Research in Developmental Disabilities 94 (2019) 103458

Contents lists available at ScienceDirect

Research in Developmental Disabilities

journal homepage: www.elsevier.com/locate/redevdis

Efficacy of a psychoeducational intervention in caregivers of people with intellectual disabilities: A randomized controlled trial (EDUCA-IV trial)

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ARTICLE INFO

Number of reviews completed is 2
This paper belongs to the regular issue

Keywords:
Intellectual disability
Caregiver
Mental health
Psychoeducative program
Randomised controlled trial

ABSTRACT

Background: People with intellectual disabilities (ID) are usually cared for by their own parents, families or informal caregivers. Caring for a person with ID can have a negative impact on caregivers' mental health (burden, depression, anxiety). The main aim of the EDUCA-IV trial was testing the efficacy of a psychoeducational intervention program (PIP) versus standard practice and to see whether the PIP intervention would reduce the caregiver's burden at post-intervention (4 months) and at follow-up (8 months).

Method: This was a multi-centre randomised controlled trial including 194 caregivers (96 randomised to PIP, 98 to control condition). PIP intervention consists of 12 weekly group sessions. The control group received treatment as usual. Primary outcomes measured included the Zarit Burden Interview (ZBI). Secondary outcomes were caregivers' mental health (GHQ-28), anxiety (STAI) and depression (CES-D).

Results: The decrease of ZBI scores was not significant at 4 months. There was significant decrease in the GHQ scores at 4 and 8 months. CES-D showed relevant results at follow-up. Intention to treat analyses showed similar results.

Conclusions: The PIP intervention seems not to be effective reducing burden, but appears to have a positive result on general mental health. The program was well received and valued by caregivers.

What this paper adds?

Relatives are the ones who usually care for people with intellectual disability (ID), and, as informal caregivers, they may

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https://doi.org/10.1016/j.ridd.2019.103458
Received 21 January 2019; Received in revised form 2 August 2019; Accepted 5 August 2019
Available online 13 September 2019

experience negative psychological consequences (i.e. caregiver burden, mental health issues, depression or anxiety). Despite this issue being well-known and described, there is a lack of well-designed studies assessing psychological interventions that could help caregivers cope with demanding/stressful situations. In this randomised controlled study we tested the efficacy of a new psychological intervention programme (PIP) delivered in group form over 12 weekly sessions versus treatment as usual. Although the PIP was well accepted by caregivers and was rated as "very interesting and useful", our results showed that it failed in reducing burden, depression or anxiety at 4 months. Only caregivers' mental health outcome showed significant results in favour of the intervention arm. At follow-up period (8 months) only depression (CES-D) showed significant results. Intention-to-treat analyses were more conservative in all outcomes. Secondary analyses indicated that those caregivers with higher levels of burden at baseline benefited more from the PIP. These results, and the heavy strain that ID puts on the informal caregiver, remind of the importance of carrying out new trials for testing different intervention strategies aimed at reducing burden, improving caregivers' psychological well-being and teaching new strategies to cope with stress.

1. Introduction

Worldwide prevalence of intellectual disability (ID) is estimated at around 1% (Maulik, Mascarenhas, Mathers, Dua, & Saxena, 2011). Higher rates are detected in low and middle-income countries (Fitaw & Boersma, 2006; Njenga, 2009; Stein, Belmont, & Durkin, 1987; World Health Organization, 2018) or in urban slum/mixed rural-urban areas (Fitaw & Boersma, 2006). ID is associated with higher costs in terms of time and other resources (i.e. financial, etc.) for caregivers and society as a whole (Lakin, Prouty, Polister, & Smith, 2002).

In most cases, the care for and attention to people affected by ID is directly assumed by their parents or direct relatives. Caregivers should coordinate this role (including tasks related to health care assistance, special education, and special needs) with other family, work and personal responsibilities (Silver, 1999). Although family caregiving is not necessarily a negative experience (Summers, Behr, & Turnbull, 1989; Yoong & Koritsas, 2012), this circumstance may cause a plethora of negative consequences for caregivers (stress, poorer mental and physical health, reduced psychological well-being, lower quality of life…). In general, evidence from population-representative data suggested that informal caregivers of people with ID reported a higher impact on their personal life compared to caregivers of other chronic conditions or parents of children with normal development (Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001; Hastings et al., 2005; Woodman, Mawdsley, & Hauser-Cram, 2015). Population based studies have reported a similar negative impact on their personal life but with some nuances: when comparisons with other caregivers of patients with different chronic health conditions were controlled by socio-demographic (subjective poverty) and caring profile (caring load), providing care to a person with ID was not associated with reduced quality of life or poorer health status (Totsika, Hastings, & Vagenas, 2017).

If caregivers cannot cope effectively with these negative consequences, they are prone to suffer from chronic exhaustion and overburden (Lazarus, 1974).

Caregiver's burden was originally described in studies involving caregivers of people with dementia (Zarit, Reever, & Bach-Peterson, 1980), but it has also been also widely found in caregivers of people with long-term mental disorders and other chronic conditions, including ID (Kayadjanian, Schwartz, Farrar, Comtois, & Strong, 2018; Martín-Carrasco et al., 2016; Niemeier, Kreutzer, Marwitz, & Sima, 2018). This psychological construct has traditionally been approached from a bi-dimensional concept that contains two main factors: subjective burden (caregiver´s subjective assessment, feelings and attitudes) and objective burden (real events and activities) (Montgomery, Stull, & Borgatta, 1985; Vitaliano, Young, & Russo, 1991). Nevertheless, there is some evidence suggesting it can be analysed as a multidimensional construct (Ballesteros et al., 2012; Irazábal et al., 2012).

As far as ID is concerned, several studies have related higher caregivers' burden to different factors including presence of organic, affective, psychotic or behavioural disorders, severity of disability (functional level, social functioning and intelligence quotient), lack of support, or the caregiver's functionality level (Heller & Factor, 1991; Irazábal et al., 2012; Maes, Broekman, Dosen, & Nauts, 2003; Pinquart & Sörensen, 2006). In the same way, caregiver burden may results in negative consequences for the care recipient. Decreased caregiver´s psychological well-being is a predictor of lower quality of life among children with ID (Cramm & Nieboer, 2011) and could increase the risk of institutionalization (Essex, Seltzer, & Krauss, 1997; Miltiades & Pruchno, 2001).

These findings point at an important need to develop intervention strategies to tackle this problematic situation. A literature review reveals that, unlike other chronic mental illness (dementia or severe mental illness), there is a dearth of studies proposing psychosocial interventions to help caregivers of people with ID to acquire coping strategies, knowledge about the disease, support, or training in relaxation techniques (Magaña, Li, Miranda, & Paradiso de Sayu, 2015; McConkey, Kelly, & Craig, 2011). Most studies focus on improving the care recipient's situation through programs related to behaviour analysis techniques, cognitive rehabilitation, training, support, communication skills or physical activation (De Souza & Rehfeldt, 2013; Perez-Cruzado & Cuesta-Vargas, 2017; Reichow, Servili, Yasamy, Barbui, & Saxena, 2013). Studies related to improving caregivers' psychological well-being have put forth different psychosocial and educational interventions that have had mixed results related to depression, burden, self-efficacy and stress (Hassiotis et al., 2012; Hu et al., 2010; Magaña et al., 2015; Picard, Morin, & De Mondehare, 2014; Shahrier, Islam, & Debroy, 2016; Sofronoff, Jahnel, & Sanders, 2011; Yıldırım, Hacıhasanoğlu Aşılar, & Karakurt, 2013).

EDUCA project was developed with two main goals in mind: increasing knowledge of the caregiver burden and creating new psychoeducational interventions that could help informal caregivers of people with different chronic illnesses. These psychoeducational interventions have been created following the model proposed by REACH study, and their intervention Coping with Caregiving (CWC) (Wisniewski et al., 2003). The results obtained for caregivers of people diagnosed with dementia and schizophrenia have been favourable to intervention versus treatment as usual (Martín-Carrasco et al., 2009, Martín-Carrasco, Domínguez-

Panchón, González-Fraile, Muñoz-Hermoso, & Ballesteros, 2014; Martín-Carrasco et al., 2016).

The present study aims at assessing the efficacy of a standardised group psychoeducational intervention program (PIP) versus standard care (waiting list) on the reduction of caregivers' burden at the end of the trial (4 months from baseline), and at follow-up (8 months from baseline) for caregivers of people diagnosed with ID. Secondary objectives were to evaluate the efficacy of the PIP on caregivers' mental health, anxiety and depression.

2. Methods

2.1. Hypothesis and design

The principal hypothesis of the EDUCA-IV trial is that informal caregivers allocated to the PIP condition would show less burden at post-intervention endpoint (4 months), and at follow-up (8 months) than caregivers allocated to control condition (wait list). The secondary hypothesis was that caregivers allocated to PIP condition would also show a significant improvement in mental health conditions (well-being, depression, and anxiety) after intervention and at follow-up compared to caregivers allocated to control condition (wait list).

The study design was carried out as an interventionist, multicentre, single-blind, randomised controlled trial with individual randomisation to one of two conditions: PIP or standard care (waiting list). A total of 7 special educational research centres collaborated in the recruitment of the sample. Each research site has two independent researchers: one responsible for the application of the PIP, and another who assessed the outcomes and was blinded to the allocated intervention.

2.2. Ethics

The study was approved by the Ethical and Scientific Research Committees of Navarra (Project 2015/54), Spain, and by the Ethical Committee of each research centre. Participants were recruited via advertisements in research sites and by telephone calls to potential participants. First, they were informed of the aim of the study, the experimental design (randomization process), the potential benefits of the intervention, the voluntary participation and the freedom to dismiss participation. Both respondents (caregivers and care receivers) provided written informed consent forms before their inclusion and randomisation. If any participant with ID was legally incapacitated, the signature of the legal guardian was required.

2.3. Participants, inclusion and exclusion criteria

The study population included caregivers of people diagnosed with ID. To be eligible to participate in the study, caregivers had to be: (i) be men or women (of 18+ years); ii) care for a relative diagnosed with intellectual disability (DSM-V); (iii) be an informal caregiver (not paid); iv) spend a minimum of 4 h/week caring for their relative. In turn, the participants with ID should be residents in the community and receiving appropriate outpatient care.

Exclusion criteria were: (i) being caregivers without time to participate in the weekly intervention sessions; (ii) being caregivers currently receiving or having received in the last year any standardised intervention similar to the one administered in the trial; and (iii) being people with ID living in a residential care unit.

The selection of the centres was made according to the following criteria: i) schools or educational centres attended exclusively by people diagnosed with ID, ii) located in Spain, iii) possessing an educational program adapted and oriented to the special needs of their students, iv) not a residential care unit, v) interested in the development and implementation of the intervention in their centres. The participating centres were located in Spain (Galicia, Granada and Tenerife).

The following criteria for ending the trial before completion were established: (i) caregiver's decision, (ii) change from outpatient to inpatient status or residential care for the care receiver, and (iii) protocol deviations detected by the central research committee.

2.4. Interventions

Caregivers allocated to the intervention group received the treatment as usual, plus the PIP. The PIP was developed by a group of experts in mental health (psychiatrists and psychologists specialised in intellectual disabilities and developmental disorders) with on-hands knowledge in psychosocial interventions. It was formulated following the principles of the psychoeducational program developed by previous studies of the EDUCA project. It was adapted to the general needs of caregivers of people with intellectual disabilities and the special characteristics and difficulties related to their difficulties. Previously, 244 families had completed a survey on their opinions and interests in relation to their situation as caregivers. The answers helped us focus and better adapt the contents of the intervention. Caregivers showed a great interest in behavioural disturbances and problem-solving.

Caregivers received standardised information about the clinical course of the ID and were trained in cognitive and behavioural skills. These included general caregiving abilities, communication skills, the ability to seek and enjoy pleasant events, to seek support, as well as relaxation techniques. The PIP required active participation from caregivers (i.e. role playing, applying new skills to conflict, etcetera). It applied cognitive- behavioural techniques focused on identifying and challenging negative beliefs and developing a new style to deal with caregiving demands. It was administered over 12 weekly group sessions lasting 90–120 min each. Each session included a time-break of 15 min to prevent participant fatigue or inattention.

All sessions were delivered in the educational centres participating in the study (normal classrooms) in a group format and had

Table 1
Contents of the Psychoeducational Intervention Program (PIP).

Session	Content	Topics developed
1	Who are we?	Information about EDUCA project (aims and related issues). The real meaning of intellectual disabilities. The importance of our needs, care and self-care
2	My life has changed.	Adapting to change How it can affect to our daily living Evaluating our rights and compromises
3	Stress & well-being	Tension, emotion & stress Coping with stress Relaxation techniques
4	Take care of oneself	Effect of burden in the caregiver Learning to take care of oneself Changing routines
5	Importance of pleasant activities	Pleasant activities and mood Identifying & planning pleasant activities
6	Knowledge of support & helping services	Seeking support Seeking services
7	Improving my communication	How to talk with the patient How to talk with the health services Relaxation by mental distraction
8	Importance of thinking	Identifying beliefs & changing negative beliefs Muscular relaxation
9	Solving problems	Identifying and defining the problem Seeking solutions Choosing the solution Applying the solution Evaluation of results Relaxation by imaginary
10	Behaviour problems	ABC of behaviour How to manage behaviour problems To make a plan to change behaviour Relaxation by mental distraction
11	Approaching demanding situations	Improving the situation Strategies for managing behaviour Demanding situations Relaxation by imaginary
12	Planning the future	Worries about the future Assistance, health services & laws About the future Wrapping summary and program evaluation by the caregivers

the same formal structure: (i) an introduction to review the progress of the homework tasks, (ii) information about the topic to be dealt with, and (iii) exercises to practice the new knowledge or skills. The PIP was applied by mental health professionals (psychologists or psychiatrists) trained in the program by the same researchers who developed it. To ensure faithful delivery, they received a copy of the manual and special indications to follow and adhere to the contents. Caregivers received similar manual to follow the PIP throughout the trial. Table 1 presents the contents of the PIP by session.

In addition, the therapist manual included a checklist with the names of the participants to assess adherence to the intervention. Therapists scored the full assistance to each session. To improve the loyalty of the participants and to facilitate their attendance, sessions were delivered during class schedule and they received a notification reminding them of the day and hour of the next session. A full description of the methodology aspects related to the intervention application is provided following the TIDieR proposal (Hoffmann et al., 2014) in the appendix.

Caregivers allocated to the control group received the usual support from their outpatient centre, and had periodical interviews and information requests about the state and clinical course of the person diagnosed of ID.

2.5. Data collection and outcome assessment

Educa-IV trial included three visits conducted at baseline, post-intervention after finishing the PIP intervention (˜4 months since trial inception), and follow-up (˜8 months since trial inception). To avoid potentially being influenced by the researcher or similar biases, all outcomes were based on self-reported scales and were applied by a researcher blinded to intervention allocation.

2.6. Primary outcome measures

The main hypothesis was tested by the change scores since baseline to post-intervention endpoint (˜4 months) and follow-up endpoint (˜8 months) in the caregiver burden assessed by the Zarit Burden Interview (ZBI) (Zarit et al., 1980). It contains 22 items

graded in a 0–4 Likert scale (total score range from 0 to 88), that rates subjective caregiver burden. ZBI was originally developed to be used with caregivers of patients with dementia, but several studies have demonstrated its validity with caregivers of other pathologies including intellectual disability, palliative care, heart failure, brain injury or schizophrenia (Bhatia, Srivastava, Gautam, Saha, & Kaur, 2015; Goncalves-Pereira et al., 2017; Higginson, Gao, Jackson, Murray, & Harding, 2010; Martín-Carrasco et al., 2016; Naoki et al., 2017). ZBI was included in the assessment protocol due to its sensitivity to change (Chien & Lee, 2008; Martín-Carrasco et al., 2016). ZBI has been translated into Spanish and validated for Spanish populations (Martín-Carrasco et al., 2010).

2.7. Secondary outcome measures

Secondary hypothesis were tested using the change scores of three different psychological constructs: psychological distress with the General Health Questionnaire (GHQ-28) (Goldberg & Hillier, 1979), depression assessed by the Center for Epidemiologic Studies Depression Scale (CES-D) (Radloff, 1977) and anxiety using the State-Trait Anxiety Inventory (STAI) (Spielberger, Gorsuch, & Lushene, 1970). The GHQ-28 consists of 4 subscales (somatic symptoms, anxiety-insomnia, social dysfunction, and severe depression), including 7 items each. Scoring items present a 0–3 Likert scale. Total score and total subscale score range from 0 to 84 and 0 to 21 respectively. The higher score means a higher level of psychological distress; in turn, negative score changes mean beneficial effects. The GHQ-28 has been validated in Spanish (Lobo, Perez-Echeverria, & Artal, 1986). The CES-D was developed to measure depressive symptoms in the general population and includes 20-items that rate depressive feelings and related behaviours during the past week. Items are presented in a 0–3 Likert scale. The higher score means a higher level of depressive symptoms, thus negative score changes mean better outcomes. We used the Spanish versions of CES-D (Soler et al., 1997). The State-Trait Anxiety Inventory (form x) consists of 40 questions based on a 1-4-point Likert scale. Twenty questions are related to the current state of anxiety, and 20 to the trait of anxiety as a stable and personal characteristic. Range scores of each subtest vary from 20 to 80. The higher score indicates greater anxiety. The inventory has been validated for the Spanish population (Fonseca-Pedrero, Paino, Sierra-Baigrie, Lemos-Giraldez, & Muñiz, 2012).

We collected information about each care receiver and their adaptative behaviour through the Inventory for Client and Agency Planning (ICAP) (Bruininks, Hill, Weatherman, Woodcock, & DLM Teaching, 1986). This tool includes four scales (motor skills, social skills, personal life skills and community-living skills) and a general independence scale that provides information on services and support available to the individual. The Spanish version of the ICAP has good internal consistency (Cronbach alpha, 0.85) and high inter-rater reliability (Cohen kappa statistic, > 0.75) (Montero, 1996).

In addition, caregivers assigned to intervention arm responded to a brief qualitative questionnaire designed *ad hoc* to evaluate their degree of satisfaction with the program. The questionnaire included some items related to their individual opinion about the full program, the adequacy of the contents, the most interesting issues, difficulties or problems detected, and their desire to continue receiving similar interventions.

2.8. Sample size

Sample size was calculated assuming a baseline ZBI score of 22 with SD of 15, and a moderate effect size (standardised mean difference = 0.40) on the inter-group comparison in the ZBI scale. Calculations indicated a total sample size of 200 caregivers (randomised in 1:1) to attain 85% power with α level of 5%.

2.9. Randomization

The randomization was done individually for each research site. The sequence generation was obtained by block randomisation using block sizes of 1–4 using the R package "*blockrand*" (Snow, 2012). The process of the randomization was blinded for all the site researchers who participated in the study. The randomisation sequence was only reported to the researcher who developed the PIP. The researcher responsible for assessing the caregiver self-reported outcomes at visits #1 to #3 was blind to the allocation of the dyads.

2.10. Statistical methods

Continuous variables were described by means and SDs and category variables by frequencies and percentages. Main results were analysed by between-group differences in the change scores since baseline (visit #1) at post-intervention (visit #2) and at follow-up (visit #3). Two analyses were performed, one including only those caregivers with all available information [complete case analysis (CC)], whereas the other used an intention-to-treat analysis (ITT) with multiple imputations by chained equations (White, Royston, & Wood, 2011). Results are reported by mean differences, SDs and/or 95% confidence intervals (CI), and standardised effect sizes (SMD). Cohen's cut-off points were used to interpret SMD estimates: values ˜0.2 for a small effect, values ˜0.5 for a medium effect, and values ˜0.8 for a large effect (Cohen, 2013). We analysed the interaction of intervention and visits using a mixed linear model for repeated measures (MLM) including all visits. In addition to the main analysis, a subgroup analysis within the experimental group was conducted to find clinical, sociodemographic and methodological variables associated with the reduction of caregiver burden. The a priori selected variables were the ZBI and GHQ-28 scores at baseline, the number of PIP sessions attended by the caregivers, gender, age, educational level, marital status of the caregiver, the research centre, the time the caregiver was providing care as well as the weekly hours dedicated. After simple and multiple regression analysis, a backward stepwise linear regression analysis was

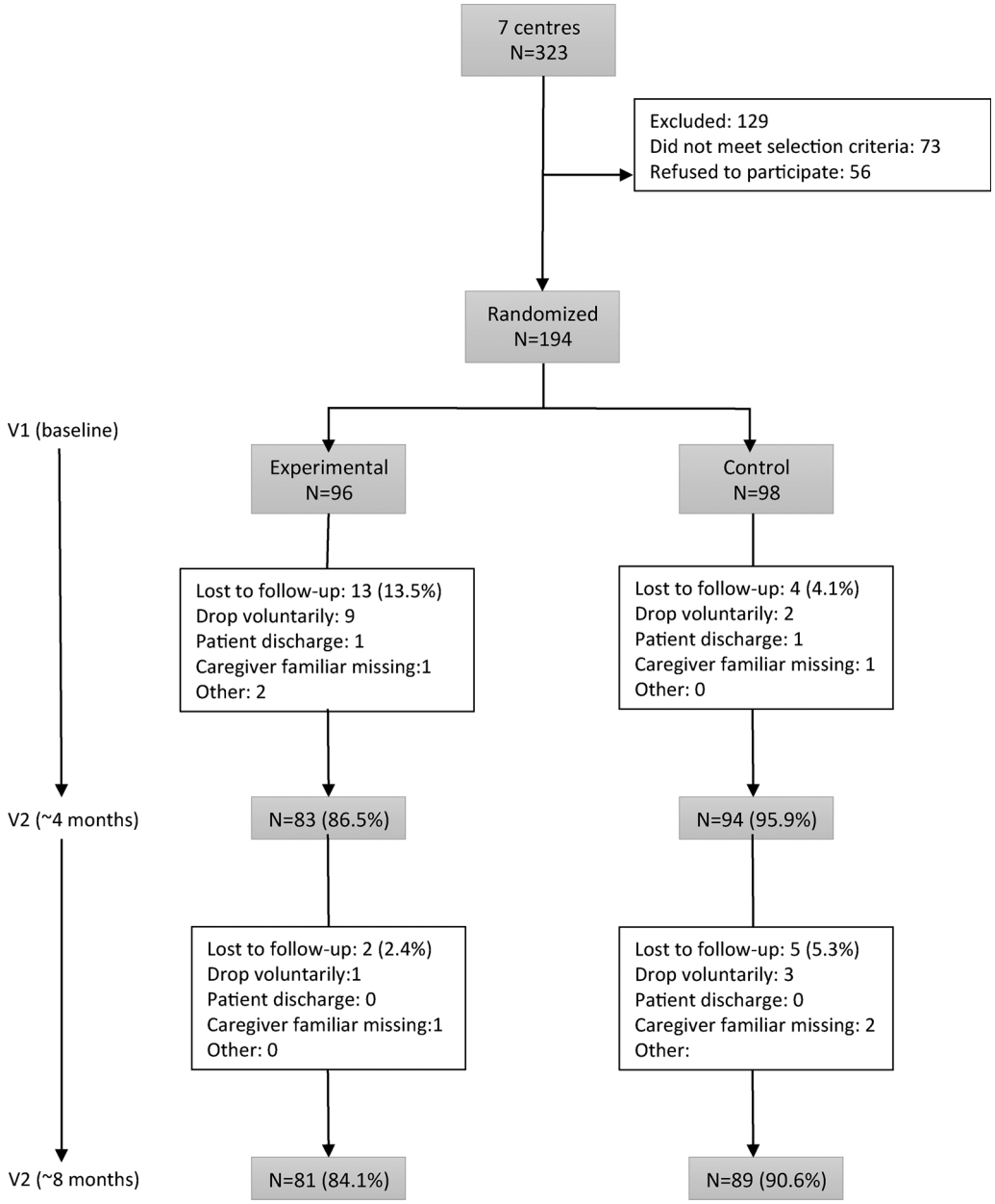

Fig. 1. Trial flowchart.

performed (P-value to retain of 0.10) to obtain the most parsimonious model able to explain the variability in the change of ZBI scores from baseline to end of trial at 8 months.

An independent and blinded statistician (MacCoun & Perlmutter, 2015) performed analyses using Stata v14 (StataCorp, College Station, TX, 2015) and R v3.3.3 (R Foundation for Statistical Computing, Vienna, Austria, 2017).

3. Results

3.1. Recruitment, losses to follow-up and fidelity

A total of 323 dyads were assessed for eligibility. 73 did not meet selection criteria, and 56 refused to participate. The remaining 194 caregivers were recruited to participate and randomised (96 to PIP and 98 to control condition), from October 2015 to January 2016. The last visit took place throughout the period June – September 2016. Fig. 1 shows the trial flowchart. Data from 7 research sites were included in the analysis. The median recruitment by research site was 27.7 caregivers (range from 9 to 73).

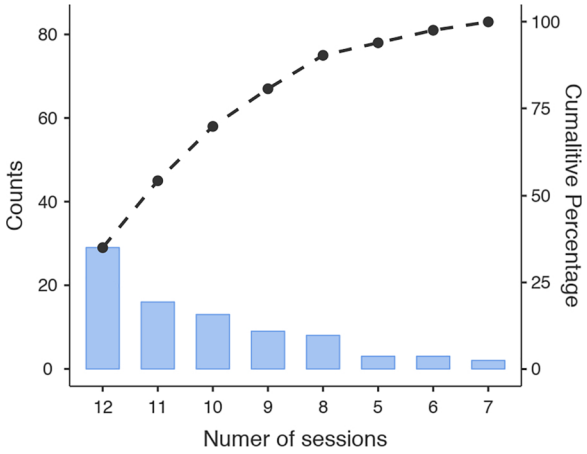

Fig. 2. Participants adherence to the program.

In the PIP group, 13 caregivers (13.5%) were lost to follow-up at visit #2. The corresponding figure in the control group was 4 (4.1%). The difference was significant (χ^2 test, $P = 0.021$). At visit #3, 2 additional caregivers in the PIP group were lost to follow-up (2.4%). In the control group 5 were lost (5.3%). The difference of drop-outs was not significant (Fisher exact test, $P = 0.27$).

Participants adherence to the program was high: 29 (34.93%) participants received the full program (12 sessions), 16 (19.27%) attended 11 sessions, 13 (15.66%) 10 sessions, and the rest of participants attended 9 or less sessions (Fig. 2). The average attendance was 10.19 sessions (SD = 1.94).

3.2. Sample description

The gender of the participants with ID was similar (50 [51.5%] were men and 47 [48.5%] women in the PIP and control arm respectively) and had a mean age of 26.3 years (SD = 12.5) in the PIP group and 28.23 years (SD = 12.4) in the control group. Most participants presented a primary diagnosis of Intellectual disability (86 [89.6%] in the PIP group and 92 [93.9%] in the control group). The rest had a primary diagnosis related to autism spectrum disorder conccurring with other neurodevelopmental disorders. Although most of them had good verbal communication skills (129 of the total sample [67.1%]), the majority had no access to writing or reading or had severe difficulties to read simple texts (79 [83.2%] in the PIP group and 72 [73.4%] in the control group).

ICAP scores showed a level of general independence corresponding to a functional age of 4 years and a general index of behavioural problems of -7.21 (DE = 10.55) that can be translated as “low severity”. Service score indicates that users needed a “frequent

Table 2
Sociodemographic description of the ID sample.

Variables	Total (N = 194)	PIP (n = 96)	Control (n = 98)	P value
Age, mean (SD)	27.26 (12.46)	26.31 (12.53)	28.15 (12.39)	0.333
Female gender, n (%)	97 (50)	50 (51.5)	47 (48.5)	0.566
Reading access, n (%)				
No access	115 (59.6)	60 (63.2)	55 (56.1)	
Simple	36 (18.7)	19 (20)	17 (17.3)	0.237
Normal	42 (21.7)	16 (16.9)	28 (26.5)	
Communication skills, n (%)				
None	36 (18.8)	19 (20.2)	17 (17.3)	
Alternative language	27 (14.1)	14 (14.9)	13 (13.3)	0.440
Normal	129 (67.1)	61 (64.9)	68 (69.4)	
Legally incapacitated, n (%)				
No	79 (41.4)	36 (37.9)	43 (44.8)	
Partial	3 (1.6)	3 (3.2)	0	0.157
Total	109 (57.1)	56 (58.9)	53 (55.2)	
ICAP, mean (SD)				
Motor skills	3.18 (2.31)	2.99 (2.17)	3.37 (2.44)	0.255
Social and communicative skills	3.80 (3.43)	3.47 (2.76)	4.13 (3.96)	0.181
Personal life skills	5.10 (4.10)	4.78 (3.17)	5.40 (4.85)	0.296
Community-living skills	4.34 (3.43)	4.10 (2.70)	4.58 (4.02)	0.335
General independence	4.04 (3.65)	3.75 (2.47)	4.33 (4.51)	0.278
GIBP	−7.21 (10.55)	−8.57 (12.04)	−5.89 (8.74	0.078
Service score	51.95 (21.06)	49.25 (22.39)	54.60 (19.42)	0.078

ICAP: Inventory for Client and Agency Planning; GIBP: General Index of Behaviour Problems.

Table 3
Sociodemographic and baseline clinical data of caregivers.

Variables	Total (N = 194)	PIP (N = 96)	Control (N = 98)	P value
Age, mean (SD)	55.58 (11.13)	54.48 (10.01)	56.61 (11.97)	0.210
Female gender, n (%)	178 (91.8)	93 (96.9)	85 (86.7)	0.010
Time of care (years), mean (SD)	24.27 (11.17)	22.88 (11.26)	25.60 (10.98)	0.093
Hours of care (weekly), n (%)				
15–21 h	4 (2)	–	4 (4)	0.261
22–28 h	4 (2.1)	2 (2.1)	2 (2)	
> 28 h	186 (95.9)	94 (97.6)	92 (93.9)	
Education level, n (%)				
Without degree	6 (3.1)	2 (2.1)	4 (4.1)	0.521
Primary	65 (33.5)	29 (30.2)	36 (36.7)	
Secundary (high school)	65 (33.5)	32 (33.3)	33 (33.7)	
College, university	58 (29.9)	33 (34.3)	25 (25.5)	
Marital status, n (%)				
Single	15 (7.7)	9 (9.4)	6 (6.1)	0.775
Married	128 (66)	62 (64.6)	66 (67.3)	
Separated/divorced	29 (14.9)	14 (14.6)	15 (15.3)	
Widow/er	22 (11.3)	11 (11.4)	11 (11.2)	
Relation to patient, n (%)				
Parent	170 (87.6)	83 (87.4)	87 (88.8)	0.534
Brother/sister	14 (7.2)	9 (9.5)	5 (5.1)	
Other	10 (5.1)	3 (3.1)	6 (6.1)	
External assistance provided, n (%)				
Professional	46 (25.6)	24 (27)	22 (24.2)	0.802
Financial	65 (36.1)	33 (37.1)	32 (35.2)	
None	69 (38.3)	32 (36)	37 (40.7)	
Working situation, n (%)				
Working	72 (37.1)	33 (34.4)	39 (39.8)	0.392
Unemployed	28 (14.4)	14 (14.6)	14 (14.3)	
Studing	1 (0.5)	1 (1)	–	
Housewife	81 (41.8)	44 (45.8)	37 (37.8)	
Retired	9 (4.6)	2 (2.1)	7 (7.1)	
Disabled	3 (1.5)	2 (2.1)	1 (1)	
ZBI score, mean (SD)	29.24 (15.15)	29.64 (15.81)	28.84 (14.56)	0.715
GHQ-28 score, mean (SD)	23.59 (11.86)	23.97 (11.67)	23.22 (12.10)	0.659
Somatic symptoms	6.81 (4.11)	6.9 (3.87)	6.73 (4.36)	0.773
Anxiety and insomnia	7.19 (4.33)	7.33 (4.4)	7.06 (4.27)	0.663
Social dysfuntion	7.10 (2.56)	7.15 (2.6)	7.05 (2.55)	0.776
Depression	2.47 (3.54)	2.58 (3.53)	2.37 (3.52)	0.687
CES-D score, mean (SD)	14.34 (11.01)	14.07 (11.02)	14.61 (11.06)	0.734
STAI score, mean (SD)				
State	19.37 (12.20)	10.17 (13.12)	18.59 (11.23)	0.367
Trait	19.67 (10.62)	20.40 (10.97)	18.94 (10.26)	0.341

ZBI: Zarit Burden Interview; GHQ-28: General Health Questionnaire-28 items; CES-D: Center for Epidemiologic Studies-Depression Scale; STAI: State-Trait Anxiety Inventory.

supervision". There were no statistical differences between groups (Table 2).

Table 3 presents the baseline characteristics of the caregivers by arm. Overall, caregivers were predominantly married women caring for their child, with a mean age of 55.58 years (SD = 11.13), with a low to medium level of educational, and presented a mean caring exposure of 24.27 years. The current caring load was higher than 28 h per week and more than a half received external help (professional or financial). The average score in the ZBI showed moderate burden (Stagg & Larner, 2015). The CES-D scores indicated that caregivers of both groups were below the cut-off (Vilagut, Forero, Barbaglia, & Alonso, 2016).

The comparison between those participants who dropped out versus completers did not reveal significant demographic or clinical baseline differences.

3.3. Primary outcomes

Table 4 shows the ZBI differences among interventions through time by complete cases (CC). The CC analyses for the ZBI were not significant at 4 and 8 months since trial inception (SMD = −0.09 and −0.01). The MLM analysis did not reveal a significant interaction of intervention arm by time ($P = 0.663$). ITT analyses (Table 5) were similar, presenting null effects at 4 months (SMD = −0.09) and 8 months (SMD = −0.00).

Table 4
Efficacy Outcomes (change scores from baseline). Complete Cases Analyses and Mixed Linear Model for Repeated Measures.

	Visit 2 (4 months since baseline)				Visit 3 (8 months since baseline)				
Outcomes	PIP (N = 83)	Control (N = 94)	Mean Difference (95% CI)	SMD (95% CI)	PIP (N = 81)	Control (N = 89)	Mean Difference (95% CI)	SMD (95% CI)	*p*-values*
Zarit Total Score, mean (SD)	−3.18 (13.74)	−2.12 (10.02)	1.05 (−2.489;4.59)	0.09 (−0.21;0.34)	−1.25 (19.50)	−1.11 (19.54)	0.15 (−5.77;6.06)	0.01 (−0.29;0.31)	0.663
GHQ-28 Total Score, mean (SD)	−6.54 (13.68)	−1.78 (8.96)	4.75 (1.35;8.15)	0.42 (0.10;0.70)	−6.35 (13.88)	−0.97 (14.17)	5.38 (1.12;9.63)	0.38 (0.07;0.68)	0.036
GHQ-28 Somatic Symptoms	−2.09 (5.04)	−0.45 (3.76)	1.64 (0.32;2.94)	0.37 (0.06;0.66)	−2.01 (5.07)	0.07 (5.77)	2.09 (0.43;3.74)	0.38 (0.08;0.69)	0.026
GHQ-28 Anxiety & Insomnia	−2.19 (4.73)	−0.8 (3.6)	1.38 (0.14;2.62)	0.33 (0.02;0.62)	−2.17 (5.21)	−0.44 (5.19)	1.72 (0.14;3.30)	0.33 (0.02;0.63)	0.039
GHQ-28 Social Dysfunction	−1.42 (3.63)	−0.09 (2.56)	1.33 (0.4;2.25)	0.43 (0.11;0.71)	−1.01 (2.95)	−0.24 (3.16)	0.77 (−0.16;1.69)	0.25 (−0.05;0.55)	0.275
GHQ-28 Depression	−0.83 (2.86)	−0.42 (2.33)	0.41 (−0.36;1.17)	0.16 (−0.14;0.45)	−1.16 (4.07)	−0.35 (4.35)	0.8 (−0.48;2.08)	0.19 (−0.11;0.49)	0.730
CES-D Total Score, mean (SD)	0.62 (10.88)	1.41 (8.93)	0.79 (−2.15;3.73)	0.08 (−0.21;0.37)	−4.25 (14.37)	2.14 (12.12)	6.41 (2.39;10.42)	0.48 (0.17;0.78)	0.230
STAI, mean (SD)									
STAI-State	−3.73 (14.34)	−5.40 (12.35)	1.67 (−2.29;5.62)	0.13 (−0.17;0.42)	−2.35 (12.65)	0.32 (13.95)	2.68 (−1.36;6.73)	0.20 (−0.10;0.50)	0.413
STAI-Trait	−2.73 (10.19)	−5.19 (10)	2.42 (−0.54;5.45)	0.24 (−0.05;0.54)	−1.83 (12.24)	0.53 (13.37)	2.38 (−1.51;6.27)	0.19 (−0.11;0.48)	0.255

PIP: Psychoeducational Intervention Program; SMD: Standardized Mean Difference; CES-D: Center for Epidemiologic Studies – Depression Scale; GHQ-28: General Health Questionnaire Scaled (28 items).
* *p*-Values for the interaction of intervention by time from the mixed linear model for repeated measures.

Table 5
Efficacy Outcomes (change scores from baseline). Intention to treat analyses and Mixed Linear Model for Repeated Measures.

	Visit 2 (4 months since baseline)				Visit 3 (8 months since baseline)				
Outcomes	PIP (N = 96)	Control (N = 98)	Mean Difference (95% CI)	SMD (95% CI)	PIP (N = 96)	Control (N = 98)	Mean Difference (95% CI)	SMD (95% CI)	*p*-values*
Zarit Total Score, mean (SD)	−2.74 (12.82)	−1.96 (9.87)	0.77 (−2.465;4.01)	0.06 (−0.21;0.35)	−1.05 (17.81)	−1.02 (18.71)	0.03 (−5.14;5.20)	0.00 (−0.28;0.28)	0.793
GHQ-28 Total Score, mean (SD)	−5.66 (12.91)	−1.79 (8.81)	3.87 (0.747;6.99)	0.35 (0.07;0.63)	−5.03 (12.89)	−0.89 (13.57)	4.41 (0.66;8.16)	0.31 (0.03;0.59)	0.044
GHQ-28 Somatic Symptoms	−1.80 (4.74)	−0.45 (3.68)	1.35 (0.15;2.55)	0.31 (0.03;0.60)	−1.68 (4.69)	−0.07 (5.52)	1.75 (0.3;3.20)	0.31 (0.03;0.60)	0.031
GHQ-28 Anxiety & Insomnia	−1.86 (4.48)	−0.82 (3.56)	1.03 (−0.10;2.18)	0.25 (0.01;0.54)	−1.81 (4.83)	−0.41 (4.97)	1.40 (0.12;2.79)	0.28 (0.00;0.57)	0.047
GHQ-28 Social Dysfunction	−1.25 (3.41)	−0.112 (2.52)	1.13 (0.28;1.99)	0.37 (0.10;0.66)	−0.84 (2.72)	−0.21 (3.03)	0.61 (−0.19;1.43)	0.22 (−0.06;0.50)	0.345
GHQ-28 Depression	−0.75 (2.69)	−0.40 (2.28)	0.34 (−0.36;1.05)	0.13 (−0.14;0.42)	−0.96 (3.74)	−0.32 (4.17)	0.63 (−0.48;1.76)	0.16 (−0.12;0.44)	0.762
CES-D Total Score, mean (SD)	0.57 (10.12)	1.43 (8.78)	0.86 (−1.81;3.55)	0.09 (−0.37;0.19)	−3.55 (13.21)	1.96 (11.62)	5.25 (2;9.05)	0.13 (−0.15;0.41)	0.292
STAI, mean (SD)									
STAI-State	−3.38 (13.43)	−5.15 (12.16)	1.76 (−1.85;5.39)	0.13 (−0.42;0.14)	−2.96 (11.58)	0.29 (13.35)	2.26 (−1.26;5.8)	0.21 (−0.07;0.49)	0.501
STAI-Trait	−2.35 (9.53)	−5.10 (9.86)	2.74 (−0.001;5.49)	0.28 (−0.57;0.00)	−1.53 (11.19)	0.49 (12.80)	2.03 (−1.37;5.43)	0.09 (−0.20;0.37)	0.293

PIP: Psychoeducational Intervention Program; SMD: Standardized Mean Difference; CES-D: Center for Epidemiologic Studies – Depression Scale; GHQ-28: General Health Questionnaire Scaled (28 items).
* *p*-Values for the interaction of intervention by time from the mixed linear model for repeated measures.

3.4. Secondary outcomes

Tables 4 and 5 also present the GHQ-28, CES-D and STAI differences among interventions and visits. For the GHQ-28 (total score) the analysis showed a significant effect favouring the PIP with a small-medium size at 4 (CC, SMD = −0.42; ITT, SMD = −0.35) and 8 months (CC, SMD = −0.38; ITT, SMD = −0.31). The MLM analysis displayed a significant interaction of intervention arm by time (CC, $P = 0.03$; ITT, P = 0.044). The subscales of somatic symptoms and anxiety and insomnia showed positive and significant results at 4 and 8 months in CC and ITT analyses. Social dysfunction subscale found a medium effect size at 4 months (CC, SMD = 0.43; ITT SMD = 0.37), that decreased at 8 months (CC, SMD = 0.25; ITT, SMD = 0.22). MLM analysis for this subscale indicates no significant effects of intervention arm by time (CC, $P = 0.27$; ITT P = 0.345). There was no significant impact on the depression subscale at any time.

The analysis for the CES-D at 4 months showed a small increase in scores (higher depression) in both groups. Comparisons by arm did not detect significant differences (CC, SMD = 0.08; CI 95% [−0.21 to 0.37]; ITT, SMD = 0.09; CI 95% [−0.37 to 0.19]). Nevertheless, at 8 months the PIP showed an important decrease in depression while it remained in the control group. These results were significant for CC, but not for the ITT approach (CC, SMD = 0.48; CI 95% [0.17–0.78]; ITT, SMD = 0.13; CI 95% [0.15–0.41];). The MLM analyses were not significant (CC, $P = 0.23$; ITT, P = 0.29).

Results for the STAI-State subscale showed a great reduction in both groups at 4 months that disappeared at 8 months. The MLM analyses were not significant (CC, $P = 0.413$; ITT, P = 0.501). STAI-Trait subscale showed small and no significant effect sizes at 4 months (CC, SMD = 0.24; ITT, SMD = 0.28) and at 8 months (CC, SMD = 0.19; ITT, SMD = 0.09).

3.5. Subgroup analysis

Only the baseline ZBI score was significantly associated with the improvement in ZBI. The ZBI at baseline is positively associated with the reduction of burden, which means that those caregivers experiencing more burden at baseline will also be also those who will experience greater improvement by the end of trial (0.19 [95% CI: 0.15–0.38]).

The qualitative questionnaire designed to evaluate the degree of satisfaction of the caregivers yielded interesting results. The 94.3% of the respondents indicated that the program was “very interesting”, while the rest answered the question as “interesting”. The best-rated contents were those related to relaxation techniques (41.09%), communication role-playings (31.5%), and active training to cope with the behavioural problems (16.43%). Caregivers expressed that the programme had helped them better understand some difficult situations, the link between thoughts and emotions, and the importance of taking care of oneself to be able to take better care of others. Some of the detected problems were the extension and complexity of some contents, the difficulty to maintain adherence and attendance to all sessions and homework. In general, program acceptance was high and the entire sample was interested in continuing to receive similar interventions.

4. Discussion

Parents providing ongoing care to people with intellectual disabilities are found to be at risk of adverse mental health outcomes such as burden, distress, depression or anxiety. The current study endorses this problem (baseline scores) and has tested the efficacy of a psychoeducational intervention in a sample of informal caregivers of people with intellectual disabilities. According to our results, we cannot confirm the primary hypothesis related to the efficacy of our intervention.

Measures in the secondary outcomes suggested that PIP intervention is useful for reducing distress, somatic symptoms, social dysfunction, and anxiety and insomnia (GHQ-28). Depression assessed by GHQ-28 and CES-D threw mixed results. These differences could be explained because GHQ depression items assess severe and likely unchangeable symptoms, and the CES-D items include other more changeful dominions (i.e. motor and physiological symptoms). Anxiety change scores (STAI) were small and not significant. ITT analyses approach showed similar but more conservative results. Focusing on the caregivers who had the PIP intervention (subgroup analysis), the result was the same as in a previous study with caregivers of people with schizophrenia (Martín-Carrasco et al., 2016), where those caregivers with higher levels of burden at the beginning of the intervention were more likely to reduce their burden.

Our results are in accordance with other studies following similar interventions and design (randomised controlled trials). Studies that assessed the efficacy of psychological interventions on caregiver´s burden found positive but no significant (Bilgin & Gozum, 2009; Hassiotis et al., 2012; Magaña et al., 2015) and those authors who used depression questionnaires obtained mixed results (Isabelle, Diane, & Laurence, 2014; Magaña et al., 2015; Sofronoff et al., 2011; Yıldırım et al., 2013). Only two studies assessed the effects on anxiety without significant results (Isabelle et al., 2014; Sofronoff et al., 2011). In this sense, we have run online searches in different scientific databases confirming an important lack of relevant evidence. There are few studies that apply interventions focused on the caregiver´s needs and those that are published have small sample sizes.

4.1. Limitations and strengths of the research

Despite our efforts to control methodological aspects related to the validity (internal consistency) of the study, the latter must be

considered within the context of several limitations. First, in this kind of studies with non-pharmacological interventions it is very difficult to mask and blind participants and professionals. It would have been desirable to have an active control condition to blind them. Maybe it may have affected the self-perception of the caregivers and the answers to the questions posed in the assessment questionnaires. Second, the experimental design (randomisation process) and the length of the study produced an initial attrition bias (56 participants refused to participate before the study started). Maybe those caregivers under worse conditions (lack of time or support, or with high levels of distress) refused to participate and are not represented in this study. Once the study started, 24 recruited participants dropped out the study. Despite the fact that we did not find differences between completers and non-completers at baseline, the majority of drop outs (62.5%) occurred in the experimental arm during the intervention period. Possibly, a shorter programme or a different format of delivery (individually or online) would have been desirable in order to avoid this attrition bias and increase adherence. We aimed at controlling attrition bias by using intention-to-treat analysis but this led to very similar results. In addition, small sample size (less than calculated in previous analysis) could impact the representation of the entire population of parents of children with ID.

Third, caregivers participanting in of this study (those randomised to experimental or control arm) were receiving usual attention in their centres during their participation in this study. This attention included quarterly meetings with the staff, regular follow-up and management of each case, and access to professionals to ask relevant questions related to their relatives. It would have been interesting to have recorded this information in order to avoid an overlap of the programme across the two groups. This situation could annul the differences between groups or a ceiling effect where there was less possibility of improving usual attention.

Fourth, the group format and the extension of the contents could have produced a limiting effect. Despite our desire to apply the intervention in an individual format and to extend the contents to produce a lasting effect, there were some limitations related to the usual clinical practice that make it impossible.

Notwithstanding these limitations, this study presents a psychoeducational program intervention that is in accordance with current guidelines (Catalano, Holloway, & Mpofu, 2018) and that had demonstrated its efficacy to improve the mental health of caregivers of people with intellectual disabilities. The empirical results of this study and the limitations detected need to be added to the current evidence to develop new strategies for helping parents of children with intellectual disabilities or related problems.

5. Conclusions

Our study shows that informal caregivers of people with intellectual disabilities may present negative consequences that affect their mental health (burden, distress, risk of depression and anxiety) and should receive assistance, especially those with a higher burden. We developed a psychoeducational intervention (12 group weekly sessions) to improve their psychological wellbeing. Using an experimental methodology, we found positive results on general mental health but not on caregiver burden, depression or anxiety. The intervention was well tolerated and valued by caregivers. New strategies related to sample participation (recruitment, retention, attendance), implementation of the intervention (format of delivery, adherence and intensity), and evaluation of the results (acceptability, variables associated with the reduction on the caregiver burden) must be further explored to obtain better results.

Declaration of Competing Interest

None.

Acknowledgements

The EDUCA-IV trial was funded by Sisters Hospitallers. The role of the funding source was limited to economic support. It was not involved in the study process, preparation or submission of the manuscript.

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Appendix A

Table A1

Table A1
Information to include when describing an intervention and the location of the information.

Item number	
	BRIEF NAME
1.	**Provide the name or a phrase that describes the intervention.** PIP (Psychoeducational Intervention Program)
	WHY
2.	**Describe any rationale, theory, or goal of the elements essential to the intervention.** It is based on the theoretical model of the stress of the caregiver defined by Lazarus and subsequently completed by Pearlin, that puts emphasis on the weight of environmental variables, on the resources and in the supports received to explain the overload and to guide prevention.
	WHAT
3.	**Materials: Describe any physical or informational materials used in the intervention, including those provided to participants or used in intervention delivery or in training of intervention providers. Provide information on where the materials can be accessed (e.g. online appendix, URL).** Intervention was applied through a printed manual with the contents of the intervention. The therapist has his own manual with the contents and the answers of the proposed activities and exercises. The participants had the same manual but without the answers. These manuals can be accessed contacting with the corresponding author. The sessions took place in standard rooms adapted with teaching elements (dashboard, chalks and chairs for the assistants). The therapist training was two months before the study starts in the same way.
4.	**Procedures: Describe each of the procedures, activities, and/or processes used in the intervention, including any enabling or support activities.** PIP included general caregiving abilities, communication skills, the ability to seek and enjoy pleasant events, to seek support, as well as relaxation techniques. The PIP required active participation from caregivers (i.e. role playing, applying new skills to conflict, etcetera). It applied cognitive-behavioural techniques focused on identifying and challenging negative beliefs and developing a new style to deal with caregiving demands. It was administered over 12 weekly group sessions lasting 90-120 minutes each. Each session included a time-break of 15 minutes to avoid fatigue or inattention. All sessions had the same formal structure: (i) an introduction to review the progress of the homework tasks, (ii) information about the topic to be deal with, and (iii) exercises to practice the new knowledge or skills. Session titles and contents are described below: 1. Who are we? Information about EDUCA project (aims and related issues). The real meaning of intellectual disabilities. The importance of our needs, care and self-care 2. My life has changed: Adapting to change. How it can affect to our daily living. Evaluating our rights and compromises. 3. Stress & well-being: Tension, emotion & stress. Coping with stress. Relaxation techniques. 4. Take care of oneself: Effect of burden in the caregiver. Learning to take care of oneself. Changing routines. 5. Importance of pleasant activities: Pleasant activities and mood. Identifying & planning pleasant activities. 6. Knowledge of support & helping services: Seeking support. Seeking services. 7. Improving my communication: How to talk with the person with ID. How to talk with the health services. Relaxation by mental distraction. 8. Importance of thinking: Identifying beliefs & changing negative beliefs. Muscular relaxation. 9. Solving problems: Identifying and defining the problem. Seeking solutions. Choosing the solution. Applying the solution. Evaluation of results. Relaxation by imaginary. 10. Behavior problems: ABC of behavior. How to manage behavior problems. To make a plan to change behavior. Relaxation by mental distraction. 11. Approaching demanding situations: Improving the situation. Strategies for managing behavior. Demanding situations. Relaxation by imaginary 12. Planning the future: Worries about the future. Assistance, health services & laws. About the future. Wrapping summary and program evaluation by the caregivers
	WHO PROVIDED
5.	**For each category of intervention provider (e.g. psychologist, nursing assistant), describe their expertise, background and any specific training given.** The PIP was provided by mental health professionals (psychologist or psychiatrist) trained in the program by the same researchers who developed the PIP.
	HOW
6.	**Describe the modes of delivery (e.g. face-to-face or by some other mechanism, such as internet or telephone) of the intervention and whether it was provided individually or in a group.** Face-to-face in group format.
	WHERE
7.	**Describe the type(s) of location(s) where the intervention occurred, including any necessary infrastructure or relevant features.** The intervention took place in the same research centres in the classrooms of each centre.
	WHEN and HOW MUCH
8.	**Describe the number of times the intervention was delivered and over what period of time including the number of sessions, their schedule, and their duration, intensity or dose.** The intervention was composed by 12 sessions of 90-120 minutes applied weekly. The full program needs a 3 months to be applied.
	TAILORING
9.	**If the intervention was planned to be personalised, titrated or adapted, then describe what, why, when, and how.** To avoid possible bias related to the therapist (different ways to apply it or personal interpretations), the intervention was very manualized and applied following the instructions. The therapist had to follow the contents and explanations described in the manuals. This issue was mentioned during the training period.
	MODIFICATIONS

(*continued on next page*)

Table A1 (*continued*)

Item number	
10.	**If the intervention was modified during the course of the study, describe the changes (what, why, when, and how).** No. The intervention did not suffer any modifications during the course of the study. **HOW WELL**
11.	**Planned: If intervention adherence or fidelity was assessed, describe how and by whom, and if any strategies were used to maintain or improve fidelity, describe them.** The intervention adherence was assessed including a checklist with the names of the participants in the therapist manual. To improve the fidelity of the participants, they received a notification reminding them the hour and day of the next session.
12.	**Actual: If intervention adherence or fidelity was assessed, describe the extent to which the intervention was delivered as planned.** Participants adherence to the program was high: 29 (34.93%) participants received the full program (12 sessions), 16 (19.27%) attended 11 sessions, 13 (15.66%) 10 sessions, The rest of participants attended 9 or less sessions (Figure 2). The average attendance was 10.19 sessions (SD = 1.94).

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