, 2007; Ariely, 2008) dictates that when one party is willing to make concessions, the other one will reciprocate.

The kind of "discussion" described by Fogarty, basically corresponds to what argumentation scholars call "pragmatic argumentation," which happens when the parties need to agree on the solution to a problem, and discuss the validity of a course of action based primarily on its consequences (Perelman, 1959). In this use of argumentation, the positive or negative evaluation of the consequences is transferred to the causes, which are accordingly accepted or rejected.

In more concrete terms, during the medical encounter one therapeutic regimen may be preferred over another because it

is believed to obtain more positive consequences. Clearly, in order to align each other's criteria for the evaluation of the consequences it is necessary for both parties to be able to express their preferences during the discussion. Moreover, in order to put into practice suggestions 1. and 2. patients should be able to put forward their own proposals or perspectives on the provider's suggestions. In order for this communicative process to be effective, it cannot be left entirely to the good will or talent of providers. Its inner workings, potential and risks should be laid out and explained. In the next section, this is done by resorting to the model of the deliberation dialogue (Walton, 1989; Walton and Krabbe, 1995).

ADVANTAGES OF USING A MODEL OF DELIBERATION

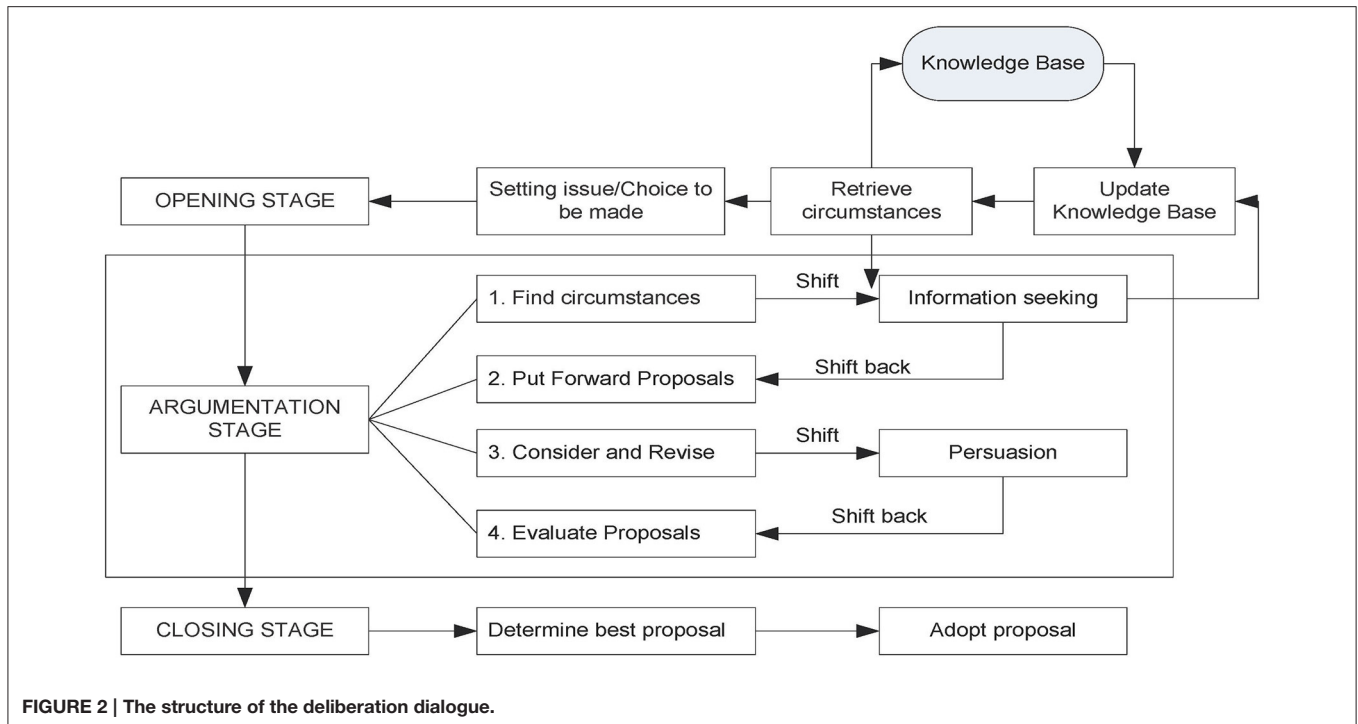
The model of the deliberation dialogue has been developed within an approach that aims at representing and analyzing types of dialogue as communicative intentions within a verbal interaction (Walton and Krabbe, 1995; Walton and Macagno, 2007). As such, the representations of the types of dialogue are abstract, normative frameworks capturing shared dialogical intentions. The deliberation dialogue is one among seven types of dialogues, described according to the intentions of and the initial relationship between the interlocutors: information-seeking, persuasion, deliberation, inquiry, negotiation, and eristics.

The structure of the deliberation dialogue outlines the most effective dialogical moves aimed at finding an acceptable course of action to achieve a certain goal (Walton, 2010; Walton et al., 2010). This kind of dialogue usually takes place when there is no compelling objective way of coping with a problem and parties discuss their reasons for proposing a certain solution; in this sense, it is a representation of pragmatic argumentation. It is therefore appropriate for the representation of the deliberative process occurring within a medical encounter when providers and patients need to discuss the acceptability of a therapeutic regimen or of a specific behavior.

One important premise for deliberation dialogues is that parties are out to reach a collective goal, which can be contrary to or different from the individuals' personal goals (Walton et al., 2010). This is true also of the kind of deliberations occurring within medical encounters. Indeed, the arousal of reactance can be partly explained as resistance to a potential threat that is perceived due to a misalignment between the parties' intentions and preferences. Part of the deliberative effort is precisely to set a shared goal in a collaborative way (Bigi, 2014a), which is basically what Fogarty (1997) suggests when proposing that patients' collaboration be explicitly invited.

The structure of deliberation dialogues is represented in **Figure 2** (Walton et al., 2014).

As shown in **Figure 2**, deliberation dialogues usually develop in three stages: the opening stage, the argumentation stage and the closing stage. The stages of the dialogue do not correspond to phases in the structure of the medical encounter; their names refer to the development of the deliberative process and they are not meant to represent the chronological order in which stages



appear in real-life interactions, even if they do represent the most logical order in which they should appear. In other words, they represent the ideal organization of an optimal deliberation dialogue.

Deliberation in this model is represented as a complex dialogue type, resulting from the combination and intersection between the persuasion and the information seeking dialogues.

In the opening stage, the parties share relevant information regarding the problem at issue. In this stage information is functional to the argumentative process that will follow, therefore it is crucial that parties share what they know about the problem, but also their preferences, values, circumstances that may in any way bear an import on the kind of solution that can be found. The aims of this stage remind of some of the suggestions for practice put forward by Fogarty (1997), in particular the indication to spend time with patients before the actual encounter, listening to their concerns of personal, non-medical nature (Fogarty, 1997, p. 1283). For Fogarty this procedure is aimed at keeping patients involved in the process of care, thereby limiting their perceptions of threat to their freedom. However, from a dialogical perspective, this is also a precious indication for the achievement of more effective deliberations. Indeed, proposals for therapeutic regimes or behaviors will be much more tailored to patients' actual conditions and abilities if providers are aware of them (in line with the suggestion to keep treatments as simple as possible, from the patient's point of view; to take into consideration patients' abilities to do certain things; and to find the best way to fit the prescribed regimens into patients' lifestyles).

The opening stage then gives way to the argumentation stage, which is the heart of deliberation. Here the parties start putting forward proposals for the solution of the problem, based on

the previously shared information and on the shared goals of the interaction. The relevance of this stage in relation to the issue of reactance is very high. First, it is assumed that both parties should put forward at least one proposal to solve the problem. This is coherent with the indication to engage patients in the discussion in order to reach a negotiated regimen that is acceptable to both parties. Second, it is also foreseen by the model that if new information emerges during the argumentation stage this might imply that the parties revise their initial proposals. This is coherent with the suggestion to providers to be willing to make concessions and to offer more than one option for patients' choice.

Upon finding an option that is acceptable for both parties, the dialogue enters its closing stage. Here it is important that patients make explicit commitments to carrying out the chosen behavior or therapy. Providers' commitment to providing expert advice and the best options available are presupposed by the institutional context within which the interaction takes place.

This structure helps to clarify the role information and argumentation play within the deliberative process and their very close connection and interdependence. Moreover, at least at a theoretical level, it would seem that carrying out deliberations by following the structure and premises of the deliberation dialogue should prevent or limit the arousal of reactance. Indeed, the model foresees that both parties are fully engaged in the deliberative process at all stages, which basically realizes Fogarty's suggestion to keep patients involved in the process of care so as to limit reactance by softening the strength of threat.

Thus, theoretically speaking, this model of deliberation could be proposed to clinicians as a "deliberation protocol" that would allow to overcome sloppy or naïve—and thus,

ineffective—realizations of deliberation. However, before discussing practical applications of the model, the role of information in relation to reactance should be discussed.

THE ROLE OF KNOWLEDGE IN RELATION TO REACTANCE

As already mentioned, patient education is discussed by Fogarty (1997, pp. 1284–1285) as a way to limit patient reactance and non-adherence. Moreover, the findings by Dillard and Shen (2005) show that information should be framed very carefully also depending on the potential strength of threat implied by certain health topics. Thus, the use of knowledge as a means to achieve adherence should be managed carefully. Indeed, more information can have the boomerang effect of increasing reactance, because by knowing more about their disease individuals are also more aware of possible further limitations on their freedom, thus behave in ways that try to prevent these limitations. Another problem with knowledge is that it may interfere with patients' Locus of Control: patients who wish to remain in control of their lives may feel excessively threatened by education programs that enhance their awareness of the many ways in which the disease and its treatments will "take control away" from them (Fogarty, 1997).

In order to better understand the role of information in the provider-patient encounter and in relation to the arousal of reactance, a general distinction that can be made is between information as the means to achieve patient education on the one hand, and as a component of the process of pragmatic argumentation on the other. This distinction can be useful because it allows to collocate the provision of information within two rather distinct dialogical processes, thus more easily identifying its role, potential and risks in relation to the arousal of reactance.

As a means to achieve patient education, the provision of information should be considered first of all from an institutional point of view. Indeed, the medical encounter is by definition an institutional context of interaction, with predefined roles, aims and norms that regulate the communicative exchange. Depending on the design of each healthcare system, these institutional elements may vary, but generally speaking providing information to patients regarding their health condition is considered to be one of the aims of the encounter. Therefore, providers cannot avoid this step and if knowledge can produce reactance, it is important to understand how it can be provided in a way that may limit or prevent its arousal.

Based on the considerations presented above referring to the boomerang effect of knowledge, it seems necessary to move away from a conception of education as mere transmission of information. In this respect, helpful insights may be gained from studies in the field of science education, in which it is shown experimentally that argumentative processes favor learning and understanding (Schwartz and Asterhan, 2010; Felton et al., 2015). More specifically, when students are given the task of "finding agreement" on an issue, they seem to be more willing to open up to different points of view and understandings of the problem.

Moreover, argumentative practices seem fundamental for the achievement of conceptual change, i.e., a radical reframing of knowledge, which is often necessary in the case of lay beliefs and misrepresentations of health issues that patients often bring to the encounter and that impact on patients' expectations and on the ways they make decisions or form commitments. The "exercise" of using argumentative strategies for patient education in analogy to experiments with students could be done during group-work with patients, where peer-to-peer interaction may also favor the expression of personal beliefs. On the other hand, the "exercise" of reaching radical conceptual change regarding wrong beliefs or lay prejudices about diseases should probably be conducted during the one-to-one encounter with the physician.

In both cases, experimental interventions need to be carried out before it is possible to offer providers specific techniques for achieving patient education through argumentation.

In general, the role of information within a process of education is different from its role within a deliberative process. As discussed previously, in the latter situation information is relevant only if it informs the argumentative component of deliberation. This means that, if during the opening stage of deliberation, the provider realizes the patient has wrong beliefs or information about the disease, the dialogue may shift to an education dialogue and then shift back to the deliberation. However, the potential for threat to freedom seems different in the two cases: it may be higher during an education dialogue, because the provision of information entails expectations for further limitations to freedom; it should be lower during a deliberation dialogue, where patients should be fully involved in the decision, thus maintaining a fair degree of control over what is being decided and what they will have to commit to. An example of the first situation is provided by the following excerpt, from a consultation in a diabetes outpatient clinic (Bigi, 2014b). The patient (P) and the doctor (D) are discussing the patient's health situation and the patient starts asking questions about the correct choice of food:

P: what about beans, peas, can I eat those?

D: of course

P: but I noticed that they raise my glucose values

D: well, yes, you can eat them but appropriate quantities. So, for example, if you want to have pasta with beans you will add less pasta than when you have pasta with butter

P: also vegetable soup?

D: eh, of course because

P: if I make vegetable soup I noticed it [the glycemia] increases

D: vegetable soup, excellent question, it contains potatoes or carrots. Potatoes for example have a higher glycemic index so...

P: also carrots?

D: also carrots, but less than potatoes

P: I see...

This can be described as an education dialogue, because it has been triggered by a question from the patient, aimed at integrating a knowledge gap. The doctor replies by making examples of correct behaviors and by explaining the characteristics of different kinds of food. The doctor's

explanations carry implications for the patient's behavior: if the patient is informed that potatoes contain more sugar than carrots, from now on he cannot just go on eating as many potatoes as he wants, because he can be held accountable for such behavior. In this sense, an education dialogue can contain a threat to freedom.

A different case is the one of the deliberation dialogue, like the following, also an excerpt from a consultation in a diabetes outpatient clinic (Bigi, 2014b). The doctor (D) has explained to the patient (P) that she really needs to be careful about her weight, because it could impact negatively on her diabetes. She starts a deliberation dialogue in which she invites the patient to decide how she wants to cope with the situation:

D: ok, so from my point of view I can't suggest much. You are already taking strong medications for your diabetes, which means that if the three branches of a therapy are physical activity, medications and eating habits, I am already pretty high on medications. It would be better to act on the other two levels. Only one, both, a bit of both, you have to tell me. What do you think you can do?

P: I would like to...

D: not I would like

P: no, I would like to, really...

D: ok, what we would like is the ideal situation, it's perfection, but what is it that you can actually do at this point of your life?

P: I don't know what I will be able to do...

P's daughter: why don't you come to the gym with me, Mom? Three months?

P: ok, let's go, let's try...

D: three months at the gym, ok then, 3 months at the gym, and we could add to that no restrictions on eating, but a very careful management of sweets

P: no..., look, I don't mind giving up sweets, but don't make me give up fruit

D: that's ok, I'm telling you, let's negotiate. Let's choose two things, 3 months at the gym, no sweets and you can have fruit. Let's try and see how it works, ok?

Also in this case, the doctor's opening turn contains an explanation that implies restrictions on the patient's freedom, but the deliberation dialogue that follows actively involves the patient in the process of decision making regarding which freedoms to give up and which to retain. In this particular case, the doctor is very good at allowing the patient to express her preferences, at the same time putting forward proposals for action that she can agree to or refuse. In this way, the patient retains control over the actions that are decided upon and she can freely decide what she wants to commit to.

So, while in the case of education, special attention should be paid to the way information is presented, in the case of deliberation it is the "procedure" that should be followed carefully in order to ensure patients' full participation, thereby limiting reactance. By "procedure" is meant the structure of the deliberation dialogue as described in **Figure 2**.

In both cases, the adoption of new strategies in the clinical practice entails a challenge at the institutional level of context, which is what we turn to in the following section.

INTRODUCING PRO-ENGAGEMENT STRATEGIES IN THE CLINICAL PRACTICE: A CHALLENGE FOR HEALTHCARE SYSTEMS

As mentioned in the introductory section of the paper, the definition itself of patient engagement entails an opening to the wider context of the healthcare system within which providers and patients interact. The focus on very specific dialogical processes proposed in this paper does not imply a different perspective, quite the contrary.

The description of the model of the deliberation dialogue (see **Figure 2**) allows to visualize the components of this complex dialogical process, and to ponder their roles in relation to the achievement of the final goal of a participatory and collaborative decision (itself a pre-requisite for behavior change). In particular, the role of information has been discussed in relation to the aims of patient education and deliberation. The crucial role of argumentative strategies has been pointed out in relation to the prevention or softening of reactance, and the support to processes that may lead to full patient engagement.

The previous discussion has been conducted at a theoretical level and has proceeded from a top-down approach. The conclusions that can be drawn at this level are still in the form of hypotheses and will have to be confirmed through experiments and interventions. However, even at this theoretical level, the awareness of the optimal realization of crucial dialogical processes within the medical encounter allows some reflections regarding the setting in which such processes take place. In order to make the discussion more concrete, the Italian healthcare system will be used as an example, given the familiarity of the author with its structure (Bigi, 2012).

The Italian Healthcare System

Italy's health care system as we know it today was officially born in 1978 (Centro di ricerca sulle amministrazioni pubbliche "V. Bachelet", 2008, pp. 4–12). The system is founded on a principle of "universality," which means that minimal levels of healthcare should be guaranteed to everyone. In coherence with this principle, medications and exams are mostly paid for by the healthcare system with resources collected through taxation. Citizens may be required to contribute for a smaller part to the expenses. As a consequence, the system is quite easily accessible, at least at the level of general practice, which is free and managed by single practitioners through appointments. Disadvantages of the system are that not all the 20 regions in which Italy is divided are able to manage resources optimally, which leads to bad imbalances in the provision of healthcare. Moreover, in case of economic crises, the central government may decide to cut the healthcare budget, which of course has an impact on the quality of care provided.

One important implication of healthcare being managed centrally is that all providers working for the public healthcare system are employees of the public administration and, as such, have the juridical status of "public officials." This means

that in their capacities as healthcare professionals they are the representatives of the central government, which delegates to them the safeguard of health intended as a public good (Costituzione della Repubblica italiana [Constitution of the Italian Republic], 2009, art.32). In this context, prescriptions and certificates signed by providers are public instruments, which, if improperly written or signed, can cause providers to undergo a penal trial.

In case of malpractice, the criteria that the judge uses to evaluate a healthcare professional's conduct are the ones of diligence and information. In the case of diligence, providers have to do everything and the best that is in their possibilities, having considered the situation of the patient (Bianca, 1965). In the case of information, professionals must provide all the relevant information to patients regarding the procedures they will undergo, along with potential risks and advantages. The Constitution (art. 32) states that nobody can be made to undergo a treatment unless they freely accept to. This leads to the obligation to provide and require explicit Informed Consent from patients, in all of the cases that entail procedures in which the patient is a passive subject (e.g., surgical operations). In the case of drug prescriptions, e.g., in outpatient clinics or in general practice, formal Informed Consent is not required because patients can always refuse treatment, but the obligation to provide thorough information remains. In any case, the only criterion that must guide providers' decisions is patients' quality of life and wellbeing.

A Challenge for the Healthcare System

The description of the structure of the Italian healthcare system and of providers' juridical obligations reveals the degree of complexity of providers' task. As public officials, providers have an obligation to exercise their profession with diligence, which entails the obligation to information giving. On the other hand, in any circumstance patients are free (by law) to refuse or interrupt therapeutic regimens. The reasons for not wanting to take care of one's own health are not as improbable as they may seem. Incorrect information collected on the Internet, through acquaintances, via the mass media, may lead patients to become suspicious of certain treatments or to decide not to take any "chemical drugs." Wrong beliefs regarding the definition of healthy food or healthy nutrition may lead patients to opt for unhealthy eating habits, thus damaging their health (see, Bigi and Pollaroli, 2016 for a discussion on this topic). The asymmetrical social roles predefined by the context of interaction may also play against the construction of trust between patients and providers, thus complicating even more the achievement of the institutional goals of patients' wellbeing.

Wrong information, badly informed beliefs, the tension created by misaligned expectancies related to each other's roles within the encounter may concur to create more opportunities for the arousal of reactance rather than patient engagement. Very recent data regarding patient non-adherence to treatments in Italy (Italian Medicines Agency, 2015) cannot be ascribed entirely

to these causes, but certainly indicate that the problem exists and needs attention.

The discussion conducted in the preceding sections has pointed out the crucial role played by argumentation in both the process of patient education and of shared deliberation. In both cases, the potential for the reduction of reactance is very high and should be tested experimentally. However, in consideration of the institutional constraints presented in the previous section, there is one more reason to urge the system to innovate. Indeed, the obligation for providers to provide information to patients combined with findings on the high potential for a boomerang effect of information would suggest that providers be not only trained to provide information appropriately, but also be put in the conditions to do so. The suggestion of allowing more time and resources for trying out argumentative strategies in the context of patient education goes in this direction. Moreover, if patients are free to interrupt treatment at any time and providers cannot force them, it is also true that they can try to persuade them, if continuing treatment is good for patients' health. In this case too it is an issue of appropriate training in argumentation (Bigi, 2015 for a discussion on this point), but also of having the opportunity of spending time with patients, either personally or as a team.

As the issue of time is very common and cuts across national boundaries, a consideration regarding chronic conditions can be made. Indeed, the time for individual encounters is not very long and it is not possible to educate, motivate, and train patients to the use of devices, while listening to them and doing the paperwork required in the space of 15–18 min. However, chronic conditions have an "advantage," which is that they last in time. The opportunity of meeting with patients on a regular basis for years, offers the possibility to distribute the various tasks over different encounters. One time it might be a priority to conduct a session of education on a certain topic, and the following time it might be more important to focus on motivating the patient.

Finally, as already argued by other scholars (Wagenaar, 2006), sometimes healthcare facilities should be designed more appropriately, in order to limit the perception of alienation (and thus, loss of freedom) in patients. Entering very complex buildings, with no clear signs and getting lost; interacting with dismissive or non-professional personnel; waiting for a long time in a stuffy corridor, with lots of people, while sitting on an uncomfortable chair: all this may not be conducive to collaborative and trusting relationships during the encounter.

Therefore, the challenge for any healthcare system, also in times of fewer resources and sustainability emergency, is not only to reduce budgets and cut on expenses, but also to rethink or restructure some crucial points in the system that hinder engagement and collaboration among all the actors in the system.

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The author confirms being the sole contributor of this work and approved it for publication.

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An Educational Intervention to Train Professional Nurses in Promoting Patient Engagement: A Pilot Feasibility Study

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Introduction: Growing evidence recognizes that patients who are motivated to take an active role in their care can experience a range of health benefits and reduced healthcare costs. Nurses play a critical role in the effort to make patients fully engaged in their disease management. Trainings devoted to increase nurses' skills and knowledge to assess and promote patient engagement are today a medical education priority. To address this goal, we developed a program of nurse education training in patient engagement strategies (NET-PES). This paper presents pilot feasibility study and preliminary participants outcomes for NET-PES.

Methods: This is a pilot feasibility study of a 2-session program on patient engagement designed to improve professional nurses' ability to engage chronic patients in their medical journey; the training mainly focused on passing patient engagement assessment skills to clinicians as a crucial mean to improve care experience. A pre-post pilot evaluation of NET-PES included 46 nurses working with chronic conditions. A course specific competence test has been developed and validated to measure patient engagement skills. The design included self-report questionnaire completed before and after the training for evaluation purposes. Participants met in a large group for didactic presentations and then they were split into small groups in which they used role-play and case discussion to reflect upon the value of patient engagement measurement in relation to difficult cases from own practice.

Results: Forty-six nurses participated in the training program. The satisfaction questionnaire showed that the program met the educational objectives and was considered to be useful and relevant by the participants. Results demonstrated changes on clinicians' attitudes and skills in promoting engagement. Moreover, practitioners demonstrated increases on confidence regarding their ability to support their patients' engagement in the care process.

Conclusions: Learning programs teaching nurses about patient engagement strategies and assessment measures in clinical practice are key in supporting the realization of patient engagement in healthcare. Training nurses in this area is feasible and accepted and might have an impact on their ability to engage patients in the chronic care journey.

Due to the limitation of the research design, further research is needed to assess the effectiveness of such a program and to verify if the benefits envisaged in this pilot are maintained on a long-term perspective and to test results by employing a randomized control study design.

Keywords: patient engagement, patient engagement measurement, nursing staff, hospital, professional-patient relation, empowerment, educational program, patient activation

INTRODUCTION

Effectively managing chronic diseases and the burden they put on patients and families, clinicians, and healthcare organizations is currently a major concern for health policy makers (Vogeli et al., 2007). Promoting patient engagement is increasingly acknowledged as a way to address the challenge of chronic conditions (Hibbard and Greene, 2013; Graffigna et al., 2014) and it has been widely advocated as a crucial component of patient-centered models of sustaining healthcare innovation (Thomson et al., 2005; Washington and Lipstein, 2011). Moreover, embracing the patient engagement paradigm is an ethical priority for healthcare systems across countries (Sherman and Hilton, 2014; Solomon et al., 2016; Weil, 2016): it is well-known as a key strategy to include the patients' preferences and expectations in services' design and delivery thus maximizing their clinical effectiveness (World Health Organization, 2002; Eldh et al., 2004; Koloroutis, 2004; Elwyn et al., 2007; Jordan et al., 2008; Eaton et al., 2015; Fisher et al., 2016).

Research has validated this perspective and it has been demonstrated that enhancing patient engagement might increase patients' motivation toward treatments and the care process (Mosen et al., 2007), improve treatment outcomes (Hibbard and Greene, 2013), and generate greater satisfaction for received care (Tobiano et al., 2015). Lastly, engaging patients in their care might contribute to the system's sustainability through a reduction of healthcare services use (Remmers et al., 2009).

Nursing: A Key to Patient Engagement

In the midst of healthcare transformation and evolution, where patient engagement is an imperative to catalyze the delivery system reform (Carman et al., 2013; Fisher et al., 2016), nursing plays a critical role in the effort to make patients fully engaged in their disease management (Lammon et al., 2010; Barello et al., 2012). Professional nurses across Western countries have a well-established tradition of engaging patients in the medical course and are trained to embrace clinical models which endorse "autonomous and collaborative care of patients of all ages, families, groups, and communities, sick or well and in all clinical settings" (International Council of Nurses, 2010; Pelletier and Stichler, 2013). The concept of relationship-based care constitutes a milestone of the nursing care paradigm with several theories supporting the nurse-patient partnership as the foundation for effective care delivery (Koloroutis, 2004; McCormack and McCance, 2006; Sahlsten et al., 2007; Barello and Graffigna, 2015b). Through the relational and educational aspects of nursing care, nurses have the opportunity to build care relationships that support patient engagement, which ultimately

lead to improved patients' quality of life even when life-threatening chronic conditions occur (Gruman et al., 2010; Jerofke et al., 2014; Barello and Graffigna, 2015a; Jenerette and Mayer, 2016). In their clinical encounters, nurses are recognized to be key persons whose strategies might hugely affect the patient's level of engagement in the care process (Jordan et al., 2008; Kutney-Lee et al., 2009; Laschinger et al., 2010; Deyo et al., 2016; Morath and Braaten, 2016). Research on the patients' perspective showed how the patients' perception of professionals' positive attitude toward their self-management behaviors is associated with higher level of patient engagement (Graffigna et al., 2016) and adherence to medical prescriptions (Chan et al., 2009; Schmidt et al., 2012) thus confirming the crucial role of nurses who are often in charge of self-care interventions.

Particularly, scholars (Laschinger et al., 2010; Pelletier and Stichler, 2013) agree that nurses can engage patients by enacting different behaviors and relational strategies: (1) sensitizing patients to be aware of their active role in managing their health and in playing an active part in their treatment plans and decisions; (2) providing patients with trustable information, resources, and support in order to enhance their level of health literacy; (3) stimulating the patients' autonomy in treatment decision making; (4) facilitating exchanges within the formal and informal patients' care network (health providers and family); (5) coordinating care delivered by other providers. For these reasons, nurses are today increasingly valued as professionals qualified to provide personalized and flexible care, which are fundamental for a patient engagement approach to healthcare (Needleman and Hassmiller, 2009; Naylor et al., 2011; Lewis et al., 2016).

Barriers to Patient Engagement in the Patient-Nurse Relationship

Despite the growing awareness that engaging patients in their care process should be a nursing priority, considerable barriers related to its effective realization still exist and are reported in literature. For instance, Wellard (2001), Wellard et al. (2003) showed how poor communication between patients and nurses, limited privacy, and task-focused nursing practice might hinder the patients' possibility to engage in effective disease management. According to other scholars (Eldh et al., 2006), patients tend to be disengaged in their care when they do not feel in an equal relationship, when there is a lack of information about their health condition or they experience a paternalistic attitude by their health providers. Previous research also identified patients' dissatisfaction with the limited opportunities given to them to be actual partners in their own healthcare management (Eaton et al., 2015). Probably, this is due to the fact that some health practitioners do have a vision for partnership,

but frequently lack the practical resources or pragmatic skills required to actualize this principle in their daily practice (Wellard and Street, 1999; Rasmussen et al., 2001; Wellard and Rushton, 2002; Wellard et al., 2003). Then, although professionals might value and embrace the engagement philosophy, they may experience some tensions between their professional role and the raising patients' desire for autonomy and participation in the care decisions. This may limit professionals in sustaining patient engagement as well as it may be perceived as a potential attack to the established patient-clinician relationship (Kennedy et al., 2007). Moreover, lack of theoretical knowledge might impact nurses' awareness about the need to plan deliberate strategies for effectively engaging patients along their care journey (Sahlsten et al., 2008).

Collecting Patients' Experience of Engagement: The Value of Patient Engagement Measures to Support Nurses' Action

Modern healthcare systems are increasingly recognizing the need for paying attention to the patients' engagement expectations in order to best align services with patients' needs (Elwyn et al., 2007; Black, 2013; Graffigna and Barello, 2016). Patient engagement measures have the potential to drive changes in how the healthcare system is organized and delivered and might contribute to actually realize the "patient engagement promise." Moreover, patient engagement measurements—initially developed for use in research (Hibbard et al., 2005; Graffigna and Barello, 2016)—have recently started to be considered a mandatory requirement for health professionals to enhance clinical management (Graffigna and Barello, 2016; Hibbard et al., 2016). Adopting this view also implies to consider that patients' engagement expectations might vary depending of the phase of the patients' medical journey (Barello et al., 2015b). Moreover, the patients' conception of what it means to be engaged might change widely, from being welcomed to be an active part of treatment decision and care plans (Thompson, 2007). This means that professionals' skills and actions need to be balanced and attuned with the expertise and expectations of patients. Capturing the patient engagement experience, for this reason, cannot be limited to the professionals' sensitiveness to patients' illness experience; rather, it should be systematically conducted by adopting dedicated assessment tools which are able to collect patient engagement levels. Despite resistances to their adoption still exist, clinicians increasingly recognize the benefits of collecting their patients' engagement expectations. Particularly, the call for patient engagement measures adoption has been sustained by evidences that show they can be of benefit for multiple purposes (Simmons et al., 2014; Graffigna and Barello, 2016; Hibbard et al., 2016; Roberts et al., 2016): (1) a process or outcome measure to determine the patients' characteristics that may predict their level of engagement and their risk for dis-engagement, (2) a tool to personalized interventions based on the individual's ability for self-management (3) an outcome measure for evaluating the performance and effectiveness of healthcare interventions,

comparing pre- to post-intervention assessments of one patient's level of engagement; (4) an instrument to optimize and tailor professionals' relational and communicational strategies and, potentially, to improve the effectiveness of their actions.

In the light of these premises, the "measurement act" can become a precious occasion for nurses to feel empowered in their clinical effectiveness and to have concrete guidelines about how to achieve positive relationship with their patients and provide more patient-centered care. However, the innovation potentials offered by the adoption of patient engagement measurements can only be realized if clinicians can understand the values of these data in patient care and start collecting and utilizing this information in their daily practice. Indeed, a key issue limiting successful implementation of patient engagement measures is the clinician's lack of knowledge and awareness about how to effectively utilize these data in the clinical management of their patients. All these factors appear to stress the need to train professional nurses in introducing and benefit from patient engagement measures in their daily clinical practice.

The Present Study

In order to enhance professional nurses' ability to make their patients effectively engaged in their medical course, the authors (GG and SB) developed a novel program designed to train nurses in patient engagement strategies and to effectively adopt patient engagement measures in their routine practice (NET-PES program). From a theoretical point of view, the implementation of patient engagement measurement in routine patient care represents a significant change to clinical practices of both individual clinicians and healthcare organizations. Changing the organizational culture of healthcare systems is well-known to be a challenge. The phases of the change process, namely dissemination, adoption, implementation, and continuation (Fleuren et al., 2004), can be applied specifically to the process of implementing patient engagement measures in clinical practice, in the light of specific organizational and clinical issues. The training of clinicians represents the first step of dissemination.

The NET-PES program is based on the assumption that clinicians who are sensitized to the value of patient engagement, who have adequate theoretical and practical knowledge about this concept, who are provided with communication and relational skills and have reasonable confidence in their own abilities to engage patients are more likely to correctly assess and thereby to better respond to patients' engagement needs and expectations. Moreover, a central asset of the course is that without providing professionals with reliable measures for capturing their patient engagement level, they cannot succeed in tailoring their own actions on patients' care expectations and preferences. The course was developed according to extensive research about patient engagement and the factors enabling it and designed to be highly participative and interactive among participants (Hibbard et al., 2005; Graffigna and Barello, 2015a). This is a report of feasibility testing of the program along with examining potential impact and participant satisfaction.

MATERIALS AND METHODS

We conducted a pre-post pilot study evaluating the feasibility of the NET-PES program. Self-report questionnaires were completed before and after the workshop for evaluation purposes. To guarantee data confidentiality and assure anonymity, each participant was assigned with a random identification number in order to track data from pre- and post-questionnaires. All nurses working with chronic patients at the Azienda Sanitaria Universitaria Integrata di Trieste (Italy) were contacted and recruited through e-mail invitations sent to the hospital units, to service medical directors and to nurse managers and they were invited to participate in the course. Only nurses having work experience with chronic patients were considered eligible for the training program. Eligible nurses interested in learning more were invited to an information session. Recruitment materials emphasized the voluntary nature of the program.

Due to the fact that it is an observational design with education as the intervention it didn't require any ethical approval by the hospital institutional review board according to the institutional and national requirements. However, we collected by all participants written informed consent for confidentiality issues. We confirm that all ongoing and related trials for this observational educational intervention will be submitted to Ethical approval and registered in ClinicalTrials.gov.

The NET-PES Program Description

According to the principles of patient engagement and activation theories (Hibbard et al., 2005; Graffigna and Barello, 2015a), a theoretically-driven educational training for nurses working with chronic patients entitled "Nurses Engagement Training in Patient Engagement Strategies" (NET-PES) was designed, delivered, and tested by researchers who have experience in adult training programs and patient engagement theories. Moreover, specific inputs were given from clinicians (GP, MM, MC, LP), which contributed to outline the main educational needs of the target participants, reviewed didactic materials and training scenarios. To guarantee adequate participation, a maximum number of 25 participants was set for each edition of the workshop. Teaching methods included adult teaching learning principles (Colliver, 2000) and provided an experiential learning environment where nurses were encouraged to share their professional experience and reflect on patient engagement strategies they enact in their daily clinical practice.

The program content was articulated into four main themes: (1) *setting the scene*: a didactic overview covering the core principles of patient engagement and activation theories and the evidence base for improving nurses' knowledge and skills to engage patients in their care; (2) *the whys for adopting patient engagement measures*: an outline of the main evidence supporting the value of patient engagement measures and the benefits of using them in clinical practice; (3) *acquiring practical skills to use patient engagement measures in clinical practice*: familiarization with the measures, understanding of clinically important differences among the patient engagement

profiles, how to adapt communication and relational style to different levels of engagement, and developing shared strategies to guide potential nurses' actions based on patients' scores; (4) *from theory to practice*: overview of the most effective patient engagement interventions currently available in the scientific literature and tools for guiding clinical actions aimed at promoting patient engagement. **Table 1** describes in details the contents, the learning objectives and the main educational techniques featuring the program.

Didactic presentations were constantly alternated with interactive discussions of clinical cases of real patients with different clinical conditions. The cases included a summary of the medical history, patient engagement profiles' card, and linkage of the patient engagement profiles to patients' clinical parameters. These allowed reflecting upon the value of collecting patient engagement measures to address the patients' needs across patient engagement profiles and, consequently, to tailor the interventions. Moreover, simulated clinical consultations enacted by a patient-actor followed by debriefing with facilitators allowed participants to share knowledge, to receive live peer feedback and to experience a collaborative learning context with colleagues working in different clinical units. The interactive fashion of the program was encouraged by facilitators and provided rich contents for the course.

The supporting materials used in the program included: the main measures of patient engagement currently available in the scientific literature (i.e., Patient Activation Measure by Hibbard et al., 2005; Patient Health Engagement Scale by Graffigna et al., 2015b), the case studies discussed and memory-aid materials including the scales' scoring system, guidance on changes in patients' needs across the patient engagement levels, and related suggested actions, and scientific articles discussing the evidence base that oriented the program's contents.

Participants' Outcomes and Program Evaluations

Feasibility was evaluated including process measures (such as participants' attendance, completion rates of assessment measures, and satisfaction with the program). Nurses engagement attitudes and skills were assessed using self-report measures administered before (T_0) and at the end of the program (T_1). The second day of the course was delivered 1 month after the first session. Socio-demographic and professional characteristics (i.e., age, gender, discipline, year of professional experience,) were collected before the delivery of the program.

Pre- and post-evaluation surveys included the following measures:

- The 13-item version of the *Clinician Support Patient Activation Measure* (CS-PAM) (Rademakers et al., 2015), which assesses clinicians' beliefs and attitudes toward the importance of patient self-management behaviors and his/her active role in the care process. The items of the CS-PAM are aimed at assessing the degree to which practitioners value patient activation behaviors as important (Hibbard et al., 2010). Particularly CS-PAM items might be grouped into four

TABLE 1 | Program sessions' description.

Sessions	Content	Learning objectives	Learning activities
Day 1	Setting the scene A didactic overview covering the core principles of patient engagement and activation theories and the evidence base for improving nurses' knowledge and skills to engage patients in their care	<ul style="list-style-type: none"> Enhance nurses' motivation and sensitiveness toward the value of engaging patients in the care process Introduce the theoretical underpinnings at the base of the patient engagement paradigm Make nurses aware of their crucial role in realizing patient engagement and catalyzing organizational change 	Didactic theoretical presentations, group discussion, narratives Role-playing, clinical case discussions, group and facilitators feedbacks, wrap-up
	The whys for adopting patient engagement measures An outline of the evidences supporting the value of patient engagement measures (PAM and PHE-s)	<ul style="list-style-type: none"> Provide nurses with the evidence base for adopting patient engagement measures in the clinical practice Discuss the clinical and relational value of including patient engagement measures in the routine patients' assessment along with other clinical parameters 	
Day 2	Acquiring practical skills to use patient engagement measures in clinical practice Experience-based session dedicated to make nurses experience the use of patient engagement measures and pragmatically reflect upon their use in the relationship with patients	<ul style="list-style-type: none"> Familiarize nurses with the main patient engagement measures and make them experience how to use data obtained through them Teach nurses how to use patient-reported outcomes as a relational instrument within the interaction with patients Promote nurses' competences in patient centered communication and relational skills basing on the patient engagement profiles Sensitize nurses to align their communication and relational style to the level of patient engagement 	Didactic theoretical presentations, standardized patient exercises, vignettes, clinical case discussions
	From theory to practice Description of the main interventions currently available for promoting patient engagement of provision of a toolbox for orienting clinical practice aimed at sustaining patient engagement	<ul style="list-style-type: none"> Provide nurses with validated and evidence-based intervention strategies grounded in the literature of patient activation and engagement Let nurses experience tools and exercise with simulated patients 	Didactic theoretical presentations, role-playings, wrap-up

categories of increasing level of patient's activation behaviors that clinicians might endorse (items 1, 4, 5, 8: "*patient should have the knowledge and behave in order to prevent or minimize symptoms associated with their health condition*" (adherence); items 2, 6, 7: "*patient can make independent judgment and action*" (independency); items 9, 10, 11: "*patient is able to take an active role during the consultation*" (partnership); items 3, 13: "*patient should be an independent information seeker*" (information seeker). Each item is scored using a 5-point Likert scale (1, Strongly disagree; 2, Disagree; 3, Agree; 4, Strongly agree; 5, Not applicable).

- **Clinicians Competence in Patient Engagement Strategies (CC-PES)** is an ad-hoc scale developed for the aims of the program that scores clinicians on 9 items, which equate to their perceived confidence in enacting 9 key strategies to promote patient engagement. Each item is scored using a 5-point Likert scale (1, Not at all confident; 2, Not very confident; 3, Neutral; 4, Confident; 5, Very confident). Competences assessed are the followings: (1) *assess the level of patients' engagement*; (2) *generally support patient engagement*; (3) *motivate patients in following medical prescriptions*; (4) *inform patients about disease and treatments*; (5) *assess patient's health literacy*; (6) *empathize with patients*; (7) *assess and manage difficult patients' emotions*; (8) *effectively communicate with patients and their families*; (9) *effectively relate with patients and their families*.
- **Participants' experience and satisfaction questionnaire:** the post-questionnaire was created to obtain rating and qualitative

reports for participants on their experience and satisfaction with the NET-PES program and recommendations for future improvements. Particularly the survey consisted of six items, including six questions rated with a 10-point Likert scale assessing nurses' satisfaction with the course in term of general satisfaction, interest, usefulness, quality of teaching, quality of didactic materials, and quality of practical sessions. Open-ended questions were also used to gather information on what participants liked most and least about the program (in terms of both didactic contents and teaching methods) and to obtain specific recommendations to improve the program itself.

Fidelity Assessment

The research team monitored the program fidelity by implementing monthly supervision and protocol review sessions including the principal investigators, co-investigators, and program facilitators. In addition, the principal investigator attended at least one session of the course and provided feedbacks to program facilitators. Ongoing review of the program sessions through the study period revealed both the strengths and weakness of the protocol design and intervention delivery. This pilot feasibility work has been intended to form the basis for subsequently modifying the design of the NET-PES program for a larger clinical trial.

Data Analysis

Participants' socio-demographic and professional features and satisfaction items were analyzed through descriptive statistics. To determine the internal consistency of the adopted measures, Cronbach's α was used. As the CC-PES was ad-hoc developed for the purposes of the present study, the factorial structure was assessed with a principal component analysis (PCA). Paired-sample t -tests were used to compare T_0 and T_1 mean scores of CS-PAM and CC-PES. Moreover, two separate multivariate analyses of variance (MANOVAs) were used to assess pre-to-post changes in CS-PAM scales and CC-PES items. To adjust for multiple testing, consistent with recent literature (Provenzi et al., 2015), the Benjamini-Hochberg false discovery rate (FDR) procedure was used, setting $q < 0.05$. Basic assumptions for the use of MANOVA were checked, including assessment of normal distribution of variables through the Kolmogorov-Smirnov test. The Statistical Package for Social Sciences (SPSS) version 21.0 for Windows was used for data analysis. Open-ended responses were transcribed and analyzed using a content analysis approach. Two researchers (SB and GG) independently read and coded the open responses in order to detect recurring themes representative of the participants' viewpoints and recommendations for future improvements.

RESULTS

Preliminary Results

In the following paragraphs, data collected from two subsequent editions of the program are presented.

Participants' Characteristics

Overall, 49 nurses participated in the educational program. Among them, 46 completed both pre- and post-questionnaires and attended both sessions of the program. Of the three missing questionnaires, three pre-questionnaires were not filled because participants arrived late. Participants with missing questionnaires did not differ from included subjects for socio-demographic and professional characteristics. Mean age of included professionals was 47 years ($SD = 7.08$, range 25–58). Mean years of professional experience was 23.9 ($SD = 8.64$, range 3–35). The majority of participants were female (78.3%).

Instruments Reliability

The CS-PAM pre- and post-training Cronbach's alpha was 0.89 and 0.85, respectively. Cronbach's alpha for the CC-PES was 0.87 (T_0) and 0.89 (T_1). As such, reliability was high for both instruments. The PCA revealed a one-factor structure for the CC-PES, with PC1 explaining 51.2% of variance (eigenvalue = 4.61).

Program Outcomes

The mean scores of CS-PAM, $t_{(45)} = 4.05$, $p = < 0.001$, and CC-PES, $t_{(45)} = 3.48$, $p = 0.001$, increased from T_0 to T_1 .

CS-PAM Outcomes

The attitudes of participants toward patient activation and self-management behaviors changed from the beginning to the end of the program, $F_{(4, 42)} = 5.08$, $p = 0.002$, $\eta_p^2 = 0.33$. Scores

improved for each of the CS-PAM categories (Table 2). Testing for False Discovery Rate (FDR) did not affect the results and no significant mean comparison was excluded.

CC-PES Outcomes

Participants also reported improvements in their confidence toward enacting patient engagement strategies, $F_{(9, 37)} = 2.26$, $p = 0.039$, $\eta_p^2 = 0.36$. Specifically, an increase was observed in 8-out-of-9 skills depicted by the CC-PES (see Table 3). Testing for FDR did not affect the results and no significant mean comparison was excluded.

Participants' Experiences and Satisfaction

Overall, on a scale from 1 to 10, satisfaction with the program was high (mean = 8.62), as well as the level of participants' interest toward the course's contents (mean = 8.76), the perceived course usefulness (mean = 8.65), the quality of teaching (mean = 9.07), the quality of the didactic materials (mean = 8.77) and of the interactive sessions (mean = 8.79).

Moreover, from the qualitative analysis of participants' responses, several themes emerged to describe the most useful aspects of the program. Participants gave highly positive feedback after the program. Participants found the course to be interesting, useful, enjoyable, informative, and highly relevant to their own nursing practice and for teaching junior staff.

Summarizing and analyzing participants' written reports resulted in the following achievements:

Learning through sharing. Particularly, nurses reported highly positive feelings related to the opportunity to *share and discuss their experience* with colleagues belonging to different clinical units within a safe and stimulating learning environment. The heterogeneous group nature of the program was, in this sense, particularly valued as a fundamental factor to promote a safe climate of discussion and feedback.

Learning by doing. Moreover, participants highly valued the possibility to participate in role-playing and case scenarios with simulated patients. This allowed them to directly experience contents explained during the didactic sessions and to reflect upon them by receiving feedbacks from other participants and facilitators.

Valuing the nursing professionalism. The valorization of the nursing profession and the capitalization on the past professional experience was then considered a core aspect of the program and was highly appreciated by participants who felt professionally recognized. The contents of the course were perceived as a systematization and valorization of the daily work of nurses.

Time for self-reflection. Furthermore, the possibility to reflect while learning allowed nurses to *question and discuss daily practices with patients and clinical hidden assumptions*. Particularly, participants reported as a value of the program the space given to reflect upon clinical models and cultures that often are enacted without any critical thinking. Nurses commented on the importance to build a culture of patient engagement, which emphasize patients' autonomy, and is respectful of the patients' needs along the care process.

TABLE 2 | Descriptives for CS-PAM scales and mean comparison statistics.

Scales	T ₀				T ₁				Mean comparisons	
	Min	Max	Mean	SD	Min	Max	Mean	SD	F	p
Adherence	2.75	4.00	3.58	0.43	2.50	4.00	3.76	0.37	7.72	0.008
Independency	2.00	4.00	3.38	0.51	2.67	4.00	3.72	0.36	17.29	0.000
Partnership	1.75	4.00	3.49	0.55	2.75	4.00	3.73	0.38	8.58	0.005
Information seeker	1.00	4.00	3.05	0.62	2.50	4.00	3.38	0.38	11.83	0.001

TABLE 3 | Descriptives for CC-PES items and mean comparison statistics.

Items	T ₀				T ₁				Mean comparisons	
	Min	Max	Mean	SD	Min	Max	Mean	SD	F	p
Assessing the level of patient engagement	3.00	5.00	3.76	0.52	3.00	5.00	4.09	0.66	7.31	0.010
Promoting patient engagement	2.00	5.00	3.78	0.63	3.00	5.00	3.96	0.59	3.04	0.090
Motivating patients in following medical prescriptions	2.00	5.00	3.65	0.71	3.00	5.00	4.02	0.54	10.58	0.002
Informing patients about disease and treatments	2.00	5.00	3.80	0.62	3.00	5.00	4.09	0.51	5.64	0.022
Assessing patient's health literacy	2.00	5.00	3.72	0.72	3.00	5.00	4.02	0.54	6.04	0.018
Empathizing with patients	2.00	5.00	3.93	0.68	3.00	5.00	4.24	0.60	8.08	0.007
Assessing and managing difficult patients' emotions	2.00	5.00	3.50	0.75	3.00	5.00	3.80	0.65	6.04	0.018
Effectively communicating with patients and families	2.00	5.00	3.80	0.69	3.00	5.00	4.09	0.55	4.68	0.036
Developing/maintaining relationship with patients and families	3.00	5.00	4.15	0.56	3.00	5.00	4.37	0.53	4.48	0.040

Innovation for the nursing paradigm. Finally, the course was also perceived useful due to the *innovativeness* of its contents. This was perceived to be a step forward from the epistemology of patient-centeredness to concrete strategies of action to actually promote the centrality of people along the care process. The possibility to experience and understand the *relational value* of patient engagement measures was perceived as a useful aspect of the training. Having assessment measures that concretely helps clinicians in detecting the patients' needs and expectation of being involved in their care allowed clinicians to reflect upon their relational strategies to really address the needs of patients. *Suggestions for improvements.* Regarding the least favorite aspects of the program and recommendation for the future, participants least liked its brief duration, and reported to desire more individual feedback on their patient engagement during the course and in their implementation in the actual clinical practice. Finally, participants recommended that the program in the future include the testimony of nurses who successfully employed patient engagement strategies in the clinical work with chronic patients and additional guidance on communication and relational strategies to improve the engagement of both patients and their caregivers.

Selected quotes and overall descriptive themes from open-ended questions exploring participant satisfaction and recommendation for future improvements are listed in **Table 4**.

DISCUSSION

In this paper we described the preliminary examination of the feasibility of a 2-session program (NET-PES) designed to

train nurses in adopting patient engagement measures to enact patient engagement strategies. Healthcare quality improvement efforts are increasingly focused on chronic illness care, and there will undoubtedly be heightened interest in monitoring the clinicians' performance in supporting patient engagement in the management of their health condition; at the same time, healthcare organizations need to train their clinicians in order to provide them knowledge and skills to effectively support the patient active role. Within this context, nurses are crucial actors for sustaining this paradigmatic revolution and promoting the imperative of patient engagement. Training the next generation of clinicians—and of nurses as one of the main patient's navigators along the complexities of the current healthcare environment—means not only to emphasize the relevance of supporting patients in self-care and self-management skills, but also to sustain the acquisition of skills and strategies to monitor and encourage patient engagement along the medical course.

This pilot feasibility study demonstrated that NET-PES was associated with observable improvements in nurses' attitudes and skills for promoting patients' engagement in chronic care management. Also, qualitative feedback from participants revealed that the program made them more confident in delivering patient engagement actions. These findings, despite the small sample size, suggest preliminary evidence that a brief educational intervention designed to train nurses in patient engagement assessment and improvement is not only feasible but holds promise toward potentially improve clinicians' patient engagement strategies. The achieved results also highlighted the potential for the NET-PES program to contribute to efforts aimed at improving the quality of healthcare for persons with chronic conditions. To date, interventions in the area of patient

TABLE 4 | Selected quotes and overall descriptive themes representing nurses satisfaction with the program and recommendation for future improvements.

Domains	Themes	Sample quotes
Favorite aspects	Learning through sharing	<i>Among the most useful aspect of this course I identify the group as a fundamental source of exchange and feedback (nurse, male, 27 years of professional experience). This is a different—but really effective—way to learn something new (nurse, female, 15 years of professional experience). The group allowed us to share our daily practice, by valuing each one experience (nurse, female, 25 years of professional experience)</i>
	Learning by doing	<i>I really appreciated the experiential moment. It was useful to observe encounters between a nurse and a patient from outside (nurse, female, 8 years of professional experience). It was really useful to experience a simulated consultation with a patient. It allows me to actually put in the patient shoes (nurse, female, 29 years of professional experience)</i>
	Valuing the nursing professionalism	<i>I appreciated that in this course emerged a strong valorization of the role of nurses... (nurse, female, 17 years of professional experience). This course helped me in systematizing what we (the nurses) just do when we have to motivate and engage patients in the medical course (nurse, male, 13 years of professional experience). I felt recognized as a key player in the care team (nurse, female, 4 years of professional experience)</i>
	Time for self-reflection	<i>I learned the importance of taking time to reflect upon our own clinical practice (nurse, male, 30 years of professional experience). I took home a great awareness about the complexity of the patient inner world. This means that what we (i.e., nurses) perceive is not necessarily aligned with the real patient experience (nurse, female, 19 years of professional experience)</i>
	Innovation for the nursing paradigm	<i>I did not expect that this course could offer concrete tools for the nursing practice (nurse, female, 22 years of professional experience). We are used to learn theories about putting the patient at the center of their care...it is much rarer to discuss and experience concrete strategies (nurse, male, 8 years of professional experience). I did not expect that this course could offer concrete tools for the clinical practice; I was not used to conceive patients' assessment as a relational instrument (nurse, female, 14 years of professional experience). I understood that patients' assessment is not a mere bureaucratic act...it could be useful in the clinical daily practice (nurse, female, 34 years of professional experience)</i>
Least favorite aspects	Brief duration Not enough personal feedback	<i>I would have preferred this program was longer (than two sessions). I wanted to talk more on each subject (nurse, female, 22 years of professional experience). I was never sure if the role-playings we enacted in the sessions was right (nurse, male, 19 years of professional experience). I would like to share my actual clinical experience to see how much I have improved my skills after the course (nurse, male, 26 years of professional experience)</i>
Recommendations for future improvements	Peer testimony	<i>To listen to successful implementation experience of others (nurses) who employed in their practice the acquired patient engagement skills. I would find their voice inspiring (nurse, female, 12 years of professional experience)</i>

engagement have largely concentrated on improving health providers' communication and relational behaviors and their empathy in the delivery of care (Kennedy et al., 2005; Morriss et al., 2006; Levinson et al., 2010; Otero-Sabogal et al., 2010; Cunico et al., 2012; Légaré et al., 2012). NET-PES was aimed to complement these established efforts by focusing on training engagement assessment measures particularly useful to identify the patients at high risk of disengagement and to adapt the health professionals' relational strategies of the basis of the patients' priorities and engagement needs.

Consistent with prior studies about interventions aimed at educating providers on supporting patient activation and engagement (Greene et al., 2007; Lamiani et al., 2012; Barello et al., 2015a), our findings may indirectly suggest that this program might be potentially helpful to enhance patients' healthcare outcomes. From this perspective, previous studies have demonstrated a positive correlation between the patients' perception of their clinicians' support for autonomy in self-care and the patients' level of engagement in self-management

behaviors and adherence to treatments (Graffigna et al., 2016). There are also studies that discussed the role of health practitioners' beliefs about the importance of patients being active agents in managing their health condition (Greene and Yedidia, 2005; Blakeman et al., 2006). The encouraging results of this study, unless preliminary, support the value of designing, and implementing educational initiatives dedicated to nurses among the actions to realize the patient engagement imperatives. In line with other experiences across countries, nurses may be envisaged as a powerful resource to promote organizational care models where this professional figure can absolve and foster the implementation of integrative approach to patient's care.

Despite the promising nature of these results, there are a number of limitations that warrant consideration. First, the use of pre-post design prohibits any inference of program effectiveness due to the lack of a comparison group and due the challenges to internal validity of this kind of research design as suggested by Vockell and Asher (1995). Moreover the single site study limits results' generalizability. This study design—frequently adopted in

research testing the feasibility and potential impact of educational interventions (Hulsman et al., 1999; Berger et al., 2010; Lamiani et al., 2011; Meyer et al., 2011; Bartels et al., 2013; Ledford et al., 2014; Arnold et al., 2015; Shah et al., 2015; Robinson et al., 2016; Viau et al., 2016) is appropriate for the demonstration of feasibility and for initial proof of concept, it is cost effective, and it is pragmatic because it allows for “real world” variability in variables. Consistent with the intent of a pilot study, our goal was to examine feasibility and potential impact. A determination of effectiveness will require a larger and appropriately selected sample, ideally employing a randomized controlled trial with follow-up measures to estimate the maintenance of achieved results across time. Finally, participants in our pilot study were recruited using a convenience sample. As attendees were largely self-selected, the surveyed group might be more open to reflection and self-improvement than a randomly selected group. Moreover, also the Hawthorne effect could have had an impact on the nurses’ engagement behaviors. Hence, we are unable to generalize our findings to a different population of nurses. Also the clinicians’ self-rating of self-efficacy and attitudes can be considered as a methodological weakness because of the risk of response bias. The nurses involved were not blinded to the intervention and the increase in their self-ratings can therefore reflect a willingness of the responders to show that the training course had been useful. Moreover, it should be noted that nurses’ T_0 scores were high in both CS-PAM and CC-PE (mean pre-training score = 3.43, $SD = 0.45$; mean post-training score = 3.68, $SD = 0.31$): this could be related to a self-selection bias because of voluntary participation and the risk of an over-representation of participants who believed in and supported patient engagement in care should not be completely neglected. Also, the majority of participants were women, and it is possible that men and women may respond differently to the program’s stimuli: previous research on patient-centeredness has indeed demonstrated that gender may affect the attitudes of clinicians toward being more or less open to involve patients in the care process (Roter and Hall, 2004; Sandhu et al., 2009). More inquiry is needed to determine how nurses implement teaching principles in their practice and how patient-related factors may affect that implementation.

CONCLUSIONS

These findings provide preliminary evidences that a brief educational program designed to improve nurses’ competences in promoting patient engagement is not only feasible, but holds promise toward potentially implementing this training in nursing educational curriculum. If proven in a future empirical trial, NET-PES program might provide a useful contribution to translate the principle of patient engagement from theories into actual practice.

Today’s care models require nurses to play many different roles: these include providing high quality and compassionate nursing care; the ability to adopt a multidisciplinary perspective to the patients’ care; partnering with patients and their families

(Barello et al., 2014, 2015a); valuing the patients’ perspective by collecting their care experience in an ecological way (Graffigna et al., 2011, 2015a). For these reasons, embracing a patient engagement care model and the necessary skills to enact it become vital components for the nurses’ education. However, to effectively realize these goals, clinicians have to be provided with reliable measures to assess their patient engagement levels and trained to effectively use them. There is an increasing need to provide more resources for effective training in this area if patient engagement is to improve. Such initiatives are also essential to ensure the professional well-being of nurses themselves by valuing and recognizing their crucial role in addressing the challenges of the current healthcare environment. If demonstrated effective, learning programs teaching clinicians how to use and act on patient engagement measures in clinical practice might be a key steps in supporting the realization of patient engagement in healthcare (Barello et al., 2015a; Graffigna et al., 2016). Researchers and clinicians from different clinical areas should collaborate to share good practices and develop guidelines to advance the field of patient engagement and shift from theory into practice.

The concrete implementation of patient engagement interventions and the adoption of patient engagement measures is a dynamic and challenging process, in which different factors play a role at both individual (professional, patients, caregivers) and organizational levels. We focused here on nurse’s training, but it is necessary to acknowledge the importance of other factors. First, patients and caregivers’ trainings in gaining an active role in their care are of paramount importance for patient engagement successful implementation. Studies exploring patients and caregivers’ attitudes clearly state that if their clinicians are open to patient’s autonomy and self-care behaviors, this will encourage patients’ active involvement in care (Graffigna and Barello, 2015b). Second, an organizational revision of care models (Bosio et al., 2012) and a pragmatic support for collecting and integrating patient engagement measurement into patient records and in the clinical workflow is a required step. Finally, using opinion leaders and specific organizational change strategies also appear pivotal to maximize the possibilities of innovation’s adoption.

AUTHOR CONTRIBUTIONS

GG and SB developed the research design, coordinated data collection and wrote the manuscript. LP performed statistical analyses and give clinical supervision about the contents. GP, MM, and MC revising the work critically for important intellectual content. The authors have approved the article and agree to be accountable for all aspects of the work.

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Conflict of Interest Statement: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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