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ORIGINAL CONTRIBUTION

Alexithymia and tic disorders: a study on a sample of children and their mothers

Paola R. Silvestri[1] · Flavia Chiarotti[2] · Sandra Giustini[3] · Francesco Cardona[1]

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Abstract
Tic disorders are neurodevelopmental disorders characterised by the presence of motor or phonic tics, or both. Patients with tic disorders commonly report premonitory urges of tics. Alexithymia is a psychological trait characterised by a difficulty in identifying and expressing one's own feelings and by an externally oriented thinking. We aimed to explore alexithymia in children with tic disorders and in their mothers. Global alexithymia scores of both children with tic disorders and of their mothers did not differ from those of the participants from the control group. In the tic disorder group, however, both children and their mothers showed a cognitive style characterised by operational thinking and a lack of imaginative abilities. The mothers of children with tic disorder reported significantly higher parental stress. Alexithymia was not predictive of tic severity but was predictive of the severity of the premonitory urges. The implications of these findings are discussed.

Keywords Tourette · Tic disorders · Premonitory urges · Alexithymia · Children · Mothers

Introduction

Tic disorders are neurodevelopmental disorders characterised by the presence of sudden, rapid, recurrent, nonrhythmic motor movements (motor tics) or vocalisations (phonic tics). Tourette disorder is diagnosed when multiple motor tics and one or more phonic tics occur, lasting for at least 1 year since the first tic occurred, with an onset before age 18. Chronic motor or vocal tic disorder is diagnosed when one or multiple motor or phonic tics occur (but not both motor and phonic tics), lasting for at least 1 year since the first tic occurred, with an onset before age 18 [1].

Tic disorders typically have an onset at the age of 6–8 years, with an estimated 3–4:1 male-to-female ratio and a prevalence ranging 5–7 cases per 1000 in children aged 6–18 years [2].

Patients often show comorbid mental disorders, mainly attention deficit hyperactivity disorder (ADHD) and obsessive–compulsive disorder (OCD) [3, 4]. Depressive and anxiety disorders are also common in these patients [5, 6].

Patients with tic disorders commonly report premonitory urges of tics. These urges can be located in areas involved in tics or can spread to the whole body. They reach a peak just before the tic onset and are temporary relieved by performing tics [7].

Recently, authors found that interoceptive awareness (i.e., the awareness of internal processes of the body) is a strong predictor of the severity of the premonitory urges [8].

Premonitory urges were reported to be positively related to the activity of the right temporo-parietal junction during a task of social reasoning [9, 10]. Interestingly, the temporo-parietal junction plays a crucial role in processes like multisensory integration, bodily awareness, social cognition and self-other differentiation [11].

The severity of tics is influenced mainly by the severity of the premonitory urges, but emotional states and stress factors also play a role [12].

Alexithymia (from the ancient Greek: "a" for lack, "lexis" for word and "thymos" for emotion, "no words for emotions") is a psychological trait characterised by a difficulty in identifying and expressing one's own feelings and by a cognitive style called "externally oriented thinking" (described

✉ Francesco Cardona
francesco.cardona@uniroma1.it

[1] Department of Human Neurosciences, Sapienza University of Rome, Via dei Sabelli 108, 00185 Rome, Italy

[2] Center for Behavioral Sciences and Mental Health, Istituto Superiore di Sanità, Rome, Italy

[3] Department of Internal Medicine and Medical Specialties, Sapienza University of Rome, Rome, Italy

as an operational thinking style associated with poor imaginative abilities).

In the early 1970s, Nemiah and Sifneos [13, 14] identified this psychological trait in patients with psychosomatic disorders. The role of alexithymia was also highlighted in medical diseases, especially those where the chronic inflammation and the immune dysregulation have a pathogenic role [15–17].

Later, alexithymia was also observed in many mental disorders, such as in anxiety [18, 19], depressive and obsessive–compulsive disorders [20, 21], as well as in eating [22], gambling [23] and substance use disorders.

Alexithymic traits were also found in movement disorders, such as Parkinson's disease [24] and Huntington's disease [25]. In Parkinson's disease, it is noteworthy that alexithymic traits were reported in patients with coexistent depressive disorder and that the severity of the depressive symptoms was predictive of higher levels of alexithymia.

The development of alexithymic traits in adulthood was related to the extent to which emotions were expressed in the family of origin. Authors observed that the alexithymia scores in college students were correlated with their mothers' alexithymia scores and hypothesised an intergenerational transmission of alexithymia [26].

There is increasing evidence that neurobiological influences may play a role in social cognition and in the development of alexithymic traits, and the role of many brain regions in the processing of emotions was recently highlighted. Alexithymia was found to be associated with higher activation in the dorsal anterior cingulate cortex, with a decreased activity of the amygdala, of the supplementary motor, premotor and dorsomedial prefrontal cortices when processing negative emotional stimuli, and with a diminished activity of the right insula and pre-cuneus when processing positive emotional stimuli [27].

To date, only one study investigated the presence of alexithymia in tic disorders. Authors did not find differences in the alexithymia total scores between a group of adults with Tourette syndrome and a control group [28].

To our knowledge, no studies have investigated alexithymia either in children and adolescents with tic disorders or in their families.

Aim of the study

It is well known that tic severity is influenced by the subject's emotional status. Since tics and premonitory urges are highly related to each other, it is likely that the individual's emotional status can have an influence on his/her premonitory urges, which, in turn, can influence tic severity. In the clinical practice with children with tic disorders and their families, a peculiar communication style—that is, rich in details of tics and premonitory urges and lack of the co-occurring emotional dimensions—can be observed.

Rising from these observations, we hypothesised that both children/adolescents with tic disorders and their families can show alexithymic traits, such as the difficulty in distinguish between bodily sensations and associated emotions (which is part of the alexithymic trait *difficulty in identifying feelings*), a difficulty in expressing one's own feelings and a communication style focused on describing symptoms details and lacking of the expression of the individual's emotions.

Therefore, the present study was aimed first to explore alexithymia in children and adolescents with tic disorders (Tourette's disorder or persistent tic disorder) and in their mothers. Moreover, the study aimed to explore the relationship occurring between children's alexithymia levels and the core features of the disorder (severity of tics, intensity of premonitory urges). Finally, the relationship between the mothers' alexithymia levels and both the parental stress and the mothers' perception of the presence of emotional–behavioural symptoms in children was explored.

Methods

This study received approval from the local Ethics Committee of the University Hospital (Policlinico Umberto I, Rome, Italy).

Participants

Forty-five patients (35 males) diagnosed with Tourette's disorder or persistent motor tic disorder according to the DSM 5 criteria [1] were recruited in the tic disorder group among children and adolescents in the outpatient clinic of the Paediatric Neurology Unit, Department of Human Neurosciences, Sapienza University of Rome, Italy. The mothers were also enrolled in the study.

Inclusion criteria included the following: a diagnosis of Tourette's disorder or persistent tic disorder (motor or vocal subtype) and an age ranging between 8 and 13 years. Exclusion criteria included the following: diagnosis of intellectual disability, autism spectrum disorder or schizophrenia spectrum disorder and chronic illnesses, such as diabetes mellitus, epilepsy, renal failure or heart disease.

Forty-five patients (29 males) were recruited in the control group among children visited in the outpatient clinic of the Paediatric Dermatology Unit, Department of Internal Medicine and Medical Specialties, Sapienza University of Rome. Inclusion criteria included the following: accessing the outpatient clinic for regular follow-up of moles in asymptomatic children. Exclusion criteria included the following: diagnosis of tic disorders, intellectual disability, autism spectrum disorder or schizophrenia spectrum

disorder, positive history of tics, chronic illnesses such as diabetes mellitus, epilepsy, renal failure or heart disease. The mothers were also enrolled in the study.

Procedure

Children and their parents were asked to participate in the study at the end of the medical visits and received age-appropriate informative material about the study. Then parents were asked to sign written informed consent for the enrolment of their child in the study, and his/her mother was additionally asked to sign written informed consent of her own enrolment in the study. Children and adolescents were asked to give verbal assent to participate to the study. None of the invited families refused to participate to the study.

Inclusion and exclusion criteria were then reviewed, and demographic information was collected (age of the child and of his/her mother, education of the child and of his/her mother, marital status of the parents of the child). Clinical files of participants with tic disorders were examined to collect information about the main diagnosis and the associated disorders.

Children from the two groups were asked to fill out a questionnaire assessing alexithymia (Alexithymia Questionnaire for Children, AQC) [29]. Their mothers were asked to fill in a questionnaire assessing alexithymia (20-item Toronto Alexithymia Scale, TAS-20) [30] and a questionnaire assessing perceived parental stress (Parenting Stress Index, short form, PSI) [31].

We administered clinical interviews about tics (Yale Global Tic Severity Scale, YGTSS) [32] and obsessive–compulsive symptoms (Children Yale-Brown Obsessive–Compulsive Scale, CY-BOCS) [33] to both children from the tic disorder group and their mothers. These latter were asked to fill in a questionnaire about their child's emotional and behavioural adjustment (Child Behavior Checklist, CBCL) [34]. In the tic disorder group, children were asked to rate their premonitory urges (Premonitory Urges for Tics Scale, PUTS) [7], their anxiety (Multidimensional Anxiety Scale for Children, MASC) [35] and depressive (Children's Depression Inventory, CDI) [36] symptoms.

Psychological tools

- Alexithymia Questionnaire for Children (AQC): A 20-item self-report questionnaire that assesses alexithymia in children aged 8–14 years. The questionnaire has a three-factor structure that explores the dimensions "Difficulty Identifying Feelings (DIF)", "Difficulty Describing Feelings (DDF)" and "Externally Oriented Thinking (EOT)". The agreement with each statement (e.g., "I am often confused about the way I am feeling inside") is rated on a 3-point scale.
- 20-item Toronto Alexithymia Scale (TAS-20): A 20-item self-report questionnaire that assesses alexithymia in adults. The questionnaire has a three-factor structure that explores the dimensions "Difficulty Identifying Feelings (DIF)", "Difficulty Describing Feelings (DDF)" and "Externally Oriented Thinking (EOT)". The agreement with each statement (e.g., "I am often confused about what emotion I am feeling") is rated on a 5-point scale.
- Parenting Stress Index (PSI), short form: A 36-item questionnaire that focuses on three major domains of stress in the parent–child system: parental distress (PD), parent–child dysfunctional interaction (P-CDI) and difficult child (DC).
- Yale Global Tic Severity Scale (YGTSS): A clinician-rated scale that explores number, frequency, intensity, complexity and interference of motor and phonic tics in the week preceding the assessment.
- Children Yale-Brown Obsessive–Compulsive Scale (CY-BOCS): A clinician-rated scale that explores the presence of obsessions and compulsions reported in the last 2 weeks.
- Premonitory Urges for Tics Scale (PUTS): A 10-item self-report questionnaire that measures premonitory urges of tics. The agreement with each statement (e.g., "Right before I do a tic I feel like my insides are itchy") is rated on a 4-point scale.
- Multidimensional Anxiety Scale for Children (MASC): A 39-item self-report questionnaire that assesses anxiety symptoms in children aged 8–19 years.
- Children's Depression Inventory (CDI): A self-report questionnaire that assesses depressive symptoms in children aged 8–17 years.
- Child Behavior Checklist for ages 6–18 (CBCL 6–18): A parent-rated questionnaire that assesses the presence of behavioural and emotional problems in children aged 6–18 years. The score is expressed in eight syndrome scales and two broad scales (Internalizing Problems and Externalizing Problems). The agreement with each statement (e.g., "Act too young for his/her age") is rated on a 3-point scale.

Statistical analyses

Qualitative data are summarised by absolute and percent frequencies and quantitative data by means and standard deviations (SD).

Differences between groups with respect to demographic qualitative variables were tested by the Fisher exact probability test. With respect to quantitative data, these were first tested for normality using the Shapiro–Wilk test, which was separately performed in each subgroup, and for homogeneity of variance between subgroups using the Levene test. Variable distribution did not differ significantly from normality

for most variable/subgroup combinations, and variances were homogeneous across subgroups for all variables. Therefore, differences between groups were assessed by parametric *t* tests (in case of two groups) or analysis of variance (ANOVA, in case of more than two groups). Non-parametric tests (Mann–Whitney *U* test in cases of two groups and Kruskal–Wallis test in cases of more than two groups) were used to validate results of parametric tests when variable distribution was non-normal in at least one subgroup. Since results of non-parametric and parametric tests were in agreement, only parametric test results are reported. Differences were considered significant for $p < 0.05$.

In the tic disorder group, Pearson linear correlation coefficient *r* was used to assess the association between variables. Correlation was considered meaningful for *r* values greater than or equal to 0.30. Multiple regression analysis was performed to assess the effect of TAS 20 and AQC scores on PUTS, PSI, CBCL and YGTSS (Total Tic Score) scores (in separate analyses), adjusting for child gender and mother education. Independent variables were considered significantly affecting the outcome for $p < 0.05$; regression coefficients with 95% confidence intervals are reported.

Statistical analyses were performed using STATA Statistical Software, Release 8.1 [37].

Results

In the tic disorder group, 27 children had been diagnosed with Tourette's disorder (among those, four had received an additional diagnosis of ADHD and two had received an additional diagnosis of OCD) and 18 children had been diagnosed with persistent motor tic disorder (among those, two had received an additional diagnosis of OCD). Three children were taking medications for tics or associate disorders. The severity of motor tics was moderate. The severity of phonic tics was mild as well as global tic severity. Thirty-five participants reported premonitory urges. Almost half of the patients reported mild–moderate anxiety symptoms (see Table 1).

The two groups did not differ significantly for gender, age and education of the participants and for age, education and marital status of their mothers (see Table 2). None of the mothers in the two groups reported positive history of tics.

Parametric *t* tests were performed for between-group comparisons (see Table 3).

Mean total and sub-scores obtained by the two groups of children (tic disorder and control groups) in the AQC were compared. The tic disorder group showed higher scores in the EOT factor of the AQC. No differences were found between the two groups in the total, DIF and DDF scores.

Mean total and sub-scores obtained by the two groups of mothers in the 20-item TAS-20 were compared. In the tic disorder group, the mothers showed higher scores in the EOT factor of the TAS-20. No differences were found between the two groups in the total, DIF and DDF scores.

Table 1 Clinical features of the children with tic disorders

Measures	Mean	Median	SD	Range
Motor YGTSS score	7.96	8	3	0–14 (out of 25)
Phonic YGTSS score	4	4	4.52	0–15 (out of 25)
Total YGTSS Tic Score	11.89	11	5.86	0–29 (out of 50)
PUTS score	15.96	15	6.47	9–33 (out of 36)
Total MASC score[a]	51.24	52	9.7	37–71 (out of 90)
CDI score	8.67	7	6.84	1–31 (out of 52)
Internalizing CBCL score[a]	59.28	61	9.41	34–83 (out of 100)
Externalizing CBCL score[a]	54.97	54	8.72	33–79 (out of 100)
Total CBCL score[a]	57.82	58	9.85	31–76 (out of 100)

YGTSS Yale Global Tic Severity Scale, *PUTS* Premonitory Urges for Tics Scale, *MASC* Multidimensional Anxiety Scale for Children, *CDI* Children's Depression Inventory, *CBCL* Child Behavior Checklist

[a]MASC and CBCL scores are expressed as *T* scores

In the tic disorder group, three mothers fell in the clinical range (total TAS 20 score above 61) and eight fell in the subclinical range (total TAS 20 scores ranging 50–60) for alexithymia, whereas in the control group one mother fell in the clinical range and six fell in the subclinical range for alexithymia.

Mean total and sub-scores obtained by the two groups of mothers in the PIS questionnaire were compared. In the tic disorder group, the mothers showed higher scores in all the sub-scales of the questionnaire.

Linear correlation analyses were performed in the tic disorder group.

No correlations were found between total and subtotal AQC scores and total and subtotal TAS-20 scores. No correlations were found between the Total YGTSS Tic Score and total and subtotal AQC scores or between the Total YGTSS Tic Score and total and subtotal TAS-20 scores.

Moderate direct correlations were found between the DIF score of the AQC and the PUTS score, between the DIF factor of the AQC and the CY-BOCS total score, between the DIF factor of the AQC and the MASC total score, and between the DDF score of the AQC and the CDI total score (see Table 4).

Moderate direct correlations were found between the DIF score of the TAS-20 and PD, P-CDI, DC and total scores of the PSI questionnaire. Moderate direct correlations were also found respectively between the DIF score of the TAS-20 and Internalizing Problems, Externalizing Problems and Total Problems CBCL scores. As expected, high CBCL scores were correlated with high PSI scores (see Table 5).

Table 2 Comparison of age and education between the two groups

Measures	Clinical group			Control group			Statistics	
	Mean	SD	Median	Mean	SD	Median	*t*	*p*
Children's age	10.18	1.72	10	9.84	1.69	10	0.93	0.36
Children's education[a]	4	1.64	4	3.87	1.63	4	0.39	0.70
Mothers' age	43.07	4.59	43	42.89	5.12	43	0.16	0.87
Mothers'education[a]	12.73	2.78	13	13.02	3.18	13	−0.46	0.65

[a]Education is expressed as grades completed

Table 3 Comparison of AQC, TAS-20 and PSI scores between the two groups

Measures	Clinical group			Control group			Statistics	
	Mean	SD	Median	Mean	SD	Median	*t*	*p*
AQC-DIF	11.53	2.91	11	12.18	3.02	12	−1.03	0.3054
AQC-DDF	9.22	2.35	9	9.36	2.49	9	−0.26	0.7946
AQC-EOT	16.31	2.25	17	14.53	2.55	15	3.50	**0.0007**
Total AQC	37.07	5.92	38	36.07	5.23	36	0.85	0.3982
TAS 20-DIF	12.89	5.30	12	12.38	5.64	11	0.44	0.6588
TAS 20-DDF	10.80	4.50	10	10.29	4.12	10	0.56	0.5751
TAS 20-EOT	18.73	6.44	19	16.29	4.98	17	2.015	**0.0469**
Total TAS 20	42.42	13.74	41	38.91	10.19	37	1.38	0.1721
PSI-PD	59.20	27.66	65	42.40	25.23	35	2.96	**0.0039**
PSI-P-CDI	64.89	30.11	70	41.91	27.82	35	3.70	**0.0004**
PSI-DC	73.75	27.83	85	44.86	28.47	40	4.79	**0.0001**
Total PSI	69.20	27.87	75	44.98	26.71	45	4.14	**0.0001**

AQC-DIF Alexithymia Questionnaire for Children-Difficulty Identifying Feelings, *AQC-DDF* Alexithymia Questionnaire for Children-Difficulty Describing Feelings, *AQC-EOT* Alexithymia Questionnaire for Children-Externally Oriented Thinking, *TAS 20-DIF* 20-item Toronto Alexithymia Scale-Difficulty Identifying Feelings, *TAS 20-DDF* 20-item Toronto Alexithymia Scale-Difficulty Describing Feelings, *TAS 20-EOT* 20-item Toronto Alexithymia Scale-Externally Oriented Thinking, *PSI-PD* Parenting Stress Index-Parental Distress, *PSI-PCDI* Parenting Stress Index-Parent–Child Dysfunctional Interaction, *PSI-DC* Parenting Stress Index-Difficult Child

Statistically significant values are in bold

Table 4 Correlations between results of alexithymia questionnaires (AQC and TAS 20) and clinical scales (PUTS, CY-BOCS, MASC, CDI, total YGTSS Tic Score) in children with tic disorders

	PUTS	CY-BOCS	MASC	CDI	YGTSS
TAS 20-DIF	−0.03	−0.15	0.00	−0.13	0.19
TAS 20-DDF	0.12	−0.19	0.03	−0.04	−0.04
TAS 20-EOT	0.06	−0.13	0.05	−0.06	−0.03
Total TAS 20	0.06	−0.18	0.03	−0.09	0.05
AQC-DIF	**0.41** ($p=0.005$)	**0.30** ($p=0.04$)	**0.37** ($p=0.02$)	0.26	0.06
AQC-DDF	0.08	0.21	0.24	**0.39** ($p=0.02$)	−0.00
AQC-EOT	−0.25	0.18	−0.04	0.14	−0.03
Total AQC	0.11	**0.30** ($p=0.04$)	**0.37** ($p=0.02$)	**0.36** ($p=0.04$)	0.04

Correlations are expressed as Pearson's *r* coefficient

PUTS Premonitory Urges for Tics Scale; *CY-BOCS* Children Yale-Brown Obsessive Compulsive Scale (total score); *MASC* Multidimensional Anxiety Scale for Children; *CDI* Children's Depression Inventory; *YGTSS* Yale Global Tic Severity Scale, Total Tic Score; *AQC-DIF* Alexithymia Questionnaire for Children-Difficulty Identifying Feelings; *AQC-DDF* Alexithymia Questionnaire for Children-Difficulty Describing Feelings; *AQC-EOT* Alexithymia Questionnaire for Children-Externally Oriented Thinking; *TAS 20-DIF* 20-item Toronto Alexithymia Scale-Difficulty Identifying Feelings; *TAS 20-DDF*: 20-item Toronto Alexithymia Scale-Difficulty Describing Feelings; *TAS 20-EOT* 20-item Toronto Alexithymia Scale-Externally Oriented Thinking

Statistically significant values are in bold

Table 5 Correlations between results of alexithymia questionnaires (AQC and TAS 20) and parent-rated questionnaires (PSI and CBCL) in children with tic disorders

	PSI PD	PSI PCDI	PSI DC	PSI TOT	CBCL INT	CBCL EXT	Total CBCL
TAS 20-DIF	**0.35**	**0.33**	**0.35**	**0.37**	**0.45**	**0.38**	**0.43**
TAS 20-DDF	0.25	0.28	**0.32**	**0.34**	0.05	−0.03	−0.05
TAS 20-EOT	0.2	0.13	0.05	0.19	0.09	0.07	0.04
Total TAS 20	**0.31**	0.28	0.26	**0.34**	0.23	0.17	0.16
AQC-DIF	0.27	0.01	0.08	0.17	0.03	0.17	0.14
AQC-DDF	0	−0.23	−0.09	−0.14	−0.1	−0.14	−0.14
AQC-EOT	−0.13	−0.15	−0.22	−0.2	−0.18	−0.29	−0.22
Total AQC	0.03	−0.17	−0.11	−0.09	−0.13	−0.19	−0.15
PSI-PD					**0.34**	**0.45**	**0.47**
PSI-PCDI					**0.33**	**0.48**	**0.5**
PSI-DC					**0.33**	**0.52**	**0.51**
Total PSI					**0.37**	**0.56**	**0.54**

Correlations are expressed as Pearson's *r* coefficient

TAS 20-DIF 20-item Toronto Alexithymia Scale-Difficulty Identifying Feelings, *TAS 20-DDF* 20-item Toronto Alexithymia Scale-Difficulty Describing Feelings, *TAS 20-EOT* 20-item Toronto Alexithymia Scale-Externally Oriented Thinking, *AQC-DIF* Alexithymia Questionnaire for Children-Difficulty Identifying Feelings, *AQC-DDF* Alexithymia Questionnaire for Children-Difficulty Describing Feelings, *AQC-EOT* Alexithymia Questionnaire for Children-Externally Oriented Thinking, *PSI-PD* Parenting Stress Index-Parental Distress, *PSI-P-CDI* Parenting Stress Index-Parent–Child Dysfunctional Interaction, *PSI-DC* Parenting Stress Index-Difficult Child, *CBCL-INT* Child Behavior Checklist-Internalizing Problems, *CBCL-EXT* Child Behavior Checklist-Externalizing Problems

Statistically significant values are in bold

Multiple regression analyses were performed in the tic disorder group (see Table 6).

The DIF score of the AQC was predictive of the PUTS score, adjusting for the gender of the child, mothers' education, the PSI total score and the DIF score of the TAS-20.

The DIF score of the TAS-20 was predictive of the PSI total score, adjusting for the gender of the child, tic severity, mothers' education and the DIF score of the AQC. The DIF score of the TAS-20 was also predictive of Internalizing Problems CBCL scores, adjusting for the gender of the child, mothers' education, tic severity and the DIF score of the AQC.

Neither the total TAS-20 score nor the total AQC score was predictive of the tic severity (Total YGTSS Tic Score), adjusting for the gender of the child and mothers' education.

Discussion

To our knowledge, this is the first study aimed at exploring alexithymia in children with tic disorders, as well as alexithymic traits in their mothers.

Comparing our findings with those from the one study that focused on alexithymia in adults with Tourette syndrome [28], the present study confirmed that global alexithymia scores of the participants with tic disorders did not differ from those of the control group in the developmental age, too. In addition, the global alexithymia scores of the mothers from the tic disorder group did not differ from those of the mothers from the control group.

The differences between the two groups in the EOT factor of the AQC and of the TAS-20 scales was highly significant, however, the difference between the two groups of mothers was still significant but close to borderline significant. This finding suggests that a peculiar cognitive style—an operational thinking style and poor imaginative abilities—is common to both children with tic disorders and their mothers. A possible explanation of this finding is that an intergenerational transmission of alexithymic dimensions may occur in these families, as suggested by Fukunishi and Paris [26] when they observed that the alexithymia scores in college students were correlated with their mothers' alexithymia scores. Moreover, data from the literature suggest a correlation between developing alexithymic traits in adulthood and the extent to which emotions were expressed in the family of origin, leading to the hypothesis that alexithymia is the result of a dysfunctional model of expressing emotions learned during infancy [38–40].

In the tic disorder group, the mothers reported significantly higher parental stress when compared with the mothers in the control group. This finding can easily be explained by the observation that tic disorders have a heavy impact on the child's family members. Because these disorders are chronic, their prognosis is uncertain and often causes stigmatisation. However, our data also showed that the score obtained by the mothers of the children with tic disorders in the DIF factor of the TAS-20 was predictive of the mothers'

Table 6 Linear multiple regression analyses between results of alexithymia questionnaires (AQC and TAS 20), PUTS and parent-rated questionnaires (PSI and CBCL) in children with tic disorders

	PUTS				Total PSI				CBCL INT			
	Regression coefficient	95% CI	Beta	*p*	Regression coefficient	95% CI	Beta	*p*	Regression coefficient	95% CI	Beta	*p*
TAS 20-DIF	−0.12	−0.51 to 0.27	−0.10	0.533	**1.60**	**0.00 to 3.20**	**0.31**	**0.050**	**0.75**	**0.20 to 1.31**	**0.42**	**0.009**
AQC-DIF	**0.87**	**0.18 to 1.56**	**0.39**	**0.014**	1.04	−1.87 to 3.95	0.11	0.474	−0.26	−1.29 to 0.77	−0.08	0.609
Mothers' education	−0.01	−0.70 to 0.68	−0.01	0.973	−1.07	−4.02 to 1.87	−0.11	0.465	0.01	−1.01 to 1.04	0.01	0.977
Child gender	1.22	−3.40 to 5.84	0.08	0.596	6.04	−13.85 to 25.93	0.09	0.542	4.76	−1.66 to 11.18	0.22	0.141
Total PSI	0.03	−0.05 to 0.11	0.12	0.450								
Total YGTSS Tic Score					0.56	−0.89 to 2.00	0.12	0.440	0.30	−0.21 to 0.80	0.18	0.240

TAS 20-DIF 20-item Toronto Alexithymia Scale-Difficulty Identifying Feelings, *AQC-DIF* Alexithymia Questionnaire for Children-Difficulty Identifying Feelings, *PSI* Parenting Stress Index, *YGTSS* Yale Global Tic Severity Scale, *PUTS* Premonitory Urges for Tics Scale, *CBCL-INT* Child Behavior Checklist-Internalizing Problems

Statistically significant values are in bold

parental stress. This finding may be suggestive of a greater tendency toward complaining of parental stress in those mothers whose abilities to get in touch with their own feelings are lacking. Previous studies have widely highlighted the relation occurring between alexithymia and chronic stress [16].

Alexithymia was not found to be predictive of tic severity. However, the score obtained by the children in the DIF factor of the AQC scale was predictive of the severity of the premonitory urges, suggesting that the difficulty in getting in touch with one's own feelings can directly determine how strongly the premonitory urges of tics are perceived and can perhaps indirectly influence the global severity of the tic disorder through the role that premonitory urges play in triggering tics.

A study by Ganos et al. [8] showed that interoceptive awareness is highly predictive of the severity of premonitory urges in Tourette patients. Therefore, we hypothesise that the tendencies of Tourette patients to pay attention to interoceptive stimuli may be part of a greater difficulty in distinguish between emotions and bodily sensations that is typical of alexithymic individuals. If this hypothesis is true, the study by Ganos indirectly proves the correlation we highlighted between the difficulty in identifying feelings (which includes also the difficulty in distinguish between emotions and bodily sensations) and the premonitory urges.

However, since the cross-sectional design of the study does not allow to define the direction of the association between alexithymia and premonitory urges, other hypotheses should be taken into account.

Deficits in social cognition have been highlighted in TS individuals, and tics are viewed as an effort to cope with emotional reactivity and with dysfunctional social cognition.

A reduced ability to take other people's perspectives and an elevated personal distress when interacting with people in emotional crises was found in subjects with TS. Tic severity was negatively related to personal distress ratings, indicating that tics and emotion regulation are inversely associated each other [41]. The authors suggested two possible explanations of this finding: either that TS patients with more severe tics feel more frequently uncomfortable in social situations and consequently cope with unpleasant emotional experiences becoming less emotionally sensitive, or that, since TS subjects are prone to emotional dysregulation, they may act tics in the effort to relieve their inner tension [41].

In another paper from the same author, the reduced sense of agency is thought to lead TS subjects to act tics as a conditioned response to regain internal control and self-other distinction [42].

In the tic disorder group, the DIF score of the AQC scale correlated with the severity of the obsessive–compulsive and anxiety symptoms. Moreover, the DDF score of the AQC correlated with the severity of the depressive symptoms.

These results are in line with data from the literature: indeed, alexithymic traits have already been described in anxiety [18, 19], depressive and obsessive–compulsive disorders [20, 21].

However, we confirmed that the relationship between alexithymic traits and the dimensions of anxiety, depression and OC behaviours exists also in TS patients.

We suggest two possible explanations of the correlation between alexithymic traits and anxiety, depressive and OC symptoms: either the comorbid disorders/symptoms enhance the levels of alexithymia in TS patients, or the TS patients' alexithymic traits act as risk factors for developing other comorbid disorders/symptoms.

The existing literature about the relationship between alexithymia and depression supports both hypotheses.

Allen et al. [43] found that the relationship between the two subscales of alexithymia (DIF and DDF) and somatization was significantly mediated by depression in a sample of healthy children.

On the other side, Rieffe et al. [44] explored the best fitting model of the relationship between alexithymia, mood and internalizing symptoms (worry/ruminations and somatization) in a sample of children. They found that somatization shared an independent relationship with alexithymia.

Lumley [45] commented on the relationship between alexithymia, anxiety and depression and suggested that, since alexithymia has been defined as a deficit in regulating emotions through cognitive strategies [46], this deficit enhances the risk of developing a broad range of mental disorders related to poor affect regulation, including mood and anxiety disorders.

In the tic disorder group, the score obtained by the mothers in the DIF factor was predictive of their perception of the presence of behavioural and emotional problems in their children, suggesting a greater tendency toward reporting his/her child's behaviour as dysfunctional in those mothers whose abilities to get in touch with their own feelings are poor. Another possible explanation is that the children of the mothers with alexithymic traits can show more dysfunctional behaviours when compared to the children of mothers without those traits.

Limitations of this study come mainly from the use of self-report questionnaires to assess alexithymia rather than using clinician-rated interviews. Data from the literature have shown that the TAS-20 can underestimate alexithymic traits when compared to a clinician-rated interview in parents of children with learning disorders [47] and in parents of girls with eating disorders [48].

Further limitations also come from the cross-sectional design of the study, that does not allow to ultimately define the direction of the relationships highlighted from the data analyses.

It is also important to note that the reliability of the EOT subscale of the TAS-20 and the AQC scales was found to be low and decreasing with age [49]. Some authors also found that the two-factor structure of those scales, without the EOT subscale, showed a better fit compared to the three-factor structure [50].

Male gender was found to be linked to alexithymic traits [51]. This has to be taken into account when interpreting the results of this study, since our sample was composed mainly by boys with tic disorders and only by mothers: the extent to which the male gender could have influenced the levels of alexithymia in the sample of children with tic disorders is difficult to define.

Future studies that include clinician-rated interviews in the assessment of alexithymia are needed.

Conclusions

Our data suggest that school-age children with tic disorders and their mothers show a peculiar cognitive style characterised by an operational thinking style and poor imaginative abilities.

The difficulty in identifying feelings—a core feature of the alexithymic construct—seems to influence the severity of the premonitory urges, which, in turn, is a key determinant of the severity of tics. Taking these findings into account can be helpful in the clinical practice with children with tic disorders and with their families.

Compliance with ethical standards

Conflict of interest On behalf of all authors, the corresponding author states that there is no conflict of interest.

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