



Original Article

Socio-Behavioral Development in Siblings of Autistic Children; A Comparative Case-Control Study

Elham Niazi¹, Soroor Arman^{1*}¹ Department of Psychiatry, Isfahan University of Medical Sciences, Isfahan, Iran

*Corresponding author: Soroor Arman

Email: soroorarman@yahoo.com

ABSTRACT

Background: Autism spectrum disorder (ASD) is a psychiatric disorder with a spectrum of symptoms, including impaired social interactions, impaired verbal and non-verbal communications, and limited and repetitive patterns of behaviors. The incidence of social and emotional disorders in siblings of an autistic child and relationship with having an autistic sibling is a matter of debate. The current study is aimed to assess the socio-behavioral development in the sibling of an autistic child (Sib-A).

Methods: The current case-control study has been conducted on 51 Sib-A and 138 children without any autistic sibling (Sib-H). In order to assess social development, the Social Responsiveness Scale (SRS) that consists of 5 subscales of social awareness, social cognition, social communication, social motivation, and restricted interests and behavior was used. The behavioral status was assessed using the Strength and difficulties questionnaire (SDQ) in 5 subscales of emotional symptoms, conduct problems, hyperactivity, peer communications problems, and prosocial behaviors. Eventually, the findings of the two groups were compared.

Results: The mean score of SRS in cases was 43.35 ± 36.84 and in controls 29.69 ± 11.20 (P-value = 0.012). Besides, the two groups were different in all subscales (P-value < 0.05) except social motivation (P-value = 0.1). The comparison of SDQ revealed a significant difference between the cases with a mean score of 5.93 ± 5.53 and controls with 3.26 ± 2.57 (P-value = 0.011). Besides, the two groups were different in all subscales (P-value < 0.05) except for conduct problems (P-value = 0.1).

Conclusion: The siblings of autistic children showed more socio-behavioral problems than the controls. The genetically or nurturing of these problems should be investigated.

Keywords: Autism; Autism Spectrum Disorder; Siblings

Citation: Niazi E, Arman S. Socio-Behavioral Development in Siblings of Autistic Children; A Comparative Case-Control Study. Caspian J Health Res. 2020;5(1):??-??.

ARTICLE INFO

Received: August 25, 2019

Accepted: January 26, 2020

ePublished: March 10, 2020

Introduction

Autism Spectrum Disorder (ASD) is a psychiatric disorder with ranges of symptoms, including impaired social interactions, failure in verbal and non-verbal communications, and limited and repetitive patterns of behaviors. This disorder consists of Asperger syndrome, childhood disintegrative disorder, and pervasive developmental disorder(1, 2). ASD usually develops within 12-24 months of age and commonly has a progressive course. Up to 30% of ASD cases represent a regressive pattern; therefore, a child with the ability to communicate

verbally, develops disability to speak and becomes isolated (3-5). The diagnosis of ASD has dramatically increased in recent years, and even the prevalence of 10-17% has been reported in the literature (1).

The etiology of this disorder remains unknown, but the genetic footprint strongly exemplifies in ASD (6). Thus exposure to communication and social disorders in other family members is not a weird phenomenon. Moreover, the presence of an autistic child in a family deviated the parents' attention toward the sick kid, a fact that reinforces the communication- and social-related disorders among the siblings (7).

Considering the distinctive features of autism, all of the family members, including parents, siblings, and second-degree families, may be affected. The family system theory represents that the relations in a family unit consist of four subunits of marital, parental, sibling, and extended family relations. Each of these subunits affects other family members, family function, and familial interactions. Therefore, psychiatrists should assess the influences of ASD on all of these subunits, such as siblings, as it can incline the supports toward the ASD child and negatively affect the familial interactions. Most of the studies in the literature have assessed the influence of ASD on parents, and limited ones have evaluated the effect of ASD child presence on siblings (8). Numerous scientists have insisted on the critical role of siblings in the developmental pattern of a healthy child. The continuous interaction of siblings with each other, not only improves the conception of a sibling but also affects the social abilities, cognition, and conduct (9). Therefore, by consideration of the siblings' interactions, scientists have raised studies to assess the psychological adjustment ability in siblings of a disabled child. It seems that siblings of disease children are at increased risk for developing adjustment disorders as compared to the siblings of healthy children; however, reports have demonstrated controversial results. Some of the studies have confirmed this theory, whereas the others represented no significant differences between these two groups, and studies are showing the superiority of children with a diseased sibling to those with healthy ones regarding adjustment ability (10).

Based on the Powell and colleagues' study, the effect of a diseased child on the siblings can be represented as an axis with two extreme spectra, on a hand very adverse outcomes and, on the other hand, very positive ones (11). Furthermore, investigations showed that the ability of adjustment is not a one-dimensional structure, as a child may present diverse reactions with wide ranges of functions at different times (12). These outcomes may occur due to the complexity of behaviors and unpredictable and indeterminate symptoms expressed by an autistic child that, in turn, has led to different adjustment advantages and disadvantages among siblings of autistic children as compared to siblings of cases with other disorders. The evaluation of behavioral disorders, social competence, and self-esteem among the siblings of autistic children is a critical issue as these areas have important implications for the overall psychological well-being of children (13, 14). Therefore, in the current case-control report, it is aimed to assess and compare the behavioral and social disorders among siblings of children with and without ASD.

Methods

Study population

The current case-control study was aimed to assess the socio-behavioral status among the siblings of autistic children, referring to the Autism Center of Isfahan from April 2017 to June 2019. A total of 51 children (brothers or sisters) from the families with at least one autistic child (Sib-A) were included as case groups, and 138 children without autistic siblings (Sib-H) were included as the control group.

The Ethical Committee of Isfahan University of Medical

Sciences approved the study protocol. Then, the protocol was explained for the legal guardians of the children, and they were reassured about the confidentiality of their personal information. Eventually, written consent was obtained from the legal guardians.

The 6-12 years old children with at least one autistic sibling were included in the study if they had both biologic parents, and they had no history of chronic physical disease. Besides, in order to match the cases and controls, the participants of control groups, who did not have an autistic sibling, were recruited from the classmates of the case group.

The parents' reluctance for the participation of their child in the study, adoption or being a stepbrother/ stepsister, impairment in the checklist completion, chronic physical diseases, and not living with biological parents were considered as the exclusion criteria. Those subjects with more than 20% deficit in the questionnaires' responses were excluded from the study.

The diagnosis of autism was made based on the Diagnostic and Statistical Manual of Mental Disorders, fifth edition (DSM-5) (15). The case and controls were included in the study through non-randomized and convenience sampling until achieving the desired numbers of participants.

Data collection tools

Social Responsiveness Scale (SRS): The social symptoms was measured using Social responsiveness scale (SRS), a questionnaire consisted of 65 questions assessing the parents' observations about the child's behaviors. This questionnaire evaluated five subscales, including social awareness, social cognition, social communication, social motivation, and restricted interests and behaviors. The response to each item was recorded using Likert type scale form 1 (never) to 4 (almost always). The higher scores show the worse conditions.

The reliability and validity of this questionnaire were measured in the range of 75-91% for all of the subscales regardless of the child's intelligence quotient. This scale assesses the attitude toward typical social content and can quantitatively demonstrate impulsivity severity and autistic symptoms; therefore, it can efficiently differentiate ASD from other psychiatric disorders (16). The validated Persian version of this questionnaire has been published by Tehrani-Doost et al. with acceptable reliability and validity in 2018 (17).

Strength and Difficulties Questionnaire (SDQ): In order to assess the behavioral symptoms of the participants, the Strength and difficulties questionnaire (SDQ) was utilized. This instrument consists of 25 questions about the parent observations in 5 entities, including emotional symptoms, conduct problems, hyperactivity, peer communications problems, and prosocial behaviors. Five questions are allocated to each of the subscales, with three alternatives designed in the Likert method. The higher scores demonstrate the worse behavioral disorder condition. This questionnaire has been validated with Cronbach's alpha of 0.73 (18). The validated Persian version of the questionnaire was raised in 2009 with the Cronbach's alpha range of 0.69-0.79 for each of the subscales (19).

The cases and control demographic data, including age, gender, and familial relation with the autistic person (sister/ brother) was entered in the study checklist. Both

questionnaires of SRS and SDQ were filled out by the parents of cases and control group.

Statistical analysis

Data were analyzed using SPSS version 23. The descriptive data were presented in mean, standard deviation, percentages, and absolute numbers. Normal distribution was assessed using graphical method and Kolmogorov-Smirnov test. Independent T-test or Mann-Whitney u test was used to compare the two groups, as appropriate. P-value of less than 0.05 was considered as a significant level.

Results

In this study, 51 cases and 138 controls were evaluated. The mean age of cases was 9.61 ± 2.28 years (range: 6-15 years) and in controls was 8.31 ± 1.10 years (range 6-10 years). Twenty-eight children (54.9%) in case group and 65 children (47.1%) in control group were male. There was not meaningful difference between the two groups regarding demographic features (P-value > 0.05). Figure 1 and 2 shows histogram of SRS and SDQ data, respectively. The data did not follow normal distribution (P-value < 0.001).

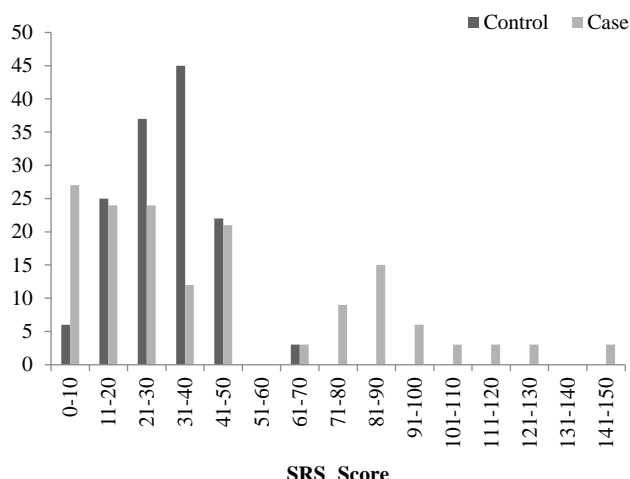


Figure 1. Histogram of Social Responsiveness Scale in Cases and Controls (P-value < 0.001)

The mean score of SRS in the case group was 43.35 ± 36.84 , that was significantly higher than the control (29.69 ± 11.20 , P-value = 0.02). The two groups were remarkably different

in terms of all subscales except for social motivation (P-value = 0.163) (Table 1).

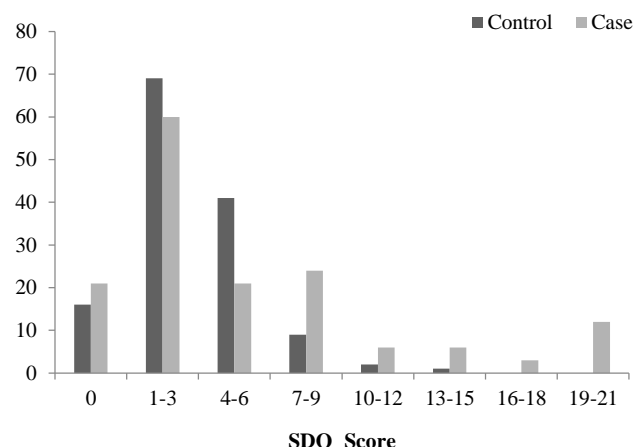


Figure 2. Histogram of Strengths and Difficulties Questionnaire in Cases and Controls (P-value < 0.001)

The comparison of the mean score of SRS in case group stratified by sex revealed an insignificant difference (47.68 ± 35.10 (median: 31) for males, 38.98 ± 38.09 (median: 39.5) for females; P-value = 0.29). A similar pattern was found among controls, as well (31.06 ± 11.29 (median: 30) for males, 28.47 ± 11.05 (median: 32) for females; P-value = 0.139) (Table 2).

The comparison of SDQ showed a significant difference between the two groups with a mean score of 5.93 ± 5.53 in cases versus 3.26 ± 2.57 in controls (P-value = 0.02). Besides, the detailed assessments of subscales revealed statistically significant differences between the two groups regarding emotional symptoms (P-value = 0.01), peer communications problems (P-value = 0.001) and prosocial behaviors (P-value = 0.04), while two groups were similar in terms of conduct problems (P-value = 0.74) and hyperactivity (P-value = 0.81) (Table 3).

The gender-based assessments of SDQ revealed insignificant differences in both cases and controls as follows; the mean score of 5.9 ± 5.4 (median: 5) was found in females versus 5.1 ± 6.6 (median: 4) in males of the cases group (P-value = 0.21), and the mean score of 3.57 ± 2.6 (median: 3) in females versus 2.99 ± 2.5 (median: 3) in males of the control group (P-value = 0.17).

Table 1. The Comparison of the Social Responsiveness Scale between Cases and Controls

Variable		Median	Mean	Standard deviation	P-value*
Social awareness	Case	9	8.82	6.75	0.034
	Control	6	6.33	3.37	
Social cognition	Case	8	9.76	7.86	0.026
	Control	7	7.17	3.70	
Social communications	Case	12	13.14	12.36	0.04
	Control	8	8.32	4.67	
Social motivations	Case	3	3.35	2.44	0.163
	Control	3	2.74	1.87	
Restricted interests and behavior	Case	6	7.22	9.23	0.036
	Control	4	4.33	2.38	
Total	Case	39	43.35	36.84	0.02
	Control	31	29.69	11.20	

*P-value is based on Mann Whitney U test.

Table 2. The Comparison of Strength and Difficulties Questionnaire between Cases and Controls

Variable		Median	Mean	Standard deviation	P-value*
Peer communications problems	Case	1	1.25	1.28	0.001
	Control	0	0.56	0.71	
Prosocial behaviors	Case	1	1.98	2.69	0.04
	Control	0	0.92	1.20	
Emotional symptoms	Case	1	0.67	1.03	0.01
	Control	0	0.33	0.58	
Conduct problems	Case	0	0.63	1.04	0.74
	Control	0	0.51	0.64	
Hyperactivity	Case	1	1	1.25	0.81
	Control	1	0.96	1.06	
Total	Case	4	5.93	5.53	0.02
	Control	3	3.26	2.57	

*P-value is based on Mann Whitney U test.

Discussion

Based on the findings of current study, Sib-A, in comparison to Sib-H, was involved with more socio-behavioral disorders remarkably. Their social disorders were independent of gender and were found in entities, including social awareness, cognition, social communication, and restricted interests and behavior, while only social motivation was not different between the two groups. The evaluation of behavioral development based on SDQ showed significant differences in all aspects except for conduct problems.

There are other investigations with similar principles, among which some confirmed our findings and the others opposed. Hastings et al. assessed the effect of behavioral supports on the behavioral adjustment among siblings of children with autism. Similar to our result, they represented impaired behavioral development among the Sib-A, which was directly correlated with the severity of autism. They revealed the role of family support as a modulator of behavioral development, not a director or a compensator (20). Meyer et al. They found that the socio-behavioral disorders in Sib-A were correlated with the severity of autism in the affected child, the mother's depression, and even the depression severity (21).

In Constantino et al. compared the social development of siblings in families with more than one autistic child with two other groups, including siblings of children with pervasive development disorder and siblings of children with other psychiatric disorders other than ASD. The worst social developmental status was found among siblings in families with more than one ASD child, then siblings of children with pervasive development disorder. These findings strongly support the theory of the genetic basis of ASD (13).

There are also some contradictory findings. Walton et al. compared the behavioral development of Sib-A with Sib-H using SDQ as well as maternal mood status. They showed that Sib-A had a significantly better capability of representing appropriate behaviors when exposed to stressors and are less violent, less involved, and more avoidance toward their autistic sibling (22).

In a case-control study by Quintero et al., the academic, behavioral, and social adjustment of 6-10-year-old Sib-A was compared with Sib-H. These assessments were performed on siblings older than the autistic child. Outcomes of this study showed insignificant difference

between the two groups. Nevertheless, the depression and anxiety status of mothers that was more severe among those with an autistic child as compared to controls remarkably influenced the trend of socio-behavioral development in siblings (23). These findings may have achieved partially due to the age of healthy cases, as the assessed siblings of this study were older than the autistic child; therefore they were born before the exposure of family to the autism-related stress and depression, particularly as the maternal mood status affected the socio-behavioral development of the children.

In Verte et al. study, they found better socio-behavioral development among Sib-A as compared to Sib-H. They hypothesized that the siblings who experience living with an autistic child had better competence, more self-esteem, and more appropriate empathy that all together led to better social behaviors expression. (10).

The socio-behavioral problems in siblings of autistic cases seem to be more prominent in childhood, while eventually, with the reduction in the family role during adolescence and the ability to better controlling of the behaviors and better performance of the social activities, the siblings would present less socio-behavioral disorders.

In terms of comparing these two groups, precise considerations should be made because not only the selection of cases and controls may be affected by selection bias, the mothers' responses may be affected by the comparison of a healthy child with the ASD one, as well.

Another finding of this study was the lack of relationship between social and behavioral disorders and gender that was confirmed in the study of Pourbagheri et al. (14); however, Hesse and colleague presented more severe social disorders in girl Sib-A than boys and argued that the social developmental pattern was affected by maternal anxiety toward her autistic child (24).

Conclusion

This study showed that the socio-behavioral problems in siblings of autistic children were significantly more than the control group regardless of their gender. These disorders may be related to genetic factors or might be acquired during life that need to be investigated in further studies.

Acknowledgements

We are grateful to Psychiatry officials of Autism Center of Isfahan.

Ethical consideration

This study has been approved with Ethical Committee of Isfahan University of Medical Sciences, Isfahan, Iran.

Conflicts of interests

Authors declared no conflict of interest.

Funding

The study was sponsored by Isfahan University of Medical Sciences.

References

1. Meadan H, Stoner JB, Angell ME. Review of literature related to the social, emotional, and behavioral adjustment of siblings of individuals with an autism spectrum disorder. *J Dev Phys Disabil*. 2010;22(1):83-100. doi: 10.1007/s10882-009-9171-7.
2. Raza S, Sacrey LAR, Zwaigenbaum L, Bryson S, Brian J, Smith IM, et al. Relationship between early social-emotional behavior and autism spectrum disorder: a high-risk sibling study. *J Autism Dev Disord*. 2020;50(7):2527-2539. doi: 10.1007/s10803-019-03977-3.
3. Sadock BJ, Sadock VA, Ruiz P. Kaplan & Sadock's comprehensive textbook of psychiatry. 10th ed. Philadelphia, PA: Lippincott Williams & Wilkins (LWW); 2017.
4. Martin A, Volkmar FR. Lewis's child and adolescent psychiatry: a comprehensive textbook. 5th ed: Philadelphia, PA: Lippincott Williams & Wilkins (LWW); 2018.
5. Sadock BJ, Sadock VA, Ruiz P. Kaplan and Sadock's Synopsis of psychiatry. 11th ed: Lippincott Williams & Wilkins (LWW); 2014.
6. Tordjman S, Cohen D, Coulon N, Anderson GM, Botbol M, Canitano R, et al. Reframing autism as a behavioral syndrome and not a specific mental disorder: Implications of genetic and phenotypic heterogeneity. *Neurosci Biobehav Rev*. 2017;80:210. doi: 10.1016/j.neubiorev.2017.01.030.
7. Toth K, Dawson G, Meltzoff AN, Greenson J, Fein D. Early social, imitation, play, and language abilities of young non-autistic siblings of children with autism. *J Autism Dev Disord*. 2007;37(1):145-157. doi: 10.1007/s10803-006-0336-2.
8. Aparicio RR, Mínguez RT. Behavioral, emotional, and social adjustment in siblings of children with autism spectrum disorder. a theoretical review. *Papeles del Psicólogo*. 2015;36(3):189-197.
9. McHale SM, Updegraff KA, Feinberg ME. Siblings of youth with autism spectrum disorders: Theoretical perspectives on sibling relationships and individual adjustment. *J Autism Dev Disord*. 2016;46(2):589-602. doi: 10.1007/s10803-015-2611-6.
10. Verté S, Roeyers H, Buysse A. Behavioural problems, social competence, and self-concept in siblings of children with autism. *Child Care Health Dev*. 2003;29(3):193-205. doi: 10.1046/j.1365-2214.2003.00331.x.
11. Gallagher PA, Powell TH, Rhodes CA. Brothers & sisters--a special part of exceptional families. Baltimore, Md: Paul H. Brookes Pub. Co; 2006.
12. Goodman R, Meltzer H, Bailey V. The strengths and difficulties questionnaire: a pilot study on the validity of the self-report version. *European Child & Adolescent Psychiatry*. 1998;7:125-130.
13. Constantino JN, Lajonchere C, Lutz M, Gray T, Abbacchi A, McKenna K, et al. Autistic social impairment in the siblings of children with pervasive developmental disorders. *Am J Psychiatry*. 2006;163(2):294-296. doi: 10.1176/appi.ajp.163.2.294.
14. Pourbagheri N, Mirzakhani N, Akbarzadehbaghban AR. A comparison of emotional-behavioral problems of siblings at the age range of 3-9-year-old children with autism and down syndrome. *Iran J Child Neurol*. 2018;12(2):73-82. doi:10.22037/ijcn.v12i2.15849.
15. Grzadzinski R, Huerta M, Lord C. DSM-5, and autism spectrum disorders (ASDs): an opportunity for identifying ASD subtypes. *Mol Autism*. 2013;4(1):12. doi: 10.1186/2040-2392-4-12.
16. Bölte S, Poustka F, Constantino JN. Assessing autistic traits: cross-cultural validation of the social responsiveness scale (SRS). *Autism Res*. 2008;1(6):354-363. doi: 10.1002/aur.49.
17. Tehrani-Doost M, Shahrivar Z, Torabi N, Ansari S, Haji-Esmaelzadeh M, Saeed-Ahmadi S. Cross-cultural validation and normative data of the social responsiveness scale in a group of Iranian general child population. *J Autism Dev Disord*. 2018;50(7):2389-2396. doi: 10.1007/s10803-018-3773-9.
18. Goodman R. Psychometric properties of the strengths and difficulties questionnaire. *J Am Acad Child Adolesc Psychiatry*. 2001 Nov;40(11):1337-1345. doi: 10.1097/00004583-200111000-00015.
19. Alavi A, Mohammadi M, Mahmoudi J, Tehranidoost M, Shahrivar Z, Saadat S. The Farsi Version of the strength and difficulties questionnaire self report form: the normative data and scale properties. *Iran J Child Neurol*. 2009;3(1):27-34. doi: 10.22037/ijcn.v3i1.990.
20. Hastings RP. Behavioral adjustment of siblings of children with autism engaged in applied behavior analysis early intervention programs: The moderating role of social support. *J Autism Dev Disord*. 2003;33(2):141-150. doi: 10.1023/a:1022983209004.
21. Meyer KA, Ingersoll B, Hambrick DZ. Factors influencing adjustment in siblings of children with autism spectrum disorders. *Res Autism Spectr Disord*. 2011;5(4):1413-1420. doi: 10.1016/j.rasd.2011.01.027.
22. Walton KM, Ingersoll BR. Psychosocial adjustment and sibling relationships in siblings of children with autism spectrum disorder: Risk and protective factors. *J Autism Dev Disord*. 2015;45(9):2764-2778. doi: 10.1007/s10803-015-2440-7.
23. Quintero N, McIntyre LL. Sibling adjustment and maternal well-being: An examination of families with and without a child with an autism spectrum disorder. *Focus Autism Other Dev Disabil*. 2010;25(1):37-46. doi: 10.1177/1088357609350367.
24. Hesse TL, Danko CM, Budd KS. Siblings of children with autism: Predictors of adjustment. *Res Autism Spectr Disord*. 2013;7(11):1323-1331. doi: 10.1016/j.rasd.2013.07.024.