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PREVALENCE OF STEREOTYPIES AMONG ADULTS WITHOUT NEUROLOGICAL DISORDERS AND INTELLECTUAL DECLINE

Stereotypies, along with tics, are the most common hyperkineses, especially among children. It has been esTableished that stereotypies can occur among children with normal development. At the same time, we found only one article on the prevalence of stereotypies among relatively healthy adults. **The aim of the study** is to assess the prevalence and nature of stereotypies among adults without neurological and intellectual disorders and to determine its relationship with tics and associated psychiatric symptoms. Material and methods. We have created an online questionnaire that includes 32 questions about the presence of movement disorders and their characteristics, assessment of anxiety, depression and attention level. 80 people were studied, including 21 men and 59 women, the median age was 23.0 [22.0; 26.0] years. **The results of the study.** Of the 80 people who participated in the study, signs of stereotypies, motor and/or vocal tics were identified in 60 people (75%). Each of them had from 1 to 15 symptoms (median – 3.5 [3.0; 6.0]). In 59 (73.8%) people, the revealed motor symptoms were regarded as stereotypies, in 10 (12.5%) people – motor tics, in 6 (7.5%) – vocal tics. In 4 people (5.0%), motor tics were combined with vocal tics, which is typical for Tourette's syndrome. Participants with violent movements were more likely to have a high level of anxiety (53.3% vs. 10%), which often indicated obsessive-compulsive symptoms (41.7% vs. 15.0%), and they also performed worse on the Schulte Table test (40.0 sec vs. 31.5 sec). In-person examination of respondents demonstrated a high level of sensitivity and specificity of the questionnaire in relation to stereotypies (100% and 66.7%, respectively), but a low level of sensitivity to tics (33.3%) with 100% specificity. **Conclusion.** Stereotypies are quite common among the adult population, even in the absence of organic brain damage and cognitive decline, and in about half of the cases they are combined with obsessive-compulsive symptoms and anxiety.

Keywords: stereotypies, motor tics, vocal tics, obsessive-compulsive symptoms, anxiety.

Stereotypies, along with tics, are the most common hyperkineses, especially among children [6]. Stereotypies are repetitive and aimless movements that have a certain pattern and can be stopped by distraction. These include both simple movements in the form of stamping with your foot, twisting your hair, biting your nails, and complex movements – flapping your arms, rotating your hands, swaying your torso, orofacial movements, as well as self-harming behavior [4, 11, 13].

Stereotypies are often observed in various mental disorders and diseases of the nervous system. Thus, according to the meta-analysis, 21.9–97.5% (median – 51.8%) of children with autism spectrum disorder (ASD), regardless of gender, have stereotypies, and they are associated with a younger age, lower level of intelligence and severity of ASD [16]. Other causes of the development of stereotypies can be schizophrenia, affective disorders, oligophrenia, genetic diseases (most often Rett syndrome), as well as neurological diseases such as epilepsy, Tourette syndrome, Parkinson's disease [14].

However, simple stereotypies can be observed in 20-70% of cases, and complex stereotypies – in 3-4% of cases of normally developing children [13]. They do not differ phenotypically from pathological stereotypies, they are most often manifested by thumb sucking, body swaying and nail biting and are more often identified in boys (3:2 ratio) [7]. Such stereotypies, observed more often in children under 2 years of age, are associated with the maturation of the neuromuscular pathways and insufficient maturity of the inhibitory effect of the cortex, especially the frontal lobes [1].

If the prevalence and phenomenology of stereotypies in normally developing children have been studied well enough, then this cannot be said about adults. Stereotypical motor disorder in intellectually intact adults without mental disorders was first described in 1996 by Castellanos F. et al. So, out of 20 respondents, 12 people had signs consistent with DSM-IV SMD. 11 of the 12 subjects had a history of affective or anxiety disorder [8].

Tics are semi-spontaneous, sudden, rapid, repetitive, irregular movements or vocal tics in response to an imperative urge, a sensory feeling that leads to the need to make a particular movement (the so-called "premonitory urge") [2]. Motor and vocal tics are the basis of Tourette's syndrome and occur mainly in children, adolescents and young people (under 18 years of age) and, as a rule, tend to regress. They are more common in boys than in girls, but in adulthood this predominance is less pronounced [9].

The aim of the study is to assess the prevalence and nature of stereotypies among adults without neurological and intellectual disorders and to determine its relationship with tics and associated psychiatric symptoms.

Materials and methods of research. The study was conducted on the premises of the Department of Neurology and Psychiatry of the Medical Institute of the M.K. Ammosov North-Eastern Federal University. Based on the aim of the study, we created an online questionnaire that includes 32 questions and is divided into the following sections:

1. General information: gender, age, ethnicity, employment, information about parents and siblings, information about previously diagnosed neurological and somatic diagnoses, information about taking medications, information about bad habits, information about academic performance at school and university.
2. Signs of stereotypies and tics. The patients had to note certain motor phenomena that they observed. The questions were based on the Yale Global Tic Severity Scale (Leckman et al., 1989), in addition, willpower suppression, the presence of premonitory urge, duration and frequency of motor phenomena, cessation during distraction, influence on

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daily activity, aggravating and reducing factors were evaluated.

3. Obsessive-compulsive symptoms based on the Yale Brown Obsessive-Compulsive Scale (Goodman et al., 1989).

4. Assessment of anxiety and depression on the HADS scale (Hospital Anxiety and Depression Scale).

5. Evaluation of the attention function according to the Schulte Table test.

Participants had the opportunity to provide contact details to send the results of their research, as well as to clarify the specified information.

The object of the study were students of years 4-6 of a medical university. Before starting the survey, each respondent was instructed on the basic terminology.

Criteria for inclusion in the study: 1) age 18 and older; 2) voluntary consent to participate. Exclusion criteria: 1) age under 18 years; 2) the presence of epilepsy and other paroxysmal disorders; 3) the presence of cognitive impairment that prevents the analysis of the data obtained; 4) the presence of diseases of the nervous system accompanied by the development of chorea, dystonia, tremor, paresis, and paralysis.

A total of 80 people participated in the study, including 21 men (26.3%) and 59 women (73.7%). The median age of the respondents was 23.0 [22.0; 26.0] years, all were university students, two participants were employed at the same time (2.5%). By ethnicity, the majority of participants were Yakuts (66 people, 82.5%), Russians – 5 people (6.3%), another ethnic group – 9 people (11.2%).

Based on the analysis of these motor symptoms, all participants were divided into two groups. The first (main) group consisted of persons with motor symptoms that can be attributed to tics and/or stereotypies, the second (control) group consisted of persons without motor phenomena.

During the study, none of the respondents were recommended to take medications and other therapeutic manipulations. If a motor phenomenon was present, the report included the nature of hyperkinesis (stereotypies, tics), the presence of associated mental symptoms (obsessive-compulsive symptoms, signs of anxiety and depression) and the recommendation of an in-person consultation with a specialist (neurologist or psychiatrist).

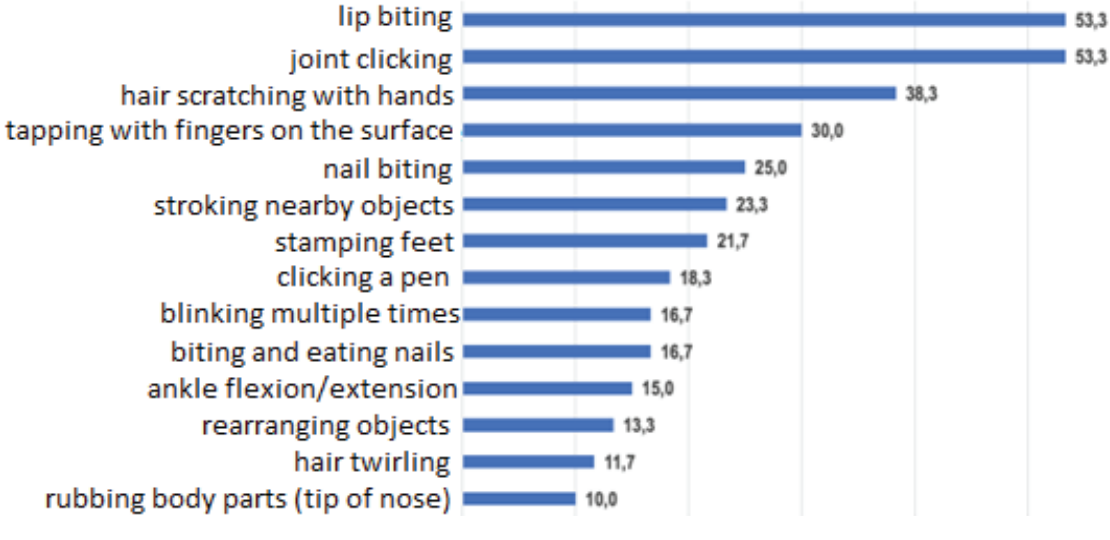
To check the sensitivity and specificity of the created questionnaire from among the respondents, we invited a group of volunteers for an in-person examination, who underwent a neurological examination and a thorough analysis of existing hyperkinesis.

Statistical processing was carried out in the SPSS Statistica 25.0 program. Since the distribution of most of the studied quantitative indicators differed from the normal one, their descriptive statistics are given in the form of the median, the 25th and 75th quartiles (Me [Q1; Q3]), and the Mann-Whitney U-test was used to compare the two groups of quantitative data. Qualitative data are presented in the form of frequencies, for comparison of binary data, a contingency Table was used with the calculation of the Pearson $\chi 2$ criterion or the Fisher exact criterion, depending on the assumed minimum value. To determine the strength of the connection between two nominal variables, Cramér's V criterion was calculated. The differences at $p \leq 0.05$ were considered statistically significant.

The results of the study. Of the 80 people who participated in the study, signs of stereotypies, motor and/or vocal tics were identified in 60 people (75%). Each of them had from 1 to 15 symptoms (median – 3.5 [3.0; 6.0]). The most common motor symptoms were lip biting (53.3%), joint clicking (53.3%), hair scratching with hands (38.3%) and tapping with fingers on the surface (30.0%). Figure 1 shows the frequency of the identified hyperkineses.

In 59 (73.8%) people, the revealed motor symptoms were regarded as stereotypies, in 10 (12.5%) people – motor tics, in 6 (7.5%) – vocal tics. In 4 people (5.0%), motor tics were combined with vocal tics, which is typical for Tourette's syndrome. However, none of the respondents with this combination of signs had such a diagnosis, one participant was diagnosed with residual encephalopathy, and another was diagnosed with intracranial hypertension. A combination of motor tics and stereotypies was found in 9 (11.3%) people, and a combination of all three signs (motor tics, vocal tics, stereotypies) – in 3 (3.8%) people.

The respondents with hyperkineses did not differ from the representatives of the control group in age and the age of the mother at the time of birth. In the main group, 17 (28.3%) people had at least one neurological diagnosis in their medical history, and in the control group, 3 (15.0%) people ($p = 0.234$). Moreover, in both groups, intracranial hypertension was most often diagnosed: in 10 (16.7%) people of the main group and 2 (10.0%) people of the control group ($p = 0.47$). Attention deficit hyperactivity disorder



Frequency distribution of hyperkineses (in % of all responses).
Note: symptoms observed in less than 10% of patients are not included.

(ADHD) was noted in only one respondent with stereotypies.

The respondents of both groups did not differ statistically in the frequency of rheumatism, helminthiasis, and viral hepatitis. At the same time, anemia was noted by 32 (53.3%) representatives of the main group and only 3 (15.0%) representatives of the control group (p = 0.003; V= 0.335). Of the respondents in the main group, 1 (1.7%) people took antidepressants, 1 (1.7%) people took neuroleptics, 3 (5.0%) people took tranquilizers. Among the respondents of the control group, the use of these drugs was not registered. Although smoking was more often registered in the main group (40% versus 20%), no statistically significant difference was achieved.

Representatives of the main group more often ended their semester with good and excellent grades (93.3% vs. 75.5%, V = 0.25; p = 0.025), although they did not differ in academic performance from representatives of the control group.

The analysis of associated psychiatric disorders showed (Table 2) that the respondents of the main group had a statistically significantly higher level of anxiety, and anxiety was detected in 32 (53.3%) people, whereas among the control group – only 2 (10.0%) people. Obsessive-compulsive symptoms were found in 25 (41.7%) representatives of the main group and only in 3 (15.0%) representatives of the control group (p = 0.023). The most common symptoms among the main group were the desire to double-check (13 people, 21.7%), the need for symmetry (9 people, 15.0%) and the need for excessive cleanliness (8 people, 13.3%). Of the 25 people with obsessive-compulsive symptoms, 23 people noted the meaninglessness of rituals, but could not get rid of them on their own. In addition to anxiety and obsessive-compulsive symptoms, the respondents of the main group were slower at passing the Schulte Table test (40.0 [33.0; 54.5] sec versus 31.5 [27.5; 51.0] sec, p = 0.032).

Out of 80 people, 10 people agreed to take part in an in-person neurological examination. In 7 people who, according to the results of an online survey, hyperkineses were regarded as stereotypies, as a result of the analysis of complaints, analysis, neurological examination, the nature of hyperkinesis was confirmed. On the contrary, among the three people who were assigned to the control group, one respondent revealed stereotypies during an in-person examination. Thus, the sensitivity of our questionnaire in relation to stereotypies was 100%, the specificity was 66.7%.

The questionnaire's sensitivity to ticks was significantly lower. Thus, tics were identified in only one patient out of three volunteers with hyperkineses. But at the same time, none of the control group had been identified with tics. Consequently, the sensitivity was 33.3%, and the specificity was 100%.

Discussion. Our study shows the widespread prevalence of stereotypies among adults without neurological diseases and intellectual disabilities. Tics and stereotypies are two of the most common non–directional motor behaviors, which in some cases can coexist. If in mild manifestations they decrease over time and do not require active intervention, in severe cases they can persist further into adulthood and affect daily activity [15].

A recent systematic review revealed that 23% of children aged 8 to 31 years with stereotypical motor disorder have concomitant tics, 37.6% of patients have ADHD and 16.5% have obsessive-compulsive symptoms. And 8% of children with Tourette's syndrome have stereotypies [6]. Consequently, stereotypies, tics, ADHD and obsessive-compulsive disorder may have a common pathophysiological basis. According to our data, a

Table 1

General characteristics of the respondents

Parameter	Main group (n=60)	Control group (n=20)	p-level
Age, years	23.0 [22.0; 25.0]	25.5 [23.0; 27.0]	p = 0.043
Males, abs. (%)	14 (23.3)	7 (35.0)	p = 0.304
Age of the mother at the time of the respondent's birth, years	29.0 [24.2; 35.75]	31.0 [22.5; 36.75]	p=0.854
Neurological diagnosis since childhood, abs. (%)	7 (11.7)	2 (10.0)	p = 0.838
Neurological diagnosis in medical history, abs. (%)	17 (28.3)	3 (15.0)	p = 0.234
Somatic diagnosis in medical history, abs. (%)	39 (65)	8 (40)	p = 0.05
Smoking, abs. (%)	24 (40)	4 (20)	p = 0.104
Excellent and good grades as of last semester, abs. (%)	56 (93.3)	15 (75)	p = 0.025; V = 0.251

Table 2

Associated psychiatric symptoms in respondents

Parameter	Main group (n=60)	Control group (n=20)	p-level
HADS, anxiety, points	7.0 [5.0; 10.0]	4.0 [2.25; 5.75]	p < 0.001
Anxiety, abs. (%)	32 (53.3)	2 (10.0)	p = 0.001; V = 0.38
HADS, depression, scores	5.0 [3.25; 8.0]	4.0 [2.0; 6.75]	p=0.151
Depression, abs. (%)	22 (36.7)	5 (25.0)	p = 0.339
Obsessive-compulsive symptoms, abs. (%)	25 (41.7)	3 (15.0)	p=0.023
Schulte Table test, sec.	40.0 [33.0; 54.5]	31.5 [27.5; 51.0]	p=0.032

combination of tics and stereotypies was observed in 11.3% of cases.

Tics and stereotypies, along with akathisia, restless legs syndrome, obsessive-compulsive disorder, tardive dyskinesia and some levodopa-induced dyskinesias, are combined into the general term "acasitic spectrum disorders", since their development is based on an imperative need to make movements and, probably, a single pathophysiological mechanism [3].

Long-term observation of stereotypies in children without mental retardation and ASD showed that in the vast majority of cases (81%) they develop before 24 months and are more often associated with ADHD (30%), tics (18%) and OCD (10%) [12]. In almost all cases, stereotypies have a protracted course. Thus, a survey of 49 children and adolescents aged 9 to 20 years revealed that during the entire observation period (range from 6.8 to 20.3 years) stereotypies persist in 48 (98%) people, including 9 (19%) people - for more than 15 years. However, 37 people noted an improvement in symptoms, and 10 people's symptoms remained at the same level [17].

Tics in the vast majority of cases begin at the age of 5 to 8 years, occur three times more often in men, characterized by cranio-caudal spread with the first tics in the form of blinking, movement of the nose, face, whereas the muscles of the trunk and limbs are involved later, the peak of tics occurs at the age of 10 to 13 years, after then their severity gradually decreases [5].

Freeman R. and colleagues observed 42 children (11 girls and 31 boys) with a stereotypical motor disorder, but without self-harming disorder, mental retardation, and ASD. Among them, ADHD was found in 16 people, tics in 18 people, including Tourette's syndrome in 11 people, OCD in 2, obsessive-compulsive behavior in 3 people. The first signs of stereotypies appeared at the age of 17 months, and the diagnosis was esTableished on average at the age of 6 years. The authors observed 39 children for more than 6 months, and 25 people's symptoms improved, including 4 children whose stereotypies were completely stopped. The family history was positive in 13 children [10]. According to the results of our study, representatives of the main group, which mainly included patients with stereotypies, were more likely to have obsessive-compulsive symptoms and anxiety, and were slower at passing the Schulte Table tests.

Attention is drawn to the lack of a diagnosis of Tourette's syndrome in individuals who note all the symptoms of the disease, such as motor and vocal tics.

An interesting finding of the study was the more frequent indication of anemia in the main group of individuals. We conducted a search in PubMed databases, eLibrary.ru, Google Academy, however, we did not find any scientific articles discussing the relationship of anemia with stereotypies or tics. The only disease from the group of "acasitic" disorders that can be caused by iron deficiency is restless legs syndrome [18].

The strength of our research is the high sensitivity of the created questionnaire in relation to stereotypies. Undoubtedly, most respondents, especially when identifying anxiety or obsessive-compulsive symptoms that reduce daily activity, need to consult a neurologist or psychiatrist for an in-person examination. In our study, we were able to show a fairly wide prevalence of stereotypies among adults.

The study also has a number of limitations. First of all, it is the identification of signs using a questionnaire. At the same time, in-person examinations revealed its rather high sensitivity to stereotypies, but low sensitivity to tics. Secondly, we did not take into account the age of hyperkineses development. Thirdly, we limited ourselves to obsessive-compulsive symptoms. However, the last point is quite justified, since the diagnosis of obsessive-compulsive disorder undoubtedly requires face-to-face consultation with a psychiatrist.

Conclusion. Stereotypies are identified not only among children with normal development, but are also quite common among the adult population, even in the absence of organic brain damage and cognitive decline, and in about half of all cases they are combined with obsessive-compulsive symptoms and anxiety. Although active treatment may not be required in all cases, with the influence of stereotypies, tics, obsessive-compulsive symptoms on daily activity, the development of stigmatization, pharmacotherapy or psychotherapy may be options for improving a person's condition.

References

1. Attaeva L.Zh., Makarov I.V. Stereotipii u detei [Stereotypes in children]. Sotsial'naya i klinicheskaya psikhiatriya [Social and clinical psychiatry]. 2021;31(2):79-85 (in Russ.)

2. Zalyalova Z.A. Granitsy neproizvol'nosti tikoznykh giperkinezov [Boundaries of involuntary tic hyperkinesis]. Byulleten' Natsional'nogo obshchestva po izucheniyu bolezni Parkinsona [Bulletin of the National Society for the Study of Parkinson's Disease]. 2022; 2:61-65 (in Russ.) DOI: 10.24412/2226-079X-2022-12436

3. Akhmadulina A.O., Chimagomedova A.Sh., Levin O.S. [et al.]. Klinicheskaya fenomenologiya i patofiziologicheskie mekhanizmy dnevnykh i nochnykh imperativnykh dvizhenii [Clinical phenomenology and pathophysiological mechanisms of daytime and nighttime imperative movements]. Zhurnal nevrologii i psikhiatrii im. S.S. Korsakova [S.S. Korsakov Journal of Neurology and Psychiatry]. 2022;122(11):67-72 (in Russ).] DOI: 10.17116/jnevro202212211267

4. Hoch J., Spofford L., Dimian A. [et al.].A direct comparison of self-injurious and stereotyped motor behavior between preschool-aged children with and without developmental delays. J Pediatr Psychol. 2016;41(5):566-572. DOI: 10.1093/jpepsy/jsv102

5. Martino D., Madhusudan N., Zis P. [et al.]. An Introduction to the Clinical Phenomenology of Tourette Syndrome. International Review of Neurobiology. Amsterdam: Elsevier Inc., 2013:1-33. DOI: 10.1016/B978-0-12-411546-0.00001-9

6. Cavanna A.E., Purpura G., Riva A. [et al.]. Co-morbid tics and stereotypies: a systematic literature review. Neurol Sci. 2023;45(2):477-483. DOI: 10.1007/s10072-023-07095-y

7. Bonnet C., Roubertie A., Doummar D. [et al.]. Developmental and benign movement disorders in childhood. Mov Disord. 2010;25(10):1317-1334. DOI: 10.1002/mds.22944

8. Castellanos F.X., Ritchie G.F., Marsh W.L. [et al.]. DSM-IV stereotypic movement disorder: persistence of stereotypies of infancy in intellectually normal adolescents and adults. J Clin Psychiatry. 1996;57(3):116-122.

9. Szejko N., Robinson S., Hartmann A. [et al.]. European clinical guidelines for Tourette syndrome and other tic disorders-version 2.0. Part I: assessment. Eur Child Adolesc Psychiatry. 2022;31(3):383-402. DOI: 10.1007/s00787-021-01842-2

10. 6Freeman R.D., Soltanifar A., Baer S. Stereotypic movement disorder: Easily missed. Dev Med Child Neurol. 2010;52(8):733-738. DOI: 10.1111/j.1469-8749.2010.03627.x

11. Gal E., Dyck M.J., Passmore A. The relationship between stereotyped movements and self-injurious behavior in children with developmental or sensory disabilities. Res Dev Disabil. 2009;30(2):342-352. DOI: 10.1016/j.ridd.2008.06.003

12. Harris K.M., Mahone E.M., Singer H.S. Nonautistic Motor Stereotypies: Clinical Features and Longitudinal Follow-Up. Pediatr Neurol. 2008;38(4):267-272. DOI: 10.1016/j.pediatrneurol.2007.12.008

13. Katherine M. Stereotypic Movement Disorders. Semin Pediatr Neurol. 2018;25:19-24. DOI: 10.1016/j.spen.2017.12.004

14. Maltête D. Adult-onset stereotypical motor behaviors. Rev Neurol (Paris). 2016;172(8-9):477-482. DOI:10.1016/j.neurol.2016.07.002

15. Martino D., Hedderly T. Tics and stereotypies: A comparative clinical review. Park Relat Disord. 2019;59:117-124. DOI: 10.1016/j.parkreldis.2019.02.005

16. Melo C., Ruano L., Jorge J. [et al.]. Prevalence and determinants of motor stereotypies in autism spectrum disorder: A systematic review and meta-analysis. Autism. 2020;24(3):569-590. DOI: 10.1177/1362361319869118

17. Oakley C., Mahone E.M., Morris-Berry C. [et al.]. Primary complex motor stereotypies in older children and adolescents: Clinical features and longitudinal follow-up. Pediatr Neurol. 2015;52(4):398-403.e1. DOI: 10.1016/j.pediatrneurol.2014.11.002

18. Gossard T.R., Trotti L.M., Videnovic A. [et al.]. Restless Legs Syndrome: Contemporary Diagnosis and Treatment. Neurotherapeutics. 2021;18(1):140-155. DOI: 10.1007/s13311-021-01019-4