



## Research article

## Development of 5-day hikikomori intervention program for family members: A single-arm pilot trial



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## ABSTRACT

**Backgrounds:** Hikikomori, a severe form of social withdrawal, is increasingly a serious mental health issue worldwide. Hikikomori is comorbid with various psychiatric conditions including depression, social anxiety and suicidal behaviors. Family support is encouraged as a vital first step, however evidence-based programs have yet to be established. Mental Health First Aid (MHFA) is one of the most well-validated educational programs encouraging lay people such as family members, to support close persons suffering from various psychiatric conditions such as depression, anxiety and suicidal behaviors.

**Methods:** We newly developed an educational program for family members of hikikomori sufferers mainly based on MHFA and 'Community Reinforcement and Family Training (CRAFT)' with role-play and homework. As a single-arm trial, 21 parents (7 fathers and 14 mothers) living with hikikomori sufferers participated in our program with five once-a-week sessions (2 h per session) and six monthly follow-ups, and its effectiveness was evaluated using various self-rated questionnaires.

**Results:** Perceived skills toward a depressed hikikomori case vignette, stigma held by participants, and subscales of two problematic and one adaptive behaviors of hikikomori sufferers were improved throughout the sessions and follow-ups. In addition, positive behavioral changes of hikikomori sufferers such as improved social participation were reported by participants.

**Limitations:** Single-arm design and evaluation using self-rated questionnaires are the main limitations of the present study.

**Conclusions:** Our newly developed program has positive effects on family members in their contact and support of hikikomori sufferers. Future trials with control groups are required to validate the effectiveness of this program.

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## 1. Introduction

Hikikomori, a severe form of social withdrawal, has been highlighted in Japan since the late 1990s (Saito, 1998; Kato et al., 2011, 2018). A survey by Japan's Cabinet Office in 2016 estimated that hikikomori sufferers aged between 15 and 39 number 540,000 in Japan (Cabinet Office, 2016). Moreover, the same office has recently estimated the number of hikikomori between 40 and 65 to be 610,000 in Japan (Cabinet Office, 2019). Sufferers with hikikomori have been reported not only in Japan but also in many other countries and it is increasingly considered to be serious worldwide condition (Kato et al., 2011, 2012, 2016b, 2018, 2019; Teo et al., 2015a, 2015b; Wu et al., 2019).

Hikikomori is characterized as 1) staying at home for most of the day, 2) avoidance of social participation (e.g. working and schooling), and 3) these conditions continue at least six months (Saito, 2010a). Hikikomori cases are often comorbid with a variety of psychiatric disorders such as mood disorders (especially major depressive disorder), schizophrenia, social anxiety disorders, and personality disorders (Kondo et al., 2013; Teo et al., 2015b). Furthermore, a recent epidemiological survey has shown that hikikomori sufferers have a higher risk of suicide (Yong and Nomura, 2019).

A recent survey has revealed that the average age of people with hikikomori was 34.4 years old and the average duration of the hikikomori condition was 9.6 years (KHJ, 2018). Prolonged and aging hikikomori is becoming a novel social issue, called the 80–50 problem (hikikomori sufferers in their 50s living with parents in their 80s). Support for people with hikikomori condition is prone to delay. Only 6.6 % of hikikomori sufferers initiate the search for help by themselves, and in 72.2 % of cases it is a parent or relative who is first to consult support institutions (Ito, 2003). In addition, it takes 4.4 years on average for people with hikikomori to utilize support. In 13.0 % of hikikomori cases, it takes more than 10 years to initial support (Saito, 2010b).

Therefore, more effective intervention approaches for hikikomori are warranted. In 2010, Japan's Ministry of Health, Labor, and Welfare published "Guidelines for assessment and support of hikikomori," and proposed a four grade step-by-step approach. Family support/approach is encouraged as the first step (Saito, 2010a). However, an evidence-based family intervention approach to deal with mental health problems has not been developed. Family members (mainly parents) have difficulty in approaching hikikomori sufferers mainly due to a lack of knowledge (about mental health problems and hikikomori condition in particular) and stigma towards such problems (Kato et al., 2017a). In many cases, family members tend not to provide appropriate support, and may "turn a blind eye" for many years (Kato et al., 2017a). We suppose that delay of direct support for hikikomori sufferers is partly caused by such parental behaviors. Thus, it is important for family members, especially parents, to acquire appropriate knowledge and skills for dealing with hikikomori sufferers directly.

Mental Health First Aid (MHFA) is a mental health educational/training program for non-professionals of mental health such as school teachers, firefighters and lay people, originally developed in Australia in 2000 (Kitchener and Jorm, 2002; Langlands et al., 2008). The MHFA program helps participants not only to increase mental health literacy, but also to obtain practical skills to provide help to persons with mental health problems by learning the five-step MHFA principles (Kitchener and Jorm, 2002). The five-step MHFA principles (3rd version) are as follows: Step 1) Approach the person, assess and assist with any crisis; Step 2) Listen non-judgmentally; Step 3) Give support and information; Step 4) Encourage the person to get appropriate professional help; and Step 5) Encourage other support (Kitchener et al., 2013). The 12-h standard MHFA program utilizes the same five-step principles targeted to four psychiatric conditions (depression, anxiety, psychosis, substance abuse) (Kitchener et al., 2013).

Hikikomori sufferers often have a variety of psychiatric conditions, thus we hypothesize that the MHFA five-step approach is applicable to most hikikomori cases.

In addition, the cognitive behavioral approach, especially Community Reinforcement and Family Training (CRAFT), is considered a candidate approach for family members of hikikomori sufferers (Saito, 2010a). CRAFT was originally developed for substance use disorders as a family intervention method (Smith and Meyers, 2004). Sakai and his colleagues modified the original CRAFT program for parents with hikikomori sufferers to encourage help-seeking behaviors (Nonaka et al., 2013; Sakai and Nonaka, 2013). The CRAFT program targeting family members of hikikomori sufferers is conducted as both individual (Sakai et al., 2015) and group sessions (Nonaka et al., 2013). In this program, functional analysis, an assessment method based on behavioral therapy, is applied to alter dysfunctional family interactions (Sakai and Nonaka, 2013). Functional analysis of behavior therapy (FABT) helps to clarify the chronological relationship among targeting behaviors (e.g. aggressive behaviors by hikikomori sufferers), prior events to the targeting behaviors (e.g. parental criticism) and following results (e.g. parents avoid hikikomori sufferers due to aggressive behaviors). FABT emphasizes that changing prior events (e.g. exchange criticism for calm words) can alter the targeting behaviors and following results for more appropriate interactions among family members (Hanley et al., 2003). In addition, positive communication skills are introduced by the CRAFT program for hikikomori so that parents can alter dysfunctional prior events such as criticizing, scolding, or vague words (Sakai and Nonaka, 2013).

We herein hypothesize that the combination of MHFA and CRAFT (mainly FABT) provides a helpful option in supporting family members with hikikomori sufferers. The purpose of the present study is to develop a 5-day family intervention program combining MHFA and CRAFT, and to evaluate the effectiveness of the program as a single-arm open trial.

## 2. Materials and methods

### 2.1. Ethics

This study was approved by the ethics committee of Kyushu University, and was registered at the UMIN Clinical Trials Registry (UMIN000029643; [https://upload.umin.ac.jp/cgi-open-bin/ctr/ctr\\_view.cgi?recptno=R000033862](https://upload.umin.ac.jp/cgi-open-bin/ctr/ctr_view.cgi?recptno=R000033862)).

### 2.2. Program development

The original MHFA is 12-hour program (Kitchener and Jorm, 2002). Previously we have developed short-time MHFA-based intervention programs so that participants can learn the essence and basic skills of MHFA efficiently for specific settings such as a medical setting for clinical patients and an ordinary company setting for office workers (Kato et al., 2010; Suzuki et al., 2014; Kubo et al., 2018; Nakagami et al., 2018).

We herein developed a family intervention program consisting of five once-a-week sessions (2 h per session) including lectures, group-work (i.e. role-playing) and homework based on the MHFA (the five-step MHFA principles of depression and suicide intervention), and CRAFT programs (FABT) to encourage participants to practice communication with hikikomori sufferers in daily life (Figure 1). We introduced basic knowledge about mental health literacy including the biopsychosocial background and therapeutic approach for hikikomori (Kato et al., 2011, 2012, 2016a, 2017b, 2018; Hayakawa et al., 2018; Teo et al., 2018). Next, we introduced the basics of MHFA, especially focusing on MHFA skills for depression and suicidal behavior in the supporting hikikomori sufferers. In addition, we introduced the positive methods of communication practiced in CRAFT program (Sakai and Nonaka, 2013). Detailed contents of each session are shown in Table 1.

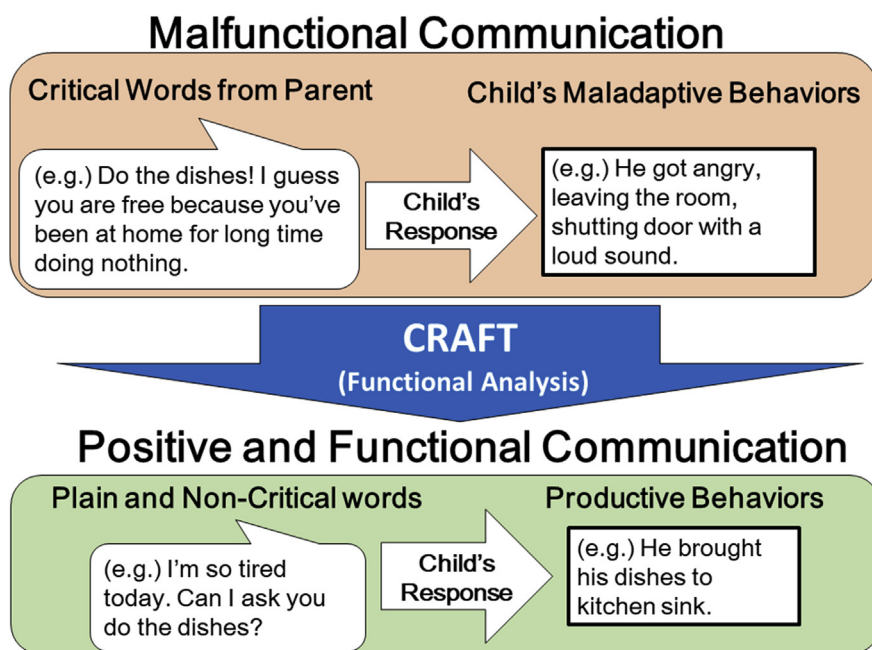


Figure 1. Group work of communication based on CRAFT using FABT.

### 3. Measurements

#### 3.1. Demographic data

Demographic data of participants themselves and hikikomori sufferers were collected. The data included the age, gender and relationship of participants and hikikomori sufferers, and the duration of the hikikomori condition. The present condition of hikikomori sufferers (e.g. how often they went out or how frequently they utilized support) and their life history were also collected indirectly from participants.

#### 3.2. Perceived skill

To evaluate perceived skills based on MHFA for early intervention of hikikomori cases, an original questionnaire using a case vignette of hikikomori was developed as shown below:

*“Mr. A is 30 years old. After graduating university, he was employed at a company. However, he quit his job six months after employment. Then, he got part-time job, however, he was unable to continue to work. He changed his part-time job several times. Afterwards, he didn't work or seek a job for a year. Now, he seldom goes out of his room. When you occasionally see him in the hallway, he returns to his room. He seems to avoid family members, and his face is vacant and gloomy. You (respondent), a parent of Mr. A, are worrying about him.”*

Respondents are asked to imagine themselves as a parent of Mr. A, and to answer nine questions rating the possibility of performing behaviors in accordance with MHFA: (1) leave Mr. A alone for a while (This is an inappropriate behavior based on MHFA; assessed as a reversal score), (2) tell Mr. A that you are concerned about him at an early stage, (3) consult a support center about Mr. A, (4) greet Mr. A when you see him, (5) ask about Mr. A's physical condition such as appetite or sleep, (6) ask about Mr. A's feelings of depression, (7) directly convey the possibility of depression when Mr. A's depressed mood proved to be serious, (8) directly ask about suicidal thoughts when you think Mr. A probably suffers from depression, (9) recommend Mr. A visit a psychiatric or psychosomatic department.

The possibility of each behavior was rated using five-point scale indicating 0 (absolutely no), 1 (probably not), 2 (don't know), 3 (probably yes), and 4 (absolutely yes).

#### 3.3. Confidence

The confidence level in management of people with hikikomori expressing depressive symptoms was evaluated by six original questions

Table 1. Contents of the 5-day hikikomori intervention program for family members.

<p>Session 1</p> <p>A psychiatrist (TAK) introduced 1) knowledge about mental health literacy including the biopsychosocial background and therapeutic approach for hikikomori, 2) the MHFA 5-step principles, and 3) information about supporting resources for people with hikikomori.</p> <p>For homework, we required participants to record the daily behaviors of their children (greetings, conversation, asking for help, calling for dinner, etc.).</p>
<p>Session 2</p> <p>To organize more appropriate interactions between the parent and their children, we introduced the MHFA approach for hikikomori and FABT.</p> <p>For homework, we required participants to classify the behavior of their children into desirable (greetings, reply to parent's proposal, going out, etc.) and undesirable (ignoring family members, abusive words to family members, shut-in, etc.) behaviors.</p>
<p>Session 3</p> <p>At first, to obtain listening skills, role-playing of active listening (i.e. Step 2 of MHFA; listen non-judgmentally) was introduced. A pair of participants played listener and speaker roles alternately. Next, FABT-based practice to enhance desirable behaviors was conducted to alter interactions between the parent and their children (see Figure 1).</p> <p>Positive communication based on CRAFT includes plain, short, and positive way of speaking rather than using vague, confusing, or negative words.</p> <p>For homework, we required participants to think of positive words suitable with CRAFT communication to enhance desirable behaviors of their children.</p>
<p>Session 4</p> <p>To acquire practical skills in encouraging their children to utilize professional support, a pair of participants alternately played the case scenario of a parent and a hikikomori sufferer based on MHFA.</p> <p>For homework, we required participants to list up supporting resources such as psychiatric clinics and hikikomori support centers.</p>
<p>Session 5</p> <p>We reviewed the whole of the program and demonstrated breathing technique as a MHFA self-help strategy. Next, Q &amp; A time was introduced to discuss the difficulty in approaching their children with hikikomori condition. Finally, we informed the participants of individual and group follow-up sessions.</p>

based on the MHFA: (1) approach a person with hikikomori, (2) listen non-judgmentally, (3) tell the possibility of clinical depression, (4) give support and information, (5) ask about “suicidal thoughts,” (6) encourage the person to obtain appropriate professional help. Respondents were asked to answer these questions on a five-point scale, ranging from 0 (not confident at all) to 4 (very confident).

### 3.4. Stigma

The Japanese version of 12-item Link's Devaluation-Discrimination Scale was conducted to measure respondents' stigma toward mental health problems (Shimotsu et al., 2006).

### 3.5. Problematic behaviors of hikikomori

Hikikomori Behavior Checklist (HBCL) was conducted to measure problematic behaviors by the family members (children) of participants (Sakai et al., 2004). This scale is consisted of 45 items and has 10 factors as sub-scales: (1) Aggressive behavior, (2) Social anxiety, (3) Obsessive-compulsive behavior, (4) Avoidance from family members, (5) Depression, (6) Absence of activities of daily living, (7) Incomprehensible maladapted behavior, (8) Absence of social participation, (9) Decreased activity, and (10) Irregular life pattern. Respondents were asked regarding behavior within the last three months with a four-point scale.

### 3.6. Adaptive behavior of hikikomori

Adaptive Behaviors Scale for Hikikomori (ABS-H) was conducted to assess the adaptive behavior of the hikikomori sufferer (Nonaka et al., 2018). This scale is consisted of 26 items and has four factors as sub-scales: (1) Interaction, (2) Family, (3) Value, and (4) Social participation. Respondents were asked to answer regarding the behaviors of their family members (children) within the last three months.

### 3.7. Depressive symptoms of participants

The Japanese version of the Patient Health Questionnaire (PHQ-9) was conducted to assess the severity of depressive symptoms within the last two weeks (Kroenke et al., 2001; Muramatsu et al., 2007).

### 3.8. Stress responses of participants

Stress Response Scale-18 (SRS-18) was conducted to measure psychological stress responses of respondents (Suzuki et al., 1997). This scale is consisted of 18 items and has three factors as sub-scales: (1) Depression-Anxiety, (2) Irritability-Anger, and (3) Helplessness.

### 3.9. Hikikomori condition

We evaluated a retrospective assessment on hikikomori condition at the final follow-up session (Jun 2018). Participants responded to a self-rated question whether their family member (child) was suffering from hikikomori during the period of pre-participation (Oct 2017) and during the period of the final follow-up session (Jun 2018), respectively.

## 4. Participants

Participants were recruited through advertisements issued by the municipal government. Regarding eligibility criteria, duration of the hikikomori period had to exceed six months. Participants were informed of the aims and methods of the present study and that their participation was completely voluntary. 21 parents (7 fathers and 14 mothers) living with a child with hikikomori participated in the present study. Participants who agreed to participate in the study then registered as study participants with written informed consent. All the participants (N = 21) agreed to join the present study.

## 5. Procedure

From November to December 2017, we implemented the newly developed family intervention program at Kyushu University hospital. The five-session program was held weekly as group learning. An experienced psychiatrist who is a MHFA-Japan trainer (TAK) conducted the session, and two clinical psychologists (HKU and HU) facilitated the session and conducted the group work.

During October 2017 (one month before the program), a 1-h individual interview was conducted to collect demographic data. In addition, we conducted six monthly follow-ups. The first follow-up, implemented in January 2018, was a 1-h individual interview. In the follow-up interview, we asked participants about problems in approaching their child. For the second to sixth follow-ups, along with the review of the program, we conducted group-work discussing the difficulties in approaching hikikomori sufferers in daily life, and coping with stress among participants.

Self-rated questionnaires were conducted just before Session 1 (#1), immediately after Session 5 (#5), and each follow-up sessions (from FU1 to FU6). SRS-18 was conducted at each time point (five sessions and six follow-ups). An illustration of the procedure is shown in Figure 2.

## 6. Statistical analysis

Data obtained from 21 participants was longitudinal for each self-rated questionnaire. Scores of SRS-18 were obtained at 11 time points (conducted every session from #1 to FU6); other scores were obtained at 8 time points (#1, #5 and FU1-FU6). Linear mixed-effects models were used to evaluate the longitudinal changes in each measurement. We included time (each time point) as a fixed effect; and we included participants, including drop-out patterns and missing scores, as a random effect. In addition, we reported marginal  $R^2$  (variance explained by fixed factors) as effect size (Nakagawa and Schielzeth, 2013).

In this study, 4 pairs of couples (both father and mother) jointly participated (total 8 participants). Thus, we conducted group comparison analysis between joint participation group (both parents have participated) and single participation group (either father or mother has participated). For group comparison, linear mixed-effects models were conducted as well as longitudinal changes of measurement mentioned above. Time (each time point) and parental participation type (joint or single participation) were included as fixed effects. In order to evaluate the effects of the present program on parents, we included perceived skills, confidence, stigma, depressive symptoms, and stress response among participants as group comparison analysis.

All analysis was performed using R version 3.5.1 (R Core Team, 2018). We used the lme4 package for fitting linear mixed-effect models and MuMIn package for calculating  $R^2$ .

## 7. Results

### 7.1. Demographic data of participants and hikikomori sufferers

21 parents (7 fathers and 14 mothers) participated in the present study. Among the 21 participants, 4 couples (husband and wife) participated. The average age of the participants was  $61.62 \pm 6.42$  (ranging 49–76). Their children with hikikomori condition were 13 males and 4 females, and the average age of the child was  $27.59 \pm 6.24$  (ranging 19–39). Average duration of hikikomori condition was  $100.94 \pm 59.37$  months (ranging 12–228 months). Participants' demographic data was shown in Table 2, and hikikomori sufferers' demographics were shown in Table 3.

### 7.2. Rate of attendance in the program

20 of 21 participants attended all five sessions. One participant did not attend Session 2 (#2). The average rate of attendance of the five-



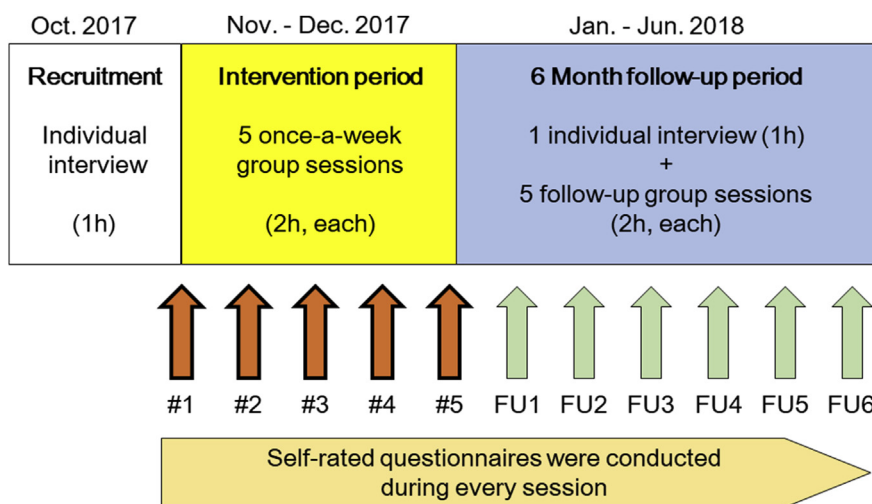


Figure 2. Procedure of the present study.

session program was 99.05 %. As for the six follow-ups, 16 participants attended all the follow-ups; one participant did not attend the follow-ups at all, one attended one follow-up, two participants (a couple) attended three follow-ups, and one attended five follow-ups. The average rate of attendance of six follow-ups was 85.71 %.

7.3. Effectiveness of the program

The average scores of each time point of the self-rated questionnaires were shown in Table 4. Mixed-effects modeling of the present program was shown in Table 5. Although effect sizes are low, some scores of self-rated questionnaires proved to show significant changes throughout program sessions and follow-ups as shown below.

The score of perceived skills in early intervention for depressive symptoms of a hikikomori case based on MHFA was significantly improved throughout the sessions and follow-ups (Slope = 0.25, 95% CI = [0.05, 0.45], p = 0.02). Average scores of each sub-item and their mixed-effects modeling were shown in Supplement 1. Some scores of the sub-items were significantly improved. Throughout the sessions and follow-ups, participants tended not to leave hikikomori sufferers alone and showed significant improvement on perceived skills in approaching and asking depressive feelings and suicidal thoughts (these skills are recommended alongside with MHFA).

In addition, stigma toward mental health problems was significantly reduced throughout the sessions and follow-ups (Slope = -0.29, 95% CI = [-0.42, -0.15], p < 0.001) (Table 5).

Furthermore, some hikikomori sufferers' behaviors reported by participants were significantly improved (Table 5). Specifically, adaptive

Table 2. Demographic data of participants (parents).

	N	%
Total	21	
Sex		
Male	7	33.3
Female	14	66.7
Age		
40–49	1	4.8
50–59	8	38.1
60–69	10	47.6
70–79	2	9.5
Mean	61.62 (SD = 6.42)	

Note: Among the 21 participants, 4 pairs of couples (husband and wife) participated.

behavior of “value” score assessed with ABS-H were significantly improved (Slope = 0.12, 95% CI = [0.03, 0.21], p = 0.01). Value score of ABS-H represents “acting/trying to approach the ideal/goal (Nonaka et al., 2018).” Two problematic hikikomori behaviors based on HBCL, “obsessive-compulsive behavior” and “decreased activity”, were significantly reduced throughout the sessions and follow-ups (Slope = -0.09, 95% CI = [-0.15, -0.03], p = 0.01; Slope = -0.06, 95% CI = [-0.12, 0.00], p = 0.04, respectively).

Hikikomori condition (reported indirectly by parents) were shown in Table 6. Two out of 18 respondents reported that their child's hikikomori condition improved at last follow-up compared with pre-participation in the present study.

7.4. Group comparison between joint participation group and single participation group

Among 21 participants, 4 couples (8 participants) have jointly participated in the present study. We have compared the effects of the present program on parents between joint participation group and single participation group. As for perceived skills, confidence and stigma, no significant differences were observed between two groups. Total score of PHQ-9, two stress response sub-scales (Depression-Anxiety and Helplessness) and total score of SRS-18 were significantly lower among joint participation group compared to single participation group from before participation (#1) to the end of the follow-ups (FU6) (shown in Supplement 2).

Table 3. Demographic data of hikikomori sufferers.

	N	%
Total	17	
Sex		
Male	13	76.5
Female	4	23.5
Age		
10–19	1	5.9
20–29	10	58.8
30–39	6	35.3
Mean	27.59 (SD = 6.24)	
Duration of hikikomori (months)		
12–59	6	35.3
60–119	5	29.4
120–	6	35.3
Mean	100.94 (SD = 59.37)	

**Table 4.** Average scores of each time point of the self-rated questionnaires.

		#1	#5	FU1	FU2	FU3	FU4	FU5	FU6			
<b>Perceived skills</b>												
	N	21	21	20	18	19	19	19	17			
Total	Mean	22.71	26.10	25.65	24.83	23.74	24.95	25.58	26.47			
	SD	5.09	4.86	4.63	4.57	6.50	5.51	4.68	4.86			
<b>Confidence</b>												
	N	21	21	20	19	19	19	19	16			
Total	Mean	10.52	11.76	11.35	11.21	11.68	11.05	12.74	11.75			
	SD	4.03	3.38	3.54	3.53	5.14	4.29	6.26	3.77			
<b>Stigma</b>												
	N	20	21	20	19	18	18	19	17			
Total	Mean	31.70	30.14	29.25	28.84	29.78	29.39	29.63	28.47			
	SD	6.03	5.41	6.10	6.10	5.69	6.66	6.19	6.45			
<b>HBCL</b>												
	N	20	20	20	19	16	17	18	16			
Aggressive behavior	Mean	7.00	6.90	6.80	6.16	7.19	7.06	7.22	6.75			
	SD	5.10	5.37	5.37	5.07	5.91	4.95	5.14	4.97			
Social anxiety	Mean	6.25	6.05	5.89	6.44	6.65	6.44	6.31	6.10			
	SD	2.66	3.14	2.63	2.76	2.72	2.19	2.05	2.70			
Obsessive-compulsive behavior	Mean	4.20	4.20	3.89	3.63	3.76	3.44	3.69	4.25			
	SD	2.62	2.29	2.51	2.20	1.83	2.06	2.34	2.64			
Avoidance from family members	Mean	5.45	5.95	5.58	5.94	5.76	5.56	5.56	5.50			
	SD	3.51	3.46	3.56	3.40	2.98	3.00	2.87	3.58			
Depression	Mean	3.10	3.65	2.32	4.50	2.82	2.83	2.63	3.40			
	SD	3.21	3.09	2.96	3.43	2.59	2.79	2.60	2.91			
Absence of activities of daily living	Mean	6.95	7.65	7.21	7.94	7.29	7.39	7.25	6.95			
	SD	4.02	3.89	3.61	3.93	3.46	3.11	3.91	3.90			
Incomprehensible maladapted behavior	Mean	2.70	2.85	2.74	3.38	3.29	2.78	2.50	2.55			
	SD	1.49	2.08	1.62	2.12	1.77	1.72	1.46	1.60			
Absence of social participation	Mean	7.75	7.75	7.58	8.06	7.65	7.50	7.38	7.55			
	SD	1.26	1.51	1.46	1.30	1.53	1.38	1.49	1.47			
Decreased activity	Mean	5.70	5.75	5.63	5.81	5.47	5.50	5.44	5.90			
	SD	1.55	1.61	1.90	1.33	1.42	1.50	1.97	1.70			
Irregular life pattern	Mean	5.50	6.10	5.11	6.44	6.47	6.06	5.63	6.10			
	SD	2.42	2.47	2.36	2.18	1.85	1.90	2.45	2.43			
Total	Mean	54.50	56.75	52.11	59.31	56.24	54.72	53.13	55.30			
	SD	17.12	17.96	17.37	17.03	13.19	12.85	14.25	15.57			
<b>ABS-H</b>												
	N	18	19	20	19	17	18	16	16			
Interaction	Mean	10.17	9.26	10.20	9.58	9.88	10.22	8.56	10.81			
	SD	5.80	5.46	6.73	5.73	6.59	5.96	5.71	7.25			
Family	Mean	6.94	7.32	6.85	7.53	7.29	7.67	6.75	7.63			
	SD	2.91	2.96	2.57	2.74	2.67	2.85	2.54	2.50			
Value	Mean	3.78	3.95	4.50	4.37	4.35	4.50	3.75	4.69			
	SD	2.17	2.06	2.60	3.34	3.03	3.04	2.05	2.73			
Social participation	Mean	2.17	1.16	1.90	2.79	2.24	1.72	1.69	1.75			
	SD	1.89	1.18	2.26	2.12	2.26	2.21	1.99	2.17			
Total	Mean	23.06	21.68	23.45	24.26	23.76	24.11	20.75	24.88			
	SD	9.78	8.66	10.84	10.14	10.22	10.73	8.73	12.52			
<b>PHQ-9</b>												
	N	21	21	20	18	18	19	19	17			
Total	Mean	4.43	4.43	4.55	2.72	4.22	3.95	4.00	3.12			
	SD	5.40	4.47	5.14	2.94	4.54	4.39	4.29	5.20			
<b>SRS-18</b>												
		#1	#2	#3	#4	#5	FU1	FU2	FU3	FU4	FU5	FU6
	N	20	20	20	21	20	20	19	19	19	19	16
Depression-Anxiety	Mean	4.60	3.75	3.00	2.62	2.95	4.15	3.74	4.26	4.32	3.42	3.56
	SD	4.20	3.90	2.93	2.77	2.94	3.85	3.35	4.64	4.27	3.90	4.33
Irritation-Anger	Mean	2.30	2.25	1.45	1.33	1.95	2.20	1.32	2.84	2.37	2.00	1.75
	SD	2.69	2.61	1.66	1.70	2.33	2.64	2.00	4.17	3.01	3.09	2.61

(continued on next page)

Table 4 (continued)

		#1	#5	FU1	FU2	FU3	FU4	FU5	FU6		
Helplessness	Mean	3.30	3.90	3.10	2.95	2.85	2.70	2.37	3.37	3.68	3.26
	SD	3.58	3.55	2.74	2.80	2.87	3.24	2.52	3.51	4.14	3.96
Total	Mean	10.20	9.90	7.55	6.90	7.75	9.05	7.42	10.47	10.37	8.68
	SD	9.44	8.76	6.67	6.25	7.29	9.13	7.08	11.69	10.42	10.32

8. Discussion

In the present study, we newly developed a five-day intervention program, combining MHFA and CRAFT, for family members (mainly parents) who live with a child suffering from hikikomori. The trial program was conducted for five weekly sessions and six monthly follow-up sessions, for 21 parents who live with a child suffering with hikikomori,

Table 5. Estimated treatment effect from the linear mixed-model analysis.

Outcome	Slope	Standard error	p value	95% CI	R <sup>2</sup>
Perceived skills					
Total	0.25	0.10	*0.02	(0.05, 0.45)	0.02
Confidence					
Total	0.07	0.08	0.38	(-0.08, 0.22)	0.00
Stigma					
Total	-0.29	0.07	***0.00	(-0.42, -0.15)	0.02
HBCL					
Aggressive behavior	-0.03	0.05	0.50	(-0.13, 0.06)	0.00
Social anxiety	0.02	0.04	0.59	(-0.05, 0.09)	0.00
Obsessive-compulsive behavior	-0.09	0.03	**0.01	(-0.15, -0.03)	0.01
Avoidance from family members	-0.02	0.04	0.50	(-0.10, 0.05)	0.00
Depression	-0.05	0.04	0.25	(-0.13, 0.03)	0.00
Absence of activities of daily living	-0.02	0.05	0.65	(-0.11, 0.07)	0.00
Incomprehensible maladapted behavior	0.03	0.03	0.36	(-0.04, 0.10)	0.00
Absence of social participation	-0.01	0.02	0.61	(-0.05, 0.03)	0.00
Decreased activity	-0.06	0.03	*0.04	(-0.12, -0.00)	0.01
Irregular life pattern	-0.03	0.03	0.29	(-0.09, 0.03)	0.00
Total	-0.27	0.22	0.24	(-0.70, 0.17)	0.00
ABS-H					
Social interaction	0.08	0.08	0.32	(-0.08, 0.25)	0.00
Family	0.06	0.03	0.05	(0.00, 0.12)	0.00
Value	0.12	0.05	*0.01	(0.03, 0.21)	0.02
Participation in society	-0.02	0.04	0.62	(-0.11, 0.06)	0.00
Total	0.24	0.14	0.08	(-0.03, 0.51)	0.00
PHQ-9					
Total	0.01	0.06	0.85	(-0.10, 0.12)	0.00
SRS-18					
Depression-Anxiety	0.06	0.04	0.15	(-0.02, 0.14)	0.00
Irritation-Anger	0.02	0.04	0.54	(-0.05, 0.10)	0.00
Helplessness	0.03	0.04	0.50	(-0.05, 0.10)	0.00
Total	0.11	0.09	0.24	(-0.07, 0.30)	0.00

CI: confidence interval.

\* p < .05, \*\* p < .01, \*\*\* p < .001.

and the present program has at least some positive effects on family members living with hikikomori sufferers.

The present program has improved participants' perceived skills for the management of a hikikomori sufferer with depressive symptoms (based on the case vignette), and reduced stigma toward mental health problems. MHFA is known to be effective in improving knowledge of mental health problems including psychiatric disorders, reducing stigmatizing attitudes, and increasing supportive behaviors (Hadlaczky et al., 2014). Hikikomori cases are often comorbid with psychiatric disorders (Kondo et al., 2013; Teo et al., 2015b). Thus, this program in encouraging the acquisition of MHFA-based knowledge and skills in dealing with mental health problems is suggested to be useful especially in hikikomori cases comorbid with psychiatric conditions. As for each sub-item score of perceived skills, asking "feelings of depression" and "suicidal thoughts" were improved (see Supplement 1). These results are consistent with previous studies that evaluated the effectiveness of short version of MHFA-based intervention programs for specific settings such as a medical setting for clinical patients and an ordinary company setting for office workers (Kato et al., 2010; Suzuki et al., 2014; Kubo et al., 2018; Nakagami et al., 2018). It is suggested that participants of the present study can obtain practical MHFA-oriented skills for management in hikikomori sufferers. In addition, Nakamura et al. reported that reduction in stigma toward mental health problems among family members enhances help-seeking behavior (Nakamura et al., 2006). The present program is expected to improve such aspects, and further validation is needed. These changes seen in self-rated questionnaires seemed to occur just after taking the program (between #1 and #5), and such changes were kept throughout follow-ups. We have conducted monthly follow-up sessions. It is possible that regular follow-ups would also have a positive effect on participants.

Perceived skills in approaching a child with hikikomori based on MHFA improved throughout the sessions and follow-ups, however participants' confidence in management of hikikomori cases was unchanged. We provided a large variety of content every week in the program, which may have been excessive. It is possible that participants felt difficulty in attaining sufficient success or competency in approaching their child due to the complexity of the program content. Thus, longer intervals between sessions (e.g. every other week) would give participants enough time to try newly obtained skills in their home with hikikomori sufferers. Furthermore, revising the program design and especially cutting down superfluous content would also be helpful.

The present program, consisted of five 2-h weekly sessions, showed improvement in obsessive-compulsive behavior and decreased activity. Previously, Sakai and Sakano developed a 12-session, 2-h group intervention program for family members of hikikomori sufferers, and evaluated the effectiveness of the program by assessing the changes in problematic behaviors (Sakai and Sakano, 2010). From 9th to 11th session of their intervention program, functional analysis of problematic behaviors based on behavior therapy and behavioral psychoeducation

Table 6. Hikikomori condition (indirectly reported by parents).

	Pre-participation	Last follow-up
Hikikomori condition		
Do you think your child is in hikikomori condition?		
Yes	17	15
No	1	3

were conducted, and outcomes between intervention group and control group were compared using self-rated questionnaires including HBCL among their participants. According to HBCL, decreased activity was improved in the intervention group while obsessive-compulsive behaviors worsened (Sakai and Sakano, 2010). On the other hand, Nonaka et al. implemented a monthly CRAFT program consisted of six 2-h sessions as a single-arm trial for parents of hikikomori cases, and they revealed that aggressive behaviors, avoidance of family members, incomprehensible maladapted behaviors and obsessive-compulsive behaviors were improved (Nonaka et al., 2013). The present study validated the improvement of obsessive-compulsive behaviors by CRAFT-oriented interventions.

A World Health Organization epidemiological survey in Japan between 2002 and 2006 targeting individuals aged between 15 and 49 years reported that 1.2% of the population has experienced hikikomori for a period greater than 6 months (Koyama et al., 2010). According to this survey, the lifetime prevalence of hikikomori is higher in males (1.8%) than in females (0.4%) (Koyama et al., 2010). In the present study, the number of male sufferers was approximately three times higher than females (13 males and 4 females). Thus, this construction would represent hikikomori's general characteristics. On the other hand, a series of surveys by Japan's Cabinet Office reported that hikikomori sufferers aged between 40 and 65 were estimated to exceed those of aged between 15 and 39 (Cabinet Office, 2016, 2019). As represented by the 80–50 problem, aging hikikomori is an urgent social issue in Japan. However, no participant living with a hikikomori sufferer more than 40 years of age participated in the present study. Further study targeting family members living with middle aged hikikomori is warranted to evaluate the effectiveness of the present program.

In the present study, aversive effects were not reported except for a decrease in time spent with children (worsening rate was 5.6 %). It is presumed that the present program is safe and of low invasiveness in supporting family members and their children with hikikomori. Furthermore, the attendance rate of the program was high (99.05% of five sessions; 85.71% of six follow-ups). This implies that our newly developed program is suitable for parental needs.

The MHFA program is reported to have positive effects on participants' mental health (Kitchener and Jorm, 2002). However, in the present trial, the mental health conditions of participants themselves including depressive symptoms and stress responses were not significantly changed throughout the sessions and follow-ups. The average total score of PHQ-9 (depressive symptoms) at each time point (#1, #5 and FU1-FU6) was ranged 2.72–4.55; SRS-18 (stress response) at each time point (#1-FU6) was ranged 6.90–10.47 (Table 4). These scores indicate that participants in this study have not expressed a clinical level of depressive symptoms or stress response (Suzuki et al., 1997; Kroenke et al., 2001; Muramatsu et al., 2007). Future studies targeting hikikomori family members with high depressive symptoms and/or psychological stress are warranted.

In the present study, joint participation group showed lower depressive symptoms and stress response than single participation group at baseline (#1). This result implies that parents who participated in this study as a couple may have a more positive marital relationship in their daily life. During the course of this program, we have directly observed positive behaviors by jointly participating fathers and mothers such as; 1) some participants talked together on their paternal/maternal roles in concerning with their hikikomori sufferer, and 2) some participants reviewed their previous marital relationships and tried to reform their roles vis-a-vis each other. Based on the above-preliminary observations, joint participation may have merits especially in rebuilding family relationships and enhancing appropriate paternal/maternal roles. Larger studies with additional scales should be conducted to confirm our hypothesis.

The present study has some limitations. First, all assessments were conducted based on self-rated questionnaires among participants (parents), thus actual behavioral changes in participants and their children

with hikikomori were not directly evaluated. In addition, behavioral changes of their children were assessed as a retrospective evaluation at final follow-up. Hence, recall bias would have affected participants' reports. Moreover, follow-up period was 6 months; behavioral changes of hikikomori sufferers would occur at larger time frame (e.g. not a monthly basis but a year basis). Longitudinal and longer evaluation of behavioral changes is preferable in future trials. Finally, the present study was a single-arm design with no control groups. In addition, effect size of the observed effectiveness was low (effect sizes of self-rated questionnaires were 0.01–0.02 at  $R^2$ ). Alongside with revision of the program, future randomized controlled trials are required to validate the effectiveness of the program. Hikikomori is becoming a serious issue not only in Japan but also worldwide (Kato et al., 2011, 2012, 2016b, 2018; Teo et al., 2015a, 2015b), thus it is vital to develop effective family intervention programs internationally.

## Declarations

### Author contribution statement

H. Kubo: Conceived and designed the experiments; Performed the experiments; Analyzed and interpreted the data; Contributed reagents, materials, analysis tools or data; Wrote the paper.

H. Urata: Performed the experiments; Analyzed and interpreted the data.

M. Sakai, S. Nonaka, K. Saito, M. Tateno, K. Kobara, N. Hashimoto, D. Fujisawa, Y. Suzuki and K. Otsuka: Contributed reagents, materials, analysis tools or data.

H. Kamimae, Y. Muto, T. Usami and Y. Honda: Performed the experiments.

J. Kishimoto: Analyzed and interpreted the data.

T. Kuroki and S. Kanba: Conceived and designed the experiments.

T. Kato: Conceived and designed the experiments; Performed the experiments; Analyzed and interpreted the data; Contributed reagents, materials, analysis tools or data; Wrote the paper.

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### Competing interest statement

The authors declare no conflict of interest.

### Additional information

The clinical trial described in this paper was registered at UMIN Clinical Trials Registry under the registration number UMIN000029643.

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