

8-30-2016

Subjective versus Objective Measures of Tic Severity in Tourette Syndrome – The Influence of Environment

Meirav Barnea
Bar-Ilan University, Israel

Noa Benaroya-Milshtein
Tel Aviv University

Eva Gilboa-Sechtman
Bar-Ilan University, Israel

Douglas W. Woods
Marquette University, douglas.woods@marquette.edu

John Piacentini
University of California, Los Angeles

See next page for additional authors

Accepted version. *Psychiatry Research*, Vol. 242 (August 30, 2016): 204-209. DOI. © 2016 Elsevier.

Used with permission.

Douglas W. Woods was affiliated with Texas A&M University at the time of publication.

Authors

Meirav Barnea, Noa Benaroya-Milshtein, Eva Gilboa-Sechtman, Douglas W. Woods, John Piacentini, Silvana Fennig, Alan Apter, and Tamar Steinberg

Psychology Faculty Research and Publications/College of Arts and Sciences

This paper is NOT THE PUBLISHED VERSION; but the author’s final, peer-reviewed manuscript. The published version may be accessed by following the link in the citation below.

Psychiatry Research, Vol. 242, (2016): 204-209. [DOI](#). This article is © Elsevier and permission has been granted for this version to appear in [e-Publications@Marquette](#). Elsevier does not grant permission for this article to be further copied/distributed or hosted elsewhere without the express permission from Elsevier.

Contents

Abstract	3
Keywords	3
1. Introduction.....	3
2. Methods	4
2.1. Sample	4
2.2. Instruments	4
2.2.1. Effect of environment on tic expression	4
2.2.2. Presence and characteristics of tics	5
2.2.3. Co-morbidities	5
2.2.4. Inclusion criterion.....	6
2.3. Procedures and video measures	7
2.4. Data collection and analysis	7
3. Results	8
3.1. Prediction of tic expression by subjective measures	8
3.2. Prediction of tic expression in environmental situations by the FAI, child and parent reports.....	9
3.3. Influence of the environment on tic characteristics	11
4. Discussion	12
Acknowledgment.....	13
References	13

Subjective versus objective measures of tic severity in Tourette syndrome – The influence of environment

Meirav Barnea

The Harry Freund Neuro-Psychiatric Clinic, Schneider's Children's Medical Center of Israel, Petach Tikva, Israel

Bar-Ilan University, Ramat Gan, Israel

Noa Benaryoa-Milshtein

The Harry Freund Neuro-Psychiatric Clinic, Schneider's Children's Medical Center of Israel, Petach Tikva, Israel

Sackler Faculty of Medicine, Tel Aviv University, Tel Aviv, Israel

Eva Gilnoa-Sechtman

Bar-Ilan University, Ramat Gan, Israel

Douglas W. Woods

Texas A&M University, Commerce, TX

John Piacentini

University of California-Los Angeles, Los Angeles, CA

Silvana Fennig

The Harry Freund Neuro-Psychiatric Clinic, Schneider's Children's Medical Center of Israel, Petach Tikva, Israel

Alan Apter

The Harry Freund Neuro-Psychiatric Clinic, Schneider's Children's Medical Center of Israel, Petach Tikva, Israel

Tamar Steinberg

The Harry Freund Neuro-Psychiatric Clinic, Schneider's Children's Medical Center of Israel, Petach Tikva, Israel

Abstract

The objective of this study was to examine the influence of environmental challenges on tic expression by subjective and objective measures. The study group consisted of 41 children aged 6–18 years ($M=10.15$, $SD=2.73$) with a primary diagnosis of Tourette syndrome. Subjective measures included the Functional Assessment Interview developed for this study and three standard validated instruments. The objective measure was a video-recording of the patients in five daily-life situations: watching television, doing homework, being alone, receiving attention when ticcing, and talking to a stranger. In addition, the effect of premonitory urges on assessment of tic expression was evaluated. The associations between the subjective and objective measures of tic expression were moderate to low. A significantly higher number of tics were observed in the television situation, and a significantly lower number in the alone situation, compared to the other situations. Higher levels of premonitory urge were associated with greater awareness of objectively measured tic expression. In conclusion, tic expression is significantly influenced by the environment. Subjective measures of tic expression may be misleading. These results have implications for refining the clinical assessment of tics, improving research methodology, and developing new therapeutic strategies.

Keywords

Premonitory urge, Video-recording, Self-report, Tic expression, Environmental situations

1. Introduction

Tourette syndrome, a relatively common neuropsychiatric tic disorder, serves as a model for understanding brain-environment interactions (Murphy and Eddy, 2013). Although tics are an observable motor phenomenon driven by disturbances in underlying brain circuitry, they are accompanied by subjective sensory and psychological manifestations and are highly sensitive to environmental influences (Conelea and Woods, 2008). Apart from their theoretical implications, these factors are important considerations in the clinical assessment. In addition to history, comorbidities, and observation, tic intensity, frequency, and impact must be precisely calibrated for clinicians to fully understand and appropriately treat the disorder (Woods et al., 2007). However, most of the currently available instruments for quantifying tic characteristics consist of subjective self-report questionnaires, parent- and teacher-rated scales, and semi-structured clinician-administered interviews (Cavanna and Piedad, 2013).

Several groups have attempted to develop more objective tools, usually based on video-recordings (Himle et al., 2006, Pappert et al., 2003), but these are limited to measurements of tic expression, without accounting for sensory or psychological phenomena. In addition, technical and logistic issues prevent their widespread use, and video-recording itself have been found to influence tic expression (Piacentini et al., 2006). It is also noteworthy that the role of premonitory urges in tic expression has hardly been addressed. Theoretically, because premonitory urges predict tic occurrence, patients with a greater awareness of urges would be expected to display greater awareness of tics, making their subjective reporting more accurate.

The sensitivity of tic expression to environmental influences may be related to both internal and external factors and to various types and levels of activity. To identify and analyze the conditions under which tic expression increases, and thereby achieve better behavioral management of the disorder, clinicians use functional assessment procedures (Gresham et al., 2001, Piacentini et al., 2010). However, function-based assessments harbor methodological problems. Most of the research so far has been done on small samples or in artificial environments unrelated to the patient's daily life. In addition, few efforts have been made to understand the mechanisms underlying the effect of the environment on tic expression, apart from those related to learning theory and broad emotional effects (Conelea and Woods, 2008). One plausible theory is that an environment with highly stimulating properties can interfere with motor inhibition and thereby exacerbate tic expression (Belluscio et al., 2011).

The aim of the present study was fourfold: 1. To test the ability of common subjective measures of tics to predict objective tic expression; 2a. To test the ability of the Functional Assessment Interview (FAI), a semi-structured instrument designed by our group, to systematically assess the influence of environmental situations on tic levels 2.b To test the FAI validity, by examining its ability to predict tic expression across research situations; 3. To examine the effect of premonitory urges on the associations between subjective and objective measures of tic expression; and 4. To compare the effect of different daily-life situations with different levels of stimulation on tic expression.

The following hypotheses were tested: 1. Subjective measures of tic expression accurately predict objective recordings of tic activity; 2a. Children and parents report different levels of tic expression in different environmental situations; 2b. The FAI is an accurate measure of expected tic expression in different environmental situations; 4. The intensity of premonitory urges affects the strength of the association between patient self-reports and objective assessments of tics; 5. Tic expression differs by type and degree of environmental stimulation.

2. Methods

2.1. Sample

Forty-one children and adolescents with a primary diagnosis of Tourette syndrome participated in the study. All participants were patients in a child and adolescent Tourette syndrome clinic at a university-affiliated children's hospital. Consecutive admissions to the clinic were recruited to the study, and when both parent and child gave permission, they were admitted to the study. Children with IQ less than 80 were excluded ($M=102.48$, $SD=18.87$).

2.2. Instruments

2.2.1. Effect of environment on tic expression

2.2.1.1. *Functional Assessment Interview (FAI)*

The FAI is a semi-structured clinical interview constructed specifically for the present study. It was designed to identify the effect of five common, daily-life environmental challenges on tic expression: being alone, watching television, doing homework, talking to a stranger, receiving attention when ticcing. All these situations have a precedent in the literature (Conelea and Woods, 2008) and are relatively easy to simulate in the laboratory. Children and parents (separately) rate the frequency of

ticcing experienced when each situation is encountered in everyday life on a scale of 1 (low frequency) to 4 (high frequency).

2.2.2. Presence and characteristics of tics

2.2.2.1. Yale Global Tic Severity Scale (YGTSS) (Leckman et al., 1989)

The YGTSS is a clinician-rated instrument designed to assess various tic characteristics (severity, number, frequency, intensity, interference, and complexity), each scored on a 5- point scale. Separate scores are obtained for vocal tics and motor tics, and these are summed to yield a total severity score (range 0–50). The scale also includes an impairment rating, scored on a 5-point scale (range 0–50). The YGTSS has demonstrated excellent psychometric properties with solid internal consistency, inter-rater reliability, and convergent and divergent validity (Leckman et al., 1989, Storch et al., 2005). The internal reliability in this study (Cronbach's α) was 0.86 for the motor scale and 0.90 for the vocal scale.

2.2.2.2. Parent Tic Questionnaire (PTQ) (Chang et al., 2009; Woods et al., 2007)

The PTQ is a parent-report questionnaire designed to assess the presence of 14 common motor and vocal tics, in addition to their frequency (1=weekly, 2=daily, 3=hourly, 4=constantly) and intensity (1=lowest intensity, 4=highest intensity). The intensity and frequency scores for each item are summed, yielding a total score of 0 (no tics) to 8 (constant and intense tics). Motor and vocal tics may be evaluated separately, and the scores summed for a total score. The PTQ was found to have excellent validity against other tic-severity measures (Cavanna and Piedad, 2013).

2.2.2.3. Tourette Syndrome Clinical Global Impression (TS-CGI) (Leckman et al., 1988)

The TS-CGI is a clinician-rated 5- item scale designed to assess the severity of Tourette syndrome, obsessive-compulsive disorder (OCD), anxiety disorder, depression, and attention deficit-hyperactivity disorder (ADHD). Each item is scored on an ordinal scale from 1 (normal) to 7 (very severe); a score of 4 or more indicates a significant clinical disorder. The TS-CGI shown excellent reliability and validity (Walkup et al., 1992) and has been widely used (Kwon et al., 2011, Pringshem and Steeves, 2011).

2.2.2.4. Premonitory Urge for Tics Scale (PUTS) (Woods et al., 2005)

The PUTS is a 9- item self-report questionnaire designed to assess the presence and frequency of premonitory sensory urges. Each item is rated on an ordinal scale from 1 (not at all true) to 4 (very much true); the total score ranges from 9 to 36. The PUTS has demonstrated high internal consistency and convergent validity (Woods et al., 2005). The Hebrew version was found to have good psychometric properties (Steinberg et al., 2010). The internal reliability in this study (Cronbach's α) was 0.83.

2.2.3. Co-morbidities

2.2.3.1. The Children's Yale Brown Obsessive Compulsive Scale (CYBOCS) (Scahill et al., 1997)

The CYBOCS is a clinician-administered semi-structured interview for the assessment of the severity of obsessive-compulsive symptoms in children. The contents of the obsessions and compulsions are divided into 5 sub-domains, each rated on a scale from 0 (none) to 4 (severe): duration of the

symptoms throughout the day, symptom-induced interference, level of stress, level of resistance, and control. Separate scores are obtained for obsessions and compulsions; their sum yields a total severity score (range 0–40); 16 is the cutoff total score for a diagnosis of OCD. The CYBOCS has demonstrated acceptable internal consistency and convergent validity (Gallant et al., 2008, Storch et al., 2006). The internal reliability in this study (Cronbach's α) was 0.89 for the obsession scale, 0.93 for the compulsion scale, and 0.93 for the total score.

2.2.3.2. Screen for Child Anxiety Related Emotional Disorders (SCARED) (Birmaher et al., 1997)

The SCARED is a self-report scale for children with anxiety disorders. It contains 38 items describing different emotions and behaviors. Children rate the frequency of each on a 3-point scale from 0 (never) to 2 (often). The sum of all items yields a total anxiety score; 25 is the cutoff total score for a diagnosis of anxiety disorder. The SCARED has demonstrated acceptable internal and test-retest consistency and divergent validity (Birmaher et al., 1999, Rassin et al., 2000). The internal reliability in this study (Cronbach's α) was 0.93.

2.2.3.3. Child Depression Inventory (CDI) (Kovacs, 1982, 1985)

The CDI is a 27-item self-report scale that measures the emotional, cognitive, behavioral, and somatic symptoms that characterize depression in children and adolescents. Children rate each item from 0 to 2, with scores corresponding to their selection of the particular statement among those listed that describes their behavior or feeling. The sum of scores for all items yields a total severity score. The CDI is widely used, and has been proposed as the tool of choice for the assessment of depression in children and adolescents with Tourette syndrome (Robertson and Orth, 2006). The internal reliability in this study (Cronbach's α) was 0.81.

2.2.3.4. Conners' Parent Rating Scale IV (CPRS) (Conners et al., 1998)

The CPRS is a 10-item scale designed to assess the presence of ADHD. Each item contains a behavioral description, and parents select the degree to which the behavior characterizes their child on a scale from 0 (never) to 3 (frequently). The sum of the items yields a total severity score (range 0–30); a total score of 15 is the cutoff for a diagnosis of ADHD. The internal reliability in this study (Cronbach's α) was 0.88.

2.2.4. Inclusion criterion

2.2.4.1. Wechsler Abbreviated Scale of Intelligence (WASI) (Psychological Corporation, 1999)

The WASI is a measure of intellectual functioning for use in individuals aged 6–89 years. It is based on the Wechsler Intelligence Scale and includes 4 of its subscales (Matrix reasoning, Similarities, Vocabulary, and Block design). These subscales were taken from the Hebrew adaptation of Wechsler Intelligence Scale for Children (WISC IV^{HEB}) which was standardized in Israel by *PsychTech*[®]. The mean score is 100, with a standard deviation of 15.

2.3. Procedures and video measures

The study was approved by the hospital's ethics committee, and performed in accordance with the Declaration of Helsinki. Written informed consent was obtained from each child and his parent after the nature of the procedures had been fully explained.

Each study participant attended two separate 3-hour sessions over the course of one week. During the initial session, children and parents completed the comprehensive diagnostic/assessment battery and the children were videotaped, with their knowledge, alone in a room for 10 min to accustomize themselves to the camera and to view a videotaped sample of their tics. They were instructed to sit in a chair facing the camera, to remain seated without covering their face, and to tic as much or as little as needed.

The second session included the experimental procedure, in which the child was videotaped under the five environmental situations presented in the FAI: being alone, doing homework, watching television, talking to a stranger, and receiving attention when ticcing. Each situation was filmed twice, for 10 min each, in random order, for a total of 100 min of film for each child. Like in the test session, children were asked to sit in a chair facing the camera, to remain seated without covering their face, and to feel free to tic as much or as little as needed.

During the alone condition, children were recorded while seated in a room by themselves; during the homework condition, they were seated by themselves next to a table and asked to do a simple age-appropriate academic exercise (to read a short story and answer five open questions about it); during the television condition, children were seated by themselves in front of a television playing a popular age-appropriate program; during the stranger condition, children were seated in a room with a research assistant and instructed to talk about anything but tics (in order to neutralize the effect that a tic-related conversation might have on tic frequency (Woods et al., 2001)); during the feedback condition, children were seated in a room with the experimenter who talked about anything but tics and signaled to them, with their pre-knowledge, each time a tic occurred.

The level of stimulation inherent to each of the situations was scored subjectively before onset of the study by four independent judges blinded to the study hypotheses. Inter-rater agreement, measured with the interclass coefficient (ICC) was 75% before discussion and 100% after.

Following the first session, a list of operationally defined tics was created for each child. The videotapes were scored for the presence of tics using an event-recording method. Each segment was scored separately for motor tics, vocal tics, and total tics using the Multi-Option Observation System for Experimental Studies (MOOSES), a computer program capable of recording and time-stamping computer key strokes (Tapp et al., 1995). The number of tics in each condition was calculated as the mean of the two videotaped segments. Scoring was done by research assistants trained in direct observation.

2.4. Data collection and analysis

Multiple regression analyses were conducted to assess the ability of the subjective measures to predict tic number on the video-recordings. Hierarchy regression analyses were used to assess the influence of

premonitory urges. Repeated-measures analyses of variance (ANOVAs) were used to test differences in tic number among the five research situations. Of the total 41 videotapes obtained (100 min each), 10 (24.4%) were scored by two coders, and inter-rater reliability was determined by ICC analysis. Pearson's correlation was used to determine test-retest reliability.

3. Results

The study group consisted of 34 boys (83%) and 7 girls (17%); 20 (58%) were regularly taking a steady dose of anti-tic medication or stimulants. Common comorbidities were ADHD (48.8%), anxiety disorders (26.8%), and OCD (14.6%). Corresponding mean scores on the co-morbidity measures were as follows: CPRS, 13.37 (SD 5.68); SCARED, 8.43 (SD 5.68); CYBOCS total scale, 4.19 (SD 4.62) obsession scale, 5.34 (SD 5.61) compulsion scale, 9.73 (SD 9.23). Mean score on the CDI was 8.43 (SD 5.68).

3.1. Prediction of tic expression by subjective measures

A series of multiple regression analyses were performed with scores on the PTQ, YGTSS, and CGI as the predictor variables, and tic expression (motor and vocal separately) in the five situations served as the outcome measure. The results are shown in Table 1.

Table 1. Standardized regression coefficients (beta) from a series of multiple regression analysis predicting tic expression for different subjective measures (N=41).

	TVm	TVv	Fm	Fv	Sm	Sv	HWm	HWv	Am	Av
YGTSSm	0.23	0.08	0.09	-0.53 [†]	0.14	-0.47 [†]	0.13	0.05	0.10	-0.04
YGTSSv	0.36 [*]	0.35 [*]	0.13	0.44 [†]	0.05	0.39 [*]	0.01	0.54 [‡]	0.07	0.30
PTQm	-0.15	-0.31	0.11	0.00	0.15	-0.11	0.08	-0.33 [*]	0.37 [*]	0.14
PTQv	0.01	0.12	-0.40 [*]	-0.21	-0.27	-0.01	0.10	-0.25	0.01	-0.17
CGI	0.14	0.40 [†]	0.41 [*]	0.60 [‡]	0.19	0.37 [*]	0.39 [*]	0.44 [†]	0.12	0.21
R²	0.27 [†]	0.42 [‡]	0.27 [*]	35.0 [‡]	0.14	0.25 [*]	0.36 [‡]	0.42 [‡]	0.31 [†]	0.17

TV=watching television; F=receiving attention when ticcing; S=talking with a stranger; HW=doing homework; A=being alone; m=motor; v=vocal.

YGTSS=Yale Global Tic Severity Scale; PTQ=Parent Tic Questionnaire; CGI=Clinical Global Impression.

[†] $p < 0.05$.

^{*} $p < 0.10$.

[‡] $p < 0.01$.

The PTQ motor scale (M=21.95, SD=17.76) nearly predicted objective motor tic expression in the alone situation, but not in any of the other situations. Moreover, the PTQ motor scale was *inversely* related to tic expression in the homework situation, and the PTQ vocal scale (M=12.03, SD=14.54) was inversely related to tic expression in the feedback situation.

The YGTSS motor scale (M=13.84, SD=4.76) was a poor predictor of motor tic frequency in all environmental situations. Moreover, the motor scale score was *inversely* related to vocal tic number in the feedback and stranger situations. However, the YGTSS vocal scale (M=9.12, SD=5.31) predicted the number of vocal tics in the feedback and homework situations with statistical significance, and in the television and stranger situations with near-statistical significance. The YGTSS vocal scale also predicted the number of motor tics in the television situation.

The CGI (M=4.17, SD=1.28) was the most efficient predictor of objective tic expression, significantly predicting total tic expression in the feedback and homework situations, and vocal tic expression in the television, feedback, homework, and stranger situations.

On multiple regression analysis of the ability of the frequency subscales of the YGTSS and PTQ to predict objective tic frequency, none of the findings was statistically significant. To analyze the influence of the strength of premonitory urge on tic prediction, we performed a series of multiple regressions with tic frequency as the outcome measure. The first step included the scores of the individual subjective measures (PTQ, YGTSS, CGI), and the second step included the interactions of the subjective measures with the PUTS scale. There were no significant effects of any of the predictive variables on the outcome measure.

3.2. Prediction of tic expression in environmental situations by the FAI, child and parent reports

For repeated-measures ANOVA, we employed the Greenhouse-Geisser correction for degrees of freedom because the preliminary analysis showed that the data violated the sphericity assumption. As shown in Fig. 1, tic level differed among the different environmental situations [$F(3.36, 134.36)=4.58$, $p<0.01$, $\eta p^2=0.01$]. On post hoc Bonferroni test for multiple comparisons, tic expression was rated higher in the feedback situation than in the television, homework, and stranger situations. It was also rated higher in the alone than the homework situation. There was a main effect of responder identity ($F(1,40)=10.77$, $p<0.01$, $\eta p^2=0.21$), with parents reporting higher tic levels than children (M=2.57, SE=0.06; M=2.29, SE=0.08, respectively). There was no interaction between responder identity and environmental situation.

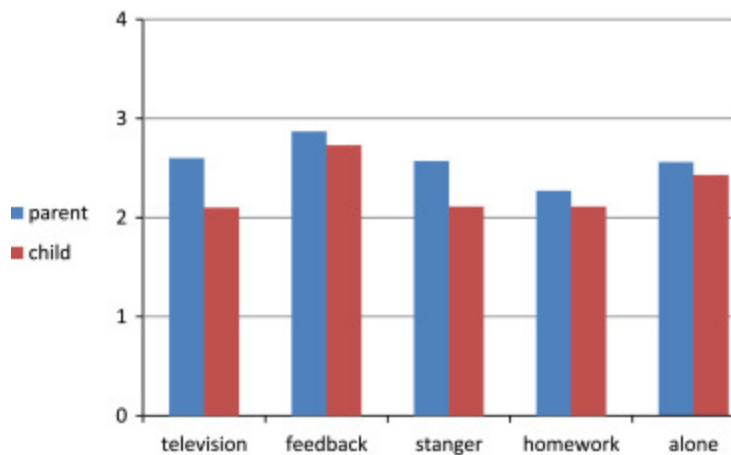


Fig. 1. Child and parent reports on FAI.

PUTS scores ($M=18.35$, $SD=6.50$) did not predict objective tic expression in any of the five situations.

Table 2 shows the ability of the FAI to predict tic number and the role of premonitory urges. In the homework and feedback situations, the children's report predicted objective tic expression. Using the method reported by Preacher et al. (2006) to examine interactions within a multiple regression, we found that when the level of premonitory urges was high (PUTS score >1 SD of the sample mean), the children's rating of tic level predicted tic expression in the alone and television situations ($b=22.26$, $p=0.08$; $b=58.87$, $p=0.06$, respectively). This was not true when the level of premonitory urges was low (PUTS score <1 SD of the sample mean). In the homework situation, the parental ratings of tic number was *inversely* related to the objective tic number ($b=-67.09$, $p < 0.01$).

Table 2. Standardized regression coefficients (beta) from a series of hierarchical multiple regression analysis for predicting Tic number by child and parent reports on FAI and premonitory urge ($N=41$).

		TV	F	S	H	A
Step 1	FAI child	0.28	0.37*	0.09	0.36 [†]	0.15
	FAI parent	0.07	-0.09	0.26	-0.41 [†]	-0.03
Step 2	PUTS	0.29	-0.09	-0.22	0.16	0.22
	FAI child×PUTS	0.40*	-0.09	-0.16	0.00	0.39*
	R ²	0.8	0.09	0.09	0.11	0.01
	ΔR ²	0.14*	0.10	0.02	0.18 [†]	0.11
	Total R ²	46	0.43	0.33	0.53*	0.34

TV=watching television; F=receiving attention when ticcing; S=talking with a stranger; HW=doing homework; A=being alone; FAI=Functional Assessment Interview; PUTS=Premonitory Urge for Tics Scale.

* $p < 0.10$.

† $p < 0.05$.

3.3. Influence of the environment on tic characteristics

On video-tape scoring by the two coders, the ICC for motor tics ranged from 0.82 through 0.98, and for vocal tics, from 0.73 through 0.98. Pearson's correlation yielded a test-retest reliability range of 0.78 through 0.93 ($M=0.86$). There was a high correlation of tic level in all situations (range 0.59–0.92, $p < 0.001$).

For repeated-measures ANOVA of tic expression in the five situations, we used the Greenhouse-Geisser correction for degrees of freedom, because a preliminary analysis showed that the data violated the sphericity assumption. The different situations were associated with differential amounts of tic expression ($F(2.26, 85.86)=5.70, p < 0.01, \eta p^2=0.13$) (Fig. 2). On post hoc Bonferroni test for multiple comparisons, the children had a greater tendency to tic in the television situation (high stimulation) than in the other four situations ($p < 0.01$ for all). There was no statistically significant difference in tic expression among the other four situations, although a significant linear trend was noted from the television situation through the feedback, stranger, homework and alone situations, in that order.

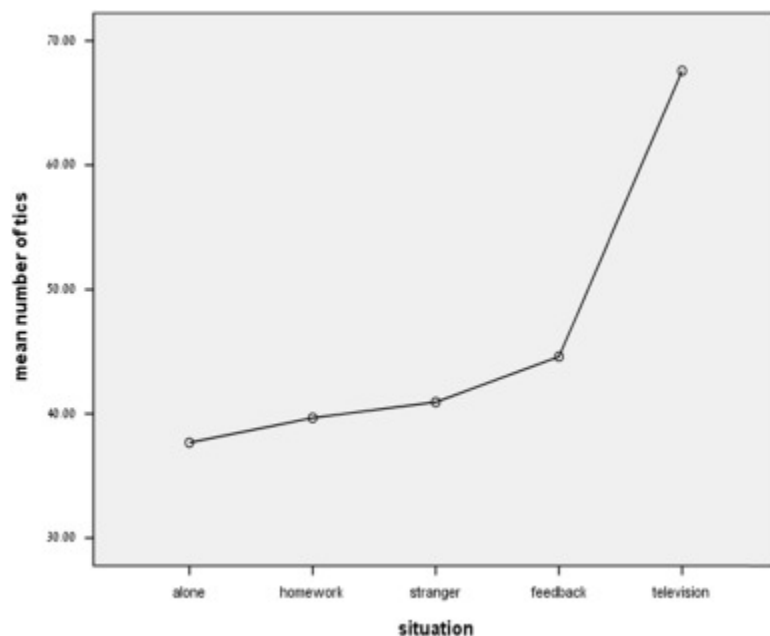


Fig. 2. Tic expression in the five environmental situations.

Hierarchical regression analyses revealed that the child's age had no effect on the associations between FAI scores and video-recordings. On multiple regression analysis, the presence of co-morbid conditions

(ADHD, OCD, and anxiety, as measured by CPRS, CY-BOCS and SCARED, respectively) had no significant predictive value on objective tic expression, but the individual groups were too small to draw definitive conclusions.

4. Discussion

The assessment of tic expression is challenging because tics vary over time and in location and intensity, and may be aggravated by environmental conditions and associated psychological and sensory factors.

Perhaps the most striking finding of this study is the marked alteration in tic frequency across environmental situations. Although this has long been clinically suspected, the present study is probably the most comprehensive and controlled investigation of this phenomenon to date. None of the subjects showed an undifferentiated pattern of tic number, and only two parents and three children reported no influence of the study situations on tic level. Watching television was associated with the highest tic expression of all the situations with 59% of the children experiencing most of their tics while watching television. Among the multiple explanations for this finding, the most plausible seems to be that watching television increases sensory stimulation. Accordingly, all four of our blinded reviewers found television to be the most stimulating situation of those included in the experiment.

The impact of television on the brain was shown by Engel et al. (2005) in a study of the *in vivo* activation of different brain regions during surgery in epileptic patients. However, electroencephalography studies of brain activation in subjects watching television commercials reported statistically significant activity in the fronto-parietal cortical areas, which are known to be involved in the process of tic suppression. Leckman et al. (2013) used the term “site sensitization” to describe the tendency of individuals with Tourette syndrome to be acutely aware of, distracted, and distressed by even faint sensory stimuli, and Belluscio et al. (2011) reported heightened sensitivity to external stimuli in all five sensory modalities.

It is noteworthy that the patterns of environmental effects differed between the subjective and objective measures. Our findings suggest that child self-reports predict real-life tic expression in some but not all contextual situations (Table 2). For example, 39% of the children reported that being alone was associated with the strongest tics, whereas the objective study showed that this was true in only 22%. Most of the children (56%) and many parents (49%) reported the highest number of tics for the feedback situation, but on objective study, this was true for only 19% of the children. This is further evidence of the apparent unreliability of subjective reports of tic severity, which form the basis of most of the clinical and research evaluations to date, and it has important implications for treatment. Many different treatments seem to diminish tic expression, but since tics classically wax and wane in severity and move from location to location, clinical impressions can be misleading. This problem is traditionally met by the use of structured interviews such as the YGTSS in which the clinician systematically asks about tics in their various forms. The validity of such interviews is based on the assumption that the subject is able to accurately report on his/her own tics. From our findings, however, it appears that this might not always be the case.

Although the PUTS score did not predict the objective expression of tics, premonitory urges appear to mediate the accuracy of the children's subjective reports of tic frequency within specific environmental

challenges (Table 2). The subjective reports may have been more reliable in the presence of strong premonitory urges, perhaps because the urge increases awareness of the tic. This might explain the lack of a statistically significant effect of the PUTS on the correlation between parent/clinician assessment and video observation, given that parents and clinicians are not privy to the urge sensation.

The relatively good performance of the CGI is relevant to this issue, because the CGI takes into account the overall general impression of the clinical state of the child based on direct observation as well as symptom elicitation, without detailed itemizing of the various clinical features. This is important in therapy research, where outcome is often based on symptom counts derived from self-report questionnaires or structured interviews. Although the YGTSS also takes the direct observation of the clinician into account, it includes a detailed itemization partially based on the subject's report, which may actually interfere with the accuracy of this measurement. Despite the obvious limitations of the YGTSS, at present there is no consensus on a better measure. However there is definitely a need to try and produce such an instrument. Perhaps using novel techniques such as smartphone applications might give the answer.

The major advantages of the present study are the comparatively large sample and experimental control. In addition, insofar as we were able to ascertain, this is the first attempt to use a structured interview to assess environmental effects on tic expression and to validate the findings against objective video-recordings. Himle et al. (2006) compared the YGTSS findings to video-observations in the home and clinic, but they did not focus on specific environmental challenges or use a specific instrument for this purpose.

This study was limited by its cross-sectional design. Further studies might use a single-case design to test the separate effects of contextual variables on tic expression. A behavioral model would predict individual variations that are washed out in group analysis. In addition, despite the relatively large sample size, it was still too small to account for the effects of different comorbidities, age, and anti-tic medications. Furthermore, the ability to generalize results to clinical practice is limited. However with the increasing availability of video using smartphones hopefully in many clinics this difficulty can be overcome.

Acknowledgment

The authors acknowledge the support for this study from the TSA and the Marie Curie Actions. We gratefully acknowledge the children and parents for their willingness to participate in this study.

References

- Belluscio et al., 2011. B.A. Belluscio, L. Jin, V. Watters, T.H. Lee, M. Hallett **Sensory sensitivity to external stimuli in Tourette syndrome patients** *Mov. Disord.*, 26 (2011), pp. 2538-2543
- Birmaher et al., 1999. B. Birmaher, D.A. Brent, L. Chiappetta, J. Bridge, S. Monga, M. Baugher **Psychometric properties of the Screen for Child Anxiety Related Emotional Disorders (SCARED): a replication study.** *J. Am. Acad. Child Adolesc. Psychiatry*, 38 (1999), pp. 1230-1236
- Birmaher et al., 1997. B. Birmaher, S. Khetarpal, D. Brent, M. Cully, L. Balach, J. Kaufman, *et al.* **The Screen for Child Anxiety Related Emotional Disorders (SCARED): scale construction and psychometric characteristics.** *J. Am. Acad. Child Adolesc. Psychiatry*, 36 (1997), pp. 545-553

- Cavanna and Piedad, 2013. A.E. Cavanna, J.C. Piedad **Clinical Rating**. D. Martino, J.F. Leckman (Eds.), *Instruments in Tourette Syndrome, Tourette Syndrome*, Oxford University Press, New York (2013), pp. 411-438
- Chang et al., 2009. S. Chang, M.B. Himle, B.T.P. Tucker, D.W. Woods, J. Piacentini **Initial psychometric properties of a brief parent - report instrument for assessing tic severity in children with chronic tic disorders**. *Child Fam. Behav. Ther.*, 31 (2009), pp. 181-191
- Conelea and Woods, 2008. C.A. Conelea, D.W. Woods **The influence of contextual factors on tic expression in Tourette's syndrome: a review**. *J. Psychosom. Res.*, 65 (2008), pp. 487-496
- Conners et al., 1998. C.K. Conners, G. Sitarenios, J.D. Parker, J.N. Epstein **The revised Conners' Parent Rating Scale (CPRS-R): factor structure, reliability, and criterion validity**. *J. Abnorm. Child Psychol.*, 26 (1998), pp. 257-268
- Engel et al., 2005. A.K. Engel, C.K.E. Moll, I. Fried, G.A. Ojemann **Invasive recordings from the human brain: clinical insights and beyond**. *Nat. Rev. Neurosci.*, 6 (2005), pp. 35-47
- Gallant et al., 2008. J. Gallant, E.A. Storch, L.J. Merlo, E.D. Ricketts, G.R. Geffken, W.K. Goodman, *et al.* **Convergent and discriminant validity of the Children's Yale-Brown Obsessive Compulsive Scale-Symptom Checklist**. *J. Anxiety Disord.*, 22 (2008), pp. 1369-1376
- Gresham et al., 2001. F.M. Gresham, T.S. Watson, C.H. Skinner **Functional behavior assessment: principles, procedures, and future directions**. *Sch. Psychol. Rev.*, 30 (2001), pp. 156-172
- Himle et al., 2006. M.B. Himle, S. Chang, D.W. Woods, A. Pearlman, B. Buzzella, L. Bunaciu, *et al.* **Establishing the feasibility of direct observation in the assessment of tics in children with chronic tic disorders**. *J. Appl. Behav. Anal.*, 39 (2006), pp. 429-440
- Kovacs, 1982. M. Kovacs **The Children's Depression Inventory: a Self-rated Depression Scale for School-aged Young-sters**. University of Pittsburgh School of Medicine, Pittsburgh, PA (1982)
- Kovacs, 1985. M. Kovacs **The children's depression inventory**. *Psychopharmacol. Bull.* (1985), pp. 995-998
- Kwon et al., 2011. H.J. Kwon, W.S. Lim, M.H. Lim, S.J. Lee, J.K. Hyun, J.H. Chae, *et al.* **1-Hz low frequency repetitive transcranial magnetic stimulation in children with Tourette's syndrome**. *Neurosci. Lett.*, 492 (2011), pp. 1-4
- Leckman et al., 2013. J.F. Leckman, M.H. Bloch, D.G. Sukhodolsky, L. Scahill, R.A. King **Phenomenology of Tics and Sensory Urges: The Self under Siege**. D. Martino, J.F. Leckman (Eds.), *Tourette Syndrome*, Oxford University Press, New York (2013), pp. 3-25
- Leckman et al., 1989. J.F. Leckman, M.A. Riddle, M.T. Hardin, S.I. Ort, K.L. Swartz, J. Stevenson, *et al.* **The Yale Global Tic Severity Scale: initial testing of a clinician-rated scale of tic severity**. *J. Am. Acad. Child Adolesc. Psychiatry*, 28 (1989), pp. 566-573
- Leckman et al., 1988. J.F. Leckman, K.E. Towbin, S.I. Ort **Clinical assessment of tic disorder severity**. D.J. Cohen, R. Bruun, J.F. Leckman (Eds.), *Tourette's Syndrome and Tic Disorders: Clinical Understanding and Treatment*, John Wiley and Sons, New York (1988), pp. 212-224
- Murphy and Eddy, 2013. T. Murphy, C.M. Eddy. **Neuropsychological Assessment in Tourette Syndrome**. D. Martino, J.F. Leckman (Eds.), *Tourette Syndrome*, Oxford University Press, New York (2013), pp. 439-467
- Pappert et al., 2003. E.J. Pappert, C.G. Goetz, E.D. Louis, L. Blasucci, S. Leurgans. **Objective assessments of longitudinal outcome in Gilles de la Tourette's syndrome**. *Neurol*, 61 (7) (2003), pp. 936-940
- Piacentini et al., 2006. J. Piacentini, M.B. Himle, S. Chang, D.E. Baruch, B.A. Buzzella, A. Pearlman, *et al.* **Reactivity of tic observation procedures to situation and setting**. *J. Abnorm. Child Psychol.*, 34 (2006), pp. 649-658
- Piacentini et al., 2010. J. Piacentini, D.W. Woods, L. Scahill, S. Wilhelm, A.L. Peterson, S. Chang, *et al.* **Behavior therapy for children with Tourette disorder: a randomized controlled trial**. *J. Am. Med. Assoc.*, 303 (2010), pp. 1929-1937

- Preacher et al., 2006. K.J. Preacher, P.J. Curran, D.J. Bauer. **Computational tools for probing interactions in multiple linear regression, multilevel modeling, and latent curve.** Anal. J. Educ. Behav. Stat., 31 (2006), pp. 437-448
- Pringshem and Steeves, 2011. T. Pringshem, T. Steeves. **Pharmacological treatment for Attention Deficit Hyperactivity Disorder (ADHD) in children with comorbid tic disorders.** Cochrane Database Syst. Rev. (2011). CD007990
- Psychological Corporation, 1999. Psychological Corporation, 1999. Wechsler Abbreviated Scale of Intelligence. San Antonio, TX.
- Rassin et al., 2000. E. Rassin, P. Muris, H. Schmidt, H. Merckelbach. **Relationships between thought-action fusion, thought suppression and obsessive-compulsive symptoms: a structural equation modeling approach.** Behav. Res. Ther., 38 (2000), pp. 889-897
- Robertson and Orth, 2006. M. Robertson, M. Orth. Behavioral and Affective Disorders in Tourette Syndrome, Williams And Wilkins, Philadelphia, Lippincott (2006)
- Scahill et al., 1997. L. Scahill, M.A. Riddle, M. McSwiggin-Hardin, S.I. Ort, R.A. King, W.K. Goodman, *et al.* **Children's Yale-Brown Obsessive Compulsive Scale: reliability and validity.** J. Am. Acad. Child. Adolesc. Psychiatry, 36 (1997), pp. 844-852
- Steinberg et al., 2010. T. Steinberg, S. Shmuel Baruch, A. Harush, R. Dar, D. Woods, J. Piacentini, *et al.* **Tic disorders and the premonitory urge.** J. Neural Transm., 117 (2010), pp. 277-284
- Storch et al., 2006. E.A. Storch, T.K. Murphy, J.W. Adkins, A.B. Lewin, G.R. Geffken, N.B. Johns, *et al.* **The children's Yale-Brown obsessive-compulsive scale: psychometric properties of child- and parent-report formats.** J. Anxiety Disord., 20 (2006), pp. 1055-1070
- Storch et al., 2005. E.A. Storch, T.K. Murphy, G.R. Geffken, M. Sajid, P. Allen, J.W. Roberti, *et al.* **Reliability and validity of the Yale Global Tic Severity Scale.** Psychol. Assess., 17 (2005), pp. 486-491
- Tapp et al., 1995. Tapp, J.H. Wehby, D. Ellis. **MOOSES: a multi option observation system for experimental studies.** Behav. Res. Methods Instrum. Comput. (1995), pp. 25-31
- Walkup et al., 1992. J.T. Walkup, L.A. Rosenberg, J. Brown, H.S. Singer. **The validity of instruments measuring tic severity in Tourette's syndrome.** J. Am. Acad. Child Adolesc. Psychiatry, 31 (1992), pp. 472-477
- Woods et al., 2005. D.W. Woods, J. Piacentini, M.B. Himle, S. Chang. **Premonitory Urge for Tics Scale (PUTS): initial psychometric results and examination of the premonitory urge phenomenon in youths with Tic disorders.** J. Dev. Behav. Pediatr., 26 (2005), pp. 397-403
- Woods et al., 2007. D.W. Woods, J.C. Piacentini, M.B. Himle. **Assessment of Tic Disorders.** D.W. Woods, J.C. Piacentini, J.T. Walkup (Eds.), Treating Tourette Syndrome and Tic Disorder: A Guide for Practitioners, Guilford Press, New York (2007), pp. 22-38
- Woods et al., 2001. D.W. Woods, T.S. Watson, E. Wolfe, M.P. Twohig, P.C. Friman. **Analyzing the influence of tic-related talk on vocal and motor tics in children with Tourette's syndrome.** J. Appl. Behav. Anal., 34 (2001), pp. 353-356