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IS THE PROGRESS OF CHILDREN WITH ASD IN A BEHAVIOURAL THERAPY PROGRAMME INFLUENCED BY PARENTS' HYPER-SYSTEMIZING?

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Abstract

Background. In an attempt to find genetic explanations for the heterogeneous characteristics of autistic patients, research has shown that parents of autistic children are more likely than parents of neurotypical children to exhibit autistic-like characteristics, meeting the criteria for the so-called "broad autism phenotype" . Subclinical autistic traits have been identified in the families of children with ASD, in both fathers and mothers, but the way in which specific parental phenotypes influence the child's pathology remains unclear.

Methods. This study aimed to analyse the progress that a group of children with Autism Spectrum Disorders (ASD) have made during a behavioural therapy programme and the way their evolution has been influenced by the parents' systemizing level. 52 participants (aged 2 to 5 years old) diagnosed with an Autism Spectrum Disorder and both of their parents were included in the study. The severity of ASD symptomatology was assessed using the ADOS-G instrument, at the beginning of the study (T0) and after one year of

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behavioural therapy as well (T1), while the parents' systemizing level was evaluated using the self-report Systemizing Quotient (SQ).
Results. The ADOS-G scores showed a significant improvement after one year of therapy in both Communication and Social Interaction domains. Only the fathers` systemizing level (SQF) had a significant effect on the ADOS-G scores after one year of therapy, with greater improvements reported for children having fathers with higher SQF scores.
Conclusion. The significance of these findings is discussed in relation to the empathizing-systemizing (E-S) theory. We consider that it's particularly important to continue investigating the way that specific parents' traits, including their systemizing level or their possible broad autism phenotype, can influence the severity of their children's ASD or the outcome of the behavioural intervention.

Keywords: Autism Spectrum Disorder, childhood, parents' systemizing level, behavioural therapy.

Introduction

Autism Spectrum Disorders (ASD) are a group of clinically and etiologically heterogeneous developmental disorders. The classical triad of symptoms (social interaction and communication deficits as well as stereotypical behaviour and resistance to change) depict several levels of severity which result in different levels of disability (Dobrescu, 2016; Fuentes, 2014).

In an attempt to explain both the common characteristics of ASD as well as the individual characteristics of the spectrum, respectively the individual differences such as the remarkable results in certain fields, in 2003 Baron Cohen and collaborators proposed The theory of hyper-systemizing as an explanatory model for ASD (Cohen, 2003; Cohen, 2006). This theory provides an explanation for difficulties in adapting to change, explains why they have a preference for predictable systems driven by laws and why they become 'unable' when faced with complex systems such as social relationships or conversations. Although a high level of systemizing becomes a disability when it comes to social contexts, it is this capacity that enables ASD individuals to have remarkable results in systematized domains (Cohen, 2017).

According to the Theory of hyper-systemizing developed by Baron-Cohen in 2003 ASD is the result of high levels of systemizing. This theory explains the need for routines, their preference for predictable systems which vary very little or at all, as well as significant incapacity to less law-governed systems like social relationships along with the conversation and feelings involved (Cohen, 2003; Cohen, 2017).

The theory of hyper-systemizing introduced by Baron-Cohen in order to explain ASD, has the origin in the hypothesis according to which the brain has a systemizing mechanism (SM) which has different levels of functioning. It is thought that in individuals with ASD this level is set too high (Cohen, 2006; Cohen, 2017). The level to which the systemizing is set is genetically determined and conditioned by biological factors such as levels of foetal testosterone. Kickmeyer and collaborators (2005) have demonstrated that in utero exposure to foetal testosterone is associated with poor social interactions (boys get lower scores than girls on scales that measure social interaction). This theory could also explain why ASD is more common in males (Knickmeyer, 2005).

In some individuals the systemizing level is set too high, which makes them search for rules and structures in every system. In other people systemizing is set to a medium level and thus they occasionally present difficulties when it comes to systemizing, while people with a low or inexistent level of systemizing cannot function by rules or follow them (Cohen, 2006; Greenberg, 2018). According Baron-Cohen's theory, in the general population there are 8 levels of systemizing, most people having this level set at 2 (more frequently in females) or 3 (more frequently in males) (Cohen, 2006). Scientists, as well as parents of children with ASD (both mothers and fathers) have a systemizing level above average and have a borderline, subclinical phenotype (Cohen, 2006; Golden, 2013). A systemizing level above 5 is specific to individuals with ASD and a high level correlates with the severity of core symptoms (Cohen, 2006). Baron-Cohen provides proof according to which the level of systemizing is genetically determined and that the genes involved could be the same ones that are involved in the aetiology of ASD (Cohen, 1997; Cohen, 2006; Golden, 2013).

Autistic spectrum traits of subclinical intensity or the "borderline" phenotype for ASD have been identified in the families of children with ASD in both fathers and mothers by Constantino et al. (2005) and by Baron Cohen (2006) (Constantino, 2005; Cohen, 2006). According to Baron-Cohen's theory, Autism Spectrum Disorder may be the genetic result of two parents with a high systemizing level and a low empathy level (assorted marriage).

Objectives

The purpose of this paper is to analyse the progress of children diagnosed with ASD who are enrolled in a behavioural therapy programme in relation to the systemizing level of their parents.

The specific objectives of the paper are the following:

- Analysis of the progress accomplished by children diagnosed with ASD one year after the initiation of the behavioural therapy programme;

- Determining the parents' systemizing level and establishing its connection with the child's progress.

Methods

Participants

In order to achieve the aforementioned objectives, we designed an observational study on a group of 52 subjects and their parents, subjects diagnosed with Autism Spectrum Disorder, monitored in the Child and Adolescent Psychiatry Clinic of 'Prof. Dr. Alexandru Obregia' Clinical Psychiatry Hospital in Bucharest, Romania.

Each of the subjects included in the clinical group followed an applied behavioural therapy programme for 1 year, coordinated by a psychotherapist in a regime of 2 hours / day, 5 days / week. In order to achieve the standardisation of behavioural intervention amongst the subjects, the supervision of intervention programmes was directed by the same psychologist for all of the subjects, in the Child and Adolescent Clinic in the hospital.

Inclusion criteria:

- Age between 2 and 5 years old
- Clinical diagnosis of Autism Spectrum Disorder (Autistic Disorder/PDD-NOS) according to the DSM IV-TR diagnostic criteria.
- Availability, and financial and time resources to attend the applied behavioural therapy programme (2 hours/day, 5 days/week, for one year).

Exclusion criteria:

- Significant mental comorbidities (intellectual disability) or somatic comorbidities (neurological disorders- epilepsy, etc., genetic disorders, severe chronic somatic disorders)
- Psychotropic pharmacological treatment prior to the inclusion in the study or necessity to initiate treatment at any time during the study.

Initially, 63 families having a child diagnosed with ASD were willing to participate in the study, but 11 of the participants dropped out due to their lack of availability for taking part in the therapy program, at the established frequency. Out of the remaining 52 families, none withdrew by the end of the study. The study got approval from the Ethics and Research Committee in 'Prof. Dr. Alexandru Obregia' Clinical Psychiatry Hospital. Verbal consent from both parents of a child was obtained before the inclusion in the study, as well as written informed consent from at least one of the parents or the legal caregiver.

Instruments

For the evaluation of the children we used the ADOS-G test („Autism Diagnostic Observation Schedule – Generic", Lord C., Rutter M., DiLavore P.C., Risi, S. 1999) which represents a standardised protocol for observing the child suspected of Autism Spectrum Disorder. The test was applied by an examiner (medical doctor/psychologist) with special training in applying it. We used Module 1 for all the subjects, which is a module designed for children with no acquisition of expressive language. We present the cut-off values in Table 1. We reported and used in the analysis the following ADOS-G scores: ADOS-Communication score ("ADOS-C"), ADOS-Social Interaction score ("ADOS-SI") and ADOS-Communication+ Social Interaction score ("ADOS-C+SI).

Table 1. ADOS-G Module 1- Cut-off values

	Infantile autism	Atypical autism
Communication (C)	4	2
Social Interaction (SI)	7	4
Total C+SI	12	7

For the evaluation of the parents we used the SQ („Systemizing Quotient") (Baron-Cohen S. & Wheelwright S. 2003), an instrument developed by researchers at Autism Research Centre, Cambridge University, designed to measure the level of systemizing for adults. Depending on the total score, the systemizing level is categorised in one of 4 levels of severity: Low (L)/ Average (A)/ Above Average (AA)/ Very High (VH) (Table 2). The abbreviations "SQF" and "SQM" will be used for defining the SQ scores of the fathers (F) and of the mothers (M) and "SQF-L/ -A/ -AA / -VH" and "SQM- L/ -A/ -AA/ -VH" for defining the fathers' and mothers' systemizing severity level.

Table 2. SQ- levels of severity

Level of systemizing	SQ (systemizing)
Low (L)	0-19
Average (A)	20-39
Above average (AA)	40-50
Very high (VH)	51-80

Data collection involved a longitudinal direction by evaluating the main symptomatic areas of ASD in children included in the clinical group, both at the time of inclusion in the study (T0) and after one year of behavioural therapy (T1). The instruments used for the evaluation of the parents were administered exclusively at inclusion (T0).

Data analysis

To analyse the evolution of ASD symptomatology after 1 year of therapy, t tests for paired-samples were carried out using the ADOS-G scores. The procedure used to determine the effect of parents' systemizing level on the evolution of the subjects after 1 year of therapy involved running two-way mixed ANOVA tests, with one between-subject factor (group based on the severity of the SQ scores), one within-subjects factor (time defined by T0 and T1 moments) and the ADOS-G scores as the dependent variable. The homogeneity of variances was assessed using the Levene's test of homogeneity of variance and homogeneity of covariances using the Box's test of equality of covariance matrices. The two-way interaction (SQ severity group * time) on the dependent variable are reported using the F test result, the statistical significance and the effect size (partial η2). If the interaction is significant, simple main effects for each of the factors (SQ severity group and time) are reported as well as the results of multiple comparisons using Tukey HSD test if simple main effects are present.

Results

a) Progress of children diagnosed with Autism Spectrum Disorder

There were statistically significant differences between ADOS-G scores at T0 and T1 as shown by the results of t tests for paired-samples. The scores were lower both in the total domain of Communication + Social Interaction (M ADOS-C+SI (T0) = 12.33± 3.62; M ADOS-C+SI (T1)= 9.88± 3.83; t(51)= 6.44, p= .00) as well as separately in each of the two domains (M ADOS-C (T0)= 4.37± 1.63; M ADOS-C (T1)= 3.40± 2.67; t(51)= 5.22, p= .00 and M ADOS-SI (T0)= 7.98± 2.18; M ADOS-SI (T1)= 6.48± 2.46; t(51)= 5.74, p= .00) (Table 3).

Table 3. Differences between ADOS-G scores at T0 and T1- t test for paired samples

	Paired Differences							
				95% CI diff				
	Mean	Std. Dev	Std.er Mean	Lower	Upper	t	df	Sig 2 tailed
ADOS-C+SI(T0) – ADOS C+SI(T1)	2.442	2.733	.379	1.682	3.203	6.445	51	.000
ADOS-C(T0) - ADOS-C(T1)	.962	1.328	.184	.592	1.331	5.222	51	.000
AD0S-SI(T0) - ADOS-SI(T1)	1.500	1.884	.261	.976	2.024	5.742	51	.000

b) The effect of parents' systemizing level (SQF/SQM) on the evolution of ADOS-G scores

The mean SQF score was 35.27± 12.98 95% CI [31.65, 38.88] and the mean SQM was 26.69± 11.48 95% CI [23.49, 29.89] (Figure 1). Most of the subjects, both males (F) and Females (M) had an SQ score falling in the Average severity group (20-39 points).

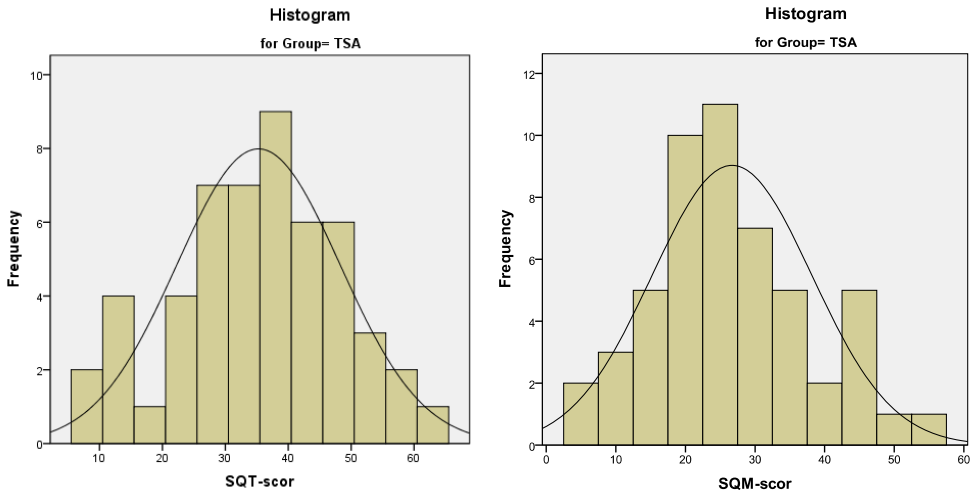

Figure 1. The distribution of SQF and SQM

The interaction between the fathers' level of systemizing and time (SQF group * time) on the ADOS-G scores was significant (F(3,48)= 3.504, p= .022, partial η^2= .180) (Table 4A). The dependent variable was defined as the ADOS-G total score (Communication + Social interaction), time as the within-subjects factor with two levels (T0 and T1) and SQF as the between-subjects factor with four levels (L, A, AA, VH). There was homogeneity of variances, as assessed by Levene's test of homogeneity of variance (p=.208), as well as homogeneity of covariances, as assessed by Box's test of equality of covariance matrices (p=.218).

The simple main effect of the SQF group on ADOS-G scores at each of the two moments of time was studied (Table 4B). The univariate analysis showed no significant difference between the 4 groups based on SQT severity level neither at T0 (F (3,48) = 2.74, p=. 053, partial η^2 = .146), nor at T1 (F (3,48) = 2.69, p=. 057, partial η^2= .144).

The simple main effect of time on the ADOS-G score analysed separately in the 4 categories showed statistically significant differences from T0 to T1 in SQF-A, AA, VH and no difference in the SQF-L category level, with consistent increase of the difference between T0 and T1 along with the increase of the systemizing level in parents (Table 4C).

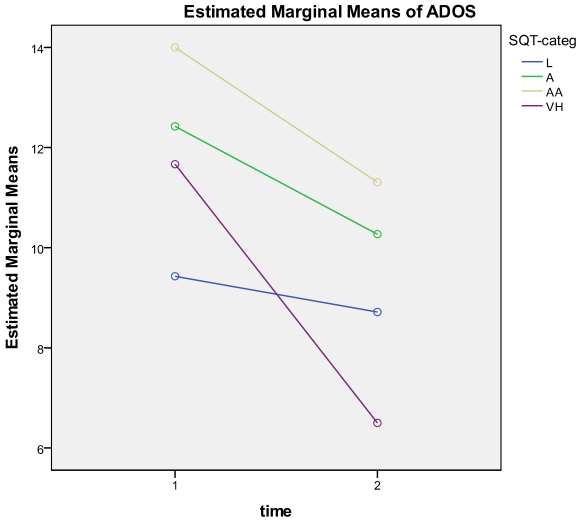

Figure 2. Progression of ADOS-G scores from T0 to T1 based on SQF severity group

Table 4. The effect of SQF severity category on the progression of ADOS-G scores
A. Interaction between the factors (two-way mixed ANOVA)

ADOS-G		Type III Sum of Squares	df	Mean Square	F	Sig.	Partial Eta Squared
time * SQF	Sphericity Assumed	34.206	3	11.402	3.504	.022	.180
	Greenhouse-Geisser	34.206	3.000	11.402	3.504	.022	.180
	Huynh-Feldt	34.206	3.000	11.402	3.504	.022	.180
	Lower-bound	34.206	3.000	11.402	3.504	.022	.180
Error(time)	Sphericity Assumed	156.208	48	3.254			
	Greenhouse-Geisser	156.208	48.000	3.254			
	Huynh-Feldt	156.208	48.000	3.254			
	Lower-bound	156.208	48.000	3.254			

Table 4. The effect of SQF severity group on the progression of ADOS-G scores
B. Mean ADOS-G scores at T0 and T1 for each SQF severity group

	SQF group	Mean	Std. Deviation	N
ADOS-C+SI (T0)	L	9.43	3.409	7
	A	12.42	3.501	26
	AA	14.00	3.937	13
	VH	11.67	1.366	6
	Total	12.33	3.623	52
ADOS-C+SI (T1)	L	8.71	1.799	7
	A	10.27	4.104	26
	AA	11.31	3.945	13
	VH	6.50	1.871	6
	Total	9.88	3.843	52

Table 4. The effect of SQF severity group on the progression of ADOS-G scores
C. Differences of ADOS-G scores at T0 and T1, separately
in each of the 4 SQF severity groups (t test for paired samples)

Differences ADOS-C+SI (T0) – ADOS-C+SI (T1) – Paired samples								
				95% CI Diff				
	Mean	Std. Deviation	Std. Error Mean	Lower	Upper	t	df	Sig. (2-tailed)
SQF-L	.714	3.302	1.248	-2.340	3.768	.572	6	.588
SQF-A	2.154	2.525	.495	1.134	3.174	4.350	25	.000
SQF-AA	2.692	2.359	.654	1.267	4.118	4.115	12	.001
SQF-VH	5.167	2.041	.833	3.025	7.309	6.200	5	.002

There was no interaction effect between the mothers' systemizing levels and time (SQM group * time) on the ADOS-G scores (F (2,48)= .479, p= .622, partial η^2= .020). The SQM-VH group was dropped from the analysis as it contains only one participant. The main effect of the SQM group on ADOS scores at each of the two moments was not significant (at T0 F(2,48)= .640, p=. 532, partial η^2= .026; at T1 F(2,48)= .948, p=. 395, partial η^2= .038).

Discussion

Behavioural therapy significantly improves ASD-specific symptoms amongst children with ASD

The statistically significant differences of ADOS scores in T0 and T1 indicate the positive progress of ASD symptoms after 1 year of therapy. The differences emerge both in the total domain ADOS-C+SI as well as independently in each of the domains. The mean difference between the two moments is 2.44±2.72.

In 2014, Weitlauf and colleagues published a report on the effectiveness of behavioural therapy in ASD (Weitlauf, 2014). They concluded that scientific literature studies bring clear evidence on the improvement of cognitive and language skills after intensive behavioural therapy, but the results of the studies are not as consistent in terms of social interaction and adaptation. The different outcome in individuals depends on the individual particularities of each child, as well as on the intervention technique (type, frequency, duration), although it is still unclear how these factors modulate the progress (Turner, 2006).

The level of systemizing of the fathers has a direct effect on the evolution ASD severity under therapy

In the analysis of the effect had by the parents' phenotype on the symptoms of ASD, the systemizing level (low, average, above average or very high) was taken into account. The linear model we used showed us that the father systematization coefficient (SQF) exerts a direct effect on evolution of the children's ASD severity

in therapy, with a more pronounced trend of decreasing ADOS-G scores after 1 year of therapy among children whose fathers have a higher level of systemizing. One possible explanation for the meaning of this interaction, from a clinical perspective, could be that a hyper-systemizing parent will apply the rules of behavioural therapy in a very rigorous manner, extremely precise and will follow the programme and instructions provided by the therapist, thus offering a consistency of the therapeutic programme, unlike a parent with a chaotic parenting style.

Our results do not necessarily overlap with the general knowledge existing in the scientific literature regarding the influence of autistic traits in parents, defined by us from the perspective of Baron-Cohen's theory in terms of hyper-systemizing, on the progress of children with ASD. The presence of hyper systematization in parents should be, at least theoretically, a factor that increases the severity of ASD symptoms in children (measured by the severity of ADOS score) and negatively affect the child's evolution.

The existence of BAP (Broad Autism Phenotype) in parents has not been correlated with the child's intellectual level, regression in acquisitions or sex, as noted by Gerdts and Bernier in a literature review from 2011 (Gerdts and Bernier, 2011). Families in which both parents experience social reciprocity difficulties, even at a subclinical level, are at risk to have a child with ASD. It seems that low social responsivity in fathers could be a reliant predictor for low social responsivity in descendants. Gerdts and Bernier conclude that the severity of autism spectrum characteristics correlates with BAP particularities in fathers (Gerdts and Bernier, 2011). From our point of view identifying BAP in parents can influence the objectives of the therapy, the motivation of the parent to participate in the behavioural process and openness to change, as well as the ability of the parent to implement the therapeutic techniques. Regarding the objectives pursued in the therapy, they can be greatly influenced by the parent's perception. Thus, for a parent who exhibits autistic traits and who himself is less interested in social relationships, the goals of therapy will be rather aimed at language skills improvement or decreasing stereotypical behaviours and less at developing the child's ability to interact socially. On the other hand, a parent without BAP characteristics, with a typical social interaction pattern will aim to improve social relationships. At the same time, as noted before, a hyper-systemizing parent will not be successful in stimulating improvement of social skills considering that social interaction is not something of interest for the parent either.

In 2014, Vivanti considered that at the time there was not enough evidence in the scientific literature to conclude how the progress of the child wth ASD is influenced by the parents. Parental motivation, difficulties and strengths (communication skills, flexibility) are factors that could influence the child's development (Vivanti, 2014). Most of the researchers agree that more studies are needed in order to identify the predictive factors of ASD progression, as it is still unclear why in some children the symptoms change over time.

It is certain that the results obtained by us must be seen from the perspective of the limitations of this study and mainly from the perspective of the small number of subjects included. Other important limitations could result from the exclusion of patients with severe neurological / somatic conditions as well as those who required psychotropic medication at the time of study inclusion or at any time during the study. Thus, it is the possible to have included less severe cases whose evolution in therapy has been favourable also due to a lower severity at the time of the enrolment.

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