Profile Of Tourette Syndrome In Children And Adolescents From A Tertiary Care Child Psychiatry Centre In India

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Abstract

Introduction: Tourette Syndrome is a type of tic disorder that is not as rare as it was once thought to be. There is a paucity of literature on Tourette syndrome in children and adolescents in the Indian setting. The objectives of this study were to elucidate sociodemographic and clinical profile of children and adolescents with Tourette syndrome.

Material And Method: The present study is a retrospective chart review of children and adolescents up to age 18 years diagnosed with Tourette Syndrome in a tertiary care centre, India. This sample is from a subset of a larger sample of case records of patients with a diagnosis of tic disorder, coded as F 95, according to ICD 10 from 1st January 2014 to 31st December 2017 (4 years duration).

Results: The sample included 28 children. All of them were male, and the mean age of onset was 8.2 years. The period prevalence of Tourette syndrome was 0.22%. The entire sample received pharmacotherapy and the most common medication prescribed was Risperidone followed by Clonidine, Aripiprazole, Haloperidol, and Tetrabenazine. Majority (64.28%) of the sample had a comorbid condition. The most common comorbid conditions were Hyperkinetic Disorders followed by Obsessive Compulsive Disorder Specific and Specific Developmental Disorder of Scholastic skills.

Conclusion: Tourette syndrome can be a disabling condition in school-going children. There is a need for population-based studies for estimating its prevalence in Indian subcontinent, and there is also a great need to increase awareness and sensitize the public to seek help to improve the quality of life of children and adolescents with Tourette syndrome.

Keywords: Adolescent Child, India, Tic, Tourette

INTRODUCTION

According to DSM 5 classification, Tourette syndrome (TS) is a neurodevelopmental disorder characterized by multiple motor and one or more vocal/phonic tics, lasting at least one year, with onset during childhood or adolescence.1 The first patient with Tourette’s syndrome was described in the medical literature by Itard in 1825.2 Georges Edouard Albert Brutus de la Tourette, published a cohort of nine cases in 1885.3 For many the experience of tics may only be transient, but for those with TS it is chronic and may be a debilitating mental health problem. Although initially considered to be rare, TS is more common than previously expected, with a suggested overall prevalence of 1/200 in children. TS is reported worldwide and is more common in boys than in girls with a ratio of 1.6:1 to 9:1.4,5 Most patients will have a complete or nearly complete remission of the disorder after 21 years of age. In 10-20% of cases, TS symptoms fluctuate, persist, or even worsen.6,7,8 The available evidence suggests that neurodevelopmental comorbidities in TS are the rule, rather than the exception with attention deficit/hyperactivity disorder (ADHD) being the most frequent.9 In addition, the disorder is often accompanied by obsessive-compulsive disorder (OCD), self-injurious behavior, depression, or specific learning disabilities. The use of medications is reserved for moderate to
severe tics causing severe impairment in quality of life or when medication responsive psychiatric comorbidities are present that target both tic symptoms and comorbid conditions. A case series reported in 1992 from India suggested cross-cultural similarity in symptomatology, perhaps emphasizing the biological factors in the aetiology of Tourette syndrome. However, there is limited literature on Tourette syndrome in children and adolescents in the Indian setting. Therefore, we studied the sociodemographic and clinical profile of children and adolescents with Tourette syndrome who presented to the Child and Adolescent Psychiatry (CAP) services of National Institute of Mental Health and Neuro Sciences (NIMHANS), Bengaluru.

MATERIAL AND METHOD
The study design is a retrospective chart review of the cases that presented from 1st January 2014 to 31st December 2017 (four years). This sample is from a subset of a larger sample of case records of patients with a diagnosis of tic disorder, coded as F95-F95.9 according to ICD 10. Study site was the Department of child and adolescent psychiatry (DCAP), NIMHANS, Bengaluru, India which provides services to children and adolescents up to age 18. The DCAP at NIMHANS caters to children and adolescents up to age 18. There is no definite catchment area for the CAP services although a majority is from the state of Karnataka and the neighboring states. All patients undergo a detailed clinical evaluation according to a prespecified topical format based on an unstructured psychiatric interview with both the child and the parent(s). The topical format includes sociodemographic information, presenting complaints, a history of present illness in chronological order of complaints, developmental history, temperament, family history, treatment details, and a mental status examination that involves a direct interview with the child or the adolescent. Diagnosis and management are confirmed by a consultant child and adolescent psychiatrist.

Ethics committee approval was obtained from Institutional Ethics Committee with reference number EC(BEH.SC.DIV) 13th Meeting/2018 of the Institute. The department offers out-patient and in-patient services to children and adolescents up to age 18. All patients are evaluated in detail by junior residents using a semi-structured psychiatric interview. Typically the informants are child’s parent(s). Diagnosis and management are confirmed either by a consultant or a senior resident.

A total of 85 case records with a diagnosis of Tic Disorder as per ICD 10 were reviewed. Out of the 85 case records with Tic Disorder there were 28 children/adolescents with Tourette syndrome. Data was analyzed using descriptive statistics.

RESULT
A total of 12,662 patients attended the out-patient services during the 4-year period. Eighty-five children were identified as having a diagnosis of Tic disorder, out of which 28 (32.94%) of them had a diagnosis of Tourette syndrome. The 4-year prevalence of TS in child and adolescent psychiatry population at this centre was 0.22%.

| Characteristics   | Frequency (%) |
|-------------------|--------------|
| **Age at presentation** (in years) |  |  |
| Less than 12      | 12 (42.8)    |
| More than 12      | 16 (57.1)    |
| **Gender**        |  |  |
| Male              | 28 (100)     |
| Female            | -            |
| **Education**     |  |  |
| Primary school    | 10 (35.7)    |
| Middle school     | 10 (35.7)    |
| High school       | 8 (28.5)     |
| **Family type**   |  |  |
| Nuclear           | 19 (67.8)    |
| Joint             | 9 (32.1)     |
| **Locality**      |  |  |
| Urban             | 21 (75)      |
| Rural             | 7 (25)       |

The mean age of presentation was 11.75 (SD-2.84). More than half of the sample (57.1%, n=16) presented to our service after 12 years of age. All patients were male and were going to school. Majority (67.8%, n=19) were from a nuclear family set up and three-fourths (n=21) were from an urban locality. Family history of tics was present in one patient. Table 1 shows sociodemographic profile of patients.
Table 2: Phenomenology Of Tics

| Motor tics                          | Frequency | Vocal tics                  | Frequency |
|-------------------------------------|-----------|----------------------------|-----------|
| Shoulder shrugging                  | 14        | Throat clearing            | 14        |
| Eye blinking                        | 12        | Coughing                   | 7         |
| Head jerk                           | 9         | Repeating words            | 4         |
| Facial grimacing                    | 8         | Sniffing                   | 4         |
| Flexing and extending the arm       | 7         |                            |           |
| Chin touching shoulder              | 3         |                            |           |
| Feet tapping                        | 2         |                            |           |
| Jerky movement of arms              | 1         |                            |           |
| Lip smacking                        | 1         |                            |           |
| Extension of arm                    | 1         |                            |           |
| Nose twitching                      | 1         |                            |           |
| Neck jerking                        | 1         |                            |           |
| Head nodding                        | 1         |                            |           |

Table 2 illustrates phenomenology of motor and vocal tics in the sample. The most common type of motor tic was shoulder shrugging (n=14) followed by eye blinking (n=12), head jerk (n=9), facial grimacing (n=8) and flexing and extending the arm (n=7). The most common type of vocal tic was throat clearing (n=14), coughing (n=7) and repeating words and sniffing (n=4).

Table 3: Clinical Profile Of Patients (N=28)

| Characteristics                  | Frequency (%) | Mean ±SD       |
|----------------------------------|---------------|----------------|
| Age of onset (years)             |               |                |
| Preschool (less than 5 years)    | 1 (3.5)       | 8.2±2.1        |
| School going (6-12 years)        | 25 (89.2)     |                |
| Adolescence (12-18 years)        | 2 (7.14)      |                |
| Duration of tics (in months)     |               | 41.14±25.0     |
| 12-60 months (1-5 years)         | 21 (75)       |                |
| >60 months (5yrs)                | 7 (25)        |                |
| Medications                      |               |                |
| Risperidone                      | 16 (57.1)     |                |
| Clonidine                        | 6 (21.4)      |                |
| Aripiprazole                     | 2 (7.14)      |                |
| Haloperidol                      | 2 (7.14)      |                |
| Tetrabenazine                    | 2 (7.14)      |                |
| Duration of follow up (in months)|               | 9.9±8.22       |

Majority of patients (89.2%; n=25) had onset of tics between 6 to 12 years of age i.e. middle childhood, with the mean age of onset being 8.2 (SD-2.1). Three-fourths of the sample (n=21) had duration of tics between 1 to 5 years. The most common medication prescribed was Risperidone (57.1%; n=16) followed by Clonidine (21.4%; n=6), Aripiprazole, Haloperidol and Tetrabenazine. The mean duration of follow up of the sample was 9.9(SD-8.22) months. Clinical profile of patients is summarized in Table 3.

Table 4: Pattern of comorbidities in patients with Tourette Syndrome (n=28)

| Comorbidities                      | Frequency (%) |
|------------------------------------|---------------|
| Hyperkinetic disorders             | 8 (28.57)     |
| OCD                                | 4 (14.28)     |
| SDDSS                              | 3 (10.7)      |
| Hyperkinetic disorders + OCD       | 2 (7.14)      |
| Hyperkinetic disorders + Social Phobia | 1 (3.57)   |
| Conduct disorder                   | 1 (3.57)      |
| Pervasive developmental disorders  | 1 (3.57)      |
| Dysthymia                          | 1 (3.57)      |

* OCD - Obsessive compulsive disorder, SDDSS – Specific Developmental Disorder of Scholastic Skills

Table 4 illustrates the comorbidities. Majority (64.28%, n=18) of the sample had a comorbid condition. Most common comorbid conditions were Hyperkinetic disorders (28.75%; n=8), followed by OCD (14.28%; n=4) and SDDSS (10.7%; n=3).

DISCUSSION:

Crude prevalence of 0.22% is lower than 0.4-1% reported in the west. While the estimates in west are from epidemiological studies, ours is a clinic-based population. This explains the lower prevalence in our sample. The National Mental Health Survey (2015-2016) did not mention prevalence of tic disorders, so prevalence in general population in the Indian context is unknown. TS has a typical course of progression and is known to peak in severity between 10-12 years and the mean age of presentation of our cases was 11.75 years. Age at help-seeking corresponds to the age of worsening of the condition. While literature suggests a male to
female ratio of 1.6:1 to 9:1, all the cases in our sample were males. Perhaps, this can be explained by referral bias. The majority of the patients were from nuclear families and an urban setting. The hospital is situated in one of the major urban cities of India and its location can explain the predominant sociodemographic characteristics of the sample.

Majority of the sample (89.2%) had an onset between 6-12 years of age i.e. middle childhood, as compared to the typical onset between 4-6 years described in literature. This difference in age of onset could partly be due to recall bias with a possible tendency to underreport subtle tics in the initial stages. However, there here is a need to explore if this is true to population in the Indian sub-continent.

Tics can manifest as any movement or sound that human body can make, which can range from simple/inarticulate movements/sounds to very complicated forms. The phenomenology of our sample is similar to that reported across the globe, typically characterised by eye-blinking, shoulder shrugging, head jerks, facial grimacing, throat clearing, coughing, sniffing and repeating words. The more complex presentations such as flexing extending arm and feet tapping were also noted as biological underpinnings. Around three fourth of the sample had duration of 1-5 years of illness at the time of presentation which suggests delay in seeking services but detail information about visit to other centres or services could not be obtained and cannot be ruled out.

Risperidone followed by clonidine were the commonly prescribed medications, which is in line with the European Child and Adolescent Psychiatry guidelines 2011, that haloperidol, pimozide, risperidone, alpha-2 agonists such as clonidine and guanfacine have level of evidence A for use in tic disorders. Haloperidol and pimozide are the only two FDA approved treatments for tics, although they are not currently recommended as the first-line pharmacotherapy because of their adverse side-effect profile. Treatment should begin with psychoeducation followed by pharmacotherapy or non-pharmacotherapeutic interventions such as habit reversal training (HRT). Apart from medications, in our sample psychoeducation with HRT was provided to all.

The average duration of follow up of approximately 10 months in this population is not long enough, given its worsening course described in this age-group. This suggests that often children and families are lost to follow-up. Therefore, there is a need to sensitize the public/improve awareness about this condition in the population, so it can help in identification of children enduring this illness and offer treatment accordingly.

CONCLUSION:
Tourette syndrome can be disabling in school-going children that can affect their general well-being and school performance. However, its prevalence in general population remains unknown. This calls for studies for example in schools to estimate its true prevalence. The later age at onset noted in our sample has important clinical and research implications. This finding needs to be replicated in diverse and larger population studies. The short duration of follow-up is alarming for a clinician, there is a need to improve awareness about this condition in general population.

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