OFFICIAL JOURNAL OF THE ITALIAN SOCIETY OF PSYCHOPATHOLOGY

Journal of PSYCHOPATHOLOGY

Editor-in-chief: Alessandro Rossi

VOL. 27 - 2021

NUMBER 2

Cited in: EMBASE - Excerpta Medica Database ▪ Index Copernicus ▪ PsycINFO ▪ SCOPUS ▪ Google Scholar ▪ Emerging Sources Citation Index (ESCI), a new edition of Web of Science

CLARIVATE ANALYTICS EMERGING SOURCES CITATION INDEX INDEXED

SOPSI SOCIETÀ ITALIANA DI PSICOPATOLOGIA

www.jpsychopathol.it

PACINI EDITORE MEDICINA

Periodico trimestrale POSTE ITALIANE SpA - Spedizione in Abbonamento Postale - D.L. 353/2003 conv.in L.27/02/2004 n°46 art.1, comma 1, DCB PISA - Aut. Trib. di Pisa n. 9 del 03/06/95 - June - ISSN 2284-0249 (Print) ISSN 2499-6904 (Online)

OFFICIAL JOURNAL OF THE ITALIAN SOCIETY OF PSYCHOPATHOLOGY

Journal of
PSYCHOPATHOLOGY

Editor-in-chief: *Alessandro Rossi*

International Editorial Board
D. Baldwin (University of Southampton, UK)
D. Bhugra (Emeritus Professor, King's College, London, UK)
J.M. Cyranowski (University of Pittsburgh Medical Center, USA)
V. De Luca (University of Toronto, Canada)
B. Dell'Osso ("Luigi Sacco" Hospital, University of Milan, Italy)
A. Fagiolini (University of Siena, Italy)
N. Fineberg (University of Hertfordshire, Hatfield, UK)
A. Fiorillo (University of Campania "Luigi Vanvitelli", Naples, Italy)
B. Forresi (Sigmund Freud Privat Universität Wien GmbH, Milan, Italy)
T. Ketter (Professor Emeritus, Stanford University, Stanford, USA)
G. Maina (University "San Luigi Gonzaga", Turin, Italy)
V. Manicavasagar (Black Dog Institute, Randwick, Australia)
P. Monteleone (University of Salerno, Italy)
D. Mueller (University of Toronto, Canada)
S. Pallanti (Stanford University Medical Center, USA)
C. Pariante (King's College, London, UK)
J. Parnas (University of Copenhagen, Denmark)
S. Pini (University of Pisa, Italy)
P. Rucci ("Alma Mater Studiorum" University of Bologna, Italy)
N. Sartorius (Emeritus Professor, University of Geneva, Switzerland)
J. Treasure (King's College, London, UK)

Advisory Board
E. Aguglia (University of Catania, Italy)
M. Amore (University of Genoa, IRCCS Ospedale Policlinico San Martino, Genoa, Italy)
L. Bellodi (Ospedale San Raffaele, Milan, Italy)
A. Bertolino (University of Bari "Aldo Moro", Azienda Ospedaliero Universitaria Consorziale Policlinico, Bari, Italy)
M. Biondi (Sapienza University of Rome, Italy)
B. Carpiniello (University of Cagliari, Italy)
F. Catapano (University of Campania "Luigi Vanvitelli", Naples, Italy)
D. De Ronchi (University of Bologna, Italy)
L. Dell'Osso (University of Pisa, Italy)
M. Di Giannantonio (University of Chieti, Italy)
A. Favaro (University of Padova, Italy)
S. Galderisi, University of Campania "Luigi Vanvitelli", Naples, Italy)
D. La Barbera (University of Palermo, Italy)
M. Maj (University of Campania "Luigi Vanvitelli", Naples, Italy)
P. Rocca (University of Turin, Italy)
R. Roncone (University of L'Aquila, Italy)
A. Rossi (University of L'Aquila, Italy)
A. Siracusano (University of Rome Tor Vergata, Italy)
A. Vita (ASST Spedali Civili, Brescia, Italy)

Italian Society of Psychopathology
Executive Council
President: Eugenio Aguglia • *Past President:* Alessandro Rossi
President Elect: Silvana Galderisi • *Secretary:* Andrea Fagiolini • *Treasurer:* Paola Rocca
Councillors: Mario Amore, Bernardo Carpiniello, Massimo Di Giannantonio, Alberto Siracusano, Massimo Biondi, Cristina Colombo, Palmiero Monteleone, Antonio Vita

Founders: Giovanni B. Cassano, Paolo Pancheri

Honorary Councillors: Giovanni B. Cassano, Luigi Ravizza

Editorial Coordinator: Francesca Pacitti

Editorial Staff: Roberto Brugnoli, Milena Mancini

Editorial and Scientific Secretariat: Valentina Barberi, Pacini Editore Srl, Via Gherardesca 1, 56121 Pisa • Tel. 050 3130376 • Fax 050 3130300 • journal@jpsychopathol.it

© Copyright by Pacini Editore Srl

Publisher: Pacini Editore Srl, Via Gherardesca 1, 56121 Pisa • www.pacinimedicina.it

VOL. 27 - 2021
NUMBER 2

Cited in:
EMBASE - Excerpta Medica Database ▪ Index Copernicus
PsycINFO ▪ SCOPUS ▪ Google Scholar ▪ Emerging Sources Citation Index (ESCI), a new edition of Web of Science

SOPSI
SOCIETÀ ITALIANA DI PSICOPATOLOGIA

PACINI
EDITORE
MEDICINA

www.jpsychopathol.it

Contents

Original articles

Case Report

JOURNAL OF PSYCHOPATHOLOGY 2021;27:71-80
DOI: 10.36148/2284-0249-346

Original article

Implementing individual and placement support for patients with severe mental illness: findings from the real world

Maria Lorena Ficarelli[1], Elisa Troisi[2], Elisabetta Vignali[1], Simona Artoni[1], Maria Cristina Franzini[1], Serenella Montanaro[1], Maria Vittoria Andreoli[1], Sara Marangoni[3], Elizabeth Ciampà[3], Diana Erlicher[3], Simona Pupo[4,5], Lorenzo Pelizza[1,6]

[1] Department of Mental Health and Pathological Addiction, Azienda USL-IRCCS di Reggio Emilia, Italy; [2] School of Psychiatric Rehabilitation Techniques, Università di Modena e Reggio Emilia, Italy; [3] E.N.A.I.P. Foundation, Reggio Emilia, Italy; [4] Intensive Care Unit, Guastalla Civil Hospital, Azienda USL-IRCCS di Reggio Emilia, Italy; [5] Anesthesia and Resuscitation Service, Azienda Ospedaliero-Universitaria di Parma, Italy; [6] Department of Mental Health and Pathological Addiction, Azienda USL di Parma, Italy

Received: August 19, 2019
Accepted: March 1, 2021

Correspondence
Lorenzo Pelizza
c/o Centro "Santi", via Vasari 13, 43100 Parma, Italy. Tel. +39 0521 396512. Fax +39 0521 293482. E-mail: lorpelizza@ausl.pr.it

Conflict of interest
The Authors declare no conflict of interest

How to cite this article: Ficarelli ML, Troisi E, Vignali E, et al. Implementing individual and placement support for patients with severe mental illness: findings from the real world. Journal of Psychopathology 2021;27:71-80. https://doi.org/10.36148/2284-0249-346

SUMMARY

Objective

Individual Placement and Support (IPS) is a psychosocial intervention with a considerable body of evidence for its effectiveness in helping people with Severe Mental Illness (SMI) to obtain and maintain competitive job. However, little is known about IPS model in Italy, a country with a different socioeconomic climate than the USA and the UK. Aim of this study was to investigate the effect of IPS in Italian patients with SMI, assessing the main competitive employment outcomes and drop-out rates during a 3-year follow-up period.

Methods

Participants (n = 46) were recruited from the 7 adult Community Mental Health Centers (CMHCs) of the Reggio Emilia Department of Mental Health. Together with drop-out rates, we examined job acquisition (employment in the labor market for at least 1 day during the follow-up), job duration (total number of days worked), job tenure (weeks worked on the longest-held competitive job), and total hours per week worked.

Results

A crude competitive employment rate of 50% and a crude drop-out rate of 34.8 over the 3-year follow-up period were found. Using a Kaplan-Meyer survival analysis, the cumulative employment rate reached a 73% percentage at 36 months.

Conclusions

This study shows the feasibility and the utility of an implementation strategy for applying the IPS approach in the public mental health care system in Italy.

Key words: supported employment, individual placement and support, vocational rehabilitation, psychiatric rehabilitation, mental health services, outcomes

Introduction

Despite most individuals suffering from Severe Mental Illness (SMI) (i.e. schizophrenia and bipolar disorder) would actually like to work in the competitive labor market, this condition induces a significantly higher risk of unemployment in adults of working age than other disabilities [1]. Indeed, only 15% of people affected by SMI are regularly employed in the competitive employment world [2].

Individual Placement and Support (IPS) is a psychosocial intervention to help people with SMI in achieving and maintaining competitive jobs in the labor market[3]. IPS has proven to be an evidence-based practice, showing a higher effectiveness over other vocational rehabilitation approaches [4]. In this respect, a selective meta-analysis of 15 randomized controlled trials found an overall employment rate of 55% for people receiving IPS compared to 23% for controls [5].

IPS in Italy

Since the seminal work of Basaglia in the sixties, programs aimed at employment have always been considered hallmarks of good practice in Italian community psychiatry [6]. They mostly include traditional "train and place" approaches provided in different settings, such as sheltered workshops and/or training stages by public/private employers or by social enterprises, known as cooperatives [4].

The Italian system also incorporates the statutory provision of law 68/1999, establishing a "quota of working places" for disabled citizens by private or public employers. Usually, all these activities are associated with temporary employment grants (generally a sum of approximately 4 euro/hour) that are flexible and sometimes rapid instruments, but they are overprotective and quite stigmatizing, leave little choice to users in the type of occupation or work, and often keep clients out of competitive employment for a long time [7].

The pervasive economic crisis and a clearer awareness of personal rights have boosted the demand for employment services by people with SMI. During the past ten years, the number of individuals who entered traditional vocational rehabilitation programs in Italy almost doubled, despite the rate of people entering competitive jobs halved from 10 to 5% [4]. Thus, Italian mental health services became interested in innovative interventions, such as IPS.

After the EQOLISE trial (that was the first European trial on effectiveness of IPS [in which Rimini was one of the six European sites involved] confirming the excellent findings of US researches, despite ample differences in culture of psychiatric services and in labor market regulations), Emilia-Romagna Region put IPS in its policy and financed a program for its implementation in all regional department of mental health. In 2014, 20 of out 41 Community Mental Health Centers (CMHC) in the region began offering IPS to their users [4]. Among them, there were all of the 7 adult CMHCs of the Reggio Emilia Department of Mental Health.

Because the EQOLISE study was not powered to test the IPS effectiveness for the separate countries, new evidence is needed, particularly in European countries as Italy, where, differently to the US socioeconomic climate, labor market is less flexible, there is a stronger social security system (for which labor and disability policies can impede returns to work) [8], and the employment opportunities are rather limited [4]. Indeed, it has been widely reported that IPS effectiveness was restrained in those countries where unemployment benefits offer a more secure income than potential jobs [7].

Starting to this background, *aim* of the current study was two-fold: (1) to assess the effect of IPS in patients with SMI, examining the main competitive employment outcomes (i.e. days to first job, job acquisition, and job duration) and drop-out rates during a 3-year follow-up period; and (2) to explore any relevant association of these outcomes with working history, sociodemographic and clinical characteristics. To the best of our knowledge, this is the first, entirely Italian study in the literature addressed to replicate the IPS effectiveness in the "real world" and to shed more light on its long-term effects, using a 36-month follow-up design.

Methods

Participants

Participants (n = 46) were recruited from clients receiving treatment for SMI in one of the 7 adult CMHCs of the Reggio Emilia Department of Mental Health, a semi-urban catchment area of approximately 550.000 inhabitants, in the northern Italy [9]. Enrollment started on 1 January 2015 and ended on 30 June 2018.

For the purpose of this study, inclusion criteria were: (a) working age (18-60 years), (b) presence of a SMI (i.e. schizophrenia and bipolar disorder as defined in the Diagnostic and Statistical Manual of mental disorders, IV Edition, Text Revised [DSM-IV-TR]) [10] with a major role dysfunction in the previous 12 months, (c) to be in contact with CMHC for a minimum of 6 months and expected to remain in outpatient follow-up, (d) unemployment status at the time of study admission and in the preceding year, (e) expressed desire for competitive job in the open market, (f) at least a 3-month stabilization period before study entry, (g) ability and willingness to give informed consent, and (h) residence in the catchment area. Specifically, all the participants underwent an extensive diagnostic assessment using the Structured Clinical Interview for axis I mental disorders (SCID-I) [11].

Exclusion criteria were: (a) absence of a primary diagnosis of mental retardation (known Intelligence Quotient < 70), dementia or other organic mental disorders, or substance/alcohol abuse, (b) absence of significant medical conditions (such as end-stage cancer) that would preclude working during the follow-up period, (c) full-time hospitalization, and (d) engagement in another traditional vocational rehabilitation trajectory.

All adults entering the study protocol agreed to participate to the research and gave their informed consent before interview engagement. Relevant local ethical approvals were sought for the study. The current research has been carried-out in accordance with the Code of Ethics of the World Medical Association (Declaration of Helsinki) for experimental protocols including humans.

Procedures

Clients of the participating CMHCs were informed about the study in various ways (e.g. directly by mental health team members or through local information meetings). To assess eligibility, each client interested in participation and expressing a wish for paid employment was interviewed by IPS independent local coordinators, who were trained to evaluate eligibility. Clients who met the study criteria were accepted as participants of the study and referred for baseline assessment.

All the participants were trained by job coaches in accordance with the IPS supported employment model, which is based on the following key principles: (a) focus on competitive employment in work settings integrated into a community's economy and the open job market, (b) support in rapid job search (i.e. clients are expected to obtain job directly, without lengthy pre-employment training), (c) integration of vocational services with psychiatric services (i.e. rehabilitation is considered as an integral component of mental health intervention rather than a separate service), (d) attention to client's job preferences and choices, (e) individualized job support with employment specialists' engagement in systematic and active job development, (f) continuous assessment based on real work experiences, (g) time-unlimited support, (h) no exclusion criteria (i.e. motivation for obtaining competitive employment is the only necessary and sufficient condition for IPS enrollment), and (i) financial counselling about social security benefits [12].

All the participants were assigned to an IPS employment specialist, added to multidisciplinary community mental health team, and were followed-up during a 3-year period. Data were collected through interview on vocational outcomes at baseline and every 6 months to compare with previous IPS studies. Prior to start-up of the IPS program, IPS specialists received at least 4-month internal training and supervision on the IPS model and its "place-and-train" approach to job rehabilitation from a team of IPS trainers consisting of expert on supported employment that collaborated with IPS model developers in the "IPS Employment Center" at Lebanon, New Hampshire [3]. Each IPS specialist met regularly with his allocated CMHC to raise awareness of the service, and relied on CHMC staff members to refer potential patients. Referred patients were assessed by the IPS specialist for their motivation for obtaining employment before being offered the service, as well as for their work preferences, past work experiences, past experiences of traditional vocational rehabilitation, duration of taking charge at CMHC (i.e. before IPS enrollment), social benefits (e.g. disability pension, unemployment insurance), current skills, and tolerance for type and intensity of job demands.

Similarly to the EQOLISE trial, in each CMHC, the IPS model followed the structured and manualized approach focused on the immediate support of a job coach and a direct integration into competitive employment. IPS specialist supported the client by searching for vacant jobs, assisting applications, as well as coaching the client in working situations[8]. In some cases, participants accepted newly created competitive jobs developed in collaboration between the specialist and local employers. Once employed, "on the job" training and follow-along support were provided to help the individual in retaining the job for as long as possible. IPS specialists provided time-unlimited support before, during, and after periods of employment, operating in close collaboration with the other community mental health team members.

To assess the quality of the IPS implementation, we also conducted a *fidelity* assessment using the IPS-25 Fidelity Scale [13]. This scale measures adherence to the IPS principles and has shown good pr1edictive and discriminative validity in previous US studies. It has also been used in Italy [14] and the older version of the scale was applied in EQOLISE trial, supporting its predictive validity even in the European context [15]. In this regards, it has been widely reported that the lack of adequate technical assistance and training for staff members leads to IPS substandard implementation, attenuated effectiveness of the IPS program, and relevant impairment of the quality of the resulting evaluation [5]. Total score of the IPS-25 Fidelity Scale ranges from 25-125. According to Reme and co-workers [16], the critical cut-off point for being recognized as IPS was > 74. Each CMHC was assessed at three time points during the follow-up period (i.e. at baseline and after 3 and 6 months). Teams of trained evaluators followed detailed instructions, and the ratings were done based on interviews, team meeting observations, and document reviews. At the initial evaluation, two of the seven CMHC teams were just below the critical cut-off, but on the second and third assessment, all teams scored fair (> 85) on the fidelity scale. Therefore, all of the 7 teams improved their fidelity scores steadily throughout the follow-up period.

According to Bond and colleagues [17], competitive employment was defined as paid jobs in work settings integrated into the open job market. In the present research, we examined the following five competitive employment *outcomes*: (a) job acquisition (i.e. employment in the labor market for at least 1 day during the 36-month follow-up period), (b) job duration (i.e. total number of days worked), (c) job tenure (defined as weeks worked on the longest-held competitive job), (d) total hours per week worked, (e) days to first job (defined as the number of days from IPS admission to first competitive job), and (f) ever working ≥ 20 hours per week (defined as working at least 20 hours per week at some time during the follow-up period). The measure of days to first job is a negative indicator of successful employment: that is, the longer the duration, the poorer the outcome [17]. Job acquisition and ever working ≥ 20 hours per week were dichotomous measures; the others were continuous parameters. All employment outcomes were prospectively assessed during the 36-month follow-up period at baseline and every 6 months. Self-reported information was derived from interviews and cross-checked through chart records, which were maintained by support center staff having every week contact with all the participants. Finally, we also determined drop-out rates during the 3-year follow-up period.

In the total sample, we firstly observed crude competitive employment rate (i.e. employment at any time during the follow-up period) and crude drop-out rate (i.e. number of participants who discontinued IPS service during the follow-up period). Secondly, we calculated competitive employment rates and drop-out rates every 6 months throughout the follow-up period, using a survival analysis method. Finally, we examined any relevant association of both job acquisition and drop-out rate with working history (i.e. years of previous work, presence of past work experiences, presence of past experiences in traditional rehabilitation, presence of social benefits), sociodemographic and clinical characteristics (i.e. gender, age, years of education, and duration of taking charge at CMHC).

Statistical analysis

Data were analyzed using the Statistical Package for Social Science (SPSS) for Windows, version 15.0 [18]. Descriptive data included mean value ± standard deviation, median and interquartile range for quantitative variables, as well as absolute frequencies and percentages for categorical variables. There were no missing data. All tests were two-tailed with level of significance set at 0.05. Due to non-normality in all explorations (Kolmogorov-Smirnov test, with Lilliefors significance correction: $p < 0.05$), non-parametric statistics were used [18]. Categorical data in between-group comparisons were analyzed with Chi-square or Fisher's exact test, as appropriate (i.e. when any expected frequency was < 1 or 20% of expected frequency was ≤ 5). The Mann-Whitney U test was used to compare ordinal variables. Finally, we performed a Kaplan-Meier survival analysis to take into account the different duration of follow-ups and individuals who dropped-out. The primary aim of survival analysis is the modeling and analysis of "time-to-event" data: that is, data that have as end-point the time when an event occurs [19]. In this regards, events are not limited to death, but may include other significant events for the research such as job acquisition and participants who dropped-out. We specifically calculated cumulative survival and cumulative proportion of job acquisition and subjects who dropped-out (i.e. 1 - cumulative survival) every 6 months during the 36-month follow-up period.

Results

Over the course of the study, 46 individuals (26 [56.5%] males, 40 [95.2%] white Caucasians, median age = 32.56 years [interquartile range = 17.54 years]) were consecutively provided with the IPS service in one of the CMHCs of the Reggio Emilia Department of Mental Health. Clinical and sociodemographic characteristics of the total sample are reported in the Table I.

At the baseline, SCID-I[11] showed that 78.3% (n = 36) of the participants fulfilled the DSM-IV-TR diagnostic criteria for schizophrenia, while 21.7% (n = 10) were categorized as bipolar disorder. Forty-one (89.1%) patients reported having at least one previous competitive work experience (median of years worked = 7.50 [interquartile range = 15.75 years]), 14 (30.4%) at least one past experience of traditional vocational rehabilitation, and 20 (43.5%) a social benefit at IPS enrollment (Tab. I).

Employment outcomes

The crude competitive employment rate along the 36-month follow-up period (i.e. job acquisition) was 50% (n = 23) (Tab. I). Using a Kaplan-Meier survival analysis, cumulative employment rates were 41% at 6 months, 48% at 12 months, 60% at 18 months, 66% at 24 months, and 73% at 36 months (Tab. II).

The other main employment outcomes of IPS participants obtaining job during the 3 years of follow-up (i.e. IPS worker subgroup [n = 23]) are shown in the Table I. In details, median of days to first job was 102 (interquartile range = 135 days), median of total days employed was 215 (interquartile range = 278 days), median of weeks worked on the longest-held competitive job (i.e. job tenure) was 28 (interquartile range = 37 weeks), and median of hours per week worked was 20 (interquartile range = 10 hour/week). Moreover, 15 (65.2%) of the 23 IPS participants obtaining job were ever working ≥ 20 hours per week at some time during 36-month follow-up period (Table I). Finally, no significant association of job acquisition with working history, sociodemographic and clinical characteristics was found (Tab. III).

Drop-out rate

The crude drop-out rate across the 3-year follow-up period was 34.8% (n = 16) (Tab. I). Using a Kaplan-Meier survival analysis, cumulative drop-out rates were 18% at 6 months, 36% at 12, 18 and 24 months, and 52% at 36 months (Tabs. IV-V). No significant association of "drop-out" condition with working history, sociodemographic and clinical characteristics in the IPS total sample was found.

Discussion

First aim of the current study was to assess the long-term effect of IPS approach in patients with

TABLE I. *Employment outcomes, work history, and sociodemographic/clinical characteristics in the IPS total sample (n = 46).*

Variables	
Gender (males)	26 (56.5%)
Ethic group (Caucasian)	40 (95.2%)
Age	32.56 (17.54)
Years of education	13.00 (2.00)
DSM-IV-TR diagnosis	
Schizophrenia	36 (78.3%)
Bipolar disorder	10 (21.7%)
Duration (in years) of taking charge at CMHC (i.e. before IPS enrollment)	3.25 (6.72)
Work history (i.e. before IPS enrollment)	
Previous work experiences	41 (89.1%)
Years of previous work	7.50 (15.75)
Past experience of traditional vocational rehabilitation	14 (30.4%)
Social benefits	
Disability pension	20 (43.5%)
Unemployment insurance	16 (34.8%)
	4 (8.6%)
Job acquisition	
Crude cumulative employment rate	
6-month cumulative employment rate	23 (50.0%)
12-month cumulative employment rate	41%
18-month cumulative employment rate	48%
24-month cumulative employment rate	60%
36-month cumulative employment rate	66%
	73%
Drop-outs	
Crude cumulative drop-out rate	16 (34.8%)
6-month cumulative drop-out rate	18%
12-month cumulative drop-out rate	36%
18-month cumulative drop-out rate	36%
24-month cumulative drop-out rate	36%
36-month cumulative drop-out rate	52%
Employment outcomes in IPS worker subgroup (n = 23)	
Days to first job	102 (135)
Total days employed	215 (278)
Job tenure (in weeks)	28 (37)
Hours per week worked	20 (10)
Ever working ≥ 20 hours per week	15 (65.2%)

Legend. IPS: Individual Placement and Support; CMHC: Community Mental Health Center; IPS worker subgroup: IPS participants obtaining job during the follow-up period. Frequencies, percentages, median and interquartile range are reported

SMI attending to adult CMHCs of an Italian Department of Mental Health, directly examining the most used competitive employment outcomes in "the real world" (i.e. in the daily practice of a public mental health care service in Italy). Indeed, as the EQOLISE study was not powered to test the IPS effectiveness for the separate countries, new evidence is needed in Europe, particularly in countries as Italy, where, differently to the US labor economics, there is a stronger social security system and the employment opportunities are rather limited [4].

Competitive employment outcomes

During the 36-month follow-up period, we found a *crude competitive employment rate* of 50%. This result is in line with what observed in the EQOLISE study (55% [in

TABLE II. *Kaplan-Meier survival analysis and cumulative employment rates in the IPS total sample (n = 46).*

Time (in months)	IPS individuals	Employment (yes) (n)	Censured individuals (n)	Proportion surviving	Cumulative survival	Cumulative employment rate
0-3	46	11	9	0.76	0.76	0.24
3-6	26	6	4	0.77	0.59	0.41
6-9	16	1	2	0.94	0.56	0.44
9-12	13	1	3	0.92	0.52	0.48
12-15	9	2	0	0.77	0.40	0.60
15-18	7	0	0	1	0.40	0.60
18-21	7	0	0	1	0.40	0.60
21-24	7	1	1	0.86	0.34	0.66
24-27	5	1	0	0.80	0.27	0.73
27-30	4	0	0	1	0.27	0.73
30-33	4	0	0	1	0.27	0.73
30-36	4	0	4	1	0.27	0.73

Legend. IPS: Individual Placement and Support; Censured individuals = IPS participants lost to a specific month interval of the follow-up without obtaining a job; Proportion surviving on a specific month interval = 1 – (number of subjects obtaining job/number of IPS participants in this time interval); Cumulative survival = proportion surviving in a defined month interval multiplied cumulative survival from the previous step; Cumulative employment rate = cumulative proportion of obtained job (i.e. 1 – cumulative survival)

TABLE III. *Associations of job acquisition with working history, sociodemographic and clinical characteristics in the IPS total sample (n = 46).*

Variables	Job acquisition		
	(yes; n = 23)	(no; n = 23)	(Z/χ^2)
Gender (males)	12 (52.2%)	14 (60.9%)	0.35
Age	33.34 ± 9.80	35.80 ± 10.17	-0.74
Age group (18-35 years)	15 (65.2%)	11 (47.8%)	1.42
Years of education	13.17 ± 3.10	12.48 ± 3.68	-0.64
Duration (in years) of taking charge at CMHC	4.64 ± 4.97	5.69 ± 6.29	-0.69
Years of previous work	11.30 ± 10.15	10.04 ± 9.19	-0.43
Past work experiences	21 (91.3%)	20 (87.0%)	0.22
Past traditional rehabilitation	8 (34.8%)	6 (26.1%)	0.41
Social benefit	10 (43.5%)	10 (43.5%)	0.00

Legend. IPS: Individual Placement and Support; CMHC: Community Mental Health Center. Frequencies, percentages, mean ± standard deviation, Chi-squared (χ^2) test and Mann-Whitney test (Z) values are reported

a 18-month follow-up period]) [7], but slightly higher than those reported in some Northern European trials conducted in Sweden (46% [in a 18-month follow-up period]) [20], The Netherlands (44% [in a 30-month follow-up period]) [21], and Norway (41% [in a 12-month follow-up period]) [16], and significantly greater than that (22%) observed in a 24-month IPS trial conducted in the UK [22]. However, it is not still comparable to job acquisition rates reported in a meta-analysis on IPS model in the US studies (62.1%) [5], and in a Hong Kong (70% [in a 12-month follow-up period]) [23] and an Australian (64% [in a 6-month follow-up period]) [24] IPS trial. Diminished effectiveness for IPS in Europe has been typically ascribed to disability and labor policies that can prevent the return to work (i.e. what Burns referred as the "benefit trap") [7].

However, our survival analysis results showed increasing cumulative employment rates ranging from 48% at 1 year to 66% at 2 years and 73% at 3 years. These findings are more consistent with those reported in the US studies, which also suggested a higher effectiveness over the other vocational rehabilitation approaches[4]. In details, rigorous evaluation of IPS found that 60% or more of IPS clients obtained competitive jobs compared to approximately 25% of those who received other types of vocational assistance [17]. In this regards, Bond and co-workers[5] suggested that almost 1/4 of patients who express an interest in competitive employment will suc-

TABLE IV. *Kaplan-Meier survival analysis and cumulative drop-out rates in the IPS total sample (n = 46).*

Time (in months)	IPS individuals	Drop-out (n)	Censured individuals (n)	Proportion surviving	Cumulative survival	Cumulative drop-out rate
0-3	46	5	4	0.89	0.89	0.11
3-6	37	3	5	0.92	0.82	0.18
6-9	29	3	3	0.90	0.74	0.26
9-12	23	3	2	0.87	0.64	0.36
12-15	18	0	2	1	0.64	0.36
15-18	16	0	2	1	0.64	0.36
18-21	14	0	1	1	0.64	0.36
21-24	13	0	3	1	0.64	0.36
24-27	10	0	2	1	0.64	0.36
27-30	8	0	0	1	0.64	0.36
30-33	8	2	0	0.75	0.48	0.52
30-36	6	0	6	1	0.48	0.52

Legend. IPS: Individual Placement and Support; Censured individuals: IPS participants lost to a specific month interval of the follow-up without being dropped out; Proportion surviving on a specific month interval = 1 – (number of dropped-out subjects/number of IPS participants in this time interval); Cumulative survival = proportion surviving in a defined month interval multiplied cumulative survival from the previous step; Cumulative drop-out rate = cumulative proportion of dropped out individuals (i.e. 1 – cumulative survival)

TABLE V. *Associations of "drop-out" condition with working history, sociodemographic and clinical characteristics in the IPS total sample (n = 46).*

Variables	Drop-out		
	(yes; n = 16)	(no; n = 30)	(Z/χ^2)
Gender (males)	11 (68.8%)	15 (50.0%)	1.49
Age	36.18 ± 10.38	33.72 ± 9.82	-0.67
Age group (18-35 years)	8 (50.0%)	18 (60.0%)	0.42
Years of education	12.13 ± 3.12	13.20 ± 3.51	-1.04
Duration (in years) of taking charge at CMHC	2.75 ± 3.84	5.37 ± 5.58	-1.13
Years of previous work	12.60 ± 9.58	9.97 ± 9.68	-0.88
Past work experiences	15 (93.8%)	26 (86.7%)	0.54
Past traditional rehabilitation	3 (18.8%)	11 (36.7%)	1.58
Social benefit	4 (25.0%)	16 (53.3%)	3.40

Legend. IPS: Individual Placement and Support; CMHC: Community Mental Health Center. Frequencies, percentages, mean ± standard deviation, Chi-squared (χ^2) test, Fisher exact test, and Mann-Whitney test (Z) values are reported

ceed in obtaining a job in diverse vocational program (or even without any vocational services), but IPS helps an additional 35% of the target group who otherwise remain unemployed.

Furthermore, our findings confirm the absolute inconsistency of concerns that several clinicians often raised about the potential detrimental impact of the IPS model. In this regards, many mental health professionals worried that IPS (i.e. rapid job searching attempts and the efforts to hold a competitive employment) might lead to increased anxiety and uncertainty in patients with long-term mental disorders because of the threat of returning to the workplace without a protracted period of preparation [2]. Such clinicians frequently discouraged patients from applying for competitive employment because they were convinced that a stressful surrounding would have led to a destabilization of the subject. Overall, no evidence supports these concerns and a deterioration in mental or social functioning at final follow-up in the IPS compared with other vocational services [25]. In contrast, On the contrary, it has been demonstrated that finding employment correlated with an increase in global functioning, symptoms, and social adjustment [8]. As job acquisition (i.e. the percentage of participants who gain competitive job during follow-up) has been criticized as a crude indicator, other competitive employment outcomes have been suggested as measure of IPS effectiveness, including time to first job, job duration, and job tenure. In the present research, the median of *days to first job* (102 days) is lower than what reported in other IPS comparable study (i.e. 136 days [5], 137 days [25], and 126 days [17], respectively). Together with the evidence that 19 (82.6%) out of 23 IPS participants obtaining job during the 3-year follow-up period did so within their first 12 months, this finding seems to confirm that little is lost in terms of job acquisition by limiting the duration of involvement in IPS services to 1 year. In this regards, Burns and colleagues [25] recently proposed an overcoming of IPS "no-discharge" policy and its active support that is not withdrawal although patients may disengage from services. As this "no-discharge" policy powerfully restricts access to IPS in resource-limited public services, the authors suggested that there is a merit on a time limit to avoid persisting with participants who are currently unlikely to succeed. In the current study, a limit of the duration of the support to 12 months does not appear to significantly reduce the number of subjects obtaining competitive employment. Thus, given current difficulties in implementing IPS in times of austerity, a time-limited model could be the first choice for new services [25].

In the present research, the median of *total days employed* (215 days) is in line with what reported in the EQUOLISE trial (i.e. 214 days in a 18-month follow-up period) [7] and in the most US IPS studies (e.g. 199 days in a 12-month follow-up period [13] and 215 days in a 18-month follow-up period [17]), but much higher than those observed in other European IPS researches (e.g. 74 days in a 12-month follow-up period [25] and 123 days in a 30-month follow-up period [21]). Similarly, in the current study, both median of *hours per week worked* (20 hours/week) and *job tenure* (28 weeks) are consistent with what reported in the US studies (e.g. respectively 19 hours/week and 25 weeks in a 18-month follow-up period [13], and 23 hours/week and 25 weeks in another 18-month follow-up period [17]), but slightly higher than those observed in a UK 12-month follow-up trial (respectively 15 hours/week and 18 weeks [25]). Finally, similarly to what reported in other IPS comparable studies [5,17], almost two-thirds of our IPS participants obtaining job worked 20 hours or more per week at some time during the 36-month follow-up period. Few IPS clients worked full-time, likely due to preferences, limited stamina, and/or fear of losing health insurance or other benefits.

In conclusion, our findings on primary and secondary employment outcomes appear to excel in the European context and to be comparable with the evidence of a higher effectiveness of the IPS model than the traditional vocational approach in rehabilitating people with SMI shown in several US trials. However, despite the encouraging results that emerged in this research, a transfer of IPS methodology to Europe requires certain structural changes, as European countries differ from USA in terms of job market, labor economics, and welfare system [2]. According to Fioritti and co-workers [4], a controversial issue in Italy concerns the fact that some clients find jobs in the informal "black labor market", which, differently to the set of values in the Italian constitution (defining work as a right of the individual), represents 10-to-50% of all employment opportunities in different regions, mostly comprising jobs requiring non-specialized manpower. Moreover, a second controversial issue is the precarious nature of jobs in Italy. Indeed, most participants find part-time employment in 6-to-12 month contact, which is very far from the gold standard of full-time and forever that traditional Italian regulations would require (though almost never ensure) [6]. Precarious jobs represent approximately 50% of employment opportunities for all young workers in Italy. Conversely, social enterprise and temporary grant jobs are often not precarious, as they tend to last forever, but they are not open labor market jobs and are an economically protected niche. For these reasons, many mental health clients may be apprehensive about participating in a new vocational program aimed at competitive employment, and they may opt for continuing in the more familiar and comfortable environment of existing services [26]. Finally, Fioritti and colleagues [4] also suggested that a third important controversial aspect regards the 40-to-50% of users who do not find a job with IPS and still demand work. In this sense, perhaps it is useful to provide IPS model along with other treatment option, possibly in a stepwise order. Strategies supporting the individual in entering mainstream jobs should be used at first, especially for first-episode psychosis [27], and for a sufficient duration (at least 12-18 months), before entering the subsidy system and sheltered approach.

In the present research, no significant association of job acquisition with work history, sociodemographic and clinical variables was found. These findings suggests that employment rate appear to be independent from gender, age, level of education, duration of taking charge at CMHC, past work experiences, previous traditional rehabilitation program, and social benefits. In contrast, Metcalfe and coworkers [28] showed that a recent work history and a less time on the Social Security rolls are associated with greater probability of employment. Differently, our results do not seem to support the risk-adverse of the benefit trap and the perverse incentives of social security system.

Drop-out rate

In the current study, our drop-out rates are higher than those reported in several IPS comparable trials (approximately 10% in most US studies)[10]. In details, we found a crude drop-out rate of approximately 35% and cumulative drop-out rates of 18% at 6 months and 36% at 1 and 2 years. However, these results are substantially in line with what (43% in a 30-month follow-up period) reported in a relatively recent IPS study conducted in the Netherlands [21] and in an early review noting a high drop-out rate (about 40%) among supported employment clients [12]. Finally, although no association of drop-out condition with work history, sociodemographic and clinical characteristics was found, the large majority of participants dropping-out the IPS program did so within their first 12 months. In times of limited resources, this result further supports a time-limited IPS model as first choice for new services [25].

Limitations

Several limitations of this study should be acknowledged. Firstly, we have focused exclusively on competitive job, and the impact of supported employment on nonvocational measures of psychiatric symptoms and quality of life was not evaluated. Thus, further research in Italy is required to investigate the relationships between vocational and nonvocational outcomes, including clients' motivation to work, self-esteem, and self-efficacy. Moreover, measures of job quality are needed, as are measures of job satisfaction [25].

Secondly, in the present research, measuring job tenure has been problematic because several participants are employed at the end of follow-up (i.e. many job tenure periods are right-censored). According to Bond and co-workers [25], perhaps the literature consistently underestimates job tenure. The optimal solution would be to conduct long-term follow-up studies.

Finally, another relevant limitation was the limited size of IPS worker subgroup (n = 23). This probably reduces the generalizability of our results, which must be replicated in larger IPS samples.

Conclusions

This study documents the feasibility of an IPS implementation strategy for introducing a new service model in a traditional public mental health care system in Italy, which has not always welcomed change. Indeed, a nationwide introduction of IPS not only might lead to beneficial changes for clients, but also might precipitate system changes towards the development of a recovery-oriented system. Moreover, this research adds evidence to the growing literature on the positive effect of IPS in promoting employment among people with SMI, also in a European country with a socioeconomical climate that differs and is more protective than that in the US. However, future studies on subjective outcomes, process evaluations, and cost effectiveness are needed.

Acknowledgements

This research received no specific grant from funding agencies in the public, commercial or not-for-profit sectors. IPS implementation in the Reggio Emilia Department of Mental Health and Pathological Addiction is partly financed through a special regional fund provided by Emilia Romagna Region. Our warmest thank to all the participants.

References

1. Schneider J, Akhtar A. Implementation of individual placement and support: the Nottingham experience. Psychiatr Rehabil J 2012;35:325-332. https://doi.org/10.2975/35.4.2012.325.332
2. Viering S, Bartsch B, Obermann C, et al. The effectiveness of individual placement and support for people with mental illness new on social benefits: a study protocol. BMC Psychiatry 2013;13:195. https://doi.org/10.1186/1471-244x-13-195
3. Becker DR, Drake RE. A working life for people with severe mental illness. New York: Oxford University Press 2003.
4. Fioritti A, Burns T, Hilarion P, et al. Individual placement and support in Europe. Psychiatr Rehabil J 2014;37:123-128. https://doi.org/10.1037/prj0000065
5. Bond GR, Drake RE, Becker DR. Generalizability of the Individual Placement and Support (IPS) model of supported employment outside the US. World Psychiatry 2012;11:32-39. https://doi.org/10.1016/j.wpsyc.2012.01.005
6. Fioritti A, Bassi M, De Girolamo G. Italian psychiatry, 25 years of change. Intern Psychiatry 2003;2:14-16.
7. Burns T, Catty J, Becker T, et al. The effectiveness of supported employment for people with severe mental illness: a randomised controlled trial. Lancet 2007;370:1146-1152.
8. Burns T, Catty J, White S, et al. The impact of supported employment and working on clinical and social functioning: results of an international study on individual placement and support. Schizophr Bull 2009;35:949-958. https://doi.org/10.1093/schbul/sbn024
9. Pelizza L, Azzali S, Paterlini F, et al. The "Reggio Emilia At-Risk Mental States" program: a diffused, "liquid" model of early intervention in psychosis implemented in an Italian Department of Mental Health. Early Interv Psychiatry 2019;54:254-263. https://doi.org/10.1111/eip.12851
10. American Psychiatric Association (APA). Diagnostic and statistical manual of mental disorders, IV Ed., Text Revised. Washington DC: American Psychiatric Association Press 2000.
11. First MB, Spitzer RL, Gibbon M, et al. Structured clinical interview for Axis I DSM-IV-TR disorders, Research Ed. New York: New York State Psychiatric Institute 2002.
12. Bond GR. Principles of the individual placement and support model: empirical support. Psychiatr Rehabil J 1998;22:11-23. https://doi.org/10.1037/h0095271
13. Bond GR, Peterson AE, Becker DR, et al. Validation of the Revised Individual Placement and Support Fidelity Scale (IPS-25). Psychiatr Serv 2012;63:758-763. https://doi.org/10.1176/appi.ps.201100476
14. Berardi D, Fioritti A. Individual placement and support: manuale italiano del metodo per il supporto all'impiego delle persone con disturbi mentali. Bologna: Bononia University Press 2017.
15. Catty J, Lissouba P, White S, et al. Predictors of employment for people with severe mental illness: results of an international six-centre randomised controlled trial. Br J Psychiatry 2008;192 :224-231. https://doi.org/10.1192/bjp.bp.107.041475
16. Reme SE, Monstad K, Sveinsdottir V, et al.

A randomized controlled multicenter trial of individual placement and support for patients with moderate-to-severe mental illness. Scand J Work Environ Health 2019;45:33-41. https://doi.org/10.5271/sjweh.3753

[17] Bond GR, Drake RE, Campbell K. Effectiveness of individual placement and support supported employment for young adults. Early Interv Psychiatry 2016;10:300-307. https://doi.org/10.1111/eip.12175

[18] SPSS Inc. Statistical Package for Social Science (SPSS) for Windows, version 15.0. Chicago, IL: SPSS Inc. Press 2010.

[19] Jager K, van Dijk P, Zoccali C, et al. The analysis of survival data: the Kaplan-Meier method. Kidney Int 2008;74:560-565. https://doi.org/10.1038/ki.2008.294

[20] Bejerholm U, Areberg C, Hofgren C, et al. Individual placement and support in Sweden: a randomized controlled trial. Nord J Psychiatry 2015;69:57-66. https://doi.org/10.3109/08039488

[21] Michon H, van Busschbach JT, Stant AD, et al. Effectiveness of individual placement and support for people with severe mental illness in The Netherlands: a 30-month randomized controlled trial. Psychiatr Rehabil J 2014;37:129-136. https://doi.org/10.1037/prj0000061

[22] Heslin M, Howard L, Leese M, et al. Randomized controlled trial of supported employment in England: 2 year follow-up of the Supported Work and Needs (SWAN) study. World Psychiatry 2011;10:132-137. https://doi.org/10.1002/j.2051-5545-2011-tb00035.x

[23] Kin Wong K, Chiu R, Tang B, et al. A randomized controlled trial of a supported employment program for persons with long-term mental illness in Hong Kong. Psychiatr Serv 2008;59:84-90. https://doi.org/10.1176/ps.2008.59.1.84

[24] Killackey E, Jackson HJ, McGorry PD. Vocational intervention in first-episode psychosis: individual placement and support v. treatment as usual. Br J Psychiatry 2008;193:114-120. https://doi.org/10.1192/bjp.bp.107.043109

[25] Burns T, Yeeles K, Langford O, et al. A randomized controlled trial of time-limited individual placement and support: IPS-LITE trial. Br J Psychiatry 2015;207:351-356. https://doi.org/10.1192/bjp.bp.114.152082

[26] Oshima I, Sono T, Bond GR, et al. A randomized controlled trial of individual placement and support in Japan. Psychiatr Rehabil J 2014;37:137-143. https://doi.org/10.1037/prj0000085

[27] Pelizza L, Paterlini F, Azzali S, et al. The approved Italian version of the comprehensive assessment of at-risk mental states (CAARMS-ITA): field test and psychometric features. Early Interv Psychiatry 2019;13:86-94. https://doi.org/10.1111/eip.12669

[28] Metcalfe JD, Riley J, McGurk S, et al. Comparing predictors of employment in individual placement and support: a longitudinal analysis. Psychiatry Res 2018;264:85-90. https://doi.org/10.1016/j.psychres.2018.03.050

JOURNAL OF PSYCHOPATHOLOGY 2021;27:81-83
DOI: 10.36148/2284-0249-390

Original article

Impact of waiting time on the outcome of a group therapy intervention for patients with functional neurological disorders

Mohammed K. Quraishi[1], Hammad Sadique[1], Andrea E. Cavanna[1,2,3]

[1] *Department of Neuropsychiatry, BSMHFT and University of Birmingham, Birmingham, United Kingdom;* [2] *School of Life and Health Sciences, Aston Brain Centre, Aston University, Birmingham, United Kingdom;* [3] *Sobell Department of Motor Neuroscience and Movement Disorders, Institute of Neurology and University College London, London, United Kingdom*

SUMMARY

Objective

The clinical management of patients with functional neurological disorders can be challenging and often involves neuropsychiatric input. Relatively little is known about factors affecting clinical outcomes following treatment interventions in this patient population. This retrospective study evaluated the care pathway based on group therapy intervention for adult patients with functional neurological disorders attending a specialist neuropsychiatry clinic.

Methods

We retrospectively reviewed the care pathways of 67 consecutive adult outpatients referred to group therapy sessions for functional neurological disorders, focusing on outcome predictors.

Results

The mean length of the care pathway (from referral to neuropsychiatry to first contact with therapists) in patients rated as clinically improved was significantly lower than the duration of the care pathway of patients who did not show any improvement: 37.8 weeks compared to 52.1 weeks, respectively ($p < 0.03$). There were no other significant differences between the groups in either demographic or clinical variables.

Conclusions

Longer waiting times were found to negatively affect clinical outcomes of group therapy sessions for functional neurological disorders in a neuropsychiatry setting. Clinicians should be aware of the possible impact of waiting times on the care pathways of patients with functional neurological disorders. Streamlined care pathways for early intervention in this clinical population should be prioritized.

Key words: functional neurological disorders, group therapy, neuropsychiatry, waiting times

Received: May 18, 2020
Accepted: January 5, 2021

Correspondence
Andrea E. Cavanna
Department of Neuropsychiatry, National Centre for Mental Health, 25 Vincent Drive, Birmingham B15 2FG, United Kingdom
E-mail: a.e.cavanna@bham.ac.uk

Conflict of interest
The Authors declare no conflict of interest

How to cite this article: Quraishi MK, Sadique H, Cavanna AE. Impact of waiting time on the outcome of a group therapy intervention for patients with functional neurological disorders. Journal of Psychopathology 2021;27:81-83. https://doi.org/10.36148/2284-0249-390

Introduction

Functional neurological disorders are condition in which patients experience medically unexplained neurological symptoms, such as weakness, movement disorders, sensory symptoms, and blackouts [1,2]. It has been estimated that these symptoms account for about 10% of primary care presentations, however their diagnosis and treatment can be particularly challenging [3]. Patients with functional neurological disorders are often referred to neuropsychiatry clinics for specialist assessment and management [4]. Treatment strategies include psychoeducation and psychological interventions, usually based on cognitive behavioral therapy principles, which can be administered either alone or in combination with pharmacotherapy [5]. Predictors of better outcome include early diagnosis, psychoeducation and patient acceptance, along with appropriate referrals to specialist services [6,7], where shorter time to treatment through streamlined care pathways were shown to have the potential to amplify the therapeutic effect [8].

We set out to conduct a retrospective study evaluating the care pathway based on group therapy intervention for adult patients with functional neurological disorders attending a specialist neuropsychiatry service in the United Kingdom. We focused on the possible impact of waiting times on the clinical outcomes of this patient population, in order to identify areas for improvement in neuropsychiatry service provision [9].

Methods

We retrospectively reviewed the care pathways of 67 consecutive outpatients diagnosed with functional neurological disorders at the specialist neuropsychiatry clinic, Department of Neuropsychiatry, National Centre for Mental Health, Birmingham, United Kingdom. Following clinical assessment and diagnosis confirmation as per the Diagnostic and Statistical Manual of Mental Disorders criteria[1], patients with functional neurological disorders were referred to an information and management intervention, consisting of five group therapy sessions. The duration of each session was one hour and the frequency of the sessions was weekly. The group therapy sessions were delivered by trained liaison nurses and occupational therapists, and involved elements of psychoeducation, with the use of presentations, hand-outs, discussion, and personal reflection. The aims of the sessions were to increase patients' understanding of their diagnosis, to help them manage their own condition and symptoms, to increase understanding of potential triggers, to provide support and to reduce feelings of isolation and hopelessness. The group therapy intervention was followed by a clinical assessment by the referring consultant, who rated clinical outcomes according to three major categories: improvement, improvement with further therapy recommended, or lack of improvement.

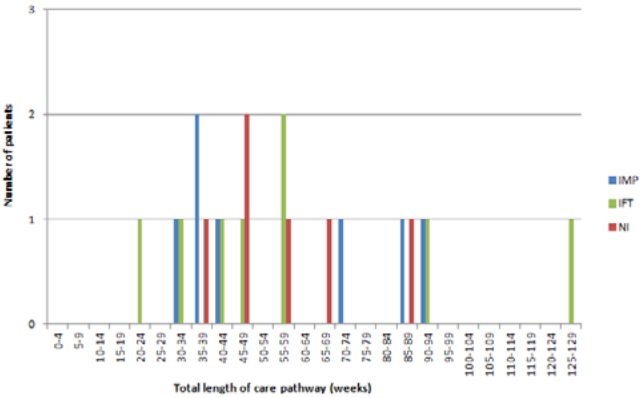
The timeline of the care pathway was assessed for each patient, with focus on its total duration from referral to neuropsychiatry to first contact with therapists. Intermediate points, including first neuropsychiatric assessment, confirmation of diagnosis, and referral to group therapy intervention, were also examined. Student's t-tests were used to assess possible differences in demographic and clinical variables, as well as mean durations of care pathways between the groups.

Results

Of the 67 patients with a diagnosis of functional neurological disorders, 47 were females (70%). The median age at referral to the specialist neuropsychiatry clinic was 43 years (range 16-69). Referrals mainly originated from secondary care (neurologists: n = 38, 57%), followed by primary care (general practitioners: n = 18, 27%). The mean time from referral to first assessment at the neuropsychiatry clinic was 17.5 (± 1.5) weeks. The mean time between neuropsychiatric assessment and diagnosis was 7 (± 2.0) weeks, with 84% of the patients being diagnosed on their first assessment. Following discussion in clinic, 34 patients (51%) were referred to the group therapy sessions for functional neurological disorders. The mean waiting time from referral to first contact with the therapists was 24.8 (± 2.4) weeks.

At their follow-up appointment following the intervention, 7 patients (20%) were classified as clinically improved, 8 patients (23%) as improved with further therapy recommended, and 6 patients (18%) as not improved. The remaining 13 patients showed poor compliance with the treatment intervention or were still attending the therapy sessions at the time of data collection (Fig. 1).

There were no significant differences in either demographic or clinical variables between the outcome groups. However, the mean total length of the care pathway (from referral to neuropsychiatry to first contact with therapists) in patients rated as clinically improved was significantly lower than the duration of the care pathway of patients who did not show any improvement: 37.8 (± 2.2) weeks compared to 52.1 (± 5.2) weeks, respectively ($p < 0.03$). This difference was mainly driven by the shorter waiting time between referral to the group therapy sessions and first contact with therapists in the group of patients who reported a clinical improvement: 22.9 (± 3.8) weeks versus 39.1 (± 4.4) weeks ($p < 0.02$). When comparing the group of patients rated as improved with further therapy recommended and



Abbreviations. IMP: improved; IFT: improved with further therapy recommended; NI: not improved

FIGURE 1. *Clinical outcomes following group therapy sessions for functional neurological disorders, according to waiting time before active intervention.*

the group of patients rated as not improved, the waiting time between referral to the group therapy sessions and first contact with therapists showed a significant difference (25.3 ± 5.9 weeks *versus* 39.1 ± 4.4 weeks; $p < 0.05$), whereas the total length of the care pathway was not significantly different.

Discussion

This retrospective study found evidence for the impact of waiting times on a care pathway involving group therapy intervention for patients with functional neurological disorders in a neuropsychiatry setting. In our sample, clinical improvement was associated with more streamlined care pathways and shorter waiting times. The total duration of the care pathways exceeded 37 weeks for all the patients who showed no improvement following the group therapy intervention. The group of patients with poor outcome was characterized by longer waiting times between referral to neuropsychiatry and confirmation of diagnosis (at least 10 weeks) and longer waiting times between referral to group therapy intervention and first contact with therapists (at least 25 weeks).

Our findings are to be interpreted in the light of several limitations, as the relatively small sample size and the specialized nature of the neuropsychiatric clinic limit their generalizability. The nature of the treatment intervention (group therapy sessions) might also be a factor that limits the generalizability of the findings from our study. For example, the results of a study on patients with psychogenic movement disorders showed that there was no specific benefit from short term psychodynamic psychotherapy either early or late as opposed to neurological observation and support [10]. Moreover, selection bias might have operated at the time of patient referral to our group therapy intervention. Finally, we had no control group of patients without active intervention.

Overall, our results confirm previous findings suggesting that the duration of the care pathway can have an impact on the clinical outcome of interventions for functional symptoms [8] and prompt further work to investigate these effects beyond group therapy interventions for functional neurological disorders. Likewise, work toward the development guidelines on care pathways across neuropsychiatric disorders focusing on timeliness of intervention and identification of gaps in healthcare service provision should be prioritized.

References

1. American Psychiatric Association. Diagnostic and statistical manual of mental disorders, 5th ed. (DSM-5). Arlington VA: American Psychiatric Publishing 2013.
2. Voon V, Cavanna AE, Coburn K, et al.; On behalf of the American Neuropsychiatric Association Committee for Research. Functional neuroanatomy and neurophysiology of functional neurological disorders (conversion disorder). J Neuropsychiatry Clin Neurosci 2016;28:168-190. https://doi.org/10.1176/appi.neuropsych.14090217
3. Kroenke K, Rosmalen JG. Symptoms, syndromes, and the value of psychiatric diagnostics in patients who have functional somatic disorders. Med Clin North Am 2006;90:603-626. https://doi.org/10.1016/j.mcna.2006.04.003
4. Arambepola NMA, Rickards H, Cavanna AE. The evolving discipline and services of neuropsychiatry in the United Kingdom. Acta Neuropsychiatrica 2012;24:191-198. https://doi.org/10.1111/j.1601-5215.2012.00655.x
5. Kroenke K. Efficacy of treatment for somatoform disorders: a review of randomized controlled trials. Psychosom Med 2007;69:881-888. https://doi.org/10.1097/PSY.0b013e31815b00c4
6. Oyama O, Paltoo C, Greengold J. Somatoform disorders. Am Fam Physician 2007;76:1333-1338.
7. Durrant J, Rickards H, Cavanna AE. Prognosis and outcome predictors in psychogenic non-epileptic seizures. Epilepsy Res Treat 2011:274736. https://doi.org/10.1155/2011/274736
8. Mayou R. Are treatments for common mental disorders also effective for functional symptoms and disorder? Psychosom Med 2007;69:876-880. https://doi.org/10.1097/PSY.0b013e31815b00a6
9. Agrawal N, Fleminger S, Ring H, et al. Neuropsychiatry in the UK: Planning the service provision for the 21st century. Psychiatr Bull 2008;32:303-306. https://doi.org/10.1192/pb.bp.107.018424
10. Kompoliti K, Wilson B, Stebbins G, et al. Immediate vs delayed treatment of psychogenic movement disorders with short term psychodynamic psychotherapy: Randomized clinical trial. Parkinsonism Relat Disord 2014;20:60-63. https://doi.org/10.1016/j.parkreldis.2013.09.018

JOURNAL OF PSYCHOPATHOLOGY 2021;27:84-89
DOI: 10.36148/2284-0249-410

Original article

Cognitive development and adaptive functions in children with Down syndrome at different developmental stages

Renata Nacinovich[1,2,3], Monica Bomba[1], Silvia Oggiano[2],
Simona Di Guardo[2], Fiorenza Broggi[2], Andrea E. Cavanna[4,5,6]

[1] Clinic of Child and Adolescent Neuropsychiatry, San Gerardo Hospital, Monza, Italy; [2] Department of Medicine and Surgery, University of Milano-Bicocca, Milan, Italy; [3] Milan Center for Neuroscience (Neuro-MI), Milan, Italy; [4] Department of Neuropsychiatry, BSMHFT and University of Birmingham, United Kingdom; [5] Sobell Department of Motor Neuroscience and Movement Disorders, Institute of Neurology and University College London, United Kingdom; [6] School of Life and Health Sciences, Aston Brain Centre, Aston University, Birmingham, United Kingdom

SUMMARY

Background

Children with Down syndrome (DS) have learning difficulties resulting in mild to severe intellectual disability, whereas their adaptive functions are generally more preserved. Little is known about the developmental trajectories of cognitive and adaptive functions in this population. In the present study, cognitive and adaptive functions were assessed in children with DS at different developmental stages.

Methods

Cognitive and adaptive functions were assessed in a total of 53 children with DS: 20 children aged 2 to 6 and 33 children aged 10 to 15. Cognitive development was assessed using the Griffiths Mental Development Scales 2-8 for younger children and the Wechsler Intelligence Scale for Children-Fourth Edition for older children. Adaptive functions were evaluated with the Vineland Adaptive Behavior Scale in both age groups.

Results

Among cognitive functions, working memory was the most significantly affected, whereas the visuo-spatial component was relatively preserved. In terms of adaptive functions, children reported the lowest mental age in the expressive communication domain, and the highest mental age in the daily living skills. Adaptive functions were comparatively worse in the older group, whereas cognitive profiles were impaired to a similar degree between the two age groups.

Conclusions

Adaptive functions appear to be relatively more impaired than cognitive functions in older children with DS. The increasing demands from the environment that children have to deal with during pre-adolescence and adolescence might contribute to selectively affect their adaptive skills.

Key words: adaptive functions, cognitive development, down syndrome, intellectual disability

Received: October 21, 2020
Accepted: November 24, 2020

Correspondence
Andrea E. Cavanna
Department of Neuropsychiatry, National Centre for Mental Health, 25 Vincent Drive, Birmingham B15 2FG, United Kingdom
E-mail: a.e.cavanna@bham.ac.uk

Conflict of interest
The Authors declare no conflict of interest

How to cite this article: Nacinovich R, Bomba M, Oggiano S, et al. Cognitive development and adaptive functions in children with Down syndrome at different developmental stages. Journal of Psychopathology 2021;27:84-89. https://doi.org/10.36148/2284-0249-410

Background

Down syndrome (DS) is the most common genetic cause of intellectual disability across all ethnic and socio-economic groups [1]. Despite the consistent increase in maternal age in developed Countries, the prevalence of DS births has remained stable as a result of increasing use of prenatal diagnostic procedures [2].

Children with DS have learning difficulties which result in mild to severe intellectual disability [3]. The mental age is rarely over 8 years old, although a few cases of normal Intelligence Quotient (IQ) in children with DS have been reported [4]. IQ scores are known to progressively decrease with age [5,6], especially in the first decade of life [7-12], before reaching a plateau in adolescence which persists throughout adulthood [7]. Interestingly, cognitive functions do not appear to be equally affected: children with DS report more severe deficits in language compared to visuo-spatial skills, compared to children of the same mental age with mental retardation of different etiology or with typical development [13].

Fewer studies have focused on adaptive functions in children with DS. A study by Dykens et al. [14] on 80 children with DS aged between 1 and 11.5 years showed more significant deficits in communication skills (especially expressive language) compared to daily living and socialization skills. A subsequent study by Dressler et al. [3] on 75 individuals with DS aged between 4 and 53 years showed similar findings, although in this study socialization was the most severely impaired domain in the age group 4-10 years. Overall, children with DS were shown to have a good degree of adaptive functioning within the broader population of individuals with mental retardation.

Studies on the development of adaptive functions in relation to age in individuals with DS have reported controversial results. Dressler et al. [3] found that adaptive functions gradually and steadily increase with age up to 30 years, after which they undergo a progressive decline, whereas earlier studies had showed that adaptive functions worsen with age [7,15,16]. The findings of Dykens et al. [14] supported the hypothesis of an advance-plateau pattern of adaptive development [17,18]: these authors observed that while children aged 1 to 7 showed significant age-related gains in adaptive behaviour, older subjects (7 to 11.5 years) showed no association between chronological age and adaptive behaviour.

The aim of the present study was to assess cognitive development and adaptive functions in two groups of children with DS at different developmental stages (2-6 versus 10-15 years of age). This should provide a better understanding of the trajectory of cognitive development and adaptive skills in children with DS, thus contributing to the implementation of more effective and targeted educational strategies and treatment interventions.

Methods

Participants

A total of 53 children with DS aged 2 to 15 (35 males, 66%) participated in this cross-sectional study conducted at the outpatient Child and Adolescent Mental Health Clinic, San Gerardo Hospital, Monza, Italy. In our clinical sample there were two separate age groups: 20 children aged 2 to 6 years (12 males, 60%; mean age 3.8 ± 1.6) and 33 children aged 10 to 15 years (23 males, 70%; mean age 12.6 ± 1.5). These age groups correspond to two important stages of psychomotor development: from early childhood to the beginning of school age, when children have to cope with new social and academic demands, and from pre-adolescence to adolescence, when conflicts related to their identity and physical changes begin to emerge.

All children were born in the northern part of Italy. The ethnic distribution was homogeneous: a Mediterranean European origin was reported by 90% children in the younger group (where two parents were from Asia and one from Africa) and 94% in the older group (where two parents were from Eastern Europe and one from Central Europe). The socioeconomic level was evaluated using the Hollingshead index, with values ranging from 1 (low socioeconomic level) to 5 (highest socioeconomic level)[19]. The socioeconomic level of the children in the younger group had the following distribution: 73% level 4, 16% level 2, 11% level 3. The socioeconomic level of the children in the older group 2 had a similar distribution, with the majority of participants in the intermediate socioeconomic layers: 50% level 4, 25% level 2, 14% level 1, 11% level 3.

Written informed consent was obtained from the participants' parents or guardians. The study protocol was approved by the San Gerardo Hospital Ethics Committee.

Methods

A standardized battery of psychometric instruments was administered by trained psychologists, psychomotor therapists and neuropsychiatrists according to the instructions provided in the instruments' manuals.

Cognitive development was assessed using the Italian translation of the Griffiths Mental Development Scales (GMDS-ER) 2-8 for the younger group of children [20] and the Italian version of the WISC-IV for the older group of children [21]. Both instruments yield standardised z scores for the analysis of GMDS-ER General Quotient (GQ) and WISC-IV core subtests.

Adaptive functions were evaluated by administering the Vineland Adaptive Behavior Scales (VABS) to one of the parents [22]. The parents of one child in the younger group and seven children of the older group did not give their consent to complete the questionnaire; therefore VABS scores were collected for 19 children in the younger group and 26 children in the older group. The difference between chronological and mental age (DELTA parameter) was calculated in order to assess the patterns of change of adaptive skills with increasing age.

Statistical analysis

Data distribution was assessed by the Anderson-Darling test, Shapiro-Wilk test and Kolmogorov-Smirnov test. The non-parametric Wilcoxon test was used to compare the WISC-IV indices, with the exception of normally distributed scores, which were analysed using the paired samples t test. The bivariate correlation of Spearman was used to assess the correlation between continuous variables, whereas the relationship between DELTA and chronological age was analysed using the second-order polynomial regression model because of the quadratic trend in data distribution. The level of statistical significance was set at $p < 0.05$ and all statistical analyses were performed using the statistical software R version 3.0.1.

Results

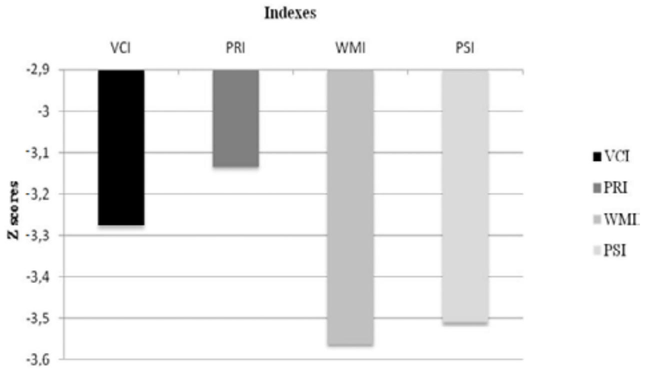
Cognitive functions

Griffiths Mental Development Scales (GMDS-ER) 2-8

All the subjects in group 1 had a GQ < -2.33, except for one who scored -1.77 (almost two standard deviations below the mean). Most of the children had a degree of intellectual disability between mild and moderate (14 children out of 20, 70%); 4 children (20%) had severe intellectual disability and 2 children (10%) had profound intellectual disability (Tab. I).

Wechsler Intelligence Scale for Children – Fourth Edition (WISC-IV)

Only 4 out of the 33 subjects had an IQ ≥ 40 (two children scored 40, one 43 and one 49). Most children's z scores were consistent with an intellectual disability range between mild and moderate according to DSM guidelines (26 children out of 33; 79%). The remaining 7 children (21%) had severe intellectual disability (Tab. II).



Abbreviations. VCI: Verbal Comprehension Index; PRI: Perceptual Reasoning Index; WMI: Working Memory Index; PSI: Processing Speed Index

FIGURE 1. *Four indexes of the Wechsler Intelligence Scale for Children – Fourth Edition (WISC-IV).*

Among the four indexes (Verbal Comprehension Index, VCI; Perceptual Reasoning Index, PRI; Working memory Index, WMI; and Processing Speed Index, PSI), the highest scores were reported in PRI, whereas the lowest scores were reported in WMI (Fig. 1).

The results of the Wilcoxon test showed statistically significant differences between PRI and WMI (p-value = 0.048), and between PRI and PSI (p-value = 0.006).

Adaptive functions

Vineland Adaptive Behavior Scales (VABS)

In group 1 the mean mental age was 2 years and 4 months, with an average difference between chrono-

TABLE I. *Degrees of intellectual disability in children with Down syndrome aged 2 to 6 years.*

Degrees of intellectual disability	IQ levels	z scores (mean)	Number of subjects	z scores of participants (mean ± sd)
Profound	< 20-25	< -5.0	2 (10%)	-7.7 ± 0.9
Severe	from 20-25 to 35-40	≥ -5.0 and < -4.0	4 (20%)	-4.8 ± 0.2
Moderate	from 35-40 to 50-55	≥ -4.0 and < -3.0	10 (50%)	-3.7 ± 0.3
Mild	from 50-55 to 70	≥ -3.0 and < -2.0	4 (20%)	-2.6 ± 0.4

TABLE II. *Degrees of intellectual disability in children with Down syndrome aged 10 to 15 years.*

Degrees of intellectual disability	IQ levels	z scores (mean)	Number of subjects	z scores of participants (mean ± sd)
Profound	< 20-25	< -5.0	0	
Severe	from 20-25 to 35-40	≥ -5.0 and < -4.0	7 (21%)	-4.1 ± 0.1
Moderate	from 35-40 to 50-55	≥ -4.0 and < -3.0	17 (52%)	-3.4 ± 0.2
Mild	from 50-55 to 70	≥ -3.0 and < -2.0	9 (27%)	-2.7 ± 0.3

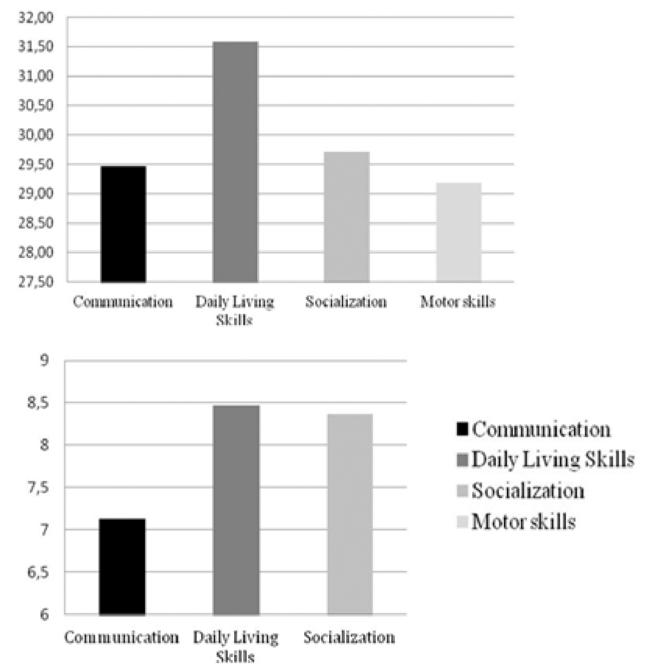


FIGURE 2 *(above). Vineland Adaptive Behavior Scales (VABS) scores in children with Down syndrome aged 2 to 6 years; (below). Vineland Adaptive Behavior Scales (VABS) scores in children with Down syndrome aged 10 to 15 years.*

TABLE III. *Mental age in scales and subscales of the Vineland Adaptive Behavior Scales (VABS).*

Scales and subscales	Mental age (in years) Mean ± SD
Communication	7.1 ± 2.8
- Receptive	7.0 ± 2.9
- Expressive	5.8 ± 2.3
- Written	8.0 ± 2.8
Daily Living Skills	8.5 ± 2.5
- Personal	7.3 ± 1.7
- Domestic	11.3 ± 3.4
- Community	7.1 ± 2.6
Socialization	8.4 ± 3.3
- Interpersonal Relationships	8.2 ± 4.7
- Play and Leisure Time	7.5 ± 3.5
- Coping Skills	9.2 ± 3.0
Mental age	7.5 ± 2.8

logical age and mental age of approximately 12 months (± 10.6 SD). In this group most children scored below the baseline on all scales (< 18 months) (Fig. 2; above). In group 2 the mean mental age was 7 years and 5 months, with an average difference between chronological age and mental age of approximately 5 years (± 2.0 SD). Figure 2 (below) and Table III show the scores reported in all scales and subscales, respectively (the Motor Skills scale was not analysed because this domain is assessed only in children younger than 6 years old, unless they present with motor skills deficit).

The Expression subscale was the most affected domain within the Communication scale. In the Daily Living Skills domain, both the Community subscale and the Personal subscale were more affected than the Domestic subscale. There were minor differences in the three subscales of the Socialization scale. Finally, the overall IQ deviation score was 113 (± 8.0 SD).

Clinical correlates of adaptive functions

Spearman bivariate correlation analysis showed a statistically significant positive correlation between DELTA values (difference between chronological and mental age) and chronological age (p-value = 0.000), whereas there was no statistically significant correlation between z scores and chronological age (p-value = 0.168). Results of the second-order polynomial regression analysis showed that DELTA values increase with chronological age until approximately 12 years, after which they become stable (Fig. 3).

Discussion

Children with DS have different degrees of intellectual disability and impairment in their capacity to cope with the demands of the social context in which they live. A both intellectual trajectories and life demands change

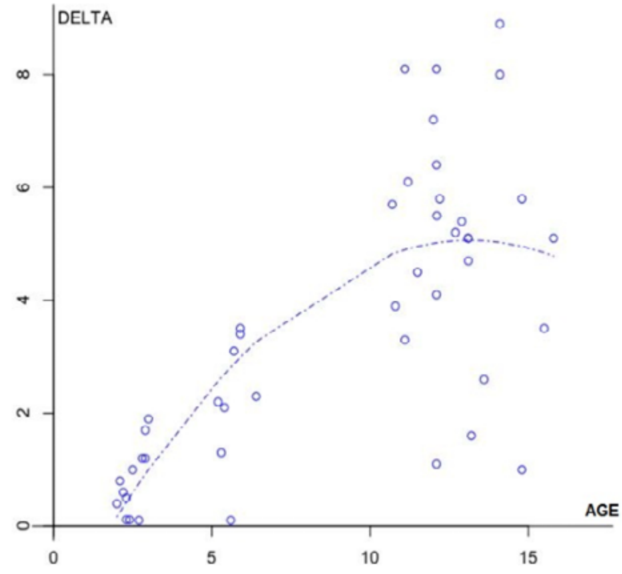


FIGURE 3. *Second-order polynomial regression analysis of DELTA values (difference between chronological and mental age) in children with Down syndrome.*

with age, this study investigated cognitive and adaptive functions in children with DS at different developmental stages. Our study groups had a degree of intellectual disability ranging from mild to severe, in line with existing data from other studies in the DS population [13]: 90% of children aged 2-6 years and 100% of children aged 10-15 years had a degree of disability which was rated as less than profound.

Moreover, results from the WISC-IV test in the older group of children with DS showed that the most severely affected cognitive domain was working memory. Specifically, children reported marked difficulties in those subtests (Digit Span and Letter Number Sequencing) that involve verbal short term memory. Conversely, PRI scores showed that the visuo-spatial component was relatively well preserved. These findings were consistent with the results of previous studies [23-27].

Although children with DS can have substantial language deficits [13,24,28], especially in verbal production[29], differences between verbal comprehension and perceptual reasoning performances in our study did not reach statistical significance. This could be due to the fact that the subtests that make up the VCI scale of the WISC-IV assess the comprehension component of language rather than verbal production. Consistent with the results of previous studies [3,13,14], we found a significant difference between comprehension and verbal production on the VABS: subjects had the lowest mental age in the communication scale, with the most pronounced impairment in the expressive subscale.

In terms of adaptive functions, our results replicated previous findings about higher levels of functioning within the daily living skills domain [3,14]. In our study, children with DS reported relatively high scores on measures of social competence related to daily living capabilities, which rely more heavily on implicit memory and are frequently expressed within the family context. Specifically, the mean mental age in the domestic subscale was relatively preserved, possibly due to the fact that children with DS remain dependent on the attachment figures longer than their peers, both physically and psychologically.

Children in our study reported higher scores on the socialization subscale than on the communication scale, contrary to the findings of the study by Dressler et al. [3]. These differences could be explained at least in part by the high level of social integration in our sample: all children were attending school and were involved in sports or recreational activities with peers during or after school. They also benefitted from a strong social network and continuing family support. The average deviation IQ score showed that the participants in our study had overall good adaptive functions within the population of individuals with intellectual disabilities.

Finally, our study explored the correlation between intellectual disability, adaptive functions and chronological age. We found that adaptive functions, but not cognitive deficits, were relatively more compromised in the older group of children: these findings suggest that the progressively widening disparity between the development of children with DS and those with typical development could be more significantly affected by the adaptation component than cognition. Specifically, we found that the difference between chronological age and mental age increases with age, until it comes to a plateau, at about 12 years of age. It is possible that the physical and psychological changes of pre-adolescence and adolescence, along with the increasing environmental demands (school and other activities), might play a negative role on the older children's adaptive skills. These results have important clinical implications, also in consideration of the new criteria for intellectual disabilities described in the DSM-5, which emphasize the need to assess the severity of impairment on the basis of adaptive functioning, rather than relying on IQ scores alone [1]. A better understanding of the functioning profile of young patients with DS, with particular attention to their skills in everyday life activities, would allow to implement more targeted intervention strategies to improve their health-related quality of life.

Our study has limitations. The cross-sectional design allowed us to describe the status of cognitive and adaptive skills at a single time point and to compare these data between two different age groups, however a longitudinal evaluation over time would have been optimal to evaluate the patterns of change with age. A second limitation of our study was the relatively small sample size, as well as its source: we enrolled only children from the northern part of Italy, who had been referred to the local health care services and whose medical history was fully known. Further studies with larger and more representative samples would be needed to confirm our findings using different research paradigms. Further limitations are related to the choice of the psychometric instruments. For the assessment of cognitive functions, we chose to use two different, yet comparable, tools because a single scale for the comprehensive assessment of cognitive development across childhood and adolescence is not currently available. However intelligence testing in children with intellectual disabilities poses considerable challenges, because the most widely used tools are highly subject to floor effect: having been designed for use in individuals with typical development, these psychometric instruments do not provide a sensitive measurement of cognition in the low ability range. As expected, we found a significant floor effect for both GMDS-ER and WISC-IV: these findings are consistent with the results from a previous study on children with fragile X syndrome and represent a major limitation of standardised intelligence testing [30]. Contrary to the WISC-IV and GMDS-ER, the VABS is a subjective tool assessing adaptive functions based on the parents' perspective, which can overestimate the difficulties of their children in a period of time in which the difference with their peers is more evident.

Despite these limitations, our findings that adaptive functions can be relatively more impaired than cognitive functions in older children with DS contribute to inform therapeutic interventions focused on strengths and weaknesses across different age groups.

Aknowledgments

The authors are grateful to the late Prof. Arturo Orsini for providing means and standard deviations of the Italian standardization sample for the 10 core subtests of the WISC-IV. Gratitude is also expressed to all participants and their families for their cooperation.

References

1. American Psychiatric Association. Diagnostic and statistical manual of mental disorders, 5th ed. Arlington, VA: American Psychiatric Publishing 2013.
2. Cocchi G, Gualdi S, Bower C, et al. International trends of Down syndrome 1993-2004: births in relation to maternal age and terminations of pregnancies. Birth Defects Res A Clin Mol Teratol 2010;88:474-9. https://doi.org/10.1002/bdra.20666
3. Dressler A, Perelli V, Feucht M, et al. Adaptive behaviour in Down syndrome: a cross-sectional study from childhood to adulthood. Wien Klin Wochenschr 2010;122:673-80. https://doi.org/10.1007/s00508-010-1504-0
4. Epstein CJ. Down syndrome. In: Scriver CR, Beaudet AL, Sly WS, et al., Eds. The metabolic basis of inherited disease. New York, NY: McGraw-Hill 1989, pp. 291-396.
5. Pennington BF, Moon J, Edgin J, et al. The neuropsychology of Down syndrome: evidence for hippocampal dysfunction. Child Dev 2003;74:75-93. https://doi.org/10.1111/1467-8624.00522
6. Vicari S, Bates E, Caselli MC, et al. Neuropsychological profile of Italians with Williams syndrome: an example of a dissociation between language and cognition? J Int Neuropsychol Soc 2004;10:862-76. https://doi.org/10.1017/s1355617704106073
7. Brown FR 3rd, Greer MK, Aylward EH, et al. Intellectual and adaptive functioning in individuals with Down syndrome in relation to age and environmental placement. Pediatrics 1990;85:450-2.
8. Carr J. Longitudinal research in Down syndrome. Int Rev Res Ment Retard 1992;18:197-223.
9. Crombie M, Gunn P. Early intervention, families, and adolescents with Down syndrome. Int J Disabil Dev Educ 1998;45:253-81.
10. Sigman M, Ruskin E. Continuity and change in the social competence of children with autism, Down syndrome, and developmental delays. Monogr Soc Res Child Dev 1999;64:1-114. https://doi.org/10.1111/1540-5834.00002
11. Hauser-Cram P, Warfield ME, Shonkoff JP, et al. Children with disabilities: a longitudinal study of child development and parent well-being. Monogr Soc Res Child Dev 2001;66:1-131.
12. Patterson T, Rapsey CM, Glue P. Systematic review of cognitive development across childhood in Down syndrome: implications for treatment interventions. J Intellect Disabil Res 2013;57:306-18. https://doi.org/10.1111/jir.12037
13. Vicari S. Motor development and neuropsychological patterns in persons with Down syndrome. Behav Genet 2005;36:355-64. https://doi.org/10.1007/s10519-006-9057-8
14. Dykens EM, Hodapp RM, Evans DW. Profiles and development of adaptive behavior in children with Down syndrome. Downs Syndr Res Pract 2006;9:45-50. https://doi.org/10.3104/reprints.293
15. Melyn MA, White DT. Mental and developmental milestones of noninstitutionalized Down's syndrome children. Pediatrics 1973;52:525-45.
16. Morgan SB. Development and distribution of intellectual and adaptive skills in Down syndrome children: implications for intervention. Ment Retard 1979;17:247-9.
17. Loveland KA, Kelley ML. Development of adaptive behavior in adolescents and young adults with autism and Down syndrome. Am J Ment Retard 1988;93:84-92.
18. Loveland KA, Kelley ML. Development of adaptive behavior in preschoolers with autism or Down syndrome. Am J Ment Retard 1991;96:13-20.
19. Hollingshead AA. Four-factor index of social status. New Haven, CT: Yale University 1975.
20. Cianchetti C, Sannio Fancello G, Eds. Griffiths Mental Development Scales, extended revised: 2-8 anni. Manuale di somministrazione. Firenze: Giunti O.S. 2006.
21. Orsini A, Pizzuti L, Picone L. Wechsler Intelligence Scale for Children-IV. Firenze: Giunti Organizzazioni Speciali 2004.
22. Sparrow SS, Balla DA, Cicchetti DV. Vineland Adaptive Behavior Scales. Intervista, forma completa. Giunti Organizzazioni Speciali, Firenze 2003.
23. Chapman RS, Hesketh LJ. Language, cognition, and short-term memory in individuals with Down syndrome. Downs Syndr Res Pract 2001;7:1-7. https://doi.org/10.3104/reviews.108
24. Seung HK, Chapman R. Digit span in individuals with Down syndrome and in typically developing children: temporal aspects. J Speech Lang Hear Res 2000;43:609-20. https://doi.org/10.1044/jslhr.4303.609
25. Seung HK, Chapman R. Sentence memory of individuals with Down's syndrome and typically developing children. J Intellect Disabil Res 2004;48:160-71. https://doi.org/10.1111/j.1365-2788.2004.00526.x
26. Kittler PM, Krinsky-McHale SJ, Devenny DA. Semantic and phonological loop effects on verbal working memory in middle-age adults with mental retardation. Am J Ment Retard 2004;109:467-80. https://doi.org/10.1352/0895-8017(2004)109<467:SAPLEO>2.0.CO;2.
27. Lanfranchi S, Mammarella IC, Carretti B, et al. How to improve spatial-simultaneous working memory in down syndrome: a computerized training. ECIDD, Sweden 2014.
28. Vicari S, Carlesimo GA. Short-term memory deficits are not uniform in Down and Williams syndromes. Neuropsychol Rev 2006;16:87-94. https://doi.org/10.1007/s11065-006-9008-4
29. Vicari S, Caselli MC, Tonucci F. Asynchrony of lexical and morphosyntactic development in children with Down syndrome. Neuropsychology 2000;38:634-44. https://doi.org/10.1016/s0028-3932(99)00110-4
30. Hessl D, Nguye DV, Green C, et al. A solution to limitations of cognitive testing in children with intellectual disabilities: the case of fragile X syndrome. J Neurodev Dis 2009;1:33-45. https://doi.org/10.1007/s11689-008-9001-8

JOURNAL OF PSYCHOPATHOLOGY 2021;27:90-98
DOI: 10.36148/2284-0249-379

Original article

Mental health and rural communities: prevalence of psychopathology among children and preadolescents in a mountainous area of Italy

Alessio Gori[1], Marco Giannini[1], Eleonora Topino[2], Marco Cacioppo[2], Giulia Palmieri[3], Elena De Sanctis[3], Yura Loscalzo[1], Catia Burgassi[4], Cristiana Manzi[4], Paola Giovannelli[4], Giulio Morganti[4], Mauro Camuffo[4], Giuseppe Craparo[5]

[1] Department of Health Sciences, University of Florence, Florence, Italy; [2] Department of Human Sciences, LUMSA University of Rome, Rome, Italy; [3] University of Florence, Florence, Italy; [4] AUSL Toscana Sud-Est (ex AUSL 9 Grosseto), Grosseto, Italy; [5] University of Enna "Kore", Enna, Italy

Received: February 26, 2020
Accepted: March 25, 2021

Correspondence
Alessio Gori
Department of Health Sciences, University of Florence, via di San Salvi 12, pad. 26, 50135 Florence, Italy. E-mail: alessio.gori@unifi.it

How to cite this article: Gori A, Giannini M, Topino E, et al. Mental health and rural communities: prevalence of psychopathology among children and preadolescents in a mountainous area of Italy. Journal of Psychopathology 2021;27:90-98. https://doi.org/10.36148/2284-0249-379

SUMMARY

Objective

There is substantial evidence of an increase in depression and other psychopathological aspects during adolescence, and this phenomenon is even more evident in rural contexts. Considering the predictive role that emotional disorders in youth could have on future mental health, this study aims to investigate differences in psychopathology between a group of children and pre-adolescent resident in a rural context (Amiata, Italy) and a homogeneous group by age of non-resident in a mountainous area.

Methods

This study involved teachers and parents for the assessment of 510 children and preadolescents (49.8% male, 50.2% female; Mean age = 8.44; SD = 1.44) from the rural context, and a normative sample of 1201 urban youth. Regarding the latter, the Child Behavior Checklist (CBCL) scores reported in the Achenbach manual in its Italian edition were considered, while for the "rural" sample, a group of parents and teachers children living in Amiata (Italy) completed respectively the Child Behavior Checklist (CBCL) and Teacher's Report-Form (TRF) scales.

Results

Firstly, results showed a good correlation between CBCL and TRF scales, indicating consistency in the parents' and teachers' detection of rural youth problems. Compared to the normative sample the group of residents in the mountainous area got higher scores on some of the CBCL scales, especially with regards to Social Problems, Cognitive Problems and Somatic Complaints. Finally, the Amiata girls obtained a lower total score on TRF and CBCL syndrome scales, and, in parallel, better academic competence that could be considered as a protective factor.

Conclusions

The comparisons of parents' reports with reports by others, such as teachers, seemed to be helpful for assessing the consistency of problems on syndromes such as anxiety/depression, somatic complaints, and attention problems to document the need for medical assessment or referral for mental health services. Specifically, the findings of this study suggest that subjects residing in the mountainous area tended to adopt fewer externalizing behaviors and they had more inner-directed disorders, especially somatic complaints: some aspects of youth psychopathological features can be influenced by a rural context of life. Therefore, the research analyzes the psychopathological features of children and pre-adolescents living in a rural environment (Amiata, Italy), highlighting important elements on which it may be important to intervene therapeutically to prevent the rural adolescent disorders, well documented by the Scientific literature.

Key words: emotional disorders, depression, adolescence, assessment, rural contexts

Introduction

Increases in prevalence rates of depression in adolescence have been found in clinical [1] and in community [2] samples, across different cultural settings [3]. Furthermore, more and more studies have identified depressive symptoms even in childhood, a condition that could be predictive of the subsequent depressive disorder (*e.g.,* Mazza and colleagues [4]). These increases have been found independent of gender, although girls have proven more likely to develop depression than boys [5]. The causes of emotional disorders and depression in childhood and adolescence are still not clear. Genetic [6], personality [7], hormonal [8], as well as cognitive [9] and interpersonal [10] factors have shown to be non-negligible predictors of these disorders. Besides, youth depression was found to be impacted by parenting style and parental mental health [11]. Specifically, paternal depression is associated with cognitive vulnerability to depression during middle childhood, a link that was also found with maternal criticism [12], closely related to psychopathological states: depressed mothers, in fact, perceive the behavior of their children more negatively than those who are not depressed [13]. Several studies indicate that stressful life events may have both short-term and long-term effects on the onset as well as on the course of depression [14]. There are also indications that stressful life events interact with gender and timing of pubertal transition [15], and that it may be more significant on the first onset than in the relapse of depressive episodes [16]. Thus, data from cross-sectional and longitudinal studies have documented a clear association between depression and recent stressful life events in adolescents, but also the predictive force of childhood symptoms on subsequent depressive disease [17,18]. However, prevention of depression in these life periods has received little attention in spite of the considerable evidence that depression symptomatology is not simply a transitory condition, but a pernicious form of the disorder that severely impacts with long-term consequences [19,20]. The prevalence of major depression disorders is of 1.9% in childhood and increases to 11.0-7.5% in adolescence (with rates of 3.0-2.3% for more severe conditions), depending on the age of the subjects and the instrument used for the assessment [21-23]. The implications of adolescent depression include developmental lags, suicidality, non-suicidal self-injury, and possible sensitization to recurrent depression [24,25]. Only a few longitudinal studies have nevertheless identified risk factors for emotional distress in adolescents [26] and none addressed screening for risk factors of depression or depressive symptoms. The risk factors for emotional distress in adolescents include environmental factors of school and other demographics, familial factors, and individual characteristics. More specifically, living in a rural as opposed to urban or suburban area, being female, and to have parents receiving welfare were demographic risk factors for emotional distress. Furthermore, feeling disconnected from school and family, combined with low self-esteem and lack of strong religious or spiritual beliefs were also risk factors [27]. Whereas demographic factors are easily identified, feelings of connectedness, spirituality and self-esteem are rather difficult to observe without formal testing. Study of rural adolescents has much to contribute to the field of adolescence and to social policy; recently, in fact, a lot of research about rural youth, within the sociological and psychological literature, has increased. In sociology, there is a long and distinguished tradition of research on rural-urban differences [28], but typically these studies have focused on adults or on the rural population as a whole, with little attention to adolescents as an important subgroup. Conversely, within psychology, adolescents are recognized as a distinct developmental subgroup, but there has been little attempt to determine how and to what extent rural youth differ from their urban and suburban counterparts. From an ecological perspective [29,30], differences would be expected, because rural settings differ from metropolitan settings in important ways, creating distinct contexts for development. Comparisons between rural and non-rural adolescents to date have pointed to several differences that have implications for the development of rural youth. Some of these represent a rural advantage (e.g., in terms of social capital), whereas others suggest that rural youth are at risk. Rural/non-rural comparisons of specific psychological dimensions yield somewhat equivocal findings. Several studies have examined differences in self-image, producing mixed results. Some research [31] found that rural adolescents have lower self-image than do their non-rural counterparts, while previous results did not find any differences [32] or at the opposite end, they found greater self-esteem in rural youth [33]. However, in more recent studies, there are also some suggestions that rural adolescents have more psychological symptoms than non-rural youth, such as depression and anxiety [34]. Indeed, as with metropolitan areas, depression in rural children and adolescents is related to family financial stress and low socioeconomic status [34]. Given that poverty rates are higher in rural areas than in non-rural areas, depression may be of particular concern for rural youth. Thus, based on the scientific literature proposed above, this study aims to investigate psychopathology in children and pre-adolescents resident in a rural context and comparing them to a group of children non-resident in a mountainous area.

Method

Participants and procedure

This study involved the evaluation of 510 children and preadolescents from a rural context, also comparing them with a normative sample of 1201 ones from an urban environment. The first ones (49.8% males, 50.2%

females) attended elementary and middle schools (M_{age} = 8.44; SD = 1.44; ranging 6-11 years) and were recruited from the rural context of Amiata area in Tuscany, while the others were a group of non-residents in a mountainous area, homogeneous by age to the first[35]. Regarding the latter, the Child Behavior Checklist (CBCL) scores reported in the Achenbach manual in its Italian edition were considered[35]. Concerning the "rural" sample, parents and teachers of these children completed, respectively, the Child Behavior Checklist (CBCL) and Teacher's Report-Form (TRF) scales. Each scale requires ten/fifteen minutes to be completed. The teachers completed 502 reports about their students (M_{age} = 8.46; SD = 1.45; age range 6-11), for a total of 255 females (M_{age} = 8.35; SD = 1.43; age range 6-11) and 247 males (M_{age} = 8.57; SD = 1.46; age range 6-11). The parents completed 446 reports about their children (M_{age} = 8.46; SD = 1.46; age range 6-11), for a total of 230 females (M_{age} = 8.37; SD = 1.44; age range 6-11) and 216 males (M_{age} = 8.56; SD = 1.47; age range 6-11): of these, 232 are among those who were also evaluated by a teacher.

Participants were recruited in the schools where data were collected: all the teachers and parents present at the time of administration joined. All participants provided information about age, sex and gender of the youth and completed an informed consent form after the intake assessment.

Measures

The *Achenbach System of Empirically Based Assessment* (ASEBA)[35-37] for school-age children is a standardized instrument including three measures for assessing emotional and behavioral problems: the Child Behavior Checklist (CBCL), Youth Self-Report (YSR), and Teacher's Report Form (TRF).

Specifically, in this study two of these questionnaires were used: the *Teacher's Report Form,* which evaluates behavioral problems that a child may display in school, and the *Child Behavior Checklist*, *i.e.* a parent measure that also evaluates behavioral problems a child may display and was collected at the same time as the TRF, which measures the same constructs. Both are considered to be extremely reliable measures of behavioral problems. Multi- axial system of CBCL and TRF includes a lot of information coming from different sources and it aims to give a complete description of competence and problems of each subject. This multi-axial assessment makes use also of additional instruments such as: neurological test, medical test, cognitive assessment, psychometric assessment and much more. Besides it includes different observers. So, it permits a five-axis evaluation: Axis 1) Parents' information; Axis 2) Teachers' information; Axis 3) Cognitive assessment; Axis 4) Physical conditions; Axis 5) Direct evaluation of subject.

Teacher's Report Form (TRF)

The TRF[36,37] is a behavioral assessment scale which is completed by teachers to obtain his/her perception of child's academic performance, adaptive functioning, and behavioral problems over the past two months in a standardized format. Time required to complete this scale is about ten minutes. The first section of the Teacher Report Form (TRF) requests background information about the student and the respondent's role at the school, and permit to explore the child's adaptive functioning, by asking to rate academic performance and four adaptive characteristics (Working Hard, Behaving, Learning and Happy). The remaining items comprise a behavioral problem checklist. These items consisted of three broad-band scales (Internalizing, Externalizing, and Total Problems) and eight syndrome scales (Withdrawal, Somatic Complaints, Anxious/Depressed, Social Problems, Thought Problems, Attention Problems, Rule-Breaking Behavior, and Aggressive Behavior)[35].

Child Behavior Checklist (CBCL)

The CBCL[35-37] is a standardized form that parents fill out to describe their children's behavioral and emotional problems. The version for ages 4 to 18 years (CBCL/4 to 18) includes competence and problems items. The problem items can be completed by most parents in about 10 minutes, and the (optional) competence items require an additional 5 to 10 minutes. Problem items on the CBCL/ 4 to 18 resemble those on the CBCL/2 to 3, except that parents rate the CBCL/4 to 18 problem items for the preceding 6 months instead of the 2 months specified on the CBCL/2 to 3. Competence items on the CBCL/4 to 18 assess the child's activities, social relations, and school functioning. The data obtained with the CBCL are summarized on a profile that displays the parent's ratings of each item. The profile also displays the child's standing on syndromes of problems that were derived from statistical analyses of CBCL's filled out for large numbers of clinically referred children. Each syndrome consists of problems that were found to occur concomitantly.

Data analysis

The statistical analyses were performed using the SPSS for Windows version 15.0. First, correlations between CBCL scales and TRF scales were calculated using the Pearson's r coefficient. Descriptive statistics, including means and standard deviations, were calculated to characterize the total sample and subsamples based on gender variable. Therefore, a One-Sample T-Test statistical analysis was used to compare the CBCL scores between the "rural" youth and the "urban" ones of the normative sample[35]. Moreover, to explore the differences in the CBCL and TRF scores based on gender, an Independent

Samples T-Test was carried out. Results were regarded as statistically significant for p-values < 0.05.

Results

To verify correlation between CBCL and TRF scales we used the Pearson's *r* coefficient. Table I indicates that Total Capacity scale of TRF correlates with Total Competence of CBCL ($r = .24$, $p < 0.01$).

TRF's Internalizing scale and CBCL's Internalizing scale demonstrate a sufficient correlation ($r = .16$, $p < 0.01$), instead there is a good correlation among Externalizing scale of each questionnaires ($r = .39$, $p < 0.01$); Total Score of TRF demonstrates a high correlation with Total score of CBCL ($r = .33$, $p < 0.01$). Furthermore, there are negative correlations between Attention Problems scale of CBCL and Working Hard of TRF ($r = -.43$, $p < 0.01$), and between CBCL's Attention Problems and Scholastic Behavior of TRF ($r = -.41$, $p < 0.01$). Finally, the two scales Attention Problem of each questionnaire show a strong correlation ($r = .47$, $p < 0.01$).

In Table II were reported Means and Standard Deviations for the CBCL scores concerning the total sample, while in Table III and IV were showed those of the CBCL and TRF scores regarding the "rural" subsamples based on gender. Concerning the differences between the rural and normative samples in CBCL scales, the first ($M = 20.83$, $SD = 15.92$) showed significantly lower Total

TABLE I. *Correlations between CBCL scales and TRF scales in the rural sample.*

	CBCL-A	CBCL-SO	CBCL-SC	CBCL-TOT	CBCL-A/D	CBCL-W	CBCL-S/C	CBCL-S/P	CBCL-T/P	CBCL-A/P	CBCL-R/B	CBCL-A/B	INT	EXT	TOT
TRF-Acc/P	.095	.231**	.339**	.205**	-.170**	-.036	-.136**	-.236**	-.169**	-.307**	-.237**	-.116*	-.166**	-.160**	-.233**
TRF-W/H	.056	.204**	.346**	.168**	-.115*	-.060	-.225**	-.190**	-.170**	-.433**	-.313**	-.291**	-.177**	-.325**	-.330**
TRF-B	.023	.156**	.232**	.113*	-.078	-.056	-.106*	-.152**	-.089	-.413**	-.269**	-.293**	-.105*	-.314**	-.276**
TRF-L	.088	.205**	.341**	.200**	-.197**	-.091	-.166**	-.252**	-.197**	-.374**	-.316**	-.205**	-.210**	-.255**	-.311**
TRF-H	-.055	.101	.140**	.034	-.073	-.097	-.079	-.137**	-.064	-.125*	-.097	-.017	-.107*	-.040	-.097
TRF-TOT	.096	.272**	.313**	.236**	-.088	-.049	-.165**	-.195**	-.081	-.405**	-.269**	-.225**	-.137**	-.260**	-.277**
TRF-A/D	-.047	-.121*	-.249**	-.109*	.071	.062	.186**	.164**	.015	.108*	.117*	.115*	.137**	.127**	.146**
TRF-W	-.085	-.193**	-.269**	-.179**	.054	.075	.219**	.142**	.028	.079	.084	.072	.145**	.083	.115*
TFR-S/C	-.017	-.056	-.177**	-.052	.045	.031	.159**	.121*	.040	.181**	.104*	.083	.102*	.097*	.135**
TRF-S/P	-.075	-.177**	-.345**	-.159**	.101*	.079	.139**	.268**	.061	.259**	.194**	.174**	.142**	.196**	.221**
TRF-T/P	-.061	-.148**	-.229**	-.130*	.134**	.107*	.220**	.241**	.116*	.337**	.310**	.320**	.201**	.349**	.331**
TRF-Att/P	-.023	-.184**	-.384**	-.133*	.105*	.027	.168**	.245**	.125**	.472**	.300**	.327**	.139**	.352**	.326**
TRF-R/B	.066	-.132**	-.272**	-.038	.127**	.044	.217**	.188**	.173**	.371**	.295**	.339**	.178**	.361**	.317**
TRF-A/B	.011	-.142**	-.278**	-.081	.090	.027	.169**	.259**	.107*	.365**	.299**	.361**	.130**	.379**	.310**
INT	-.067	-.166**	-.297**	-.151**	.071	.079	.235**	.178**	.026	.128**	.122*	.111*	.162**	.125**	.158**
EXT	.029	-.145**	-.289**	-.071	.106*	.034	.192**	.250**	.132**	.384**	.312**	.371**	.152**	.391**	.327**
TOT	-.028	-.194**	-.384**	-.141**	.115*	.055	.223**	.276**	.116*	.406**	.299**	.324**	.176**	.349**	.329**

$^{*}p < .05$ $^{**}p < .01$

TRF-Acc/P: Academic Performance; TRF-W/H: Working Hard; TRF-B: Behaving; TRF-L: Learning; TRF-H: Happy; TRF-TOT: Total of Competence; TRF-A/D: Anxious/Depressed; TRF-W: Withdrawal; TRF-S/C: Somatic Complaints; TRF-S/P: Social Problems; TRF-T/P: Thought Problems; TRF-Att/P: Attention Problems; TRF-R/B: Rule-Breaking Behavior; TRF-A/B: Aggressive Behavior; INT: Internalizing; EXT: Externalizing; TOT: Total Problems; CBCL-A: Activities; CBCL-SO: Social; CBCL-SC: School; CBCL-TOT: Total of Competence; CBCL-A/D: Anxious/Depressed; CBCL-W: Withdrawal; CBCL-S/C: Somatic Complaints; CBCL-S/P: Social Problems; CBCL-T/P: Thought Problems; CBCL-A/P: Attention Problems; CBCL-R/B: Rule-Breaking Behavior; CBCL-A/B: Aggressive Behavior; INT: Internalizing; EXT: Externalizing; TOT: Total Problems

TABLE II. *Comparison of CBCL scores between rural sample and normative sample.*

Variable	mean ± SD		T	*P*
	Rural sample (N_{total} = 510)*	Normative sample N_{total} = 1201		
CBCL-A	6.14 ± 3.32	6.40 ± 1.75	-1.52	ns
CBCL-SO	6.87 ± 2.24	6.90 ± 2.05	-0.30	ns
CBCL-SC	5.14 ± 0.64	5.20 ± 5.78	-1.88	ns
CBCL-TOT	18.20 ± 5.03	18.60 ± 3.46	-1.56	ns
CBCL-A/D	2.81 ± 2.97	3.25 ± 3.21	-3.11	.01
CBCL-W	1.16 ± 1.59	1.90 ± 1.95	-9.71	.001
CBCL-S/C	1.75 ± 2.13	0.90 ± 1.47	8.37	.001
CBCL-S/P	2.39 ± 2.31	1.95 ± 1.80	3.97	.001
CBCL-T/P	1.00 ± 1.53	0.50 ± 0.95	6.92	.001
CBCL-A/P	3.09 ± 2.98	2.89 ± 3.22	1.40	ns
CBCL-R/B	1.19 ± 1.49	1.39 ± 1.57	-2.88	.01
CBCL-A/B	4.32 ± 4.42	7.58 ± 5.53	-15.53	.001
INT	5.71 ± 5.16	5.91 ± 5.14	-0.80	ns
EXT	5.50 ± 5.38	8.97 ± 6.60	-13.56	.001
TOT	20.83 ± 15.92	23.63 ± 15.56	-3.70	.001

** N_{range}: 379-446; Missing responses: N = 46 for the scale CBCL-A; N = 28 for the scale CBCL-SO; N = 18 for the scale CBCL-SC; N = 67 for the scale CBCL-TOT; N = 3 for the scale CBCL-A/D; N = 3 for the scale CBCL-W; N = 4 for the scale CBCL-S/C; N = 3 for the scale CBCL-S/P; N = 3 for the scale CBCL-T/P; N = 3 for the scale CBCL-A/P; N = 4 for the scale CBCL-R/B; N = 4 for the scale CBCL-A/B; N = 4 for the scale INT; N = 4 for the scale EXT; N = 4 for the scale TOT.*

CBCL-A: Activities; CBCL-SO: Social; CBCL-SC: School; CBCL-TOT: Total of Competence; CBCL-A/D: Anxious/Depressed; CBCL-W: Withdrawal; CBCL-S/C: Somatic Complaints; CBCL-S/P: Social Problems; CBCL-T/P: Thought Problems; CBCL-A/P: Attention Problems; CBCL-R/B: Rule-Breaking Behavior; CBCL-A/B: Aggressive Behavior; INT: Internalizing; EXT: Externalizing; TOT: Total Problems.

scores than the urban sample (*M* = 23.63, *SD* = 15.56): *t*(442) = -3.70, *p* > 0.001 (see Table II).

Rule-Breaking Behavior scale (t_{441} = -2.88, *p* < 0.01), Aggressive behaviour scale (t_{442} = -15.53, *p* < 0.001), and Externalizing scale (t_{442} = -13.56, *p* < 0.001) were found significantly lower in rural sample (*M* = 1.19, *SD* = 1.49; *M* = 4.32, *SD* = 4.42; *M* = 5.50, *SD* = 5.38, respectively), than in the normative one (*M* = 1.39, *SD* = 1.57; *M* = 7.58, *SD* = 5.53, *M* = 8.97, *SD* = 6.60, respectively). Contrary, significantly higher scores on Somatic Complains scale (t_{441} = 8.37, *p* < 0.001), Social Problems scale (t_{442} = 3.97, *p* < 0.001) and Cognitive Problems scale (t_{442} = 6.92, *p* < 0.001) were shown in the rural sample (*M* = 1.75, *SD* = 2.13; *M* = 2.39, *SD* = 2.31; *M* = 1.00, *SD* = 1.53, respectively), compared whit the normative one (*M* = 0.90, *SD* = 1.47; *M* = 1.95, *SD* = 1.80; *M* = 0.50, *SD* = 0.95, respectively).

Finally, in the Anxious/Depressed (t_{442} = -3.11, *p* < 0.01) and Withdrawal (t_{442} = -9.71, *p* < 0.001) scales the rural sample (*M* = 2.81, *SD* = 2.97; *M* = 1.16, *SD* = 1.59, respectively) obtained higher scores than the normative one (*M* = 3.25, *SD* = 3.21; *M* = 1.90, *SD* = 1.95, respectively).

Comparing scores by gender, we found that boys (*M* = 7.10, *SD* = 2.28) obtained higher scores than girls (*M* = 6.66, *SD* = 2.19) on CBCL's Social scale: *t*(416) = 2.02, *p* < 0.05, see Table III). Furthermore, the CBCL's total scores of competence where significantly higher in males (*M* = 18.72, *SD* = 5.07) than in females (*M* = 17.71, *SD* = 4.96): *t*(377) = 1.95, *p* < 0.05. Besides, boys (*M* = 3.59, *SD* = 3.19) showed significantly higher scores in Attention Problems than girls (*M* = 2.62, *SD* = 2.69): *t*(441) = 3.47, *p* < 0.001.

Regarding TRF scales (see Table IV), significant lower scores on Working hard scale (t_{496} = - 2.96, *p* < 0.01), Behaving scale (t_{493} = - 6.71, *p* < 0.001) and Total Competence scale (t_{425} = -3.15, *p* < 0.01) were found in male subsample (*M* = 3.90, *SD* = 1.32; *M* = 3.77, *SD* = 1.24; *M* = 15.76, *SD* = 3.54, respectively), compared with the female one (*M* = 4.24, *SD* = 1.26; *M* = 4.50, *SD* = 1.18; *M* = 16.87, *SD* = 3.71, respectively). Male subsample got mean scores significantly higher on all of TRF syndrome scales, except for Withdrawal. Consistently, no significant gender differences were found in Internalizing scores: *t*(499) = 1.59, *p* = .112.

TABLE III. *Comparison of CBCL scores between male and female youth belonging to the rural sample.*

Variable	Gender		T	*P*
	Male (N_{total} = 216)*	Female (N_{total} = 230)°		
CBCL-A	6.40 ± 3.38	5.92 ± 3.26	1.45	ns
CBCL-SO	7.10 ± 2.28	6.66 ± 2.19	2.02	.05
CBCL-SC	5.11 ± 0.69	5.17 ± 0.60	-0.91	ns
CBCL-TOT	18.72 ± 5.07	17.71 ± 4.96	1.95	.05
CBCL-A/D	2.77 ± 3.05	2.85 ± 2.92	-0.27	ns
CBCL-W	1.19 ± 1.57	1.14 ± 1.62	0.28	ns
CBCL-S/C	1.68 ± 2.02	1.82 ± 2.23	-0.69	ns
CBCL-S/P	2.48 ± 2.43	2.30 ± 2.20	0.80	ns
CBCL-T/P	1.09 ± 1.53	0.92 ± 1.53	1.21	ns
CBCL-A/P	3.59 ± 3.19	2.62 ± 2.69	3.47	.001
CBCL-R/B	1.30 ± 1.59	1.07 ± 1.38	1.62	ns
CBCL-A/B	4.69 ± 4.53	3.98 ± 4.30	1.69	ns
INT	5.61 ± 4.93	5.81 ± 5.37	-0.40	ns
EXT	5.99 ± 5.58	5.05 ± 5.16	1.85	ns
TOT	21.72 ± 15.75	19.99 ± 16.07	1.15	ns

** N_{range}: 183-214; Missing responses: N = 23 for the scale CBCL-A; N = 16 for the scale CBCL-SO; N = 7 for the scale CBCL-SC; N = 33 for the scale CBCL-TOT; N = 2 for the scale CBCL-A/D; N = 2 for the scale CBCL-W; N = 3 for the scale CBCL-S/C; N = 2 for the scale CBCL-S/P; N = 2 for the scale CBCL-T/P; N = 2 for the scale CBCL-A/P; N = 2 for the scale CBCL-R/B; N = 2 for the scale CBCL-A/B; N = 2 for the scale INT; N = 2 for the scale EXT; N = 2 for the scale TOT.*

° N_{range}: 196-229; Missing responses: N = 23 for the scale CBCL-A; N = 12 for the scale CBCL-SO; N = 11 for the scale CBCL-SC; N = 34 for the scale CBCL-TOT; N = 1 for the scale CBCL-A/D; N = 1 for the scale CBCL-W; N = 1 for the scale CBCL-S/C; N = 1 for the scale CBCL-S/P; N = 1 for the scale CBCL-T/P; N = 1 for the scale CBCL-A/P; N = 2 for the scale CBCL-R/B; N = 1 for the scale CBCL-A/B; N = 1 for the scale INT; N = 1 for the scale EXT; N = 1 for the scale TOT.

CBCL-A: Activities; CBCL-SO: Social; CBCL-SC: School; CBCL-TOT: Total of Competence; CBCL-A/D: Anxious/Depressed; CBCL-W: Withdrawal; CBCL-S/C: Somatic Complaints; CBCL-S/P: Social Problems; CBCL-T/P: Thought Problems; CBCL-A/P: Attention Problems; CBCL-R/B: Rule-Breaking Behavior; CBCL-A/B: Aggressive Behavior; INT: Internalizing; EXT: Externalizing; TOT: Total Problems.

Discussion

The rural environment exposes a series of risk factors for psychiatric disorders that endanger the mental health of its residents [38] and many stressors that are not present in urban areas: geographic isolation, for example, limits social networks and community resources, including healthcare [39]. In particular, the many barriers to obtaining essential mental-health services in rural further areas complicates the early identification and treatment of psychological symptoms. Several studies (see, for a review, Smalley and colleagues [40]) described how the unique characteristics of rural areas contribute to the difficulty in accessing care and services. Rural areas are often less affluent and are less populous: this limits the government allocations and, consequently, the service provisions in schools, which are a traditional avenue for families who would not otherwise receive assistance in mental health. In addition, there is a shortage of mental-health professionals, which tend to be concentrated in urban areas because of client availability and professional contacts, thereby limiting access of rural residents to needed services. Furthermore, there is often a difficulty in accessing these services both in terms of mobility and financial constraints. Finally, in addition to these "tangible" barriers, there are also the "perceived" ones, such as the limited insight into the usefulness of mental-health services and stigma [41]: the latter is more difficult to fight, because the prejudice towards mental health problems is internalized at such a level that people are convinced that they have to overcome their difficulties alone. This creates a vicious circle in which individuals feel isolated and powerless [42]. It is therefore clear that without better access to mental health care, mental health problems will worsen with negative consequences on the quality of life of patients, their families, and the community [40]. Thus, there is a strong case for the development of a method to identify rural children and adolescents at risk for depression. In an age of cost-effectiveness and time limitations, this article will provide a map to easily identify those adolescents who could benefit from an intervention to prevent depression

TABLE IV. *Comparison of TRF scores between male and female youth belonging to the rural sample.*

Variable	Gender		T	*P*
	Male (N_{total} = 247)*	Female (N_{total} = 255)°		
TRF-Acc/P	3.03 ± 0.74	3.07 ±.71	-0.50	ns
TRF-W/H	3.90 ± 1.32	4.24 ± 1.26	-2.96	.01
TRF-B	3.77 ± 1.24	4.50 ± 1.18	-6.71	.001
TRF-L	4.07 ± 1.24	4.15 ± 1.24	-0.65	ns
TRF-H	3.99 ± 0.78	4.10 ± 0.89	-1.32	ns
TRF-TOT	15.76 ± 3.54	16.87 ± 3.71	-3.16	.01
TRF-A/D	2.52 ± 3.32	1.95 ± 2.79	2.10	.05
TRF-W	1.16 ± 2.02	1.22 ± 2.19	-0.29	ns
TFR-S/C	0.53 ± 1.18	0.27 ± 0.71	3.03	.01
TRF-S/P	1.77 ± 2.58	1.31 ± 2.33	2.10	.05
TRF-T/P	0.52 ± 1.27	0.26 ± 1.02	2.50	.05
TRF-Att/P	8.92 ± 9.88	4.65 ± 7.69	5.39	.001
TRF-R/B	1.72 ± 2.47	0.76 ± 1.81	4.93	.001
TRF-A/B	4.02 ± 6.45	1.47 ± 3.54	5.50	.001
INT	4.19 ± 5.43	3.46 ± 4.74	1.59	ns
EXT	5.64 ± 8.36	2.24 ± 4.92	5.51	.001
TOT	21.45 ± 23.58	12.34 ± 18.17	4.83	.001

** N_{range}: 212-247; Missing responses: N = 3 for the scale TRF-Acc/P; N = 3 for the scale TRF-W/H; N = 4 for the scale TRF-B; N = 5 for the scale TRF-L; N = 35 for the scale TRF-H; N = 35 for the scale TRF-TOT; N = 1 for the scale INT; N = 1 for the scale EXT, N = 1 for the scale TOT;*

° N_{range}: 214-255; Missing responses: N = 2 for the scale TRF-Acc/P; N = 1 for the scale TRF-W/H; N = 3 for the scale TRF-B; N = 5 for the scale TRF-L; N = 40 for the scale TRF-H; N = 41 for the scale TRF-TOT; N = 1 for the scale TOT.

TRF-Acc/P: Academic Performance; TRF-W/H: Working Hard; TRF-B: Behaving; TRF-L: Learning; TRF-H: Happy; TRF-TOT: Total of Competence; TRF-A/D: Anxious/Depressed; TRF-W: Withdrawal; TRF-S/C: Somatic Complaints; TRF-S/P: Social Problems; TRF-T/P: Thought Problems; TRF-Att/P: Attention Problems; TRF-R/B: Rule-Breaking Behavior; TRF-A/B: Aggressive Behavior; INT: Internalizing; EXT: Externalizing; TOT: Total Problem.

and/or further monitor for depressive symptom development. Amiata, the location of this study, represents a large and important area characterized by a rural way of life, in a central region of Italy. This research provides that, in the rural sample, there are good correlations between CBCL and TRF scales, so information given by teachers and parents are in line with each other.

Compared to the normative sample, we found that CBCL's total scores indicates a lower level of psychological diseases among rural youth, although some scales are more problematic in this latter sample: more specifically, Amiata's sample got higher scores on Social Problems, Cognitive Problems and Somatic Complaints scales. These can be read as expression of a discomfort that could be lower or latent in rural childhood and may become relevant in adolescence. In other words, the myth that rural life is idyllic and stress-free has begun to be dispelled [43] or conceptualized as a protective factor only in very first years of a children life: stressful life events that are commonly experienced by rural families were positively associated with feelings of depression and worthlessness. Indeed, a previous study about rural way of life by Burgassi and colleagues [31] shows significant levels of depression in middle school students, specifically in scores of irritable mood, insecurity, low self-esteem and guilt. Furthermore, it also highlighted that parents reported a lower level of problems than their children, a result that could indicate a tendency to minimize and speak less about mental suffering in rural environments [44]. Consistently, scientific literature [45] reported higher levels of depressive symptoms in rural adolescents than urban peers, and this appears to be related to personal and interpersonal variables, such as gender, low family support, parent-child conflict, negative friend behaviors and negative peer relationships.

Furthermore, data showed that scores on Aggressive Behavior and Externalizing scales were inferior than in the normative sample; it demonstrates that our rural sample should be less action oriented, in accordance with previous research [31].

Comparing male and female subsample we have discovered that Amiata's girls obtained lower total score at TRF and CBCL syndrome scales. This is not in line with literature [5]: perhaps it demonstrates better girl's strategies to handle possible difficulties of rural environment. Furthermore, girls have demonstrated better academic competence that could be considered as protection factors for emotional and behavioral disorders [46].

This research has several limitations that should be considered. First, a unique and coherent definition of "rural" has not been established yet [47]: the present study determined rurality on the basis of geographical and social characteristics. However, this limit could hamper the generalizability of the results and future research could overcome this problem with more varied rural samples and cross-cultural analyses. Furthermore, the rural sample of this study was compared with a normative urban one. Future research could get two different samples (rural and urban) in a common period of time and longitudinally or cross-sectionally analyse them. Finally, in light of the stigma against mental health problems documented by the scientific literature in rural environment (*e.g.,* Parr and colleagues [44]), it is important to highlight that social desirability bias have not been controlled, so participants may have tried to give answers that would make them look good to the researcher. However, the results seem to show coherence between the different perspectives (*e.g.,* teachers and parents) and they underline several problem areas on which it may be important to intervene. Indeed, as discussed above, the adolescent period is a vulnerable to depression and this risk is exacerbated in rural environments [45]. Hence, early interventions focusing on the first symptoms that this study highlighted in the childhood and pre-adolescence phases could be fundamental to prevent subsequent more serious disorders.

Conclusions

In conclusion, the comparisons between parents' reports and the one given by others, such as teachers, seem to be helpful for assessing the consistency of problems on syndromes such as an anxiety/depression, somatic complaints, and attention problems to document the need for medical assessment or referral for mental health services. Specifically, in the present study, correlations between the CBCL and TRF scales have demonstrated the capability of the two forms to identify children's behavioral and emotional problems. Moreover, Amiata children have showed higher levels of somatic complaints, together with social and thought problems. This suggests that some aspects of adolescent depression may have origins in youth problems linked to a rural context of life.

Ethical consideration

The study was conducted in accordance with the recent and international ethical standards for research involving human subjects.

Acknowledgement

None.

Funding

This research received no external funding.

Conflict of interest

The Authors declare no conflict of interest

Author contributions

Conceptualization: AG and MG;
Methodology: AG, MG and ET;
Formal analysis: ET and AG;
Investigation: AG, MC, MG, YL, CB, CM, PG, GM and MC;
Data curation: CB, CM, PG, GM and MC;
Writing – original draft preparation: GP, EDS and ET;
Writing – review and editing: ET, AG and YL;
Supervision: AG, MG, GC and MC
All authors have read and agreed to the published version of the manuscript.

References

1. Williamson DE, Birmaher B, Frank E, et al. Nature of life events and difficulties in depressed adolescents. J Am Acad Child Adolesc Psychiatry 1998;37:1047-1057. https://doi.org/10.1097/00004583-199810000-00015
2. Mojtabai R, Olfson M, Han B. National trends in the prevalence and treatment of depression in adolescents and young adults. Pediatrics 2016;138:e20161878. https://doi.org/10.1542/peds.2016-1878
3. Weissman MM, Bland RC, Canino GJ, et al. Crossnational epidemiology of major depression and bipolar disorder. JAMA 1996;276:293-299. https://doi.org/10.1001/jama.1996.03540040037030
4. Mazza JJ, Abbott RD, Fleming CB, et al. Early predictors of adolescent depression: a 7-year longitudinal study. J Early Adolesc 2009;29:664-692. https://doi.org/10.1177/0272431608324193
5. Wade TJ, Cairney J, Pevalin DJ. Emergence of gender differences in depression during adolescence: National panel results from three countries. J Am Acad Child Adolesc Psychiatry 2002;41:190-198. https://doi.org/10.1097/00004583-200202000-00013
6. Rice F, Riglin L, Thapar AK, et al. Characterizing developmental trajectories and the role of neuropsychiatric genetic risk variants in early-onset depression. JAMA 2019. https://doi.org/10.1016/j.euroneuro.2018.08.402
7. Pruneti C, Fontana F, Fante C, et al. Corrispondenza tra manifestazioni depressive e tratti di personalità in un campione di studenti di scuola media superiore [Correspondence between depressive symptomatology and personality traits in a sample of high school students]. Journal of Psychopathology 2012;18:35-39.
8. Ellis R, Fernandes A, Simmons JG, et al. Relationships between adrenarcheal hormones, hippocampal volumes and depressive symptoms in children. Psychoneuroendocrinology 2019;104:55-63. https://doi.org/10.1016/j.psyneuen.2019.02.016
9. Kertz SJ, Petersen DR, Stevens KT. Cognitive and attentional vulnerability to depres-

sion in youth: a review. Clin Psychol Rev 2019;71:63-77. https://doi.org/10.1016/j.cpr.2019.01.004

[10] Fiorilli C, Capitello TG, Barni D, et al. Predicting adolescent depression: the interrelated roles of self-esteem and interpersonal stressors. Front Psychol 2019;10. https://doi.org/10.3389/fpsyg.2019.00565

[11] Bufferd SJ, Dougherty LR, Olino TM, et al. Predictors of the onset of depression in young children: a multi-method, multi-informant longitudinal study from ages 3 to 6. J Child Psychol Psychiatry 2014;55:1279-1287. https://doi.org/10.1111/jcpp.12252

[12] Hayden EP, Olino TM, Mackrell SV, et al. Cognitive vulnerability to depression during middle childhood: Stability and associations with maternal affective styles and parental depression. Pers Individ Dif 2013;55:892-897. https://doi.org/10.1016/j.paid.2013.07.016

[13] Oyserman D, Bybee D, Mowbray C. Influences of maternal mental illness on psychological outcomes for adolescent children. J Adolesc 2002;25:587-602. https://doi.org/10.1006/jado.2002.0506

[14] Hamilton JL, Stange JP, Abramson LY, et al. Stress and the development of cognitive vulnerabilities to depression explain sex differences in depressive symptoms during adolescence. Clin Psychol Sci 2015;3:702-714. https://doi.org/10.1177/2167702614545479

[15] Conley CS, Rudolph KD, Bryant FB. Explaining the longitudinal association between puberty and depression: sex differences in the mediating effects of peer stress. Dev Psychopathol 2012;24:691-701. https://doi.org/10.1017/s0954579412000259

[16] Elsayed NM, Fields KM, Olvera RL, et al. The role of familial risk, parental psychopathology, and stress for first-onset depression during adolescence. J Affect Disord 2019;253:232-239. https://doi.org/10.1016/j.jad.2019.04.084

[17] Luby JL, Gaffrey MS, Tillman R, et al. Trajectories of preschool disorders to full DSM depression at school age and early adolescence: continuity of preschool depression. Am J Psychiatry 2014;171:768-776. https://doi.org/10.1176/appi.ajp.2014.13091198

[18] Twenge JM, Nolen-Hoeksema S. Age, gender, race, socioeconomic status, and birth cohort difference on the children's depression inventory: a meta-analysis. J Abnorm Psychol 2002;111:578. https://doi.org/10.1037/0021-843x.111.4.578

[19] Garaigordobil M, Bernarás E, Jaureguizar J, et al. Childhood depression: relation to adaptive, clinical and predictor variables. Front Psychol 2017;8:821. https://doi.org/10.3389/fpsyg.2017.00821

[20] Keenan-Miller D, Hammen CL, Brennan PA. Health outcomes related to early adolescent depression. J Adolesc Health 2007;41:256-262. https://doi.org/10.1016/j.jadohealth.2007.03.015

[21] Kelleher KJ, Taylor JL, Rickert VI. Mental health services for rural children and adolescents. Special Issue: child and adolescent mental health. Clin Psychol Rev 1992;2:841-852. https://doi.org/10.1016/0272-7358(92)90005-s

[22] Wittchen HU, Nelson CB, Lachner G. Prevalence of mental disorders and psychosocial impairments in adolescents and young adults. Psychol Med 1998;28:109-126. https://doi.org/10.1017/s0033291797005928

[23] Avenevoli S, Swendsen J, He JP, et al. Major depression in the national comorbidity survey–adolescent supplement: prevalence, correlates, and treatment. J Am Acad Child Adolesc Psychiatry 2015;54:37-44. https://doi.org/10.1016/j.jaac.2014.10.010

[24] Orri M, Galera C, Turecki G, et al. Association of childhood irritability and depressive/anxious mood profiles with adolescent suicidal ideation and attempts. JAMA psychiatry 2018;75:465-473. https://doi.org/10.1001/jamapsychiatry.2018.0174

[25] Gatta M, Rago A, Dal Santo F, et al. Non-suicidal self-injury among Northern Italian high school students: emotional, interpersonal and psychopathological correlates. Journal of Psychopathology 2016;22:185-190.

[26] Galambos NL, Leadbeater BJ, Barker ET. Gender differences in and risk factors for depression in adolescence: a 4-year longitudinal study. Int J Behav Dev 2004;28:16-25. https://doi.org/10.1080/01650250344000235

[27] Rasic D, Kisely S, Langille DB. Protective associations of importance of religion and frequency of service attendance with depression risk, suicidal behaviours and substance use in adolescents in Nova Scotia, Canada. J Affect Disord 2011;132:389-395. https://doi.org/10.1016/j.jad.2011.03.007

[28] Adair-Toteff C. Ferdinand Tonnies: utopian visionary. Sociol Theory 1994;13:58-65. https://doi.org/10.2307/202006

[29] Bronfenbrenner U. The ecology of human development: experiments by nature and design. Cambridge, MA: Harvard University Press 1979.

[30] Ianni FA. The search for structure: a report on American youth today. New York: Free Press 1989.

[31] Burgassi C, Giannini M, Giovannelli P, et al. Il disagio adolescenziale nel territorio dell'Amiata grossetana. Nuova Rassegna Di Studi Psichiatrici 2017;15 (http://www.nuovarassegnastudipsichiatrici.it/index.php/numeri-precedenti/volume-15/disagio-adolescenziale-territorio-amiata-grossetana. Accessed February 25, 2020).

[32] Prendergrast P, Zdep SM, Sepulveda P. Self-image among a national probability sample of girls. Child Study J 1974;4:103-114.

[33] Trowbridge N. Self concept and socioeconomic status in elementary school children. Am Educ Res J 1972;9:525-537. https://doi.org/10.3102/00028312009004525

[34] Mishra SK, Srivastava M, Tiwary NK, et al. Prevalence of depression and anxiety among children in rural and suburban areas of Eastern Uttar Pradesh: a cross-sectional study. J Family Med Prim Care 2018;7:21. https://doi.org/10.4103/jfmpc.jfmpc_248_17

[35] Frigerio A, Ed. Child Behavior Checklist 4-18, Manuale. Bosisio Parini, Italy: IRCCS Eugenio Medea, Editore Ghedini Libraio 2001.

[36] Achenbach TM. Integrative Guide to the 1991 CBCL/4-18, YSR, and TRF Profiles. Burlington, VT: University of Vermont, Department of Psychology 1991.

[37] Achenbach TM, Rescorla LA. Manual for the ASEBA preschool forms and profiles (Vol. 30). Burlington, VT: University of Vermont, Research center for children, youth, & families 2000.

[38] Jensen EJ, Mendenhall T. Call to action: family therapy and rural mental health. Contemp Fam Ther 2018;40:309-317. https://doi.org/10.1007/s10591-018-9460-3

[39] Zaheer J, Links PS, Law S, et al. Developing a matrix model of rural suicide prevention: a Canada-China collaboration. Int J Ment Health 2011;40:28-49. https://doi.org/10.2753/imh0020-7411400403

[40] Smalley KB, Yancey CT, Warren JC, et al. Rural mental health and psychological treatment: a review for practitioners. J Clin Psychol 2010;66:479-489. https://doi.org/10.1002/jclp.20688

[41] Boyd CP, Hayes L, Sewell J, et al. Mental health problems in rural contexts: a broader perspective: response to Jackson et al., 2007. Mental health problems in rural contexts: what are the barriers to seeking help from professional providers? Aust Psychol 2008;43:2-6. https://doi.org/10.1080/00050060701711841

[42] Robinson WD, Springer PR, Bischoff R, et al. Rural experiences with mental illness: through the eyes of patients and their families. Fam Syst Health 2012;30:308. https://doi.org/10.1037/a0030171

[43] Puskar K, Tusaie-Mumford K, Boneysteele G. Rurality and advanced practice nurses. J Multicult Nurs Health 1996;2:43-47.

[44] Parr H, Philo C, Burns N. Social geographies of rural mental health: experiencing inclusions and exclusions. Trans Inst Br Geogr 2004;29:401-419. https://doi.org/10.1111/j.0020-2754.2004.00138.x

[45] Smokowski PR, Evans CB, Cotter KL, et al. Ecological correlates of depression and self-esteem in rural youth. Child Psychiatry Hum Dev 2014;45:500-518. https://doi.org/10.1007/s10578-013-0420-8

[46] Millings A, Buck R, Montgomery A, et al. School connectedness, peer attachment, and self-esteem as predictors of adolescent depression. J Adolesc 2012;35:1061-1067. https://doi.org/10.1016/j.adolescence.2012.02.015

[47] Health Resources and Services Administration (HRSA). Mental health and rural America: 1994-2005. Rockville: MD 2005. https://doi.org/10.4135/9781412971942.n186

JOURNAL OF PSYCHOPATHOLOGY 2021;27:99-105
DOI: 10.36148/2284-0249-381

Original article

Perceived public stigma towards schizophrenia among healthcare students: the relationship with diagnostic labelling and contact with people with schizophrenia

Lucia Sideli[1], M. Valentina Barone[2], Laura Ferraro[2], Serena Giunta[3], Giuseppe Mannino[3], Fabio Seminerio[2], Crocettarachele Sartorio[2], Giuseppe Maniaci[2], Cristina Guccione[2], Francesca Giannone[4], Daniele La Barbera[2], Caterina La Cascia[2]

[1] Department of Human Sciences, LUMSA University, Rome, Italy; [2] Section of Psychiatry, Department of Biomedicine, Neuroscience, and Advanced Diagnostic, University of Palermo, Palermo, Italy; [3] Department of Law, LUMSA University, Palermo, Italy; [4] Department of Psychological and Educational Sciences, University of Palermo, Palermo, Italy

SUMMARY

Objectives

This study aimed at investigating the relationship between perceived public stigma towards people with schizophrenia (PWS) and their family members in a large sample of medical and psychology students. We hypothesised that: a) schizophrenia labelling would be related to greater perceived public stigma; b) contact with PWS would be related with lower perceived stigma; c) perceived public stigma would be similar between medical and psychology students and would be higher among students attending the clinical stage compared to their pre-clinical colleagues.

Methods

Participants were 592 students attending either the pre-clinical or clinical stage of coursework in Medicine and Psychology, at the University of Palermo (Italy) (Tab. I). Study measures included a short socio-demographic questionnaire, the Devaluation of Consumers Scale (DCS), and the Devaluation Consumers Families Scale (DCFS).

Results

Students who identified schizophrenia in an unlabelled clinical description expressed greater perceived public stigma towards PWS ($t = -2.895$, $p = 0.004$) and their family members ($t = -2.389$, $p = 0.017$). A trend-level association was found between previous contact with PWS and lower perceived public stigma ($t = 1.903$, $p = 0.058$), which became significant for those students who had a more extensive contact (Mann-Whitney $z = 2.063$, $p = 0.039$). Compared to medical students, psychology students perceived greater public stigma towards PWS. No difference was observed between students at different stages of their academic coursework (Tab. II). In a multivariate linear regression model, schizophrenia labelling and degree course predicted perceived public stigma towards severe mental disorders.

Conclusions

This study replicated previous findings on the relationship between public stigma towards PWS, schizophrenia labelling, and contact with PWS. Perception of public stigma was similar among pre-clinical and clinical students and greater among psychology students. The findings suggest the importance of promoting a critical awareness of negative stereotypes towards schizophrenia among healthcare students, since the beginning of their coursework. In addition to correct information about schizophrenia, anti-stigma intervention should include contact with PWS who live in the community.

Key words: perceived public stigma, stereotype, schizophrenia, psychotic disorders, healthcare students

Received: March 9, 2020
Accepted: May 12, 2021

Correspondence
Lucia Sideli
Department of Human Sciences, LUMSA University, via di Porta Castello, 44, 00193 Rome, Italy. E-mail: lucia.sideli@gmail.com

How to cite this article: Sideli L, Barone MV, Ferraro L, et al. Perceived public stigma towards schizophrenia among healthcare students: the relationship with diagnostic labelling and contact with people with schizophrenia. Journal of Psychopathology 2021;27:99-105. https://doi.org/10.36148/2284-0249-381

Introduction

An increasing body of literature has recognised the high prevalence and the detrimental effects of stigma towards severe mental illness and, in particular, towards schizophrenia spectrum disorders [1-6]. Common stereotypes related to these conditions include pessimism about recovery, desire for social distance, and attribution of the responsibility of the disorder to these patients [1]. A systematic review found that the economic burden of mental health stigma substantially affects the possibility of a stable employment, the personal and family income, and the use of healthcare services by people affected with mental disorders [2]. Compared to individuals with depression or anxiety [7,8], people with schizophrenia (PWS) appeared particularly exposed to stigmatisation, as schizophrenia is often associated with stereotypes of dangerousness and unpredictability [9]. According to a worldwide multicentric study [10], 47% of PWS have experienced some discrimination in establishing or maintaining friendship, and 43% believed they were treated differently by family members because of their mental illness. Furthermore, 29% of PWS reported that they have been disadvantaged in finding or keeping a job and 27% in intimate or sexual relationships [10].

Research has consistently demonstrated that public stigma towards severe mental disorders significantly contribute to increase barriers to care for PWS [3] and to worsen the symptom and functional outcome of schizophrenia. In general, stigmatisation has been consistently related to a delayed search for psychiatric care by PWS – because of the fear of being labelled as mentally ill – and to poor adherence to pharmacological and psychosocial treatments [4-6]. Specifically, PWS who perceived higher levels of public stigma towards severe mental disorders reported lower self-esteem, poorer quality of life, and more severe depressive symptoms [11,12]. The effect of perceived public stigma on self-esteem and quality of life was mediated by stress-related stigma and self-stigma, i.e. by the extent to which PWS recognised to be part of a stigmatized group [11,13,14]. Furthermore, among college students with self-reported mental disorders, perceived public stigma predicted reduced treatment-seeking behaviours, and this effect was also mediated by self-stigma [15]. Studies on the general and clinical populations suggests that the relationship between perceived public stigma, low self-esteem, and barriers to care are relatively independent by the potential confounding effect of ethnicity [12,16]. There are some indications that perceived public stigma towards severe mental disorders might be higher among women and older people [17,18].

Stigma towards PWS has been also associated with delayed or inadequate care for physical health problems, such as cardiovascular, metabolic, and infectious diseases [19]. For instance, schizophrenia label was related to low availability of general practitioners to have PWS on their practice list and respond to their need for care [20]. Furthermore, healthcare providers who endorsed a stereotyped view of schizophrenia were more concerned about the adherence to treatment of PWS and were less likely to make specialist referrals and medical prescriptions when needed [21].

Along with PWS, their family members are often exposed to negative stereotypes and stigmatising behaviours in the community [22-26]. This so-called "affiliate stigma" can be expressed in the form of social distancing, attribution of responsibility for the condition of their family member affected with schizophrenia, and comments reflecting lack of knowledge and fear of the disorder [22]. Furthermore, caregivers of PWS complained about the lack of consideration for their worries, needs, and competences by healthcare providers, inadequate information flow, and low participation in healthcare decisions [22]. Several studies have suggested that affiliate stigma was greater among caregivers of PWS than among caregivers of people with depressive and bipolar disorders [24,25], although less stronger than those affecting caregivers of people with substance use disorders [27]. Affiliate stigma was consistently related to reduced self-esteem, increased family burden, lower perceived social support, and increased psychopathological symptoms [24-26, 28].

Given the relevance of public stigma towards PWS and their family members and its impact on pathways to care, it is crucial to assess to what extent future healthcare professionals perceived these stereotypes, in view of possible sensitisation interventions. Evidence suggests that stigmatising attitude towards severe mental disorders tend to establish early in healthcare professionals [5]. Studies found that medical and psychology students hold a more negative view of PWS than people with other mental disorders [29-35]. However, this view was found to be improved by interventions involving education and social contact with PWS [26,27,36,37].

Only a few comparative studies assessed stigma towards PWS among different degree course and across different stages of education. A German study on medical and psychology students observed no difference between the two groups [38]. According to two cross-sectional studies, students attending the fifth and the sixth year of the medical school perceived greater social distance towards PWS, compared to students attending the first and the second year [29,30,39,40]. However, another longitudinal study reported a reduction of students' stereotypes from the beginning to the end of their medical studies [41].

This study aimed at investigating the relationship between perceived public stigma towards PWS and their family members, the identification of schizophrenia in an unlabelled clinical vignette (diagnostic labelling), and any previous knowledge or contact with PWS in a sample of medical and psychology students. Furthermore, the study aimed at comparing the perception of public stigma between medical and psychology students at different stages of their study course. We hypothesised that: a) schizophrenia labelling would be related to greater perceived public stigma; b) contact with PWS would be related with lower perceived public stigma; c) perceived public stigma would be similar between medical and psychology students and would be higher among students attending the clinical stage, compared to their pre-clinical colleagues.

Materials and methods

Participants and procedure

This was a cross-sectional study on a convenience sample of pre-clinical and clinical students attending the coursework in Medicine and Psychology, at the University of Palermo (Italy). Pre-clinical students were third-year medical and psychology students who have not yet attended an academic course in Psychiatry. Clinical students were fifth- and sixth-year medical students and fourth- and fifth-year psychology students who have attended a Psychiatry course providing information about the diagnosis, causes, and treatment of mental disorders. Students were approached at the end of their classes, informed about the study aims and methods, and ensured about the anonymity of data collection. Those willing to participate were invited to read an unlabelled description o<f schizophrenia according to ICD 10 criteria and to complete a short demographic questionnaire (including gender, age, and occupation of the main parental figure – which was considered as a proxy for family social class), the Devaluation of Consumers Scale (DCS), and the Devaluation Consumers Families Scale (DCFS) [23,42,43]. The Ethical Committee of the University Hospital "P. Giaccone" of Palermo (Italy) approved the study.

Measures

The Devaluation Consumers' Scale (DCS) and the Devaluation Consumers Families Scale (DCFS) were used to assess public stigma towards PWS and their family members [23,42,43]. Both questionnaires were rated on a 4-point scale, from "strongly disagree" to "strongly agree", with higher score indicating greater perceived public stigma. Within the DCS, five items assessed the perceived diminished patients' status in the society ("status reduction"; e.g., "Most people feel that entering psychiatric treatment is a sign of personal failure"), two items the reduced possibility to find a job or a stable relationship ("role restriction"; e.g., "Most employers will not hire a person who once had a serious mental illness if he or she is qualified for the job"), and one item difficulties in establishing friendship ("friendship refusal"; e.g., "Most people would not accept a person who once had a serious mental illness as a close friend"). Within the DCFS, four items assessed the social distancing from family members of PWS ("community rejection"; e.g., "Most people would rather not visit families that have a member who is mentally ill"), two items blaming parents for the disease of their family member ("causal attribution"; e.g., "Most people do not blame parents for the mental illness of their children"), and one item to the belief that these parents of were less responsible and caring than other parents ("uncaring parents", e.g., "Most people believe that parents of children with a mental illness are not as responsible and caring as other parents"). Mean scores of the total DCS and DCFS scales and of their subscales were calculated.

Schizophrenia labelling was defined as the ability to identify "schizophrenia" (*vs* depression, or anxiety, or nervous breakdown, or other unspecified mental or physical disorder) in a clinical description of schizophrenia according to ICD-10 criteria [40]. Contact with PWS was assessed using two questions. The first question asked the participants if they knew someone affected with the disorder described in the clinical vignette and was followed by a specifying question, which allowed for multiple responses. Responses to the specifying question were then classified as "family member", "partner", "friend", or "acquaintance" and were coded as "yes" or "no". The second question asked the participants if they had ever lived with someone affected with that disorder, who was not a family member.

Analyses

Demographic characteristics of the sample were compared using χ^2, Student's t-test, and ANOVA, as appropriate. Association of perceived public stigma with type and stage of their coursework, as well as with schizophrenia labelling and personal knowledge or previous contact with PWS were analysed using Student's t-test or Mann-Whitney z test, for variables non-normally distributed. Multivariate linear regression was used to investigate the effect of degree course on perceived public stigma (dependent variable), accounting for the effect of socio-demographic differences between medical and psychology students (i.e., gender and family social class). Significance level was set out at 0.05 for the total score of DCS and DCFS and to 0.006 for the subscale scores, applying Bonferroni correction. Analyses were carried out using STATA V12.0 IC.

Results

Participants in the study were 234 Medical students and 358 Psychology students. Three-hundred-eight were pre-clinical students and 284 were clinical students. Demographic characteristics of the sample were reported in Table I. Compared to psychology students, medical students were mostly male and belonging to an upper social class.

Three-quarter of the sample (445, 76.86%) identified schizophrenia in an unlabelled clinical description. These students showed a greater perceived public stigma towards PWS (schizophrenia 2.89 (0.55) *vs* other diagnoses 2.73 (0.56); $t = -2.895$, $p = 0.004$) and their family members (schizophrenia 2.54 (0.57) *vs* other diagnoses 2.40 (0.55); $t = -2.389$, $p = 0.017$).

One-quarter of the sample (147, 25%) knew someone affected with schizophrenia, but only a trend-level association was found with perceived public stigma (previous knowledge 2.76 (0.56) *vs* no previous knowledge 2.86 (0.55); $t = 1.903$, $p = 0.058$). No difference in perceived public stigma was found between students who had previous contact with either a relative (56, 9.4%), or a partner (16, 2.5%), or a friend (56, 9.4%), or an acquaintance (35, 24%), compared to those who had not. However, the few students that have lived with PWS (17, 2.8%) reported lower perceived public stigma towards PWS (sum of rank = 168,996 *vs* 3,582, Mann-Whitney $z = 2.063$, $p = 0.039$).

Compared to medical students, psychology students perceived greater public stigma towards PWS and their family members (see Table II), while no difference was found between students at different stages of their academic course (i.e. pre-clinical *vs* clinical). The association between degree course and perceived public stigma was still evident after controlling for socio-demographic differences between the two groups (DCS total score: $F(4,561) = 3.80$; $p = 0.005$; $\beta = 0.18$, $p = 0.001$; DCFS total score: $F(4,551) = 7.99$; $p < 0.001$; $\beta = 0.23$, $p < 0.001$). When schizophrenia labelling and personal knowledge of PWS were also included in the regression model, perceived public stigma towards patients was positively predicted by degree course ($F(6, 541) = 4.02$; $p < 0.001$; $\beta = 0.19$, $p < 0.001$) and schizophrenia labelling ($\beta = 0.12$, $p = 0.004$). Perceived public stigma towards family members was positively predicted by degree course ($F(6, 525) = 5.77$; $p < 0.001$; $\beta = 0.23$, $p < 0.001$).

Discussion and conclusions

The aim of this study was to investigate the relationship between perceived public stigma, schizophrenia labelling, and contact with PWS. We found that schizophrenia labelling was associated with greater public stigma towards people with severe mental disorders and their family members. This is consistent with previous studies among healthcare students reporting an association between schizophrenia label and negative stereotypes, such as dangerousness, unpredictability, and incurability [30,31,35,40,44,45]. In addition, the schizophrenia label was related to a negative view of caregivers of PWS, such as social distancing and attribution of responsibility for the disease of their family member. The findings suggest

TABLE I. *Demographic characteristic of the sample.*

	Pre-clinical students		
	Medical (n = 98)	**Psychology (n = 210)**	**χ^2/Student's t**
Gender **Male n (%)**	48 (48.98)	24 (11.43)	52.601 (< 0.001)
Age **M (SD)**	21.71 (0.11)	22.28 (0.24)	-1.548 (0.098)
Family social class			50.842 (< 0.001)
High n (%)	44 (46.83)	18 (9.47)	
Middle n (%)	34 (35.42)	106 (55.79)	
Low n (%)	17 (17.89)	66 (34.74)	
	Clinical students		
	Medical (n = 136)	**Psychology (n = 148)**	**χ^2/Student's t**
Gender **Male n (%)**	62 (45.59)	18 (12.16)	39.135 (< 0.001)
Age **M (SD)**	24.15 (2.62)	24.09 (10.65)	0.056 (0.955)
Family social class			30.090 (< 0.001)
High n (%)	54 (40.0)	17 (11.64)	
Middle n (%)	53 (39.26)	77 (52.74)	
Low n (%)	28 (20.74)	51 (35.17)	

that, in order to promote a collaborative relationship between healthcare professionals and family members, affiliate stigma should also be part of anti-stigma interventions for healthcare students [46,47].

We found that contact with PWS was related to a lower perceived public stigma. Specifically, we found that students who knew someone with schizophrenia and those who shared time and experience with him/her (e.g., by living in the same place) tend to perceive a less stigmatising attitude from the community. The findings are in line with some previous studies on high school and university students regarding the positive effect of social contact on mental health stigma. For instance, a cross-sectional study on pre-clinical, post-clinical rotation medical students, and psychiatrists, which observed that the more extensive was the contact to people with severe mental disorders, the lower were the stigmatising attitudes [48]. Moreover, a recent qualitative study on a four-days cohousing experience, involving patients with severe mental disorders and high school students, documented to what extent daily life interactions positively influenced mental health stereotypes and provided the students with a more realistic and integrated view of people affected with severe mental disorders [49].

The second aim of this study was to investigate whether perceived public stigma varied across the different academic programs and different stages of coursework. We found that perceived public stigma tend to be relatively stable over time, with no significant differences between students in the middle and the final stage of education. Previous cross-sectional studies on medical students found that perceived social distance towards PWS has increased between early (first/ second) and late years (fifth/ sixth) of their coursework [39,40]. Compared to these studies, the lack of differences observed in our sample might be due to the fact that we compared students at the final stage of the Medical and Psychology schools with students in the middle stage of their coursework, when beliefs about the social consideration of PWS might have been already established and might be less susceptible to change than at the beginning. Furthermore, we found that psychology students reported greater perceived public stigma, compared to medical students. Previous studies on college students found that perceived public stigma does not fully overlap with personal stigma, with participants reporting greater perceived public stigma than personal stigma [50,51], particularly if women [52]. In line with a study on social distancing towards PWS among university students [53], it might be also speculated that the different perception of public stigma towards mental disorders between medical and psychology students might have been also influenced by their future professional choices. In this regard, the prevision of future professional contacts with PWS and their families, which may be more common among psychology than among medical students, may have increased psychology students' perception of public stigma.

In summary, this study replicated previous findings on the relationship between schizophrenia labelling, social contact, and perceived public stigma towards PWS. In addition, the study found that perceived public stigma was greater among psychology than medical students and was substantially similar between pre-clinical and clinical students. These results should be interpreted in light for several limitations: first, the use of a convenience sample might have reduced the generalizability of the findings; second, the cross-sectional study design prevented any inferences on the relationship between academic program and perceived public stigma; third, the relationship between perceived public stigma and schizophrenia labelling and personal contact might have been confounded by other factors (e.g. the students' personal values and beliefs), here not assessed.

Given the influence that public stereotypes exert on the personal attitudes of the individuals [54], increasing students' awareness of such stereotypes is the first step

TABLE II. *DCS and DCFS distribution between the two samples.*

	Medical (n = 234)	Psychology (n = 358)	Student's t (p)
DCS total score M (SD)	2.74 (0.64)	2.91 (0.47)	-3.675 (< 0.001)*
DCS status reduction	2.78 (0.68)	2.98 (0.50)	-4.148 (< 0.001)*
DCS role restriction	2.78 (0.72)	2.92 (0.59)	-2.406 (0.016)
DCS friendship refusal	2.48 (0.77)	2.55 (0.71)	-1.101 (0.271)
DCFS total score M (SD)	2.36 (0.62)	2.60 (0.51)	-4.930 (< 0.001)*
DCFS community rejection	2.39 (0.65)	2.59 (0.55)	-3.958 (< 0.001)*
DCFS causal attribution	2.39 (0.70)	2.66 (0.60)	-4.862 (< 0.001)*
DCFS uncaring parents	2.20 (0.84)	2.50 (0.76)	-4.427 (< 0.001)*

*DCS: Devaluation Consumers' Scale; DCFS: Devaluation Consumers' Families Scale; *Associations statistically significant after Bonferroni correction for multiple testing* to develop a more positive view. As the next step, students' opinions towards PWS should take advantage of specific anti-stigma interventions, involving both information and contact with PWS [55-57]. Furthermore, as public stigma may affect self-stigma [54,58], and the onset of schizophrenia commonly occurs in late adolescence/early adulthood [59], studies suggested that assessing and discussing stigmatising beliefs among college and university students may contribute to reduce the barriers to care for those students who are experiencing themselves severe mental health problems [52,60].

Ethical consideration

The study was approved by the IRB of the University Hospital "P. Giaccone" of Palermo (Italy)

Acknowledgement

We are grateful to all the students who participated in the study.

Funding

None.

Conflict of interest

The authors declare to have no conflict of interest

Author contributions

All the authors significantly contributed to study conception, data acquisition, data analysis, or interpretation. All the authors participated in drafting the article or revising it for important intellectual contents.

References

1. Corrigan PW, Watson AC. Understanding the impact of stigma on people with mental illness. World Psychiatry 2002;1:16-20.
2. Sharac J, McCrone P, Clement S, et al. The economic impact of mental health stigma and discrimination: a systematic review. Epidemiol Psichiatr Soc 2010;19:223-232. https://doi.org/10.1705/524.6266
3. Henderson C, Evans-Lacko S, Thornicroft G. Mental illness stigma, help seeking, and public health programs. Am J Public Health 2013;103:777-780. https://doi.org/10.2105/AJPH.2012.301056
4. Thornicroft G. Stigma and discrimination limit access to mental health care. Epidemiol Psichiatr Soc 2008;17:14-19. https://doi.org/10.1017/s1121189x00002621
5. Corrigan P. How stigma interferes with mental health care. Am Psychol 2004;59:614-625. https://doi.org/10.1037/0003-066X.59.7.614
6. Clement S, Schauman O, Graham T, et al. What is the impact of mental health-related stigma on help-seeking? A systematic review of quantitative and qualitative studies. Psychol Med 2015;45:11-27. https://doi.org/10.1017/S0033291714000129
7. Morselli P, Cantarelli C, Rizzi M, et al. Cosa ci dicono i pazienti? Rapporto preliminare sui dati italiani del "GAMIAN International Survey". J Psychopathol 1999 (https://www.jpsychopathol.it/article/cosa-ci-dicono-i-pazienti-rapporto-preliminare-sui-dati-italiani-del-gamian-international-survey).
8. Angermeyer MC, Matschinger H. Public beliefs about schizophrenia and depression: similarities and differences. Soc Psychiatry Psychiatr Epidemiol 2003;38:526-534. https://doi.org/10.1007/s00127-003-0676-6
9. Angermeyer MC, Holzinger A, Carta MG, et al. Biogenetic explanations and public acceptance of mental illness: systematic review of population studies. Br J Psychiatry 2011;199:367-372. https://doi.org/10.1192/bjp.bp.110.085563
10. Thornicroft G, Brohan E, Rose D, et al.; INDIGO Study Group. Global pattern of experienced and anticipated discrimination against people with schizophrenia: a cross-sectional survey. Lancet 2009;373:408-415. https://doi.org/10.1016/S0140-6736(08)61817-6
11. Kao Y-C, Lien Y-J, Chang H-A, et al. Evidence for the indirect effects of perceived public stigma on psychosocial outcomes: the mediating role of self-stigma. Psych Res 2016;240:187-195. https://doi.org/10.1016/j.psychres.2016.04.030
12. van Zelst C, van Nierop M, Oorschot M, et al. Stereotype awareness, self-esteem and psychopathology in people with psychosis. PLoS One 2014;9:e88586. https://doi.org/10.1371/journal.pone.0088586
13. Norman RM, Windell D, Lynch J, et al. Parsing the relationship of stigma and insight to psychological well-being in psychotic disorders. Schizophr Res 2011;133a:3-7. https://doi.org/10.1016/j.schres.2011.09.002
14. Rüsch N, Corrigan PW, Heekeren K, et al. Well-being among persons at risk of psychosis: the role of self-labeling, shame, and stigma stress. Psych Serv 2014;65:483-489. https://doi.org/10.1176/appi.ps.201300169
15. Jennings KS, Cheung JH, Britt TW, et al. How are perceived stigma, self-stigma, and self-reliance related to treatment-seeking? A three-path model. Psychiatr Rehabil J 2015;38:109-116. https://doi.org/10.1037/prj0000138
16. Tsai J, Whealin JM, Pietrzak RH. Asian American and Pacific Islander military veterans in the United States: health service use and perceived barriers to mental health services. Am J Public Health 2014;104:S538-S547. https://doi.org/10.2105/AJPH.2014.302124
17. Böge K, Zieger A, Mungee A, et al. Perceived stigmatization and discrimination of people with mental illness: a survey-based study of the general population in five metropolitan cities in India. Indian J Psychiatry 2018;60:24-31. https://doi.org/ 10.4103/psychiatry.IndianJPsychiatry_406_17
18. Guttikonda A, Shajan AM, Hephzibah A, et al. Perceived stigma regarding mental illnesses among rural adults in Vellore, Tamil Nadu, South India. Indian J Psychol Med 2019;41:173-177. https://doi.org/10.4103/IJPSYM.IJPSYM_297_18
19. Thornicroft G, Rose D, Kassam A. Discrimination in health care against people with mental illness. Int Rev Psychiatry 2007;19:113-122. https://doi.org/10.1080/09540260701278937
20. Lam TP, Lam KF, Lam EWW, Ku YS. Attitudes of primary care physicians towards patients with mental illness in Hong Kong. Asia Pac Psychiatry 2013;5:e19-28. https://doi.org/10.1111/j.1758-5872.2012.00208.x
21. Corrigan PW, Mittal D, Reaves CM, et al. Mental health stigma and primary health care decisions. Psychiatry Res 2014;218:35-38. https://doi.org/10.1016/j.psychres.2014.04.028
22. Angermeyer MC, Schulze B, Dietrich S. Courtesy stigma – a focus group study of relatives of schizophrenia patients. Soc Psychiatry Psychiatr Epidemiol 2003;38:593-602. https://doi.org/10.1007/s00127-003-0680-x
23. Struening EL, Perlick DA, Link BG, et al. Stigma as a barrier to recovery: the extent to which caregivers believe most people devalue consumers and their families. Psychiatr Serv 2001;52:1633-1638. https://doi.org/10.1176/appi.ps.52.12.1633
24. Chang C-C, Yen C-F, Jang F-L, et al. Comparing affiliate stigma between family caregivers of people with different severe mental illness in Taiwan. J Nerv Ment Dis 2017;205:542-549. https://doi.org/10.1097/NMD.0000000000000671
25. Grover S, Avasthi A, Singh A, et al. Stigma experienced by caregivers of patients with severe mental disorders: a nationwide multicentric study. Int J Soc Psychiatry 2017;63:407-417. https://doi.org/10.1177/0020764017709484
26. Bipeta R, Yerramilli SS, Pillutla SV. Perceived stigma in remitted psychiatric patients and their caregivers and its association with self-esteem, quality of life, and caregiver depression. East Asian Arch Psychiatry 2020;30:101-107. https://doi.org/10.12809/eaap1943

27 Chang CC, Su JA, Chang KC, et al. Perceived stigma of caregivers: psychometric evaluation for Devaluation of Consumer Families scale. Int J Clin Health Psychol 2018;18:170-178. https://doi.org/10.1016/j.ijchp.2017.12.003

28 Hsiao C-Y, Lee C-T, Lu H-L, et al. Living with schizophrenia: health-related quality of life among primary family caregivers. J Clin Nurs 2017;26:5151-5159. https://doi.org/10.1111/jocn.14063

29 James BO, Omoaregba JO, Okogbenin EO. Stigmatising attitudes towards persons with mental illness: A survey of medical students and interns from Southern Nigeria. Ment Illn 2012;4:32-34. https://doi.org/10.4081/mi.2012.e8

30 Schenner M, Kohlbauer D, Gunther V, et al. Communicate instead of stigmatizing – does social contact with a depressed person change attitudes of medical students towards psychiatric disorders? A study of attitudes of medical students to psychiatric patients. Neuropsychiatr 2011;25:199-207.

31 Magliano L, Read J, Rega S, et al. The influence of causal explanations and diagnostic labeling on medical students' views of schizophrenia. Acad Med 2011;86:1155-1162. https://doi.org/10.1097/ACM.0b013e318226708e

32 Serafini G, Pompili M, Haghighat R, et al. Stigmatization of schizophrenia as perceived by nurses, medical doctors, medical students and patients. J Psychiatr Ment Health Nurs 2011;18:576-585. https://doi.org/10.1111/j.1365-2850.2011.01706.x

33 Magliano L, Read J, Rinaldi A, et al. The influence of causal explanations and diagnostic labeling on psychology students' beliefs about treatments, prognosis, dangerousness and unpredictability in schizophrenia. Community Ment Health J 2016;52:361-369. https://doi.org/10.1007/s10597-015-9901-5

34 Lyndon AE, Crowe A, Wuensch KL, et al. College students' stigmatization of people with mental illness: familiarity, implicit person theory, and attribution. J Ment Heal 2016;28:1-5. https://doi.org/10.1080/09638237.2016.1244722

35 Yang LH, Anglin DM, Wonpat-Borja AJ, et al. Public stigma associated with psychosis risk syndrome in a college population: Implications for peer intervention. Psychiatr Serv 2013;64:284-288. https://doi.org/10.1176/appi.ps.003782011

36 Corrigan PW, Morris SB, Michaels PJ, et al. Challenging the public stigma of mental illness: a meta-analysis of outcome studies. Psychiatr Serv 2012;63:963-973. https://doi.org/10.1176/appi.ps.201100529

37 Thornicroft G, Mehta N, Clement S, et al. Evidence for effective interventions to reduce mental-health-related stigma and discrimination. Lancet 2016;387:1123-1132. https://doi.org/10.1016/S0140-6736(15)00298-6

38 Lincoln TM, Arens E, Berger C, et al. Can antistigma campaigns be improved? A test of the impact of biogenetic vs. psychosocial causal explanations on implicit and explicit attitudes to schizophrenia. Schizophr Bull 2008;34:984-994. https://doi.org/10.1093/schbul/sbm131

39 Ay P, Save D, Fidanoglu O. Does stigma concerning mental disorders differ through medical education? A survey among medical students in Istanbul. Soc Psychiatry Psychiatr Epidemiol 2006;41:63-67. https://doi.org/10.1007/s00127-005-0994-y

40 Magliano L, Read J, Sagliocchi A, et al. Differences in views of schizophrenia during medical education: A comparative study of 1st versus 5th-6th year Italian medical students. Soc Psychiatry Psychiatr Epidemiol 2013;48:1647-1655. https://doi.org/10.1007/s00127-012-0610-x

41 Esen Danacı A, Balıkçı K, Aydın O, Cengisiz C, Uykur AB, Bildik T, et al. The effect of medical education on attitudes towards schizophrenia: a five-year follow-up study. Turk Psikiyatri Derg 2016;27:176-184. https://doi.org/10.5080/u7194.1

42 Link BG, Struening EL, Neese-Todd S, et al. Stigma as a barrier to recovery: the consequences of stigma for the self-esteem of people with mental illnesses. Psychiatr Serv American Psychiatric Publishing 2001;52:1621-1626. https://doi.org/10.1176/appi.ps.52.12.1621

43 Sideli L, Mulè A, La Cascia C, et al. Validation of the Italian version of the Devaluation consumers' Scale and the Devaluation Consumers Families Scale. Journal of Psychopathology 2016;22:251-257.

44 Economou M, Peppou LE, Louki E, et al. Medical students' beliefs and attitudes towards schizophrenia before and after undergraduate psychiatric training in Greece. Psychiatry Clin Neurosci 2012;66:17-25. https://doi.org/10.1111/j.1440-1819.2011.02282.x

45 Magliano L, Read J, Sagliocchi A, et al. Effect of diagnostic labeling and causal explanations on medical students' views about treatments for psychosis and the need to share information with service users. Psychiatry Res 2013;210:402-407. https://doi.org/10.1016/j.psychres.2013.07.022

46 Angermeyer MC, Millier A, Rémuzat C, et al. Continuum beliefs and attitudes towards people with mental illness: results from a national survey in France. Int J Soc Psychiatry 2015;61:297-303. https://doi.org/10.1177/0020764014543312

47 Read J, Haslam N, Sayce L, et al. Prejudice and schizophrenia: a review of the 'mental illness is an illness like any other'approach. Acta Psychiatr Scand 2006;114:303-318. https://doi.org/10.1177/0020764017735865

48 Eksteen H-C, Becker PJ, Lippi G. Stigmatization towards the mentally ill: perceptions of psychiatrists, pre-clinical and post-clinical rotation medical students. Int J Soc Psychiatry 2017;63:782-791. https://doi.org/10.1177/0020764017735865

49 Palacios-Ceña D, Martín-Tejedor EA, Elías-Elispuru A, et al. The impact of a short-term cohousing initiative among schizophrenia patients, high school students, and their social context: a qualitative case study. PLoS One 2018;13:e0190895. https://doi.org/10.1371/journal.pone.0190895

50 Eisenberg D, Downs MF, Golberstein E, et al. Stigma and help seeking for mental health among college students. Med Care Res Rev 2009;66:522-541. https://doi.org/10.1177/1077558709335173

51 Calear AL, Griffiths KM, Christensen H. Personal and perceived depression stigma in Australian adolescents: magnitude and predictors. J Affect Disord 2011;129:104-108. https://doi.org/10.1016/j.jad.2010.08.019

52 Pedersen ER, Paves AP. Comparing perceived public stigma and personal stigma of mental health treatment seeking in a young adult sample. Psychiatry Res 2014;219:143-150. https://doi.org/10.1016/j.psychres.2014.05.017

53 Arens EA, Berger C, Lincoln TM. Stigmatisierung von patienten mit schizophrenie: prägt das studium die einstellungen angehender psychologen und mediziner? [Stigmatization of patients with schizophrenia: the influence of university courses on the attitudes of prospective psychologists and doctors]. Nervenarzt 2009;80:329-339. https://doi.org/10.1007/s00115-008-2646-4

54 Corrigan PW, Watson AC, Barr L. The self-stigma of mental illness: implications for self-esteem and self-efficacy. J Soc Clin Psychol 2006;25:875-884. https://doi.org/10.1521/jscp.2006.25.8.875

55 Magliano L, Read J, Sagliocchi A, et al. Social dangerousness and incurability in schizophrenia: results of an educational intervention for medical and psychology students. Psychiatry Res 2014;219:457-463. https://doi.org/10.1016/j.psychres.2014.06.002

56 Magliano L, Rinaldi A, Costanzo R, et al. Improving psychology students' attitudes toward people with schizophrenia: a quasi-randomized controlled study. Am J Orthopsychiatry 2016;86:253-264. https://doi.org/10.1037/ort0000161

57 Del Casale A, Manfredi G, Kotzalidis GD, et al. Awareness and education on mental disorders in teenagers reduce stigma for mental illness: a preliminary study. Journal of Psychopathology 2013;19:208-212.

58 Vogel DL, Bitman RL, Hammer JH, et al. Is stigma internalized? The longitudinal impact of public stigma on self-stigma. J Couns Psychol 2013;60:311-316. https://doi.org/10.1037/a0031889

59 Mulè A, Sideli L, Capuccio V, et al. Low incidence of psychosis in Italy: confirmation from the first epidemiological study in Sicily. Soc Psychiatry Psychiatr Epidemiol 2017;52. https://doi.org/10.1007/s00127-016-1322-4

60 Wu IHC, Bathje GJ, Kalibatseva Z, et al. Stigma, mental health, and counseling service use: a person-centered approach to mental health stigma profiles. Psychol Serv 2017;14:490-501. https://doi.org/10.1037/ser0000165

JOURNAL OF PSYCHOPATHOLOGY 2021;27:106-114
DOI: 10.36148/2284-0249-412

Original article

Hikikomori, problematic internet use and psychopathology: correlates in non-clinical and clinical samples of young adults in Italy

Simone Amendola[1], Rita Cerutti[1], Fabio Presaghi[2], Valentina Spensieri[1], Chiara Lucidi[3], Elisa Silvestri[3], Vassilij Di Giorgio[3], Fabio Conti[3], Alessandro Martorelli[3], Gaia Izzi[3], Alan Teo[4-6]

[1] Department of Dynamic and Clinical Psychology, Sapienza University of Rome, Rome, Italy; [2] Department of Psychology of Development and Socialization Processes, Sapienza University of Rome, Rome, Italy; [3] Psychiatric Residential Structure "Casa di cura Villa Armonia Nuova", Rome, Italy; [4] Center to Improve Veteran Involvement in Care (CIVIC), VA Portland Health Care System, Portland, OR, USA; [5] Department of Psychiatry, School of Medicine, Oregon Health & Science University, Portland, OR, USA; [6] School of Public Health, Oregon Health & Science University and Portland State University, Portland, OR, USA

Received: January 11, 2021
Accepted: March 25, 2021

Correspondence
Simone Amendola
Department of Dynamic and Clinical Psychology, Faculty of Medicine and Psychology, Sapienza University of Rome, via degli Apuli 1, 00185 Rome, Italy. Tel. +39 06 4991 7936. Fax +39 06 4991 7910
E-mail: simone.amendola@uniroma1.it

How to cite this article: Amendola S, Cerutti R, Presaghi F, et al. Hikikomori, problematic internet use and psychopathology: correlates in a non-clinical and clinical sample of young adults in Italy. Journal of Psychopathology 2021;27:106-114. https://doi.org/10.36148/2284-0249-412

SUMMARY

Objectives

The aims of this study were to explore hikikomori (prolonged social withdrawal) as well as its relationship with problematic internet use and other psychopathology.

Methods

A total of 66 young adults in Italy were recruited for this study consisting of: a non-clinical sample recruited through an online survey (n = 47), and a clinical sample of patients with a psychiatric disorder at onset (n = 19).

Results

Our findings demonstrated the occurrence of hikikomori in both the non-clinical and clinical samples (n = 5). Brief episodes of social withdrawal (i.e., duration between one and three months) were also reported by participants (n = 10). Hikikomori symptoms were associated with overall personality dysfunction in both samples (r = .643, p < .001; r = .596, p < .01, in the non-clinical and clinical sample, respectively). Problematic internet use was related to interpersonal sensitivity (r = .309, p < .05) and depression (r = .475, p < .05) in the non-clinical and clinical samples, respectively.

Conclusions

We demonstrated the occurrence of hikikomori in both non-clinical and clinical samples of Italian young adults. Clinical features of psychopathology (e.g., self- and other-directed aggressive behaviors, substance misuse) were more prevalent among hikikomori participants of the clinical sample. Moreover, symptoms of hikikomori showed strong associations with overall personality dysfunction. Our results highlighted the need to disentangle the intricate relation between hikikomori and psychopathology and they were discussed considering scientific advances. Finally, findings of this study suggested that online survey is a useful methodology to identify young adults with hikikomori. Further research with larger sample sizes is needed to confirm our data.

Key words: technology use, hikikomori, internalizing symptoms, emerging adulthood, personality functioning

Introduction

Social withdrawal is an "umbrella term" [1] that refers to the voluntary behaviour of isolating oneself from the interactions with peers. Its roots may lie in biological aspects and parenting behaviours [1,2]. The changes that occur in adolescence can increase social isolation and constitute a risk factor for psychological distress subsequently in life [3-5]. Although it is mainly considered a symptom of other psychopathological disorders, in the last decade the interest of researchers has been focusing on social withdrawal itself [6,7]. The present study focused attention on brief and prolonged episodes of social withdrawal to increase knowledge about the phenomenon in young adulthood.

Hikikomori (prolonged social withdrawal)

The scientific debate concerning the definition and conceptualization of *hikikomori*, a specific type of social withdrawal also commonly referred to as *prolonged social withdrawal* or *pathological social withdrawal*, has been enhanced by some recent studies. Kato, Kanba and Teo [8,9] revised the diagnostic criteria for hikikomori as "a form of pathological social withdrawal or social isolation whose essential feature is physical isolation in one's home" (p. 117). Marked social isolation in one's home (criterion 1), duration of continuous social isolation of at least 6 months (criterion 2) and significant functional impairment or distress associated with the social isolation (criterion 3) must be endorsed to diagnose hikikomori. Importantly, withdrawal severity may be specified, according to the degree to which the subject leaves his home (i.e., *mild* = 2-3 days/week; *moderate* = 1 day/week or less; *severe* = to rarely leave the single room). Individuals with a duration of continuous social withdrawal of at least 3 but less than 6 months should be considered *pre-hikikomori* [8,9]. The authors have also specified other (not mandatory) aspects to better characterize the condition (e.g., social participation, in-person social interaction, indirect communication, loneliness, co-occurring condition).

The first aim of our study was to examine the occurrence and the characteristics of hikikomori phenomenon among Italian young adults. While hikikomori was initially reported in Japan, it has since been described as a syndrome worldwide [10-14]. Researchers have also shown presence in Western countries that are culturally distinct from Japan, such as France [11], Spain [13], Ukraine [12], and the USA [14]. To the best of our knowledge, only case reports of hikikomori have been reported in Italy [15,16].

Following the suggestion of Kato and colleagues [9] regarding the need to consider this condition as a public and mental health concern, the present study aimed to expand scientific knowledge on hikikomori and its characteristics in a western sociocultural context. Specifically, a non-clinical and clinical sample of Italian young adults were considered for this investigation.

Symptoms of hikikomori and problematic internet use

The second aim of our study was to examine the relationship between symptoms of hikikomori and problematic internet use. Despite growing acknowledgement concerning the different activities and functions of digital technologies, recent research has shown that the problematic use of internet technology has a negative impact on physical health as well as on emotional and behavioural functioning [17]. The advent of the internet and new technologies (e.g., computer, smartphone, videogame console) may contribute to a shift from direct to indirect forms of play and communication. These changes may have an influence on the psychological and behavioural development of young individuals, affecting the worldwide emergence of hikikomori phenomenon [8]. Symptoms of internet addiction have been investigated in 41 socially withdrawn youth referred to mental health centres and several psychiatric clinics in Korea [18]. Approximately 55% were at high risk for Internet addiction and 10% were shown to be addicted. Furthermore, around 30% of a sample comprising 190 Spanish adults treated at home for social withdrawal showed Internet addiction behaviours [13]. Finally, two recent studies demonstrated an association between problematic internet use and symptoms of social withdrawal in non-clinical young adults [19,20].

In light of the above considerations, we hypothesized to find evidence of hikikomori occurrence both in a non-clinical and clinical sample of Italian young adults according to the criteria defined by Kato and colleagues [8,9]. Additionally, we expected an association between symptoms of hikikomori and problematic internet use.

Materials and methods

Participants and recruitment method

In this cross-sectional study, data were collected from two samples, one a non-clinical sample and the other a clinical sample. The non-clinical sample consisted of young adults who completed an online survey in October 2019. An age between 18 and 25 years was the inclusion criterion, while a diagnosis for psychiatric disorders was the exclusion criterion. The clinical sample consisted of young adults with a psychiatric disorder at onset and in the acute phase. They were recruited between September 2018 and November 2019 at the Psychiatric Residential Structure "Casa di Cura Villa Armonia Nuova" in Rome, Italy. An age between 18 and 25 was the inclusion criterion. Exclusion criteria included: neurological disease, inability to understand written Italian or to participate in the procedure, and moderate to severe mental retardation.

Data procedures

After the exclusion of one participant to avoid inclusion of possible outliers due to extreme values on two of the questionnaires, 47 young adults constituted the non-clinical sample. To advertise the survey, two social media platforms were used (i.e., WhatsApp and Facebook). The respondents' consent was obtained online by reading and approving an informed consent, before proceeding to the compilation of the questionnaire battery. To limit response bias, the study was presented as a survey on young people's Internet use and the related risky online behaviours. Research data was encoded and stored on password protected drives to ensure respect for data protection and privacy. Ownership and access to data was limited to the research team.

For the clinical sample, one participant was excluded from analysis due to extreme values on the questionnaires that indicated unreliability of the responses, resulting in nineteen young adults. Informed consent was obtained from both participants and their parents before enrolment in the study. After providing informed consent, and once the acute symptomatology phase was over, the questionnaires were briefly presented to the participants individually. The administration lasted approximately 40 minutes and participants individually completed the questionnaires together with the clinical psychologist of the Psychiatric Residential Structure. This study was approved by the Ethics Committee of the Department of Dynamic and Clinical Psychology, Sapienza University of Rome.

Measures

Hikikomori (prolonged social withdrawal) diagnosis

This study used the definition of hikikomori proposed by Kato and colleagues [8,9]. The following symptoms were investigated for "present" and "past" episodes of hikikomori through a questionnaire designed for this study: (1) marked social isolation in one's home (i.e., leaving the house 3 days/week or less), and (2) duration of social withdrawal of at least six months. Isolation caused by physical illness (e.g., with the impossibility of walking or moving), pregnancy or childbirth, working from home, or the need to stay at home to take care of children, were considered exclusion criteria of hikikomori. Lack of social participation (i.e., 3 days/week or less), lack of in-person social interaction (i.e., 3 days/week or less), self-reported withdrawal motivation, other-directed aggression, self-injury (without suicidal intent) and suicidal behaviour during episodes of social withdrawal were also investigated only in participants who reported episodes of social withdrawal to further characterize the phenomenon.

The Web survey was conducted in October 2019 in order to identify and reach socially withdrawn youth according to Liu, Li, Teo, Kato, and Wong [21] study methodology.

Hikikomori symptoms

We explored characteristics of individuals with a duration of the social withdrawal behaviour less than six months in order to better delineate the phenomenon of social withdrawal. Specifically, participants were considered to have experienced brief social withdrawal if they indicated to staying at home almost every day for at least one month but less than three.

Furthermore, symptoms of social withdrawal were examined using two self-report measures.

The clinical sample completed the Hikikomori Symptoms Scale (HSS) firstly used in the survey of the Cabinet Office of the Government of Japan [22]. It consists of four items that explore the symptoms of social withdrawal (e.g., "I sometimes have in my mind an idea that I want to keep at home or in my room"; "When bad things happen, I do not feel like I want to go outside"). For each item, the respondent is presented with six response choices (from 1 = "absolutely not true" to 6 = "absolutely true"). The scale showed good psychometric properties [22]. In the present study, Cronbach's alpha for the scale was.66.

The non-clinical sample completed the Hikikomori Questionnaire (HQ-25), a new validated instrument with good psychometric properties to evaluate symptoms of hikikomori [23]. This questionnaire was not available when the research involving the clinical sample was planned and conducted. The HQ-25 is a self-report questionnaire including 25 items that evaluate the severity of hikikomori symptoms over the preceding 6 months. Typical psychological features and behavioural patterns of hikikomori syndrome, such as socialization, isolation, emotional support, and a sense of alienation from society, are investigated. Participants respond on a 5-point Likert scale (from 0 = "strongly disagree" to 4 = "strongly agree"). The HQ-25 has a score range of 0-100. Higher values indicate higher symptomatology. Teo and colleagues [23] reported good psychometric properties and proposed a cut off score of 42 to discriminate individuals at risk for hikikomori. In the present study, Cronbach's alpha for the questionnaire was.92.

Psychopathology

Information on the diagnoses of psychiatric disorders were determined by consulting the medical records of the clinical sample participants and asking specific questions to the non-clinical sample participants during the online survey (i.e., "Do you suffer from a medical or psychological disorder that has been diagnosed by a health professional?"; "If yes, please indicate what the disorder is and how long you have been diagnosed with the disorder."). Information on substance use (i.e., psychoactive substances and alcohol) in the last six months was recorded. Three subscales of the *Brief Symptom Inventory* (BSI) [24] were also administered to participants. The BSI is a 53-item self-report instrument that assesses nine primary psychological symptom dimensions during the past seven days. For the purposes of the present study, interpersonal sensitivity, depression, and anxiety dimensions were used. Respondents rated each item on a 5-point Likert scale (from 0 = "never" to 4 = "always"). The reliability of the BSI dimensions of interest proved to be good in a sample of Italian adults [25]. In the present study, Cronbach's alpha of the three dimensions were.70,.84,.78 (i.e., anxiety, depression, and interpersonal sensitivity, respectively) for the non-clinical sample and.88,.88,.76, for the clinical sample.

The *Internet Disorder Scale* (IDS-15) [26] is a self-report scale composed of 15 items. It assesses the severity and impact of internet addiction by focusing upon users' online leisure activity from any device with internet access over the past year. Four distinct domains related to internet addiction are investigated: escapism and dysfunctional emotional coping; withdrawal symptoms; impairments and dysfunctional self-regulation; and dysfunctional Internet-related self-control. The respondents rated each item on a 5-point Likert scale (from 1 = "strongly disagree" to 5 = "strongly agree"). The total score can range from 15 to 75, with higher scores being an indication of higher degrees of internet addiction. The Italian version of the IDS-15 demonstrated good psychometric properties [27]. In the present study, Cronbach's alpha for the scale was.85 for the non-clinical sample and.94 for the clinical sample.

Finally, we explored the main activity for which the subject used the internet, classifying whether it was social (e.g., social network, online videogame) or non-social (e.g., listening to music, watching TV series or movies).

Maladaptive personality functioning

The *Personality Inventory for DSM-5 Brief Form* (PID-5-BF) [28] is a scale that evaluates five maladaptive trait domains of personality functioning including Negative Affect, Detachment, Antagonism, Disinhibition, and Psychoticism. The PID-5-BF includes 25 items, five for each domain, with four response alternatives distributed on a 4-point Likert scale (from 0 = "very false/often false" to 3 = "very true/often true"). The maladaptive functioning of the individual, linked to the domains and to the total score, increases as the score increases. In the present study, the scale showed good internal consistency (a Cronbach's α of.86 and.74 for the non-clinical and clinical samples respectively).

Statistical analysis

Statistical analysis was performed using SPSS version 25.0 (IBM SPSS Statistics, Armonk, NY). Given the small sample size, statistical analysis was limited to descriptive statistics and Pearson correlations to examine relationships between the variables of interest. To explore the relationship between social withdrawal and the other variables, Pearson correlation analysis were performed. We set statistical significance at $p < 0.05$.

To determine whether the sample size was large enough for the correlation analyses, we ran an a priori power analysis using the "G*Power 3.1" [29]. Results showed that the minimum samples size to detect a medium-to-large effect size (i.e., ρ of.4), given a power of 0.80 and an alpha of.05 for two-tailed significance, was n = 44. Instead the minimum samples size to detect a large effect size (i.e., ρ of.5), given a power of 0.80 and an alpha of.05 for two-tailed significance, was n = 26. Based on these calculations, the non-clinical sample appeared to be adequate to detect a medium-to-large effect size, while the size of the clinical sample was less than that indicated by the a priori power analysis to detect a large effect.

Results

Focusing on participants' education, in the non-clinical sample consisting of 47 participants (38.3% males, n = 18), ten (21.3%) had obtained an early secondary school education, 27 (57.4%) completed high school, and ten (21.3%) had a university degree. In the clinical sample consisting of nineteen participants (52.6% males, n = 10), one (5.3%) had obtained elementary education, eight (42.1%) an early secondary school education, and ten (52.6%) completed high school.

No participants in the non-clinical sample were diagnosed with a personality or clinical disorder by a mental health professional.

34 and 68.4% of participants reported substance use in the non-clinical and clinical sample, respectively. Furthermore, 27.7 and 15.8% of the non-clinical and clinical samples used the internet for non-social purposes.

Regarding scores on the questionnaires exploring social withdrawal, the mean score on the HQ-25 for the non-clinical sample was 22.25 (SD = 15.07) while the clinical sample showed a mean score of 4.5 (SD = 0.62) on the HSS. Table I presents the descriptive statistics for the non-clinical and clinical samples.

Hikikomori and brief social withdrawal

12.8% (n = 6) of participants of the non-clinical sample and 47.4% (n= 9) of participants of the clinical sample reported a lifetime episode of social withdrawal (i.e., hikikomori or brief social withdrawal).

In particular, as shown in Table I, in the non-clinical sample, two (4.3%) participants had experienced a condition of hikikomori and four (8.5%) reported a brief episode of social withdrawal. Regarding the period of social withdrawal episodes, the two hikikomori participants were in a current condition of prolonged social withdrawal. Instead, of the four participants who reported brief social withdrawal, one and three experienced current and past episodes of isolation, respectively.

According to the cut-off score for the HQ-25 proposed by Teo and colleagues [23], six subjects (12.8%; males = 2) from non-clinical sample screened positive for hikikomori. The cut-off of the HQ-25 correctly identified the two subjects who met the diagnosis of hikikomori. On the contrary, none of the subjects who reported a brief episode of social withdrawal showed a total score higher than the cut-off on the HQ-25.

In the clinical sample, 3 (15.8%) participants reported hikikomori and 6 (31.5%) showed a brief episode of social withdrawal. Regarding the period of social

TABLE I. *Participants' characteristics (n = 66).*

	Non-clinical sample (n = 47) n (%)	Clinical sample (n = 19) n (%)
Demographics		
Age (M ± SD)	20.49 ± 2.64	21.05 ± 2.07
Male	18 (38.3)	10 (52.6)
Education		
Elementary	0	1 (5.3)
Early secondary school	10 (21.3)	8 (42.1)
High school	27 (57.4)	10 (52.6)
University degree	10 (21.3)	0
Substance use	16 (34.0)	13 (68.4)
Non-social internet use	13 (27.7)	3 (15.8)
Maladaptive personality (M ± SD)		
Overall personality dysfunction	0.81 ± 0.41	1.20 ± 0.36
Psychopathology (M ± SD)		
Interpersonal Sensitivity	0.81 ± 0.84	1.21 ± 0.97
Depression	0.78 ± 0.62	1.17 ± 0.90
Anxiety	0.77 ± 0.59	1.16 ± 1.02
Problematic internet use	38.08 ± 9.31	39.37 ± 13.53
Lifetime episode of social withdrawal		
Hikikomori	2 (4.3)	3 (15.8)
Brief social withdrawal	4 (8.5)	6 (31.5)

Note. Participants were evaluated for a current or past social withdrawal episode

withdrawal episodes, two of the three participants who reported hikikomori were in a current condition of prolonged social withdrawal. Instead, five of the six participants who reported brief social withdrawal experienced a past episode of isolation.

Patients who reported hikikomori received the following diagnoses: Psychotic Disorder Not Otherwise Specified (n = 1); Mood Disorder Not Otherwise Specified (n = 1); Schizoid Personality Disorder and Mood Disorder Not Otherwise Specified (n = 1) (Tab. II).

Regarding the prevalence of characteristics related to social withdrawal, non-clinical and clinical samples showed similarity in lack of social participation and in-person social interaction but also differences in self-reported withdrawal motivations. Participants of the clinical sample who reported episodes of social withdrawal during the isolation period demonstrated self- and other-directed aggressive behaviours. Moreover, non-social internet use was more prevalent among participants of the clinical sample who reported social withdrawal episodes.

Correlations between symptoms of social withdrawal, problematic internet use, and psychopathology

Symptoms of social withdrawal were not found to be significantly associated to problematic internet use (Tabs. III-IV). However, a trend toward a positive correlation between social withdrawal and problematic internet use (r = .278, p = .059) was found in the non-clinical sample.

A positive association between symptoms of social withdrawal and overall personality dysfunction was reported in the two samples despite symptoms of social withdrawal were measured though different questionnaires.

In the non-clinical sample, symptoms of social withdrawal also showed significant positive associations with depression and interpersonal sensitivity. Furthermore, interpersonal sensitivity was associated to problematic internet use. In addition, a trend toward positive correlations were also found between social withdrawal and anxiety (r = .287, p = .050).

In the clinical sample, social withdrawal symptoms were not found to be significantly associated to other variables. However, near significant positive correlations

TABLE II. *Characteristics of partipants who reported a lifetime period of social withdrawal (n = 15) (i.e., hikikomori or brief social withdrawal), arranged by sample.*

Variables	Non-clinical sample		Clinical sample	
	Hikikomori (n = 2)	**Brief social withdrawal** (n = 4)	**Hikikomori** (n = 3)	**Brief social withdrawal** (n = 6)
	n (%)	n (%)	n (%)	n (%)
Male	1 (50)	2 (50)	1 (33.3)	3 (50)
Substance use	0	1 (25)	2 (66.7)	3 (50)
Non-social internet use	1 (50)	2 (50)	2 (66.7)	5 (85)
Social withdrawal characteristics				
Lack of social participation	2 (100)	4 (100)	3 (100)	6 (100)
Lack of in-person social interaction	2 (100)	3 (75)	3 (100)	5 (83.3)
Physical aggression towards others	0	0	1 (33.3)	0
Self-injury	0	0	3 (100)	3 (50)
Suicidal ideation	0	1 (25)	3 (100)	4 (67)
Suicidal attempt	0	0	0	1 (15)
Self-reported withdrawal motivation				
Low self-esteem	1 (50)	2 (50)	0	0
Feeling safe only at home	0	1 (25)	0	3 (50)
Feeling difficulty in doing things	1 (50)	0	0	2 (33)
Bullying episodes	0	1 (25)	1 (33.3)	0
Psychiatric disorder	0	0	2 (66.7)	0
Psychiatric hospitalization for				
Severe withdrawal	-	-	1 (33.3)	2 (33)
Suicide attempt	-	-	1 (33.3)	4 (67)
Positive symptoms	-	-	1 (33.3)	2 (33)
Anhedonia	-	-	1 (33.3)	0
Current psychiatric diagnosis				
Mood disorder not otherwise specified	-	-	2 (66.7)	3 (50)
Psychotic disorder not otherwise specified	-	-	1 (33.3)	1 (15)
Schizoid personality disorder	-	-	1 (33.3)	0
Schizoaffective disorder	-	-	0	1 (15)
Major depressive disorder	-	-	0	1 (15)

Note. Participants were evaluated for a current or past social withdrawal episode. Reason for psychiatric hospitalization and diagnosis percentages total more than 100% as participants may have more than 1 reason and diagnosis as reported in medical records. Characteristics and diagnosis with a frequency of zero are not reported.

were also found between social withdrawal and interpersonal sensitivity (r = .416, p = .077) as well as between social withdrawal and anxiety (r = .398, p = .091). Finally, problematic internet use positively correlated with depression in the clinical sample.

Discussion and conclusions

This study was the first to examine hikikomori and its relationship with problematic internet use in non-clinical and clinical Italian young adults. Our main finding suggested that episodes of hikikomori have been reported in both non-clinical and clinical samples, providing initial empirical evidence on the existence of the hikikomori among Italian young adults. These results are consistent with those of previous studies that reported cases of primary and secondary hikikomori in adult populations of western countries [11-14].

TABLE III. *Pearson's correlations between the variables of interest in the non-clinical sample.*

	1.	2.	3.	4.	5.	6.
1. Social withdrawal symptoms	1					
2. Overall personality dysfunction	.643‡	1				
3. Interpersonal sensitivity	.729‡	.555‡	1			
4. Depression	.716‡	.690‡	.733‡	1		
5. Anxiety	.287	.471†	.446†	.517‡	1	
6. Problematic internet use	.278	.228	.309*	.212	.281	1

Note. Social withdrawal symptoms as evaluated by the Hikikomori Questionnaire (HQ-25).
* $p < .05$; † $p < .01$; ‡ $p < .001$.

TABLE IV. *Pearson's correlations between the variables of interest in the clinical sample.*

	1.	2.	3.	4.	5.	6.
1. Social withdrawal symptoms	1					
2. Overall personality dysfunction	.596†	1				
3. Interpersonal sensitivity	.416	.352	1			
4. Depression	.294	.407	.767‡	1		
5. Anxiety	.398	.205	.765‡	.790‡	1	
6. Problematic internet use	-.163	.182	.127	.475*	.242	1

Note. Social withdrawal symptoms as evaluated by the Hikikomori Symptoms Scale (HSS).
* $p < .05$; † $p < .01$; ‡ $p < .001$.

Findings showed that 12.8% (n = 6) of participants from the non-clinical sample reported a lifetime episode of social withdrawal (i.e., hikikomori or brief social withdrawal). Namely, 4.3% (n = 2) revealed hikikomori phenomenon. Importantly, the HQ-25 cut-off score correctly identified them. Therefore, the present study emphasised the usefulness of the online survey as an effective methodology to reach young adults at risk of social withdrawal, in line with a previous study conducted through online survey that showed a prevalence of 6.6% for hikikomori (i.e., 9 subjects among 137 participants) [21].

Focusing on the clinical sample, results highlighted that approximately half (n = 9) of participants reported a lifetime episode of social withdrawal (i.e., hikikomori or brief social withdrawal). In particular, 15.8% (n = 3) of participants reported hikikomori.

No difference in prevalence of social withdrawal lifetime episodes (i.e., hikikomori or brief social withdrawal) according to gender was detected in both samples. This finding is in line with that of another study [12] although several studies have reported a higher prevalence in males [11,13,14].

As for clinical characteristics of social withdrawal, self- and other-directed aggressive behaviours were reported by participants of the clinical sample, in line with findings of previous studies [11-13]. Substance use was also more common among participants of the clinical sample. These clinical features could help clinicians to differentiate between participants with social withdrawal associated to clinical disorders, indicating poorer mental health [7]. Frankova [12] hypothesized that hostility and aggressive behaviours among hikikomori represent maladaptive emotion regulation strategies to deal with high levels of resentment, inner tension, self-doubt and low quality of life. These conditions were more frequent among participants with hikikomori associated with clinical disorders rather than among those who only showed hikikomori, indicating a personality trait (excitability and impulsivity) rather than other conditions. Moreover, clinical diagnoses of Mood Disorder Not Otherwise Specified and Psychotic Disorder Not Otherwise Specified of participants who reported prolonged social withdrawal could also indicate the difficulty in framing the phenomenon of hikikomori using diagnostic categories from the main diagnostic manuals currently available [7].

The results of this study revealed that in both non-clinical and clinical samples, symptoms of social withdrawal as evaluated by the HQ-25 and HSS were strongly associated to overall personality dysfunction. This finding was in line with those of previous studies [13,14,30]. As suggested by Suwa and Suzuki [31], two type of hikikomori may be differentiated: a primary type, with no other psychopathology, and a secondary type, with clinical disorders in comorbidity. Malagon-Amor et al. [13] showed that hikikomori condition was comorbid with different psychiatric disorders, including personality disorders. Specifically, 33 of 190 adults with hikikomori fulfilled the criteria for a personality disorder. Furthermore, Teo et al. [14] provided evidence for the existence of both primary and secondary hikikomori. The authors reported that the majority of 22 participants with a history of hikikomori, 87% (n = 17), were diagnosed with psychiatric disorders. Avoidant and paranoid personality disorders were among the first and the third more commonly observed (i.e., in 41 and 32% of the sample, respectively). On the other hand, five participants did not show other clinical disorders in comorbidity [14]. Our study's findings suggest that the overall personality dysfunction conceptualized as a dimensional *continuum* is associated with hikikomori, with high dysfunction representing a risk factor. Hikikomori condition may represent the outcome of the dysfunctional adaptation process of young adults with high overall personality dysfunction to the increased social and environmental demands typical of young adulthood. Psychotherapy and intervention aimed at increasing psycho-social skills are recommended [13].

In the non-clinical sample, symptoms of social withdrawal were related to interpersonal sensitivity and depression. On the contrary, no significant associations were observed between symptoms of social withdrawal and other symptoms of psychopathology in the clinical sample, except for overall personality dysfunction. In comparison, previous studies have reported significant relationships between social withdrawal and symptoms of affective disorders mainly in clinical samples [11-14]. Regarding the present results, a possible reason is that there was no real association between variables of interest. Another explanation is that there might be an association, but this study was underpowered to detect it because there were not enough participants to detect small-to-medium effects. In fact, near significant positive correlations between social withdrawal, interpersonal sensitivity and anxiety in the clinical sample were also reported.

Furthermore, near significant positive correlations between social withdrawal, anxiety and problematic internet use in the non-clinical sample were found. Considering the fact that symptoms of internet addiction were associated to symptoms of social withdrawal in previous studies conducted in non-clinical and clinical samples [13,18-20], our findings may still be clinically important and warrant further consideration although they did not meet statistical significance. On the other hand, the lack of association between symptoms of social withdrawal and problematic internet use may also indicate that young adults with symptoms of social withdrawal are less motivated to pay attention to the use of the internet and, as a consequence, to not being problematically involved in it. In line with this hypothesis, Teo et al. [14] showed that none of 22 participants with a history of hikikomori scored in the severe category of the Internet Addiction Test. Longitudinal studies should focus their attention on the relationship between problematic internet use and the risk for prolonged social withdrawal in young adults. Findings of the present study seem to suggest that non-social internet use is frequently reported by youth with a lifetime episode of social withdrawal.

Finally, problematic internet use was associated with interpersonal sensitivity (i.e., feelings of personal inadequacy and inferiority in comparison with others) in the non-clinical sample, and to depression (i.e., symptoms of dysphoric mood and affect as well as lack of motivation and loss of interest in life) in the clinical sample. These results are in line with previous studies that showed complex relationships between self-esteem, depression and problematic internet use and the role as risk factors of the former towards the last [32,33].

Findings of this study demonstrated that online surveys are a useful methodology to identify young adults at risk of social withdrawal and decreased mental health. In addition, our findings have possible significant clinical implications for the prevention and treatment of socially withdrawn youths, highlighting the relationship between symptoms of social withdrawal and overall personality dysfunction, in order to improve mental health and prevent a possible public health emergency.

Our results should be interpreted while keeping some limitations in mind. First, the study had a cross-sectional design; therefore, it was not possible to make causal inferences. Second, data are not representative of the entire young adult national population due to the recruitment methods and the small sample size. Third, social desirability response bias may also have affected the results.

We found evidence for hikikomori in non-clinical and clinical samples of Italian young adults. Clinical features of psychopathology (e.g., self- and other-directed aggressive behaviours, substance misuse) associated to social withdrawal could indicate poorer mental health. Moreover, symptoms of hikikomori showed strong association with overall personality dysfunction. Finally, findings of this study suggested that online survey is a useful methodology to identify young adults with hikikomori.

Ethical consideration

All procedures performed in studies involving human participants were in accordance with the 1964 Helsinki declaration and its later amendments or comparable ethical standards, and with the ethical standards of the Ethics Committee of the Department of Dynamic and Clinical Psychology, Sapienza University of Rome.

Acknowledgement

The authors would like to thank the individuals who generously shared their time and experience for the purposes of this project.

Funding

No funding was received for this specific study.

Conflict of interest

The authors declare to have no conflict of interest

Informed consent

Informed consent was obtained from all individual participants included in the study.

Author contributions

AS, RC, and FP contributed to the conceptualization of the study. AS, RC, VS, CL, ES, VDG, FC, AM, and GI contributed to participant recruitment and data collection. AS, RC, FP, VS and AT analyzed the data. AS wrote the original draft of the manuscript. RC and AT provided critical revision of the manuscript. Finally, all the authors have approved the final version of the manuscript.

References

[1] Rubin KH, Coplan RJ, Bowker JC. Social withdrawal in childhood. Annu Rev Psychol 2009;60:141-171. https://doi.org/10.1146/annurev.psych.60.110707.163642

[2] Rubin KH, Coplan RJ, Bowker JC, et al. Social withdrawal and shyness. In: Smith PK, Hart CH, Eds. The Wiley-Blackwell Handbook of Childhood Social Development. Oxford, UK: Wiley-Blackwell 2011, pp. 434-452. http://doi.wiley.com/10.1002/9781444390933.ch23

[3] Barzeva SA, Meeus WHJ, Oldehinkel AJ. Social withdrawal in adolescence and early adulthood: measurement issues, normative development, and distinct trajectories. J Abnorm Child Psychol 2019;47:865-879. https://doi.org/10.1007/s10802-018-0497-4

[4] Coplan RJ, Bowker JC. All alone: multiple perspectives on the study of solitude. In: Coplan RJ, Bowker JC, Eds. The Handbook of Solitude. New York, NY: Wiley 2013, pp. 1-13. https://doi.org/10.1002/9781118427378.ch1

[5] Laursen B, Hartl AC. Understanding loneliness during adolescence: developmental changes that increase the risk of perceived social isolation. J Adolesc 2013;36:1261-1268. https://doi.org/10.1016/j.adolescence.2013.06.003

[6] Baek S-B. Psychopathology of social isolation. J Exerc Rehabil 2014;10:143-147. https://doi.org/10.12965/jer.140132

[7] Teo AR, Nelson S, Strange W, et al. Social withdrawal in major depressive disorder: a case-control study of hikikomori in japan. J Affect Disord 2020;274:1142-1146. https://doi.org/10.1016/j.jad.2020.06.011

[8] Kato TA, Kanba S, Teo AR. Hikikomori: multidimensional understanding, assessment and future international perspectives. Psychiatry Clin Neurosci 2019;pcn.12895. https://doi.org/10.1111/pcn.12895

[9] Kato TA, Kanba S, Teo AR. Defining pathological social withdrawal: proposed diagnostic criteria for hikikomori. World Psychiatry 2020;19:116-117. https://doi.org/10.1002/wps.20705

[10] Bommersbach T, Millard H. No longer culture-bound: hikikomori outside of Japan. Int J Soc Psychiatry 2019;65:539-540. https://doi.org/10.1177/0020764019859379

[11] Chauliac N, Couillet A, Faivre S, et al. Characteristics of socially withdrawn youth in France: a retrospective study. Int J Soc Psychiatry 2017;63:339-344. https://doi.org/10.1177/0020764017704474

[12] Frankova I. Similar but different: psychological and psychopathological features of primary and secondary hikikomori. Front Psychiatry 2019;10:558. https://doi.org/10.3389/fpsyt.2019.00558

[13] Malagón-Amor Á, Martín-López LM, Córcoles D, et al. A 12-month study of the hikikomori syndrome of social withdrawal: clinical characterization and different subtypes proposal. Psychiatry Res 2018;270:1039-1046. https://doi.org/10.1016/j.psychres.2018.03.060

[14] Teo AR, Stufflebam K, Saha S, et al. Psychopathology associated with social withdrawal: Idiopathic and comorbid presentations. Psychiatry Res 2015;228:182-183. https://doi.org/10.1016/j.psychres.2015.04.033

[15] Ranieri F. When social withdrawal in adolescence becomes extreme: the "hikikomori" phenomenon in Italy. Psychiatr Psychol Klin 2015;15:148-151. https://doi.org/10.15557/PiPK.2015.0023

[16] Ranieri F, Luccherino L. Hikikomori: debating a XXI century phenomenon from a clinical point of view. Scand J Child Adolesc Psychiatry Psychol 2018;6:72-79. https://doi.org/10.21307/sjcapp-2018-002

[17] Cerutti R, Spensieri V, Presaghi F, et al. An exploratory study on Internet addiction, somatic symptoms and emotional and behavioral functioning in school-aged adolescents. Clin Neuropsychiatry 2017;14:374-383.

[18] Lee YS, Lee JY, Choi TY, et al. Home visitation program for detecting, evaluating and treating socially withdrawn youth in Korea: visitation program for withdrawn youth. Psychiatry Clin Neurosci 2013;67:193-202. https://doi.org/10.1111/pcn.12043

[19] Tateno M, Teo AR, Ukai W, et al. Internet addiction, smartphone addiction, and hikikomori trait in Japanese young adult: social isolation and social network. Front Psychiatry 2019;10:455. https://doi.org/10.3389/fpsyt.2019.00455

[20] Stavropoulos V, Anderson EE, Beard C, et al. A preliminary cross-cultural study of hikikomori and internet gaming disorder: the moderating effects of game-playing time and living with parents. Addict Behav Rep 2019;9:100137. https://doi.org/10.1016/j.abrep.2018.10.001

[21] Liu LL, Li TM, Teo AR, et al. Harnessing social media to explore youth social withdrawal in three major cities in China: cross-sectional web survey. JMIR Ment Health 2018;5:e34. https://doi.org/10.2196/mental.8509

[22] Umemura T, Watanabe M, Tazuke K, et al. Secure base script and psychological dysfunction in Japanese young adults in the 21st century: using the attachment script assessment. Dev Psychol 2018;54:989-998. https://doi.org/10.1037/dev0000471

[23] Teo AR, Chen JI, Kubo H, et al. Development and validation of the 25-item hikikomori questionnaire (HQ-25). Psychiatry Clin Neurosci 2018;72:780-788. https://doi.org/10.1111/pcn.12691

[24] Derogatis LR. Brief symptom inventory. Baltimore, MD: Clinical Psychometric Research 1975.

[25] Adawi M, Zerbetto R, Re TS, et al. Psychometric properties of the Brief Symptom Inventory in nomophobic subjects: insights from preliminary confirmatory factor, exploratory factor, and clustering analyses in a sample of healthy Italian volunteers. Psychol Res Behav Manag 2019;12:145-154. https://doi.org/10.2147/PRBM.S173282

[26] Pontes HM, Griffiths MD. The development and psychometric evaluation of the Internet Disorder Scale (IDS-15). Addict Behav 2017;64:261-268. https://doi.org/10.1016/j.addbeh.2015.09.003

[27] Monacis L, Sinatra M, Griffiths MD, et al. Assessment of the Italian Version of the Internet Disorder Scale (IDS-15). Int J Ment Health Addict 2018;16:680-691. https://doi.org/10.1007/s11469-017-9823-2

[28] American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th ed. Washington, DC 2013.

[29] Faul F, Erdfelder E, Buchner A, et al. Statistical power analyses using G*Power 3.1: tests for correlation and regression analyses. Behav Res Methods 2009;41:1149-1160. https://doi.org/10.3758/BRM.41.4.1149

[30] Kondo N, Sakai M, Kuroda Y, et al. General condition of hikikomori (prolonged social withdrawal) in Japan: psychiatric diagnosis and outcome in mental health welfare centres. Int J Soc Psychiatry 2013;59:79-86. https://doi.org/10.1177/0020764011423611

[31] Suwa M, Suzuki K. The phenomenon of "hikikomori"(social withdrawal) and the socio-cultural situation in Japan today. Journal of Psychopathology 2013;19:191-198.

[32] Aalbers G, McNally RJ, Heeren A, et al. Social media and depression symptoms: a network perspective. J Exp Psychol Gen 2019;148:1454-1462. https://doi.org/10.1037/xge0000528

[33] Sevelko K, Bischof G, Bischof A, et al. The role of self-esteem in Internet addiction within the context of comorbid mental disorders: findings from a general population-based sample. J Behav Addict 2018;7:976-984. https://doi.org/10.1556/2006.7.2018.130

JOURNAL OF PSYCHOPATHOLOGY 2021;27:115-121
DOI: 10.36148/2284-0249-404

Original article

Knowledge on the COVID-19 pandemic and the nursing role influence anxiety and depression levels: a descriptive correlational study between nurses and general population

Elsa Vitale[1], Vito Galatola[2], Rocco Mea[3]

[1] Department of Mental Health, Local Healthcare Company Bari, Italy; [2] Occidental Hospital in Castellaneta, Local Health Company Taranto, Italy; [3] San Carlo Hospital, Potenza, Italy

SUMMARY

Objectives

To perform if the knowledge on the COVID-19 pandemic and the nursing role could influence anxiety and depression levels in nursing and general population participants.

Methods

A descriptive correlational study was conducted. Participants were divided into nursing and general population groups, respectively. General knowledge on the COVID-19 pandemic was assessed and then, thanks to the Hospital Anxiety and Depression Scale (HADS), anxiety and depression levels were also performed.

Results

400 subjects were enrolled in this study. Nurses and general population recorded the similar trend in anxiety levels ($p = .265$). Nurses recorded higher frequency in normal depressive score than general population ($p = .006$). Significant correlations were reported between: anxiety and depression levels ($p < .001$), anxiety levels and the pandemic knowledge ($p = .024$), anxiety levels and the nursing role ($p = .005$), depression levels and the nursing role ($p < .001$).

Conclusions

The pandemic knowledge and the nursing role might be protective factors both in anxiety and depressive disorders.

Key words: anxiety disorder, depression disorder, disease outbreak, nursing role

Received: October 14, 2020
Accepted: April 29, 2021

Correspondence
Elsa Vitale
Department of Mental Health, Local Healthcare Company Bari, via X marzo, 43, 70026 Modugno, Bari, Italy
E-mail: vitaleelsa@libero.it

How to cite this article: Vitale E, Galatola V, Mea R. Knowledge on the COVID-19 pandemic and the nursing role influence anxiety and depression levels: a descriptive correlational study between nurses and general population. Journal of Psychopathology 2021;27:115-121. https://doi.org/10.36148/2284-0249-404

Introduction

On 31 December 2019, a new viral pneumonia originating in Wuhan, China, was announced to the World Health Organization [1]. Since the beginning of the pandemic, the number of confirmed cases in the world were 6.991.920 with 403.128 deaths [2].

The pandemic brought a rapid and unprecedented changes in daily life, as the cases of soaring viruses, the death toll and the drastic measures to contain the spread of the disease continuing to be impressive. Despite considerable attention to measures identifying infected people and also mental health needs had relatively neglected [3]. Already in previous mass tragedies, particularly caused by infectious diseases, general population felt fear and anxiety with a negative impact on the psychological well-being [4,5]. In fact, literature reported that in the earliest stage of the pandemic, many psychiatric symptoms were recorded, such as persistent depression, anxiety, panic attacks and even self-harm [6], and there were higher levels of depression in people who had themselves or their families and friends in quarantine or suspected of being infected [7].

Moreover, numerous evidences stated that both general population [8-10] and nurses [11-13] experienced panic, disappointment and anger, especially those who had confirmed or suspected SARS-CoV-2 infection; that quarantined people could experience anxiety, boredom, loneliness; and that mental health support should be provided to the needy public. Hence, psychological counseling services quickly spread to provide psychological support to general population [14] every day from 8 to 24 [15].

Many questions related to the pandemic without precise answers, such as when it will end and questions related to the methods of treatment; constant exposure to a flow of information on the pandemic and its effects; the decrease in social relationships due to the pandemic; and the drastic measures taken, might adversely affect people's mental health. Symptoms such as anxiety, depression, fear, stress and sleep disorders are most frequently observed during the COVID-19 pandemic [16-21]. A study involving 253 individuals in one of the regions most affected by the COVID-19 pandemic in China reported a 7% incidence of post-traumatic stress symptoms 1 month after the outbreak of the pandemic [22].

The aim of the present study was to understand if a good knowledge on the COVID-19 pandemic and the nursing role could influence anxiety and depression levels between nurses and general population groups. Specifically, pandemic knowledge focused on: the transmission process of the SARS-CoV-2 infection, the close contact definition, the main prevention measures to consider important in this period, treatments available in the SARS-CoV-2 infection.

Materials and methods

Design

A descriptive correlational study was conducted. The on-line questionnaire was developed in an anonymous form through the Google function: Google Modules and is administered in the period from 25 April 2020 to 30 May 2020 through some pages and nursing groups present on the following Facebook and Instagram pages: #noisiamopronti, Nurse health professional, Professional nurse, Nurses by passion, NurseTimes, Nurse24.it, Nurse Specialist, Nurseallface, Nursing research, NursesInProgress, Nurses, Active Nurses, Nurses Italy, Nurses supporting health, Nursing Mobility, Nursing Competitions, Informed Nurses (Instagram).

Questionnaire

An "ad hoc" questionnaire was created including three sections. The first part concerned the socio-demographic data collection, particularly:

- gender, as female and male;
- age, divided into 5 classes: from 0 to 20 years, from 21 to 40 years, from 41 to 60 years, from 61 to 80 years and over 80 years;
- the educational level, as: elementary school, middle school, diploma and degree;
- the Region of Italy belonging: whether the north, center or south Italy,
- if the participant was a student or a worker;
- if the participant was a nurse or other, indicating by "general population".

The second part of the questionnaire included a total of 25 items that investigated basic knowledge related to SARS-CoV-2 infection (Appendix I). All the information processed was reproduced from the official website of the Italian Higher Institute of Health, on the internet page reserved for the COVID-19 pandemic [23]. Specifically, the basic knowledge was assessed on the method of transmission of the SARS-CoV-2 infection, on the prevention measures to be adopted to contain the pandemic, on the treatments available to date. For each proposed item a Linkert scale was associated with the answers given, attributing the value zero to the correct answers and 1 to the wrong answers.

The third and final section of the questionnaire included the assessment of anxiety and depression levels by administering the Hospital Anxiety and Depression Scale (HADS) [24]. Participants were asked to indicate how they have been feeling during the COVID-19 pandemic period among the different options presented. The answers were associated with a numerical value, the sum of which totaled a profile for both anxiety and depression. For values between zero and 7 the conditions of anxiety and depression were to be considered normal, for values between 8 and 10 the conditions of anxiety and depression were to be considered borderline, for values between 11 and 21 the conditions of anxiety and depression were considered pathological.

Participants

The questionnaire was administered in an online version. Participation was voluntary and no form of personal restitution of the results obtained was involved.

All the information collected had no diagnostic purpose and the results were treated confidentially, guaranteeing complete anonymity, and as such the information acquired cannot in any case be traced back to the natural person who completed the questionnaire.

Data analyses

The collected data were sorted in an Excel sheet and processed with the statistical program SPSS version 20. For the entire first part of the questionnaire concerning the characteristics of the sample collected, all data were grouped by numbers and percentages. For the second part of the questionnaire, data were grouped by numbers and percentages of correct and incorrect answers given, dividing the participants according to their nursing and general population role. The multiple linear regression model was calculated to verify the existence of any statistically significant differences between the two groups. The levels of anxiety and depression were assessed, considering the 3 subgroups obtained from the sum of the responses obtained, namely: normal, borderline, abnormal. The chi-square test was used to evaluate the differences between two groups of participants. Finally, by adding the basic knowledge values on the SARS-CoV-2 topic, Spearman's correlation was calculated to assess how anxiety and depression levels depended on the participant's role and related basic knowledge. All the inferential statistics values with $p < 0.05$ were considered statistically significant.

Results

A total of 400 subjects participated in this investigation (Tab. I). Of these, 320 were females and 80 males. 4 aged until 20 years, 244 aged between 21 and 40 years, 144 were between 41 and 60 years old, 8 aged between 61 and 80 years. Most of the participants stated that they had a diploma as educational qualification (69.25%) and belonged to one of the Regions of north of Italy (52.25%). 94% of participants declared that they were workers and among them the 67.75% were nurses. As regards general knowledge assessment on the COVID-19 pandemic between nurses and general population, some questions on the pandemic were given (Appendix I). There were not many statistically significant differences ($p > 0.05$) between the right and wrong answers between the two groups of participants, with the exception of item no.4 ($p = 0.029$), within the definition of "close contact", in which 105 subjects including 83 nurses and 22 general subjects responded incorrectly. Almost all of the participants answered item no.22 in the right way ($p = 0.008$) and therefore "… in case of fever, cough or breathing difficulties or suspected contact with a positive subject to COVID-19" they knew they must stay at home and not to go to the emergency room and to all medical ambulatories but to contact the doctor or family pediatrician or medical guard by phone. Finally, there was a statistical significance in the reduced number of incorrect responses for item no.24 ($p = 0.019$), as the response to the main treatment for COVID-19 which "remained mainly a symptomatic approach, providing supportive therapies".

TABLE I. *Sample characteristics (n = 400).*

Characteristics	N	%
Sex:		
Female	320	80
Male	80	20
Age:		
0-20 years	4	1
21-40 years	244	60
41-60 years	144	36
61-80 years	8	2
Over 80 years	0	0
Educational qualification:		
Primary school	5	1.25
Middle school	77	19.25
Diploma	277	69.25
Graduation	41	10.25
Region of Italy:		
North	209	52.25
Centre	106	26.5
South	85	21.25
Profession:		
Student	24	6
Worker	376	94
Role:		
Nurses	271	67.75
General population	129	32.25

Additionally, by considering the anxiety and depression levels detecting between the two groups of participants (Tab. II), nurses reported higher normal levels in anxiety disorders (33.75%) than general population (11.25%). However, the anxiety level trend was not statistically significant ($p = 0.265$). As regards the depression levels, there was a statistical significance between the depression values recorded between the nurses and general population ($p = 0.006$). In fact, data reported that only 22 nurses and 22 general population participants recorded abnormal depressive scores comparing to 210 nurses and 78 subjects belonging to general population who registered normal depressive values.

By correlating the levels of anxiety and depression, the state of basic knowledge on the topic and the professional role (Tab. III) there were significant correlations between anxiety and depression levels ($p < 0.001$), between anxiety levels and knowledge on the topic "SARS-CoV-2 infection" ($p = 0.024$), between anxiety levels and the nursing role ($p = 0.005$). Also, depression levels significantly correlated with the nursing role ($p < 0.001$).

Discussion

The aim of the present study was to understand if a good knowledge on the COVID-19 pandemic and the nursing role could positively influence anxiety and depression levels, differentiating them between nurses and general population groups. In the literature, studies comparable to ours for purpose and methodology were not present. Anyway, previous studies have just

TABLE II. *Anxiety and Depression scores between nurses and general population.*

	Anxiety levels				Depression levels			
	Normal scores: 0-7 n(%)	Borderline scores: 8-10 n(%)	Abnormal scores: 11-21 n(%)	P-value	Normal scores: 0-7 n(%)	Borderline scores: 8-10 n(%)	Abnormal scores: 11-21 n(%)	P-value
Nurses	135 (33.75%)	69 (17.25%)	67 (16.75%)	0.265	210 (52.5%)	39 (9.75%)	22 (5.5%)	0.006*
General population	45 (11.25%)	40 (10%)	44 (11%)		78 (19.5%)	29 (7.25%)	22 (5.5%)	

p < 0.05 is statistically significant

TABLE III. *Correlations between anxiety and depression levels, the knowledge condition and the working roles of participants.*

	Anxiety levels	Depression levels	Knowledge condition	Roles: nurses/ general population
Anxiety levels	----	< 0.001*	0.024*	0.005*
Depression levels	< 0.001*	----	0.720	< 0.001*
Knowledge condition	0.024*	0.720	----	0.642
Nurses/general population	0.005*	<0.001*	0.642	----

p < 0.05 is statistical significant

demonstrated important associations between anxiety and depression disorders and COVID-19 [17-21,25], as anxiety and mood disorders were the most common mental health problems in the general population around the world [26].

The present findings reported that the pandemic knowledge and the nursing role influenced depressive conditions (p = .006), as nurses recorded more normal scores (52.5%) than general population (19.5%). On the other hand, data recorded no statistical significance between nurses and general population as concern anxiety disorders (p = .265). Additionally, significant correlations were found between knowledge and anxiety levels (p = .024), nursing and general population role and anxiety levels (p = .005), too. The present findings were in agreement to current literature. In fact, a study conducted in China looked at the psychological and mental health impact of the general Chinese population within the first two weeks of the COVID-19 pandemic established the prevalence of psychiatric symptoms and identified the risk and protective factors that contributed to psychological stress [27]. The respondents' depression, anxiety and stress levels registered 12.2% of participants with a moderate depression and 4.3% with a severe and extremely severe depression. For the anxiety subscale, 20.4% of participants recorded a moderate anxiety disorders and 8.4% an extremely severe anxiety disorder, respectively. Also the same study investigated knowledge about COVID-19 and highlighted that the most common perceived route of transmission was through droplets (92.1%), followed by contaminated objects (73.7%) and airborne transmission (60.5%). The most common source of health information for COVID-19 came from the Internet (93.5%). Most respondents (75.1%) were very satisfied or fairly satisfied with the amount of health information available. However, this study did not correlate knowledge level with anxiety a,nd depressive disorders, so an ad hoc comparison with our findings was not possible to perform. Furthermore, another study conducted in Turkey investigated depression and anxiety levels during the COVID-19 pandemic. The study also considered the effect on these variables of some factors that might influence them such as age, gender, place of residence, the presence of a chronic disease, a positive COVID-19 friend or relative and a current history for previous psychiatric illness. Data found that women and people living in urban areas, with a COVID-19 patient among friends or relatives, previously or currently undergoing psychiatric treatment and with at least one accompanying chronic disease, were most severely affected [28]. However, knowledge level on the COVID-19 pandemic was not assessed and discussed. From literature, in Italy, there were several studies conducted on the psychological impact that COVID-19 had both for nurses [29,30] and general population [31,32], but few investigations focused on association to mental disorders and knowledge to the pandemic [33]. For example, the Pagnini et al. [33] study suggested that during the first week of the COVID-19 outbreak in Italy, general population were well informed and had a relatively stable level of worries. This aspect could be in agreement with our findings, however this study did not associate knowledge to anxiety and depression disorders, but it focused on mental well-being challenges associated with more cognitive rigidity and emotional instability.

Research limitations

Data were collected on-line and there was no form of iteration with the participants. The greater shortcoming of this study was the instrument of assessment of anxiety and depression as literature considered the HADS a self-assessment scale developed for detecting states of depression and anxiety in the setting of a hospital medical outpatient clinic. However, it contributed to evidence on anxiety and depression differences between nurses and general populations.

Conclusions

The present study reported that knowledge and the nursing role might be protective factors both in anxiety and depressive disorders. However, future studies with a higher number of subject will be desirable in order to confirm and generalize the data of our observational study which, at least for the Italian reality, could be considered as pilot.

Ethical consideration

All participants voluntary agreed to participate to the on-line survey. The current research has been carried out in accordance with the Code of ethics of the World Medical Association (Declaration of Helsinki).

Acknowledgement

We thank all the participants.

Funding

None.

Conflict of interest

The authors declare to have no conflict of interest

Author contributions

Conceptualization, methodology, software, validation, data curation formal analysis, writing-original draft preparation and writing-review and editing: VE; data collection: GV and MR. All authors have read and agreed to the published version of the manuscript.

References

[1] World Health Organization (WHO). (11 March 2020). WHO Director-General's opening remarks at the media briefing on COVID-19 March 2020 (https://www.who.int/dg/speeches/detail/who-director-general-s-opening-remarks-at-themedia-briefing-on-covid-19).

[2] Ministero della salute. COVID-19 - Situazione nel mondo. Ultimi dati OMS. Fonte: Health Emergency Dashboard, 2020 (http://www.salute.gov.it/portale/nuovocoronavirus/dettaglioContenutiNuovoCoronavirus.jsp?lingua=italiano&id=5338&area=nuovoCoronavirus&menu=vuoto).

[3] Dell'Osso L, Muti D, Toschi D, et al. Contagion: society, brain and culture. Journal of Psychopathology 2020;26:121-123. https://doi.org/10.36148/2284-0249-394

[4] Balaratnasingam S, Janca A. Mass hysteria revisited. Curr Opin Psychiatry 2006;19:171-174. https://doi.org/10.1097/01.yco.0000214343.59872.7a

[5] Vitale E, Moretti B, Noternicola A, et al. How the Italian Nursing students deal the pandemic COVID-19 condition. Acta Biomed 2020;91:e2020007. https://doi.org/10.23750/abm.v91i12-S.9860

[6] Liu YB, Chen XY Miao GD, et al. Recommendations on diagnostic criteria and prevention of SARS-related mental disorders. J Clin Psychol Med 2003;13:188-191. https://doi.org/10.24869/psyd.2020.32

[7] Talevi D, Pacitti F, Socci V, et al. The COVID-19 outbreak: impact on mental health and intervention strategies. Journal of Psychopathology 2020;26:162-168. https://doi.org/10.36148/2284-0249-393

[8] Hossain MM, Tasnim S, Sultana A, et al. Epidemiology of mental health problems in COVID-19: a review. F1000Res 2020;9:636. https://doi.org/10.12688/f1000research.24457.1

[9] Aguglia E. Living in the era of COVID-19: new challenges for psychopathology. Journal of Psychopathology 2020;26:183-185. https://doi.org/10.36148/2284-0249-391

[10] Braun-Lewensohn O, Abu-Kaf S, Kalagy T. Hope and resilience during a pandemic among three cultural groups in Israel: the second wave of COVID-19. Front Psychol 2021;12:637349. https://doi:10.3389/fpsyg.2021.637349

[11] Maben J, Bridges J. COVID-19: supporting nurses' psychological and mental health. J Clin Nurs 2020;29:2742-2750. https://doi.org/10.1111/jocn.15307

[12] Turale S, Meechamnan C, Kunaviktikul W. Challenging times: ethics, nursing and the COVID-19 pandemic. Int Nurs Rev 2020;67:164-167. https://doi.org/10.1111/inr.12598

[13] Galehdar N, Kamran A, Toulabi T, et al. Exploring nurses' experiences of psychological distress during care of patients with COVID-19: a qualitative study. BMC Psychiatry 2020;20:489. https://doi.org/10.1186/s12888-020-02898-1

[14] China NHCO. Principles for emergency psychological crisis intervention for the new coronavirus pneumonia, 2020 (http://www.nhc.gov.cn/jkj/s3577/202001/6adc08b966594253b2b791be5c3b9467.shtml).

[15] Liu S, Yang L, Zhang C, et al. Online mental health services in China during the COVID-19 outbreak. Lancet Psychiatry 2020;7:e17-e18. https://doi.org/10.1016/S2215-0366(20)30077-8

[16] Torales J, O'Higgins M, Castaldelli-Maia JM, et al. The outbreak of COVID-19 coronavirus and its impact on global mental health. Int J Soc

Psych 2020;66:317-320 https://doi.org/10.1177/0020764020915212

17 Vitale E, Mea R, Di Dio F, et al. Anxiety, insomnia and body mass index scores in Italian nurses engaged in the care of COVID-19 patients. Endocr Metab Immune Disord Drug Targets 2020;20:1. [Epub ahead of print]

18 Vitale E, Galatola V, Mea R. Exploring within and between gender differences in burnout levels in Italian nurses engaged in the COVID-19 health emergency: a cohort observational study. Minerva Psichiatr 2020;61:162-170.

19 Vitale E, Galatola V, Mea R. Observational study on the potential psychological factors that affected Italian nurses involved in the COVID-19 health emergency. Acta Biomed Health Professions 2021;92:e2021007.

20 Vitale E, Casolaro S. Anxiety, burnout and depression levels according to sex and years of work experience in Italian nurses engaged in the care of COVID-19 patients. Journal of Evidenced-Based Psychotherapies 2021;21:83-96.

21 Vitale E. Anxiety, depression and insomnia conditions in Italian nurses during the first and the second waves of the COVID-19 pandemic. Journal of Evidenced-Based Psychotherapies 2021;21:69-82.

22 Liu N, Zhang F, Wei C, et al. Prevalence and predictors of PTSS during COVID-19 outbreak in China hardest-hit areas: gender differences matter. Psychiatry Res 2020;287:112921. https://doi.org/10.1016/j.psychres.2020.112921

23 Speciale COVID-19. Cosa sapere, 2020 (http://www.ISS per COVID-19 – ISS).

24 Bjelland I, Dahl AA, Haug TT, et al. The validity of the Hospital Anxiety and Depression scale. An updated literature review. J Psychosom Res 2002;52:69-77.

25 Coughlin SS. Anxiety and depression: linkages with viral diseases. Public Health Rev 2012;34:1-17. https://doi:10.1007/BF03391675

26 Prince M, Patel V, Saxena S, et al. No health without mental health. Lancet 2007;370:859. https://doi.org/10.1016/S0140-6736(07)61238-0

27 Wang C, Pan R, Wan X, et al. Immediate psychological responses and associated factors during the initial stage of the 2019 Coronavirus Disease (COVID-19) epidemic among the general population in China. Int J Environ Res Public Health 2020;17:1729. https://doi.org/10.3390/ijerph17051729

28 Ferrucci R, Averna A, Marino D, et al. Psychological impact during the first outbreak of COVID-19 in Italy. Front Psychiatry 2020;11:559266. https://doi.org/10.3389/fpsyt.2020.559266

29 Siracusano A, Di Lorenzo G, Longo L, et al. Building a resilient hospital in Tor Vergata: the role of emotional defusing for health care workers during COVID-19 pandemic. Journal of Psychopathology 2020;26:131-133. https://doi.org/10.36148/2284-0249-403

30 Pelizza L, Pupo S. Public mental health outpatient service at the time of the COVID-19 pandemic: did we have any other choice? Journal of Psychopathology 2020;26:186-187. https://doi.org/10.36148/2284-0249-398

31 Özdin S, Özdin ŞB. Levels and predictors of anxiety, depression and health anxiety during COVID-19 pandemic in Turkish society: the importance of gender Int J Soc Psychiatry 2020. https://doi:10.1177/0020764020927051 [Epub ahead of print]

32 Rossi R, Socci V, Talevi D, et al. COVID-19 pandemic and lockdown measures impact on mental health among the general population in Italy. Front Psychiatry 2020;11:790. https://doi.org/10.3389/fpsyt.2020.00790.

33 Pagnini F, Bonanomi A, Tagliabue S, et al. Knowledge, concerns, and behaviors of individuals during the first week of the coronavirus disease 2019 pandemic in Italy. JAMA Netw Open 2020;3:e2015821. https://doi.org/10.1001/jamanetworkopen.2020.15821

Appendix I. Items concerned general knowledge on the Covid-19 pandemic

Items

Transmission mode:
Item 1: Some coronaviruses can be transmitted from person to person, usually after close contact with an infected patient, for example between family members or in a healthcare setting. The new Coronavirus responsible for respiratory disease Covid-19 can also be transmitted from person to person through close contact with a probable or confirmed case.

Item 2: The new Coronavirus is a respiratory virus that mainly spreads through close contact with a sick person. The primary route of contamination are the droplets of the breath of infected people, for example through: saliva, coughing and sneezing, direct personal contacts, hands, for example by touching the mouth, nose or eyes with contaminated (not yet washed) hands. In rare cases, infection can occur through faecal contamination. Normally respiratory diseases are not transmitted with food, which in any case must be handled respecting good hygiene practices and avoiding contact between raw and cooked food.

Definition of close contact
Item 3: A person who lives in the same house as a COVID-19 case;
Item 4: A person who has had direct physical contact with a COVID-19 case (eg handshake);
Item 5: A person who has had unprotected direct contact with the secretions of a COVID-19 case (for example, touching used paper tissues with bare hands);
Item 6: A person who has had direct contact (face to face) with a case of COVID-19, at a distance of less than 2 meters and lasting longer than 15 minutes;
Item 7: A person who has been in a closed environment (for example classroom, meeting room, hospital waiting room) with a case of COVID-19 for at least 15 minutes, at a distance of less than 2 meters;
Item 8: A healthcare professional or other person providing direct assistance to a COVID-19 case or laboratory personnel handling samples of a COVID-19 case without using the recommended PPE or using unsuitable PPE;
Item 9: A person who has traveled by plane sitting in the two adjacent seats, in any direction, of a COVID-19 case, travel companions or carers and crew members assigned to the section of the plane where the index case was seated (if the index case has a severe symptomatology or has moved within the plane, causing greater passenger exposure, consider all passengers seated in the same section of the plane or in the whole as close contacts airplane);
Item 10: The epidemiological link may have occurred within a period of 14 days before or after the onset of the disease in the case under consideration.

Prevention: what measures to take
Item 11: Wash your hands often. In all public places (gyms, supermarkets, pharmacies) and other meeting places, the use of hydro-alcoholic solutions for hand washing is recommended;
Item 12: Avoid close contact with people suffering from acute respiratory infections;
Item 13: Avoid hugs and handshakes;
Item 14: Maintain, in social contacts, an interpersonal distance of at least one meter;
Item 15: Practice respiratory hygiene (sneezing and/or coughing in a handkerchief avoiding hand contact with respiratory secretions);
Item 16: Avoid the promiscuous use of bottles and glasses, particularly during sports activities;
Item 17: Do not touch your eyes, nose and mouth with your hands;
Item 18: Cover your mouth and nose if you sneeze or cough;
Item 19: Do not take antiviral drugs and antibiotics, unless they are prescribed by your doctor;
Item 20: Clean the surfaces with chlorine or alcohol based disinfectants;
Item 21: It is strongly recommended in all social contacts to use respiratory protection as an additional measure to other personal hygiene-health protection measures;
Item 22: If you experience a fever, cough or breathing difficulties and you suspect that you have been in close contact with a person with COVID-19 respiratory disease, stay indoors, do not go to the emergency room or doctor's office but call the doctor on the phone. family, pediatrician or medical guard.

Treatment:
Item 23: At present, there is no specific treatment for the disease caused by the new coronavirus;
Item 24: Treatment remains mainly based on a symptomatic approach, providing supportive therapies (eg oxygen therapy, fluid management) to infected people, which can be very effective;
Item 25: Several clinical trials are underway to treat COVID-19 disease. The Italian Medicines Agency (AIFA) provides information on its website about the drugs that are made available to Covid-19 patients.

JOURNAL OF PSYCHOPATHOLOGY 2021;27:122-124
DOI: 10.36148/2284-0249-386

Case report

On the edge of Capgras' syndrome

Andrea Turano[1], Cecilia Caravaggi[2], Giuseppe Ducci[2], Silvia Bernardini[2], Pier Luca Bandinelli[2]

[1] *Department of Pyschiatry, Sapienza University of Rome, Sant'Andrea Hospital, Rome, Italy;*
[2] *Department of Mental Health ASL Roma 1, Rome, Italy*

SUMMARY

Capgras' syndrome, the delusional belief in the existence of doubles of others or of oneself, belongs to the delusional misidentification syndromes (DMSs), a group of syndromes characterized by delusional misidentification of oneself and/or of other people. These syndromes are not codified as diagnoses per se on the DSM-5 or on the ICD-11, and are usually seen as specific presentations of broader psychiatric disorders. Capgras' syndrome has been shown on both psychiatric and non-psychiatric disorders, thus not being a manageable tool in helping clinicians to define a diagnosis. Presenting what we believe is a special case of Capgras' syndrome, we aim to propose a characterization of such syndrome very specific of schizophrenic – and thus psychiatric – conditions, which may turn especially useful in clinical pictures where no other psychiatric or medical symptoms are found and can help defining a diagnosis.

Key words: Capgras' syndrome, delusional misidentification syndromes, differential diagnosis

Received: April 30, 2020
Accepted: December 10, 2020

Correspondence
Pier Luca Bandinelli
Department of Psychiatry, SPDC San Filippo Neri, via G. Martinotti 20, 00135 Rome, Italy
Tel. +39 3394878959
E-mail: pierluca.bandinelli@aslroma1.it

Conflict of interest
The Authors declare no conflict of interest

How to cite this article: Turano A, Caravaggi C, Ducci G, et al. On the edge of Capgras' syndrome. Journal of Psychopathology 2021;27:122-124. https://doi.org/10.36148/2284-0249-386

> *Non c'è uomo che a forza di portare una maschera,*
> *non finisca per assimilare a questa anche il suo vero volto.*
>
> (Nathaniel Hawthorne; La lettera scarlatta)

Introduction

In classic psychopathology there was a tendency toward giving names to specific presentations of delusional symptoms, mainly basing on their content. To name a few, in 1880 Jules Cotard gave a lesson on what is now known as the Cotard's syndrome, in which the subject believes he/she is dead, does not exist or has lost one or more of his internal organs [1]; in 1921 Gaëtan Gatian de Clérambault described the erotomanic delusion, today also known as the De Clérambault's syndrome [2]; in 1923 Capgras and Reboul-Lachaux wrote the first paper on the so called Capgras' syndrome, in which the subject believes one or more of the people in his life have been replaced by perfect doubles [3]; in 1927 Courbon and Fail described the case of a patient who believed that her two persecutors continuously changed aspect taking those of her dear ones, and called it Fregoli's syndrome after the Italian actor Leopoldo Fregoli [4]. The present paper focuses on Capgras' syndrome. In today's psychopathology both Capgras' and Fregoli's syndromes belong to the delusional misidentification syndromes (DMS), a group of syndromes characterized by delusional misidentifications of oneself and/or other people. In the beginning, the DMS encompassed four forms of delusions, namely Capgras, Fregoli, subjective doubles and intermetamorphosis syndromes, but in later years were also included reverse Capgras, reverse Fregoli, reverse subjective doubles and reverse intermetamorphosis [5].

In a paper from 1996, Mojtabai showed how following classic definitions of these syndromes only a small number of delusional patients met their criteria, while a broader definition of the DMS allowed for a greater inclusion of blurred cases [6]. Such effort to broaden and redefine the classic criteria of the DMS was due to a growing dissatisfaction with the limitations of their classification, which seemed to fail to adequately represent the diversity of misidentification phenomena observed [7,8].

The definition of Capgras' syndrome that most accurately describes cases similar to that of the original paper from Capgras and Reboul-Lachaux comes from Berson: "The delusional belief in the existence of doubles of others or of oneself or of both" [9]. However, since misidentification may occur even in the absence of the belief in doubles, Capgras' syndrome is sometimes used to describe patients who believe that someone (usually one or a few close relatives) are simply impostors. We believe the case described in the present report can be regarded as a special form of Capgras' syndrome, where the patient does not state his parents are doubles but still regards them as impostors.

Besides the aforementioned common concern on the narrow definitions of DMS, we also believe that the specific characteristics of single presentations of such syndromes have not been described closely enough, hence leading to the creation of a group of syndromes very unspecific of a defined psychiatric disorder. While several authors consider these specific psychotic conditions as possible clinical manifestations of well-defined psychiatric disorder [10,11], other authors wonder whether they could be different disorders or unusual presentations of pre-existing DSM-5 conditions [10]. Furthermore, the analytical description of these psychiatric conditions is absent in the modern diagnostic categories [12]. As a matter of fact, though classically regarding such delusional contents as "syndromes", clinicians never use them in psychiatric practice as diagnoses per se, simply regarding them as useful tools to diagnose a broader psychiatric disorder (like schizophrenia, to name one). A recent systematic review of 255 published cases of Capgras' syndrome [13] reported that the three most frequent diagnoses in which it manifested were schizophrenia (32%), organic psychosis (19%) and dementia (15%). We believe there is some specificity in the clinical presentation and pattern of creation of these syndromes, and that the exploration of their characteristics may shed light on the underlying disorder in cases where the DMS is the only manifest symptom. Therefore, we present the case of a patient whom, though not being in his first psychotic episode and already having a diagnosis of schizophrenia, embodies what we believe are peculiar traits of schizophrenic Capgras' syndrome.

Case report

The patient was a 44-year-old man with untreated chronic schizophrenia, admitted to the ward due to an acute psychotic episode. Angelo, as we will call him, was in a state of confusion, perplexity, a mild mixed affective state, and frankly delusional. He declared how he had recently discovered that his parents were both of the opposite sex, i.e. his father was actually a woman and his mother was actually a man, and that therefore they were not his real parents. When asked why his parents would have had to lie to him all this time, paranoid thoughts emerged, as he claimed that their true goal was to obstruct him from pursuing the catholic faith. Angelo also told us the story of how he had the huge revelation: a couple of months earlier he received what he described as a gentle, delicate look from his father, and this clearly showed that he was a woman, since (as he said) men don't give this kind of looks; then, after a few weeks, his mother fell and hit her knee on the ground, and while showing the injured part to Angelo, he clearly understood (though not directly seeing them) that she had male genitalia between her legs. During the hospitalization, as the medications slowly showed effect, Angelo became progressively less confused and paranoid, and by the time of the discharge (22 days after the admission) he no longer believed his parents were impostors, nor of the opposite sex.

During the hospitalization, extended laboratory tests did not reveal any significant abnormality. Cranial MRI, EEG and ECG were all unremarkable. Clinical internal and neurological examinations were both normal. Results of the MMPI-2 mainly showed a lack of dominance, a low perceived need for psychiatric treatment, a weak sense of his assertiveness, a difficulty in being in contact with his desires and last a slight degree of psychopathy and paranoia. During the permanence in our ward the patient was treated with paliperidone 9 mg/die, lithium carbonate 600 mg/die, citalopram 10 mg/die and clonazepam 10 ml 2.5 mg/ml 30gtt/die. The choice of therapy was motivated by the fact that in the conditions of acute delusion, in addition to the obvious choice of an antipsychotic drug, it is also necessary to control the patient's behavior and stabilize the mood condition that often provides the substrate for the delusional symptoms.

Discussion

As mentioned earlier, Capgras' delusion is not specific of a defined psychiatric or medical condition. During the twentieth century, Capgras' delusion was thought to occur mainly in schizophreniform psychoses [14], but after the publication of Ellis and Young's famous paper in 1990 [15] the proportion of cases reporting the delusion in the context of medical (mainly neurological) disorders notably increased. Still, schizophrenia survives as the most common diagnosis. A systematic review written by Pandis et al. in 2019 [13] pointed out the underlying diagnoses of 255 cases of Capgras' syndrome. Of these, 144 cases occurred in the context of functional psychiatric disorder, while 111 had an identified organic etiology. The most frequent psychiatric diagnoses were schizophrenia (32%), schizoaffective disorder (6%) and bipolar disorder (6%), while the most common medical conditions were organic delusional disorder (19%) and dementia (15%). The clinical features of Capgras' delusion in the mentioned disorders were somewhat different: though no difference emerged in gender, organic delusions had a statistically significant prevalence of misidentifications concerning the spouse and inanimate objects, while the misidentification of a parent was significantly more frequent in psychiatric conditions. Intuitively so, symptoms associated with Capgras' delusion also differed between functional and organic cases: the presence of paranoid thoughts, auditory hallucinations, formal thought disorders and aggression were all statistically predominant in psychiatric syndromes, while visual hallucinations were more frequent in organic causes of the delusion.

The delusional misidentification syndrome we described shows in our opinion a pattern of development typical of schizophrenic conditions. Though Capgras' syndrome has been classically associated in some cases with prosopagnosia [16], given that Capgras himself saw at its core a natural dissociation between recognition and identification of familiar faces [17], most cases of purely psychiatric Capgras' syndrome show a pattern of formation that goes beyond a static and permanent inability to assign familiarity to known faces. We state that true psychiatric misidentification delusions stem from acute episodes characterized by pre-delusional mood or deep mood oscillations that generate "rumor", i.e. temporary interference in perception that impair one's ability to recognize familiar faces [18]. Once perception is altered, once it becomes "delusional", even from intact logical abilities can develop a delusional misidentification syndrome [18]. This developmental pattern of certain cases of Capgras' syndrome, as the one described earlier, seems to us very specific of schizophreniform disorders, and thus could lead to more precise diagnosis and treatment in cases where no other psychiatric or medical symptoms occur.

References

1. Debruyne H, Portzky M, Van den Eynde F, Audenaert K. Cotard's syndrome: a review. Curr Psychiatry Rep 2009;11:197-202. https://doi.org/10.1007/s11920-009-0031-z
2. Berrios GE, Kennedy N. Erotomania: a conceptual history. Hist Psychiatry 2002;13:381-400. https://doi.org/10.1177/0957154X0201305202
3. Capgras J, Reboul-Lachaux J: L'illusion des "sosies" dans un délire systématisé chronique. Ann Med Psychol (Paris) 1923;81:186.
4. Ellis HD, Whitley J, Luauté JP. Delusional misidentification. The three original papers on the Capgras, Frégoli and intermetamorphosis delusions. (Classic Text No. 17). Hist Psychiatry 1994;5:117-46. https://doi.org/10.1177/0957154X9400501708
5. Rodrigues AC, Banzato CE. Delusional misidentification syndrome: why such nosologic challenge remains intractable. Psychopathology 2006;39:296-302. PubMed PMID: 16974136.
6. Mojtabai R. Identifying misidentifications: a phenomenological study. Psychopathology 1998;31:90-5. PubMed PMID: 9561552.
7. Odom-White A, de Leon J, Stanilla J, et al. Misidentification syndromes in schizophrenia: case reviews with implications for classification and prevalence. Aust N Z J Psychiatry 1995;29:63-8. PubMed PMID: 7625978.
8. Banov MD, Kulick AR, Oepen G, et al. A new identity for misidentification syndromes. Compr Psychiatry 1993;34:414-7. PubMed PMID: 8131387.
9. Berson RJ. Capgras' syndrome. Am J Psychiatry 1983;140:969-78. PubMed PMID: 6869616.
10. Atta K, Forlenza N, Gujski M, et al. Delusional misidentification syndromes: separate disorders or unusual presentations of existing DSM-IV categories? Psychiatry (Edgmont) 2006;3:56-61. PMID: 20975828; PMCID: PMC2963468.
11. Barrelle A, Luauté JP. Capgras syndrome and other delusional misidentification syndromes. Front Neurol Neurosci 2018;42:35-43. https://doi.org/10.1159/000475680
12. Greener, M. New insights into delusional misidentification syndromes. Progress in Neurology and Psychiatry 2017;21:33-5.
13. Pandis C, Agrawal N, Poole N. Capgras' delusion: a systematic review of 255 published cases. Psychopathology 2019;52:161-73. https://doi.org/10.1159/000500474
14. Bienenfeld D, Brott T. Capgras' syndrome following minor head trauma. J Clin Psychiatry 1989;50:68-9. PMID: 2914883.
15. Anderson DN. The delusion of inanimate doubles. Implications for understanding the Capgras phenomenon. Br J Psychiatry 1988;153:694-9. https://doi.org/10.1192/bjp.153.5.694
16. Ventriglio A, Bhugra D, De Berardis D, et al. Capgras and Fregoli syndromes: delusion and misidentification. Int Rev Psychiatry 2020;32:391-5. https://doi.org/10.1080/09540261.2020.1756625
17. Luauté JP, Bidault E. Capgras syndrome: agnosia of identification and delusion of reduplication. Psychopathology 1994;27:186-93. https://doi.org/10.1159/000284868
18. Bandinelli PL, Palma C, Penna MP, et al. Logic and context in schizophrenia. In: Minati G, Pessa E, Abram M, Eds. Systemics of emergence: research and development. Boston, MA: Springer 2006. https://doi.org/10.1007/0-387-28898-8_8