



# HHS Public Access

Author manuscript

*Disabil Health J.* Author manuscript; available in PMC 2015 April 13.

Published in final edited form as:

*Disabil Health J.* 2013 January ; 6(1): 26–35. doi:10.1016/j.dhjo.2012.10.002.

## Tourette syndrome, parenting aggravation, and the contribution of co-occurring conditions among a nationally representative sample

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### Abstract

**Background**—Previous research suggests that parents of a child with Tourette Syndrome (TS) have lower self-concepts, higher caregiver burden, and more difficulties with home activities. However, the contributions of TS and mental, emotional, or behavioral (MEB) conditions to family functioning are difficult to identify from previous research due to relatively small TS sample sizes and high rates of co-occurring conditions within samples of children with TS.

**Objective**—The current study hypothesized that families of children with TS would report significantly more family functioning difficulties (more parenting aggravation, more difficulty with coping with the child's care, less parent–child communication, and less consistent family routines). Specifically, co-occurring conditions would contribute substantially to reported parenting aggravation.

**Method**—Parent-reported data from the 2007 National Survey of Children's Health were analyzed, including whether the child had been diagnosed with TS or an MEB. Weighted analyses were restricted to US children 6–17 years of age ( $n = 64,034$ ) and adjusted for child age, sex, race and ethnicity.

**Results**—Parents of children with TS were more likely to fall into the high parenting aggravation index category compared with parents of children without TS (aPR = 3.8, 95% CI: 2.2–6.6). Controlling for the co-occurring MEB conditions attenuated the relations between TS and parenting aggravation; however, a significant effect for TS remained in some cases.

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The authors do not have any conflicts of interest to declare. Results from this study were previously presented as a poster symposium at the 2010 Society for Behavioral Developmental Pediatrics Annual Meeting. The findings and conclusions in this report are those of the authors and do not necessarily represent the official position of the Centers for Disease Control and Prevention.

**Conclusion**—Parents of children with TS may face significant challenges in raising their children, leading to increased parenting aggravation; these challenges appear to be primarily associated with the presence of co-occurring MEB conditions.

### Keywords

Tourette syndrome; Family functioning; Mental; emotional; behavioral conditions

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### Introduction

Mental, emotional, and behavioral conditions (MEBs) occur among 14–20% of children and adolescents and the financial burden of these disorders is estimated at \$247 billion. Tourette syndrome (TS) is a neurobehavioral disorder with onset in childhood, characterized by multiple persistent motor tics and at least one vocal tic<sup>2</sup> that affects 3–8/1000 U.S. children.<sup>3,4</sup> TS has social costs that impact public health through greater use of education and health care services, and potential impacts on employment, as well as an impact on family functioning.<sup>5–10</sup>

Family functioning is the family's ability to promote and support the health and wellbeing of its family members. Process models of parental and family functioning suggest that transactions within the family context impact child adjustment with contextual sources of stress and support as barriers and facilitators, respectively, to family functioning.<sup>11,12</sup> Although originally conceptualized to describe risk for child maltreatment, a transactional-ecological model of risk and resilience<sup>13</sup> posits that interacting social contexts impact a child's functioning over time; this model can be used to describe family functioning and child adjustment more broadly.

The transactional-ecological model contends that child and family outcomes are most at risk when risk factors outweigh protective factors. Nurturing, responsive, and consistent positive interactions within the parent–child relationship are associated with long-term child health<sup>14</sup>; parenting stress can impair the quality of these relationships and is bi-directionally associated with poorer child functioning.<sup>15–18</sup> Elements of the family's social context such as emotional support and consistent family routines are factors that can promote child development; conversely, poor parental mental health, lack of neighborhood social capital, and poor parental coping potentiate family functioning risk and negatively impact child health and adjustment<sup>1,11,14,19,20</sup>

Having a child with a chronic medical condition can increase parenting stress which in turn can exacerbate child symptomatology and management of the child's condition.<sup>21,22</sup> Family stressors that have been associated with TS are similar to those associated with other chronic medical conditions, and include: parental guilt and difficulty coping with the child's diagnosis, challenges of implementing consistent discipline and parenting strategies in the context of the unpredictability of the condition, and strained family dynamics and functioning.<sup>23,24</sup> These family stressors are complicated by personal stressors experienced by the family and child with TS, including social stigma and the common misconception that tics are controllable.<sup>24</sup> Although, the body of literature examining the impact of TS on family functioning is limited, previous reports suggest that parents of a child with TS have

lower self-concepts,<sup>25</sup> higher caregiver burden,<sup>26</sup> more difficulties with home activities,<sup>27</sup> and more parent–child communication difficulties.<sup>28</sup> Increased TS severity has also been associated with increased TS family stress; specifically parents of children with more severe TS report more difficulties caring for their child.<sup>10</sup>

The high rate of co-occurring MEBs with TS has been reported consistently in different sample populations (e.g., clinic, community, administrative data); attention-deficit/hyperactivity disorder (ADHD) is often cited as the most common.<sup>6,29</sup> Although TS has a high rate of co-occurrence with other MEBs, TS and MEBs (such as obsessive compulsive disorder, OCD, and ADHD) are distinct clinical diagnoses as defined by the Diagnostic and Statistical Manual of Mental disorders, 4th Edition, Text Revision<sup>2</sup> with overlapping but different neural<sup>30,31</sup> and genetic pathways.<sup>32,33</sup> The presence of co-occurring conditions may impact family stress directly, and through the need for additional health care and educational services.<sup>5,6,34</sup> Research supports an association between family functioning difficulties and MEBs such as ADHD, oppositional defiant disorder, autism spectrum disorders (ASD), and depression.<sup>1</sup> Therefore the independent contributions of TS and MEBs to family functioning are difficult to identify particularly due to relatively small TS sample sizes and high rates of co-occurring conditions within samples of children with TS. Several studies suggest minimal effects of TS on family functioning when considering the contribution of disruptive behavior disorders and ADHD; however, TS that co-occurs with disruptive behavioral disorders has been associated with poorer family functioning.<sup>27,35,36</sup> Greater rejecting parenting style,<sup>35</sup> greater impact of the disorder on the family,<sup>36</sup> and more problematic family relations, less cohesion, less organization, less open expression, and more conflict,<sup>37</sup> are specific family functioning difficulties associated with TS with co-occurring MEBs. In a study of children with TS, 52% of caregivers attributed significant difficulties in their child's social, school, or home activities to tics, versus 70% attributing impairment to a co-occurring condition.<sup>27</sup>

While all children with TS may not require treatment, evidence-based treatments for TS are available, including habit reversal training and pharmacotherapy.<sup>35</sup> Children with TS and a co-occurring MEB may require additional health care services, and the presence of co-occurring conditions may complicate treatment.<sup>35</sup> In one study, over 73% of children with TS filled at least one prescription for a psychotropic medication in the previous year; mental health treatment was also common among these children.<sup>6</sup> No formal assessments of combined psychotropic treatment for TS and co-occurring MEBs have been reported.<sup>34</sup>

The relatively small sample size of previous studies (range = 59–100) has not allowed for comprehensive examination of the unique contributions of different types of MEBs on TS family functioning relations. Previous studies have focused on either externalizing problems broadly or a few of the most common conditions that co-occur with TS (e.g., ADHD and conduct problems). The current study addresses some of the limitations of previous studies by examining the relationship of TS and major co-occurring MEBs, specifically depression, anxiety, ADHD, behavior or conduct problems and ASD, with family functioning characteristics using data from the National Survey of Children's Health, a national, population-based telephone survey. This large national data set allows for further examination of the contribution of TS in family functioning while beginning to identify the

contribution of common co-occurring conditions. Based on the previous literature, we hypothesized that families of children with TS would report significantly more family functioning difficulties (more parenting aggravation, more difficulty with coping with the child's care, less parent-child communication, and less consistent family routines) than families without a child with TS. Furthermore, we hypothesized that co-occurring MEB conditions would contribute substantially to one indicator of family functioning, reported parental aggravation.

## Methods

Data from the 2007–2008 National Survey of Children's Health (2007 NSCH) were analyzed. The 2007 NSCH was the first national, population-based survey that included questions on TS.<sup>38</sup> Telephone interviews were conducted with 91,642 parents or guardians of children from birth through 17 years of age between April 2007 and July 2008; the majority of respondents were mothers (73.5%).<sup>38</sup> Participants were from all 50 states and the District of Columbia and were chosen through a random-digit dial method of landline telephone numbers. One child was randomly selected from each household as the focus of the interview. The overall response rate was 46.7%, representing the percentage of households that completed the interview among all eligible households, including those that were not successfully contacted. The overall cooperation rate was 66%, representing the percentage of eligible households participating among those that were contacted. Analyses for this study were limited to children aged 6–17 years ( $n = 66,034$ ) because TS is not typically diagnosed at younger ages.<sup>2,39</sup> Prior to public release, the data were weighted to account for the unequal probability of selection of each household and child, for non-response, and for households without landline telephones. Weights were also adjusted so that estimates reflected the demographic distribution of noninstitutionalized U.S. children based on the 2007 American Community Survey of the U.S. Census Bureau. Data and documentation for the NSCH are publically available and can be downloaded from the CDC website (<http://www.cdc.gov/nchs/slait/nsch.htm>).

## Independent variable

Parent respondents were asked a series of questions about different health conditions affecting the study child that were worded: "Has a doctor or other health care professional ever told you that [your child] had [condition]?" Conditions analyzed in the current study included Tourette Syndrome (TS); ADHD; behavioral or conduct problems such as oppositional defiant disorder or conduct disorder, behavioral or conduct problems; depression; anxiety problems; and ASD. Categories of TS status were created: children without TS (no TS), all children with TS (TS), TS with a specified co-occurring MEB (ADHD, behavioral or conduct problems, depression, anxiety problems, and ASD; TS + MEB), children with TS only (no co-occurring MEB; TS only), children with TS and an internalizing problem (depression or anxiety problems; TS + INT), children with TS and an externalizing problem (behavioral or conduct problems or ADHD; TS + EXT), and children with TS and ASD (TS + ASD). Because of the high rate of co-occurring conditions among children with TS, there was significant overlap between TS + INT, TS + EXT and TS + ASD; additionally, all these groups met criteria for inclusion in the TS + MEB category.

## Dependent variables

Parents were also asked questions about their general family functioning (Table 1). Specifically, parents were asked about the frequency of family meals, their ability to share and have meaningful conversations with their child, and their ability to cope with the daily demands of parenting. These three questions are indicators of different elements of family functioning: the frequency of family meals is an indicator of consistent family routines, the ability to share conversations is an indicator of parent–child communication, and the ability to cope with demands of parenting is an indicator of parental coping.

Three additional questions asked parents to report the past month frequency of the following: feeling that it was harder to care for their child than others of the same age, the child doing things that really bothered them, and feeling angry with the child. These three questions dichotomized as described here,<sup>40</sup> were analyzed independently and also combined into a single aggravation index using established criteria for the Aggravation in Parenting Scale.<sup>41</sup> Parenting aggravation is an indicator of family functioning that measures the level of parenting stress and frustration a parent experiences during care for her child. The Aggravation in Parenting Scale was created using items from the Parenting Stress Index and the Child-rearing Scale.<sup>38</sup> Previous research indicates parents of children with autism have higher levels of parenting aggravation.<sup>40,42</sup> Consistent with research on parenting stress,<sup>43,44</sup> the Aggravation in Parenting Scale is also related to poverty, lower parental education, and single-parenthood.<sup>38</sup>

The original Aggravation in Parenting Scale<sup>41</sup> included four questions; however, the 2007 NSCH survey did not include the question about “parent report of giving up more of their life than expected to meet the child’s needs” and therefore the scoring was revised, as described here.<sup>40</sup> Responses were coded from 1 (never/rarely) to 4 (always) and the response values were averaged to create a parenting aggravation index score. Parents who answered usually or always to all outcomes, answered always to at least two questions, or answered always to one outcome, usually to one outcome, and sometimes to one outcome were classified as “high aggravation”.

In addition, potential protective factors that support family functioning were examined to determine whether these variables (emotional support, social capital, and parental mental health) were associated with TS status, Table 1. Parents answered yes or no to the emotional support question. A social capital index (a measure of social cohesion, trust, and reciprocity) was created using established criteria<sup>45</sup> with the following 4 questions: 1) People in this neighborhood help each other out; 2) We watch out for each other’s children in this neighborhood; 3) There are people I can count on in this neighborhood; and 4) If my child were outside playing and got hurt or scared, there are adults nearby who I trust to help my child. Responses were coded from 1 (definitely agree) to 4 (definitely disagree), and the response values were summed to create an index score.<sup>38</sup> Moderate to high social capital was defined as a composite score of  $<7$ .<sup>40</sup> Factor analysis confirms the use of a single scale.<sup>46</sup>

Maternal and paternal mental health were also measured using the following question: “Would you say that, in general, (mother/father) mental and emotional health is excellent,

very good, good, fair, or poor?” Answers were dichotomized with excellent, very good, and good as good mental health; and fair or poor as poor mental health.

### Data analysis

Data were analyzed using SAS 9.2 (SAS Institute Inc., Cary, NC, USA) and SUDAAN 10.0.1 (Research Triangle Institute International, Research Triangle Park, NC, USA) to account for the complex survey sampling design. Demographic variables analyzed included child age (6–11 years vs. 12–17 years), sex, race (non-white vs. white), Hispanic ethnicity (Hispanic vs. non-Hispanic), family structure (2 biological or adoptive parents; 1 biological and one step-parent in the home, single parent, and all other [kin, legal guardians, co-habiting, and same sex couples]), highest level of education achieved by parent in household (high school diploma or less vs. at least some college or technical school), and household income ( $\leq 200\%$  of federal poverty level [FPL],  $>200\%$  of FPL). Demographic variables and the prevalence of co-occurring MEBs were examined by TS status. Chi-square tests were used to compare percentages and calculate prevalence ratios examining demographic characteristics by TS status (with children without TS as the comparison group). Prevalence ratios for family structure were calculated using logistic regression. Family functioning and protective factors were dichotomized and examined by TS status. Multivariable logistic regression was used to calculate adjusted prevalence ratios of the association between family functioning variables and TS status, controlling for child age, sex, race, and ethnicity. In addition, multivariable logistic regression was used to calculate prevalence ratios, adjusted for demographic variables, for the association between parenting aggravation and TS status, controlling for different MEBs. The prevalence ratio (PR) is appropriate for analyses using cross-sectional data, and may be more conservative than odds ratios.

Because the sample size for some groups was small, relative standard errors (RSE) were calculated (standard error/prevalence estimate  $\times 100$ ) for each prevalence estimate. All RSEs that are greater than 30% are reported because estimates may be statistically unreliable.

### Results

Demographic characteristics are reported in Table 2. Children diagnosed with TS were more likely to be older and male. There was no difference in parent education, family structure, race or ethnicity, or household income by TS status.

Among children with TS, 78.7% had at least one other co-occurring MEB: 52.5% had an internalizing condition or problem, 70.1% had a co-occurring externalizing condition or problem, and 26.2% had an ASD. The most common co-occurring MEB was ADHD (63.8%), followed by behavioral or conduct problems (43.0%), anxiety problems (39.7%), depression (36.2%) and ASD (26.2%). These prevalence rates were all significantly greater than among the non-TS group.

There was no difference observed between the number of times a family ate dinner together in the past week, shared ideas with each other, or how well the parent was coping with the

day to day demands of parenthood, between parents of children with or without TS, Table 3. Compared to parents of children without TS, parents of children with TS were more likely to respond “usually or always” that their child was much harder to care for than most children his/her age, that their child did things that really bothered the parent a lot, and that the parent felt angry with the child. Parents of children with TS were also more likely to fall into the high aggravation index category compared with parents of children without TS, Table 3. To compare dichotomized results to a continuous score for parenting aggravation, the association of “higher aggravation” associated with TS was confirmed with an ANOVA comparing the continuous aggravation score by TS status ( $p = 0.001$ ).

There were no differences by TS status for emotional help, the social capital index, or paternal mental health. Because there were no significant findings for these variables, they were not controlled for in the multivariable models below.

To examine the contributions of co-occurring MEBs to parenting aggravation, analyses were also run separately examining the relations between TS and parenting aggravation, controlling for each of the following: any MEB, internalizing conditions, externalizing conditions, ASD, ADHD, behavioral or conduct problems (BEH), anxiety (ANX), and depression (DEP), Table 4. Overall, controlling for the co-occurring conditions attenuated the relations between TS and parenting aggravation. However, in some cases (TS + DEP and TS + ASD) there was still a significant effect for TS. In other analyses that included co-occurring conditions (TS + MEB, TS + INT, TS + ANX, TS + EXT, TS + ADHD, TS + BEH), the association between TS and parenting aggravation was no longer statistically significant (although all aPRs were  $\leq 1.5$ ). Nonetheless, the associations between each of the co-occurring condition variables and the parenting aggravation outcome was strong (aPRs  $\leq 3.9$ ) and statistically significant, suggesting the total association between TS and parenting aggravation (aPR = 3.8 from original model) was strongly influenced by the co-occurring conditions (Table 4). In particular, behavioral or conduct problems (aPR 7.6, 95% CI: 6.5–8.8) and ASD (aPR 4.7 95% CI: 3.6–6.1) were the individual conditions with the strongest relative associations with parenting aggravation.

In an attempt to examine the contribution of MEBs not directly measured by the NSCH, additional analyses examined the relationship between TS and parenting aggravation controlling for “any kind of emotional, developmental, or behavioral problem for which [he/she] needs treatment or counseling.” As observed in the TS + MEB analyses, after controlling for this co-occurring condition variable, there no longer was an association between TS and parenting aggravation (aPR 1.4, 95% CI: 0.7–2.8; RSE 35.7%); additionally, the association of the co-occurring problems and parenting aggravation was much stronger and statistically significant (aPR 6.6, 95% CI: 5.7–7.6).

## Discussion

This study showed increased parenting aggravation related to parenting a child with TS using nationally representative data. Furthermore, we were able to explore the relationships between co-occurring conditions to start to identify the factors affecting family functioning for children with TS and their families. Others have reported the impact of co-occurring

externalizing problems on family functioning;<sup>27,35,36</sup> the current study additionally reports a relationship between TS with co-occurring internalizing conditions and a similar pattern of effects for individual MEBs (anxiety, depression, ADHD, behavioral or conduct problems, and ASD). Thus, these results can inform treatment of TS and co-occurring conditions to improve family functioning.

Results from this study support earlier findings that parents of children with TS may experience increased stress and difficulties parenting their children; however, these difficulties may be associated with the conditions that co-occur with the TS and not the TS symptomatology itself. In particular, ASD and behavioral or conduct problems were the individual conditions most strongly associated with parenting aggravation. Previous research with children with ASD<sup>40</sup> and children with behavior and conduct problems<sup>47</sup> supports this finding. Two small trials have shown that children with TS and disruptive behavioral disorders, and their families, might benefit from parent-management (parent focused) or anger-control (adolescent focused) training.<sup>48,49</sup>

Secondary data analysis of national survey data presents both challenges and opportunities for research. Given that TS is a less common condition and large sample sizes of TS are often difficult to collect, the NSCH allowed us to obtain a large enough sample to generate national estimates of TS prevalence<sup>3</sup> and to examine some factors within TS. However, there are some limitations associated with secondary data analysis in general and these data specifically. First, although the sample size among the TS group was relatively large, the TS only group had limited size due to high rates of co-occurring conditions among children with TS, preventing definitive conclusions in some cases. Despite this limitation, analyses consistently showed higher parenting aggravation among the TS groups (even when controlling for some co-occurring conditions, Table 4). Furthermore the limits of the TS sample size precluded further examination into potentially interesting demographic modifiers of the relations between TS and parenting aggravation (such as developmental stage, sex, race, parental diagnoses, and ethnicity). In addition, the sample size was not large enough to detect reliable differences between children with current TS and those who had ever had TS, but not currently.

A second set of limitations of these data are related to the questions collected on the NSCH. As with all secondary data analyses, we were limited by the data that was collected and a more extensive collection of family functioning indicators could have some advantages. However, the questions asked in this data set were still able to provide a broad picture of family functioning for children with TS. In addition, there was not a direct question addressing OCD on the NSCH; therefore, unmeasured confounding by co-occurring conditions not specifically measured in the NSCH may be contributing to the findings around the TS and MEB group. We attempted to address this limitation by examining the relationship between children with TS and any mental, emotional, behavioral, or developmental problems and parenting aggravation, controlling for the co-occurring problems. The pattern of effects for this group was consistent with the TS and MEB group further supporting the contribution of co-occurring conditions to parenting aggravation.



The NSCH relied on parental report that a doctor or health care provider told the parent their child has a condition instead of the use of direct assessment or clinical diagnosis. This methodology is subject to recall bias of parental report and also error related to inaccurate diagnoses. Nevertheless, previous research has supported the validity of parental recall of diagnosis for neurobehavioral conditions.<sup>40,50</sup> The prevalence estimate of TS was in the range of community-based studies of TS using diagnostic criteria.<sup>4</sup> Finally, the results are subject to non-coverage biases associated with the exclusion of households without landlines and low response rates of telephone-based surveys relative to face-to-face surveys.<sup>38</sup>

Despite these limitations, these data make a meaningful contribution to our understanding of the family context for children with TS. Family functioning and interactions within the family context are related to children's adjustment and life-long health.<sup>11–13,51</sup>

Developmental trajectories of internalizing<sup>52</sup> and externalizing problems<sup>53</sup> may persist and may be maintained by family stress and problematic parenting interactions. Because these relationships are bidirectional, reducing family stress and problematic parent-child interactions may also improve behavioral symptomatology. In a sample of children with TS, Carter and colleagues<sup>37</sup> report that family functioning is associated with behavioral adjustment above and beyond the child's TS or ADHD diagnoses. At the extreme end of the continuum of family functioning, parental stress and parental perception of the child's behavior as problematic are critical factors that can place a child at higher risk for child maltreatment.<sup>54</sup> Moreover, the conditions that commonly co-occur with TS, such as ADHD<sup>55</sup> and general behavioral problems, have been identified as placing a child at risk for child maltreatment.<sup>56</sup> Given the high rates of TS with co-occurring MEBs and the impact of these MEBs on family functioning, there is a significant need for evidence-based treatments that address these more complex cases. Thus, addressing externalizing and internalizing disorders with evidence-based interventions may mitigate parenting aggravation associated with TS, improve family functioning and also potentially prevent more serious escalation of negative parenting behaviors and child symptomatology.

In summary, parents of children with TS may face significant challenges in raising their children, leading to increased parenting aggravation. These challenges appear to be primarily associated with the presence of co-occurring MEB conditions. Increasing awareness and understanding by parents, clinicians and educators, of the significant impact of TS and co-occurring MEBs on family functioning provides a first step in addressing this challenge. Attention to and management of the co-occurring conditions, may help alleviate some aspects of family stress associated with these conditions and improve the overall adjustment of children with TS. From a public health perspective, this could have impacts in the use of medical and education services, and improve long-term outcomes among youth with TS.

## References

1. National Research Council and Institute of Medicine. Preventing Mental, Emotional, and Behavioral Disorders Among Young People: Progress and Possibilities. Washington DC: The National Academies Press; 2009.
2. American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders. 4th ed.. Washington, DC: American Psychiatric Association; 2000. Text Revision ed

3. Centers for Disease Control and Prevention. Prevalence of diagnosed Tourette syndrome in persons aged 6–17 years – United States, 2007. *Morb Mortal Wkly Rep.* 2009; 58(21):581–585.
4. Scahill L, Sukhodolsky DG, Williams SK, Leckman JF. Public health significance of tic disorders in children and adolescents. *Adv Neurol.* 2005; 96:240–248. [PubMed: 16383223]
5. Debes N, Hjalgrim H, Skov L. The presence of Attention-Deficit Hyperactivity Disorder (ADHD) and Obsessive-Compulsive Disorder worsen psychosocial and educational problems in Tourette syndrome. *J Child Neurol.* 2010; 24(12)
6. Olfson M, Crystal S, Gerhard T, Huang C, Walkup JT, Scahill L. Patterns and correlates of tic disorder diagnoses in privately and publicly insured youth. *J Am Acad Child Adolesc Psychiatry.* 2011; 50(2):119–131. [PubMed: 21241949]
7. Shady G, Broder R, Staley D, Furer P, Papadopolos RB. Tourette syndrome and employment: descriptors, predictors, and problems. *Psychiatr Rehabil J.* 1995; 19(1):35–42.
8. Packer LE. Tic-related school problems: impact on functioning, accommodations, and interventions. *Behav Modif.* 2005; 29(6):876–899. [PubMed: 16204421]
9. Dodel I, Reese JP, Muller N, et al. Cost of illness in patients with Gilles de la Tourette's syndrome. *J Neurol.* 2010; 257(7):1055–1061. [PubMed: 20179960]
10. Lee MY, Chen YC, Wang HS, Chen DR. Parenting stress and related factors in parents of children with Tourette syndrome. *J Nurs Res.* 2007; 15(3):165–174. [PubMed: 17806033]
11. Belsky J. The determinants of parenting: a process model. *Child Dev.* 1984; 55:83–96. [PubMed: 6705636]
12. Bronfenbrenner U. Ecology of the family as a context for human development: research perspectives. *Dev Psychol.* 1986; 22(6):723–742.
13. Cicchetti D, Lynch K. Toward an ecological/transactional model of community violence and child maltreatment: consequences for children's development. *Psychiatry.* 1993; 56:96–118. [PubMed: 8488217]
14. Shonkoff, J.; Philips, D. *From Neurons to Neighborhoods: The Science of Early Childhood Development.* Washington, DC: National Academy Press; 2000.
15. Crnic KA, Gaze C, Hoffman C. Cumulative parenting stress across the preschool period: relations to maternal parenting and child behaviour at age 5. *Infant Child Dev.* 2005; 14(2):117–132.
16. Pinderhughes EE, Dodge KA, Bates JE, Pettit GS, Zelli A. Discipline responses: influences of parents' socioeconomic status, ethnicity, beliefs about parenting, stress, and cognitive-emotional processes. *J Fam Psychol.* 2000; 14(3):380–400. [PubMed: 11025931]
17. Gershoff ET, Aber JL, Raver CC, Lennon MC. Income is not enough: incorporating material hardship into models of income associations with parenting and child development. *Child Dev.* 2007; 78(1):70–95. [PubMed: 17328694]
18. Benzie KM, Harrison MJ, Magill-Evans J. Parenting stress, marital quality, and child behavior problems at age 7 years. *Public Health Nurs.* 2004; 21(2):111–121. [PubMed: 14987210]
19. Fiese BH, Foley KP, Spagnola M. Routine and ritual elements in family mealtimes: contexts for child well-being and family identity. *New Dir Child Adolesc Dev.* 2006; 2006(111):67–89. [PubMed: 16646500]
20. Drukker M, Kaplan C, Feron F, van Os J. Children's health-related quality of life, neighbourhood socio-economic deprivation and social capital. A contextual analysis. *Soc Sci Med.* 2003; 57(5): 825–841. [PubMed: 12850109]
21. Raina P, O'Donnell M, Rosenbaum P, et al. The health and well-being of caregivers of children with cerebral palsy. *Pediatrics.* 2005; 115:e626–e636. [PubMed: 15930188]
22. Armstrong B, Mackey ER, Streisand R. Parenting behavior, child functioning, and health behaviors in preadolescents with type 1 diabetes. *J Pediatr Psychol.* 2011; 36(9):1052–1061. [PubMed: 21828111]
23. Ginsburg, GS.; Kingery, JN. *Management of Familial Issues in Persons with Tourette Syndrome. Treating Tourette Syndrome and Tic Disorders: A Guide for Practitioners.* New York, NY, US: Guilford Press; 2007. p. 225-241.
24. Cohen, DJ.; Ort, SI.; Leckman, JF.; Riddle, MA.; Hardin, MT. *Family Functioning and Tourette's Syndrome. Tourette's Syndrome and Tic Disorders: Clinical Understanding and Treatment.* Oxford, England: John Wiley & Sons; 1988. p. 179-196.

25. Edell-Fisher BH, Motta RW. Tourette syndrome: relation to children's and parent's self-concepts. *Psychol Rep.* 1990; 66(2):539–545. [PubMed: 2349344]
26. Cooper C, Robertson MM, Livingston G. Psychological morbidity and caregiver burden in parents of children with Tourette's disorder and psychiatric comorbidity. *J Am Acad Child Adolesc Psychiatry.* 2003; 42(11):1370–1375. [PubMed: 14566175]
27. Storch EA, Lack CW, Simons LE, Goodman WK, Murphy TK, Geffken GR. A measure of functional impairment in youth with Tourette's Syndrome. *J Pediatr Psychol.* 2007; 32(8):950–959. [PubMed: 17522110]
28. Matthews M, Eustace C, Grad G, Pelcovitz D, Olson M. A family systems perspective on Tourette's Syndrome. *Int J Fam Psychiatry.* 1985; 6(1):53–66.
29. Kadesjo B, Gillberg C. Tourette's disorder: epidemiology and comorbidity in primary school children. *J Am Acad Child Adolesc Psychiatry.* 2000; 39(5):548–555. [research]. [PubMed: 10802971]
30. Makki MI, Govindan RM, Wilson BJ, Behen ME, Chugani HT. Altered fronto-striato-thalamic connectivity in children with Tourette syndrome assessed with diffusion tensor MRI and probabilistic fiber tracking. *J Child Neurol.* 2009; 24(6):669–678. [PubMed: 19491113]
31. Sukhodolsky DG, Landeros-Weisenberger A, Scahill L, Leckman JF, Schultz RT. Neuropsychological functioning in children with Tourette syndrome with and without attention-deficit/hyperactivity disorder. *J Am Acad Child Adolesc Psychiatry.* 2010; 49(11):1155–1164. [PubMed: 20970703]
32. Debes NM, Hjalgrim H, Skov L. Predictive factors for familiarity in a Danish clinical cohort of children with Tourette syndrome. *Eur J Med Genet.* 2010; 53(4):171–178. [PubMed: 20457287]
33. Grados MA, Mathews CA. Latent class analysis of Gilles de la Tourette syndrome using comorbidities: clinical and genetic implications. *Biol Psychiatry.* 2008; 64(3):219–225. [PubMed: 18359008]
34. Kurlan R. Clinical practice. Tourette's Syndrome. *N Engl J Med.* 2010; 363(24):2332–2338. [PubMed: 21142535]
35. Cohen E, Sade M, Benarroch F, Pollak Y, Gross-Tsur V. Locus of control, perceived parenting style, and symptoms of anxiety and depression in children with Tourette's syndrome. *Eur Child Adolesc Psychiatry.* 2008; 17(5):299–305. [PubMed: 18301938]
36. Wilkinson B, Newman MB, Shytle RD, Silver AA, Sanberg PR, Sheehan D. Family impact of Tourette's Syndrome. *J Child Fam Stud.* 2001; 10(4):477–483.
37. Carter AS, O'Donnell DA, Schultz RT, Scahill L, Leckman JF, Pauls DL. Social and emotional adjustment in children affected with Gilles de la Tourette's syndrome: associations with ADHD and family functioning. *Attention Deficit Hyperactivity Disorder. J Child Psychol Psychiatry.* 2000; 41(2):215–223. [PubMed: 10750547]
38. Blumberg SJ, Foster EB, Frasier AM, et al. Design and operation of the national survey of children's health, 2007. *Vital Health Stat 1.* 2012:55.
39. Leckman JF, Zhang H, Vitale A, et al. Course of tic severity in Tourette syndrome: the first two decades. *Pediatrics.* 1998; 102(1 Pt 1):14–19. [PubMed: 9651407]
40. Schieve LA, Boulet SL, Kogan MD, et al. Parenting aggravation and autism spectrum disorders: 2007 National Survey of Children's Health. *Disabil Health J.* 2011; 4(3):143–152. [PubMed: 21723521]
41. Macomber, JE.; Moore, KA. *NSAF Benchmarking Measures of Child and Family Well-Being.* Washington, D.C: Urban Institute; 1999. 1997
42. Schieve LA, Blumberg SJ, Rice C, Visser SN, Boyle C. The relationship between autism and parenting stress. *Pediatrics.* 2007; 119(suppl 1):S114–S121. [PubMed: 17272578]
43. Raikes A, Thompson RA. Efficacy and social support as predictors of parenting stress among families in poverty. *Infant Ment Health J.* 2005; 26:177–190.
44. McLoyd, VC.; Wilson, L. The strain of living poor: parenting, social support, and child mental health. In: Huston, AC., editor. *Children in Poverty.* New York: Cambridge University; 1991. p. 105-135.

45. Singh G, Kogan M, Siahpush M, van Dyck P. Independent and joint effects of socioeconomic, behavioral, and neighborhood characteristics on physical inactivity and activity levels among us children and adolescents. *J Community Health*. 2008; 33(4):206–216. [PubMed: 18373183]
46. Singh GK, Kogan MD, Van Dyck PC, Siahpush M. Racial/ethnic, socioeconomic, and behavioral determinants of childhood and adolescent obesity in the united states: analyzing independent and joint associations. *Ann Epidemiol*. 2008; 18(9):682–695. [PubMed: 18794009]
47. Kolko DJ, Dorn LD, Bukstein O, Burke JD. Clinically referred ODD children with or without CD and healthy controls: comparisons across contextual domains. *J Child Fam Stud*. 2008; 17(5):714–734.
48. Scahill L, Sukhodolsky DG, Bearss K, et al. Randomized trial of parent management training in children with tic disorders and disruptive behavior. *J Child Neurol*. 2006; 21(8):650–656. [PubMed: 16970865]
49. Sukhodolsky DG, Vitulano LA, Carroll DH, McGuire J, Leckman JF, Scahill L. Randomized trial of anger control training for adolescents with Tourette’s syndrome and disruptive behavior. *J Am Acad Child Adolesc Psychiatry*. 2009; 48(4):413–421. [PubMed: 19242384]
50. Russ SA, Larson K, Halfon N. A national profile of childhood epilepsy and seizure disorder. *Pediatrics*. 2012; 129(2):256–264. [PubMed: 22271699]
51. Shonkoff JP, Boyce WT, McEwen BS. Neuroscience, molecular biology, and the childhood roots of health disparities: building a new framework for health promotion and disease prevention. *J Am Med Assoc*. 2009; 301(21):2252–2259.
52. Herrenkohl TI, Kosterman R, Hawkins JD, Mason WA. Effects of growth in family conflict in adolescence on adult depressive symptoms: mediating and moderating effects of stress and school bonding. *J Adolesc Health*. 2009; 44(2):146–152. [PubMed: 19167663]
53. Campbell SB, Pierce EW, Moore G, Marakovitz S. Boys’ externalizing problems at elementary school age: pathways from early behavior problems, maternal control, and family stress. *Dev Psychopathol*. 1996; 8(4):701–719.
54. Stith SM, Liu T, Davies LC, et al. Risk factors in child maltreatment: a meta-analytic review of the literature. *Aggress Violent Behav*. 2009; 14(1):13–29.
55. Ouyang L, Fang X, Mercy J, Perou R, Grosse SD. Attention-deficit/hyperactivity disorder symptoms and child maltreatment: a population-based study. *J Pediatr*. 2008; 153(6):851–856. [PubMed: 18619612]
56. Sullivan PM, Knutson JF. Maltreatment and disabilities: a population-based epidemiological study. *Child Abuse Negl*. 2000; 24(10):1257–1273. [PubMed: 11075694]

**Table 1**

National Survey of Children's Health 2007 (2007 NSCH) question, family functioning indicator and protective factor, study variable name, response choices, and dichotomized variable

2007 NSCH question	Family functioning indicator or protective factor	Variable	Answer options	Dichotomous variable
During the past week, on how many days did all the family members who live in the household eat a meal together?	Family routines	Family mealtimes	0–7	0–4, 5–7
How well can you and [your child] share ideas or talk about things that really matter?	Parent–child communication	Share ideas with the child	Very well, somewhat well, not very well or not very well at all	Very well; Somewhat well, not very well or not very well at all
In general, how well do you feel like you are coping with the day to day demands of [parenthood/raising children]?	Parental coping	Coping with parenting demands	Very well, somewhat well, not very well or not very well at all	Very well; Somewhat well, not very well or not very well at all
During the past month, how often have you felt [your child] is much harder to care for than most children [his/her] age?	Parenting aggravation	Harder to care for the child	Never, rarely, sometimes, usually or always	Never, rarely, sometimes; usually or always
During the past month, how often have you felt [he/she] did things that really bother you a lot?	Parenting aggravation	Bothered by the child	Never, rarely, sometimes, usually or always	Never, rarely, sometimes, usually or always
During the past month, how often have you felt angry with [him/her]?	Parenting aggravation	Angry with the child	Never, rarely, sometimes, usually or always	Never, rarely, sometimes, usually or always
Is there someone that you can turn to for day to day emotional help with [parenthood/raising children]?	Emotional support	Emotional support	Yes, no	Yes, no
People in this neighborhood help each other out	Social capital	Helping out	Definitely agree, somewhat agree, somewhat disagree, definitely disagree	Moderate to high social capital was defined as a composite score of <7
We watch out for each other's children in this neighborhood	Social capital	Neighborhood monitoring	Definitely agree, somewhat agree, somewhat disagree, definitely disagree	
There are people I can count on in this neighborhood	Social capital	Count on	Definitely agree, somewhat agree, somewhat disagree, definitely disagree	
If my child were outside playing and got hurt or scared, there are adults nearby who I trust to help my child.	Social capital	Trusted neighbors	Definitely agree, somewhat agree, somewhat disagree, definitely disagree	
Would you say that, in general, (mother/father) mental and emotional	Parental mental health	Parental mental health	Excellent, very good, good, fair, or poor	Excellent, very good, good; fair, or poor

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<b>2007 NSCH question</b>	<b>Family functioning indicator or protective factor</b>	<b>Variable</b>	<b>Answer options</b>	<b>Dichotomous variable</b>
health is excellent, very good, good, fair, or poor				

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**Table 2**

Percentage distribution and prevalence ratios of child and family demographic factors among children with Tourette syndrome (including co-occurring conditions) and without Tourette syndrome from the National Survey of Children's Health 2007

	No Tourette syndrome % (CI) (unweighted <i>N</i> = 63,809)	All Tourette syndrome % (CI) (unweighted <i>N</i> = 225)	Prevalence ratio (CI)
Age (years)			
6–11	48.6 (47.6–49.6)	30.6 (20.1–43.5)	Referent
12–17	51.4 (50.4–52.4)	69.5 (56.5–79.9)	1.4 (1.1–1.6)
Sex			
Female	48.9 (48.0–49.9)	24.6 (13.7–40.3)	Referent
Male	51.1 (50.1–52.1)	75.4 (59.8–86.4)	1.5 (1.2–1.8)
Race			
Non-white	27.8 (26.9–28.8)	16.9 (9.0–29.5)	Referent
White	72.2 (71.3–73.1)	83.1 (70.5–91.0)	1.2 (1.0–1.3)
Hispanic ethnicity			
Hispanic	19.3 (18.3–20.3)	10.3 (5.7–18.1)	Referent
Non-Hispanic	80.7 (79.7–81.7)	89.7 (81.9–94.3)	1.1 (1.0–1.2)
Family structure <sup>a</sup>			
2 biological/adoptive parents	62.7 (61.7–63.7)	59.7 (45.1–72.7)	Referent
2 parents (1 step-parent)	10.2 (9.6–10.8)	7.4 (4.1–13.1)	0.8 (0.4–1.5) <sup>c</sup>
Single parent	20.1 (19.3–20.9)	16.3 (10.0–25.2)	0.9 (0.5–1.5) <sup>c</sup>
Other family structure	7.1 (6.6–7.6)	16.6 (6.9–34.8) <sup>c</sup>	2.5 (0.9–6.8) <sup>c</sup>
Highest level of education achieved by parent in household			
High school diploma or less	32.8 (31.8–33.8)	38.8 (25.1–54.4)	Referent
At least some college or technical school	67.2 (66.2–68.2)	61.2 (45.6–74.9)	0.9 (0.7–1.2)
Household income			
≤200% of federal poverty level	37.2 (36.2–38.2)	37.1 (23.3–53.4)	Referent
>200% of federal poverty level	62.8 (61.8–63.8)	62.9 (46.6–76.7)	1.0 (0.8–1.3)
Co-occurring MEB <sup>b</sup>			
Any co-occurring MEB	16.4 (15.7–17.1)	78.7 (68.4–86.3)	4.8 (4.3–5.4)
Any internalizing condition	7.7 (7.2–8.3)	52.5 (38.4–66.2)	6.8 (5.1–9.0)
Depression	4.7 (4.3–5.1)	36.2 (22.7–52.3)	7.8 (5.0–11.9)
Anxiety	5.4 (5.0–5.9)	39.7 (27.4–53.5)	7.3 (5.2–10.4)
Any externalizing condition	12.5 (11.9–13.1)	70.1 (57.1–80.5)	5.6 (4.7–6.7)
Attention-deficit/hyperactivity disorder	10.6 (10.0–11.2)	63.8 (50.5–75.3)	6.0 (4.9–7.4)
Behavioral or conduct problems	5.3 (4.9–5.8)	43.0 (28.9–58.2)	8.1 (5.7–11.7)
Autism spectrum disorder	1.8 (1.5–2.1)	26.2 (14.4–42.8)	14.8 (8.4–26.1)

MEB = mental, emotional, or behavioral condition; CI = confidence interval.

<sup>a</sup>Family structure PRs calculated using logistic regression (all others analyzed with chi-square tests).

<sup>b</sup>Percentages for no MEBs not reported.

<sup>c</sup>Relative standard error (RSE) > 30% (Range = 31–42%); RSEs for estimates and PRs are reported if over 30% because these estimates may not be reliable.

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**Table 3**

Percentage distribution and prevalence ratios of family functioning indicators among children with and without Tourette syndrome from the National Survey of Children's Health 2007

	No Tourette syndrome % (CI)	Tourette syndrome % (CI)	Adjusted prevalence ratio, aPR (CI)
Family mealtimes			
0–4 times a week	37.7 (36.8–38.7)	40.2 (26.9–55.1)	1.0 (0.7–1.5)
5–7 times a week	62.3 (61.3–63.2)	59.8 (44.9–73.1)	
Share ideas with the child			
Somewhat well/not very well/not well at all	30.2 (29.3–31.1)	48.7 (34.6–63.0)	
Very well	69.8 (68.9–70.7)	51.3 (37.0–65.4)	0.8 (0.6–1.0)
Coping with parenting demands			
Somewhat well/not very well/not well at all	43.1 (42.1–44.1)	49.6 (35.5–63.8)	
Very well	56.9 (55.9–57.9)	50.4 (36.2–64.5)	0.9 (0.7–1.2)
Harder to care for the child			
Usually/always	6.7 (6.1–7.3)	25.7 (14.0–42.3)	4.2 (2.4–7.5)
Sometimes/rarely/never	93.3 (92.8–93.9)	74.3 (57.7–86.0)	
Bothered by the child			
Usually/always	5.4 (5.0–5.9)	12.9 (7.9–20.3)	2.5 (1.5–4.0)
Sometimes/rarely/never	94.6 (94.1–95.1)	87.1 (79.7–92.1)	
Angry with the child			
Usually/always	3 (2.6–3.4)	13.9 (4.5–35.7) <sup>a</sup>	4.9 (1.7–14.6) <sup>a</sup>
Sometimes/rarely/never	97 (96.6–97.4)	86.1 (64.3–95.5)	
Aggravation index			
High	7.3 (6.7–7.9)	26.3 (14.6–42.8)	3.8 (2.2–6.6)
Low	92.8 (92.2–93.3)	73.7 (57.2–85.5)	
Emotional help			
Yes	87.1 (86.3–87.8)	83.2 (70.4–91.2)	0.9 (0.8–1.04)
No	12.9 (12.2–13.7)	16.8 (8.8–29.6) <sup>a</sup>	
Social capital			
Moderate to high	73.7 (72.7–74.6)	69.6 (53.1–82.2)	0.9 (0.7–1.1)
Low	26.3 (25.4–27.3)	30.4 (17.8–46.9)	
Maternal mental health			
Excellent, very good, good	92.2 (91.6–92.8)	80.7 (66.7–89.8)	0.9 (0.7–1.00)
Fair, poor	7.8 (7.3–8.4)	19.3 (10.2–33.3)	
Paternal mental health			
Excellent, very good, good	94.6 (94.0–95.2)	88.9 (70.6–96.4)	0.9 (0.8–1.1)
Fair, poor	5.4 (4.8–6.0)	11.1 (3.6–29.4) <sup>a</sup>	

Adjusted for age, sex, race, ethnicity (95% CI); CI = Confidence Interval.

<sup>a</sup> Relative standard error (RSE) > 30% (Range = 32–56%); RSEs for estimates and aPRs are reported if over 30% because these estimates may not be reliable.

**Table 4**

Adjusted prevalence ratios (aPR) for Tourette syndrome and co-occurring mental, emotional and behavioral (MEB) conditions predicting the parenting aggravation index score controlling for each other and demographic variables, National Survey of Children's Health 2007

Conditions included in separate models predicting parenting aggravation index	Tourette syndrome aPR (CI)	Co-occurring MEB condition aPR (CI)
Model including Tourette syndrome, but not MEBs as independent variables (original aPR)	3.8 (2.2–6.6)	
Model including Tourette syndrome plus:		
Any MEB <sup>a</sup>	1.7 (0.9–3.2) <sup>b</sup>	5.2 (4.5–6.1)
Any externalizing condition	1.7 (0.9–3.1) <sup>b</sup>	5.5 (4.8–6.4)
Attention-deficit/hyperactivity disorder	1.9 (1.0–3.6) <sup>b</sup>	4.4 (3.8–5.1)
Behavioral or conduct problems	1.5 (0.8–2.9) <sup>b</sup>	7.6 (6.5–8.8)
Any internalizing condition	1.9 (1.0–3.6) <sup>b</sup>	4.3 (3.7–5.1)
Depression	2.2 (1.1–4.2) <sup>b</sup>	4.4 (3.6–5.3)
Anxiety	2.3 (1.0–5.3) <sup>b</sup>	3.9 (3.3–4.6)
Autism spectrum disorder	2.4 (1.4–4.0)	4.7 (3.6–6.1)

*Note:* Each row represents a separate model; PR adjusted for age, sex, race, ethnicity (95% CI); CI = Confidence Interval.

<sup>a</sup> Mental, emotional or behavioral problems include attention-deficit/ hyperactivity disorder, behavioral or conduct problems, depression, anxiety and autism spectrum disorders.

<sup>b</sup> Relative standard error (RSE) > 30% (Range = 32–44%); RSEs for aPRs are reported if over 30% because these estimates may not be reliable.