Systematic review

Occupational Therapy's efficacy in children with Asperger's syndrome: a systematic review of randomized controlled trials

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Abstract

This systematic review of randomized controlled trials (RCTs) aims at evaluating the efficacy of Occupational therapy (OT) interventions in Asperger's syndrome (AS) pediatric patients. We conducted a systematic review according to PRISMA guidelines. The screening of the literature was carried out on PUBMED, SCOPUS, WEB OF SCIENCE and OT SEEKER databases, TO December 2018. We selected three RCTs having the common objective to evaluate whether children with AS can improve their social skills thanks to OT treatments. The interventions targeted by the three selected studies were: LEGO therapy, Social Use of Language Programme, Let's Face It! software and Parent Training combined with the administration of risperidone. All the studies showed that the intervention groups improved their overall social ability. This review shows that OT interventions can help concretely AS children in overcoming their social issues. Nevertheless, more RCTs are needed to better understand the OT's benefits and limitations in AS patients. Clin Ter 2019; 170(5):e382-387. doi: 10.7417/CT.2019.2164

Key words: Reahbilitation, Occupational therapy, Asperger's syndrome, Autism

Introduction

The Asperger's Syndrome (AS) is named after the Austrian physician Hans Asperger, who first identified and described the behavioral profile of a group of children having issues in social interaction and communication skills.

The fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM, 2013) has classified Asperger's Syndrome, Autistic Disorder, and Pervasive Developmental Disorder-Not Otherwise Specified within a single diagnostic category called Autism Spectrum Disorder (ASD). According to the DSM-V diagnostic criteria, we refer to ASD when there are 'persistent deficits in social communication and social interaction across multiple contexts' and 'restricted, repetitive patterns of behavior, interests, or activities', both present since childhood (DSM-V, 2013).

Some studies consider the prevalence of AS being higher than the autism (Kadesjo et al. 1999, Ehlers and Gillberg 1993, Schopler, Mesibov, Kunce 1998). In contrast, other trials on autism observed a ratio of the prevalence of autism on the AS being greater than the unit (Sponheim and Skjeldal 1998, Taylor et al. 1999, Baird et al. 2000, Powell et al. 2000, Chakrabarti and Fombonne 2001, 2005, Fombonne 2006, Ellefsen et al. 2007, Latif and Williams 2007). Fombonne et al. (2009) estimated the AS prevalence being 6 / 10,000, though recognising the strong limitations of the existing data. Worldwide one person out of 160 is affected by ASD; the median value of the ASD's prevalence is 6.2 / 1000 in Europe and 6.5 / 1000 in US (www.epicentro.iss.it).

Nowadays, the issues related to AS are neither treated by standardized interventions nor by a specific pharmacological therapy, although some medications can improve symptoms, such as anxiety, depression or hyperactivity (Malone et al. 2005). Timing and quality of tailored therapeutic strategies are essential for prevention and management of maladaptive behaviors and social issues. In this context, to improve the social skills and reduce invalidating behaviors, such as obsessions, anger, panic attacks and anxiety disorders, it is possible to intervene through cognitive and behavioral techniques. Some of these methods are described in case reports and case series available in literature (Rodger et al. 2007, 2008, Rodger and Brandenburg 2009, Sofronoff et al. 2005, 2007, McConachie et al 2013). One of the most popular cognitive methods is the Cognitive Orientation to daily Occupational Performance (CO-OP), which aims at improving the performance of daily activities with the help of effective cognitive strategies (SITE Company Italian Scientific Technical of Occupational Therapy, 2015). Rodger et al. reported two cases of pediatric patients with SA, successfully treated with CO-OP method (Rodger et al. 2007, 2008, Rodger and Brandenburg 2009). Among other available behavioral interventions, it is important to mention: the Applied Behavior Analysis (ABA), that aims at improving intellectual abilities, language and adaptive behaviors in children with ASD; and the Cognitive Behavioral Therapy (CBT), that is mainly used (Guidelines 2011) to treat the ASD with comorbid anxiety (Sofronoff et al. 2005,

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2007) and to improve affective communication (Andrews et al. 2013). Moreover, interventions mediated by parents having received a specific Parent Training (PT), can improve the quality of social interactions, in terms of happiness, stress reduction and better quality of communication style (McConachie et al 2013).

Although a number of occupational therapy (OT) interventions on AS children are described in the literature, to our knowledge there are no reviews evaluating the OT's effectiveness in AS patients. In addition to this gap in the literature, there is a gap in the guidelines, since the approach to the AS from the OT side is neither optimized nor standardized.

The aim of this systematic review is to evaluate the efficacy of Occupational therapy (OT) interventions in Asperger's syndrome (AS) pediatric patients.

Materials and methods

The screening of the literature was conducted to 18 December 2018, searching on PUBMED, SCOPUS, WEB OF SCIENCE and OTseeker databases, by using the keywords: "Asperger Syndrome AND occupational therapy", "Asperger Syndrome AND therapy", "Asperger Syndrome AND rehabilitation".

Selection of studies

Before starting the review duplicate papers were filtered out using Endnote. Following guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement two authors (RG, GG) first screened titles and abstracts using the following inclusion criteria: last 10 years as time of publication; between 0 and 18 years as age of patients; occupational therapy as therapeutic field; randomized controlled trial (RCT) as study-design.

Data collection and analysis and assessment of risk of bias

A common table has been developed to summarize the data extracted from the selected studies, providing the following information: (1) references (authors and year of publication); (2) participants' features (number, age, diagnosis); (3) duration of the study; (4) presence of the follow up; (5) interventions performed on cases and controls (type, duration and frequency); (6) scales for outcome evaluation; (7) results.

To assess the quality of the studies, the Jadad score was calculated for each trial (Clark et al. 1999).

Results

The search through the four databases produced 1792 (Table 1) citations in the first place; after having applied the filters concerning study design and time, 696 papers out of 718 were excluded, because not matching the requirements. The full texts of the 22 remaining studies were read to assess eligibility: 19 papers did not match the inclusion criteria of age, therapeutic field and study design (Fig. 1), therefore a

final amount of 3 studies (Scahill et al. 2012, Owens et al. 2008, Tanaka et al. 2010) were selected and included in the present review (Table 2).

Subjects

The sample size ranges from a minimum of 61 (Owens et al. 2008) to a maximum of 124 children (Scahill et al. 2012) with pervasive developmental disorders, including AS patients, from 4 to 12 years of age.

Treatments

The treatments were from 18 (Owens G. el at. 2008) to 36 hours (Schaill et al. 2012) long. The follow-up period was 6 months long in the studies by Owens et al. (2008) and Scahill et al. (2012), and 19 weeks long in the trial by Tanaka et al. (2010).

The therapeutic interventions targeted by the three selected studies are: 1) LEGO therapy (Owens et al. 2008), that is used for the acquisition of social and communication skills; 2) Social Use of Language Programme (SULP) (Owens et al. 2008), that is an educational programme promoting social communication and behaviors; 3) Let's Face It! (LFI!) software (Tanaka et al. 2010), that trains the user to process and recognize people's facial expressions; 4) intervention mediated by parents who received specific Parent Training (PT) (Scahill et al. 2012).

The control groups were treated with standard approach (Tanaka et al. 2010), pharmacological treatment (Scahill et al. 2012) or no treatment (Owens et al. 2008).

Evaluation of the outcomes

The rating scales used in the three studies to evaluate the outcomes, are the following:

- SI-GARS (Gilliam Autism Rating Scale Social Interaction Subscale): it is used to identify autism and assess symptoms' severity in individuals between 3 and 22 years of age; in this review it was used by Owens et al. (2008);
- VABS (Vineland Adaptive Behavior Scale): it is used to assess social skills in real life situations, and cognitive abilities in the management of everyday autonomy; in the present review it is used by Owens et al. (2008) and Scahill et al. (2012);
- ADOS (Autism Diagnostic Observation Schedule): it is used to assess social and communicative behaviors in ASD; in this review it is used by Tanaka et al. (2010);
- ADI (Autism Diagnostic Interview): it is a semi-structured interview administered to the parents of the patient, evaluating abnormalities and developmental delays in the areas of social interaction, language, communication and imagination; in the present review it is used by Tanaka et al. (2010);
- ABC (Aberrant Behavior Checklist): it is a diagnostic scale for ASD; in this review it is used by Scahill et al. (2012).

Table 1. Data extraction table (Owens et al. 2008, Tanaka et al. 2010, Scahill et al. 2012)

Reference Sample	IG treat- ment	CG treat- ment	Duration of treatment	Outcome measures	Results	Lost to follow-up	Ja- dad
			and lollow- up				aioos
61 children Age: 6-11yo Diagnosis: HFA, AS, ASC, A IG 1= 23 IG 2= 22 CG= 16	1G 1: LEGO 1G 2: SULP	ment ment	Treatment Th/week, for 18 weeks Follow-up 5,5-6 months	1) GARS-SI 2) VABS: Maladaptive behaviors Communication Socialization 3) Child enjoyment and parent satisfaction 4) Frequency of self-initiated social contact with peers and duration of social interactions with peers	ut no	7 in 1G1 7 in 1G2	-
117 children Age: 10-12 yo Diagnosis: AS, PDD-NOS, A IG= 65 CG= 52	=======================================	Standard treatment	100 min/ week, for a total of 20.2 h in 19 weeks Follow-up 19 weeks	LFII skills battery: Face subtests Object subtests	Significant results were obtained only in one of the Face subtests " <u>Parts/Wholes Identity test"</u> : IG showed a significant improvement of the scores comparing to the CG (p < 0,003); Particularly, the IG showed improvements in the analytic recognition of mouth features (p < 0,05) and holistic recognition of a face based on its eyes features (p<0,001).	23 in lG 15 in CG	-
124 children (M 105, F 19) Age: 4-10 yo Diagnosis: AS, PDD-NOS, A IG= 75 (AS n.4, PDD-NOS n.22, A n.49) CG= 49 (AS n.4, PDD-NOS n.13, A (32)	Risperido- ne + PT (COMB)	Risperido- ne (MED)	75-90 min/ PT session, for a total of 11,4 PT sessions Follow-up 6 months	1) HSQ 2) ABC-I 3) VABS: Daily Living Skills Socialization Communication Adaptive Behavior 4) Noncompliance Index (from the Vineland Daily Living Skills domain)	1) HSQ: Both groups showed improvement 2) ABC-I: Both group showed improvement 3) VABS: Both showed improvement on Vineland Standard and Age Equivalent scores across all domains; Both showed improvement on Vineland Standard scores for Socialization domain and Adaptive Composite were significantly greater for COMB versus MED. (p=0.01 e 0.05 respectively); On the mean Age Equivalent scores (also adjusted for IQ), the Socialization and Communication domains showed greater improvement in COMB versus MED (p=0.03 e 0.05 respectively). 4) Noncompliance Index: Significant reduction in the IG in comparison to the CG (p=0.03)	17 out of 124	0

Legend: A: autism/Autistic disorder; ABC-I: Aberrant Behavior Checklist Irritability Subscale; AS: Asperger syndrome; ASC: autism spectrum condition; CG: control group; COMB: combined therapy (PT + Risperidone); F: females; GARS-SI: Gilliam Autism Rating Scale Social Interaction Subscale; H: hours; HFA: high functioning autism; HSQ: Home Situations Questionnaire; IG: intervention group; PI: Parent Training; IQ: intelligence quotient; LFII: Let's Face It!; M: males; MED: medication alone (Risperidone); MIN: minutes; PDD-NOS: Pervasive Developmental Disorder-Not Otherwise Specified; PT: Parent Training; SD: Standard Deviation; SULP: Social Use of Language Programme; VABS: Vineland Adaptive Behavior Scales; VO: years old;

Tabella 2

Data base	Key word	Number of records
Pubmed	"Asperger Syndrome" AND "occupational therapy" "Asperger Syndrome" AND "therapy" "Asperger Syndrome" AND "Rehabilitation"	36 557 125
Scopus	"Asperger Syndrome" AND "occupational therapy" "Asperger Syndrome" AND "therapy" "Asperger Syndrome" AND "Rehabilitation"	57 661 60
Web of Science	"Asperger Syndrome" AND "occupational therapy" "Asperger Syndrome" AND "therapy" "Asperger Syndrome" AND "Rehabilitation"	13 231 244
OtSeeker	"Asperger Syndrome"	8

Risk of bias

We assessed the methodological quality of the selected studies by applying the Jadad score to each one of them. This score takes into account the description of the key aspects of a high quality trial: randomization, blindness and subjects lost to follow-up. None of the studies included in the present review achieved a score equal to or greater than 3 (on a scale from 0 to 5), that is the score of a good quality study. Two studies obtained a final score of 1 (Owens et al., 2008; Tanaka et al. 2010), while one obtained 0 (Scahill et al. 2012).

Discussion

To our knowledge, the present systematic review is the first in literature to examine, through an analysis of RCTs, the efficacy of the OT in pediatric patients with AS.

This review examines three different RCTs, conducted by three independent research groups (Owens et al. 2008, Tanaka et al. 2010, Scahill et al. 2012) having the common goal to evaluate the effectiveness of OT strategies to improve the social dimension of ASD children. The patients assigned after randomization to the intervention groups showed improvements in social behavior and skills after the treatment, in comparison to the control groups.

Owens et al. (2008) observed statistically significant improvements (p < 0.05) in social interaction in the group treated with LEGO therapy (23 patients), in comparison to SULP (22 patients) and control groups (16 patients). Nevertheless, these findings need to be confirmed by further studies on bigger samples.

Tanaka et al. (2010) have observed statistically significant improvements (p < 0.003) in parts/wholes face recognition in the intervention group (65 patients), treated with the software Let's Face It!, in comparison to the control group (52 patients). These findings indicate that even a relatively short-term intervention programme (19 weeks) can produce measurable effects on facial expressions recognition skills in ASD children.

Scahill et al. (2012) have observed significant improvements in socialisation (p=0,01) and overall adaptation (p=0.05) in the intervention group (75 patients), treated with risperidone and Parent Training (PT), over medication alone in the control group (49 patients).

Notwithstanding their low Jadad scores, these three RCTs clearly indicate that OT interventions can improve social skills and, as a result, the quality of life in AS children.

For this kind of patients, the possibility of being treated with the OT approach during childhood represents a better chance for a good social functioning during adulthood, avoiding difficulties in work activities and potential psychiatric comorbidities.

The occupational therapist is a key figure in the process of preventing adult social issues by successfully treating child social impairments, that can affect present and future quality of life. OT interventions are person-centered and tailored to the needs of the child affected by AS, thereby supporting his daily activities towards toward an increased independent functioning.

Limitations of the study

The present work is not free from limitations, mainly due to the gap in the literature of RCTs on OT in AS children. The three factors that reduced the number of the studies included in this review were mainly: 1) the study design, 2) the diagnosis, and 3) the OT's relevance.

As for the first limitation (RCT study design), while carrying out the literature review, we identified other studies than RCTs on OT in AS children, mainly case reports (Rodger et al. 2007, 2008, Rodger and Brandenburg 2009, Prečín et al. 2010, Gutman et al. 2010, Arikawa et al. 2013) and case series (Carter et al. 2004, Hutchins et al. 2015). Nevertheless, the number of the studies present in the literature is still low if compared to the many possibilities of treatment OT can offer to AS patients. As a result, more studies, especially RCTs, are needed to deliver scientific evidence on the efficacy of OT interventions in AS patients.

As far as the second limitation (diagnosis of AS), the initial aim of this study was to assess the effectiveness of the OT specifically in AS children, but the selected RCTs include not only individuals with AS but also with Autism and Pervasive Developmental Disorder-Not Otherwise Specified; thus the specificity of the diagnosis was not fulfilled by the present review. It is worth to mention that the number of studies on AS in the literature was indeed influenced by the DSM-V's classification changes, according to which AS was absorbed into the diagnosis of Autism Spectrum

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Disorder (ASD). It is therefore not a case that all the studies included in this review are anterior to 2013, hence before the release of DSM-V.

With reference to the third limitation (OT's relevance), the literature related to the OT interventions is still scant. More studies in the field are needed, in order to share scientific evidences within the OT community and set guidelines to optimize the therapeutic approach.

In addition to the gap identified in the literature, another limitation that affects the quality of this review are the low Jadad scores of the selected studies (Owens et al. 2008, Tanaka et al. 2010, Scahill et al. 2012).

These combined factors prevented the present work to reliably assess the efficacy of the OT in AS patients.

Conclusions

Despite the abovementioned limitations, the key message of the present review is twofold: 1) OT interventions demonstrated their potential in helping concretely AS children in overcoming their social issues; 2) a larger number of RCTs evaluating OT treatments in AS patients are needed to confirm OT's effectiveness in such a complex and delicate pediatric disorder.

The screening of the literature did not show the presence of systematic reviews of RCTs with the objective to evaluate the efficacy of the OT in AS pediatric patients. Thus, to our knowledge, this review is the first to systematically assess this and, as such, the results of this work need to be confirmed by further studies.

Competing interests

The authors declare that there are no competing interests regarding the publication of this paper.

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