THE SOCIOCULTURAL CONTEXT OF PSYCHOSOCIAL INTERVENTIONS

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THE SOCIOCULTURAL CONTEXT OF PSYCHOSOCIAL INTERVENTIONS

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Across diverse academic fields, scholars and practitioners are engaged in developing interventions to promote outcomes like health and quality of life. Indeed, such is the apparent efficacy of such interventions, that there are many policy-led initiatives to implement these at national and international scales. However, few scholars or practitioners have thought in any systematic and critical way about the importance of contextualizing these interventions, i.e., considering how the impact of such interventions may be affected and mediated by specific sociocultural factors (from gender, to ethnicity and socio-economics).

The aim of the Research Topic "The Sociocultural Context of Psychosocial Interventions" was to address this lacuna. As such, we tried to help bringing a more 'contextual' mindset to the implementation of health and wellbeing interventions. This may help to shift the way such interventions are designed and implemented, both at a granular local level (i.e., influencing individual practitioners) and at a large-scale macro level (e.g., influencing policy makers).

Themes within this Research Topic have concerned both macro-sociocultural as well as meso-and micro-layers, and the peculiarities of implementing real world research based on these levels. There has been room for physical and mental health, for family relationships, for educational contexts and even for the effects of crime. Some works have included interesting methodological discussions on the integration of different ecological layers or the modal distribution of our interests.

For us it has been very important to work giving a greater diffusion to these issues since, considering psychosocial interventions in the context in which they occur, goes beyond an epistemological or methodological discussion. Rather, these considerations seriously affect the ability of practitioners to really reach the people who need their interventions, listening to their needs and respecting their preferences. For the editors of this book, then, the contextualization of interventions means considering the people who receive them as full citizens immersed in complex societies where factors such as social justice and health or well-being do not float apart in space but affect each other dialectically. We therefore think that the duty of both academics and practitioners is not to forget that it is as important to evaluate the direct effect of our interventions as the influence we have in the society as a whole when we carry them out.

We hope you enjoy reading these works and that their dissemination stimulates new lines of research committed to both good practise and social transformation.

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EDITORIAL
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Editorial: The Sociocultural Context of Psychosocial Interventions

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Editorial on the Research Topic

The Sociocultural Context of Psychosocial Interventions

During the last decades several authors have criticized the arrival of evidence-based practice (EBP) approaches due to its contribution to the decontextualization of psychosocial and behavioral health interventions (Coghlan and Brydon-Miller, 2014). The term decontextualization here refers to the fact that, although it is not something explicit in its foundations (Leach, 2006), EBP researchers have in practice largely prioritized the internal over the external validity of evaluations. In other words, the problem is not the use of evidence against tradition, intuition or power; but that what has been considered "good" evidence has been narrowed to be aligned with the dogma of behavioral sciences' neutrality. In relation to this alleged neutrality, Cushman (1990) already discussed the historical configuration of the "self" concept. Psychology, he argued, has the role of "*healing the empty self (. . .) without being able to address its historical causes,*" (pp. 599) thus being responsible in part for the very process of "emptying" which it claims, and aims, to address. Cushman argues that psychology strives to appear as a neutral science, while its interests—its subjects, ideologies and businesses—have "historical antecedents, economic constituents, and political consequences" (p. 600). In this regard, Sampson (1989), gives evidence that the transformation of psychology as a discipline in which the functional unit of social order has moved from the community and the household to the individual level, as postmodernism and globalization have exerted their influences. The theory of the individual as a rational and self-directed entity has produced several industries that are responsible for ensuring the proper functioning of a newly disembodied self. Among these, mental health, which has focused on eliminating symptoms regardless of their source, has become one of the fastest developing industries during the past century.

It would be unfair not to recognize countless theoretical and empirical efforts carried out especially in the last 30 years by researchers who have delved into the covariates of assessment and intervention success related to class, gender, race, or culture. However, many of these aspects remain underrepresented in the training of practitioners, research budgets and outreach activities aimed at the general public. All these strategic areas are still highly biased toward individualistic (Thomas, 2016), brain-centric (Pérez Álvarez, 2011), and positivistic (Williams and Garner, 2002) views of evaluation. In spite of this, there have been well-established calls for the integration of biological, psychological and social determinants of health and wellbeing (Engel, 1977; Cloninger, 2006) and for the mixture of methods of inquiry (Poses and Isen, 1998), however the reality today is far from being balanced.

The aim of this research topic is to bring a more "contextual" mindset to the implementation and evaluation of health and wellbeing interventions. Our main objective is to contribute to the shift in the way in which such interventions are designed and implemented, both at a granular local level (i.e., influencing individual practitioners) and at a large-scale macro level (e.g., influencing policy makers).

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In the selection of articles for this special topic we have considered the need to promote self-critical research on underrepresented contexts and innovations that provide greater awareness of excluded groups.

Firstly, some of the articles give special importance to the macro-sociocultural context where interventions take place. In this sense, Giordano et al. present a group analytic intervention conducted within a 'Community Based Participatory Research' in an area oppressed by the Mafia. Their qualitative findings revealed the development of an awareness process that allowed the participants to recognize the strong emotional impact related to the Mafia's presence in their lives. Additionally, Eiroa-Orosa and Rowe reflect on how to transfer the principles and practice of Citizenship in Mental health to different sociocultural contexts. Citizenship is a novel approach to mental health based on people's connections to the rights, responsibilities, roles, resources and relationships that society offers to them through public and social institutions. The authors evaluate the multiple possibilities that this approach has, without forgetting the neo-colonialist risks of exporting ideas from a prestigious academic institution to daily reality in a distant context. Macro factors such as gender have also been addressed in this special topic. Wang et al. performed a metanalysis on gender differences when reporting depressive symptoms in non-clinical populations. Their results appear to confirm the so-called "female preponderance" in the level of self-reported depressive symptoms in the general population, giving support to social gender role theories in explaining these differences.

On another level, other articles on this special topic address meso-levels such as schools and the peculiarities of implementing real world research in these settings. In this line, Orosz et al. present a self-critique of their Growth Mindset intervention whose positive effects were not as long lasting as they previously expected. These authors discuss how contextual factors, such as the focus on non-problematic students or the implementation of the intervention in a non-transition stage may have been the reasons for its lack of effectiveness. Similarly, Andrés-Rodríguez et al. adapted their evaluation methods in order to be conducted by teachers in the school context. This involved a curricular intervention including education and social contact with persons with lived experience of mental disorders. They discuss the advantages and limitations of implementing a flexible methodology in the classroom including social contact with stakeholders.

A topic that has attracted much attention is the immediate environment of people affected by physical and mental health problems. Considering the importance of family care, Delle Fave et al. explore happiness, goals, and meanings among persons with multiple sclerosis and their caregivers, including socio-cultural aspects that may crucially contribute to their functioning. The authors stress at positive adjustment to the disease evolves as a result of the development of personal and family resources both among persons affected as well as their caregivers. Shi et al. examine the association between family function and self-esteem of Chinese university students with and without grandparenting experience, exploring the moderating effects of social support within this relation as well. The same authors also evaluated the effect of Systemic Therapy as they compared supportive therapy to a 6-month treatment for students who were at a clinically higher risk of developing psychosis. Although no time by treatment interactions were found, the authors discuss the possibilities of an intervention that fosters a resource-oriented mindset focusing on solutions rather than problems. Moreover, Tang et al. examine the linkages between family factors at the whole, dyadic, and individual levels and two dimensions (affective and behavioral) of Oppositional Defiant Disorder symptoms among Chinese children. In their analyses, the most proximal factors (parent-child relationship and child emotion regulation, which were directly related to subsequent child internalizing problems) were significantly related to child behavioral defiant symptoms.

Finally, in this special topic there is also room for methodological discussions. Trying to integrate two very different worlds, Ray et al. explore the possibilities of functional neuroimaging at interpersonal and group levels. These authors attempt to embed their findings within classic concepts such as 'collective consciousness' or 'crowd'. Furthermore, Buckley et al. report results from a post-programme survey of participants in a non-profit outdoor health programme predominantly targeted at women with families. In this study they test their hypothesis that the population-scale distribution of interest in nature exposure may be bimodal rather than unimodal. Under the bimodal view some individuals would be heavily addicted to nature-based outdoor activities, and others indifferent or indeed repelled by them.

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Promoting Awareness about Psychological Consequences of Living in a Community Oppressed by the Mafia: A Group-Analytic Intervention

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The effects of the Mafia have been extensively studied from sociological, economic, and historical points of view. However, little research has investigated the influence of the Mafia on individuals and communities in terms of its psychological and social impact. In order to contribute to the advancement of our understanding of the psychological effects of the Mafia on individuals and communities and to promote a participative process of social change, a group analytic intervention was conducted within a Community Based Participatory Research carried out in Corleone, a small Sicilian town with a historically recognized role in the evolution of the Mafia, as well as in the fight against its control. Qualitative findings from the group intervention revealed the development of an awareness process that allowed participants to become aware of their social unconscious anxieties and defenses and to recognize and manage the strong emotional impact related to the Mafia's presence in their lives. Highlighting how psychological processes can have negative impacts on individual and collective capacity to pursuit transformation and resilience, this article provides important insight on how clinical psychology may operate in socio-cultural contexts to promote the reconstruction of the traumatic social dimensions in the community.

Keywords: Mafia, Antimafia, social unconscious, group analysis, group process

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INTRODUCTION

The Mafia is a criminal organization that uses the strength of intimidation to commit crimes and to acquire the management and control of economic activities, public contracts, and services, in order to enhance the control of territory and economic profits (Paoli, 2003; Finckenauer, 2005; Lavorgna and Sergi, 2014). The Mafia's presence has been negatively affecting social and economic development in Italy from more than 150 years (Gambetta, 2000; Daniele and Marani, 2011; Dalla Chiesa, 2014). Specifically, by means of violent and illegal instruments, the Mafia hinders Italian financial, cultural, and social development through its members' infiltration in the most important institutional and political networks (Ruggiero, 2010; Pinotti, 2015; Savona, 2015).

Similarly to other criminal organizations, the Mafia shares in particular with gangs some social and cultural features as the achievement of a higher status, power, respect and a sense of protection from social threats (Taylor et al., 2008; Goldman et al., 2014; Travaglino et al., 2014). With regard to southern Italian countries, criminal organizations provide a distinctive social identity and a sense of belonging that may be particularly appealing to youth that confront high levels of job insecurity, unemployment, and weak youth policies (Travaglino et al., 2014; Di Blasi et al., 2016). Nevertheless, the Sicilian Mafia shows some distinctive characteristics such as a more formal interne structure, well-defined economic activities and political relationships, specific codes of conduct, especially when compared to gangs (Paoli, 2003; Travaglino et al., 2014). Moreover, the Mafia maintains the monopoly of violence and the control of a territory since it illegally ensures several services that the State is unable to provide to citizens (Gambetta, 1993; Paoli, 2003, 2004; Schneider and Schneider, 2005). It should also be considered that the Mafia requires swearing a lifelong allegiance to its membership (Paoli, 2003; Lo Verso and Lo Coco, 2004).Violence is used to ensure the absolute obedience of the affiliates and to punish those who betray or disrespect the rules and the authority of the group. The menace or the effective use of violence is also employed to impose obedience on the communities in which Mafia exerts its influence and to frighten or eventually eliminate those who oppose the power and the economic activities of the criminal organization (Paoli, 2003; Travaglino et al., 2014).

Through an extensive infiltration of the vital fabric of the communities, the Mafia exerts a form of social control that can be described as a "private security system" that ensures protection, while imposing the respect of the criminal organization's rules (Gambetta, 1993). This social control implies submission and *omertà* (the law of silence) for both affiliates and citizens (Paoli, 2003; Lo Verso and Lo Coco, 2004). *Omertà* requires the duty of loyalty, obedience, and silence of affiliates and when these rules are transgressed members are punished through the use of violent and cruel acts (Paoli, 2003). However, the law of silence applies to everyone else and implies to avoid the collaboration with the forces of law and order if being victims or witnesses of illegal activities (Lo Verso and Lo Coco, 2004).

Travaglino et al. (2014, 2015, 2016, 2017), in their recent studies, clearly highlighted how in the Italian criminal organizations *omertà* is a key socio-cultural dimension linked to the concepts of honor and masculinity. The link between honor and masculinity is evident in linguistic expressions such as "men of honor" (Paoli, 2003), which describe the affiliates to the Mafia. A "man of honor" is required to be able to avenge crimes and offenses without the help of the authorities and police. Specifically, individuals' higher acceptance of *omertà* and honor-related ideology was related to both the perception of greater legitimacy of the criminal organizations and fewer collective action intentions against them.

As highlighted in recent literature (Natale et al., 2013; Di Blasi et al., 2015; Nardin et al., 2016; Travaglino et al., 2017), the Mafia affects the Italian context not only from the economic and political points of view but it also weakens the whole community system compromising social capital, values, social identities, and mental representations. This study aimed to deepen these aspects and to show the need for a psychological intervention in social contexts in order to weaken the negative impact of the Mafia on individuals and communities. While the effects of the Italian organized crime have been widely examined from sociological, economic, and historical points of views, little research has focused on the influence that the Mafia exerts on individuals and communities in terms of its psychological and social impact. Over the past two decades, our research team has been studying the Mafia from a psychological perspective in order to achieve a deeper understanding of the culture and mentality underlying the Mafia behaviors from interviews with relatives and affiliates and clinical reports of Mafia members' psychotherapies. These studies have highlighted how the Mafia generates suffering and distress both in its family's members and in its victims and how the identity of Mafia members is inextricably nested in the culture of the families and the community to which they belong (Lo Verso and Lo Coco, 2004; Giordano and Di Blasi, 2012; Giordano and Lo Verso, 2015; Mannino et al., 2015). Moreover, these studies suggest that the identification with the group represents a core dimension in order to explain affiliates' and citizens' attitudes, behaviors, and the overall Mafia's culture (Lo Verso and Lo Coco, 2004; Schimmenti et al., 2014; Mannino et al., 2015). Finally, prevalent denial defense mechanisms to cope with emotions, inflated self-representations, and the lack of guilt observed in Mafia members suggest the presence of specific psychological traits (Lo Verso and Lo Coco, 2004; Schimmenti et al., 2014; Giordano and Lo Verso, 2015).

Recent literature has also provided additional psychological and sociological contributions that examine processes of social change in opposition to the Mafia's control promoted by activists, citizens, and associations. Specifically, the anti-racketeering campaign promoted by Addiopizzo's in Palermo from 2004 represents a significant attempt to carry out long-lasting cultural and social changes involving the civil society and the public administration. Addiopizzo in a unique experience of a bottom-up Antimafia strategy aimed to promote extortion resistance through entrepreneurs' public refusal to pay the racket to the local Mafia and encouraging consumers to practice a critical consumption buying good from firms that joint Addiopizzo. Recently, cultural and social determinants of firms' decision to join Addiopizzo have been investigated (Vaccaro, 2012; Giannone and Ferraro, 2015; Vaccaro and Palazzo, 2015), in parallel with other studies that explored values that can prompt entrepreneurs and consumers to engage in anti-racketeering actions (Natale et al., 2013; Gunnarson, 2015; Elsenbroich, 2016; Marin and Russo, 2016). Nonetheless, little has been written about the Mafia from a psychological perspective taking into account the individual and collective consequences of living in a community oppressed by the presence of a criminal organization.

This article would address this paucity of research showing how the concepts of social unconscious and group processes elaborated by group-analysis may help to understand how social constraints imposed by the Mafia's control over the community impinge on the individual psychological functioning. The group-analytic perspective focuses on the interrelations between social, interpersonal and the intrapsychic levels of individual life experience (Foulkes, 1948). Recently, the concept of the social unconscious has increased in importance in the group-analytic literature. Although Foulkes (1964) mentioned this concept to describe the social and communicative forces affecting interpersonal and transpersonal processes, he did not elaborate further on this concept. The social unconscious has been subsequently defined as a dimension inherent to the social level and structured by social power relations: it is co-constructed by members of the same society or culture and contains the shared unconscious fantasies, myths, anxieties, defenses, and memories of a specific community (Dalal, 2001; Weinberg, 2007). Hopper provided a detailed definition of this concept: "The concept of the social unconscious refers to the existence and constraints of social, cultural, and communicational arrangements of which people are unaware. Unaware, in so far as these arrangements are not perceived (not "known"), and if perceived not acknowledged ("denied"), and if acknowledged, not taken as problematic ("given"), and if taken as problematic, not considered with an optimal degree of detachment and objectivity" (2001, p. 10). Additionally, Brown (2001) stated that the most powerful defenses to avoid pains or deep conflicts linked to social traumatic experiences or painful historical events are denial, projection, and the repression of memory.

Recent research developments have focused on the analysis of the social unconscious in relation to social trauma and transgenerational enactments, as in the case of the Holocaustus (Wilke, 2016), the Palestinian and Israeli conflict (Doron, 2014; Even-Tzur, 2016), and to social inequalities linked to cultural hierarchies of sexism, racism, classism (Layton, 2007; Geyer, 2017).

These studies addressed the question of how enactments of the social unconscious can be studied on a wider social level in clinical and non-clinical settings and showed how trauma or social, cultural, and political events stay alive in the collective consciousness of groups and communities by means of shared stories and narratives. In line with these studies, we hypothesized that the presence of the Mafia in the vital fabric of a community, as a permanent painful dimension affecting social power relations, requires specific social, cultural, and communicational arrangements of which people are unaware, or that are not known or denied. The obedience to the law of silence (*omertà*), strengthened by the Mafia through the massive use of violence and intimidation, contribute to maintain not only the *status quo* but also to the enactments of social unconscious elements that weaken citizens' capacities and intentions to oppose the Mafia. Adopting this point of view may help recognize psychological, cultural, and social dimensions that may increase individual and collective support for the status quo (Travaglino et al., 2017), and overcome reductive perspectives according to which Sicilian people display a collective passivity in opposing the Mafia only due to legitimization processes (Sciarrone and Storti, 2014) or to widespread forms of social consensus linked to ethical bases such as amoral familism (Banfield, 1958).

According to Foulkes (1948, 1964), each individual is inevitably determined by the world in which he lives, by the community, the group of which he forms a part and common social perceptions and experience linked to this belonging penetrate the inmost being of the individual personality.

As within the group analytic approach the group allows the analysis of both the individual and social unconscious, it is a very appropriate instrument to help people develop an understanding of their psychic and social experience. Providing a theoretical and clinical container to elaborate thoughts and feelings (Foulkes, 1964), the group experience promotes a change by creating a holding environment for analyzing, understanding, and becoming aware of the social unconscious dimensions in the group and in society (Weinberg, 2007).

The present study reports a group analytic intervention aimed to enhance participants' awareness through a shared reflective process about the social unconscious dimensions, in terms of affects and defenses, linked to the presence of the criminal organization in the territory. The intervention took place within a Community Based Participatory Research (CBPR) conducted in Corleone, Sicily (Italy), a territory with a significant historical role both in the evolution of the Sicilian Mafia and in the fight against Mafia organization. Specifically, during the years 1970-1980, the clan of Corleonesi, headed by Luciano Liggio and Salvatore Riina, became dominant within Cosa Nostra. In the early 1980s, Riina and Provenzano decimated other Mafia clans through a fierce mafia war that caused hundreds of murders including heads of rival clans and members of Antimafia (Andretta, 2005; Lupo, 2010). Many of these victims were children and women who were often murdered by horrifying methods, such as being liquefied into acidic compounds (Schimmenti et al., 2014). On the other hand, the Anti-mafia movement in Corleone has an important tradition that since the nineteenth century began with trade union activists and cooperatives of agrarian workers (Santino, 2009; Rakopoulos, 2017; Scalia, 2017). With this regard, the story of Placido Rizzotto, a union trade activist killed by the Mafia in 1948, is emblematic since his mutilated body was found only in 2009. The figure of Rizzotto has been evoked in numerous films and television series and a red wine produced by the Antimafia cooperative "Libera Terra" still bears his name. Moreover, the first confiscated good, belonged to Riina the leader of the Sicilian Mafia, was a vineyard located in Corleone. In 1999, the plot was conferred to the "Lavoro e Altro" cooperative. As Rakopoulos (2017) underlined, this was the beginning of the formation of an Antimafia cooperativism system in Corleone territory that today involves about 10 agrarian cooperatives across Italy. As mentioned above, in order to understand the relationship between Corleone citizens and the Mafia, it is important to consider that social and historical trauma or/and cultural and political events can persist in the collective consciousness of groups and communities producing a damning impact in terms of individual and collective psychological functioning (Mohatt et al., 2014; Geyer, 2017). Moreover, to avoid conflicts and anxieties linked to these social traumatic events, powerful defenses as denial, projection, and the repression of memory are activated at a social unconscious level (Brown, 2001). These unconscious psychological arrangements are reinforced by some cultural, political and anthropological aspects such as *omertà* and a widespread distrust in institutions and law enforcement, too often ineffective or colluding with the criminal organization. The interplay between psychological, socio-cultural, and political elements affects the capacity of Corleone, and overall Sicilian, citizens to successfully oppose to the Mafia's control over their lives.

METHODS

The present study sought to analyze a group-analytic intervention carried out in order to promote participants' awareness of the adverse effects of the criminal organization on their lives. Group meetings took place within a CBPR conducted in Corleone, Sicily. The CBPR started in January 2011 and ended up in January 2012 and was developed through three interrelated steps: the context analysis, the group meetings, and the final public meeting. The partnership was composed by the academic team (two senior researchers, who were also the conductors of the groups, and two facilitators), the Municipality of Corleone, and a group of community members (local administrators, teachers, college students, housewives, and representatives of Antimafia associations).

The Context

Since the Mafia spreads differently depending on socio-cultural features that characterize a specific geographical area, a strong focus was primarily maintained on the request for the intervention and on the link between such a request and the current features of the local community. Specifically, the request for the research was introduced to the team by a psychology student at the University of Palermo who had previously met the City Mayor. Following the request reported by the student, the team met the mayor, and other local administrators individually in order to clearly define the motivations underlying the request for the intervention.

As mentioned above, the Mafia has affected the history of Corleone and its citizens' social imagery, and some relevant events had occurred at the time when the intervention was requested. Some months earlier, one of the mafia boss Salvatore Riina's sons returned to Corleone to undergo preventive special surveillance, after he had been imprisoned for 8 years and 10 months for Mafia association crime. This event upset Corleone's inhabitants: once again they were forced to share their everyday living spaces with a Mafia bosses' family members and experienced worries about the risk of overbearing behaviors and threats to individual freedom and safety. In fact, only a few bosses' family members have chosen to live outside Corleone during their relative's hiding or imprisonment. Moreover, many bosses' siblings, children, grandchildren, wives, and sisters-in-law currently live in Corleone. As a consequence, Corleone's citizens often meet Riina's wife or Provenzano's children during their daily commitments. Acting on behalf of most Corleone citizens, the mayor took a very courageous stance. He declared to the press: "Riina's junior is not welcome here." First, such a strong public declaration produced the mayor's awareness that he was not opposing only an individual (Riina's junior) but a criminal organization as a whole. With regard to citizens, they had to take a stance: either with the mayor or with the Mafia. Most of the citizens had no doubt over which side to choose and felt represented by the mayor's words, however, it is likely that some felt unable to decide because the conflict was too strong. Furthermore, for a small minority, the mayor's statement was probably interpreted as an offense to the criminal organization.

Five meetings with the mayor and local administrators allowed the team to better understand the meaning of the intervention request: on one hand, the mayor and municipality, at a sensitive time in the town's history, wished to strengthen their protection network by giving voice to many honest Corleone citizens' consensus; on the other, they wished to provide citizens with a space where they could elaborate feelings prompted by the presence of the boss's son in town. Drawing on these lines, after the meetings, the following goals were planned: (a) helping Corleone citizens to share emotions and fears linked to their cohabitation with the Mafia; (b) promoting an emancipation process from Mafia oppression, starting from an increased awareness of the thoughts, behaviors, and feelings that contact with the Mafia arouses in people; and (c) analyzing the psychological and social consequences produced by organized crime in the local population and detecting consensual future developments.

Moreover, the psychology student, who acted as a facilitator, was in charge of inviting people to participate in the CBPR introductory meeting during which the team announced that it would be possible to register for discussion group meetings so as to actively participate in the CBPR. Given that some people might need more time to decide to actively intervene when a relevant them such as the Mafia is dealt with, researchers added that people could attend the first group meeting even though they had not previously enrolled.

Group Meetings

The CBPR approach conceptualizes community as a dimension of collective and individual identity in which individuals share common symbolic systems, social norms, interests, and needs(Israel et al., 1998; Wallerstein and Duran, 2008).Consistent with this theoretical framework, the group analytic approach focuses on the interrelations between individuals, families, community, and social network (Foulkes, 1948). Specifically, group analysis is based on the principle that human beings are deeply social by nature since their lives are inextricably linked in many ways. Given that group membership reflects the wider norms and values of society, the group is a privileged tool to explore social unconscious dimensions and promote deep lasting attitude changes by facilitating interaction and for exploring social unconscious (Dalal, 2001, 2011).Consistent with other participatory research studies utilizing psychodynamic groups in educational and work settings (Leclerc and Maranda, 2002; Newton and Goodman, 2009; Giannone et al., 2015), the group work promotes participants' interactions, aiming at an intersubjective analysis of their experiences and feelings. The conductor's capacity for holding is crucial to creating a safe communicative space (Newton and Goodman, 2009) in which feeling and emotions can help participants in collectively reconsidering their relations to each other and to their environment.

As previously pointed out, one of the biggest difficulties in the fight against the Mafia is *omertà,* defined as an implicit law of silence that originates from a fear of retaliation, and conveys a direct or indirect solidarity with the authors of a crime. As a consequence, participating in a group where publicly talking about the Mafia is required means, first of all, breaking the law of *omertà*. An individual who expresses his/her point of view against the Mafia is, at the same time, engaged in an action of strong political, social, and individual significance, thus experiencing deep unpleasant emotions. For this reason, it is particularly important to build a relationship based on trust with the partnership, as highlighted in the literature (Christopher et al., 2008; Shalowitz et al., 2009; Lucero et al., 2016).

In this study, the group meetings took place at the Mafia boss Bernardo Provenzano's house, a seized asset and the seat of some cultural associations committed to the fight against the Mafia, which promotes awareness campaigns and training courses. Currently named the Legality Lab, it is located close to the house where Provenzano's brother still lives. This location contributed to making the experience emotionally intense due to repeated intimidating acts which have been occurring over the last 20 years in Sicily and Corleone against the managers and users of seized and confiscated assets.

The team expected a numerically poor participation to the group. During the introductory meeting, only 14 citizens had registered to participate in group meetings. Despite being the first time that research against the Mafia involving citizens' active participation had been proposed in Corleone, unexpectedly, 31 citizens attended the first group meeting. Since the number of participants was greater than fifteen, the research team decided to set up two groups, each with a conductor and a facilitator. Only one of the two groups was entirely recorded and transcribed after consent from all participants was obtained, while the second group could not be registered because two participants did not give their consent. In the latter group, the facilitator acted as a recorder, transcribing the most significant sentences and aspects of the process. The group's work included three meetings of two sessions, lasting for an uninterrupted 75 min with one break of 20 min, using an intensive group format over 3 days. Two senior researchers, who are also group analytic therapists, acted as conductors.

The first group consisted of 16 participants (9 female), aged between 18 and 62, while the second group consisted of 15 participants (7 female), aged between 19 and 58. Two group-analytic therapists lead the groups, encouraging the establishment of a free-flowing discussion. The group analytic therapist tends to be referred to as a "conductor" rather than a "leader" (Foulkes, 1948). This reflects both a strong democratic principle that underlies the group analytic approach, and an understanding that the therapist's role is not to start a discussion or overly direct the group, but to interact in order to help the group make sense of the stories that are told, the feelings that resonate, and the resonances and dissonances that arise.

RESULTS AND DISCUSSION

The present study reports the experience of a group analytic intervention aimed to enhance people's awareness through a shared reflective process about the social unconscious dimensions linked to the presence of the criminal organization in the community of Corleone. In this section, we sought to highlight some significant passages characterizing the group process that led participants to recognize and share unconscious or unknown feelings and emotions linked to the influences of the criminal organization on their lives. During the early phase of the first group meeting, discussion of the Mafia took place at a shallow level, highlighting both the need for putting aside an early personal and emotional involvement and the presence of denial defenses to avoid pains or deep conflicts linked to the social unconscious dimensions related to the Mafia (Brown, 2001; Hopper, 2001). Thereafter, starting from some interventions about personal daily experiences, the law of *omertà* was broken, thus allowing most of the members to express doubts and difficulties in sharing everyday spaces with bosses and their relatives. These shared narratives represented the first important passage allowing participants to voice the "unthought known" (Bollas, 1989) about the Mafia and showed the presence of specific social and cultural constraints of which participants were unaware before (Hopper, 2001). The possibility to consciously think about their past and current experiences allowed participants to make a further progress during the second meeting, coming into contact with painful feelings and emotions, and making them available for thought. Through this second passage, participants could start to recall traumatic memories and events and recognize that they had built strong defenses and developed specific behaviors to avoid the sufferance the Mafia had caused in their lives (Weinberg, 2007). During the third group meeting, the previous emotive and cognitive engagement helped participants to recall and re-interpret their current and past experiences, increasing awareness of the limitations of freedom and expression that the Mafia imposes on their lives and their community. Becoming aware of this influence and sharing ideas about the modalities available for coping with it was the third and final step of the group process.

First Group Meeting*:* Voicing the "Unthought Known"

At the beginning of the first group session, the conductor asked the participants to introduce themselves and explicate their expectations with respect to the experience they were about to start. Everybody initially assumed a "passive" position with respect to the group experience. Many participants seemed to be pushed by curiosity and intellectual interest, rather than the need to share lived experiences and thoughts connected to the Mafia. Moreover, as the following quotes show, great diffidence and skepticism initially seemed to linger in the group, and many participants expressed a stronger need to listen rather than intervene.

> *My name's Laura, I'm an office worker at school. The topic we are talking about is interesting; however, I'm not quite sure things will change. Sometimes it seems that everything is going to change but eventually it remains the same... In short, I count on young people...*
>
> *My name's Mario, today I'm here as a listener, may I?*
>
> *My name's Piero, I'm an estate agent and curious to listen to all of you.*

> *I'm a police commissioner. I'm here because I've been invited. I'm not here to talk because in that case I should be authorized by the police Headquarters...*

However, some young participants started to show a greater propensity to confront and share their experience from the beginning.

> *My name's Paola and I'm from Corleone. Going into this theme is such a great pleasure for me because I'd like to have a confirmation about the stories my parents have been telling me. I'm pleased to talk about this theme with people of my age, as well as with all the people present here.*
>
> *My name's Maria, I've got a degree in Educational Science and I'm working in Palermo. I chose to participate because I consider this topic very interesting and also wish to talk about my experience.*

Thereafter, some participants started to discuss the Mafia through a narration of their personal experiences, specifically talking about the "law of silence." They spoke about the trouble of living in a city where they often feel observed not only by the Mafia but also by law enforcement agencies and others citizens. They also reported episodes that highlight the anxieties and difficulties caused by sharing daily life spaces with Mafia members' families, as the quotes of three participants below illustrate.

> *I wish to talk about my everyday life in Corleone. As well as being an employee, I also work here as a municipal councilor. My father is a retired police officer and, even though my parents are not from Corleone, I was born and grown up here and I hope to continue to live in Corleone (...) Since I have been living in this town, I know who belongs to the Mafia and who doesn't and I also know how to behave, even though it's not always easy... (...) Every morning I go to a café to have breakfast; sometimes it can happen that some people connected to Mafia families enter that café so I have to choose, in a heartbeat, if I have to say hello to them or not. Sometimes I realize that some policemen are coming in at the same time. I feel cornered, at a turning point, and wonder: What if I don't say hello, close my eyes, play dumb and go away? Should I say hello even if I know that the policeman could write in a report "Today the municipal councilor said hello to the dude at the café"? Almost every morning I usually find myself at this turning point.*
>
> *My fiancée is not from Corleone, so she doesn't know the houses where Mafia family members live. Sometimes she points her finger at a house and says: "Look how beautiful that house is!". Aware of the fact that Mafia members are living in that house, I beg her to lower her finger. (...) It's great to talk about the Mafia but it's really difficult to talk about the Mafia in Corleone.*
>
> *Let me tell you one of my experiences... When I was at school, one day a teacher told us: "Guys, let me inform you that in our class there's going to be a new student. I would highly recommend you have to behave properly and, starting today, we will not talk about issues relating to the Mafia". My new classmate was the daughter of a boss. My classmates and I were shocked, we were afraid. However, we started a more or less friendly relationship with her, despite the fact that we knew that we had to be careful what we could or could not say when we were talking to her.*

During the first group meeting, a lot of questions, doubts, and dilemmas emerged. Moreover, the quietest participants nodded while hearing other people's tales of their experiences. It seemed clear that all participants had found themselves at a challenger turning point at least once in their life: "Should or shouldn't I say hello to the boss's relative? Should I welcome him into my house? How can I conquer my fear of talking about the Mafia if, since I was a child, adults have always taught me not to speak about or oppose to the presence of the Mafia in my town?". Painful conflicts not clearly perceived before that time or not "known" (Hopper, 2001) linked to the experience of sharing spaces of everyday life with Mafia members' children and relatives started to emerge and became an important theme during this meeting. In this first meeting, the main conductor's tasks were to create a safe communicative space, a holding environment in which both cognitive and emotional unconscious or unknown feelings in relation to the Mafia could be listened to. Such a holding environment supported the emergent understanding of an important passage: from the narration of the Mafia as an external criminal organization far from citizens' lives to the narration of a present and close Mafia who observe, impose silence, and influence community members' public and private behaviors and lives. To summarize, the group process started to work for participants as a holding environment in which they could analyze, understand and become aware of the social unconscious processes they shared as a result of living in a close contact with the Mafia.

Second Group Meeting: Voicing Emotions and Feelings

The second group meeting was characterized by the possibility of sharing emotions mainly connected to past events of Corleone's history within the group. The holding environment built in the first meeting allowed participants to reveal, and later elaborate on, the painful emotions that they had experienced in relation to the Mafia. As Weinberg (2007) pointed out, the group process allows members to unconsciously re-live and re-enact in the here-and-now relationships emotions related to painful or traumatic past events. The role of the conductor was mainly oriented to facilitate reflective and constructive encounters with the past.

The word "fear" was often present in several interventions. For example, a mother talks about the fear for her children's safety that she had been feeling for many years:

> *I felt scared when my children, when they were kids, lingered on the street with friends. Sometimes they could have been talking with a boss's son... and I was afraid they could be involved in an ambush...even if you were not the target, you could be killed... It could have been fatal!".*

In another example, a young woman remembers her frustration during adolescence, presenting the following episode:

> "*Once, while I was joking with my friends, I pronounced the name of a boss... They glared at me and looked around scared to death... From that day on, I understood that I had to give up my spontaneity, the spontaneity of my language*"

During this second group meeting, some participants could also tell the group about memories connected to Mafia violence.

> *I remember that there was a local festival and many people around. Suddenly, we heard a woman yelling "Bastards, bastards". My sister and I got closer to the place these voices were coming from. A woman was shouting "They killed him". We stopped in front to the place of the homicide, I saw a white car—I'll never forget—and as soon as my sister understood, she immediately pushed me in such a way that I couldn't see anything!*
>
> *... Years ago there was a murder and my father, who was working at the Town Hall (...), was called very early in the morning because he was asked to clean the roads. When he came back, he told us about this river of blood which was flowing down... I was still too young to understand these things, but I could see my mother's fear every time my father had to go and pick some panjandrums at the airport or take the mayor somewhere... I really remember this fear...*

These narratives opened a space of resonance and mirroring (Foulkes, 1948) in the communication process of the group thus activating a growing emotional flow. Participants could share emotions and anxieties connected to the Mafia and recognize the relevant psychological and social effects that organized crime had caused in local people.

Almost at the end of the second meeting, a young woman's intervention moved the group members, who could finally recognize the suffering the Mafia produces in citizens and the community.

> *For me, the Mafia is pain, an immense dark pain. The pain I feel is not abstract... (...) one of my schoolmates suddenly disappeared: he was first killed and then chewed up by dogs. Then they put him in a jeep and set it on fire. (...) From that moment onwards I escaped and stayed away from Corleone for many years; I didn't want to live there any more... (...)*
> *Mafia is pain, a strong pain which is also present in Mafia relatives. (...) Sometimes, when I speak with these people, I realize the immense dark pain they feel inside; they'd like to talk to you, but cannot; they do not reveal much, but they have passed down a huge pain.*

During the second group meeting, the holding environment enabled participants to recognize their shared painful emotions linked to the violence of Mafia, which they had had to keep silent for a long time. In a recent review on the impact of historical trauma, Mohatt et al. (2014) proposed a perspective that interprets trauma as a psychological process independent from the specific traumatic event, as a narrative representation of the past that contains both personal and collective components and that continuously affect present-day representations of people. Through the possibility of sharing public narratives about violent and traumatic social unconscious dimensions participants could recognize the psychological suffering the Mafia had produced over the years, as well as the pain rising from the awareness of feeling oppressed by a hidden power that was, at the same time, clearly visible in many inhabitants' faces and stories.

Finally, it is important to note that at least two elements contributed to promote the willingness to explore these social unconscious dimensions in the group. First, the capacity of the conductor to encourage and contain the emotions that surfaced. Second, the emotional group dynamics are also influenced by the affective dimension of group identities and by people's specific interest in a particular situation (Hoggett and Thompson, 2002). As a consequence, the affective and current factual dimension of the Mafia in the group culture of Corleone's citizens made them passionate and willing enough to express their feelings in a safe communicative space.

Third Group Meeting: From Emotions to Awareness

Sharing unconscious emotions and aspects connected to the Mafia allowed participants to gradually become aware of the power that the Mafia has exerted over them and to reflect on personal and collective initiatives to cope with the threat of the Mafia in everyday life. Two participants stated:

> *I paradoxically came here to meet the Mafia through your stories (...). After these group meetings, I realized I had seen, met the Mafia, but I didn't think that the Mafia also meant this... I met it, let's say, in many small things, in many little moments of my life (...) I realized, for example, that the family who was living next door when I was on holiday was a Mafia family.*
>
> *Once, I was in the garage with my boyfriend (...) he comes from a different town (...) he is more free from such concerns (...) a child, son of the Boss living in my apartment building, deliberately stepped on my boyfriend's foot with his bicycle. My boyfriend gave him a slap and I've been scared for a week (...). I realize now how much I have been conditioned.*

The quotes mentioned above clearly show how the frequent contact with Mafia's member or relatives in everyday life caused anxieties and pains against which participants had built strong defenses in order to protect themselves. As highlighted by Weinberg: "Uncovering these traumatic memories or the way they unconsciously impact on a society is the essence of the Social Unconscious." (2007, p. 316). Mutual resonance and mirroring facilitated in the group the emergence of analogous narratives increasing participants' awareness growth.

Subsequently, a participant shared with the group a narrative about his parents' ways of coping with the perceived threat exerted by the Mafia that provides an example of the reenactment of the memory of a suffered humiliation:

> *My father is a customs officer and his station has recently been transferred near to Totò Riina's house (...) Once, he told me he had to leave his station with his service car but he found another car closing the exit (...) he began to honk and, after a really long time, Riina's wife and their daughter (...) she moved her car apologizing*

> *(...) my father is a big man and he was driving his service car (...) Do you think that he was able to affirm the government power? (...) but would he be able to do it as an ordinary man?*

During the third group meeting, participants could also recognize that the Mafia strongly limits their personal freedom. For example, noting this negative influence, two participants said:

> *I live in a special apartment building where Mafia families also live. Every day I have to deal with people who literally limit my freedom!*

> *I lived in Palermo to attend the university (...) I come from a town in which the presence of the Mafia is relevant and nothing shocking has ever happened to me. However, I've reached a deeper understanding of many events of my life in Sicily by participating in this group (...). My father used to bring me to Palermo with his car and to park it in a small street near my house (...). He always found a flat tire or a rear-view mirror (...) It did not happen by accident, we could not park the car in that street (...) A girl living in my building also found her car to be burned twice.*

The above reported quotes represent a shared anxiety among Corleone's citizens, and they mirror the original motivation for the intervention request. The return of the Boss' son to Corleone seems to have created an emotional imbalance in its citizens. His homecoming compelled them to face the painful experience of living once more with the frustration of being silent, obeying the Mafia codes, and being limited in one's own freedom of expression and action.

At the end of the third group meeting, the conductor pointed out some aspects which highlighted the possibility of giving a shared meaning to social unconscious emotions felt outside and inside the group.

> *One might think that the idea that the son of the boss came back and we do not want his presence here is, like,... an ideological or political opinions... In contrast, in the course of these meetings, you have spoken of a strong emotional imbalance. As though Corleone had suddenly lost a bit of serenity and integrity laboriously acquired before. The story told by one of the participants can be considered a strong metaphor of this process. Just like Maria's father, Corleone had to clean its roads from the blood that the Mafia had spilled! Therefore, the idea that the boss's son is returning causes an imbalance, makes you feel the smell of blood once more. In my opinion, during these group meeting, a mostly emotional process took place which had its peak, when you spoke about your pain, the deep pain that Corleone has been experiencing and that strongly touched us, mostly because it's as if you had spoken about the most brutal aspect of the Mafia, that is its inhumanity, the annihilation of freedom which had corroded the community's social networks.*

During this last meeting, the experience of a safe communicative space allowed the expression of the unconscious and unthinkable about the Mafia and made it possible for participants to contemplate new thoughts and hope for a less problematic future. Below, a participant's intervention explains his perspective on the group meeting experience:

> *I had the feeling that we did something which in Corleone had never been carried out (...) because each one in the group has given evidence of his or her experience or anyway told his or her personal opinion and had the chance to confront with other citizens about the Mafia. (...) For those who were present, this has been something very strong which has left a seed. I'm sure that this work is really useful, way more than any face-to-face meeting, demonstration, book presentation, because I had the feeling that we were willingly putting ourselves in the first person and this allowed us to make a step up. In my opinion, this has been important for all participants and for our community.*

This last quote highlights how the enactment of social unconscious aspects through public narratives of significant shared painful experiences could sustain resilient responses. As suggested by Mohatt et al. (2014), historical trauma narratives could inhibit psychological growth and collective future-oriented aspiration or sustain lasting community transformation and resilience. One of the most important challenges that clinical psychology has to deal with operating in socio-cultural contexts is to investigate and identify the conditions that could sustain the change and the reconstruction of the social traumatic and conflictual wounds in the community.

CONCLUSIONS

As Geyer (2017) suggested, the group-analytical oriented group provides a window into the social unconscious and the unconscious power relations that sustain it, showing to be a very appropriate tool for research issues analyzing social change processes. In this paper, we sought to broadly report the most salient passages of a group-analytic intervention highlighting how the enactment of social unconscious dimensions allowed participants to become aware of the negative influences exerted on them and on the community of Corleone by the presence of the Mafia. This experience provided to participants a greater ability to manage the negative impact caused by the Mafia in their lives and this could promote a future-oriented capacity to oppose the criminal organization. Indeed, during the group meetings, participants could experience a holding environment in which they could become aware of their social unconscious anxieties and defenses and find new ways for re-thinking and interpreting their cognitive and emotional experiences, as well as discussing further collective actions to oppose the oppression of the Mafia. Although for the first time, this intervention gave participants the chance to be aware of the ways in which the Mafia oppresses their lives, one important limitation needs to be acknowledged. Due to the lack of financial resources, this intervention was time-limited and involved a small number of citizens while a more effective action would have required a more extensive participation and a long-term intervention. Nevertheless, we think that this intervention could represent a useful model to analyze the mutual influences between social phenomena and processes and subjective experience and to inspire future psychological intervention to promote lasting changes in the fight against the Mafia. Because no single approach by itself can fully be effective to promote social change against a complex phenomenon such as the Mafia, this article sought to highlight that psychological processes can have damaging impacts on the capacity of individual and collectivity to pursuit transformation and resilience. This evidence also provides important insight on how clinical psychology may operate in socio-cultural contexts to promote the reconstruction of the traumatic social dimensions in the community.

ETHICS STATEMENT

This study was carried out in accordance with the recommendations of the Ethical Code of the University of Palermo and of the Code of Ethics approved by the General Assembly of the Italian Association of Psychology held on March 27, 2015. All subjects gave written informed consent in accordance with the Declaration of Helsinki. The real names of participants have been replaced with pseudonyms to ensure confidentiality and anonymity.

AUTHOR CONTRIBUTIONS

CG, GC, CT, LP, and MD had substantial contribution to the conception of this work. CG and MD designed the study. GC, CT, and LP collected and analyzed the data and all authors had substantial contribution to the interpretation of the data. CG and MD drafted a previous version of this article and all authors critically revised it for important intellectual input and finally approved of the version to be published. All authors 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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METHODS
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Taking the Concept of Citizenship in Mental Health across Countries. Reflections on Transferring Principles and Practice to Different Sociocultural Contexts

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Transferring principles and practices to different sociocultural and professional contexts in the field of mental health can be very complex. Previous research on public health policy points to difficulties in different areas such as the understanding the new concepts, their applicability in different health systems, and suitable approaches to its effective implementation. The purpose of this article is to describe and analyze the process of transferring the concept of Citizenship, from its United States origins in mental health outreach work with persons who are homeless to Catalonia, Spain. We define Citizenship as promoting the rights, responsibilities, roles, resources and relationships of persons with mental illnesses, along with a sense of belonging that is validated by other citizens. The process of this transition involves embedding Citizenship in the mental health "first-person" (internationally known as Consumer/Survivor/Peer) movement in Catalonia. The paper includes a discussion of the concept of transference, including a case example of the adoption of the concept of mental health recovery in different countries. Following this, we describe the United States Citizenship model and key elements of its development. We then turn to Spain and the evolution of its mental health system, and then to Catalonia for a brief case history of transference of the principles and practices of Citizenship to that region. The "take home message" of this work is that concepts being brought from one sociocultural and national context to another, must focus on contextualization in the 'adoptee's' practices, including the balance between personal involvement and professional rigor, the involvement of key actors, and ongoing evaluation of actions taken.

Keywords: transference, citizenship, public mental health, user involvement, recovery, mental health reform, Catalonia

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INTRODUCTION: PRINCIPLES AND PRACTICE TRANSFER BETWEEN CONTEXTS AND REGIONS

Transferring innovative principles and practices to different sociocultural contexts has often been analyzed from the policy transfer and mobility perspectives, i.e., the scientific study of the implementation of public policies in places not of their origin (Stone, 2012; Cook, 2015). Similar methods have been employed to study the transfer of health systems strengthening strategies

(El-Jardali et al., 2014), general (Simms, 2010) and mental health reform toward patient-centered services (Hickie et al., 2014). Previous research points to difficulties during the process of transference in areas such as the understanding of novel concepts, their applicability in a different environment, and the most suitable strategies and approaches to their effective implementation (Tervonen-Gonçalves and Lehto, 2004; Simms, 2010). In addition, resistance to and lack of motivation for change often confronts systems reformers who introduce novel concepts in different settings (Saraceno et al., 2007; Makhashvili and van Voren, 2013).

Less discussed in the literature and practice of adapting innovations to different sociocultural settings is the distinction between 'top down' administrative-directed change (Tervonen-Gonçalves and Lehto, 2004) and 'bottom up' or street-level change (Rowe et al., 1998) seen in cross-national transformation (Ovuga et al., 2007). When we talk about bottom-up approaches, we talk of processes in which users or practitioners have initiated actions to try to promote change. When we talk about top-down approaches, we refer to actions in which policymakers or health managers have initiated change. These approaches include differences in the motivation for change that the diverse actors have, and in the ways and directions in which change occurs. Processes by which policymakers and managers decide to borrow health concepts, practices, and policies of another, usually more developed, country have little to do with processes by which citizen and professional mobilizations occur. Top-down changes are guided by the interests of policy-makers and managers, which do not always coincide with collective aspirations, while bottom-up transformations must be promoted and accepted by service users and practitioners. For example, the profound influence of Paulo Freire's *Pedagogy of the Oppressed* on the internationalization of the empowerment concept, can be understood from the critical insights that professionals borrowed for their own work. Particularly, that inequalities in access to health and education services had their origin in social inequalities and could be changed through collective action (Campbell, 2014). On the other hand, psychiatric deinstitutionalization can be understood as a top-down transformation. While stakeholders' protests may have brought the need into public view, the 'heavy lifted' was done by administrators whose distance from the issues in play may partly explain some of its failures, including the increase of the homeless population with mental disorders (Lamb, 1984) and the failures of its implementation in non-Western Contexts (Shek and Pietilä, 2016).

Another case example relevant to our theme is the implementation and transference of the Recovery model, essentially adopted by the Clinton administration in the United States in 1999 with the Surgeon General's Report on Mental Health (Davidson et al., 2006). The recovery model emphasizes the participation of consumers and their families in service planning and the promotion of recovery not as a clinical fact or occurrence but as a process of change through which individuals improve their health and wellness, live self-directed lives, and strive to reach their full potential (Substance Abuse and Mental Health Services Administration, 2012). The introduction of this model in the United States has had a clear impact on the policy and practice of mental health care. Perhaps its signal accomplishment and characteristic is to advocate for and support the participation of service users as "peer specialists" or "peer mentors" for persons with mental health disorders, and for their participation in systems reform (Davidson, 2016).

The transfer of the recovery concept and its derived practices soon spread to the rest of the Anglo-Saxon world, especially the United Kingdom, New Zealand, Australia, and Canada, as well as other developed countries (Ramon et al., 2007). Although to our knowledge there is no specific literature that analyzes the strategy of transfer of the movement toward Recovery as a public policy, we know that the consumer movement was strongly involved in protesting current practice and advocacy for change, along with academic, researchers, policy makers, and practitioners. The advocacy activities carried out by these groups and their ability to connect internationally contributed mightily to its dissemination and adoption around the world. Small-scale implementation strategies have included "top-down" activities such as training and organizational change activities for professionals (Mabe et al., 2016), sometimes provided by people with lived experience of mental disorder (e.g., Simpson, 2002), and bottom-up participatory processes such as involving service users in the design of Recovery-in-Action initiatives (Park et al., 2014).

Although the international expansion of the Recovery movement has been instrumental in deepening the processes of extension of community rehabilitation practices that, in retrospect, could have enhanced the process and outcome of psychiatric deinstitutionalization, the "colonization" of its vocabulary by policymakers has been criticized, even leading to movements such as *Recovery in the Bin*. Service users in the recovery movement, and others, have accused policy makers of harnessing the values of empowerment and mutual support that are part of the recovery movement and message to cut services and blame people for their mental ailments (Thomas, 2016). For these reasons, critical analyses aiming to support the autonomy, empowerment, and full citizenship of people with mental health problems remain a topic of discussion.

The purpose of this article is to describe and analyze the preliminary process of transferring the concept of Citizenship, a novel approach to the social inclusion and community membership of people with mental health problems, from the United States to other countries. We employ a case example of a Citizenship project developed in Catalonia, Spain as what we hope we will also serve as an instructive example of the challenges and possibilities for dissemination of the Citizenship model internationally.

CITIZENSHIP: A NOVEL APPROACH TO MENTAL HEALTH

Citizenship, involves the strength of people's connections to the rights, responsibilities, roles, resources and relationships that society offers to people through public and social institutions (Rowe et al., 2001), and a sense of belonging as full, participating members in society that is validated by one's fellow citizens (Rowe, 2015). The strengths and limitations of outreach work, including the finding that helping people find housing does not, in itself, lead to their community integration or full membership, led a group of scholars and practitioners based at Yale to develop the theoretical framework of Citizenship (Rowe and Pelletier, 2012). Although the Citizenship framework was developed during the period of the Recovery movement (Slade et al., 2014), and analyses show that higher levels of Recovery are usually found in persons who are better able to exercise their rights as full citizens (Pelletier et al., 2015), the two approaches evolved independently.

The first Citizenship intervention was the Citizens Project, implemented at the Yale Program for Recovery and Community Health. The concept of Citizenship (Rowe et al., 2001) was used as a framework (Rowe et al., 2009) for opening up opportunities for social participation to members of stigmatized groups. In this program, rather than viewing individuals with mental illness as problems to be addressed through the intervention of others, participants are "students" and "citizens." Hence, they are viewed as experts on many of their own problems and difficulties, on identifying solutions to them, and who are capable of learning not only how to maintain themselves stably in their communities but to see themselves as and take actions to become valued members of their communities. Citizens Project participants are persons with mental illnesses including, for many, the dual disorder of substance misuse, and previous criminal charges. A randomized clinical trial comparing the Citizenship intervention to usual care for the target group showed that it successfully reduced alcohol and other substance use, and increased quality of life for participants (Clayton et al., 2013). Following this study, participatory action research methods including peers (service users) as researchers were employed to develop an individual measure of Citizenship (Rowe et al., 2012) with seven dimensions of Citizenship—Personal Responsibilities, Government and Infrastructure, Caring for Others in Community, Civil Rights, Legal Rights, Choices and Stewardship. This measure has also been validated for use with persons with mental illness receiving public care (O'Connell et al., 2017).

The use of Citizenship as a psychosocial intervention rooted in collaborative work with and among people affected by mental health problems is related to other similar conceptualizations. A meeting point between all these conceptualizations is that Citizenship should be thought as negotiated and enacted rather than given (Stevenson et al., 2015). For example, Barnes et al. (2004) link Citizenship with the dynamics of membership and its legitimacy. They show how the entitlements associated with the category citizen are embedded in the dynamics of inclusion and exclusion of rights. Continuing with the idea of Citizenship as the legitimation of rights entitlement, and adding a transformative dimension, Renedo and Marston (2015) developed the concept of participatory Citizenship in the context of patient and public involvement in the healthcare system. They propose a dynamic view of Citizenship comprising the participation of different actors in negotiating and acting on their rights and responsibilities as health service users and drivers of change.

Similarly to our conceptualization of Citizenship, the idea of agency as opposed to prescribed interventions has emerged as an argument against pathological approaches to trauma and distress (Veronese et al., 2015). Specifically, there are lines of research that may help us understand how the structure and protocols used by mental health institutions subvert the agency of social inclusion of their clients (Watson, 2012). In general, the conceptualization of agency on which our view of Citizenship is based is systemic, with individual actions understood within the broad contexts and power relations (Diewald, 2007; Stetsenko, 2007), rather than Bandura's (1977) conceptualization of individual beliefs of self-efficacy and Rotter's (1954) notion of locus of control.

Our Citizenship framework is now being adapted for use in other countries and sociocultural contexts. Current Citizenship research and practice is being undertaken in Quebec (Canada), Scotland (United Kingdom) and more recently, Catalonia (Spain). After contextualizing our work within the Spanish mental health system evolution, we will discuss some of the implications of transferring the concept of Citizenship in mental health to the latter territory.

THE SPANISH CASE OF MENTAL HEALTH SYSTEM EVOLUTION AND STAKEHOLDERS' INVOLVEMENT

Spain has had some differences with respect to other Western European countries in terms of the implementation of the last two major waves of mental health care reform, namely deinstitutionalization and recovery. Due to the duration of Franco's fascist dictatorship, the deinstitutionalization of the psychiatric system did not begin until the 1980s, when alternative resources to the great psychiatric hospitals and cooperation with primary care began to be developed (Vázquez-Barquero and García, 1999). The evolution of structural changes in the Spanish Public Mental Health system has been marked by the division of Spanish psychiatry into two major professional associations: The Spanish Society of Psychiatry (*Sociedad Española de Psiquiatría*, SEP, biologically oriented) and The Spanish Association of Neuropsychiatry (*Asociación Española de Neuropsiquiatría*, AEN, community and psychotherapy-oriented). While the latter has worked together with other professional bodies (e.g., the Spanish Federation of Psychosocial Rehabilitation, *Federación Española de Rehabilitación Psicosocial* in Spanish, FEARP) and consumer-led (relatives and more recently, "first-person" consumers') to support the implementation of community-based resources, the former has always had an ambiguous role marked by its conflicts of interest with the pharmaceutical industry (Escobar, 2009).

The arrival of the Recovery movement to Spain has been limited to community rehabilitation services and its extension to other care settings occurred only recently through specific and somewhat isolated projects. In 2005, for example, the European-wide EMILIA (acronym for *Empowerment of Mental Illness Service Users: Life Long Learning, Integration and Action*) project (Ramon et al., 2011) was one of the first to systematize joint training actions between professionals, users and their families. This project, together with community-oriented professional bodies such as the AEN and FEARP, served as an outpost of the Recovery movement in Spain. However, the arrival of the crisis in 2008 and the cuts in funding for public health that entailed led to a total withdrawal of funding for this type of projects, as the Spanish public mental health system retreated to "survival" funding for mental health care. Based on preliminary results of focus groups carried out as part of this Citizenship transfer project (see 'Methodological Implications' section), mental health professionals agree that in recent years they have barely been able to provide minimum services to users, who must endure long waiting lists for care. Many professionals say that this was not the time to implement "any kind of novelty," as they are simply overwhelmed. Over the same time period there has been an intense media debate on the influence of the pharmaceutical industry, as the primary care protocols for the treatment of the increased anxiety-depressive symptoms reported in these services, caused by the economic difficulties of the population (Gili et al., 2013), have mainly involved use of antidepressants and anxiolytics.

An exception to the shortage of mental health care resources has been the incorporation of individualized monitoring programs (Programas de Seguimiento Individualizados, PSIs) staffed by professionals who provide care in community environments. Although these practitioners are not fully trained in Recovery principles, many of them are familiar with its practices. Furthermore, elements derived from other orientations such as Community Assertive Treatment share some principles with those of Recovery. Yet, as mentioned above, the Recovery approach has had a very narrow scope for practice, being confined to community rehabilitation services. The implementation of Recovery principles in places which are more difficult to influence such as hospitalization services, is still almost non-existent in Spain.

An ominous event regarding the potential to implement the Citizenship framework in Spain was the attempt to introduce "special security measures" for people diagnosed with mental disorders in the Spanish Penal Code by the conservative government that took office in late 2011. This 2013 bill produced an immediate and massive reaction from mental health advocacy groups. The campaign against the bill revived old synergies among groups that had been relatively inactive for a few years, giving additional support to the first-person (consumers/survivors/peer) movement. In Spain, this movement is formed by associations that operate in diverse contexts, from activism and policy reform to leisure and art. The Spanish Federation of Relatives of Persons with Mental Illness changed its name to "Confederation Mental Health Spain,"[1] intending to integrate first-person organizations within the same structure. However, many organizations believed that the best approach was to construct differentiated spaces for families and those who have lived the experience themselves. A first-person federation already existed in Andalusia since 2009 and a new one was created in Catalonia in 2014. Less populated communities such as Asturias, Balearic Islands, Canary Islands, Madrid, Navarra, or Valencia have active associations. A negotiation to create a Spanish-wide federation is in process.

Andalusia and Catalonia are territories especially active in the progressive approaches to mental health care and supports because of (1) having federations that exercise the institutional representation of people with lived experience of mental disorders ("In first person,"[2] *En primera persona* in Spanish, and "Voices," *Veus*[3] in Catalan, respectively), independently of relative's and users' associations' federations (Andalusian Federation of Relatives and People with Mental Illness[4], and Mental Health Catalonia[5]), and 2) having territory-specific anti-stigma campaigns. The executive boards of these campaigns are formed by relatives' and users' first-person federations, and by representatives of public and private mental health service providers and local governments. The Andalusian campaign is called *1 de cada 4*[6] ("one in four," referring to the fact that 25% of the world's population will suffer from a mental disorder during their lifetimes) and the Catalan, "Obertament"[7] ("openly"). In addition to its activities to prevent stigma, the "1 de cada 4" campaign has translated materials and openly supports the implementation of the Recovery paradigm in the Andalusian public mental health care system. Obertament, for its part, trains first-person activists to carry most of their activities, and is designing a campaign to educate and influence health care professionals. In the case of Catalonia, as a result of the collaboration of both federations, a project has recently been launched (*Activa't per la salut mental*,[8] literally "get involved in mental health") which aims to promote the recovery model through information, psychoeducation, empowerment and mutual support spaces.

Having reappeared under the threat of hardening of the penal code for persons diagnosed with a mental health disorder, the "first-person" movement made the struggle for rights a core objective of its work. At the same time, the institutional anti-stigma "1decada4" and Obertament campaigns, and the relatives' movement also supported prioritizing defense of the rights of people diagnosed with mental health problems.

FROM BIG CONCEPTS TO DAILY WORK WITHIN THE CATALAN MENTAL HEALTH NETWORK

Figure 1 shows the structure of the Catalan mental health network. As can be seen at the top of the figure, there are three main political representation groups. The local administration promotes executive (centered in the health system) and comprehensive (coordinated with other departments such as work or justice) mental health plans. Additionally, the

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[1] http://consaludmental.org/

[2] http://enprimerapersona.org/

[3] http://veus.cat/

[4] http://feafesandalucia.org/

[5] http://www.salutmental.org/

[6] http://www.1decada4.es/

[7] http://obertament.org/

[8] http://activatperlasalutmental.org/

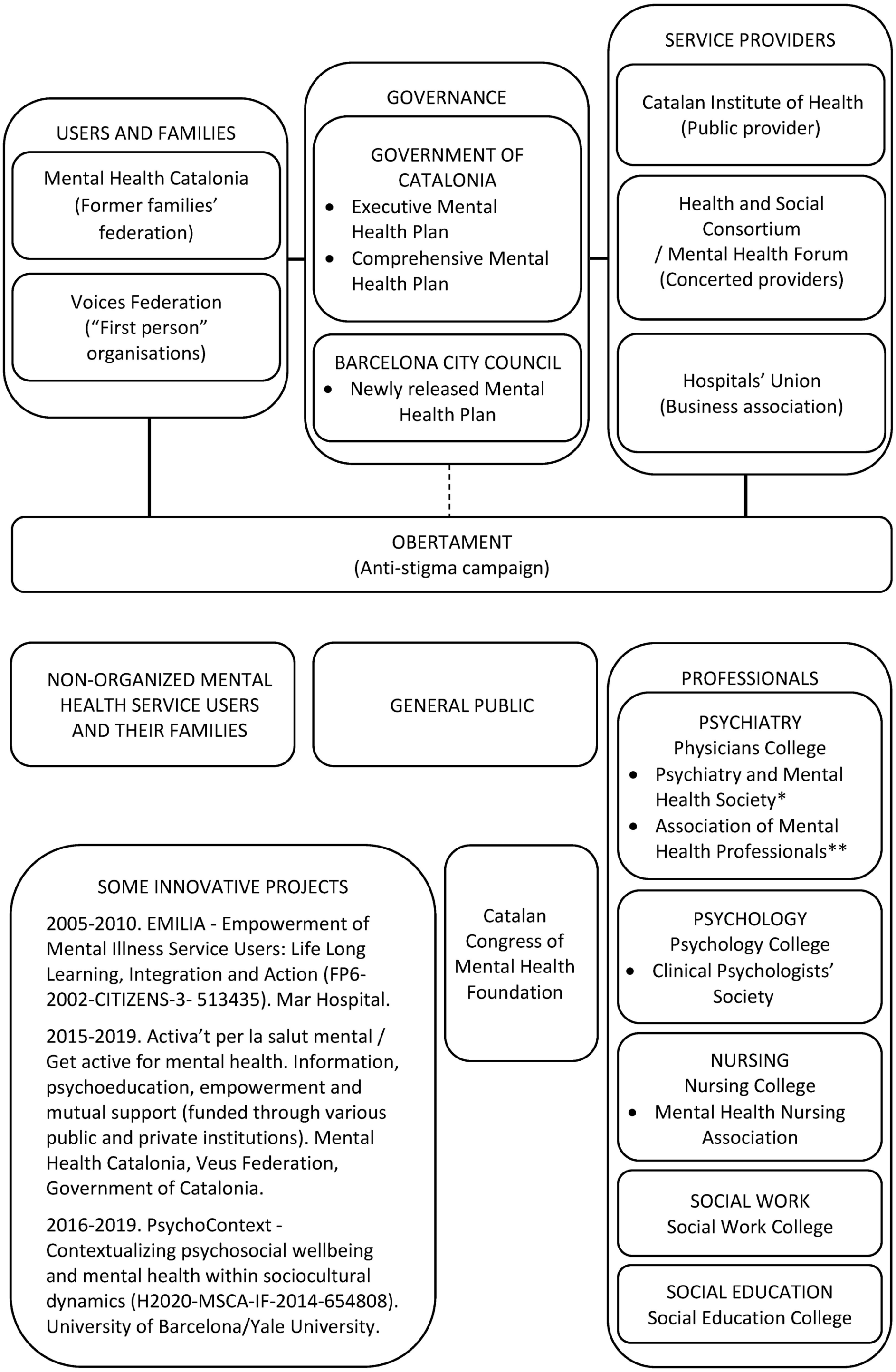

FIGURE 1 | Principal Mental Health Stakeholders in Catalonia. *The Catalan Psychiatry and Mental Health Society is related to the Spanish Society of Psychiatry, of neurobiological orientation, although it is not an organic part of it. **The Association of Mental Health Professionals is the local branch of the Spanish Association of Neuropsychiatry, with psychotherapeutic and community orientation, and admits the participation of psychologists, nurses, social workers, educators and other professionals. It has recently begun to welcome the debate on the affiliation of service users.

city of Barcelona has launched its first local mental health plan. In collaboration with these administrations, users' and relatives' representatives collaborate with service providers in the governance of the *Obertament* campaign against stigma.

At the bottom of the figure we see the three groups receiving messages from the Obertament campaign, and the remainder of activities involving the implementation and improvement of mental health services and societal wellbeing provided by users' and relatives' federations, local administration and service providers. Their messages are obviously not always univocal. While unorganized service users, families and society in general receive these messages in passively, it is important to note that mental health professionals have complex organizational structures independent of the service providers' representation structures. In addition to the professional colleges of all direct care professions, there are specific professional associations in the cases of psychiatry, clinical psychology and mental health nursing. In Catalonia, there are two associations of psychiatry with different philosophical orientations regarding mental health intervention. The Catalan Association of Mental Health Professionals, as the local branch of the Spanish Association of Neuropsychiatry. The latter accepts the participation of psychologists, nurses, social workers, educators and other direct care professionals, as well as service users and their relatives. The Catalan Society of Psychiatry and Mental Health, without being a local headquarters of the Spanish Society of Psychiatry, shares some of its scientific values, framed in a biomedical vision of mental disorders. In addition, the Catalan Congress of Mental Health Foundation, which emerged from the organization of an annual congress, brings together different professionals with a focus on human rights.

In this context, and inspired by the model of Citizenship originated in New Haven, the first author of this article began his journey as an agent of change of the Catalan Mental Health system in mid-2015. In the first place, his positioning regarding participation in participative spaces both for professionals and service users was considered important. As an academic and certified psychotherapist, he contacted some of the aforementioned professional bodies, which showed some interest and invited him to give lectures to explain the project in public, with the understanding that more concrete activities must follow. Examples of these are training and educational activities, the design of which will be developed at the same time as the project itself. As a former mental health service user, the first author initiated activist training for the *Obertament* campaign in Barcelona. The involvement of the "first-person" associations in this campaign facilitated his contact with the self-managed movement, where he was recognized as a first-person participant like others, a fact that facilitated the process. In addition, his recognition and inclusion as a mental health academic provided a source of additional human capital, and he soon became involved in tasks where his academic skills were useful.

As we have said before, the struggle for the rights of people diagnosed with a mental disorder is a key objective of the first-person mental health movement. Thus, a key strategic action for incorporating Citizenship into the spectrum of mental health in Catalonia was participation in a campaign to reduce or eliminate mechanical restraints in mental health units.

The other two strategic actions that have been considered appropriate to foster within the present project and that match the objectives of other users', families,' and professionals' groups, are training and hiring peer support staff and carrying out awareness activities and training for professionals. In this regard, some of the associations within the Veus federation already provide training to moderators of mutual support groups and perform different awareness-raising activities for mental health professionals, including a yearly lecture series for all mental health trainees in Catalonia. It is understood that these activities must be enhanced to make them accessible to all professionals, regardless of their or their institutions,' philosophical and practice orientation. In a context of partial implementation of the recovery approach, the incorporation of peer-support staff only makes sense if mental health professionals are motivated to accept staff with lived experience of a mental disorder. As such, we hope to learn not only from the successes but also the challenges of countries such as the United States that has been implementing recovery-oriented policies for the last two decades (Davidson, 2016). Finally, people offering peer support need to be able to conduct other activities, such as helping people navigate the mental health system including psychiatric hospitals.

Methodological Implications

Our project to transfer practices and approaches based on Citizenship is built on strong personal and professional involvement and a strategy with concrete objectives. Hence, although it has the strength of close collaboration and personal involvement within the main stakeholders involved, it is critical that we also maintain scientific rigor in evaluating all interventions carried out through this initiative.

In general terms, starting from the theoretical elaboration of the sociocultural dynamics of well-being and distress, our project aims to promote Citizenship practices through continuous training of professionals and users of mental health services and their relatives. This initiative will benefit from the experience of the Yale program, but must also attend to the characteristics and priorities of local stakeholders. **Figure 2** shows a representation of a proposed cycle of development of our transference project within a complex mental health system. The three columns represent sets of tasks that are repeated cyclically: preparation, involvement, and evaluation of the results. The three states of activity boxes reflect current and ongoing work.

In the first stage, international models and allies were sought. In this phase, the first author met the second, a senior academic director of the Yale Citizens Project. Once funding and ethical approval were obtained (University of Barcelona's ethics approval reference IRB3099), focus groups were held with local stakeholders. Fifteen focus groups including 70 mental health professionals, 40 service users and 10 relatives were conducted. Our choice of strategic actions was supported by the preliminary results of these focus groups obtained through thematic analysis (Braun and Clarke, 2006). These results show how speaking about Citizenship and its most intuitive components, rights and responsibilities, implies a change that can be facilitated by user participation in the design and implementation of interventions. **Table 1** shows selected quotes from interviewees supporting the need for self-determination, one of the main themes arising from the analysis.

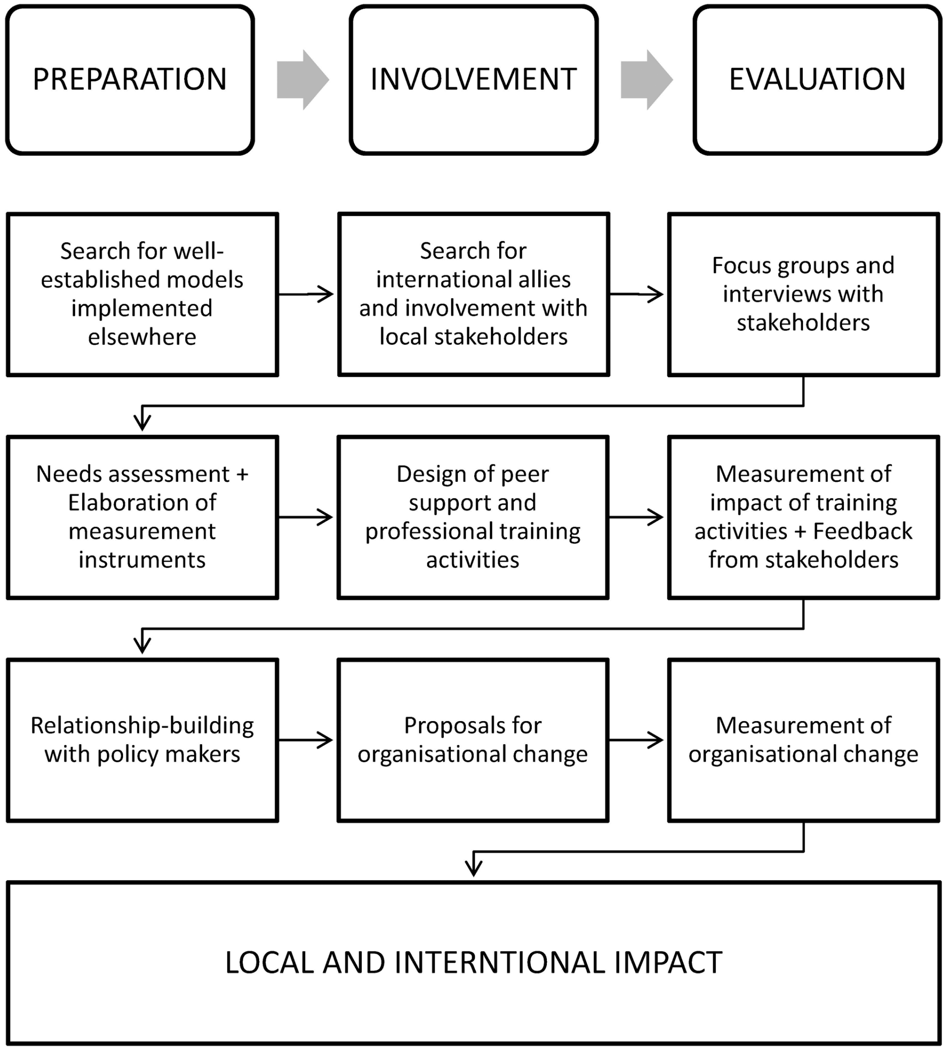

FIGURE 2 | Proposed cycle of development of a transference project within a mental health system.

Once this exploratory phase was completed, needs assessments were carried out and valid measurement instruments were designed to evaluate activities of mutual support and training of professionals. The proper evaluation of these activities has been a source of arduous discussions. The use of controlled experimental designs entails the exclusion of some users and professionals from training activities, due to their disagreement with the fact of starting a process in which they may end up in a control group. Therefore, we have decided to evaluate all our activities, where possible, through wait-list-controlled randomized experimental designs. Thus, all participants complete a baseline evaluation. Only half of these (selected randomly) have immediate access to the training activity. Nevertheless, once the first follow-up has been carried (approximately 1 month after the activity is completed) the second half can be involved in the activity.

Wait-list-controlled randomized experimental designs allow for two very important flexibility features. The first is the possibility of randomizing individuals or blocks, as in some cases activities must be offered to whole clinical teams or service user groups, but in others, individual participation is more appropriate. The second is that it is not necessary to wait until all the participants have been included to carry out the randomization. Rather, blocks of participants based on natural groupings may be included until the capacity of two training activities is completed. Thus, an activity will take place immediately after enrollment and randomization and the same activity will take place a month later, for participants who were randomized "out" at baseline. This also allows for evaluation of possible biased expectations on the part of different participant groups since, once the activity has been carried out, the changes in the evaluation measures should be identical both in the first group that carried out the activity and among the members of the waiting list group.

The final project phase includes an intense activity to interact with a diverse range of health managers and policy makers with the intention of proposing profound organizational changes.

TABLE 1 | Quotations related to self-determination themes.

Service users	*"For me being a citizen means being able to express myself freely and without any fear: To be able to use my principles and my ideas and express my needs, feelings… ".*
	"In my process one of the most important moments, the one I remember with most affection, it is when a friend, who had never been diagnosed, told me that he had also had a bad moment and told me how he bounced forward doing things with his friends, keeping himself alive. Feeling that someone understands you as an equal."
Relatives of service users	*"So, for you to be able to make decisions about your own life you should be totally autonomous, right? Because making decisions about your own life when you depend on another person is a bit relative."*
	"So, I think that the tendency now is to try to make the person who has the disorder have their own autonomy, their own life, dispose, make, and have their own rights."
Mental health professionals	*"I believe that the human right that is least protected is the right to decide. I think the professionals still decide for them: in an office, in the white coat, we inform the decision ... But we do not have direct contact with the person."*
	"Talking about a decision, about their life, they ask you: "How do you see it?" Or "I'll talk to my mother to see what she thinks" or "I'll talk to my psychiatrist to see what she thinks." "No, no, but ... What do you want?" I think we have to work this issue a lot, from empowerment, right?"

This phase, however, only makes sense if at the same time, we are offering stakeholders' feedback, in the form of scientific evaluation of their activities, and carrying out training and awareness activities.

Engagement and Implications of Strategic Activities

All the tasks described above involve the professional and personal involvement of many people, beyond professionals and volunteers involved in these changes. They also involve the resolution of complex conflicts and the careful choice of allies. In our case, as university faculty, doing such a job involves maintaining the balance between the requirements of an academic institution and the objectives of the first-person movement. The former is focused on finding meaningful results from rigorous evaluation and publication in prestigious peer-reviewed journals. The latter is focused on physical presence, personal relationships, and constancy in the struggle, a militancy that must go beyond standard professional commitment, as the potential interpersonal conflicts that can occur in the context of these struggles are often related to intimate and sometimes traumatic experiences.

Despite the prioritization of implementation projects including training and awareness activities facilitated by funding that had been absent during the 1st years of crisis, the need to build bridges with local policy makers is a key activity. Of the three projects mentioned (mechanical restraint reduction, peer support and training/awareness activities for professionals), the reduction of mechanical restraints requires the greatest amount of collaboration with, and support from policy makers, including meetings and spaces where all voices, including those of both service users and professionals, will be heard. Thus, following a series of meetings with the director of the Executive Plan of Mental Health and Addictions in 2016, this project has been finally articulated through the Service of the Promotion of Patient Safety of the Government of Catalonia's Department of Health, which also includes the reduction of mechanical restraints in minors and older adults in different restrictive contexts. Recently, the Veus Federation of mental health entities in first person appeared before the Health Commission of the Parliament of Catalonia (2017) to explain this project, which aims to eliminate mechanical restraints in all mental health care facilities in Catalonia before 2025. The first author of this article elaborated a collection of arguments in favor of the elimination of these coercive practices that was delivered to the deputies of the commission (Eiroa-Orosa, 2017).

The second strategic action that would benefit from institutional support is peer support training and implementation, involving funding to make this work possible and the institutional commitment this funding would represent for subsequent recruitment of trainees. A plan for peer support incorporation in the mental health workforce is included in the Executive Mental Health Plan Working Groups for the period 2017–2020. However, given the experience of the EMILIA project, which has advocated to establish peer support training for a decade, it has been considered important to develop actions without explicit institutional support. As such, workshops have been developed with the collaboration of relatives and professionals for the training of "mutual support technicians," in collaboration with the Mar University Hospital. Furthermore, the Veus federation, of which EMILIA is part, supports the development of self-managed spaces for peer support training.

Regarding the third strategic action—training and awareness-raising activities—practitioner associations in charge of training residents in psychiatry, forensic medicine, clinical psychology and nursing (see **Figure 1**), have included these activities in response to advocacy by the federations of Catalan relatives' and service user associations. Additionally, funding has been achieved by the Veus federation and its associations to offer a training activity called: *"Beyond Recovery: toward a rights-based mental health care."* This activity will be offered to mental health professionals regardless of their level of experience, orientation or place of work.

While this prioritization of activities has been undertaken with consideration of the interests of key stakeholders, the adequacy of the Citizenship framework has become evident. In all these strategic actions, the importance of the main components of citizenship—rights, responsibilities, roles, resources and relationships—is incorporated to show that full recovery is not possible if people are not considered full citizens with the right to choose their own way. The aim is to enhance the rights of people with mental health disorders through increasing the responsibility they have over their lives, allowing them to take on meaningful roles and helping them gain access to resources that will enable them to be full and valued members of their communities and society in conditions of equality.

Although the differences between the systems of support for people with mental health problems between the United States and European countries with a public health system go beyond the scope of this article, we consider it fundamental to discuss the implications that these differences may have on a process of conceptual and practice transfer. The existence of a purely public and universal system of health care in a country such as Spain ensures that all residents in the territory receive adequate attention, with a life expectancy of 83 compared to 79 in the United States (Kontis et al., 2017). This said, the possibilities of innovation in a highly bureaucratized system in serious financial crisis, as is the case in Spain, are lower. This applies, for example, to arduous efforts to hire mutual support (peer) staff, with innovation filtered through many layers of bureaucratic prioritizing and decision making. In addition, the forums in which such decisions are made, depend on the ideological orientation of political power and the ability of policymakers to release budget allocations.

One of the characteristics that facilitated the emergence of the movement toward recovery in the United States was the increase in funding made available from the closing of state hospitals, though by no means the automatic transfer of it to community services. Even so, mental health activists and reform-minded mental health professionals had the opportunity to lobby for the use of these resources for community mental health care. In Spain, reformist professionals carried out the process of deinstitutionalization with the approval of the health authorities.

Furthermore, in the 1990s the Spanish relatives' movement began to manage a part of the psychosocial rehabilitation services. However, the first-person movement so far has not reached a level of management capacity comparable to that of the Anglo-Saxon countries. For this reason, and probably also because of the prejudices about the capacity of people with mental health problems, their voices have largely been absent when service planning decisions are made. The project of transferring models such as citizenship, we argue, will only take root if the efforts and results of our evaluations are made available for debate in public spaces with full participation and empowerment of all interest groups, including those with lived experience of mental illness.

TAKE-HOME MESSAGE

The beginning of the transference of a concept such as Citizenship from Connecticut in the United States to Catalonia in Spain, involves the work of academics and activists but also that of many others who have long advocated for the rights of people diagnosed with mental disorders. The creation of different synergies among first-person, relatives' and professional organizations has been important in planning strategic actions for a successful transfer process. We think that a former service user being one of the main advocates for and actors in this initiative is important, as indicated by a growing body of literature on the contributions of mental health professionals who have experienced mental illness (Richards et al., 2016; Spaulding, 2016).

This initiative involves four key (groups of) actors—users, families, professionals and policy makers. Although the involvement of the first author is greater with the first and third group, he constantly attends meetings and provides as much as possible feedback and personal implication to the other two actors. Acting otherwise would limit the Citizenship movement to only one of the actors in this process and therefore, would have insignificant impact on the lives of users.

The overlap of Citizenship efforts with the incomplete implementation of recovery-based care in Spain has at times made it difficult for us to identify ways in which this project supports citizenship, or recovery, or both. In any case, both movements are dedicated to transforming the understanding, care of and support for people with mental health diagnoses to assure that they are the leading actors in their own recovery processes, and in their achieving full citizenship. We are convinced that both fostering Recovery and Citizenship practices are desirable. However, with an eye to criticisms of Recovery discussed earlier, this transfer process is not intended to be an exercise in neocolonialism. Our intention is not to implement an idea coming from an institution (Yale) whose prestige has obvious connotations, but to benefit from the evaluation possibilities that the process has had in a resource-rich context. The fact that the project has been initiated in a prestigious institution with access to resources and carrying a certain imprimatur, facilitates the justification and legitimacy of transfer. The use of focus groups with the groups involved as the first research tool and the professional and professional involvement of the principal investigator of the United States project, help to support the academic viability of the project in the Spanish context.

We are aware that many innovations do not occur through participatory processes such as we describe here. Our main objective is that innovations such as the Citizens Project can reach the maximum number of practitioners, with adaptation as needed for different contexts. Doing so might facilitate the extension of these concepts not only to the psychosocial rehabilitation sector, but also to other mental health care sectors that must promote and safeguard the dignity and agency of their users.

AUTHOR CONTRIBUTIONS

Both authors wrote and reviewed the whole paper. The content's responsibility is shared.

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"Female Preponderance" of Depression in Non-clinical Populations: A Meta-Analytic Study

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Clinical observations and research suggest a female preponderance in major depressive disorder. However, it is unclear whether a similar gender difference is found for the reporting of depressive symptoms in non-clinical populations. The present meta-analysis was conducted to address this issue. We searched for published papers targeting non-clinical populations in which the 21-item Beck Depression Inventory (BDI) was used. Eighty-four papers (91 studies) published between 1977 and 2014 were included in the final meta-analysis, which comprised 23,579 males and 29,470 females. Females in the general population reported higher level of depressive symptoms than males ($d = -0.187$, corresponding to 1.159 points in the 21-item BDI). This pattern was not found to influence by years of publication, socioeconomic status, or version of the BDI used. Using age group as a moderator, studies with adolescents and young adults were found to show a smaller effect size than studies with older participants. Our results appear to confirm the "female preponderance" in the level of self-report depressive symptoms in the general population, and support the social gender role theory in explaining gender difference over biological susceptibility theory and evolutionary theory.

Keywords: gender difference, depression, age, social gender role, economic status, BDI

INTRODUCTION

While results of several large-scale studies seem to suggest a higher level of depressive symptoms in females than males in the general population (Nolen-Hoeksema et al., 1999; Koivumaa-Honkanen et al., 2004; Aalto et al., 2012; Lopez Molina et al., 2014), consistent results are not always reported (Hammen and Padesky, 1977; Kim et al., 2011). However, due to the apparent agreement of this idea with the widely accepted "female preponderance" of major depressive disorder (Weissman and Klerman, 1977), this notion has been widely accepted, despite the lack of any systematic investigation in the general population.

Three representative theories purport to explain this gender difference in the level of depressive symptoms in the general population. First, the social gender role theory is based on females' long-standing disadvantaged social status which exists across many different cultures (Gove, 1977; Mirowsky, 1996). The theory states that lower status in the workplace and in heterosexual relationships cause chronic strain, decrease the sense of mastery and stimulate the use of a rumination strategy for emotional coping, thus contributing to a higher vulnerability to depression in females. Second, the biological susceptibility theory is based on the observation that clinical depression tends to be associated with events in the female reproductive cycle. Thus, the effect of female hormones has been proposed as a potential cause for the observed gender difference in depression (Janowsky and Rausch, 1985). Third, evolutionary psychologists have noted that males tend to value physical appearance more than females when choosing a partner. In modern societies, the mass media has repeatedly promoted the images of idealized attractive females and these images are readily accepted by both males and females. Thus, females may become increasingly dissatisfied with their own physical appearances due to increased pressure for mate selection, and as a result are more likely to become depressed when their appearances fall short of the ideal (Buss, 2003). All of these theories have been elaborated extensively (Weissman et al., 1993), but a direct comparison between them has not been carried out.

Unlike the ratio of gender difference in patients with diagnosed major depressive disorder (about 2:1), which is supported by many community surveys, epidemiological studies, clinical observations, and meta-analyses (Weissman and Klerman, 1977; Boyd and Weissman, 1981; Ayuso-Mateos et al., 2001; Hankin and Abramson, 2001; Seedat et al., 2009; Luppa et al., 2012), empirical support for the difference in level of depressive symptoms in the general population is less clear (Hill and Needham, 2013). In fact, some large scale studies have not found any gender difference in the level of depressive symptoms in non-clinical samples (Hammen and Padesky, 1977; Kim et al., 2011). Depressive symptoms are frequently measured by self-report measures, such as the Beck Depression Inventory (BDI; Beck et al., 1961). By combining results from different studies, a meta-analysis can answer not only whether there is a gender difference in the level of depressive symptoms in non-clinical populations, but also assess the magnitude of this difference. In view of the widely observed gender difference in diagnosed major depressive disorder (Weissman and Klerman, 1977; Boyd and Weissman, 1981; Blazer et al., 1998; Hankin et al., 1998; Ayuso-Mateos et al., 2001; Seedat et al., 2009; Luppa et al., 2012) and the continual nature of depression, we expected to find a gender difference in the level of depressive symptoms in non-clinical populations as well.

Using moderator analyses, it is also possible to examine what factors might be related to gender difference in the level of depressive symptoms. For example, the association between depression and economic status has been well-established (McGrath et al., 1990; Steptoe et al., 2007; Kessler et al., 2009). In addition to a stable ratio of gender difference in major depressive disorder (especially in industrialized countries), people living in poverty have been found to show a higher prevalence of diagnosed depression than those of a higher economic status in the same country (Kessler et al., 2009). However, whether gender difference in the level of depressive symptoms in the general population, if it exists, is influenced by socioeconomic status is largely unknown. Similarly, epidemiological surveys and family studies suggest that the rate of depression has been increasing over the past few decades (Angst, 1985; Klerman and Weissman, 1989; Cross-National Collaborative Group, 1992; Weissman et al., 1993), which may also influence gender difference in the level of depressive symptoms. For example, with epidemiological survey data collected in the United States, Canada, Germany, and New Zealand, Weissman et al. (1993) suggested that gender difference in the rate of depression is decreasing due to the increased prevalence of depression in men. This question has not been addressed in the general population. By including year of publication and socioeconomic status of countries/regions where the study was carried out as moderators, a meta-analysis could address both of the above questions. In addition, taking year of publication as a potential moderator offers us an opportunity to observe whether the expected gender difference in the general population is decreasing or not, since such a trend has been suggested in diagnosed depression (Klerman and Weissman, 1989).

Finally and theoretically, taking participants' age as a moderator in the meta-analysis offers the opportunity to test which of the three theories mentioned above best explain gender difference in the level of depressive symptoms in the general population. If the social gender role theory is valid, we would expect gender difference in the level of depressive symptoms to increase with gender socialization processes, and to remain unchanged when an individual attains the socially accepted gender role. If female hormones best explain this phenomenon, we would expect gender difference in the level of depressive symptoms to decrease in the age period when the production and release of female hormones decrease (Mirowsky, 1996), especially in the elderly group. Finally, if gender difference in the level of depressive symptoms was mainly due to increased dissatisfaction of physical appearance and increased mate selection pressure, we would expect the largest gender difference to be found in young adulthood when an individual's prime concern is to develop intimacy and find a mate (Erikson, 1975).

In summary, the primary aim of the present research was to conduct a meta-analysis to investigate whether there is gender difference in the level of depressive symptoms in the general population and to examine how this gender difference, if it exists, varies with time and socioeconomic status of a country/region. Finally, we aimed to examine which of the three aforementioned theories best explains this gender difference, if it exists, by including age as a moderator. The findings of the current meta-analysis may contribute to our understanding of clinical depression, since depression has been suggested as a spectrum ranging from subthreshold depressive symptoms to clinically diagnosed depression (Seligman, 1978; Flett et al., 1997; Bowins, 2015).

MATERIALS AND METHODS

Literature Search

First developed in 1961, the Beck Depression Inventory (Beck et al., 1961) is one of the best known and most widely used instruments for assessing the presence and severity of depressive

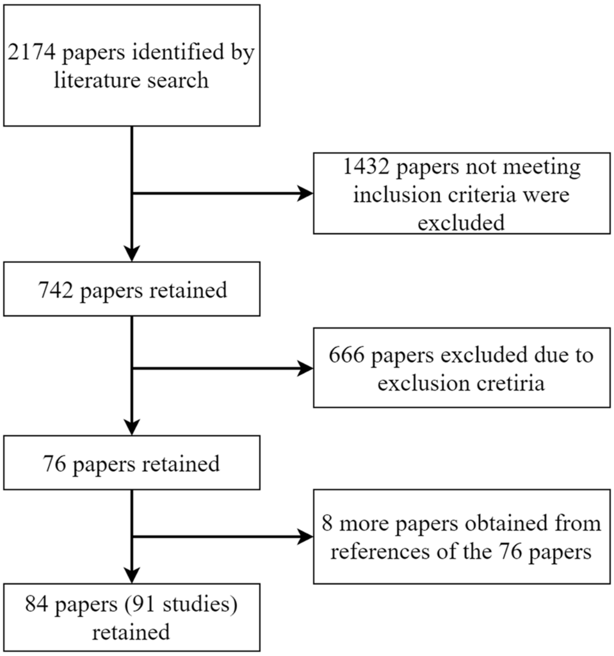

FIGURE 1 | Flowchart for the inclusion of published data for the current meta-analysis.

symptoms in individuals aged 13 years and above (Beck et al., 1996). It is considered a gold standard of self-rating scales of depressive symptoms (Cusin et al., 2010). Good agreement between the BDI and a clinical diagnosis of depression has been repeatedly demonstrated (Beck et al., 1996; Schotte et al., 1997; Lasa et al., 2000; Gomes-Oliveira et al., 2012; Araya et al., 2013). The cross-cultural acceptance of the BDI also makes it a good tool to use in studying the underlying construct of depression in different cultural settings. The BDI has several versions, with the 21-item version most frequently adopted in either English or non-English languages. Thus, in the present meta-analysis, we only selected studies that had used the 21-item BDI for a better cross-cultural comparison of depressive symptoms.

Potential journal articles were identified from PubMed, EBSCO (PsychINFO, PsychARTICLE), and Web of Knowledge between 1961 and February 2014. The keywords used were ‘Beck Depression Inventory’ or “BDI” in the text but not “patient” in either the title or the abstract of a paper. The search yielded an initial pool of 2174 articles.

The 2174 papers were checked against criteria for inclusion and exclusion (**Figure 1**).

The inclusion criteria were:

(1) Published in English (106 studies excluded);
(2) Participants included both genders (507 studies excluded);
(3) Participants were selected from the normal population (e.g., not elite athletes, transsexuals, cancer survivors, or cross-culturally married couples), and were free from physical and mental disorders (e.g., eating disorder, depression), addiction of any kind or obvious stress (e.g., care givers, survivors of accidents, victims of sexual assault, or recently bereaved; 800 studies excluded);
(4) The 21-item BDI was used (19 studies excluded).

After this step, a total of 742 papers remained and they were then subjected to the following six exclusion criteria:

(1) Studies that included data already reported in a previous study (one study excluded);
(2) Data for meta-analysis (means and SDs for both genders or exact *t* values or *F* values for gender means comparison) were not available (651 studies excluded);
(3) The BDI was used to measure the effect of a manipulation (10 studies excluded);
(4) Participants were asked to respond to the BDI under faked conditions (one study excluded);
(5) Not all 21 items of the BDI were used in the study (one study excluded);
(6) Case study (two studies excluded).

After this step, 76 papers were retained. A review of the reference list of these 76 papers generated another eight additional papers which were included in the meta-analysis. Consequently, the final sample included 84 papers, seven of which had two participant groups. Thus, the final meta-analysis contained 91 studies.

The socioeconomic status of a country/region where the study was carried out was measured using two indices: the gross domestic product (GDP) per capita and the Gini index. GDP data for 2013 (the most recent available data) were retrieved from the World Bank online database[1]. The Gini index reflects the income distribution of a nation’s residents, and is the most commonly used measure of inequality (Gini, 1909). Since the Gini index data for each country/region provided by the World Bank contained many missing data, we retrieved the relevant data from Quandl[2], a search engine with financial, economic, and social studies. Nevertheless, the Gini indices for four studies were not available (one from the United Arab Emirates, one from Bahrain and two from Saudi Arabia). We then compared the Gini indices of regions/countries included in the present analysis with data available from the World Bank and found that the data tallied well.

It has been suggested that different versions of the BDI used across studies may be the cause of inconsistent findings in gender difference in the level of depressive symptoms in the general population (O’Hara et al., 1998). For this reason, we also investigated whether gender difference in the level of depressive symptoms may be influenced by the BDI version used (English vs. non-English; for English version: BDI-I vs. BDI-II). This measure was taken as a potential moderator for analysis. Overall, potential moderators in this meta-analysis included the age range of the participants, year of publication, the BDI version used, the GDP per capita, and the Gini index of the country/region where the study was carried out.

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[1] http://data.worldbank.org/indicator/NY.GDP.PCAP.CD

[2] http://www.quandl.com/demography/gini-index-by-country

Meta-Analysis Procedure

The present meta-analysis was carried out using the Comprehensive Meta-Analysis (version 2.0) software package (Borenstein et al., 2005). Since we only included studies using the 21-item BDI, the mean difference (the mean BDI score in the male sample minus the mean BDI score in the female sample) was also available as an effect size, together with the widely used Cohen's *d*. For both measures, a negative difference score indicates a higher BDI depression score in females than in males. The random model was used for calculating the effect sizes due to expected heterogeneity. A funnel plot was used to illustrate potential publication bias and quality of individual studies. In addition, the fail-safe-N, the number of additional unpublished studies with negative effect that would be needed to increase the *p* value for the meta-analysis to above 0.05, was also calculated to estimate publication bias (Rosenberg, 2005).

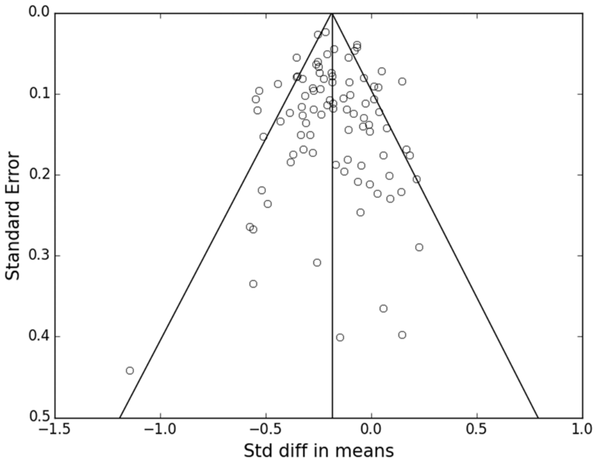

FIGURE 2 | Funnel plot of standard errors as a function of standard difference in the means.

RESULTS

Feature of Studies

The 91 studies were published between 1977 and 2014, with 38 published before 2000 and 53 published in 2000 or later. These studies involved 23,579 males and 29,470 females in 22 different countries/regions from six continents (1 in Africa, 11 in Asia, 25 in Europe, 40 in North America, 3 in Oceania, and 11 in South American). The socio-economic status of these countries varies substantially, with the GDP per capita ranging from USD 4838.50–67458.40 and the Gini index from 27.79 to 63.14. In relation to the versions of the BDI used, 47 studies used the English version of the BDI, among which 22 clearly stated using the BDI-I and 13 stated using the BDI-II. The other 44 studies were carried out with a non-English version of the BDI. Based on the age range of the participants and the sampling approach used, the studies were classified into five groups, adolescents (13 studies), young adults (exclusively university students, 45 studies), middle-aged adults (30–50 year old, five studies), older adults (six studies), and general population aged above 13 years (15 studies). The remaining seven studies could not be classified into any of these groups due to insufficient information. The mean depression score in 86 studies (five did not report the mean total scores, but the *t* or *F* value for between-gender comparison on BDI total score were available for meta-analysis) ranged from 2.08 to 19.30 [five reported a BDI mean total score higher than 13, the cut-off for mild depression on BDI-II (Beck et al., 1988)] for males, and 2.86–19.00 (eight reported a BDI mean total score higher than 13) for females.

General Analysis

The effect size for gender difference of each study varied from −1.146 to 0.227. Taken together, a random effect size of −0.187 ($SE = 0.017$, 95% $CI = [-0.220, -0.155]$) was found. The small but significant ($Z = -11.70$, $p < 0.001$) effect size indicated that on average females scored about 1.159 points ($SE = 0.112$, 95% $CI = [-1.379, -0.939]$) higher on the 21-item BDI than males. Given the large number of studies, it could be expected that the 91 studies were not homogeneous [$Q(90) = 223.218$, $p < 0.001$, $I^2 = 59.681$]. Closer inspection showed that the study reported by Goodrich and Weaver (1998) generated a very large effect size of −1.146 (the second largest was −0.576). The sample included only 11 men and 13 women. Removing this study from the pool had a negligible effect on the mean effect size (Cohen's $d = -0.186$). This study was therefore excluded from further analysis.

Publication bias was evaluated by the fail-safe-*N* and funnel plot. The fail safe N was 6987. **Figure 2** shows the funnel plot with each circle representing an individual study included in the meta-analysis. The circles were generally symmetrical in their distribution, indicating the absence of publication bias. Additionally, most of the circles were located near the top of the funnel, indicating a relatively small standard error, suggesting good methodological quality of these studies.

Potential Moderator Analysis

Taking age as a moderator, we compared the effect sizes of the five groups of studies, namely adolescents, young adults, middle-aged adults, general population, and older adults. Significant effect sizes (**Table 1**) were generated from each group. Studies in the older adult group, the general population group, and the middle-aged adult group were homogenous. Additionally, the effect sizes generated from these three groups were not significantly different [$Q(2) = 4.089$, $p = 0.13$]. Studies in the adolescent group and the young adult group were heterogeneous, and generated relatively small but significant effect sizes (adolescents: $d = -0.188$; early adults: $d = -0.157$). To test whether the effect sizes of the two younger groups were smaller than those of the three older groups, we created a "young" group including studies with adolescents and young adults, and an "old" group with studies involving middle-aged adults, older adults, and the general population. The 58 studies in the "young" group generated a small but significant effect size of −0.166 ($SE = 0.024$, 95% $CI = [-0.212, -0.119]$, $Z = -11.66$, $p < 0.01$), which corresponded to −1.063 ($SE = 0.167$, 95% $CI = [-1.390, -0.736]$) points in the total BDI

TABLE 1 | Effect sizes of gender difference in BDI (Beck Depression Inventory) total score across age groups.

Age	K	NM	NF	Cohen's *d*	*SE*	95% CI	Z	Heterogeneity		Fail-safe-N
								Q	I^2	
Adolescents	13	4640	4852	−0.188	0.042	−0.271, −0.105	−4.44**	39.745**	69.808	199
Early adults	45	6599	9389	−0.157	0.029	−0.214, −0.100	−5.37**	109.94**	59.978	773
Middle adults	5	3497	4966	−0.251	0.052	−0.352, −0.149	−4.83*	5.567	28.150	66
General population	15	7436	8474	−0.208	0.024	−0.255, −0.162	−8.75**	22.93	38.939	505
Elderly	6	776	1032	−0.315	0.048	−0.409, −0.220	−6.55**	4.265	0.000	20

*K, number of studies used for meta-analysis; NM, number of males; NF, number of females; **p < 0.01; *p < 0.05.*

score. The fail-safe N was 1804. The 26 papers in the "old" group generated an effect size of −0.224 ($SE = 0.019$, 95% $CI = [-0.262, -0.187]$, $Z = -11.76$, $p < 0.01$), which corresponded to −1.33 ($SE = 0.117$, 95% $CI = [-1.562, -1.103]$) points in the total BDI score. The fail-safe N was 1283. The *Q*-test indicated that the effect sizes in the "old" group were larger than those in the "young" group [$Q(1) = 3.702$, $p = 0.05$]. Studies that recruited from the general population contained individuals belonging to both the "old" and "young" groups. For this reason, we excluded these studies from the "old" group and found a similar group difference [$Q(1) = 5.270$, $p < 0.05$].

To test whether the observed gender difference in depressive symptoms systematically varied with time, a correlation analysis between publication year and effect size of the 91 studies was carried out. This revealed no significant correlation ($r = -0.11$, $p = 0.30$). In addition, we classified all the 91 studies into two groups, those published before 2000 and those published after 2000. The 38 studies published before 2000 generated an effect size of −0.181 ($SE = 0.032$, 95% $CI = [-0.243, -0.119]$, $Z = -5.730$, $p < 0.001$), with a fail-safe number of 688. The studies included were heterogeneous [$Q(37) = 86.372$, $p < 0.01$, $I^2 = 57.162$]. Similarly, the 53 studies published in 2000 or later also generated a small but significant effect size of −0.190 ($SE = 0.020$, 95% $CI = [-0.229, -0.151]$, $Z = -9.54$, $p < 0.001$), and the studies included were heterogeneous [$Q(52) = 135.672$, $p < 0.01$, $I^2 = 61.672.162$]. The fail-safe number was 3218. The effect sizes of the two groups were not significantly different [$Q(1) = 0.056$, $p = 0.82$]. Using the 85 studies that reported the mean BDI score for each gender, correlation analyses were carried out to test whether depressive symptoms increased with publication year. No significant correlation was found (male: $r = 0.064$, $p = 0.55$; female: $r = 0.13$, $p = 0.23$. Please see **Supplementary Figure S1** for a scatter plot between depressive symptoms and year of publication for both genders).

Next, the two measures of socioeconomic status were included as moderators for analysis. First, correlation analyses between socioeconomic indices (GDP per capita and Gini index) and BDI scores were carried out separately for both genders for the 87 studies. GDP per capita correlated negatively and significantly with BDI scores for both genders (females: $r = -0.428$, $p < 0.01$; males: $r = -0.414$, $p < 0.01$), indicating that studies carried out in areas with higher GDP per capita generated a lower mean BDI score. Similar significant correlations were observed between the Gini index and BDI scores (females: $r = 0.336$, $p < 0.01$; males: $r = 0.356$, $p < 0.01$), indicating a higher BDI score in countries/regions with a higher Gini index. To test whether gender difference in the level of depressive symptoms also varied with socioeconomic status, correlation analyses between the two socioeconomic indices and the respective effect sizes were carried out. No significant correlation was found (GDP: $r = 0.006$, $p = 0.96$; Gini index: $r = 0.056$, $p = 0.61$). **Figure 3** illustrates the relationship between the BDI score of each gender, effect sizes, and GDP. All the data were standardized.

The last moderator examined was the BDI version used. The wordings used in the BDI of a certain language may contribute to the observed gender difference in the level of depressive symptoms. As such, we separated all the studies into two groups: one group consisting of studies using an English version of the BDI and the other group consisting of studies using a non-English version of the BDI. The 47 studies using the English version of the BDI generated a small but significant effect size of −0.155 ($SE = 0.027$, 95% $CI = [-0.208, -0.102]$, $Z = -5.725$, $p < 0.01$). These studies were heterogeneous [$Q(46) = 89.701$, $p < 0.01$, $I^2 = 48.718$]. The large fail-safe-N of 710 indicated that this small effect was reliable. Similarly, the 44 studies using non-English versions of the BDI also generated a significant

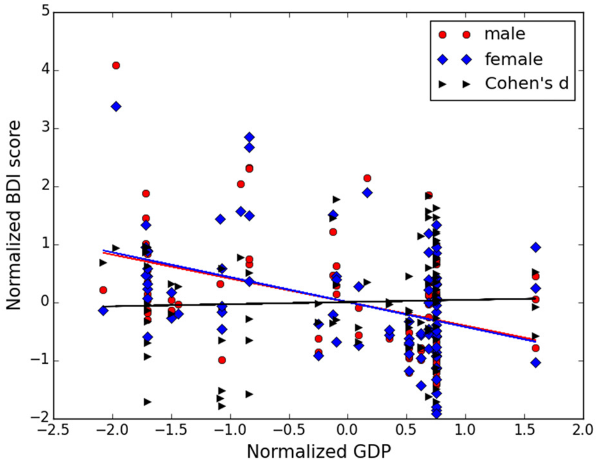

FIGURE 3 | Scatter plots between GDP (gross domestic product) per capita and male BDI (Beck Depression Inventory) score (red), between GDP per capita and female BDI score (blue), and between GDP per capita and Cohen's *d* (black). All data were normalized for illustration purpose.

effect size of −0.211 ($SE = 0.021$, 95% $CI = [-0.252, -0.170]$, $Z = -10.097$, $p < 0.01$). These studies were heterogeneous [$Q(43) = 121.616$, $p < 0.01$, $I^2 = 64.643$]. The fail-safe-N was 9162. There was no significant difference between the effect sizes of the two groups [$Q(1) = 2.754$, $p = 0.10$].

In studies using the English version of the BDI, we also tested whether different versions of the BDI contributed to the observed gender difference in the level of depressive symptoms. Among the 47 studies adopting the English version of the BDI, 22 clearly stated that they used the original BDI-I (published in 1961) and 13 used the BDI-II. The 22 studies using the BDI-I generated a significant effect size of −0.172 ($SE = 0.041$, 95% $CI = [-0.253, -0.091]$, $Z = -4.149$, $p < 0.01$). The studies were heterogeneous [$Q(21) = 38.36$, $p = 0.01$, $I^2 = 45.255$] and the fail-safe-N was 142. Similarly, a small but significant effect size of −0.117 ($SE = 0.041$, 95% $CI = [-0.198, -0.037]$, $Z = -2.848$, $p < 0.01$) was generated from the 13 studies using the BDI-II. These studies were heterogeneous [$Q(12) = 22.091$, $p < 0.01$, $I^2 = 45.679$]. The fail-safe-N was 42. These two effect sizes were not significantly different [$Q(1) = 0.865$, $p = 0.35$], indicating that gender difference in the level of depressive symptoms was not likely to be influenced by the version of the BDI used.

DISCUSSION

The main aim of the present study was to use the meta-analytic method to ascertain whether there is a gender difference in the level of depressive symptoms in the general population. With 91 studies from five continents, we found that females did report a significantly higher level of depressive symptoms than males, with female scoring on average about 1.159 points more on the BDI than males (the highest possible BDI total score is 63). Importantly, the studies in the present meta-analysis were largely homogeneous, except those that included adolescents and young adults. This finding is interesting because most of the previous studies that did not find a gender difference in the level of depressive symptoms in non-clinical populations were based on adolescent (Teri, 1982; Martin et al., 1995; Kim et al., 2011) or university student samples (Hammen and Padesky, 1977; Gould, 1982; Bryson and Pilon, 1984; Endler et al., 1992; Steer and Clark, 1997; O'Hara et al., 1998; Pillay et al., 2002; Erol et al., 2006; Thomas and Altareb, 2012). Moreover, the gender difference in the level of depressive symptoms appeared to be independent of socioeconomic status, year of publication, and version of the BDI used. Overall, our results appear to confirm that there is a "female preponderance" in the level of depressive symptoms in the general population.

By taking into consideration two socioeconomic indices as moderators, we found a negative relationship between socioeconomic status and the level of depressive symptoms when socioeconomic status was assessed at the country/region level. Previous research on the relationship between socioeconomic status and depression was mainly carried out by assessing socioeconomic status at the individual level (Kessler et al., 2009). Taking the findings together, it can be concluded that poor socioeconomic status has a negative impact on a person not only at the individual level, but also at a country/region level. If we take depression as a continuum (Flett et al., 1997; Bowins, 2015), with healthy individuals showing mild symptoms at one end and those with a clinical diagnosis at the other end, this finding speaks against the notion that depression is a disease of modernization (Hidaka, 2012), and supports that poverty is "the pathway to depression"(McGrath et al., 1990). It is possible that in developed countries/regions, people have more knowledge about and better acceptance of mental illness, and more opportunity to have their depression diagnosed and treated. As a result, the prevalence of depression in developed countries/regions presented may be artificially inflated[3]. In contrast, in less developed countries/regions, due to poorer knowledge and acceptance of mental illness and less opportunity for diagnosis and treatment, more depressed individuals are left undiagnosed and untreated, presenting a "lower" prevalence rate.

Depression in China, where people's acceptance of depression is not high (Xu, 1987), might offer some insight for the above point of view. The prevalence of depression in China is relatively low, with a 1-year prevalence of 1.8% and a life-time prevalence of 3.6% according to a WHO survey (Lee et al., 2009), which is much lower than the USA (Kessler et al., 2003). In another study, researchers randomly selected 50 general hospitals in Beijing in 2003–2004. A total of 73 cases who met the DSM-IV criteria for major depression were screened out among all the outpatients. Only 10 of them (14%) were identified by physicians, and only four (5%) sought help from psychologists or psychiatrists (Zhang et al., 2006). In other words, the existence of many unidentified patients may artificially "decrease" the prevalence of depression in China. This possibility is also compatible with our analysis using year of publication as a moderator. We observed that level of depressive symptoms did not vary with year of publication in the last few decades, while epidemiological studies have shown an "increasing" prevalence of depression (Joyce et al., 1990; Kessler et al., 1994; Murphy et al., 2000; Lee et al., 2007). It is possible that the increasing prevalence rate was due to an increasing number of people being diagnosed and treated secondary to increased awareness and acceptance of depression.

Another interesting finding regarding the influence of socioeconomic status on the level of depressive symptoms is that these indices had no relationship with gender difference in depressive symptoms. This suggests that women in both non-industrialized and industrialized countries/regions report a higher level of depressive symptoms than males. Previously, it has been suggested that the gender ratio of major depressive disorder in non-industrialized countries is smaller than in industrialized countries (Weissman et al., 1993). One piece of evidence that supports this viewpoint is that treated cases of depression in some non-industrialized countries indicated either equal gender ratio [e.g., Bulaways in Rhodesia (Republic of Zimbabwe after 1980) and Baghdad in Iraq; see Table 1 in Weissman and Klerman, 1977] or even a higher proportion of males than females (e.g., Dakar in Guinea, Madurai, Madras, and New

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[3] In cases of countries/regions where a hunter-gather lifestyle is dominant and contact with the "outside" world is nearly absent, there may be another explanation. However, none of the reviewed studies were conducted in such counties/regions.

Delhi in India, see Table 1 in Weissman and Klerman, 1977). Considering the economic situation as well as the level of medical service in these countries/regions, it is possible that depression in females in these countries/regions may be even more underdiagnosed than males. This issue warrants further research in these countries/regions.

The final aim of the present meta-analysis was to evaluate the validity of the three theories purporting to explain the gender difference in depression. To do this, analyses were conducted using age as a moderator. Similar to the finding previously reported in a study using patients with diagnosed depressive disorders (Angold et al., 1999), gender difference in the level of depressive symptoms was found in our youngest group, that is, the adolescents. A similar extent of gender difference in the level of depressive symptoms was also observed in young adults. The gender difference increased significantly when the group reached 30 years of age. The change in gender difference magnitude with age is consistent with the social gender role theory of depression, which is based on the disadvantaged social status that females experience relative to males (Gove, 1977; Mirowsky, 1996). In particular, it supports the age increment prediction of this theory as originally proposed by Mirowsky. Smaller gender differences are expected in adolescence since the majority of women and men would be in school and relatively few would be in paid employment. Gender difference in levels of depressive symptoms emerges in adolescence and increases significantly in middle adulthood due to the emergence of inequities in employment, housework, family responsibilities, and other factors.

The change in gender difference with age is not consistent with the biological or evolutionary theories. A biological account of gender difference, which suggests that the influence of female hormones as the cause (Janowsky and Rausch, 1985), is not consistent with the similar gender difference in the level of depressive symptoms in the middle-age and the older adult groups. We would expect female hormone levels to be different in these two periods of the human lifespan. Similarly, the evolutionary theory, which highlights females concerns about their own appearance and body image in mate selection, would predict a peak in gender difference in the level of depressive symptoms in the two younger age groups. However, our results did not support this prediction.

The embodiment of unequal social gender for adult females has been well described by those who believe that an unfavorable social gender role causes females to be at a higher risk of depression (Barnett et al., 1992; McIntosh et al., 1994). A quote from the recent Human Development Report summarized the poor situation experienced by many females worldwide: "Women experienced many kinds of disadvantage and discrimination in health, education, and employment" (United Nations Development Programme, 2014, p. 39). Thus, it appears that there is a significant link between the present finding of females scoring 1.159 points higher than males on the 21-item BDI and the observed 8% lower score of females on the Human Development Index (a comprehensive index which takes into consideration life expectancy, education, and economy) as reported by the recent Human Development Project (United Nations Development Programme, 2014). Importantly, the present findings suggest that the link may be important even at the sub-clinical level of depression in the general population.

It might be expected that the gender difference in depression would show a decline over recent decades, given improvements in education, technology, and life expectancy (United Nations Development Programme, 1990). Moreover, changes in society have begun to address the disadvantages experienced by women, particularly in industrialized countries. However, correlation analysis between year of publication and the magnitude of gender difference in the level of depressive symptoms did not support this prediction. Similar conclusions of stable gender differences in mathematical and science achievements have also been reported (Reilly et al., 2015), which further reinforces the conclusion that recent societal changes asserting women's rights did not seem to reduce gender gaps. Such findings suggest that more specific factors may be important for the gender difference in psychological functions (Reilly and Neumann, 2013).

A limitation of the present meta-analysis is that we cannot exclude the possibility that the observed gender difference in the level of depressive symptoms is due to systematic bias in participants' responses to certain items of the BDI. For example, several studies have shown that females are more likely to respond positively to items such as "crying" (O'Hara et al., 1998), "self-dislike," "fatigability," and "somatic preoccupation" (Gorenstein et al., 1999). Future studies are needed to explore and address this possibility. Additionally, the results of the present study might have been influenced by the exclusion of certain studies.

AUTHOR CONTRIBUTIONS

KW designed the study; HL and KW searched the literature and analyzed data; KW wrote the first version of the manuscript; other authors contributed on writing and revising the manuscript.

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SUPPLEMENTARY MATERIAL

The Supplementary Material for this article can be found online at: http://journal.frontiersin.org/article/10.3389/fpsyg.2016.01398

FIGURE S1 | The scatter plots between mean BDI (Beck Depression Inventory) score and year of publication for female (circle) and male (square).

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How Not to Do a Mindset Intervention: Learning from a Mindset Intervention among Students with Good Grades

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The present study examined the effectiveness of a Growth Mindset intervention based on Dweck et al.'s (1995) theory in the Hungarian educational context. A cluster randomized controlled trial classroom experiment was carried out within the framework of a train-the-trainer intervention among 55 Hungarian 10th grade students with high Grade Point Average (GPA). The results suggest that students' IQ and personality mindset beliefs were more incremental in the intervention group than in the control group 3 weeks after the intervention. Furthermore, compared to both the baseline measure and the control group, students' amotivation decreased. However, no intrinsic and extrinsic motivation change was found. Students with low grit scores reported lower amotivation following the intervention. However, in the second follow-up measurement—the end of the semester—all positive changes disappeared; and students' GPA did not change compared to the previous semester. These results show that mindset beliefs are temporarily malleable and in given circumstances, they can change back to their pre-intervention state. The potential explanation is discussed in the light of previous mindset intervention studies and recent findings on wise social psychological interventions.

Keywords: good grades, grit, growth mindset, incremental theory of intelligence, social psychological intervention

INTRODUCTION

The Incremental theory of intelligence—Growth Mindset since the book of Dweck (2006)—deals with beliefs influencing responses to challenges and setbacks (Dweck and Leggett, 1988; Hong et al., 1999). These beliefs refer to theories of students concerning the nature of intelligence. Students may believe that intelligence cannot be changed and it can be represented as a limited, stable entity (entity theory). Alternatively, they may believe that it can be developed (incremental theory) and further improved (for meta-analysis see Burnette et al., 2013). According to previous results, such beliefs have serious implications in terms of reactions to challenges, motivations, and choosing or giving up demanding activities (Dweck and Leggett, 1988; Mueller and Dweck, 1998). From the perspective of the societal significance of this theory, growth mindset can reduce the negative effect of poverty on academic achievement (Claro et al., 2016). From the perspective of cognitive neuroscience, growth mindset induction contributes to better cognitive control (Schroder et al., 2014). In the present study, we carried out a Mindset intervention which aimed to create incremental beliefs of intelligence among Hungarian high school students.

Based on Dweck's Mindset theory, several intervention studies were conducted with positive and promising results, mainly among underprivileged or minority groups. In one of the first interventions testing this theory, African-American students were encouraged to perceive their intelligence as a malleable rather than a fixed capacity (Aronson et al., 2002). This intervention made the performance of these students—compared to the control group—less vulnerable to stereotype threat, helped them in maintaining their academic engagement and also led to higher GPA. In a further study, Good et al. (2003) found that African-American, Hispanic and low income students' stereotype threat was reduced as a consequence of mentoring college students. These mentors encouraged the participants to see their intelligence malleable and helped them in attributing their academic problems to the novelty of educational context they were in. In another intervention among lower achiever students, focusing on the creation of incremental beliefs of intelligence had positive impact on classroom motivation, and it stopped the decline in mathematics grades compared the control group (Blackwell et al., 2007). More recently, Paunesku et al. (2015) carried out a large scale online field experiment in which they measured the effectiveness of a short (45 min) Mindset training. They found that exposing the incremental theory of intelligence raised the GPA among high school students who were at risk of dropping out. Consequently, these above-mentioned studies showed that the mindset intervention was mainly effective among those students who are African-American, Hispanic, lower achiever, who had lower income or who are at risk of drop-out.

The question arises whether Mindset trainings can be effective in a fundamentally different cultural and educational context: the Hungarian educational system (Csapó, 2015). In Hungary, efforts in general do not have a good reputation. For example, according to the fifth wave of the World Value Survey, only 7% of the Hungarians agreed that hard work pays off in the long run which is much lower than the total average of 19% (WVS, 2009). Furthermore, dissimilarly to previously measured high schools in the United States, the Hungarian public education can be characterized by rather conservative values, stronger state-level control of the national curriculum, and less supportive climate concerning change. In the present study, we aimed to test the effectiveness of growth mindset interventions in a fundamentally different school context as the US one. Furthermore, besides the potential less supportive educational context, we aimed to examine whether growth mindset can be effective among students who have relatively good grades.

In the present study, a Growth Mindset intervention was carried out for high-school students and its effect on academic motivations and grades was measured. The intervention protocol was similar to Aronson et al. (2002), Good et al. (2003), Blackwell et al. (2007) and Paunesku et al. (2015) and it was based on the precise and well-founded theory of the incremental theory of intelligence (Hong et al., 1999). Similarly to Blackwell et al.'s (2007), we intended to create a brief, offline intervention in which five classes (45 min) were spent with integrating the knowledge of incremental intelligence. Similarly to this study, it was a train-the-trainer intervention where the authors trained the teachers. We were mainly interested in the dynamics of the mindset beliefs and their short vs. long-term effects on the motivations of among students with good grades and the impact of intervention on their GPA. We expected that—as a result of the Growth Mindset intervention—students will be more motivated (H1) and will have higher GPA (H2).

Growth mindset beliefs are general beliefs that have a pervasive effect on different fields (see the meta-analysis of Burnette et al., 2013). Furthermore, previous interventions showed that there is a way to change them (Blackwell et al., 2007; Paunesku et al., 2015; Yeager et al., 2016). However, as far as we know, we have less accumulated knowledge about more stable individual differences—as personality traits—that might influence the effectiveness of these beliefs. One of the potential, proximal and stable individual differences or personality characteristics is Grit which refers to the perseverant and passionate striving toward long-term goals (Duckworth et al., 2007). Previous studies suggested that this characteristic contributes to academic success and success in many fields and in different age-groups (Duckworth et al., 2007; Duckworth and Quinn, 2009). We expected that the Growth Mindset intervention will be more effective among low Grit students compared to high Grit students (Duckworth and Quinn, 2009). We supposed that a Growth Mindset intervention—focusing on the importance of efforts and good strategies—can be less effective in terms of changing motivations and grades among those who are persistent and already make a lot of effort and strive for long-term goals compared to low Grit students whose motivations and grades can be improved by enhancing the importance of efforts (H3).

MATERIALS AND METHODS

Participants

A total of 123 Hungarian public high school students and their parents were told that they had the opportunity to participate in a 5-week workshop. In the case of the control group, they were informed that they could participate in an intervention related to helping behavior. In the experimental group, they were informed about the possibility to participate in a workshop about intelligence and learning.

Five parents and six students indicated that they did not wish to participate in the training. Therefore, 112 students participated in the pre-test, 79 students participated in the post-test and 55 students participated in the second post-test in this study in spring 2015 (from February to June). Causes of drop-out were non-participation in each round of the intervention (49 students), same code-names for different students (2 students), or code-names could not be matched with the students' real identity (6 students). These students were recruited from two Hungarian high schools from the countryside. Their average GPA right before the intervention was 3.72 ($SD = 0.94$) based on a

1 (unsatisfactory) to 5 (excellent) grading system. Students were 10th graders aged between 15 and 18 years ($M_{\text{age}} = 16.00$; $SD_{\text{age}} = 0.58$). A total of 26 students (11 female, 42.31%) were included in the intervention group and a total of 29 students (18 female, 62.07%) were in the control group.

To ensure the ethical treatment of human participants, this study was carried out with the approval of the local university's ethical board and it was conducted in accordance with the Declaration of Helsinki. Participation was entirely voluntary, and the consent of both students and parents were obtained in advance. Furthermore, the directors of the schools also approved the intervention in advance.

Measures

All measures used in this study were translated to Hungarian from the original scales using the protocol of Beaton et al. (2000).

Intelligence Mindset Scale

We used the Implicit Theory of Intelligence Scale (Dweck et al., 1995) to assess the participants' beliefs of intelligence changeability. The scale contains eight items; four of them are reverse coded. Respondents indicated their answers on a ten-point Likert scale (from 1—"strongly disagree" to 10—"strongly agree"). This scale had good internal consistencies ($\alpha_{\text{pre}} = 0.79$; $\alpha_{\text{post}} = 0.89$; $\alpha_{\text{post2}} = 0.90$). Higher scores on this scale indicate higher levels of intelligence changeability beliefs

Personality Mindset Scale

We used the Implicit Theory of Intelligence Scale (Dweck et al., 1995) to assess the participants' beliefs of personality changeability. The scale contains 12 items; six of them are reverse coded. Respondents indicated their answers on a ten-point Likert scale (from 1—"strongly disagree" to 10—"strongly agree"). This scale had good internal consistencies ($\alpha_{\text{pre}} = 0.81$; $\alpha_{\text{post}} = 0.93$; $\alpha_{\text{post2}} = 0.93$). Higher scores on this scale indicate higher levels of personality changeability beliefs.

Academic Motivation Scale

We used the Academic Motivation Scale (Vallerand et al., 1992; Orosz et al., 2013) to assess the students' level of amotivation, intrinsic and extrinsic motivation toward studying. The scale contains 11 items; four items belong to the amotivation and extrinsic motivation factors and the intrinsic motivation included three items. Respondents indicated their answers on a seven-point Likert scale (from 1—"doesn't correspond at all" to 7—"corresponds exactly"). This scale had good internal consistencies (amotivation: $\alpha_{\text{pre}} = 0.86$; $\alpha_{\text{post}} = 0.91$; $\alpha_{\text{post2}} = 0.91$; extrinsic motivation: $\alpha_{\text{pre}} = 0.78$; $\alpha_{\text{post}} = 0.80$; $\alpha_{\text{post2}} = 0.73$; intrinsic motivation: $\alpha_{\text{pre}} = 0.82$; $\alpha_{\text{post}} = 0.78$; $\alpha_{\text{post2}} = 0.78$).

Short Grit Scale

We used the Short Grit Scale (Duckworth and Quinn, 2009) to assess the participants' level of persistence. The scale contains eight items. Respondents indicated their answers on a five-point scale (from 1—"very much like me" to 5—"not like me at all"). This scale had good internal consistencies ($\alpha_{\text{pre}} = 0.77$; $\alpha_{\text{post}} = 0.85$; $\alpha_{\text{post2}} = 0.85$). Higher scores on this scale indicate higher levels of persistence.

GPA

GPA data was available from the school's electronic diary. GPA in Hungary includes all grades including main (Mathematics, Hungarian literature and grammar, history, and foreign language) and peripheral subjects (Physics, Biology, Chemistry, Geography, Music, Informatics, PE, Arts) as well. The average GPA can be calculated based on a 1 (unsatisfactory) to 5 (excellent) grading system by aggregating and averaging the grades of the above-mentioned subjects.

Procedure

Dissimilarly to the study of Blackwell et al. (2007) in which research was carried out by the experts, it was a train-the-trainer intervention in which one of the authors showed the training exercises to the randomly assigned teachers during 4 h and then they implemented it without further supervision. During the intervention, students were not exposed to direct persuasion regarding the content. The intervention was not framed as a helping session, but as an opportunity to learn about how one can react to difficulties.

We started the intervention at the beginning of a spring semester. The mindset intervention was held in five 45-min sessions (one per week), during a homeroom class which is designated to studies-related activities that are independent from specific classes, such as recording the attendance. In average 25 students participated in each of the classes. Homeroom teachers (teachers who have a certain class assigned to look after) of these classes were randomly assigned to experimental vs. control group. Before the first session, we assessed the students' intelligence and personality beliefs, their academic motivations, and individual differences in Grit. **Table 1** provides details about the intervention protocol which borrowed some elements form the guidelines of the Heroic Imagination Project's *"The Growth Mindset: The psychology of motivation and success"* (Dickerson et al., unpublished manuscript) workshop schedule. Compared to previous interventions (Blackwell et al., 2007; Paunesku et al., 2015), in the present case, we put more emphasis on the everyday academic aspects of the incremental theory of intelligence instead of the functioning of the brain. Neuroplasticity got a larger emphasis only in the third session.

The control group got the same type of tasks as the intervention group, but regarding the bystander effect. The pre-test was carried out 1 week before the intervention, the post-test was 3 weeks after the 5-week long intervention and the second post-test was at the end of the school year (2 months later than the first post-test and 4 months later than the pre-test).

Statistical Analysis

To test our hypothesis that the mindset intervention would change the intelligence, personality beliefs of students, we performed 2 × 3 mixed model analyses of variance (ANOVA) with CONDITION (Growth Mindset intervention vs. control) as a between-subjects factor, and TIME (pre-intervention, post-intervention and second post-intervention) as a within-subjects factor.

First, we tested whether the growth mindset intervention changed the intelligence and personality beliefs of students

TABLE 1 | Summary of the intervention protocol.

Sessions	Intervention group—mindset	Control group—bystander effect
1	Warm up, Introducing the growth mindset, exercise about a personal failure sharing it in pairs, discussions about short videos in which failures were exposed, Close up.	Warm up, Introducing the bystander effect, (1st exercise) Imagining a situation in which a person is lying on the pavement—what would you do?, (2nd exercise) Watching a short video about the bystander effect and talking about it in small groups, Close up.
2	Warm up, (1st exercise) Demonstration of a short Mindset video and based on it explanation of the difference between fixed mindset and growth mindset in terms of (a) reactions to setbacks, (b) reactions to challenges, (c) the role of effort, (d) the role of challenges, (e) reactions to criticism, (f) reactions to others' success with the active group-level activity of students. (2nd exercise) Pair then group discussion about someone who had positive expectations, personal story sharing, analysis of this behavior, its relationship with growth mindset, if you are a leader why it is important to have a growth mindset. (3rd exercise) Pair discussion about a time when the student was bad at something and now is good with the following questions: How she/he get better? How much work was needed for it? Close up, Homework.	Warm up, (1st exercise) Demonstration of a short video on lack of activity in an emergency situation and talking about the content of the video, the possible feelings and thought of the characters. (2nd exercise) Every student recorded and wrote down two own stories: one about helping someone in need and another one about not helping. Small group discussion was carried out about the stories and the feelings concerning the situations. Then these stories were anchored to the concept of pluralistic ignorance, diffusion of responsibility, and the spotlight effect which can prevent from helping in emergency situation. Close up, Homework.
3	Warm up, (1st exercise) Group level simulation of a debate in one's head to give up or work harder after a failure, gathering the good arguments to make effort despite the setback. (2nd exercise) Exposing (videos), writing down and discussing the main obstacles and writing down personal plans to overcome them. Later pair, finally group discussion. Close up, Homework.	Warm up, (1st exercise) Group level simulation of a debate in one's head to help a younger student bullied by an older one or not to help him, gathering the pro and contra arguments. (2nd exercise) Gathering the obstacles that occur in a helping situation and find the solutions to them. Close up, Homework.
4	Warm-up, (1st exercise) Writing personal plan for responding with a growth mindset every time they experience challenge or setback. Then pair, finally group discussion about these plans giving advices to each other about coping with difficulties. (2nd exercise) Writing word associations about Mindset, then gathering in group and explaining with their own words in group setting, discussing on the most interesting and surprising aspects. Close up, Homework.	Warm up, (1st exercise) Writing a personal plan for helping one of your classmates who is bullied. Then pair, finally group discussion about these plans—with further refinements in terms of planning—giving advices to each other about coping with these kind of situations. (2nd exercise) Thinking about the learnt skills and writing down the most important keywords and associations about the bystander effect. Close up, Homework.
5	Warm up, (1st exercise) Students think of someone (family member, friend) who can benefit from the mindset knowledge. First pair, then group discussion about what he would say to this person and imagination of the reactions. What is the most interesting to you regarding Mindset? (2nd exercise) Writing postcards to themselves with recommendations. Close up, Homework.	Warm up, (1st exercise) Students think of someone (family member, friend) who can benefit from the bystander effect knowledge. Write it down and some students share their "message" with the class. (2nd exercise) Summarizing the main points of helping others. (3rd exercise) Gathering what we have to take into consideration when we help others. Close up, Homework.

by conducting pre-post-post2 comparisons on each mindset measure (intelligence and personality) among participants who participated or did not participate in the mindset intervention. Second, we tested whether the intervention changed the academic motivations and the GPA scores by conducting pre-post-post2 comparisons on the academic motivation measure (amotivation, intrinsic and extrinsic motivation) and GPA scores among participants who participated or did not participate in the mindset intervention.

Finally, we created groups based on the Grit scores (high Grit and low Grit groups) to test whether the initial extent of persistence could modify the results of the intervention. We performed 2× 2 × 3 mixed model analyses of variance (ANOVA) with CONDITON (Growth Mindset intervention vs. control) and Grit (high or low Grit scores) as between-subjects factors, and TIME (pre-intervention, post-intervention and post-intervention 2) as a within-subjects factor. We performed the same analyses as mentioned above with adding the Grit variable.

RESULTS

Statistical analyses were performed using SPSS 22. Means and standard deviations are provided in **Table 2**. The normality of the data was investigated with multiple indicators. First, the Shapiro-Wilk test indicated that the data was statistically non-normal in the case of all measurement point of amotivation ($p < 0.001$), in the case of intrinsic motivation post-test 1 ($p = 0.033$), in the case of extrinsic motivation pretest ($p = 0.006$), the second post-test ($p = 0.042$), and regarding IQ mindset post-test 1 ($p = 0.033$). Contrarily, Skewness and Kurtosis values were between ±1.5: Personality Mindset ($\text{Kurtosis}_{\text{pre}} = -0.24$; $\text{Kurtosis}_{\text{post1}} = -0.50$; $\text{Kurtosis}_{\text{post2}} = -0.16$; $\text{Skewness}_{\text{pre}} = 0.30$; $\text{Skewness}_{\text{post1=}}-0.30$; $\text{Skewness}_{\text{post2}} = 0.36$) and IQ Mindset ($\text{Kurtosis}_{\text{pre}} = -0.70$; $\text{Kurtosis}_{\text{post1}} = -0.37$; $\text{Kurtosis}_{\text{post2}} = -0.46$; $\text{Skewness}_{\text{pre}} = -0.19$; $\text{Skewness}_{\text{post1}} = -0.56$; $\text{Skewness}_{\text{post2}} = -0.26$) variables as well as for the intrinsic motivation ($\text{Kurtosis}_{\text{pre}} = -0.65$; $\text{Kurtosis}_{\text{post1}} = -0.29$; $\text{Kurtosis}_{\text{post2}} = -0.55$; $\text{Skewness}_{\text{pre}} = -0.10$; $\text{Skewness}_{\text{post1}} =$

TABLE 2 | Descriptive statistics of measures in relation to each target group.

Scale	Type of intervention	Mean (SD)			Observed range		
		Pre	Post	Post 2	Pre	Post	Post 2
Intelligence mindset	Intervention	6.98 (1.93)	7.39 (1.78)	6.70 (2.04)	2.88–10	2.88–9.8	2–9.5
	Control	7.32 (1.63)	5.99 (2.02)	5.77 (1.69)	4.5–10	1.88–9.38	2–8.63
Personality mindset	Intervention	5.20 (1.24)	6.13 (1.62)	5.30 (2.01)	3.33–8.41	3–8.33	2.63–8.75
	Control	5.05 (1.47)	4.33 (1.50)	4.62 (1.50)	2.42–8.17	1.08–6.75	1.83–9
Amotivation	Intervention	2.32 (1.50)	1.40 (0.82)	1.98 (1.22)	1–6	1–4	1–4.50
	Control	1.97 (1.38)	2.67 (1.77)	2.42 (1.57)	1–5.75	1–6.75	1–5.50
Intrinsic motivation	Intervention	3.92 (1.76)	4.29 (1.08)	3.95 (1.29)	1–7	2–6	2–6.67
	Control	4.34 (1.38)	4.44 (1.65)	4.39 (1.49)	2–7	1.33–7	1–7
Extrinsic motivation	Intervention	4.98 (1.46)	4.93 (1.27)	4.88 (1.30)	2.5–7	2–7	1.5–7
	Control	5.48 (1.20)	5.30 (1.16)	5.28 (1.26)	2.75–7	3–7	2–7
GPA	Intervention	3.90 (.82)	–	3.87 (.82)	2.29–5.00	–	2.41–5.00
	Control	4.15 (.82)	–	4.19 (.77)	2.53–5.00	–	2.80–5.00

-0.52; $\text{Skewness}_{\text{post2}} = 0.14$), extrinsic motivation ($\text{Kurtosis}_{\text{pre}} = -1.03$; $\text{Kurtosis}_{\text{post1}} = -0.42$; $\text{Kurtosis}_{\text{post2}} = 0.09$; $\text{Skewness}_{\text{pre}} = -0.34$; $\text{Skewness}_{\text{post1}} = -0.23$; $\text{Skewness}_{\text{post2}} = -0.62$) and amotivation ($\text{Kurtosis}_{\text{pre}} = 0.47$; $\text{Kurtosis}_{\text{post1}} = 1.23$; $\text{Kurtosis}_{\text{post2}} = -0.74$; $\text{Skewness}_{\text{pre}} = 1.22$; $\text{Skewness}_{\text{post1}} = 1.43$; $\text{Skewness}_{\text{post2}} = 0.82$), and Grit ($\text{Kurtosis}_{\text{pre}} = -0.05$; $\text{Kurtosis}_{\text{post1}} = -0.33$; $\text{Kurtosis}_{\text{post2}} = -0.51$; $\text{Skewness}_{\text{pre}} = -0.21$; $\text{Skewness}_{\text{post1}} = -0.20$; $\text{Skewness}_{\text{post2}} = 0.12$). These Kurtosis and Skewness values are within the absolute values recommended by Curran et al. (1996) as they suggested a value of 2.0 for skewness and 7.0 for kurtosis. However, considering the results of the Shapiro-Wilk test, log10 transformation was applied for amotivation, intrinsic motivation and IQ Mindset scores.

Effectiveness of the Mindset Intervention

The CONDITION * TIME ANOVA predicting the beliefs of the changeability of intelligence revealed significant main effects of TIME, $F_{(2, 106)} = 4.34$, $p = 0.023$, $\eta_p{}^2 = 0.08$, power = 0.70; however no significant main effect of CONDITION, $F_{(1, 53)} = 2.52$, $p = 0.118$, $\eta_p{}^2 = 0.05$, power = 0.34 was found. The interaction of CONDITION * TIME was significant, $F_{(2, 106)} = 4.17$, $p = 0.027$, $\eta_p{}^2 = 0.07$, power = 65. Although intelligence mindset scores did not significantly differ between the intervention and control groups at baseline ($p = 0.381$), intelligence mindset scores differed significantly in the post-test among groups ($p = 0.015$), however, the scores did not differ significantly between the second post-tests ($p = 0.188$) (see **Figure 1A**).

Pairwise comparisons revealed that students who participated in the intervention group did not report significantly higher intelligence mindset scores between the pre- and post-test ($p = 0.383$), while significant difference was found between the post- and the second post-test scores ($p = 0.005$), there was no significant difference between the pre- and the second post-test ($p = 0.526$). While students in the control group showed significantly lower intelligence mindset scores between pre- and post-test ($p = 0.010$) and between pre- and second post-test ($p = 0.004$), while no significant difference was found between the post- and the second post-test scores ($p = 0.847$).

The CONDITION * TIME ANOVA predicting the beliefs of the changeability of personality did not reveal significant main effects of TIME, $F_{(2, 106)} = 0.58$, $p = 0.54$, $\eta_p{}^2 = 0.01$, power = 0.14; however, main effect of CONDITION, $F_{(1, 53)} = 8.11$, $p = 0.006$, $\eta_p{}^2 = 0.13$, power = 0.80 was found. The interaction of CONDITION * TIME was significant, $F_{(2, 106)} = 5.47$, $p = 0.006$, $\eta_p{}^2 = 0.09$, power = 0.80. Although personality mindset scores did not significantly differ between the intervention and control groups at baseline ($p = 0.70$), personality mindset scores differed in the post-test among groups ($p < 0.001$), however in the second post-test again, there was no significant difference between the intervention and the control group ($p = 0.164$) (see **Figure 1B**).

Pairwise comparisons revealed that students who participated in the intervention group reported significantly higher personality mindset scores between the pre- and post-test ($p = 0.023$) and significant difference was found between the post- and the second post-test scores ($p = 0.017$), while there was no significant difference between the pre- and the second post-test ($p = 0.80$). Students in the control group showed no significant differences between pre- and post-test ($p = 0.054$), between pre- and second post-test ($p = 0.33$), and between the post- and the second post-test scores ($p = 0.25$).

Effectiveness of the Mindset Intervention in Academic Motivations and GPA scores

In the academic amotivation scores the analysis did not reveal significant main effects of TIME, $F_{(2, 106)} = 0.52$, $p = 0.543$, $\eta_p{}^2$

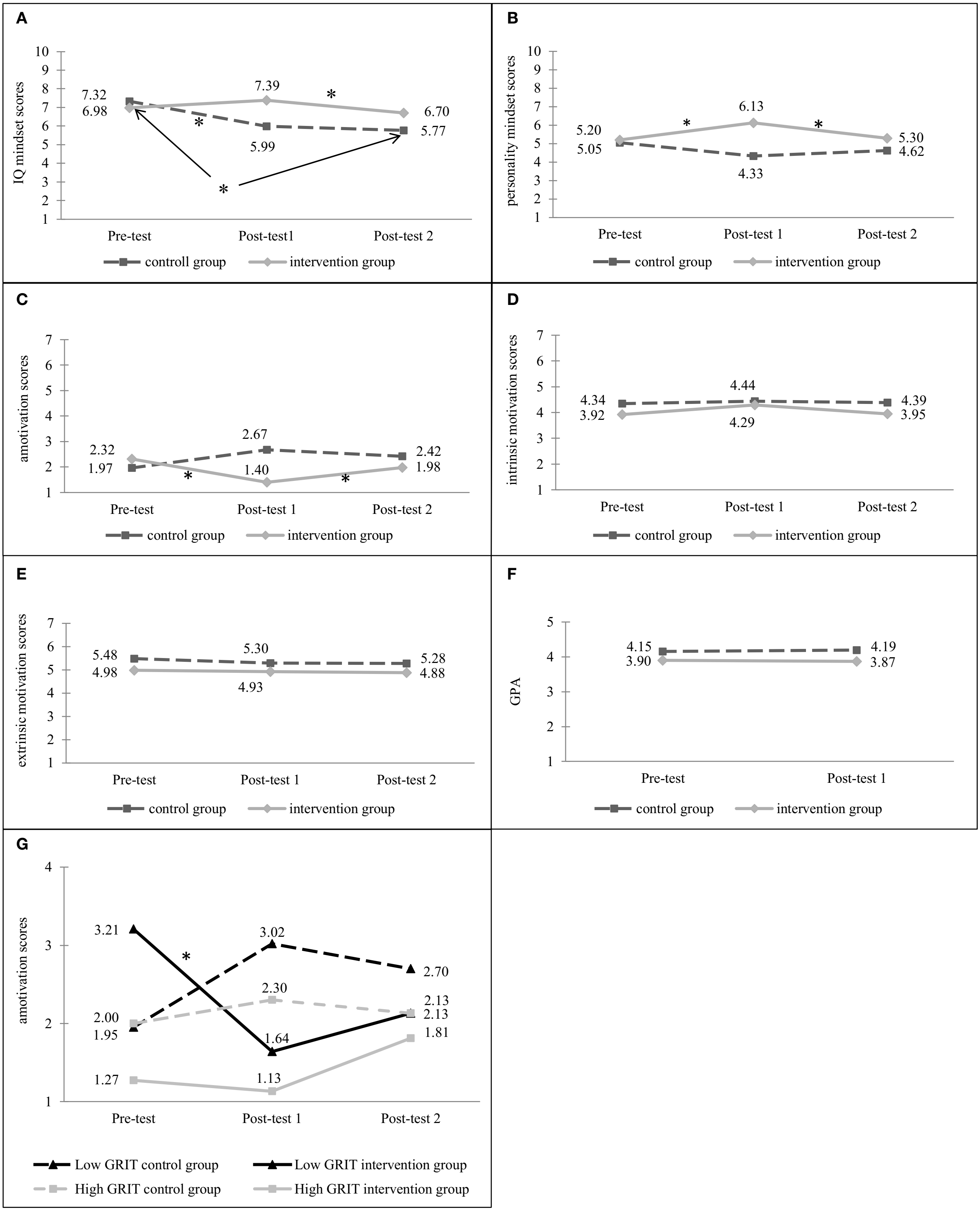

FIGURE 1 | (A) IQ mindset scores in the intervention and control group. **(B)** Personality mindset scores in the intervention and control group. **(C)** Amotivation scores in the intervention and control group. **(D)** Intrinsic motivation scores in the intervention and control group. **(E)** Extrinsic motivation scores in the intervention and control group. **(F)** GPA scores in the intervention and control group. **(G)** Amotivation scores in High and Low GRIT intervention and control groups. $^*p < 0.05$.

$= 0.01$, power $= 0.12$ and CONDITION, $F_{(1, 53)} = 2.32$, $p = 0.134$, $\eta_p{}^2 = 0.04$, power $= 0.32$. The interaction of CONDITION * TIME was significant, $F_{(2,106)} = 6.19$, $p = 0.007$, $\eta_p{}^2 = 0.10$, power $= 80$. Although academic amotivation scores did not significantly differ between the intervention and control groups at baseline ($p = 0.341$), amotivation scores differed in the post-test among groups ($p = 0.001$), however, in the second post-test again, there was no significant difference between the intervention and the control group ($p = 0.358$) (see **Figure 1C**).

Pairwise comparisons revealed that students who participated in the intervention group reported significantly lower amotivation scores between the pre- and post-test ($p = 0.005$) and significant difference was found between the post- and the second post-test scores ($p = 0.006$), while there was no significant difference between the pre- and the second post-test ($p = 0.446$). Students in the control group showed no significant differences between pre- and post-test ($p = 0.089$), between pre- and second post-test ($p = 0.275$), and between the post- and second post-test scores ($p = 0.300$).

In the intrinsic motivation scores the analysis did not reveal significant main effects of TIME, $F_{(2, 106)} = 0.63$, $p = 0.494$, $\eta_p{}^2 = 0.01$, power $= 0.14$ and CONDITION, $F_{(1, 53)} = 1.19$, $p = 0.280$, $\eta_p{}^2 = 0.02$, power $= 0.19$. Also, the interaction of CONDITION * TIME was not significant, $F_{(2, 106)} = 0.95$, $p = 0.368$, $\eta_p{}^2 = 0.02$, power $= 0.19$ (see **Figure 1D**).

In the case of extrinsic motivation, the analysis did not reveal significant main effects of TIME, $F_{(2, 106)} = 0.27$, $p = 0.719$, $\eta_p{}^2 = 0.01$, power $= 0.09$ and CONDITION, $F_{(1, 53)} = 2.91$, $p = 0.094$, $\eta_p{}^2 = 0.05$, power $= 0.39$. Also, the interaction of CONDITION * TIME was not significant, $F_{(2,106)} = 0.07$, $p = 0.903$, $\eta_p{}^2 < 0.001$, power $= 0.06$ (see **Figure 1E**).

In the GPA scores the analysis did not reveal significant main effects of TIME, $F(_{1, 72)} = 0.07$, $p = 0.79$, $\eta_p{}^2 = 0.00$, power $= 0.06$ and CONDITION, $F_{(1, 72)} = 2.35$, $p = 0.13$, $\eta_p{}^2 = 0.03$, power $= 0.33$. Also, the interaction of CONDITION * TIME was not significant, $F_{(1, 72)} = 2.60$, $p = 0.11$, $\eta_p{}^2 = 0.04$, power $= 0.36$ (see **Figure 1F**).

Effectiveness of the Mindset Intervention in Low and High GRIT Score Groups

We examined whether participants' level of persistence (high/low GRIT) would moderate the effectiveness of mindset interventions. Based on GRIT scores measuring the extent of persistence ($M = 28.15$; $SD = 5.30$), the sample was split into two groups (median split) to distinguish between participants who had lower level of persistence ($M < 28$) and those who had higher level of persistence ($M \geq 28$). We then conducted a 2 (CONDITION) * 3 (TIME) * 2 (GRIT: High/Low) ANOVA to predict the changes in mindset beliefs, in academic motivation and in GPA. There were 14 participants in the low GRIT intervention group, 12 in the high GRIT intervention group, 14 in the low GRIT control group and 15 in the high GRIT control group.

In the case of academic amotivation, beyond the effects for CONDITION and TIME reported above, this analysis revealed significant main effect for GRIT, $F_{(1, 51)} = 7.81$, $p = 0.007$, $\eta_p{}^2 = 0.13$, power $= 0.78$ (see **Figure 1G**). GRIT did not significantly interact with either TIME, $F_{(2, 102)} = 1.24$, $p = 0.27$, $\eta_p{}^2 = 0.02$, power $= 0.19$ or CONDITION, $F_{(1,51)} = 1.52$, $p = 0.22$, $\eta_p{}^2 = 0.03$, power $= 0.23$. However, the three-way interaction between CONDITION, TIME, and GRIT was significant, $F_{(2, 102)} = 3.51$, $p = 0.048$, $\eta_p{}^2 = 0.06$, power $= 0.56$.

Pairwise comparisons revealed that students who participated in the intervention group with lower persistence (low GRIT group) reported significantly lower amotivation scores between the pre- and post-test ($p = 0.009$), however, no significant difference was found between the post- and the second post-test scores ($p = 0.051$) and between the pre- and the second post-test ($p = 0.102$). Pairwise comparisons also revealed that students who participated in the intervention group with higher persistence (high GRIT group) reported no significantly lower amotivation scores between the pre- and post-test ($p = 0.075$), between the post- and the second post-test scores ($p = 0.065$), and between the pre- and the second post-test ($p = 0.185$). Moreover, pairwise comparisons revealed that students who participated in the control group with lower persistence (low GRIT group) did not report significantly higher amotivation scores between the pre- and post-test ($p = 0.096$), whereas the difference was not significant between the post- and the second post-test scores ($p = 0.311$), and between the pre- and the second post-test ($p = 0.297$). Pairwise comparisons revealed that students who participated in the control group with higher persistence (high GRIT group) reported no significantly higher amotivation scores between the pre- and post-test ($p = 0.568$), between the post- and the second post-test scores ($p = 0.770$), and between the pre- and the second post-test ($p = 0.688$). In the case of IQ mindset, personality mindset, intrinsic motivation, extrinsic motivation, and GPA-change, no effects of GRIT can be observed.

DISCUSSION

Compared to previous successful Growth Mindset interventions (Aronson et al., 2002; Good et al., 2003; Blackwell et al., 2007; Paunesku et al., 2015; Yeager et al., 2016), the present one did not achieve its goals in terms of belief-, motivation- and GPA-change. Regarding the short-term positive consequences, the present form of growth mindset intervention resulted in more incremental personality beliefs, and it sustained the initial level of the amotivation and IQ mindset scores. However, in the control group, the personality mindset scores stayed at the initial level, and students in this condition also reported more fixed intelligence mindset scores and higher level of amotivation. Therefore, the short-term effect of this growth mindset intervention kept IQ mindset high and amotivation low, while at the same time inducing more incremental personality beliefs. However, all of these effects disappeared by the end of the semester. Furthermore—according to a further analysis focusing the role of Grit as a more stable personality characteristic—we found that the short term drop of amotivation can be attributed to those students who are less persistent. Among the low Grit students who participated in the growth mindset treatment, we measured a short-term drop of amotivation, but this effect also disappeared by the end of the semester. Finally, the growth mindset intervention could not improve the GPA of the students. In sum, we found some short-term belief-related and motivational effects, but no long-term effects were sustained.

Some previous studies did not support the beneficial effect of the growth mindset interventions on motivation, academic performance beliefs, study skills, and math performance (Dommett et al., 2013). However, at least Dommett et al.'s (2013) study could demonstrate a small but significant long-term effect with the increased incremental intelligence beliefs. Donohoe et al.'s (2012) used a similar growth mindset intervention in an online setting and they had similarly short-term positive results in terms of resiliency and sense of mastery that disappeared after 3 months. Dommett et al.'s (2013) results, along with the present ones, show that mindset beliefs—in the field of personality and intelligence—are temporal malleable in given circumstances and they can return to their initial state in the long run.

One of the most interesting results is related to the question as to why growth mindset beliefs of the control group participants became more fixed over time. One of the potential explanation is related to the fluid aspect of the school context (Spitzer and Aronson, 2015). It is possible that when the end of semester approached, students experienced several situations—criticism, failures, setbacks—which made them doubt their abilities. Furthermore, it is also possible that their grades and the feedbacks of their teachers did not change in the expected way or to the expected extent which in turn had a negative effect on their growth mindset beliefs.

As researchers with less experience in social psychological interventions, first, we interpret these results in the light of the guidelines of more experienced experts of this field. Second, we aim to share our experience with this specific intervention, hoping that it might be valuable for similarly less experienced researchers.

Garcia and Cohen (2011) used the interpretation of Lewin (1948, 1951) concerning the classroom which is a relatively stable tension system including dynamic forces of interaction. Social psychological interventions can modify this tension system in a successful way if they take into consideration core elements identified by previous research (Garcia and Cohen, 2011; Yeager and Walton, 2011; Walton, 2014). These seven core elements—psychological precision, targeting specific groups, appropriate timing, creating recursive processes, embedding in the appropriate context, avoiding persuasive appeals and framing as learning opportunity—are depicted in **Table 3** in the light of the present study design. The present results suggest that an intervention can be a holistic context in which changing one parameter can lead to different results.

Psychological Precision

The present intervention was based on the Mindset theory of Dweck et al. (1995). Although this training focused on the analogy of the plasticity of the brain to some extent, it was not as emphasized as in the case of both previous online (Paunesku et al., 2015) and offline Mindset interventions (Blackwell et al., 2007). We rather focused on more applied aspects of growth mindset, such as what students can do with potential obstacles in difficult situations (see **Table 1**, second session) as well as we demonstrated the internal dialog between growth and fixed mindset in order to provide practical guidelines for using growth mindset in different ways in diverse situations (see **Table 1**, third session). The lack of efficacy of the intervention can be attributed to the lack of such analogy which can be easily objectified by the students. Besides this alternation, the intervention was based on solid theoretical background and intended to change only one specific belief. In sum, besides the solid theoretical background, recurring, very illustrative analogies—such as the brain-muscle one in the original Growth Mindset interventions—can also contribute to the effectiveness.

Targeting Specific Groups

In most of the Mindset interventions, specific target groups were low achievers (Blackwell et al., 2007; Paunesku et al., 2015), low income students (Good et al., 2003) or minority students (Aronson et al., 2002). In these studies, the effect of the intervention was visibly more salient than among other experimental group members. In the present case, students with relatively good grades with no minority background were in the focus of investigation. Among these students, the training was effective in short-term regarding their amotivation, whereas this improvement diminished by the end of the semester.

In the present case, neither extrinsic nor intrinsic motivation changed in long-term, similarly to GPA. Very probably, in the case of these students, measuring challenge-taking might be more beneficial. These students were possibly less exposed to failures than students with worse grades. However, in this explanation, we cannot ignore the importance of subjective experience of failure. Further investigations are needed to support that students with good grade are less exposed to failures. It is possible to expect that students with good grades in the Hungarian educational context have comparable results to underprivileged students in the US. However, our results did not support this assumption.

Appropriate Timing

The third point refers to the appropriate timing. In the present case, it was not at the beginning of the high school (9th grade), but at the beginning of the second semester of the 10th grade. According to both Yeager and Walton (2011) and Garcia and Cohen (2011), interventions in the transition points are more effective. We assume that the stressful nature of the transition context is important because it can allow context-dependent learning. If a well-targeted intervention (beliefs in change through effort) can help the student with immediate positive feedbacks in these transitional circumstances (i.e., positive feedbacks such as less failure in longer term, better grades attributed to more effort or better strategy, etc.), then the recently learnt strategies can be reinforced and more probably used in further stressful situations. In the present case, coping routines related to high school context had already stabilized. Supposedly, changing these already established attributions might need more in-depth and lengthier interventions. In sum, in the present case, the timing was sub-optimal. Future interventions should focus more on the appropriate timing.

TABLE 3 | Comparing the requirements of social psychological interventions (Garcia and Cohen, 2011; Yeager and Walton, 2011; Walton, 2014) with the elements of the present study.

Requirements of a good social psychological intervention	How it appeared in the present study
1. Psychologically precise (theory and tools)	It used the Mindset theory and valid measurements
2. Targets a specific group	High achievers were targeted
3. Appropriate timing	Second semester of second year in high school
4. Recursive processes	Weekly, 1-h session for 5 weeks
5. Appropriate context	Classroom context in which the form master conducted the intervention and each student participated in the sessions
6. Not using direct persuasive appeal	Students shared their experiences with each other, not direct, one-way lecture was given about the subject
7. Not help, but give an opportunity	The training was framed as a learning opportunity

Creating Recursive Processes

The fourth element refers to recursive processes. Both one session (Paunesku et al., 2015), two-session (Yeager et al., 2016) and multiple session (Blackwell et al., 2007) interventions can be effective. The recursive nature of the intervention can appear through the number of direct interventions and the processes catalyzed by previous session. Obviously, it is more economic if one session can catalyze further long-term recursive processes which leads to far-reaching positive consequences. In the present case, compared to Blackwell et al.'s (2007) eight sessions, we had fewer sessions (five) with less results. There are many reasons why it might have been less effective.

Paradoxically, longer interventions do not help in reinforcing the recursive processes, because they can provide "too many" information, allowing room for diverse interpretations. For example, in the present case, thanks to the numerous activities, some students might put more emphasis on the role of efforts, while others might put more emphasis on choosing the appropriate problem-solving strategies. Therefore, as a consequence of this biased representation of Growth Mindset, their effort beliefs can become rather different. This belief bias might influence further catalyzing processes, because resolving a problem attributed to efforts vs. careful strategy choice can reinforce more efforts and less experimenting with newer strategies. The more components of the Growth Mindset are in the focus of a longer intervention, the more alternative (mis)interpretation and bias can be expected. According to Dweck (2016), such misinterpretations can lead to a false growth mindset, including elements such as effort is more important than asking help from others or trying out new strategies. In order to identify the specific effect of the intervention, differentiated measurement of these belief-facets (i.e., efforts, strategies, asking help from others) might be beneficial in future studies. Possibly, physical tools—such as posters in the classrooms depicting infographics on the appropriate content of the Growth Mindset—could be an adequate reminder for this purpose. In sum, in the case of social psychological interventions, less can be more.

Embedding in the Appropriate Context

The fifth element refers to the appropriate context. In the present case, the teachers started to implement the mindset module exercises in their school context. The training neither aimed at changing the school climate, nor directly changing the teacher's teaching practices; it only provided knowledge about Mindset theory through the above mentioned exercises. Naturally, we did not intend to deeply change the school context in itself. Teachers—who volunteered for participating in this study and who were randomly assigned to groups—could have teaching practices which reflect on fixed mindset and growth mindset as well. Unfortunately, teachers' initial mindset beliefs were not assessed in the present study. However, these initial beliefs can be critical in terms of teaching practices and how they conduct the mindset intervention in the classroom.

Another important issue is that we supposed that a 4-h long training can change the teachers' intelligence beliefs toward the growth mindset which can be transmitted through the exercises described above. Many factors can influence this context: (a) students' attitudes toward the teacher who led the training, (b) how much growth mindset appears the given teacher's educational practices which can validate the message of the training, (c) the attitude of the other teachers at the school concerning the notions of the growth mindset, (d) the presence or lack of standardized testing in terms of grading, (e) peer norms concerning Growth Mindset, etc. As far as we know, only a few studies examined these factors in details in the light of the effectiveness of the interventions. Future studies are needed in order to identify the relative importance of these (and other) contextual factors.

Avoiding Persuasive Appeals and Framing as Learning Opportunity

Concerning the last two characteristics of wise social psychological interventions—the avoidance of using persuasive appeals and not framing the intervention as helping but as an opportunity—the present Mindset training met these requirements (Yeager and Walton, 2011; Walton, 2014). The exercises summarized in **Table 1** are neither directly persuasive, nor framed as helping.

One might think that a student (or an adult) can hold growth mindset irrespectively to the environmental context or feedback. Despite the weaknesses of the present study, it became evident that Mindset beliefs can temporarily change in a beneficial direction, but in the long run, they can change back to their pre-intervention level. Further research is needed to explore the interactions between the relevant social situations and the temporal changes on the continuum of fixed and growth mindset beliefs. However, it appears that the present train-the-trainer intervention did not lead to a long term positive mindset change. In sum, Mindset beliefs are malleable and not only in the good direction. Future research is needed to explore which contextual variables can change back an intervention-induced growth mindset to the baseline.

The Role of Grit in Growth Mindset Interventions among Students with High GPA

Despite the similar theoretical roots of mindset (Hong et al., 1999) and Grit (Duckworth et al., 2007) theories in terms of persistence and effort, as far as we know, no previous research was conducted to measure the link between them in an intervention study. Despite low Grit students' intrinsic, extrinsic motivations and grades did not differ, their amotivation decreased temporarily more than amotivation of high Grit students. Therefore, we assume that high-Grit good-grader students' amotivation basically stayed on a stable low level. On the other hand, those good-graders who were low on Grit measure felt less that they were wasting their time at school after 3 weeks of the intervention than before. This effect was quite large: the averages of the amotivation scores dropped by almost 40% by the first post-test. If this level of amotivation could be stabilized among low Grit good-graders, then these students could benefit a lot from such interventions. Unfortunately, according to the results, this effect also disappeared by the end of the semester. Furthermore, it is important to note that the pre-test amotivation scores of the high-grit and low-grit intervention group were different which may also bias the results.

Limitations And Future Directions

One of the merits of the present study that it opens several questions; however, besides the above-mentioned principle-based issues, we can mention more specific methodological mistakes and limitations. These can also be instructive to those—similarly to the first four authors of the present study—who start to implement similar social psychological interventions in educational context. First, we found significant changes only in the case of self-reporting; further studies should use similar behavioral measures as the Yeager et al. (2014, 2016) studies used. Furthermore, it would be important to measure the perceived change reported by teachers, parents and peers (as norms how the class can accept failures). Second, high drop-out rate could be avoided by online data gathering (instead of paper and pencil solutions) and using the participants' name instead of code-names. Third, it might be a fruitful way to give more in-depth trainings to the teachers instead of 4-h trainings and measure how much their teaching practices changed. We might think that their beliefs changed, but this could not manifest in their instructional behavior. Supposedly, teachers who provide such knowledge via trainings should be trained to the implementation of these beliefs in her/his instructional practices. Furthermore, they might be trained to recognize the signs (of communication or behavior) when they teach with fixed mindset. Fourth, future interventions should choose randomization of students instead of teachers or classrooms, similarly to Paunesku et al. (2015). Fifth, we assume that a good intervention can be implemented not only in the case of students, but to the parents and teachers as well. This multidirectional intervention could create a supportive social context in which growth mindset can be catalyzed in a more efficient way. Sixth, the sample size hand in hand with the power were low, therefore it is important to have a larger initial sample size and in the present case be cautious regarding even with the short term results.

CONCLUSION

The goal of the present study was to assess the effectiveness of a train-the-trainer mindset intervention among Hungarian 10th grade students with high GPA. Teachers led the intervention after a 4-h long training which highlighted the most important aspects of Hong et al. (1999) mindset theory. The results suggest that growth beliefs of personality changed in a positive way (i.e., students had more growth mindset) 3 weeks after the intervention. Growth mindset IQ scores were sustained as a result of the intervention, whereas we found a significant drop of this measure in the control group after 3 weeks. Furthermore, the amotivation score of the intervention group reduced compared to both the baseline measure and the control group. However, no intrinsic and extrinsic motivational change was measured. Students with good grades and low Grit scores reported lower amotivation compared to grittier high achievers in the first post test. However, by the end of the semester, these positive changes disappeared. Furthermore, their GPA did not improve as a result of the intervention. These results show that mindset beliefs can change back to their pre-intervention state. Several possible explanations exist as to why this intervention was not successful in the long run; therefore, future research is needed to explore the boundaries of the effectiveness of growth mindset interventions.

AUTHOR CONTRIBUTIONS

GO designed the study, analyzed the data, interpreted the data and drafted the manuscript. SP designed the study, collected data, and drafted the manuscript. BB analyzed the data, drafted the manuscript and interpreted the data. IT analyzed the data, drafted the manuscript and interpreted the data. RB designed the study, interpreted the data and drafted the manuscript. All authors commented on the draft and contributed to the final version, approved the publication of the manuscript, and agreed to be accountable for all aspects of the work.

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Effectiveness of the "What's Up!" Intervention to Reduce Stigma and Psychometric Properties of the Youth Program Questionnaire (YPQ): Results from a Cluster Non-randomized Controlled Trial Conducted in Catalan High Schools

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Mental disorders are highly prevalent in the general population, and people who experience them are frequently stigmatized. Stigma has a very negative impact on social, academic/professional, and personal life. Considering the high rates of mental disorders among children and adolescents (13.4%) and how critical this age is in the formation of nuclear beliefs, many campaigns to combat stigma have been developed in the last decade, with mixed results. The OBERTAMENT initiative has produced various anti-stigma campaigns in Catalonia (Spain). In the present study, the main objective was to report on the effectiveness of the OBERTAMENT "*What's up!*" intervention, a curricular intervention including education and social contact conducted by the teachers in the classroom with teenagers aged between 14 and 18. Prior to this, we examined the psychometric properties of the Youth Program Questionnaire (YPQ), our main outcome measure, in terms of dimensionality, reliability, and validity. A cluster non-randomized controlled trial was conducted to assess this intervention, which was tested in nine high schools situated in the Barcelona region. A convenience sample of 261 students formed the intervention group and 132 the control group (52% women, mean age = 14, *SD* = 0.47). The assignment to study conditions was conducted by Departament d'Ensenyament (Department of Education), Generalitat de Catalunya (Catalan Government). Participants were evaluated at baseline, post-intervention, and 9-month follow-up. The main outcome measure of this study was the YPQ. The Reported and Intended Behavior Scale (RIBS) was used as secondary outcome measure. The statistical analysis indicated that the YPQ possesses a two-factor structure (stereotypical attitudes and intended behavior) and sound psychometric properties. The multilevel mixed-effects models revealed statistically significant interactions for both study measures and *post hoc* intragroup analyses revealed a significant but small improvement in the YPQ and RIBS scores in the intervention group. Overall, our results indicate that "*What's up!*" produced statistically significant, albeit small improvements in stereotypical attributions and intended behavior toward people with mental disorders. Some methodological limitations and the relatively low levels of stigma observed in our sample may undermine our results. The implications of our results are discussed in relation to stigma research.

Keywords: stigma, mental disorders, non-randomized controlled trial, Youth Program Questionnaire, Reported and Intended Behavior Scale

INTRODUCTION

Mental disorders have a high prevalence in the general population. According to the most recent evidence, 38.2% of the general European population experiences at least one mental disorder, which corresponds to an estimated 164.7 million people (Wittchen et al., 2011). In Spain, the European Study of the Epidemiology of Mental Disorders (ESEMeD; Haro et al., 2003) concluded that approximately one out of every five people who were surveyed had presented a mental disorder at some point of their life, and 8.5% of the population had experienced a mental disorder in the previous year. Moreover, children and teenagers are a high-risk population since a worldwide prevalence of 13.4% for any mental disorder has been recently reported (Polanczyk et al., 2015).

It is noteworthy, that people experiencing mental disorders frequently deal with stigma associated with their condition (Hugo et al., 2003; Ando et al., 2013; Loch et al., 2014). Stigma is a multidimensional phenomenon that includes cognitive and behavioral aspects (Corrigan and Shapiro, 2010). The first one is, in turn, formed out of two constructs: *stereotypes*, which are related to knowledge (Thornicroft et al., 2007); and *prejudices*, which are the generalized attitudes toward members of a social group. These cognitive aspects of stigma begin to form and consolidate during adolescence (Flavell et al., 2002; Schulze et al., 2003) and their consequence is *discrimination*, the behavioral aspect of stigma. In the case of mental disorders, a common example would be an employer who, based on the belief that people with a mental disorder are violent (stereotype), has a negative feeling -fear, anxiety- toward them (prejudice) and, thus, decides not to hire a person who experiences one (discrimination) (Crespo et al., 2008; Ke et al., 2015).

Stigma has many consequences: it contributes to low self-esteem and quality of life (Livingston and Boyd, 2010) and has a negative effect on adequate housing, work and financial status (Rüsch et al., 2005; Sharac et al., 2010); from a clinical perspective, it has a negative influence on symptom severity and compliance with treatment (Li et al., 2014) and may trigger suicidal ideation and behavior (Rüsch et al., 2014). When stigma is internalized (i.e., self-stigma), it is associated with rejection of help, avoidance of treatment and limited prospects of recovery, among other damaging consequences (Hing and Russell, 2017). These are some reasons why stigma has been identified as one of the greatest challenges facing mental health (Hing and Russell, 2017).

In Spain, the level of stigma toward mental disorders is in line with or slightly lower than in other European countries, with mean Reported and Intended Behavior Scale (RIBS; Evans-Lacko et al., 2011) score corresponding to the 76th percentile (Aznar-Lou et al., 2016), which indicates relatively low levels of stigma. However, this does not mean that the Spanish population should not be subjects of anti-stigma interventions as, for instance, knowledge of mental disorders has been found to be low. A study conducted by López-Ibor concluded that 83% of the surveyed Spanish population knew nothing about schizophrenia and 33% did not know about the origin and causes of the disorder even though 44% affirmed that schizophrenia is not a curable illness. In other countries, such as the United Kingdom, it was reported that the general population, and youngsters particularly, had many misconceptions about mental disorders (Crisp et al., 2000). Similarly, Crespo et al. (2008) reported a moderate degree of knowledge about the treatment of mental disorders, the work options for people who experience them, and the causes and degree of awareness of the disorder in a general population sample.

Nonetheless, knowledge about mental disorders is not the only aspect to be improved regarding stigma, as there is evidence that public attitudes have not changed over the last two decades, or have even become worse regarding people with psychotic disorders (Hansson and Markström, 2014). Some studies have identified several personal characteristics that are associated with having a higher degree of stigma toward people who experience mental disorders, such as being male (Stickney et al., 2012), having had no previous contact with people with mental conditions (Batterham et al., 2013), lower educational levels and living alone (Coppens et al., 2013). In Spain, some factors such as having completed secondary or university education and having had contact with people with mental problems are related to a better attitude toward mental disorders and more favorable intended behavior (Aznar-Lou et al., 2016).

Bearing this in mind, various approaches have attempted to reduce stigma toward mental disorders through education and social contact. Corrigan and Penn (1999) considered educational interventions to be one of the most effective ways of reducing stigma. Educational approaches often challenge inaccurate stereotypes of mental disorders, replacing them with factual information (Corrigan and Shapiro, 2010; Economou et al., 2012). These interventions are frequently addressed to children and teenagers as they constitute one of the most indicated populations for many reasons: studies have proved that children do not yet have a consolidated idea of what mental illness denotes (Corrigan and Watson, 2007), and the personality traits that constitute the foundation for stereotype endorsement are not well-entrenched until adolescence (Flavell et al., 2002), considering that this stage lasts, approximately, from 13 to 18 years old. Besides, as previously mentioned, adolescents form a population which is severely affected by mental disorders, in part due to the great challenges that this life stage entails (e.g., identity definition, sexual role and body changes, academic goals).

Therefore, interventions targeted at these age groups seem to be especially useful, not only with preventive aims but also to combat the stigma that children and adolescents already begin to experience and that frequently damages their social and academic development. Some countries, such as England, have prioritized the topic of improving mental health knowledge and reducing stigma in schools and colleges. In this case, the Public Health England and the Children & Young People's Mental Health Coalition produced the Promoting Children and Young People's Emotional Health and Wellbeing (2015) guide, with key evidence-based guidelines for teachers and college principals to create a whole-school approach to promoting emotional health and well-being. This approach does not simply involve a workshop on mental health, but a series of regular practices to be carried out in every school space. By using this model, it may be possible to overcome the limitations of the short-duration educational interventions that have been commonly used until date, which have generally been related to small and short-term improvements in attitudes toward mental disorders in adolescents (Bulanda et al., 2014). These interventions' effects have been reported to last over 1–6 months (Pinfold et al., 2003; Schulze et al., 2003), and regarding its content, it has been assessed that adding social contact can either imply lower effects for adolescents (Corrigan et al., 2012) or no difference at all in comparison to education alone (Schulze et al., 2003; Warner, 2005; Yamaguchi et al., 2011).

The evaluation of the effectiveness of educational interventions is generally carried out by self-reported measures that evaluate both the cognitive and behavioral components of stigma. Many different measures have been developed, but most present some methodological issues which question their adequacy, and there is no consensus on which one to use (Sakellari et al., 2011). Furthermore, validations of stigma measures oriented to teenagers are scarce worldwide (Ochoa et al., 2015). In this regard, the Youth Program Questionnaire (YPQ), an instrument developed for the campaign "*Opening Minds*" which was conducted in Canada in 2013 (Stuart et al., 2014), is considered to be a good instrument as it measures both the cognitive and behavioral aspects of stigma and is addressed specifically to adolescents. Nonetheless, its psychometric properties have yet to be established.

In Catalonia, the OBERTAMENT initiative, created in 2010, has developed various campaigns to fight stigma associated with mental disorders. The present study has the main objective of evaluating the effectiveness of "*What's up!*" on reducing perceived stigma in a Catalan sample of adolescent students using the self-report YPQ as primary outcome and the RIBS (Evans-Lacko et al., 2011) as a secondary outcome. "*What's up!*" is a curricular intervention which includes both education and social contact, created by the OBERTAMENT initiative and addressed to 14–18 year-old Catalan students to reduce stigma levels. Since the YPQ has not yet been validated, its psychometric properties in our Catalan adolescent sample are also reported.

MATERIALS AND METHODS

Study Design

This is a cluster non-randomized controlled trial designed to assess the intervention "*What's up!*" in nine high schools in the region of Barcelona (Spain) with pre–post and follow up evaluations.

This study was performed in accordance with ethical standards established in the 1964 Declaration of Helsinki and its subsequent updates and established in the World Psychiatric Association Declaration of Madrid. The study protocol was approved by the ethics committee at the Sant Joan de Déu Foundation (CEIC PIC-107-14; Esplugues de Llobregat, Spain). We report this non-randomized controlled trial following Transparent Reporting of Evaluations with Non-randomized Designs guidelines (TREND; Des Jarlais et al., 2004).

Participants

A total of 446 students aged between 14 and 18 years were recruited from nine different high schools: five (Pere Vives i Vich, Montbui, Salvador Claramunt, Escola Pia, and Maristes) were assigned to the intervention group, and four (Pla de les Moreres, Escola Anoia, IES Joan Mercader, and IES Guinovarda) to the control group. There were no other inclusion/exclusion criteria for participation in this study. All centers were located in the Barcelona region. These high schools were selected by convenience by the Catalan Government (Generalitat de Catalunya) Department of Education. This public entity performed the allocation to study conditions and matched high schools considering the number of students and whether they were private (Escola Pia, Maristes and Escola Anoia) or public centers (Pere Vives i Vich, Montbui, Salvador Claramunt, Pla de les Moreres, IES Joan Mercader and IES Guinovarda). All nine high schools are coeducational and have a maximum of 25 students per class.

Intervention

The "*What's up!*" intervention consists of a multifocal action to combat stigma from the classroom. The teachers were given a manual with nine didactic units which contained information, examples, and exercises about mental health problems, and they were in charge of using these units in their classes, with the freedom to choose how and when to do so. This intervention model was conceived to make "*What's up!*" truly implementable in the real context of high schools. It was indicated that every unit had to be implemented for 1 week. The materials that were selected for each unit were chosen by the OBERTAMENT team and based on evidence from previous effective interventions. The protocol explained in detail different types of exercises that the students should do both individually and in groups for every didactic unit. The teachers were given instructions to employ at least three of the nine didactic units, with the first and last considered to be "essential."

The nine didactic units corresponded to different school subjects: Language (Catalan and Spanish), Foreign Language (English and French), Sciences (Biology and Geology), Mathematics, Physical Education, and Culture and Ethical Values. It was in this last unit that a person with a mental disorder reported firsthand experiences to the students in the classroom, providing the social contact component of the intervention. **Table 1** shows which units were applied in each high school.

Although the teachers did not receive any specific training from OBERTAMENT on how to provide support on mental health disorders, general knowledge about what mental disorders are and how to manage classrooms with heterogeneous needs are part of every teacher's regular academic training.

Further information on this project is available here (in Spanish): https://obertament.org/es/educacion/proyecto-what-s-up.

Procedure

After an initial meeting with the education department and all the heads of the high schools, the "*What's up!*" project team presented the curricular project protocol and the study goals to the teachers who would implement it.

Data from study participants were obtained at three different times: before the intervention, after the intervention (1–3 months, depending on the high school), and at follow-up 9 months from study commencement. Students were aware of their participation in a study but they did not know to which condition their high school was allocated. However, teachers administering the interventions and those assessing outcomes were not blind to study allocation. All three evaluations were conducted in the participants' classrooms using a paper and pencil self-administered questionnaire. As they were not adults, informed consent was signed by their parents or legal guardians prior to the first evaluation. The same version of the battery of measures was administered in all high schools.

The YPQ was translated from English into Catalan by two native bilingual English/Catalan speakers. Any discrepancies between the Catalan and English versions were resolved by agreement.

Measures

To evaluate previous contact and experience with people with a mental disorder, four ad hoc questions in dichotomous format (yes/no) were included, along with a further question regarding personal experiences of having a mental disorder.

- The main outcome measure was the YPQ adapted to Catalan, an instrument developed by the Canadian initiative *"Opening minds"* (Stuart et al., 2014) which has two 11-item scales; the Stereotype Scale (YPQ-SS) which measures stereotypic attributions (controllability of the illness, potential for recovery, and potential for violence and unpredictability) and the Social Acceptance Scale (YPQ-SAS) which measures behavioral intentions related to social acceptance. The YPQ is rated on a five-point Likert scale (from 1 to 5) where lower scores indicate lower levels of stigma toward people with a mental disorder. Items 14, 16, 19, 20, 21, and 22 are reverse scored. This study uses the total mean and subscales scores. The psychometric characteristics of this questionnaire are described below. Two exemplary items of this questionnaire are "People with mental illnesses need to be locked away" (YPQ-SS) and "If I knew someone had a mental illness I would not date them" (YPQ-SAS). All the items can be found in the table added as Supplementary Material.
- The Catalan version of the RIBS (Evans-Lacko et al., 2011) was also used to measure intended behavior in relation to future contact with people with a mental disorder (intention to live with, work with, live nearby and continue a relationship with someone with a mental disorder). The four items are rated on a five-point Likert scale from 1 (strongly agree) to 5 (strongly disagree) so that lower scores indicate more favorable intended behavior. In the present study, we use the mean item score. In this project, internal consistency (Cronbach's α) of RIBS was 0.81, indicating adequate reliability ($\alpha \geq 0.8$ is defined as "good"; Nunnally, 1978). An example of an item in this scale is "In the future,

TABLE 1 | Units of the curricular intervention and their implementation in every high school.

Unit	Escola Pia ($n = 90$)	Institut Montbuí ($n = 54$)	Maristes Igualada ($n = 54$)	Escola Pere Vives ($n = 24$)	Salvador Claramunt ($n = 57$)
The standup kid (English)	One week	One week	One week	One week	One week
I'm Michael (English)	One week	–	–	–	One week
Donne lui la parole (French)	–	One week	One week	–	–
What do you know? (Catalan)	One week	–	One week	Partially implemented	One week
Health dimension (Biology)	One week	–	One week	One week	One week
What do we now about health? (Maths)	One week	One week	One week	One week	One week
Activate your well-being (P.E.)	One week	One week	Partially implemented	–	–
Let's fight stigma (Culture and Ethic Values)	One week	One week	One week	One week	One week
Testimonials (Culture and Ethic Values)	One week	One week	One week	One week	One week

Partially implemented means that not all the materials of the didactic unit where applied.

I would be willing to live nearby to someone with a mental health problem."

Data Analyses

Student's t-test for continuous values and χ^2 tests with continuity corrections for categorical values were computed on baseline measures and socio-demographic variables to examine differences between the two groups.

Firstly, to analyze the effectiveness of an intervention, we need outcome measures that are well-designed and have empirically proven their reliability and validity. Therefore, we analyzed the psychometric properties of the YPQ in terms of factor structure, internal consistency, and construct validity. To this end, CFA was conducted to test the fit of the two-factor model proposed by the Mental Health Commission of Canada (MHCC; Stuart et al., 2014). In addition, we tested a two-factor model with method effects (model 2 as respecification of model 1) that incorporated correlated error terms on the negatively phrased items. The weighted least squares mean and variance (WLSMV) was used as an estimation method to test the fit of the factor models. The following indices were examined to evaluate model fit (Schumacker and Lomax, 2010): chi-square (non-significance reflects good fit), the Tucker-Lewis index (TLI $\geq$ 0.9), the comparative fit index (CFI $\geq$ 0.9), and the root means square error of approximation (RMSEA $\leq$ 0.08). Internal reliability of the YPQ and its subscales was explored with Cronbach's α coefficient. Test–retest reliability was also evaluated in the Control group by means of the intraclass correlation coefficient. Pearson correlation with RIBS was used to explore the convergent validity of the YPQ.

Regarding the effectiveness of the "*What's up!*" intervention, to examine differences in primary and secondary outcome measures between the intervention and control groups, mixed-effects models (Gueorguieva and Krystal, 2004) were performed using Restricted Maximum Likelihood (REML) to estimate the parameters. One of the advantages of mixed models is that these methods account for the correlation between the repeated measurements (baseline, post-intervention, follow-up) for an individual. Since different high schools were assigned to study conditions, the statistical analyses were computed at the group level, where mixed effects models were used to account for random subject effects within each high school. Age, gender, and items concerning previous contact with people with a mental disorder, and having experienced a mental disorder were included as covariates. We tested the effect of a treatment group (intervention vs. control), time, and the interaction term (Group × Time). A separate model was estimated for each of the four outcome measures. Analyses were conducted separately for intervention completers and for the intent-to-treat (ITT) sample. We used multiple-imputation to impute the missing values.

CFA was conducted using the statistical software MPlus 7.4. For the other analyses, SPSS v.22 and STATA13.1 were used.

RESULTS

Participants' Characteristics

Figure 1 shows the study flowchart. A total of 446 students were recruited and non-randomly assigned to the two study conditions. Their ages ranged from 14 to 17 years old (16 in

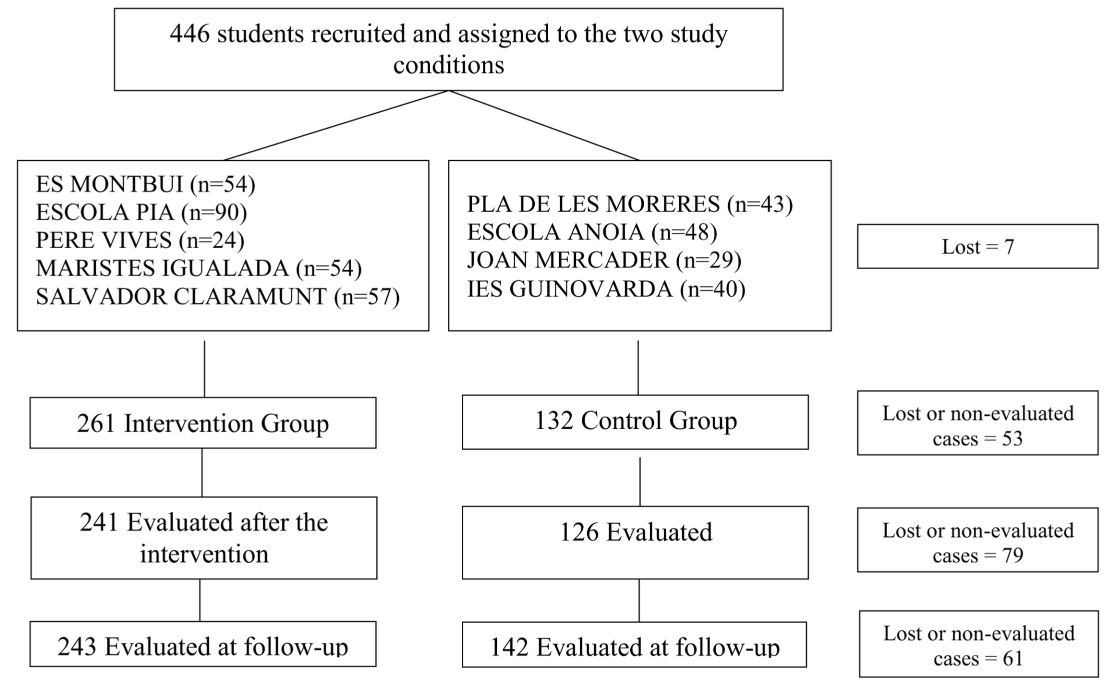

FIGURE 1 | Flow chart of the "*What's up!*" study; distribution of sample size by high school.

the intervention group). Baseline evaluation was carried out in 393 students (88.12% of the initial sample); post-intervention evaluation in 367 students (82.29%) and follow-up was conducted in 385 students (86.32%). As these evaluations were carried out in the ordinary classrooms, absent students did not undergo the corresponding evaluation.

We did not find significant differences in baseline characteristics between study arms (see **Table 2**). Distribution by gender was very similar (52% women) and the mean age was approximately 14 years old. A fifth of the students (21%) had experienced some kind of mental health problem.

No statistically significant differences were found between those students who answered at follow-up and those who did not for either the entire sample or both groups considered separately (all $p > 0.05$).

Psychometric Analysis of the YPQ

CFA yielded significant χ^2 values for both models (model 1 = 727.86, $p < 0.01$; model 2 = 434.99, $p < 0.001$). The other fit indices of model 2 indicated a more adequate fit to the data (CFI = 0.936; TLI = 0.924; RMSEA = 0.058, 90 and CI 0.051–0.065) than the first model (CFI = 0.863; TLI = 0.848; RMSEA = 0.082, 90 and CI 0.075–0.088). The reliability of the original version of the YPQ-SS was 0.79 and for YPQ-SAS it was 0.85 (Stuart et al., 2014).

In Model 2, the statistically significant correlated residuals were as follows: $\theta_{14-19} = 0.29$; $\theta_{14-20} = 0.18$; $\theta_{14-21} = 0.23$; $\theta_{14-22} = 0.21$; $\theta_{19-20} = 0.53$; $\theta_{19-21} = 0.33$; $\theta_{20-21} = 0.57$; $\theta_{20-22} = 0.33$; $\theta_{21-22} = 0.36$. A table with all the factor loadings can be found as Supplementary Material.

The overall internal consistency of the YPQ in our sample, based on Cronbach's alpha among all items, was 0.84. We also calculated internal consistency at the scale level using the α "if item deleted" option. The alpha value for the total score ranged from 0.83 to 0.85. Regarding YPQ sub-scales, both present acceptable values (α = 0.73 for YPQ-SS and α = 0.8 for YPQ-SAS). Examining the α "if item deleted" option, the YPQ-SS and YPQ-SAS obtained values that ranged from 0.69 to 0.72; and 0.77 to 0.81, respectively. The test–retest reliability for the YPQ total score was 0.84, while the YPQ-SS and the YPQ-SAS both obtained 0.80. Convergent validity was also satisfactory as statistically significant correlations were found between the YPQ and its two factors, and the other measure used in this study (RIBS). The YPQ global score obtained correlations of $r = 0.63$ with RIBS; the YPQ-SS presented lower correlations, yet significant ($r = 0.47$). Correlations of $r = 0.62$ with RIBS were found for the YPQ-SAS.

Effectiveness of the "What's Up!" Intervention

Table 3 presents descriptive statistics for the study measures by group throughout the study. Statistical analyses revealed that the interaction Group (Intervention vs. Control) × Time (pre, post, and follow up) was significant for all measures (all $p < 0.01$). **Table 4** shows all the fixed and random effects of the models, which proved to be statistically significant for all measures (Chi squared: YPQ-SS = 259.42, $p < 0.01$; YPQ-SAS = 297.38, $p < 0.01$; RIBS = 217.02, $p < 0.01$). These interaction results imply that the groups differed in rate and manner of change over the course of the study. The YPQ sub-scale scores and RIBS scores in the intervention group improved significantly from baseline to post-intervention ($p < 0.01$ for YPQ-SS and $p = 0.03$ for YPQ-SAS; $p = 0.01$ for RIBS) and from baseline to follow-up ($p < 0.01$ for YPQ-SS and $p = 0.01$ for YPQ-SAS; $p = 0.02$ for RIBS). Interestingly, no statistically significant difference was found between the post-intervention evaluation and follow-up (all $p > 0.05$), which indicates that the changes observed after the intervention were maintained over time. In contrast, no differences were found from baseline to post-treatment in the control group ($p = 0.06$ for YPQ-SS, $p = 0.16$ for YPQ-SAS; $p = 0.10$ for RIBS) or between baseline and follow-up ($p = 0.08$ for YPQ-SS and $p = 1.00$ for YPQ-SAS; $p = 1.00$ for RIBS). As can be observed in **Figure 2** and **Table 3**, with the exception of the YPQ-SS, all measures worsened at the post-treatment evaluation and returned to baseline values at follow-up.

Despite the significant interactions Group × Time, *post hoc* analysis revealed that groups did not differ significantly in any of the study periods in any of the study outcomes (all $p > 0.05$), which indicates that the change achieved by the intervention was small.

Analysis of covariates indicated that both stereotypical attitudes and intended behavior were significantly better in women than in men (all $p < 0.01$), and attitudes, measured with the YPQ-SS, were less stigmatizing in those students who had a close relative or a friend with mental health problems ($p = 0.03$; $p = 0.04$, respectively). Intended behavior measured with RIBS also presented significant effects of the covariates "having a close relative with a mental disorder" ($p = 0.01$), "having a friend with a mental disorder" ($p = 0.01$), and "having experienced

TABLE 2 | Baseline characteristics of the sample by condition (Intervention and Control).

Variables		Total	Intervention	Control	*p*-Values
Gender (*n*, % women)		203 (51.7%)	137 (52.5%)	66 (50.0%)	0.67
Age (*M*, *SD*)		14.24 (0.47)	14.21 (0.43)	14.31 (0.54)	0.59
Own experience of a mental disorder (*n*, % Yes)		80 (20.6%)	46 (17.9%)	34 (25.8%)	0.09
Contact with mental disorders (*n*, % Yes)	A close relative	60 (15.4%)	42 (16.3%)	18 (13.7%)	0.56
	Another relative	87 (22.3%)	60 (23.3%)	27 (20.5%)	0.61
	A friend	164 (42.1%)	107 (41.5%)	57 (43.2%)	0.75
	An acquaintance	255 (65.2%)	172 (66.4%)	83 (62.9%)	0.50

mental disorders” ($p = 0.01$). The other covariates did not present significant effects for this scale.

Following multiple imputation of missing values (data available upon request), results were very similar to those obtained in the completers’ approach. Only some aspects changed: the effect of the follow-up measure became significant for the YPQ-SS ($p < 0.01$) but, surprisingly, the interaction Group × Time lost its significance at follow-up for the YPQ-SAS and the RIBS ($p = 0.26$ and $p = 0.15$, respectively). Furthermore, some of the covariates which presented no significant effect changed after the multiple imputation of missing values, such as age, which became significant for all the outcome measures (all $p < 0.01$).

DISCUSSION

The main objective of this study was to analyze the effectiveness of the “*What’s up!*” intervention on mental illness stigma in a large adolescent Catalan sample and, secondly, to examine, for the first time, the psychometric properties of the YPQ in a Catalan-speaking sample. The results can be summarized as follows: statistically significant but small improvements were found in the intervention group, and the psychometric analysis of the main outcome measure (YPQ) revealed a two-factor structure, good reliability coefficients for both dimensions (YPQ-SS and YPQ-SAS), and adequate convergent validity.

To our knowledge, this is the first study to report a full validation of the YPQ, as the original authors only assessed its internal reliability. A CFA was conducted, assuming that the two YPQ scales (i.e., YPQ-SS and YPQ-SAS) described by the authors would imply a two-factor structure. This analysis indicated that the YPQ shows a two-factor structure and similar psychometric properties to those reported by the designers of the measure (Stuart et al., 2014). A common result in CFA of psychological instruments composed of direct and reverse scored items is to obtain an inadequate fit because positively phrased items are prone to load on one factor and negatively phrased items on another (Woods, 2006). We decided to covariate the error terms of the reverse scored items to resolve this methodological problem and we obtained a better fit than without covariance. The YPQ had good internal consistency and convergent validity with the RIBS. When taking the questionnaires’ two scales separately, both correlated significantly with the RIBS. All items loaded correctly on their respective factor with the exception of item 4 (“People with a mental illness could snap out of it if they wanted to”), which presented a very low λ for both models.

TABLE 3 | Mean scores and standard deviations for the study measures (YPQ and RIBS).

	Intervention			**Control**		
	Baseline	**Post**	**Follow-up**	**Baseline**	**Post**	**Follow-up**
YPQ-SS	2.41 (0.59)	2.07 (0.57)	2.07 (0.62)	2.43 (0.72)	2.31 (0.85)	2.32 (0.83)
YPQ-SAS	2.06 (0.62)	1.94 (0.64)	1.94 (0.65)	2.11 (0.81)	2.21 (0.93)	2.15 (0.90)
RIBS	2.16 (0.91)	1.94 (0.90)	1.94 (0.91)	2.00 (1.11)	2.18 (1.41)	2.06 (1.24)

TABLE 4 | Mixed effects models for YPQ-SS, YPQ-SAS, and RIBS (completers’ approach).

Parameter	**YPQ-SS coefficient (*SE*)**	**YPQ-SAS coefficient (*SE*)**	**RIBS coefficient (*SE*)**
Fixed effects			
Group	0.03 (0.08)	0.00 (0.11)	0.20 (0.12)
Time			
T2	−0.10* (0.05)	0.10* (0.05)	0.18* (0.07)
T3	−0.09 (0.05)	0.06 (0.05)	0.08 (0.07)
Group × Time			
G1 T2	−0.23** (0.06)	−0.21** (0.06)	−0.38** (0.09)
G1 T3	−0.23** (0.06)	−0.19** (0.06)	−0.28** (0.09)
Age	0.07 (0.05)	0.03 (0.05)	0.09 (0.07)
Gender (1 = woman)	−0.17** (0.05)	−0.33** (0.05)	−0.31** (0.07)
Close relative with a mental disorder (1 = yes)	−0.14* (0.07)	−0.08 (0.07)	−0.23* (0.10)
Other relative with a mental disorder (1 = yes)	−0.06 (0.06)	−0.06 (0.06)	−0.03 (0.08)
Friend with a mental disorder (1 = yes)	−0.10* (0.05)	−0.07 (0.05)	−0.17* (0.07)
Acquaintance with mental disorders (1 = yes)	−0.08 (0.05)	−0.05 (0.05)	−0.07 (0.07)
Personal experience of a mental disorder (1 = yes)	−0.11 (0.06)	−0.08 (0.07)	−0.23* (0.09)
Random effects			
High school	0.01 (0.01)	0.02 (0.01)	0.01 (0.01)
Students	0.12 (0.02)	0.15 (0.02)	0.28 (0.04)

$^{*}p < 0.05$, $^{**}p < 0.01$; G1 = Intervention group; T2 = Post-Intervention; T3 = Follow-up.

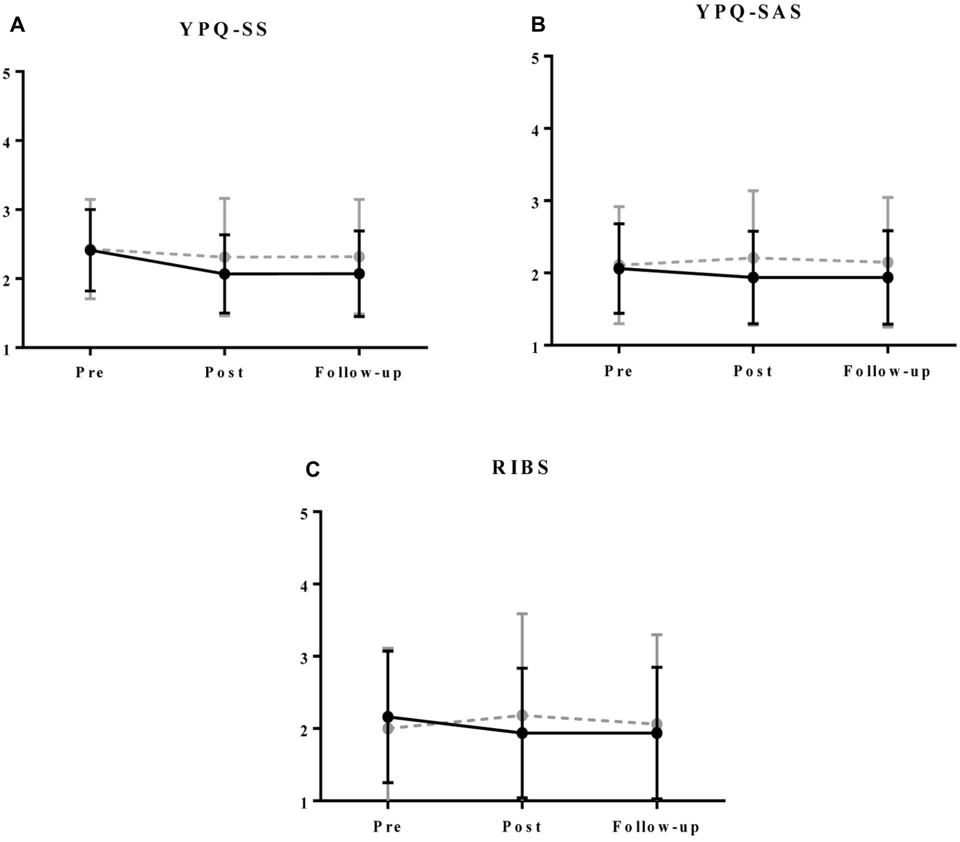

FIGURE 2 | Mean scores and standard deviations for YPQ-SS **(A)**, YPQ-SAS **(B)**, and RIBS **(C)** by group throughout the study. Black and continuous line is for intervention group. Gray and dotted line is for control group. Higher scores in YPQ and RIBS represent higher stigma levels.

Without ignoring that our sample may not reflect the whole variability of adolescence as its age variability is very low, our results show that the YPQ is a valuable instrument for use in further studies addressed to secondary students. Its value lies not only in its sound psychometric properties but also in the adequacy of the language and the aspects evaluated, which are directly related to the reality of adolescence (e.g., sitting next to a classmate with a mental disorder or being taught by a teacher with mental health problems). This favors the YPQ in comparison with other commonly used measures such as the Community Attitudes toward Mental Illness (CAMI; Taylor and Dear, 1981), whose items contain questions related to the health system or the economic costs of people with a mental disorder, areas which are not often well-known by teenagers. Moreover, the YPQ items reflect the realities of today's teenager, such as bullying or volunteering. Many measures were designed for previous studies, but not all of them were reliable and valid (Sakellari et al., 2011). Consequently, the present study provides validation of the YPQ to help overcome the difficulty of choosing which measure to use when delivering an anti-stigma intervention to a teenage population.

Regarding the effects of the intervention, we can conclude that "*What's up!*" was able to improve the attitudes and intended behavior of the students. Despite being significant, the changes achieved by the intervention were small, similar to those observed in some previous studies (Mino et al., 2001; Altindag et al., 2006; Kerby et al., 2008; Lincoln et al., 2008), although the reason why these interventions did not have a bigger impact was considered to be the fact that they were addressed to medical students, a population with such high levels of knowledge about mental disorders that they focused more on the negative aspects of these conditions, making their stigma stronger (Corrigan and Watson, 2007; Angermeyer et al., 2011). Nonetheless, this cannot be considered the reason why "*What's up!*" has not presented more significant effects on reducing stigma, as our sample was constituted of high school students who did not have such a degree of knowledge. In all likelihood, the fact that the students already presented very low levels of stigma before the intervention (floor effect) was in part responsible for the small effects of the intervention. This is in line with previous findings showing the Spanish population reporting lower levels of stigma than those of other European and American countries (Aznar-Lou et al., 2016), which would imply that it could be more efficient to address interventions such as "*What's up!*" to students who report high levels of stigma.

To our knowledge, this is the first time that the effectiveness of a curricular intervention addressed to fight stigma toward mental disorders has been explored. Previous approaches were commonly based on short but very immersive workshops which used materials such as presentations, videos, role-plays, talks, and contact with people with mental disorders. Some were reported to have achieved a greater impact on stigma than "*What's up!*" (Petchers et al., 1988; Mound and Butterill, 1993; Esters et al., 1998; Rahman et al., 1998; Ng and Chan, 2002; Pinfold et al., 2003; Schulze et al., 2003; Watson et al., 2004; Stuart, 2006; Spagnolo et al., 2008; Essler et al., 2009), mostly on knowledge about mental health problems (Sakellari et al., 2011), an aspect that was not evaluated in our study. However, many of these studies had methodological shortcomings such as the absence of a control group (Mound and Butterill, 1993; Pinfold et al., 2003; Stuart, 2006; Essler et al., 2009) and only three conducted a follow-up, so these results should be considered with caution. Additionally, a very common limitation of previous interventions was that they seldom reported sustained improvements over time (Economou et al., 2012; Thornicroft et al., 2016), which implied that they had short-term effects that were not maintained at follow-up. In contrast, our results indicate that the improvements in stigmatizing attitudes and intended behavior achieved by "*What's up!*" were maintained at 9-month follow-up. It is possible that the model of intervention used for this campaign, where the teachers include the anti-stigma material in their classes, makes it easier for the students to get involved with the information than in those interventions in which an external professional brings the material to the adolescents in a workshop format. This may produce a greater immediate reduction in stigma as it is more immersive than doing their regular classes with some didactic units on mental health, but its effects may be more superficial and easily lost with the passage of time.

These results suggest that the debate on which approach is more effective to fight stigma is open. The literature provides different examples of interventions addressing this issue (workshops, whole-school approaches, and now, curricular interventions). Considering the characteristics of the target population and the available resources to conduct the intervention, the most adequate alternative is chosen. In the case of curricular interventions such as "*What's up!*," our results suggest that they can be effective in promoting sustained effects regarding stigmatizing attitudes and intended behavior toward people with mental disorders. These improvements have been found in a sample with relatively low levels of stigma, which may imply that other populations with similar levels may benefit from the intervention as well. Since the results of "*What's up!*" were maintained after a 9-month follow-up, it may be also considered as an affordable extension to other approaches that have already proved to be effective but only in the short-term.

In another vein, it is also interesting to discuss the significant influence of the covariates in the effectiveness findings. For instance, being female was a significant covariate for all the measures, which is in line with previous studies (Stickney et al., 2012). Some studies have found that adolescent girls had more knowledge and experience about mental disorders, reported less stigma toward people with them and felt more likely to use mental health services (Barrett and Raskin White, 2003). The statistical analysis did not find a significant effect of being female on the response to the intervention, but this effect has been found in previous studies (Thornicroft et al., 2007). A possible explanation for this could be that traditional ideals of masculinity (e.g., success, power, competition, emotional repression) influence teenage boys in such a way that messages designed to combat stigma with respect to mental disorders have a smaller impact than they do on teenage girls (Chandra and Minkovitz, 2006).

Two other covariates that also appeared to be significant in all the measures but the YPQ-SAS were having a close relative with a mental disorder and having a friend with a mental disorder. Again, no significant effect of these variables on the response to the intervention was found, so it cannot be concluded that these adolescents are more likely to decrease their stigma. However, it can be asserted that their levels of stigma were significantly lower than those reported by the teenagers who did not have friends or close relatives with a mental disorder. It may be the case that having a close relationship with someone with a mental disorder makes it easier for teenagers to feel empathy with them and so to decrease their levels of stigma more significantly than adolescents who have no friends or close relatives with this kind of problem, as it has been observed that empathy is a key individual factor influencing attitude change toward mental disorders (Couture and Penn, 2003).

Strengths and Limitations

To our knowledge, this is the first study to report the effectiveness of a curricular intervention to combat stigma associated with mental disorders. This is a controlled study, but not randomized, so there could be some selection bias. In this respect, it is notable that the comparative analyses in the basal phase showed no significant differences between the intervention and the control group in any of the study measures. Due to the low number of independent variables of this study, no mediation analysis was conducted.

Some limitations about the intervention itself must be considered: on the one hand, the instructions given to the teachers were very flexible, requiring the application of just three of the nine didactic units proposed by the project, although remarking that the first and the last (which contained the social contact component of the campaign) were considered "essential." It can be concluded that a little more systematic orientation in applying the intervention is needed to evaluate intervention effectiveness, although it should be noted that all high schools implemented at least five of the nine didactic units and an important strength of the intervention was that this degree of flexibility made "*What's up!*" a truly implementable intervention in the real context of high schools.

We must take into account that the timing was not the same for all the high schools at the post-intervention measure. Although all started within 2 weeks of each other, one class took the post-intervention measure only 25 days after baseline assessment, while the rest of the classes were evaluated between 77 and 105 days after baseline. These differences may have had an impact on the results as it seems reasonable to suppose that the more time spent on didactic units in class, the more impact they would have on the students. This could be easily corrected by adjusting the evaluation periods and conducting assessments at roughly the same time in all schools. Additionally, during application of the intervention there was no register of external events that could interfere with it (e.g., to include an item in the socio-demographic scale registering contact with fictional characters with mental health problems either in movies, TV shows, or novels).

Regarding the external validity of the intervention, it should be borne in mind that, although the measures used for this study presented strong psychometric properties, their items might be susceptible to social desirability and more objective measures should be developed to ensure that the assessment of stigma is as close to actual behavior as possible. It would also have been interesting to include a qualitative measure in the evaluation of the impact of the intervention to determine which aspects had the biggest effect on the participants and why. This could have been especially useful considering that "*What's up!*" consisted of nine distinct units implemented by different teachers, which, as mentioned previously, introduced a high degree of variation to the intervention, and not all the units could impact equally for every student. Future studies with a larger sample size could use a "dismantling design" to examine which of the multiple didactic units included in the "*What's up!*" intervention are the active ingredients of change. One typical method for conducting this type of study is to compare single intervention components with the global intervention.

As noted above, an important strength of this study is the use of reliable and valid outcome measures. The YPQ emerged as a useful instrument that measures both attitudes and intended behavior and whose psychometric attributes have been found to be positive. However, it is true that its convergent validity was only studied with RIBS, which is an intended behavior measure, and it would also be interesting to study it with a measure of stigmatizing attitudes such as the CAMI. It could also be argued that this measure is not very sensitive to change, but as similar results were found for RIBS, we must conclude that the intervention did not have the impact required to produce more significant changes, maybe due to some of its previously explained limitations. Moreover, we should point out that our results may not be totally generalizable to the whole adolescent population, as our sample presented a very low degree of age variability.

CONCLUSION

"What's up!" is a promising, original intervention model in which teachers are the responsible for introducing campaign materials into regular didactic units, without modifying the school curriculum and offering the students the opportunity to learn about mental health through education and social contact. Although significant improvements were observed, these results were small. To overcome the shortcomings of this intervention, further anti-stigma interventions could focus on reducing time differences between classes and registering external variables that could affect students' stigma levels. Additionally, it may be more appropriate to address the intervention especially to those students who report higher levels of stigma at baseline to counter the "floor effect."

On the other hand, the present work has satisfactorily validated the YPQ, an instrument that had only been used in one previous intervention and whose psychometric properties had not yet been reported. This questionnaire evaluates stereotypical attitudes and intended behavior and presents good internal consistency, test–retest reliability and convergent validity and can thus be considered as a valuable measurement instrument for use in further studies of stigma in adolescent populations.

AUTHOR CONTRIBUTIONS

AP-A and LA-R made substantial contribution to the analysis and to the interpretation of the data, drafted the manuscript, provided 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. JL, IA-L, MR-V, and AF-S made substantial contributions to the conception and the design of the study, drafted the manuscript, provided 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. AS-B helped out in the interpretation of data for the work, revised the manuscript critically for important intellectual content, provided 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. MJ and AT revised the manuscript critically for important intellectual content, provided 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. AB revised the manuscript critically for important intellectual content, provided 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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SUPPLEMENTARY MATERIAL

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Beyond Disease: Happiness, Goals, and Meanings among Persons with Multiple Sclerosis and Their Caregivers

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The experience of persons with multiple sclerosis (MS) and their caregivers is usually investigated in terms of emotional distress and health-related quality of life, while well-being indicators remain largely underexplored. In addition, findings are often interpreted from the clinical perspective, neglecting socio-cultural aspects that may crucially contribute to individuals' functioning. At the methodological level, most studies rely on scaled instruments, not allowing participants to freely express their needs and resources. Based on the bio-psycho-social perspective endorsed by the International Classification of Functioning, well-being indicators were investigated among 62 persons with MS (PwMS), their 62 caregivers and two control groups, matched by age and gender. Participants completed the Positive Affect Negative Affect Schedule (PANAS), the Satisfaction with Life Scale (SWLS), and the Eudaimonic and Hedonic Happiness Investigation instrument (EHHI). EHHI provides information on participants' happiness, goals and meanings through scaled and open-ended questions, contextualized within major life domains. No relevant differences emerged among PwMS and caregivers, compared with the respective control groups, as concerns life domains associated with happiness, goals and meaning. Participants across groups prominently mentioned family, highlighting its intrinsic value and its relevance as a sharing context; health did not represent a major theme for PwMS; community, society and religion/spirituality issues were substantially neglected by all participants. PwMS and caregivers reported lower levels of positive affect than their control groups, while no substantial differences emerged for negative affect, happiness and meaningfulness levels in life and across most domains. Results suggest that the experience of MS is associated with well-being in relevant life domains, such as family and close relationships. Although PwMS and caregivers identified a lower number of goals and meaning-related opportunities compared to control groups, they showed a positive adjustment to disease through the development of personal and family resources. These assets are often undervalued by health professionals and social institutions, while they could be fruitfully exploited through the active involvement of PwMS and their families as expert and exemplary informants in initiatives aimed at promoting the well-being of individuals and communities

Keywords: multiple sclerosis (MS), well-being, caregiving, daily living, meaning-making, goals, mixed method, psychosocial interventions

INTRODUCTION

The technological advancements in the medical domain fostered the representation and interpretation of health-related phenomena through rigorously quantitative approaches, leading to the affirmation of the biomedical model. Despite the shortcomings entailed by this narrow perspective, and the classical definition of health as "a state of complete physical, mental and social well-being" (World Health Organization, 1946, p. 1), both research and clinical practice are still permeated by a strictly biological view of human functioning.

A laudable effort to overcome this limitation and to promote the adoption of a bio-psycho-social view of health and disease is represented by the International Classification of Functioning, Disability and Health (ICF; World Health Organization, 2001). ICF is aimed at comprehensively assessing the physical, mental and social functioning of any individual, including persons with diseases and disabilities. It presents a marked shift in terminology, from focusing on the consequences of disease (impairments, disabilities, handicaps) to investigating the components of health and functioning, classified as structures and functions, daily activities, and social participation. Far from endorsing a pure linguistic convention, the ICF stems from a relevant conceptual change, leading to the evaluation of health conditions from a constructive perspective. It is based on the assumption of a dynamic interplay between individual features (body functions, activities, and participation) and environmental aspects that can facilitate or hinder the person's functioning (Dixon et al., 2008; Bodde and Seo, 2009). However, the implementation of this model into practice is problematic, as it requires a multidisciplinary integration effort involving researchers and practitioners from the healthcare, education and policy domains (Stucki et al., 2017). In addition, the assessment of psychosocial functioning through the current version of ICF poses several challenges. Psychological and environmental dimensions are collapsed into the heterogeneous domain of "contextual factors"; the psychological aspects are not explicitly identified through a checklist; and the components of community participation are difficult to evaluate (Chang et al., 2013). Despite these limitations, only the ICF currently offers an integrated approach to human functioning which can be used by professionals working in different sectors.

From the ICF perspective, it is not surprising that people with chronic diseases or disabilities describe themselves as ordinary persons who cope with extraordinary circumstances (Saravanan et al., 2001) that include biological impairments as well as material, social and institutional barriers. Despite these disadvantages, the mobilization of personal, relational and environmental resources allows these people to attain high levels of mental health (Arnold et al., 2005; Andrykowski et al., 2008; Delle Fave et al., 2015). Similarly, caregivers experience the coexistence of caring burden and limitations in daily opportunities with psychological and social resources (Song and Singer, 2006; Olsson et al., 2008; Fianco et al., 2015).

The study of mental health and well-being has received great impulse during the last two decades, through theories and empirical models deriving from two complementary perspectives. Within the hedonic perspective, well-being is operationalized as the predominance of positive over negative affect in daily experience and a globally positive life evaluation, defined as satisfaction with life (Ryan and Deci, 2001). From the eudaimonic perspective well-being is instead conceptualized as a dynamic growth process, that includes a wide range of constructs such as goal setting and pursuit, meaning-making, self-expressiveness, self-determination, self-acceptance, skill development and mastery, trust in relationships, and social integration (Huta and Waterman, 2014). Both these approaches proved to be useful in exploring protective resources, adaptation processes and adjustment outcomes that allow persons with chronic diseases and their caregivers to attain positive functioning (Cummins, 2005; Diener and Chan, 2011).

This avenue of research is however characterized by an emphasis on individual processes, while the societal and cultural factors influencing the person's daily functioning are substantially neglected (Di Martino et al., 2017). Demographic and contextual features, when investigated, are treated as components of the person's stable identity, despite the changes that both the environment and the individual ceaselessly undergo (Slife and Richardson, 2008).

The Individual and Family Experience of Multiple Sclerosis: Challenges and Resources

Multiple sclerosis (MS) is a chronic neurodegenerative disease, characterized by largely unpredictable symptoms and course, and currently lacking of curative treatment. Therefore, besides physical impairments, MS poses a number of psychological, behavioral and social challenges to both affected persons and their caregivers (Dennison et al., 2009; Ackroyd et al., 2011). The progressive course of the disease requires a constant re-adjustment over time (Bogosian et al., 2017), leading the person to gradually scale back or adapt daily activities to the new condition, find new occupations and interests, but also disengage from the adjustment process and withdraw from active life.

While the negative consequences of MS were extensively explored, research on the positive aspects is still limited. Studies have been conducted to investigate the eudaimonic process of meaning making, a key individual resource to attain well-being under unfavorable and irreversible conditions (Hicks and King, 2009). Meaning making allows individuals to integrate the problematic condition into a global and constructive view of their present and future life. The qualitative exploration of meaning making among persons with MS shed light on its components, which include disease acceptance, personal and relationship growth, and positive lifestyle changes (Pakenham, 2008a). Quantitative studies highlighted the positive association of sense making with life satisfaction, and its negative association with depression. Sense making was identified as a key predictor of positive adjustment also among caregivers (Pakenham, 2008b). Similar findings were detected as concerns perceived illness coherence, a construct closely related to meaning making; illness coherence was positively associated with eudaimonic and hedonic well-being indicators in both persons with MS and their caregivers (Bassi et al., 2016).

Benefit finding, consisting in the ability to identify positive consequences in an otherwise negative situation (Lechner et al., 2009), was also investigated among persons with MS and their family caregivers. Cross-sectional and longitudinal studies highlighted its positive association with meaning-based coping strategies, and its direct effects on positive adjustment outcomes, such as good dyadic relationships and subjective well-being (Pakenham, 2005, 2007, 2008b; Pakenham and Cox, 2009). Related findings support the idea of a "communal search for meaning" where persons with MS and their caregivers experience the trauma of a chronic illness and subsequently find positive aspects together (Pakenham, 2005). Further evidence of this dyadic process emerged from the investigation of post-traumatic growth and illness perceptions among persons with MS and their partners (Ackroyd et al., 2011).

These studies also showed that the disruption of life goals caused by MS progression can be counterbalanced by the development of new meanings and life purposes (Joseph and Linley, 2006). This process entails a complex interplay between disengagement from previous goals and reengagement in new ones (Neter et al., 2009). It also requires the implementation of adaptive tasks (Bensing et al., 2002), such as defining new challenges, acknowledging one's limits, maintaining emotional balance and self-esteem, facing uncertain future, cultivating social relationships, and looking at the bright side of life. Besides these individual resources, other factors located at the intersection between persons and their context may foster positive adaptation; they include facilitators of mobility/independence, social support, and social comparisons (Dilorenzo et al., 2008).

A frequent problem in studies investigating well-being and perceived quality of life among persons with MS is the lack of objective measures of disease severity; this limitation prevents researchers to exclude that positive adjustment may simply reflect a condition of less severe illness (Dennison et al., 2009). The assessment of demographic and clinical dimensions however entails methodological and interpretive challenges, as showed by the contrasting findings obtained in different studies. For example, in some studies demographic variables such as education and employment emerged as the strongest predictors of health-related quality of life (HR-QoL), together with clinical ones like depression and disability (Patti et al., 2003, 2007b). In other studies, HR-QoL itself emerged as an independent predictor of disability progression (Benito-Léon et al., 2013). Satisfaction with life was primarily predicted by disease severity and social support in one study (Ryan et al., 2007), while in another study age and education level emerged as the strongest predictors of hedonic and eudaimonic well-being (Bassi et al., 2014). Finally, a comparative study did not highlight significant differences in personal growth (a eudaimonic dimension) between persons with MS and healthy participants, while life satisfaction (a hedonic dimension) was significantly lower among the former (Barak and Achiron, 2011).

The investigation of well-being among caregivers of persons with MS is even less frequent, as studies are primarily focused on the emotional impact of caring-related burden and stress (Patti et al., 2007a; Rivera-Navarro et al., 2009), and on the daily choices and free time restrictions imposed by the caring role (Becker, 2011). Studies investigating satisfaction with life did not detect differences between caregivers of persons with MS and samples derived from the general population or carers of persons with other chronic diseases (Waldron-Perrine et al., 2009; Bassi et al., 2014). Eudaimonic well-being dimensions such as meaning making, benefit finding and illness coherence were more often investigated among caregivers, as reported in the previous paragraphs (Pakenham, 2007; Pakenham and Cox, 2009; Bassi et al., 2016). In a recent qualitative study involving Turkish caregivers (Topcu et al., 2016), participants mentioned both individual and social resources, such as motivation to care and perceived support.

Acknowledging the existence and adaptive role of well-being dimensions in the experience of MS surely represents an advancement. At the same time, most studies suffer from two conceptual limitations. The first one is the lack of a comprehensive bio-psycho-social perspective, and especially the neglect of the societal context surrounding persons with MS and their caregivers. Only few studies were conducted within the ICF framework. One of them was aimed at identifying a core set of ICF categories specific to MS that could be used in healthcare practice (Khan and Pallant, 2007). Another study proposed the Multiple Sclerosis Impact Profile (MSIP; Wynia et al., 2008) an instrument designed to evaluate the perceived impact of MS on functioning. A recent review (Dorstyn et al., 2017) highlighted the association between depressive symptoms and reduced social participation in persons with MS. A qualitative study exploring the impact of ICF contextual factors on the daily functioning of Jordanian persons with MS (Hamed et al., 2012) identified adequacy of financial and medical resources, religion and community awareness as facilitators, and social stigma as a barrier. Finally, the environmental and personal facilitators of social participation and satisfaction with parenting were investigated among mothers with MS (Farber et al., 2015). Findings suggested the importance of incorporating both categories of facilitators into treatment. The relevance of environmental factors also emerged from the investigation of unmet needs among persons with MS. These needs most often refer to contextual dimensions, such as family and social support, healthcare services, everyday life management, and the relationship with physicians (Galushko et al., 2014). Interestingly, representations of the unmet needs of persons with MS differ among health professionals (Golla et al., 2012): while physicians emphasize limitations in access to care due to poor financial resources, nurses and social workers are more aligned with patients' perceptions, quoting family support, social relationships and daily life management.

A second limitation characterizing the literature on MS is the emphasis on disease and related adjustment processes: participants and caregivers are rarely solicited to freely reflect on aspects of their life and daily experience potentially unrelated to disease or caregiving respectively. This limitation emerged in one of the studies investigating unmet needs of persons with MS (Galushko et al., 2014): participants expressed the need to be viewed as distinct individuals, not constrained by and identified through their health conditions.

The Present Study

The first aim of the present study was to investigate different dimensions of well-being among persons with MS and their caregivers through a mixed method approach and from a bio-psycho-social, ICF informed perspective. Answers obtained through open-ended and scaled questions were jointly analyzed, in the attempt to contextualize findings within participants' global outlook of their own life and environmental opportunities. The well-being dimensions investigated in the study included happiness (its definition and recent related situations), hedonic well-being (positive and negative affect, satisfaction with life) and eudaimonic well-being (perceived meaningful things and goals). In addition, information was collected on happiness and meaningfulness levels in life in general, as well as in the specific domains of work, family, standard of living, interpersonal relations, health, personal growth, leisure, spirituality/religion, community, and society. Overall, the mixed method potentials are still underexploited in the psychological literature; moreover, to the best of our knowledge no studies have yet been conducted on these topics and through this methodological approach in the MS domain.

The second aim was to compare the findings collected among persons with MS and their caregivers with those obtained from two groups of participants, matched by age and gender, but with no history of chronic disease or caregiving experience. From an ICF informed perspective, this comparison was expected to shed light on group differences in perceived personal and contextual facets of well-being, including daily activities and opportunities, relational networks, and social participation. Attention was also paid to the role of employment status and education level in predicting well-being dimensions. These two demographic features represent crucial indicators of participation, classified in the ICF as contextual personal factors influencing human functioning (World Health Organization, 2002; Martins, 2015). The level of disability was also taken into consideration in the analysis of data collected among persons with MS.

Based on the available literature, some hypotheses were formulated. In line with studies on adjustment to MS (Pakenham, 2008a; Bassi et al., 2016) we expected that, when describing happiness, goals and meaningful things, a significantly higher percentage of persons with MS would refer to health compared with a control group. We also expected persons with MS to associate health with significantly lower levels of happiness and higher levels of meaningfulness, compared with a control group. As concerns caregivers, based on previous evidence (Pakenham, 2008b; Becker, 2011; Mausbach et al., 2011) we expected that a significantly higher percentage of participants would refer to family and a lower percentage to leisure, compared with a control group. We also expected caregivers to report significantly lower levels of happiness with leisure, and higher levels of meaningfulness in relation to family. As concerns ratings of affect and life satisfaction (the two components of hedonic well-being), the contrasting findings detected across studies (Barak and Achiron, 2011; Diener et al., 2017) did not allow us to formulate specific hypotheses. Finally, based on the literature highlighting the relevant role of education and employment in predicting individual well-being (Keyes, 2007; Patti et al., 2007b; Diener et al., 2017), we hypothesized that these two demographic features would provide a specific contribution to hedonic and eudaimonic well-being values across all groups. The same hypothesis was formulated as concerns the role of severity disease among persons with MS.

MATERIALS AND METHOD

Participants

The study involved 248 Italian adults divided into four groups, each of them comprising 62 participants: persons with MS (PwMS), their caregivers, and two control groups of adults with no history of chronic disease or caregiving experience, selected from a larger study aimed at investigating well-being in the general population. Inclusion criteria for PwMS were being at least 18 years of age, having a clinically definite MS diagnosis for at least 3 years and having a caregiver; exclusion criteria comprised the presence of additional neurological or psychiatric disorders, severe cognitive impairment, MS in the active phase, and a condition of very severe disability, corresponding to a score above 8 on the Extended Disability Status Scale (EDSS; Kurtzke, 1983). The majority of PwMS involved in the study (59.68%) showed an EDSS score between 3.5 and 8 (indicating increasing levels of motor impairment), while 40.32% scored below 3.5 (indicating a high level of autonomy). No significant differences were detected between PwMS with low and high levels of disability for any of the variables examined in this study; therefore disability level will not be further considered in the following sections.

Caregivers were predominantly partners (59.67%) or parents (22.57%) of PwMS, siblings or children (each accounting for 6.46%), friends (3.23%), or professional caregivers (1 participant, 1.61%). Participants in the control groups were randomly extracted from a sample of 691 healthy Italian adults; age, gender and education level were used as filters to match the control groups with the respective comparison groups – PwMS and caregivers. It was however not possible to obtain a complete match between caregivers and their control group as concerns education level. The decision to include two different control groups was based on the significant age difference between PwMS and their caregivers ($t = 2.85$, $p < 0.005$). The demographic features of the four groups of participants are reported in **Table 1**.

No differences emerged between PwMS and their control group (control 1) for age, gender, marital status and education, while a significantly higher percentage of PwMS was unemployed ($\chi^2 = 21.81$, $p < 0.001$). As concerns caregivers and their control group (control 2), differences were detected for employment and education, both higher in the control group ($\chi^2 = 7.52$, $p < 0.01$, and $\chi^2 = 4.61$, $p < 0.05$ respectively).

Materials

Data were collected through the following instruments:

A *socio-demographic questionnaire* provided information on participants' age, employment status, education level, and civil status. Clinical information including time from disease onset, level of disability, type of treatment and co-morbidities was collected for PwMS. Caregivers also reported their type of bond with PwMS.

Eudaimonic and Hedonic Happiness Investigation inventory (EHHI; Delle Fave et al., 2011, 2016). This mixed-method instrument allows researchers to collect qualitative and quantitative information on different components of well-being. Through open-ended questions, participants are asked to define happiness in their own words; to list the three future goals they consider most important, and the three things they consider most meaningful in their present life; and to briefly describe three situations associated with intense happiness during the last 6 months. In addition, participants are asked to rate on two sets of scaled items, ranging from 1 (extremely low) to 7 (extremely high), the levels of happiness and meaningfulness associated to 10 major life domains (work, family, standard of living, interpersonal relations, health, personal growth, leisure, spirituality/religion, community, and society) and to life in general.

Satisfaction with Life Scale (SWLS; Diener et al., 1985; Goldwurm et al., 2004). This widely used measure of well-being comprises five items on scales ranging from 1 (strongly disagree) to 7 (strongly agree). The items investigate the individuals' overall cognitive evaluation of their global life conditions and achievements.

Positive Affect and Negative Affect Schedule (PANAS; Watson et al., 1988; Terracciano et al., 2003). The instrument assesses the overall perceived intensity of positive and negative affect during daily life, through 10 items measuring components of positive affect (PA) and 10 items measuring components of negative affect (NA). Scales range from 1 (very slightly or not at all) to 5 (extremely).

TABLE 1 | Demographic characteristics of the four groups.

	PwMS ($N = 62$)	Control 1 ($N = 62$)	Caregivers ($N = 62$)	Control 2 ($N = 62$)
Age[a]	40.1 ± 9.7	40.5 ± 10.9	45.7 ± 12.0	45.7 ± 11.4
Age range	21–63	20–60	19–81	22–81
Gender (% Female)	69.35	69.35	58.06	56.45
EDUCATION (%)[b]				
High school or less	82.26	75.81	85.48	69.35
University	17.74	24.19	14.52	30.65
Work/study (%)	57.38	93.55	75.81	93.55
CIVIL STATUS (%)				
Married/cohabiting	66.13	51.61	77.42	72.13
Single/divorced/widowed	33.87	48.39	22.58	27.87

[a] *Means and Standard Deviations are reported.*

[b] *Education was dichotomized: "High school or less" includes elementary, middle, and high school; "University" includes graduation, post-graduation and PhD.*

Procedure

This study involved seven different academic and clinical institutions; therefore, the protocol was submitted to the ethical committees of each institution. After approval from all committees, participants with MS and their caregivers were recruited at six MS centers in different Italian regions, in the context of a broader project aimed at investigating clinical, psychological and relational aspects of MS (Bassi et al., 2014, 2016). PwMS were contacted by the centers' personnel during check-ups or by phone, and were invited to identify their primary caregiver. Data from participants in the two control groups were selected from a larger study that had been approved by the ethical committee of the first author's institution.

Upon their expression of interest in joining the study, participants received detailed information on the project from a researcher involved in the study. They signed an informed consent in compliance with Italian privacy rules, and were provided with the battery of questionnaires. They could inspect the questionnaires, pose general and specific questions to the researcher, and express their doubts and concerns. PwMS and their caregivers were free to decide whether to complete the questionnaires immediately at the MS center, or at a time and place convenient to them. They could hand in their responses, or send them via mail. Before data processing researchers removed the consent form from each battery of questionnaires, thus guaranteeing participants' anonymity throughout the phases of data coding, storing and analysis. Data were stored in password protected computers. Participation to both the MS related study and the general survey was voluntary, and participants were free to leave the study at any time. Persons with MS were explicitly assured that refusal to participate or withdrawal from participation in the study would in no way interfere with the long-term healthcare services they were receiving at the MS center.

Coding Procedure

Answers to the open-ended questions of the EHHI required an accurate coding work, using a coding system originally developed by Delle Fave et al. (2011), and gradually expanded through additions from various studies, in line with the bottom-up approach guiding the EHHI research project. In this coding system, answers are grouped into broad categories corresponding to the major life domains: work, family, standard of living, interpersonal relationships, health, personal/psychological life, leisure, spirituality/religion, society and community issues, and more general/unspecific life aspects. Multifaceted categories such as work, family, relationships, leisure, spirituality/religion, community/society and personal life are further subdivided into more fine-grained sub-categories (Delle Fave et al., 2013a). The categories family and interpersonal relationships were organized into the subcategories intrinsic value (e.g., having a family; a partner; children; friends), sharing (e.g., spending time with children; sharing life projects with partner; sharing good and bad experiences with friends), personal contribution (caring for elder parents; raising children; helping friends in need), family well-being/harmony, balance (e.g., health of family members; children's positive development; harmony in relationships) and personal reward (e.g., gratification from partner, children; gratitude expressions from siblings; satisfaction with relations). A similar partition characterizes community/society issues. In the psychological/personal life category, some subcategories refer to eudaimonic well-being dimensions (growth/engagement, purpose, competence/mastery, autonomy, self-actualization, meaning/value, harmony/balance, fullness/awareness, optimism), and others to hedonic well-being ones (satisfaction/achievement, positive emotions, and absence of negative feelings); a less specific subcategory, labeled as "positive experiences/states", includes answers generically referring to happiness as "inner well-being", "a stable state", or "a way of being."

The coding procedure for each answer comprises first the identification of a category in which the answer can be included, and then a specific numeric item to which the answer can correspond, if available. If an answer content does not fit any available item, a new item will be added to the category. As concerns happiness definitions, participants often report complex descriptions including different components of happiness. Each of these facets is treated as a specific semantic unit and coded separately; up to six answer units are retained for each participant. Based on the specific formulation of the questions concerning goals, meaningful things, and recent situations of most intense happiness, three answer units are retained for each question. In the present study, reliability in the coding process was established through the involvement of two expert coders providing independent ratings for each answer; divergences were clarified through a subsequent discussion; unsolved discrepancies and answers potentially requiring the inclusion of new items in the coding system were further discussed with the first author.

As concerns the scaled questions included in the EHHI, and the data collected through PANAS and SWLS, the numeric values corresponding to the perceived levels of each variable were reported. The EHHI items assessing the levels of happiness and meaningfulness associated with personal/psychological life are labeled as "personal growth"; this decision was originally based on the assumption that growth and development could best represent the positive and meaningful sides of this life domain (Delle Fave et al., 2013a).

Statistical Analyses

The analysis of qualitative data collected through the EHHI was aimed at identifying the life domains predominantly mentioned by participants across groups in their descriptions of happiness, major goals, meaningful things, and recent sources of intense happiness. Results were compared between groups (respectively PwMS and control 1, and caregivers and control 2). Since a high percentage of participants across groups indicated family as prominent goal, meaningful thing, and source of recent happiness, analyses were performed on family sub-categories. The same in-depth analysis was conducted for the psychological definitions of happiness.

Coded answers were first grouped into the corresponding categories and sub-categories; subsequently, considering that each participant provided more than one answer for each question, the number of participants reporting at least one answer in each category and subcategory was calculated across groups. This approach allowed us to compare the percentage of participants referring to each answer category and subcategory between groups through 2 × 2 frequency tables by means of the χ^2 procedure. The procedure was not considered as reliable when the number of participants in one or more cells was below 5. Through Spearman coefficients, correlations were then calculated between participants' distribution in answer categories and their demographic and group features. Logistic regression analyses allowed us to verify whether demographic or group features predicted a specific pattern of answers.

Quantitative data were first analyzed using descriptive statistics. Correlations of hedonic and eudaimonic dimensions of well-being with participants' group and demographic features were calculated through Pearson and Spearman coefficients. Hierarchical regression analyses provided information on the specific contribution of participants' group and demographic features to well-being dimensions.

Given the large number of group comparisons performed on quantitative and qualitative variables, we took a Bonferroni approach, adjusting the critical alpha value for significance to the number of t-tests and χ^2 comparisons performed on the same dataset (25 and 61 respectively). More specifically, to achieve $\alpha < 0.05$ with 25 t-tests, the alphas obtained from each data set had to score below 0.002 ($\alpha < 0.05/25$, two-tailed); in order to reject a null hypothesis, the test statistic had to exceed critical $t = 3.16$, corresponding to $p < 0.002$ with 122 degrees of freedom (for two groups, df $= N$ participants-2).

The same approach was used for frequency table comparisons ($N = 61$ in each data set). To achieve $\alpha < 0.05$ with 61 comparisons, the alphas obtained from the data set had to score below 0.0008 ($\alpha < 0.05/61$); in order to reject a null hypothesis, the test statistic had to exceed critical $\chi^2 = 11.34$, corresponding to $p < 0.0008$ with 1 degree of freedom, since df = (N columns-1) (N rows-1). As for correlations, regardless of the statistical significance only coefficient values equal to or higher than 0.30 were considered as adequate to interpret associations as meaningful (Hinkle et al., 2003). More specifically, values between 0.30 and 0.50 indicated low correlation, values between 0.50 and 0.70 moderate correlation, and values above 0.70 high correlation.

RESULTS

This section illustrates the qualitative and quantitative findings presented for each group separately, in order to allow for comparisons.

Qualitative Findings

The findings obtained from the open-ended questions of the EHHI are presented as percentages of participants across groups who provided at least one answer in the different categories and subcategories. The answers referring to psychological dimensions are grouped into the category labeled as "psychological definitions" for the definitions of happiness, and into the category "personal life" for goals, meaningful things and recent situations of intense happiness.

Happiness Definitions

Participants were invited to answer the question "What is happiness for you?" in their own words, without specific constraints. It is worth noticing that 9 PwMS and 8 caregivers either did not provide any answer, or stated that "happiness does not exist," while only 2 participants in each control group did so. Moreover, control groups 1 and 2 reported on average a higher number of answer units in their happiness definitions (2.65 and 2.80 respectively) compared with PwMS and caregivers (2.25 and 2.19 respectively). These differences were however not statistically significant. **Table 2** shows for each group the percentage distribution of the participants who provided at least one answer in the different categories and subcategories. Results of group comparisons are also reported.

Overall, a substantial similarity emerged between groups across most categories. Only two significant differences were detected between PwMS and control 1: a lower percentage of PwMS reported psychological definitions of happiness, and a lower percentage of controls reported family-related ones. Despite these overarching differences, the percentage distribution of participants across subcategories of both psychological and family-related definitions did not differ between the two groups. Among psychological definitions, participants predominantly referred to eudaimonic constructs, such as harmony/balance, self-actualization, personal growth, and optimism. Since a remarkably high percentage of participants specifically mentioned harmony/balance, related findings are reported separately in **Table 2**. As concerns family, most participants in both groups provided answers related to sharing (experiences, activities, projects). Personal rewards followed as subcategory; answers included receiving love expressions, support, acknowledgment from family members, and satisfaction with family. Fewer participants quoted the other domains, and a negligible percentage in both groups mentioned leisure, spirituality/religion, community/society issues and standard of living. A similar answer pattern was detected among caregivers and control 2, but no significant group differences emerged.

TABLE 2 | Definitions of happiness: percentage of participants mentioning each category and subcategory by group, and comparisons between groups.

	PwMS	Control 1	χ^2	Caregivers	Control 2	χ^2
Psychological definitions	54.72	93.33	22.51**	66.67	80.00	2.61
Harmony/balance	68.97	60.71	0.56	63.89	72.92	0.78
Other eudaimonic	24.14	39.29	1.95	25.00	41.67	2.52
Hedonic	41.38	46.43	0.19	27.78	33.33	0.30
Positive states	10.34	10.71		2.78	8.33	
Family	45.28	15.00	12.48*	51.85	28.33	6.58
Intrinsic value	28.00	11.11		13.79	11.76	
Sharing	48.00	55.56	0.69	44.83	58.82	0.83
Personal contribution	4.00	11.11		10.34	0	
Family well-being	12.00	22.22		34.48	23.53	
Personal reward	28.00	22.22		20.69	23.53	
Interpersonal relations	15.09	23.33	1.22	12.96	30.00	4.82
Health	11.32	8.33	0.29	12.96	15.00	0.10
Work	11.32	1.67		5.56	8.33	
Standard of living	1.89	1.67		7.41	3.33	
Leisure	5.66	0		0	1.67	
Spirituality, Religion	3.77	1.67		3.70	6.67	
Community, Society	7.55	5.00		0.00	3.33	
Life in general	0	13.33		3.70	8.33	
N participants[a]	53	60		54	60	

[a] *Each participant could provide more than one answer; Bonferroni adjusted alpha *$p < 0.05$; **$p < 0.01$; χ^2-values are not reported if the no. of participants in one or more cells falls below 5.*

Goals and Meaningful Things

While all participants in the control groups identified some important future goals, 2 PwMS and 4 caregivers did not. In addition, 59 participants in control 1 reported up to three goals compared with 45 PwMS ($\chi^2 = 11.68$, $p < 0.05$); similarly, 59 participants in control 2 reported up to three goals compared with 44 caregivers ($\chi^2 = 12.89$, $p < 0.05$). **Table 3** depicts the percentage distribution of the participants who provided at least one answer across categories in each group.

The majority of participants in all groups mentioned family, specifically referring to its intrinsic value (having a family; having or adopting children; having grandchildren; finding the right partner) and to the well-being of family members (physical health, self-actualization or goal achievement of children, grandchildren, partner, siblings, and parents). Health, work, and personal life were mentioned by lower percentages of participants; spirituality/religion and community/society were marginally represented, together with interpersonal relations. The only significant difference concerned the higher percentage of PwMS mentioning health, compared to control 1.

As regards the most meaningful things in the present life, 7 caregivers did not identify any, compared to 4 PwMS and no participants in the control groups ($\chi^2 = 13.22$, $p < 0.05$). Moreover, 60 participants in control 2 reported up to three meaningful things, compared with only 46 caregivers ($\chi^2 = 12.74$, $p < 0.05$). Nevertheless, as illustrated in **Table 4**, participants' percentage distribution across answer categories was largely overlapping across groups.

Participants in all groups almost unanimously quoted family as one of the most meaningful things in their lives

TABLE 3 | The most important future goals: percentage of participants mentioning each answer category by group and comparisons between groups.

	PwMS	Control 1	χ^2	Caregivers	Control 2	χ^2
Personal life	26.67	38.71	2.00	15.52	20.97	0.59
Family	68.33	67.74	0.004	79.31	64.52	3.23
Intrinsic value	51.22	52.38	0.01	39.13	40.00	0.007
Sharing	14.63	4.76		10.87	25.00	2.96
Personal contribution	12.20	23.81	1.89	23.91	12.50	1.84
Family well-being	31.71	35.71	0.15	43.48	45.00	0.02
Personal reward	4.88	0		0	2.50	
Interpersonal relations	5.00	6.45		1.72	6.45	
Health	53.33	22.58	12.28*	39.66	32.26	0.71
Work	45.00	59.68	2.63	37.93	58.06	4.86
Standard of living	6.67	16.13	2.69	29.31	22.58	0.71
Leisure	10.00	19.35	2.12	8.62	16.13	1.54
Spirituality, Religion	3.33	0.00		3.45	6.45	
Community, Society	3.33	3.23		0.00	14.52	
Life in general	3.33	12.90		6.90	14.52	
N participants^a	60	62		58	62	

*^{a}Each participant could provide more than one answer; Bonferroni adjusted alpha *$p < 0.05$; χ^2-values are not reported if the no. of participants in one or more cells falls below 5.*

TABLE 4 | The most meaningful things in present life: percentage of participants mentioning each answer category by group and comparisons between groups.

	PwMS	Control 1	χ^2	Caregivers	Control 2	χ^2
Personal life	12.07	20.97	1.71	25.45	17.74	1.03
Family	91.38	79.03	3.58	83.64	82.26	0.04
Intrinsic value	86.79	83.67	0.20	78.26	84.31	0.59
Sharing	1.89	12.24		13.04	5.84	
Personal contribution	1.89	6.12		6.52	3.92	
Family well-being	7.55	0		6.52	7.84	
Personal reward	7.55	2.04		2.17	1.96	
Interpersonal Relations	27.59	40.32	2.16	23.64	38.71	3.06
Health	48.28	30.65	3.90	32.73	33.87	0.02
Work	43.10	30.65	2.00	43.64	50.00	0.47
Standard of living	5.17	12.90		9.09	14.52	
Leisure	5.17	17.74		3.64	9.68	
Spirituality, Religion	8.62	6.45		5.45	9.68	
Community, Society	5.17	11.29		3.64	1.61	
Life in general	0	13.33		5.45	4.84	0
N participants^a	58	62		55	62	

^{a}Each participant could provide more than one answer; χ^2-values are not reported if the no. of participants in one or more cells falls below 5.

(specifically referring to its intrinsic value); progressively lower percentages of participants mentioned work, health, interpersonal relationships and personal life, without significant group differences. Spirituality/religion, leisure, standard of living and community/society were mentioned by less than 10% of the participants across groups.

Recent Situations of Intense Happiness

As illustrated in **Table 5**, only 39 caregivers (62.9%) provided answers to this question, while the remaining 23 (37.10%) could not remember any recent situation of intense happiness. This answer distribution pattern was significantly different from those detected for the other three groups ($\chi^2 = 29.93$, $p < 0.001$): only 12 participants (19.35%) among PwMs, 5 (8.1%) in control 2 and 2 (3.23%) in control 1 did not report recent situations of intense happiness.

No group differences instead emerged in the percentage of participants who provided at least one answer across categories. Family ranked first again; within this category, the majority of participants across groups referred to sharing positive events and experiences, such as anniversaries and other celebrations. A lower percentage of participants mentioned events promoting the well-being of family members (positive school and work achievements, disease recovery or health improvements). Leisure predominantly included the practice of arts and crafts, sports, travels and media fruition. Interpersonal relationships and work followed in rank across groups. A higher percentage of PwMS referred to health related situations, but after Bonferroni adjustment the difference with control 1 was not significant. Spirituality/religion and community/society were again reported by a negligible percentage of participants across groups, together with personal life and standard of living.

TABLE 5 | Recent situations of intense happiness: percentage of participants mentioning each answer category by group and comparisons between groups.

	PwMS	Control 1	χ^2	Caregivers	Control 2	χ^2
Personal life	6.00	8.33		0	10.53	
Family	74.00	73.33	0.006	82.05	78.95	0.14
Intrinsic value	10.81	22.73	1.99	12.50	15.56	
Sharing	70.27	68.18	0.04	56.25	46.67	0.69
Personal contribution	0	4.55		6.25	0	
Family well-being	35.14	29.55	0.29	50.00	55.56	0.23
Personal reward	13.31	13.64		3.13	11.11	
Interpersonal relations	22.00	36.67	2.79	20.51	22.81	0.07
Health	18.00	3.33	6.52	5.13	1.75	
Work	20.00	21.67	0.05	12.82	29.82	3.79
Standard of living	4.00	6.67		7.69	10.53	
Leisure	34.00	31.67	0.08	38.46	33.33	0.27
N participants^a	50	60		39	57	

^{a}Each participant could provide more than one answer; χ^2-values were not reported if one or more cells included less than 5 participants; answer categories cited by less than 5 participants across groups were excluded from analysis.

Quantitative Findings

Ratings of Happiness, Meaningfulness, and Hedonic Well-Being

Table 6 shows the mean ratings of happiness and meaningfulness across groups, and the results of the comparisons between PwMS and control 1, and between caregivers and control 2.

Participants across groups reported the highest levels of happiness and meaningfulness in the domains of family, health, interpersonal relations, life in general, and personal growth (with slight variations in domain order across groups). In contrast, spirituality/religion, community and society issues were associated with the lowest levels of happiness and meaningfulness across groups. The only difference between PwMs and control

1 concerned happiness with health, with the former reporting significantly lower values. The comparison between caregivers and control 2 highlighted two significant differences: Caregivers reported higher values of happiness in relation to community issues, and they attributed lower meaningfulness to the domain of leisure.

TABLE 6 | Levels of happiness and meaningfulness in life domains and comparisons between groups.

	PwMS *M* (*sd*)	Control 1 *M* (*sd*)	*t*	Caregivers *M* (*sd*)	Control 2 *M* (*sd*)	*t*
HAPPINESS						
Work	4.50 (1.96)	4.47 (1.31)	0.11	4.25 (1.72)	4.56 (1.25)	1.13
Family	5.90 (1.47)	5.60 (1.34)	1.18	5.56 (1.35)	5.84 (1.20)	1.13
Standard liv.	4.74 (1.34)	4.90 (1.30)	0.69	4.41 (1.28)	4.88 (1.28)	2.00
Relations	5.00 (1.55)	5.10 (1.34)	0.37	4.70 (1.42)	5.33 (1.06)	2.75
Health	4.37 (1.72)	5.45 (1.10)	4.17**	5.21 (1.14)	5.54 (1.30)	1.40
Personal Growth	5.05 (1.41)	4.92 (1.35)	0.52	4.87 (1.43)	5.18 (1.16)	1.31
Leisure	4.15 (1.48)	4.66 (1.45)	1.96	3.98 (1.72)	4.41 (1.66)	1.41
Spirituality	3.93 (2.09)	4.04 (1.70)	0.29	4.02 (2.04)	4.89 (1.56)	2.60
Community	3.64 (1.41)	4.23 (1.44)	2.18	3.56 (1.67)	4.49 (1.31)	3.29*
Society	3.71 (1.60)	3.62 (1.43)	0.29	3.70 (1.73)	3.72 (1.20)	0.08
Life in general	5.03 (1.31)	5.08 (1.12)	0.22	5.10 (1.25)	5.31 (0.93)	1.05
MEANINGFULNESS						
Work	5.28 (1.76)	5.21 (1.29)	0.24	5.36 (1.67)	5.66 (1.23)	1.12
Family	6.45 (1.08)	6.58 (0.82)	0.75	6.72 (0.77)	6.69 (0.82)	0.19
Standard liv.	5.15 (1.34)	5.03 (1.19)	0.50	4.93 (1.28)	5.29 (1.19)	1.60
Relations	5.56 (1.35)	6.02 (0.98)	2.13	5.34 (1.28)	5.97 (0.96)	3.10
Health	6.32 (1.02)	6.44 (0.84)	0.67	6.53 (0.88)	6.81 (0.44)	2.19
Personal Growth	5.74 (1.25)	5.91 (1.08)	0.85	5.51 (1.37)	5.90 (1.31)	1.64
Leisure	4.87 (1.52)	5.10 (1.17)	0.93	4.55 (1.42)	5.55 (1.04)	4.48**
Spirituality	4.01 (2.12)	4.00 (2.12)	0.04	4.61 (2.06)	5.08 (1.79)	1.35
Community	4.26 (1.37)	4.45 (1.67)	0.70	4.19 (1.60)	4.64 (1.45)	1.63
Society	4.29 (1.54)	4.25 (1.62)	0.14	4.26 (1.68)	4.88 (0.99)	2.53
Life in general	5.98 (1.06)	6.08 (0.80)	0.57	5.90 (1.25)	6.27 (0.85)	1.93
N participants	62	62		62	62	

*Bonferroni adjusted alpha *$p < 0.05$; **$p < 0.01$.*

More relevant differences were detected between groups as concerns the hedonic well-being dimensions. As illustrated in **Table 7**, both PwMS and caregivers scored significantly lower on positive affect compared with their respective control groups. Satisfaction with life ratings were significantly lower among PwMS than among participants in control 1. No group differences were instead detected for negative affect.

Demographic and Group Predictors of Well-Being Dimensions

Correlations and regression models were finally calculated within each group set-PwMS and control 1, and caregivers and control 2 respectively -, in order to investigate the role of group type and demographic features as predictors of qualitative and quantitative evaluations of well-being. Age, gender and marital status were not considered, as these characteristics did not differ between PwMS and control 1, and between caregivers and control 2. Attention was instead paid to education level and employment status, as group differences were detected for these two features, as reported in the comments on **Table 1**. **Table 8** illustrates Spearman correlations among predictors (demographics and group classification) and between predictors and well-being dimensions.

For the sake of synthesis, analyses were performed only for the dimensions showing significant differences between groups. As reported in the method section, correlations higher than 0.30 were deemed as meaningful. Overall, group classification showed the most numerous and strongest correlations with the well-being dimensions. In the PwMS/control 1 group set, only employment status showed a low positive correlation with satisfaction with life; together with group classification, it showed a low positive correlation with happiness with health as well. No meaningful correlations were instead identified for education level. In the caregivers/control 2 group set, group type showed low to moderate correlations with most well-being dimensions, except for satisfaction with life and negative affect. A low positive correlation emerged between education level and positive affect, while employment status did not show any relevant correlation with well-being dimensions.

TABLE 7 | Levels of affective and cognitive dimensions of hedonic well-being, and their comparison between groups.

	PwMS *M* (*sd*)	Control 1 *M* (*sd*)	*t*	Caregivers *M* (*sd*)	Control 2 *M* (*sd*)	*t*
Positive affect	3.01 (0.81)	3.59 (0.57)	4.62**	3.01 (0.71)	3.87 (0.48)	7.83**
Negative affect	2.15 (0.93)	2.51 (0.82)	2.29	2.03 (0.89)	2.22 (0.66)	1.35
Satisfaction with life	3.80 (1.55)	4.65 (1.30)	3.30*	4.14 (1.51)	4.87 (1.15)	3.06
N participants	62	62		62	62	

*Bonferroni adjusted alpha *$p < 0.05$; **$p < 0.01$.*

TABLE 8 | Correlations among demographic and group predictors, and between predictors and variables showing significant differences in the two group sets.

	Employment	Education	Group
PwMS/CONTROL 1			
Employment	–	0.11	0.42***
Education	0.11	–	0.08
Positive affect	0.28**	0.22*	0.39***
Negative affect	−0.004	−0.15	0.20*
Satisfaction with life	0.35***	0.24**	0.29**
Happiness with health	0.31*	0.05	0.33*
Happiness def.—psychological	0.16	0.13	0.49***
Happiness def.—family	−0.04	−0.09	−0.29**
Goals—health	−0.19*	−0.03	−0.30***
CAREGIVERS/CONTROL 2			
Employment	–	0.12	0.25
Education	0.12	–	0.19*
Positive affect	0.26**	0.30***	0.58***
Negative affect	0.11	0.03	0.12
Satisfaction with life	0.16	0.15	0.27**
Happiness with community	0.06	0.21*	0.30**
Leisure meaningfulness	0.23**	0.12	0.38***

**p < 0.05; **p < 0.01; ***p < 0.001. "Group" is a dummy variable with 1 for PwMS or caregivers and 0 for the respective control groups.*

Based on these results, linear regressions with stepwise selection were carried out to investigate the role of employment status, education level and group classification as predictors of the hedonic well-being dimensions: positive affect, negative affect, and satisfaction with life. In addition, linear regressions (with scaled outcome variables) and binary logistic regressions (with categorical outcome variables) were performed to assess the role of demographic features in the findings obtained through the EHHI for which group differences were identified. More specifically, for the PwMS/control 1 group set regressions were conducted for happiness with health, psychological and family-related definitions of happiness, and health-related goals. For the caregivers/control 2 group set regressions were conducted for happiness with community and meaningfulness of leisure.

As concerns PwMS and control 1, for the dependent variable positive affect the two predictors entered in the model were group type and education level, together explaining 18% of the variable variance ($r^2 = 0.143$, $F = 20.13$, $p < 0.001$, and $r^2 = 0.039$, $F = 5.41$, $p < 0.05$ respectively). Negative affect was significantly predicted by group type ($r^2 = 0.041$, $F = 5.27$, $p < 0.05$), however explaining only 4.4% of the variable variance. Satisfaction with life was significantly predicted by employment status and education level ($r^2 = 0.121$, $F = 16.69$, $p < 0.001$, and $r^2 = 0.042$, $F = 5.82$, $p < 0.05$ respectively), but not by group type; the two significant predictors together explained 16.3% of the variable variance. The level of happiness with health was predicted by group type ($r^2 = 0.126$, $F = 17.47$, $p < 0.001$), and employment status ($r^2 = 0.048$, $F = 7.04$, $p < 0.01$), together explaining 17.4% of the variable variance. The distribution of participants citing psychological and family-related happiness definitions was predicted by group type only ($B = -1.27$, $\mathrm{OR} = 0.079$, Wald $\chi^2 = 22.63$, $p < 0.001$; and $B = 0.67$, $\mathrm{OR} = 3.820$, Wald $\chi^2 = 9.04$, $p < 0.01$ respectively). Group type was also the sole predictor of health-related goals distribution ($B = 0.66$, $\mathrm{OR} = 3.783$, Wald $\chi^2 = 11.20$, $p < 0.001$).

In the caregivers/control 2 group set, positive affect was significantly predicted by group type and education level, together explaining 36.9% of the variable variance ($r^2 = 0.334$, $F = 61.25$, $p < 0.001$, and $r^2 = 0.035$, $F = 6.75$, $p < 0.05$ respectively); no significant predictors were instead identified for negative affect. Group type emerged as the only significant predictor for the three remaining variables: satisfaction with life ($r^2 = 0.071$, $F = 9.36$, $p < 0.01$, explaining 2.2% of the variable variance); happiness with community ($r^2 = 0.088$, $F = 10.80$, $p < 0.01$, explaining 2.6% of the variable variance) and leisure meaningfulness ($r^2 = 0.141$, $F = 20.04$, $p < 0.001$, explaining 4.2% of the variable variance).

DISCUSSION

This study aimed at investigating different facets of well-being among persons with MS and their caregivers from an integrated perspective. At the conceptual level, the eudaimonic dimensions of goal pursuit and meaning-making were jointly investigated with the hedonic ones of affect and life satisfaction. In addition, based on a bio-psycho-social and ICF informed perspective, the investigation of well-being was contextualized within the major life domains, and findings were compared with those reported by two groups of participants sharing similar demographic features, but not exposed to experiences of chronic illness or caregiving. At the empirical level, the adoption of a mixed method approach gave participants the opportunity to freely describe their present evaluations and future expectations.

The Private Context of Well-Being: A Shared Perspective

The information collected through the EHHI allowed us to contextualize individuals' perceived well-being within the major life domains. Regardless of group inclusion, in both qualitative and quantitative answers participants primarily referred to few life domains, basically circumscribed to the private sphere. Family, personal life and health distinctly emerged across groups as the predominant contexts of happiness, meaningfulness and goal investment. A substantially lower relevance was attributed to the broader contexts of work and interpersonal relations; finally, the public domains of community and society, and the transcendent sphere of spirituality/religion were almost absent from participants' qualitative answers, and they scored lowest in rank on the scales. Overall, these findings largely confirmed previous EHHI studies conducted across countries on adult samples belonging to the general population (Delle Fave et al., 2011; Delle Fave et al., 2013b).

Across groups, participants who referred to family in their definition of happiness mainly focused on the dimension of sharing. This finding is consistent with the models emphasizing the primacy of relational connectedness in humans (Richardson, 2012). As specifically concerns persons with MS and their caregivers, these results are also consistent with the evidence of a shared process of adaptation to illness, based on communal growth and search for meaning (Pakenham, 2005; Pakenham and Cox, 2009; Ackroyd et al., 2011). Among the psychological definitions of happiness, eudaimonic dimensions were predominant; inner harmony was reported by the highest percentage of participants across groups, followed by self-actualization and personal growth. This finding, consistent with previous evidence (Delle Fave et al., 2016), provides further support to the view of happiness as connectedness, at both the inner and relational levels (Kjell, 2011). Most of the research hypotheses concerning happiness definitions were however not confirmed. Against expectations, the percentage of PwMS referring to health and the percentage of caregivers referring to family were not significantly higher, compared to their respective control groups. Similarly, against expectations no difference was detected between caregivers and control 2 in the percentage of participants quoting leisure. Only two unexpected differences emerged between PwMS and control 1: a lower percentage of PwMS provided psychological definitions of happiness, and a higher percentage mentioned family related ones. In both cases, group classification emerged as the only significant predictor. The overall relevance of family has been widely documented among persons with MS (Ryan et al., 2007; Pakenham, 2008a). It is however worth noting that in this study persons with MS, similarly to participants in the other groups, emphasized sharing rather than receiving support and rewards from family—as it could be expected from individuals facing a progressively increasing dependence on their caregivers.

Across groups, the majority of participants mentioned family as a major future goal, primarily referring to its intrinsic value and to the well-being of family members. Work followed in rank, while marginal relevance was attributed to extra-family relations, and to social, community and spirituality issues. As hypothesized, a significantly higher percentage of persons with MS quoted health, compared to control 1; group classification was the only predictor of this result. All the other hypotheses were not confirmed. It is worth noticing that, compared to their respective control groups, a significantly higher percentage of PwMS and caregivers reported less than three future goals. This result can be related to the perceived uncertainty highlighted in the MS literature, and in studies involving people with other chronic diseases (Bensing et al., 2002; Tams et al., 2016). Uncertainty leads individuals to focus on the present rather than on future planning; this aspect can be even more relevant in diseases such as MS, which entails a higher margin of unpredictability, compared with other chronic and degenerative conditions (Alschuler and Beier, 2015).

Family also emerged as a key meaningful thing; the vast majority of participants across groups referred to its intrinsic value—having a family, a partner, children, siblings were identified as valuable components of life *per se*. The primacy of intimate relationships in the meaning making process largely confirmed previous evidence obtained with a variety of samples (Baumeister and Leary, 1995; Lambert et al., 2010; Taubman – Ben-Ari et al., 2012). Work, health and interpersonal relations followed across groups, while only a negligible percentage of participants mentioned spirituality/religion, community/society, and leisure. A significantly higher percentage of caregivers reported less than three meaningful things compared to control participants; this difference may be related to the narrower range of daily opportunities that characterizes the caring role (Mausbach et al., 2011).

When describing recent situations of intense happiness, most participants across groups referred to events shared with the family: birthdays, marriages, holidays, but also receiving a good news concerning family members. Leisure and interpersonal relationships, following in rank, were mentioned by a relatively high percentage of participants across groups only in the context of this answer. Finally, and in line with expectations, health was quoted almost exclusively by PwMS. It is also important to note that over one third of the caregivers could not identify any recent situation of intense happiness.

Extraordinary Circumstances, Ordinary Experiences of Well-Being

Research has repeatedly emphasized the negative psychological consequences of living with MS as a person or a caregiver, especially at the emotional level. Our findings were only partially consistent with the literature, rather emphasizing the "ordinariness" of PwMS and caregivers (Saravanan et al., 2001) in their quantitative ratings of global and domain-related well-being.

As concerns domain-related happiness and hedonic well-being (assessed as positive and negative affect, and satisfaction with life), PwMS scored significantly lower than control participants in happiness with health (thus confirming expectations) and in positive affect. Although participants' group emerged as the strongest predictor of the two variables, unemployment further contributed in predicting lower happiness with health, and lower education level in predicting lower positive affect. PwMS also reported significantly lower levels of life satisfaction, but employment status and education, rather than presence of MS, emerged as significant predictors of this result. No differences instead emerged between PwMS and control participants for negative affect, despite the (weakly) significant predictive role of group classification. To this purpose, it is worth mentioning that negative affect values were on average higher in the control group. The comparison between caregivers and their control group highlighted a significant difference in positive affect, with the former scoring significantly lower. Group type was the strongest predictor of positive affect; the additional though limited contribution of lower education level replicated the findings detected between PwMS and control 1. Group type emerged as significant predictor of satisfaction with life as well, though with marginal explanatory relevance.

The lower levels of positive affect reported by PwMS and caregivers were consistent with the literature highlighting the emotional burden of chronic disease; nevertheless, education emerged in both cases as an additional environmental predictor, suggesting that emotions are—at least partially—socially constructed. In particular, this finding provides support to the role of education as a major objective indicator of hedonic or subjective well-being, regardless of health conditions (Kroll and Delhey, 2013). The same consideration can be made as regards satisfaction with life, whose levels were not predicted by group type, but by the social opportunities derived from employment status and education, thus confirming previous evidence (Oishi and Diener, 2014). The lack of group differences in negative affect levels confirmed instead the conceptual and empirical independence of positive and negative affect (Seib-Pfeifer et al., 2017), as well as the importance to consider well-being and ill-being as partially independent domains of experience, rather than as opposite poles of a single continuum (Keyes, 2007).

In this study eudaimonic well-being was quantitatively assessed as the level of meaningfulness associated with different life domains. Findings did not support the study hypotheses, as PwMS and caregivers did not differ from control groups in family and health related meaningfulness. An unexpected difference emerged between caregivers and control 2, with the former attributing significantly lower meaningfulness to the leisure domain. This result, solely predicted by participants' group, can be related to the restrictions in daily life opportunities experienced by family caregivers (Becker, 2011). Since under these circumstances leisure activities get often sacrificed first, downplaying their meaning and relevance can help caregivers adjust to the related constraints (Pakenham and Cox, 2009).

Patients and Caregivers: Social Assets Beyond Clinical Labels

This study highlighted that overall persons with MS and their caregivers do not differ from healthy people in their experience of hedonic and eudaimonic well-being. These findings are consistent with the literature showing that individuals and families mobilize a variety of resources in order to adjust to disease conditions. At the same time, they offer further suggestions. Not only have these people adjusted to disease; they pursue value-driven goals, cultivate inner harmony and balance, invest their energies in meaningful activities and relationships. Persons with MS do not seem to be primarily focused on their own health and related needs; they are rather actively engaged in sharing experiences and collaborating to the promotion of family well-being (Bogosian et al., 2017). As for caregivers, their life trajectory—although forcefully disengaged from extra-family socialization and leisure—is grounded in personal and relational values, despite the costs emerging at the hedonic well-being level. In general, the present study did not highlight group differences in participants' level of engagement in public roles and social activities, thus showing that active involvement in community and society is related to cultural dimensions rather than health conditions.

Overall, these findings can be considered as a provocative claim for a change in perspective, as concerns the social representation of health. This claim, consistent with the ICF model, is based on theoretical and empirical evidence. As highlighted by studies investigating resilience (Walsh, 2015), individuals and families experiencing chronic diseases should be valued and appreciated for their ability to develop personal and communal competences, rather than being considered as weak and low-performing members of the society. Their psychological and relational competences, laboriously built over time, could be rather shared to the benefit of others. Community Based Rehabilitation (CBR) programs are rooted in this view, aimed at empowering persons with disabilities through the promotion and acknowledgment of their active community role (Khasnabis et al., 2010). Across countries, people experiencing disease are often founders or active members of associations, promoters of fundraising campaigns in support of biomedical research, civil rights activists. Therefore, health professionals could approach them as experts who can offer first-person knowledge of a specific condition, and not only as patients to treat and caregivers to instruct (Greenhalgh, 2009). Their social involvement could be extended to educational programs and other community initiatives, allowing them to share their resources and enjoy recognition as full-fledged members of the society. Although this change implies an overall revision of the health culture, the advantages would be remarkable, as efforts in this direction could lead to a more inclusive and participative society.

Study Strengths and Limitations

The major strength of the present study is the investigation of hedonic and eudaimonic dimensions of well-being among persons with MS, their caregivers and two control groups through a mixed method approach. The complexity of this research design, too often neglected by researchers (Morales-Gonzales et al., 2004), allows for contextualizing scaled ratings within qualitative, semantically richer answers. In this study, the domain-related ratings of happiness and meaningfulness could be combined with a fine grained description of the same domains, their present and future relevance, and their relation to well-being. The findings provided an integrated representation of the daily activities, contexts and relationships in which participants' meaning-making process, goal pursuit and happiness experiences took place. To our knowledge, no studies of this kind are available in the psychological literature on MS, and more generally on chronic diseases.

This study has limitations as well. First of all, the cross-sectional design prevented from identifying causal relationships among variables. Although the circumscribed observation field allowed for an in-depth analysis of well-being, the negative impact of disease and caregiving was not explored. The sample sizes were relatively small. Disability levels of the persons with MS did not reflect the whole range of progression stages: the inclusion of participants with very severe disability could lead to different results, at both the qualitative and quantitative levels. All participants were Italians, thus belonging to a specific socio-cultural context: this feature increases reliability in the comparison of results across samples, but it prevents from generalizing results to countries characterized by different healthcare, welfare and value systems.

Future Directions

The findings from this study shed light on participants' experience of well-being, in the context of their daily activities and social roles. It is however important to consider that both individuals and their contexts are dynamic entities, changing over time while interacting with each other; this further level of complexity, endorsed by the ICF model, can be satisfactorily evaluated only through the collaboration of researchers from different disciplinary fields. As specifically concerns psychology, relevant contributions could derive from community and cultural psychology, with their focus on the interaction dynamics between socio-cultural practices, individual experience and collective behaviors (Christopher and Hickinbottom, 2008; Di Martino et al., 2017).

A stronger interdisciplinary collaboration is especially needed in the light of a specific result emerged from this study: persons with MS, their caregivers and the control groups reported low levels of happiness and meaningfulness in community and society issues; in addition, these domains were almost absent from their lists of goals, meaningful things, and occasions for happiness. This finding may be interpreted as an alarming sign of civic disengagement. However, it confirms evidence obtained in other studies conducted in individualistic societies (Delle Fave et al., 2011). We consider this result as a general warning for researchers, practitioners and policy makers, highlighting the pressing need to promote a culture of interconnectedness (Prilleltensky, 2005), in order to contrast the deterioration of community networks presently emerging across nations.

AUTHOR CONTRIBUTIONS

ADF and MB provided substantial contributions to the conception and design of the work; the acquisition, analysis, and interpretation of data; drafting the work and revising it critically for important intellectual content; final approval of the version to be published; and agreement 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. All the other co-authors provided substantial contributions to the acquisition and interpretation of data; critical revision of the work at the conceptual level; final approval of the version to be submitted; and agreement 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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Family Function and Self-esteem among Chinese University Students with and without Grandparenting Experience: Moderating Effect of Social Support

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This study examines the association between family function and self-esteem of Chinese university students with grandparenting experience, and explores the moderating effects of social support in this link. Two thousand five hundred thirty university students (1372 males and 1158 females) from a Chinese university completed the Perceived Social Support Scale, the Rosenberg's Self-esteem Scale, and the Family Assessment Device (FAD). Six hundred and forty-five (25.69%) students reported grandparenting experience and they reported lower scores on self-esteem and social support than the students raised only by their parents. The grandparenting group scored higher on such dimensions of family functioning as Communication, Role, Affective Involvement, Affective Responsiveness, and General Family Function (GF) than their counterpart group. For both groups, self-esteem scores were positively correlated with social support scores, while negatively correlated with FAD all sub-scale scores. Hierarchical regression analysis showed that for the students with grandparenting experience the social support moderated the relationship between GF and self-esteem. When students reported a high level of social support, those with low GF score reported higher scores in self-esteem than those with low self-esteem. However, in case of low social support, there were no differences in self-esteem between groups with high and low GF scores. These findings suggest that social support plays a positive role to relieve the adverse impact of poor family function on self-esteem of the adolescents with grandparenting experience. In addition, the significance and limitations of the results will be discussed.

Keywords: social support, self-esteem, family function, Chinese university students, grandparenting

INTRODUCTION

Since the 1980s, due to the radical development of the society resulting from China's "reform and opening," there were many significant changes within the Chinese family. One of the major changes was the introduction of grandparenting which became an important family rearing pattern. Grandparenting refers to the situation of grandparents rearing and educating their grandchildren.

A nationwide survey in China has shown that in the largest cities, such as Beijing, Shanghai, and Guangzhou, among children under the age of six those accepting grandparenting accounted for more than 50% (Wu, 2010). Therefore, grandparenting has become an important regular condition of children's growth in China. In the European countries and United States of America, the reasons for grandparenting are mainly based on the parents' marital status (divorce, separation, premarital pregnancy, etc.) and economic reasons, while in China there are unique reasons for this phenomenon: first of all, Chinese traditional culture has formed strong and unique family ethics, as part of this moral code of conduct, grandparents view raising grandchildren as their responsibilities, and through this it is considered to obtain happiness and satisfaction. In addition, the young parents spend most of the time and energy on working or further studying, which means that they cannot fully take care of their children, therefore the children are handed over to the grandparents to be raised. Prevailing studies on grandparenting are mainly concerned with the impact on the physical and mental health of grandparents (Muller and Litwin, 2011; Grundy et al., 2012; Arpino and Bordone, 2014; Burn et al., 2014; Zhang and Han, 2016). Other research is focusing on the influence of grandparenting on the psychological and social development of the children, reporting a negative effect on their self-esteem. The children being raised by their grandparents show more psychological problems and difficulties with social adaptation in family and school life (Qi, 2011; Edwards and Daire, 2006; Li and Li, 2006; Zhang and Lu, 2017). Compared with children who were raised by their parents, they showed more emotional and behavior problems (Edwards, 2006; Edwards and Daire, 2006; Wang, 2007; Qi, 2011; Zhang and Lu, 2017), they were shy and had little skills in communicating with others (Fuller-Thomson and Minkler, 2005; Xin, 2010; Han and Guo, 2016). However, the main research objects in this study were pre-school or primary school children who currently accept grandparenting. Due to a lack of retrospective studies on the influence of grandparenting on adolescents and young adults, we raised the following question: Do children with a background of being brought up by their grandparents up to adolescence or adulthood show different levels of self-esteem compared to their peers without the experience of grandparenting?

Self-esteem refers to the evaluation and experience related to self-value, the perception of self ability as well as the acceptance of the whole self, which an individual obtains during the process of socialization (Rosenberg, 1965; Leary and MacDonald, 2005). According to Maslow (1987) a person with high self-esteem feels more confident, is more competent, and hence more productive; while an individual with low self-esteem often feels inferior, hopeless, even desperate and tends to get neurosis. An individual with high self-esteem has a high level of mental health status and self-harmony (Peng et al., 2013). Moreover, self-esteem could positively affect the general well-being (Shim et al., 2013; Tian et al., 2013). Self-esteem might also contribute to a higher success motivation (Pang et al., 2011). In addition, it might affect the ability to achieve goals or increase the ability to cope with problems, while low self-esteem may lead to avoidance (Hendy et al., 2003). Self-esteem can also affect decision-making, which may have an impact on the individual's entire life (D'Amico and Cardaci, 2003). Self-esteem is one of the basic needs for all individuals especially in adolescence. College Students are in the transitional period from adolescents to adults, they inevitably experience great changes during the shift from middle school students to college freshmen. They should independently manage their college life and cope with challenges. Both family and society put high expectations on them, and at the same time they must adjust to the new environment. Although their intellectual and physical development has already been completed, their psychological development is not yet fully mature, and in addition their self-consciousness is still developing. Therefore the college students are easily to experience inner conflicts and ambivalence. Self-esteem as an important trait of the self-system has a positive impact on the mental health, personality development and social adaptation of the individual (Leary and MacDonald, 2005).

The formation and development of one's self-esteem is not only the result of the socialization process, but also the product of the interaction with one's sociocultural and familial environment as well as school education. Most of the research findings demonstrated that children's self-concepts are related to parent-child relationships, and family characteristics (Brown et al., 2009). Brown et al. (2009) found that the interplay of familial relationships has an impact on a child's developing sense of self: In the families that show more harmonious interactions, the children describe themselves as being more adventurous. However, it is more likely that in the families which experience high levels of discord the children view themselves as more fearful and less agreeable. The results of another study showed that the parents of the children with low self-esteem tend to punish the children severely, whereas in the group of the children with high self-esteem the family exhibited a more democratic way of interaction (Peng and Fan, 2007; Heaven and Ciarrochi, 2008). Various researchers examined the relationship between family function and self-esteem, they consistently found that there is a close relationship between self-esteem and family function, and the results have cross-cultural consistency: Wang et al. (2010) for example found that there is a significant correlation between the development of adolescent self-esteem and parental rearing styles. Other data showed that there is a significant positive correlation between a high level of family intimacy as well as emotional expression and a high level of self-esteem and self achievement of adolescents (Li et al., 2006); the extent of closeness of family members and the parental expectations might affect the self-esteem and self-concept of the college freshmen (Wu and Ye, 2009). However, grandparenting experience might affect the parent-child relationship. Li (2010) examined the parent-child relationship of college students with grandparenting experience during childhood and found that intimacy between parents and children was lower compared to those with non-grandparenting experience; the students felt distant from their parents and the parents did not trust in their children's capability due to a lack of initiative communication between parents and children. Hence, it is necessary to explore whether and how the family function of the students with grandparenting experience influences their self-esteem? Compared with students solely raised by their parents, in case of existing factors, which ones do have a moderating effect on the relationship between family function and self-esteem of the students with grandparenting experience?

Social support can be defined as an individual's perception or experience that their social network will provide effective emotional and substantial support during the times of need (Wills, 1991). The consistent finding in many researches show that social support is positively related to self-esteem (e.g., Barrera and Garrison-Jones, 1992; Wentzel, 1994; Sweeting and West, 1995; Goodwin et al., 2004; Birndorf et al., 2005; Zhang, 2007; Denissen et al., 2008). In adolescence significant changes to social relationships will happen. In particular, the transition to the university needs more and more independence from the family and the development of a new social network. Data showed that there was a positive relationship between the utilization of social support and self-esteem of university students (Peng et al., 2003; Ma, 2005). Moreover, it is suggested by some authors that during early adolescence the peer relationships take on increased importance (Furman and Buhrmester, 1992). The results of previous studies (Fan and Fang, 2004; Lian et al., 2016) showed that support from the peer group significantly influences the self-esteem of adolescents, especially the emotional support positively predicted the level of self-esteem.

Further data demonstrated that adolescents' social relations outside the family might be a moderator for the connection of relationships with parents and adolescent adjustment. For example, Beyers and Seiffge-Krenke (2007) longitudinal study found that adolescents' relationships with parents influenced later problems of behavior when interacting with friends and romantic partners. To our knowledge, no studies exist which examine the moderator effect of social support on the relationship between family function and self-esteem. Therefore, in the current study the relationship between the three factors was explored. The aim of this study is to examine the difference in family functioning and self-esteem between university students with and without grandparenting experience and to investigate the moderating effect of social support in the relationship between family functioning and self-esteem based on retrospective data from a group of university students.

Hypothesis

First, we assumed that grandparenting experience would have influence on family functioning and level of self-esteem. Second, we assumed that social support would have moderating effect in the relationship between family functioning and self-esteem in the students with grandparenting experience; Third, from a perspective of moderation model, we hypothesized that in the relationship between family function and self-esteem it would show a significant difference between persons with high social support and low social support.

MATERIALS AND METHODS

Participants

A sample of 2530 freshmen and sophomores (1372 were males, 1158 were females) was recruited at a university in Shanghai, a large city in the eastern part of China. The students' ages ranged from 16 to 20 years, with an average of 18.79 years ($SD = 0.02$). Written informed consent was obtained from all the participants prior to the study. The written informed consent from the caretakers on behalf of the participants under 18 years was also obtained.

Measures

Family Assessment Device (FAD)

The Family Assessment Device (FAD) is a well established measure to assess family functioning based on the theoretical concept of the McMaster Model of Family Functioning (MMFF). The term "family functioning" refers to the ability of the family to work together as a unit to satisfy the basic needs of its members (Ryan and Keitner, 2009). The MMFF emphasizes the interrelatedness of the family members and the family system, and represents six dimensions of family functioning that are considered of high relevance in clinical practice. It is made up of seven scales that assess six dimensions of the MMFF as well as the General Family Functioning. Among the six dimensions there are Problem Solving (PS), Communication (CM), Roles (RL), Affective Involvement (AI), Affective Responsiveness (AR), and Behavioral Control (BC) (Shek, 2001): (1) problem solving (the capability of the family to cope with problems in order to keep effective family functioning); (2) CM (the way of exchanging information between family members); (3) RL (whether the family assign certain tasks to guarantee implementation of family functions); (4) AR (to which extent the family members emotionally react to stimulation); (5) AI (to which extent the family members show concern to each other); and (6) behavior control (the behavioral models that the family establishes to cope with stressful situations). The instrument consists of 60 statements about a family; the requirement is to rate to which extent the description of each statement accords with their own family. All items are rated on a 4-point Likert scale, with answer choices from "1 = strongly agree" to "4 = strongly disagree." Sample items for example include, "You can't tell how a person is feeling from what they are saying," "If someone is in trouble, the others become too involved," "Making decisions is a problem in our family," and, "We confront problems involving feelings." The higher scores the respondents rate, the worse levels of family functioning indicate. The validity and reliability of the Chinese FAD has been demonstrated by Shek (2001, 2002). The test-retest reliability is 0.53–0.81, and coefficient alpha ranges from 0.53 to 0.94.

Rosenberg Self-esteem Scale

The Rosenberg Self-esteem Scale (SES; Rosenberg, 1965) was applied to assess self-esteem. This instrument consists of 10 items. It is a self-report measure of global self-esteem. All items are rated on a 4-point Likert scale with answer choices from "1 = strongly disagree" to "4 = strongly agree." Sample items for example include "I am able to do things as well as most other people." and "I take a positive attitude toward myself." The total scores range from 10 (low level of self-esteem) to 40 (high level of self-esteem). Good levels of reliability and validity of the Chinese RSES have been demonstrated in the previous studies (Zhao et al., 2012; Kong and You, 2013). In this study, the Cronbach alpha coefficient for the RSES was 0.84.

Perceived Social Support Scale

The Perceived Social Support Scale (PSSS) was developed by Zimet et al. (1988). The scale consists of 12 items designed to assess perceived social support from three sources: Family, Friends, and Significant Others. Items are scored on a 7-point rating scale ranging from 1 (very strongly disagree) to 7 (very strongly agree) with possible scores ranging from 12 to 84. The current total perceived social support status can be measured according to the total score for the 12 items. The Chinese version of this scale has been widely used and exhibited good level of validity and reliability (Wang et al., 1999; Kong and You, 2013). In this study, the Cronbach's a coefficient for the PSSS was 0.72.

Grandparenting Experience

The definition of grandparenting experience in this study refers to that individual during their childhood left their parents and was raised by grandparents for at least 1 year. The grandparenting experience was evaluated using a questionnaire designed by the authors that included sex, age, whether left parents and was raised by others in the childhood, the duration and caregiver. The students who reported that they left their parents and were raised by grandparents for at least 1 year during their childhood were defined as grandparenting group.

Procedure

All of the participants volunteered to participate at the end of the mental health education course. They signed an informed consent form at first. Participants were instructed to complete a questionnaire survey including measures of family functions, self-esteem, and perceived social support as well as grandparenting information in a quiet classroom environment after informing consent. It took approximately 20 min for the students to complete all the instruments.

Data Analysis

All statistical procedures were conducted using the Statistical Package for the Social Sciences (SPSS 17.0). Independent sample *t*-test was used to compare the level of self-esteem, family function score and social support score between the two groups with and without grandparenting experience; Pearson correlation was calculated to explore how levels of self-esteem and family functions as well as perceived social support were related; Hierarchical regression analysis was applied to examine the moderating effect of social support on the relationship between family function and self-esteem.

RESULTS

Demographic Background of the Participants

Six hundred and forty-five (25.69%) students reported that they left their parents and were raised by grandparents for at least 1 year during their childhood; 1764 (70.25%) students were brought up only by their parents; and 121 (4.06%) students reported that they had experience of being raised by other persons, who were not their own parents or grandparents. Among the students who were only raised by their parents the percentage of only children was higher compared to the grandparenting group (80% vs. 74.1%; $X^2 = 9.648$, $P = 0.002$); there was no statistic difference in proportion of gender between two groups (Parents-raising: male 53.5% vs. Grandparenting: male 57.1%; $X^2 = 2.463$, $P = 0.117$).

Comparison of the Level of Self-esteem, Family Function Scores and Social Support Scores between the Two Groups with and without Grandparenting Experience

The students with grandparenting experience scored lower on self-esteem and total score of social support as well as support from family compared to the students raised only by their parents, while on the sub-scales of family function such as CM, RL, AR, AI, and GF the grandparenting group scored higher than their counterpart group (see **Table 1**).

Correlation between Family Function, Social Support and Self-Esteem

In both of the two groups there are obviously significant positive correlations between self-esteem and social support and obviously significant negative correlations between self-esteem and all the sub-scales of family function. The sub-scales of family function are positively inter-correlated (see **Table 2**).

Hierarchical Regression Analysis on the Moderating Effect of Social Support on the Relationship between Family Function and Self-esteem

In order to test the moderating effects of social support on the relationships between family function and self-esteem, hierarchical regression procedures were performed as recommended by Baron and Kenny (1986). Before testing the moderating effects, the two predictor variables (social support and family function) were standardized to reduce problems associated with multicollinearity between the interaction term and the main effects (Frazier et al., 2004). Thus, *z*-scores were calculated for social support and family function.

The hierarchical multiple regression analyses were conducted in each group. In the hierarchical regression model, the order of entry was as follows. At step 1, the control variables (age, gender, and "only child status") were entered into the regression

TABLE 1 | Comparison of the level of self-esteem, social support and family function scores between two groups ($\bar{x} \pm s$).

Variables	Grandparenting group (*n* = 645)	Parents-raising group (*n* = 1764)	*t*-value	*P*-value
Self-esteem	29.90 ± 4.256	30.28 ± 4.132	2.000	0.046
Social support				
Total	66.75 ± 10.523	67.90 ± 10.319	2.399	0.017
Family	22.41 ± 4.342	23.05 ± 4.127	3.3.15	0.001
Friends	22.39 ± 4.143	22.61 ± 3.979	1.190	0.234
Others	21.94 ± 3.948	22.21 ± 3.805	1.544	0.123
Family function				
PS	12.56 ± 2.091	12.38 ± 2.260	−1.821	0.069
CM	19.04 ± 3.847	18.59 ± 3.921	−2.500	0.012
RL	23.22 ± 3.711	22.77 ± 3.598	−2.681	0.007
AR	13.10 ± 2.923	12.82 ± 3.032	−2.010	0.045
AI	14.30 ± 2.798	13.89 ± 2.785	−3.229	0.001
BC	20.24 ± 2.755	20.14 ± 2.715	−0.780	0.436
GF	22.29 ± 4.942	21.61 ± 4.877	−2.996	0.003

PS, problem solving; CM, communication; RL, role; AR, affective responsiveness; AI, affective involvement; BC, behavioral control; GF, general functioning.

equation. At step 2, the predictor variable (all sub-scales of family function) was entered with the method of stepwise ($\alpha = 0.05$) into the regression equation. At step 3, the moderator variable (social support) was entered into the regression equation. At step 4, the interaction of each dimension of family function × social support was added with the method of stepwise ($\alpha = 0.05$). Significant change in R^2 for the interaction term indicates a significant moderator effect.

The results exploring predictors of self-esteem showed that for the grandparenting group three variables – RL ($\beta = -0.170$, $P < 0.01$), social support ($\beta = 0.243$, $P < 0.01$), and BC ($\beta = -0.078$, $P < 0.05$) had a significant effect on the self-esteem. In this model, lower family function score and higher social support score were associated with higher self-esteem score. Most importantly, there was a significant interaction between general family functioning and social support ($\beta = -0.123$, $P < 0.01$). These results suggest that social support moderated the relationship between general family functioning and self-esteem. In the parent-raising group, six significant predictors of self-esteem were found, these were social support ($\beta = 0.212$, $P < 0.01$), RL ($\beta = -0.115$, $P < 0.01$), CM ($\beta = -0.111$, $P < 0.01$), AI ($\beta = -0.080$, $P < 0.01$), BC ($\beta = -0.065$, $P < 0.01$), and PS ($\beta = -0.062$, $P < 0.05$). However, there was no significant interaction between family function and social support. Results of these analyses are presented in **Table 3**.

To illustrate the general family functioning × social support interaction for self-esteem in the grandparenting group, we plotted the regression of self-esteem on general family functioning at high and low levels of social support (see **Figure 1**). According to the procedures posed by Aiken and West (1991), we used the simple slope for the regression of self-esteem on general family function by using the high (one standard deviation above the mean) and low (one standard deviation below the mean) values for social support. There was a significant negative relation between general family functioning and self-esteem at high levels of social support ($\beta = -0.452$, $P < 0.01$). However, at low levels of social support, the relation between general family functioning and self-esteem was non-significant ($\beta = -0.096$, $P > 0.05$). Thus, among students with high social support, general family functioning was a significant determinant of self-esteem. In contrast, at low levels of social support, general family function did not influence self-esteem.

DISCUSSION

The current study was designed to examine the difference in family functioning and self-esteem between university students with and without grandparenting experience and the important role of social support on the relationship between family function and self-esteem in a sample of Chinese university students. We found that the students who have the experience of grandparenting showed a lower level of self-esteem than the students raised by their parents. Grandparents raising children is a common phenomenon in China, which is related to the country's social and cultural background. The parents are both too busy to take care of the child or they are working in another city far away from their hometown, so the child is sent back to the grandparents to be raised until he or she goes to kindergarten or school (Wang, 2012). Chinese culture is collectivist and deeply influenced by Confucianism, while many western cultures are more individualistic. Collectivism emphasizes common interests, conformity, cooperation, and interdependence. These cultural traits strengthen extended families and collectives where each person takes responsibility for fellow members of their group (Hofestede, 2001). Therefore, in China many grandparents in retirement take over the responsibility of raising the grandchildren by their own choice. Song (2004) found that for late adolescents, parental attachment contributed significantly to both self-liking and self-competence. According to Bowlby (1982), the period from birth to 3 years old is crucial to establish attachment between the infant and main caregiver; individual's fundamental self-perception and

TABLE 2 | Correlation between family function, social support and self-esteem in two groups (r).

Variable	SS		PS		CM		RL		AR		AI		BC		GF	
	G	P	G	P	G	P	G	P	G	P	G	P	G	P	G	P
PS	−0.355**	−0.394**														
CM	−0.416**	−0.461**	0.510**	0.581**												
RL	−0.338**	−0.425**	0.410**	0.439**	0.579**	0.604**										
AR	−0.405**	−0.442**	0.377**	0.449**	0.669**	0.651**	0.535**	0.543**								
AI	−0.320**	−0.401**	0.339**	0.344**	0.603**	0.576**	0.627**	0.636**	0.488**	0.541**						
BC	−0.237**	−0.263**	0.290**	0.323**	0.269**	0.325**	0.434**	0.451**	0.341**	0.345**	0.393**	0.406**				
GF	−0.491**	−0.524**	0.613**	0.630**	0.753**	0.765**	0.668**	0.662**	0.643**	0.673**	0.688**	0.675**	0.415**	0.395**		
SE	0.297**	0.384**	−0.263**	−0.310**	−0.334**	−0.382**	−0.361**	−0.380**	−0.318**	−0.314**	−0.241**	−0.354**	−0.247**	−0.264**	−0.327**	−0.399**

G, grandparenting group; P, parent-raising group; SS, social support; SE, self-esteem.
** $P < 0.05$, ** $P < 0.01$.*

self-evaluation derive from the experience of interaction between the child and caregiver in the early childhood. The stable and health parent-child relationship makes the child feel that he or she is a favorite and capable person, and otherwise the unstable parent-child relationship makes the child feel unwelcome and worthless. The grandparenting experience could result in failure of establishment of secure attachment, and hence exert a negative influence on the self-esteem development. The children who left their parents and were raised by grandparents in infancy fail to establish an attachment relationship with their parents in their early childhood; instead they may become more attached to their grandparents. However, the lack of parental care and companionship means that when they came back to live with their parents, both the parents and the children would feel distant from each other, and the children could fail to gain strong sense of security and belonging, which might affect their self-awareness and self-evaluation (Song, 2004). Furthermore, the rearing style of grandparents is different from the parents; the grandparents may look after the grandchildren too carefully to give them enough exploration space; according to Erikson's psychosocial stages, it would hinder their development of sense of autonomy and self-worth.

In addition, compared to their counterpart group the grandparenting group reported worse family function in terms of role, CM, AI as well as AR and general family function. In non-grandparenting families, the children and parents can develop a close parent-children relationship and the children can establish a good sense of trust and security in the core family, whereas in grandparenting families, the parents may be both too busy to take care of the child or they are working in another city far away from their hometown, they cannot fully take over the responsibilities of rearing the child. In that case the grandparents instead bear a certain responsibility of raising and educating the grandchildren, it is therefore possible that the children fail to develop a close attachment relationship to their parents. As a result, this could lead to less emotional exchange and low confidence between parents and children when the children came back to their core family. Li (2010) has investigated the parent-child relationship in the college students with grandparenting experience, it was found poor intimacy, lack of trust on each other and deficient initiative CM. Therefore, worse family function might be exhibited within the grandparenting family compared to the families where the children are raised by their parents.

The students with grandparenting experience also reported lower perceived social support than the students raised by their parents. Perceived social support can be understood as a personal subjective evaluation that he or her social network will provide effective help when needed (Lakey and Scoboria, 2005). It can be distinguished from the received support, which means that actual support is provided within a specific time frame (Uchino, 2009). Family support is an important part of social support. Compared to the students raised by their parents, the perceived family support of students with grandparenting experience could be generally lower due to their distant parent-child relationship.

As we expected, the results of correlative analysis showed that self-esteem had a significant positive relationship with social support and a negative relationship with family function.

TABLE 3 | Hierarchical regression analysis predicting self-esteem from family function and social support.

Group	Variables	R^2	ΔR^2	F	β	t
Grandparenting	Gender	0.000	0.000		−0.114	−3.329**
	Age	0.000	0.000		0.034	1.026
	Only child status	0.010	0.010	2.502	0.029	0.868
	RL	0.139	0.129	29.244**	−0.170	−3.839**
	AR	0.165	0.026	28.494**	−0.076	−1.610
	BC	0.177	0.012	22.144**	−0.078	−2.120*
	CM	0.182	0.005	19.949**	−0.080	−1.624
	SS	0.209	0.027	23.677**	0.243	6.059**
	GF × SS	0.222	0.013	22.773**	−0.123	−3.536**
Parent-raising	Gender	0.000	0.000		−0.069	−3.182**
	Age	0.000	0.000		−0.048	−2.195*
	Only child status	0.017	0.017	9.870**	0.013	0.588
	RL	0.186	0.169	96.862**	−0.115	−3.678**
	CM	0.195	0.009	82.203**	−0.111	−3.500**
	BC	0.201	0.006	71.005**	−0.065	−2.648**
	PS	0.203	0.002	61.758**	−0.062	−2.296*
	AI	0.206	0.003	55.153**	−0.080	−2.664**
	SS	0.237	0.031	66.069**	0.212	8.386**

$^*P < 0.05$, $^{**}P < 0.01$.
SS, social support; PS, problem solving; CM, communication; RL, role; AR, affective responsiveness; AI, affective involvement; BC, behavioral control; GF, general functioning.

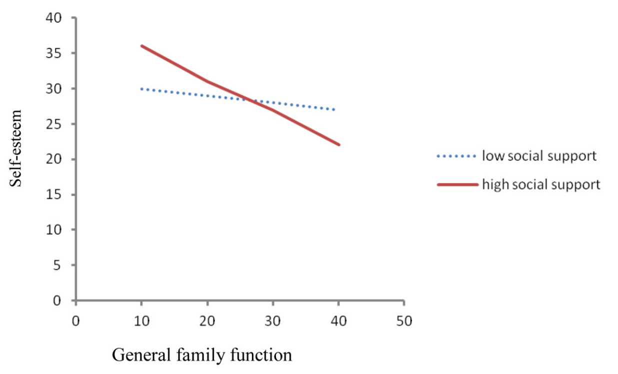

FIGURE 1 | Effects of general family function and social support on self-esteem scores.

These results accord with previous studies on the relationships between self-esteem and social support (e.g., Goodwin et al., 2004; Birndorf et al., 2005; Zhang, 2007; Denissen et al., 2008; Lian et al., 2016; Chen et al., 2017) and family function (e.g., Heaven and Ciarrochi, 2008; An et al., 2010; Guo, 2012; Yen et al., 2013).

The most important finding of this study is that social support moderated the influence of general family function on self-esteem of the students with grandparenting experience. When these students reported a high level of social support, those with a low score of general family function reported a higher level in self-esteem. However, there were no differences in self-esteem between high and low general family function scores when they were confronted by a low level of social support. These results suggest that among the students who perceived great social support, more positive the family functioning were evaluated, higher level of self-esteem the students showed; whereas among the students who perceived poor social support, family functioning exert slight influence on the level of self-esteem. It seems that in the current study for the first time to report that social support influences on the association between general family function and self-esteem of students with grandparenting experience. We suggest that the results of our study can be ascribed to the self-development connected to the extent of early attachment. According to attachment theory, the children start to establish their basic self representation in reaction to the accessibility and sensitivity of parents and other caregivers in childhood, and these concepts are revised in the whole life (Bowlby, 1982; Bretherton, 1991). Therefore, if the caregiver is sensitive and accessible to the child, the child's self model is valuable and worthy of being loved. On the contrary, if the parents are not available and sensitive to the child, the child's self model is invaluable and is unworthy of being loved. Studies generally supports the idea that positive self-concept, including high levels of self-esteem and self-efficacy are associated with secure attachment to parents in infancy, childhood, and adolescence (Thompson, 1999; Arbona and Power, 2003). In Zhao's (2011) study it was found that the proportion of secure attachment among the children with grandparenting experience was less than that of the children raised by their parents, instead the disorganized relationship was more. Those children who were often separated from their parents in early childhood to be raised by their grandparents, missed this crucial period during childhood to build a long-lasting attachment with their parents. Although the children may establish secure attachment with the grandparents, when they came back to their parents, they tended to feel a lack of care from their parents, they did not feel loved, and this kind of thought probably made the children feel unworthy (Li, 2010).

It is supposed that support from family, peer groups, and significant others, like teachers, or romantic partners for example is especially important for fostering identity and self-development of the college students who exhibit a lack of secure attachment to their parents (Allen and Land, 1999; Hazan and Zeifman, 1999; Fass and Tubman, 2002; Furman et al., 2002). The senses of self-importance, achievement and power gaining from the life experience are necessary for the construction of self-esteem (Yang and Zhang, 2003). If the college students are exposed to stressful situations, support from others might enable them to feel acceptance and strength, and thus enhance self-confidence and eventually lead to a higher self-esteem. Therefore, those students with a perceived lower social support as a result for being raised by their grandparents for at least 1 year, though reporting greater family function, will have difficulties in enhancing self-esteem.

Social support didn't show moderating effect in the relationship between family function and self-esteem among the students without grandparenting experience, the possible reason could be that in the parents-raising family the influence of the parents may play more crucial role in the development of self-esteem of the children across the lifespan from infancy to adolescence than that of other family members as well as significant others outside of the family.

CONCLUSION

In the current study some limitations should be mentioned. Firstly, in this study we used self-report measures to collect data, so it could have a threat to internal validity. We suggest using multiple methods for evaluation (e.g., peer reports) in the future study, which in contrast might reduce the effect of subjectivity. The second limitation lies in quantitative methodology of the study, in which it is difficult to make deep analysis of the impact of grandparenting experience on attachment and hence on self-esteem. In future research we suggest using experimental studies or qualitative studies to test the moderating model. The third limitation is that this sample came from a university population, so it limits the extent to which these findings could be generalized to other educational environments and geographic locations in China. Despite these limitations, there are important contributions of this study.

In conclusion, all the three hypotheses in this study are fully supported. The current study makes a significant contribution to the study of the family functioning and self-esteem in youth with grandparenting experience. It provides an empirical framework for researchers through testing the moderating effects of social support between family function and self-esteem in a sample of Chinese college students. The findings show that, social support is an important contributor in the relationship between family functioning and self-esteem in the students with grandparenting experience. Support from family members and significant others outside of the family are especially important for fostering identity and self-development of the college students who exhibit a lack of secure attachment to their parents. In future studies, first, more exploration may be needed of how the quality of early attachment and the time of the grandparenting (e.g., infancy, toddlerhood, early childhood, or middle childhood) influence the self-esteem of the students with grandparenting experience; second, further investigation may be need of how they develop friendships with peers and significant others that helps them improve self-esteem and efficacy. In addition, these findings may provide empirical reference to help to establish effective psychosocial interventions with the aim of improving social support and family function in university students with low self-esteem, and hence improving mental health status.

ETHICS STATEMENT

This study was approved by the Institutional Review Board of Tongji University. All participants provided written informed consent prior to the study. For the participants under 18 years old, one parent provides written consent.

AUTHOR CONTRIBUTIONS

JS contributed to study design, recruitment of participants, data analysis and interpretation and writing of the manuscript. LW contributed to recruitment of participants and interpretation of results. YY and NS contributed to recruitment of participants. XZ contributed to study design and interpretation of results. CZ contributed to recruitment of participants. All authors have approved the final manuscript.

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Systemic Therapy for Youth at Clinical High Risk for Psychosis: A Pilot Study

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Psychosocial intervention trials for youth at clinical high risk (CHR) for psychosis have shown promising effects on treating psychotic symptoms but have not focused on psychosocial functional outcomes, and those studies have been conducted among help-seeking patients; there is a lack of research on non-clinical young CHR individuals. Systemic therapy (ST) is grounded in systemic-constructivist and psychosocial resilience theories. It has a number of advantages that makes it attractive for use with CHR individuals in non-clinical context. The present study evaluated the effect of ST for students at CHR on reducing symptoms and enhancing psychosocial function. This was a single-blind randomized controlled trial for CHR young people comparing ST to supportive therapy with a 6-month treatment. Psychotic and depressive symptoms (DS) as well as self-esteem and social support (SS) were assessed at pre- and posttreatment. 26 CHR individuals were randomly divided into intervention group ($n = 13$) and control group ($n = 13$). There were no significant differences in severity of symptoms, level of SS and self-esteem at baseline between the two groups ($P > 0.05$). At posttreatment, significant improvements in positive and DS as well as SS and self-esteem were observed in the ST group ($P < 0.05$); in the control group, these improvements were not significant ($P > 0.05$). The findings indicated that systemic intervention for university students at CHR for psychosis may have a positive effect on symptoms and self-esteem as well as SS in short term. More long-term research is needed to further evaluate this intervention.

Keywords: systemic therapy, intervention, clinical high risk, psychosis, youth

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INTRODUCTION

Young people are at greater risk of developing mental illness as they transition from childhood to adulthood (1). Psychosis typically emerges in late adolescence or early adulthood and can disrupt social and psychological development, including the attainment of educational goals and relationship skills, thus seriously impairing their quality of life. Prior to the first onset of psychotic disorder, 80–90% of individuals experience attenuated psychotic symptoms, and this stage was conceptualized as a prodromal phase, or at clinical high risk (CHR) (2, 3). The CHR for psychosis state is characterized by the presence of low intensity/frequency psychotic symptoms, brief limited psychotic episodes, and/or familial risk and/or schizotypal personality disorder in the presence of psychosocial functional decline, and with increased risk of developing psychosis (4). The CHR criteria provide an important opportunity for early intervention in preventing or delaying the onset of psychosis and reducing the social and economic burden associated with long-term mental health problems.

Systemic therapy (ST) has shown particular promise in improving adolescent and adult mental health problems (5–7). ST is based on system theory and controlling theory. It emphasizes on viewing problems in developing, comprehensive, positive, and diverse ways and focuses on understanding the individual symptoms within the system of interpersonal relationships. In ST, the function and significance of the symptoms is much valued. Attention will be paid to the interaction between individual and his/her environment. System therapy is suitable for individuals and families. Its value orientation is positive psychology, namely, resource orientation, treating the patients/clients as experts of solving their problems; it is assumed that the patients/clients do have the resource for solving the problem. ST emphasizes and explores the individual strengths, ability, ideas, and social resources, focusing on extending the space beyond the problem, to bring new and diverse perspectives for individuals and families, and thus to promote changes from the inner and interpersonal levels (8). Previous research reported that ST had shown positive effects on improving the symptoms and psychosocial functioning of schizophrenia patients (9). In addition, ST considers changes outside of the treatment; all in all, it is an efficient, economical, short-term treatment with a long interval.

To our knowledge, there was no specific manual for the treatment of clinical risk of psychosis available at the time of research, therefore a manual of ST was developed by the experts for ST, integrating a broad range of systemic methods, for the CHR university students.

We reviewed ST manuals for psychotic disorders in adult psychotherapy (10) and well-established ST manuals for various disorders (11–13). We used general ST concepts (8), integrating constructivist, solution-oriented methods (14), in addition to paying attention to the disorder-specific relational systemic dynamics (15, 16). According to the literature and to our experiences of treating patients, the aim of ST is to contextualize attenuated psychotic symptoms by addressing an individual's social system to which he/she attaches importance. Although the social system members could not attend the meeting physically, circular questions included them on a cognitive level. The analysis of transgenerational relationships and of past and present interpersonal interactions help to develop a new understanding of the important roles, places, and resources of all system members. Under the systemic model, the therapy sessions alike are held individually but with a strong focus on relationship issues. The ST manual differentiates between four phases, which are described in detail in the Section "Materials and Methods."

We found only seven systemic intervention RCTs for psychotic disorder (17–23), and only one for patients at CHR state (22). This study ($n = 40$, China) compared only medication treatment with 10 sessions of structural family therapy (one school of ST) plus medication. The outcome relied on the family function, severity of prodromal symptoms, and treatment compliance. This trial demonstrated the structural family therapy group with a higher reduction of severity of symptoms, higher treatment compliance, and a higher improvement in family function than the control group ($P < 0.05$).

To our knowledge, there is a lack of studies, which explore the effectiveness of ST for CHR individuals among general populations. Most studies focus on the primary outcome as a dichotomous one of transition to psychosis, rather than the dimensional domains of psychosocial functioning. Previous studies indicate that the adolescents and young adults at CHR might show poor psychosocial functioning. In some CHR studies, it was reported that the subjects had shown a lower level of self-esteem and social support (SS) in comparison to healthy controls (24, 25), and the levels of self-esteem and SS were negatively associated with the severity of the prodromal psychotic symptoms (25–28). However, many studies indicated that such psychosocial factors could play a positive role in mental health (29–31). Moreover, it is considered as an important resource for coping with psychotic symptoms (32). Thus, a positive self-perception and a strong sense of control would prevent a negative perception of daily stress; self-esteem also has a stress-buffering effect, which protects youth from the harmful effects of stress on mental health (33). In psychotic disorders, low self-esteem has been demonstrated in both the development of delusions (34) as well as in the maintenance of psychotic symptoms (35). SS as another important personal resource has been improved, having positive effects on mental health by either directly enhancing self-esteem or indirectly by protecting individuals against the adverse impact of exposure to stress and trauma (36). SS may exert such positive effects both prior to and at the onset, as well as during the course of disorder, operating to reduce both risks of onset and relapse.

Therefore, this study was designed to investigate the effectiveness of ST for CHR individuals among general youth populations on both symptom and psychosocial functioning outcome. We hypothesize that compared to the control group, ST would reduce symptom severity while improving self-esteem as well as SS.

MATERIALS AND METHODS

Study Design

This study was a single-blind RCT of ST compared to supportive therapy conducted at university with a 6-month treatment. The treatment began within 2 weeks of completion of the baseline assessment and was available for up to 10 sessions over a 6-month period. Assessments were conducted after the treatment. Clinical raters were blind to treatment groups.

Participants

In the study participated 26 university students at CHR (12 males and 14 females; age 18.85 ± 1.120). The 26 CHR subjects were screened out from 2,800 students of the first and second grades. The status of CHR was diagnosed through a two-stage assessment, consisting of screening with the self-report Chinese version of 16-item Prodromal Questionnaire (CPQ-16) (37) and an assessment interview according to the Structured Interview for Psychosis-Risk Syndromes (SIPS). The criteria of prodromal symptoms require that individuals meet at least one of the three clinical criteria: (1) attenuated positive symptom prodromal syndrome, defined as recent occurrence of attenuated positive psychotic symptoms with sufficient frequency or severity; (2) brief intermittent psychosis prodromal syndrome, defined as recent presenting of psychotic symptoms with spontaneous remission within 1 week; and (3) genetic risk and deterioration prodromal syndrome, defined as coexisting of genetic risk and recent functional decline (38). This study had the ethics approval from the institutional review board of Tongji University. Prior to the study, each participant provided written informed consent.

Procedure

In order to select the potential individuals at CHR for psychosis, at first stage, the CPQ-16 was given to 2,800 university students. In the current study, the positive threshold for the CPQ-16 was set at 6, according to the study conducted by Ising et al. (39). Totally, 611 students reached this cut-off score, and they were invited to take part in our second stage for SIPS. We received agreement from 529 participants who accepted the SIPS, which was operated by trained psychiatrists. Of the 529 students referred to the trial, 32 were screened and 26 were randomized for treatment, 13 to the ST group and 13 to the control group. The 26 students at CHR completed the 6-month treatment.

Measures

Symptoms

The severity of positive and negative symptoms (NS) was measured with the Scale of Prodromal Syndromes from 0 (absent) to 6 (psychotic) and a total symptom score was created (38). The Montgomery–Åsberg Depression Rating Scale was used to assess depressive symptoms (DS) (40). The overall functioning was rated by the Global Assessment of Functioning scale (GAF) (41).

Self-Esteem

Self-esteem was rated with the self-reported Rosenberg Self-esteem Scale (SES) (42), which includes 10 items with a measure of global self-esteem. Each item is rated on a 4-point Likert scale ranging from 1 = strongly disagree to 4 = strongly agree. It has been demonstrated that the RSES has good reliability and validity (43, 44).

Social Support

Social support was measured with the Perceived Social Support Scale (45), which is based on the unique social and cultural conditions in China. The scale is made up by 12 items designed to assess subjective SS rated on a 7-point Likert scale ranging from 1 = strongly disagree to 7 = strongly agree. The higher scores reflect higher SS. The scale has been widely used among Chinese populations and proved to have a good validity and reliability (44, 45).

Interventions

Systemic therapy for CHR students followed the manual developed according to Carr (7) by experts on ST, ranging from 10 to 30 years of experience. It is grounded in systemic-constructivist (46) and psychosocial resilience theories (47) and based on a solution focused model that prioritizes a careful clarification of therapeutic goal. It is solution and resource oriented, reframing one's problem from functional and meaningful perspectives, using a variety of questioning techniques to enrich perspectives toward identified problems, exploring and strengthening the resource of the clients and creating more space and possibilities to solve the problem, homework tasks were usually given to help clients gain new insights and experiences. The treatment comprises four phases, each phase including 2 to 5 sessions. First phase: establishment of therapeutic relationship, collection of information, and clarification of therapeutic goals; second phase: understanding the context of the identified problem as well as interactive patterns around the identified problem, reconstruction of the problem, and exploring resources and solutions, making use of the resources and putting the solution into practice; third phase: reinforcement and deepening of changes; fourth phase: relapse prevention. Each session lasted 50 min. During the process of treatment, the treatment interval gradually extended from weekly to monthly. The details of sessions are listed in **Table 1**.

Supportive therapy is conducted only using general counseling techniques: warm, empathic, and non-judgmental face-to-face contact and supportive listening.

All therapy sessions including ST and supportive therapy were delivered by a qualified systemic therapist with 10 years experience. It has the advantage of controlling for non-specific aspects of treatment (e.g., therapist age, sex, personality, therapist experience). The therapist received expert and peer supervision regularly.

Data Analysis

All statistical procedures were conducted using the Statistical Package for the Social Sciences (SPSS 17.0). Chi-square tests were applied to compare categorical demographic variables between the two groups. *T*-tests for independent samples were used to assess differences in self-esteem score and SS score between the two groups. *T*-tests for paired samples as well as effect sizes were calculated to assess differences in self-esteem score, SS score, symptom score, and functioning (GAF) between baseline and posttreatment in two groups. End of intervention scores on various outcome measures between the ST group and the control groups were compared using ANOVA of repeated measures to explore the impact effects of time and intervention.

RESULTS

Social Demographic Characteristics and Baseline Data

There were no significant differences in age, gender, whether only child and family history between the two groups ($P > 0.05$). There were no significant differences in severity of psychotic symptoms, overall functioning, level of supports, and self-esteem at baseline between the two groups ($P > 0.05$). Social demographic characteristics and baseline data are presented in **Table 2**.

Effectiveness of Systemic Intervention on Severity of Symptoms and GAF

Participants in the ST group demonstrated significant decreases in severity of positive symptoms (PS) and DS comparing to that at baseline ($P = 0.005$); however, the participants in the control group had shown no significant changes in severity of PS and DS ($P > 0.05$). Improvements were also observed in NS and GAF

TABLE 1 | Topics and key concepts of systemic therapy.

Therapeutic phases	Topics	Key issues and techniques	Homework
First phase (2 sessions)	Introduction and join in	Buildup rapport	Write a strength and resource list, including at least 50 points
	Collection of information	Positive listening, systemic questioning, buildup the first hypotheses, draw genogram	Write down 10 things, which the clients want to do most in the next 3 years
	Clarification of therapeutic goals	Inquiry about the expectations, using systemic questioning to clarify the therapeutic goal, which is clear, feasible, and in a positive way of formulation	
Second phase (5 session)	Understanding the context of the identified problem as well as interactive pattern around the identified problem	Shifting the pathology from symptoms to relations. Understanding the meaning and function of the identified problem in an interpersonal system; systemic questioning such as circular questions, exception questions, scaling questions; family boards and timelines	On odd days, the client should act as if the problems become more serious and, on even days, the client should act as if the problems disappear, and meanwhile, he or she observes the reaction of others
	Reconstruction of the problem and exploring resources and solutions	Finding out and creating diverse possibilities; challenging the certainty of the knowledge of the identified problem; rewriting the self-narrative and reframing, positive connotation	
	Making use of the resource and putting the solution into practice	Homework	
Third phase (2 session)	Reinforcement and deepening of changes	Reflecting and reviewing the progress and changes; expand the details of the changes; emphasize the client's efforts and abilities to make changes; discuss about how to maintain the changes	Observe and write down the sympathetic behaviors
Fourth phase (1 session)	Relapse prevention	Considering the risks of relapse and building up a treasure box of strategies	Build up a treasure box of strategies

TABLE 2 | Baseline clinical and social demographic characteristics of two groups.

	ST ($n = 13$)	Control ($n = 13$)	t/X^2	P
Age mean (SD)	18.85 (0.987)	18.85 (1.281)	0.000	1.000
Female (%)	9 (64.2)	5 (38.5)	2.476	0.116
Only child (%)	11 (84.6)	10 (76.9)	0.248	0.619
Family history (%)	0 (0.0)	1 (7.7)	1.040	0.308
PS	6.85 (3.460)	7.62 (3.477)	−0.565	0.577
NS	4.54 (4.666)	3.92 (3.499)	0.380	0.707
DS	6.62 (5.455)	7.08 (6.849)	−0.190	0.851
Global assessment of functioning scale	73.62 (5.546)	72.85 (6.453)	0.326	0.747
Self-esteem	26.54 (4.824)	28.08 (4.192)	−0.868	0.394
Social support	56.38 (12.868)	62.46 (8.491)	−1.421	0.168

ST, systemic therapy; PS, positive symptoms; NS, negative symptoms; DS, depressive symptoms; SE, self-esteem; SS, social support.

in both groups, but the changes were not significant ($P > 0.05$). The results are presented in **Table 3**. Furthermore, the statistic analysis results of ANOVA of repeated measures didn't show significant impact effects of the time factor, interaction between time and intervention, as well as intervention factor on the changes. These interactions are graphed in **Figure 1**. The statistic results are presented in **Table 4**.

Effectiveness of ST on Level of Self-Esteem and SS

Obvious improvements in level of SS and self-esteem in participants in ST were evident at posttreatment ($P = 0.013$, $P = 0.011$); the level of SS as well as self-esteem of the individuals in the control group had no significant changes at posttreatment ($P > 0.05$). The results are presented in **Table 3**. Furthermore, the statistic analysis results of ANOVA of repeated measures didn't show significant impact effects of the time factor, interaction between time and intervention, as well as intervention factor on the changes. These interactions are graphed in **Figure 2**. The statistic results are presented in **Table 4**.

Clinical Significance

At posttreatment, attenuated psychotic symptoms in eight participants (61.5%) in the ST group and six participants (46.2%) in the control group were reduced to a level of remission from an initial CHR status as defined by the SIPS. However, the difference was not significant ($X^2 = 0.619$, $P = 0.431$). One CHR individual in the control group developed an onset of the psychotic manic episode of bipolar I disorder during the follow-up period. The conversion rate was 3.8% (1/26).

DISCUSSION

The goals of this study were (1) to evaluate the effectiveness of systemic family therapy focusing specifically on the change in youth-attenuated psychotic symptoms and improvements in psychosocial functioning, and (2) to evaluate the effectiveness of systemic family therapy for CHR individuals in a non-clinical context. Most previous systemic intervention studies evaluated its impact on symptoms and treatment compliance. Although in clinical context to reduce the symptoms and enhance the

TABLE 3 | Comparison of changes between groups.

Measures		Pre-intervention	Post-intervention	Cohen's *d*	*t*	*P*
Positive symptoms	Systemic therapy (ST)	6.85 (3.46)	4.54 (5.08)	0.53	3.426	0.005
	Control	7.62 (3.48)	5.23 (3.90)	0.65	1.934	0.077
NS	ST	4.54 (4.67)	3.85 (4.78)	0.15	0.454	0.658
	Control	3.92 (3.50)	3.85 (4.67)	0.02	0.054	0.958
Depressive symptoms	ST	6.62 (5.46)	3.00 (4.08)	0.75	3.065	0.010
	Control	7.08 (6.85)	6.46 (8.24)	0.08	0.211	0.837
Global Assessment of Functioning scale	ST	73.62 (5.55)	77.00 (9.57)	0.43	−1.206	0.251
	Control	72.85 (6.45)	77.15 (10.01)	0.51	−1.510	0.157
SE	ST	26.54 (4.82)	29.08 (3.73)	0.59	−2.980	0.011
	Control	28.08 (4.19)	28.23 (5.09)	0.03	−0.148	0.885
SS	ST	56.31 (12.76)	62.54 (14. 81)	0.45	−2.916	0.013
	Control	62.46 (8.49)	62.08 (12.47)	0.04	−0.464	0.901

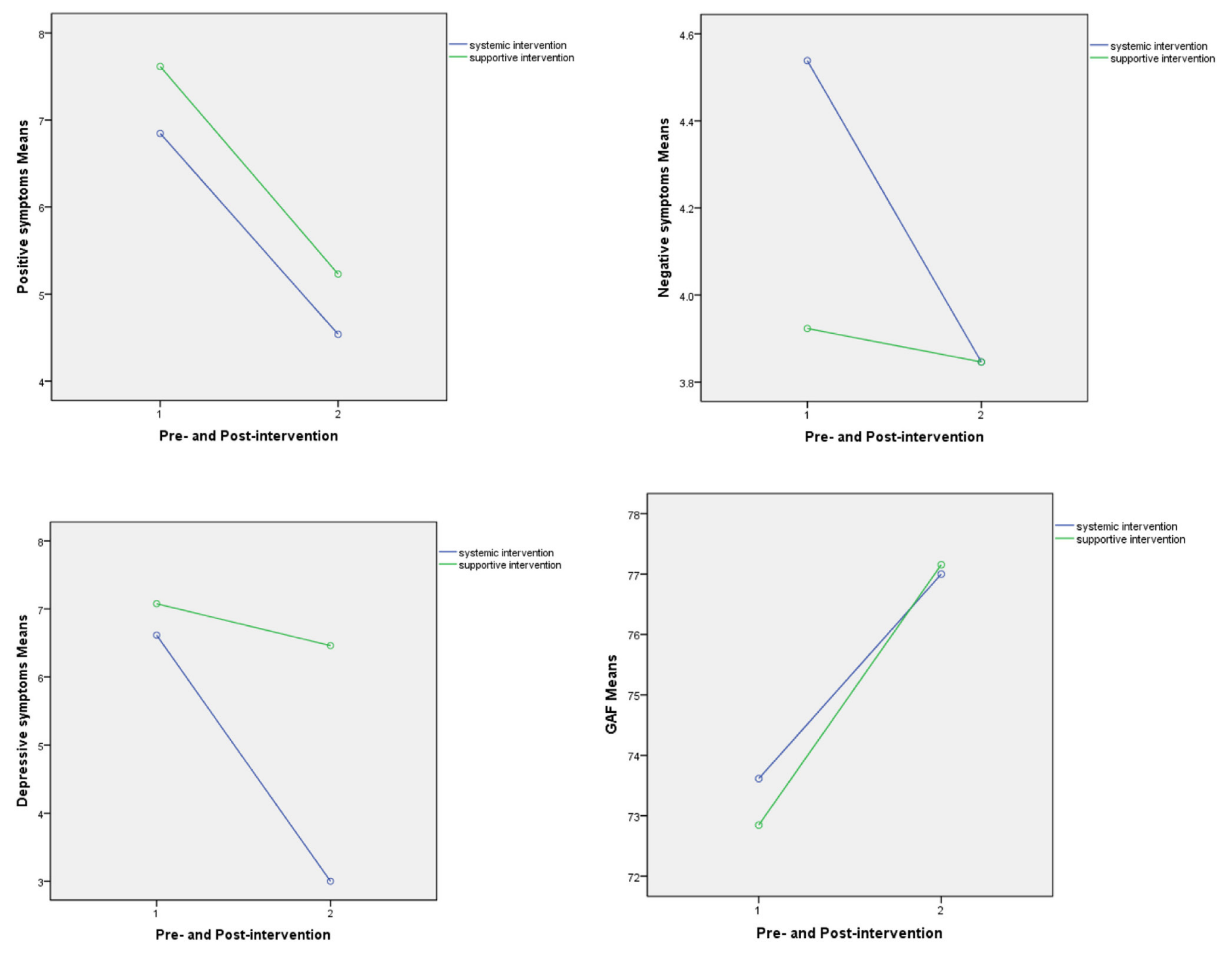

FIGURE 1 | Improvements in scores of severity of symptoms.

treatment compliance has a significant benefit for youth and families, it is important to know if ST enhances the psychosocial functioning level and the feasibility in non-clinical context. These issues provided the impetus for the present study. As far as we know, this trial is the first RCTs of ST for young people at CHR for psychosis. The trial targeted a non-clinical young sample, evaluated the treatment effect of ST, and employed an active control condition.

As expected, we found that ST led to a significant reduction in positive and DS and to an obvious improvement in self-esteem and SS comparing between the pre-and posttreatment measure outcomes. The ecological model, which underpins ST proposes that individuals' problems become problems only in the context and different social systems. The uniqueness of ST lies in its innovative nature of reconstructing "problems" in developing, comprehensive, positive, and diverse ways, treating the patients/clients respectfully as experts of solving their problem, and suggesting creative, unusual homework tasks, which function as perturbacion in behavioral and interactive patterns. These systemic therapeutic concepts are quite suitable for the non-clinical context. First, ST focuses on resources rather than on deficits, and it recognizes the expert status of the young people at CHR. This is in line with the view of resilience in CHR individuals that focuses on protective factors against adversity (48). Second, in a systemic constructivist perspective, psychotic symptoms are regarded as phenomena, which are neither from the beginning as surely existent confirmed nor as definitively non-existent considered. Symptoms could be considered as psychological and biological phenomena, but also as deriving from social interaction constructed reality. Therefore, this understanding about the symptoms comes as a great relief to the students at CHR and suggests that they could influence their symptoms. Third, the approach moves the focus from problems to solutions by setting limited and clearly defined goals, and it promotes an early and positive relationship between students and therapist. ST can give them support in resolving or at least reducing such problems through a structured and focused way, emphasizing the individual's unique contribution. In systemic intervention, the students at CHR could gradually develop a resource-oriented mindset: focusing on resources rather than deficits, on solutions rather than problems, and that could contribute to enhancing the SS and self-esteem, and thus, they are playing a positive role in improvement of the symptoms.

From **Table 4**, it could be seen that there are no statistically significant differences when considering the interaction between the time and group. There are several potential explanations for it. First, there may have been a "floor effect" with little room for self-reported, treatment-related improvement, because youth did not perceive themselves to have clinically significant problems. Second, because of the small sample size, the quantitative study design and statistical analyses could not fully reflect the difference in quality of changes between groups. Third, the natural recovery process is an essential factor to influence the clinical outcomes. In some cases, personal qualities, such as resilience might play a more important role to protect a subject's mental health (49).

All in all, we can say that the results of this pilot study support at least the possibility of using a systemic intervention for young people at CHR in non-clinical context. Nevertheless, it is necessary to do further research in this area with larger sample sizes, standardized measures, prolonged follow-up assessments, and an exploration of effective therapeutic factors on improvement of self-esteem as well as SS. Specifically, we suggest to investigate how self-esteem and SS reduce symptoms or increase positive reactions and to compare this with other established therapies.

TABLE 4 | ANOVA of repeated measures.

Measures	Difference between pre- and post (time)		Time × group		Difference between groups	
	F	***P***	***F***	***P***	***F***	***P***
Positive symptoms	11.157	0.003	0.003	0.957	0.266	0.611
NS	0.137	0.715	0.087	0.770	0.049	0.827
Depressive symptoms	1.806	0.192	0.908	0.350	1.033	0.320
Global Assessment of Functioning scale	3.695	0.067	0.053	0.820	0.015	0.902
SE	4.025	0.056	3.158	0.088	0.045	0.833
SS	4.797	0.038	2.154	0.155	0.665	0.423

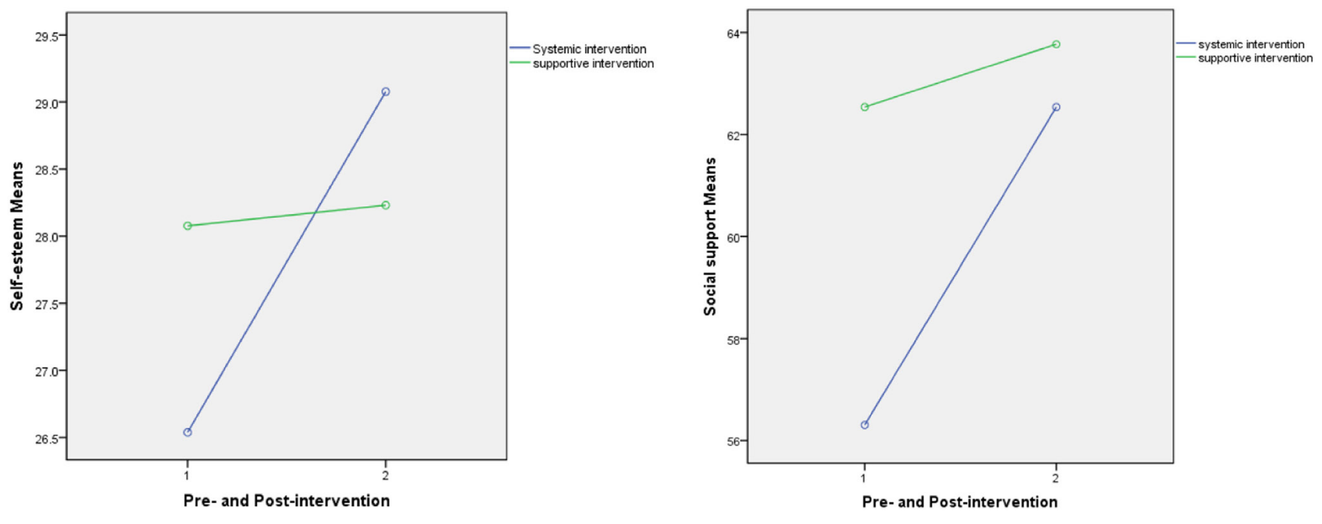

FIGURE 2 | Improvements in level of self-esteem and social support.

ETHICS STATEMENT

Our study had ethics approval from the School of Medicine Ethics Committee, Tongji University, Shanghai, China. Reference number: 2014YXY16, and we acquired written informed consent from participants of the study.

AUTHOR CONTRIBUTIONS

JS contributed to study design, recruitment of participants, data analysis and interpretation, and writing of the manuscript. LW contributed to recruitment of participants and interpretation of results. YY, CZ, and NS contributed to recruitment of participants. XZ contributed to study design and interpretation of results.

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Multi-Level Family Factors and Affective and Behavioral Symptoms of Oppositional Defiant Disorder in Chinese Children

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Given the important role of family environment in children's psychological development, the objective of this study was to examine the linkages between family factors at the whole, dyadic, and individual levels and two dimensions (affective and behavioral) of Oppositional Defiant Disorder (ODD) symptoms in Chinese children. Participants comprised of 80 father-child dyads and 169 mother-child dyads from families with ODD children. The results indicated that multilevel family factors were differently associated with children's affective and behavioral ODD symptoms. All the family factors at the dyadic and individual levels were significantly associated with child affective ODD symptoms. However, only the most proximal factors (parent-child relationship and child emotion regulation, which were directly related to child) were significantly related to child behavioral ODD symptoms. The present study extends the current knowledge regarding the relationships between family factors and two dimensions of child ODD symptoms by testing the comprehensive multilevel family factors model. This study also recommends that future interventions for ODD children should consider the multi-level family factors to enhance intervention efficacy.

Keywords: child with ODD, multilevel family factors, child affective ODD symptoms, child behavioral ODD symptoms

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INTRODUCTION

Oppositional Defiant Disorder (ODD), one of the most commonly-occurring disorders in childhood (Egger and Angold, 2006; Lavigne et al., 2009), was defined by the DSM-5 as a recurrent pattern of angry/irritable mood, negativistic/defiant behavior, or vindictiveness toward authority figures (American Psychiatric Association, 2013). Children with ODD display persistent resistance, argumentation, and acts of aggression that disrupt interactions with peers and family members, instead of releasing occasional outbursts that result from intermittent frustration or negative events (American Psychiatric Association, 2013).

Previous studies suggested that ODD symptoms could be divided into two different dimensions, including affective and behavioral dysregulation (Burke and Loeber, 2010). Recent studies also provided empirical support for the two-dimension structure of ODD symptoms. For example, Lavigne et al. (2015) examined the metric and scalar of existing models to identify whether ODD consisted of multiple dimensions. The results suggested that the two-dimension model optimally represented the dimensionality of ODD symptoms (Lavigne et al., 2015). Symptoms of affective dimension included being touchy, short-tempered, spiteful, and resentful to others. On the other hand, symptoms of behavioral dimension included deliberately acting to annoy others, refusing to comply with majority's requests or consensus-supported rules, frequent arguing, and blaming others for their own mistakes. (American Psychiatric Association, 2013).

Links between Family Factors and ODD Symptoms

Previous studies have well-documented the significant linkage between family context and child psychological development (Hetherington and Martin, 1986), particularly in families with children having potential affective and behavioral problems (e.g., Cox and Paley, 2003; Smeekens et al., 2007; Lavigne et al., 2014). A wealth of literature has identified numerous family factors that placed children at increased risk of developing ODD, including poor family function, low marital quality, parental maladaptive behavior, paternal substance abuse, and low quality parent-child relationship (Burke et al., 2002; Greene et al., 2002; Marmorstein et al., 2009; Matthys and Lochman, 2010).

Both cross-sectional and longitudinal studies also proved the robust associations between family factors and child ODD symptoms. For example, Lavigne et al. (2014) found that higher scores on family risk factors (family conflict, parent hostility in parenting, child emotion temperament) were positively associated with child ODD symptoms in a cross-sectional study. Similarly, Smeekens et al. (2007) found that multiple family factors, like parent-child interaction, parent-child attachment, and various parental, child, and contextual characteristics, served as predictors of child later externalizing behavior problem in a longitudinal study.

Three Levels of Family Factors

Although previous studies have identified a wide range of family factors linked to child ODD symptoms, the majority of these studies focused almost exclusively on family factors at either one level or mixed levels. Different associations of multi-level family factors and child ODD symptoms remained unclear. Cox and Paley (2003) proposed that family was a dynamic and interactive system consisting of interdependent subsystems, including whole-family subsystem, parent-child subsystem, co-parenting subsystem, and marital subsystem. Factors at these subsystems served as important contexts in understanding child development (Cook and Kenny, 2006). Based on this theory, Lavigne et al. (2012) investigated the relations between family subsystems and child ODD symptoms. They proposed a multi-domain model of family factors for ODD symptoms encompassing contextual factors, parental depression, parenting, and child characteristics. Furthermore, they assessed the pathways of these multi-domain factors on child ODD symptoms. However, this model was too complex to clearly demonstrate the hierarchy of these family factors. Considering the complexity of Lavigne's model, Lin et al. (2013) proposed a concise three-level model on the basis of the Family System Theory (Cox and Paley, 2003) and the Bioecological Model (Bronfenbrenner, 2009), which delineated the mechanism of ODD development. This model divided family factors into three levels, including the whole level, the dyadic level (including couple dyadic and parent-child dyadic levels) and the individual level (including parental individual and child individual levels). In the present study, we adopted this comprehensive model to test the associations between family factors and two dimensions of ODD symptoms.

Family Factors and Child's Affective and Behavioral Problems

Previous research highlighted the crucial role of family function at the whole level in predicting children's affective and behavioral symptoms (Slee, 1996; Lucia and Breslau, 2006). Family cohesion, as one aspect of family function, was negatively correlated with child internalizing and externalizing problems (Lucia and Breslau, 2006). Children in less cohesive families were more likely to develop conduct disorder and delinquent symptoms (Leflore, 1988; Slee, 1996). In cohesive and well-adapted families, members were prone to interact with each other in a harmonious manner, which further promoted parental and children's emotion regulation abilities. Thus, children in such families were less likely to develop affective and behavioral problems.

Among the numerous dyadic level factors, marital quality, and parent-child relationship were noted as the most influential factors on children's psychological problems within the family (Lin et al., 2013). A considerable body of research has linked marital quality directly and indirectly to children's internalizing and externalizing problems (Cummings and Davies, 2002). Lower marital quality was directly related to subsequent child internalizing problems. Exposure to marital conflict was upsetting to children and appeared to elicit child maladjustment in both direct and indirect ways (Zimet and Jacob, 2001; Erath and Bierman, 2006). Additionally, poor parent-child relationship appeared to be a robust risk factor of children's behavioral adjustment (Masten and Garmezy, 1985). Negative parent–child relationships were significantly associated with child externalizing disorders (Waschbusch, 2002), namely attention-deficit/hyperactivity disorder (ADHD), conduct disorder (CD), and ODD (Burt et al., 2005). Secure parent-child relationships protected children from adverse developmental outcomes (Groh et al., 2014), whereas insecure parent-child relationship was a risk factor of child anxiety and other internalizing problems (Kerns and Brumariu, 2014).

Besides the factors at the whole and dyadic levels, specific individual characteristics of parents and children also accounted for the development of ODD symptoms (Eisenberg and Fabes, 2006; Cunningham et al., 2009). Smith (2010) suggested that parental emotion regulation played a vital role in child developmental outcomes. Parental dysregulated emotion contributed to inappropriate emotional expressions or behaviors toward children, which further predicted children's maladaptive emotional outcomes (Muhtadie et al., 2013). Moreover, parents with difficulty in emotion regulation exhibited poor emotional coaching, which would positively predict children's disruptive behavior, especially when children demonstrated high levels of emotion liability/negativity (Dunsmore et al., 2013). Previous studies also found that children's deficits in emotion regulation contributed to the manifestation of externalizing and internalizing symptoms (Southam-Gerow and Kendall, 2002; Kim and Cicchetti, 2010), whereas children with better emotion regulation demonstrated less subsequent internalizing and externalizing problems (Blandon et al., 2010; Kim and Cicchetti, 2010).

Parenting in Chinese Context

Previous studies indicated that ecological context played a crucial role in shaping family functioning, parenting styles, and child development (Schleyer-Lindenmann, 2006). Currently, the majority of studies examining the associations between parenting styles and child ODD symptoms were based on Western samples. The information on how parenting in non-Western societies, such as Mainland China, affect children's psychological development remains scarce. Moreover, the implementation of the one-child policy before 2015 introduced changes into parenting styles. Some families adopted the "child-centered" approach into child-rearing, particularly among well-educated parents (Chang et al., 2003). The "child-centered" parenting style linked children and parents together closely, but it also increased the possibility that children become spoiled. Meanwhile, most parents put high expectations on their children, expecting children to be obedient, respectful, and excellent in schoolwork. Consequently, parents would be under a lot of pressure, particularly when their children misbehaved. In the present study, we examined how the specific family context and parenting styles in China contribute to children's ODD symptoms.

The Present Study

We proposed a comprehensive framework concerning how multi-level family factors are differently related to children's ODD symptoms. Specifically, we included family function as the whole level family factor, marital quality, and parent-child relationship as the dyadic level family factors, and emotional regulation of both parent and child as the individual level family factors. Our study aimed to specify the different associations of the three-level family factors with child affective and behavioral oppositional defiant disorder symptoms. We hypothesized: (1) Among families with children identified with ODD, there are significant associations between three levels of family factors and children's ODD symptoms; (2) Three levels of family factors exhibit different associations with ODD symptoms. Specifically, family function, as the whole level factor and the most distal factor, is less correlated with ODD symptoms than family factors at the dyadic and individual levels. (3) Multi-level family factors exhibit different associations with two dimensions of children's ODD symptoms in that all family factors are more closely linked to affective ODD symptoms than to behavior ODD symptoms.

METHOD

Participants and Sample Procedure

Data in this study was derived from a large research project on family risk and protective factors of ODD children in China. Between 2013 and 2014, 14 elementary schools in northern (Beijing), eastern (Shandong province), and southwestern (Yunnan province) parts of China participated in this study. School psychologists in these 14 primary schools invited all the class master teachers who taught first grade through fifth grade to nominate the children in their classes who exhibited ODD symptoms. The nomination was based on the ODD symptoms assessment checklist, which was derived from the *Diagnostic and Statistical Manual of Mental Disorders* (DSM-IV-TR; American Psychiatric Association, 2000). In total, 187 class master teachers nominated 360 students from 7,966 children in the participating schools.

After the initial nomination, two clinical psychologists from Beijing Normal University further confirmed the assessment, using a semi-structured interview guide in interviewing class master teachers. The interview was based on the DSM-IV-TR's ODD diagnostic criteria. After the interview, two clinical psychologists discussed their assessments for each child to ensure accordance. Only children with both clinical psychologists' diagnoses of ODD were recruited into the current study. Ultimately, 305 children identified as displaying the symptoms of ODD were invited to participate in the research (3.8% of the children in the participating schools), and 282 parent-child dyads agreed to join the project. Thirty-three families were excluded from the analyses because more than 20% of the items on one questionnaire were missing.

The final ODD sample consisted of 249 parent-child dyads, including 80 father-child dyads and 169 mother-child dyads. Ages of parents ranged from 25 to 53 (paternal age $M = 38.35$, $SD = 5.08$; maternal age $M = 36.64$, $SD = 4.28$). All children (180 boys and 69 girls) aged between 6 and 13 ($M = 9.59$, $SD = 1.59$). Among these children, 80% were the only child in their families. Approximately, 141 families (56.6%) had a monthly income over 5,000 Chinese Yuan (the average monthly income for Chinese urban families is about 5,485 Chinese Yuan; National Health and Family Planning Commission of the PRC, 2015). More than half of the participants, including 151 fathers (60.6%) and 141 mothers (56.6%), reported that they completed junior college education or higher education.

Survey Procedure

After signing the informed consents, each participating child was asked to deliver a parent survey package to their father/mother. Parents were invited to fill in the parent survey and return the completed surveys to the class master teachers in 1 week.

All study procedures, including informed consent and child assent, were approved by the Institutional Review Board of Beijing Normal University. Each participant received a token of appreciation to acknowledge their participation in the study. All the 305 children who met the criteria for ODD were provided with the opportunity for treatment from psychiatrists in Beijing Anding Hospital, psychological counselors, and family therapists from the Center of Family Study and Therapy at Beijing Normal University.

Measures

Two Dimensions of ODD Symptoms

Two dimensions of ODD Symptoms were assessed by an 8-item scale derived from the eight ODD symptoms indicated in

DSM-IV-TR. This scale measures child ODD symptoms through two subscales of negative affect and oppositional behavior, each subscale includes 4 items that had the highest item total correlation coefficient (e.g., ODD affective symptoms "He/she is touchy or easily annoyed," ODD behavioral symptoms "He argues often and blames others for their own mistakes"). Child ODD symptoms were collected in family settings. Parents evaluated children's ODD symptoms, using a dichotomous measure (0 = *no*, 1 = *yes*). A higher total score indicates more ODD symptoms. In the current study, the Cronbach's α for the negative effect and the oppositional behavior were 0.78 and 0.71.

Whole Level Measures

Family Function

Parents reported on their family function using the Family Adaptability and Cohesion Evaluation Scale (FACES-II; Olson, 2000), which has proved to be an appropriate measure for Chinese families (Chen et al., 2011). FACES-II assesses the family function in two dimensions: Adaptability (14 items; e.g., "In solving problems, the children's suggestions are followed"; α = 0.74 for this sample) and Cohesion (16 items; e.g., "Family members like to spend free time with each other"; α = 0.71 for this sample). Father/mother reported their perception of actual conditions in the family, using a 5-point scale ranging from 1 (*almost never*) to 5 (*almost always*). A higher total score on FACES-II indicated better adaptability and cohesion in the family. In the current study, the Cronbach's α for FACES-II was 0.84.

Dyadic Level Measures

Marital Quality

Parents reported on their own marital quality using the Dyadic Adjustment Scale (DAS; Spanier, 1976), which was a valid measure of marital relationship quality for Chinese couples (Shek, 1995; Gau et al., 2012). Father/mother completed four subscales of DAS: Dyadic Consensus (13 items; e.g., "Career decisions"; α was equal to 0.77), Dyadic Satisfaction (10 items; e.g., "Are you confident in your mate?"; α was equal to 0.83), Affectional Expression (4 items; e.g., "Demonstrations of affection"; α was equal to 0.60), and Dyadic Cohesion (5 items; e.g., "Work together on a project"; α was equal to 0.83). Most items of the DAS were scored using a 6-point scale, but some items were scored in 2-point and 5-point scale to better express the meaning. Higher total scores indicated higher marital quality. The Cronbach's α for the DAS total scale in the current sample was 0.90.

Parent-Child Relationship

The Parent Stress Index-Short Form (PSI-SF; Abidin, 1995) was utilized to measure the parent-child relationship. Parents were asked to rate 36 items from various aspects of their perceived interaction stress with their children, using a 5-point scale (1 = *strongly agree*, 5 = *strongly disagree*). The PSI-SF has three subscales: Parental Distress (12 items, indicating the distress resulting from personal factors such as depression or conflict with a partner and from life restrictions due to the demands of child-rearing), Parent-Child Dysfunctional Interactions (12 items, indicating parents' dissatisfaction with interactions with their children and the degree to which parents find their children unacceptable), Difficult Child Characteristics (12 items, measuring parents' perceptions of their children's self-regulatory abilities), and a Defensive Responding scale (7 items) that indicates the degree to which the parent might be attempting to deny or minimize problems. Higher scores overall indicated lower levels of parental stress and better parent-child relationship. Internal consistency was good for the PSI-SF total scale (α = 0.94).

Individual Level Measures

Parental Emotional Regulation

The Difficulties in Emotion Regulation Scale (DERS; Gratz and Roemer, 2004) was used to assess parental emotion regulation ability in the present study, which has been previously used in Chinese samples (Yan et al., 2015). The DERS is a 36-item self-report questionnaire providing a comprehensive assessment of parents' overall emotion regulation difficulties as well as six specific dimensions: (a) Non-acceptance of negative emotional responses (6 items; α = 0.71), (b) Difficulties engaging in goal-directed behaviors when experiencing negative emotions (5 items; α = 0.54), (c) Difficulties controlling impulses when experiencing negative emotions (6 items; α = 0.78), (d) Limited access to emotion regulation strategies perceived as effective (8 items; α = 0.78), (e) Lack of clarity of emotional responses (α = 0.682) (5 items; α = 0.65), and (f) Lack of awareness of emotional responses (6 items; α = 0.41). The Awareness subscale was deleted from DERS in this current study because of low factor loading ($\beta = -0.07$, $p = 0.31$). Father/mother rated the items on a 5-point scale (1 = *almost never*, 5 = *almost always*). Total scores were summed for DERS, with higher scores indicating more difficulties in emotional regulation. Internal consistency was good for the DERS total scale in the current sample (α = 0.84).

Child Emotion Regulation

Children's emotion regulation ability was assessed using the Emotion Regulation Checklist (ERC; Shields and Cicchetti, 1997), which demonstrated good internal reliability in China (Chang et al., 2003). The ERC is comprised of 23 items that target processes central to children's negative emotionality and regulation. The ERC yields two subscales: Liability/Negativity subscale (15 items) and Emotion Regulation subscale (8 items). Because this study targeted the children's overall management of emotions, the Emotion Regulation subscale was used (e.g., "Can modulate excitement in emotionally arousing situations"). Items are scored on a 4-point scale ranging from 1 (*never*) to 4 (*almost always*). A higher total score was reflective of better emotion regulation of child. In the current study, the Cronbach's α of the Emotion Regulation subscale was 0.82.

Data Analysis Procedures

All data in the current study was based on self-reports from parents. To reduce concerns about common method variance, we conducted Harman's one-factor test using Exploratory Factor analysis with a principal axis factoring method of extraction. The first extracted component accounted for 16.48% of the total variance, indicating that the common method variance was not of great concern (Aulakh and Gencturk, 2000).

Then, the preliminary analyses were performed to evaluate the descriptive statistics and correlations between the study variables. Preliminary data analyses were performed using SPSS 20.0. Bivariate correlations were conducted to test initial relations between variables (see **Table 1**).

Finally, the structural equation model (SEM, see **Figure 1**) was tested using Mplus Version 7.0 (Muthé and Muthén, 2010). Model fit criteria used in this study were chi-square statistic (χ^2), the goodness-of-fit index (CFI), the Tucker-Lewis index (TLI), the root mean square error of approximation (RMSEA), and the standardized root mean residual (SRMR). A model is typically considered as acceptable fit the data when the CFI and TLI values are larger than 0.90, the RMSEA value < 0.08, and the SRMR value is no > 0.08 (Hu and Bentler, 1999).

RESULTS

Preliminary Analysis

Descriptive statistics and bivariate correlational analyses were presented in **Table 1**. As shown in **Table 1**, basically each dimension of family function, marital quality, parent-child relationship, parent emotion regulation, child emotion regulation, and ODD symptoms was significantly correlated with each other ($ps < 0.01$).

SEM Analysis

Before testing the hypothesized structural model, the preliminary analyses examined the gender, primary school grade, number of siblings, as well as household income differences in two dimensions of child oppositional defiant disorder symptoms (see **Table 2**). Among all these demographic variables, only the children's gender was significantly associated with observed variables. Hence, children's gender was placed into the subsequent path model test as covariates, yet it did not show any significant effect in the models presented below.

Table 2 presented the means and standard deviations for ODD affective and behavioral symptoms across basic demographic variables. Further, ANOVA was conducted to compare two dimensions of child ODD symptoms in various demographic groups. There was no significant difference of child affective ODD symptoms [$F_{(1,\,247)} = 1.17$, $p = 0.28$] and child behavioral ODD symptoms [$F_{(1,\,247)} = 0.06$, $p = 0.81$] between families with only one child and families with siblings. Families with different monthly incomes did not show significant difference in reporting child affective ODD symptoms [$F_{(1,\,247)} = 2.38$, $p = 0.12$] and child behavioral ODD symptoms [$F_{(1,\,247)} = 0.36$, $p = 0.55$]. Children from different primary grades did not score differently on child affective ODD symptoms [$F_{(2,\,246)} = 2.08$, $p = 0.13$] and child behavioral symptoms [$F_{(2,\,246)} = 0.59$, $p = 0.56$]. However, it is noteworthy that there were significant differences in boy's and girl's affective and behavioral ODD symptoms. Hence, children's gender was placed into the subsequent model test as covariates, yet it did not show significant effect on either dimension of children's ODD symptoms in the model presented below. A model of proposed relationships among the study variables, controlling for child gender, was presented in **Figure 1**.

Model results indicated that the proposed model fit the data very well ($\chi^2 = 236.23$, $df = 114$, CFI = 0.95, TLI = 0.93, RMSEA = 0.06). Results of the model indicated that on the whole level, family function was not significantly associated with either dimensions of child ODD symptoms. When testing the association of dyadic level factors on child ODD symptoms, we found that both marital quality ($\beta = 0.19$, $p < 0.05$) and parent-child relationship ($\beta = -0.64$, $p < 0.001$) were significantly linked to the ODD affective symptoms. However, only the parent-child relationship was significantly related to the ODD behavioral symptoms ($\beta = -0.66$, $p < 0.001$). At the individual level, both parental emotion regulation ($\beta = -0.17$, $p < 0.05$) and child emotion regulation ($\beta = -0.25$, $p < 0.001$) were significantly correlated with child ODD affective symptoms, but only child emotion regulation was negatively linked to ODD behavioral symptoms ($\beta = -0.19$, $p < 0.01$).

DISCUSSION

The current study tested the multilevel family factors model proposed by Lin et al. (2013), trying to provide a comprehensive understanding of the associations between multilevel family factors and two dimensions of child ODD symptoms. Specifically, the current study examined family factors at the whole, dyadic, and individual levels in explaining child ODD behavioral and affective symptoms. Consistent with our hypotheses, three levels of family factors showed different associations with affective and behavioral ODD symptoms in Chinese Mainland families. Family factors at the whole level were less correlated with ODD symptoms than factors at the dyadic and individual levels. Also, multi-level family factors exhibited different associations with two dimensions of child ODD symptoms in that all family factors were more closely linked to affective ODD symptoms. The present study highlighted the value of studying child affective and behavioral ODD symptoms in the family context.

Model results indicated that multilevel family factors were differently associated with child ODD symptoms. The findings in the current study attracted our attention to the distal and proximal ends of the socio-biological environment in child ODD development (Lin et al., 2013). As the most distal factor, family function was not significantly associated with either dimension of child ODD symptoms. This finding was consistent with our hypothesis, as well as findings of previous studies (e.g., Grant et al., 2006). Representing the wholeness and higher order of family environment, family function was not directly related to child psychopathological outcomes (Grant et al., 2006). In the proximal end of family environment, dyadic and individual levels family factors exhibited stronger correlation with child ODD symptoms than the distal factors. All the factors in the dyadic and individual levels were significantly linked to child affective ODD symptoms, but only the most proximal factors (the factors directly related to children—parent-child relationship and child emotion regulation) showed significant linkages with child behavioral symptoms. The findings indicated that the family

TABLE 1 | Variable descriptions and correlations among variables for children with ODD.

	M	*SD*	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16
1. Family-adaptability	71.86	10.17	–															
2. Family-cohesion	48.79	7.84	0.76**	–														
3.PSI-parental distress	38.36	7.46	0.37**	0.46**	–													
4. PSI-dysfunction interaction	43.74	7.65	0.43**	0.49**	0.62**	–												
5. PSI-difficult child	37.77	8.58	0.39**	0.44**	0.57**	0.73**	–											
6. DAS-dyadic satisfaction	45.82	6.15	0.59**	0.44**	0.29**	0.35**	0.33**	–										
7. DAS-dyadic consensus	56.55	10.61	0.52**	0.45**	0.28**	0.31**	0.34**	0.64**	–									
8. DAS-dyadic cohesion	17.20	4.27	0.51**	0.43**	0.25**	0.34**	0.29**	58**	0.49**	–								
9. DAS-affective expression	11.93	2.27	0.54**	0.42**	0.31**	0.33**	0.33**	0.67**	0.69**	0.45**	–							
10. DERS-nonacceptance	11.78	4.01	−0.22**	−0.27**	−0.35**	−0.25**	−0.18**	−0.18**	−0.08	−0.13**	−0.15**	–						
11. DERS-goals	9.45	2.85	−0.32**	−0.33**	−0.32**	−0.33**	−0.16**	−0.22**	−0.17**	−0.22**	−0.23**	0.35**	–					
12. DERS-impulse	11.00	3.51	−0.35**	−0.36**	−0.41**	−0.37**	−0.28**	−0.20**	−0.12	−0.12	−0.20**	0.51**	0.57**	–				
13. DERS-strategies	14.81	4.50	−0.35**	−0.34**	−0.46**	−0.42**	−0.24**	−0.28**	−0.17**	−0.20**	−0.22**	0.58**	0.65**	0.70**	–			
14. DERS-clarity	9.82	2.97	−0.34**	−0.38**	−0.31**	−0.43**	−0.28**	−0.23**	−0.21**	−0.29**	−0.21**	0.38**	0.51**	0.50**	0.56**	–		
15. Child emotion regulation	26.34	3.50	0.33**	0.31**	0.25**	0.49**	0.40**	0.30**	0.22**	0.29**	0.26**	−0.28**	−0.25**	−0.35**	−0.40**	−0.44**	–	
16. ODD affective symptoms	1.41	1.39	−0.28**	−0.31**	−0.35**	−0.49**	−0.62**	−0.20**	−0.16**	−0.14*	−0.15*	0.19**	0.11	0.22**	0.21**	0.17**	−0.43**	–
17. ODD behavioral symptoms	1.23	1.33	−0.31**	−0.35**	−0.43**	−0.54**	−0.64**	−0.23**	−0.23**	−0.22**	−0.20**	0.21**	0.16**	0.28**	0.31**	0.24**	−0.44**	0.71**

**p < 0.05; **p < 0.01.*

TABLE 2 | The characteristics of children with ODD.

	ODD affective symptoms *M* (*SD*)	ODD behavioral symptoms *M* (*SD*)
CHILD GENDER		
Boy ($n = 180$)	1.53(1.37)	1.35(1.34)
Girl ($n = 69$)	1.09(1.40)	0.91(1.28)
F	5.24*	5.50*
η^2	0.03	0.02
SIBLINGS		
Only child ($n = 199$)	1.46(1.38)	1.24(1.31)
One or more sibling ($n = 50$)	1.22(1.40)	1.19(1.42)
F	1.17	0.06
η^2	0.00	0.00
FAMILY INCOME		
< 5,000 RMB per month ($n = 112$)	1.26(1.39)	1.17(1.33)
Over 5,000 RMB per month ($n = 137$)	1.53(1.38)	1.28(1.34)
F	2.38	0.36
η^2	0.01	0.00
CHILD PRIMARY SCHOOL GRADE		
Junior grade ($n = 70$)	1.64(1.39)	1.36(1.36)
Middle grade ($n = 117$)	1.41(1.33)	1.22(1.30)
Upper grade ($n = 62$)	1.15(1.46)	1.11(1.37)
F	2.08	0.59
η^2	0.02	0.00

*$p < 0.05$.
Junior grade, 1st and 2nd grade in primary school;
Middle grade, 3rd and 4th grade in primary school;
Upper grade, 5th and 6th grade in primary school.

factors in the proximal end of family environment exhibited robust association with child ODD symptoms. The family factors in the distal end of family environment might exert its effect indirectly, through its effect on the proximal factors, on child psychological development (Grant et al., 2006).

The findings in the current study also indicated that family factors were more significantly related to children's affective ODD symptoms than to children's behavioral ODD symptoms. All the family factors at the dyadic and individual levels exhibited significant associations with child affective ODD symptoms, but only the most proximal factors (the factors directly related to children—parent-child relationship and child emotion regulation) showed significant association with child behavioral ODD symptoms. The result indicated that child affective ODD symptoms were more susceptible to family factors than child behavioral ODD symptoms. It was explainable that the behavioral problems served as the outer form of child affective problems, which were usually motivated by affective problems (Carver and Scheier, 2004; Aldao and Christensen, 2015). Deficits in emotion regulatory abilities were known to contribute to the manifestation of externalizing symptoms (Southam-Gerow and Kendall, 2002; Yap et al., 2007) Additionally, a network analysis of ODD symptoms also suggested that affective symptoms appeared to be relatively central to the homogenous ODD symptoms network while behavioral symptoms fell along the periphery (Smith et al., 2016). For children with ODD, affective ODD symptoms placed them at a greater risk of developing into ODD behavioral symptoms. Family risk factors exerted effects on child affective ODD symptoms, and went further on child behavioral ODD symptoms. Aggravation of affective symptoms might lead to further deterioration of the overall ODD symptoms.

The findings of the current study demonstrated the different associations between family factors and two dimensions of child ODD symptoms. Making sense of these findings, future intervention should focus on family factors at the dyadic and individual levels, which might firstly decrease child affective ODD symptoms and further decrease child behavioral ODD symptoms. The different associations of family factors with child affective and behavioral ODD symptoms in the current study were generally consistent with previous literature in that ODD symptoms consisted of two separate dimensions. This finding further lend credit to the two-dimension structure of ODD symptoms (Lavigne et al., 2012). ODD symptoms should be divided into two separate dimensions that differentially depict affective and behavioral ODD symptoms.

Several limitations of the current study should be addressed when interpreting the data. First, all data in the current study was based on self-reports from the child's father/mother. Although the Harman's one-factor test indicated that the common method variance was not of great concern, further study should try different measures. Second, this study adopted a cross-sectional study method; therefore, causal relationships could not be established from this study. The associations between family factors and child ODD symptoms were also very likely to be bi-directional and transactional, the multi-level family factors might not be paratactic as well. Distal family factors (e.g., family function) might predict dyadic level factors (e.g., marital relationship and parent-child relationship) and further influence parental and children's individual characteristics (e.g., parental and child emotion regulation), which would finally result in ODD. Further research with longitudinal design is encouraged to examine the direction of the associations between multilevel family factors and child ODD symptoms and the potential mediating effect. Some paths (marital quality and parental emotion regulation to child affective and behavioral ODD symptoms) in the SEM model also exhibited opposite patterns with the correlation results. Currently the underlying mechanism remains unclear, further studies are needed to explore the underlying mechanism.

Despite these limitations, the current study provided a comprehensive understanding of the associations between multilevel family factors and two dimensions of child ODD symptoms. It also provided useful applications for intervention in decreasing children's ODD symptoms from the perspective of multilevel family environment. Instead of focusing solely on parent-child interaction, which was widely adopted in most interventions for ODD (Lavigne et al., 2012), understanding child affective and behavioral ODD symptoms in the broader family context is critical to develop the best possible interventions for child ODD. Additionally, professionals should consider the

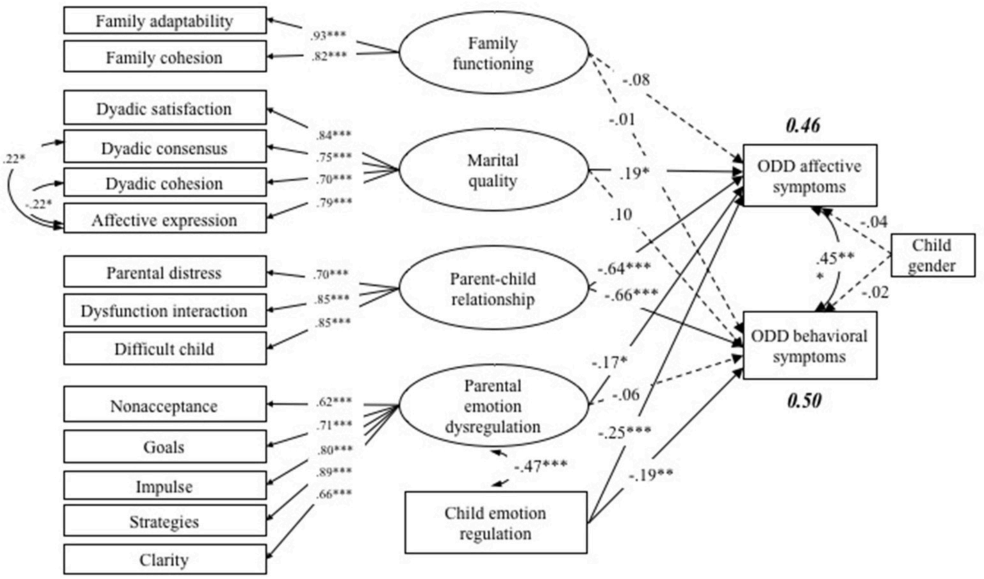

FIGURE 1 | Model of multilevel family factors associated with ODD affective and behavioral symptoms. This is a standardized model, $^*p < 0.05$, $^{**}p < 0.01$, $^{***}p < 0.001$. The interrupted line means the path is not statistically significant.

type of child ODD symptoms and the different associations between multilevel family factors and affective and behavioral oppositional defiant disorder symptoms when designing interventions. If a child exhibits affective oppositional defiant disorder symptoms, interventions and counseling strategies should focus more on strengthening parent-child relationships, marital relationship, and improving parent and child emotion regulation. On the other hand, if a child exhibits behavioral oppositional defiant disorder symptoms, interventions and counseling strategies should focus more on strengthening parent-child relationships and improving child emotion regulation.

AUTHOR CONTRIBUTIONS

Each of the five authors contributed a lot to the current manuscript. YT: Substantial contributions to the conception or design of the work; the acquisition, analysis, and interpretation of data for the work; drafting the work and revising it critically for important intellectual content; XL: Substantial contributions to the conception or design of the work; revising the draft critically for important intellectual content; PC, QZ, and XH: editing the whole paper and revising the language; ensuring the work are appropriately investigated and resolved. Each author read the final version of the current manuscript and support its submission.

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Neural Substrate of Group Mental Health: Insights from Multi-Brain Reference Frame in Functional Neuroimaging

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Contemporary mental health practice primarily centers around the neurobiological and psychological processes at the individual level. However, a more careful consideration of interpersonal and other group-level attributes (e.g., interpersonal relationship, mutual trust/hostility, interdependence, and cooperation) and a better grasp of their pathology can add a crucial dimension to our understanding of mental health problems. A few recent studies have delved into the interpersonal behavioral processes in the context of different psychiatric abnormalities. Neuroimaging can supplement these approaches by providing insight into the neurobiology of interpersonal functioning. Keeping this view in mind, we discuss a recently developed approach in functional neuroimaging that calls for a shift from a focus on neural information contained *within* brain space to a multi-brain framework exploring degree of similarity/dissimilarity of neural signals *between* multiple interacting brains. We hypothesize novel applications of quantitative neuroimaging markers like inter-subject correlation that might be able to evaluate the role of interpersonal attributes affecting an individual or a group. Empirical evidences of the usage of these markers in understanding the neurobiology of social interactions are provided to argue for their application in future mental health research.

Keywords: inter-subject correlation, functional neuroimaging, multi-body neuroscience, mental health, collective consciousness, interpersonal space

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MENTAL HEALTH IN INDIVIDUALS AND BEYOND

> All actual life is encounter
>
> —Buber, 1923/1970

Contemporary mental health paradigm limits mental disorders to problems within an individual. DSM-5 defines mental disorder as "a syndrome characterized by clinically significant disturbance in an individual's cognition, emotion regulation, or behavior" (American Psychiatric Association, 2013). Accordingly, mental health interventions mostly focus on treatment of individual patients. Psychopharmacotherapy targets the neurochemical processes taking place inside individual brains. Psychotherapy usually has a focus on the psychological processes in an individual: for example, cognitive behavioral therapy focuses on an individual's maladaptive patterns of thought and behavior, and psychodynamic psychotherapy focuses on unconscious contents of the psyche of an individual (Leichsenring and Leibing, 2007; Beck, 2011). Even interpersonal psychotherapy (IPT), though based on the recognition of the crucial role played by interpersonal factors in the genesis of mental illness, concentrates on individual sufferers in practice. During IPT sessions, a therapist guides an individual to learn appropriate emotional expressions and other skills to address problems in the relationship with significant others or to modify his/her expectation of the relationship (Markowitz and Weissman, 2004). Thus, with the exception of family and group therapies, in all major therapeutic approaches in mental health including IPT, "intra-personal" space is held as the main site of anomaly. Processes taking place in the interpersonal space are discussed as mere risk factors/predisposing factors that can causally affect the intra-personal space.

Contrary to the individual-centric approaches, an interpersonal perspective of mental health is built on the assumption that the interpersonal functioning may be deranged, independent of the abnormalities in individual sufferers. Thus, it aims to bring disturbed interpersonal relationships into the clinic and direct medical interventions toward the improvement of the same. Hence, interpersonal perspective necessitates a direct evaluation of the functioning of the interpersonal space comprising of the patient, related individuals, and their interactions rather than the assessment of the social and interpersonal skills of the patient as employed in approaches that focus on the individual patient. A broader clinical scope of services can be offered as impaired interpersonal spaces are present in several mental illnesses. For example, persons with paranoid personality disorder are characterized by pervasive, irrational mistrust, and suspicion of other people (Bernstein et al., 1993). The abnormality in this disorder is thus contingent on the company of other individuals and the corresponding interpersonal spaces. Likewise, an individual suffering from antisocial personality disorder has a pervasive pattern of disregard for the rights of others (Swanson et al., 1994). Dependent personality disorder is marked by an excessive psychological dependence on others (Disney, 2013). A person with social anxiety disorder has a fear of being closely watched and criticized by others (Stein and Stein, 2008). Autism spectrum disorder (ASD) is characterized by an impaired ability to communicate with other people (Lord et al., 2000). In sexual sadism disorder, sexual gratification is derived from inflicting psychological or physical sufferings on others (Hamilton and Rosen, 2016). In all these examples, pathology lies in the interpersonal space and hence a prime focus on the individual is bound to be inadequate for comprehensive understanding of the disorder in question.

Certainly, there are other disorders where abnormality can be defined within an individual or in other words in the intra-personal space. Nevertheless, all mental disorders unfold in an interpersonal context. Accordingly, interpersonal processes shape the symptomatology, therapeutic alliance, rehabilitation, and other factors that ultimately determine the course of the disease and treatment outcomes. Thus, even in treating individual patients, a direct evaluation of the interpersonal functioning may reveal important insights.

There has been a growing recognition of the importance of interpersonal functioning in mental health and the necessity to evaluate it (King-Casas and Chiu, 2012; Schilbach et al., 2013; Schilbach, 2016). A handful of recent studies have borrowed game theoretic paradigms from behavioral economics to probe interpersonal dynamics in mental health disorders. An example being the study by King-Casas et al. (2008) in which an iterated version of the economic exchange "trust" game is employed to probe the cooperation in bipolar personality disorder (BPD). Participants included normal controls paired with either another normal individual or an individual with BPD, resulting in two types of "dyads" or pairs. Compared to a normal–normal dyad, a dyad with a BPD individual showed a consistent tendency to rupture cooperation and an impaired ability to repair cooperation once it was ruptured. Another study has found greater chances of mutual defection in dyads that include high-psychopathic individuals in a cooperation game known as the "prisoner's dilemma" (Rilling et al., 2007). Lack of facilitation of donation choices when being observed by others has also been reported employing a modified dictator game in the group comprising of autistic individuals (Yoshida et al., 2010). Neuroimaging studies have also been employed to investigate neural underpinnings of disordered social interaction. However, most of these studies, following the conventional way of reporting neuroimaging results, describe neural activity in a coordinate system constructed around a single brain, i.e., an individual brain reference frame is employed. For example, in the same BPD study described above, the researchers found decreased anterior insular cortex activation in BPD patients relative to their typically developing (TD) counterparts in the backdrop of a faltering cooperation with their partners. In spite of initial promises and more than 20 years of worldwide research efforts, neuroimaging under the individual brain-centered regime has yet to achieve the desired effect in the domain of mental health research (Linden, 2012). Although neuroimaging provides even more sensitive measures of brain structures and functions relevant for human cognitive processes, it has failed, so far, to come up with a single reliable marker for diagnosis of major mental disorders (Deacon, 2013). An attractive postulate is that much relevant information regarding mental health is contained in the interpersonal interactions that cannot be fully captured by limiting our search of markers to individual brains. This possibility calls for employing a multi-brain reference frame in functional imaging studies where interactions among multiple brains are the main object of study, rather than individual brain activations. For example, in the King-Casas et al. study, an fMRI investigation incorporating the multi-brain framework could have explored how the interdependence of the hemodynamic signals from the normal-BPD dyads differ from that of the normal–normal dyads. In the remainder of this article, we would like to draw the attention of mental health researchers, to some exciting new developments in the field of neuroimaging that hold tremendous promise to capture the interpersonal attributes of mental health. We briefly present the history of the evolution of reference frame in functional neuroimaging from single region-of-interest (ROI) to multiple brains in section "Functional Neuroimaging: Evolution of Reference Frame." We then identify potential application areas of multi-brain neuroimaging in mental health research (section "Multi-Brain Reference Frame of Functional Neuroimaging and Mental Health Research: Some Potential Application Areas"). Following this, we further demonstrate some theoretical implications of this quantitative "description" of the interpersonal perspective of mental health and discuss why this should be seen as a compliment, *rather than a substitute*, for the existing rich body of qualitative research on this topic (section "Collective Mental State and Multi-Body Reference Frame of Functional Neuroimaging: Some Theoretical Implications").

FUNCTIONAL NEUROIMAGING: EVOLUTION OF REFERENCE FRAME

The primary research strategy of functional neuroimaging in the late 20th century was the localization of a particular brain function to a distinct region of the brain (Brett et al., 2002). For example, in fMRI, the goal was to identify brain voxels showing significant relative blood oxygenation level change compared to a baseline as an indirect measure of brain activity in a particular sensory, motor, or cognitive task (Forster et al., 1998) (see **Figure 1A**). For the last two decades, however, there is a gradual shift toward a paradigm giving equal importance to functional integration of information over distinct brain areas. This shift of the reference frame from brain regions to the whole brain (see **Figure 1B**, Hong et al., 2013) is an acknowledgment of the fact that different aspects of a particular brain function are sub served in distinct brain areas but the overall performance is dependent on integration among the distinct modules across multiple spatiotemporal scales (Friston, 1994). The recognition that spatiotemporal integration of brain activity has functional implications leads to its exploration using functional connectivity analysis that measures statistical dependence in neuronal activation patterns of anatomically separated brain regions (e.g., "region A correlates with region B") (Rogers et al., 2007). Research studies have employed effective connectivity analysis that explores how different parts of the brain impart causal influences on each other ("region A drives region B") (Friston, 1994). The functional and effective connectivity is typically quantitated in EEG/magnetoencephalography (MEG) recordings or BOLD signals on the basis of correlation, coherence, Granger causality (GC) or transfer entropy between time series of different regions (Li et al., 2009). Several other sophisticated statistical tools including graph theoretical analysis have also been employed (Toppi et al., 2015). From the perspective of mental health, localization of functional neuroimaging, so far, has failed to identify pathology in a single brain region that is causally related to any of the major mental illnesses (Deacon, 2013). The emergence of the connectivity analysis encourages the neuroscience community to explore the possibility that more information regarding neural correlates of mental illnesses may be found at the level of the interaction of distributed neural systems rather than in discrete brain regions (Garrity et al., 2007; Greicius et al., 2007; Broyd et al., 2009; Woodward and Cascio, 2015).

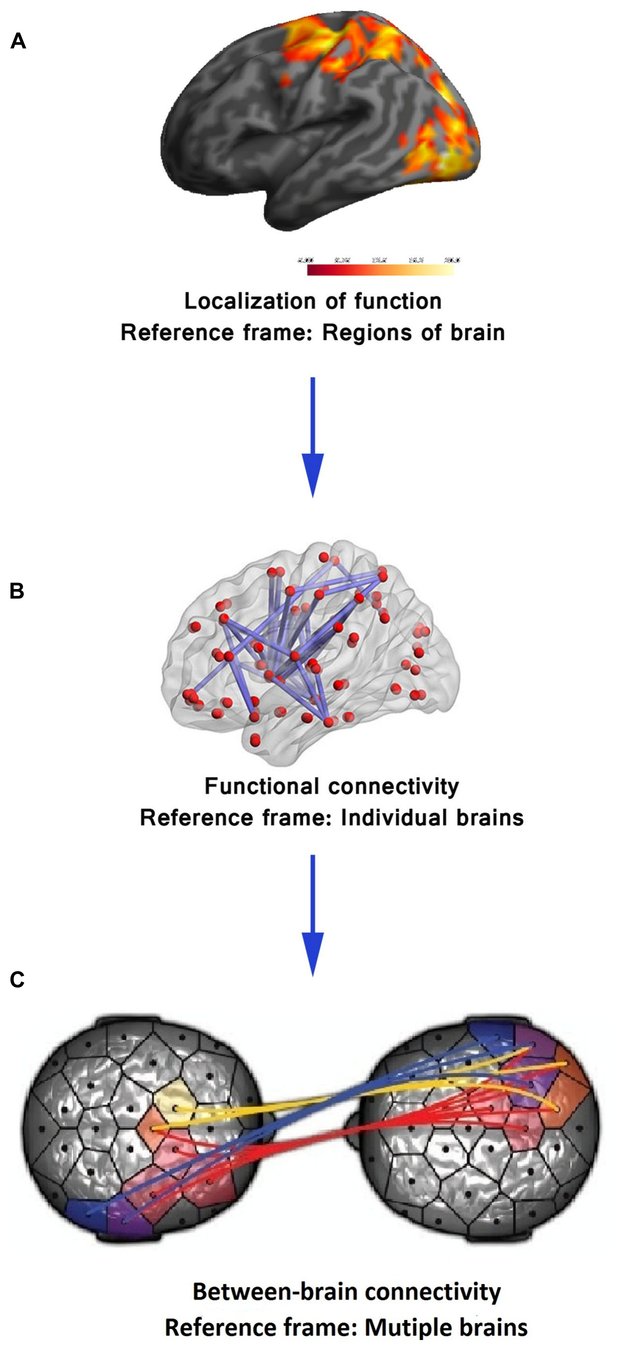

FIGURE 1 | Functional neuroimaging: evolution of reference frame. **(A)** In its early years, the sole focus of functional neuroimaging techniques (e.g., fMRI) was to localize brain function to distinct brain regions. **(B)** The first paradigm shift took place with experimental validation of functional integration over distinct brain areas using measures of functional connectivity and shifting of the reference frame to the whole of the brain. Adapted from "Decreased functional brain connectivity in adolescents with internet addiction," by Hong et al. (2013). Copyright 2013 by Hong et al. used under Creative Commons Attribution License. **(C)** The emerging techniques to assess between-brain functional connectivity calls for a second paradigm shift from a single brain to a multi-brain reference frame. Adapted from "Toward a two-body neuroscience," by Dumas (2011). Copyright 2011 by Landes Bioscience. Adapted with permission.

At present, the domain of functional neuroimaging may be on the brink of a second paradigm shift – quantifying the brain interactions *between* individuals transcending the boundary of the skull (Tognoli et al., 2007; Dumas, 2011; Hasson et al., 2012). In the arena of cognitive neuroscience, there is a growing recognition of the common knowledge that, in most social settings, the human brain works in interaction with other brains. In many daily cognitive tasks, from the simplest of verbal interactions between two individuals to playing games, shopping in the market, teaching in the classroom, or kissing, more than one brain cooperate/compete with each other establishing a "coupling" between themselves (Tognoli et al., 2007; Dumas, 2011; Hasson et al., 2012). The structure of this coupling shapes and constrains the activity of the individual brains to a certain extent. Therefore, a proper understanding of brain activity requires putting it in an interpersonal context. This realization is paralleled by the emergence of inter-brain connectivity analyses in functional neuroimaging (see **Figure 1C**) analogous to within-brain connectivity analysis. Various developments in neuroimaging hardware and experimental paradigm have made this shift to multi-brain reference frame possible. In fMRI studies, two subjects are typically scanned simultaneously either in two different scanners (see **Figure 2A**, Casebeer, 2003) connected over the internet (Montague et al., 2002) while they interact with each other using computer interface or in a single scanner using specially designed double-head volume coil (Lee et al., 2012) (**Figure 2B**) making face-to-face interaction possible. In EEG [and Functional Near-Infrared Spectroscopy (fNIRS)] experiments, either multiple devices are employed for recording simultaneous data from multiple subjects (Babiloni et al., 2006) or a single multichannel device is used (see **Figure 2C**, Baker et al., 2016) with a portion of the channels dedicated to each participant (Liu et al., 2016). In MEG, simultaneous recordings are usually performed in separate scanners (see **Figure 2D**, Hirata et al., 2014). However, for tasks that do not need online time-to-time interaction, e.g., studies involving social perception, simultaneous recording is not an essential requirement for studying interpersonal processing. A person's brain activity can be recorded while she performs some socially relevant behavior (e.g., narrates a story, expresses some emotion, shows some gestures) that can be audio or videotaped and played to another person (perceiver) who is recorded in turn. This setup acts as a substitute for the live computer interface used in simultaneous "hyperscanning" paradigms described above. Thereafter, various time series analysis techniques can be used to quantify the degree of togetherness and dissimilarity between signals obtained from the two individuals.

Inter-Subject Correlation (ISC)

The simplest measure imaginable to quantify inter-brain neural processes is a correlation. Consequently, a multitude of studies have employed ISC analysis as a measure of information exchange between two brains. For example, in a 2010 study (Stephens et al., 2010), fMRI was recorded when a speaker narrated an unrehearsed life story within the MRI scanner. The story was recorded and later played for listeners while further fMRI scans were done. The study found strong ISC between time-locked neural dynamics of speaker's and listener's brain in various regions, including low-level auditory processing areas, auditory comprehension areas, sound production areas, and areas known to be involved in processing semantic and social information (**Figure 3A**). Moreover, ISC measures correlated well with the level of understanding of the story, and disappeared when there was a failure of communication. Similar neural-functional coupling has been observed during non-verbal communication involving gestures to communicate meaning (Schippers et al., 2010). Expanding the mirror neuron theory, first proposed for monkey premotor cortex (Rizzolatti et al., 1996), a more general framework suggests that understanding of the action, emotion, and sensation of other individuals crucially depends on simulation of inner state of others through activation of specific brain regions or neurons in the perceiver's brain (Keysers and Gazzola, 2009). Thus, as the above examples show, ISC can be seen as a neural marker of successful inter-brain communications.

Understandably, among all inter-brain communications, communication of emotion is the most important process for the purpose of mental health. Several neuroimaging studies have supported the idea that emotional expression and perception are intimately related (Jackson et al., 2005; Jabbi et al., 2008). These observations have been further extended using ISC analysis. In an fMRI study, female participants were asked to freely express their emotional feeling within the scanner while their facial expressions were videotaped and shown to their romantic partners who were also being scanned (Anders et al., 2011). The subsequent ISC analysis showed significant coupling of neuronal activity between the female and male partners during sending and receiving affective cues. Recently, another study (Nummenmaa et al., 2012) found that while watching movies depicting unpleasant, neutral, and pleasant emotions, participants' brain activity was synchronized not only in lower- and higher-order sensory areas but also in emotion processing, visual attention, and mental simulation networks (**Figure 3B**). This may reflect the neurobiological processes that help in mental simulation of other's emotional, attentional, and sensory states by synchronizing the brain activities of specific neural circuits. This notion was further validated by the observations from the same study (Nummenmaa et al., 2012) that the ISC in the posterior middle temporal gyrus, an area that has been proposed to encode the intentions of an agent's actions, was positively associated with self-reported empathy, i.e., the capacity to understand other people's feelings. In other words, the higher the self-reported empathy scores were the more similar were the MTG time courses among those participants.

Methods Beyond ISC

Correlation is a relatively crude measure of the relationship between two random variables. Consequently, other more sophisticated measures of connectivity have been borrowed from the intra-brain analysis to the inter-brain analysis. For example, several fNIRS studies (Dommer et al., 2012; Holper et al., 2012) have used wavelet transform coherence (WTC)

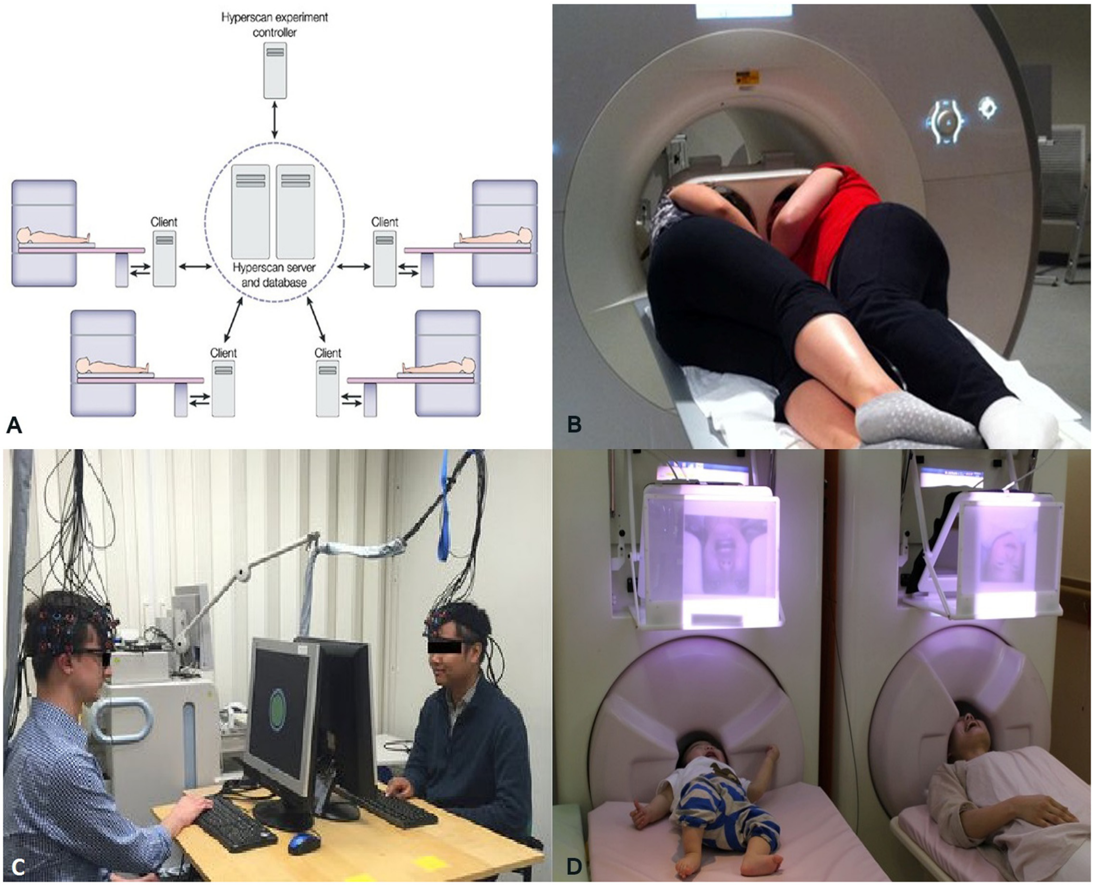

FIGURE 2 | (A) A schematic representation of simultaneous fMRI recording using multiple scanners. Adapted from "Moral cognition and its neural constituents," by Casebeer (2003). Copyright 2003 by Nature Publishing Group. Adapted with permission. **(B)** A specially designed dual-head coil for fMRI hyperscanning in a single scanner. Adapted from "Decoupled circular-polarized dual-head volume coil pair for studying two interacting human brains with dyadic fMRI," by Lee et al. (2012). Copyright 2011 by Wiley Periodicals, Inc., Adapted with permission. **(C)** Simultaneous recording of Functional Near-Infrared Spectroscopy (fNIRS) data from two individuals participating in computer-based cooperation tasks using a single recording device. Adapted from "Sex differences in neural and behavioral signatures of cooperation revealed by fNIRS hyperscanning," by Baker et al. (2016). Copyright 2016 by Macmillan Publishers Limited. Used under Creative Commons Attribution 4.0 International License. **(D)** Magnetoencephalography (MEG) hyperscanning of a mother and her infant. A mother and her infant look at each other's facial expressions while simultaneous MEG recordings are performed in two separate scanners. Adapted from "Hyperscanning MEG for understanding mother-child cerebral interactions," by Hirata et al. (2014). Copyright 2014 by Hirata et al. used under Creative Common Attribution ("CC BY") version 4.0 license.

for measuring between-brain connectivity in hyperscanning paradigms. WTC (Chang and Glover, 2010) is a method for analyzing the coherence between two time series as a function of both time and frequency. WTC is capable of detecting significant coherence between two time series even in the face of low common power. Similarly, researchers (Yun et al., 2012) have employed phase locking value (PLV), a measure of consistency in the relationship of the phases of two EEG signals with respect to time (Lachaux et al., 1999), as a marker of social interaction. In contrast to analyses based on frequency specific spectral changes, phase synchronization is said to be particularly suitable for quantifying long-range functional connectivity which may be critical for understanding neural mechanisms during interpersonal interactions. In many social situations, the directed asymmetric influence of one individual to another individual, rather than simple connectivity estimates, might be of particular interest and hence causality analysis may provide further useful information. Accordingly, both GC (Holper et al., 2012) and frequency domain variances of GC like partial directed coherence (Dumas et al., 2010; Astolfi et al., 2011) have been undertaken in inter-brain analyses. Additionally, as it is well established that several functional aspects of a complex system like the brain show strong non-linearities across levels of organizations, some of the tools of non-linear dynamics have been proposed to investigate the neural dynamics of inter-brain interactions (Tognoli and Kelso, 2009). In particular, transfer entropy, a model free information theoretic measure of effective connectivity, has been suggested (Liu and Pelowski, 2014) to be well-suited for this purpose. The graph theory approach has also been extended to describe the functional topology formed by multiple interacting brains (Toppi et al., 2015).

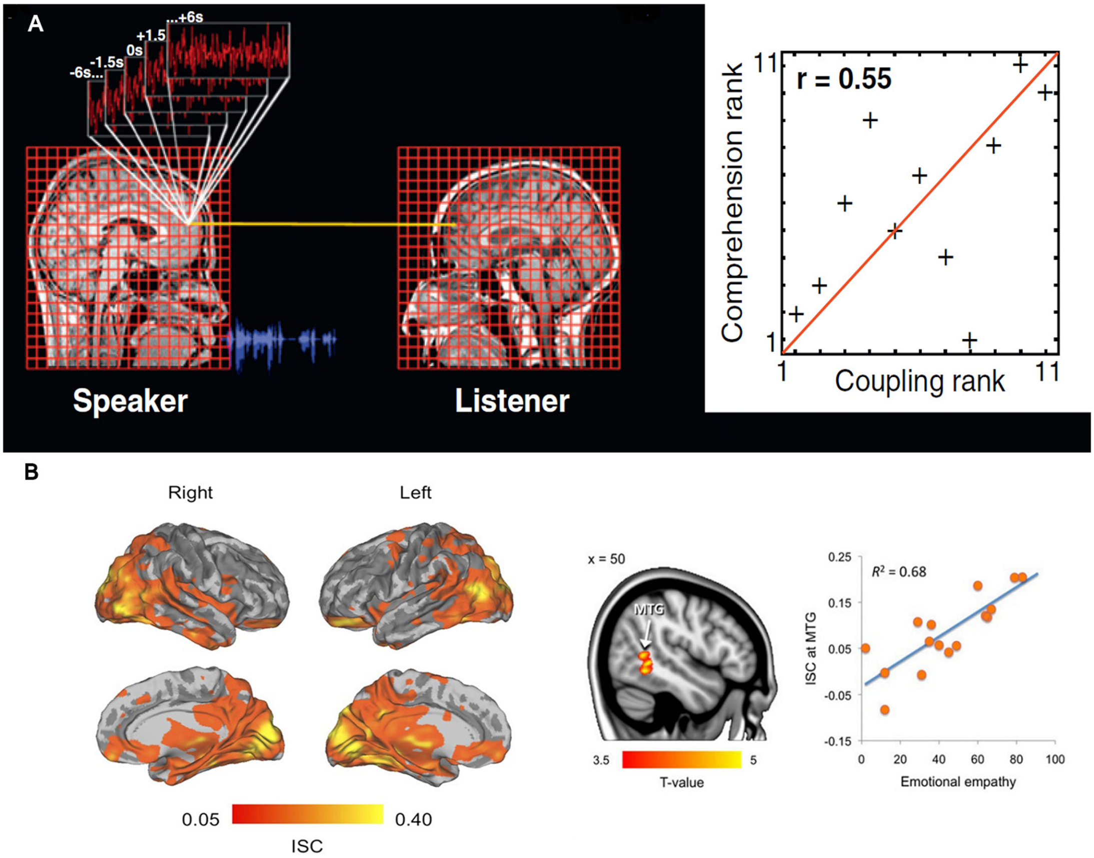

FIGURE 3 | (A) Inter-subject correlation (ISC) between hemodynamic responses of a speaker listener pair during storytelling. The neural coupling was assessed through the use of a general linear model in which the time series in the speaker's brain are used to predict the activity in the listeners' brains. The second part of the figure shows that, the greater the neural coupling between a speaker and listener the better the understanding. Adapted from "Speaker-listener neural coupling underlies successful communication," by Stephens et al. (2010). **(B)** Inter-subject correlation while watching movies depicting unpleasant, neutral, and pleasant emotions. Brain regions showing statistically significant [$P < 0.05$, false discovery rate (FDR) corrected] group-level ISCs during viewing of film clips. Tendency to catch others' emotions as indexed by Measure of Emotional Empathy in individual subjects was associated with ISC in the right middle temporal gyrus ($P < 0.05$, FDR corrected). ISC scores in the right panel are averages from an 8-mm sphere drawn around the peak voxel. Adapted from "Emotions promote social interaction by synchronizing brain activity across individuals," by Nummenmaa et al. (2012).

MULTI-BRAIN REFERENCE FRAME OF FUNCTIONAL NEUROIMAGING AND MENTAL HEALTH RESEARCH: SOME POTENTIAL APPLICATION AREAS

We propose that the development of inter-brain connectivity analyses may have important implications for mental health research. Recognizing other individual's affective, sensory, and attentional states provides the key to predicting their intentions and actions. Emotional intelligence (EI), sometimes described as the ability to identify and manage our own emotions and the emotions of others (Goleman, 2006), has become of widespread interest to psychological research in recent years (Hertel et al., 2009). Both EI and empathy have been reported to be associated with mental health status both at the individual (Hertel et al., 2009; Castro, 2011) and organizational (Slaski and Cartwright, 2003) levels. Measures of inter-brain neural processing can be used as objective markers for mutual empathy and interpersonal understanding between individuals. Though there are several commentaries on inter-brain connectivity analyses, their implications in mental health research are yet to be properly discussed. In what follows, we identify certain research areas in mental health discourse that include both the mental health conditions and other related issues like rehabilitations and therapeutic alliance where the interpersonal perspective is prominent and hence inter-brain connectivity analyses may find some applications. To the best of our knowledge, this is the first paper to offer a framework for the application of this novel approach in mental health research. Understandably, at present, most of the hypothetical applications proposed below lack any empirical validity. Still, they sub serve two main purposes here: they provide real life hypothetical examples for a better understanding of the concepts presented in this paper; and they also propose a roadmap for a meaningful future collaboration between mental health and neuroimaging researchers to explore this state-of-the-art methodology.

Mental Health Disorders

Depression

A rich body of literature exists discussing the interpersonal perspective in depression and other mood disorders. Several studies have identified, for example, basic behavioral differences between depressed and non-depressed individuals in interpersonal contexts (Hames et al., 2013). Depressed individuals have been found to be less likely to initiate social interactions (Libet and Lewinsohn, 1973), to speak more slowly with less volume and voice modulation (Youngren and Lewinsohn, 1980), engage in less eye contact (Kazdin et al., 1985), hold their head downward and engage in more self-touching (Ranelli and Miller, 1981), and use fewer gestures (Kazdin et al., 1985) compared to non-depressed individuals while communicating with others. Thus, hyperscanning depression patients and their friends/relatives during social interactions can be a useful tool to identify neural markers of impaired interpersonal communication in depression. Particularly, it can be tested as a tool to monitor progress in patients under interpersonal therapy. In addition, inter-brain connectivity analysis can also be helpful in the case of depression contagion. There is empirically validated evidence that shows close contacts, such as roommates of patients suffering from depression have higher chance to get affected, even after controlling for the shared negative life stress (Haeffel et al., 2014). Most interestingly, in the context of mental health, studies have empirically verified that caregiver's emotional empathy is positively correlated with their mental health outcomes (Lee et al., 2001; Ruiz-Robledillo et al., 2015). Inter-brain connectivity, as a neuroimaging marker of mutual empathy, has the potential to become a key tool in the research on depression contagion.

Autism Spectrum Disorders (ASD)

A core feature of ASD is impairment of reciprocal social interactions (Kanner, 1968). Therefore, it is not surprising that ASD is perhaps the only mental disorder where ISC analysis has already been applied. An fMRI study using ISC analysis revealed that activity in the right inferior frontal gyrus (IFG) is diminished in TD–ASD pairs compared to TD–TD pairs during joint attention task (Tanabe et al., 2012). Another fMRI study showed that during watching a movie portraying social interaction, ASD participants show diminished ISC in brain regions implicated in processing social information, including the insula, posterior and anterior cingulate cortex (PCC/ACC), caudate nucleus, precuneus lateral occipital cortex, and supramarginal gyrus (Salmi et al., 2013). Both studies employed only adult ASD patients. Nonetheless, inter-brain connectivity approach can also be particularly useful in child ASD subjects as several studies have stressed the need for early diagnosis of ASD. At present, ASD is usually detected at 3–6 years of age (Landa, 2008) and can be diagnosed as early as the age of two (Charman and Baird, 2002). Several existing studies have suggested that an detection and intervention notably improves prognosis (Nadel and Poss, 2007; Dasgupta et al., 2016). Inter-brain connectivity measures can be employed to explore social interaction between children and their caregivers for this purpose. Recent development of a hyperscanning system to examine real-time brain-to-brain interaction between a mother and her child (**Figure 2D**) may prove useful in this regard (Hirata et al., 2014).

Schizophrenia

According to a theory first proposed in 1992 (Frith, 1992), a wide range of symptoms in schizophrenia can be explained in terms of compromise in the theory of mind (ToM). For example, inability to attribute mental state to own behavior has been postulated to be behind delusion of alien control, command hallucination, and other "passivity" symptoms (Brüne, 2006). Similarly, an abnormal mentalizing of other person's thoughts and intentions has been suggested to be associated with symptoms like delusions of reference and persecution (for an alternative theory of impairment of ToM in schizophrenia, see Hardy-Bayle et al., 1994). For example, Abu-Akel and Bailey (2000) suggest that in some schizophrenic patients with positive symptoms, hyper-ToM with over-attribution of intentions to themselves and others may be the main psychopathology. Consequently, several studies have used different ToM tasks in schizophrenic subjects to explore these theories. Inter-brain connectivity measures can further add to this line of research by providing a neuroimaging marker of "theory of other mind" (ToOM) deficit in schizophrenia. Brain to brain connectivity can also play important part in research aiming at early diagnosis of schizophrenia as several studies have suggested that impaired interpersonal skill in childhood and adolescence is an early predictor of the development of schizophrenia in adult life (Dworkin et al., 1993; Stanghellini, 2004).

Personality Disorders

As stated by Sullivan (1953), personality is "the relative enduring pattern of recurrent interpersonal situations which characterize a human life." Naturally, interpersonal factors have been suggested to form the core of the personality psychopathology (Hopwood et al., 2013). In addition to the three personality disorders mentioned in the beginning, interpersonal perspective is crucial for conceptualizing many other personality disorders. Borderline personality disorder is characterized by a pervasive pattern of instability in interpersonal relationships (Leichsenring and Leibing, 2007). Histrionic personality disorder is marked by exaggerated attention-seeking behavior (Weiner and Craighead, 2010). Narcissistic personality disorder is characterized by pervasive grandiosity, an excessive need for admiration and a lack of empathy for others (Ritter et al., 2011). Persons with avoidant personality disorder persistently show social inhibition, feelings of inferiority, and extreme sensitivity to negative criticism or rejection by others (Weiner and Craighead, 2010). Thus, it will be interesting to investigate any differences in inter-brain connectivity patterns in the context of different personality disorders. Additionally, a study (Alden and Capreol, 1993) on avoidant personality disorder has found that treatment response is significantly influenced by differences in patterns of interpersonal behavior among patients. Thus, between-brain analysis can be tested as a tool for treatment choice in personality disorders.

Social Anxiety Disorder

Interpersonal context is prominent in social anxiety disorder. Positive qualities in friendship or romantic relationship are interpersonal factors that can protect against social anxiety whereas relational victimization or negative interactions can predict high social anxiety (La Greca and Harrison, 2005). More interestingly, studies have demonstrated that high socially anxious persons show elevated mentalizing and empathic abilities. There is some suggestion that a unique social-cognitive ability profile is behind their preoccupation with the other person's impression of them (Tibi-Elhanany and Shamay-Tsoory, 2011). Understandably, inter-brain connectivity analyses can be applied to further explore this hypothesis.

Somatic Symptom Disorder

Somatic symptom disorder (SSD) is characterized by excessive thoughts, feelings, and behaviors that focus on physical symptoms such as pain or fatigue. These symptoms may or may not be related to any physical cause (Kanaan et al., 2010; Rowe, 2010). Despite tremendous theoretical and research efforts, proper understanding of the psychopathology of this disorder remains elusive (Rofé and Rofé, 2013). There are some suggestions that interpersonal factors play a significant role in it. The somatizing patient often seeks the "sick" role, which provides them relief from stressful interpersonal expectations as a "primary gain," often accompanied by attention, caring, and sometimes even monetary reward ("secondary gain") (Moran et al., 2010). Consistent with this view, others have found desire for interpersonal closeness combined with the fear of being rejected is the most prominent internal representation of relationships in majority of patients with SSD (Landa et al., 2012). Accordingly, an interpersonal neuroimaging marker can be envisaged as a useful research tool for this mysterious disorder.

Eating Disorders

Eating disorders are among the list of conditions, besides depression, where the application of interpersonal therapy has been extended (Rieger et al., 2010). This is based on the recognition that interpersonal factors play the central role in the development and maintenance of eating disorder such as anorexia, bulimia, and binge eating disorder. The core behavioral patterns of eating disorders, i.e., distorted body image, preoccupation with control of body shape and weight, etc., are intimately associated with social settings in which these disorders take place, e.g., the "cult of thinness" and body shaming of contemporary popular culture (Hesse-Biber, 1996; Hesse-Biber et al., 2006). In parallel, studies have revealed significant interpersonal attributes, non-assertivity, submissive interpersonal styles, and social inhibition in patients with eating disorders (Hartmann et al., 2010). Thus, probing the neural coupling between the patient and persons in their immediate surroundings can be proved useful as a research tool in eating disorders.

Sexual Dysfunctions

In recent years, there is an increased research interest in various sexual dysfunctions including erectile dysfunction, arousal disorders, and sexual pain disorders. In spite of the moderate efficacy rates of the available treatment options, the discontinuation rates of treatments are equally impressive (Althof, 2002). Among the factors responsible for discontinuation, relationship issues occupy an important place. They are so intimately related to sexual functioning that in most cases it is impossible to determine which came first, a non-intimate relationship leading to sexual dysfunction or sexual dissatisfaction leading to mutual antipathy (McCabe et al., 2010). Despite this difficulty to ascribe causality, literature clearly suggests that addressing issues with interpersonal relationship is associated with better long-term outcome (Althof, 2002). This is also the premise of application of interpersonal therapy in sexual dysfunctions. Inter-brain connectivity analyses have multiple potential research applications here: from identifying the interpersonal problems behind sexual dysfunctions to shaping an outcome measure of treatment including monitoring interpersonal therapy.

Suicide

The presence of a mental disorder including mood disorder, schizophrenia, personality disorder, and substance abuse is an important risk factor for suicide. According to some estimate, diagnosable mental illness is present in 98% of those who commit suicide (Bertolote and Fleischmann, 2002). Assessment of suicidal risk in turn determines treatment choice and urgency of intervention in mental illnesses. Interpersonal theory of suicide (Joiner, 2007) has recently proposed an empirically validated theoretical framework that explains suicidal behavior and guide risk assessment and intervention in clinical settings. Thwarted belongingness and perceived burdensomeness are two interpersonal risk factors for suicide, as identified by this theory. Inter-brain connectivity analyses can be employed to explore the interpersonal factors between mentally ill patients and family members/friends to identify persons at risk of suicide and guide treatment strategy.

Other Issues Related to Mental Health

Therapeutic Alliance

Therapeutic alliance, broadly defined as collaborative bond between patient and therapist (Krupnick et al., 1996), is considered as "the quintessential integrative variable" (Wolfe and Goldfried, 1988) of psychotherapies as mounting evidence suggests that it is a better predictor of outcome across a wide range of psychotherapeutic modalities and many other sophisticated factors (Horvath and Symonds, 1991; Martin et al., 2000; Castonguay et al., 2006). Remarkably, a few studies have provided evidence supporting the significant impact of quality of therapeutic alliance on the clinical outcome of pharmacotherapy as well (Krupnick et al., 1996; Zilcha-Mano et al., 2015). These findings stress the need for early detection and repair of "rupture" of the therapeutic alliance. Here, inter-brain connectivity between the patient and the therapist can be evaluated as a key measure of deterioration in the relationship. An objective assessment of the alliance is particularly important keeping in mind the evidence showing a marked difference in the perception of rupture between the patient and the therapist (Safran et al., 2002).

Rehabilitation

The goal of psychiatric rehabilitation is to help persons suffering from severe mental illnesses to assimilate in the social life seamlessly with the least amount of professional support (Rössler, 2006). The role of rehabilitation in mental health has become particularly important after the de-institutionalization movement. Interpersonal aspects play an important role in rehabilitation and this provides scope for the brain-to-brain neuroimaging. It may be investigated as a tool to assess the interpersonal skill of the patient to guide the selection of rehabilitation strategy (Goldsmith and McFall, 1975; Halford and Hayes, 1991). Moreover, inter-brain connectivity between the patient and near ones can be explored as a marker of social support as a part of resource assessment (Buchanan, 1995).

COLLECTIVE MENTAL STATE AND MULTI-BODY REFERENCE FRAME OF FUNCTIONAL NEUROIMAGING: SOME THEORETICAL IMPLICATIONS

> The truth is the whole.
>
> —Hegel, 1807/1977

Throughout history theorists from disparate disciplines have engaged with the concept of groups as the bearer of mental states. In the modern era, Durkheim (1893) formulated his concept of "conscience collective" (translated as collective consciousness or collective conscience) as a set of shared beliefs, ideas, and moral attitudes that operate as a unifying force within the society. Durkheim contended that the concept of "*collective consciousness*" must be designated by a special term, simply because the states which constitute it differ specifically from those which constitute the individual consciousnesses. Based on the same concept, Durkheim methodically explored, perhaps for the first time, the social rather than individual causes of suicide including lack of social integration and solidarity, mismatch of the individual and collective moral values, excessive social control, and so on (Durkheim, 1897). Le Bon (1896) proposed that in a crowd the individual psychology is submerged in the collective mentality that completely transforms individual behavior. List of related early theoretical approaches that deal with the mental states beyond individual level includes "group psychology" by Freud (1922), "collective unconscious" by Jung (2014), "collective madness" by Borkenau (1981), etc. In spite of these early interests, the concept of collective mental state, until recently, has fallen into neglect for several reasons. One major obstacle has been the lack of direct objective measures of "collective consciousness." As one commentator notes, "the obvious methodological problem of how such an entity could be tested empirically has been such as to place it outside modern social science discussion, which is predominantly quantitative" (Bostock, 2002, p. 2).

Development of neuroscience more firmly established the correlation of mental states with the brain processes. As a group does not possess a brain and as no neurological correlates of group mental state could be envisaged, the usage of terms like group mental state or collective mental illness became a mere metaphor with no real resemblance with the "actual" mental states or mental illnesses "taking place within a brain." As a natural consequence, in scientific discourse, for a considerable period of time, the idea of collective consciousness had more metaphysical and transcendental connotation.

From what we have discussed so far in the present article, there are ample reasons to believe that inter-subject brain connectivity analysis (see "Glossary") can provide some novel insights into these debates. By shifting the functional neuroimaging to the multi-brain reference frame, it can furnish an objective measure of the collective mental state, thus making it more empirically accessible. The group mental state can be investigated recruiting the same analytical tools (e.g., correlation, coherence of neural dynamics) that are employed in individual brain. By doing so, inter-brain connectivity analysis creates some sort of neural correlates of group mental state, thus giving it a firm grounding in material reality. More generally, by providing support for a neural substrate of the collective mental state our proposal contributes to the endeavor to discard dualism in the context of mental health (Kelso and Engstrøm, 2006). The repudiation of dualism can only be seen as a part of the search for a more comprehensive approach to mental health. An objective neuromarker of collective mental state can never belittle the importance of a thorough subjective assessment of psychiatric patients, nor can it ever provide the exclusive information that is contained in the individual or ROI level neuroimaging. On the contrary, by putting the individual in the interindividual context and by providing a quantitative correlate of subjective experience, it can add new dimensions to both individual-centered and qualitative research programs. We fully agree with Lewontin et al. (1984, p. 282) when they commented that

> The biological and the social are neither separable, nor antithetical, nor alternatives, but complementary. All causes of behavior of organisms, in the temporal sense to which we should restrict the term cause, are simultaneously both social and biological, as they are available to analysis at many levels. All human phenomena are simultaneously social and biological, just they are simultaneously chemical and physical. Holistic and reductionist accounts of phenomena are not "causes" of those phenomena but merely "descriptions" of them at particular levels, in particular scientific languages. The language to be used at any time is contingent on the purpose of the description.

What we propose in the present article thus should be conceptualized as a novel addition to the rich array of "descriptions" of mental health phenomena. In the context of mental health, the insights that inter-brain connectivity analyses bring should be properly contextualized within the social, biological, and psychological determinants of mental health, as a complementary tool to other methods. Our ultimate goal should be to open up the discussion to a plurality of explanatory principles.

Glossary

Blood Oxygen Level Dependent (BOLD) Signal

An indirect measure of neuronal activity employed in fMRI studies that relies on regional differences in cerebral blood flow and oxy-hemoglobin level.

Correlation

A statistical measure that quantifies the strength of interaction between two random variables (e.g., neural signals from two different areas).

Coherence (Signal Processing)

Frequency domain description of correlation (above).

Functional Near-Infrared Spectroscopy (fNIRS)

A non-invasive, low-cost functional neuroimaging tool to monitor brain activation in the pre-frontal cortex by measuring changes in near-infrared light.

Functional Integration

The process by which segregated brain regions at various levels work together to process information and effect responses.

Granger Causality

A statistical measure of directionality between two random variables. According to GC, if a signal X "Granger-causes" a signal Y, then past values of X should contain information that helps predict Y above and beyond the information contained in past values of Y alone.

Graph Theory

A mathematical formalism concerned about how networks can be symbolically represented and accounted statistically.

Hyperscanning

A functional neuroimaging method for measuring brain activity simultaneously from two (or more) subjects. This can become a key tool in the context of studying group mental health and needs to be explored by future investigations. Please also refer to Montague et al. (2002) for details.

Inter-Subject Brain Connectivity

A key measure that can be defined by computing correlation, coherence, and other statistical properties between brain signals coming from two different brains. This is a fundamental marker for the multi-brain reference frame hypothesis and can become a clinical marker.

Localism (Neuroscience)

A philosophical approach to neuroscience that emphasizes the functional specificity of individual brain regions.

Magnetoencephalography (MEG)

A non-invasive neurophysiological technique for recording the magnetic fields generated by neuronal populations at macroscopic scale.

Partial Directed Coherence

A frequency domain description of GC between multivariate time series represented by autoregressive model.

Phase Locking Value

A measure of consistency in the relationship of the phases of two signals with respect to time.

Transfer Entropy

An information-theoretic measure of directionality between two random variables.

Wavelet Transform Coherence

Coherence as a function of both time and frequency.

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DRa and AB conceived the original idea. DRa wrote the manuscript with support from AB, DRo, PS, and BS. All authors provided critical feedback. AB and DRo helped supervise the project.

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Social Mechanisms to Get People Outdoors: Bimodal Distribution of Interest in Nature?

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We report results from a post-program survey ($n = 930$) of participants in a non-profit outdoor health program targeted principally at women with families in Australia's metropolitan cities. We analyze communications, motivations, experiences, satisfaction, and intentions. The program involves 3 months' outdoor training in scenic locations, culminating in a single-day event. Training includes social opportunities and peer-group support. Event entry is in teams and includes charitable fundraising and personal challenges. Drop-out rates are very low, and repeat sign-up high. There are 2,000–3,600 places per event, and the most recent sold out in <24 h. We propose that for urban residents of developed nations, individual interest in exposure to nature may be bimodal rather than unimodal. Programs of this type target individuals most likely to shift from low-interest to high-interest mode, using a set of social levers to change attitudes and behaviors. This contrasts with most public outdoor health programs, which assume a unimodal distribution and aim for small lifestyle changes at population scale. We suggest that the bimodal hypothesis is relevant to the sociocultural context of psychosocial interventions in a public health context, and merits direct testing.

Keywords: policy making, psychology, social, outdoors, exercise therapy, nature relatedness

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INTRODUCTION

For urban residents of wealthy temperate developed nations, who normally work indoors, outdoor exercise in unpolluted natural areas improves both physical health (1–7) and mental health (8–16). Mental health gains from exposure to nature include reduced stress, anxiety, and depression (17, 18); and improved sleep (19, 20) and cognition (21–23). These mental health improvements are non-specific: evidence indicates gains across all age groups (24–28), for both women and men (29); and across countries and cultures (27, 30, 31).

From the perspective of public health agencies and health insurers, nature provides free prevention and treatment for many health conditions: free, because other government agencies manage parks and greenspace, and individuals bear the financial costs of accessing them. Therefore, health agencies and insurers in many countries have repeatedly adopted programs intended to get people outdoors, including educational campaigns, publicly funded outdoor exercise programs, and so-called green prescription systems linked to health insurance and clinical medicine systems (32–34). Local governments also include and fund outdoor activities in their urban parks, and national parks agencies promote programs such as "Healthy Parks, Healthy People" (35) and

"Every Kid in a Park" (36). Such publicly run programs have been broadly positive, but both uptake and outcomes have been rather limited in scope and duration.

Here, we present an evaluation of an alternative approach run by a non-profit organization and suggest possible reasons for its outcomes to date. The evaluation is based on post-program questionnaires completed by participants, so it relies on their perceptions and recollections, rather than paired measurements prior and subsequent to participation. As with many such studies, however, it is these perceptions and recollections that influence subsequent participant attitudes and behaviors, including repeat enrollments, so this design is appropriate for the question addressed. The possible reasons for success include the broad hypothesis that individual interest in nature may be bimodal rather than unimodal. We put forward this hypothesis for rigorous testing in future.

MATERIALS AND METHODS

Program, Site, and Scale

The program evaluated here is known as Coastrek. It is a privately operated portfolio of annual walking events and prior training programs, held along public hiking trails around major metropolitan centers in Australia. The annual events are 30–60 km in length. The formal training programs are conducted over 3 months prior to each event. Marketing approaches are closely linked into a multi-tier network of small-scale health and fitness providers in each region. The overall approach combines multiple social measures that (a) target individuals likely to change their lifestyles to increase their exposure to nature and (b) encourage them to make this change.

The Sydney Coastrek started in 2009 with 800 individual participants, increased to 3,600 in 2016. The Melbourne Coastrek began in 2015 with 1,300 participants and increased to 2,400 in 2016. The Sunshine Coastrek will start in 2017 with 2,000 places. Overall, 90% of participants in the Coastrek program are females. Participants sign up 6 months in advance of the walk, and places are taken rapidly. For Melbourne in 2016, for example, online registration to the entire event sold out to previous Coastrek participants within 24 h, so the organizers had to negotiate additional places to provide for new participants.

Data and Analysis

We distributed an online post-event survey to all 3,600 participants in the 2016 Sydney Coastrek. The survey contained 20 multiple-choice questions developed by the authors, assessing the key factors associated with the event, namely communications, motivations, experiences, satisfaction, and intentions. Responses were in categorical or rating-scale (Likert-type) format, and the Likert-type questions contained five response categories. We received $N = 930$ completed responses, a response rate of 26%. Not all respondents answered all the questions, but overall there was a minimum per-question response rate of 87%. We analyzed this dataset using straightforward parametric statistics, to identify overall response patterns, and significant associations between the various components of the questionnaire.

RESULTS

Communications, Motivations, Satisfaction, and Intentions

In total, 69% of respondents ($n = 642$) reported that they had initially heard of the Coastrek program from friends and family members, i.e., *via* direct word of mouth. Repeat Coastrek participants comprised 15% ($n = 140$) of the total sample. The two key motivations for enrolling in the Coastrek program were taking part with friends (45%, $n = 419$) and the personal challenge of completing the event (41%; $n = 381$). Over 95% of respondents ($n = 884$) reported their overall experience in either the highest ("awesome") or second-highest ("good") Likert categories, with <1% of respondents scoring jointly in the two below-average categories. The event received high commendations (85 to 95% rated "good" or "very good") for organization, registration, atmosphere, support, facilities, and checkpoints, respectively; and 77% ($n = 716$) of participants indicated they planned to participate again the following year. Eight different communication channels were used to maintain contact with participants, including various email formats, websites, social media, phone assistance, and briefings in person. All of these communication channels received high commendations.

Training and Prior Participation

Across all respondents, 40% ($n = 372$) participated in the 12-week pre-event training programs, either through online instructions or through associated trek guiding companies in Sydney. A similar proportion (39%, $n = 363$) reported that they had no prior experience in any similar event. The proportion of participants taking advantage of Coastrek-sponsored training options was 49% ($n = 456$) for those who had no prior experience in similar events, and 38% ($n = 353$) for those who did have prior experience. This association is significant at $p = < 0.001$ (Fisher's Exact Test, $n = 567$). That is, prior experience was a statistically significant but relatively weak determining factor in whether or not participants used training programs offered or endorsed by Coastrek. The remaining 60% of Coastrek participants carried out any training independently. A similar proportion (61%) reported that they had previously participated in at least 1 of 10 broadly similar walks, runs, or similar events. However, less than 1% ($n = 9$) of respondents indicated they were competitive trail-runners. That is, runners do not consider Coastrek to be a race event.

Marketing

Marketing is targeted particularly at women of moderate fitness with limited time, including women with families. The Coastrek program is run by an Australian-based womens' fitness and adventure company, whose marketing tagline is that it "inspires women to transform their lives" (37). The training program and the event are marketed as enjoyable self-paced social occasions in scenic locations, with ample support, sharing, and opportunities for refreshment. The relevant text on the home page (37) states: "When you sign up for Coastrek, you embark on months of adventure – planning, preparation, training, fundraising, chatting, walking, shopping….” The event is run as a challenge, not a race. It raises funds for a well-regarded and non-political international charity, but fundraising is non-competitive. Entry is in teams of four, with at least two women per team; and with social support for training, group cohesion through branded clothing, and peer pressure against dropping out.

This combination of factors creates multiple encouragements and mechanisms to sign up; multiple social incentives to continue; multiple disincentives to drop out; and multiple individual rewards, including improved physical and mental health, social opportunities and social capital, altruistic “warmglow” factor, and enhanced self-esteem. A single enthusiast in each team can convert three other individuals from a negative or neutral attitude to nature exposure, to a positive attitude and behavior. The event home page (37) states: “Your energy and enthusiasm will radiate from you as you are motivated and inspired by your challenge.”

DISCUSSION

Past public policy measures, intended to increase individual exposure to nature, have implicitly assumed that the distribution of individual interest in nature-based outdoor activities across the population concerned is unimodal (**Figure 1**). They assume that population-scale education, encouragement, or incentives will lead everyone to increase exposure to nature by a small marginal amount, with a large aggregate net effect. These get-into-nature programs thus have the same underlying rationale as successful public health initiatives such as infant vaccinations and fluoridation of drinking water.

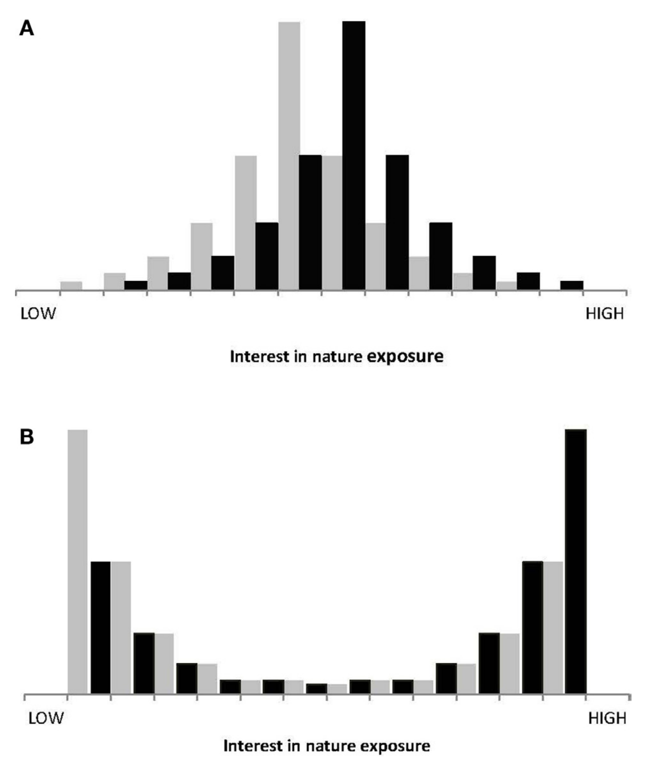

FIGURE 1 | Conceptual distributions of individual in nature exposure. Gray: prior to public health intervention. Black: after intervention. **(A)** Current unimodal view. **(B)** Alternative bimodal view. All bar heights are conceptual only.

It is possible, however, that in fact the population-scale distribution of individual interest in nature may be highly bimodal, with some individuals heavily addicted to nature-based outdoor activities, and others indifferent or indeed repelled by them (**Figure 1**). This hypothesis has apparently not been advanced previously, and has not yet been tested, either by this study or independently.

If the bimodal hypothesis proves correct, then policy initiatives would perhaps prove more successful if they focused on moving individuals from the negative to the positive mode: a large change in attitude for a small proportion of the population, rather than a small change for a large proportion. This approach would require first, identifying and targeting specific individuals most likely to make that move; and second, identifying and applying the most effective and cost-efficient social levers to induce them to actually do so. That is a very different policy design from those adopted historically and indeed currently.

In line with this hypothesis, we suggest that one possible reason for Coastrek’s success to date may be that all of its components operate jointly to move individuals from a negative to a positive mode of attitude to nature exposure. Most Coastrek participants are busy urban women with families, who allocate little time to outdoor activities. The Coastrek program provides them with incentives, personal rewards, peer support, and social justification to include outdoor activities in nature as part of their regular schedule, displacing a part of their previously higher priorities.

None of the individual marketing approaches is new, but the combination appears to be especially effective. The 3-month preparation period creates sustained behavioral change, with nature-based adventures becoming part of participants’ regular lifestyles. For many participants, this change appears to be multi-year in length, as shown both by stated intentions, and by the 100% take-up of the 2,000 initial places in the 2016 Melbourne Coastrek, by previous Coastrek participants.

CONCLUSION

From a theoretical perspective, the success of Coastrek lends at least preliminary support to our hypothesis that the population-scale distribution of interest in nature exposure may be bimodal rather than unimodal. It would now be valuable to test this hypothesis directly. This would require construction, validation, and trialing of an attitudinal and stated-behavior scale related specifically to nature-based outdoor activities; and application of this scale across large-scale random population samples, together with standard socioeconomic and demographic parameters, individual history of outdoor activities, information on outdoor activities by family, close friends, and colleagues; and data on outdoor nature-based opportunities nearby.

From a practical public health perspective, the popularity of the Coastrek model among its participants, and its ability to change their lifestyles to a more active outdoor mode, indicate the value of expanding and emulating elsewhere. In particular, its focus on adult women of moderate fitness, and particularly those women with children, enhances its public health outcomes by creating changes in diet and activity schedules for their entire families. We suggest that Coastrek provides a model that can be scaled up, expanded internationally, extended into different outdoor activities, and adopted broadly in public health policy.

AUTHOR CONTRIBUTIONS

RB conducted analyses and is the principal author. DW provided access to participants for data collection and assisted in interpretation. PB provided literature and expertise and contributed to writing.

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