

# LIFE SPAN AND DISABILITY

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Psychology  
Social issues  
Education  
Rehabilitation  
Habilitation

- Effects of a non-intensive tact training on the emission of spontaneous verbal operants
- Early numerical skills in individuals with Down Syndrome
- Burden and professional quality of life of caregivers: The clinical role of suppression and resilience
- The link between apathy and Alzheimer's Disease: Psychometric tools and implications for treatment
- Level-balanced psychomotor support program for preschool children with ID
- Religious coping among caregivers of differently-abled children



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**Unit of Psychology**  
Oasi Research Institute – IRCCS, Troina, Italy



## LIFE SPAN AND DISABILITY

Psychology, Social issues, Education, Rehabilitation, Habilitation

Founded in 1998, Life Span And Disability promotes interdisciplinary research on psychological, social, educational, rehabilitative and neuro-psychological aspects of the human life span. The aim is to disseminate scientific studies tapping on cognitive, emotional and interpersonal – transient or permanent – problems that may occur during the individual's life span (e.g., adolescence, unemployment, retirement, fertility drop, normal and pathological aging), causing uneasiness or permanent disability. Neuropsychological and social aspects of Intellectual Disability, as well as rehabilitation strategies to improve the cognitive and adaptive functions and the quality of life of these persons are the target of scientific papers included in the Journal. Attention is focused on potential or residual skills and competences that might be enhanced to promote individual's fulfillment and cognitive development. Personal skills and abilities are also considered from educational, social, environmental point of views, in relation with the bio-psychological bases and/or data derived from empirical research. Both quantitative and qualitative methodological approaches are welcomed.

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## Summary

Effects of a non-intensive tact training on the emission of spontaneous verbal operants in different non-instructional settings <i>Silvia Iacomini, Antonella Diano, Luca Vascelli &amp; Vanessa Artoni</i>	7
Early numerical skills in individuals with Down Syndrome <i>Silvia Stefanelli, Maristella Scorza &amp; Giacomo Stella</i>	29
Burden and professional quality of life of caregivers: The clinical role of suppression and resilience <i>Emanuele M. Merlo, Federica Sicari, Fabio Frisone, Angela Alibrandi &amp; Salvatore Settineri</i>	55
The link between apathy and Alzheimer's Disease: The role of psychometric tools and the possible implications for treatment <i>Giovanna Furneri, Grazia Razza, Mario Santagati, Filippo Drago, Santo Di Nuovo &amp; Filippo Caraci</i>	85
Level-balanced psychomotor support program for preschool children with Intellectual Disabilities <i>Viktor Hladush, Alla Symko, Oleksiy Havrilov &amp; Viera Šilonová</i>	113
Religious coping among caregivers of differently-abled children <i>Mubashir Gull &amp; Akbar Husain</i>	133





# Effects of a non-intensive tact training on the emission of spontaneous verbal operants in different non-instructional settings

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## Abstract

*The study evaluates the effects of a tact training on the emission of three verbal operants (i.e., tacts, mands and conversational units) in non-instructional contexts. The participants were a 16-year-old boy with autism and a 14-year-old girl with severe intellectual disability. Both students had achieved the listener repertoire and presented a level of verbal development defined as speaker and emergent speaker, respectively. They were selected on the basis of their difficulties in the production of spontaneous language in different non-instructional settings. A delayed multiple probe design across participants was used for the study. The number of pure mands, pure tacts and conversational units was measured in three non-instructional settings as dependent variables. The tact training consisted of an increase of 100 learn units in the emission of tacts compared to the average daily number of learn units received by each participant. Results show that for both students the procedure was effective in increasing the number of pure tacts and conversational units emitted in non-instructional settings.*

**Keywords:** Tact; Mand; Conversational Unit; Autism; Intellectual Disability.

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## 1. Introduction

Language acquisition occurs spontaneously in the first years of the typical life of developing children. In fact, the quantity of words acquired is not attributable to direct teaching (Greer & Ross, 2008). Children learn easily to communicate by responding to environmental antecedents and by interacting spontaneously in the presence of natural Establishing Operations (EOs; Pistoljevic, Cahill, & Casarini, 2010).

Functional communication is compromised in children with autism (DSM-5; American Psychiatric Association – APA, 2013). It has been estimated that only half of the children with autism and related disabilities use speech as a form of communication (Lord & Paul, 1997). Typically, a child with autism, in the presence of a non-verbal stimulus, does not emit any form of spontaneous interaction. The child remains silent and speaks only if verbally stimulated by another person, for example when a question is asked (Ingenmey & Van Houten, 1991). Communication is also one of the areas commonly compromised in Intellectual Disability (ID) (Kurtz, Boetler, Jarmolowicz, Chin, & Hagopian, 2011; Matson, Horovitz, Kozlowski, Sipes, Worley, & Shoemaker, 2011; Matson, Sipes, Horovitz, Worley, Shoemaker, & Kozlowski, 2011; van der Schuit, Segers, van Balkom, & Verhoeven, 2011). This condition is especially true for people with severe ID because, in most cases, they do not possess any verbal skills (Matson, Terlonge, Gonzalez, & Rivet, 2006). As far as expressive language is concerned, it seems that individuals with ID only use language to communicate simple requests for objects or rejections rather than to request the attention of other people (Cascella, 2005).

Overall, it is important to teach children with autism and/or ID to emit tacts (i.e., denominations) for those objects or events that are under the control of a non-verbal antecedent (Matson, Sevin, Box, Francis, & Sevin, 1993; Williams, Carnerero, & Pérez-González, 2006; Greer & Ross, 2008) to promote the emergence of spontaneous language.

Children with communication deficits often benefit from an intensive behavioral intervention that offers many opportunities to compensate their mental delay (Delgado & Oblak, 2007). This procedure involves teaching verbal operants, such as mands and tacts.

Skinner (1957) defines spontaneous verbal interactions as pure mands and pure tacts since they are emitted under the control of a non-verbal antecedent stimulus. In children with developmental disabilities, a possible explanation for a deficit in the emission of pure mands and pure tacts can be

found in their learning history (Nuzzolo-Gomez & Greer, 2004). The learning of tacts may have been introduced under the control of an intraverbal (i.e., an antecedent vocal) stimulus and, consequently, examples of spontaneous speech may have never been taught (Partington, Sundberg, Newhouse, & Spengler, 1994). While the mand repertoire is vitally important for an individual's independence (Michael, 1988), tacts are mostly essential for the expansion of verbal repertoires and for the acquisition of more complex social behaviors (Lamarre & Holland, 1985). Research suggests that tact development represents a crucial stage for the further acquisition of a fluent-speaking repertoire. Tacts are necessary for the acquisition of naming (Greer, Stolfi, Chavez-Brown, & Rivera-Valdes, 2005), conversational units (Donley & Greer, 1993), reading (Greer & Speckman, 2009), and as a support for phonemic control considered as a source of derived relationships between naming, reading and writing (Reilly-Lawson, 2008). Several studies have identified effective procedures for the teaching of functional verbal operants, such as a) the use of echoic prompts to increase the spontaneous emission of tacts and mands (Kodak, Clements, & Ninness, 2009); b) the application of the mand-tact teaching protocol to facilitate the rapid acquisition of tacts (Arntzen & Almås, 2002), as well as c) the administration of an Intensive Tact Training (ITT) to increase the spontaneous emission of tacts, mands and intraverbals (Greer, 2002).

More specifically, the ITT protocol involves the presentation of 100 daily learn units in which the student has the opportunity to be exposed to tact visual stimuli (e.g., cards with two-dimensional pictures), in addition to a general educational instruction (Pistoljevic & Greer, 2006). Learn units outline the student-teacher interactions and define the antecedent of behavior, the desired response, and the consequence of its correct or incorrect emission (Greer & McDonough, 1999). For example, in verbal behavior, a learn unit occurs when the teacher asks the student "Do you want a cookie?" (i.e., antecedent); the student answers with a mand (i.e., desired response) and the teacher reinforces the mand by providing access to the cookie as a consequence. More in detail, Pistoljevic and Greer (2006) measured the effects of an ITT on the number of verbal vocal operants emitted in non-instructional settings by three preschoolers with autism who emitted a very low number of spontaneous tacts and mands. In the described study, students were provided with a picture of a stimulus to emit a tact. A correct response was reinforced with generalized reinforcers, such as verbal approvals. In case of an incorrect response or non-response, the

experimenter provided a correction consisting of having the student echo the correct response, for which no reinforcement was provided. The training consisted of delivering five sets of stimuli, each consisting of four stimulus pictures for five categories (i.e., community helpers, transportation, animals, instruments, and food). Following the mastery of a set, that is the emission of 90 correct tacts out of 100 in two sessions or 100 correct tacts out of 100 in one session, the authors measured the number of mands and tacts emitted in non-instructional settings, such as the playground, the cafeteria, or the hallway. The results showed that intensive daily instruction was effective in increasing the number of spontaneous verbal operants emitted. These results were replicated by Delgado and Oblak (2007).

Greer and Du (2010) examined whether the increase of pure mands and tacts in non-instructional settings was due to the ITT or to the increase in the number of learn units presented. The authors compared the number of pure tacts, mands, and intraverbals emitted during a basic teaching session (e.g., numbers, calendar, letters) to those produced during an ITT session. In both conditions, 100 additional learn units were presented to the participants. The results of the study confirmed the existence of a functional relationship between the intensive tact and the increase in the emission of pure tacts, pure mands, and intraverbals.

Schauffler and Greer (2006) evaluated the effects of the ITT on the acquisition of tacts and conversational units in two middle-school students with emotional and behavioral disorders, applied to a classroom setting. The results showed that the daily presentation of 100 learn units could significantly increase the production of appropriate language.

Lydon and colleagues examined the effects of an ITT on the emission of pure tacts, pure mands and conversational units. The authors showed that, although this training did not affect the number of pure mands and conversational units, it increased the emission of pure tacts (Lydon, Healy, Leader, & Keohane, 2009).

Individuals presenting autism spectrum disorder and/or ID experience marked delays and deficits in the reciprocal and spontaneous use of language and in communication.

Compared to neurotypical age-mates who develop language spontaneously and independently without any direct intervention (i.e., without the presentation of environmental antecedents), individuals with autism spectrum disorder and/or ID typically require an intensive intervention to develop the skills necessary to display spontaneous and generative verbal behavior (Ross, Nuzzolo, Stolfi, & Ntarelli, 2006). The

speaker is dependent on the presence of the contrived EOs and the presence of the person (discriminative stimulus) and a majority of teaching trials occur under these particular teaching and reinforcement conditions (Pistoljevic, 2008).

## 2. Aims and hypothesis

The current research partially replicates the study conducted by Lydon and colleagues (2009) although the participants in this study were a 16-year-old boy with autism and a 14-year-old girl with severe ID, diagnosed according to the DSM-5 criteria (APA, 2013) and were thus much older than the two preschool students, as described by the previous authors. Though the ITT has been widely investigated on younger populations, no studies to the best of our knowledge have used this procedure thus far with adolescents characterized by neurodevelopmental disorders. In the current study, tact training was not delivered daily (i.e., intensively), but according to the students' frequency of attendance at the learning center. Therefore, for each participant in this study, only two sets of stimuli (i.e., 200 stimuli divided into 10 categories) were used rather than the five of the intensive tact protocol. Additionally, the stimuli used in this study were from similar but not identical categories to those from Lydon and colleagues (2009).

The purpose of this study was to evaluate the effectiveness of a non-intensive tact training (NITT) on the number of pure tacts, pure mands and conversational units in non-instructional settings emitted by adolescents with autism and ID who have communication deficits.

## 3. Materials and methods

### *3.1. Participants*

Two participants were selected for this study.

Participant A was a 16-year-old male diagnosed with autism and severe ID (DSM-5; APA, 2013). He attended the third year of secondary school in a suburban area of northern Italy, five days a week, for a total of 20 hours per week. He received individualized teaching based on an Applied Behavior Analysis (ABA) for three hours a week in the afternoon. According to the student's level of functioning, a CABAS<sup>®</sup> International Curriculum and Inventory of Repertoires for Children from Pre-School through Kindergarten (C-PIRK<sup>®</sup>) (Greer & McCorkle, 2003) was

administered prior to the onset of the study; assessment results showed that the participant functioned at an emergent speaker level of verbal development (Greer & Ross, 2008). The student was able to communicate his basic needs through the emission of one to two-word requests and could have brief conversational exchanges, but only when engaged by other people and in a structured environment (i.e., emitting yes/no or one to two-word responses to the requests of others).

Participant B was a 14-year-old female diagnosed with severe ID (DSM-5; APA, 2013) who attended the third year of secondary school in a suburban area of northern Italy, six days a week, for a total of 30 hours per week. The student had a teacher or an educator as personal support during her whole school timetable. She received individualized teaching based on an ABA, for four hours a week in the afternoon. The C-PIRK<sup>®</sup> (Greer & McCorkle, 2003) was administered for this student as well, due to her level of functioning; assessment results showed that the participant functioned at an emergent speaker level of verbal development (Greer & Ross, 2008). She was able to repeat single words and presented a very limited number of spontaneous requests.

Participants were selected for this study as they emitted a very low number of spontaneous verbal operants, such as pure mands and tacts and conversational units, in non-instructional settings.

Informed consent was obtained from the parents of the participants for the participation in this study.

### *3.2. Dependent variables*

The dependent variables were the numbers of pure tacts, pure mands and conversational units emitted by each participant during five-minute probe sessions, which were conducted across three non-instructional settings.

A pure tact was defined as a verbal operant under the control of a non-verbal antecedent and reinforced by generalized social reinforcement (Greer, 2002). An example of a tact was when a participant said "That's a cat" upon looking at a picture of a cat and received generalized reinforcement from a listener (i.e., "That's right, it's a cat!").

A pure mand was defined as a verbal operant under the control of a relevant motivating operation (Cooper, Heron, & Heward, 2007) in which the response was reinforced by a characteristic consequence (i.e., the specified item). For example, participants could say "Chocolate" or "Juice"

and the emitted mand was reinforced by the delivery of the item (i.e., the participant receiving chocolate or juice).

A conversational unit was defined as a full exchange in which participants had the opportunity to alternate speaker and listener responses (Donley & Greer, 1993). More in detail, a participant emitted both a speaker response, which was reinforced by another student responding as a listener and, in turn, a listener response to the other person's response (Greer, 2002).

### *3.3. Independent variable*

The independent variable in this study was represented by the NITT, consisting of an increase of 100 tact learn units additionally to other types of learn units delivered daily for each participant. The mastery criterion for the training sessions was defined as 90% correct responses for two consecutive sessions, or 100% for one session. Each time the student mastered the criterion in a category, the stimuli were replaced with a new category to maintain the number of learn units presented at 100. The NITT ended when the participant mastered a total of five categories.

### *3.4. Setting and materials*

The study took place, for both participants, in a learning center in northern Italy. Probe sessions were conducted across three non-instructional settings: activity break, snack time, and free play. The activity break probe was conducted in a room of the center, during the interval of time between the end of one activity and the beginning of another. The snack time probe was recorded in a room where there was a rectangular table with four chairs. Finally, during the free play probe, the students had access to a locker that contained several games (i.e., board games, cards) to play with other students. The materials used for the probes were a clipboard, a data collection sheet, a pencil, and a timer.

The training phase took place in a room of the learning center, where students were engaged in individualized instructional programs and afternoon homework. A rectangular table and two chairs were used. The stimuli (i.e., pictures) were presented through computer slideshows. For each participant, the material used consisted of 200 stimuli divided into 10 categories (see Tab. 1 and Tab. 2). The categories included four target stimuli from one of the following classes: food, animals, transportation, school objects and household objects. Each class, in turn, was repeated twice

so that two categories were created for each class of stimuli (i.e. two categories for Animals, Food etc.). Each of the four target stimuli was repeated within the category, for five different exemplars (see Tab. 1 and Tab. 2).

Table 1 – *Categories and stimuli used during tact training sessions for Participant A*

Categories	Stimuli
1. Animals	Cheetah, Horse, Lizard, Octopus
2. Food	Mortadella, Pear, Peach, Broccoli
3. Household objects	Nightstand, Tub, Broom, Wardrobe
4. School objects	Diary, Eraser, Desk, Map
5. Transportation	Tractor, Jet ski, Tram, Subway
6. Animals	Giraffe, Lion, Penguin, Bull
7. Food	Meat, Fish, Popcorn, Chips
8. Household objects	Knife, Spoon, Mug, Napkin
9. School objects	Glue, Scissors, Smock, Backpack
10. Transportation	Hot air balloon, Roller skates, Ski, Surfboard

Table 2 – *Categories and stimuli used during tact training sessions for Participant B*

Categories	Stimuli
1. Animals	Rabbit, Butterfly, Snake, Mouse
2. Food	Chocolate, Biscuits, Candy, Cake
3. Household objects	Armchair, Toilet, Bed, Sink
4. School objects	Chalk, Markers, Rubber, Correction fluid
5. Transportation	Scooter, Helicopter, Minibus, Raft
6. Animals	Cheetah, Horse, Lizard, Octopus
7. Food	Mortadella, Pear, Peach, Broccoli
8. Household objects	Nightstand, Tub, Broom, Wardrobe
9. School objects	Diary, Eraser, Desk, Map
10. Transportation	Tractor, Jet ski, Tram, Subway



A clipboard, a data collection sheet, and a pencil were used for data collection.

### *3.5. Experimental design*

A delayed multiple probe design across participants (Horner & Baer, 1978) was used to compare the number of verbal operants emitted by each participant in non-instructional settings before and after the acquisition of mastery for each category. Probe sessions were conducted before and after the intervention to measure the effects of the tact training procedure on the emission of pure tacts, pure mands and conversational units for each participant.

### *3.6. Procedure*

The intervention was conducted by psychologists and educationalists with a post-graduate specialization in Applied Behavior Analysis under the supervision of a Board Certified Behavior Analyst<sup>®</sup>. All practitioners had been working for at least nine months as teachers for the organization that owned the learning center, which provided different services for individuals with disabilities or specific educational needs. The teachers served as experimenters and data collectors.

During the implementation of the NITT, the supervisor collected data on treatment integrity through a checklist containing all the steps required in order to provide an accurate instruction.

#### *3.6.1. Pre probes in non-instructional settings*

Prior to the introduction of tact training, 15-minute probe sessions, divided into three five-minute observation intervals, were conducted across three different non-instructional settings: activity break, snack time and free play. The activity break probe consisted of a transition time between the end of an activity (e.g., completing an instructional program) and the beginning of another one.

The experimenter sat at the table with the student and then said "Now let's take a break, what would you like to do?"; the student agreed with the experimenter upon an activity to be done (e.g., watching a video on the computer or listening to a song) and, once it had started, the experimenter proceeded with the observation. During the snack time probe, once the target

student was seated at the table with at least another student, the experimenter told them they could start eating and began the observation procedure. The free play probe was carried out at a specific moment of the daily routine during which all students were engaged in leisure activities. The experimenter invited the target student to decide, together with one or more peers, which game to play among those available. Once the student was engaged in the game activity, the experimenter began the observation procedure. The experimenter used an event recording procedure (Cooper *et al.*, 2007) to register the number of tacts, mands and conversational units emitted by the participants. Tacts were reinforced through verbal approvals and praise. Following the emission of a mand, the requested item was delivered to the participants. Finally, conversational units were reinforced through the alternation between listener and speaker functions.

### *3.6.2. Non-Intensive Tact Training (NITT)*

At the beginning of the training, the experimenter showed to the student the four pictures of the stimuli from one of the categories and provided the participant with an echoic prompt for each stimulus. Independent tact training could begin once the student's response had reached a point-to-point vocal correspondence with the experimenter's model. The experimenter needed to be sure the student paid attention (e.g., by calling him/her by name) before presenting the antecedent, that is, the stimulus picture on the computer. Once the experimenter was sure that the picture to be named was observable, the student was given an opportunity to respond, followed by a consequence (reinforcement or correction). A correct response was defined as the emission of a tact corresponding to the picture shown, within three seconds from the presentation of the stimulus. For example, the experimenter showed the image of a horse to the student and he/she emitted the "horse" tact within three seconds from the picture presentation. Correct responses were reinforced through generalized reinforcers, like praise and attention from the teacher. Other responses, or the emission of a non-response, within three seconds were recorded as incorrect. In this case, the experimenter provided the student with a correction, by repeating the name of the stimulus. The student had to repeat, in turn, the name of the stimulus presented and this repetition was not reinforced; the experimenter then introduced the next learn unit. Twenty learn units were presented daily for five categories for a total of 100 learn units. Mastery criterion was set at 90% correct responses emitted by each participant for two consecutive

sessions (Lydon *et al.*, 2009). After the student had achieved the mastery criterion for a specific category, targets were replaced with another category in order to keep the number of learn units presented to a total of 100. The training ended when the student mastered five categories in total. Instruction was carried out twice a week for Participant A and once a week for Participant B and following the same procedure for both participants.

### *3.7. Post probes in non-instructional settings*

Post probes were conducted sixty minutes after achieving the criterion for one category by applying the same observation procedure as in the pre probes in non-instructional settings.

## **4. Data collection**

During pre-probe sessions, data was collected through five-minute systematic observations in three different non-instructional settings, for a total of 15 minutes. During this time, none of the stimuli targeted during tact training were present in the environment. In the activity break condition, the experimenter activated the timer as soon as the student started the activity he/she had previously chosen (e.g., watching a video or listening to a song), or in the time interval between the end of an activity and the beginning of another one. During the snack time probe, the experimenter turned on the timer as soon as the student sat at the table with at least one other student. During the game probe, the timer was activated as soon as the participant began to play one of the games, chosen among those available, with at least another student. Data was recorded through an event recording procedure (Cooper *et al.*, 2007). Each instance of the emission of vocal behaviors was recorded on a special notation sheet with an X. During probes, the experimenter did not provide any prompts to facilitate the emission of verbal operants by the participants. At the end of the observation sessions, the total number of mands, tacts and conversational units was counted and data was recorded on a graph.

During the tact training phase, responses were recorded on a 20-learn unit (both correct and incorrect) data sheet, marking with a plus (+) if the student correctly named the picture, or with a minus (-) if the student did not respond within three seconds, or responded incorrectly. The collected data was then recorded on a chart.

Post-probe sessions were conducted sixty minutes after the achievement of mastery criterion during the training sessions.

#### *4.1. Treatment Integrity and Inter-Observer Agreement*

Treatment integrity data was collected for 45% of tact training sessions, with a mean value of 98%. Inter-Observer Agreement (IOA) was calculated by having a second observer, who independently and simultaneously recorded the frequency of verbal operants emitted during probe sessions in non-instructional settings. Agreements were divided by agreements plus disagreements then multiplied by 100 (Cooper *et al.*, 2007). The IOA was collected for 30% of the treatment sessions and for 50% of the probe sessions with an agreement of 100%.

### **5. Results**

Figure 1 shows the number of verbal operants, emitted during the 15-minute probe sessions for each participant, in the three non-instructional settings (refer also to Tab. 3 for a summary of the total number of verbal operants emitted by the two participants).

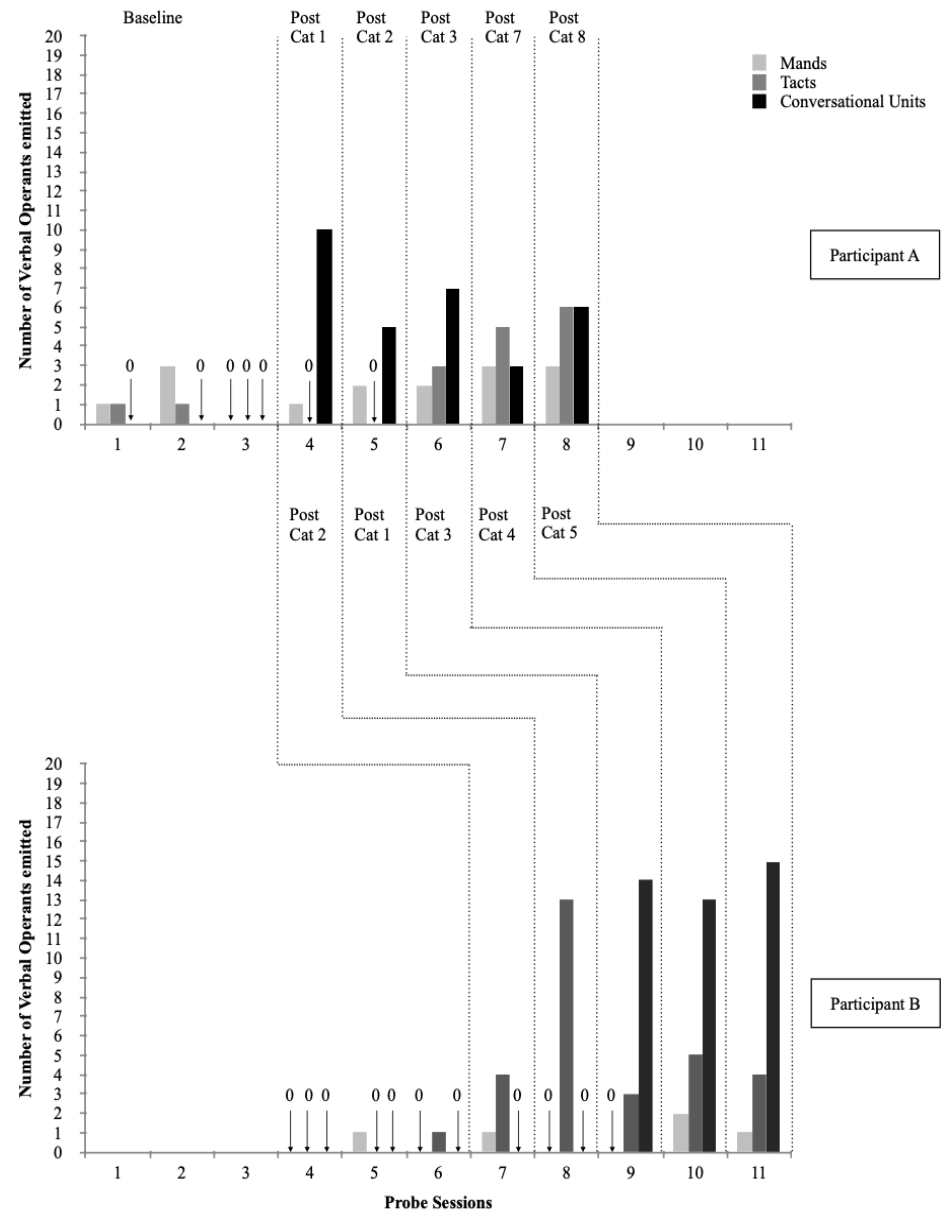
During the three baseline observations, Participant A emitted, respectively, 1, 3, and 0 mands ( $M = 1.3$ ); 1, 1, and 0 tacts ( $M = .6$ ), and 0 conversational units for each session ( $M = 0$ ). After mastering the criterion for Category 1, Participant A emitted a total of 1 mand, 0 tacts, and 10 conversational units. Following the acquisition of Category 2, he emitted 2 mands, 0 tacts, and 5 conversational units. Once the criterion for Category 3 was mastered, the subject emitted 2 mands, 3 tacts, and 7 conversational units. Finally, the individual emitted 3 mands, 5 tacts, and 3 conversational units and 3 mands, 6 tacts, and 6 conversational units, following the acquisition of Category 7 and 8, respectively (see Fig. 1 and Tab. 3).

During the three baseline observations, Participant B emitted, respectively, 0, 1, and 0 mands ( $M = .3$ ); 0, 0, and 1 tact ( $M = .3$ ), and 0 conversational units for each session ( $M = 0$ ). After mastering the criterion for Category 2, Participant B emitted a total of 1 mand, 4 tacts, and 0 conversational units. Once the criterion for Category 1 was reached, she emitted 0 mands, 13 tacts, and 0 conversational units. Following the acquisition of Category 3, the individual emitted 0 mands, 3 tacts, and 14 conversational units. Finally, the subject emitted 2 mands, 5 tacts and 13

conversational units and 1 mand, 4 tacts, and 15 conversational units once the criterion for Category 4 and 5 was met (see Fig. 1 and Tab. 3).

Table 4 summarizes the number of learn units to criterion for each category for each participant.

Figure 1 – *Number of verbal operants emitted by Participants A and B during pre- and post-probes in non-instructional settings*



This data shows that Participant A's learn units to criterion through the five categories were 40, 40, 40, 140 and 140, respectively. Participant A required 400 learn units ( $M = 80$ ) to master the five categories during tact training. Participant B's learn units to criterion were, instead, 40, 60, 120, 140 and 160, respectively. Participant B required a total of 520 learn units ( $M = 104$ ) to master the five categories.

Table 3 – *Summary of the total number of verbal operants emitted*

<i>Participant</i>	<i>Probe sessions</i>	<i>Total number of verbal operants emitted</i>
Participant A	Baseline	2
	Baseline	4
	Baseline	0
	Post Cat 1	11
	Post Cat 2	7
	Post Cat 3	12
	Post Cat 7	11
	Post Cat 8	15
Participant B	Baseline	0
	Baseline	1
	Baseline	1
	Post Cat 2	5
	Post Cat 1	13
	Post Cat 3	17
	Post Cat 4	20
	Post Cat 5	20

Table 4 – *Learn units to criterion for each stimulus set for Participants A and B*

<i>Participant A</i>	<i>Learn units to criterion</i>	<i>Participant B</i>	<i>Learn units to criterion</i>
Cat 1	40	Cat 2	40
Cat 2	40	Cat 1	60
Cat 3	40	Cat 3	120
Cat 7	140	Cat 4	140
Cat 8	140	Cat 5	160

## 6. Discussion

The present study evaluated the effectiveness of a NITT on the emission of pure mands, pure tacts and conversational units in three different non-instructional settings. The students were selected for this study because they emitted a limited number of spontaneous verbal operants. The intervention represents a partial replication of the experiment conducted by Lydon and colleagues (2009). In fact, the stimuli used in this study were similar but not identical to those originally adopted by the aforementioned authors and, for each participant, only two sets (i.e., ten categories) were used, instead of the five provided in the original procedure. Furthermore, participants in this study were older than the participants from Lydon's group study.

According to previous results (Pistoljevic & Greer, 2006; Schaufler & Greer, 2006; Lydon *et al.*, 2009; Greer & Du, 2010), the study shows a functional relationship between the implementation of NITT and the increase in the emission of spontaneous verbal operants, for both participants. More specifically, both participants showed an increase in the number of tacts following the intervention (see Fig. 1). For participant A, there was an increase in the production of pure tacts in non-instructional settings only after the achievement of criterion for Category 3. Participant B began to produce an increased number of tacts following the acquisition of Category 1. These differences in response across participants might be explained by their different levels of verbal development. It would seem that, following the implementation of NITT, both students had learned to emit tacts as a means of achieving generalized reinforcement in the form of social attention from adults (Pistoljevic & Greer, 2006). In fact, the increase in the number of learn units presented was directly proportional to the

increase of reinforcers. This means that the attention of adults could have been progressively conditioned as a reinforcer for the students. This would have led them to emit more tacts in their natural environment. This result is consistent with previous studies (Pistoljevic & Greer, 2006; Delgado & Oblak, 2007; Lydon *et al.*, 2009). Interestingly, tacts emitted by the participants during post-probe sessions were not the same the students had learned during the intervention; students began to name stimuli that were present in their natural environment, thus emitting an overall higher number of verbal behaviors.

In contrast, a NITT in both participants did not produce significant effects on the emission of pure mands in non-instructional settings. It is likely to be hypothesized that the increase in the emission of tacts, along with the generalized reinforcement obtained through the teachers' social approval for the correct tacts, could have reduced the motivational conditions for the emission of mands during post probes. A further explanation could be that the short duration of post-probe sessions (i.e., 15 minutes) represented a limitation for the participants compared to the number of mands they could have emitted. Therefore, data collected in the present study seems to corroborate the results reported by Lydon and colleagues (2009) and the functional independence of verbal operants (Skinner, 1957).

A significant increase in the production of conversational units was revealed in this study, for both participants, following the introduction of a NITT. A possible explanation could be that the students started to produce conversational units because of the reinforcement obtained both as a speaker and listener (Greer & Ross, 2008). These findings are not in line with the original results of the study by Lydon and colleagues (2009), in which there seemed to be no functional relationship between the ITT and the emission of conversational units in non-instructional settings. In order to produce conversational units, individuals need the relevant EOs for conducting conversations: a) the listener's control, b) the reinforcement for listening behavior, and c) the reinforcement for the behavior of speaking. In the study by Lydon and colleagues (2009) students were younger (i.e. 3 and 4 years old) than those in the present study (i.e. 16 and 14 years old). Therefore, it can be assumed that participants, despite their language delay involving a restricted vocabulary, had verbal skills that allowed them to produce conversational units. In fact, the participants emitted verbal exchanges with others either because they were interested or because their behavior was reinforced by the response of others, thus allowing a hypothesis about their



acquisition of the social control that was determined by the reinforcement of listening behavior (Greer & Ross, 2008).

The study presents some methodological limitations that should be underlined. It would have been advantageous for both participants to implement a tact training with a higher frequency and to present a higher number of stimulus sets. In fact, the data reported in the literature suggests that individuals who received more learn units showed a more significant increase in the production of verbal operants (Pistoljevic & Greer, 2006; Delgado & Oblak, 2007; Lydon *et al.*, 2009). A crucial methodological limitation was the use of different target stimuli for the two participants. This choice was motivated by the differences in the participants' level of verbal development. A further limitation was the choice of carrying out the probes after the participants had mastered the criterion in different categories and not for the same stimuli, as in the original study by Lydon and colleagues (2009). This decision, together with the fact of ending the intervention when the participants had mastered five categories, was determined by the fact that the training was not provided daily but based on the students' frequency of attendance at the learning center. Another limitation was undoubtedly the low number of participants in the study, which did not allow to generalize the results obtained. Using a multiple probe design, three baselines would have been recommended to demonstrate a functional relationship.

The expansion of tact and conversational repertoires in 14- and 16-year-old students in a relatively short time seems to suggest that this procedure could have even more significant effects with younger children. It is possible that the same effects reported in this study could be obtained by simply increasing the number of learning opportunities for students, regardless of whether they are intended for tacts or other teaching objectives. Further research is needed to evaluate the positive effects of a NITT on the emission of other verbal operants and to investigate other student populations.

Despite the limitations mentioned above, the results are promising. The NITT described in this study could represent an effective strategy to increase the emission of verbal operants for students with autism or language delays (Hart & Risley, 1995). The present study, together with previous studies (Pistoljevic & Greer, 2006; Delgado & Oblak, 2007; Lydon *et al.*, 2009), suggests that a NITT could increase the students' capability to gain reinforcement by speaking. When this EO becomes relevant, it can allow students to acquire the naming capability.

Future research should be promoted to investigate the effects of increasing the tact repertoire and the expansion of complex verbal skills, in order to support families and schools in promoting the emergence of spontaneous language in children with autism or language delays. This would offer students a greater number of opportunities to participate in verbal exchanges and to feel more integrated in the social life of the community.

## References

- American Psychiatric Association (2013). *Diagnostic and Statistical Manual of Mental Disorders (5<sup>th</sup> ed.)*. Arlington, VA: American Psychiatric Publishing.
- Arntzen, E., & Almås, I. K. (2002). Effects of mand-tact versus tact-only training on the acquisition of tacts. *Journal of Applied Behavior Analysis*, 35 (4), 419-422.
- Cascella, P. W. (2005). Expressive communication strengths of adults with severe to profound intellectual disabilities as reported by group home staff. *Communication Disorders Quarterly*, 26, 156-163.
- Cooper, J. O., Heron, T. E., & Heward, W. L. (2007). *Applied Behavior Analysis*. Upper Saddle River, NJ: Prentice Hall.
- Delgado, J. A. P., & Oblak, M. (2007). The effects of daily intensive tact instruction on the emission of pure mands and tacts in non-instructional settings by three preschool children with developmental delays. *Journal of Early and Intensive Behavior Intervention*, 4 (2), 392-411. <http://dx.doi.org/10.1037/h0100381>.
- Donley, C. R., & Greer, R. D. (1993). Setting events controlling social verbal exchanges between students with developmental delays. *Journal of Behavioral Education*, 3 (4), 387-401.
- Greer, R. D. (2002). *Designing teaching strategies: An applied behavior analysis systems approach*. San Diego, CA: Academic Press.

Greer, R. D., & Du, L. (2010). Generic instruction versus intensive tact instruction and the emission of spontaneous speech. *The Journal of Speech and Language Pathology – Applied Behavior Analysis*, 5 (1), 1-19. <http://dx.doi.org/10.1037/h0100261>.

Greer, R. D., & McCorkle, N. P. (2003). *CABAS® International curriculum and inventory of repertoires for children from preschool to kindergarten*. Yonkers, NY, CABAS® and the Fred S. Keller School.

Greer, R. D., & McDonough, S. H. (1999). Is the learn unit a fundamental measure of pedagogy? *The Behavior Analyst*, 22 (1), 5-16.

Greer, R. D., & Ross, D. E. (2008). *Verbal Behavior Analysis: Inducing and Expanding New Verbal Capabilities in Children with Language Delays*. New York, NY: Allyn and Bacon.

Greer, R. D., & Speckman, J. (2009). The integration of speaker and listener responses: A theory of verbal development. *The Psychological Record*, 59 (3), 449-488.

Greer, R. D., Stolfi, L., Chavez-Brown, M., & Rivera-Valdes, C. (2005). The emergence of the listener to speaker component of naming in children as a function of multiple exemplar instruction. *The Analysis of Verbal Behavior*, 21 (1), 123-134.

Hart, B., & Risley, T. R. (1995). *Meaningful differences in the everyday experience of young American children*. Baltimore, MD: Paul H. Brookes Publishing Company.

Horner, R. D., & Baer, D. M. (1978). Multiple-probe technique: A variation of the multiple baseline. *Journal of Applied Behavior Analysis*, 11 (1), 189-196.

Ingenmey, R., & Van Houten, R. (1991). Using time delay to promote spontaneous speech in an autistic child. *Journal of Applied Behavior Analysis*, 24 (3), 591-596.

- Kodak, T., Clements, A., & Ninness, C. (2009). Acquisition of mands and tacts with concurrent echoic training. *Journal of Applied Behavior Analysis*, 42 (4), 839-843.
- Kurtz, P. F., Boetler, E. W., Jarmolowicz, D. P., Chin, M. D., & Hagopian, L. P. (2011). An analysis of functional communication training as an empirically supported treatment for problem behavior displayed by individuals with intellectual disabilities. *Research in Developmental Disabilities*, 32, 2935-2942.
- Lamarre, J., & Holland, J. G. (1985). The functional independence of mands and tacts. *Journal of the Experimental Analysis of Behavior*, 43 (1), 5-19.
- Lord, C., & Paul, R. (1997). Language and communication in autism. In D. Cohen & F. Volkmar (Eds.), *Handbook of Autism and pervasive developmental disorders* (pp. 195-225). New York: Wiley.
- Lydon, H., Healy, O., Leader, G., & Keohane, D. D. (2009). The effects of intensive tact instruction on three verbal operants in non-instructional settings for two children with autism. *The Journal of Speech and Language Pathology – Applied Behavior Analysis*, 3 (2-3), 173-184. <http://dx.doi.org/10.1037/h0100242>.
- Matson, J. L., Horovitz, M., Kozlowski, A. M., Sipes, M., Worley, J. A., & Shoemaker, M. E. (2011). Person characteristics of individuals in functional assessment research. *Research in Developmental Disabilities*, 32, 621-624.
- Matson, J. L., Sevin, J. A., Box, M. L., Francis, K. L., & Sevin, B. M. (1993). An evaluation of two methods for increasing self-initiated verbalizations in autistic children. *Journal of Applied Behavior Analysis*, 26 (3), 389-398.
- Matson, J. L., Sipes, M., Horovitz, M., Worley, J. A., Shoemaker, M. E., & Kozlowski, A. M. (2011). Behaviors and corresponding functions addressed via functional assessment. *Research in Developmental Disabilities*, 32, 625-629.

Matson, J. L., Terlonge, C., Gonzalez, M. L., & Rivet, T. (2006). An evaluation of social and adaptive skills in adults with bipolar disorder and severe/profound intellectual disability. *Research in Developmental Disabilities*, 27, 681-687.

Michael, J. (1988). Establishing operations and the mand. *The Analysis of verbal behavior*, 6 (1), 3-9.

Nuzzolo-Gomez, R., & Greer, R. D. (2004). Emergence of untaught mands or tacts of novel adjective-object pairs as a function of instructional history. *The Analysis of Verbal Behavior*, 20 (1), 63-76.

Partington, J. W., Sundberg, M. L., Newhouse, L., & Spengler, S. M. (1994). Overcoming an autistic child's failure to acquire a tact repertoire. *Journal of Applied Behavior Analysis*, 27 (4), 733-734.

Pistoljevic, N. (2008). *The effects of multiple exemplar and intensive tact instruction on the acquisition of naming in preschoolers diagnosed with autism and other language delays* (Order No. 3317598). Available from British Nursing Database; Health Research Premium Collection. (304622605). Retrieved from <https://search.proquest.com/dissertations-theses/effects-multiple-exemplar-intensive-tact/docview/304622605/se-2?accountid=31745>.

Pistoljevic, N., Cahill, C., & Casarini, F. (2010). Effects of a speaker immersion procedure on the production of verbal operants. *The Journal of Speech and Language Pathology – Applied Behavior Analysis*, 5 (2), 191-206. <http://dx.doi.org/10.1037/h0100273>.

Pistoljevic, N., & Greer, R. D. (2006). The effects of daily intensive tact instruction on preschool students' emission of pure tacts and mands in non-instructional setting. *Journal of Early and Intensive Behavior Intervention*, 3 (1), 103-120. <http://dx.doi.org/10.1037/h0100325>.

Reilly Lawson, T. L. (2008). *Phonemic control as the source of derived relations between naming and reading and writing* (Doctoral dissertation, ProQuest Information & Learning).

Ross, D. E., Nuzzolo, R., Stolfi, L., & Natarelli, S. (2006). Effects of speaker immersion on independent speaker behavior of preschool children with verbal delays. *Journal of Early and Intensive Behavior Intervention*, 3 (1), 135-150. <http://dx.doi.org/10.1037/h0100327>.

Schauffler, G., & Greer, R. D. (2006). The effects of intensive tact instruction on audience- accurate tacts and conversational units. *Journal of Early and Intensive Behavior Intervention*, 3 (1), 121-134. <http://dx.doi.org/10.1037/h0100326>.

Skinner, B. F. (1957). *Verbal Behavior*. Acton, MA; Copley Publishing Group. *Journal of Behavioral Education*, 12, 185-206.

van der Schuit, M., Segers, E., van Balkom, H., & Verhoeven, L. (2011). How cognitive factors affect language development in children with intellectual disabilities. *Research in Developmental Disabilities*, 32, 1884-1894.

Williams, G., Carnerero, J. J., & Pérez-González, L. A. (2006). Generalization of tacting actions in children with autism. *Journal of Applied Behavior Analysis*, 39 (2), 233-237.

# Early numerical skills in individuals with Down Syndrome

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## Abstract

*The aim of the present work was to explore early mathematical competences in individuals with Down Syndrome (DS). Intellectual Disability has been identified as one of the most important features in this population. The behavioral phenotype of individuals with DS is characterized by deficits in cognitive functions and learning abilities. A numerical battery was administered to a group of 11 individuals with DS and 11 Typically Developed (TD) children matched for mental age, as assessed with the Logical Operations Test. The findings revealed that early numerical skills of individuals with DS were well aligned to mental age: the two groups presented similar competences in counting, in mental calculation and cardinality. Moreover, individuals with DS read better Arabic numbers than the control group. Data concerning the discrimination of numbers in individuals with DS was also taken into*

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*consideration. Certainly, more research on children with DS is needed although the findings here presented have implications to understand the development of numerical skills in DS and to improve the neuropsychological assessment of children with this genetic condition.*

**Keywords:** Intellectual Disabilities; Down Syndrome; Logical thinking; Early numerical skills; Mental age.



## 1. Introduction

Down Syndrome (DS) is a genetic condition caused by an extra copy of chromosome 21, featuring peculiar somatic traits, a distinctive neurofunctional architecture (Pennington, Moon, Edgin, Stedron, & Nadel, 2003; Edgin, Tooley, Demara, Nyhuis, Anand, & Spanò, 2015) and atypical developmental patterns. DS is the most frequent form of intellectual disability among genetically determined forms (Kittler, Krinsky-McHale, & Devenny, 2008; Parker, Mai, Canfield, Rickard, Wang, Meyer *et al.*, 2010; Daunhauer, Fidler, Hahn, Will, Lee, & Hepburn, 2014). According to the World Health Organization (WHO), DS affects 1 out of every 1.000 - 1.100 living children in the world (WHO, 2015), encompassing different ethnicities and genders. Apart from a poor intellectual functioning, individuals with DS also show compromised cognitive functions as well as impairments in terms of adaptive behavior and learning abilities (Jarrold, Baddeley, & Hewes, 1999; Chapman & Hesketh, 2000; Pennington *et al.*, 2003; Vicari, Marotta, & Carlesimo, 2004; Rowe, Lavender, & Turk, 2006; Iacono, Torr, & Wong, 2010; Lanfranchi, Jerman, Dal Pont, Alberti, & Vianello, 2010; Lee, Fidler, Blakely-Smith, Daunhauer, Robinson, & Hepburn, 2011; APA, 2013). Although the literature describes the typical features of individuals with DS, the interaction between epigenetic, environmental and chromosomic variables triggers a series of individual differences, on a genetic, neurofunctional and cognitive level, which can lead to very different neuropsychological profiles (Vianello, 2006; Karmiloff-Smith, Al-Janabi, D'Souza, Groet, Massand, Mok *et al.*, 2016). In spite of the fact that intellectual disability is currently and usually identified as one of the most important characteristics of individuals with DS (Vicari, Bellucci, & Carlesimo, 2005; Contestabile, Benfenati, & Gasparini, 2010), only a handful of studies have globally analyzed the degree of intelligence in this population of individuals. More in general, the peak of intellectual functioning of individuals with DS is comparable to that of a 7-year-old child: a mental age (MA) beyond 7 years old has been demonstrated only in very few studies, even in adult subjects (Dykens, Hodapp, & Finucane, 2000). Researches revealed that Intellectual Disability might vary from mild to severe in this population (e.g.: Määttä, Kaski, Taanila, Keinänen-Kiukaanniemi, & Iivanainen, 2006; Vianello, 2006; Contestabile *et al.*, 2010; Lott & Dierssen, 2010; Orsini, Pezzuti, & Picone, 2012; Grieco, Pulsifer, Selingsohn, Skotko, & Schwart, 2015). These conflicting results are also due to discussed methodological issues, concerning the

neuropsychological assessment of individuals with intellectual disabilities (Vicari, 2004; Vianello, 2006; Edgin, Mason, Allman, Capone, DeLeon, Maslen *et al.*, 2010; Patterson, Rapsey, & Glue, 2013; Pulina, Vianello, & Lanfranchi, 2019). There are various national and international scientific studies concerning the intellectual functioning of individuals with DS; many authors administered psychometric instruments based on intelligence as constructs of a factor analysis, while few studies actually focused their attention on the quality of thinking of children with DS.

By employing tools based on the Piagetian theory, Moniga (2007) and Pizzoli and colleagues (Pizzoli, Lami, & Stella, 2001) analyzed the sensorimotor competences of children with DS during the first three years of their lives. Vianello and co-workers (Vianello, Lanfranchi, & Moalli, 2006) analyzed the logical operations of 189 children, aged between 8 and 17, using the Logical Operations Test (Vianello & Marin, 1997). The authors found that the mental ages of these individuals varied from 4 years and 10 months to 5 years and 7 months and they were able to successfully solve logical operations at a double chronological age (CA) compared to their TD counterparts (Vianello *et al.*, 2006). It has been demonstrated that logical operations are associated, even in non-typically developing children, to subsequent mathematical skills (Van de Rijt & Van Luit, 1998). Some studies reported that subjects with DS show difficulties in logical operations and in numerical cognition, exhibiting a delay of about two years in this domain compared to other learning abilities (Gelman & Cohen, 1988; Porter, 1999; Nye, Fluck, & Buckely, 2001; Buckley, 2007). Math performances of individuals with DS would seem to be inferior to their reading and writing performances and they generally do not meet 2<sup>nd</sup>-grade school requirements (Rynders, 1999). Through the CA-MT test (Cornoldi, Lucangeli, & Bellina, 2002), Sestili and collaborators (Sestili, Moalli, & Vianello, 2006) observed lower numerical skills in individuals with DS, as opposed to those of TD children at the beginning of the primary school. Scientific studies on the numerical cognition in DS are limited and, therefore, require further research, given the impact that mathematical skills have on each individual's daily life activities and personal autonomy. Subjects with DS thus present severe difficulties in mathematical learning, vulnerabilities that are not directly ascribable to the child's general functioning (Marotta, Viezzoli, & Vicari, 2006). Authors observed a significant gap in mathematical skills between children with DS and TD children of the same chronological age (Brigstocke, Hulme, & Nye, 2008), in particular in numerical knowledge, in counting and calculation (Porter,

1999; Nye *et al.*, 2001). The origin of these difficulties is a debated topic. Some researchers support the *developmental hypothesis* (Zigler, 1969), suggesting that the mathematical difficulties of individuals with DS stem from their low general cognitive level (e.g., Caycho, Gunn, & Siegal, 1991). Others support the *difference hypothesis* (e.g., Gelman & Cohen, 1988; Nye *et al.*, 2001) by showing poorer performance of individuals with DS in comparison to TD children of the same mental age. Sella and co-workers (Sella, Lanfranchi, & Zorzi, 2013) investigated the numerical estimation in children with DS. They compared a group of 21 participants with DS ( $M_{\text{age}}$  14 years and 2 months) to two control groups of TD children, matched for verbal MA (Peabody Picture Vocabulary Scale-Revised – PPVT-R; Dunn & Dunn, 1997;  $M_{\text{MA}} = 5;4$ ,  $SD_{\text{MA}} = 0;6$  months) and CA. The children with DS showed a specific deficit (even after accounting for MA), which emerged when they had to distinguish between small numbers, up to 4 (and particularly when comparing 2 and 3, or 3 and 4). The discrimination between larger number sets was similar between the DS group and the control group of similar MA. Considering the two core systems responsible for numerical skills, the *approximate number system* (ANS) and the *object tracking system* (OTS), the authors suggested that the OTS was compromised in subjects with DS, while the ANS, and thus their ability to compare large number sets, seemed to be in line with the MA. Other studies supported these findings: using the *preferential looking* paradigm, Paterson and colleagues showed a deficit in the discrimination of two or three objects in a sample of 30-month mentally-aged children (Paterson, Girelli, Butterworth, & Karmiloff-Smith, 2006). The OTS system, but not the ANS, evaluated with the test of Molin and co-workers (Molin, Poli, & Lucangeli, 2007), correlated with numerical cognition in individuals with DS and TD-MA children (Sella *et al.*, 2013). Following these authors, other studies were also in support of an non-compromised ANS system in children with DS: children with DS from 5 to 8 years of age could discriminate between large number sets and they were more competent when the ratio (the difference) between the two sets was significantly large, as in TD subjects (Izard, Sann, Spelke, & Streri, 2009). They also showed some difficulties in tasks concerning dots discrimination with a 2:3 dot ratio (Camos, 2009; Abreu-Mendoza & Arias-Trejo, 2015). The debate of a similarity in the ANS system between individuals with DS and subjects with TD-MA or TD-CA remains open. Lanfranchi and colleagues also administered numerical estimation tasks (number-to-position with interval: 1-10 and 1-100) to a group of adolescents with DS (Lanfranchi, Berteletti, Torrisi, Vianello, &

Zorzi, 2015). The performances on these tasks were similar between the group of DS and TD-MA children.

Furthermore, the development of preverbal numerical skills to more complex ones, related to linguistical and socio-cultural aspects (Geary, 1994, 2000), could be difficult for children with DS too. In 1986, Gelman and Gallistel registered lower performances in counting and cardinality tasks in individuals with DS compared to pre-schooler subjects having the same MA. Caycho and colleagues showed competences in cardinality tasks in this population also with sufficient linguistical skills (Caycho *et al.*, 1991). Through the “*give-a-number*” task, Nye and collaborators (2001) found that only a third of the participants with DS was able to use the cardinality’s principle, while other authors argued that the latter competence is in line with the MA (Bashash, Outhred, & Bochner, 2003; Sella *et al.*, 2013). Some authors investigated the other two counting principles in the population with DS: they were able to use the one-on-one correspondence and the stable order principles (Caycho *et al.*, 1991; Bashash *et al.*, 2003). Sella and collaborators described counting as less fluent in individuals with DS compared to TD subjects (Sella *et al.*, 2013). In this respect, the analysis of Abdelahmeed (2007) on DS’ counting competences showed severe difficulties in this domain but also emphasized the important role of interventions. Gelman and Cohen (1988) recognized counting issues in the population with DS and, according to several authors, this is restricted to procedural counting. It would seem that subjects with DS are not aware of their errors in the counting sequences: they tend to forget number-words and/or omit words or objects during enumeration (Porter, 1999). In 1974, Cornwell noticed children with DS were not able to complete their tasks when they interrupted the sequences, or they needed to restart counting from the very beginning. This was probably due to rote learning. Hanrahan and Newman (1996) also claimed that children with DS master counting and recognition of numbers from 1 to 10 through mere repetition. Finally, Nye and colleagues found that children with DS showed shorter counting sequences and could enumerate fewer objects than TD-MA children (Nye *et al.*, 2001). Some authors found severe difficulties in the calculation process in children with DS as well (Marotta *et al.*, 2006). Hence, there is substantial evidence on neuropsychological deficits in individuals with DS; however, findings in some domains, such as numerical cognition, remain unclear.

## 2. Aims and hypothesis

The main purpose of the present study was to analyze the early numerical competences in individuals with DS and to compare them to the MA-matched TD group, to identify strengths and weaknesses in counting, calculation and lexical and semantic processes concerning the MA, as evaluated with the Logical Operations Test – OL18 (Vianello & Marin, 1997). We hypothesized that subjects belonging to different populations but of equal mental age show the same skills in all of the numerical tasks under examination. The question addressed was whether the numerical competences displayed by individuals with DS were tied to the overall cognitive level (indexed by mental age) or whether these individuals showed specific deficits even concerning the MA.

## 3. Methods

### 3.1. Sample

The participants recruited in the study were eleven subjects with DS (6 males, 5 females) with a mean CA of 10 years and 4 months ( $SD = 4.3$  years; age range = 5 years and 7 months to 17 years and 11 months) and with a mean MA of 4 years and 6 months ( $SD = 4$  months; age range: 48 to 59 months; see Tab. 1 for more details). All our participants were Italian native speakers and were still attending school and all of them were included in regular schools. The selection criteria were the following: a MA between 48 and 59 months, a CA between 5 and 17.11 years and the absence of hearing problems. Subjects with DS and with a MA lower than 48 months or higher than 59 months were thus excluded from the study sample ( $n = 8$ ). Concerning the overall socioeconomic status (SES), as estimated from parents' highest level of educational attainment, we found that 7 (64%) mothers and 8 (73%) fathers had a middle/low level of education (they had completed high school or at least a form of basic education), while 4 (36%) mothers and 3 (27%) fathers had a high educational level (they had completed a Bachelor/Master's degree). The mean age of the mothers at the time of their offspring's birth was 40 ( $SD = 5$ , age range = 33-50). The mean age of the fathers at the time of their offspring's birth was 41 ( $SD = 4$ , age range = 36-50; refer to Tab. 1 for more details).

A control group of typically developed children was recruited and they were matched for mental age to the DS group. The TD-MA group was

comprised of eleven TD children (6 males, 5 females) with a mean CA of 4 years and 5 months ( $SD = 3$  months; age range = 4 years and 2 months to 4 years and 9 months) and with a mean MA of 4 years and 6 months ( $SD = 4$  months; age range = 48 to 59 months; see Tab. 1 for more details). All participants were Italian native speakers, attending the second year of the Republic of San Marino's kindergartens (RSM). The selection criteria were the following: the absence of certified disabilities, a Fluid Reasoning Index (FRI), as measured by the Wechsler Preschool and Primary Scale of Intelligence – Fourth Edition (WPPSI-IV), scoring higher than 70 (Wechsler, 2012; Saggino, Stella, & Vio, 2019) and a CA between 4 and 4.11 years. Concerning the overall socioeconomic status (SES), as estimated from parents' highest level of educational attainment, we found that 5 (45.5%) mothers and 8 (73%) fathers had a middle/low level of education (they had completed high school or at least a form of basic education), while 6 (54.5%) mothers and 3 (27%) fathers had a high educational level (they had completed a Bachelor/Master's degree). The mean age of the mothers at the time of their offspring's birth was 31 ( $SD = 4$ , age range = 24-36). The mean age of the fathers at the time of their offspring's birth was 34 ( $SD = 5$ , age range = 25-39; see Tab. 1 for more details).

These two groups differed significantly on CA [ $t(20) = -5.854$ ,  $p < .001$ ] and on parents' CA [mother,  $t(19) = -4.448$ ,  $p < .001$ ; father,  $t(18) = -3.368$ ,  $p = .03$ ]. The two groups did not differ significantly on the parents' educational level [father,  $\chi^2(1, N = 20) = .808$ ,  $p = .59$ ; mother  $\chi^2(1, N = 22) = .733$ ,  $p = .39$ ].

Table 1 – *Socio-demographic characteristics of the TD-MA and DS groups*

			TD-MA	DS
			<i>n</i> = 11	<i>n</i> = 11
Infant's gender	Male	<i>N</i> (%)	6 (60)	6 (60)
	Female		5 (40)	5 (40)
Infant's age (years)		<i>M</i> ( <i>SD</i> ); <i>range</i>	4.5 (0.3); 4.2-4.9	10.4 (4.3); 5.7-17.11
Maternal age (years)		<i>M</i> ( <i>SD</i> ); <i>range</i>	31 (4); 24-36	40 (5); 33-50
Maternal education	Low/medium	<i>N</i> (%)	5 (45.5)	7 (64)
	High		6 (54.5)	4 (36)
Paternal age (years)		<i>M</i> ( <i>SD</i> ); <i>range</i>	34 (5); 25-39	41 (4); 36-50
Paternal education	Low/medium	<i>N</i> (%)	8 (73)	8 (73)
	High		3 (27)	1 (9)

A pairing criterion by gender and mental age was chosen for the purpose of this study. Groups were individually matched on gender and on MA, as assessed by the OL18 (Vianello & Marin, 1997). The test, based on Piaget's cognitive theory, is comprised of 18 tasks that assess areas of logical thinking, such as seriation, numeration, and classification. Each group's MA mean was 4 years and 6 months ( $SD = 4$  months; range: 48 to 59 months). There weren't statistically significant differences between the two groups [seriation:  $U = 55.50$ ,  $z = -.36$ ,  $p = .75$ ,  $r = -.08$ ; numeration:  $U = 71$ ,  $z = .73$ ,  $p = .52$ ,  $r = .15$ ; classification:  $U = 55$ ,  $z = -.61$ ,  $p = .75$ ,  $r = -.13$ ].

Moreover, in order to also have a measure of fluid intelligence, the recent WPPSI-IV (Wechsler, 2012; Saggino *et al.*, 2019) was administered to both the DS and TD-MA groups. The raw scores of the DS and TD-MA groups to both tests are presented in Table 2. There weren't statistically significant differences between the two groups (see Tab. 2 for details).

The participants to this survey were children and this study was conducted in compliance with the WMA Declaration of Helsinki's latest revision. Parental consent forms describing the project's objectives, the research procedures followed as well as information concerning data retrieval were presented to the parents and obtained before testing.

Table 2 – *Fluid intelligence (WPPSI-IV FRI): comparison between the TD-MA and DS groups*

WPPSI-IV variables	TD-MA		DS		Mann-Whitney			
	$M (SD)$	$Mdn$	$M (SD)$	$Mdn$	$U$	$z$	$p$	$r$
Matrix Reasoning	10.91 (3.08)	11	9.54 (4.32)	10	50	-.69	.52	-.15
Picture Concepts	8.27 (3.52)	8	6.10 (4.53)	7	48	-.82	.44	-.17
FRI	19.18 (4.58)	18	16.45 (7.95)	17	44.50	-1.06	.30	-.23

### 3.2. Instruments

The tools described in the following section were appropriate for our sample of individuals with DS because the average mental age was 5 years old.



### *3.2.1. The Numerical Intelligence Scale for children aged between 4 and 6 – BIN 4-6 (Molin et al., 2007)*

It provides a measurement of numerical and counting skills. It provides specific indexes for each area investigated, and specifically the lexical, counting and semantic processes.

Lexical tasks assess the knowledge of the names of numbers and of the stable sequence of numbers. In Arabic numeral reading, the child must say the name of the number presented, which is shown in Arabic numerals. In Arabic number recognition, the child must recognize and choose the Arabic-coded number (one among three), which has been pronounced by the examiner. In the correspondence between the Arabic number and quantity, the child must specify the exact quantity of dots corresponding to the Arabic number presented. A point is attributed to each correct item. In each task, the minimum score is 0 and the maximum score is 9.

Counting tasks assess the ability to count (i.e. counting 1-20: the child must count out loud from 1 to 20 using the correct sequence). The minimum score is 0 and the maximum score is 20. Errors are recorded and then subtracted from the total scoring. The total time (in seconds) is also recorded.

Semantic tasks assess the ability to understand the link between numbers and their quantity representations. In the discrimination of dots, the child must choose which set contains more dots from a two-piece set. There are 10 difficulty-scalable items, which include comparisons between dot sets spanning different sizes (congruent and incongruent situation) and same size set comparisons (neutral situation). A point is attributed to each correct item. In each task, the minimum score is 0 and the maximum score is 10. The total time (in seconds) is also recorded.

### *3.2.2. Counting (1-10)*

The child must count out loud from 1 to 10 in the correct sequence. A point is attributed to each correct number. The minimum score is 0 and the maximum score is 10. The total time (in seconds) is also recorded.

### *3.2.3. Backward counting (5-1)*

The child must count out loud from 5 to 1 in the correct sequence. The classic “5, 4, ...” example is given to get the child started. A point is attributed to each correct number located in the correct backward sequence. In the case of the repetition of the suggested sequence (5, 4), the attributed score is equals to zero.



#### 3.2.4. *Number Sense: Prerequisites – SNUP* (Tobia, Bonifacci, & Marzocchi, 2017)

It assesses early numeracy skills and can be administered to children from 4 to 6.9 years old. The *Quantity Comparison Test* comprises 24 items, divided into two separate subtests, evaluating simple stimuli quantity comparison and complex stimuli quantity comparison, respectively. In this test, children are asked to quickly indicate the box with a greater number of elements, choosing between two illustrated baskets of fruit. The number of elements varies from 3 to 20, and the differences between sets are from 1 to 6 units. A point is attributed to each correct item. The minimum score is 0 and the maximum score is 24.

#### 3.2.5. *“Give-a-number” task* (ad hoc, based on Wynn’s model, 1990, 1992)

The child has 10 tiny pale wooden cubes, measuring  $2 \times 2 \times 2$  cm each, and a small transparent box, measuring  $5 \times 15,5 \times 9$  cm, in front of him. He/she is asked to put an ever-changing number of cubes in the small box (e.g.: “Put two cubes in the box”) and to say “done/finished” when his/her assignment is completed (meanwhile the operator covers his/her eyes). An example item is provided. Quantities are exposed in the following order: 2-, 6-, 9-, 4-, 3-, 7-, 1-, 5-, and 8-. A point is attributed to each correct item. The minimum score is 0 and the maximum score is 9.

#### 3.2.6. *Mental calculations < 5 task* (ad hoc)

The child must answer 4 simple additions orally proposed by the examiner. The experimental items include the calculations  $1+1=2$ ;  $3+1=4$ ;  $1+2=3$  and  $2+2=4$ . Children are allowed to answer verbally or to show the result with their fingers. A point is attributed to each correct item. The minimum score is 0 and the maximum score is 4.

#### 3.2.7. *WPPSI – Wechsler Preschool and Primary Scale of Intelligence – Fourth Edition* (Wechsler, 2012; Saggino et al., 2019)

The WPPSI-IV is an innovative measure of cognitive development for preschoolers and young children. The WPPSI-IV model reflects contemporary structural theories, such as the CHC (Cattell-Horn-Carroll) theory. *Matrix Reasoning* (MR) and *Picture Concepts* (PC) are administered to the participants. Every single subtest provides a raw score (MR range: 0-26; PC range: 0-27) and, given the sum of the two subtests’ weighed scores, it is possible to obtain a composite score, the FRI with a mean value of 100

( $SD = 15$ ). The FRI may be conceptualized as measuring fluid and inductive reasoning, broad visual intelligence, simultaneous processing, conceptual thinking, and classification ability. We referred to the manual for the method of administration and scoring.

### 3.2.8. Logical Operations Test – OL 18 (Vianello & Marin, 1997)

It assesses the development of logical thinking. The test, based on the Piagetian cognitive theory, is comprised of 18 tasks that assess areas of logical thinking, such as seriation, numeration, and classification. The test is standardized for the Italian population aged between 4 and 9. A score of 1 is given for each task performed correctly. The grand scoring total is hence 18. This raw score can then be turned into a mental age score. This test seems particularly appropriate to match children with DS with TD children on a central intelligence component while limiting the influence of culture and linguistic ability (for a review, see Vianello & Marin, 1997).

## 4. Procedure

Participants of the DS group were contacted through several associations for people with DS in Emilia Romagna (Italy) and the Republic of San Marino (RSM). Participants of the TD-MA group were contacted in some kindergartens of the Republic of San Marino (RSM) during the 2018-2019 term. All participants were exposed to areas of logical thinking, fluid non-verbal reasoning, and early numerical competences. All the tasks were administered individually in two sessions, separated by approximately 1 week, with each session lasting approximately 30 minutes. Each session was performed in a well-lit and quiet room.

## 5. Data analysis

All statistical analyses were carried out using SPSS 22.0 for Windows with  $\alpha = .05$ . The experimental design involved two groups, a TD-MA and a DS group, at a data collection moment ( $t_0$ ). Prior to conducting analyses, data was checked for violation of assumptions using the Kolmogorov-Smirnov test. Because distributions for some of the communicative behaviors were non-normal, nonparametric Mann-Whitney tests were conducted to assess potential differences in early numerical competences between the DS and TD-MA groups. Effect sizes ( $r$ ) for Mann-Whitney U tests were calculated using the formula  $r = \frac{z}{\sqrt{N}}$ , in which  $z$  is  $z$ -

the score that SPSS produces and  $N$  is the size of the study on which  $z$  is based. The standard values of  $r$  for medium and large effect sizes are .3 and .5, respectively (Field, 2018).

Raw scores for the DS and TD-MA children in early numerical competence tests are shown in Table 3.

### *5.1. Lexical process*

Individuals with DS showed a significantly higher score in the Arabic numbers reading task: they were more accurate than the children of the TD-MA group. The other numerical lexical task did not differ in the two groups, although individuals with DS showed a higher performance in the recognition of Arabic numbers. The participants could also associate about 5 out of 9 Arabic numbers to their numerosities: the total scores did not significantly differ in the two groups (see Mann-Whitney test in Tab. 3).

### *5.2. Mental calculations*

The performances of a simple additions task did not differ in the two groups, although the  $p$ -value was near significance ( $p = .06$ ; see Mann-Whitney test in Tab. 3). The percentage of children of the TD-MA group (36%) who completed at least one mental additive operation was lower than the DS group percentage (64%) but differences between the two groups were not statistically significant [ $\chi^2(1, N = 22) = 1.64, p = .20$ ].

### *5.3. Counting*

The performances of counting did not differ in the two groups, although the children of the TD-MA group were more accurate than the individuals of the DS group. Moreover, the TD-MA group was significantly quicker in counting from 1 to 10 compared to the DS group ( $p = .02$ ; see Mann-Whitney test in Tab. 3). In order to control this data, the same analysis was conducted a second time only selecting the participants who were able to correctly count from 1 to 10 ( $n = 14$ ) and from 1 to 20 ( $n = 10$ ): the TD-MA group confirmed to be quicker than the DS group also in this particular case but the difference was not statistically significant. The scores for the backward counting test did not significantly differ in the two groups (refer to Tab. 3 for more details).

#### 5.4. Semantic processes

Although individuals with DS showed a lower accuracy and time scores in the BIN 4-6 task (Molin *et al.*, 2007), the performance for the dot comparison task did not significantly differ in the two groups. Instead, there were significant differences in the quantity test (SNUP; Tobia *et al.*, 2017), particularly on a basic quantity comparison subtest (same-sized element sets). Individuals with DS showed, in fact, a significantly lower score in this task: they were less accurate than the children of the TD-MA group ( $p = .04$ ; see Mann-Whitney test in Tab. 3). Finally, the performances in the *give-a-number* task showed that participants could identify the cardinality of about 5 out of 9 numbers (mean): the total scores did not significantly differ in the two groups (see Mann-Whitney test in Tab. 3).

Table 3 – *Early numerical skills: comparison between the TD-MA and DS groups*

Variables	TD-MA		DS		Mann-Whitney			
	<i>M (SD)</i>	<i>Mdn</i>	<i>M (SD)</i>	<i>Mdn</i>	<i>U</i>	<i>z</i>	<i>p</i>	<i>r</i>
<b>Numbers reading</b>	5.27 (2.90)	6	7.91 (1.92)	9	96.50	2.45	<b>.02</b>	.52
Numbers recognition	7.18 (2.14)	8	8.64 (.67)	9	83.50	1.70	.13	.36
Numbers-quantities correspondence	5.27 (1.79)	5	5.82 (1.40)	5	69.50	.61	.56	.13
Mental additions < 5	.45 (.69)	0	2 (1.79)	2	89.50	2.05	.06	.44
Counting 1-20 (accuracy)	17.91 (2.16)	19	14.64 (5.87)	16	43.00	-1.18	.27	-.25
Counting 1-20 (seconds)	14.82 (6.26)	13	23 (19.21)	17	76.50	1.05	.30	.22
Counting 1-20 (seconds) ( $n = 10$ )	15.83 (7.47)	14	28.5 (29.08)	15	14.5	.53	.61	.17
Counting 1-10 (accuracy)	10 (0)	10	9 (1.90)	10	38.50	-2.15	.15	-.46
<b>Counting 1-10 (seconds)</b>	5.09 (2.74)	3	11.73 (8.89)	10	96.50	2.38	<b>.02</b>	.51
Counting 1-10 (seconds) ( $n = 14$ )	5.14 (3.08)	3	7.28 (4.31)	5	34.00	1.23	.26	.33
Counting 5-1 (accuracy)	2.27 (2.61)	0	2.73 (2.61)	5	66.00	.42	.75	.09

Dot discriminations (accuracy)	8.36 (1.12)	8	7 (1.79)	7	34.50	-1.75	.08	-.37
Dot discriminations (seconds) ( $n = 20$ )	19.70 (15.72)	12.5	18.30 (4.88)	17	70.50	1.56	.12	.35
<b>Quantity Comparison (accuracy)</b>	20.45 (2.16)	21	18.18 (3.03)	18	29.50	-2.05	<b>.04</b>	-.44
Give-a-number task	5 (2.19)	5	5.64 (2.84)	7	68.50	.53	.61	.11

## 6. Discussion

The present work was aimed at analyzing the early numerical competences in individuals with DS. The participants in our study did not show an understanding of the reversibility concept. They presented irreversible mental representations, which are typical of the pre-operational stage. Participants with DS were indeed able to perform the one-to-one correspondence tasks, as has already been noted in this population by Caycho and colleagues (1991), but, in our case, only one subject out of two was able to use it as a strategy to infer numbers that were no longer visible. No subject with DS was able to understand that, when something changes in number or appearance, it is still the same, a concept known as conservation. They were not capable to dissociate spatial information from number-related information, showing they did not acquire awareness that actions can be reversed. In this study, individuals with DS and TD children presented similar prelogical operations. Our results show that the performances of individuals with DS on numerical tasks were well aligned with that of TD children matched for MA. DS and TD-MA groups did not differ on most tasks of numerical cognition: they indeed presented similar competences in counting, in mental calculation and cardinality. Concerning the latter skill, the performances of individuals with DS paralleled that of TD-MA children: all participants were able to identify the number's cardinality of a restricted number of objects (about 5 out of 9), confirming the same results reached by other authors (Caycho *et al.*, 1991; Bashash *et al.*, 2003; Sella *et al.*, 2013). Likewise, our results revealed that all the participants of the sample showed difficulties in a verbal calculation task. Lanfranchi and colleagues (Lanfranchi *et al.*, 2010; Lanfranchi, Baddeley, Gathercole, & Vianello, 2012) claimed that some problems with mental operations could be due to deficits in other functions, like working memory and attention, which are both considered important for the numerical development. As regards counting, Abdelahmeed's review (2007) described the presence of

significant difficulties in this area in individuals with DS. Our results were in line with previous research by Porter (1999) and revealed that participants with DS completed the sequences of counting as TD-MA children, but they made a greater number of errors and omissions. Therefore, the performances of individuals with DS on forward and backward counting, both in terms of accuracy and time, were well aligned with that of the TD-MA group. Some authors have recently analyzed the ANS and OTS systems in the DS population and have underlined, in particular, a deficit in the latter (Paterson *et al.*, 2006; Sella *et al.*, 2013). The present study also investigated the ANS system, through two quantity comparison tasks; the results show that dots discrimination competences of individuals with DS were well aligned to those of TD children matched for MA. Indeed, the scores in the dot discrimination task of the BIN 4-6 test (Molin *et al.*, 2007) did not show significant differences in the ANS between both groups. Nevertheless, we noted that subjects with DS performed more errors when they were asked to discriminate between two sets of dots and when there was inconsistency between the number of dots and the size of dots (e.g.: the set with the greater number of dots is composed of smaller dots) compared to the TD-MA group. We also noticed that the performances of participants were different by changing the presentation materials, namely by exposing subjects to an objects comparison task instead of a dot comparison task. Individuals with DS performed worse than TD-MA children in the quantity comparison test, in which they had to indicate the group with a greater number of elements, choosing between two illustrated baskets of fruit. In this test (SNUP; Tobia *et al.*, 2017), quantity discrimination in individuals with DS did not appear to be in line with mental age. In this respect, our data was in line with previous studies (Camos, 2009; Sella *et al.*, 2013; Abreu-Mendoza & Arias-Trejo, 2015): more research is needed but our results indicate that the ability to discriminate large non-symbolic numerosities in individuals with DS could be in line with the mental age, although it could be sensible to visual stimuli properties. Finally, we noted that the lexical knowledge of numbers was more developed in participants with DS than in TD-MA children. More specifically, we found that the clinical group performed significantly better than TD-MA children in the Arabic numbers reading task. The latter result could be a positive consequence of the longer exposure to the numbers due to their years of education and CA (“*experience effect*”, Fidler, Daunhauer, Will, & Schworer, 2018).

In sum, the present study reveals that early numerical competences of individuals with DS were well aligned to mental age, measured by a logical

thinking test: they can recognize numbers and associate them to their corresponding quantity, count from 1 to 20 and from 5 to 1 and they are sufficiently competent in using the cardinality principle. They are stronger in Arabic number reading, while we found inconsistent data concerning numerosity discrimination, leaving the debate still open.

More research is needed that would involve more individuals with DS: one limitation of this study was its small sample size. According to Edgin and colleagues (2010), there are some issues to consider when assessing the cognitive abilities of individuals with DS. As underlined by Pulina and colleagues (2019), some tasks may be too difficult for individuals with DS (*floor effect*). Moreover, the instruments generally used in research and clinical practices are standardized on TD individuals: they do not allow comparing results with normative data. Another issue could be linked to the *experience effect* (Fidler *et al.*, 2018): the scores of participants with DS could be a positive consequence of the environment ("*experience effect*"). The limitations and strengths of the current study lend themselves to several future research directions. It could be interesting to compare a group of children with DS and a group of TD with the same CA. It could also be interesting to monitor participants longitudinally with the purpose of analyzing predictive factors of numerical skills in atypical development. Moreover, it could also be useful to include different age groups or populations (i.e.: Williams Syndrome) and/or to compare performances of 4-year-old children with those of adolescents with DS, to understand the influence of education and environmental context on numerical cognition.

In conclusion, these results have implications to understand the development of numerical skills in individuals with DS. Moreover, the present findings suggest that numerical cognition and logical thinking should also be included in the assessment of the numerical skills of children with DS. During testing and evaluation, clinicians should use tools to analyze fluid reasoning (e.g.: IQ score) and the mental structures (e.g.: logical thinking). The mental age is a very important data in neuropsychological practices to underline the main strengths and weaknesses in each cognitive profile. Starting from these points, it is possible to plan and carry out neuropsychological interventions for most of the weaknesses. Moreover, projects of inclusion and well-being in both school and daily life could promote the main strengths in individuals with DS.

## References

- Abdelahmeed, H. (2007). Do Children with Down Syndrome Have Difficulty in Counting and Why? *International Journal of special education*, 22 (2), 129-139.
- Abreu-Mendoza, R. A., & Arias-Trejo, N. (2015). Numerical and area comparison abilities in Down syndrome. *Research in Developmental Disabilities*, 41, 58-65. <https://doi.org/10.1016/j.ridd.2015.05.008>.
- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders* (5<sup>th</sup> ed.). Arlington, VA: American Psychiatric Publishing.
- Bashash, L., Outhred, L., & Bochner, S. (2003). Counting skills and number concepts of students with moderate intellectual disabilities. *International Journal of Disability, Development and Education*, 50 (3), 325-345. <https://doi.org/10.1080/1034912032000120480>.
- Brigstocke, S., Hulme, C., & Nye, J. (2008). Number and arithmetic skills in children with Down syndrome. *Down Syndrome: Research and Practice*, 74-78. <https://doi.org/10.3104/reviews/2070>.
- Buckley, S. (2007). Teaching numeracy. *Down syndrome. Research and Practice*, 12 (1), 11-14. <https://doi.org/10.3104/updates.2031>.
- Camos, V. (2009). Numerosity discrimination in children with Down syndrome. *Developmental Neuropsychology*, 34 (4), 435-447. <https://doi.org/10.1080/87565640902964557>.
- Caycho, L., Gunn, P., & Siegal, M. (1991). Counting by children with Down syndrome. *American Journal on Mental Retardation*, 95 (5), 575-583.
- Chapman, R. S., & Hesketh, L. J. (2000). Behavioral phenotype of individuals with Down syndrome. *Mental Retardation and Developmental Disabilities Research Reviews*, 6 (2), 84-95. [https://doi.org/10.1002/1098-2779\(2000\)6:2<84::AID-MRDD2>3.0.CO;2-P](https://doi.org/10.1002/1098-2779(2000)6:2<84::AID-MRDD2>3.0.CO;2-P).



Contestabile, A., Benfenati, F., & Gasparini, L. (2010). Communication breaks-Down: from neurodevelopment defects to cognitive disabilities in Down syndrome. *Progress in Neurobiology*, 91 (1), 1-22. <https://doi.org/10.1016/j.pneurobio.2010.01.003>.

Cornoldi, C., Lucangeli, D., & Bellina, M. (2002). *AC-MT: test di valutazione delle abilità di calcolo e soluzione di problemi - gruppo MT* [AC-MT 6–11 – Test to assess calculation and problem solving skills]. Trento: Centro Studi Erickson.

Cornwell, A. C. (1974). Development of language, abstraction, and numerical concept formation in Down's syndrome children. *American Journal of Mental Deficiency*, 79 (2), 179-190.

Daunhauer, L. A., Fidler, D. J., Hahn, L., Will, E., Lee, N. R., & Hepburn, S. (2014). Profiles of everyday executive functioning in young children with Down syndrome. *American Journal on Intellectual and Developmental Disabilities*, 119 (4), 303-318. <https://doi.org/10.1352/1944-7558-119.4.303>.

Dunn, L. M., & Dunn, L. M. (1997). *Peabody picture vocabulary test*. Circle Pines: American Guidance Service.

Dyken, E. M., Hodapp, R. M., & Finucane, B. M. (2000). *Genetics and mental retardation syndromes*. Baltimore, MD: Paul H. Brookes Publishing Company.

Edgin, J. O., Mason, G. M., Allman, M. J., Capone, G. T., DeLeon, I., Maslen, C., Reeves, R. H., Sherman, S. L., Nadel, L. (2010). Development and validation of the Arizona cognitive test battery for down syndrome. *Journal of Neurodevelopmental Disorders*, 2 (3), 149-164.

Edgin, J. O., Tooley, U., Demara, B., Nyhuis, C., Anand, P., & Spanò, G. (2015). Sleep Disturbance and Expressive Language Development in Preschool Age Children With Down Syndrome. *Child Development*, 86 (6), 1984-1998. <https://doi.org/10.1111/cdev.12443>.

Fidler, D. J., Daunhauer, L. A., Will, E. A., & Schworer, E. (2018). Working memory in Down Syndrome. Methodological considerations and intervention implications. In T. P. Alloway (Ed.), *Working Memory and Clinical Developmental Disorders: Theories, Debates and Interventions* (pp.157-179). London: Routledge.

Field, A. (2018). *Discovering statistics using IBM SPSS statistics 5<sup>th</sup> edition*. London: Sage.

Geary, D. C. (1994). *Children's mathematical development: Research and practical applications*. Washington, DC: American Psychological Association.

Geary, D. C. (2000). From infancy to adulthood: The development of numerical abilities. *European Child & Adolescent Psychiatry*, 9 (2): S11.

Gelman, R., & Cohen, M. (1988). Qualitative differences in the way Down syndrome and normal children solve a novel counting task. In L. Nadel (Ed.), *The psychobiology of Down syndrome*. Massachusetts: MIT Press.

Gelman, R., & Gallistel, C. R. (1986). *The child's understanding of number*. Harvard University Press.

Grieco, J., Pulsifer, M., Seligsohn, K., Skotko, B., & Schwartz, A. (2015). Down syndrome: Cognitive and behavioral functioning across the lifespan. *American Journal of Medical Genetics Part C: Seminars in Medical Genetics*, 169 (2), 135-149. <https://doi.org/10.1002/ajmg.c.31439>.

Hanrahan, J., & Newman, T. (1996). Teaching addition to children. In B. Stratford & P. Gunn (Eds.), *New approaches to Down syndrome* (pp. 300-308). London: Continuum International Publishing Group.

Iacono, T., Torr, J., & Wong, H. Y. (2010). Relationships amongst age, language and related skills in adults with Down syndrome. *Research in Developmental Disabilities*, 31 (2), 568-576. <https://doi.org/10.1016/j.ridd.2009.12.009>.

Izard, V., Sann, C., Spelke, E. S., & Streri, A. (2009). Newborns infants perceive abstract numbers. *PNAS Proceedings of the National Academy of Sciences of the United States of America*, 106 (25), 10382-10385. <https://doi.org/10.1073/pnas.0812142106>.

Jarrold, C., Baddeley, A. D., & Hewes, A. K. (1999). Genetically dissociated components of working memory: Evidence from Downs and Williams syndrome. *Neuropsychologia*, 37 (6), 637-651. [https://doi.org/10.1016/S0028-3932\(98\)00128-6](https://doi.org/10.1016/S0028-3932(98)00128-6).

Karmiloff-Smith, A., Al-Janabi, T., D'Souza, H., Groet, J., Massand, E., Mok, K., Startin, C., Fisher, E., Hardy, J., Nizetic, D., Tybulewicz, V., & Strydom, A. (2016). The importance of understanding individual differences in Down syndrome. *F1000Research*, 5, F1000 Faculty Rev-389. <https://doi.org/10.12688/f1000research.7506.1>.

Kittler, P. M., Krinsky-McHale, S. J., & Devenny, D. A. (2008). Dual-task processing as a measure of executive function: A comparison between adults with Williams and Down syndromes. *American Journal of Mental Retardation*, 113 (2), 117-132.

Lanfranchi, S., Baddeley, A., Gathercole, S., & Vianello, R. (2012). Working memory in Down syndrome: is there a dual task deficit? *Journal of Intellectual Disability Research*, 56 (2), 157-166. <https://doi.org/10.1111/j.1365-2788.2011.01444.x>.

Lanfranchi, S., Berteletti, I., Torrisi, E., Vianello, R., & Zorzi, M. (2015). Numerical estimation in individuals with Down syndrome. *Research in Developmental Disabilities*, 36, 222-229. <https://doi.org/10.1016/j.ridd.2014.10.010>.

Lanfranchi, S., Jerman, O., Dal Pont, E., Alberti, A., & Vianello, R. (2010). Executive function in adolescents with Down syndrome. *Journal of Intellectual Disability Research*, 54 (4), 308-319. <https://doi.org/10.1111/j.1365-2788.2010.01262.x>.

Lee, N. R., Fidler, D. J., Blakely-Smith, A., Daunhauer, L., Robinson, C., & Hepburn, S. (2011). Parent-report of executive functioning in population-based sample of young children with Down syndrome. *American Journal on Intellectual and Developmental Disabilities*, 116, 290-304. <https://doi.org/10.1352/1944-7558-116.4.290>.

Lott, I. T., & Dierssen, M. (2010). Cognitive deficits and associated neurological complications in individuals with Down's syndrome. *The Lancet Neurology*, 9 (6), 623-633. [https://doi.org/10.1016/S1474-4422\(10\)70112-5](https://doi.org/10.1016/S1474-4422(10)70112-5).

Määttä, T., Kaski, M., Taanila, A., Keinänen-Kiukaanniemi, S., & Iivanainen, M. (2006). Sensory impairments and health concerns related to the degree of intellectual disability in people with Down syndrome. *Down Syndrome Research and Practice*, 11 (2), 78-83.

Marotta, L., Viezzoli, D., & Vicari, S. (2006). Le abilità numeriche e di calcolo nella Disabilità Intellettiva. *Età Evolutiva*, 83, 71-78.

Molin, A., Poli, S., & Lucangeli, D. (2007). *BIN 4-6. Batteria per la valutazione dell'intelligenza numerica in bambini dai 4 a 6 anni* [BIN 4-6 – Test to assess numerical and counting skills in children from 4 to 6 years old]. Trento: Centro Studi Erickson.

Moniga, S. (2007). Profilo di sviluppo sensomotorio in bambini con sindrome di Down: indici prognostici e interventi riabilitativi. *Psichiatria dell'Infanzia e dell'Adolescenza*, 74 (1), 99.

Nye, J., Fluck, M., & Buckley, S. (2001). Counting and cardinal understanding in children with Down syndrome and typically developing children. *Down Syndrome Research and Practice*, 7 (2), 68-78.

Orsini, A., Pezzuti, L., Picone, L. (2012). *WISC-IV. Contributo alla taratura italiana*. Florence: Giunti O.S.

Parker, S. E., Mai, C. T., Canfield, M. A., Rickard, R., Wang, Y., Meyer, R. E., Anderson, P., Mason, C. A., Collins, J. S., Kirby, R. S., & Correa, A. (2010). Updated national birth prevalence estimates for selected birth defects in the United States, 2004-2006. *Birth Defects Research Part A: Clinical and Molecular Teratology*, 88, 1008-1016. doi:10.1002/bdra.20735.

Paterson, S. J., Girelli, L., Butterworth, B., & Karmiloff-Smith, A. (2006). Are numerical impairments syndrome specific? Evidence from Williams syndrome and Down's syndrome. *Journal of Child Psychology and Psychiatry*, 47 (2), 190-204. <https://doi.org/10.1111/j.1469-7610.2005.01460.x>.

Patterson, T., Rapsey, C. M., & Glue, P. (2013). Systematic review of cognitive development across childhood in Down syndrome: implications for treatment interventions. *Journal of Intellectual Disability Research*, 57 (4), 306-318. <https://doi.org/10.1111/j.1365-2788.2012.01536.x>.

Pennington, B. F., Moon, J., Edgin, J., Stedron, J., & Nadel, L. (2003). The neuropsychology of Down syndrome: Evidence for hippocampal dysfunction. *Child Development*, 74 (1), 75-93. <https://doi.org/10.1111/1467-8624.00522>.

Pizzoli, C., Lami, L., & Stella, G. (2001). Le prime tappe dello sviluppo psicomotorio: aspetti cognitivi. In A. Contardi & S. Vicari (Eds.), *Le persone Down. Aspetti neuropsicologici, educativi e sociali*, (pp. 15-26). Milano: FrancoAngeli.

Porter, J. (1999). Learning to count: A difficult task? *Down Syndrome: Research and Practice*, 6 (2), 85-94.

Pulina, F., Vianello, R., & Lanfranchi, S. (2019). Cognitive profiles in individuals with Down syndrome. *International Review of Research in Developmental Disabilities*, 56, 67-92. <https://doi.org/10.1016/bs.irrdd.2019.06.002>.

Rowe, J., Lavender, A., & Turk, V. (2006). Cognitive executive function in Down's syndrome. *British Journal of Clinical Psychology*, 45 (1), 5-17. <https://doi.org/10.1348/014466505X29594>.

Rynders, J. E. (1999). Favorire la competenza educativa degli studenti con Sindrome di Down. In J. A. Rondal, J. Perera, & L. Nadel (Eds.), *Down's syndrome: A review of current knowledge* (pp. 75-92). London: Whurr Publishers.

Saggino, A., Stella, G., & Vio, C. (2019). *Wechsler Preschool and Primary Scale of Intelligence – Fourth Edition*. Italian version. Florence: Giunti O.S.

Sella, F., Lanfranchi, S., & Zorzi, M. (2013). Enumeration skills in Down syndrome. *Research in Developmental Disabilities*, 34 (11), 3798-3806. <https://doi.org/10.1016/j.ridd.2013.07.038>.

Sestili, M., Moalli, E., & Vianello, R. (2006). Pensiero logico, lettura, scrittura e abilità di calcolo in ragazzi con Sindrome di Down di II e III media. *La Sindrome di Down*, 151-164.

Tobia, V., Bonifacci, P., & Marzocchi, G. M. (2017). *SNUP. Senso del numero: prerequisiti*. [SNUP – Test to assess number sense: prerequisites]. Florence: Hogrefe.

Van De Rijt, B. A., & Van Luit, J. E. (1998). Effectiveness of the Additional Early Mathematics program for teaching children early mathematics. *Instructional Science*, 26 (5), 337-358.

Vianello, R. (2006). *La sindrome di Down: Sviluppo psicologico e integrazione dalla nascita all'età senile*. Bergamo: Edizioni Junior

Vianello, R., Lanfranchi, S., & Moalli, E. (2006). Lo sviluppo del pensiero logico in bambini e ragazzi con sindrome di Down. In R. Vianello (Ed.), *La sindrome di Down* (pp. 142- 145). Bergamo: Edizioni Junior.

Vianello, R., & Marin, M. L. (1997). *OL18. Operazioni Logiche Progetto MS* [Logical operations test – OL18]. Bergamo: Edizioni Junior.

Vicari, S. (2004). Sindromi genetiche ed abilità cognitive: aspetti differenziali. Presentazione: disturbi neurocognitivi e sindromi genetiche. *Psicologia Clinica dello Sviluppo*, 8 (3), 419-436.

Vicari, S., Bellucci, S., & Carlesimo, G. A. (2005). Visual and spatial long-term memory: differential pattern of impairments in Williams and Down syndromes. *Developmental Medicine & Child Neurology*, 47 (05), 305-311. <https://doi.org/10.1017/S0012162205000599>.

Vicari, S., Marotta, L., & Carlesimo, G. A. (2004). Verbal short-term memory in Down's syndrome: An articulatory loop deficit? *Journal of Intellectual Disability Research: JIDR*, 48 (2), 80-92. <https://doi.org/10.1111/j.1365-2788.2004.00478.x>.

Wechsler, D. (2012). *Wechsler Preschool and Primary Scale of Intelligence – Fourth Edition*. San Antonio, TX: The Psychological Corporation.

World Health Organization. (2015). *World health statistics 2015*. World Health Organization. <https://apps.who.int/iris/handle/10665/170250>.

Wynn, K. (1990). Children's understanding of counting. *Cognition*, 36 (2), 155-193. [https://doi.org/10.1016/0010-0277\(90\)90003-3](https://doi.org/10.1016/0010-0277(90)90003-3).

Wynn, K. (1992). Children's acquisition of the number words and the counting system. *Cognitive Psychology*, 24 (2), 220-251. [https://doi.org/10.1016/0010-0285\(92\)90008-P](https://doi.org/10.1016/0010-0285(92)90008-P).

Zigler, E. (1969). Developmental versus difference theories of mental retardation and the problem of motivation. *American Journal of Mental Deficiency*, 73, 536-556.





# Burden and professional quality of life of caregivers: The clinical role of suppression and resilience

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## Abstract

*Background: the caregiver's role is fundamental in the context of healthcare and the well-being of caregivers is often undermined by traumatic experiences and suffering. The aim of this study was to evaluate the relationship among the caregiver's resilience, burden, suppressive mechanisms and professional quality of life. Goals and methods: Participants were 665 caregivers, 458 women and 207 men aged between 18 and 81 years old. In order to highlight the above-mentioned relationship, caregivers were asked to complete a Suppression Mental Questionnaire (SMQ), a Resilience Scale for Adults (RSA) self-report, a Caregiver Burden Inventory (CBI) and a Professional Quality of Life Measure (ProQOL-5). A correlation and linear regression*

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*analysis was thus performed on the results obtained. Results: The Suppression Mental factors were positively correlated with both Resilience and Burden, with the exception of Regression in the Service of the Ego. Resilience was also positively correlated with Secondary Traumatic Stress and Burnout but negatively correlated with Compassion Satisfaction. Both significant correlations and linear regressions emerged in terms of the relationship between caregiver's variables and total scores. Conclusions: Burnout and Secondary Traumatic Stress can affect the caregiver's well-being, so that resilience and suppressive mechanisms are fundamental. These components should be considered with a particular reference to the current health emergency.*

**Keywords:** Burden; Caregivers; Clinical psychology; Compassion Satisfaction; Fatigue; Resilience; Quality of Life.

## 1. Introduction

Caregivers offer assistance and support to patients with different medical conditions and their psychophysical well-being is often undermined by prolonged conditions of stress, which can even affect their quality of life (Collins & Swartz, 2011).

Although assisting another person could represent the occasion to reach different benefits, including self-realization as well as the satisfaction of alleviating the suffering of others, the possibility to develop burdens is high to the point of compromising the quality of the care provided (Meier, Back, & Morrison, 2001; Shanafelt, Bradley, Wipf, & Back, 2002; Aldrich, 2011).

Previously published research indicated that the continuous exposure to experiences of suffering disease and death, may lead to the onset of trauma and burnout syndromes in caregivers (McCloskey & Taggart, 2010; Hamilton, Tran, & Jamieson, 2016; Catalano, Sardella, Bellone, Lasco, Martino, & Morabito, 2019; Rosa, Tomai, Lauriola, & Martino, 2019; Settineri, Frisone, Alibrandi, & Merlo, 2019; Merlo, McNabney, Frisone, Sicari, Paunica, Motofei *et al.*, 2020; Merlo, Stoian, Motofei, & Settineri, 2020), understood as a physical, emotional and mental process characterized by an exhaustion evolving over time in response to excessive workloads and to high performance requirements (Figley, 1995; Platania, Di Nuovo, Caruso, Digrandi, & Caponnetto, 2020). It can be defined as a condition in which the perception of personal needs exceeds the perception of resources (Flarity, Gentry, & Mesnikoff, 2013) and whose key dimensions foresee emotional exhaustion, depersonalization and a deep sense of ineffectiveness (Maslach, Schaufeli, & Leiter, 2001). These symptoms can manifest themselves through dissociative phenomena, which involve exclusion and dissatisfaction in terms of personal and social aspects (Benau, 2020).

Another factor involved is Secondary Traumatic Stress (STS), which concerns, instead, a condition of maladjustment deriving from the contact with other people's suffering, sorrow and traumatic experiences and that culminates in a decrease of compassionate feelings. Unlike burnout, this condition emerges suddenly and sharply, accompanied by confusion and a sense of helplessness (Figley, 2002; Neumann, Edelhäuser, Tauschel, Fischer, Wirtz, Woopen *et al.*, 2011; Sinclair, Raffin-Bouchal, Venturato, Mijovic-Kondejewski, & Smith-MacDonald, 2017). In both conditions, it is clear that the level of "burden" sustained by the caregiver is quite high (Hoenig & Hamilton, 1966; Lim, Cheah, Ali, Han, Anthony, Chan *et al.*, 2014; Velutti, Pavesi, Poggio, Carettoni, Saetta, Arcanà *et al.*, 2017;

Widowati, Devi, & Nihayati, 2018) in terms of temporal, evolutionary, physical, social and emotional impact (Kosberg, Cairl, & Keller, 1990), as is supported by several studies (Collins & Long, 2003; Sabin-Farrell & Turpin, 2003; Elwood, Mott, Lohr, & Galovski, 2011; Beaumont, Durkin, Martin, & Carson, 2016; Duarte & Pinto-Gouveia, 2017; Win, Chong, Ali, Chan, & Lim, 2017).

Despite the obvious risk, research has demonstrated that many caregivers do not develop high levels of psychopathological issues when motivated by a sense of satisfaction, allowing them to engage in meaningful interactions (Ray, Wong, White, & Heaslip, 2013; Parker Oliver, Patil, Benson, Gage, Washington, Kruse *et al.*, 2017). This construct, called Compassion Satisfaction (CS), mitigates the impact of STS and of the burnout syndrome (Haley, LaMonde, Han, Burton, & Schonwetter, 2003; Lynch, Shuster, & Lobo, 2018; Allday, Newell, & Sukovskyy, 2020), which together represent opposite but intrinsic possibilities linked to the quality of professional life (Stamm, 2005).

In addition to CS, numerous studies also enhanced the role of resilience as a protective factor; this construct, which is understood as the ability to reach adaptation with reference to changes and to cope with negative events (Jackson, Firtko, & Edenborough, 2007; Bonanno, 2012; Palacio, Krikorian, & Limonero, 2018), would be negatively related to Burnout and positively related to Compassion Satisfaction, suggesting the importance of its role in caregiver's professional and private life (Gito, Ihara, & Ogata, 2013; Burnett & Wahl, 2015; Olson, Kemper, & Mahan, 2015; Rushton, Batcheller, Schroeder, & Donohue, 2015).

In this sense, it is clear that caregivers are particularly vulnerable to psychological suffering: the need to achieve a sort of adaptive process (Cramer, 1998) determines the adoption of defensive mechanisms in order to avoid unacceptable mental states. More specifically, the mechanism of suppression, which allows subjects to consciously suppress unpleasant emotional contents (Settineri, Frisone, Merlo, Geraci, & Martino, 2019), takes a fundamental role in this context. Starting from its main dimensions, it seems that a moderate implementation of these mechanisms favors the above-mentioned adaptation and helps the subject to dominate traumatic or stressful situations. An excessive use of these defenses could, instead, compromise the caregiver's mental and physical well-being and the quality of the care provided (Hyphantis, Tomenson, Bai, Tsianos, Mavreas, & Creed, 2010; Di Giuseppe, Ciacchini, Micheloni, Bertolucci, Marchi, & Conversano, 2018; Saeed, Salehi, Alavi, Ajdarkosh, Kashaninasab, &

Esfahani, 2019), thus again affecting the patient's representation in terms of affective symbolization (Caputo, 2013).

### *1.1. Objective and hypotheses*

The study was aimed at highlighting the relationship among a series of variables associated with the quality of life and well-being of caregivers. In this study we hypothesized that: (1) There were significant correlations between the suppressive tendencies of the caregiver and resilience; (2) There were significant correlations between the suppressive tendencies of the caregiver and the burden associated with the workload; (3) There were significant correlations between resilience and the variables descriptive of the caregivers' professional quality of life; (4) There were significant correlations among the personal characteristics of the caregiver and the overall factors relating to: burden, repressive tendencies, resilience and professional quality of life, and (5) there were causal relationships among the two main predictors, identified as Age and Gender, and factors such as: burden, repressive tendencies, resilience and professional quality of life.

## **2. Methods**

### *2.1. Procedure and participants*

The sample consisted of 665 subjects, 458 women (68.9%) and 207 men (31.1%). The age of participants ranged from 18 to 81 years old ( $M_{\text{age}} = 41.74$ ;  $SD = 13.61$ ). The questionnaires were filled out in paper and pencil and each participant was informed about the anonymous nature of the methods of data processing before signing the informed consent.

### *2.2. Statistical analysis*

Numerical data was expressed by mean and standard deviation and the categorical variables as number and percentage. Spearman correlations were computed among the demographic variables and empirically validated through inventories to determine the potential associations between these factors.

Multivariate linear regression was used to assess the dependence of each burden-related outcome (Time Dependence Burden, Developmental Burden,

Physical Burden, Social Burden, Emotional Burden and Total CBI score) on a set of independent predictors (Age and Gender).

Statistical analyses were performed using SPSS 26 for Windows package. A *p*-value smaller than .050 was considered to be statistically significant.

### 2.3. Instruments of observation

The participants were asked to provide some personal data, such as gender, age, educational qualification and the amount of time dedicated to the profession, expressed in days, hours and years of work. In addition, they were exposed to the four instruments of observation described, as follows:

- The *Suppression Mental Questionnaire (SMQ)* (Settineri, Merlo, Frisone, Alibrandi, Carrozzino, Diaconu *et al.*, 2019) is a self-report instrument consisting of 18 items based on a 5-point Likert scale. The original validation study included a factor analysis whose results highlighted the presence of three main factors: Repressive function, Regression in the service of the Ego, and Rationalization. The first function concerns the removal of disturbing contents; the second consists of a temporarily regressive activity based on imagery and fantasies, in order to manage conflicts; the third function concerns a logical process of ordering contents and actions. The adequacy of sampling ( $KMO = .648$ ) was demonstrated in a preliminary study (Saeed *et al.*, 2019), suggesting the following alpha coefficients: Repressive function = .742; Regression in the service of the Ego = .804; Rationalization = .698. The items referring to each factor were: Factor 1, items 3, 4, 7, 8, 10, 14, 15, 16, 17, 18; Factor 2, items 5, 6, 9, 11, 12; Factor 3, items 1, 2, 7, 13. A subsequent study (Tauschke, Merskey, & Helmes, 1990) introduced an app-based version of the tool, so that Cronbach's alpha coefficients corresponded to: .74 - .73 for the first factor, .80 - .77 for the second, and .70 - .76 for the third.
- The *Resilience Scale for Adults (RSA)* is a 33-item self-report scale based on a 5-point Likert scale, assessing resilience factors, defined as the ability to deal with traumatic events in a positive way (Hjemdal, Friborg, Martinussen, & Rosenvinge, 2001; Friborg, Hjemdal, Rosenvinge, & Martinussen, 2003; Friborg, Barlaug, Martinussen, Rosenvinge, & Hjemdal, 2005). The Italian adaptation study

(Capanna, Stratta, Hjemdal, Collazzoni, & Rossi, 2015) produced the following results: Perception of Self ( $\alpha = .74$  by Cronbach) referred to self-confidence and ability; Planned future ( $\alpha = .73$ ) was related to the sense of confidence in the opportunity for success; Social competence ( $\alpha = .83$ ) regarded the fact of feeling comfortable in a social context; Structured style ( $\alpha = .80$ ) was relative to the orientation of the objective; Family cohesion ( $\alpha = .80$ ) concerned shared values and mutual appreciation and Social resources ( $\alpha = .80$ ) referred to social support (Friborg *et al.*, 2005; Hjemdal, Friborg, Stiles, Rosenvinge, & Martinussen, 2006). The weights of the items, referring to the six factors described above, provided the following assignment: Factor 1, items 1, 7, 13, 19, 25, 29; Factor 2, items 2, 8, 14, 20; Factor 3, items 3, 9, 15, 21, 26, 30; Factor 4, items 6, 12, 18, 24; Factor 5, items 4, 10, 16, 22, 27, 31; Factor 6, items 5, 11, 17, 23, 28, 32, 33.

- The *Caregiver Burden Inventory (CBI)* (Novak & Guest, 1989; Italian version by Conti, Clari, Garrino, Maitan, Scivoletto, Cavallaro *et al.*, 2019) assesses the caregiver's burden, in reference to five dimensions: Time Dependence Burden associated with the time required for assistance; Developmental Burden concerns the caregiver's feeling of not having the same opportunities as peers; Physical Burden describes the load that pours on the body; Social Burden refers to the perception of a role conflict and Emotional Burden describes the mood resulting from the patient's conduct. The CBI consists of 24 items measured through a 5-point Likert scale. The reliability indicators were as follows: .96 for the total scale, .91 for the time dependence burden, .92 for the developmental burden, .88, .89 and .93 for the physical, social and emotional burden, respectively. In a second study, published by Conti and collaborators (2019), the total score index was .91 and ranged from .76 to .91 for the above-mentioned factors. The items referring to each factor were divided as follows: Factor 1, items 1, 2, 3, 4, 5; Factor 2, items 6, 7, 8, 9, 10; Factor 3, items 11, 12, 13, 14; Factor 4, items 15, 16, 17, 18, 19; Factor 5: 20, 21, 22, 23, 24.
- The *Professional Quality of Life Scale (ProQOL-5)* (Stamm, 2005; Italian adaptation by Palestini, Prati, Pietrantonio, & Cicognani, 2009), is a self-report scale aimed at evaluating the caregiver's well-being and suffering, associated with the nursing experiences. The instrument is composed of three scales: Compassion Satisfaction, Burnout and

Secondary Traumatic Stress. ProQOL-5 involves 30 items, based on an ordinal 5-point Likert scale. Satisfaction of compassion, reliability of the alpha scale = .88; Burnout, reliability of the alpha scale = .75; Secondary traumatic stress, alpha scale reliability = .81. The weights of the items referred to the three factors provided for the following assignment: Factor 1, items 3, 6, 12, 16, 18, 20, 22, 24, 27, 30; Factor 2, items 1, 4, 8, 10, 15, 17, 19, 21, 26, 29; Factor 3, items 2, 5, 7, 9, 11, 13, 14, 23, 25, 28.

### 3. Results

The caregivers' personal data and characteristics, as well as means and standard deviations of all factors evaluated, are summarized in Table 1.

Table 1 – *Descriptive statistics: characteristics of the sample and factors evaluated*

	<i>M</i>	<i>SD</i>
Age	41.74	13.61
Years of study	13.88	3.45
Working days per week	5.66	1.39
Working hours for week	43.82	30.73
Years of work	8.92	9.1
<i>Caregiver Burden Inventory</i>		
Time Dependence Burden	11.31	5.73
Developmental Burden	7.45	5.99
Physical Burden	7.93	5.62
Social Burden	4.51	4.63
Emotional Burden	2.7	3.74
Caregiver Burden Inventory Total Score	32.07	18.92
<i>Professional Quality of Life Measure</i>		
Compassion Satisfaction	36.68	7.41
Secondary Traumatic Stress	23.39	7.58
Burnout	23.95	6.68
Professional Quality of Life Scale Total Score	84.03	10.79
<i>Suppression Mental Questionnaire</i>		
Repressive Function	23.34	6.88
Regression in the Service of the Ego	13.04	4.35
Rationalization	10.89	3.23
Suppression Mental Questionnaire Total Score	50.1	8.08



*Resilience Scale for Adults*

Perception of Self	14.04	2.36
Planned Future	12.16	2.42
Social Competence	17.46	2.78
Structured Style	11.42	2.08
Family Cohesion	18.35	3.04
Social Resources	19.25	2.94
Resilience Scale for Adult Total Score	96.07	10.16

*3.1. Hypothesis 1*

The first hypothesis focused on the relationship between the caregivers' suppressive tendencies and their resilience, which was assessed through the Suppression Mental Questionnaire and the Resilience Scale for Adults self-report, respectively (Tab. 2).

Table 2 – *Correlation coefficients between the Suppression Mental Questionnaire (SMQ) and the Resilience Scale for Adults (RSA) self-report*

	Perception of Self	Planned Future	Social Competence	Structured Style	Family Cohesion	Social Resources	Resilience Scale for Adults Total Score
Repressive Function	.154**	.169**	.072	.064	.070	.089*	.157**
Regression in the Service of the Ego	-.184**	-.089*	-.013	-.066	-.035	.019	-.084*
Rationalization	.175**	.097*	-.019	.027	-.021	.013	.054
Suppression Mental Questionnaire Total Score	.039	.067	.040	.034	.032	.091*	.080*

\* $p < .05$  (two-tailed); \*\* $p < .01$  (two-tailed)

The results highlight several significant correlations: the total SMQ score was positively correlated with the overall scores of the Resilience questionnaire and with one of its components (i.e. Social Resources). The

first result suggests that the mechanism of suppression was in line with resilience since both can be considered two conscious cognitive functions. If suppression is considered a high-level defense, it could be inferred that it is associated with a psychological well-being and is supported by resilience. The significant relationship between suppression and the social resources of the caregiver can thus be explained in this sense.

With reference to the dimensions of the Suppression Mental Questionnaire, the Repressive Function was positively correlated with the overall score of the RSA and with some of its components, such as the Perception of Self, Planned Future and Social Resources. The Repressive Function allows the subject to consciously banish disturbing thoughts; this could, therefore, be moderately used as a strategy to deal with stressful situations in a positive way, as was proven by the high overall RSA scores. In this perspective, its use can be closely linked to domains such as self-efficacy, optimistic vision of the future and social support.

Regarding the Regression in the Service of the Ego, in this case negative correlations emerged with the total RSA score, the Perception of Self and Planned Future. This could suggest that resilient, self-effective and future-oriented individuals rely on more functional strategies to face reality, which do not require appeal to imagination.

Finally, with reference to Rationalization, positive correlations emerged with the Perception of Self and Planned Future. This would suggest that caregivers with higher levels of self-confidence and of planned future could benefit from the rationalization of contents and representations, managing the stress deriving from the burden of nursing through this mechanism.

### *3.2. Hypothesis 2*

The second hypothesis (Tab. 3) regarded the relationship between the scores of the Suppression Mental Questionnaire (SMQ) and the factors of the Caregiver Burden Inventory (CBI). More in detail, the SMQ total score was positively correlated to all the variables with the only exception of the Time Dependence Burden.

As regards these results, it can be assumed that the subjects used the suppression mechanism as a way to deal with stressful situations and manage emotions more in general. This modality could be implemented especially by caregivers who perceive few social opportunities, by experiencing a physical fatigue and a high social burden, which may be

related to conflicts of role and of time-sharing between work and family activity.

Table 3 – *Correlation coefficients between the Suppression Mental Questionnaire (SMQ) and the Caregiver Burden Inventory (CBI)*

	Time Dependence Burden	Development al Burden	Physical Burden	Social Burden	Emotional Burden	Caregiver Burden Inventory Total Score
Repressive Function	.077*	.280**	.254**	.244**	.229**	.276**
Regression in the Service of the Ego	.029	-.036	-.020	-.069	-.043	-.017
Rationalization	.098*	.179**	.171**	.137**	.087*	.185**
Suppression Mental Questionnaire Total Score	.065	.188**	.166**	.133**	.100**	.091*

\* $p < .05$  (two-tailed); \*\* $p < .01$  (two-tailed)

The correlation with the total CBI score also suggested the need for the subjects with high levels of burden to escape from grueling nursing activities.

With reference to the other dimensions, a positive correlation emerged between the Repressive Function and all the domains of the CBI. This data suggests that caregivers, oppressed by the nursing activities, face these stressful situations through repressive mechanisms.

Rationalization was also positively correlated to all the dimensions of the CBI, suggesting the need for subjects with a high burden to manage suffering through logical constructs.

No significant results emerged with reference to the Regression in the Service of the Ego.

### 3.3. Hypothesis 3

The third hypothesis (Tab. 4) concerned the relationship between the Professional Quality of Life Measure (ProQOL-5) and the Resilience Scale for Adults (RSA). The score related to the Compassion Satisfaction was negatively correlated with the overall RSA scores and with the components Planned Future, Social Competence and Social Resources. The results could suggest that subjects with higher levels of Compassion Satisfaction developed resilience, gratified by their work activities: a capacity that implies "self-repair" deriving from the perceived workload. This result could also explain the negative relationships with the domains related to future planning, skills and social resources.

Table 4 – *Correlation coefficients between the Professional Quality of Life Measure (ProQol-5) and the Resilience Scale for Adults (RSA)*

	Perception of Self	Planned Future	Social Competence	Structured Style	Family Cohesion	Social Resources	Resilience Scale for Adults Total Score
Compassion Satisfaction	-.071	-.203**	-.093*	.009	-.028	-.131**	-.145**
Secondary Traumatic Stress	.021	.257**	.069	-.026	.048	.112**	.143**
Burnout	.052	.293**	.084*	.002	.066	.114**	.154**
Professional Quality of Life Scale Total Score	.003	.230**	.025	-.017	.075	.068	.104**

\* $p < .05$  (two-tailed); \*\* $p < .01$  (two-tailed).

As for the second domain (i.e. Secondary Traumatic Stress), it emerged that this variable was positively correlated with the total RSA scores and with the factors related to Planned Future and Social Resources, suggesting both an increase in resilience in the most traumatized caregivers and an inversion of compassionate feelings towards them. In fact, we consider STS

as deriving from a sense of exhaustion that culminates in a decrease of compassionate feelings.

With reference to the Burnout domain, positive correlations emerged with the total RSA scores, with Planned Future, Social Competence and Social Resources, suggesting that individuals with high psychophysical exhaustion could develop greater resilience in the above-mentioned domains.

Finally, with reference to the last domain, the overall ProQol-5 score was positively correlated with the overall RSA score and with the Planned Future component. In line with previous data, these results suggest that subjects exposed to Burnout or Secondary Traumatic Stress can develop resilience and set new adaptive perspectives.

### 3.4 Hypothesis 4

In the fourth hypothesis, the personal characteristics of the caregiver were related to the total scores of all the scales administered (Tab. 5).

Table 5 – *Correlation coefficients between the caregivers' personal data and the CBI, SMQ, RSA and ProQol-5 scales*

	CBI Total Score	SMQ Total Score	RSA Total Score	ProQol-5 Total Score
Age	.254**	.019	-.013	.123**
Years of study	-.189**	-.113**	-.009	-.033
Working days for week	.292**	.112**	.084*	.160**
Working hours for week	.188**	.121**	.059	.113**
Years of work	.189**	.005	-.053	.123**

\* $p < .05$  (two-tailed); \*\* $p < .01$  (two-tailed)

Age was positively correlated with the CBI and with the ProQol-5, suggesting that, with an increase in age of the caregivers, the burden related to treatment is perceived more severely and caregivers are more subject to weakness and stress. The years of work factor was negatively correlated with the CBI and the SMQ, suggesting that more intensive training could represent the acquisition of functional coping strategies, which do not require the use of suppression.

The weekly working days were positively correlated with the all four total scores (CBI, SMQ, RSA, ProQol-5). A higher number of working days could be associated with the perception of a healthcare burden, exposing the subject to a greater risk of STS or burnout. At the same time, the greater experience of caregivers could correlate with the development of functional strategies to manage stress, including suppression and resilience.

The working hours per week were positively correlated with the CBI, SMQ and ProQol-5. Again, it appeared that the hours dedicated to work could have an effect on the professional health of the caregiver, exposing him/her to burnout or STS. Suppression could represent a mechanism useful to dominate suffering and exhaustion.

Finally, the years of work were positively correlated with the CBI and ProQol-5 scores, also suggesting in this case that the time spent working over the years could be associated with a higher burden perceived and to the risk of STS or burnout.

### *3.5 Hypothesis 5*

The fifth hypothesis concerned the relationship between the independent variables of age and gender and the factors referring both to the overall test scores and to the individual domains (Tab. 6).

With reference to the overall scores, the results show that age was positively correlated with both burden and professional quality of life, suggesting that older caregivers could be more affected by burden with negative effects on their mental and physical well-being. Gender, on the other hand, was significantly associated with both the suppressive tendencies of the caregiver and with resilience: this result suggests that being a female could affect the use of the two mentioned strategies, useful to deal with highly stressful situations.

Regarding the relationship between age and the individual CBI domains, positive relationships emerged with the: Time-Dependence Burden, Developmental Burden and Physical Burden. This suggests that the burden assessed could be affected by the advancement of age, with particular reference to the perception of: time restrictions, lack of social opportunities and sensations of chronic fatigue. As for gender, a significant impact emerged on the Social Burden dimension, suggesting that being a male could determine the perception of a role conflict with the professional activity.

Table 6 – Results of the linear regression analysis

	Age		Gender	
	B (95% C.I.)	p	B (95% C.I.)	p
CBI Total Score	.365 (.262; .467)	.000	-1.340 (-4.347; 1.668)	.382
ProQol-5 Total Score	.119 (.059; .179)	.000	1.337 (-.418; 3.092)	.135
SMQ Total Score	.009 (-.037; .054)	.711	1.318 (-.011; 2.646)	.050
RSA Total Score	.001 (-.055; .058)	.966	2.446 (.782; 4.111)	.004
Time Dependence Burden	.131 (.101; .162)	.000	.091 (-.806; .988)	.842
Developmental Burden	.107 (.075; .140)	.000	-.470 (-1.428; .487)	.335
Physical Burden	.117 (.087; .148)	.000	.136 (-.752; 1.024)	.763
Social Burden	.024 (-.001; .050)	.064	-.759 (-1.519; .001)	.050
Emotional Burden	.006 (-.015; .027)	.590	-.519 (-1.134; .096)	.098
Compassion Satisfaction	-.090 (-.131; -.049)	.000	.424 (-.780; 1.628)	.489
Secondary Traumatic Stress	.113 (.072; .155)	.000	1.427 (.209; 2.644)	.022
Burnout	.096 (.059; .132)	.000	-.517 (-1.598; .564)	.348
Repressive Function	-.043(-.081; -.005)	.028	1.614 (.491; 2.738)	.005
Regression in the Service of the Ego	.061 (.037; .084)	.000	-.810 (-1.512; -.108)	.024
Rationalization	-.014(-.032; .004)	.120	.854 (.327; 1.381)	.002
Perception of Self	-.018(-.031; -.005)	.006	.571 (.186; .956)	.004
Planned Future	.023 (.009; .036)	.001	.443 (.049; .837)	.027
Social Competence	.011 (-.004; .027)	.149	.350 (-.107; .807)	.133
Structured Style	-.010 (-.022; .002)	.960	-.031 (-.374; .311)	.857
Family Cohesion	-.015 (-.032; .002)	.085	.989 (.494; 1.484)	.000
Social Resources	.010 (-.006; .027)	.219	.182 (-.303; .666)	.462

\* $p < .05$

With reference to the relationship between age and the individual ProQol-5 domains, a positive relationship emerged with the domains of Burnout and Secondary Traumatic Stress and a negative relationship with the Compassion Satisfaction domain, suggesting that advancing age favors the appearance of Secondary Traumatic Stress and of the burnout syndrome while younger people feel more satisfied and gratified by the healthcare profession. In terms of secondary trauma, a significant relationship also emerged with the female gender, suggesting the possibility that women are more affected by the contact with the patient's suffering.

Regarding the relationship between age and the SMQ domains, a positive correlation was found with Regression in the Service of the Ego while a negative relationship emerged with the Repressive Function. The results suggest that younger subjects use the second mechanism to remove

distressing thoughts, while older subjects prefer the first mechanism, regressing to fantasy and imagination. As for gender, it also seems that women use the Repressive and Rationalization Function, in contrast to men who use the Regression in the Service of the Ego.

Finally, the regressions regarding age and the individual RSA domains were also significant: more specifically, positive associations emerged with the Planned Future variable while negative associations were found with the Perception of Self domain. This suggests that younger people are more self-efficient while caregivers tend to focus more on their future as they age. The female gender was positively associated with Perception of Self, Planned Future and Family Cohesion, suggesting the tendency to perceive oneself better, to set new goals and to count on the presence of shared values.

#### 4. Discussion

The results of this study focus on the analysis of the strategies adopted by caregivers to support the cost of healthcare and the factors that can affect their well-being (Pinquart & Sörensen, 2007; Wong, Lam, Chan, & Chan, 2012; Wood, Taylor-Stokes, Smith, Chirita, & Chaib Torralba, 2018; Price, Surr, Gough, & Ashley, 2020).

With regard to the aforementioned strategies, it emerged that resilient and socially supported subjects use suppressive mechanisms and, in particular, the Repressive Function to cope with the cost associated to nursing activities. We must consider, in fact, that resilient caregivers are able to cope with stressful and traumatic events also through the use of these defensive mechanisms, which support them in managing environmental and internal requests. As an evidence of their adaptive meaning, it emerged that even subjects with high social support tend to use these strategies suggesting their effectiveness in managing the care burden, as reported by the studies of Metzger (2014). Furthermore, the investment in the interpersonal domain could also respond to a repair strategy that allows the subject to rebuild a relationship between him/herself and the outside world (Caputo, 2019).

We also found that caregivers with a positive perception of themselves and their future use the Repressive Function and Rationalization more, at the expense of Regression in the Service of the Ego. In fact, individuals normally respond to unpleasant thoughts and emotions by trying to control or modify them (Horowitz, 1993): our results confirm that the functional and integrated use of the first two mechanisms allows caregivers to manage distressing thoughts or emotions through the use of logic, without the need



to take refuge in fantasy (Di Giuseppe, Ciacchini, Piarulli, Nepa, & Conversano, 2019; Di Giuseppe, Di Silvestre, Lo Sterzo, Hitchcott, Gemignani, & Conversano, 2019).

However, our studies also highlighted a use of suppressive mechanisms, in particular of the Repressive Function and Rationalization in subjects exposed to high levels of burden. More in detail, it seems that caregivers with poor social opportunities, who feel a sense of physical and emotional wear and tear and who perceive a conflict between the working and social or family spheres, try to manage the high burden through the aforementioned strategies. It is not surprising that these defense mechanisms are implemented in particularly stressful situations even if their chronic use can increase the risk of psychopathologies since, in the long term, the lack of expression of emotions and the lack of understanding of the distress can interfere with adaptation (Deburge, 2001; Caputo, 2013; Carrozzino, Costabile, Patierno, Settineri, & Fulcheri, 2019; Merlo, 2019a; Settineri, Frisone, & Merlo, 2019; Manfredi, 2020).

Interesting results emerged with reference to resilience and its relationship with some dimensions of the professional quality of life. In particular, it appears that Compassion Satisfaction is negatively correlated with resilience. We must consider that resilience is a dynamic process (Luthar, Cicchetti, & Becker, 2000), used to cope with adverse conditions or to "repair" following an injury suffered, while CS regards feelings of pleasure and gratification in relation to one's work (Ray *et al.*, 2013). This could suggest that people with CS are already satisfied by their profession and don't show a capacity that develops in adverse or stressful conditions. This could also justify the negative relationship between CS and factors regarding future planning, skills and social resources.

These results are also consistent in light of the positive correlations between some domains of resilience and the conditions of burnout and Secondary Traumatic Stress, a relationship that has been confirmed by several studies (David, 2012; Burnett & Wahl, 2015; Kutluturkan, Sozeri, Uysal, & Bay, 2016). In fact, caregivers with high levels of mental and physical exhaustion or trauma may have developed resilience to manage suffering.

With reference to the personal characteristics of the caregiver, our results suggest that older subjects are more exposed to burnout or STS, consistent with the research conducted by Craig & Sprang (2010). This data highlights the possibility that the burden of nursing will become increasingly

exhausting over time, in contrast to younger subjects who instead are gratified by their work, feeling satisfied and effective.

In fact, age seems to have a significant impact on the cost of care perceived by the caregiver, who feels ever-increasing time restrictions, the lack of social opportunities and a chronic feeling of tiredness (Bharti & Bhatnagar, 2017). Even the increase in working shifts, expressed in terms of hours, days and years, are perceived in an increasingly burdensome way and faced with suppressive mechanisms. In this sense, older caregivers mostly adopt Regression in the Service of the Ego to get away from a traumatic context while younger caregivers use the Repressive Function to banish distressing thoughts (Settineri, Frisone, Alibrandi, Pino, Lupo, & Merlo, 2018).

Our analysis also showed that intensive professional training could instead be a protective factor for the caregiver, who seems to be less exposed to burden and less inclined to use suppressive mechanisms.

With reference to gender, it seems that female caregivers use more suppressive mechanisms, such as Rationalization and Repressive Function, in contrast to male caregivers who take refuge in imagination to face the stress deriving, in particular, from the perception of a high conflict between their professional and family/social role.

In women there are also higher levels of resilience, probably also due to a greater exposure to Secondary Traumatic Stress, in line with research by Cieslak and co-workers (2014). In fact, female caregivers in close contact with suffering tend to develop resources that allow them to face adverse situations; females also seem to be supported by some protective factors, such as a better self-perception, an orientation towards the future and the existence of supportive family values.

## 5. Conclusions

Overall, our study highlights the numerous factors that can affect the caregiver's well-being and the strategies adopted to face and deal with traumatic conditions. In particular, the protective role of resilience was highlighted, which can help the subject to manage the burden deriving from the healthcare of patients. Even a moderate use of psychodynamic defense mechanisms can temporarily help the subject to cope with stressful situations but their prolonged use can lead to maladjustment, burnout and Secondary Traumatic Stress, which occur mainly in older caregivers and with the increase in work shifts.

These constructs, which are especially important in the context of the current health emergency (Di Giuseppe, Miniati, Miccoli, Ciacchini, Orrù, Sterzo *et al.*, 2020), highlight the risks to which operators are even more exposed, considering the high probability of incurring in chronic diseases with serious consequences also on their nursing ability (Conversano, 2019; Martino, Langher, Cazzato, & Vicario, 2019). The caregiver can be considered the “backbone” of the healthcare system and, as such, it is necessary to provide integrated preventive or clinical interventions, which offer a deep elaboration of the inner world (Merlo, 2019b) and help him/her to manage suffering efficiently and effectively.

## References

- Aldrich, N. (2011). CDC seeks to protect health of family caregivers. National Association of Chronic Disease (CDC). Retrieved from: [https://www.chronicdisease.org/resource/resmgr/healthy\\_aging\\_critical\\_issues\\_brief/ha\\_cib\\_healthoffamilycaregiv.pdf](https://www.chronicdisease.org/resource/resmgr/healthy_aging_critical_issues_brief/ha_cib_healthoffamilycaregiv.pdf).
- Allday, R. A., Newell, J. M., & Sukovskyy, Y. (2020). Burnout, compassion fatigue and professional resilience in caregivers of children with disabilities in Ukraine. *European Journal of Social Work*, 23 (1), 4-17. <https://doi.org/10.1080/13691457.2018.1499611>.
- Beaumont, E., Durkin, M., Martin, C. J. H., & Carson, J. (2016). Compassion for others, self-compassion, quality of life and mental well-being measures and their association with compassion fatigue and burnout in student midwives: A quantitative survey. *Midwifery*, 34, 239-244. <https://doi.org/10.1016/j.midw.2015.11.002>.
- Benau, K. (2020). Shame, Pride and Dissociation: Estranged Bedfellows, Close Cousins and Some Implications for Psychotherapy with Relational Trauma Part I: Phenomenology and Conceptualization. *Mediterranean Journal of Clinical Psychology*, 8 (1), 1-35. <https://doi.org/10.6092/2282-1619/mjcp-2155>.
- Bharti, J., & Bhatnagar, P. (2017). Personality and creativity as predictors of psychological well-being in caregivers of person with chronic mental illness. *Indian Journal of Positive Psychology*, 8 (2), 148-153.

Bonanno, G. A. (2012). Uses and abuses of the resilience construct: Loss, trauma, and health-related adversities. *Social Science and Medicine*, 74 (5), 753-756. <https://doi.org/10.1016/j.socscimed.2011.11.022>.

Burnett Jr, H. J., & Wahl, K. (2015). The compassion fatigue and resilience connection: A survey of resilience, compassion fatigue, burnout, and compassion satisfaction among trauma responders. <https://doi.org/10.4172/1522-4821.1000165>.

Capanna, C., Stratta, P., Hjemdal, O., Collazzoni, A., & Rossi, A. (2015). The Italian validation study of the resilience scale for adults (RSA). *BPA-Applied Psychology Bulletin*, 63 (272), 16-24.

Caputo, A. (2013). Health demand in primary care context: What do people think about physicians? *Psychology, Health & Medicine*, 18 (2), 145-154. <https://doi.org/10.1080/13548506.2012.687828>.

Caputo, A. (2019). Psychodynamic insights from narratives of people with amyotrophic lateral sclerosis: A qualitative phenomenological study. *Mediterranean Journal of Clinical Psychology*, 7 (2). doi: 10.6092/2282-1619/2019.7.2009.

Carrozzino, D., Costabile, A., Patierno, C., Settineri, S., & Fulcheri, M. (2019). Clinical Psychology in School and Educational Settings: Emerging Trends. *Mediterranean Journal of Clinical Psychology*, 7 (1). doi: 10.6092/2282-1619/2019.7.2138.

Catalano, A., Sardella, A., Bellone, F., Lasco, C. G., Martino, G., & Morabito, N. (2019). Executive functions predict fracture risk in postmenopausal women assessed for osteoporosis. *Aging Clinical and Experimental Research*, 32 (11), 2251-2257.

Cieslak, R., Shoji, K., Douglas, A., Melville, E., Luszczynska, A., & Benight, C. C. (2014). A meta-analysis of the relationship between job burnout and secondary traumatic stress among workers with indirect exposure to trauma. *Psychological Services*, 11 (1), 75-86. <https://doi.org/10.1037/a0033798>.

Collins, S., & Long, A. (2003). Working with the psychological effects of trauma: consequences for mental health-care workers—a literature review. *Journal of Psychiatric and Mental Health Nursing*, 10 (4), 417-424. <https://doi.org/10.1046/j.1365-2850.2003.00620.x>.

Collins, L. G., & Swartz, K. (2011). Caregiver care. *American Family Physician*, 83 (11), 1309-1317.

Conti, A., Clari, M., Garrino, L., Maitan, P., Scivoletto, G., Cavallaro, L., Bandini, B., Mozzone, S., Vellone, E., & Frigerio, S. (2019). Adaptation and validation of the Caregiver Burden Inventory in Spinal Cord Injuries (CBI-SCI). *Spinal Cord*, 57 (1), 75-82. <https://doi.org/10.1038/s41393-018-0179-7>.

Conversano, C. (2019). Psychological common factors in chronic diseases. *Frontiers in Psychology*, 10: 2727. <https://doi.org/10.3389/fpsyg.2019.02727>.

Craig, C. D., & Sprang, G. (2010). Compassion satisfaction, compassion fatigue, and burnout in a national sample of trauma treatment therapists. *Anxiety, Stress, & Coping*, 23 (3), 319-339. <https://doi.org/10.1080/10615800903085818>.

Cramer, P. (1998). Coping and defense mechanisms: What's the difference? *Journal of Personality*, 66 (6), 919-946. <https://doi.org/10.1111/1467-6494.00037>.

David, D. (2012). *Resilience as a protective factor against compassion fatigue in trauma therapist* (Unpublished doctoral dissertation). Walden University, Minneapolis, MN.

Deburge, A. (2001). La levée de la répression en psychosomatique. *Revue Française de Psychanalyse*, 65 (1), 11-27. <https://doi.org/10.3917/rfp.651.0011>.

Di Giuseppe, M., Ciacchini, R., Micheloni, T., Bertolucci, I., Marchi, L., & Conversano, C. (2018). Defense mechanisms in cancer patients: A systematic review. *Journal of Psychosomatic Research*, 115, 76-86. <https://doi.org/10.1016/j.jpsychores.2018.10.016>.

- Di Giuseppe, M., Ciacchini, R., Piarulli, A., Nepa, G., & Conversano, C. (2019). Mindfulness dispositions and defense style as positive responses to psychological distress in oncology professionals. *European Journal of Oncology Nursing*, 40, 104-110. <https://doi.org/10.1016/j.ejon.2019.04.003>.
- Di Giuseppe, M., Di Silvestre, A., Lo Sterzo, R., Hitchcott, P., Gemignani, A., & Conversano, C. (2019). Qualitative and quantitative analysis of the defensive profile in breast cancer women: A pilot study. *Health Psychology Open*, 6 (1). <https://doi.org/10.1177/2055102919854667>.
- Di Giuseppe, M., Miniati, M., Miccoli, M., Ciacchini, R., Orrù, G., Sterzo, R. L., Di Silvestre, A., & Conversano, C. (2020). Defensive responses to stressful life events associated with cancer diagnosis. *Mediterranean Journal of Clinical Psychology*, 8 (1). <https://doi.org/10.6092/2282-1619/mjcp-2384>.
- Duarte, J., & Pinto-Gouveia, J. (2017). The role of psychological factors in oncology nurses' burnout and compassion fatigue symptoms. *European Journal of Oncology Nursing*, 28, 114-121. <https://doi.org/10.1016/j.ejon.2017.04.002>.
- Elwood, L. S., Mott, J., Lohr, J. M., & Galovski, T. E. (2011). Secondary trauma symptoms in clinicians: A critical review of the construct, specificity, and implications for trauma-focused treatment. *Clinical Psychology Review*, 31 (1), 25-36. <https://doi.org/10.1016/j.cpr.2010.09.004>.
- Figley, C. R. (1995). Compassion fatigue as secondary traumatic stress disorder: An overview. In C. R. Figley (Ed.), *Brunner/Mazel psychological stress series, No. 23. Compassion fatigue: Coping with secondary traumatic stress disorder in those who treat the traumatized* (p. 1-20). Brunner/Mazel.
- Figley, C. R. (2002). Compassion fatigue: Psychotherapists' chronic lack of self care. *Journal of Clinical Psychology*, 58 (11), 1433-1441. <https://doi.org/10.1002/jclp.10090>.
- Flarity, K., Gentry, J. E., & Mesnikoff, N. (2013). The effectiveness of an educational program on preventing and treating compassion fatigue in emergency nurses. *Advanced Emergency Nursing Journal*, 35 (3), 247-258. <https://doi.org/10.1097/TME.0b013e31829b726f>.

Friborg, O., Barlaug, D., Martinussen, M., Rosenvinge, J. H., & Hjemdal, O. (2005). Resilience in relation to personality and intelligence. *International Journal of Methods in Psychiatric Research*, 14, 29-42. <https://doi.org/10.1002/mpr.15>.

Friborg, O., Hjemdal, O., Rosenvinge, J. H., & Martinussen, M. (2003). A new rating scale for adult resilience: What are the central protective resources behind healthy adjustment? *International Journal of Methods in Psychiatric Research*, 12, 65-76. <https://doi.org/10.1002/mpr.143>.

Gito, M., Ihara, H., & Ogata, H. (2013). The relationship of resilience, hardiness, depression and burnout among Japanese psychiatric hospital nurses. *Journal of Nursing Education and Practice*, 3 (11), 12-18. <https://doi.org/10.5430/jnep.v3n11p12>.

Haley, W. E., LaMonde, L. A., Han, B., Burton, A. M., & Schonwetter, R. (2003). Predictors of depression and life satisfaction among spousal caregivers in hospice: application of a stress process model. *Journal of Palliative Medicine*, 6 (2), 215-224. doi: <https://doi.org/10.1089/109662103764978461>.

Hamilton, S., Tran, V., & Jamieson, J. (2016). Compassion fatigue in emergency medicine: the cost of caring. *Emergency Medicine Australasia*, 28 (1), 100-103. <https://doi.org/10.1111/1742-6723.12533>.

Hjemdal, O., Friborg, O., Martinussen, M., & Rosenvinge, J. H. (2001). Preliminary results from the development and validation of a Norwegian scale for measuring adult resilience. *Journal of Norwegian Psychological Assessment*, 38, 310-317.

Hjemdal, O., Friborg, O., Stiles, T. C., Rosenvinge, J. H., & Martinussen, M. (2006). Resilience predicting psychiatric symptoms: A prospective study of protective factors and their role in adjustment to stressful life events. *Clinical Psychology & Psychotherapy*, 13, 194-201. <https://doi.org/10.1002/cpp.488>.

Hyphantis, T. N., Tomenson, B., Bai, M., Tsianos, E., Mavreas, V., & Creed, F. (2010). Psychological distress, somatization, and defense mechanisms associated with quality of life in inflammatory bowel disease patients. *Digestive Diseases and Sciences*, 55 (3), 724-732. <https://doi.org/10.1007/s10620-009-0762-z>.

Hoening, J., & Hamilton, M. W. (1966). The schizophrenic patient in the community and his effect on the household. *International Journal of Social Psychiatry*, 12 (3), 165-176.

Horowitz, M. J. (1993). Stress-response syndromes. A review of posttraumatic stress and adjustment disorders. In J. P. Wilson & B. Raphael (Eds.), *International handbook of traumatic stress syndromes. The Plenum Series on Stress and Coping* (pp. 49-60). Boston, MA: Springer. [https://doi.org/10.1007/978-1-4615-2820-3\\_4](https://doi.org/10.1007/978-1-4615-2820-3_4).

Jackson, D., Firtko, A., & Edenborough, M. (2007). Personal resilience as a strategy for surviving and thriving in the face of workplace adversity: a literature review. *Journal of Advanced Nursing*, 60 (1), 1-9. <https://doi.org/10.1111/j.1365-2648.2007.04412.x>.

Kosberg, J. I., Cairl, R. E., & Keller, D. M. (1990). Components of burden: Interventive implications. *The Gerontologist*, 30 (2), 236-242. <https://doi.org/10.1093/geront/30.2.236>.

Kutluturkan, S., Sozeri, E., Uysal, N., & Bay, F. (2016). Resilience and burnout status among nurses working in oncology. *Annals of General Psychiatry*, 15 (1): 33. <https://doi.org/10.1186/s12991-016-0121-3>.

Lim, W. S., Cheah, W. K., Ali, N., Han, H. C., Anthony, P. V., Chan, M., & Chong, M. S. (2014). Worry about performance: a unique dimension of caregiver burden. *International Psychogeriatrics*, 26 (4), 677-686. <https://doi.org/10.1017/S1041610213002445>.

Luthar, S. S., Cicchetti, D., & Becker, B. (2000). The construct of resilience: A critical evaluation and guidelines for future work. *Child Development*, 71 (3), 543-562. <https://doi.org/10.1111/1467-8624.00164>.



Lynch, S. H., Shuster, G., & Lobo, M. L. (2018). The family caregiver experience—examining the positive and negative aspects of compassion satisfaction and compassion fatigue as caregiving outcomes. *Aging & Mental Health*, 22 (11), 1424-1431. doi: <https://doi.org/10.1080/13607863.2017.1364344>.

Manfredi, P. (2020). Anger: what we know and what we don't want to know. Scientific contributions and social representation. *Mediterranean Journal of Clinical Psychology*, 8 (1).

Martino, G., Langher, V., Cazzato, V., & Vicario, C. M. (2019). Psychological factors as determinants of medical conditions. *Frontiers in Psychology*, 10: 2502. doi: <https://doi.org/10.3389/fpsyg.2019.02502>.

Maslach, C., Schaufeli, W. B., & Leiter, M. P. (2001). Job burnout. *Annual Review of Psychology*, 52 (1), 397-422. <https://doi.org/10.6092/2282-1619/mjcp-2326>.

McCloskey, S., & Taggart, L. (2010). How much compassion have I left? An exploration of occupational stress among children's palliative care nurses. *International Journal of Palliative Nursing*, 16 (5), 233-240. <https://doi.org/10.12968/ijpn.2010.16.5.48144>.

Meier, D. E., Back, A. L., & Morrison, R. S. (2001). The inner life of physicians and care of the seriously ill. *Jama*, 286 (23), 3007-3014. [10.1001/jama.286.23.3007](https://doi.org/10.1001/jama.286.23.3007).

Merlo, E. M. (2019a). Adolescent phobia as a “mask object”. *Mediterranean Journal of Clinical Psychology*, 7 (1). <https://doi.org/10.6092/2282-1619/2019.7.2241>.

Merlo, E. M. (2019b). Opinion Article: The role of psychological features in chronic diseases, advancements and perspectives. *Mediterranean Journal of Clinical Psychology*, 7 (3). <https://doi.org/10.6092/2282-1619/2019.7.2341>.

Merlo, E. M., McNabney, S. M., Frisone, F., Sicari, F., Paunica, M., Motofei, C., & Settineri, S. (2020). Compassion and suppression in caregivers: twin masks of tragedy and joy of caring. *Journal of Mind and Medical Sciences*, 7 (1), 61-68. doi: [10.22543/7674.71.P6168](https://doi.org/10.22543/7674.71.P6168).

Merlo, E. M., Stoian, A. P., Motofei, I. G., & Settineri, S. (2020). Clinical Psychological Figures in Healthcare Professionals: Resilience and Maladjustment as the “Cost of Care”. *Frontiers in Psychology, 11*: 607783. <https://doi.org/10.3389/fpsyg.2020.607783>.

Metzger, J. A. (2014). Adaptive defense mechanisms: function and transcendence. *Journal of Clinical Psychology, 70* (5), 478-488. <https://doi.org/10.1002/jclp.22091>.

Neumann, M., Edelhäuser, F., Tauschel, D., Fischer, M. R., Wirtz, M., Woopen, C., Haramati, A., & Scheffer, C. (2011). Empathy decline and its reasons: a systematic review of studies with medical students and residents. *Academic Medicine, 86* (8), 996-1009. <https://doi.org/10.1097/ACM.0b013e318221e615>.

Novak, M., & Guest, C. (1989). Application of a multidimensional caregiver burden inventory. *The Gerontologist, 29* (6), 798-803. <https://doi.org/10.1093/geront/29.6.798>.

Olson, K., Kemper, K. J., & Mahan, J. D. (2015). What factors promote resilience and protect against burnout in first-year pediatric and medicine-pediatric residents? *Journal of Evidence-based Complementary & Alternative Medicine, 20* (3), 192-198. <https://doi.org/10.1177/2156587214568894>.

Palacio, C., Krikorian, A., & Limonero, J. T. (2018). The influence of psychological factors on the burden of caregivers of patients with advanced cancer: Resiliency and caregiver burden. *Palliative & Supportive Care, 16* (3), 269-277. doi: <https://doi.org/10.1017/S1478951517000268>.

Palestini, L., Prati, G., Pietrantoni, L., & Cicognani, E. (2009). La qualità della vita professionale nel lavoro di soccorso: Un contributo alla validazione italiana della Professional Quality of Life Scale (ProQOL). *Psicoterapia Cognitiva e Comportamentale, 15* (2), 205-227.

Parker Oliver, D., Patil, S., Benson, J. J., Gage, A., Washington, K., Kruse, R. L., & Demiris, G. (2017). The effect of internet group support for caregivers on social support, self-efficacy, and caregiver burden: a meta-analysis. *Telemedicine and e-Health*, 23 (8), 621-629. <https://doi.org/10.1089/tmj.2016.0183>.

Pinquart, M., & Sörensen, S. (2007). Correlates of physical health of informal caregivers: a meta-analysis. *The Journals of Gerontology Series B: Psychological Sciences and Social Sciences*, 62 (2), 126-137. <https://doi.org/10.1093/geronb/62.2.P126>.

Platania, S., Di Nuovo, S., Caruso, A., Digrandi, F., & Caponnetto, P. (2020). Stress among university students: the psychometric properties of the Italian version of the SBI-U 9 scale for Academic Burnout in university students. *Health Psychology Research*, 8: 9209. <https://doi.org/10.4081/hpr.2020.9209>.

Price, M. L., Surr, C. A., Gough, B., & Ashley, L. (2020). Experiences and support needs of informal caregivers of people with multimorbidity: a scoping literature review. *Psychology & Health*, 35 (1), 36-69. <https://doi.org/10.1080/08870446.2019.1626125>.

Ray, S. L., Wong, C., White, D., & Heaslip, K. (2013). Compassion satisfaction, compassion fatigue, work life conditions, and burnout among frontline mental health care professionals. *Traumatology*, 19 (4), 255-267. <https://doi.org/10.1177/1534765612471144>.

Rosa, V., Tomai, M., Lauriola, M., & Martino, G. (2019). Body mass index, personality traits, and body image in Italian pre-adolescents: An opportunity for overweight prevention. *Psihologija*, 52 (4), 379-393.

Rushton, C. H., Batcheller, J., Schroeder, K., & Donohue, P. (2015). Burnout and resilience among nurses practicing in high-intensity settings. *American Journal of Critical Care*, 24 (5), 412-420. <https://doi.org/10.4037/ajcc2015291>.

Sabin-Farrell, R., & Turpin, G. (2003). Vicarious traumatization: implications for the mental health of health workers? *Clinical Psychology Review*, 23 (3), 449-480. [https://doi.org/10.1016/S0272-7358\(03\)00030-8](https://doi.org/10.1016/S0272-7358(03)00030-8).

Saeed, F., Salehi, M., Alavi, K., Ajdarkosh, H., Kashaninasab, F., & Esfahani, F. N. (2019). Defense Mechanisms in Patients with Irritable Bowel Syndrome and Their Relationship with Symptom Severity and Quality of Life. *Middle East Journal of Digestive Diseases*, 11 (3), 158-165. <https://doi.org/10.15171/mejdd.2019.143>.

Settineri, S., Frisone, F., Alibrandi, A., & Merlo, E. M. (2019). Vulnerability and physical well-being of caregivers: what relationship? *Journal of Mind and Medical Sciences*, 6 (1), 95-102. doi: 10.22543/7674.61.P95102.

Settineri, S., Frisone, F., Alibrandi, A., Pino, G., Lupo, N. J., & Merlo, E. M. (2018). Psychological Types and Learning Styles. *Mediterranean Journal of Clinical Psychology*, 6 (3). doi: 10.6092/2282-1619/2018.6.2005.

Settineri, S., Frisone, F., & Merlo, E. M. (2019). The Mask object in psychotherapy: Presentation and Representation. *Mediterranean Journal of Clinical Psychology*, 7 (1). <https://doi.org/10.6092/2282-1619/2019.7.2232>.

Settineri, S., Frisone, F., Merlo, E. M., Geraci, D., & Martino, G. (2019). Compliance, adherence, concordance, empowerment, and self-management: five words to manifest a relational maladjustment in diabetes. *Journal of Multidisciplinary Healthcare*, 12, 299-314. doi: 10.2147/JMDH.S193752.

Settineri, S., Merlo, E. M., Frisone, F., Alibrandi, A., Carrozzino, D., Diaconu, C. C., & Pappalardo, S. M. (2019). Suppression Mental Questionnaire App: a mobile web service-based application for automated real-time evaluation of adolescent and adult suppression. *Mediterranean Journal of Clinical Psychology*, 7 (1). doi: <https://doi.org/10.6092/2282-1619/2019.7.2056>.

Shanafelt, T. D., Bradley, K. A., Wipf, J. E., & Back, A. L. (2002). Burnout and self-reported patient care in an internal medicine residency program. *Annals of Internal Medicine*, 136 (5), 358-367. <https://doi.org/10.7326/0003-4819-136-5-200203050-00008>.

Sinclair, S., Raffin-Bouchal, S., Venturato, L., Mijovic-Kondejewski, J., & Smith-MacDonald, L. (2017). Compassion fatigue: A meta-narrative review of the healthcare literature. *International Journal of Nursing Studies*, 69, 9-24. <https://doi.org/10.1016/j.ijnurstu.2017.01.003>.

Stamm, B. H. (2005). *The ProQOL manual: The professional quality of life scale: Compassion satisfaction, burnout & compassion fatigue/secondary trauma scales*. Baltimore, MD: Sidran.

Tauschke, E., Merskey, H., & Helmes, E. (1990). Psychological defence mechanisms in patients with pain. *Pain*, 40 (2), 161-170. [https://doi.org/10.1016/0304-3959\(90\)90067-N](https://doi.org/10.1016/0304-3959(90)90067-N).

Velutti, L., Pavesi, C., Poggio, C., Carettoni, B., Saetta, A., Arcanà, C., De Maio, A. P., Lopane, D., Speciale, T., Mazziere, S., Rimassa, L., & Santoro, A. (2017). Caregiver stress: Clinical evaluation and intervention strategies for caregiver burden prevention. *Journal of Clinical Oncology* 2017 35: 31\_suppl, 22-22. [https://doi.org/10.1200/JCO.2017.35.31\\_suppl.22](https://doi.org/10.1200/JCO.2017.35.31_suppl.22).

Widowati, S., Devi, S. R., & Nihayati, H. E. (2018). Caregiver Burden, Quality of Life and Empowerment Intervention in Caregiver Family with Schizophrenia. In *Proceedings of the 9th International Nursing Conference – INC* (pp. 547-551). doi: 10.5220/0008328305470551.

Win, K. K., Chong, M. S., Ali, N., Chan, M., & Lim, W. S. (2017). Burden among Family caregivers of Dementia in the Oldest-Old: an exploratory study. *Frontiers in Medicine*, 4: 205. <https://doi.org/10.3389/fmed.2017.00205>.

Wong, D. F. K., Lam, A. Y. K., Chan, S. K., & Chan, S. F. (2012). Quality of life of caregivers with relatives suffering from mental illness in Hong Kong: roles of caregiver characteristics, caregiving burdens, and satisfaction with psychiatric services. *Health and Quality of Life Outcomes*, 10 (1): 15. doi: <https://doi.org/10.1186/1477-7525-10-15>.

Wood, R., Taylor-Stokes, G., Smith, F., Chirita, O. C., & Chaib Torralba, C. (2018). The humanistic burden of advanced non-small cell lung cancer: What are the key drivers of caregiver burden? *Journal of Clinical Oncology*, 36 (7): 149-149. doi: 10.1200/JCO.2018.36.7\_suppl.149.



# The link between apathy and Alzheimer's Disease: The role of psychometric tools and the possible implications for treatment

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## Abstract

*Alzheimer's Disease (AD) represents a critical challenge because of its increasing neuropsychological impairment with progressive cognitive decline accompanied by Behavioral and Psychological Symptoms of Dementia (BPSD) in almost 90% of patients. BPSD represent relevant clinical problems, leading to a worsening in the general conditions of patients. More specifically, apathy is often associated with a poor response to treatment, a faster cognitive and functional decline and an increased mortality rate.*

*Apathy can be considered as a common symptom of AD and as an early marker of cognitive decline and transition from mild cognitive impairment to dementia. Recent studies have shown different neurobiological and clinical links between apathy and AD. Evidence discussed in the present article suggests a strong clinical link between apathy and AD as well as the relevance of psychometric tools, such as the Apathy Evaluation Scale (AES), to better diagnose and treat apathy in patients with AD. The aim of this review was thus to provide a general overview of the neurobiological and clinical links between apathy and*

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*AD, with the purpose of evaluating the impact of apathy on the health of AD patients, focusing on the role of psychometric tools and the possible implications for treatment. A multimodal intervention should be promoted as an innovative approach for the future treatment of apathetic AD patients.*

**Keywords:** Apathy; Alzheimer; Psychometric tools; Treatment.



## 1. Introduction

Alzheimer's Disease (AD) can be defined as a progressive and remediless neurodegenerative disease that affects an increasing number of subjects over the age of 65 (Albert, 2007). It is the most common form of dementia (Herrera, Caramelli, Silveira, & Nitrini, 2002; Hirtz, Thurman, Gwinn-Hardy, Mohamed, Chaudhuri, & Zalutsky, 2007) and in recent years it has been demonstrated (Isaacson, Ganzer, Hristov, Hackett, Caesar, Cohen *et al.*, 2018) that late-onset AD begins decades before a diagnosis, with a long preclinical and prodromal phase often beginning in midlife (Frisoni, Winblad, & O'Brien, 2011). AD represents a critical challenge to the health care system with the aging population (Robert, Ferris, Gauthier, Ihl, Winblad, & Tennigkeit, 2010) because of its increasing neuropsychological impairment with a progressive decline of memory, language, executive function and visuospatial skills (Cummings & McPherson, 2001). The characteristics of the disease, of cognitive and functional decline, are accompanied by Behavioral and Psychological Symptoms of Dementia (BPSD) in almost 90% of patients (Guimarães, Levy, Teixeira, Beato, & Caramelli, 2008). BPSD are symptoms of disturbed perception, thought content, mood, and behavior that occur frequently in patients with dementia; they are common precipitators of institutional care

(Mitchell, Herrmann, & Lanctôt, 2011) and significantly contribute to decreasing the quality of life of caregivers (Lyketsos, Lopez, Jones, Fitzpatrick, Breitner, & Dekosky, 2002; Banerjee, Smith, Lamping, Harwood, Foley, Smith *et al.*, 2006). BPSD, including apathy, agitation, delusion, irritability, anxiety, disinhibition and hallucinations (Cummings & McPherson, 2001), represent significant clinical problems, leading to an accelerated functional decline, the distress of caregivers and aggression towards patients and possibly to an increased mortality rate, as reviewed by Guimarães and colleagues (2008).

Apathy can be defined as a specific dimension of behavior, characterized by a lack of motivation (Marin, Firinciogullari, & Biedrzycki, 1993), in which motivation is a superordinate concept referring to the psychological structure and the determinants of goal-directed behavior (Bindra, 1959; Madsen, 1973). Apathy is often associated with a poor response to treatment, the reliance on caregivers to start activities, a more rapid cognitive and functional decline and an increased mortality rate (Ruslan, Teerenstra, Smalbrugge, Vernooij-Dassen, Bohlmeijer, Gerritsen *et al.*, 2013). For clinical purposes apathy is often considered as a symptom of

other syndromes, such as depression or dementia (Marin *et al.*, 1993). However, it can be considered as a syndrome in itself and conceptualized as a syndrome of reduced motivation in which the lack of motivation is not attributable to a reduced level of consciousness, emotional distress, or cognitive deficits (Marin, 1991). Apathy is a multidimensional construct that can include both the emotional and the social sphere and presents three separate dimensions consisting of cognitive, affective and behavioral symptoms (Robert, Onyike, Leentjens, Dujardin, Aalten, Starkstein *et al.*, 2009) although there is heterogeneity in the degree in which each dimension could be present. The prevalence of apathy is well documented in different neuropsychiatric disorders (Padala, Frederick, & Subhash, 2005), especially in Mild Cognitive Impairment (MCI) and AD. In fact, from an epidemiological point of view, apathy can be considered as a common symptom of AD (Rea, Carotenuto, Fasanaro, Traini, & Amenta, 2014). Recent studies have shown that the higher the level of apathy, the more it is a predictor of the transition to dementia (Lanctôt, 2017). Most of the epidemiological studies on apathy in the context of AD refer to studies on the prevalence or change of apathy symptoms over time (Brodaty, Altendorf, Withall, & Sachdev, 2010). Apathy is predominant both in MCI and dementia and its prevalence is positively related to the severity of dementia (Lanctôt, 2017). It can determine high levels of distress in caregivers (Samus, Rosenblatt, Steele, Baker, Harper, Brandt *et al.*, 2005) and, as in the case with AD, it can provoke several conflicts and unpleasant emotions, like anger and exhaustion, between patients and caregivers. On the basis of this evidence apathy can be considered as an early marker of cognitive decline and transition to dementia. This has prompted the field to flag it as a high-value neuropsychiatric state of risk, as has been highlighted in the latest guidelines for preclinical AD from the National Institute on Aging (Sperling, Aisen, Beckett, Bennett, Craft, Fagan *et al.*, 2011). But already in 2008, The European Alzheimer's Disease Consortium had drawn guidelines for the diagnosis of apathy (Winbald, Frisoni, Frolich, Johannsen, Johansson, Kehoe *et al.*, 2008). To make a correct diagnosis of apathy the reduced motivation clinical picture must persist for no less than four weeks, and two of the following three dimensions should be present: (I) reduced goal-directed behavior, (II) reduced goal-directed cognitive activity, and (III) reduced emotions. In a recent work, Robert and collaborators (Robert, Lanctôt, Agüera-Ortiz, Aaltend, Bremonda, Defrancesco *et al.*, 2018) proposed new diagnostic criteria for apathy to be adopted both in the clinical and the research domain (see Tab. 1).

Table 1 – *New diagnostic criteria for apathy in both the clinical and research domain (Robert et al., 2018)*

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CRITERION A: A quantitative reduction of goal-directed activity either in the behavioral, cognitive, emotional or social dimensions in comparison to the patient's previous level of functioning in these areas. These changes may be reported by the patient him/herself or by the observation of others.

CRITERION B: The presence of at least 2 of the 3 following dimensions for a period of at least four weeks and present most of the time.

1. B1. BEHAVIOR & COGNITION: Loss of, or reduced, goal-directed behavior or cognitive activity as revealed by at least one of the following:
    - General level of activity: the patient has a reduced level of activity either at home or at work, he/she makes less effort to initiate or accomplish tasks spontaneously or needs to be prompted to perform them.
    - Persistence of activity: the patient is less persistent in maintaining an activity or conversation, finding solutions to problems or thinking of alternative ways to accomplish them if they become difficult.
    - Making choices: the individual has less interest or takes longer to make choices when different alternatives are presented (e.g., selecting TV programs, preparing meals, choosing from a menu, etc.).
    - Interest in external issues: the subject has less interest in or reacts less to news, either good or bad, or has less interest in doing new things.
    - Personal well-being: the patient is less interested in his/her own health and well-being or personal image (general appearance, grooming, clothes, etc.).
  2. B2. EMOTION: Loss of, or reduced, emotion as revealed by at least one of the following:
    - Spontaneous emotions: the patient shows less spontaneous (self-generated) emotions regarding his/her own affairs, or appears less interested in events that should matter to him/her or to people that he/she knows well.
    - Emotional reactions to the environment: the subject expresses less emotional reaction in response to positive or negative events in his/her environment that affect him/her or people he/she knows well (e.g., when things go well or bad, responding to jokes, or events on a TV program or a movie, or when disturbed or prompted to do things he/she would prefer not to do).
    - Impact on others: the individual is less concerned about the impact of his/her actions or feelings on the people surrounding him/her.
    - Empathy: the patient shows less empathy to the emotions or feelings of others (e.g., becoming happy or sad when someone is happy or sad, or being moved when others need help).
    - Verbal or physical expressions: the individual shows less verbal or physical reactions that reveal his/her emotional states.
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3. B3. SOCIAL INTERACTION: Loss of, or reduced, engagement in social interaction as highlighted by at least one of the following:
- Spontaneous social initiative: the patient takes less initiative in spontaneously proposing social or leisure activities to family or others.
  - Environmentally-stimulated social interaction: the individual participates less, or is less comfortable or more indifferent, to social or leisure activities suggested by people around him/her.
  - Relationship with family members: the subject shows less interest in the family members (e.g., to know what is happening to them, to meet them or to make arrangements to contact them).
  - Verbal interaction: the patient is less likely to initiate a conversation, or he/she immediately withdraws from it.
  - Homebound: the individual prefers to stay at home more frequently or longer than usual and shows less interest in getting out to meet people.

CRITERION C: The symptoms (A - B) cause clinically significant impairment in personal, social, occupational, or other important areas of functioning.

CRITERION D: The symptoms (A - B) are not exclusively explained or due to physical disabilities (e.g. blindness and loss of hearing), to motor disabilities, to a reduced level of consciousness, to the direct physiological effects of a substance (e.g. drug abuse, medication), or to major changes in the patient's environment.

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The recent evidence thus suggests a key role of apathy in AD. Along this line the aim of this review was to provide a general overview of the neurobiological and clinical links between apathy and AD, with the purpose of giving the clinicians the opportunity to evaluate the impact of apathy on the health of AD patients, focusing on the role of psychometric tools and the possible implications for treatment.

## 2. Neurobiological links between apathy and Alzheimer's Disease

In recent years, apathy has been deeply investigated, due to its high prevalence in neuropsychiatric disorders, especially in AD. Several studies

have demonstrated its anatomical and neuropsychological correlates, in particular through neuroimaging studies. An association was found, for example, between apathy and the dysfunction in both the dorsolateral and orbitofrontal areas of the prefrontal regions and sub-regions within the basal ganglia (Eslinger, Moore, Antani, Anderson, & Grossman, 2012). In particular, an association between apathy and atrophy was revealed in AD concerning many different frontal regions, such as the anterior cingulate cortex (ACC) (Apostolova, Akopyan, Partiali, Steiner, Dutton, Hayashi *et al.*, 2007; Bruen, McGeown, Shanks, & Venneri, 2008; Grambaite, Selnes, Reinvang, Aarsland, Hessen, Gjerstad *et al.*, 2011; Tunnard, Whitehead, Hurt, Wahlund, Mecocci, Tsolaki *et al.*, 2011; Mori, Shimada, Shinotoh, Hirano, Eguchi, Yamada *et al.*, 2014), the medial frontal cortex (Apostolova *et al.*, 2007; Mori *et al.*, 2014), the orbitofrontal cortex (Holthoff, Beuthien-Baumann, Kalbe, Lüddecke, Lenz, Zündorf *et al.*, 2005; Tunnard *et al.*, 2011; Mori *et al.*, 2014), the pars triangularis and insula (Tunnard *et al.*, 2011; Moon, Moon, Kim, & Han, 2014; Mori *et al.*, 2014), the lower inferior temporal cortical thickness (Donovan, Wadsworth, Lorus, Locascio, Rentz, Johnson *et al.*, 2014; Guercio, Donovan, Ward, Schultz, Lorus, Amariglio *et al.*, 2015), as well as the fronto-parietal control network connectivity (Munro, Donovan, Guercio, Wigman, Schultz, Amariglio *et al.*, 2015). Moreover, as recently reviewed by Starkstein and Brockman (2018), the decreased density of grey matter in the right superior frontal gyrus, in the bilateral middle and inferior frontal gyrus, the ACC and the basal ganglia (bilateral putamen and head of the caudate) has found to be correlated to an increased risk to develop apathy. These areas are critically involved in affective modulation and in sensory and emotional information processing (Tekin & Cummings, 2002; Levy & Dubois, 2006). A great attention has been given to the anterior cingulate cortex because it is involved in the systems of motivation and reward as a part of a frontal striatal circuit; moreover, it has been demonstrated that this circuit is correlated with symptoms of apathy (Tekin & Cummings, 2002; Blundo & Gerace, 2015). Levy and Dubois (2006) examined the correlation between the dimensions of apathy (emotional, affective, cognitive and auto-activation) and the anatomical regions of the brain. They found that three different areas of the brain could be attributed to their physiopathology; emotional-affective apathy was found to be typically associated with the damage of circuits linking the orbito-medial prefrontal and limbic cortex to the basal ganglia. Cognitive apathy was, instead, generally correlated to the dysfunction of the dorsolateral prefrontal cortex and related portions within the basal ganglia,

while auto-activation apathy was related to the associative and limbic territories of the internal portion of the globus pallidus.

Finally, the hypofunction of the dopaminergic system should also be taken into consideration as a relevant neurobiological link between apathy and AD. As reviewed by Mitchell and colleagues (2011), in AD patients there seems to be a correlation between the pathophysiological changes to the DAergic neurons in the reward system and the onset of apathy. Indeed, DA mediates feelings of motivation, but it has been shown that there is a dysfunction of the dopaminergic system at an early stage of the pathogenesis of AD with a central role in the pathophysiology of memory deficits and apathy in AD (D'Amelio, Puglisi-Allegra, & Mercuri, 2018).

### 3. Psychometric tools to evaluate apathy in AD

Whereas apathy can be considered as a preclinical manifestation of the disease and AD patients can have a more rapid cognitive decline (Starkstein, Jorge, Mizrahi, & Robinson, 2006) and a more rapid loss of autonomy in the activities of daily life, a rapid recognition and diagnosis are needed to plan an appropriate treatment. An accurate assessment is essential to improve the management of apathy and its associated symptoms and conditions. Several psychometric tools for apathy are present in the literature, but we will describe the ones, which are most widely adopted (Clarke, Ko, Kuhl, van Reekum, Salvador, & Marin, 2011) and that have been validated on the AD population (Radakovic, Harley, Abrahams, & Starr, 2015; Mohammad, Ellis, Rau, Rosenberg, Mintzer, Ruthirakuhan *et al.*, 2018; see Tab. 2 for details).

*Table 2 – Selection of validated psychometric tools to assess apathy in AD*

Full name	Abbreviation	Item n.	Authors
Apathy Evaluation Scale	AES (AES-C; AES-I; AES-S)	18	Marin, 1991
Apathy Scale	AS	14	Starkstein, 1992, 1995
Short Version Of The Apathy Evaluation Scale		10	Lueken <i>et al.</i> , 2007
Neuropsychiatric Inventory – Apathy Subscale	NPI – Apathy Subscale	8	Cummings <i>et al.</i> , 1994
Apathy Inventory	AI	3	Robert <i>et al.</i> , 2002
Dementia Apathy Interview and Rating	DAIR	16	Strauss & Sperry, 2002

The Apathy Evaluation Scale (AES) was developed by Marin and colleagues (Marin, Biedrzycki, & Firinciogullari, 1991), on the basis of the definition of apathy according to Marin, which describes a syndrome of loss of motivation as reflected by the acquired changes in affection (mood), behavior and cognition (Marin *et al.*, 1991). It is measured through a four-point Likert-scale, composed of 18 items that assess and quantify the emotional, behavioral and cognitive aspects of apathy. There are three different ways of measuring the scale: through a self-report (AES-S), an informant report (AES-I) or a clinician interview (AES-C). The AES-C, in particular, includes a semi-structured open-ended interview that helps the clinician to collect information from the patient concerning his/her typical day activities, hobbies and interests, which reveals the degree of the subject's motivation and directs the clinician in providing his/her own rating of the individual's level of apathy on each item. Every item is rated on a 4-point response scale (0 = not at all true/characteristic to 3 = very much true/characteristic). Higher scores indicate more severe apathy (Marin *et al.*, 1991). The AES-C version takes between 10-20 minutes to be completed (Marin *et al.*, 1991).

Different studies (Mohammad *et al.*, 2018) demonstrated good to excellent internal consistency of all the versions of the AES, varying the values for convergent validity and good discriminant validity. However, as Marin and colleagues noted (Marin, Biedrzycki, & Firinciogullari, 1991; see also Mohammad *et al.*, 2018), only the AES-C and AES-S versions were able to discriminate apathy from depression. Moreover, the AES has been employed in three Randomized Clinical Trials (RCTs) as a primary outcome in patients with apathy and AD (Herrmann, Rothenburg, Black, Ryan, Liu, Busto *et al.*, 2008; Rosenberg, Lanctôt, Drye, Herrmann, Scherer, Bachman *et al.*, 2013; Padala, Padala, Lensing, Ramirez, Monga, Bopp *et al.*, 2018).

Two different scales were developed on the basis of the first version of the AES. Starkstein and colleagues (Starkstein, Mayberg, Preziosi, Andrezejewski, Leiguarda, & Robinson, 1992) developed a 14-item Apathy Scale, which is a short and slightly modified version of Marin's AES, and they validated it on the stroke, Parkinson's and AD population (Starkstein, Migliorelli, Manes, Teson, Petracca, Chemerinski *et al.*, 1995; Starkstein, Jorge, & Mizrahi, 2006). Lueken and co-workers (Lueken, Seidl, Volker, Schweiger, Kruse, & Schroder, 2007), instead, developed an abbreviated version of Marin's instrument, reducing the items from 18 to 10. This scale demonstrated a moderate convergent validity with the Neuropsychiatric Inventory (NPI)-apathy and an excellent internal consistency (Mohammad *et*

*al.*, 2018). The aforementioned authors reduced the items considered to be redundant (Starkstein *et al.*, 1992) or inappropriate (Lueken *et al.*, 2007) according to experts with a special knowledge of the population being assessed. As with the original Apathy Evaluation Scale, each item is measured with a four-point Likert scale.

The NPI was developed by Cumming (Cummings, Mega, Gray, Rosenberg-Thompson, Carusi, & Gornbein, 1994; Cummings, 1997) as a multidimensional instrument to assess neurobehavioral disorders in dementia. The NPI has a specific 8-item subscale to measure apathy, which lies in a general screen item rated on a yes-versus-no basis. Its scores have a scale ranging from 0 to 12, and higher scores indicate a more severe presence of apathy (Cummings *et al.*, 1994; Cummings, 1997). The NPI and NPI-apaty subscale were used and validated on multicultural sample patients with dementia and AD (Clarke *et al.*, 2011).

The Apathy Inventory (AI) (Robert, Claret, Benoit, Koutaich, Bertogliati, Tible *et al.*, 2002) is based on Marin and colleagues' diagnostic criteria of apathy (Marin, 1991; Marin *et al.*, 1991), which means lack of emotional response, self-initiated actions, and interest in things. This is why the AI includes 3 different items for the assessment of emotional blunting, lack of initiative, and symptoms of loss of interest. The AI involves two versions: a clinician-administered interview to the caregiver (AI-caregiver) or the patient (AI-patient). The frequency is rated on a four-point Likert scale and the severity on a three-point Likert scale. The AI validation was carried out in a mixed sample consisting of normal controls, MCI patients and AD patients (Robert *et al.*, 2002). The internal consistency reported for the caregiver-based version is of good quality and the between-rater and test-retest reliability were excellent and demonstrated favorable values for all three items on the scale (Robert *et al.*, 2002; Mohammad *et al.*, 2018).

The Dementia Apathy Interview and Rating (DAIR) (Strauss & Sperry, 2002) is an informant-based unidimensional 16-item clinician-administrated scale to measure changes in motivation, engagement, and emotional response in MCI patients with apathy. The scale is administered to a caregiver using a structured interview with questions concerning apathy in the patient over the past month. Each item is rated on a 4-point scale (0 = no or almost never to 3 = yes or almost always) with higher scores corresponding to a greater severity in apathy. Only items representing a change in behavior are included in the final apathy score. The DAIR has very good internal consistency and the two-month test-retest reliability and



the inter-rater reliability also demonstrated to be very good (Strauss & Sperry, 2002).

The presence of valid and reliable apathy scales are essential to assess and plan treatment of the apathy syndrome in the AD population. As we have seen and thoroughly described, there are a lot of well-structured, valid and reliable psychometric tools for the evaluation of apathy that can be used in conjunction with other clinical tools of assessment and that provide altogether a more complex and complete clinical evaluation of the subject. However, a gold standard of apathy assessment is still lacking.

#### 4. Pharmacological treatment of apathy in AD

The pharmacological treatment of apathy appears to be essential to take care and manage AD patients, although it still represents an unmet clinical need. Different studies have investigated the role of different psychotropic drugs on the pharmacological treatment of apathy. Concerning donepezil and rivastigmine, evidence from Randomized Clinical Trials (RCTs) suggests that cholinesterase inhibitors can improve apathy (Winblad, Engedal, Soininen, Verhey, Waldemar, & Wimo, 1999; Cummings, Koumaras, Chen, & Mirski, 2005) and delay its onset (Waldemar, Gauthier, Jones, Wilkinson, Cummings, Lopez *et al.*, 2011). In a recent RCT, the authors compared the apathy scores of 113 mild to moderate AD patients treated for 24 months with donepezil plus a cholinergic precursor (choline alphoscerate) with those of patients who were administered to donepezil alone (Rea, Carotenuto, Traini, Fasanaro, Manzo, & Amenta, 2015). The results demonstrated that the combination of donepezil with choline alphoscerate was more effective than donepezil alone in reducing apathy in AD patients. Lopez and colleagues (Lopez, Mackell, Sun, Kassalow, Xu, McRae *et al.*, 2008) conducted a multi-center, open-label, 12-week study to evaluate the efficacy and safety of the administration of donepezil in 106 mild to moderate AD Hispanic patients treated with donepezil 5 mg/day for 6 weeks followed by 10 mg/day for another 6 weeks. The authors demonstrated that the NPI “apathy/indifference” subdomain showed a statistically significant improvement in donepezil-treated patients. Other studies confirmed that donepezil could improve apathy symptoms in patients with mild-severe AD (Drijgers, Aalten, Winogrodzka, Verhey, & Leentjens, 2009). A validation of the hypothesis that the rescue of the cholinergic system can be a useful approach for the treatment of apathy comes from other relevant studies conducted with cholinesterase inhibitors, such as

rivastigmine and galantamine. In an open-label, multi-center study (Gauthier, Juby, Dalziel, Rehel, Schechter, & EXPLORE Investigators, 2010) the efficacy of rivastigmine on apathy was evaluated in a sample of 4460 AD patients, demonstrating that the percentage of patients showing an improvement vs. a deterioration in apathy at month 6 was in the order of 42.8 vs. 7.2%, respectively. Galantamine can also be used for the treatment of apathy in AD. In 2004, Cummings and colleagues (Cummings, Schneider, Tariot, Kershaw, & Yuan, 2004) assessed the efficacy of galantamine at different doses (8, 16, or 24 mg/day) in a 21-week, multi-center RCT in 978 patients with mild to moderate AD. This study demonstrates a significantly less appearance of apathy in patients treated with galantamine and without specific behavioral symptoms. Monsch and colleagues (Monsch, Giannakopoulos, & GAL-SUI Study Group, 2004) investigated the effects of galantamine (escalated from 8 to 24 mg/day over 8 weeks) in a 3-month, open-label, multi-center study conducted on 124 AD patients. The authors demonstrated a 27% reduction in the apathy score, as assessed by the NPI at the end of the period of treatment. Freund-Levi and co-workers (Freund-Levi, Jedenius, Tysen-Backstrom, Larksater, Wahlund, & Eriksdotter, 2014) recruited subjects with diagnoses of probable dementia (88%) or MCI (12%) from a memory clinic. The aim of the randomized, open-label trial, was to explore the effects of the administration of galantamine and risperidone on the overall neuropsychiatric symptoms (NPS) and global function. Both galantamine and risperidone treatments were able to produce a small non-significant reduction of apathetic behaviors, as measured by the NPI-aphathy subscale, also over time.

In 2006 (Cummings, Schneider, Tariot, & Graham, 2006) an exploratory analysis of a 24-week, double-blind, placebo-controlled trial was conducted in order to compare memantine (20 mg/day) treatment with placebo in 404 moderate to severe AD patients, which were already treated with donepezil; the authors did not detect significant effects on apathy as assessed with the NPI, whereas an improvement in the NPI-aphathy score (-11.3%) was observed following treatment with memantine by Schmidt and colleagues (Schmidt, Baumhackl, Berek, Brücke, Kapeller, Lechner *et al.*, 2010) in a 16-week, open label study conducted on 53 AD patients.

Moreover, a recent 6-week, multi-center, double-blind, placebo-controlled RCT, investigating the treatment of low-dose methylphenidate (20 mg/day) for apathy in patients with mild-moderate AD, reported a significant improvement in apathy in the group of active treatment vs. placebo on a modified ADCooperative Study – Clinical Global Impression

of Change (Rosenberg *et al.*, 2013). Padala and colleagues (2018) conducted a 12-week, double-blind, randomized, placebo-controlled survey on 60 elderly veterans in a community dwelling. They used the AES-C to assess apathy as well as to evaluate the impact of the treatment on apathy. The authors demonstrated that the methylphenidate group experienced a significantly greater improvement than the placebo, both in terms of apathy (at 4, 8, and 12 weeks) and in terms of cognition, functional status, caregiver burden and depression (not until 12 weeks).

Pharmacological challenge has shown that AD patients with apathy have a blunted subjective reward following the administration of the dopaminergic agent dextroamphetamine (Lancôt, Herrmann, Black, Ryan, Rothenburg, Liu *et al.*, 2008), suggesting that a hypofunction of the dopaminergic system can contribute to the pathophysiology of apathy in the AD brain. Antidepressant drugs have not been found to improve apathy (Berman, Brodaty, Withall, & Seeher, 2012) although no specific studies have been conducted with second-generation antidepressant drugs, such as fluoxetine and bupropione, to evaluate the impact of these drugs alone or in combination with cholinesterase inhibitors.

## 5. Non-pharmacological treatment of apathy in AD

As discussed above, pharmacological therapies have demonstrated their efficacy in the management of apathy associated with AD. More specifically, donepezil and methylphenidate have demonstrated to be the most efficient drugs in reducing the level of apathy in the AD population. But alongside pharmacological treatment, several studies have investigated alternative types of interventions. Non-Pharmacological Treatment (NPT), also defined “ecopsychological intervention” (Zeisel, Reisberg, Whitehouse, Woods, & Verheul, 2016), has been recently considered as a new essential path (Theleritis, Siarkos, Politis, Katirtzoglou, & Politis, 2018) to be explored for the management of apathy. Non-pharmacological treatment includes interventions on cognitive, social, psychological, and relational aspects of the subject, using different methods, such as group activities, therapeutic dialogs, meditation, and sensory, physical, and physiological stimulation. The aim of this type of intervention is to improve the quality of life of the patient by strengthening his/her cognitive, psycho-affective, and social skills, reducing the psycho-behavioral symptoms, preserving the patient's social activity, restoring confidence and self-esteem, and promoting autonomy (Zuchella, Sinforiani, Tamburin, Federico, Mantovani,

Bernini *et al.*, 2018). A multiplicity of NPTs has shown to be effective on the treatment of apathy in AD. Occupational therapy and physical activity, inserted within a multidisciplinary intervention, have revealed to be effective in the treatment of apathy in patients suffering from dementia compared with pharmacological treatments alone (Treusch, Majic, Page, Gutzmann, Heinz, & Rapp, 2015). Maci and colleagues (Maci, Pira, Quattrocchi, Di Nuovo, Perciavalle, & Zappia, 2012) conducted an interesting study in a 14-patient RCT, which included physical activity as NPT, in order to assess the effect of a 3-month program of physical activity, cognitive stimulation and socialization versus usual activities at home. Maci and collaborators (2012) demonstrated a significant improvement in the AES scores. Moreover, in 2015, Telenius and co-workers confirmed that a high intensity functional exercise program in nursing home patients with dementia decreased the level of apathy following the intervention versus a control group (Telenius, Engedal, & Bergland, 2015). Several recent studies have considered the key role of exercise in the treatment of various neuropsychiatric disorders (Guerrera, Furneri, Grasso, Caruso, Castellano, Drago *et al.*, 2020). Physical activity, associated with drug treatment, allows to expand the range of rehabilitation interventions dedicated to demented patients.

Furthermore, art therapies are also used as NPTs due to their efficacy in improving the levels of apathy compared with learning therapy (Hattori, Hattori, Hokao, Mizushima, & Mase, 2011). Raglio and colleagues (Raglio, Bellelli, Traficante, Gianotti, Ubezio, Villani *et al.*, 2008) conducted a study in which they included 60 participants: a music therapy experimental group ( $n = 30$ ) and a control group ( $n = 30$ ). After 4 weeks, the NPI scores for apathy were significantly improved in the experimental group. Moreover, Ferrero-Arias and colleagues conducted a RCT, which included 146 patients, divided into two groups: an intervention group, with music, art therapy and psychomotor activity, opposed to a control group, which were simply asked to perform free activities in a room during the day. They used the DAIR scale to assess apathy and they found a significant difference between the intervention and control groups, especially in patients with moderated apathy. They thus came to the conclusion that a structured, non-pharmacological, short-term occupational therapy intervention can improve apathy in mild or moderate dementia patients (Ferrero-Arias, Goñi-Imízcoz, González-Bernal, Lara-Ortega, da Silva-González, & Díez-Lopez, 2011).

More recently, it has been demonstrated that a multi-component psychological intervention after 6 months allows to register an improvement

in the apathy NPI score versus a standard occupational therapy intervention (Fischer-Terworth & Probst, 2012).

In 2016, Di Domenico and colleagues recruited 26 AD patients and 26 healthy controls (Di Domenico, Palumbo, Fairfield, & Mammarella, 2016). The experimental group followed a brief emotional shaping intervention, which was developed to reduce apathy (assessed with the AES) and increase the “willingness to do” in AD patients. The results demonstrated that the patients of the experimental group showed a significant increase in motivation.

Finally, great relevance has been recently given to Information and Communication Technologies (ICT) with the aim to train cognitive functions, promote communication, reduce loneliness, improve physical functions and the emotional state in apathetic and non-aphathetic patients (Manera, Abraham, Agüera-Ortiz, Bremond, David, Fairchild *et al.*, 2020). An interesting study (Manera, Petit, Derreumaux, Orvieto, Romagnoli, Lyttle *et al.*, 2015) had the aim of demonstrating the efficacy of employing serious games (SGs), in this case "Kitchen and Cooking", for the assessment and rehabilitation of elderly people with MCI and AD. Twenty-one patients were recruited and results demonstrated that apathetic participants were motivated and interested in the activities, as the non-aphathetic group, confirming that this game was useful in the case of presence of apathy. Moreover, Moyle and colleagues (Moyle, Cooke, Beattie, Jones, Klein, Cook *et al.*, 2013) conducted a pilot cross-over RCT with 18 demented patients and used the AES to assess apathy; the authors found no improvement of apathy with the use of a robot companion. In contrast, other studies (Valentí Soler, Agüera-Ortiz, Olazarán Rodríguez, Mendoza Rebolledo, Pérez Muñoz, Rodríguez Pérez *et al.*, 2015) demonstrated that the use of social robots in 60 patients with dementia resulted in an improvement in the NPI apathy scores.

## 6. Conclusions

The neurobiological and clinical links between apathy and Alzheimer's Disease are essential when considering apathy as one of the main common neuropsychiatric symptoms of AD. Apathy was initially considered only as a symptom of the Major Depressive Disorder, but today, more than ever, it seems important to investigate apathy as a syndrome in itself, in order to be able to specifically intervene promptly on it and improve patients' quality of life. The connection between brain regions involved in both apathy and

Alzheimer's is quite clear and the common involvement of the cholinergic system can explain why drugs currently used in AD patients (i.e. cholinesterase inhibitors) can also be effective in the treatment of apathy. Evidence discussed in the present review suggests a strong clinical link between apathy and Alzheimer's Disease as well as the relevance of psychometric tools, such as the AES, to better diagnose and treat apathy.

We believe that only by considering every single aspect of the patient's clinical picture can the clinician plan a multi-component intervention that has effects on health, defined as a state of complete physical, mental and social well-being (World Health Organization). In fact, several studies have shown the efficacy in reducing apathy also through non-pharmacological treatments centered on individual autonomous, psychological and social functioning, but a major unmet question remains open as to how we can better integrate pharmacological and non-pharmacological treatments. A multimodal intervention is the innovative approach that we believe might be proposed in the next future for the treatment of apathetic AD patients.

## References

Albert, M. S. (2007). Changing the trajectory of cognitive decline? *The New England Journal of Medicine*, 357, 502-503.

Apostolova, L. G., Akopyan, G. G., Partiali, N., Steiner, C. A., Dutton, R. A., Hayashi, K. M., Dinov, I. D., Toga, A. W., Cummings, J. L., & Thompson, P. M. (2007). Structural correlates of apathy in Alzheimer's Disease. *Dementia and Geriatric Cognitive Disorders*, 24 (2), 91-97.

Banerjee, S., Smith, S. C., Lamping, D. L., Harwood, R. H., Foley, B., Smith, P., Murray, J., Prince, M., Levin, E., Mann, A., & Knapp, M. (2006). Quality of life in dementia: More than just cognition. An analysis of associations with quality of life in dementia. *Journal of Neurology, Neurosurgery and Psychiatry*, 77, 146-148.

Berman, K., Brodaty, H., Withall, A., & Seeher, K. (2012). Pharmacologic treatment of apathy in dementia. *The American Journal of Geriatric Psychiatry*, 20 (2), 104-122.

Bindra, D. (1959). *Motivation: A Systematic Reinterpretation*. New York: Ronald Press.

Blundo, C., & Gerace, C. (2015). Dopamine agonists can improve pure apathy associated with lesions of the prefrontal-basal ganglia functional system. *Neurological Sciences*, 36 (7), 1197-1201.

Brodaty, H., Altendorf, A., Withall, A., & Sachdev, P. (2010). Do people become more apathetic as they grow older? A longitudinal study in healthy individuals. *International Psychogeriatrics*, 22 (3), 426-436.

Bruen, P. D., McGeown, W. J., Shanks, M. F., & Venneri, A. (2008). Neuroanatomical correlates of neuropsychiatric symptoms in Alzheimer's disease. *Brain*, 131 (9), 2455-2463.

Clarke, D. E., Ko, J. Y., Kuhl, E. A., van Reekum, R., Salvador, R., & Marin, R. (2011). Are the available apathy measures reliable and valid? A review of the psychometric evidence. *Journal of Psychosomatic Research*, 70 (1), 73-97.

Cummings, J. L. (1997). The Neuropsychiatric Inventory: assessing psychopathology in dementia patients. *Neurology*, 48 (5 Suppl. 6), S10-S16.

Cummings, J. L., Koumaras, B., Chen, M., & Mirski, D. (2005). Effects of rivastigmine treatment on the neuropsychiatric and behavioral disturbances of nursing home residents with moderate to severe probable Alzheimer's disease: A 26-week, multicenter, open-label study. *The American Journal of Geriatric Pharmacotherapy*, 3 (3), 137-148.

Cummings, J. L., & McPherson, S. (2001). Neuropsychiatric assessment of Alzheimer's disease and related dementias. *Aging Clinical and Experimental Research*, 13, 240-246.

Cummings, J. L., Mega, M., Gray, K., Rosenberg-Thompson, S., Carusi, D. A., & Gornbein, J. (1994). The Neuropsychiatric Inventory: comprehensive assessment of psychopathology in dementia. *Neurology*, 44 (12), 2308-2314.

Cummings, J. L., Schneider, E., Tariot, P. N., & Graham, S. M. (2006). Memantine MEM-MD-02 Study Group. Behavioral effects of memantine in Alzheimer disease patients receiving donepezil treatment. *Neurology*, 67 (1), 57-63.

- Cummings, J. L., Schneider, L., Tariot, P. N., Kershaw, P. R., & Yuan, W. (2004). Reduction of behavioral disturbances and caregiver distress by galantamine in patients with Alzheimer's disease. *American Journal of Psychiatry*, 161 (3), 532-538.
- D'Amelio, M., Puglisi-Allegra, S., & Mercuri, N. (2018). The role of dopaminergic midbrain in Alzheimer's disease: Translating basic science into clinical practice. *Pharmacological Research*, 130, 414-419.
- Di Domenico, A., Palumbo, R., Fairfield, B., & Mammarella, N. (2016). Fighting apathy in Alzheimer's dementia: a brief emotional-based intervention. *Psychiatry Research*, 242, 331-335.
- Donovan, N. J., Wadsworth, L. P., Lorus, N., Locascio, J. J., Rentz, D. M., Johnson, K. A., Sperling, R. A., & Marshall, G. A. (2014). Regional cortical thinning predicts worsening apathy and hallucinations across the Alzheimer disease spectrum. *The American Journal of Geriatric Psychiatry*, 22, 1168-1179.
- Drijgers, R. L., Aalten, P., Winogrodzka, A., Verhey, F. R., & Leentjens, A. F. (2009). Pharmacological treatment of apathy in neurodegenerative diseases: a systematic review. *Dementia and Geriatric Cognitive Disorders*, 28, 13-22.
- Eslinger, P. J., Moore, P., Antani, S., Anderson, C., & Grossman, M. (2012). Apathy in frontotemporal dementia: behavioral and neuroimaging correlates. *Behavioral Neurology and Neuropsychiatry*, 25 (2), 127-136.
- Ferrero-Arias, J., Goñi-Imízcoz, M., González-Bernal, J., Lara-Ortega, F., da Silva-González, A., & Díez-Lopez, M. (2011). The efficacy of nonpharmacological treatment for dementia-related apathy. *Alzheimer Disease and Associated Disorders*, 25 (3), 213-219.
- Fischer-Terworth, C., & Probst, P. (2012). Effects of a psychological group intervention on neuropsychiatric symptoms and communication in Alzheimer's dementia. *Journal of Gerontology and Geriatrics*, 45 (5), 392-397.



Freund-Levi, Y., Jedenius, E., Tysen-Backstrom, A. C., Larksater, M., Wahlund, L. O., & Eriksdotter, M. (2014). Galantamine versus risperidone treatment of neuropsychiatric symptoms in patients with probable dementia: an open randomized trial. *American Journal of Geriatric Psychiatry*, 22, 341-348.

Frisoni, G. B., Winblad, B., & O'Brien, J. T. (2011). Revised NIA-AA criteria for the diagnosis of Alzheimer's disease: a step forward but not yet ready for widespread clinical use. *International Psychogeriatrics*, 23, 1191-1196.

Gauthier, S., Juby, A., Dalziel, W., Re'hel, B., Schecter, R., & EXPLORE Investigators (2010). Effects of rivastigmine on common symptomatology of Alzheimer's disease (EXPLORE). *Current Medical Research and Opinion*, 26 (5), 1149-1160.

Grambaite, R., Selnes, P., Reinvang, I., Aarsland, D., Hessen, E., Gjerstad, L., & Fladby, T. (2011). Executive dysfunction in mild cognitive impairment is associated with changes in frontal and cingulate white matter tracts. *Journal of Alzheimer's Disease*, 27, 453-462.

Guercio, B. J., Donovan, N. J., Ward, A., Schultz, A., Lorus, N., Amariglio, R. E., Rentz, D. M., Johnson, K. A., Sperling, R. A., & Marshall, G. A. (2015). Apathy is associated with lower inferior temporal cortical thickness in mild cognitive impairment and normal elderly individuals. *Journal of Neuropsychiatry and Clinical Neurosciences*, 27, 22-27.

Guerrera, C. S., Furneri, G., Grasso, M., Caruso, G., Castellano, S., Drago, F., Di Nuovo, S., & Caraci, F. (2020). Antidepressant Drugs and Physical Activity: A Possible Synergism in the Treatment of Major Depression? *Frontiers in Psychology*, 11: 857.

Guimarães, H. C., Levy, R., Teixeira, A. L., Beato, R. G., & Caramelli P. (2008). Neurobiology of apathy in Alzheimer's Disease. *Arquivos de Neuropsiquiatria*, 66 (2-B), 436-443.

Hattori, H., Hattori, C., Hokao, C., Mizushima, K., & Mase, T. (2011). Controlled study on the cognitive and psychological effect of coloring and drawing in mild Alzheimer's disease patients. *Geriatrics and Gerontology International*, 11 (4), 431-437.

Herrera, E. Jr, Caramelli, P., Silveira A. S., & Nitrini, R. (2002). Epidemiologic survey of dementia in a community-dwelling Brazilian population. *Alzheimer Disease and Associated Disorders*, 16, 103-108.

Herrmann, N., Rothenburg, L. S., Black, S. E., Ryan, M., Liu, B. A., Busto, U. E., & Lanctôt, K. L. (2008). Methylphenidate for the treatment of apathy in Alzheimer disease: Prediction of response using dextroamphetamine challenge. *Journal of Clinical Psychopharmacology*, 28 (3), 296-301.

Hirtz, D., Thurman, D. J., Gwinn-Hardy, K., Mohamed, M., Chaudhuri, A. R., & Zalutsky, R. (2007). How common are the "common" neurologic disorders? *Neurology*, 68, 326-337.

Holthoff, V. A., Beuthien-Baumann, B., Kalbe, E., Lüdecke, S., Lenz, O., Zündorf, G., Spirling, S., Schierz, K., Winiecki, P., Sorbi, S., & Herholz, K. (2005). Regional cerebral metabolism in early Alzheimer's disease with clinically significant apathy or depression. *Biological Psychiatry*, 57, 412-421.

Isaacson, R. S., Ganzer, C. A., Hristov, H., Hackett, K., Caesar, E., Cohen, R., Kachko, R., Meléndez-Cabrero, J., Rahman, A., Scheyer, O., Hwang, M. J., Berkowitz, C., Hendrix, S., Mureb, M., Schelke, M. W., Mosconi, L., Seifan, A., & Krikorian, R. (2018). The clinical practice of risk reduction for Alzheimer's disease: a precision medicine approach. *Alzheimer's & Dementia*, 14 (12), 1663-1673.

Lanctôt, K. L. (2017). Apathy associated with neurocognitive disorders: Recent progress and future directions. *The Journal of Alzheimer's Association*, 13 (1), 84-100.

Lanctôt, K. L., Herrmann, N., Black, S. E., Ryan, M., Rothenburg, L. S., Liu, B. A., & Busto, U. E. (2008). Apathy associated with Alzheimer disease: use of dextroamphetamine challenge. *The American Journal of Geriatric Psychiatry*, 16 (7), 551-557.

Levy, R., & Dubois, B. (2006). Apathy and the functional anatomy of the prefrontal cortex-basal ganglia circuits. *Cerebral Cortex*, 16 (7), 916-928.

Lopez, O. L., Mackell, J. A., Sun, Y., Kassalow, L. M., Xu, Y., McRae, T., & Li, H. (2008). Effectiveness and safety of donepezil in Hispanic patients with Alzheimer's disease: a 12-week open-label study. *Journal of the National Medical Association*, 100 (11), 1350-1358.

Lueken, U., Seidl, U., Volker, L., Schweiger, E., Kruse, A., & Schroder, J. (2007). Development of a short version of the Apathy Evaluation Scale specifically adapted for demented nursing home residents. *The American Journal of Geriatric Psychiatry*, 15 (5), 376-385.

Lyketsos, C. G., Lopez, O., Jones, B., Fitzpatrick, A. L., Breitner, J., & Dekosky, S. (2002). Prevalence of neuropsychiatric symptoms in dementia and mild cognitive impairment: Results from cardiovascular health study. *JAMA*, 288, 1475-1483.

Maci, T., Pira, F. L., Quattrocchi, G., Di Nuovo, S., Perciavalle, V., & Zappia, M. (2012). Physical and cognitive stimulation in Alzheimer disease. The GAIA project: a pilot study. *American Journal of Alzheimer's Disease and other Dementias*, 27 (2), 107-113.

Madsen, K. B. (1973). Theories of Motivation. In B. Wolman (Ed.), *Handbook of Psychology*. Prentice-Hall: New Jersey.

Manera, V., Abraham, S., Agüera-Ortiz, L., Bremond, F., David, R., Fairchild, K., Gros, A., Hanon, C., Husain, M., König, A., Lockwood, P. L., Pino, M., Radakovic, R., Robert, G., Slachevsky, A., Stella, F., Tribouillard, A., Trimarchi, P. D., Verhey, F., Yesavage, J., Zeghari, R., & Robert, P. (2020). Recommendations for the Nonpharmacological Treatment of Apathy in Brain Disorders. *The American Journal of Geriatric Psychiatry*, 28 (4), 410-420.

Manera, V., Petit, P. D., Derreumaux, A., Orvieto, I., Romagnoli, M., Lyttle, G., David, R., & Robert, P. H. (2015). "Kitchen and cooking", a serious game for mild cognitive impairment and Alzheimer's disease: a pilot study. *Frontiers in Aging Neuroscience*, 7: 24.

- Marin, R. S. (1991). Apathy: A neuropsychiatric syndrome. *The Journal of Neuropsychiatry and Clinical Neurosciences*, 3 (3), 243-254.
- Marin, R. S., Biedrzycki, R. C., & Firinciogullari, M. S. (1991). Reliability and Validity of the Apathy Evaluation Scale. *Psychiatry Research*, 38, 143-162.
- Marin, R. S., Firinciogullari, S., & Biedrzycki, R. C. (1993). The sources of convergence between measures of apathy and depression. *Journal of Affective Disorders*, 28, 7-14.
- Mitchell, R. A., Herrmann, N., & Lanctôt, K. L. (2011). The Role of Dopamine in Symptoms and Treatment of Apathy in Alzheimer's Disease. *CNS Neuroscience & Therapeutics*, 17 (5), 411-427.
- Mohammad, D., Ellis, C., Rau, A., Rosenberg, P. B., Mintzer, J., Ruthirakuhan, M., Herrmann, N., & Lanctôt, K. L. (2018). Psychometric Properties of Apathy Scales in Dementia: A systematic review. *Journal of Alzheimer's Disease*, 66 (3), 1065-1082.
- Monsch, A. U., Giannakopoulos, P., & GAL-SUI Study Group. (2004). Effects of galantamine on behavioural and psychological disturbances and caregiver burden in patients with Alzheimer's disease. *Current Medical Research and Opinion*, 20 (6), 931-938.
- Moon, Y., Moon, W. J., Kim, H., & Han, S. H. (2014). Regional atrophy of the insular cortex is associated with neuropsychiatric symptoms in Alzheimer's disease patients. *European Neurology*, 71, 223-229.
- Mori, T., Shimada, H., Shinotoh, H., Hirano, S., Eguchi, Y., Yamada, M., Fukuhara, R., Tanimukai, S., Zhang, M. R., Kuwabara, S., Ueno, S. I., & Suhara, T. (2014). Apathy correlates with prefrontal amyloid beta deposition in Alzheimer's disease. *Journal of Neurology, Neurosurgery and Psychiatry*, 85, 449-455.
- Moyle, W., Cooke, M., Beattie, E., Jones, C., Klein, B., Cook, G., & Gray, C. (2013). Exploring the effect of companion robots on emotional expression in older adults with dementia: a pilot randomized controlled trial. *Journal of Gerontological Nursing*, 39 (5), 46-53.

Munro, C. E., Donovan, N. J., Guercio, B. J., Wigman, S. E., Schultz, A. P., Amariglio, R. E., Rentz, D. M., Johnson, K. A., Sperling, R. A., & Marshall, G. A. (2015). Neuropsychiatric symptoms and functional connectivity in mild cognitive impairment. *Journal of Alzheimer's disease*, 46, 727-735.

Padala, P. R., Frederick, P., & Subhash, C. B. (2005). Methylphenitadate may treat apathy independent of depression. *The annals of Pharmacotherapy*, 39 (11), 1947-1949.

Padala, P. R., Padala, K. P., Lensing, S. Y., Ramirez, D., Monga, V., Bopp, M. M., Roberson, P. K., Dennis, R. A., Petty, F., Sullivan, D. H., & Burke, W. J. (2018). Methylphenidate for apathy in community-dwelling older veterans with mild Alzheimer's disease: A double-blind, randomized, placebo-controlled trial. *American Journal of Psychiatry*, 175, 159-168.

Radakovic, R., Harley, C., Abrahams, S., & Starr, J. M. (2015). A systematic review of the validity and reliability of apathy scales in neurodegenerative conditions. *International Psychogeriatrics*, 27 (6), 903-923.

Raglio, A., Bellelli, G., Traficante, D., Gianotti, M., Ubezio, M. C., Villani, D., & Trabucchi M. (2008). Efficacy of music therapy in the treatment of behavioral and psychiatric symptoms of dementia. *Alzheimer Disease and Associated Disorders*, 22, 158-162.

Rea, R., Carotenuto, A., Fasanaro, A. M., Traini, E., & Amenta, F. (2014). Apathy in Alzheimer's Disease: Any Effective Treatment? *The Scientific World Journal*, 2014: 421385.

Rea, R., Carotenuto, A., Traini, E., Fasanaro, A. M., Manzo, V., & Amenta, F. (2015). Apathy Treatment in Alzheimer's Disease: Interim Results of the ASCOMALVA Trial. *Journal of Alzheimer's Disease*, 48 (2), 377-383.

Robert, P. H., Clairet, S., Benoit, M., Koutaich, J., Bertogliati, C., Tible, O., Caci, H., Borg, M., Brocker, P., & Bedoucha, P. (2002). The apathy inventory: assessment of apathy and awareness in Alzheimer's disease, Parkinson's disease and mild cognitive impairment. *International Journal of Geriatric Psychiatry*, 17 (12), 1099-1105.

Robert, P., Ferris, S., Gauthier, S., Ihl, R., Winblad, B., & Tennigkeit, F. (2010). Review of Alzheimer's disease scales: is there a need for a new multi-domain scale for therapy evaluation in medical practice? *Alzheimer's Research & Therapy*, 2, 24.

Robert, P., Lanctôt, K. L., Agüera-Ortiz, L., Aaltend, P., Bremonda, F., Defrancesco, M., Hanong, C., Davidh, R., Dubois, B., Dujardin, K., Husain, M., Könige, A., Levyl, R., Mantuan, V., Meulien, D., Millero, D., Moebius, H. J., Rasmussen, J., Robert, G., Ruthirakuhan, M., Stellas, F., Yesavage, J., Zegharia, R., & Manera, V. (2018). Is it time to revise the diagnostic criteria for apathy in brain disorders? The 2018 international consensus group. *European Psychiatry*, 54, 71-76.

Robert, P., Onyike, C. U., Leentjens, A. F. G., Dujardin, K., Aalten, P., Starkstein, S., Verhey, F. R. J., Yesavage, J., Clement, J. P., Drapier, D., Bayle, F., Benoit, M., Boyer, P., Lorca, P. M., Thibaut, F., Gauthier, S., Grossberg, G., Vellas, B., & Byrne, J. (2009). Proposed diagnostic criteria for apathy in Alzheimer's disease and other neuropsychiatric disorders. *European Psychiatry*, 24, 98-104.

Ronsenberg, P. B., Lanctôt, K. L., Drye, L. T., Herrmann, N., Scherer, R. W., Bachman, D. L., Mintzer, J. E., & ADMET Investigators (2013). Safety and efficacy of methylphenidate for apathy in Alzheimer's disease: A randomized, placebo-controlled trial. *Journal of Clinical Psychiatry*, 74, 810-816.

Ruslan, L., Teerenstra, S., Smalbrugge, M., Vernooij-Dassen, M. J. F. J., Bohlmeijer, E. T., Gerritsen, D. L., & Koopmans, R. T. C. M. (2013). More insight into the concept of apathy: a multidisciplinary depression management program has different effects on depressive symptoms and apathy in nursing home. *International Psychogeriatric Association*, 25 (12), 1941-1952.

Samus, Q. M., Rosenblatt, A., Steele, C., Baker, A., Harper, M., Brandt, J., Mayer, L., Rabins, P. V., & Lyketsos, C. G. (2005). The association of neuropsychiatric symptoms and environment with quality of life in assisted living residents with dementia. *Gerontologist*, 45 (1), 19-26.

Schmidt, R., Baumhackl, U., Berek, K., Brücke, T., Kapeller, P., Lechner, A., Rainer, M., & Stögerer, E. M. (2010). Memantine for treatment of behavioural disturbances and psychotic symptoms in moderate to moderately severe Alzheimer dementia: a naturalistic study in outpatient services in Austria [in German]. *Neuropsychiatrie: Klinik, Diagnostik, Therapie und Rehabilitation: Organ der Gesellschaft Österreichischer Nervenärzte und Psychiater*, 24 (2), 125-131.

Sperling, R. A., Aisen, P. S., Beckett, L. A., Bennett, D. A., Craft, S., Fagan, A. M., Iwatsubo, T., Jack, C. R. Jr, Kaye, J., Montine, T. J., Park, D. C., Reiman, E. M., Rowe, C. C., Siemers, E., Stern, Y., Yaffe, K., Carrillo, M. C., Thies, B., Morrison-Bogorad, M., Wagster, M. V., & Phelps, C. H. (2011). Toward defining the preclinical stages of Alzheimer's disease: Recommendations from the National Institute on Aging-Alzheimer's Association workgroups on diagnostic guidelines for Alzheimer's disease. *Alzheimer's & Dementia*, 7 (3), 280-292.

Starkstein, S. E., & Brockman, S. (2018). The neuroimaging basis of apathy: Empirical findings and conceptual challenges. *Neuropsychologia*, 118, 48-53.

Starkstein, S. E., Jorge, R., & Mizrahi, R. (2006). The prevalence, clinical correlates and treatment of apathy in Alzheimer's disease. *European Journal of Psychiatry*, 20 (2), 96-106.

Starkstein, S. E., Jorge, R., Mizrahi, R., & Robinson, R. G. (2006). A prospective longitudinal study of apathy in Alzheimer's disease. *Journal of Neurology, Neurosurgery and Psychiatry*, 77 (1), 8-11.

Starkstein, S. E., Mayberg, H. S., Preziosi, T. J., Andrezejewski, P., Leiguarda, R., & Robinson, R. G. (1992). Reliability, validity, and clinical correlates of apathy in Parkinson's disease. *The Journal of Neuropsychiatry and Clinical Neurosciences*, 4 (2), 134-139.

Starkstein, S. E., Migliorelli, R., Manes, F., Teson, A., Petracca, G., Chemerinski, E., Sabe, L., & Leiguarda, R. (1995). The prevalence and clinical correlates of apathy and irritability in Alzheimer's disease. *European Journal of Neurology*, 2, 540-546.

Strauss, M. E., & Sperry, S. D. (2002). An informant-based assessment of apathy in Alzheimer disease. *Neuropsychology, Neuropsychiatry, and Behavioral Neurology*, 15 (3), 176-183.

Tekin, S., & Cummings, J. L. (2002). Frontal-subcortical neuronal circuits and clinical neuropsychiatry: an update. *Journal of Psychosomatic Research*, 53 (2), 647-654.

Telenius, E. W., Engedal, K., & Bergland, A. (2015). Effect of a high intensity exercise program on physical function and mental health in nursing home residents with dementia: an assessor blinded randomized controlled trial. *PLoS One*, 10 (5): e0126102.

Theleritis, C., Siarkos, K., Politis, A. A., Katirtzoglou, E., & Politis, A. (2018). A systematic review of non-pharmacological treatments for apathy in dementia. *International Journal of Geriatric Psychiatry*, 33, 177-192.

Treusch, Y., Majic, T., Page, J., Gutzmann, H., Heinz, A., & Rapp, M. A. (2015). Apathy in nursing home residents with dementia: results from a cluster-randomized controlled trial. *European Psychiatry*, 30 (2), 251-257.

Tunnard, C., Whitehead, D., Hurt, C., Wahlund, L. O., Mecocci, P., Tsolaki, M., Vellas, B., Spenger, C., Kłoszewska, I., Soininen, H., Lovestone, S., & Simmons, A. (2011). Apathy and cortical atrophy in Alzheimer's disease. *International Journal of Geriatric Psychiatry*, 26, 741-748.

Valentí Soler, M., Agüera-Ortiz, L., Olazarán Rodríguez, J., Mendoza Rebolledo, C., Pérez Muñoz, A., Rodríguez Pérez, I., Osa Ruiz, E., Barrios Sánchez, A., Herrero Cano, V., Carrasco Chillón, L., Felipe Ruiz, S., López Alvarez, J., León Salas, B., Cañas Plaza, J. M., Martín Rico, F., Abella Dago, G., & Martínez Martín, P. (2015). Social robots in advanced dementia. *Frontiers in Aging Neuroscience*, 7: 133.

Waldemar, G., Gauthier, G., Jones, R., Wilkinson, D., Cummings, J., Lopez, O., Zhang, R., Xu, Y., Sun, Y., Knox, S., Richardson, S., & Mackellet, J. (2011). Effect of donepezil on emergence of apathy in mild to moderate Alzheimer's disease. *International Journal of Geriatric Psychiatry*, 26 (2), 150-157.



Winblad, B., Engedal, K., Soininen, H., Verhey, F., Waldemar, G., & Wimo, A. (1999, Aug 15-20). *Donepezil enhances global function, cognition and activities of daily living compared with placebo in a one-year, double-blind trial in patients with mild to moderate Alzheimer's disease* [Poster presentation]. *Ninth Congress of the International Psychogeriatric Association*, Vancouver, Canada.

Winblad, B., Frisoni, G. B., Frolich, L., Johannsen, P., Johansson, G., Kehoe, P., Lovestone, S., Olde-Rikkert, M., Reynish, E., Vasser, P. J., & Vellas, B. (2008). EADC (European Alzheimer Disease Consortium) recommendations for future Alzheimer disease research in Europe. *Journal of Nutrition, Health and Aging*, 12 (10), 683-684.

Zeisel, J., Reisberg, B., Whitehouse, P., Woods, R., & Verheul, A. (2016). Ecopsychosocial inter-ventions in cognitive decline and dementia: a new terminology and a new paradigm. *American Journal of Alzheimer's Disease and other Dementias*, 31, 502-507.

Zuchella, C., Sinforiani, E., Tamburin, S., Federico, A., Mantovani, E., Bernini, S., Casale, R., & Bartolo, M. (2018). The multidisciplinary approach to Alzheimer's disease and dementia. A narrative review of non-pharmacological treatment. *Frontiers in Neurology*, 9: 1058.



## Level-balanced psychomotor support program for preschool children with Intellectual Disabilities

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### Abstract

*The article deals with applying a level-balanced psychomotor support program, appropriate to the levels of motor development of preschool children with intellectual disabilities and that could improve the orientation and research abilities of the aforementioned subjects. Applying a level-balanced psychomotor support system to children of preschool age with intellectual impairments could create the favorable conditions for the realization of the individually preserved capabilities of motor management and intellectual development. The central core of the psychomotor development of children with intellectual impairments is the ability to restructure the acquired movements and actions as well as the individual recovery of the children's emotional disorders, orientation and research abilities. It has been empirically proven that the use of a level-balanced psychomotor support program optimizes the psychomotor activity and improves the psychomotor capacity of preschoolers with intellectual impairments. These conditions allow a child's individually preserved physical health to be involved in the realization of motor function.*

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*Keywords:* Children with intellectual impairments; Support; Psychomotor system; Level-balanced psychomotor support program.

# 1. Introduction

## *1.1. Relevant scientific research and analysis of issues*

In modern psychological literature, there are a number of research works that have dealt with the problem of the systematization and complex analysis of child development.

Piaget (1927) made a significant contribution to the state of modern psychological science. Psychologists and pedagogues worldwide still apply his research findings in the field of child psychology, i.e., the features of children's speech and causal reasoning, the way in which children live and interpret their daily events and/or the moral and natural phenomena surrounding them. In the research conducted between 1921-1924, Piaget drew some conclusions concerning children's innate egocentrism and their gradual socialization while communicating with adults. In his opinion, we should not investigate social life in connection with the mind; instead, we should direct our efforts on social relationships. Piaget added the psychological factor in the context of these relationships, that is, the level of intellectual development of the individuals who interact.

As demonstrated by Vallon in his thesis "Stages and impairments of psychomotor and motor development of a child" (1967), the mental activity of a child is based on the idea of the gradual development of a child's mind. Certain stages of this development are characterized by the domination of the affective sphere over the rational sphere, while other stages show the primacy of intelligence over emotions. There is thus a gap and even a competition between the rational and emotional spheres that determine the development of a child's personality.

Bernshtein (1991), the founder of a new direction in science, i.e. the physiology of action, investigated deeply and partially described this issue. He was the first to describe and to demonstrate the hypothesis of action based on the assumption that, to make a movement, the brain must not only send commands to the muscles but it must also receive signals from the peripheral senses and give new "commands", based on the signals received. Thus, in a process of building the movements, there is not only a direct but also inverse connection between the brain and the peripheral nervous system.

Zaporozhets published a monograph "The development of arbitrary movements" (1960), which described a study of different types of movement habits as a model of mastering skills and new types of behavior in

preschoolers. This paper gave a significant contribution to the psychological theory of action construction. It also included some sections from the previously published paper entitled “Rehabilitation of Movements” (Leontiev & Zaporozhets, 1945).

Bachman (1961) made important conclusions concerning the tight connection of motor activity with three aspects of health (physical, mental, and social) and concerning the different roles played by motor activity throughout life. In his opinion, in childhood, motor activity provides the typical development of a body. It is during development that the body is most sensitive to the effects of adverse environmental factors, including restraints in motor activity, which are common for children with intellectual impairments.

The research of Vilchkovsky (1983) showed that the formation of preschooler's motor skills depends on three main group factors: genetically-determined motor inducements, the arbitrary motor activity of a child (that depends on the implementation of many movements in everyday life), and the particularly organized pedagogical influence, which stimulates the natural course of ontogenetic development of motor activity.

Lockman and Thelen (1993) also found the link of the interaction among the motor, cognitive and emotional manifestations of the mind. Understanding this psychomotor interaction could be especially important to study mental retardation as a pedagogical problem. The need for movement, especially in children, is considered one of the most important body features, which plays a significant role in the normal functioning and vital activity of the body of a human being. At all stages of the evolutionary process, the development of a human being takes place in close connection with its muscular activity, which can be considered one of the main factors ensuring the optimal stability of the internal environment of the organism and its activity in relation to environmental changes.

Wallace and Goldstein (1994) proved that the accuracy of movements is influenced by the individual-typological qualities of the nervous system, the intensity zone of the measured parameters, the possible damage to the nervous system as well as the critical periods of psychomotor development.

Another scientist working on this issue was Sally Goddard Blythe, who conducted research on the effects of neurological dysfunction in specific learning difficulties and proposed effective remedial programs. She was the author of the “Reflexes Learning and Behavior” (2005) as well as numerous other research papers and articles. The “Well Balanced Child” (2004) was a passionate manifesto for a “whole body” approach to learning, which

integrated the brain, senses, movement, and play. This fully revised edition included a new chapter with a story and movement exercises that parents could use to help children reach their potential in motility.

Practical experience and theoretical research both allow us to state that all children with intellectual impairments, regardless of the specialties of psychomotor underdevelopment and of the level of the stable organic violation in the cognitive activity, meet huge difficulties in performing tasks that need the implementation of changes in movements and actions that have already been mastered. As the ability to change and improve actions that are already mastered depends on the children's abilities to differentiate the movements of the senses (that is the subject of psychology), on the individual features of the localization of the central nervous system's damage (that is the subject of physiology) and on the technologies used in pedagogical actions, psychomotor impairments should be adjusted in accordance with all the factors mentioned above.

Nowadays, focused and effective programs for the support of psychomotor abilities of children with intellectual disorders are still lacking.

The aim of this research paper was therefore to characterize the application of the level-balanced psychomotor support in children with intellectual disorders aged 6-7 years, using a level-balanced approach to the construction of movements and studying an orientation activity of the mind.

## 2. Methods

### *2.1. Participants and procedure*

We aimed to examine the efficiency of a level-balanced program of psychomotor support for 6-7 year old children with intellectual impairments from an empirical point of view based on the research findings of Simko (2012). As described by the aforementioned author, the program includes four levels. Level A is the evaluation of the coordination in psychomotor potential of preschool children with intellectual impairments at the spinal level. Level B consists in the three-meters walking test with eyes closed, eliminating "the reference" of movements to the environment. Level C is the diagnostic determination in coordination potential of a child at the pyramidal and striatal level. Level D, finally, is the application of the "mounting-dismounting" manipulative test at the parietal and premotor level, the performance of which requires finger dexterity consisting of two-hand manipulations on objects of daily use.

The implementation of a level-balanced psychomotor support for children with intellectual impairments was carried out in three main stages: preparatory, main, and final.

The preparatory stage included the complex definition of the psychomotor development of children with intellectual impairments at different levels of the construction of movements, the development of goals, tasks, and strategies of the supportive therapy, the planning of work in forming the so-called “school” of movements at the levels involved in their construction as well as the arrangement of the necessary materials and technical basis to carry out the supportive classes.

The main stage included the direct implementation of a level-balanced psychomotor support in accordance with the structural and functional components of the model of psychomotor support of children with intellectual impairments in terms of the construction of movements. The structural and functional model of support of psychomotor abilities is a time-determined sequence of actions indicating functional relationships between different levels. In the main stage this program includes a set of physical exercises and mobile games that provide the content of the impact on the psychomotor skills supporting them.

The final stage of the program consisted in the consolidation of the positive changes in psychomotor development of children with intellectual impairments and the transfer of these changes into different activities. Our research revealed that the most effective influence on the psychomotor support of preschoolers with intellectual disorders is not given by isolated exercises but rather by complex exercises in which children were found to resort to compensatory ways of motor control.

A total of 65 children with intellectual impairments of older preschool age (experimental group) was involved in the study and recruited from the Kamyanets-Podilsky Specialized Preschool Educational Institution (Ukraine) and from the Kryvyi Rih Orphanage Kindergarten for Kids with Special Needs (Ukraine). Gender differences were not taken into account during the experiment. The importance of this correctional program was in its impact on the psychomotor development of children with intellectual disabilities.

Before the experiment, the difference between the arithmetic means was statistically significant  $p < .01$ . After the experiment, the difference between the arithmetic means was statistically significant  $p < .05$ .

Before applying the experimental program and following its approbation, five specifically trained experts, with the appropriate academic background



(obtainment of a PhD, Master, etc.) and practical expertise (5+ years of experience of work with children with intellectual impairments), estimated the psychomotor skills of the children under examination in interactive, labor, design, art, and speech activities by applying a 12-point scale (see Tab. 1 for more details).

Table 1 – *Dynamics of indicators of psychomotor skills in different types of activities (interactive, labor, design, art, speech) of children with intellectual disorders aged 6-7 years (experimental group) during the 16 weeks of the supportive classes (n = 65)*

Kinds of activity	Quantitative characteristics of growth (M $\pm$ m)		
	Before the experiment	After the experiment	Difference
Interactive	1.1 $\pm$ .05	.7 $\pm$ .04	.4 $\pm$ .02
Labor	1.0 $\pm$ .04	.7 $\pm$ .03	.3 $\pm$ .01
Design	1.3 $\pm$ .06	.6 $\pm$ .03	.7 $\pm$ .04
Art	.9 $\pm$ .04	.5 $\pm$ .02	.4 $\pm$ .02
Speech	1.4 $\pm$ .07	.8 $\pm$ .04	.6 $\pm$ .03

Methods of correction of psychomotor skills of children with intellectual disabilities of preschool age should provide effects on all the levels of movement. In addition, in the conditions of corrective actions in the group, such as in physical education classes, the volume of exercises for which children conduct different levels of construction of movements should be approximately the same. In this way, the relatively balanced technique will create favorable conditions for the correction of psychomotor skills of all the children in the group with individually unique damage to the central nervous system.

The use of the level-balanced program during the 16 weeks of the supportive classes (three per week) caused some changes in the indicators of psychomotor skills in the various activities.

### 3. Results and discussion

Preschool age is the main stage of the formation of an arbitrary motor function in children, which is ensured by the morphological maturation and the development of functional capabilities of the central and peripheral parts of the motor analyzer, as mentioned by Arkin (1968). It allows to provide an

effective supportive therapy of a child's psychomotor system in this developmental period.

Before applying the program, the manifestation of children's psychomotor capabilities in the interactive activity was  $6.0 \pm .30$  points, which grew to  $7.1 \pm .25$  points following 16 weeks of training (see Tab. 1). The difference between the arithmetic means was statistically significant ( $p < .01$ ).

The health and performance of preschoolers with violations in the intellectual sphere in many cases depends on the development of their motor skills. An optimal level-balanced motor activity strengthens the health and promotes the diversified development of a child, in particular, by improving the management of self-service labor activities. Prior to the application of the correctional program, the psychomotor skills of the subjects under study in their labor activities were estimated at  $5.7 \pm .22$  points, while after the supportive therapy at  $6.7 \pm .28$  points, a difference that was statistically significant ( $p < .01$ ).

Physical exercises also have a certain impact on the intellectual sphere of children. Sechenov (1947) noted that the muscular movements of a child favorably affect the development of its brain. Lesgaft (1952) pointed out that, by using physical exercises, we can influence the intellectual development of children. Vallon (1967) emphasized the relationship between motor and intellectual skills of a child. Here we found that psychomotor skills in design in children with intellectual impairments aged 6-7 years increased from  $6.5 \pm .29$  to  $7.8 \pm .31$  points after 16 weeks of supportive exercises with a level-balanced program (see Tab. 1). The difference between the arithmetic means was again statistically significant ( $p < .01$ ).

The support and improvement of skills in actions guided by the different levels leading to the construction of movements are also associated with the development of the qualitative aspects of the motor activity of a child with intellectual impairments. The assimilation of certain actions of the level-balanced support by preschoolers was accompanied by the significant development of their psychomotor skills in the art activities (from  $5.5 \pm .26$  points before the supportive therapy to  $6.4 \pm .30$  points following the therapy,  $p < .05$ ; see Tab. 1 for more details).

The need of particular attention was given to the development of speech skills, where, to a large extent, successful intellectual development depends on. The development of speech in children with intellectual impairments is possible only with the systematic execution of appropriate exercises with a

gradual increase in the level of their complexity. We found that after 16 weeks of applying the level-balanced psychomotor support on the first four levels (A-D), the speech skills at the fifth level (E) increased from  $5.4 \pm .32$  to  $6.8 \pm .33$  points. The difference between the arithmetic means was statistically significant  $p < .01$ . In other words, we observed a significant increase in the children's speech skills on a level that was not specifically adjusted. We can explain this with the fact that children with intellectual impairments have a weakened health, they get tired quickly, and thus do not execute their speech function effectively. The support of the coordination of movements, with the help of the level-balanced program, improves the general psychomotor abilities of children with intellectual impairments and thus creates the conditions for the improvement in the conduction of speech.

Motor activity restrictions deconditioning the body, with the exception of the important motor-visceral reflexes, lead to a violation in the metabolic processes and to significant changes in the functions and reserves of the internal organs and systems. The optimization of psychomotor activity, on the contrary, contributes to the growth of a child's psychophysical forces (Shynkariyuk, 2005).

When planning and organizing the motor activity of children with intellectual impairments, we adhered to certain requirements in the selection of physical exercises, games, and forms of entertainment, by choosing physical exercises of small- and medium-intensity having a certain corrective direction, which depended on the leading level of the construction of movements. We exposed children to a variety of exercises, also taking care of their attractiveness and playful nature, and involved different groups of muscles, depending on the content and nature of the performance.

The development of psychomotor functions in a child is determined by its motor regime in the family too. The more time children spend on motor activity during their daily routine, the healthier they are and the more they improve in physical development and psychomotor readiness. All the children of our dataset were in approximately the same family conditions.

By developing the motor function in preschoolers with intellectual impairments, we also combined the support, the dynamics of the formation of motor skills, and the development of psychomotor qualities.

The use of the level-balanced program during the 16 weeks of classes caused certain changes in the indicators of psychomotor readiness in the natural actions (see Tab. 2 for more details).

Table 2 – *Dynamics of indicators of psychomotor readiness in the natural actions of children with intellectual disorders aged 6-7 years (experimental group) during the 16 weeks of training (n = 65)*

Indicators (units of measure)	Quantitative characteristics of growth ( $M \pm m$ )		
	Before the experiment	After the experiment	Difference
Running 10 m at speed (s)	$.3 \pm .02$	$.2 \pm .01$	$.1 \pm .01$
Long jump range (cm)	$6 \pm .12$	$5 \pm .11$	$1 \pm .02$
Throwing bags with sand on a range (m)	$.89 \pm .03$	$.61 \pm .02$	$.28 \pm .01$

Preschoolers with intellectual impairments have in common the fact that they all need an early detection of their psychomotor abilities and a systemic support to prepare for daily life and study. Timely support of psychomotor development is aimed at the maximum possible overcoming of a child's violations.

The correctional impact on children with intellectual impairments in our study was carried out on the basis of a number of methodological principles: consequence, differentiation, individual approach to the child, etc. This is especially important when supporting such psychomotor qualities as strength, speed, endurance, and dexterity.

Psychomotor development is a complex dialectical process characterized by a concrete sequence, the uneven maturation of certain functions as well as their qualitative transformation at a new age stage. At the same time, each further stage of development is inextricably linked with the previous one (Shynkariyuk, 2002).

In the exercise “Running 10 m at speed”, the average result of the group before the correction was  $3.6 \pm .06$  s, while following the correction it became  $3.3 \pm .07$  s, a difference that was statistically significant ( $p < .01$ ; see also Tab. 2 for details). Such a significant increase in the speed of movements can be explained, on the one hand, by the influence of the level-balanced psychomotor support, and, on the other hand, by the fact that children of this age are sensitive to increasing the frequency of steps.

Observations prove that children with intellectual impairments are lagging behind in their development. This is due not only to the peculiarities of the damage to their central nervous system influenced by various environmental factors but also to the delivery of the correct diagnosis and beginning of the supportive therapy, which are not always timely.

Violations of psychomotor development have different dynamics. Along with the persistent deviations in the development due to organic brain damage, there are many so-called inverse variants that arise with mild cerebral dysfunction, somatic weakening, pedagogical neglect, and emotional deprivation.

These deviations can be completely overcome in case of a well-timed support. The dynamics of growth of psychomotor qualities in the process of supportive work is also different. For example, under the influence of the supportive therapy here proposed, the results in the “Long jump” increased from  $84 \pm 1.81$  cm to  $90 \pm 1.53$  cm, which was slightly less than in the exercise “Running 10 m at speed” but still statistically significant ( $p < .05$ ; Tab. 2).

Regardless of the peculiarities of motor underdevelopment, preschool age children with intellectual disabilities all slightly improved in the practice of “Throwing bags with sand at a range” after our level-balanced support. The average result of the investigated group before the supportive therapy was  $7.72 \pm .25$  m, while it became  $8.61 \pm .24$  m following the therapy. The differences between arithmetic means were again statistically significant ( $p < .05$ ).

Since the ability to move away from a movement, once it is learned and mastered, is based on the orientation and research skills of the psychomotor system of the subject, it is peculiar to the individual and the results obtained by children can be very different. The goal-oriented support of the ability to change one’s psychomotor stereotypes and the formation of effective orientation and research movements in children occurs not only in the application of the level-balanced program, but also in the process of carrying out the investigated natural actions. However, the task of improving the ability to differentiate movements is rather complicated and requires defining the appropriate psychological and pedagogical principles of this work. Therefore, a level-balanced approach, in our opinion, was effective in reaching the aforementioned goal.

To conduct this study, we were inspired by the position and discoveries of Bernshtein (1991) who considered the system of mechanisms for the management of the psychomotor system as a complex hierarchical formation composed of a number of levels of construction of movements. Therefore, the violation of the motor function in children with intellectual impairments is a consequence of the damage to the corresponding cerebral systems and orientation-and-research activities. We thus recorded the indicators of psychomotor skills in preschoolers with intellectual impairments at different

levels of construction of movements and investigated the possibility to apply a level-based program as supportive therapy (Tab. 3).

Table 3 – *Dynamics of indicators of psychomotor skills at different levels of construction of movements in children with intellectual impairments aged 6-7 years (experimental group) during the 16 weeks of supportive classes (n = 65)*

Indicators (units of measure)	Quantitative characteristics of growth (M ± m)		
	Before the experiment	After the experiment	Difference
Duration of balancing on one leg (s)	1.61 ± .18	.31 ± .04	.30 ± .03
Duration of balancing on two legs located one after another (s)	3.71 ± .16	1.12 ± .05	2.59 ± .03
Accuracy of the simulation of the 7-meter walking test (cm)	8.2 ± .81	1.9 ± .18	6.3 ± .14
Accuracy of the simulation of the 3-meter walking test (cm)	3.4 ± .28	.8 ± .07	2.6 ± .22
Accuracy of a long jump at 75% of max (cm)	1.49 ± .13	.25 ± .02	1.24 ± .11
Accuracy of a long jump at 50% of max (cm)	1.19 ± .12	.27 ± .03	.92 ± .10
Speed of “mounting” operation (s)	16.08 ± .67	11.12 ± .44	4.96 ± .19
Speed of “demounting” operation (s)	12.18 ± .65	6.48 ± .34	5.7 ± .30

We found that the use of supportive exercises to maintain certain positions with smooth but tangible changes in the kinematic and dynamic characteristics of psychomotor actions during the 16 weeks improved the coordination skills of preschoolers with intellectual impairments at the level of paleokinetic regulations. More specifically, the “Duration of balancing on one leg” significantly increased from  $3.90 \pm .41$  s to  $5.51 \pm .34$  s ( $p < .01$ ; see Tab. 3 for more details). This is explained by the fact that exercises like the “Bumblebee”, “Cranberry”, “Butterfly”, “Mill” represent a certain “school” in the control of movements for which the rubrospinal level is leading. Children master the skill to change the muscular tension, which is needed to hold different static positions, to differentiate their muscular tension. In children with an excessive muscular stiffness before the correction, the movements gained signs of alternation of stress and relaxation. In contrast, in children who experienced an excessive relaxation

of the muscles of the neck, trunk, and extremities before the supportive therapy, the movements were more dynamic and the muscle tone slightly increased. In other words, the muscle tone improved when performing psychomotor actions.

The “Duration of balancing on two legs” located one after another was also shown to increase significantly (Tab. 3). Before the experiment, this indicator was equal to  $12.59 \pm .62$  s, while following the supportive classes it rose to  $16.31 \pm .53$  s, a difference that was statistically significant ( $p < .001$ ). In this exercise, the children showed better results and the conditions for its implementation allowed them to hold the balance for a longer period. At the same time, the relative improvement of the result was also greater than in the previous exercise. This can be explained by the fact that, in our opinion, children with persistent organic violations of cognitive activity could preserve information concerning the work of their muscles in feelings and perceptions for a longer time. In other words, the sensory-perceptual system had more time for self-improvement.

The supportive exercises at level A allowed children to improve the management of muscle tone and muscle excitability according to the functionalities of the nervous system that were saved individually. The supportive tasks stimulated the preserved functions of children with intellectual disorders so that their bodies became active purposeful systems and they themselves became subjects of psychomotor activity.

The supportive tasks for the control of muscle tone and muscle excitability stand on the principle of the ring regulation of movements based on inverse relationships. The nervous system does not only give the efferent orders to the muscles but also receives afferent information from the analyzers regarding the results of their execution. A moderate variation of movements “teaches” the nervous system to change them with the aim to overcome the differences between the real and desirable characteristics of the movements. After all, the rubrospinal level of paleokinetic regulation receives a proprioceptive afferentation concerning the magnitude and direction of muscle tension, and the strength and direction of muscle pressure, which makes it possible to determine the body's position regarding the earth gravity and the location of the parts of the body.

As a consequence, at level A, the psychomotor problems for the acceptance and preservation of poses were solved and the exercises we chose contributed to improving the management of this class of semantic tasks.

In our dataset, we found that the implementation of the level-balanced psychomotor support during 16 weeks also allowed to improve the psychomotor skills of children with intellectual impairments aged 6-7 years at the thalamic pallidum level of synergies, which is the level of common movements and standard stamps. More specifically, before the experiment, the “Accuracy of the simulation of walking at 7 meters” without visual control was  $26.2 \pm 2.05$  cm, which became  $18.0 \pm 1.97$  cm following the experiment (Tab. 3). The difference between arithmetic means was statistically significant ( $p < .01$ ) also in this case. This can be explained by the fact that exercises focused on the measurement, reproduction, and differentiation of space and time (without visual control) actualize proprioceptor and tango receptor afferentation, which informs the brain about the magnitude of the angles of the joints during movement, the direction and speed of their changes, and creates the image of the linkage of the body parts. Thus, we can state that the adjusting tasks of differentiating, reproducing and measuring the spatial and temporal characteristics of motion with the eyes closed allow to improve the ability of the subject in psychomotor activity to coordinate the work of hundreds of muscles in the “body scheme” in time, as well as to repeat a variety of complex movements.

The “Accuracy of the simulation of walking at 3 meters” without visual control was also found to be significantly different before and after the supportive therapy ( $p < .05$ ; Tab. 3). Before applying the supportive therapy, the accuracy of the simulation of the 3-meter walking test was in the order of  $14.1 \pm .98$  cm, which went down to  $10.7 \pm 1.06$  cm following therapy. However, it must be underlined that the relative improvement in the exercise “Accuracy of the simulation of walking at 3 meters” was worse than in the exercise at 7 meters. This difference, in our opinion, can be explained by the fact, that, when reproducing a 7-meter walk, the sensory-perceptual system of children with intellectual impairments had more time to adjust and improve their psychomotor skills than when reproducing a 3-meter walk.

We also observed that the use of a level-balanced psychomotor support in children with intellectual impairments decreased the symptoms of asynergia and dyssynergia, stereotypia and dynamic disorders and improved the plastic arbitrary movements, which became less. At the same time, it should be emphasized that the thalamus pallidum level is not adapted to the use of information concerning the environment from the visual and auditory analyzers. The supportive exercises, for which the level B was leading, improved the coordination of movements in the “body scheme”, bringing the



sequence and duration of muscle tension together in time. Following the experiment, the subjects of the research considerably improved the ability to use inertial and reactive forces for solving certain psychomotor problems.

We should also add that the level of common movements and standard stamps, i.e. the level of synergies, was not stereotypical in itself and the variability of the characteristics of the exercises of free pulse gymnastics increased the ability of children with intellectual impairments to adapt their movements to the dynamic conditions of the environment.

As a consequence, the psychomotor problems concerning the coordination of movements of various parts of the body in space and time (in the body scheme) were solved on the thalamus pallidum level and the exercises chosen by us contributed to improving the management of this class of semantic tasks.

We found that the use of the supportive exercises in which the body, its parts and objects moved in accordance with the peculiarities of the surrounding space during the 16 weeks improved the coordination skills of children with intellectual impairments at the pyramidal striatal level of the spatial field. More specifically, the “Accuracy of the long jump at 75% of max” increased from  $4.60 \pm .30$  cm to  $3.11 \pm .42$  cm. The difference between arithmetic means was statistically significant ( $p < .01$ ; Tab. 3). This is due to the fact that certain exercises, such as “Throwing bags to the target”, “Hoop-to-the-hoop jumping”, “Running bypassing the obstacles”, “Two guardians” and others describe a certain “scale” of coordination of movements for which the pyramidal striatal level C is leading. Children with persistent organic impairments in cognitive activity acquired the ability to change their movements in accordance with changes in the spatial conditions of the psychomotor activity. We observed a reduction in the signs of distress, ataxia and paresis in preschoolers after 16 weeks of being exposed to the supportive exercises, showing that the spatial accuracy of “bindings” of movements to the environment was optimized.

A similar significant difference ( $p < .01$ ) prior and following the experiment was also observed in the “Accuracy of the long jump at 50% of max”. Before the experiment, this indicator was equal to  $3.21 \pm .27$  cm, while after the supportive therapy it was equal to  $2.02 \pm .31$  cm. The supportive exercises at the level of the spatial field contributed to the actualization of the receipt of information concerning the features of muscle work, changes in joints, balance, as well as the information from the organs of vision. On the basis of their synthesis, the subject of psychomotor activity optimized the management of complex arbitrary actions. The supportive

exercises we adopted actively involved the preserved morpho-functional formations of the central nervous system in the management of movements and, under certain conditions (requiring further neuropsychological research), contributed to the formation of new functional organs.

The supportive tasks of a spatial-target nature, addressed to the outside world, are always transposable. They are implemented on the basis of proprio-receptor, tango receptor and telereceptor afferentation. In our dataset, we observed that these exercises were characterized by considerable flexibility, i.e. a variation of kinematic and dynamic characteristics, although the spatial target remained unchanged. The supportive tasks we used helped the children with intellectual disorders to adapt to the spatial and temporal peculiarities of the subject world, bypassing the obstacles on the way to the goal.

Therefore, at the pyramidal striatal level C, psychomotor tasks, focused on the replacement of the body and objects in space, were solved and the exercises we used helped to improve the management of this class of semantic tasks.

Our results also revealed that performing our level-balanced program of psychomotor support for 16 weeks improved the coordination skills of senior preschool children with persistent organic cognitive disturbances at the sincipital premotor level of object actions or semantic chains. More specifically, the speed of the “mounting” operation before and after the experiment was  $133.84 \pm 5.10$  s and  $117.76 \pm 4.81$  s, respectively (mean difference:  $p < .05$ ).

This can be explained by the fact that the supportive exercises with objects used in everyday life actualized the information both concerning the geometric features of the object and concerning its semantic purpose, promoting the development of a major coordination of the hands and fine motor skills (fast and accurate finger movements).

Finally, the speed of the “dismounting” operation before and after the experiment was  $81.73 \pm 3.71$  s and  $69.55 \pm 4.12$  s, respectively (mean difference:  $p < .05$ ). Managing actions for which level D was the leading one also relied on the ability to create skills based on exercises. Our observations proved that the automation of locomotor composition, sensory syntheses and central components of action greatly increased their effectiveness.

Therefore, the system of supportive exercises that we proposed at the sincipital premotor level improved the coordination skills of children in actions with objects used in everyday life and the solution of the semantic tasks of this class. At the same time, it was not only the level of the subject's

actions or semantic chains D that took part in the actions with everyday objects, but also the lower levels (as backgrounds).

### *3.1. Propositions*

It is difficult to determine unequivocally how the growth of possibilities at the lower levels manifested itself in the results of the higher ones. We can assume that the coordination skills of the higher levels are more integral than the general psychomotor skills of the child.

Our empirical study showed that, in most of the children surveyed, the levels of movement construction on which the worst outcomes were recorded before psychomotor support therapy were delayed even after the support program. The development of psychomotor skills in actions with these levels leading was carried out more slowly. However, there were a few cases of significant progress in the coordination ability of the levels that were delayed prior to supportive care.

Therefore, using the level-balanced psychomotor support program, we created the conditions for the preschoolers with intellectual disabilities to implement their individually preserved abilities for movement management and intellectual development.

The central link in the psychomotor support of children with intellectual disabilities is the development of their ability to reorganize acquired actions and movements, the individually possible restoration of sensory disturbances, orientation and research possibilities. Thus, the conditions of intellectual development change.

## **4. Conclusions**

1. The psychomotor support program was constructed in such a way as to include various actions with different levels of movement formation, of approximately the same volume, leading to these actions, and their implementation provided the appropriate changes in kinematic and dynamic characteristics. Its application allowed to optimize the psychomotor activity and to improve the psychomotor skills of preschool children with intellectual disabilities. These conditions lay the foundations to contribute to the implementation of the motor function of a child's individually preserved psychophysical health.

2. Physical exercises, irrespective of their leading levels, also proved to be a kind of training for a child, in which the psychomotor qualities develop gradually, the coordination mechanisms of the psychomotor system improve and compensatory pathways are found. However, this way of psychomotor system support has shown to be less effective than the use of the level-balanced program.

## References

Arkin, E. (1968). *A child in preschool years*. Moscow: Prosveshcheniie. (In Russian).

Bachman, J. C. (1961). Motor learning and performance as related to age and sex in two measures of balance coordination. *Research Quarterly of the American Association for Health, Physical Education, & Recreation*, 32, 123-137.

Bernshtein, N. A. (1991). *About agility and its development*. Moscow: Physical culture and sport. (In Russian).

Goddard Blythe, S. (2005). *Reflexes, Learning and Behavior. A window into the child's Mind: A Non-Invasive Approach to Solving Learning & Behavior Problems*. Eugene, OR: Fern Ridge Press.

Goddard Blythe, S. (2004). *The Well Balanced Child: Movement and Early Learning*. Stroud, UK: Hawthorn press.

Leontiev, A. N., & Zaporozhets, A. V. (1945). *Rehabilitation of Movements. A Study into the Rehabilitation of Hand Movements After the War Injury*. Moscow: Sovetskaya Nauka. (In Russian.)

Lesgaft, P. F. (1952). *Collected pedagogical works, Guidelines for physical education of school-age children, Vol. 2*. Moscow: Physical culture and sport. (In Russian).

Lockman, J. J., & Thelen, E. (1993). Developmental biodynamics: Brain, body, behavior connections. *Child Development*, 64, 953-959.

Piaget, J. (1927). *La causalité physique chez l'enfant*. Paris: Félix Alcan.

Sechenov, I. M. (1947.) *Selected philosophical and psychological works*. Moscow: State Publishing House of Political Literature. (In Russian).

Simko, A. (2012). Specialties of usage of the level-balanced program of psychomotor skills correction in oligophrenic children aged 6-7 years. *The Collection of Scientific Papers of Kamianets-Podilskyi National Ivan Ohienko University. Pedagogical Series*, 17 (2), 186-197.

Shynkariyuk, A. (2002). *The development of movement and mind: problems of energy and freedom*. Kamianets-Podilskyi: Kamianets-Podilskyi state pedagogical university, information-and-publishing division. (In Ukrainian).

Shynkariyuk, A. (2005). *Psychomotor-level structure of energy and freedom of a subject*. Kamianets-Podilskyi: Oiyum. (In Ukrainian).

Vallon, A. (1967). *Mental development of a child*. Moscow: Education.

Vilchkovsky, E. (1983). *The development of children motor function*. Kyiv: Zdorovia. (In Ukrainian).

Wallace, P. M., & Goldstein, J. H. (1994). *An introduction to psychology* (3<sup>rd</sup> ed.). Cincinnati: Brown & Benchmark Publishers.

Zaporozhets, A. V. (1960). *The development of arbitrary movements*. Moscow: APN RSFSR.



## Religious coping among caregivers of differently-abled children

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### Abstract

*Caregivers of differently-abled children go through significant challenges in their lives. A number of studies have shown the impact of child disability on the caregivers, although only a handful of studies have shown the different coping strategies used by caregivers in order to buffer against their stressful life events. Hence, the aim of the present research was to study religious coping among the caregivers of differently-abled children with respect to their age and type of child disability. Two hundred caregivers from different rehabilitation schools and centers of the Kashmir valley were selected through a purposive sampling technique. The mean age and standard deviation of the caregivers were 40.04 and 5.31, respectively. Welch's test, followed by the Games-Howell Post-hoc test, were carried out to analyze the data. The results revealed that children with older caregivers (40-52 years old) scored high on religious coping as compared to children with younger caregivers (27-39 years old). Furthermore, results also revealed that all the caregivers use religious coping as a means to cope with their day-to-day stressful life events, except for caregivers with visually-impaired children, who practice the strategy less as compared to their counterparts.*

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*Keywords:* Religious Coping; Caregivers; Differently-abled children;  
Age; Types of child disability.



## 1. Introduction

Caregivers of children with disabilities go through significant challenges in their daily lives. They experience a wide range of emotions like anger, depression, denial, guilt, shame, hopelessness, shock, financial burden, marital conflict, and even withdrawal. These emotions keep them at stake and significantly impact their physical and mental health. Previous studies have examined the role of emotional and problem-focused coping strategies (Folkman & Lazarus, 1984) among caregivers of differently-abled children. Emotional coping strategies are applied by caregivers to reduce negative emotions like fear, anxiety, depression, embarrassment, and frustration, whereas problem-focused coping strategies are adopted to tackle the problem and to reduce the situation causing stress to the caregivers. Religious coping is the third strategy, besides emotional and problem-focused coping strategies, which could play a buffering role against these stressful life events. Several studies have shown a significant positive impact of religious coping on caregivers assisting patients with diseases like Alzheimer's disease, terminally ill cancer patients, patients with stroke, Thalassemia, mental illness, dementia, and many other diseases (Stolley, Buckwalter, & Koenig, 1999; Pearce, Singer, & Prigerson, 2006; Rathier, Davis, Papandonatos, Grover, & Tremont, 2015; Pearce, Medoff, Lawrence, & Dixon, 2016; Chong, Chong, Tang, Ramoo, Chui, & Hmwe, 2019; Kes & Aydin Yildirim, 2020).

Religious coping refers to the use of cognitive-behavioral techniques to manage stressful situations in the light of one's spirituality or religious beliefs (Pargament, 1997). Epidemiological studies (and the resulting meta-analyses) have shown that people frequently rely on religion to cope with life stresses, demonstrating the positive impact that religious coping may have on the general well-being of individuals (Harrison, Koenig, Hays, Eme-Akwari, & Pargament, 2001; Ano & Vasconcelles, 2005). Moreover, a smaller number of studies have found religious coping to moderate the relationship between stress and well-being, providing support for the buffering hypothesis (Tix & Frazier, 1998; Aydin, Fischer, & Frey, 2010; Carpenter, Laney, & Mezulis, 2012; Fernandez & Loukas, 2014). These findings elucidate the two mechanisms by which religious coping can affect the outcomes of adaptation (Ensel & Lin, 1991).

Researchers have identified a variety of ways in which people use religion to cope with stressful life situations, including engaging in religious practices, seeking social support through religious leaders and

congregations, and reframing stressful events in reference to their relationship with God (Boudreaux, Catz, Ryan, Amaral-Melendez, & Brantley, 1995; Pargament, Smith, Koenig, & Perez, 1998; Krägeloh, Chai, Shepherd, & Billington, 2012). Some researchers have focused more exclusively on the cognitive aspects of religious coping (Pargament *et al.*, 1998). Boudreaux and colleagues (1995) have explored the importance of outward religious practices and social support accessed via religious networks. Their results suggest that different facets of religious coping may differentially impact the outcomes of the well-being of individuals. These findings highlight the potential independence of the cognitive, behavioral, and social dimensions of religious coping.

Religious coping is a process of coping with difficult circumstances through religion that can help in a variety of ways.

Pargament (1997) posited three approaches to the issue of control in religious coping.

1. Deferring mode of relationship: Praying in order to put the problem completely in the hands of God.
2. Collaborative mode of relationship: Praying in which the supplicant and God work together on the problem.
3. Self-directive approach: Praying in which God is acknowledged but the problem requires personal rather than divine solutions.

Other authors (Maynard, Gorsuch, & Bjorck, 2001; Wong-McDonald & Gorsuch, 2004) proposed a fourth style of religious coping, called surrender, which is similar to the deferring prayer.

In spite of the wealth of knowledge on religious coping present in the literature, to the best of our knowledge, no research has been conducted on the use of religious coping by caregivers of differently-abled children.

## 2. Objectives

The objectives of the study here conducted were the following:

1. To understand if there were significant differences on religious coping among caregivers of differently-abled children that could be explained by the age of the caregivers and type of child disability.
2. To understand if there were significant differences on the dimensions of religious coping among caregivers of differently-abled children that could be affected by the age of the caregivers and type of child disability.

### 3. Hypotheses

The following constructs were hypothesized:

1. There were no differences on religious coping among caregivers of differently-abled children with respect to their age and type of child disability.
2. There were no differences on the dimensions of religious coping among caregivers of differently-abled children with respect to their age and type of child disability.

### 4. Methods

#### 4.1. Participants

Two hundred caregivers (108 fathers and 92 mothers) of differently-abled children were selected through a purposive sampling technique. Participants age ranged between 27 to 52 years old with a mean age of 40.03 years ( $SD = 5.31$ ; see Tab. 1 for details).

Table 1 – *Characteristics of the study population*

Variables	Group/Category	Frequency	Percentage
Caregiver Age	27-39	44	22.0%
	40-52	156	78.0%
Type of Disability	Autism	11	5.5%
	Visual Impairment	4	2.0%
	Cerebral Palsy	6	3.0%
	Down Syndrome	16	8.0%
	Intellectual Disability	71	35.5%
	Locomotor Disability	19	9.5%
	Microcephaly	5	2.5%
	Multiple Disability	42	21.0%
	Seizure Disorder	10	5.0%
	Speech Problems	16	8.0%

Caregivers (either the mother, father or both) whose child was less than 14 years old ( $M = 3.53$ ;  $SD = .86$ ) were included in the study. The data was collected from the Chotay Taray Foundation School (Rawat Pora Bhagat, Srinagar, India), the Composite Regional Centre (Bemina, Srinagar, India),

the Kamraz School for differently-abled children (Baramulla, Jammu and Kashmir, India) and the Zaiba Aapa Institute of Inclusive Education (Bijbehara, Anantnag, India). All the children with disability were identified by professionals (i.e. rehabilitation psychologies, clinical psychologists, etc.) working in the field of disability and rehabilitation.

#### 4.2. Measure used

##### 4.2.1. Religious Coping Scale

The Religious Coping Scale, developed by Gull and Husain (2020), was used to measure the religious coping among the caregivers of differently-abled children. The scale was developed for caregivers of differently-abled children irrespective of their cast, creed, culture, gender, and religion. This scale is an 18-item instrument, based on a 4-point ranked scale, ranging from “never” to “always”. In addition, the religious coping scale has five dimensions (see Tab. 2 and details in text). All the items were scored in a positive direction, i.e. from 1 to 4. Total scores ranged from 18 to 72. The higher the score, the more the subject tended to engage in religious practices and this applied to all the dimensions described. The internal consistency of the Religious Coping Scale for this study was  $\alpha = .84$ .

Table 2 – *Religious Coping Scale's structure and internal consistency*

Dimensions	Items	Number of items	Cronbach $\alpha$
Divinely Seeking	16, 15, 18, 17, 05	05	.80
Coping Attitude	04, 07, 06	03	.75
Religious Faith	01, 02, 03	03	.82
Sense of Possibility	14, 12, 13	03	.70
Spiritual and Social Support	09, 10, 08, 11	04	.77
Total		18	.84

The five dimensions were, as follows:

- *Divinely Seeking*: Islamic culture is based on God's support. Parents of disabled children perceive that Divinely seeking will be more beneficial in the recovery of the disability process of their children. It has been demonstrated that humans are likely to call out to God, when in distress or suffering from tribulations (Utz, 2011).

- *Coping Attitude:* For most Muslims Coping attitude is an important aspect of the coping process. Gathering in prayer and performing religious practices for the recovery of children's disability is a way of life for parents.
- *Religious Faith:* Religious faith leads to positive coping, where parents establish a connection with God, ask God for forgiveness of their sins and feel comfort in religion. The contents of the dimension suggest that these are the sources of every Muslim's faith and practice. Religious faith or belief impacts upon coping in a miraculous way.
- *Sense of Possibility:* Religious coping strategies offer a 'Sense of possibility' in parents of disabled children when all other remedies are lost. For believers, it translates in engaging/attending religious activities or services and in approaching God to provide a new direction in life.
- *Spiritual and Social Support:* Parents recognize the value of Spiritual and social support in coping with their children's disability. It is expressed in several ways, such as asking others to pray for the recovery of their children, providing spiritual support, seeking social support from family or friends, and seeking support from religious instructors.

#### 4.3. Procedure

A relationship was established with the parents prior to being exposed to the research study. The researcher first introduced him/herself and then explained the purpose of the research to the respondents. The participants were assured that their responses were strictly confidential and were thus used exclusively for the research purpose. A scheduled questionnaire was used in this study. The respondents were told to listen to the instructions carefully before responding to the questions. Each respondent took about 10-15 minutes to fill in the questionnaire.

### 5. Data analysis

The data was analyzed by using the SPSS 20 version. Welch's test, followed by a Post-hoc analysis (Games-Howell), were used to analyse the data. The level of significance was set at or below .05.

## 6. Results and discussion

The purpose of the study was to examine the difference on religious coping among caregivers of differently-abled children that could be explained by the age of the caregivers and type of child disability. A total of 200 caregivers, 108 fathers (54%) and 92 mothers (46%), was selected from various rehabilitation centers and special schools of the Kashmir Valley by applying a purposive sampling technique.

Table 3 highlights the significant differences on religious coping between the younger (27-39 years old) and older caregivers (40-52 years old).

Table 3 – *Comparison of younger (27-39 years old) and older (40-52 years old) caregivers on religious coping and its dimensions*

		Sum of Squares	df	Mean Square	F	Sig.
Divinely Seeking	Between Groups	15.52	1	15.52	5.98	.01
	Within Groups	513.50	198	2.59		
Coping Attitude	Between Groups	10.83	1	10.83	2.31	.13
	Within Groups	929.79	198	4.69		
Religious Faith	Between Groups	.60	1	.60	.61	.44
	Within Groups	195.15	198	.98		
Sense of Possibility	Between Groups	6.19	1	6.19	1.44	.23
	Within Groups	849.20	198	4.29		
Spiritual & Soc. Support	Between Groups	34.76	1	34.76	5.04	.02
	Within Groups	1364.99	198	6.89		
Religious Coping Scale	Between Groups	268.64	1	268.64	6.24	.01
	Within Groups	8518.95	198	43.02		

Table 4 – *Robust tests of equality of means*

		Statistic <sup>a</sup>	df1	df2	Sig.
Divinely Seeking	Welch	3.46	1	51.92	.07
Coping Attitude	Welch	2.25	1	67.68	.14
Religious Faith	Welch	.49	1	60.34	.49
Sense of Possibility	Welch	1.60	1	74.73	.21
Spiritual and Social Support	Welch	3.74	1	57.86	.06
Religious Coping Scale	Welch	5.16	1	61.31	.02

<sup>a</sup> Asymptotically *F* distributed.

As the results of the table suggest, significant differences were found among the caregivers concerning overall religious coping ( $F_{(1,198)} = 6.24, p = .01$ ) and its two dimensions, divinely seeking ( $F_{(1,198)} = 5.98, p = .01$ ) and spiritual and social support ( $F_{(1,198)} = 5.04, p = .02$ ). Welch's test was further conducted due to the substantial difference in variance and small sample size between the two groups (Tab. 4). The results from Welch's test indicate a significant difference between the two groups but only concerning overall religious coping ( $F_w = 5.16, p = .02$ ). The mean score of the older caregivers was higher as compared to the younger caregivers. This can be explained by the fact that younger caregivers are less religious as compared to older caregivers. When a child is diagnosed as disabled, young couples believe more on the expertise of professionals working in the field of disability for the recovery of their child besides having a religious faith. At a first stage, these young couples hope for their child's recovery but when they become aware of the fact that the chances of recovery are very low and that their child presents a lifelong disability, they become more religious and resort to praying to God for the well-being of their children. These religious practices increase with an increase in age. Our results are in line with the literature on psychology of religion, which shows a positive relationship between age and religiousness (Bengtson, Silverstein, Putney, & Harris, 2015; Murat, 2017).

Table 5 – *One way analysis of variance*

	Sum of Squares	df	Mean Square	F	Sig.
Between Groups	1138.48	9	126.49	3.14	.00**
Within Groups	7649.11	190	40.26		
Total	8787.59	199			

\*\* $p < .01$

Table 6 – *Robust tests of equality of means*

	Statistic <sup>a</sup>	df1	df2	Sig.
Welch	9.40	9	30.92	.00

<sup>a</sup> Asymptotically  $F$  distributed.

The results of the one way ANOVA, as presented in Table 5, also revealed a significant difference on religious coping among the caregivers of differently-abled children with respect to the type of child disability. The  $p$  value ( $sig.$ ) was .00, i.e. less than the minimum assumed level of

significance ( $p = .01$ ). Therefore, there was enough evidence to reject the null hypothesis number 2. Welch's robust formula was used to check the homogeneity of variance considering that the 10 child disability groups varied in terms of sample size (Tab. 6). The results from Welch's test also confirmed a statistically significant difference among the groups. A further pair-wise comparison was carried out by using the Games-Howell Post-hoc analysis (Field, 2009), as the sample size among the groups was very different (Tab. 7).

Table 7 – *Post-hoc analysis (Games-Howell). dependent variable: Religious Coping Scale*

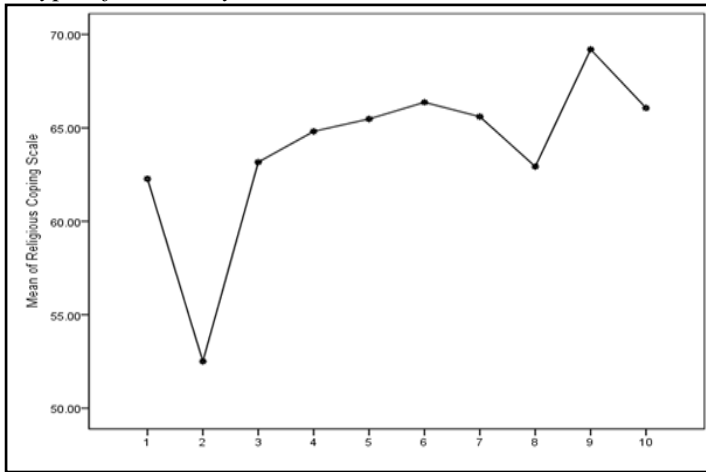
(I) Type of disability	(J) Type of disability	(I-J) Mean difference	Std. error	Sig.	95% Confidence interval	
					Lower bound	Upper bound
Autism	Visual Impairment	9.77*	2.18	.02	1.20	18.34
Down Syndrome	Visual Impairment	12.31**	2.47	.00	3.20	21.41
Intellectual Disability	Visual Impairment	12.97**	1.61	.00	4.76	21.19
Locomotor Disability	Visual Impairment	13.86**	2.04	.00	5.82	21.91
Microcephaly	Visual Impairment	13.10*	2.81	.04	.37	25.82
Multiple Disability	Visual Impairment	10.42*	1.76	.01	2.59	18.26
Seizure Disorder	Visual Impairment	16.70**	1.76	.00	8.69	24.70
Seizure Disorder	Multiple Disability	6.27**	1.44	.00	1.33	11.20
Speech Problems	Visual Impairment	13.56**	2.27	.00	4.99	22.12

\* The mean difference is significant at the .05 level; \*\* The mean difference is significant at the .01 level

The Post-hoc test revealed that religious coping among the caregivers with Autistic children or children with Cerebral Palsy, Down Syndrome, Intellectual Disability, Locomotor Disability, Microcephaly, Multiple Disability, Seizure Disorder and Speech Problems was higher as compared to caregivers with visually-impaired children (Tab. 7 and Fig. 1).



Figure 1 – *Type of disability*



Autism = 1; Visual Impairment = 2; Cerebral Palsy = 3; Down Syndrome = 4; Intellectual Disability = 5; Locomotor Disability = 6; Microcephaly = 7; Multiple Disability = 8; Seizure Disorder = 9; Speech Problems = 10

The mean score of caregivers of autistic children was higher on religious practices as compared to the mean score of caregivers having visually-impaired children (MI = 62.27; MJ = 52.50). The difference between the two groups was statistically significant (MD = 9.77;  $p < .05$ ). Likewise, the mean score of caregivers of children with down syndrome (MI = 64.81; MJ = 52.50), intellectual disability (MI = 65.48; MJ = 52.50), locomotor disability (MI = 66.36; MJ = 52.50), microcephaly (MI = 65.60; MJ = 52.50), multiple disability (MI = 62.92; MJ = 52.50), seizure disorder (MI = 69.20; MJ = 52.50) and speech problems (MI = 66.06; MJ = 52.50) was higher as compared to the mean score of caregivers of children with visual impairment. The differences between the groups were statistically significant (Tab. 7). The caregivers of visually-impaired children scored less on religious coping as compared to their counterparts. Moreover, a significant mean difference was found between the caregivers having children with seizure disorder and with multiple disabilities (MI = 69.20; MJ = 62.92). The former group scored higher on religious coping as compared to the latter.

Our results show that religious coping was a strategy applied by caregivers with children affected by all the kinds of disability tested here (9 out of 10 child disability groups), with the exception of caregivers having visually-impaired children who scored less. The results suggest that the former caregivers are more religious as compared to caregivers of children

with visual impairment. They perceive trusting in God and gathering in prayer as an effective coping strategy, which helps them to go through difficult caregiving situations. This may be due to the fact that children with such types of disabilities are more dependent on their caregivers as compared to children having a visual impairment. These results are in line with previous research (Stolley *et al.*, 1999; Leung & Li-Tsang, 2003; Malhotra & Thapa, 2015). Also depression was found to be significantly different between caregivers of differently-abled children and caregivers of normal children. Caregivers of children with disability scored a significantly higher mean score on depression as compared to caregivers having normal children (Roach, Orsmond, & Barratt, 1999; Hedov, Annere'n, & Wikblad, 2000). Further research should be promoted to explore if significant differences on depression occur among the caregivers of differently-abled children with respect to the type of child disability just as the results presented in this research appear to suggest for religious coping.

## 7. Conclusions

Caregiving in general is a very difficult task. Caregivers of differently-abled children worry about the future of their children. These caregivers can experience a range of emotions in response to their child's disability, which may include ambivalence, anger, denial, depression, hopelessness, fear of stigma, grief, guilt, shock, and even withdrawal. Most of these children are dependent on their caregivers for their "activities of daily living" and are subjected to "behavioral problems", which lead them to long-term cyclic physical and mental health problems. However, the quality and quantity of the problems incurred vary from caregiver to caregiver, depending on the type and level of severity of the child's disability. Caregivers use different strategies in order to buffer against the stressors and stressful life events of having a child with a disability, like emotional coping strategies, problem-focused coping strategies and religious coping strategies. Religious coping strategies, in particular, play a significant role when caregivers lose the hope for their child's recovery so that gathering in prayer and praying to God seem the ideal solution for the well-being of their children and to reach an inner peace for themselves.

## References

- Ano, G. G., & Vasconcelles, E. B. (2005). Religious coping and psychological adjustment to stress: A meta-analysis. *Journal of Clinical Psychology, 61* (4), 461-480.
- Aydin, N., Fischer, P., & Frey, D. (2010). Turning to God in the face of ostracism: Effects of social exclusion on religiousness. *Personality and Social Psychology Bulletin, 36* (6), 742-753.
- Bengtson, V. L., Silverstein, M., Putney, N. M., & Harris, S. C. (2015). Does religiousness increase with age? Age changes and generational differences over 35 years. *Journal for the Scientific Study of Religion, 54* (2), 363-379.
- Boudreaux, E., Catz, S., Ryan, L., Amaral-Melendez, M., & Brantley, P. J. (1995). The ways of religious coping scale: Reliability, validity, and scale development. *Assessment, 2* (3), 233-244.
- Carpenter, T. P., Laney, T., & Mezulis, A. (2012). Religious coping, stress, and depressive symptoms among adolescents: A prospective study. *Psychology of Religion and Spirituality, 4* (1), 19-30.
- Chong, L. T., Chong, M. C., Tang, L. Y., Ramoo, V., Chui, P. L., & Hmwe, N. T. T. (2019). The relationship between psychological distress and religious practices and coping in Malaysian parents of children with Thalassemia. *Journal of Pediatric Nursing, 48*, e15-e20.
- Ensel, W. M., & Lin, N. (1991). The life stress paradigm and psychological distress. *Journal of Health and Social Behavior, 32* (04), 321-341.
- Fernandez, A., & Loukas, A. (2014). Acculturation and religious coping as moderators of the association between discrimination and depressive symptoms among Mexican-American vocational students. *Journal of Immigrant and Minority Health, 16* (6), 1290-1293.
- Field, A. P. (2009). *Discovering statistics using SPSS, 3<sup>th</sup> ed.* Thousand Oaks: Sage.

Folkman, S., & Lazarus, R. S. (1984). *Stress, appraisal, and coping* (p. 460). New York: Springer Publishing Company.

Gull, M., & Husain, A. (2020). Reliability, validity, and factor structure of religious coping scale. *Iranian Rehabilitation Journal*, 18 (3), 301-308.

Harrison, M., Koenig, H. G., Hays, J. C., Eme-Akwari, A. G., & Pargament, K. I. (2001). The epidemiology of religious coping: A review of recent literature. *International Review of Psychiatry*, 13 (2), 86-93.

Hedov, G., Annere'n, G., & Wikblad, G. (2000). Self-perceived health in Swedish parents of children with Down's syndrome. *Quality of Life Research*, 9, 415-422.

Kes, D., & Aydin Yildirim, T. (2020). The relationship of religious coping strategies and family harmony with caregiver burden for family members of patients with stroke. *Brain Injury*, 34 (11), 1461-1466.

Krägeloh, C. U., Chai, P. P. M., Shepherd, D., & Billington, R. (2012). How religious coping is used relative to other coping strategies depends on the individual's level of religiosity and spirituality. *Journal of Religion and Health*, 51 (4), 1137-1151.

Leung, C. Y. S., & Li-Tsang, C. W. P. (2003). Quality of life of parents who have children with disabilities. *Hong Kong Journal of Occupational Therapy*, 13 (1), 19-24.

Malhotra, M., & Thapa, K. (2015). Religion and coping with caregiving stress. *International Journal of Multidisciplinary and Current Research*, 3, 613-619.

Maynard, E., Gorsuch, R., & Bjorck, J. (2001). Religious coping style, concept of God, and personal religious variables in threat, loss, and challenge situations. *Journal for the Scientific Study of Religion*, 40 (1), 65-74.

Murat, S. A. R. I. (2017). The impacts of the age factor on religiosity. *Firat Üniversitesi Sosyal Bilimler Dergisi*, 27 (2), 257-264.

- Pargament, K. I. (1997). *The psychology of religion and coping: Theory, research, practice*. New York: Guilford.
- Pargament, K. I., Smith, B. W., Koenig, H. G., & Perez, L. (1998). Patterns of positive and negative religious coping with major life stressors. *Journal for the Scientific Study of Religion*, 37 (4), 710-724.
- Pearce, M. J., Medoff, D., Lawrence, R. E., & Dixon, L. (2016). Religious coping among adults caring for family members with serious mental illness. *Community Mental Health Journal*, 52 (2), 194-202.
- Pearce, M. J., Singer, J. L., & Prigerson, H. G. (2006). Religious coping among caregivers of terminally ill cancer patients: Main effects and psychosocial mediators. *Journal of Health Psychology*, 11 (5), 743-759.
- Rathier, L. A., Davis, J. D., Papandonatos, G. D., Grover, C., & Tremont, G. (2015). Religious coping in caregivers of family members with dementia. *Journal of Applied Gerontology*, 34 (8), 977-1000.
- Roach, M. A., Orsmond, G. I., & Barratt, M. S. (1999). Mothers and fathers of children with Down syndrome: Parental stress and involvement in childcare. *American Journal on Mental Retardation*, 104, 422-436.
- Stolley, J. M., Buckwalter, K. C., & Koenig, H. G. (1999). Prayer and religious coping for caregivers of persons with Alzheimer's disease and related disorders. *American Journal of Alzheimer's Disease*, 14 (3), 181-191.
- Tix, A. P., & Frazier, P. A. (1998). The use of religious coping during stressful life events: main effects, moderation, and mediation. *Journal of Consulting and Clinical Psychology*, 66 (2), 411-422.
- Utz, A. (2011). *Psychology from the Islamic perspective*. Riyadh: International Islamic Publishing House.
- Wong-McDonald, A., & Gorsuch, R. L. (2004). A multivariate theory of God concept, religious motivation, locus of control, coping, and spiritual well-being. *Journal of Psychology and Theology*, 32 (4), 318-334.



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- Grossman, H. J. (Ed.). (1973). *A manual on terminology and classification in mental retardation* (Rev. ed.). Washington, DC: American Association on Mental Deficiency.
- Harter, M. R., & Aine, C. J. (1984). Brain mechanisms of visual selective attention. In R. Parasuraman & D. R. Davies (Eds.), *Varieties of attention* (pp. 293-319). New York: Academic Press.
- Lanktree, C., & Briere, J. (1991). *Early data on the trauma Symptom Checklist for Children (TSC-C)*. Paper presented at the meeting of the American Professional Society on the Abuse of Children, San Diego, CA.
- Piaget, J. (1967). *Biologie et connaissance*, Paris: Éditions de la Pléiade. (Engl. transl., *Biology and Knowledge*. Chicago: University of Chicago Press, 1971).
- Shallice, T. (1972). Dual function of consciousness. *Psychological Review*, 79, 383-393.

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