Smyrna Tıp Dergisi ***Araştırma Makalesi***

The relationship between sleep disorder, pattern of attachment style and parental quality of life in siblings of children with autism spectrum disorder
Otizm spektrum bozukluğu olan çocukların kardeşlerinde uyku bozukluğu, bağlanma stili ve ebeveyn yaşam kalitesi arasındaki ilişki

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Summary
Objective: The purpose of this study is to analyze the relationship between sleep disorders and pattern of attachment in the siblings of children with autism spectrum disorder (ASD), and to investigate the quality of life in families with children with ASD.
Material and Method: The siblings of 25 children diagnosed with ASD and their mothers and 25 healthy children and their mothers as the control group were included in the study. The mothers of all the participants were given the Sociodemographic and Clinical Data Form, Children's Sleep Habits Questionnaire (CSHQ), Quality of Life in Autism Questionnaire – Parent Version (QoLA-P) and Symptom Check List (SCL-90-R). In order to evaluate the pattern of attachment and interaction of the children and mothers in both groups, Mother Infant Multiaxial Evaluation (MIME) structured interview scale was applied.
Results: There was no significant difference between the demographic data of the healthy children in the control group and the children in the subject group. The frequency of sleep disorders and insecure attachment pattern in the children in the subject group was determined as high. However, no significant difference was found between the groups (p:0.06, 0.069). A positive and significant relationship was found between sleep disorders and insecure attachment in the children in both groups (r=0.287). The flexibility for conformity sub-group score of MIME of the parents of the subject group and the mutual attention sub-group score of the children in the subject group was found low (p:0.022, 0.008). Mean score of QoLA-P in the case group was found lower compared to the parents in the control group.
Conclusion: It was determined that there is a significant relationship between insecure attachment and sleep disorders in the siblings of children diagnosed with ASD. There is a need for future studies with more participants to be able to understand the relationship between sleep disorders and pattern of attachment in the siblings of children with autism.
Key words: Attachment, autism spectrum disorder, quality of life, sleep disorders

Özet
Amaç: Bu çalışmada otizm spektrum bozukluğu olan çocukların kardeşlerinde uyku bozukluğu, bağlanma örüntüsünün değerlendirilmesi, uyku bozukluğu ile bağlanma arasındaki ilişkinin incelenmesi ve otizmli çocuğu olan ailelerde yaşam kalitesinin araştırılması amaçlanmıştır.
Gereç ve Yöntem: Çalışmamıza OSB tanılı 25 çocuğun kardeşleri ve anneleri ile control grubu olarak 25 sağlıklı çocuk ve anneleri dahil edildi. Tüm katılımcıların annelerine Sosyodemografik ve Klinik Veri Formu, Çocuk Uyku Alışkanlıkları Anketi, Otizmde Yaşam Kalitesi Anketi – Ebeveyn Sürümü ve Ruhsal Belirti Tarama Listesi verildi. Her iki gruptaki çocuk ve annelerin bağlanma ve etkileşim örüntülerini değerlendirmek için Anne Bebek Çoğul Eksenli Değerlendirme (MIME) yapılandırılmış görüşme ölçeği uygulandı.
Bulgular: Kontrol grubundaki çocukların demografik verileri ile olgu grubundaki çocukların sosyademografik verileri arasında anlamlı bir fark yoktu. Olgu grubundaki çocuklarda uyku bozuklukları ve güvensiz bağlanma örüntüsü sıklığı yüksek olarak belirlendi. Ancak gruplar arasında anlamlı fark bulunmadı (p:0,06, 0,069). Her iki gruptaki çocuklarda uyku bozuklukları ile güvensiz bağlanma arasında pozitif yönte ve anlamlı bir ilişki bulundu (r=0,287). Olgu grubunun ebeveynlerinin MIME uygunluk alt grup puanları ve olgu grubundaki çocukların

karşılıklı dikkat alt grup puanları düşük bulundu (p:0,022, 0,008). Olgu grubundaki yaşam kalitesi puan ortalaması kontrol grubundaki ebeveynlere göre daha düşük bulundu.
Sonuç: OSB tanısı alan çocukların kardeşlerinde güvensiz bağlanma ile uyku bozuklukları arasında anlamlı bir ilişki olduğu belirlendi. Otizmli çocukların kardeşlerinde uyku bozuklukları ile bağlanma örüntüleri arasındaki ilişkinin anlaşılabilmesi için gelecekte daha fazla katılımcı ile yapılacak çalışmalara ihtiyaç vardır.
Anahtar kelimeler: Otizm spektrum bozukluğu, bağlanma, yaşam kalitesi, uyku bozuklukları

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Introductıon

It is considered that attachment and sleep are systems which develop towards the end of the first year of life (1). It is stated that waking up at night in the first year of life for infants is natural. It is observed that although most infants start sleeping throughout the night when they are 12 months old, some infants cannot sleep. Regulation of sleeping and being awake depends on the maturation of primarily biological networks but it is seen that they are associated with variables related to infants and mothers, including psychosocial factors and temperament as well. When these psychosocial factors are analyzed, it is stated that another factor considered to be associated with infants' waking up at night is the pattern of attachment between the mother and the infant (2). It is stated in a study carried out with 71 female and 63 male infants (aged 6-36 months) that there is a positive relationship between sleep problems and insecure attachment (3). In another study in which the relationship between behaviors in the infancy period and attachment is analyzed, it is stated that infants who have a secure attachment style are able to soothe themselves at night, they wake up less at night, whereas Babies with insecure attachment continue waking up at night in their second year (4). According to the theory of psychodynamics, falling asleep at night and waking up in the morning represent breaking away from the subject of attachment and reuniting with it. Since attachment studies are based on the paradigm of breaking away and getting close again, it is considered that regulation of being asleep and awake can represent a reflection of the attachment system (5). Parents have identified various sleep problems in About 25-50% of preschool children (6). Sleep problems in children diagnosed with Autism Spectrum Disorder is expressed as a common problem and more sleep disorders are seen in these children compared to children with normal development and it is stated that these problems are in the 50-80% range (7). When studies which evaluate sleep disorders in the siblings of children with autism are analyzed, it can be seen that the results are conflicting (7,8,9). In a study carried out in this area, it is stated that cases of insomnia, nightmare disorder and sleep talking are more frequent in the siblings of children with autism compared to children with normal development (7). On the other hand, it is stated in another study in which sleep disorders were studied in children with autism, children with normal development and children with developmental delays, it is stated that there is no difference between the groups (10). In addition, sleep disorders are a very important factor which influence the quality of life for children and adolescents diagnosed with ASD and their families (11). There are numerous studies of recent dates which analyze the effect of health conditions on individuals and their families (12,13). In the studies related to ASD among these, it is stated that this condition negatively affects the patient, the care giver and other individuals the patient lives with due to its nature (14). When the literature is reviewed, it can be seen that there are no studies analyzing the relationship between attachment, sleep and quality of life in the siblings of children diagnosed with ASD. Taking this as the starting point, it was aimed at analyzing the relationship between sleep, attachment and quality of life in the siblings of children diagnosed with ASD and comparing it with the healthy control group

Material and Methods

The study was carried out with the siblings aged 1-3 years of age of 25 patients being followed-up with the diagnosis of ASD and the healthy control group in the same age group in May 2018-May 2019. Approval was received from the non-interventional local ethics committee on

26.04.2018 with document no: 2018/1388. The parents were informed about the study in detail and the verbal and written consent (Informed Consent Form) of the volunteering parents were taken and they were included in the study. The patients were explained that they had the right to object to participate in the study or end the interview at any point in time and they were assured that the recordings would be kept confidential. Individuals who volunteered and were literate and could give written consent were included in the study. Children with known psychiatric/neurological diseases and those who did not wish to participate in the study were excluded. Mothers with children diagnosed with ASD and mothers who would match demographic data such as age, marital status, educational level and economic level with no current or past diagnosed psychiatric diseases and with children who were not diagnosed with any diseases were included in the study as the control group. For the study, 41 mothers with children diagnosed with ASD were interviewed. 12 of them were excluded wince they did not want to participate, 4 were excluded as their children had a medical condition which required treatment and 2 were excluded as they failed to complete forms given to them. 50 mothers-children who met the inclusion criteria were included in the study. The sociodemographic data form for all the participants was filled in by the clinician. The participants were given Children's Sleep Habits Questionnaire (CSHQ), Quality of Life in Autism Questionnaire – Parent Version (QoLA-P) and Symptoms Check List (SCL-90-R). Children and mothers in both groups were given the MIME structured interview scale.

Sociodemographic and Clinical Data Form

This form which was developed by us consists of items which question the sociodemographic characteristics (age, gender, age of mother-father, educational level of mother-father, occupation of other-father, number of siblings, duration of being nursed by mother, state of psychiatric diagnosis and corresponding diagnosis, state of medical diagnosis, medication, whether there are other people living with the family) of the children and parents of the participants.

Children's Sleep Habits Questionnaire

Children's Sleep Habits Questionnaire (CSHQ)-Abbreviated Form developed by Owens et al. in 2000 designed to analyze the sleep habits and sleep related problems of children consist of a total of 33 items. The mother-father of the children are asked to evaluate their children's sleep habits taking the previous week into consideration. The 41 points received as total is accepted as the cut-off point and the values over this are evaluated as "clinically significant" (15).

Quality of Life in Autism Questionnaire-Parent Version

The questionnaire was divided into two sub-parts (A and B) by Eapen et al. Part A consists of 28 questions which evaluate how parents perceive their own quality of life. Each question is scored with 5 point Likert scale from one (none) to give (too much/high). Part B evaluates the parents' perception as to how problematic the difficulties related to ASD their children experience is for them. The score which can be received from the scale is between 48-240, but it is suggested to score and use each part separately (16). Turkish validity and reliability study was carried out by Gürbüz Özgür B et al. (2017) (17).

Mother Infant Multiaxial Evaluation (MIME)

It is a unique method based on analyzing the quality of interaction between infants and their mothers in laboratory environment and scoring the building blocks of the interaction. It consists of 5 parts (free play, gathering together, filling in survey, structured game and breaking away and uniting). It is scored as the sum of 10 items for both the mother and the child (1: very bad, 5: very good) as 1-5 points of each item. The ten items consist of: physical engagement, emotional engagement, taking pleasure, responsiveness, reciprocity, joint attention, providing independence, flexibility About compliance, support and espousal. The validity and reliability study was carried out by Karabekiroğlu et al. (2017) (18).

Symptom Check List (SCL-90-R)

It was developed by the inventory known as Hopkins Symptom Check List (HSCL) developed by Derogatis et al. (19). HSCL's sub-scale consisting of 5 symptoms were added Somatization (SOM), Obsessive Compulsive (O-C), Interpersonal Sensitivity (INT), Depression (DEP), Anxiety (ANK) and 4 new sub-scales as Anger-Hostility (HOS), Phobic reaction (PHOB), Paranoid thought (PAR) and Psychoticism (PSY); therefore, it

consists of a total of 10 symptom groups as 9 sub-scales and an additional scale (20).

Evaluation

Firstly, the mothers were given information About the study and their approval was taken. Then, the mothers filled in the Sociodemographic and Clinical Data Form, Children's Sleep Habits Questionnaire (CSHQ), Quality of Life in Autism Questionnaire – Parent Version (QoLA-P) and Symptom Check List (SCL-90-R). The siblings of the children diagnosed with autism and the children in the control group were evaluated by the researcher child psychiatrist through the diagnostic psychiatric interview based on DSM-5 (The Diagnostic and Statistical Manual of Mental Disorders) and it was verified that they did not have any psychiatric diagnosis. Then, the structured scale Mother Infant Multiaxial Evaluation (MIME) was implemented to evaluated the quality of interaction between the children and their mothers in the subject and control groups which is based on scoring the building blocks of interaction. Prior to the application consisting of five parts, the mothers were given information. The scale consists of free play of the mother and the child, gathering together (toys), the mother's filling in the survey, structured game and breaking away and uniting parts. During the application, the mother and the child were recorded with a camera. The data obtained from the camera recording were evaluated by the child and adolescent psychiatrist, given an interaction score and the pattern of attachment was evaluated as well.

Statistical Analysis

For the statistical analysis of data, the SPSS 21.0 statistical software was used. Kolmogorov-Smirnov test was used to determine whether the constant variables displayed normal distribution. In the comparison of categorical data, Chi-Square and Fisher's Exact Chi-Square Test were used. In the comparison of constant variables in the independent groups which carried parametrical characteristics, the Independent Groups T-test (Student t-test) and in the constant variables accepted as non-parametrical, the Mann-Whitney U Test was used. The similarity of the variances between the groups in the parametrical variables which display normal distribution was evaluated with the Levene test. The relationship between the data in the study groups were analyzed with the Pearson correlation test for data which displayed normal distribution and the relationship between data not displaying normal distribution or were ordinal were analyzed with the Spearman correlation test.

Results

The mean age of the participants was calculated as follows: 28.3 months for the subject group (n=25) and 27.3 months for the children in the control group (=25) (p=0.686). 56% of the children in the subject group was male (n=14), 44% was female (n=11) and the male/female ratio was 1.27. 60% of the children in the control group was male (n=15), 40% was female and the male/female ratio was 1.5. It was determined that the children were a minimum of 13 months old and a maximum of 45 months old and that their mean age was 27.8. The mean age of the mothers in the subject group was 33.4 ±5.8, the mean age of the fathers was 36.6 ±5.8, whereas the mean age of the mothers in the control group was 29.2 ±4.7, the mean age of the fathers was 32.7 ±4.2 (successively p= 0.008, 0.014). It was determined that there was no significant relationship between the educational levels of the mothers in both groups (p=0.326). The educational level of the fathers in the subject group was significantly low (p=0.015). It was determined that there was no significant difference between the parents' smoking and drinking (alcoholic beverages) habits (p=0.637, p=0.312) (p=0.556, p=0.552). It was determined that there was no significant difference between the insecure attachment frequency of the subject group and control group children (p=0.069) (Table 1). A significant difference was not found when the frequency of sleep disorders was compared between the children in both groups (p=0.069) (Table 2). When the CSHQ sub-group scores' distribution was analyzed, it was determined that the mean score of waking up at night in the subject group children was higher (p=0.011). A significant difference was not found between the other sub-group scores of CSHQ (Table 3). A significant positive relationship was found between sleep disorders and insecure attachment in the children in both groups (r=0.287). The mean score of the mothers in the subject group was found as 37.5 ±4.5 and the mean score of the mothers in the control group was found as 39.2 ±5.9 for MIME. The mean score of the children in the subject group was found as 39.3 ±4.4 and the mean score of the children in the control group was found as 39.5 ±4.8 for MIME. It was determined that the mother-child interaction of the children in the

subject group was bad in 4% (n=1), medium level in 56% (n=14) and good in 40%, whereas the mother-child interaction of the children in the control group was medium in 60% (n=15) and good in 40% (n=10). It was determined that there was no difference between the groups in terms of mother-child interaction (p=0.349) When the sub-group score distribution was analyzed, the mean score of the mothers in the subject groups in the flexibility for compliance sub-group was statistically significantly low (p=0.022). A significant difference was not found between the other MIME sub-groups (Table 4).

In the distribution of the MIME sub-group scores, the mean score of the subject group children in the reciprocal attention sub-group was found statistically significantly low (p=0.008) (Table 5). No difference was found between the distribution of the SCL 90-R scale sub-group scores and total scores. It was determined that the quality of life mean score for the parents in the subject group was 92.8 and the quality of life mean score for the parents in the control group was 107.2 (p=0.012). A significant negative relationship was found between the SCL-90 R sub-group scores and the quality of life score and between somatization symptom sub-group score and quality of life mean score (r= 0.374**)**.

Table 1. *Comparison of attachment patterns between groups*

Attachment style	**Case** n (%)	**Control** n (%)	**Total** n (%)	χ^2	***p***
Secure	14 (56)	20 (80)	34 (68)	3.309	0.069
Insecure	11 (44)	5 (20)	16 (32)		

Table 2. *Comparison of the results of CSHQ scale of children in the case and control group*

Sleep Disorder	**Case** n (%)	**Control** n (%)	**Total** n (%)	χ^2	***p***
Absent	5 (20)	11 (44)	16 (32)	3.309	0.069
Available	20 (80)	14 (56)	34 (68)		

Table 3. *Comparison of the distribution of the CSHQ subgroup scores of the children in the case and control group*

CSHQ Subgroup Scores	**Case** Mean ±sd	**Control** Mean ±sd	**Mann-Whitney U** (p)
Bedtime	10.3 ±3.1	9.7 ±2.0	0.525
Sleep Time	3.6 ±0.7	3.8 ±1.3	0.618
Delayed Falling Asleep	1.4 ±0.6	1.4 ±1.4	0.507
Sleep Anxıety	6.7 ±2.0	6.8 ±2.0	0.936
Night Wakes	6.0 ±1.7	4.8 ±1.4	**0.011***
Parasomnias	8.7 ±1.1	8.6 ±1.7	0.509
Disruption of Breathing During Sleep	3.0 ±0.2	3.2 ±0.6	0.293
Sleepiness During the Day	9.0 ±1.6	9.0 ±2.2	0.942*

*******Student T test was used.* ***CSHQ:*** *Children's Sleep Habits Questionnaire* ***sd:*** *standart deviation*

Table 4. *Comparison of the distribution of MIME subgroup points for parents between groups*

Parent Subgroups	Case Mean ±sd	Control Mean ±sd	Mann-Whitney U (p)
Physical Engagement	4.5 ±0.6	4.3 ±0.8	0.440
Emotional Engagement	3.7 ±0.8	3.8 ±1.0	0.753
Taking Pleasure	3.5 ±0.6	3.5 ±0.9	0.746
Responsiveness	3.9 ±0.5	4.0 ±0.6	0.621
Reciprocity	3.2 ±0.6	3.6 ±0.8	0.800
Joint Attention	3.9 ±0.4	4.1 ±0.6	0.112
Providing İndependence	3.5 ±0.8	3.9 ±0.8	0.085
Flexibility About Compliance	3.6 ±0.7	4.0 ±0.8	**0.022**
Support	3.8 ±0.6	3.8 ±0.8	0.777
Espousal	3.7 ±0.7	3.8 ±0.7	0.655

MIME**:** *Mother Infant Multiaxial Evaluation (MIME)* ***sd:*** *standart deviation*

Table 5. Comparison of the distribution of MIME subgroup scores for children between groups

Child Subgroups	Case Mean ±sd	Control Mean ±sd	Mann-Whitney U (p)
Physical Engagement	4.4 ±0.6	4.5 ±0.6	0.895
Emotional Engagement	4.0 ±0.7	4.0 ±0.6	0.814
Taking Pleasure	4.3 ±0.6	4.1 ±0.7	0.395
Responsiveness	3.9 ±0.6	4.0 ±0.5	0.604
Reciprocity	3.4 ±0.8	3.8 ±0.6	0.055
Joint Attention	3.8 ±0.5	4.2 ±0.5	**0.008**
Providing İndependence	4.0 ±0.5	3.9 ±0.4	0.788
Flexibility About Compliance	4.0 ±0.7	3.9 ±0.5	0.346
Support	3.9 ±0.4	3.8 ±0.4	0.760
Espousal	3.8 ±0.6	3.9 ±0.4	0.521

MIME**:** *Mother Infant Multiaxial Evaluation (MIME)* ***sd:*** *standart deviation*

Discussion

In this study, the relationship between sleep disorders and pattern of attachment in the siblings of children diagnosed with ASD was analyzed. It was determined that the frequency of sleep disorders and pattern of insecure attachment in the subject group was higher compared to the healthy control group. In addition, it was determined that there is a positive relationship between sleep disorders and pattern of insecure attachment in both groups. No studies were found in the literature which analyzes the relationship between sleep disorders and pattern of attachment in the siblings of children diagnosed with ASD. When studies in which the relationship between sleep and attachment is analyzed in children with normal development, contradictory results can be found (21,22,23). In a study in which the relationship between sleep and attachment in the early childhood period was analyzed, a relationship was found between sleep problems and insecure attachment (24). On the other hand, Scher et al. stated that the frequency of waking up in infants with secure and insecure attachment patterns was not different (25). The findings of our study shows that secure attachment in the subject group and the healthy control group was higher. It is stated that the most frequently seen patter of attachment in children with normal development is secure attachment in the rate of 65-50% (26,27,28,29). Our findings show that the frequency of sleep disorders in the subject group children is higher. It was also determined that the mean score of waking up at night in the subject group children was high. In a research carried out in this area, it was reported that insomnia, nightmare disorder and sleep talking are more frequent in the siblings of children with autism compared to children with normal development (30). In addition, it was determined that children with autism who have sleep problems cause sleep problems in other family members who live with them as an environmental factor and that the siblings of children with autism similarly wake up frequently at night (10). The results of our study show that

there is no difference between the groups in terms of mother-child interaction. It was observed that there are very few studies in the literature in this area. It was stated in a study carried out in this area that the mother-child interaction between the siblings of children with autism and their mothers is significantly low compared to the siblings of healthy children (31). It was stated in a study in the literature that the response of children with autism in looking directly at their siblings is delayed compared to infants with normal development and that atypical response to direct glance emerges in early childhood and it is related to wide autism's phenotype characteristics (31). In our study, only the mean scores of reciprocal attention sub-group in the children in the subject group were found significantly low. It is considered that this might be related to the fact that the children in the subject group of our study display wide autism phenotype characteristics (32,33,34). In our results, a difference was not found between the SCL 90- R scale's distribution of sub-group scores and total scores. Contrary to our study, when we analyzed the studies in the literature which evaluated the psychiatric symptoms in the parents of children with autism, it was seen that the rate of anxiety, depression, obsessive compulsive disorder in the parents of children with autism is higher compared to the parents of children with normal development (35,36,37,38). In our results, the mean score of the quality of life of parents in the case group was found lower compared to the parents in the control group. The results of our study is in line with the literature (39,40). Our results should be evaluated with the consideration of certain limitations. The first of the limitations is that the number of participants is relatively low. The age of all the participants not being the same in terms of months can be listed among the limitations as well. More studies need to be carried out in larger sample groups for our results to be significant.

Conclusion

This study is the first study in which the relationship between sleep disorders and pattern of attachment in the siblings of children diagnosed with ASD is analyzed. A positive significant relationship was found between sleep disorders and insecure attachment pattern. Further studies need to be carried out todetermine the relationship between sleep disorders and attachment pattern.

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