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Personality profiles of dyslexic children: a study with the Big Five Questionnaire

Antonella Gagliano¹, Rosamaria Siracusano, Maria Boncoddo, Tiziana Calarese, Giovanna Ilardo, Domenica Fidi, Rosa Grosso, Marco Lamberti, Massimo Ciuffo, Simona Rosina, Clemente Cedro & Eva Germanò

Abstract

This study explores the personality profiles of 65 dyslexic readers (36 male and 29 female, aged between 8 and 14), and 70 normal readers using the Big Five Questionnaire (BFQ), version for children. The BFQ assesses the constellation of the traits defined by the Five Factors Theory of Personality: Agreeableness (A), Emotional Instability (I), Open mindedness (M), Energy/Extroversion (E), Conscientiousness (C). Compared to the control children, subjects with dyslexia in our sample had lower scores in M, C and A dimensions. Overall, they showed personality traits characterized by less originality and creativity, poor control over emotional reactions, changeable moods and negative affections. The dyslexic readers who have benefited from an additional specialist tuition for a longer time, showed higher scores in I, A and C dimensions (p < 0.05). It seems that an additional specialist tuition could help children with dyslexia to develop personality dimensions such as conscientiousness, agreeableness, and a stronger emotional stability. Furthermore, the I dimension correlates (p < 0.05) with the age of diagnosis: the later the diagnosis is established, the more evident the emotional instability trait becomes.

The present study suggests that dyslexia and academic failure, in the long run, could affect negatively emotional experiences and personality traits.

Keywords: Dyslexia; Children; Big Five Questionnaire; Personality Traits.

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1. Introduction

Dyslexia and other specific neurodevelopmental disorders in childhood such as persistent difficulties in learning to efficiently read, write (dysgraphia) or perform mathematical calculations (dyscalculia) despite normal intelligence, conventional schooling, intact hearing and vision, adequate motivation and socio-cultural opportunity, are based on central nervous system dysfunction and are chronic lifelong conditions (Shaywitz, 1998; Demonet, Taylor, & Chaix, 2004). It has long been assumed that children with dyslexia and other learning disabilities may be highly vulnerable to emotional consequences and comorbid psychopathology, such as anxiety and depression (Willcutt & Pennington, 2000; Carroll & Iles, 2006; Goldston, Walsh, Mayfield, Reboussin, Sergent, Erkanli et al., 2007). In fact, dyslexic children can experience high levels of stress during their school careers because of their educational and social difficulties, and more specifically in their interactions with teachers when they worry over academic examinations and test performance (Alexander-Passe, 2008).

The relationship between dyslexia and anxiety seems to be strictly associated with shared environmental factors rather than with shared genetic risks (Whitehouse, Spector, & Cherkas, 2009). Difficulties in organizing lifelong emotional experiences related to living with diagnosed dyslexia and self-esteem problems may emerge by early childhood. People with dyslexia can contend with aspects of their learning difficulty that interfere with a typical development adding a sense of emotional insecurity (McNulty, 2003).

An association between reading difficulties and subsequent problem behaviours has been also described as a consequence of reading failure (Kempe, Gustafson, & Samuelsson, 2011). Furthermore, the comorbidity of dyslexia with attention deficit disorder/hyperactivity disorder (ADHD) is frequent (Willcutt, Betjemann, McGrath, Chhabildas, Olson, DeFries et al., 2010; Germanò, Gagliano, & Curatolo, 2010), and mostly impacts the outcome of dyslexia (Mignaini, Lassi, La Malfa, & Alberini, 2009; Nelson & Gregg 2012).

Various studies report the association between clinical conditions like dyslexia and ADHD group samples of the young with Personality Disorders, (mostly cluster B – dramatic cluster – borderline, histrionic, narcissistic and antisocial) (Bleiberg, 1984; Bleiberg, 1994). Conversely, some authors provided clear evidence to support the idea that neurodevelopmental disorders in children and teenagers can be associated with DSM-IV-Axis II disorders and, in particular, with the Borderline Personality Disorder (BPD) (Gardner, Lucas, & Cowdry, 1987; Coolidge, Segal, Stewart, & Ellett, 2000; Schulze, Domes, Krüger, Berger, Fleischer, Prehn et al., 2011).
According to Kemberg (1996), a variety of psychopathological profiles could be related to an underlying borderline organization. Furthermore, neuropsychological disabilities can play an important role in the genesis of some personality disorders (Kernberg, 1990; Tremblay, Richer, Lachance, & Côté, 2010).

However, in literature there is a debate about whether dyslexia is an isolated reading deficit or rather a broader deficit affecting cognitive, metacognitive and socio-emotional domains, which may affect the personality. Evidence on the relationship between dyslexia and emotional disturbances from a few previous studies is mixed. Some studies did not reveal any significant differences in the emotional status, in self-concepts and in personality, between subjects with dyslexia and control subjects (Plaisant, 1989; Lamm & Epstein, 1992; Zeleke, 2004; Swanson & Hsieh, 2009). Other studies have reported that students with learning disabilities frequently experience low self-esteem, with negative consequences on their emotional stability (Mason & Mason, 2005).

Overall, dyslexia is presumed to have an effect on the development of personality. Peculiar personality traits in samples of subjects with neurological disabilities, such as ADHD (Nigg, John, Blaskey, Huang-pollock, Willcutt, Hinshaw et al., 2002; Ferrer, Andión, Matalí, Valero, Navarro, Ramos-Quiroga et al., 2010), and dyslexia (Flynn, Matthews, & Hollins, 2002; Nelson & Gregg, 2012) have actually been described. Furthermore, some evidence sustains the association between dyslexia and conduct disorder and antisocial behaviour (Sundheim & Voelder, 2004; Trzesniewski, Moffitt, Caspi, Taylor, & Maughan, 2006) or antisocial personality disorder (Maughan, Pickes, Hagell, Rutter, & Yule, 1996; Rasmussen, Almvik, & Levander, 2001), suggesting that reading problems are a weak predictor of antisocial behaviour but could become a long-term predictor (Simonoff, Elander, Holmshaw, Pickles, Murray, & Rutter, 2004). Particularly for personality disorders, the integrated etiological model represents the most accredited paradigm to study their genesis. As regards the antisocial personality disorder, for example, the tendency for deviant behaviour is believed to derive from individual characteristics resulting from genetic, behavioural and cognitive variables that interact with environmental influences (Lahey, Waldman, & Mcbernett, 1999). The integrated model is presumably valid for any other psychopathological condition in developmental age.

2. Aim

The aim of the present work is to assess the personality traits of young subjects with developmental dyslexia through the description of the dimensions from the Big Five Questionnaire (BFQ) regarding personality.
The BFQ was designed to assess the constellation of the traits defined by the Five Factors Theory of Personality (Digman, 1990; Goldberg, 1990; McCrae & Costa, 1997; John & Srivastava, 1999). According to Bermúdez (1995), the “Big Five” model was established as the appropriate and integrating structure to describe personality in “natural” language in the context of personality questionnaires. This penta-factorial model represents a shared framework for the structure of personality. It interprets and catalogues the individual differences of personality. Moreover, the structure of this questionnaire provides a definition of the vocabulary used for the assessment, which can favour agreement between different examiners and can reduce the variation of their assessments. At the same time, the discrepancy between the evaluation provided by the children and that provided by the parents; that is, between the self-image as described by the individual and the image depicted by other people, decreases.

The present study describes the personality profiles of a group of dyslexic children and explores the connection between the personality traits and some demographic and environmental variables (age, school attendance years, age of diagnosis, additional specialist tuition). The study aims to discuss if dyslexia could have an impact on the development of specific personality traits.

3. Methods

3.1 Sample

We included in the study 65 subjects (36 male and 29 female), aged between 8 and 14 (mean age = 11.2, SD = 2.3). All children had been referred to the Department of Child and Adolescent Psychiatry of University of Messina for an assessment of their poor school performance (academic underachievement or failure). All of them had received a diagnosis of dyslexia (reading disorder) according to the criteria of DSM IV (Academic Skills Disorder; APA 1995), and had undergone a neuropsychological assessment including standardized reading and writing tests. The time of the diagnosis had been variable, occurring when the children were aged between 6 and 14 (mean age 8.8, SD 2.0). Each child was assessed on the complete form of the Wechsler Intelligence Scale for Children (WISC-III) (Orsini & Picone, 2006); the mean IQ was in the average range (Total IQ 97.8; SD 13.3). Subjects with IQ below 85 were not included, as well as those who showed major neurological signs and sensitive and/or sensory deficits. We also excluded all those subjects who, in association with the learning disorder, presented a psychopathological disorder, or had suffered from it in the last two years.
We finally considered for how long (number of years) the dyslexic readers had received an additional specialist tuition (specific training).

The group of subjects with Dyslexia was compared to a control sample, with equivalent age, sex, IQ level, and education level. This group was made up of 70 children (30 male and 40 female), aged from 8 to 14 (mean age = 11.0, SD = 2.1), recruited among school children attending schools in Messina, and examined at school. None of them had ever presented learning difficulties or psychopathological disorders. All the subjects in the control group had undergone a psycho-diagnostic assessment including the same tests carried out with the dyslexic subjects.

3.2 Instruments

We administered the children’s version (BFQ-C) of the Big Five Questionnaire (BFQ) by Caprara, Barbaranelli, Borgogni and Perugini (1993). The children’s version (BFQ-C, Barbaranelli, Caprara, & Rabasca, 1998) constitutes a bridge between developmental and life-long personality research. This questionnaire has 65 items and two forms: BFQ-children refers to the child him/herself completing the form, and BFQ-parents refers to a parent or caregiver completing the form. It is based on the Five Factor Model: Energy/Extroversion (E), Agreeableness (A), Conscientiousness (C), Emotional Instability (I), Open mindedness (M). Table 1 summarizes them describing the psycho-behavioural connotations.

In addition to BFQ-C, the psychopathologic scale, Self-Administration Psychiatric Scale for Children and Adolescents (SAFA), was administered. This instrument, produced and validated in Italy (Cianchetti & Sannio Fancello, 2001), is an unitary instrument that allows a preliminary but sufficiently broad assessment of psychiatric conditions by means of different scales that are organized according to homogeneous criteria. This battery includes different scales and explores a wide series of symptoms and psychiatric conditions in a short period of time (30-50 minutes). Each scale consists of two different versions calibrated according to the age (for children aged 8–10 and for pre-adolescents and adolescents aged 11–18 years). On the basis of the scores obtained, it is possible to delineate a general psychopathological profile or specific profiles using each scale (Franzoni, Monti, Pellicciari, Muratore, Verrotti, Garone et al., 2009). In fact, it includes scales for Anxiety (A), Depression (D), Obsessive-compulsive disorders (O), Eating psychogenic disorders, Somatic symptoms and hypochondria (P). Each item has three possible choices (true, false, and partly true), and can be valued from 0 to 2 according to specific correction schedules. The score of each scale can be converted into T scores using reference tables for age and sex ($T = 50 + 10Z$).
T scores define different ranges: < 30: the subject is not aware of his/her characteristics in the domain investigated; 30–39: the subject does not respond to the questions that explore the domain investigated; 40–59: statistical normality; 60–69: symptoms are basically pathologic; > 69: symptoms are clearly pathologic.

The items meet two criteria: they are sufficiently indicative of the disorder, and easily understandable and adequate to the age of the subjects.

The SAFA scale is commonly used in Italy to assess psychopathological symptoms in subjects from 8 to 18 years, and the structure and the content of the items are very strictly related to the culture of the country (Franzoni et al., 2009).

3.3 Procedure

Every child in both groups, and their parents, were individually administered the children version (BFQ-C) of the Big Five Questionnaire. The BFQ-C was filled in by the dyslexic readers and their parents, separately and individually, in an individual practice office of the hospital. The normal readers and their parents filled in the BFQ questionnaire in the school building, in a silent and comfortable room used for the assessment.

SAFA was self-administered and filled in individually by the child or teenager during the observation.

4. Data analysis

Data were analyzed using the SPSS package (version 17.0; SPSS Inc.). Differences between the two groups were evaluated using the analysis of covariance (ANCOVA) and effect size scores. Correlation calculations (Pearson coefficient) between BFQ-C scores, SAFA anxiety scores and demographic characteristics have also been carried out on the sample of dyslexic readers. Also, a multiple regression was applied between specific variables related.

SAFA mean scores of children with dyslexia were all below the cut-off value (69 points) for pathological conditions (see table 2). Even if the SAFA mean score for anxiety symptoms of children with dyslexia (49.8 points) was also below the pathological cut-off value, the comparison between dyslexic readers and controls highlighted a mild but significant difference between the two groups in the A Scale (Anxiety) of SAFA.

The comparison between participants and controls with BFQ-C showed significant differences in the two groups concerning some personality dimensions (see table 3).
In particular, dyslexic readers achieved different scores compared with controls in the M and C Scales (*Open Mindedness and Conscientiousness*), both in self assessment (BFQ-children) and in parent assessment items (BFQ-parents). A significant difference emerges also in the *Emotional Instability* dimension: but in this case dyslexic readers achieved different scores compared with controls only in the BFQ-parents. Vice versa, dyslexic readers obtained different scores compared with controls in the *Agreeableness* dimension only in the BFQ-children scale.

On the whole, subjects with dyslexia express lower *Open Mindedness, Conscientiousness* and *Agreeableness* as well as a higher level of *Emotional Instability*, compared with controls.

The correlations (Pearson coefficient) between the variables, so far examined within the group of subjects with dyslexia, allow us to point out some significant points (see table 4). The A scale of the SAFA correlates (*p* < 0.05 or *p* < 0.01) with some of the personality dimensions (*Energy, Agreeableness, Open mindedness* and *Emotional Instability*) noted by the BFQ self-assessment scale (BFQ children). This means that the scores related to these traits of personality are lower in the dyslexic reader with higher anxiety level.

The E and I dimensions of the BFQ-children scale, and the A dimension of the BFQ-parents scale correlate (*p* < 0.05) with age, in such a way that the older dyslexic readers seem to have improved capacities of *Energy/Extroversion* and *Agreeableness*. They also show a greater *Emotional Instability*. Even the years of school attendance correlate with the I-children and A-parents dimensions (*p* < 0.05); this means that while the *Agreeableness* (sense of cooperation, selflessness, sense of sharing, etc.) progressively grows during the school years, the *Emotional Instability* gradually decreases. Another relevant point is that children that benefited from additional specialist tuition (specific training) for a longer time (number of years), showed higher scores in the I and C dimensions of the BFQ children (*p* < 0.05) and in the A dimension of the BFQ-parents (*p* < 0.05). Therefore, it appears that the fact that they received a specific training helped these children in the process of developing personality dimensions such as *Conscientiousness* and *Agreeableness*, and this contributed in decreasing their *emotional instability*.

The I dimension of the BFQ-children correlates (*I* *p* < 0.05) with the age of diagnosis: the later the diagnosis is established, the higher the *Emotional Instability* assessed by the BFQ-children will result. This means that a delay in the diagnosis appears to be associated with an increased emotional instability in the sample of subjects we studied.

Using a multiple regression model, we tested the independent associations between age of subjects, school attendance years, age of diagnosis, specific training
as predictor variables, and “emotional instability” as criterion variables (see Table 5). For each multiple regression model, we entered age, school attendance years and age of diagnosis scores in block 1 and co-varied age, school attendance years, age of diagnosis and specific training in block 2. Age of diagnosis in block 2 ($\beta = -0.34 \ p < .05$) predicted the personality trait “emotional instability” only when associated to a specific training ($\beta = -0.30 \ p < .05$).

5. Discussion

The dyslexic readers, compared with the control children, show lower scores in the *Open Mindedness, Conscientiousness* and * Agreeableness* dimensions as well as high scores in the Emotional Instability dimension. On the whole, the subjects with dyslexia, compared with controls, seemed more emotionally unstable, less agreeable, less conscientious, less open-minded, and less capable to invest their own cognitive resources and abilities. As the study did not include subjects with psychiatric comorbidity, the description of the personality traits, compared to the overall psycho-affective features, could be more reliable. Therefore, this sample of subjects can, in our opinion, prove to be a good model for discussion if the presence of dyslexia shows demonstrative effects on the affective dimension of an individual, as much so as in shaping personality traits.

A preliminary part of evidence, shown by the differences of BFQ scores between the two groups, is that dyslexic readers tend to have different personality traits compared to typical readers. It seems clear, looking at the differences that appear in the M and C dimensions of the BFQ-children, that children with dyslexia perceive themselves as individuals who are not very open to experience, have a narrow range of cultural interests, and are not particularly original or creative. They describe themselves as unscrupulous, inaccurate and incapable of persevering in a task through to the end. It is significant that their parents describe them in the same terms, as is revealed by the presence of a significant difference with the control group also on the BFQ-parents scale. The full agreement between children and parents in describing this dimension allows us to consider this dimension as particularly impacted by dyslexia.

It is undoubtedly difficult to understand the reason for the presence of such personality characteristics. A first interpretation could be that the perception children have of their open-mindedness and of their skill to act with accuracy and precision, could be influenced by their family background. In other words, their parents, dissatisfied by their children’s insufficient school achievements, tend to form a wholly negative opinion about their children’s ability to use their
cognitive resources efficiently and precisely. It is well known that the parents of children with specific learning disabilities experience stress in coping with their child's condition (Karande, Mehta, & Kulkarni, 2007; Karande, Kum-bhare, Kulkarni, & Shah, 2009).

At the same time, it is probable that the children themselves, faced every day with the need to adapt to study assignments often inadequate for their skills, are bound to perceive themselves as not competent, and regularly experience a sense of ineffectiveness extending to all the activities involving a cognitive effort. What’s more, this self-perception about their ability and effectiveness is often greatly reinforced by the teachers’ and other educators’ attitudes. Many dyslexic readers have an unpleasant experience at school, and feel uncared for and unsupported by their teachers. It is possible to assume that dyslexia, both for its social consequence and as cause for academic failure, can strongly weaken the sense of the cognitive self. The M and C dimensions of BFQ-C could be connected to the concept of the cognitive self and strictly linked to the weakening process occurring when personality is developing and is consequently highly influenced by experience and, above all, by the internal perceptions of experience. Other evidence, revealed by the BFQ I scale (Emotional Instability) administered to the parents, is that the dyslexic group presents a lower ability factor to manage their emotional state, to check and adjust their affective reactions and hence to control their mood stability. It is no wonder that a cognitive self shaped by such conditions favours the stabilization of dysfunctional personality traits. But we also have to consider other plausible explanations. The first is that specific personality traits exacerbate reading difficulties. If a student is emotionally unstable, not very conscientious or open minded, and is not particularly able in using his/her cognitive resources on academic activities, he/she is more likely to fall further behind in reading. It is also possible to assume that cognitive factors, such as “Effortful Control”, are strongly related to the academic performance (Allan & Lonigan, 2011) and that dyslexic readers’ coping strategies can affect a child’s self-esteem and psychopathology as well as the remedial process (Alexander-Passe, 2006). Finally, it is possible that the specific personality traits of our sample do not demonstrate fundamental long term changes of personality, but rather artefacts due to different day-to-day experiences. In fact, the difficulties that children with dyslexia have when learning to read, can affect not only the child’s ability to write and spell, but can also have the secondary effect of precluding the satisfaction linked with the activity and the optimal energy allocation for academic tasks. This can then lead to a decrease in motivation and to an active avoidance of the activity, whenever it is possible.
One last difference between dyslexics and control group, in relation to BFQ-children, regards the A dimension, describing the ability of taking care and, at the same time, of trusting other people. It is probable that dyslexic children, because of their repeatedly frustrating experiences, have somehow lost the possibility to trust others to receive help and support. Another consequence could be the similar reduction of the sense of self-effectiveness and consequently the possibility to rely on themselves. In general, the differences between dyslexics and normal readers observed in the personality profiles allow us to describe dyslexic children as insecure and emotionally unstable subjects, who invest less in their own cognitive resources and in their own abilities.

Nevertheless, a recent study showed no differences in personality traits between participants with and without dyslexia (Tops, Verguts, Callens, & Brysbaert, 2013).

This conclusion is different from our results, but it could be due to the age differences in the groups selected for the sample (adults versus children), and to the different instruments utilized for assessing the personality traits. Furthermore, we can argue that our students were younger and living in a very different cultural environment. Actually, in Italy, the awareness of the needs of dyslexic students is lower than in northern Europe. Thus we can assume that our students’ academic careers have not been facilitated as well as Tops’s and coll. students’ careers, which have been reasonably successful in their university curriculum.

The analysis of correlations between the BFQ dimensions and the demographic and environmental variables (age, school attendance years, age of diagnosis, specific training) on the sample of dyslexic readers suggests some relevant issues. The main point is that younger children appear more anxious and emotionally unstable. We can assume that learning difficulties, however they are handled, affect the emotional level of the children more in their first years of schooling. During these years, learning is often a crucial issue both for the child and his parents, as this is the moment when inclinations and skills are tested for the first time. The natural tendency to cope with dyslexia and the widening of social activities, could reduce the sense of ineffectiveness of child’s cognitive resources in the long run, and lead to an improvement in the emotional stability dimension (I) of personality and a reduction of anxiety symptoms.

A further suggestion comes from the existence of a correlation between the A dimension of BFQ-parents and age: as a matter of fact, it seems that the parents notice a progressive opening to others and a greater willingness to trust others as their children grow up, suggesting that the personality trait of extroversion increases in older dyslexic children.
This could mean that they progressively acknowledge their difficulties or become more actively involved in overcoming them. A similar connection exists between the years of schooling, the Emotional Instability dimension of BFQ, and the Anxiety scale of SAFA. We could postulate that the increased awareness of coping strategies to face difficulties normally grows progressively during school years, leading to a more effective control over anxiety.

With respect to the management of academic difficulties, the dyslexic readers that have benefited from an additional specialist tuition for a longer period of time showed higher scores in I and C dimensions of the BFQ children and in A dimension of the BFQ-parents. It seems that the children who received an additional specialist tuition, turn out to be less inhibited and more accurate, scrupulous and persevering. It is possible that a reliable specialist tuition in the afternoon can facilitate the improvement of the skills and enhances the development of compensation mechanisms. This hypothesis can be inferred also from the multiple regression analysis that highlights the importance of the specific training as a predictor, and suggests that the age of diagnosis can predict the personality trait, defined as “emotional instability”, only when associated with a specific training. In other words, it is more likely that a child who receives an early diagnosis will receive specific supports and develop positive emotional traits. It seems, therefore, that in the sample under study, the additional specialist tuition makes the children more capable of self-regulation in the management of learning activities. Some personality traits, such as scrupulousness and the determination to tackle academic tasks, can only favour a more attentive approach to the task and can offer aid in accomplishing completion of school work. This remark is strongly in contrast with expectations, since common sense suggests that to provide supplementary learning makes children more dependant on the adults and less persevering. It must be taken into account that children with learning difficulties in combination with low conscientiousness and agreeableness are more likely to be referred to specialist services than conscientious, agreeable children with only learning difficulties. Nevertheless, support in recuperating study approach strategies, provided by the adult educator, seems to favour the achievement of autonomy in school work, most likely strengthening personality traits like self-regulation, accuracy, persistence and scrupulousness. It is also true that some personality types could respond better to after school tuition than others, achieving a more organized approach to the academic work.

However, the dyslexic children in our sample who received an additional specialist tuition, seemed also more open (A dimension of BFQ-parents) with a higher level of investment than others in terms of willingness and trust. A further indication comes from the correlation between the I dimension of BFQ children...
and the age of diagnosis, meaning that the later the diagnosis is made the more evident the emotional instability trait becomes. Looking into what has been previously mentioned in the general discussion about the potential micro-traumatic effect of continuous frustration at school and about the structuring of personality, we deem particularly interesting the emergence of this interdependency between a longer period when the disorder has not yet been diagnosed and this personality trait. Indeed, the Emotional Instability dimension describes children who are emotionally weak, anxious and less self-confident. These traits are commonly observed in dyslexic subjects who, due to the fact that they were not recognized as dyslexic, have been exposed for a longer time to wrong judgement, judgemental attitudes, discomfort and frustrations. Studying the correlation between the personality traits and the psychopathological symptoms, a strong correlation between the M dimension of personality (Open Mindedness), and the presence of symptoms of anxiety seems evident in the studied sample. A low sense of self-esteem, an inadequate sense of effectiveness and demoralisation concerning the possibility of achieving satisfactory results with their mental skills, could be closely connected to the occurrence of feelings of anxiety. Something similar can be said about symptoms of anxiety, since these, too, are correlated in our sample to the E, A and I dimensions of BFQ-children, to confirm the idea that the possibility of reducing frustrating and fearful situations can probably have a protective effect on the development of personality. In other words, if we help to put dyslexic children in a cheerful mood, we can guarantee higher emotional stability, as this is a strongly adaptive personality trait, since it implies a greater ability to control aggressive and negative drives, and to reduce negative affects in the approach to experience.

6. Conclusions

Even if we can’t conclude in a definitive manner that dyslexia has a causal role in the development of dysfunctional personality traits, we can still observe that subjects with dyslexia in our sample group show scarce openness to others, while at the same time their cultural interests, originality, creativity, and control over emotional reactions remain rather limited; their mood seems changeable and their affections are negative. This could be interpreted as a possible consequence of learning difficulties and academic failure that, in the long run, could affect, negatively, both social relations and emotional experiences, and create fixed schemes of psychic functioning. It is arguable that the association between dyslexia and peculiar personality traits is partly due to a chronic lifelong
condition characterized by traumatic and frustrating experiences. It usually happens that dyslexic children experience a sense of impotence, fear of punishment, sadness, lack of self-worth, and a sense of guilt virtually every day. They could also be exposed to a series of events that affect the feeling of emotional/affective closeness to the people they can relate to, a situation endured by dyslexic children especially when their family does not support them or even inflicts punishment. It is also possible that they may suffer from a micro-traumatic condition, when a series of situations that are not traumatic per se, repeatedly occur, and produce a temporary and subjective painful experience, as it is the case when dyslexic children are frequently reproached for their inadequacies. We could assume that hidden behind the façade is what we define as school distress or family discomfort, repeated social and relational micro-traumas capable of affecting the psycho-affective equilibrium and the development of the personality.

Further research is needed to clarify if those suffering from learning difficulties of a dyslexic nature can develop peculiar personality traits or disorders.

Our findings stress, once again, the importance of establishing a diagnosis as early as possible not only to facilitate the learning process, but also to safeguard the psycho-affective development of dyslexic children as a whole. A long-lasting rigid repetition of negative events could make everyday experience and perceptions of oneself and the others less flexible, and lead the individuals to develop peculiar traits of personality. In addition, this work underlines the importance of protective environmental factors, such as early diagnosis and good academic support, to reduce the potential negative impact of the dyslexia on the psycho-affective development of the individual. Therefore, organizing a supportive and understanding environment in touch with dyslexic children can be useful in guiding their psycho-affective development towards a harmonic structuring of their personality.

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Persons with multiple disabilities exercise adaptive head responses with the support of microswitch-aided programs

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Abstract

The present two studies were aimed at extending evidence on the effectiveness of microswitch-aided programs for promoting exercise of head movements through contingent stimulation with three participants (i.e., a man and two children) with multiple disabilities. The man and the child involved in Study I were to exercise head rotation movements from a lying or sitting position. The child involved in Study II was to exercise brief head lifting movements from a supine and a prone position. Tilt, optic, and pressure microswitches were used for the three participants, respectively. Performance of the target movements led to brief periods of preferred stimulation during the intervention phases of the studies. Data showed that the intervention frequencies of those movements increased for all three participants. Implications of the studies were discussed.

Keywords: Head responses; Technology; Response exercise; Multiple disabilities.
1. Introduction

Persons with severe and profound intellectual disabilities and pervasive motor impairment are generally confined to a static position, in a wheelchair or bed, and have virtually no chances of facilitating their interaction with environmental stimuli and caring for their general condition (Mechling, 2006; Lancioni, O’Reilly, Singh, Sigafoos, Oliva, Smaldone et al., 2009; Helton, 2011; Horn & Kang, 2012; Palisano, Chiarello, King, Novak, Stoner, & Fiss, 2012). In light of this situation, daily programs are mainly focused on providing them with general stimulation (environmental enrichment) and physiotherapy (Lancioni, O’Reilly, Singh, Sigafoos, Oliva, Antonucci et al., 2008; Chen, Klein, & Minor, 2009; Lancioni, Singh, O’Reilly, Sigafoos, Didden, & Oliva 2009; Tam, Phillips, & Murdford, 2011). General stimulation is considered to be critical to ensure a sufficient level of sensory input and eventually improve their mood (Green & Reid, 1999; Dillon & Carr, 2007). Physiotherapy is considered critical to help them practice correct motor schemes and responses that could provide physical benefits and slow down the frequently inevitable degeneration of their condition (Bobath, 1980; Ketelaar, Vermeer, Hart, Van Petegem-van Beek, & Helders, 2001; Damiano & DeJong, 2009).

An alternative approach to increase stimulation and foster exercise of correct movements or body schemes might involve the use of microswitch-aided programs, in which microswitch technology serves to (a) monitor the persons’ responses targeted for exercise and (b) ensure that the occurrence of those responses are followed by brief periods of positive stimulation (Lancioni et al., 2008; Lancioni, Sigafoos, O’Reilly, & Singh, 2012). Such programs would (a) underline the persons’ active role and (b) allow repeated periods of stimulation and response exercise with limited costs on staff time, provided that the technology available and the stimulation used are suitable to the persons’ condition (Kazdin, 2001; Holburn, Nguyen, & Vietze, 2004; Chantry & Dunford, 2010; Borg, Larson, & Östergren, 2011; Shih, Shih, & Shih, 2011; Tam et al., 2011).

Three recent studies have reported the use of microswitch-aided programs to provide stimulation for adaptive head movements with four participants with extensive multiple disabilities (Lancioni, O’Reilly, Singh, Sigafoos, Oliva, Campodonico et al., 2012; Lancioni, Singh, O’Reilly, Sigafoos, Oliva, Campodonico et al., 2012; Lancioni, O’Reilly, Singh, Green, Oliva, Campodonico et al., 2013). The present two studies were aimed at extending the evidence on the effectiveness of microswitch-aided programs for combining stimulation input and exercise of head movements with three new participants (i.e., a man and two children) with multiple disabilities. The man and the child involved in Study I were to exercise head rotation movements from a lying or sitting position. The child involved in Study II was to exercise brief head lifting movements from a supine and a prone position.
2. Study I

2.1 Method

Participants

The participants (Glen and John) were 18 and 8 years old, respectively, had congenital encephalopathy with limited residual vision (Glen) or functional residual vision (John). Glen was affected by pervasive motor impairment with spastic tetraparesis, hip displacement and severe scoliosis, and had a diagnosis of epilepsy partially controlled through medication. John had a diagnosis of severe spastic tetraparesis with dystonic movements. Both participants lived with their parents at home. Glen’s intervention program was supported by care and education staff, who provided him with physiotherapy sessions including head movements, sensory and tactile stimulation, and mild massage at his home. John attended a primary school, in which his daily program was similar to that described for Glen, except for a variety of physical and verbal interactions available from other children. Neither participant possessed specific forms of communication, and self-help skills or sphincteric control. They could not handle objects, and depended on others for any interaction with their surroundings. Their levels of intellectual disability had been estimated to be in the severe/profound range, and no formal assessments appeared feasible. Caution about their functioning was recommended, due to the fact that their motor condition prevented them from almost any form of expressed behavior. One of the goals of physiotherapy was to promote head movements/rotation to counter an increasing tendency to keep a static position. The participants, however, did not seem necessarily to enjoy the maneuvers required to ensure such movements/rotation, and could show signs of resistance and discontent. A technology-aided program that could motivate them to exercise their head and neck movements on their own (i.e., through contingent positive stimulation), rather than through external intervention was considered highly beneficial. Their families had signed an informed consent for their participation in this study, which had been approved by a scientific and ethics committee.

Positions, responses, technology and stimuli

Glen lay in bed throughout the sessions of the study while John sat in his wheelchair. Head rotation responses consisted of movements of the head of about 30 degrees (a) to the left (Glen) and (b) to the right or to the left (John). Rotations could start from a right head-turned position or other positions for Glen and from an approximately midway position for John. A series of tilt microswitches attached to a headband was used for Glen.
Optic microswitches (photocells) embedded in the wheelchair’s headrest served for monitoring John’s rotation movements. The microswitches were connected to a computer system, which served for recording the responses and regulating the presentation of preferred stimuli contingent on them during the intervention phases of the study (see below).

The stimuli selected for the study included songs, familiar voices, noises, lights, and vibratory inputs for Glen, and familiar voices, noises, lights, and vibratory inputs for John. The stimuli had been recommended by parents and staff and confirmed through a stimulus preference screening procedure (Lancioni et al., 2013). The screening procedure involved 10-20 non-consecutive presentations of brief samples of the stimuli. The stimuli were selected when both research assistants involved in the screening agreed that positive reactions (e.g., orienting and smiling) occurred for more than 50% of the sample presentations.

Experimental conditions

The study was carried out according to an ABAB design in which A represented baseline phases and B intervention phases (Barlow, Nock, & Hersen, 2009). A 3-week post-intervention check was also available. Sessions lasted 5 min for Glen and 10 min for John, and were implemented 3 to 11 times a day, depending on the participants’ availability conditions. The responses were automatically recorded through the computer system. Response prompting (i.e., verbal and physical guidance) was available prior to the start of the sessions as well as during the sessions if periods of non-responding of 30-60 s occurred. The responses performed during the sessions through prompting were subtracted from the computer count (i.e., by the research assistants in charge of the sessions). Interrater agreement on recording these prompt-related responses was checked in 15 sessions for each of the two participants. Agreement, which consisted of the two raters reporting the same number of prompting instances (which could also be zero), occurred in all sessions.

Baseline I and II. The baseline phases included four and eight sessions for Glen and four and nine sessions for John. The participants were provided with their microswitch and computer system, which recorded their responses, but did not present any stimulation contingent on them.

Intervention I and II. The intervention phases included 74 and 77 sessions for Glen, and 48 and 90 sessions for John. The first intervention phase was preceded by eight and seven practice sessions for the two participants, respectively. The practice sessions could involve frequent prompting from the research assistants to help the participants gain extensive experience of the responses and their stimulus consequences (i.e., 8–10 s of preferred stimulation). During the regular intervention sessions, (a) prompting occurred as indicated in the Experimental
conditions and (b) each response was followed by 8–10 s of preferred stimulation.

Post-intervention check. Both participants continued to receive sessions such as those available during the intervention phases. Fifteen of those sessions occurring 3 weeks after the end of the second intervention phase were used as post-intervention check.

Results

Figures 1 and 2 summarize the data for Glen and John, respectively. The bars represent mean frequencies of head rotation responses per session over blocks of baseline and intervention sessions. The number of sessions included in each block/bar is indicated by the numeral above it. During Baseline I, the participants’ mean frequencies of responses per session were below two and below five, respectively. During Intervention I, their mean frequencies of responses per session increased to approximately nine and 21. Baseline II showed a decline in responding, which was apparent for both participants. Intervention II showed mean frequencies of responses per session of about 10 and 24 for the two participants, respectively. The post-intervention check (not reported in the figures) showed that both participants maintained their intervention response levels. Response prompting was largely concentrated in the first intervention phase (and the first baseline).

Figure 1. Glen’s data. The bars represent mean frequencies of head rotation responses per session over blocks of baseline and intervention sessions. The number of sessions included in each block/bar is indicated by the numeral above it.
3. Study II

3.1 Method

Participant

The participant (Vicky) was 7 years old, had a diagnosis of encephalopathy with minimal residual vision, and extensive motor impairment related to prenatal and perinatal hypoxia. She presented with spastic tetraparesis and hip displacement, had epilepsy that was largely controlled through medication, and was estimated to be functioning at the severe level of intellectual disabilities. As for the participants of Study I, caution was recommended in this area due to the impossibility of formal evaluation and the confounding motor conditions. She lived at home with her family and attended daily a regular school, in which she was provided with treatment strategies involving general stimulation, social interaction, and physiotherapy. Physiotherapy objectives for Vicky focused largely on head movements, and head-neck strengthening from supine and prone positions. A technology-aided program that could motivate her to exercise her head and neck movements on her own (i.e., though contingent positive stimulation) rather than through external intervention, was considered highly desirable and profitable. Her family had signed an informed consent for her participation in this study, which had been approved by a scientific and ethics committee.

Positions, responses, technology and stimuli

Vicky was in (a) a supine position with her head slightly lifted on a special pillow during the sessions in which head movements from such a position were targeted, and in (a) a prone position on the same special pillow, with her head slightly turned to her right, during the sessions in which her movements from such a position were targeted. The microswitch to monitor her responses in both
positions was a pressure device that was activated as soon as Vicky’s head weight was momentarily lifted from such a device (i.e., due to the responses). The microswitch was connected to a computer system, which served for recording the responses and regulating the presentation of preferred stimuli contingent on them during the intervention phases of the study (as in Study I). The stimuli selected for Study II included songs, familiar voices, noises, lights, and vibratory inputs and were selected a in Study I.

**Experimental conditions**

The study was carried out according to a multiple probe design across responses (Barlow *et al.*, 2009). After the initial baseline, intervention focused on the head responses from a supine condition. Once the frequency of these responses had increased, a new baseline and intervention occurred on the head responses from a prone position. Increases of these latter responses led to alternating groups of intervention sessions focusing on one type of responses with groups of intervention sessions focusing on the other type of responses. Three weeks after the end of the intervention a post-intervention check was carried out. Sessions lasted 5 min and were implemented three to 12 times a day. The responses were automatically recorded through the computer system as in Study I. Conditions concerning response prompting (verbal and physical guidance of the responses) and interrater agreement on recording prompt-related responses matched those of Study I.

**Baseline I: responses from both positions.** Six sessions were carried out from the supine position and two sessions were carried out from the prone position. Each session included the microswitch for the responses targeted and the computer system that recorded the response occurrences. No stimulation was available for those occurrences.

**Intervention I: responses from the supine position.** Conditions were as in the baseline sessions carried out from the same position except that (a) each response occurrence was now followed by 10 s of preferred stimulation, and (b) five practice sessions were provided prior to the 68 regular intervention sessions (see Study I).

**Baseline II: responses from the prone position.** Three baseline sessions occurred. Conditions were as in Baseline I.

**Intervention II: responses from the prone position.** Conditions were as in the baseline sessions carried out from the same position except that (a) each response occurrence was now followed by 10 s of preferred stimulation, and (b) four practice sessions were provided prior to the 28 regular intervention sessions.

**Intervention III: responses from both positions.** This phase alternated Intervention I and Intervention II conditions (each lasting for a period of two to four
sessions). The phase included totals of 23 and 21 sessions for the two conditions, respectively.

*Post-intervention check: responses from both positions.* Vicky continued to receive sessions such as those available in Intervention III, after the end of that phase. Twenty-four of those sessions, occurring 3 weeks after the end of Intervention III, served as post-intervention check. Twelve of the sessions were carried out from the supine position and 12 were carried out from the prone position.

### 3.2 Results

Figure 3 summarizes Vicky’s data throughout the baseline and intervention phases of the study. White and gray bars represent mean frequencies of responses per session from the supine position and the prone position, respectively, over blocks of baseline and intervention sessions. The number of sessions included in each block/bar is indicated by the numeral above it. During Baseline I, Vicky’s mean frequencies of responses per session were below two from each position. During Intervention I, the mean frequency of responses per session from the supine position increased to nearly 10. During Baseline II, the mean frequency of responses per session from the prone condition remained low. During Intervention II, Vicky’s mean frequency of responses per session from the prone position increased to above 15. During Intervention III, the mean frequencies of responses per session from the two positions matched or exceeded those observed within the previous intervention phases. Comparable frequencies were also maintained during the post-intervention check (not reported in Figure 3). Response prompting was concentrated in the baseline phases and at the start of Intervention I and Intervention II.

Figure 3 - Vicky’s data. White and gray bars represent mean frequencies of responses per session from the supine position and the prone position, respectively, over blocks of baseline and intervention sessions. The number of sessions included in each block/bar is indicated by the numeral above it.
4. General discussion

The data of the two studies indicate that the use of the microswitch-aided programs enabled the participants to practice/exercise adaptive head responses independently. These data, which are in line with previous research outcomes in this area (Lancioni, O’Reilly et al., 2012; Lancioni, Singh et al., 2012; Lancioni et al., 2013), may be considered very encouraging. Indeed, technology-aided programs with positive stimulation contingent on head responses may prove highly beneficial for persons with multiple disabilities to improve their motivation to independently exercise responses that they (a) do not find simple or enjoyable per se and (b) may resent practicing when forced by others to do so (Dunst, Raab, Hawks, Wilson, & Parkey, 2007; Ripat & Woodgate, 2011). The same programs may represent the only option to provide the participants sufficient exercise time without excessive burdens on physiotherapists and other staff personnel (Nicolson, Moir, & Millsteed, 2012).

The independent exercise data seem to underscore that the participants (a) found the enjoyment of the stimulation more appealing than the effort required for the responses, (b) changed from a condition of passivity and external control to a condition of self-determination and initiative, and (c) discovered a resource that could help them increase their chances of slowing down motor degeneration, and improving their social image and, probably, their level of acceptance and quality of life (Brotherson, Cook, Erwin, & Weigel, 2008; Brown, Schalock, & Brown, 2009; Carter, Owens, Trainor, Sun, & Swedeen, 2009; Sunderland, Catalano, & Kendall, 2009).

New research in this area may need to target four different goals. First, additional participants should be involved in studies matching those carried out to determine the reliability and robustness of the findings reported and thus the general applicability of the intervention approach used (Kennedy, 2005). Second, the participants’ mood during the microswitch-aided sessions should be assessed to ascertain whether they display indices of happiness during their independent exercise (Dillon & Carr, 2007). Their mood during those sessions should then be compared with the mood they display during externally directed exercise sessions or physiotherapy sessions (Lancioni, Singh, O’Reilly, Oliva, & Basili, 2005; Dillon & Carr, 2007). Third, assessment of the possible benefits of independent exercise (e.g., in terms of the rates and ranges of movements produced) should be monitored over time within and across participants also in relation to the overall exercise time (Lancioni et al., 2013). Fourth, the opinion of parents, staff, and service providers about this approach should be gathered through social validation assessments (Callahan, Henson, & Cowan,
2008). Such an opinion could become an important guideline in possible revisions of the approach aimed at improving the quality of its impact (Lancioni et al., 2008; Lancioni, Singh, et al., 2012).

References


Abstract

The aim of the current study was to test the effectiveness of imagined contact – a mental simulation of a social encounter with an outgroup member – in improving humanity perceptions of individuals with intellectual disabilities (ID). A longitudinal study was conducted, examining non-disabled adults. Humanity perceptions were assessed by using uniquely human and non-uniquely human emotions. Results showed that a not fully human status was ascribed to people with intellectual disabilities. Imagined contact, however, reduced the humanity bias. Moreover, the positive influence of imagined contact was revealed also after one month, thus providing evidence of a long-term impact. Practical implications of results are discussed.

Keywords: Imagined contact; Individuals with ID; Humanity attributions; Primary and secondary emotions; longitudinal effects.

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1. Introduction

In this paper, we present a study aimed at investigating whether imagined contact (Crisp & Turner, 2012) – a mental simulation of an encounter with an outgroup member – may ameliorate humanness attributions to people with intellectual disabilities (ID).

Intellectually disabled: A stigmatized group

The participation of persons with disabilities, including those with ID, in the educational system, work, and community activities is strongly recommended by international social policies and legislations, which stress the importance of favoring the access of the disabled to all domains of public life (see, e.g., the Convention on the Rights of Persons with Disabilities, United Nations, 2006; The World Declaration on Education for All, UNESCO, 1990). Even so, individuals with ID still represent a stigmatized group, and face prejudice, negative stereotypes, and discrimination, which hinder acceptance and inclusion (see, e.g., Werner, Corrigan, Ditchman, & Sokol, 2012). People with ID are avoided, disregarded, and teased (Pratt, 2010); they are stereotyped as aggressive (Slevin & Sines, 1996; see also Crocker, Major, & Steele, 1998), and without perspectives of change (Jahoda & Markova, 2004); their skills are underestimated (Siperstein, Norins, Corbin, & Shriver, 2003). Prejudice and discrimination toward people with ID are pervasive phenomena (see, e.g., Tachibana & Watanabe, 2004; Akrami, Ekehammar, Claesson, & Sonnander, 2006; Siperstein Parker, Norins, & Widaman, 2011), generally spread across cultures (Siperstein et al., 2003; Scior, 2011).

Imagined contact: A strategy for improving intergroup relations

Under these circumstances, how is it possible to increment prejudice reduction toward persons with ID, thus favoring their social inclusion and well-being? One of the strategies most frequently investigated in social psychological research and implemented in practical interventions, is intergroup contact (Allport, 1954; Pettigrew, 1998). According to Allport’s original formulation, repeated interactions between members of different groups, under favorable conditions (cooperation, equal status, institutional support, and common goals), can reduce prejudice and promote positive attitudes toward the outgroup. Recent meta-analytic studies have well-documented its effectiveness, by considering many target groups, different contact settings, and different cultures. In addition, contact appears to be effective also in the absence of the optimal conditions singled out by Allport (see Pettigrew & Tropp, 2006; Pettigrew,
Tropp, Wagner, & Christ, 2011). Intergroup contact theory has been further investigated by Pettigrew (1998) and Brown and Hewstone (2005), and processes underlying the effects of contact on prejudice reduction have been highlighted (see, e.g., Pettigrew & Tropp, 2008). However, only a few studies have analyzed intergroup contact as an intervention to improve attitudes toward people with ID; in general, they indicate a positive relationship between contact and more favorable attitudes (see McManus, Feyes, & Saucier, 2010; see also the review by Scior, 2011).

Research and theory on contact have been broadened to include new forms of intergroup encounter that go beyond the face-to-face interactions between members of different groups. For instance, Wright, Aron, McLaughlin-Volpe, and Ropp (1997) proposed the extended contact hypothesis, according to which merely knowing that an ingroup member has a friendly relationship with an outgroup member improves the attitudes toward the outgroup. More recently, another form of intergroup contact—imagined contact—has been proposed by Crisp and Turner (2009): it consists in a mentally simulated interaction with an unknown member of the outgroup.

Research has shown that simply imagining a positive encounter with an outgroup member can have favorable effects on many aspects of the intergroup relationship (see Crisp & Turner, 2012). Imagined contact represents a powerful tool to improve intergroup relationships, especially when the opportunities to meet outgroup members are scarce, or when encounters are likely to cause high levels of uncertainty and anxiety. Mentally simulated contact can, therefore, constitute a form of pre-contact, particularly useful for getting people prepared for actual interactions with outgroup members. Actually, imagined positive encounters contribute to developing a mental script—a contact mindset—associated with favorable feelings; this mental script can promote the intentions to actually meet members of the target outgroup. In their integrated model, Crisp and Turner detailed key principles, moderator variables, and mediation processes of the effects of imagined contact on the outcome variables. One crucial mechanism is anxiety reduction, a common mediator in literature regarding contact (see Pettigrew & Tropp, 2006, 2008; see also Swart, Hewstone, Christ, & Voci, 2011). A second basic mediator is the availability of a positive mental script channeling future interactions with outgroup members.

Concerning the beneficial effects of imagined contact, many studies have demonstrated that mental imagery of contact with outgroup members stimulates not only more tolerance at an attitudinal level, but also stronger intentions to positively interact with the outgroup. In particular, imagined contact has been found to increase: positive explicit (see, Turner, Crisp, & Lambert, 2007)
and implicit attitudes (Turner & Crisp, 2010; Vezzali, Capozza, Giovannini, & Stathi, 2012), contact self-efficacy (Stathi, Crisp, & Hogg, 2011), intentions to engage in future contact (Husnu & Crisp, 2010), behavioral approach tendencies (Turner, West, & Christie, 2013), and positive nonverbal behaviors (Turner & West, 2012). Interestingly, and relevant to the present study, imagined contact can improve humanity perceptions (Vezzali, Capozza, Stathi, & Giovannini, 2012).

A variety of intergroup relations have been examined, such as: young versus old people, straight versus gay men, Muslims versus non-Muslims, ethnic majorities versus minorities, normal-weight versus obese people (see Crisp & Turner, 2012). This suggests again the effectiveness of the imagined contact strategy for promoting harmony between groups, as well as its applicability to many intergroup settings. It is worth noting that imagined contact can have a positive impact also in ameliorating attitudes and beliefs toward highly stigmatized groups, such as people with schizophrenia (West, Holmes, & Hewstone, 2011; Stathi, Tsantila, & Crisp, 2012), a group stereotyped as dangerous, threatening, and unpredictable (see Angermeyer & Matschinger, 2005).

The typical experimental task, in the imagined contact paradigm, consists in asking participants to engage for a few minutes in a mentally simulated positive encounter with an unknown member of the target outgroup. A crucial element in the instruction set is the positivity of the interaction: participants are asked to imagine themselves interacting with the target person in a positive, relaxed, and comfortable way. In order to strengthen the manipulation, after the mental simulation, participants are required to write a description of the imagined scenario. In the control condition, instructions ask participants to imagine, and then describe, a pleasant outdoor scene.

Several factors in the instructions can enhance the effects of imagined contact. For instance, the task of imagining a scenario rich in details (degree of elaboration; Husnu & Crisp, 2010), or that of closing eyes during the mental simulation (Husnu & Crisp, 2011). In addition, taking the perspective of a third person who, as a spectator, watches the encounter may lead to infer, from attitudes and emotions experienced during the interaction, more abstract self-traits, such as, being non-prejudiced or prosocial (Crisp & Husnu, 2011). Finally, imagining the interaction partner as a typical outgroup member may favor the generalization of the positive contact effects from the partner to the whole outgroup (Stathi et al., 2011). As an intervention strategy, imagined contact is easy to implement, and suitable for different intergroup settings. It may promote more harmonious intergroup relationships in educational and organizational contexts, also when the outgroup is a stigmatized group (Crisp & Turner, 2009, 2012).
2. Aims and hypotheses

The main aim of the current study was to explore whether imagined contact can enhance humanization of individuals with ID, and whether this effect can last a certain amount of time. Initial evidence concerning the disabled as a target is offered by Cameron, Rutland, Turner, Holman-Nicolas and Powell (2011). They showed that, among young children (5 to 10 years), imagined contact reduces prejudice against physically disabled peers. However, as far as we know, people with ID have never been considered in imagined contact research. We suggest that this form of contact can be a suitable intervention strategy, which can prepare the non-disabled to encounter individuals with ID when they enter a class, a working group, or a leisure setting.

Regarding the outcome of imagined contact, we decided to consider humanness attributions. Current research in social psychology has well documented a subtle humanity bias; namely, the tendency to ascribe a lower human status to the out-group than the ingroup. In their pioneering work, Leyens and colleagues (Leyens, Demoulin, Vaes, Gaunt, & Paladino, 2007) introduced the paradigm of primary/secondary emotions. Secondary emotions (e.g., hope, remorse) are unique to human beings; primary emotions, in contrast (e.g., pleasure, anger), are shared by humans and animals. Leyens et al. (2001) showed that people tend to ascribe more secondary emotions to the ingroup than the outgroup, while primary emotions are not differently assigned to the two groups. Subsequent empirical evidence has demonstrated that this effect, along with the tendency to perceive the outgroup in animalistic or mechanistic terms (see, e.g., Capozza, Boccato, Andrighetto, & Falvo, 2009; Loughnan, Haslam, & Kashima, 2009; Capozza, Andrighetto, Di Bernardo, & Falvo, 2012), represents a pervasive bias that may strongly damage the relationships between groups. Goff, Eberhardt, Williams, and Jackson (2008, Study 5), for instance, found that the activation in White participants of the Black/ape cultural association enhanced the justification of violence against Black targets. Similarly, Greitemeyer and McLatchie (2011) showed that the denial of humanness to others can lead to an increase in aggressive behaviors (see also Waytz & Epley, 2012); dehumanization can even enhance the willingness to torture prisoners of war (Viki, Osgood, & Phillips, 2013).

Some studies have analyzed whether direct intergroup contact and extended contact can reduce the humanity bias (see, Brown, Eller, Leeds, & Stace, 2007; Capozza, Falvo, Favara, & Trifiletti, 2013; Capozza, Trifiletti, Vezzali, & Favara, 2013). However, so far only one study has investigated the effects of imagined contact on humanity attributions (Vezzali, Capozza, Stathi, et al., 2012). In the present study, we test the hypothesis that imagining a positive encounter with an
individual with ID can ameliorate the humanity perceptions of the entire category of mentally disabled. We chose to analyze humanity attributions, rather than other forms of bias, because studies investigating stigma toward intellectual disability have mostly focused on attitudes and stereotypes (see e.g., Scior, 2011; Werner et al., 2012). Furthermore, attributions of ‘humanity’ are particularly relevant when dealing with this social category, because people with intellectual disabilities have been frequently denigrated over the past through animalistic rhetoric and metaphors comparing them to animals (see Haslam, 2006). In this study, we also test whether the positive effects of imagined contact may last in time. Long term effects would provide strong support to the efficacy of this intervention strategy.

Non-disabled adults were examined. A longitudinal experimental design was used: participants were examined twice with a one-month interval. The imagined contact versus control manipulation was introduced in the questionnaire administered at Time 1 (T1). The dependent variable, measured both at T1 and T2, consisted in the attribution of primary and secondary emotions to individuals with ID.

The hypotheses are the following.

**Hypothesis 1.** A not fully human status should be ascribed to individuals with ID, namely they should be assigned more non-uniquely human (primary) than uniquely human (secondary) emotions.

**Hypothesis 2.** The tendency to deny a fully human status to individuals with ID should be reduced by imagined contact, namely the greater attribution of primary versus secondary emotions should be lower in the imagined contact condition than in the control condition.

**Hypothesis 3.** The beneficial effects of imagined contact should persist at T2.

### 3. Method

**Participants and procedure**

Participants were 164 adults, mostly living in Northern Italy (96.3%). Females were 109; the age range was between 18 and 65 years (M = 31.49, SD = 13.71; age data were missing for two participants). Eighty-two participants were randomly assigned to the imagined contact condition, and 82 to the control condition.

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5It is worthwhile to note that generally people describe one’s group or category more in terms of secondary than primary emotions, namely more in terms of the characteristics which are distinctive of the human species than in terms of the characteristics that humans share with animals (see, e.g., Cortes, Demoulin, Rodriguez, Rodriguez, & Leyens, 2005, Study 1; Demoulin et al., 2004; Eyssel & Ribas, 2012; Leyens et al., 2001, Studies 1 and 2; Whol, Hornsey, & Bennett, 2012, Studies 1 and 5). This greater attribution of uniquely than non-uniquely human features may not concern the outgroups.
Participants, individually examined, were informed that they would have to answer a first questionnaire, which included an imagination task. After a month, they would have to fill out a second questionnaire. Participants were told that the aim of the study was to analyze intergroup attitudes; detailed information about the research would have been provided at the end of the experimental session at T2. Confidentiality of responses was guaranteed. After filling in the informed consent form, participants ran through the mental simulation task.

**Experimental manipulation (T1 Questionnaire).** Participants in the imagined contact condition were instructed as follows: “We ask you to imagine, for few minutes, that you meet for the first time a person with intellectual disability. Imagine that the interaction is positive and pleasant. During the encounter you notice some pleasant, interesting, and unexpected aspects about the person. When you are imagining, think in detail about the scenario in which the encounter occurs. We ask you also to imagine the scene by taking a third-person perspective; that is, you see yourself and the disabled person as in a movie. During the imagination task, try to keep your eyes closed. Please write in the following lines the imagined encounter, reporting as many details as possible.” In the control condition, instructions were: “We ask you to imagine, for few minutes, an outdoor landscape. Try to imagine the major aspects of the scene (e.g., is it a beach, a forest, are there trees, hills, what is on the horizon). We ask you to complete the imagination task keeping your eyes closed. Please write in the following lines the imagined scene, reporting as many details as possible.”

**Manipulation check (T1 Questionnaire).** As a check of the experimental manipulation, we tested whether concepts associated with ID were more accessible in the imagined contact than in the control condition. A word-fragment completion task was applied that required participants to produce a meaningful word from each fragment proposed (e.g., d w n). Seven word-fragments were presented in a list. For each of them, participants were instructed to add as many letters as they needed to form a meaningful word. We expected that words semantically linked to disability (e.g., d w n [down]; d b e [disabled]; h c p [handicapped]) would be more frequently evoked in the imagined contact condition than control condition. As a further manipulation check, we measured whether the imagined interaction had a positive tone. Four items, used only in the imagined contact condition, asked participants whether they had perceived the imagined interaction as: a) positive, and b) pleasant; whether, during the encounter, they had perceived: c) interesting, and d) unexpected aspects about the imagined target. A 7-step scale was used, anchored by definitely false (1) and definitely true (7) with 4 as the midpoint (neither true nor false). The four items were averaged to form a reliable composite score (α = .73).
MANOVA revealed no significant main effects for group and perceived control variables, but significant group x perceived control interactions for positive parenting \[F(1, 83) = 5.60, p < .05\] and inconsistent discipline \[F(1, 83) = 6.54, p < .01\], whereas for corporal punishment the interaction does not reach statistical significance \[F(1, 83) = 3.74, p = .06\]. The figures 1 – 4 present the 2 (group) x 2 (perceived control) results for these APQ domains. In the ADHD group, parents with low PCF resulted more positive (fig. 2) than those who report high PCF \[t(34) = 1.70, p < .05\], whereas the tendency is opposite for the non-clinical group where positive parenting was higher associated with high PCF \[t(49) = –1.99, p < .05\]. ADHD parents with low PCF result more inconsistent in discipline (fig. 3) than parents with high PCF \[t(34) = 2.00, p < .01\], whereas in the non-clinical group the disciplinary incoherence, expressed by higher scores in the APQ scale, is more common in parents with high PCF \[t(49) = –1.79, p < .05\]. Finally, for ADHD group the corporal punishment frequency (fig. 4) is higher with low PCF, with decreased levels with high PCF \[t(49) = –1.99, p < .05\], but this tendency is opposite for non-clinical parents where lowest levels link with high PCF \[t(49) = –1.95, p < .05\].

**Dependent measure: Humanity attributions (T1 and T2 Questionnaires).** In order to assess the humanity attributions, we used an emotion-based measure (Demoulin, Leyens, Paladino, Rodriguez, Rodriguez, & Dovidio, 2004; see also, Leyens et al., 2007). We used three positive (hope, pride, admiration) and three negative (remorse, shame, resentment) uniquely human emotions, and three positive (pleasure, excitement, surprise) and three negative (anger, pain, sadness) non-uniquely human emotions. The 12 emotions were randomly presented in a list including also 14 filler traits (e.g., shyness, generosity, cordiality). Participants were instructed to choose the items that better described individuals with ID; they could choose as many items as they wished. For each participant, two scores were obtained, corresponding to the number of primary and secondary emotions ascribed to individuals with ID. Two scores were obtained for T1 and two for T2.

**Prior contact (T1 Questionnaire).** Finally, in the T1 questionnaire participants were asked demographic information; they also answered an item measuring the quantity of their contact with members of the target category. Specifically, the item was: “How much contact do you have with people with intellectual disabilities?”; the 6-step scale was anchored by no contact (1) and very frequent contact (6). Very little (2), little (3), some contact (4), frequent contact (5) were the other steps of the scale.
4. Results

Manipulation check. In the word-fragment completion task, it was found that completions related to ID were higher in the imagined contact than control condition: $M = 2.38$ ($SD = 0.84$) vs. $M = 1.65$ ($SD = 0.91$), $t(162) = 5.36$, $p < .001$, respectively. Therefore, intellectual disability was more cognitively accessible to participants instructed to engage in the imagined encounter compared to participants who imagined the outdoor scene.

One of the key aspects of the experimental task in the imagined contact condition was the perceived positivity of the interaction with the outgroup member. Results showed that the perceived quality of the imagined encounter was high: $M = 5.65$ ($SD = 0.94$), the mean being significantly different from the midpoint of the scale, $t(81) = 15.99$, $p < .001$. Thus, in the imagined contact condition, participants viewed the interaction as positive and pleasant, and recognized interesting and unexpected characteristics in the imagined partner (for the four items separately, mean scores ranged from 5.32 to 5.87, all being different from 4, $ts ≥ 8.89$, $ps < .001$).

Prior contact. As a preliminary analysis we checked whether quantity of prior contact with people with ID was equivalent for participants assigned to the two conditions. Mean score for the single-item measure was $M = 2.89$ ($SD = 1.51$), in the imagined contact condition, and $M = 3.12$ ($SD = 1.49$), in the control condition. The two means were not different, $t < 1$, and prior contact with people with ID was rather infrequent for both groups of participants.

Effects of imagined contact on humanity attributions. For each participant, at both waves in each condition, an index of humanity bias was created corresponding to the difference between the number of primary emotions and the number of secondary emotions assigned to the target; the higher the positive score, the more people with ID are perceived more in terms of non-uniquely human than uniquely human attributes. The mean for the control condition was $M = 1.28$ ($SD = 1.66$) at T1, and $M = 1.21$ ($SD = 1.60$) at T2. The mean for the imagined contact condition was $M = 0.77$ ($SD = 1.46$) at T1, and $M = 0.84$ ($SD = 1.49$) at T2 (see Figure 1). The four means significantly differed from 0, $ts > 4.77$, $ps < .001$, indicating that more primary than secondary emotions were assigned to people with ID. This finding supports Hypothesis 1: in both conditions, at both waves, a tendency to assign a not fully human status to disabled persons was present.
In order to test the effect of imagined contact on the reduction of the humanity bias, a condition (imagined contact vs. control) x time (T1 vs. T2) ANOVA, with repeated measures on the time factor, was conducted. The main effect of condition was significant, $F(1.162) = 4.60, p = .03, \eta^2_p = .03$: the humanity bias was lower in the imagined contact, $M = 0.80$ (SD = 1.20), than in the control condition, $M = 1.24$ (SD = 1.41). As shown in Figure 1, both at T1 and at T2, the differential attribution of primary versus secondary emotions was lower in the imagined contact condition compared to the control condition. This finding supports Hypothesis 2. Thus, imagined contact was effective in reducing the tendency to deny a fully human status to people with ID. Neither the main effect of time nor the Time x Condition interaction were significant, $Fs < 1$, these findings indicating that the positive effects of the imagined encounter lasted for about one month, and were not dissipated once out of the experimental setting. Thus, also Hypothesis 3 was confirmed. In Figure 2, we report the findings distinguishing primary from secondary emotions, and collapsing the time factor. From the figure it appears that, in the imagined contact condition, there was a tendency to ascribe less primary emotions and more secondary emotions to individuals with disabilities compared to the control condition.
5. Discussion

The aim of the present study was to investigate whether imagined contact (Crisp & Turner, 2009) can be effectively implemented to improve the perception of people with ID. In particular, we aimed at testing the beneficial effects of imagined contact on humanity attributions. A sample of Italian adults was examined in a longitudinal experimental study (one-month interval between the two waves). The task of imagined contact at T1 required participants to mentally simulate a positive encounter with an individual with ID; in the control condition, the task was to imagine a pleasant outdoor scene. To measure the humanity bias, we calculated the difference between the number of primary and secondary emotions ascribed to the target category. Findings revealed a general tendency to assign a not fully human status to people with ID, who are perceived more in terms of non-uniquely human than uniquely human emotions. These humanity attributions (see also Capozza, Di Bernardo, Falvo, Vianello, & Calò, 2013) may be one of the factors which lead to the stigma experienced in society by individuals with ID (Scior, 2011; Werner et al., 2012).
More notably, we found that the tendency to deny the outgroup a fully human status can be reduced by the mental simulation of a pleasant interaction with an outgroup member, the positive effect of contact lasting at least one month after the imagination task. It is worth noting how the present study shows, for the first time, a one-month longitudinal effect of this form of contact. In the study by Vezzali, Capozza, Stathi et al. (2012), based on an intervention with a child sample, a shorter period effect was found. Our study, therefore, extends previous longitudinal results to an adult population in which humanity perceptions are presumably more difficult to change.

Our results have practical implications. Imagined contact can be easily implemented to equip people with more positive perceptions of individuals with ID for future encounters in schools, work settings, and health care institutions. Therefore, social policies designed to favor the inclusion of people with ID in community activities could fruitfully benefit from interventions based on imagined contact. The need for effective programs emerges, for instance, from a study (Siperstein, Parker, Norins, & Widaman, 2007) in which the attitudes of US students (middle school-aged) toward the inclusion of their ID peers in regular classrooms were examined. Results showed that, despite legislation and a long period of social policies, US students did not support the inclusion of students with ID and tended to avoid interactions with their intellectually impaired peers. Similar results have been found by Siperstein et al. (2011) among Chinese students.

Likewise, for mentally ill people, direct contact with individuals with ID may cause high levels of anxiety and uncertainty. Thus, interventions based on imagined contact, rather than face-to-face contact, can represent a first step in stigma reduction, just because imagined contact creates a positive contact mindset which favors future interactions. In their research, Stathi et al. (2012) found that the mental simulation of a positive encounter with a schizophrenic person reduced negative stereotypes toward the whole group while enhancing intentions for actual encounters; these effects were mediated by reduced intergroup anxiety.

Research on direct and extended contact has widely demonstrated the crucial role played by the affective factors in reducing intergroup prejudice (Tropp & Pettigrew, 2005; Pettigrew & Tropp, 2008; see also Capozza, Falvo et al., 2013). Affective factors are reliable mediators also when imagined contact is used (for anxiety, see, e.g., Hunsu & Crisp, 2010; Turner et al., 2013); for trust, see Turner et al., 2013). In the study in which humanity attributions were used as the outcome (Vezzali, Capozza, Stathi et al., 2012), the effects of imagined contact were mediated by outgroup trust. Future research should test whether also reduced anxiety and enhanced empathy may explain the relationship between imagined contact and outgroup humanization. Which emotions are influential mediators likely depends on the target outgroup.
Another task of future research is to assess whether imagined contact can be successfully used to increase the attribution of uniquely human features (e.g., secondary emotions) to people with ID. As suggested by Capozza, Di Bernardo et al. (2013), the humanization of individuals with ID could be favored by the implementation of an imagined contact condition, in which the imagined person with ID feels secondary emotions. This strategy could generate an association between people with ID and uniquely human characteristics (for how to generate humanizing sentiments toward disabled persons, see also Haslam, 2006, p. 253).

A limitation of the study is that only one operationalization of the humanity attributions has been used. In future research, findings should be replicated by using other measures. For instance, trait-based measures or implicit tasks such as an adaptation of the Implicit Association Test (IAT; Greenwald, Nosek, & Banaji, 2003). Another limitation is that we used as target the general category of individuals with ID. Future studies should explore whether imagined contact is differently effective when targets have different degrees of intellectual impairment.

Humanization of individuals with ID may favor cooperation with these persons, reduce discrimination against them, and lead to more spontaneous approach tendencies (Capozza, Di Bernardo et al., 2013), which can pave the way toward friendly and cooperative encounters.

References


The Expressed Emotion of mothers of children with Fragile X Syndrome

Jeanine M. Coleman¹ & Karen Riley²

Abstract

Mothers of children with fragile X syndrome (FXS) face many challenges. FXS is a systemic disorder that includes cognitive, physical and psychological impairments along a continuum. However, behavioral issues remain the most challenging for most families (Epstein, Riley, & Sobesky, 2002). In contrast to mothers of children with other neurodevelopmental disorders these women have the additional challenge of a distinct phenotypic expression of their premutation carrier status, which presents its own psychological emotional challenges. The complex nature of the individuals affected by FXS and thus the interesting implications for the mother child dyad, make this population one of great interest. The purpose of this study is to explore the Expressed Emotion of mothers of children with FXS. Expressed Emotion is a construct of the familial emotional climate in which a parent (or close relative) expresses emotional involvement, hostility and/or criticism about their child (McCarty & Weisz, 2002). The primary way to measure expressed emotion is the Five Minute Speech Sample (FMSS) (Magana-Amato, 1993), which is coded through a non-scripted monologue by one of the parents. The FMSS provides a novel way of looking at the mother-child dyad in the FXS community, and provides information to guide intervention for new mothers with young children with FXS. Without pathologizing the mothers of children with FXS, the findings of this study provide insight into the expressed emotion of mothers and of children with FXS that could lead to valuable intervention strategies developed by parents and professionals. As this type of research has never been conducted generally with mothers of children with neurodevelopmental disorders or specifically with those with FXS, this study was conducted as a pilot to explore both accuracy of the tool and to gain insight into this very complicated dyad.
Keywords: Fragile X Syndrome (FXS); Five Minute Speech Sample (FMSS); Expressed Emotion.

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1. Introduction

Identifying how mothers express their emotions about their children diagnosed with Fragile X syndrome (FXS) is crucial to supporting mothers in their developing relationships with their children. Parenting a child with significant intellectual disabilities such as FXS, can lead to excessive worry and unrealistic expectations, amplifying the stress of every day caregiving (Weiss, Sullivan, & Diamond, 2003). This stress influences the mothers’ abilities to express warmth and care for their children, regardless of their feelings of love for their children (Bailey, Skinner, & Sparkman, 2003; Johnston, Hessel, Blazey, Eliez, Erba, Dyer-Friedman et al., 2003; Boger, Tompson, Pavlis, Briggs-Gowan, & Carter, 2008). The interaction between maternal stress and lack of maternal warmth is fundamental to the construct of emotional family climate, called Expressed Emotion (Vostanis & Leff, 1995). Combining the factors of emotional family climate and parenting a child with FXS was the purpose of this research.

Expressed emotion

Expressed Emotion is a complex construct that focuses on parents’ verbal and nonverbal expressions about their children and the emotional quality of the relationship (Brown, Birley & Wing, 1972; McCarty & Weisz, 2002; St. Jonn-Seed & Weiss, 2002; Calam & Peters, 2006). This construct was first used with families with adult children with mental health disorders, as a longer interview process, called the Camberwell Family Interview (CFI) (Vaughn & Leff, 1976). Expression of criticisms, excessive worry, and dissatisfaction of family members’ behavior are primary elements of Expressed Emotion (Magana-Amato, 1993; McCarty & Weisz, 2002). Brown, Birley and Wing (1972) identified several affective variables that became important components of Expressed Emotion, which were associated with symptomatic relapse of psychiatric disorders, and based on six components of the relatives’ expressed thoughts and feelings about a patient with schizophrenia: 1) critical comments; 2) hostility; 3) dissatisfaction; 4) warmth; 5) emotional over-involvement and 6) an overall index of the relative’s Expressed Emotion. The salient issue of Expressed Emotion is the balance of these affective variables (Brown & Rutter, 1966).

Fragile X syndrome (FXS)

FXS is the most common cause of inherited intellectual disabilities. This claim has been supported by a body of research that has been conducted over the past 25 years (Cornish, Turk, & Hagerman, 2008). More recently, the understanding of FXS has evolved and is viewed as a “continuum of gene effects”, resulting
in a spectrum of disorders for the whole family (McConkie-Rosell, Finucane, Cronister, Abrams, Bennett, & Petterson, 2005). FXS manifests itself in many behavioral, learning, and sensory challenges for the diagnosed individual and his/her family. There are also a number of health challenges related to FXS that include seizures, connective tissue dysplasia, vision, and cardiac problems (Hagerman, 2006a). Of interest for this particular study are two separate groups; individuals with fragile X syndrome and females with the premutation. Males with FXS, an expansion of the CGG repeat sequence on the X chromosome exceeding 200, present with overall developmental delays, significant cognitive impairment, sensory processing issues, anxiety and behavioral issues including but not limited to aggression. FXS is the leading known genetic cause of autism, and approximately 30 percent of all children with FXS demonstrate autistic-like characteristics (Bailey Jr, Hatton, Skinner, & Mesibov, 2001; Hagerman, 2006b; Hagerman, Rivera, & Hagerman, 2008). Females with FXS present with similar patterns with less involvement. Females with the premutation, or carrier females have a CGG expansion ranging from 30-200. Although development and cognition remain within typical limits these women do present with a consistent phenotypic profile, including some difficulties with math, anxiety, depression and characteristics of obsessive compulsive disorder (McConkie-Rosell et al., 2005; Hagerman et al., 2008). These women are typically highly functional, but struggle internally with their own organic issues as well as those associated with having a child or children with significant delays.

2. Aims & Hypotheses

The purpose of this pilot study was to explore the construct of Expressed Emotion with mothers of children with FXS, evaluate the use of this tool with families with children with neurodevelopmental disorders, specifically FXS, and to explore the complex dyad between carrier status mothers and their affected children. Maternal stress, emanating from lack of information and resource scarcities, arises because there is limited public awareness of FXS, and diagnosis may be delayed until the child is 3-4 years old, due to its low incidence in general populations (Guralnick, 1998; Hagerman, 2002; Skinner, Sparkman, & Bailye, 2003; Bailey, 2004; Hagerman, 2006a; Cornish et al., 2008; Hagerman, Berry-Kravis, Kauffman, Ono, Tartaglia, & Lachiewicz, 2009). Prior to diagnosis, families may have experienced unsettling behaviors (i.e. excessive tantrums, feeding, sleeping problems) and developmental delays in their children, which may have led to growing concerns and misdiagnoses.
(Hatton, Hooper, Bailey, Skinner, Sullivan, & Wheeler, 2002; Hagerman, 2006a). Ultimate accurate diagnosis may result in some sense of relief; however, it also generates new stressors related to family history and family planning due to the X-linked inheritance factors (Bailey, Skinner, Hatton, & Roberts, 2000; Bailey, 2004). In addition to the many stressors of raising children with significant intellectual and behavior disabilities, mothers of children with FXS have the emotional burden of passing on the gene mutation to their children.

There is a paucity of research on the relationship between mothers and their children with FXS, let alone research within a specified area of intellectual disabilities and Expressed Emotion. Laghezza, Mazzeschi, Di Riso, Chessa and Buratta (2010) reported in their review of literature that there is a lack of research on Expressed Emotion and children with intellectual disabilities. This information, coupled with the research on Expressed Emotion, led to the research questions for this exploratory study. It is hypothesized that the Five Minute Speech Sample (FMSS) is an appropriate tool for measuring expressed emotion (EE) of premutation carrier mothers of children with FXS. Additional research questions include defining the expressed emotion (EE) of mothers of children with FXS, and exploring the mother child dyad in this specific population (Magana-Amato, 1993).

3. Methods

3.1 Sample: Mothers, Children, and Families

Due to the low-incidence of FXS and the exploratory nature of the study, the inclusion criteria was broadly defined as any mother with a child with the full or partial mutation of the FMR1 gene, regardless of age, gender, or culture/language. Every effort was made to recruit participants that reflect diverse cultural, economic, and familial systems, although the diversity of participants was limited.

Thirty-four mothers completed the demographic questionnaire, although the entire sample for the study involved 35 mothers. Within the demographic questionnaire, the mothers provided information about themselves, their children, and families. A majority of the mothers were between the ages of 31-50 years, married, with a family household income exceeding $60,000 per year. Many of the mothers reported that they were college graduates and chose to be stay-at-home parents. Culturally and ethnically, there was little diversity in the sample, with a majority of the mothers self-selecting their race as “Caucasian”. Even though the sample was not culturally or ethnically diverse, it was geographically diverse, as the sample included mothers from across the United States and one from South America.
The children with FXS, as reported by their mothers, had a mean age of 13 years with a range of 2-40 year. Additional child information collected from the mothers included, gender, children’s ethnicity, FXS diagnosis, age at diagnosis, and additional diagnoses. Seventy-six percent of the children were male and 24% were females (Table 1). It was necessary to exclude age as a significant factor that could confound results because of the wide range of children’s ages in this sample. Please see further explanation in the analysis section.

As for the diagnosis at FXS, 95% of the mothers reported that their child was diagnosed with the full mutation, while two mothers reported a mosaic status or partial mutation. The mean age at diagnosis was 4.8 ($SD = 6.4$) corroborating earlier research that indicated that the average age of FXS diagnosis is under 5 years of age (Bailey, 2004; Hagerman, 2006a). The age of diagnosis ranged from six months to 32 years of age, with a clear association between the younger children in the sample and a younger age of diagnosis.

40% of the mothers reported that their children had two or more diagnoses. Other diagnoses included sensory processing disorder, Autism spectrum disorder (ASD), attention deficit hyperactivity disorder (ADHD), and learning disabilities.

Table 1 - Profile of Children with FXS

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Mean</th>
<th>Standard Deviation</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age (n=37)</td>
<td>13.2</td>
<td>9.2</td>
</tr>
<tr>
<td>Age of FXS Diagnosis (n=34)</td>
<td>4.8</td>
<td>6.4</td>
</tr>
<tr>
<td>Gender (n=37)</td>
<td>Frequency</td>
<td>Percent</td>
</tr>
<tr>
<td>Male</td>
<td>28</td>
<td>76%</td>
</tr>
<tr>
<td>Female</td>
<td>9</td>
<td>24%</td>
</tr>
<tr>
<td>Ethnicity (n=34)</td>
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<td></td>
</tr>
<tr>
<td>Latino/Hispanic</td>
<td>1</td>
<td>3%</td>
</tr>
<tr>
<td>African American</td>
<td>1</td>
<td>3%</td>
</tr>
<tr>
<td>Caucasian</td>
<td>32</td>
<td>94%</td>
</tr>
<tr>
<td>FXS Diagnosis (n=37)</td>
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<td></td>
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<tr>
<td>Full Mutation</td>
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<td>95%</td>
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<tr>
<td>Partial Mutation or Mosaic</td>
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<td>5%</td>
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<tr>
<td>Additional Diagnoses (n=33)</td>
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<tr>
<td>Autism Spectrum Disorder</td>
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<td>12%</td>
</tr>
<tr>
<td>Sensory Processing Disorder</td>
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<td>6%</td>
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<tr>
<td>ADHD</td>
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<td>18%</td>
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<td>2 or more diagnoses</td>
<td>13</td>
<td>40%</td>
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<tr>
<td>No other diagnoses</td>
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<td>24%</td>
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</tbody>
</table>
Within the families of the participants, 56% of the mothers reported that they had one other child, 24% reported that they had two or more children, and 20% had no other children. Of the families with other children, 41% said they had another child with FXS in their family. When asked what other transitions their family was experiencing at the time of data collection, many families reported that they were in the middle of Individualized Education Plan (IEP) reviews, additional diagnoses, and/or other family changes. Sixty-eight percent of the mothers also said that they were involved in or receiving family support through local or national FXS groups (Table 2)

Table 2 - Profile of Families with Children with FXS (N = 35)

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Frequency</th>
<th>Percent</th>
</tr>
</thead>
<tbody>
<tr>
<td>Other children in the family</td>
<td></td>
<td></td>
</tr>
<tr>
<td>No additional children</td>
<td>7</td>
<td>21%</td>
</tr>
<tr>
<td>1 additional child</td>
<td>19</td>
<td>56%</td>
</tr>
<tr>
<td>2 additional children</td>
<td>6</td>
<td>18%</td>
</tr>
<tr>
<td>3 additional children</td>
<td>2</td>
<td>6%</td>
</tr>
<tr>
<td>Other children with FXS</td>
<td>14</td>
<td>41%</td>
</tr>
<tr>
<td>Transitions</td>
<td></td>
<td></td>
</tr>
<tr>
<td>IEP Review</td>
<td>8</td>
<td>24%</td>
</tr>
<tr>
<td>Medical or educational diagnosis</td>
<td>3</td>
<td>9%</td>
</tr>
<tr>
<td>Birth of a sibling</td>
<td>1</td>
<td>3%</td>
</tr>
<tr>
<td>Parental change (i.e. employment/re-location)</td>
<td>3</td>
<td>9%</td>
</tr>
<tr>
<td>2 or more transitions</td>
<td>13</td>
<td>39%</td>
</tr>
<tr>
<td>No transitions</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Involvement of FXS Support Group</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>23</td>
<td>68%</td>
</tr>
<tr>
<td>No</td>
<td>11</td>
<td>32%</td>
</tr>
</tbody>
</table>

3.2 Instruments

The Five Minute Speech Sample (FMSS) (Magana-Amato, 1993, 2002) is a standardized instrument used to rate the Expressed Emotion (EE) of a relative towards an individual within his or her family. Expressed Emotion is the primary
construct measured by the FMSS. “The Five Minute Speech Sample (FMSS) EE coding system distinguishes the nature of the respondent-relative’s environment by identifying emotions, feelings and attitudes expressed by a respondent about a relative during a 5-minute monologue” (Magana-Amato, 1993, 2002, p. 4). The interviews were transcribed and analyzed for content and tone.

There are five scoring categories of the FMSS that include Initial Statement, Relationship, Criticisms, Dissatisfaction, and Emotional Over-involvement. The initial statement and the relationship were scored discretely as positive, neutral, or negative. The subscale scores include Emotional Over-involvement (EOI) and Criticism (CRIT), which are coded as LOW, HIGH, or BORDERLINE; and the final EE score is dichotomous, coded as either LOW or HIGH (Magana-Amato, 2002).

The guidelines provide extensive coding instructions for each of the categories with multiple caveats and examples. For example, there are guidelines for administration concerning the physical setting, adequate equipment, and verbatim instructions. In order to maintain reliability of instruction, the administrator reads following instructions read aloud to the participant:

“I’d like to hear your thoughts and feelings about (relative’s name), in your words and without my interrupting with any questions or comments. When I ask you to begin I’d like you to speak for 5 minutes, telling me what kind of person (relative’s name) is and how the two of you get along together. After you begin to speak, I prefer not to answer any questions until after the 5 minutes are over. Do you have any questions before we begin?” (Magana-Amato, 2002).

In addition, other considerations include answering questions before, during, and after the procedure; and actions the examiner should not take, such as limiting eye contacts, vocalizations or leading prompts (Magana-Amato, 2002). The guidelines provide extensive direction for coding independently and for interrater reliability, although no reliability or validity statistics are presented. Several studies have been implemented to document the reliability and validity of the FMSS with both mothers and fathers.

Beginning with two studies by Magana and colleagues (1986), in which they documented the consistency of the FMSS with the longer, and already established Camberwell Family Interview (CFI). Results of the first study revealed agreement between three raters of the FMSS and CFI classifications (Kappa statistic -.70, .70, .80), as well as a significant correlation between “proportion of variance shared by the two systems” ($r = .38$, $df = 49$, $p < .005$) (Magana, Goldstein, Karna, Miklowitz, Jenkins, & Falloon, 1986). The second study was not only a replication of the first, but also encompassed another language component, with a Mexican-American, Spanish-speaking sample. The results were very similar to the English-speaking sample.
and revealed that, “A person who is classified as high EE on the FMSS has a very high probability of receiving the same rating on the CFI” (p. 210). These two studies provided the foundation for future research on the FMSS, which was originally intended to be used as a screening instrument for the CFI, but has developed into a valid and reliable instrument in its own right. As this is a pilot study and exploratory, this study only had one coder and did not conduct interrater reliability, which will be addressed in the limitations section.

Additionally, a short demographic questionnaire (developed by the first author) was used to obtain relevant information pertaining to the participants, from mothers, their children with FXS and their families, as reported earlier.

High Expressed Emotion is assigned when one of the following is present: Critical - the presence of any of the following is necessary for a high critical rating: a negative initial statement, negative relationship rating, or one or more criticisms. Emotional Over-involvement (EOI) - the presence of any of the following statements and/or behaviors is necessary for a high EOI rating: self-sacrificing/over-involvement behavior, emotional display during the interview, any two of the following: excessive detail about the past, one or more statements of attitude, excessive praise (5 or more positive remarks). Critical and EOI:  this rating is assigned when either 1 or 2 above are present. Low Expressed Emotion: this rating is assigned when none of the ratings 1-3 above applies. Borderline Expressed Emotion: a borderline rating is given to speech samples that contain evidence for, but do not qualify as high - EE. There are two classifications of borderline rating: Critical and EOI. The score also yields a sub-rating score of criticism (CRIT) and/or Emotional Over-involvement (EOI), which can be a rating of High, Low, or Borderline.

The final EE ratings are dichotomous, either High or Low, although the Low EE rating may encompass a Borderline Critical or Borderline EOI rating. Magana-Amato (2002) stated in the guidelines that with “lower threshold” populations, relatives might be hesitant to reveal intense feelings about their children; therefore, a Borderline rating can be reassigned as High EE. Other studies have used this rule when conducting FMSS research with mothers of young children, stating “Borderline EE should be counted as High EE in samples where parents may be reluctant to express strong attitudes of criticism or over-involvement about a relative” (Jacobsen, Hibbs, & Ziegenhain, 2000, p. 901). Unfortunately, this rule is sparsely cited within the two articles and the FMSS guidelines (Stubbe, Zahner, Goldstein, & Leckman, 1993; Jacobsen et al., 2000). Taking the lead from prior research and the FMSS guidelines, the author has also applied this rule to the FXS population because mothers of children
with FXS may be more guarded in their expressions of criticisms or negative statements toward their children. This bias toward non-critical statements may be a reflection of cultural norms when talking about children with intellectual disabilities in which criticisms may seem unfair or unloving. Additionally, mothers of children with FXS are keenly aware that their children have inherited the \textit{FMR1} gene mutation from themselves, and feel guilt about their child’s condition, which may also cause the reluctance of expressed emotion. To that end, a lower threshold for EE ratings was applied. Although this rule has been applied, descriptive and inferential statistics are reported \textit{with} and \textit{without} the application of the rule to show how the rule influenced the results.

### 3.3 Procedure

Participants were recruited for voluntary participation through flyers distributed through private therapists that provide services to families with children with FXS, hospitals that serve families with children with FXS within the Denver Metropolitan area, and an international website that provides information on all aspects of FXS.

The sample was purposeful and convenient because the author targeted locations that families with children with FXS frequent. Part of the sample may have also come from a “snowball effect” in which one participant informed other mothers of the value and ease of this study (via internet list serves). This snowball effect was an important recruitment strategy because many families touched by FXS connect with other families who also have children or family members with FXS. These families were also eager to participate in studies that further the awareness, knowledge, and effects of FXS. Consequently, the recruitment of families through word of mouth from other families across the nation was an effective approach to increase participation in this study.

The mothers who agreed to participate in this study were asked to complete a short audio taped interview (i.e. Five Minute Speech Sample) and the questionnaire. The interviews were conducted in person for those mothers who lived within the Denver metropolitan area. Mothers, who lived outside of the Denver metropolitan area, completed the interview process through long-distance phone calls. Questionnaires were completed and collected via postal service ($n = 18$). Through direct communication with other researchers who have used the FMSS, it was determined that long-distance data collection through audio taped phone interviews would be valid and reliable for this measure (personal communication M. Klinnert, 2007).
4. Data Analysis

The primary question that guided this pilot study was: What is the Expressed Emotion of mothers of children with Fragile X syndrome (FXS)? To address these variables, descriptive analyses were completed on the measurement of EE, through the Five Minute Speech Sample (FMSS)(Magana-Amato, 2002) and the demographic questionnaire. Additionally, correlational analyses (r) were completed between the FMSS and the demographic questionnaire to determine significant relations between subgroups of FMSS and the sample.

Four groups based on Erikson’s stages of human development were created, and a test of analysis of variance (ANOVA) was completed to identify the existence of group differences as defined by age (Erikson, 1950). Children in the youngest group, labeled Early Childhood consisted of children aged birth to five years (n = 7). Children in the Middle Childhood group were aged 6 to 12 years (n = 14); children in the Adolescence group were 13-18 years (n = 9), and the Young Adulthood group contained 19-40 year olds (n = 7). The largest was the middle childhood group, followed by the adolescents, young adults, and early childhood. The ANOVA concluded that the only significant demographic differences between the age groups were in regards to maternal age (F = 17.02; df = 3; p < .05) and age of diagnosis (F = 8.97; df = 3; p < .05). In both of these variables, the means young adult group stood out because the mothers tended to be older and their age of diagnosis was also higher. This is also the group with the widest range of age (19-40 years). There were no significant differences for the remaining demographic variables. The youngest child in the sample was two years of age and the eldest person was 40 years old.

4.1 Five Minute Speech Sample (FMSS) Descriptive Statistics

The primary variable in this study is the Expressed Emotion of mothers of children with FXS as measured by the Five Minute Speech Sample (FMSS) and yields a dichotomous EE score of High or Low (Magana-Amato, 2002). Thirty-five mothers completed the FMSS in person (n = 14) during home visits or FXS clinic consultations or by phone (n = 21). There were no significant differences in EE scores between the mothers who completed the FMSS in person or by phone (F = 3.831, p > .05). Therefore, location of data collection was not found to be a confounding variable.

Seventy-seven percent of the mothers scored “neutral” initial statements, typified by a factual description of their child’s age and diagnosis of FXS. For example, the mothers would say, “My son is 13 years old and has fragile X syndrome”. The remaining mothers provided “positive” initial statements, expressing
a general positive remark about their child, such as “Betsy is a wonderful person. She is the sweetest person”\(^3\). None of the mothers expressed a “negative” initial statement.

The dyadic relationship within the FMSS is an overall rating within the measure, which is also coded as positive, neutral, or negative. Table 3 shows that a majority of relationship codes in the sample were “positive”, followed by 40% “neutral”, and only 6% negative. As stated above, a positive, neutral, or negative relationship was determined by statements that speak to the “quality of the relationship”, such as the quality of communication between the dyad and shared activities.

Several mothers talked about their “special relationship” with their children with FXS. Often mothers reported they had the closest relationship with their child, regardless of other family members (including fathers), even though they described themselves as the disciplinarian, while the fathers “had all of the fun”. The relationship was described as very close with mutual admiration and enjoyment. Emerging themes from the relationship included heartening statements that clearly established quality time together that led to positive relationships. The following are examples of statements that demonstrate the dyads’ positive relationships:

- “I enjoy playing with him and reading books with him”.
- “We sing together”.
- “We have a special bond”.
- “We have a unique communication with each other”.
- “We are very in sync with each other”.

Table 3 - Descriptive Statistics for Categorical Codes of FMSS Relationships (N=35)

<table>
<thead>
<tr>
<th>FMSS Variable</th>
<th>Positive</th>
<th>Neutral</th>
<th>Negative</th>
</tr>
</thead>
<tbody>
<tr>
<td>Initial Statement</td>
<td>23% (n = 8)</td>
<td>77% (n = 27)</td>
<td>0%</td>
</tr>
<tr>
<td>Relationship</td>
<td>55% (n = 19)</td>
<td>40% (n = 14)</td>
<td>6% (n = 2)</td>
</tr>
</tbody>
</table>

Table 4 illustrates FMSS categories coded as present or absent throughout the entire speech sample, although the EOI subcategory of Self-Sacrificing Overprotective Behavior (SSOP) can be coded as a borderline, with several “hints”
or evidentiary statements indicating the behavior may be present. SSOP represents statements by respondents in which they make sacrifices for the children, regardless of the impact on themselves. Some of the statements, even though they may reflect a positive relationship, indicate a lack of boundaries between the mother and child, and a loss of individuality of the child and mother. Attachment theory supports this notion of the development of an intense bond between mother and child; although as the child ages and then begins to distinguish him/herself from the mother both physically and emotionally. For a child with special health care or development needs, it is often hard for the mother to let the child explore and learn from his/her environment due to the development challenges. So the mother is cautious about letting her child develop the independent skills even though the mother remains the “secure base” for the child (Bowlby, 1982). The following statements are examples of hints of SSOP statements that were coded High or Borderline:

- “He does everything with me, and goes everywhere with me.”
- “I had to stay home a lot because I couldn’t handle him.”
- “I spend a lot of time worrying about him.”
- “Everything went into helping him.”
- “We are attached at the hip.”

Many mothers said that their lives were chaotic and frustrating, but there was little evidence that they were truly critical or dissatisfied with the behavior of their children. They understood the impact of FXS and built their daily lives around the strengths and challenges of their children. One mother stated, “I understand her because I have fragile X myself”. Statements of dissatisfaction were primarily centered on the child’s ability to “push their mother’s buttons” with challenging behaviors related to sensory overstimulation. Interestingly, mothers reported that their children’s aggressive behavior was targeted at them and not other family members, such as in this statement, “Sometimes I get hurt from his aggressive behavior, but that is ok”. Therefore, the perception of the mothers is that their children with FXS are closer to them, which allows them to feel comfortable enough in their relationship to be targets of their aggression.

Finally, Table 5 illustrates FMSS subcategories that are continuous by the use of a frequency count taken over the entire speech sample. There were very few criticisms within this sample, examples included “I get angry with him sometimes; he knows how to push buttons and when he is frustrated he will do things that are so irritating”. Criticisms are determined by content and tone, indicating some level of frustration or unfavorable view of the child.

There were several Statements of Attitude articulated by the mothers, which were coded as either present or absent (Statements of Attitude is a subcategory of Emotional Over-involvement).
The reason these statements are coded as a subcategory of EOI is that within a brief period (5 minutes), the mother verbalized statements that would indicate intense love for the child and the willingness to do anything to help the child. Such statements included, “I love him very much”, “She is my angel”, and “I love him more than anyone in the world”.

The mothers in this sample had very positive things to say about their children. They were truly proud of their accomplishments, including but not limited to adjusting to over-stimulating experiences, taking care of themselves, and communicating with peers. There was a cumulative count of 25 Positive Remarks made by mothers. The coding of “Positive Remarks” also falls under the EOI subcategory and encompasses a curvilinear result because of the frequency count. More specifically, zero to four Positive Remarks do not affect the EOI rating, although when participants express five or more Positive Remarks within five minutes, a high EOI rating results. Within this sample, the range of Positive Remarks was from 0-11, with an average of two. Five Positive Remarks is the tipping point when the coding changes from low to high. Positive Remarks include statements such, “He is a very sweet child”, “She is very sensitive”, “He has the best laugh ever”, and “He has a great sense of humor”.

<table>
<thead>
<tr>
<th>FMSS Variable</th>
<th>Present (n = 11)</th>
<th>Absent (n = 24)</th>
<th>Borderline (n = 5)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Dissatisfaction</td>
<td>31%</td>
<td>65%</td>
<td></td>
</tr>
<tr>
<td>Emotional Display</td>
<td>0%</td>
<td>100%</td>
<td></td>
</tr>
<tr>
<td>Excessive Detail</td>
<td>3% (n = 1)</td>
<td>97% (n = 34)</td>
<td></td>
</tr>
<tr>
<td>Self-Sacrificing Over Protective Behavior</td>
<td>3% (n = 1)</td>
<td>78% (n = 29)</td>
<td>14% (n = 5)</td>
</tr>
</tbody>
</table>

Table 4 - Descriptive Statistics for Categorical Codes of FMSS Subcategories (N = 35)

The mothers in this sample had very positive things to say about their children. They were truly proud of their accomplishments, including but not limited to adjusting to over-stimulating experiences, taking care of themselves, and communicating with peers. There was a cumulative count of 25 Positive Remarks made by mothers. The coding of “Positive Remarks” also falls under the EOI subcategory and encompasses a curvilinear result because of the frequency count. More specifically, zero to four Positive Remarks do not affect the EOI rating, although when participants express five or more Positive Remarks within five minutes, a high EOI rating results. Within this sample, the range of Positive Remarks was from 0-11, with an average of two. Five Positive Remarks is the tipping point when the coding changes from low to high. Positive Remarks include statements such, “He is a very sweet child”, “She is very sensitive”, “He has the best laugh ever”, and “He has a great sense of humor”.

Table 5 - Descriptive Statistics for Continuous Codes of FMSS (N = 35)

<table>
<thead>
<tr>
<th>FMSS Variable</th>
<th>Frequency</th>
<th>Range</th>
<th>Mean</th>
<th>SD</th>
</tr>
</thead>
<tbody>
<tr>
<td>Criticism</td>
<td>2</td>
<td>0-1</td>
<td>.06</td>
<td>.23</td>
</tr>
<tr>
<td>Statements of Attitude</td>
<td>7</td>
<td>0-2</td>
<td>.31</td>
<td>.67</td>
</tr>
<tr>
<td>Positive Remarks</td>
<td>25</td>
<td>0-11</td>
<td>2.11</td>
<td>2.38</td>
</tr>
</tbody>
</table>
4.2. Final FMSS EE ratings

The first set of analyses encompasses final EE ratings with a dichotomous High/Low rating reveal that 86% (n = 30) of the sample were rated Low EE, while only 14% (n = 5) were rated High EE. Figure 1 below illustrates the final FMSS rates lacking the borderline rule.

Figure 1 - Final FMSS EE Ratings lacking the Borderline Rule. The Y axis represents the number of participants (Total N=35)

In Figure 2 below, the final Low ratings included the Borderline ratings and when broken down further, 37% (n = 13) of the mothers were rated Low EE, 11% (n = 4) were rated High EE, and 51% (n = 18) were rated Borderline EE (EOI and Critical together). Figure 2 illustrates all ratings, including High, Low, and Borderline.

Figure 2 - Final FMSS EE Ratings with Borderline Rating. The Y axis represents the number of participants (Total N=35)
Alternatively, when applying the Borderline rule regarding “low-threshold” populations for a dichotomous High/Low EE rating, 57% of the sample ($n = 20$) were rated High EE, and 43% ($n = 15$) were rated Low EE (see Figure 3).

![Final FMSS EE Rating with Borderline Rule Applied](image)

Figure 3 - *Final FMSS EE Rating with Borderline Rule Applied. The Y axis represents the number of participants (Total N =35)*

Clearly, a majority of mothers fall within the Borderline EE rating, particularly from the Emotional Over-involvement subgroup. Even within this small sample, a trend of Emotional Over-involvement is emerging for mothers of children with FXS. This trend will be discussed further in the discussion.

4.3 *FMSS subgroup ratings*

The descriptive statistics for the EE subgroup ratings are broken down into eight groups, taking into account all of the possible rating combinations. The subgroups’ ratings are assigned before the final EE ratings and involve the subgroups of Low, Critical, and Emotional Over-involvement (EOI), because the subgroup rating will determine the final EE rating. Figure 4 below illustrates that 37% of the mothers in the sample were rated Low, followed by 27% rated as Borderline EOI. All Borderline EOI and Critical subgroups together constitute 52% ($n = 18$) of the sample. No participating mothers were rated with the combination of Critical and Borderline EOI (CRIT & b/EOI).
It is a surprising finding, given that specific disturbances should not affect the student’s cognitive abilities.

The values for dissatisfaction with learning processes for students with LD are very high: 27.2% of the sample stated that they were either ‘not at all’ or ‘not very’ satisfied with the capacity of the school to respond to the educational needs of students with LD, compared with 14.7% of the sample that described the educational experience of students with disability.

Evidently, teachers are expressing dissatisfaction with the way in which teaching is managed within the learning processes of these students, highlighting a lack of preparation on the part of schools to implement effective measures to compensate for disorders and ensure effective learning.

The socialisation process is more satisfying. Apart from any other factor, the number of respondents stating that they were ‘completely’ satisfied is higher.

4.4 FMSS and demographics

Finally, analyses provided significant relationships between the EE variables and demographic variables. Notably, the subcategories in the FMSS correlated with the categorical demographic variables. There was a positive relationship between the Initial Statement (coded as positive-1, neutral-2, and negative-3) and the mothers’ age ($r = .370$), indicating that younger mothers expressed more positive initial statements than older mothers. Marital status also was negatively
correlated with a number of FMSS subcategories such as Criticisms, Self-Sacrificing Over Protective Behavior (SSOP), and Emotional Over-involvement (EOI). This relationship suggests that mothers who were married (85%) were more likely to have fewer Criticisms, Self-Sacrificing Over Protective Behavior, and Emotional Over-involvement, although it may be due to the lack of variability in marital status. Ethnicity was another factor negatively correlated to FMSS subcategories, again possibly due to the lack of diversity in the sample.

There was a possible relationship between the number of additional diagnoses to FXS and the rating of Borderline EOI ($r = .316, p = .084$). This may indicate mothers are more likely to become more emotionally over-involved when their children receive additional diagnoses. Table 6 shows the significant relationships between the FMSS subcategories and demographic variables.

Table 6 - Correlations coefficients ($r$) between FMSS Subcategories and Demographics

<table>
<thead>
<tr>
<th>Variables</th>
<th>Initial Statements</th>
<th>Criticism</th>
<th>SSOP</th>
<th>Positive Remarks</th>
<th>EOI</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mothers’ Age</td>
<td>.37*</td>
<td>.04</td>
<td>.16</td>
<td>-.29</td>
<td>-.22</td>
</tr>
<tr>
<td>Marital Status</td>
<td>.00</td>
<td>-.48**</td>
<td>-.62**</td>
<td>-.01</td>
<td>-.38*</td>
</tr>
<tr>
<td>Ethnicity</td>
<td>-.15</td>
<td>.07</td>
<td>.11</td>
<td>-.49**</td>
<td>-.44*</td>
</tr>
</tbody>
</table>

Note. * Correlation is significant at the .05 (2-tailed), ** Correlation is significant at the .005 (2-tailed)

5. Discussion

5.1 Expressed Emotion and the Five Minute Speech Sample

A majority of children with FXS have inherited the FMR1 gene from their mothers and have endured an intellectual disability and other significant challenges. The mothers are well aware of their carrier status after their child’s diagnosis, and it is postulated that this knowledge limits the mothers’ Expressed Emotion in a formal setting with a standardized instrument, such as FMSS, and may, in fact, limit their expressed emotion in general. With that in mind, the application of the “borderline rule” as described above is necessary for respondents of a “lower threshold” population because they may be more “reluctant to express strong attitudes about their relatives, for example, parent of young children” (Magana-Amato, 2002, p. 45). A “lower threshold population” is a population that may be more reluctant to fully express their emotions relating to the intense relationship with their child. The argument for the application
of this rule to mothers of children with FXS is because that these mothers have a lower threshold due to the inheritability factor of the FMR1 gene mutation that causes their children’s subsequent intellectual disability. This is consistent with other “low threshold” populations, such as mothers of very young children in that mothers of children with FXS share similar parenting responsibilities that relate to the intensification of caregiving due to the children’s inability to care for themselves because of age or disability.

The presence of dissatisfactions and the frequency of critical statements add to the overall coding of the relationship and final EE ratings. Even though the relationships between mothers and their children with FXS were positive for the most part, some mothers expressed “dissatisfaction” with their child and their child’s behavior. Other mothers, while expressing dissatisfaction with the situation (i.e., diagnosis of FXS), did not attribute the dissatisfaction to their children. This form of dissatisfaction does not add to the coding of the relationship or final EE coding because the respondent does not directly “blame” the child for the situation. Therefore, these types of statements, such as “sometimes it is hard or frustrating” were not coded as “dissatisfactions”.

Similar to the lack of dissatisfaction expressed by mothers, there were also limited “criticisms”. In the entire sample, only two criticisms were coded. This may be because FXS is an inherited genetic disorder and mothers are more forgiving for the behavior of their children because of their limited intellectual ability. Another explanation could be that despite their challenges, the mothers are generally positive because they are accustomed to the children’s behaviors. Parents of children with schizophrenia and other mental health disorders tend to be more critical of their children’s behavior, which may be due to the age of onset or perceptibility of disability.

One argument supporting the discrepancy of EE ratings, especially dissatisfaction and criticisms, between children with intellectual disabilities (i.e. FXS) and children with mental health disabilities (i.e. schizophrenia), is the dichotomy of “visible” versus “invisible” disabilities. Children with FXS have a “visible” disability, even though they are born with few dysmorphic characteristics and are often not diagnosed until the later early childhood years. The visibility of FXS emerges more as they age and the developmental gap widens. Criticisms may not be attributed to children with FXS because their behavior is perceived to be no fault of their own, due to their diagnoses. Additionally, disabilities that are more noticeable may elicit patience and sympathy. Children with mental health disorders, on the other hand, have an “invisible” disability and are often criticized for their behavior because they appear “normal” at times, even though their challenges are not physically obvious. Children with
learning disabilities, attention/behavioral disorders, or sensory processing disorders encompass invisible disabilities, and the public may have higher and/or unrealistic expectations for their functioning, in relation to their actual capabilities (Mitchell, 2002). Parents and family members, while knowledgeable of their children’s diagnosis, still have higher expectations and sometimes “blame” the children for their intermittent and erratic behavior (Todd & Shearn, 1997). Society, in general, is less sympathetic and compassionate to individuals with invisible disabilities because of a common misconception that their behavior is a result of no self-control, therefore contributing to criticism, dysfunction, and discrimination (Mitchell, 2002). Historically, children and adults with visible disabilities or disfigurements were discriminated against and denied civil liberties that resulted in low expectations and adjustment to adult life.

The age at diagnosis for children with “visible” and “invisible” disabilities is also a factor and varied. Children with FXS are often not diagnosed until 3-4 years of age, which is late according to early intervention standards (Carmichael, Pembrey, Turner & Barnicoat, 1999; Bailey Jr, Roberts, Mirrett, & Hatton, 2001), but relatively younger, and remain “child-like”. However, children with mental health disorders are often diagnosed at a later age (i.e. adolescence or young adulthood), yielding less adjustment time for the parents (NIMH, 2008). Parents of these children have witnessed their children develop typically in many ways, such as physically and cognitively, and may express more dissatisfaction and criticisms when they behave inappropriately (Brown, Birley, & Wing, 1972; McCarty & Weisz, 2002). Parents of children with invisible disabilities may know intellectually that their child has a disorder that results in challenging or aberrant behaviors, but emotionally it is harder to reconcile. Therefore, they may less sensitive to their children’s behavioral outbursts. Alternatively, parents of children with visible disabilities, particularly inherited genetic disorders, may be more sensitive and accommodating to their children’s difficult behaviors. This dichotomy provides more rationale for the argument that mothers of children with FXS may be reluctant to express strong criticisms and/or dissatisfactions.

5.2 Emotional over-involvement (EOI)

Emotional over-involvement is most salient to the attitudes and characteristics of mothers of children with FXS. Research on mothers who are carriers of the FMR1 gene mutation, suggest that they have a tendency to experience more anxiety, depression, low self-esteem, social isolation, and higher levels of stress than mothers of children with other disabilities (Johnston et al., 2003; Abbeduto, Seltzer, Shattuck, Krauss, Orsmond, & Murphy, 2004; Grigsby, Brega, Engle, Leehay, Hagerman, & Tassone, et al., 2008). Previous descriptions of female
carriers are consistent with mothers in this sample who were coded Borderline and High EOI, due to increased statements of concern, worry, and anxiety about their children. Excessive worry is one of the indications that lead to the Self-Sacrificing Over Protective (SSOP) rating. Statements such as “I worry about my son constantly” or “It’s hard to go on vacation or out of town for a short time, because I worry about how she is doing”, provide evidence of an SSOP rating. Every parent worries about his/her child to some degree, but it is the “excessiveness” that is the relevant feature that can be detrimental to the emotional climate in the home environment. Mothers of children with FXS may have good reason to worry about their children due to frequent medical and behavioral problems, although it is difficult to discern whether this attribute is a result of the carrier phenotype or common concerns that come with having children with disabilities. McConkie-Rosell and colleagues (2005) reported that mothers with the FMR1 premutation may be predisposed to psychological issues such as anxiety and depression and may be exasperated by parental guilt, shame, and blame.

Most notably, more mothers in this sample tended to be rated EOI, rather than Critical due to the high expression of positive remarks. Positive Remarks are an expression by the mother that the child does something “very well” or is “very good” at something. According to the FMSS guidelines, a high frequency of positive remarks (> 5) within a short speech sample results in a borderline EOI rating. That coupled with other components of EOI, such as statements of attitude, leads to a score of High EE. Many mothers in this sample often exclaimed that their child was very good at engaging in social acts with others and that their child was popular either at school or in the community. Statements like “Everyone knows him” or “He has a friend everywhere he goes”, often followed or preceded the positive remarks. These statements are supported by the FXS literature that report children with FXS are inherently social beings although sensory over-stimulation often times gets in the way (Bailey, Mesibov, Hatton, Clark, Roberts, & Mayhew, 1998; Epstein et al., 2002; Cornish et al., 2008). These Positive Remarks may be seen as evidence of how mothers of children with FXS continually highlight the best attributes of their child.

The rating of SSOP was small but also important to recognize, because more mothers were coded as borderline SSOP rather than full SSOP. This rating indicated that mothers would sacrifice much of themselves to help their child. In many of the speech samples there were “hints” of SSOP but not enough for a borderline or full rating, but rather statements declaring that the mother-child dyad link were inextricably linked related, but that at the same time the mother did not feel she was making a sacrifice. Such statements included “we are joined at the hip”, or “He/She is closer to me than anyone else in the family”.

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5.3 Implications of Expressed Emotion

The purpose of this pilot study was to explore the construct of Expressed Emotion with mothers of children with FXS, evaluate the use of this tool with families with children with neurodevelopmental disorders, specifically FXS, and to explore the complex dyad relationship between carrier status mothers and their affected children. The results of the final ratings of EE suggest an emerging trend of Emotional Over-involvement for mothers of children with FXS. This trend has implications not only for the family but also for practitioners working with the families. The excessive worry, anxiety, and Emotional Over-involvement can lead to undesirable behaviors from the child and misperceptions of the child’s ability level by the practitioner. The research on Expressed Emotion reveals that mothers who expressed critical or emotionally over-involved views of their children are more likely to have harsher parenting practices and to be more over-protective (Vostanis & Nicholls, 1992; Calam & Peters, 2006). In addition, research indicates that when children live in a home environment with a parent who has high Expressed Emotion (either Critical or Emotional Over-involvement), their clinical treatment is more likely to be compromised, as evidenced by increased rates of schizophrenic relapse and subsequent hospitalization (Brown et al., 1972; Vaughn & Leff, 1976; Jacobsen et al., 2000). Following this Expressed Emotion research, mothers of children with FXS who are emotionally over-involved (and High EE) could be at risk of mediating the full effects of the children’s therapeutic intervention, although this claim cannot be verified without further study.

The utility of the Five Minute Speech Sample as a means for measuring this construct shows promise. The results did not indicate problems or inconsistent findings. The results actually provided insight into the feelings of this population that was either consistent with or linked to the phenotypic profile of carrier females. Additional research involving a larger sample is recommended to replicate these findings and to compare them to other measures that might provide similar insight.

Exploration of the dyad of carrier mothers with their affected children was the third goal of the study. The results provided insight into the feelings of the mother suggesting behavioral interactions and emotional connections, however the operationalization of these feelings could not be validated or even implied. As a result additional information is necessary for interactional data on the dyad.

The implications for these findings on intervention are unknown and speculative at this time; however, based on previous research, possible links could have powerful consequences. For example, children with FXS spend a limited number of hours in therapy, relative to the time they spend with their family. The skills learned in therapy may be negated because of parents’ critical or
over-involved behaviors, and the children may regress back to homeostasis because of the unintended stress by the parents. Brain research has provided evidence that when individuals experience stress, they use the more primitive parts of the brain and do not retain newly learned skills (Conger, Conger, Elder, Lorenz, & Simons, 1994; Shonkoff & Phillips, 2000; Shonkoff, Boyce, Cameron, Duncan, Fox, Greenough, et al. 2007; Howes, Burchinal, Pianta, Bryant, Early, Clifford et al., 2008). Parents are often not involved in the therapeutic sessions, and may not have a full understanding of the environmental conditions needed to elicit optimal behavior of their children and therefore unintentionally place stress on the child. By involving parents in the therapeutic sessions and/or discussing the therapeutic interventions with parents, they may understand how to reinforce intervention strategies at home in an appropriate and balanced way (i.e. not over critical or involved). This is very important for mothers in this study because they tended to be rated High EE with Emotional Over-involvement, and including them in the therapy session may only exasperate their over-involvement. Therefore, it is important for practitioners to emphasis the need for mothers to be mothers first and not therapists. When practitioners are aware of the influence of expressed emotion, particularly EOI, then they can adjust their expectations for therapeutic interventions by working closely with the parents about following through with intervention strategies and helping the parents set realistic expectations. Practitioners can also help mothers understand and balance their expressed emotion so that the therapeutic interventions can result in the best possible outcomes.

5.4 Limitations
The primary limitations of this pilot study involved the limited sample and the selected measures. This was a small sample with a subset of mothers who were inclined to participate in FXS research. Even though the sample came from across the United States, the demographic characteristics portrayed a homogeneous population of white, middle-class, stay-at-home mothers. Cultural and socioeconomic differences may affect the results and the utility of the measure and should be examined in future studies. The age range of children was also a limitation and prevented conclusions related to specific ages such as early childhood, school age, or adolescence. In future studies, it would be beneficial to study targeted age ranges independently using a larger sample to see if the expressed emotion of mothers differs according to the age of their children.

The FMSS provided rich information about the population of mothers of children with FXS, which measures a construct that has not been explored before with this population. However the FMSS instrument can be considered a
limitation. Traditionally, the FMSS was developed and used in the fields of psychology and psychoanalysis, as opposed to educational or therapeutic fields. The translation into these fields may or may not be appropriate. The instrument may be viewed as a tool that pathologizes parents, due to the connotations of the categorical labels. It may appear to judge parents even though their statements may be an appropriate reaction or situation specific. For these reasons, the FMSS may not be a tool that is ready to be used for intervention planning for children with FXS or other disabilities. Lastly, this pilot study was exploratory and had only one rater for the FMSS, and did not have the opportunity for another rater to determine inter-rater reliability.

6. Future directions

The next step for this line of research includes recruiting a larger sample to look solely at the Expressed Emotion of mothers with children with FXS to understand this construct within this population, specifically the phenotype of mothers with the \textit{FMR1} premutation. A comparison of Expressed Emotion between mothers of children with FXS and mothers of children with other developmental disabilities, such as Down syndrome, Autism, or Williams syndrome would be crucial to sort out the differences between the FXS phenotype and environmental effects within the relationship between EE and FXS, or other children with systemic delays. Further research should also include multiple coders for interrater reliability and explore how much of the variance of EE ratings are explained by the mothers’ own challenges of being a carrier of the \textit{FMR1} premutation.

7. Conclusion

This pilot study served as the initial exploration of the Expressed Emotion of mothers of children with FXS. The initial findings provide striking information on the high number of mothers with FXS who met the criteria for High EE, particularly Emotional Over-involvement. These findings may lead to intervention strategies that can help mothers balance their Expressed Emotion and improve the impact for their children’s therapeutic interventions, in light of previous research on the well being of mothers of children with FXS.

Practitioners, providers, and all types of professionals working within the FXS field need to work with parents to provide meaningful support so that their child can receive optimal benefits from treatment.
It is primarily the practitioners’ job to be objective about a child’s abilities and challenges, as opposed to the parents’ responsibility. It is difficult for any parent to view his/her child through a critical lens and provide the important intervention needed for success. Although parents are often told that they need not be a “therapist,” they are often required to implement therapeutic interventions at home. Consequently, careful interpretation of the Expressed Emotion of mothers of children with FXS is required. These findings lead to more questions, such as: how does this perception affect their child’s behavior and what level of intervention is needed to change undesirable behavioral characteristics? How do the mothers’ perceptions color their Expressed Emotion of their children? The Expressed Emotion of mothers with children with FXS is an intriguing topic that warrants further research and discussion among physicians, therapists, researchers, and families, and this research provides a beginning to that discussion.

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Expressed Emotion of mothers of children with Fragile X Syndrome


Abstract

Mental Imagery can be defined both as a reproduction of cognitive contents of the mind not actually present to sensorial perception, and as the re-elaboration and interpretation of the original perceived data. Motor and athletic rehabilitation and recovery after injuries or traumas, especially in sports, but more generally in physiotherapeutic practice, have shown to be enhanced by mental imagery processes.

Mental imagery has been assumed to be essential for the acquisition of motor skills, but to also be relevant in the involutive phases of adult cognition. In fact, empirical studies indicate that different components of imagery are selectively affected by aging.

This review discusses the neuropsychological bases of mental imagery, focusing on its possible applications to the rehabilitation of deficits from a variety of different causes, including mental deterioration in the elderly. Simulation techniques, and how they might assist in the accurate assessment of mental imagery skills in order to design optimal learning and/or training interventions, or rehabilitation of motor gestures, are also presented.

Keywords: Mental Imagery; Motor skills; Rehabilitation; Simulation.
1. Mental Imagery: definition and development.

The use of the term ‘mental imagery’ can be two-fold: it can refer to the reproduction of cognitive contents not actually detectable to sensorial perception, and to the re-elaboration and interpretation of the original perceived data.

Experimental literature (Farah, Gazzaniga, Holtzman, & Kosslyn, 1985; Denis, Logie, Cornoldi, De Vega, & Engelkamp, 2001; Pearson, De Beni, & Cornoldi, 2001; Kosslyn, Thompson, & Ganis, 2006; Joffe, Cain, & Marić, 2007), has identified the essential components of the process of mental imagery as being: the generation of images of recently perceived external stimuli that are no longer present, or from long term memory; the maintenance of an image in the visuospatial memory buffer; the inspection and verbal description of its content; the possibility of transforming and elaborating the produced image, e.g., by rotating or restructuring it by modifying its contour or shape, or by combining these actions into a mental synthesis that produces new and original models.

Psychophysiological activation often results from the imaginative process in that there are often emotions associated to images, so that the reconstruction or anticipation of images of sensations, not actually present, may generate pleasant experiences or uneasiness, and in some cases, a true pathology.

From what has been said so far, it is clear that mental imagery is not limited to a reproduction of data coming from the senses, from perception or memory; it actively contributes to the (re)construction of a reality that is different from that of the senses, and that is important for the functioning of the mind as a whole.

Imaginative function is considered essential for both spontaneous learning and for knowledge acquisition after specific training (scholastic, artistic, athletic).

The ability to imagine possibilities different from perceived reality contributes to both the cognitive, as well as emotional development of the child (Harris, 2000); and is considered the basis of creative planning and of the capacity to make new discoveries (Finke, 1990; Roskos-Ewoldsen, Intons-Peterson, & Anderson, 1993). The strengthening or enhancement of cognitive functions through mental imagery, in fact, has been studied extensively.

Mental imagery is also considered essential for the acquisition of motor and athletic skills. It creates the capacity to create or recreate an experience from information stored in memory, involving aspects that are similar to sensations and perceptions, and that are controlled willfully by the athlete, even in the absence of the stimuli normally associated to the actual experience (Morris, Spittle, & Watt, 2005). The two characteristics that make up mental imagery ability, therefore, are its vividness and controllability. The first, regards the capacity of the athlete to experience, during the actual motor execution of
the gesture. Controllability, instead, regards the facility and accuracy with which mental images are organized and re-interpreted (Moran, 1993).

Novice athletes and experts not only use mental imagery differently in their specific performance (Cumming & Hall, 2002), but also have different brain activations, as has been confirmed by a number of neurophysiological studies (Yarrow, Brown, & Krakauer 2009; Wei & Luo, 2010). Fournier, Deremaux and Bernier (2008), explain these differences in the use of mental imagery in expert athletes as opposed to novices, by referring to three elements: the content (what is being visualized), the characteristics (how it is being visualized) and the related functions (why imagery is being activated).

An extensive amount of literature regards the importance of the use of mental imagery in motor and athletic training and practice (Feltz & Landers, 1983; Murphy, 1994; Martin, Moritz, & Hall, 1999; Ahsen, 2001; Murphy & Martin, 2002; Taktek, 2004; Gregg, Hall, & Nederhof, 2005). Previous and current research findings highlight the positive effects of using cognitive and behavioral routines on athletes’ performance and on movement learning processes (Lidor, 2007). As expected, the performance ranking of the coach is significantly related to the quality of mental representations; a finding reported previously in different sports settings. Additionally, players highly qualified in a specific skill (e.g., serve in tennis) possess a similar mental representation to experts’ postulation of that skill.

If mental imagery has also been found to be important in child development, it is no less important in the involutive phases of adult cognition. Contradictory data exists in the literature regarding the deterioration of mental imagery abilities in the elderly; some authors consider it as a normal aspect of cognitive decline, while others underline how it is essentially preserved until the very advanced stages of aging (Mason & Smith, 1977; Kosslyn, Margolis, Goldknopf, Daly, & Barrett, 1990; Craik & Dirkx, 1992; Dror & Kosslyn, 1994; Brown, Kosslyn, & Dror, 1998; Palladino & De Beni, 2003; Skoura, Papaxanthis, Vinter, & Pozzo 2005; De Beni, Pazzaglia, & Gardini, 2007; Schott, 2012; Gabbard & Cordoba, 2013; Zapparoli, Invernizzi, Gandola, Verardi, Berlingeri, Sberna et al., 2013).

Many studies argue that the poor performance of imagery in the elderly regards response times rather than their accuracy, and is linked to the complexity of the stimulus when considering working memory as a mediator (Briggs, Raz, & Marks, 1999). Dror and Kosslyn (1994) studied the effects of aging on four components of mental imagery: image generation, maintenance, scanning, and transformation; moreover, image generation can be further fractionated. A representation of the to-be-imaged object can be activated from long-term memory, or the images can be composed sequentially, segment by segment (activation versus composition).
The authors found that the different components are affected selectively by aging. Older adults showed impairment in image generation, in image activation, in particular, and in rotation with respect to younger adults. In contrast, the elderly were able to compose (the process of generating the segments of a shape, one by one, and scan visual mental images) just as well as young adults. Bruyer and Scailquin (2000), extending these results, demonstrated that in image generation the deficit of the activation stage increases linearly with age, and that there is a complexity effect on accuracy that similarly increases with age. The authors conclude that age per se is not a determinant for image generation.

Kemps and Newson (2005), found that older adults were slower and less accurate than younger adults on all imagery components: image generation, maintenance, scanning and rotation, but showed that manipulations of stimulus complexity produced inconsistent, differential age effects across tasks. According to De Beni, Pazzaglia and Gardini (2007), image generation, maintenance and transformation seem to be differently affected by the type of image and aging. More recently, Kalkstein, Chechsfield, Bollinger and Gazzaley (2011), using fMRI, showed that aging disrupts neural networks that serve mental imagery, and provide evidence of this disruption as a factor in age-related memory decline. Imagery vividness has also been found relevant in normal and impaired aging (Malouin, Richards, & Durand, 2010).

The differences in the results regarding deterioration found in the previously mentioned studies most likely depend on the specific processes being evaluated and the instruments used.

2. The neuropsychological bases of Mental Imagery and the possibilities of its simulation.

Neuropsychological studies have long highlighted the complexity of cerebral activation in mental imagery activity (Farah, 1984, 1985; Denis, Engelkamp, & Richardson 1988; Hampson, Marks, & Richardson, 1990; Sergent, 1990; Tippett, 1992; Kosslyn, 1999; Thompson & Kosslyn, 2000; Kosslyn, Ganis, & Thompson, 2001; Bartolomeo, 2002; Sala, Rama, & Courtney, 2003; Kosslyn, Thompson, Shephard, Ganis, Bell, Danovitch et al., 2004; Kosslyn, 2005; Oliverio, 2013). Later studies have shown how imagery, perception and visual memory have overlapping as well as distinct brain locations (Kosslyn, Thompson, & Ganis, 2006; Gardini, De Beni, Cornoldi, Bromiley, & Venneri, 2005; Gardini, Cornoldi, De Beni, & Venneri, 2009; Thompson, Slotnick, Burrage, & Kosslyn, 2009; Slotnick, Thompson, & Kosslyn, 2012; Cichy, Heinzle,
Mental Imagery and rehabilitation through simulation

& Haynes, 2012), while Zeman, Della Sala, Torrens, Gountouna, McGonigle and Logie, (2010), have shown in a clinical case, how visuospatial perception and imagery can be functionally separated.

The relationship between the neurobiological mechanisms of perception and imagery is a controversial subject (Ferretti, 1998; Iachini, 2002). Are they analogous and interconnected systems, or are they two different parts of the same system? There is proof that visual imagery has the same neurobiological bases as vision itself (Ishai, Ungerleider, & Haxby, 2000), and that the areas that control spatial perception are also necessary for maintaining mental images active in working memory (Trojano, Conson, Maffei, & Grossi, 2006). The same is true for auditory and verbal stimuli: favorite songs, film dialogues, etc. Images are not only “seen” or “heard” with the eye or the ear of the mind, they are named, linguistically classified, assigned emotional connotations, remembered and transmitted. In fact, the verbal areas of the brain are activated during the recall and semantic qualification of an image in order for it to be memorized.

A recent line of research on mental imagery integrates neuropsychology and cognitive science, and is based on simulation models. Before this, Moulton and Kosslyn (2009) had already established that “all imagery is simulation”: imagination is not so much a specific type of mental state, but rather a simulation heuristic, a capacity to reproduce other types of mental states.

Theories in artificial intelligence have considered how mental imagery – an experience that is similar to perceptual experience, but in the absence of the appropriate external stimuli for sensorial perception – can be considered as a type of biological simulation in which the mind represents (simulates) itself in action. This itself represents a kind of off-line cognition involving proprioception that realizes an embodied cognition, especially when imagery involves movement (Jeannerod, 2001; Ziemke, Jirenhed, & Hesslow, 2005; Gallese, 2005; Iachini, 2002, 2011; Di Nuovo, Marocco, Di Nuovo, & Cangelosi, 2011).

Mental chronometry studies confirm this hypothesis, showing how the time employed in imagining a movement is correlated to the time necessary to physically execute it, suggesting that real movements and imagined ones are functionally similar (Decety & Michel, 1989; Decety, Jeannerod, & Prablanc 1989; Decety & Jeannerod, 1995; Jeannerod, 1995; Denis & Kosslyn, 1999; Calmels & Fournier, 2001; Reeds, 2002; Guillot & Collet, 2005; Calmels, Holmes, Lopez, & Naman, 2006). On a neuropsychological level, the primary motor cortex is active during the production of motor images (Jeannerod, 1994, 2001). Other neurophysiological studies have confirmed the substantial overlap between patterns of activation for real movements and imagined ones (Wuyam, Mousavi, Decety, Adams, Lansing, & Guz, 1995; Szameitat, Shen, & Sterr, 2007).
Neuroimaging studies have shown how motor areas of the brain, such as the pre-motor and parietal areas, are involved in both real as well as imagined movements (Holper, Scholkmann, Shalom, & Wolf, 2012).

Cognitive neuroscience research has also investigated the neural mechanisms of motor imagery in the control of action (e.g. de Lange, Roelofs, & Toni, 2008), and has found similar results to those concerning visual imagery and visual perception and how they recruit similar brain regions (Ganis, Thompson, & Kosslyn, 2004).

Several studies have made use of the motor expertise model to investigate the link between the action execution and action perception network (Calvo-Merino, Glaser, Grezes, Passingham, & Haggard, 2005; Calvo-Merino Grezes, Glaser, Passingham, & Haggard, 2006; Orgs, Dombrowski, Heil, & Jansen-Osmann, 2008) and motor learning (Cross, Hamilton, & Grafton, 2006).

The activity related to static movement creation showed a very similar pattern of activity to the imagery creation tasks, showing how important imagery creation is in movement creation (May, Calvo-Merino, deLahunta, McGregor, Cusack, Owen et al., 2011).

The only major difference found between movement and imagery creation was an increased activation of the right inferior frontal gyrus, which has been associated with representations of goal-directed actions in movement observation and execution (Iacoboni, Woods, Brass, Bekkering, Mazziotta, & Rizzolatti, 1999). Moreover, this area has also been related to inhibition of prepotent responses (Christopoulos, Tobler, Bossaerts, Dolan, & Schultz, 2009), but also in a multiple-demand network (Duncan, 2010), which is activated by many different cognitive demands including perceptual difficulty, novelty, and response conflict. It seems that body representation and cognitive integration of perceptual body information are strongly influenced by cognitive motor representations (Bläsing, Schack, & Brugger, 2010). Based on these and many other studies, a strongly emerging concept in our understanding of human motion is that major interfaces in the architecture of movement are cognitive in nature.

Thanks to these studies, a transition is taking place, taking us from considering “mental imagery as an internal phenomenon, unimodal and primarily visual and similar to perception, to mental imagery as a multimodal and dynamic phenomenon, based on conscious sensory-motor simulation” (Iachini, 2011, p. 1).
3. Application of Mental Imagery to rehabilitation.

3.1 Motor and athletic rehabilitation.

A related area in which the application of mental imagery to education and rehabilitation is widely used, is in the field of sports (Sheikh, Sheikh, & Molleski, 1994; Martin, Moritz, & Hall, 1999; Sordoni, Hall, & Forwell, 2000; Ahsen, 2001; Hall, 2001; Cumming & Hall, 2002; Murphy & Martin, 2002; Benchke, 2004; Gregg, Hall, & Nederhof, 2005; Vealey & Greenleaf, 2006; Fournier, Deremaux, & Bernier, 2008; Holmes & Calmels, 2008).

“Motor imagery refers to the cognitive process through which the mental representation of an action is activated. In essence, this cognitive process represents ‘motor activation without execution’. In the applied/rehabilitation context, motor imagery can be used as a standalone intervention, added to other forms of practice or embedded into another form of intervention” (Kranzioch, Zich, Schierholz, & Sterr, 2014, p. 13).

It has been known for some time that at all ages the imagined rehearsal of motor sequences improves real motor execution later on (Mulder, 2007; Maluoin & Richards, 2010). Training using mental imagery is effective in improving motor performance in that it adds, based on internal stimuli, opportune variations in the movements learned (Taktek, 2004; Munzert, Lorey, & Zentgraf, 2009). It is, therefore, applicable to motor education and rehabilitation, and in the training for various types of individual as well as team sports. For example, Smith, Holmes, Whitemore, Collins and Devonport (2001) have shown that the use of mental imagery, in the training phases of athletes engaged in throwing (e.g., a ball) towards a particular target, later improves their actual performance (Mulder, 2007; Maluoin & Richards, 2010). The imagery helps in learning and practicing the technique, in situations in which it might otherwise be impossible; for example, in contexts that are very different from those of the training scenario, or in situations of recovery after an injury (Nideffer, 1985; Morris et al., 2005; Papadelis, Kourtidou-Papadeli, Bamidis, & Albani, 2007).

When an athlete or an individual in the course of rehabilitation concentrates on mental rehearsal, his nervous system behaves in a way that is similar to when that exercise is physically executed. Rehearsal with mental imagery also has an added effect on an emotional level, because it helps to desensitize the anxiety linked to the prediction of unknown competitive situations, or that are potentially stress inducing. Imagery training is even more effective in learning and performance in motor tasks, when it provides information that would not otherwise be available to the subject (Cumming & Ste-Marie, 2001). Schuster, Hilfiker, Amft,
Scheidhauer, Andrews, Butler et al., (2011) tested the effects of motor imagery training, in association with actual physical practice, and reviewed the literature on the argument in five different disciplines: Education, Medicine, Music, Psychology and Sports. They took into consideration the detailed descriptions of the mental imagery training sessions, summarized in the acronym PETTLEP (Physical environment, Timing, Task, Learning emotion, Perspective) and the detailed temporal parameters, the duration of the study, the duration of the intervention (an average of 34 days), the number of training sessions per week (an average of 3 times for 17 minutes) and the total (an average of 178 minutes). The review covered 133 studies that discussed 141 interventions. The interventions that turned out to be the most effective were individual - supervised but not managed - and done in addition to physical practice.

Sport psychologists use the analysis of athletes’ mental representations as basis for the compilation of mental training (e.g. imagery) to control the learning process, or to evaluate the effects of a sport psychological intervention. The mental training can be considered vital for different sports, because it allows insights into the mental structure of technical and tactical skills (with or without routines) of individual players or teams.

Imagery and instructional training (routine demand) can be used for learning or optimizing performance routines (Velentzas, Heinen, Tenenbaum, & Schack, 2010).

An important use of mental imagery training is found in rehabilitation after injuries or traumas, especially in sports, but more generally in physiotherapeutic practice.

Cognitive mental imagery training has long been used to facilitate recovery and to potentiate rehabilitative exercises, control pain, and to help the patient foresee a return to normal activity, increasing the positive attitude of the subject (Cupal & Brewer, 2001; Christakou, Zervas, & Lavallee, 2007). Both visual as well as kinaesthetic images are used, adapting them to the particular characteristics of the injury and of the patient (Driediger, Hall, & Callow 2006). It is essential that trainers and rehabilitators evaluate the tendency and capacity of the subject to use motivational and cognitive imagery (Sordoni, Hall, & Forwell, 2000).

In this sector, precise guidelines in the use of mental imagery training are also necessary, so that trainers can apply well founded scientific criteria, rather than just going with exclusively practical knowledge (Bovend’Eerdt, Dawes, Sackley, & Wade, 2012). It is necessary, for example, to associate to mental imagery training, the consideration of variables such as self-efficacy and adherence to treatment (Milne, Hall, & Farwell, 2005; Wesch, Hall, Prapavessis, Maddison, Bassett, Foley et al., 2012). The images to be used need to be chosen
carefully in relation to the characteristics of the injured individual, avoiding those that in his subjective experience are associated with weakness, or that his anxiety concerning the “comeback” have more influence (Monsma, Mensch, & Farroll, 2009). Cumming and Williams (2013) have proposed a general model of imagery use in sports, dance, motor exercise and rehabilitation.

3.2 Rehabilitation of disability and deterioration

Among the uses of mental imagery training, of growing importance are those aimed at the enhancement of motor skills for those subjects with disabilities or physical and mental deterioration. For some time now, neurological rehabilitation has put mental imagery practice to good use, in that it has proven its capacity help re-organize functional cerebral processes (Jackson, Lafleur, Malouin, Richards, & Doyon, 2001). At a neuropsychological level, mental imagery practice often results in a re-organization of neuronal systems, and in the activation of plasticity in the involved systems, but the mechanisms by which this happens are yet unclear. Visual imagery systems, however, are considered to be an essential component (Jackson, Lafleur, Malouin, Richards, & Doyon, 2003; Nyberg Eriksson, Larsson, & Marklund, 2006; Olsson, Jonsson, & Nyberg, 2008; Zhang, Xu, Wang, Xie, Guo, Long et al., 2011).

Below, we discuss a number of examples of how mental imagery training has been used in the rehabilitation of deficits from a variety of different causes.

Ever since the studies conducted in the 70’s by Symmes (1971) and Lebrato and Ellis (1974), the involvement of mental imagery, among the consequences of brain damage and mental retardation, has been emphasized. Zupnick and Meyer (1975) studied the long-term effects of instructions based on images in facilitating associative learning in retarded individuals.

Courbois (1996) compared the performance on imagery tasks of two groups of adolescents with mental retardation of different etiology, and one group of subjects of normal intelligence but of the same mental age. The two groups with delay had significantly lower performance, but the major deficit was found in the group with organic causes. The author pointed out how these deficits may have repercussions on other cognitive tasks that require the use of mental representation.

The utility of mental imagery training to improve performance in cases of mental retardation has been confirmed, increasing reaction times (Suburg, 1991) as well as improving motor skills (Suburg, Porretta, & Sutlive, 1995); the effect of imagery training together with physical practice increases the accuracy of performance and reduces variability (Porretta & Suburg, 1995). Mental imagery training was used in the learning of prose in a group of children
(ages 7-12) and adults (18-57) with Down Syndrome, confirming its efficacy in improving memorization to a much greater extent than in conditions in which subjects only listened (De La Iglesia, Buceta, & Campos, 2005).

Vicari, Bellucci and Carlesimo (2006), compared two genetic syndromes, Down and Williams Syndrome, in visual perception tasks, visual and spatial imagery and working memory. Performance in the two groups was different in the different tasks, especially with regard to working memory content (visual-spatial information, or information regarding objects), confirming that these abilities are not always affected in genetic syndromes with intellectual disability, and should be evaluated on a case by case basis.

A study by Roskos-Ewoldsen, Conners, Atwell and Prestopnik (2006), showed that young adults with intellectual disability can have an adequate capacity to visualize (the task was to locate landmarks on a map of an island, and mentally travel from one landmark to another). In respect to a similar visual-perceptual task (with the stimulus present), performance was found to be slow in both individuals with disability, and in a control group made up of university students, when landmarks had to be mentally visualized. However, significant differences were not found among the two groups. Based on these results, the authors concluded that individuals with intellectual disability do not have particular deficits in this type of inspection of mental images.

Ulterior proof that the use of imagery can be fruitful in cases of intellectual disability when coupled with physical training, Hemayattalab and Movahedi (2010) found benefits in the sports training of adolescents with mental retardation, being taught how to free throw in basketball after 24 sessions of mental imagery training associated with physical practice.

3.3 Neurological rehabilitation

Even in cases of psychomotor deficits after trauma or other neurological causes, training with mental imagery has shown to be beneficial (Jackson, Lafleur, Malouin, Richards & Doyon, 2001).

The benefits of this type of rehabilitation in the therapeutic treatment of stroke patients have been found in a number of studies (Batson, 2004; Johnson-Frey, 2004; Liu, Chan, Lee, & Hui-Chan 2004; Dickstein, Dunsky, & Marcovitz, 2004; Dunsky, Dickstein, Ariav, Deutsch, & Marcovitz, 2006; de Vries & Mulder, 2007; Holmes, 2007; Page, Levine, & Leonard, 2007; Zimmermann-Schlatter, Schuster, Puhar, Siekierka, & Steurer, 2008; Nilsen, Gillen, & Gordon, 2010); the greatest effects have been found in mental imagery training coupled to relaxation (Driediger, Hall, & Callow, 2006; Evans, Hare, & Mullen, 2006).
Dijkersman, Ietswaart, Johnston and MacWalter (2004), used motor imagery training in a group of 20 patients two years after stroke incidence in which they imagined the affected limb. They found a significant improvement in almost all the motor functions treated and monitored (except the dynamometer), even if general improvement in the control of attention and perception was not found. Sharma, Pomeroy and Baron (2006) also found improvements in motor functions after mental imagery training.

Lee, Song, Lee, Cho and Lee (2011), found an increase in deambulation capacity in stroke patients, after imagery training focused on movement. The improvement, in terms of velocity of deambulation, wideness of step, and strength of support in both the paralyzed side, as well as the contralateral side, was significantly superior in respect to a control group made up of patients that had not engaged imagery training.

In yet another group of stroke patients, afflicted with chronic post-effects, Hong, Choi and Lee (2012) have shown the utility of associating mental imagery training to electromyographic stimulation; a benefit of training mediated by a brain-computer interface in the rehabilitation of these patients was shown by Prasad, Herman, Coyle, McDonough, and Crosbie (2009), and confirmed by Ortner, Irimia, Scharinger and Guger (2012). Also, home-based motor imagery training showed efficacy in gait rehabilitation in post-stroke hemiparesis (Dunsky, Dickstein, Marcovitz, Levy, & Deutsch, 2008).

Steenbergen, Crajé, Nilsen and Gordon (2009), and Steenbergen, Jongbloed-Pereboom, Spruijt and Gordon (2013), not only studied movement execution but also the programming of motor gestures after motor imagery training in a group of children afflicted with hemiplegic, unilateral cerebral paralysis. Their results suggest that beginning this type of training early is highly beneficial.

Even in cases of visual-spatial neglect, imagery training has been found effective (Smania, Bazoli, Piva, & Guidetti, 1997; McCarthy, Graham Beaumont, Thompson, & Pringle, 2002; Welfringer, Leifert-Fiebach, Babinsky, & Brandt, 2011); intensive 4 week training resulted in the long lasting improvement of visual-kinesthetic abilities and in temporal-spatial orientation (Leifert-Fiebach, Welfringer, Babinsky, & Brandt, 2013).

Mental imagery training can help increase motor functions not only in cases of brain damage, but in spinal injury as well (Cramer, Orr, Cohen, & Lacourse, 2007; Roosink & Zijdewind, 2010; Rienzo, Guillot, Rode, & Collet, 2012; Grangeon, Revol, Guillot, Rode, & Collet, 2012), in burn patients (Guillot, Lebon, Vernay, Girbon, Doyon, & Collet, 2009), in Parkinsons, and in alleviating the pain of phantom limbs (Dickstein & Deutsch, 2007; McAvinue & Robertson, 2011).
In the area of child rehabilitation, Joffe, Cain and Marić (2007), applied mental imagery training in a group of children with specific language disorders, bad readers and with low reading comprehension abilities, and in a control group of normally developing children. They obtained significant results in terms of the improvement of comprehension after just brief training.

A standardized training regimen for neuro-rehabilitation has been introduced by Wondrusch and Schuster-Amft (2013). In fact, for some time now, technology-supported protocols have been used in neurological rehabilitation (Morganti, Gaggioli, Castelnuovo, Bulla, Vettorello, & Riva, 2003).

3.4 Rehabilitation of the mentally impaired elderly

Even in cases of cognitive deterioration in the elderly, a number of studies have examined the role of imaginative functions. The importance of the evaluation of visual imagery in Alzheimer’s was emphasized by Kosslyn and Dror in 1992, and has been confirmed by our data on elderly patients with and without deterioration, presented in the previous section. Few studies, however, have proven the benefits of using mental imagery training as prevention or treatment in elderly subjects.

Zehnder, Martin, Altgassen and Clare (2009), in a meta-analytic review of the efficacy of memory training in the elderly (including imagery techniques), found contradictory results, most likely due to the different development of imagery components in working memory, that in some cases can negatively affect the efficacy of training (Schott, 2012).

Malouin, Richards and Durand (2010) have argued that even if visual-motor imagery capacity tends to decline with age, in correspondence with the decline of working memory, the vividness of motor imagery is generally preserved in the elderly, and can be exploited in imagery training even in cases of deterioration. This strategy could be extended to training in cases of deterioration, after mild cognitive impairment and in dementia, and in more severe cases, after evaluation of the residual visualization capacities, and of the possibility of applying a training regime based on them.

According to Hussey, Smolinsky, Piryatinsky, Budson and Ally (2012), even if it is true that neurocognitive deficits do not allow patients with Alzheimer caused dementia to perform complex imagery (as also Grossi, Becker and Trojano, 1994, have shown), evidence exists that residual imagery functions can be used in rehabilitation interventions to improve visual recognition abilities.
4. Possible research and intervention developments based on Mental Imagery.

Experimental research can assist in acquiring a deeper understanding, of which specific applications of mental imagery training can be the most helpful for optimal learning, training or rehabilitation of motor gestures.

For example, the possible operational applications deduced by a simulation study using the training of feedforward and recursive artificial neural networks (Di Nuovo, Marocco, Cangelosi, De La Cruz, & Di Nuovo, 2012; Di Nuovo, Marocco, Di Nuovo, & Cangelosi, 2013), are both interesting and useful to our discussion. According to these authors, the integration of mental imagery in the development of the learning of actions in the networks is valid, but primarily for actions that are not very different in complexity and/or difficulty to those actions learned without it. This means that the studying of the learning curve is essential in order to periodically integrate the mental imagery training in a way that does not create too great a discrepancy between what has been learned and what has to be inferred from imagery.

A further line of study focuses on the cognitive background of motor performance in manual action (Schack & Ritter, 2009), particularly during the rehabilitation of hand function after a stroke (Braun, Beurskens, Schack, Marchellis, Oti, Schols et al., 2007; Braun, Kleynen, Schols, Schack, Beurskens, & Wade, 2008).

Schack and Ritter (2013) learned that central costs and interference in manual actions depend solely on how these movements are represented on a cognitive level. They show that dexterity in manual action and task-related object manipulation is accompanied by order formation in memory. Such order formation in action knowledge reduces the cognitive effort required to activate relevant information. The authors underline that representation and learning in motor action may promote relevant bridges between experimental research and cognitive robotics. For an example of this approach applied to the bootstrapping of numerical cognition through motor representations of finger counting in a cognitive robot, see De La Cruz, Di Nuovo, Di Nuovo, & Cangelosi (2014).

In conclusion, mental imagery and mental simulation are fundamental cognitive capabilities that have the potential to improve the performance of complex artificial systems, going beyond simple problem solving tasks, and enhance the next generation of humanoid robots suitable for assisting rehabilitation. For a special issue on mental imagery in artificial cognitive systems and robotics, see Di Nuovo, De La Cruz and Marocco (2013).
This research should be supported by a highly interdisciplinary approach, in which the integration of neuropsychological studies on mental imagery for human action performance is blended with innovative neurocognitive models and emergent robotics technology. This will result in novel engineering principles for the design of technological aids, including humanoid robots, capable of embodied mental simulations, human-like motor performance, and adaptive social interaction through a high-level of awareness. Enhanced possibilities of rehabilitation may stem from technological improvements grounded in cognitive theories of mental imagery.

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Abstract

Over the past two years, six sets of Italian scholars have responded to questions posed by Giangreco, Doyle and Suter (2012) in a descriptive study about school demographic data pertaining to the inclusion of students with disabilities in Italy. In this reply to those responses, the authors discuss five concerns they share in common with their Italian colleagues: (a) disability identification, labeling and disproportionate placement, (b) the role of the medical model in education, (c) lack of shared definitions about inclusive education, (d) roles and capacity of general education teachers, and (e) micro-exclusions in general education schools and classrooms. The authors conclude with potential future directions to facilitate inclusive educational opportunities and outcomes for all students.

Keywords: Inclusive education; School personnel roles; Disability identification.
1. Italian and american progress toward Inclusive Education: common concerns and future directions

“Nulla si fa senza volontá!” “Without commitment nothing gets done!” This Italian proverb offers a simple truth, namely that commitment is foundational to the pursuit of achieving any worthy aim, in this case quality inclusive educational opportunities for children and youth with disabilities. This is the reason why, in 2011, the first two authors were drawn to spend a sabbatical semester studying the inclusion of children and youth with disabilities in Italy (Giangreco & Doyle, 2012), a country with a longstanding international reputation for its practice of integrazione scolastica and an ongoing commitment to including students with disabilities in general education classrooms (Canevaro & de Anna, 2010; D’Alessio, 2011). We immersed ourselves in Italian culture and schooling, positioning ourselves as learners in an effort to better understand Italian approaches to including students with disabilities and gain insights that we might apply in our own context in the United States.

As one aspect of our sabbatical visit, we collected a convenient and modest set of school demographic data from 16 schools in five regions of Italy (i.e., Lazio, Lombardia, Puglia, Sicilia, Veneto) that we analyzed and shared in Life Span and Disability (Giangreco, Doyle, & Suter, 2012). Given our emerging understanding of the Italian education system and its many intricacies, we thought it would be imprudent to make judgments about the meaning of these data through our lens as Americans. Instead, we explained our understanding of these same types of demographic data based on some of our recent research in an American context (e.g., Suter & Giangreco, 2009; Giangreco, Broer, & Suter, 2011; Giangreco, Suter, & Hurley, 2013), posed a series of questions, and invited Italian scholars to share their perspectives about what these data mean to them in the Italian context. We were grateful to receive six replies from a set of respected Italian scholars who study inclusive education and disability-related issues (Di Nuovo, 2012; D’Alessio, 2013; Ianes, Zambotti, & Demo, 2013; Soresi, Nota, Ferrari, Sgaramella, Ginevra, & Santilli, 2013; Viannello, Lanfranchi, & Pulina, 2013; Zanobini, 2013).

2. Common Concerns

The respondents to our original article addressed a wide range of important issues, many more than we can address in this forum. In the following sections
we discuss a selected subset of issues we identified as concerns we share in common with our Italian colleagues. In considering potential future directions related to these common concerns we share in common with our Italian colleagues, it is important to recognize that although both Italy and the United States have national laws ensuring the education of students with disabilities, these legal mandates are implemented differently across and within both of the countries. In other words, there is not one single Italian or American model of school inclusion for students with disabilities, rather there is variation regionally and from school to school. As highlighted by Vianello and colleagues (2013), although the Italian context is characterized by substantial autonomy, there seems to be agreement at multiple levels (e.g., Ministry of Education, researchers, local schools) on a set of foundational points: (a) specialized support teachers are available to the entire class, not exclusively assigned to support a student with a disability; (b) the creation of small special classes where students with disabilities are grouped together should be avoided as much as possible; (c) students with disabilities and those with other special educational needs should be educated in general education classrooms as much as possible; and (d) it is an expected part of a general education teacher’s role to support students with disabilities in the general education classroom.

As a matter of clarification, in response to valid points raised by D’Alessio (2013) and Soresi et al. (2013), by presenting only a small set of school-level service delivery data in our original study we did not mean to suggest that the types of structural issues illuminated by such data (e.g., personnel utilization) were the exclusive or most important areas of need related to improving access to inclusive schooling. Rather, school service delivery information was a readily accessible data set that could reasonably be collected during our sabbatical visit. We consider such service delivery data to be only one important piece of a much larger and complex puzzle that constitutes inclusive schooling (e.g., attitudes, discrimination, teacher preparation, classroom practices, supports) as highlighted in the replies to our study.

2.1 Concern 1: Disability Identification, Labeling and Disproportionate Placement

Who is disabled? Which students need extra support in schools? These seem like straightforward questions, but of course they are not; they are intertwined with a multitude of societal, political, financial, and educational issues. As Di Nuovo (2012) and D’Alessio (2013) point out, disability is socially constructed and therefore is defined differently around the world. Access to individualized supports based on disability labeling represents one of the most fundamental
differences among countries, thus it poses challenges in making international comparisons (D'Alessio & Watkins, 2009). Italy and the United States are prime examples of such differences.

In recent history Italy has certified approximately 2% of its school-aged population as disabled and eligible to access special education services (Vianello et al., 2013), whereas the United States identifies approximately 13% (U.S. Department of Education, 2013a). Despite the smaller percentage of students certified as disabled in Italy, several of the respondents expressed concern that the percentages in Italy continue to increase. It is important to recognize that in Italian schools nearly 67% of students with certified disabilities are classified as having intellectual disabilities (Vianello et al., 2013). Whereas in US schools, only about 7% of students identified as disabled and eligible for special education are classified as having intellectual disabilities (U.S. Department of Education, 2013a; 2013b). As pointed out by Vianello and colleagues (2013), in Italy the term “disability” refers to a subset of students with special educational needs, only those that might be characterized as the “most severe” (p. 220).

In both Italy and the US access to additional, often specialized, supports and services remains linked to disability labeling. This approach may inadvertently encourage the labeling of students as disabled in order to provide them with access to needed support services. Ironically, while disability labeling was put in place in an effort to help students, it may actually contribute to stereotyping and perpetuate discrimination against some individuals with disability labels (Di Nuovo, 2012).

Given the Italian practice of assigning support teachers to classrooms where students with disabilities are placed, Di Nuovo (2012) expressed concern that in some situations more students with learning disabilities (LD not certified as “disabled” in Italy) are being disproportionately placed in general education classes where a student who has a disability is enrolled so that those students with learning disabilities can have access to a support teacher in the classroom and others agreed (Vianello et al., 2013). As stated by Ianes and colleagues “... it is very common for LD students to study in classes where a support teacher is present” (2013, p. 67). However, they pointed out that this is not necessarily deliberate. There are so many support teachers it is hard to place a student with learning disabilities in room where a support teacher is not assigned.

In the US context, historically the purposeful placement of a disproportionately high percentage of students with disabilities and other special needs in the same schools (e.g., regional special classrooms for students with severe disabilities) and classes (e.g., placing a small group of students with disabilities in one general education class rather than distributing them across classes) has been referred
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to as violation of natural proportions (Brown, Nisbet, Ford, Sweet, Shiraga, York et al., 1983). When Brown and colleagues (1983) argued against the contrived and disproportionate congregation of students with disabilities and in favor of a more naturally occurring distribution, their focus was on students with severe and multiple disabilities, who in the 1980s were slowly being shifted from special education (disability-only) schools to regular schools and classes. National data are not available in either Italy or the US documenting the extent to which students with disabilities and other special or nonstandard needs might be disproportionately grouped together in general education classes. This remains a fertile opportunity for future research to explore the impact of natural proportion placements versus those that violate the naturally existing proportions to varying extents.

A contemporary concern is emerging in the US related violating natural proportions as some schools explore co-teaching models. In some cases schools are placing a disproportionately high percentage of students with disabilities or other special needs in particular classrooms to justify the assignment of a special education teacher to co-teach in those classrooms. This violates the natural proportions of students with and without special needs, thus creating new, self-inflicted challenges that may be more likely to occur under such conditions (e.g., stigma, concentration of students with more intensive needs, lower group expectations, difficulty meeting students' educational and social needs, fewer communication/language models, additional stress on school personnel).

An insidious problem associated with certifying students as disabled to access services, is that it assumes that the problems resulting from the disability are located exclusively within the person who has been labeled as having a disability. More contemporary conceptualizations of human diversity, while acknowledging the differing characteristics of individuals along many dimensions (e.g., sensory, physical, intellectual, social/behavioral), focus more on the person-environment fit and the nature of supports needed for a person participate in typical environments (Wehmeyer, Buntinx, Lachapelle, Luckasson, Schalock, Verdugo et al., 2008). So while it is important for students with disabilities to learn new skills, it is equally important for various aspects of the environment to change (e.g., peoples’ attitudes, organizational structures, classroom practices, personnel utilization) in ways that allow universal access with individually appropriate supports.

2.2 Concern 2: The Role of the Medical Model in Education

In part, the differences in disability identification and support services in Italy and the US arise from variations in how the medical and allied health systems intersect with education. What the countries share in common is an ongoing need to clarify roles and collaborate across disciplines, agencies, and services providers.
In Italy, the determination of disability, which is later used by schools as a basis to provide support services, is not made by schools but by local health agencies using the World Health Organization’s (2001) *ICF-International Classification of Functioning, Disability and Health*. Ianes and colleagues (2013) explain that a predominantly medical model still is in place related to diagnosis and eligibility for special education services in Italian schools. This differs from the US where a medical determination of disability may be just one aspect of a school-based team determining eligibility for special education services.

In Italy, specialized support services (e.g., physical therapy, occupational therapy, speech/language pathology) typically are not provided at or by schools, but rather during nonschool hours. Italian law does allow for students with severe disabilities to receive some of these therapy services during part of the school day (e.g., the first hour or two of the school day), but they are typically provided away from school at a health clinic or agency that provides services to individuals with disabilities. Ianes and colleagues (2013) expressed concern that some students with disabilities do not attend school part of the day because of this medical model of therapeutic and rehabilitative service delivery; they reported that 16.3% of students with disabilities “visited specialist or health centres for rehabilitative or therapeutic activities during school time” (p. 65). The extent to which these services support students’ education is unknown and the extent of collaboration between schools and these medical/health-related agencies varies from school to school and student to student.

In the US, under the *Individuals with Disabilities Education Act* (2004), specialized support services (referred to as “related services”) such as physical therapy, occupational therapy, and speech/language pathology, are provided at school if they are deemed to be educationally relevant and necessary, meaning that they are required in order for a student to access education and/or gain meaningful benefit from education (Giangreco, 2000; 2001). The manner in which these services are provided during the school day varies from isolated, pull-out services to integrated and consultative services in the classroom; the educational relevance, necessity, and impact of these services on the education US students with disabilities remain significant unanswered research questions.

In the US, even though the school is responsible to determine eligibility for special education and related services, potentially overlapping roles between physical/mental health agencies and schools continues to create confusion and perpetuate service fragmentation. For example, parents seeking specialized or additional supports for their children in schools often turn to physicians, special health clinics, and other allied health professionals to write prescriptions or recommend services that are sent to schools with the expectation of implementation based on assumption of the perceived authority of medical/allied health professionals.
Medical and other health professionals who are external to the school team (e.g., associated with nonschool clinics and organizations) are often not well positioned to make educationally relevant and necessary recommendations because they typically do not have sufficient contextual information (e.g., student’s educational goals, school staffing, classroom practices, potential overlap with other services).

While the ways in which the medical model and education intersect in Italy and the US are different, both countries continue to experience challenges with coordination between the health, mental health, and educational systems. D’Alessio (2013) argues strongly for rejecting ideologies, such as traditional medical models, that unnecessarily pathologize difference and contribute to abilist constructions of difference (Medeghini, D’Alessio, Marra, Vadala, & Valtellina, 2013). This perspective encourages us to continually scrutinize our approaches and view individual differences as an expected and accepted aspect of human diversity.

2.3 Concern 3: Lack of Shared Definitions about Inclusive Education

In both Italy and the US, there continues to be lack of agreement about the meaning of terminology such as: inclusion, inclusive education, and integration. Which is better? Which is more progressive? Are they the same or different? Do these terms refer to primarily students with disabilities or do they include others? To which should we aspire (or something else)? There continues to be debate about these questions between and within countries.

In the US, the Individuals with Disabilities Education Act (2004) does not use terms such as inclusion, inclusive education, or integration. Rather, it relies on the concept placing students with disabilities in the “least restrictive environment”, meaning that “to the maximum extent appropriate, children with disabilities... are educated with children who are nondisabled... special classes, separate schooling, or other removal of children with disabilities from the regular educational environment occurs only if the nature or severity of the disability is such that education in regular classes with the use of supplemental aids and services cannot be achieved satisfactorily” (Code of Federal Regulations, 2006, 300.114). Ironically, while meant to encourage inclusion, the second part of the least restrictive environment mandate, at times, has been used to justify the continued segregation of some students, especially those with the most severe disabilities (Giangreco, Dymond, & Shogren, in press; Taylor, 1988). Across the US, far too many students who have intellectual disabilities and other developmental disabilities are placed in self-contained special education classes or schools, and thus are denied opportunities to build relationships with their peers without disabilities as well as access to general education classrooms and curricula taught by qualified general education teachers.
D’Alessio (2013) argues that the historic policy of *integrazione scolastica* in Italy is not necessarily inclusive, while some others perceive the term as a synonym for inclusion. She encourages the examination of “attitudinal, structural and organizational barriers that prevent the most effective special education delivery services to promote inclusive-oriented schools” (p. 98). Her perspectives are rooted in a disability studies and human rights perspectives articulated in the *UNESCO Policy Guidelines on Inclusive Education* (2009), the *United Nations Convention on the Rights of Persons with Disabilities* (2006).

Regardless of the terminology one favors, Ianes and colleagues (2013) expressed concern that integration/inclusion for some students with disabilities in Italy may be regressing (p. 77). Zanobini (2013) expressed a related concern that students with disabilities are excluded from student assessment protocols, which may contribute to low expectations. Similar concerns exist in the US, where subgroups of students, especially those with intellectual disabilities and other severe disabilities, remain substantially excluded from general education opportunities (Doyle & Giangreco, 2013).

While both Italy and US have a history of national legislation supporting the education of students with disabilities and their access to general education environments and curriculum, such laws alone are not sufficient to ensure quality educational opportunities. It is difficult to make substantive progress toward equitable opportunities and supports for students with disabilities and other needs unless the adults supporting those students (e.g., teachers, administrators, families, self-advocates, community members) have shared meaning associated with the language they are using to discuss the ideas and practices they are seeking to advance.

2.4 Concern 4: Roles and Capacity of General Education Teachers

Each of the aforementioned concerns is inextricably linked to roles and capacity of the general education system and its perceived responsibilities to support all students. Problems arise when the only way students can gain access to otherwise unavailable supports is to be labeled disabled and when access to such specialized services may inadvertently distance a student from the benefits associated with being taught in an inclusive classroom by a qualified regular education teacher alongside classmates without disabilities. Di Nuovo (2012) explains that students with specific learning disabilities (referred to in Italy as DSA, Disturbo Specifico dell’Apprendimento) typically are not certified as disabled, yet these students often need additional instructional supports. Whereas in the US students with “specific learning disabilities” are identified as “disabled” under the Individuals with Disabilities Education Act (IDEA) (2004) and constitute, by far, the largest subcategory of disability at approxi-
mately 40% of students who labeled disabled (U.S. Department of Education, 2013b).

Zanobini (2013) points out that a recent Italian law (Law 170, 2010) now recognizes the support needs of students with specific learning disabilities, but does not address their needs under the national disability legislation because Italians are committed to the practice of general classroom teachers educating students with specific learning disabilities. The expectation is clear that the inclusion of students with learning disabilities is expected in regular classes through improved teaching strategies delivered by general education teachers (e.g., personalized learning plans with appropriate accommodations; Ianes et al., 2013). Even with the passage of this recent legislation in Italy, it has not been uniformly implemented nationwide, just as the Individuals with Disabilities Education Act has not been uniformly implemented in the US.

These issues have spawned important conversations in Italy as educators wrestle with distinguishing between students who are considered “disabled” and those who have other “special needs” that might arise from non-health characteristic reasons (e.g., immigration, non-dominant language, atypical family circumstance, socioeconomic status). Zanobini (2103) suggests referring to such needs as “non standard” (p. 88) rather than “special needs”. She points out that in some circumstances teachers may “encounter more difficulties managing cultural, linguistic, and social differences than differences linked to the presence of some disability” (p. 88). This perspective supports the points raised by D’Alessio (2013) that inclusive education is a much broader concept that extends beyond students with identified disabilities. Some of our earlier research has encouraged us to view the active engagement of the classroom teacher with his or her students who have disabilities (or other nonstandard needs) as one of the most important elements of creating classroom environments that are conducive to learning for all students (Giangreco, Broer, & Edelman, 2001); this continues to be an ongoing challenge in need of attention.

2.5 Concern 5: Micro-Exclusion

National statistics meant to capture elusive terms, such as inclusion and integration, often focus on discrete and narrow components without adequately capturing their nuances. For example, in the US, the physical placement of a student with a disability in a general education classroom is a primary marker of what many American educators might consider or refer to as “inclusive education”. Yet, it is quite clear that general education class placement, while a necessary component, does not equal inclusion. Students with disabilities can be physically placed in general education classes and yet be excluded within
the classroom either physically or programmatically, often both (Giangreco, Carter, Doyle, & Suter, 2010).

D’Alessio (2011; 2013) concurs from an Italian perspective that mere placement is not enough to be included and that micro-exclusions occur when an individual student is separated within the classroom. A classic scenario in US schools occurs when a student with a significant disability (e.g., autism, intellectual disabilities, multiple disabilities) is placed in a general education classroom and then spends a substantial amount of time physically separated from classmates at the back or side of the classroom, working primarily with a one-to-one assistant; such arrangements, while quite common, have been exposed as problematic for students (Giangreco, 2010).

Micro-exclusions are also evidenced in both countries when students with disabilities receive a substantial amount of their instruction from someone other than the classroom teacher. Zanobini (2013) expressed concern that when the “insegnante di sostegno” (support teacher) does much of the instruction of students with disabilities in the classroom it can interfere with classroom teachers becoming instructionally engaged with their students who have disabilities. This concern parallels data collected in the US where paraprofessionals (assistants) providing a substantial amount of instruction to students with disabilities are the personnel who have been identified as potentially, although inadvertently, interfering with teachers’ instructional engagement with the students with disabilities who are placed in their classes (Giangreco, Edelman, Luiselli, & MacFarland, 1997; Giangreco, Broer, & Edelman, 2001; Giangreco, 2013).

Ianes and colleagues (2013) expressed their concern about two other variations of micro-exclusion: (a) homogeneous grouping of students with some type of special need within classrooms (consist with our earlier stated concern about violating natural proportions), and (b) “pull-out phenomenon”, where students are separated from the general education classroom to receive instruction, which they characterized as “both established and widespread” (p. 60). In these micro-exclusion variations, instruction is provided by an “insegnante di sostegno” (support teacher).

While some of our Italian colleagues identified pull-out instruction as a well-known concern related to students with identified disabilities, Ianes and colleagues (2013) expressed alarm about the extent to which students with learning disabilities (LD, DSA in Italy) experienced pull-out instruction from class; they reported that “roughly 10% of LD students spend some significant part of school time separated from the rest of the class” (p. 65). They consider the extent to which pull-out services are creeping into practice for a segment of students with learning disabilities as problematic, resulting in undesirable homogeneous grou-
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Ianes and colleagues (2013) expressed additional concern that the amount of time spent in pull-out instruction increases as students get older and that this affects students with severe disabilities more extensively than those with mild disabilities. This parallels longstanding concerns in the US about pull-out instruction (Rea, McLaughlin, & Walther-Thomas, 2002). Although overall the percentage of US students with disabilities placed in general education classes has steadily increased over time, as these students with disabilities get older they spend less time in regular education classrooms, especially those with low-incidence disabilities (McLeskey, Landers, Williamson, & Hoppey, 2012; United States Department of Education, 2013b). A recent study conducted in Vermont reported that approximately 75% of all instruction provided by special education teachers to students with disabilities in inclusion-oriented schools was in a pull-out format (Giangreco, Suter, & Hurley, 2013). Italian research indicates that “…students with disability leave the classroom less often when active, cooperative, and metacognitive didactic methodologies are employed…” and “…students who spend all of their time in class obtain better results both in terms of learning and social development…” (Ianes et al., 2013, p. 59). Combined, these data and perspectives highlight ongoing concerns about the extent to which students with disabilities are being excluded from and within general education classrooms.

3. Future Directions

While many people in both Italy and the US may be justified in feeling positively about the progress made toward including more students with disabilities in general educational classrooms in certain regions and schools, it is vital to recognize that such progress is temporal and relative — much work remains to be done to ensure fully inclusive, equitable, and quality educational opportunities and outcomes for all students. While we can celebrate our collective successes, we must simultaneously be vigilant to guard against complacency that impedes progress, leads to stagnation, or in the worst-case scenario results in back sliding.

During our visit to Italy we were repeatedly impressed by the: (a) pervasive attitude of welcoming students with disabilities and other nonstandard needs, (b) expectation of their presence in general education classes, (c) expectation that general education classroom teachers should work directly with students who have disabilities in their classroom, and (d) sense that what is considered “normal” was wide. Each of these pillars is foundational to a positive future direction for
students with disabilities and other nonstandard needs. They represent antidotes to approaches that: (a) use testing and team meetings as vehicles to remove students with disabilities and other nonstandard needs from the classroom, (b) place the burden on students and families to earn their way into inclusive environments by meeting moving and ultimately arbitrary targets, (c) allow for micro-exclusions and corresponding low expectations, and (d) unnecessarily or excessively pathologize disability-related and nonstandard differences.

The path to the future will be paved by removing barriers to inclusion. While it is incumbent on us, as an educational community, to provide quality opportunities for teaching and learning, future directions should be at least as much about changes in the attitudes, expectations, and practices of people without disabilities as it is about skill development for students with disabilities and other nonstandard needs. In other words, the focus should be on improving the person-environment fit, rather than focusing exclusively or predominantly on student characteristics. Employing approaches such as Universal Design for Learning (Rose, Meyer, & Hitchcock, 2005) and personal learning plans for all students (Zanobini, 2013) are consistent with such future directions.

When tackling future challenges with optimism, an essential trait for effective problem-solvers (Parnes, 1997), the path toward a better future can appear simple and easy, we recognize that some aspects may be complex and difficult. Another notable aspect of the Italian journey toward inclusive education and a guidepost for future progress has been their collective willingness to expect the change process to be messy and embrace this disequilibrium as necessary to accomplish substantial change (Canevaro & de Anna, 2010). Italy passed legislation in the 1970s that provided few opportunities for maintaining the status quo; the only option was to move forward through the tangle that was created by a rapid change for which everyone reportedly was not ready (Vitello, 1991). As Canevaro and de Anna suggested (2010), the presence of students with disabilities and the requirement to include them provided the conditions that facilitated progress in a way that would not have been possible using an incremental approach where students were only included once everyone was prepared and felt ready. To advance requires the presence of students with disabilities; certainly there will be challenging times, yet it seems logical that the only way to advance our inclusive practices is to pursue them in context.

This context-based future will require ongoing attention to the curriculum, instruction, supports, and inclusive models of service delivery (e.g., school organization, personnel utilization, multi-tiered systems of support) in the form of empirical research and individualized data-based decision-making in ways that retain the underlying values embedded in the respective legislation and constitutions of both countries (Giangreco & Doyle, 2011).
This will require changes in teacher and school administrator preparation at universities to ensure that all school personnel have the foundational knowledge and skills to work with the full range of students with and without disabilities they will encounter in the classroom (D’Alessio, 2013; Soresi et al., 2013; Zanobini, 2013). As we pursue this multi-pronged future agenda, we must listen more often and more carefully to the perspectives of self-advocates and families (Broer, Doyle, & Giangreco, 2005; D’Alessio, 2013; Ianes et al., 2013).

As we consider future directions in both Italy and the US, the discussion needs to extend beyond graduation or compulsory schooling, but to important post school outcomes (e.g., health, social networks, employment, community engagement, culturally normative residential living options) (Di Nuovo, 2012; Soresi et al., 2013). Soresi and colleagues (2013) eloquently stated a problem that persists for too many adults with disabilities and their families:

At the end of compulsory school, many students with impairments are unfortunately placed in private vocational schools, or in special classes in health centers or in other social care contexts that, in our view, represent a sort of “institutionalization”. For many of them a “restriction” phase to their social participation starts which lasts across all the lifetime. This is the most insidious threat and the strongest “betrayal” to the inclusion principle: to let people with disabilities and their families benefit from inclusion for a few years and then force them, just when work inclusion should follow school inclusion, to accept special and separate treatments (p. 207).

Ultimately, we cannot settle for quality inclusive schooling followed by an unduly restricted adult life. The impact of inclusive schooling will not be fully realized until it leads to inclusive adult lives, where education yields more and better outcomes for our students with disabilities and other nonstandard needs. Zanobini (2013) suggests using quality of life indicators to assess progress toward meaningful post-school outcomes; we have long shared this view (Dennis, Williams, Giangreco, & Cloninger, 1993).

4. Conclusion

While the collective commitment that led us to study inclusive education efforts in Italy is foundationally important, commitment alone is not sufficient. Meaningful progress requires a vision of a better future, along with cyclical and persistent analysis, planning, and action. By exploring our common ground, we hope to encourage actions that contribute to the valuing of individuals with disabilities as full citizens and their access to the full range of educational opportuni-
ties, with appropriate supports, available to their peers without disabilities in fully inclusive environments.

Finally, we wish to extend our appreciation to our Italian colleagues for publicly sharing their perspectives, concerns, and ideas for advancement. By doing so, these national leaders have expanded and elevated what has been an ongoing intra-national conversation in Italy, primarily in the Italian language, to an international conversation in English that allows more educators and advocates around the world to benefit from their wealth of experiences and insights. Even places with well-deserved reputations for their progressive efforts related to inclusive schooling, such as those in Italy, benefit from critical self-reflection and self-analysis in order to sustain and advance their practices and policies. As Soresi and colleagues (2013) remind us, as we aspire toward being truly inclusive we must shift away from a focus on the “special”. We thank our Italian colleagues for engaging in this conversation and helping us to learn from Italy’s journey, which continues to serve as a beacon of inclusive schooling possibilities -- it still has much to teach us.

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