

Depression in Tourette Syndrome

Agnieszka Małek¹, Paulina Golińska²

¹ University of Physical Education and Sport in Gdansk,
Department of Physical Culture Pedagogy

² Department of Clinical Psychology and Neuropsychology, University of Gdansk

Summary

Aim. The aim of the study is to show relationship between severity of depressive symptoms and the severity of tics, declared feeling of stigmatization and feelings about the body.

Method. The study included 13 people with Tourette syndrome and 13 people in a comparative group – matching method, taking into account compatibility of gender, age, number of years of education, and size of the place of residence. The study used: Polish adaptation of the Yale Global Tic Severity Scale (YGTSS), Questionnaire for Measuring Depression (Kwestionariusz do Pomiaru Depresji – KPD), Perceived Stigmatization Questionnaire (PSQ), Questionnaire of feelings about the body.

Results. Wilcoxon signed-rank test analysis showed that the declared general depression rate is significantly higher in the group of people with Tourette syndrome ($Z = -2.691$; $p < 0.01$). The indicator differentiating the declared feeling of stigmatization among people with TS was the feeling of embarrassment due to other people's stare/the stare of bystanders (ZG) ($Z = -1.888$; $p < 0.05$). The general assessment of one's body image is not different in the group of people with TS and comparative group, but two important factors have been distinguished: "My body is alien to me" ($Z = -1.897$; $p < 0.05$), "It is difficult for me to understand changes in my body" ($Z = 1.950$; $p < 0.05$).

Conclusions. It is concluded that the severity of tics, the feeling of stigmatization and selected body image indexes are related to both the general severity of symptoms of depression and its individual indicators.

Key words: Tourette Syndrome, depression, psychological stress

Introduction

Tourette syndrome (TS) is a neurodevelopmental disorder characterized by co-occurring motor tics together with a minimum one vocal tic, occurring several times a day or periodically for a period of at least one year after the first episode of the disease [1]. It is estimated that the disease affects about 1% of the population [2]. Although

TS has been considered a rare disease so far, epidemiological studies show that the current incidence is on average 7 per 1000 students [3]. According to data from screening tests conducted in 2000, Tourette syndrome may affect 0.43% to 3.8% of Polish students [4]. The disease affects mostly men, the M:F ratio is estimated at around 4:1 [5]. Until now, no explicit theory explaining the pathomechanism of the disease has been specified. The hypothesis about the inherited dysfunction of the basal ganglia and the cortico-striatal-thalamic loop seems to be the most probable explanation [6].

90% of people with TS have other comorbidities, such as attention deficit disorder, attention deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), impulse control disorders (ICD), self-injurious behavior (SIB), sleep disorders, autism spectrum disorders, anxiety disorders, as well as depression [2, 7, 8].

Data on the frequency of coexistence of Tourette syndrome with affective disorders differ significantly, depending on the used research methodology. Both among children and adults with TS, the risk of developing depression is much higher than in people in the general population [5, 9]. Research on the coexistence of TS genetic mechanisms and depression did not bring definite results. One of the reasons for the occurrence of depression in patients with TS more often than in the population may be side effects of pharmacotherapy – mainly neuroleptics used to reduce tics: haloperidol, pimozide, tiapride, sulpiride or aripiprazole [5]. In addition, among the possible explanations of the increased risk of developing depression, it seems particularly likely that the cause of this phenomenon is indicated by the exclusionary and stigmatizing nature of Tourette syndrome [10]. The patients are exposed to various types of psychological stressors: social isolation, rejection by peers and being a victim of violence, which may lead to lowering their self-esteem [11]. People with Tourette syndrome declare a lower quality of life, which is probably a consequence of all the above-mentioned aspects accompanying the disease [12]. The severity of symptoms of depression may also be related to: number, frequency, severity and complexity of tics [9].

Tourette syndrome is a chronic disease characterized by varying intensity and variety of involuntary movements and vocalization, which can significantly contribute to the intensification of negative reactions and emotions accompanying the disease, consequently leading to depressive disorders [13]. In addition, lowered mood may be associated with deficits in emotional self-regulatory abilities found in people with TS [14]. Depressive disorders can occur with the course of one dominant symptom, mainly irritation, which is probably one of the manifestations of impulse control disorders [15]. The inability (or limitation) to control one's own body may result in dissatisfaction with their own corporeality, while body image disorders can contribute to the development of affective disorders [16].

It has been shown that the coexistence of affective disorders predisposes to psychiatric hospitalization [17]. Both in children and adults, emotional difficulties are associated with deterioration of health-related quality of life [12, 18, 19]. Moreover,

it has been found that depressive disorders can moderate the relationship between the severity of tics and impaired functioning in people with chronic tic disease [20].

Aim of the study

The aim of the study was to analyze the relationship between the severity of depressive symptoms and the intensity of tics, the declared feeling of stigmatization and feelings about the body. Due to the lack or ambiguity of the attitudes presented in scientific research concerning correlates between the indicated variables, the own research was exploratory in nature. The following research question was formulated: do relationships between the studied variables exist – and if they do, what and how strong are they?

Material

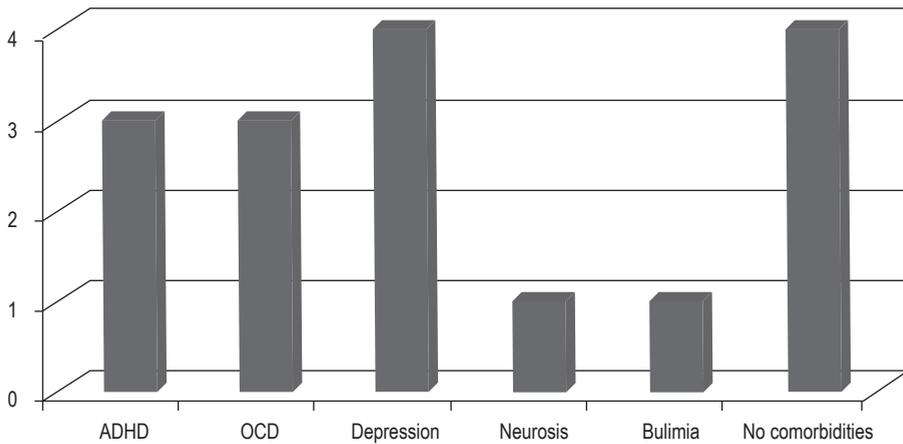
The study was approved by the Ethics Committee of the University of Gdansk. All participants were familiarized with the purpose of the study and agreed to participate in the project.

People with Tourette syndrome were qualified for the study based on a diagnosis made by a neurologist according to the DSM-5 criteria. Other neurological disorders were excluded in the medical examination. Thirteen patients took part in the project – three women and ten men. In the case of minors, both the participants and legal guardians were asked to agree to participate in the study. The mean age of people in the clinical group was 23 years ($SD = 7.5$), including two patients who were minors at the time of the study (17 years). The mean duration of the disease was 15.85 years ($SD = 8.67$).

A structured psychological interview was conducted, during which the subjects declared the number and type of comorbidities. Several participants reported the occurrence of more than one condition. The frequency of the declaration regarding the coexistence of individual diseases is presented in the Figure 1.

In addition, the subjects were asked for information on the drugs used (currently or in the past) to reduce tics and/or comorbidities. Eight people from the clinical group received medicines at the time of the study (haloperidol, sulpiride, pimozide, aripiprazole or ziprasidone), three – in the past (risperidone, clonidine, haloperidol, sulpiride), two subjects declared that they had never used pharmacological therapy. It should be noted that the main goal of taking drugs by people from the clinical group was tic reduction, however, those drugs are also widely used in the treatment of co-occurring disorders, including ADHD, OCD and anxiety disorders.

Among the persons participating in the study, four participated in the individual cognitive behavioral therapy in the past, two benefited from ad hoc psychological help; other people have never participated in any form of therapy.



ADHD – Attention Deficit Hyperactivity Disorder; OCD – Obsessive Compulsive Disorder

The procedure for selecting people for the comparative group included the matching method, taking into account gender compatibility, age, number of years of education and the size of the place of residence. Data from structured clinical interview allowed to exclude mental illnesses and neurological diseases among people from the comparative group, none of them also declared taking drugs permanently.

Method

The study used the Polish adaptation of the Yale Global Tic Severity Scale (YGTSS) [21], developed by Stefanoff and Wolańczyk [22]. The YGTSS consists of two parts. The first part contains a list of simple and complex motor and vocal tics present in the patient during the last week. The second part is used to assess the number, frequency, severity and complexity of tics, as well as the general impairment of everyday functioning; the second part of the questionnaire contains separate subscales for motor and vocal tics. Responses are scored on a scale from 0 to 5. In total, 100 points can be obtained in the Global Ticks Severity Scale. Interpreting the YGTSS, it should be assumed that the higher the score obtained in the study, the greater is the severity of tic disorders, which translates into the daily functioning of the patient. The questionnaire is characterized by good reliability, Cronbach's alpha value ranges from 0.69 to 0.75.

The declarative Questionnaire for Measuring Depression (Kwestionariusz do Pomiaru Depresji – KPD) by Łojek, Stańczak and Wójcik was used to assess the severity of depressive symptoms [23]. The questionnaire consists of 75 statements, which should be addressed by selecting the answer on a 4-point scale. The general result consists of 5 separate scales: cognitive deficits and energy loss (DPUE), thinking about death, pessimism and alienation (MSPA), guilt and anxiety (PWNL), psychosomatic symp-

toms and decrease in interest (OPSZ), self-regulation (SR)). The tool has very good psychometric properties – Cronbach’s alpha ranges from 0.60 to 0.97 for women, and from 0.87 to 0.93 for men.

The Polish adaptation of the Perceived Stigmatization Questionnaire (PSQ) [24] created by Lawrence et. al [25]. The questionnaire consists of 21 statements composed of three subscales: Absence of friendly behavior (AFB), Confused/staring behavior (CSB) and Hostile Behavior (HB). The overall result measures the subjective feeling of stigmatization. Responses are evaluated on the 5-point Likert scale, where 1 means ‘never’ and 5 – ‘always’. The questionnaire is characterized by high reliability – Cronbach’s alpha is 0.93.

The questionnaire on feelings about the body used in the study is a tool constructed by Tomkiewicz [26]. The questions concern the acceptance of biological changes taking place in the body of an adolescent, and the statements included in the questionnaire describe feelings about one’s body. Analysis of the internal consistency of the test, carried out using the Cronbach’s alpha method, indicated an alpha value of 0.89 for all scale items, which proves high reliability of the tool.

Results

The analysis of the results was carried out using the SPSS Statistics 24 software. In order to compare the functioning of the clinical and comparative group, the non-parametric Wilcoxon test was carried out – due to the lack of normal data distribution (Tables 1–3). The analysis of Spearman’s r correlation was used to separate the correlates of the declared depressive symptoms (Table 4).

Table 1. Symptoms of depression in the group of people with Tourette syndrome and the comparative group

		Clinical group		Comparative group		W	Z	p
		M	SD	M	SD			
Symptoms of depression (KPD)	Total	124.00	32.24	82.38	15.06	7	-2.691	0.01**
	DPUE	41.92	11.18	28.15	4.65	6	-2.762	0.01**
	MSPA	26.85	8.52	17.92	4.11	7	-2.510	0.01**
	PWNL	34.15	9.04	22.38	5.01	7	-2.261	0.01**
	OPSZ	21.31	7.54	13.92	3.57	12	-2.350	0.05*
	SR	39.92	6.34	44.38	4.72	24	-1.504	0.13

* $p < 0.05$; ** $p < 0.01$

KPD – Questionnaire for Measuring Depression; DPUE – cognitive deficits and energy loss; MSPA – thinking about death, pessimism and alienation; PWNL – guilt and anxiety; OPSZ – psychosomatic symptoms and decrease in interest; SR – self-regulation

Wilcoxon signed-rank test analysis showed that the declared general degree of depression is significantly higher in the group of people with Tourette syndrome ($Z = -2.691$; $p < 0.01$). Cognitive deficits and energy loss (DPUE) ($Z = -2.762$; $p < 0.01$), thinking about death, pessimism and a sense of alienation (MSPA) ($Z = -2.510$; $p < 0.01$) occur significantly more often in people with TS. The subjects with Tourette syndrome declare a significantly high level of guilt and anxiety (PWNL) ($Z = -2.2261$; $p < 0.01$). In addition, they show a decrease in interests and a significant increase in psychosomatic symptoms (OPSZ) ($Z = -2.350$; $p < 0.05$). There was no statistically significant difference between the clinical and comparative group in self-regulation skills (SR).

Table 2. **Feeling of stigmatization in the group of people with Tourette syndrome and the comparative group**

		Clinical group		Comparative group		W	Z	p
		M	SD	M	SD			
Feeling of stigmatization (PSQ)	Total	2.02	0.40	1.79	0.23	19.50	-1.533	0.13
	AFB	2.35	0.46	2.33	0.45	45	-0.035	0.97
	CSB	2.05	0.69	1.59	0.38	18.50	-1.888	0.05*
	HB	1.52	0.46	1.26	0.31	21	-1.429	0.15

* $p < 0.05$

PSQ – Perceived Stigmatization Questionnaire; AFB – Absence of friendly behavior; CSB – Confused/staring behavior; HB – Hostile Behavior

The general assessment of the feeling of stigmatization does not differ significantly in the group of patients and in the comparative group. An important indicator differentiating the declared stigmatization of people with TS is feeling of embarrassment due to other people's stare/staring behavior (CSB) ($Z = -1.888$; $p < 0.05$). Other subscales: hostile behavior and absence of friendly behaviors did not prove to be differentiating.

Table 3. **Body image in the group of people with Tourette syndrome and the comparative group**

	Clinical group		Comparative group		W	Z	p
	M	SD	M	SD			
Total	37.00	5.81	41.42	4.56	22.5	-1.609	0.11
My body makes me happy	2.54	1.33	2.54	0.97	30	-0.273	0.79
My body is alien to me	4.23	1.17	4.85	0.38	3.5	-1.897	0.05*
I am attracted by my body	3.23	1.36	3.08	1.04	25	-0.262	0.79

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My body scares me	4.08	1.04	4.39	0.87	10	-.686	0.49
I like my body	2.31	1.25	1.85	0.99	16.5	-.720	0.47
I am afraid whether I will like my body in a few years	2.85	1.57	4.00	1.15	11	-1.702	0.09
My body is close to me	1.92	1.12	2.00	1.41	30	-.271	0.79
I hate my body	4.23	1.24	4.77	0.60	4.5	-1.276	0.20
It is difficult for me to understand the changes taking place in my body	3.77	1.24	4.77	0.60	11.5	-1.950	0.05*
My body makes me feel anxious	3.92	1.26	4.69	0.63	4	-1.715	0.09
My body makes me feel ashamed	3.85	1.41	4.54	0.66	14	-1.412	0.16

* $p < 0.05$

The general assessment of one's body image is not significantly different in the group of people with TS and the comparative group. Two differentiating factors were distinguished: "My body is alien to me" ($Z = -1.897$; $p < 0.05$) and "it is difficult for me to understand the changes taking place in my body" ($Z = -1.950$, $p < 0.05$).

Table 4. **The relationship between the severity of depressive symptoms and the severity of tics, the feeling of stigmatization and the body image**

	Total	DPUE	MSPA	PWNL	OPSZ	SR
Severity of tics (YGTSS)	0.81**	0.72**	0.75**	0.65**	0.59*	n.s.
Perception of stigmatization (PSQ) – total	0.63*	0.57*	0.74*	n.s.	0.73**	n.s.
AFB	n.s.	n.s.	n.s.	n.s.	0.57*	n.s.
CSB	0.72**	0.71**	0.64*	n.s.	0.79**	n.s.
HB	n.s.	n.s.	n.s.	n.s.	n.s.	n.s.
Feelings about the body – total	0.64*	n.s.	n.s.	n.s.	n.s.	0.59*
My body makes me happy	n.s.	n.s.	n.s.	0.66*	n.s.	n.s.
My body is alien to me	-0.75**	-0.56*	-0.75**	-0.74**	n.s.	0.64*
I am attracted by my body	n.s.	n.s.	n.s.	n.s.	n.s.	n.s.
My body scares me	-0.71**	n.s.	-0.63*	-0.72**	n.s.	n.s.
I like my body	n.s.	n.s.	n.s.	0.61*	n.s.	n.s.
I am afraid whether I will like my body in a few years	n.s.	n.s.	n.s.	-0.56*	n.s.	n.s.
My body is close to me	n.s.	n.s.	n.s.	0.63*	n.s.	n.s.
I hate my body	-0.57*	n.s.	-0.57*	-0.74**	n.s.	n.s.
It is difficult for me to understand the changes taking place in my body	n.s.	n.s.	n.s.	n.s.	n.s.	0.73**

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My body makes me feel anxious	-0.59*	n.s.	n.s.	n.s.	n.s.	0.58*
My body makes me feel ashamed	n.s.	n.s.	n.s.	n.s.	n.s.	n.s.

* $p < 0.05$; ** $p < 0.01$;

YGTSS – Yale Global Tic Severity Scale; PSQ – Perceived Stigmatization Questionnaire; AFB – Absence of friendly behavior; CSB – Confused/staring behavior; HB – Hostile Behavior; DPUE – cognitive deficits and energy loss; MSPA – thinking about death, pessimism and alienation; PWNL – guilt and anxiety; OPSZ – psychosomatic symptoms and decrease in interest; SR – self-regulation; n.s. – not significant

The presented results indicate moderate or high correlation coefficients between the severity of tics and selected body image indexes, the severity of depressive symptoms and the feeling of stigmatization. Conducted statistical analysis of Spearman's rho shows significant increase in the general symptoms of depression ($r = 0.81$; $p < 0.01$), cognitive deficits and energy decrease (DPUE) ($r = 0.57$; $p < 0.01$), thinking about death, pessimism and alienation (MSPA) ($r = 0.74$; $p < 0.01$), guilt and anxiety (PWNL) ($r = 0.74$, $p < 0.01$), and psychosomatic symptoms and decrease in interests (OPSZ) ($r = 0.59$; $p < 0.05$), with the intensity of tics and the influence of tics on the impairment of daily functioning declared by patients. The higher the overall level of depression, the greater the tendency to describe the body in negative categories: “My body is alien to me” ($r = -0.75$; $p < 0.01$), “My body scares me” ($r = -0.71$; $p < 0.01$), “I hate my body” ($r = -0.57$; $p < 0.05$), “My body makes me feel anxious” ($r = -0.59$; $p < 0.05$).

Discussion

Based on the obtained results, it is concluded that the severity of tics, the feeling of stigmatization and selected body image indexes are related both to the overall severity of depressive symptoms as well as to its individual indicators. People with Tourette syndrome declared a significantly higher level of depression compared to those in the control group, which is consistent with the results obtained in other studies [27]. The applied research method allowed not only to estimate the overall level of depression, but also its individual indicators. Patients declared a decrease in activity and loss of energy, which is associated with general apathy. The subjects have deficits in cognitive functioning and psychosomatic health problems. Difficulties in intellectual capacity may be secondary to affective disorders.

The results of research on cognitive abilities are inconsistent [28–30]. It has been shown that people with TS have difficulties with visual-spatial skills and obtain worse results in the verbal fluency test, which examines the ability to create strategies for searching memory resources.

Organization of activities is one of the aspects of executive functions, i.e., the ability to initiate, inhibit and control the course of action [31]. Patients with Tourette syndrome may have difficulty with the suppression of reaction, which is probably related

to abnormalities in the fronto-striatal circuits. The same mechanism may contribute to an increased risk of other co-occurring disorders, such as obsessive-compulsive disorder [30, 32]. However, it should be born in mind that depressive disorders (especially the so-called major depression) can significantly affect the cognitive functioning of people with TS. Depression can reduce functioning in the field of broadly understood executive functions, concentration of attention and motor speed, and thus areas which are considered the most often disturbed in the course of TS [33]. Patients are more likely to think about death, which is associated with pessimism, or a negative assessment of current situations and their consequences. They report a feeling of alienation, probably due to the stigmatizing character of TS [34].

People with Tourette syndrome may have difficulties in establishing contacts with other people, and school-age children are exposed to violence from peers, which may have serious psychological consequences in the future [35]. In addition, the respondents declare a high level of general guilt. They observe a decrease in their interests. On the other hand, the self-regulation ability is not significantly different among the group of ill and healthy people and even people with significant mood disorders may have a high level of self-regulatory abilities [23].

The occurrence of depressive disorders is found to be associated with the severity of tics. Similar results were obtained in other studies, where the relationship between strength and character of tics, advanced age and coexistence of obsessive-compulsive disorders (OCD) was observed [36, 37]. It has been shown that a significant increase in tics may imply a higher prevalence of anxiety disorders [17, 38]. Studies on a large group of people based on the self-report methodology suggest that in the course of life the prevalence of anxiety disorders is at the level of 36.1%, whereas mood disorders at 29.8% in people with TS [39]. The latest research shows that in people with Tourette Syndrome, generalized anxiety disorder is diagnosed even more frequently than depressive disorders [9].

Treatment in Tourette syndrome is symptomatic and may include pharmacotherapy and psychological interventions, including psychotherapy and psychoeducation. Reports comparing the efficacy of adrenergic agonists with neuroleptics are inconsistent. Depression may be one of the side effects of the medication [40]. It has been shown that the use of dopamine antagonists may increase the risk of depression, which applies to both typical and atypical antipsychotics. The efficacy of atypical (second-generation) neuroleptics, which are generally usually better tolerated, has not been sufficiently proven in the case of tics reduction in TS patients, therefore the use of typical antipsychotics is most often preferred [41]. The relationships between the severity of depressive disorders and the use of: clonidine, haloperidol, sulpiride, tiapride, pimozide or risperidone have been proven [5] – i.e., substances declared to be used by the selected TS patients. The impact of neuroleptics on the mental state of the clinical group has not been analyzed in this study due to the inability of making

a reliable assessment of the effects of the substances, e.g., differences in the dosage of pharmaceuticals, dosage of the active substance and in the duration of administration of the medication.

Depression is diagnosed significantly more frequently in TS patients with other co-morbidities. A study by Sukhodolsky showed no differences between the incidence of depression in the general population and people suffering from the so-called pure form of Tourette syndrome (without co-occurring disorders) [42].

TS is a stigmatizing disorder, because the behavior of the ill can be critically evaluated by outside observers, which is manifested in the significantly high results achieved in the indicator “Confused/staring behavior” [35]. Studies on the image of Tourette syndrome created in the media indicate that the message is usually biased – it shows a false picture of the disease consistent with the common stereotype [43, 44]. Due to the changing nature of tics, patients with TS may have a disturbed sense of control over their own body, which together with a negative social assessment may contribute to the reduction of satisfaction with their body. A study by Schier showed that patients with TS evaluate their satisfaction with their own body at a significantly low level (the body image is perceived as the perceptions, thoughts and feelings about the body) [45]. They more often declare concern and difficulties in accepting changes in the body. In addition, research indicates a relationship between dissatisfaction with the body and depression: disorders of the body image may contribute to the development of affective disorders [16, 46], as well as depression may lead to disorders of the body image. Low acceptance of corporeality, along with anxious-ambivalent attachment style and the occurrence of mental illness in the family, is one of the predictors of the severity of depressive symptoms in women from the period of adolescence to early adulthood [47].

The presented study has some limitations. Due to the difficult access to people with Tourette syndrome, who often do not consent to the examination – probably due to the stigmatizing character of this disorder – 13 persons participated in the research project. Difficulties in recruitment resulted in the heterogeneity of the group in terms of age – two minors, at the threshold to adulthood (17 years of age), were included in the study. This fact may be important in the context of an unambiguous interpretation of the obtained results. The experience of chronic disease, such as TS, and other variables included in the study, can vary in people of different ages. Future researches should take into account more homogeneous selection criteria for the group. The study group is not adequately representative because people with TS belong to the Polish Association of Tourette Syndrome. As members of the association, they receive support, therefore their psychosocial functioning may be significantly higher compared to non-members of the society, which may explain quite high self-regulation skills and lower level of declared victimization. However, despite this, the subjects obtained high scores on the depression scale.

In addition, in the future it is worth to verify the occurrence of anxiety disorders – due to other reports of their significant prevalence among people with TS. Another limitation of the study is the inability to make an analysis of the effects of drugs used by patients – due to a very large variation in the used substances and the different duration of their use. In subsequent studies, a larger and more diverse group of subjects should be considered. The study, however, is exploratory in nature because the relationship between depression, tics and the body image has not been verified so far. In subsequent research projects, it is also worth considering this variable, due to the often disturbed feeling of body control in Tourette syndrome.

Conclusions

1. Patients suffering from TS significantly more often show symptoms of depression.
2. The severity of tics, the feeling of stigmatization, especially the feeling of being observed, and selected indicators of the body image are related to both the general severity of depressive symptoms and its individual indicators.
3. Due to the high risk of depressive disorders in people with Tourette syndrome, it is worth considering the implementation of a prophylactic program, taking into account the psychoeducation of the ill and society – due to the stigmatizing nature of this disorder.

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Address: Agnieszka Małek,
University of Physical Education and Sport in Gdansk
80-336 Gdańsk, Kazimierza Górskiego Street 1
e-mail: agnieszka.malek0@gmail.com