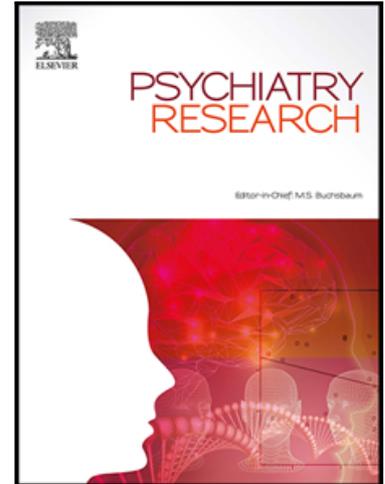


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A 12-month study of the hikikomori syndrome of social withdrawal:
clinical characterization and different subtypes proposal

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Highlights

- Hikikomori syndrome of social withdrawal is comorbid with several psychiatric disorders in our society.
- There are subgroups based on comorbidity, with differences in basal characteristics and evolution.
- The anxiety-affective subgroup presents more favorable baseline characteristics, but worse evolution at 12 months.
- These subjects present fragility and tendency to relapse and have disengagement, which is particularly relevant in the anxiety-affective subgroup.
- Long-term intensive home or hospital treatments are more effective than short outpatient treatments.

A 12-month study of the hikikomori syndrome of social withdrawal: clinical characterization and different subtypes proposal.

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Abstract

Social withdrawal is a new mental health problem increasingly common, present in different cultures, whose psychopathology and treatment is not yet established. This study aims to determine the socio-demographic and clinical features and possible clinical subtypes that predict the 12-month outcomes of cases with hikikomori syndrome, a severe form of social withdrawal. Socio-demographic and clinical data at baseline were analysed as well as data obtained for 12 months after at-home treatment in 190 cases. The inclusion criteria were: spending all time at home, avoiding social situations and relationships, significant deterioration due to social isolation, with a minimum duration of 6 months. Six major diagnostic groups were identified: affective, anxiety, psychotic, drug use, personality and other Axis I disorders. The anxiety-affective subgroup demonstrated lower clinical severity, but worse evolution. Less than half of the cases were available for medical follow-up at 12-months. Subjects undergoing intensive treatment had a higher medical follow-up rate and better social networks at 12-months. Therefore, our findings provide data to reach consensus on the specific characteristics of social isolation hikikomori syndrome. The analysis demonstrated the fragility and tendency to relapse and have disengagement, particularly relevant in the anxiety-affective subgroup, suggesting that intensive treatments are more effective.

Keywords: social isolation; comorbidity; long-term treatment; home treatment; differential diagnosis.

1. Introduction

Social isolation is a health problem that has become increasingly important in recent years, requiring studies that investigate its existence and psychopathology in different cultures. "Hikikomori syndrome", initially described in Japan by Tamaki Saito (Saito, 1998), is a type of isolation with specific characteristics. Cases are described as "those who are completely isolated from society and are in their home for more than six months, beginning in the second half of the twentieth, and in which other psychiatric disorders do not better explain the main causes of this condition".

In East Asian countries, hikikomori syndrome has been described as having a 1-2% lifetime prevalence (Koyama et al., 2010; Teo and Gaw, 2010; Wong et al., 2014), and it has been declared a public health problem in Japan. Thus, the Japanese government published guidelines with diagnostic criteria that excluded schizophrenia or other known psychopathology (Ministry of Health, 2003; Saito, 2010). Nonetheless, subsequent investigations have included associated psychiatric pathologies, such as anxiety, depressive, internet addiction, autism spectrum and other disorders (Campbell et al., 2006; Kato et al., 2011a, 2011b; Kondo et al., 2013; Koyama et al., 2010; Lee et al., 2013; Li and Wong, 2015; Malagón-Amor et al., 2015; Nagata et al., 2013; Stip et al., 2016; Suwa and Hara, 2003; Tateno et al., 2012; Teo, 2013; Teo et al., 2015a, 2015b, 2013a). It has also been suggested that patients with prodromal stages of psychotic disorders may be included (prior to a definitive diagnosis of psychosis) based on their similar characteristics (Kondo et al., 2013; Malagón-Amor et al., 2015; Stip et al., 2016). Some authors have divided hikikomori into two categories: primary and secondary (Kato et al., 2012; Suwa et al., 2003). Primary hikikomori is not associated with any psychiatric disorders, while secondary hikikomori is caused by a currently recognized psychiatric disorder. However, there is no consensus on which psychiatric disorder should be considered to exclude a diagnosis of primary hikikomori. Therefore, it is evident that there is no consensus on which comorbid psychiatric disorders should be included. In addition, given the existence of cases that do not meet the criteria of other existing psychiatric disorders, it has been suggested that severe social isolation might be considered as a new psychiatric disorder (Teo and Gaw, 2010). In an effort to standardize the diagnosis of severe social isolation-hikikomori, the following diagnostic criteria have been suggested (Teo et al., 2015b; Teo and Gaw, 2010): 1) spending most of the day and most days at home, 2) avoiding social situations, such as attending school or going to work, 3) avoiding social relationships, such as friendships or contacts with family members, 4) experiencing discomfort or significant deterioration due to social isolation, and 5) having a minimum duration of 6 months.

The description of its existence in other countries and cultures (De Michele et al., 2013; Furuhashi et al., 2012; García-Campayo et al., 2007; Gariup et al., 2008; Gondim et al., 2017; Kondo et al., 2013; Lee et al., 2013; Malagón-Amor et al., 2015; Ovejero et al., 2014; Sakamoto et al., 2005; Teo, 2013; Wong et al., 2017a) suggests that it is not only a disorder related to Japanese culture. Our group described the existence of hikikomori in Spain, where primary hikikomori less common than secondary hikikomori. It was concluded that it may not be a new diagnosis but rather a severe syndrome associated with multiple mental illnesses (Malagón-Amor et al., 2015).

Social exclusion is frequent in severe mental illness patients (Koenders et al., 2017; Linz and Sturm, 2013), and it has been described that intensive case management improves social functioning (Dieterich et al., 2017), emphasizing the need for treatments and services designed to develop social skills, as well as to create opportunities for social connection and community involvement. There has been little research investigating treatment approaches focused solely on social isolation syndrome. Psychotherapy (Teo et al., 2015a), including individual, group, and exercise-oriented psychotherapy, have been proposed as treatment options (Nishida et al., 2016). Previous studies have found that despite a long evolution of isolation, subjects presented a preference for treatment delivered in person, as opposed to telepsychiatry (Teo et al., 2015a). Pharmacological treatment would be based on the associated comorbid psychiatric pathology. These studies highlight the lack of awareness of the disorder and the subject's motivation for treatment (Kondo et al., 2013; Malagón-Amor et al., 2015), highlighting the need for specialized home care teams that have access to these subjects (Kondo et al., 2008; Lee et al., 2013; Malagón-Amor et al., 2015). Regarding the long-term treatment and prognosis of hikikomori, there is little information. Kondo (Kondo et al., 2013) have found that only 15.3% of subjects achieved social participation after 21.7 months of consultation. Tateno (Tateno et al., 2012) have suggested the division of

hikikomori into three groups that are based on comorbid pathology and therapeutic options that are based on a survey-based study of psychiatrists from different countries.

Despite the recent development of clearer diagnostic criteria, there is still considerable heterogeneity among subjects with hikikomori, and there are important knowledge gaps in the prevalence, comorbidity, and psychopathology, as well as their treatment options.

The hypothesis of our study is that social isolation is formed by a heterogeneous group of subjects with different comorbid pathologies, and depending on the comorbid pathology, there are different clinical characteristics and responses to treatment. Accordingly, this study aimed to determine the possible different subtypes of social isolation and their possible associations with the diagnostic entities already described, and finally, to analyse the evolution of the cases during 12 months after an assertive home care treatment approach.

2. Methods

2.1. Participants

A total of 190 people who were treated at home because of social withdrawal by the Crisis Resolution Home Treatment (CRHT) from 2008 to 2014 in Barcelona (Spain). The definition and diagnostic inclusion criteria for the social isolated cases were (Tateno et al., 2012; Teo et al., 2015a, 2015b; Teo and Gaw, 2010) 1) spending most of the day and most days at home, 2) avoiding social situations, such as attending school or going to work, 3) avoiding social relationships, such as friendships or contacts with family members, 4) experiencing discomfort or significant deterioration due to social isolation, and 5) having a minimum duration of 6 months. These symptoms should be the primary and predominant over any other symptoms, in case there were.

2.2. Home visitation program

The CRHT team was formed by two psychiatrists and two nurses. The target population were patients with severe mental disorders dissociated from outpatient monitoring. People with no psychiatric history who presented behavioural disorders suggestive of mental disorder were also evaluated. Cases were referred to the CRHT by primary and psychiatry outpatient teams, psychiatric emergency services and/or social workers. CRHT coordinated with these resources to assess and treat these subjects at home, and, once stabilized, linked them back to the regular psychiatric services. Since its beginning, the demand for intervention in cases of social isolation was remarkable, specializing on its assessment and treatment as a result of it. The inclusion criteria were evaluated based on the information provided by the family and/or the subjects, depending on the collaboration. In turn, they were evaluated and confirmed at the first home visit. The exclusion criteria were subjects with diagnosed cognitive disorders, such as dementia, drug dependence without other comorbid psychiatric disorders, age younger than 12 years old and subjects for whom the only treatment option was involuntary inpatient therapy. For more information on the operation of the equipment, review the previous article (Malagón-Amor et al., 2015). After home treatment was initiated and initial improvement was achieved, the CRHT home treatment was completed, and the cases were delivered to the most appropriate device in an individualized manner: outpatient psychiatric or medical centre, hospitalization or others. If stabilization was not reached, and there were criteria for involuntary treatment (suicide risk, aggression, risk of escape, altered reality judgment), an involuntary transfer was made at the referral hospital for hospitalization. For those subjects who retained the reality judgment, were aware of the seriousness of the situation, but they did not agree to carry out the treatment and receive help, it was not possible to carry out the treatment. Following this derivation, the clinical condition and situation of isolation was evaluated by contacting the subjects' professional references 4, 8 and 12 months after derivation.

2.3. Measurement instruments

In the first assessment at home, all cases were prospectively studied according to a routine computerized protocol that included demographic and clinical information by a trained clinical team. Socio-demographic data included age, gender and social network. The clinical characteristics included a personal psychiatric history, including any history of suicide, aggressiveness, and internet addiction. The socially withdrawn period and the age at onset of hikikomori were also recorded. The patient diagnoses were evaluated using the DSM-IV-TR criteria, grouping the major mental disorders into six categories: psychotic, affective, anxiety, drug abuse, personality and other Axis I diagnoses. The service referred by the CRHT was recorded. Illness severity was assessed using the Spanish version of the Severity of Psychiatric Illness (SPI) scale (Bulbena et al., 1997; Lyons et al., 1997). The Global Assessment of Functioning (GAF) and World Health Organization Disability Assessment (WHO/DAS) and Scale of Unawareness of Mental Disorder (SUMD) scores were also registered. Social network was evaluated according to criteria developed by our team based on clinical experience: 1) null relationship, 2) relationship with family with whom we live, 3) relationship with a friend outside the home, and 4) normalized social relation. For the analysis, the cases were divided into two groups: null social network vs. some relationship with family or people outside the home.

In the surveillance of the linkage at the 4, 8 and 12 months, an evaluation of the subject's connection to the mental health network was performed by contacting to the responsible medical service. The clinical status was evaluated using the GAF and WHO/DAS scales, as well as the persistence of social isolation and its severity. We recorded whether there was a change in the diagnostic orientation performed by the CRHT during the 12-month linkage, as well as the new diagnosis, if any.

2.4. Statistical analysis

First, a descriptive analysis of the subjects at baseline was performed. Second, univariate analysis between the baseline determinations and outcome measures of functioning were performed using Pearson's correlation coefficient. The outcome variables were social isolation persistence, improvement in social isolation condition, social network, responsible medical services and GAF and WHODAS scores. Third, different regression models were performed using outcome variables as the dependent variables, and clinical and socio-demographical variables were used as the independent variables. The variables with significant correlation with outcome measures in the univariate analysis were selected. In addition, diagnostics were grouped into affective-anxiety group vs others due to the baseline similarities within these groups (see Table 1). The evolution across time of measured outcomes were analysed using generalized estimating equations with an exchangeable working correlation structure. The effect of presence/absence of anxiety-affective disorder across time was assessed introducing an interaction term (time by visit) (see Figure 2). Analyses were conducted using Stata (version 14) and SPSS 24 (SPSS, Chicago, IL, USA).

2.5. Ethics statement

The study was approved by the Parc de Salut Mar Barcelona Clinical Research Ethical Committee, in accordance with the Code of Ethics of the World Medical Association (Declaration of Helsinki).

3. Results

The global socio-demographic and clinical characteristics are shown in Table 1 (first column). The 12-months follow-up results are shown in Table 2.

Regarding the social network, 57.4% of the subjects maintained relationships with their families, while 34.2% had no relationship with anyone, even with family members inside the home. No statistically significant differences were found in the 12-month follow-up for any of the outcome variables.

The influence of isolation time on severity and evolution was also evaluated without obtaining statistically significant data, and the results demonstrated that the time of isolation has not a determinant role in severity or evolution.

3.1. Comorbid pathology

Only one case had no associated pathology. In that case, internet addiction behaviour was detected. Six major diagnostic groups were identified: affective ($n = 28$), anxiety ($n = 35$), psychotic ($n = 75$), drug use ($n = 8$), personality (total $n = 33$, cluster A $n = 12$, cluster B $n = 19$, and cluster C $n = 2$) and other diagnoses ($n = 10$), including hoarding disorder ($n = 1$), autism spectrum disorder ($n = 2$) and body dysmorphic disorder ($n = 1$).

Patients in the anxiety and affective disorders diagnostic groups had similar a gender proportion (affective group male 57.1 % vs anxiety group 60 %) and outcome variables (affective group vs anxiety group: mean WHO/DAS Total 11.8 (SD 3.7) vs 11.4 (3.4); mean GAF 42.8 (SD 13.9) vs 42.6 (12.9), mean SPI total 10.3 (5.6) vs 10.4 (4.1), bonding at 12 months 37 % vs 44.1 %, improvement in social isolation state at 12 months 26.9 % vs 35.5 %), but the mean age differed (affective group 53 years vs anxiety group 35 years, $p < 0.001$). Accordingly, we decided to perform an analysis in which one group would include patients with comorbid pathologies of anxiety or affective disorders, compared to the other large group of cases with social isolation (Table 1).

3.2. Twelve-month follow-up analysis

Table 2 shows the comparative analysis of the 12-month evolution in the anxiety or affective subgroup vs other cases once discharged from CRHT and referred to outpatient or inpatient centres. Figure 1 shows the total confirmed follow-up in medical resources, once referred from CRHT team, during 12 months, of cases with social isolation. Lost cases are not included. The results of the regression models examining the influence of the diagnostic groups across time are shown in Table 3 and Figure 2.

Significant variables between the affective-anxiety group and other groups (age, gender, initial social network, history of aggressiveness, number of CRHT visits, referral device-whether or not admitted- GAF, total WHODAS, total SPI, SPI 10, SPI 8 and SPI 12) were included in the logistic regression analysis of the predictors of isolation at 12 months, with statistically significant results for the variables age ($B = 0.024$, $p = 0.098$), derivation device and cases with and without hospitalization ($B = 1.766$, $p < 0.001$) and SPI12 ($B = 0.629$, $p = 0.028$).

3.3. Collaboration with treatment

The classification of the cases based on whether they were collaborators vs non-collaborators, according to the score of the SPI sub-item 10 (opposition to treatment), showed statistically significant differences in variables such as the initial social network ($p = 0.001$) and at 8 months ($p = 0.041$), as well as the baseline referral medical service ($p = 0.009$), at 4 months ($p = 0.009$) and 8 months ($p = 0.058$), with a higher percentage of involuntary hospitalization as opposed to treatment ($n = 28$ [58.3 %] vs $n = 8$ [16.7 %], $p < 0.001$). Statistically significant differences were found, with a direct relationship between SPI 10 and total SPI ($B = 2.526$, $p < 0.001$), SUMD ($B = 0.167$, $p < 0.001$), baseline total WHODAS ($B = 1.225$, $p < 0.001$), at 4 months ($B = 0.683$, $p = 0.005$) and 8 months ($B = 0.742$, $p = 0.011$), and inverse between SPI 10 and baseline GAF ($B = -2.778$, $p = 0.001$) and GAF at 4 months ($B = -2.208$, $p = 0.014$).

3.4. Type of treatment after CRHT

Cases who required hospital admission after home treatment by CRHT compared to those who were referred for outpatient follow-up had lower isolation behaviour persistence at 12 months ($n = 21$ [61.8%] vs $n = 74$ [84.1%], $p = 0.008$), 4 months ($n = 24$ [68.6%] vs $n = 89$ [93.7%], $p < 0.001$) and at 8 months ($n = 22$ [66.7%] vs $n = 80$ [87.9%], $p = 0.006$), better psychiatric follow-up at 12 months ($n = 28$ [73.7%] vs $n = 42$ [40%], $p < 0.001$), 4 months ($n = 37$ [90.2%] vs $n = 64$ [58.7%], $p < 0.001$) and 8 months ($n = 29$ [72.5%] vs $n = 53$ [49.5%], $p = 0.013$), and better social networks at 12 months (better $n = 20$ [58.8%] vs $n = 30$ [33.3%], $p =$

0.010), 4 months (better $n = 27$ [77.1%] vs $n = 33$ [35.9%], $p < 0.001$), and 8 months (better $n = 20$ [58.8%] vs $n = 30$ [33.3%], $p = 0.008$). Overall, they had a lower relapse rate of social isolation ($n = 6$ [15.5%] vs $n = 13$ [48.1%], $p = 0.005$). In terms of baseline characteristics, they had a worse social network (poor social network $n = 22$ [23.2%] vs $n = 73$ [76.8%], $p = 0.020$), without any gender differences (men $n = 35$ [81.4%] vs $n = 82$ [71.9%], $p = 0.225$) or age (mean 39.8 vs 39.2, $p = 0.849$). There were differences in the comorbid diagnoses, with a higher proportion of hospitalization in patients with psychotic disorders ($n = 34$ [79.1%] vs $n = 27$ [23.7%], $p < 0.001$).

4. Discussion

To our knowledge, this study is the first to analyse a large sample social isolation cases in our society in terms of the global characteristics and the evolution and response to long-term treatment, thereby providing more scientific evidence about the phenomenon in Western culture. The results of this study confirm that the phenomenon exists in cultures other than in Japan (Aguglia et al., 2010; De Michele et al., 2013; Furuhashi et al., 2012; García-Campayo et al., 2007; Gariup et al., 2008; Gondim et al., 2017; Kato et al., 2012; Kondo et al., 2013; Lee et al., 2013; Malagón-Amor et al., 2015; Ovejero et al., 2014; Sakamoto et al., 2005; Teo, 2013; Wong et al., 2017a). Only one case corresponded to primary hikikomori (Koyama et al., 2010; Teo and Gaw, 2010), although the subject did demonstrate behaviour compatible with internet addiction (Teo et al., 2015b) confirming our hypothesis that isolation behaviour is not a new diagnostic category but a serious syndrome common to different mental disorders (Malagón-Amor et al., 2015). Regarding comorbid pathologies, which were diverse, our theoretical proposal of differentiation between the group of cases with affective or anxiety pathology and the remaining cases shows that the affective-anxiety subgroup presents a lower clinical severity and disability at basal level, higher disorder awareness and greater collaboration with the treatment, being possible to be linked voluntarily to outpatient treatment centres. These baseline characteristics more favourable initially contrast with a worse evolution during the 12-month follow-up, with lower bonding and higher percentages of persistence of isolation. Regarding the disease evolution, it should be noted that when the subjects underwent less intensive treatment (outpatient treatment), there was a greater disconnection and abandonment of treatment at 12 months, with worsening of the isolation. When the treatment referral was more intensive care (e.g., hospitalization), then the improvement was greater and more stable over time. These data indicate that the best type of treatment is more effective intensive treatments at home or hospital, initially focusing on creating a therapeutic link, improving the awareness of disorder and collaboration, until complete rehabilitation of the person is achieved.

4.1. Socio-demographic and clinical characteristics

As described in previous studies, social isolation was more frequent in men (Kondo et al., 2013; Koyama et al., 2010; Li and Wong, 2015; Wong et al., 2014). This fact has been related to social factors typical of Japanese culture, in which young Asian men have a strong desire for success, together with a greater need to keep up appearances when unsuccessful. Thus, they may be more sensitive to failure and are more likely to decide to withdraw from society if they feel that they have failed (Uchida, 2010; Uchida and Norasakkunkit, 2015). This sense of failure in men could also occur, though likely less intensely, in our society. This fact would not explain the phenomenon in its entirety but no other influencing factors have been described.

While hikikomori Japanese research has focused on young people and adolescents (Chan and Lo, 2014, 2013; Hattori, 2006; Kato et al., 2016; Kondo, 2001; Kondo et al., 2008, 2007; Li and Wong, 2015; Teo et al., 2015b; Yong and Kaneko, 2016) the wide sample analysed in our study, without any exclusions by age, shows that the existence of social isolation is not limited to a young population (Malagón-Amor et al., 2015). As described (Koyama et al., 2010), cases with onset in middle age might be related to particular social situations such as the loss of one's job, suggesting a different psychopathology. In addition, some had first experienced it in their teens or twenties, suggesting that the social isolation is a problem that has not been recognized at the beginning.

Regarding the social network, it was predominantly null or restricted only to the people with whom the subjects coexisted. The subjects with the worst social networks at the first evaluation were younger, highlighting the need for early detection and treatment in young patients. Furthermore, given the discrepancy between studies of type of relationships these subjects can maintain (Li and Wong, 2015), these data provide more evidence to reach a consensus that unifies diagnostic and inclusion criteria. Also, mobile health research methodology of extracting behaviorally meaningful features from sensor data may be useful to provide objective information to the professional clinical judgment and early detection on withdrawal behavior (Li et al. 2017a).

The isolation time was highly variable, without a certain influence on the development or severity of the disease, which is surprising and suggests that despite the suitability of early detection, long-term cases may also have good prognoses.

4.2. Comorbid pathology

Only one case presented no associated mental disorder, corresponding to primary hikikomori (Koyama et al., 2010; Teo and Gaw, 2010). This person presented with behaviour compatible with internet addiction (Stip et al., 2016), which is not included as a diagnostic category in DSM-5; however, there are increasing proposals that it should be included (Stip et al., 2016; Tao et al., 2010). Secondary hikikomori is predominant in our society, supporting the hypothesis that the phenomenon of social isolation is a severe syndrome common to different mental disorders and not a new diagnostic category (Malagón-Amor et al., 2015). It is likely that the different mental health practices in different countries and cultures can also influence these differences (Kato et al., 2011a), which may indicate different diagnostic and therapeutic plans for similar isolation cases.

The comorbid pathologies were diverse, with psychotic, personality, affective and anxiety disorders being predominant. Other disorders, such as autism spectrum disorder, occurred infrequently. In this sense, interest in the study of social isolation has led in recent research to determine diagnostic criteria and describe the associated comorbid pathology (Teo et al., 2015b), relating it to several very heterogeneous psychiatric disorders, without a final conclusion. The relationship between depressive disorders (Teo, 2013; Teo et al., 2013a), such as Modern Type Depression (related to Japanese culture) (Kato et al., 2016) and hikikomori has questioned. In addition, anxious disorders (Nagata et al., 2013; Teo et al., 2013b) have been linked to social isolation. Our overall analysis of the diagnostic groups showed some significant differences between the diagnostic groups, socio-demographic and clinical variables, as well as the 12-month disease evolution. Based on previous studies, as well as this global analysis, we made a theoretical proposal differentiating between the group of patients with affective or anxiety pathology and the other patients, with significant between-group differences. Thus, the anxiety-affective subgroup had a greater proportion of women, with lower clinical severity and disability at baseline, greater awareness of the disorder and greater collaboration with the treatment, making it possible to treat them voluntarily on an outpatient basis. Initially, these baseline characteristics contrasted more favourably with a worse disease evolution during the 12 months, with lower bonding, higher percentages of persistence of isolation and worsening of clinical condition. These data lead us to consider whether the unique and differentiated characteristics of the anxiety-affective subgroup would be specifically related to hikikomori social isolation syndrome. Therefore, could we exclude subjects with comorbid pathologies, such as substance use disorder, personality disorder or psychosis, from the studies of social isolation-hikikomori? Alternatively, perhaps we should evaluate the different subtypes of isolation to offer different types of treatment based on disease comorbidity.

4.3. Evolution during the twelve months: importance of the type of treatment and collaboration

To the best of our knowledge, this study is the first 12-month follow-up study of subjects with social isolation. After the home treatment by the CRHT has been completed, and the cases are referred to the outpatient or hospital centre, there is an overall tendency for cases to disconnect over the following months, to persist with their isolation behaviour and to even worsen in the isolation state. Of the cases, only 14% ended with a normalized social network. Although disconnection was a global phenomenon, it was more pronounced

in the anxiety-affective subgroup, which is surprising given its initial greater collaboration with treatment. We consider that differences in the maintenance of the psychiatric follow-up and worsening of the recovery state in this subgroup are partly related to the fact that this subgroup was more frequently referred to outpatient devices. The results suggest that subjects treated with more intensive treatment devices, such as hospitalization, achieved more rapid and stable improvement over a year, with better social networks and maintenance of the psychiatric follow-up during the 12 months. This result occurred even when the initial clinical characteristics were more unfavourable, such as poor social network and psychotic diagnosis. This fact raises the question of which treatment types are most appropriate to treat social isolation and highlights the failure of existing specialized outpatient devices used to treat these types of social isolated cases. Thus, some authors have indicated the need for early intervention (Hattori, 2006), both voluntary and involuntary hospitalization (Kato et al., 2012), with pharmacotherapy (Nagata et al., 2013), antidepressants (Teo, 2010), animal-assisted therapy (Wong et al., 2017b) or even traditional Chinese medicine (Kato et al., 2012). Others have emphasized the need to create a bond and trust with the patient (Krieg and Dickie, 2013) through which treatment can be initiated and to understand their psychosocial situations and possible attachment traumas (Hattori, 2006; Ogino, 2004; Wong, 2009). There have been described approaches to engage withdrawn young people, based on three identified reasons for changing their withdrawal behavior: rebalancing one's ideal self with reality, reconnecting with tuned-in people, and regaining momentum in life (Li et al., 2017b). In this regard, it should be noted that one of the most important problems for the management and treatment of social isolation cases is the access to these subjects and their limited collaboration (Lee et al., 2013). Cases with the worst collaboration had the worst social networks, greater severity and disability, and more frequently required hospitalization. The intensive home-based approach and the creation of a therapeutic relationship facilitates the awareness of the disorder and the need for treatment, improving the social isolation behaviour. These data make it possible to determine the type of treatment that is most appropriate for people with social isolation, and we believe that the basis for treating these subjects is their initial home access, link building and initial intensive approach, either at home or in the hospital, with prolonged treatment times focusing on social skills rehabilitation and social reintegration, until complete rehabilitation of the person is achieved. We consider that short-term treatments represent failure for this type of subject.

This study has several strengths and limitations that must be considered. Its main strength is the fact that it is the first study in Western culture to investigate a wide sample of isolation cases and to analyse its global characteristics, long-term disease evolution and response to treatment. However, the first and primary limitation is the methodology; this study is an observational descriptive study, for which it is not possible to establish causal relations between variables or to realize a true calculation of incidence. Another important limitation is the heterogeneity of the sample, in terms of the age range and comorbid pathology, although this heterogeneity has been used to evaluate different subgroups that contribute more scientific evidence on the phenomenon. In addition, some variables, such as the degree of isolation and social network, have not been evaluated objectively through standardized scales, which could be used as a reference for future research. Due to the long 12-months follow-up, some data has not been possible to obtain for all cases, such as the social network (Table 2), which supposes a remarkable loss of information.

In conclusion, this study reinforces the existence of hikikomori-isolation syndrome in our culture, supporting the hypothesis that it is a syndrome comorbid with several psychiatric disorders. In turn, possible subgroups, depending on the comorbid condition, may exist. Included in these subgroups are patients with anxiety or affective disorder who present specific differential characteristics with other heterogeneous cases that could resemble the hikikomori syndrome described in Japan and have specific treatment needs. This analysis would provide data to reach consensus on the diagnostic criteria and establish the specific characteristics of social isolation hikikomori syndrome. The 12-month follow-up analysis demonstrated the fragility and tendency to relapse and have disengagement, which was particularly relevant in the subgroup of patients with mood-anxiety disorders. Intensive treatments may be more effective than conventional outpatient treatments.

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Conflicts of interest

The authors declare no conflicts of interest.

Disclaimer

The U.S. Department of Veterans Affairs played no role in the design and conduct of the study; in the data collection, management, analysis, and interpretation; in the preparation, review, or approval of the manuscript; or in decision to submit the manuscript for publication.

The findings and conclusions in this document are those of the authors, who are responsible for its contents. The findings and conclusions do not necessarily represent the views of the U.S. Department of Veterans Affairs or the United States government.

Fig. 1. Confirmed follow-up for outpatient medical resources in cases with social isolation after being referred from the CRHT team (during the 12 months)

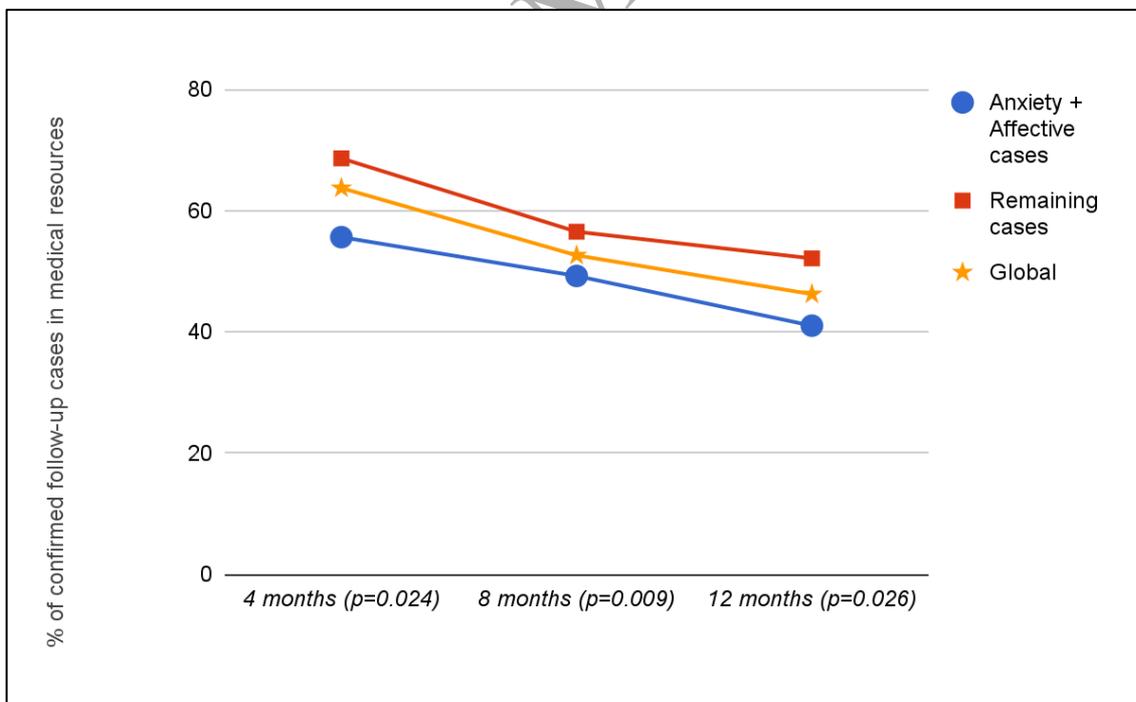


Table 1

Socio-demographic and clinical differences between the socially isolated cases with comorbid anxiety or affective disorder and cases with different comorbid disorders who were at home by CRHT

Variables	Global (<i>n</i> = 190) <i>n</i> (%)	Anxiety + affective cases (<i>n</i> = 63) <i>n</i> (%)	Remaining cases (<i>n</i> = 127) <i>n</i> (%)	<i>p</i>
Gender, Male, <i>n</i> (%)	136 (72.1)	37 (58.7)	99 (78.6)	0.004
Age, mean (<i>SD</i>)	39.1 (18.1)	43.6 (20.65)	37 (16.04)	0.018
Isolation period, months, mean (<i>SD</i>)	38.1 (52.3)	36.5 (47.4)	39.1 (54.8)	0.734
Age at onset of isolation, mean (<i>SD</i>)	36.2 (17.9)	39.8 (20.6)	34.4 (16.2)	0.073
Social network at the beginning of CRHT treatment				0.032
- None	65 (34.2)	15 (23.8)	49 (38.9)	
- People living together	109(57.4)	39 (61.8)	71 (55.6)	
- People outside the home	16 (8.4)	9 (14.3)	7 (5.6)	
Social network at the end of CRHT treatment				0.027
- None	41 (21.6)	7 (11.1)	34 (26.2)	
- People living together	118(62.1)	40 (63.5)	78 (61.9)	
- People outside the home	29 (15.3)	15 (23.8)	14 (11.1)	
- Normalized	2 (1.1)	1 (1.6)	1 (0.8)	
Psychiatric history	141 (74.7)	47 (33.3)	94 (66.7)	<0.001
- Drug use disorder	5 (3.5)	2 (4.3)	3 (3.2)	
- Psychotic disorder	53 (37.6)	3 (6.4)	50 (53.2)	
- Affective disorder	29 (20.6)	18 (38.3)	11 (11.7)	
- Anxiety disorder	32 (22.7)	22 (46.8)	10 (10.6)	
- Personality disorder	15(10.6)	0 (0)	15 (16)	
- Other	7 (5)	2 (4.3)	5 (5.3)	
Aggressiveness history	39 (20.7)	7 (11.1)	32 (25.8)	0.019
Suicide history	19 (10.1)	5 (7.9)	14 (11.2)	0.483
Internet addiction	59 (31.4)	20 (30.2)	39 (31.5)	0.851

Referred to service by the CRHT				0.006
- Medical outpatient team	26 (16.6)	8 (14.8)	18 (17.5)	
- Psychiatric outpatient team	62 (39.5)	31 (57.4)	31 (30.1)	
- Hospitalization	48 (30.6)	9 (16.7)	39 (37.9)	
- Other	21 (13.4)	6 (11.1)	15 (14.6)	
Number of visits by the CRHT, M (SD)	4.2 (3.8)	5.71 (5.37)	3.59 (2.59)	<0.001
WHO/DAS total, mean (SD)	12.8 (3.7)	11.66 (3.54)	13.45 (3.77)	0.002
GAF at the beginning of CRHT treatment, mean (SD)	40.7 (15)	42.74 (13.29)	39.37 (15.6)	0.148
GAF at the end of CRHT treatment, mean (SD)	45.7 (16.7)	50.47 (14.69)	42.98 (17.2)	0.003
SUMD Total, mean (SD)	10 (4.2)	6.44 (3.76)	10.68 (4.03)	<0.001
SPI total, mean (SD)	12.6 (4.9)	9.98 (4.75)	13.52 (4.7)	<0.001

SPI: Severity of Psychiatric Illness Scale GAF: Global Assessment of Functioning Scale; WHO/DAS: World Health Organization Disability Assessment Scale; CRHT: Crisis Resolution Home Treatment

Table 2

Comparative analysis of the 12-month evolution in the anxiety or affective subgroup vs other cases of social isolation once discharged from CRHT and referred to outpatient or inpatient treatment centres

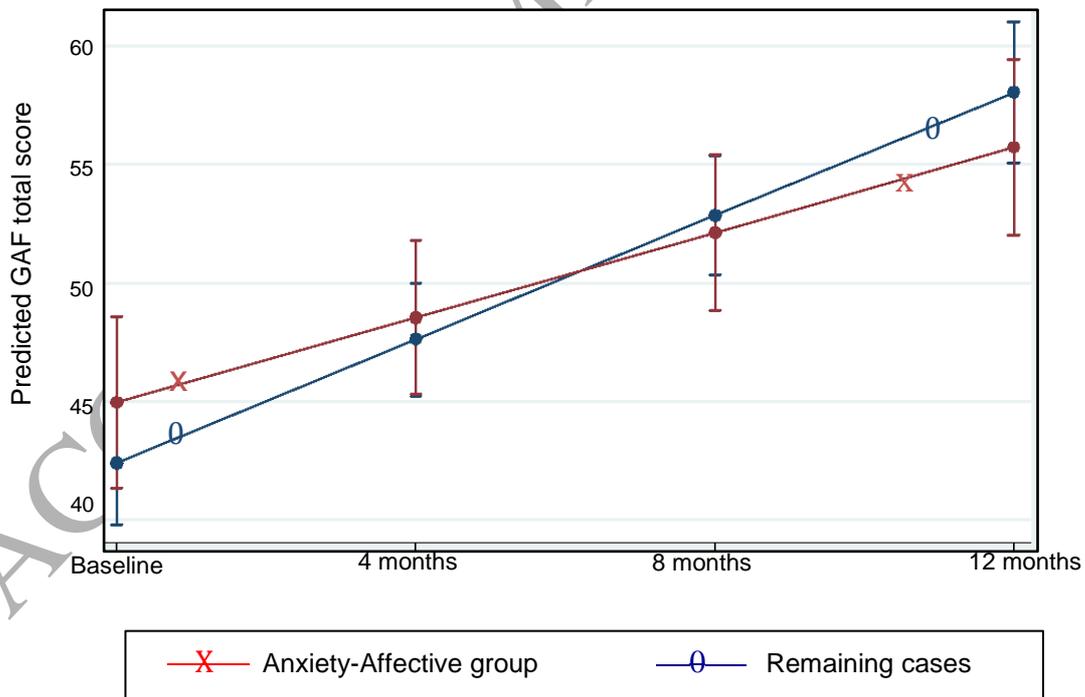
Variable	4 months			8 months			12 months		
	Anxiety + affective cases (<i>n</i> = 63) <i>n</i> (%)	Remaining cases (<i>n</i> = 127) <i>n</i> (%)	<i>p</i>	Anxiety + affective cases (<i>n</i> = 63) <i>n</i> (%)	Remaining cases (<i>n</i> = 127) <i>n</i> (%)	<i>p</i>	Anxiety + affective cases (<i>n</i> = 63) <i>n</i> (%)	Remaining cases (<i>n</i> = 127) <i>n</i> (%)	<i>p</i>
Social isolation persistence*	56 (91.8)	78 (84.8)	0.197	50 (87.7)	69 (79.3)	0.193	48 (84.2)	64 (76.2)	0.248
Better social isolation condition**	19 (32.2)	50 (54.9)	0.015	18 (31.6)	38 (47.7)	0.031	14 (24.6)	36 (42.9)	0.006
Social network, <i>n</i> (%)			0.491			0.965			0.402
- None	7 (12.1)	7 (12.1)		7 (12.3)	12 (14)		4 (7)	13 (15.9)	
- People living together	30 (51.7)	7 (7.7)		31 (54.4)	43 (50)		34 (59.6)	40 (48.8)	
- People outside home	20 (34.5)	24 (26.4)		16 (28.1)	26 (30.2)		11 (19.3)	17 (20.7)	
- Normal	1 (1.7)	2 (2.2)		3 (5.3)	5 (5.8)		8 (14)	12 (14.6)	
Responsible medical service			0.200			0.974			0.991
- Medical outpatient service	7 (11.7)	11 (10.3)		7 (11.7)	11 (11.6)		7 (11.9)	10 (11)	
- Psychiatric outpatient service	49 (81.7)	76 (71)		49 (81.7)	77 (81.1)		48 (81.4)	75 (82.4)	
- Inpatient unit	4 (6.7)	19 (17.8)		3 (5)	6 (6.3)		3 (5.1)	4 (4.4)	
- Other	0	1 (0.9)		1 (1.7)	1 (1.1)		1 (1.7)	2 (2.2)	
GAF	50.80 (14.24)	53.37 (13.4)	0.281	53.75 (14.87)	54.89 (15.5)	0.663	53.39 (16.49)	55.82 (17.7)	0.413
WHODAS Total	10.82 (3.79)	11.55 (3.73)	0.263	10.64 (4.22)	11.05 (4.44)	0.557	10.64 (4.97)	10.73 (4.03)	0.922

*Social isolation persistence: cases that maintain social isolation behaviour; **Better social isolation condition: cases with improvement in the severity of the isolation behaviour

Table 3

Regression model of the influence of anxiety-affective diagnostic group across a 12-month evolution period

	Isolation state		WHODAS Total score		GAF Total score	
	Odds ratio (SD)	<i>p</i>	Coef. (SD)	<i>p</i>	Coef. (SD)	<i>p</i>
Anxiety-affective disorder	0.27 (0.12)	0.004	-2.20 (0.70)	0.002	4.17 (2.71)	0.124
Evolution time	0.80 (0.05)	0.001	-0.86 (0.11)	<0.001	5.22 (0.49)	<0.001
Anxiety-affective disorder and evolution time	1.23 (0.13)	0.048	0.51 (0.17)	0.004	-1.62 (0.77)	0.037
Constant	1.73 (0.47)	0.043	13.96 (0.41)	<0.001	37.17 (1.6)	<0.001

Fig. 2 Influence of anxiety-affective diagnoses in total GAF score across the 12-month evolution period

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