

Case Report

Support Intervention for Siblings of Children with Developmental Disabilities: An Italian Experience

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Abstract

Siblings of children with disabilities showed a high incidence of behavior problems. Specific support programs are not provided. The aims of the present study are to assess the behavior of a group of Italian siblings of children with neurodevelopmental disabilities and to analyze the possible influence of a specific support intervention.

Siblings of children with different disabilities and their parents completed the Multidimensional Anxiety Scale for Children (MASC), the Children's Depression Inventory (CDI) and the Child Behavior Check List (CBCL). They were assessed at baseline and after a 12 weeks period of training with a group intervention improving the understanding and adjustment of sibling. Parents attended groups concurrently on parallel session. A control group performed the same assessments and used as reference data. Fifty-six siblings (mean age 9.5 ± 1.6) of children with different disabilities (autistic spectrum, cerebral palsy, intellectual disabilities) and their mothers completed the assessments. At baseline siblings showed a pathological score in Internalizing, Externalizing and Total CBCL with a statistical significant improvement at the end of the study. At MASC and CDI, siblings reported mean scores within the normal range with no statistical difference before and after the treatment. Control group showed lower scores in all the scales than the sibling group at the beginning and the end of the study.

Our results confirm an elevated rate of behavior problems in siblings of children with disabilities assessed by parents with significant improvement after the treatment. Further studies in a large cohort of children may provide additional information on the possible effect of treatment strategies.

Keywords: Siblings; Disabilities; Children; MASC; CDI; CBCL

Introduction

In the last ten years several studies have focused on siblings of children with disabilities [1-13]. The sibling relationship constitutes a longest lasting family issue and it could be different when one member of the sibling pair has a developmental disability. Well siblings of children with disabilities showed a higher risk for developing emotional and behavioral problems than siblings of healthy children [14]; they could

engage in a rich, complex set of roles that might be expected to bring developmental benefits [11]. Learning a wide variety of roles may enhance perspective-taking abilities, increasing children's abilities to understand the feelings of others [12]. On the other hand assume major childcare roles have been found to have more behavior problems, such as anxiety, depression poor self-esteem and to experience increased conflict and decreased positive interaction with their siblings [3,7,9]. Furthermore behavioral problems could be related to parents

conduct, directing more attention to children with disabilities than to typically developing siblings [12]. Siblings and parents should be assisted by service providers, emphasizing and promoting the positive aspects of living with a disabled child both to parents and to other children in the family [3,4,7,10-12]; siblings should be provided with more information about developmental disabilities for their own understanding and to explain better to other children why their brother or sister behaves differently [1].

Recently, few studies proposed group interventions to improve knowledge of disability and sibling relationship in self-selected siblings of children with disabilities with encouraging results [5,6,8,13]. Almost all studies were conducted out of the Europe and no-one in Italy. Therefore the purpose of the present study is to assess the behavioral development of an Italian sample of siblings of children with disabilities and to analyze the possible influence of a specific support intervention on their behavior.

Materials and Methods

Siblings of disabled children were prospectively collected from a regularly followed register from January 2011 to June 2013. A child neurologist (DMR) and a psychologist (VG) confirmed the diagnosis of the disabled children using the Diagnostic Statistical Manual, Fourth Edition, Text Revision and on motor and cognitive assessments [14-16]. In order to have a homogeneous cohort we only included the parents (both) living with the child, with no history of a severe or chronic medical condition (e.g. stroke, diabetes) and psychological disorder. All the siblings were attending normal education with no signs of any neurological or other chronic disorder.

All the siblings were involved in a 12 weeks period of training, for a total of 6 sessions of two hours, with a psycho-social intervention, performed by one psychologist (VG) and 2 pedagogist (PM, LF) and an adult sibling (CP). Siblings were divided in groups of 6-8 children, age selected with a difference of only one year of age for each group, irrespective of the diagnosis of the disabled sibling. Parents attended groups concurrently on parallel topics on information exchange with clinicians (psychologist and pedagogist) and parental awareness of sibling needs [6], including didactic presentation, group discussions, and an open forum with an adult sibling. Discussion topics were based on previous studies on emphasizing issues related to within family functioning, enhancing communication about a developmental disability with children, and understanding how the developmental characteristics of young children affect sibling knowledge of and coping with a developmental disability [13].

The design of the intervention for siblings was based upon a review of the literature and consultation with pediatric nurse

clinicians, nurse researchers and child neurologist, previously published [6,8,13] with group intervention improving the understanding and adjustment of sibling of children with disability. Program focused on experiential learning sessions for disability education, problem solving and incorporated role-play, puppets, self-expressive crafts, relaxation techniques, and games; sibling knowledge and family information exchange; identifying and managing sibling emotions (isolation; over-identification and overachievement; embarrassment; anger, resentment, and neglect); siblings' individual needs. At the end of the program, siblings and their disabled brother/sister performed together the last session. Siblings created a videotape about their experiences that parents reviewed.

Each of the operators (psychologist/pedagogist) performed a specific course to conduct the support intervention. The intervention was described and discussed with parents and siblings and an informed consent was signed.

Psychological assessment

All children and their caregivers completed a battery of psychological rating forms for a comprehensive evaluation of behavior, depression and anxiety.

The Multidimensional Anxiety Scale for Children (MASC), a 39-item four point Likert style self-reporting scale for children and adolescents was used to measure anxiety symptoms (17, 18). Each item is rated on a 4-point Likert-type response scale ranging from 0 (never true about me) to 3 (often true about me). It has four subscales measuring physical symptoms (12 items), social anxiety (9 items), harm avoidance (9 items) and separation/panic (9 items). The MASC total and sub-factor raw scores were converted into standard T-scores using the MASC Profile forms for males and for females. T-scores are standard scores with a mean of 50 and a standard deviation of 10 [17,18] and a T-score >65 indicated the presence of anxiety symptoms. The Italian version of Children's Depression Inventory (CDI) was used to assess depression symptoms. The CDI is a self-rating scale mostly used to assess depressive symptomatology in children and adolescents aged 8-17 years. This scale consists of 27 items scored on a three-point scale indicating increasing severity of symptoms. According to Italian validation criteria, 19-point cut-off indicates the ideal threshold for a child at risk of depression [19,20]. Children completed the CDI and MASC with the assistance of trained psychologists. When necessary, following the manual's standard instructions, questions were read and eventually explained to the children [17-20].

Siblings' behaviour was assessed using the Italian version of Child Behaviour Check List (CBCL) [21,22]. In this test behaviour problems are reported by the child primary caregiver (the person who is most responsible for the day-to-day decision making and care of the child), that was the mother in

100% of cases. The CBCL consists of 118 items on which parents rate their child's behaviour by using 3-point scales of 0 (not true), 1 (somewhat or sometimes true), and 2 (very true or often true). The CBCL provides a total behaviour problems score, 2 second-order factor scores (internalizing problems, externalizing problems), and 8 syndrome scores (aggressive behaviour, anxious/depressed, attention problems, delinquent behaviour, social problems, thought problems, withdrawn, somatic complaints). Raw scores on each clinical factor were transformed to T-scores based on published norms [21,22]. Scores >63 are considered pathological, scores between 60 and 63 as borderline, scores <60 normal. All the caregivers of the groups completed the questionnaire.

The caregiver and the children completed the CBCL, MASC and CDI two times, at the beginning of the study and at the end of the 12 weeks period of training.

A sample of families of age-matched children, with at least one sibling without neurodevelopmental disabilities, was used as control group; they were recruited via schools attending regular classes in mainstream schools with no obvious or reported sign of mental, developmental or physical disabilities according to school medical records. None of them was receiving on-going prescription medication. The children and their caregivers completed the same questionnaires of the study group and the results were used as reference data.

At the time of enrolment in the study, parents of control group gave written consent and assent.

Statistical analysis

Data were presented as mean values (standard deviations [SDs]) for continuous variables normally distributed. The comparisons between control and study group at the beginning of the study and between pre and post-treatment were performed using the non-parametric test of Mann Whitney U. The level of significance was set at $p < 0.05$.

Results

A total of 56 siblings of disabled children were enrolled; the diagnosis of the disabled children was: 31 children with autistic spectrum, 10 with cerebral palsy and 14 with intellectual disability, with a mean age of 6.3 ± 3.0 . Because of the limited number of children of the single groups (autism, cerebral palsy and intellectual disability), a statistical intergroup comparison has not been performed. In each family, only one child showed a neurodevelopmental disability. In 50% the sibling was the first-born, 46% the second and in 4% the third. Their age ranging from 7 to 12 years (mean 9.5 ± 1.6), with 31 males and 25 females.

All enrolled families completed treatment with a high rate attending all sessions (90%). Complete pre-post parent and siblings' data existed for all of the 56 siblings. At the moment of the evaluation parent's age ranging from 28 to 55 years.

A total of 56 well-being children (28 males and 28 females) were enrolled and used as control group; their age ranging from 7 to 12 years (mean 10.1 ± 1.5). No significant differences were reported for age and gender between the control and study group ($p > 0.05$).

Study group: CBCL results

At the beginning of the study, the parents (mother in 100% of cases) reported a pathological score in 39%, 18% and 27 % respectively in Internalizing, Externalizing and Total CBCL questionnaire; at the end of treatment the percentage of pathological scores were lower (16%, 4% and 12% respectively) with a statistical significance for Internalizing ($p < 0.05$) Externalizing ($p < 0.01$) and Total scores ($p < 0.05$); a specific improvement was reported on withdrawn and aggressive behavior subscores ($p < 0.05$). Details of CBCL findings reported by the primary caregiver (mother in all cases) are reported in Table 1.

Table 1 CBCL results

	Internalizing mean±SD	Externalizing mean±SD	Total mean±SD	Withdrawn mean±SD	Somatic complaints mean±SD	Anxious Depressed mean±SD	Social problems mean±SD	Thought problems mean±SD	Attention problems mean±SD	Delinquent behaviour mean±SD	Aggressive behaviour mean±SD
baseline	60.9±10.4*	54.3±10.0**	57.5±10.3*	3.7±2.9**	2.0±2.4	6.8±4.6*	3.4±2.8*	1.1±1.4	4.7±3.3*	2.7±2.9	10.4±6.7**
Post-treatment	56.2±10.3*	48.6±10.0	52.6±11.0	2.4±2.1	1.4±1.5	5.3±4.3*	2.8±2.6*	0.9±1.2	3.7±3.1*	2.5±2.0	7.9±5.3*
Control group	50.8±10.2	45.5±9.7	47.4±11.1	1.6±2.0	1.4±1.5	3.2±3.0	1.4±1.4	0.9±1.1	2.2±2.7	2.0±1.9	4.7±4.3

Baseline Vs Post-treatment: * $P < 0.05$;

Baseline Vs Control group: + $P < 0.01$

Control group Vs Post-treatment: ° $P < 0.05$

Study group: MASC and CDI results

At the beginning of the study siblings reported mean scores on MASC and CDI within the normal range; only 3 and 1 children, respectively for MASC total score and CDI, showed a pathological score. No statistical difference in all the scores ($p > 0.05$) were observed before and after treatment. Details of the scores of the MASC and CDI are reported in Table 2.

Table 2 MASC and CDI results

	MASC tot mean±SD	Physical symptoms mean±SD	Harm avoidance mean±SD	Social anxiety mean±SD	Separation Panic mean±SD	Anxiety disorder mean±SD	CDI mean±SD
baseline	53.0±10.0*	50.3±8.3*	46.1±8.4	55.4±10.6*	56.7±11.0	50.9±9.0*	8.1±4.4*
Post-treatment	52.0±12.1*	49.4±11.2*	47.6±8.4	52.9±11.9	56.0±12.6	48.7±12.2	7.9±5.6*
Control Group	45.9±9.7	44.6±5.6	43.5±10.5	47.7±11.1	52.2±9.3	45.3±9.4	4.7±3.5

MASC: Multidimensional Anxiety Scale for Children

CDI: Children's Depression Inventory

Baseline Vs Post-treatment: NS

Baseline Vs Control group: * $P < 0.05$

Post-treatment Vs Control Group: ° $P < 0.05$

Control and study group comparison

Control group showed scores within the normal range in all the scales (Table 1 and 2).

At CBCL, the control group showed significant lower scores ($p < 0.01$) than the study group at baseline for internalizing, externalizing and total score and for sub-scale withdrawn, anxiety/depression, social problems, attention problems and aggressive behavior; significant lower scores ($p < 0.05$) were observed when comparing the control group with the study group at post-treatment for internalizing, anxiety/depression, social problems, attention problems and aggressive behavior.

At MASC, the control group showed significant lower scores ($p < 0.05$) than the study group at baseline for total score, physical symptom, social anxiety, anxiety disorder; significant lower scores ($p < 0.05$) were observed when comparing the control group with the study group at post-treatment for total score and physical symptom only.

At CDI control group showed significant lower scores ($p < 0.05$) than the study group at baseline and at post-treatment assessments.

Discussion

Siblings of children with neurodevelopmental disabilities are reported by parents to be more negatively affected by behavioral problems compared to siblings of typically developing children, probably because sibling relations have a crucial importance for child development, influencing cognitive, affective and social skills [4,10].

Due to the elevated level of behavior problems among children with neurodevelopmental disabilities, siblings of these children may experience more levels of stress that could be problematic especially at young age [9]. Furthermore these siblings report other negative outcomes as more conflicted sibling-child relationships and reduced social interactions [3,7]. Although support offered by health care professionals are limited or non-existing, recent studies demonstrated that behavior problems can be significantly reduced through effective interventions [9,23,24]. These studies were conducted in countries out from Italy and, in our knowledge, there is not a structured treatment and support programs adapted to siblings specific needs.

In the present study we reported the psychological profile of 56 Italian siblings of children affected by different types of neurodevelopmental disorders assessed before and after a support intervention. The intervention was very well received, with excellent attendance and high program completion rates. At the beginning of the study, parents of disabled siblings re-

ported a higher incidence of both internalizing and externalizing problems than the control group especially in withdrawn, anxiety/depression, social, attention and aggressive problems; although within the normal range, siblings showed a higher incidence in self-reporting anxiety and depression questionnaires than the control group, confirming the judgments of parents.

Our results are confirmed by the data of literature with an elevated rate of internalizing and externalizing behavior problems for siblings of children with autism or mental retardation [25].

After a period of 12 weeks of support intervention, an improvement in all the clinical area assessed were observed; parents of siblings reported significant lower scores in internalizing, externalizing and total scores at CBCL and mainly in withdrawn and aggressive behaviors. Although in self-reporting questionnaires (MASC, CDI), siblings presented similar scores at baseline and after the intervention, when comparing siblings at post-treatment assessment with control group, they reported more similar scores in different areas of externalizing behavior than the comparison at baseline, maintaining significant higher scores for siblings in internalizing scale only.

It is possible that, due to the intervention, increasing information and exposure to disability-related topics with communications between the children might have improved sibling knowledge of the child's condition and sense of connectedness with a decrease in the heightened negative impact experienced by siblings [9]. On the other hand the parent component of the program, including parental awareness of sibling needs, group discussions and topic presentation, provided information and debate regarding typical and atypical sibling behavior and relationships. It is possible that the reductions in parent-reported behavioral problems reflected changes in parent perspectives on sibling behavior, actual changes in sibling behavior, or a combination of both [8].

In considering the validity of our conclusions, the potential effects of some methodological limitations should be considered. The principal limitation of this study is related to the fact that it was not designed to test the single or synergic effects of the sibling and parent interventions or a possible placebo effect and therefore we cannot identify which session played a more important factor to change the behavior outcome.

Another limitation is related to the absence of a real control group as the one in the present study was used only as reference data for the questionnaires. Further studies should consider two groups of siblings, one with performing the support intervention and one receiving no special sessions.

Furthermore, previous researches [8] showed that siblings of

children with autism spectrum disorder, mental retardation, and psychiatric disorder presented less knowledge of these conditions at pretreatment and their knowledge increased more from pre to post-treatment in comparison to siblings of children with physical disabilities or medical disorders. In our study siblings were grouped together without regard to the type of the diagnosis of the child and a specific treatment effects across diagnostic conditions was not evaluated.

Conclusion

Our results, although preliminary, support early interventions to promote understanding and psychological adjustment among young siblings of children with developmental disabilities. Further studies, in a large cohort of children at different ages with specific target of intervention for well siblings or for those at high risk of negative outcomes, and with long term follow up, may provide additional information on the possible effect of treatment strategies.

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