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Diagnostic Hierarchy of Tic Disorders in Real-World Clinical Practice

Yeeji Sung[1,2] and Soon-Beom Hong[1,2,3]

[1]Department of Psychiatry, Seoul National University Hospital, Seoul, Korea
[2]Department of Psychiatry, Seoul National University College of Medicine, Seoul, Korea
[3]Institute of Human Behavioral Medicine, Seoul National University Medical Research Center, Seoul, Korea

Objectives: According to the 10th revision of the International Classification of Diseases, the main categories of tic disorders (F95.0, F95.1, and F95.2) follow a diagnostic hierarchy based on the duration and diversity of tic symptoms. The present study investigated the use of this diagnostic hierarchy in real-world clinical practice.
Methods: Based on the National Health Insurance Service-National Health Information Database, the diagnosis of transient tic disorder (F95.0) made after a diagnosis of chronic motor or vocal tic disorder (F95.1) or Tourette's syndrome (F95.2) and diagnosis of chronic motor or vocal tic disorder (F95.1) made after a diagnosis of Tourette's syndrome (F95.2) were referred to as type A errors. The diagnosis of transient tic disorder (F95.0) repeated after a period of >12 months was referred to as type B error. Demographic and clinical differences according to the diagnostic error types were analyzed using analysis of variance, Student's t-tests, and chi-squared tests.
Results: Most participants (96.5%) were without errors in the diagnosis of tic disorders. Higher proportions of males (p=0.005) and antipsychotic prescriptions (p<0.001) were observed in patients with type A or B diagnostic errors. A higher proportion of health insurance holders was observed among those with type A errors (p=0.027).
Conclusion: Errors were absent in majority of the tic diagnoses in real-world clinical practice in terms of the diagnostic hierarchy.

Keywords: Tic disorders; Tourette syndrome; ICD-10; Diagnostic errors.

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Address for correspondence: Soon-Beom Hong, Division of Child and Adolescent Psychiatry, Department of Psychiatry, Seoul National University College of Medicine, 103 Daehak-ro, Jongno-gu, Seoul 03080, Korea
Tel: +82-2-2072-4208, Fax: +82-2-747-2471, E-mail: hsbmdmore@hanmail.net

INTRODUCTION

According to the 10th revision of the International Classification of Diseases (ICD-10) [1], the diagnosis of tic disorders includes transient tic disorder (F95.0), chronic motor or vocal tic disorder (F95.1), combined vocal and multiple motor tic disorder (de la Tourette's syndrome; F95.2), other tic disorders (F95.8), and tic disorder, unspecified (F95.9). The first three main categories of tic disorders follow a diagnostic hierarchy. Transient tic disorder (F95.0) is diagnosed when one or more motor or vocal tics have been present for ≤12 months. Chronic motor or vocal tic disorder (F95.1) is diagnosed when one or more motor or vocal tics, but not both, have been present for >12 months. Tourette's syndrome (F95.2) is diagnosed when both motor and vocal tics have been present for >12 months. Accordingly, a diagnosis of transient tic disorder (F95.0) cannot be made after a diagnosis of chronic motor or vocal tic disorder (F95.1) or Tourette's syndrome (F95.2), and a diagnosis of chronic motor or vocal tic disorder (F95.1) cannot be made after a diagnosis of Tourette's syndrome (F95.2). In summary, a hierarchical order is present in terms of the diversity (i.e., motor tics, vocal tics, or both) and duration (i.e., ≤12 or >12 months) of tic symptoms.

However, insufficient empirical evidence exists for substantiating the differentiation of diagnostic categories based on these three conditions. A previous study suggested that most children with transient tic disorder (F95.0) continued to have symptoms after 12 months, thus meeting the criteria for either chronic motor or vocal tic disorder (F95.1) or Tourette's syndrome (F95.2) [2]. Chronic motor or vocal tic disorder (F95.1) has also been proposed to be a mild form of Tourette's syndrome (F95.2) because the two groups do not differ in any of the clinical or demographic variables except tic severity, suggesting a symptom severity continuum [3].

Furthermore, diagnostic criteria should not only be evidence-based but also clinically practical. For example, the age criterion for attention-deficit/hyperactivity disorder (ADHD) diagnosis in the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition was revised from 7 years to 12 years

with the practical consideration that, unlike children, adults with ADHD often do not have a clear memory of their pre-7-year-old state when they visit the clinic without their parents [4,5]. Similarly, if difficulties exist in adhering to the established diagnostic criteria for tic disorders in clinical settings, the diagnostic criteria should be revised for practical purposes. Therefore, it is important to investigate whether real-world diagnoses align with the diagnostic criteria.

To conduct such investigations, it is generally necessary to compare data that reflect the diagnostic practices of clinicians in clinical settings, such as health insurance data, with systematically collected diagnostic interview results. Conducting such research requires substantial resources, including personnel and costs. However, in the case of tic disorders, it is possible to verify whether the diagnoses made by clinicians, as collected in health insurance claims data, are consistent with their own hierarchical criteria without the need for additional resources. This provides a unique opportunity to indirectly validate the practicality of the diagnostic hierarchy of tic disorders.

How well medical practitioners comply with the diagnostic hierarchy when diagnosing tic disorder is currently unclear. This study aimed to investigate the use of this hierarchy in real-world clinical practice. Specifically, we examined whether the hierarchy of tic disorder diagnoses is consistently followed in real-world clinical practice. Considering that the accuracy of diagnostic codes in claims databases is generally not very high [6], we hypothesized that there may be a significant number of diagnostic errors contradicting the hierarchy of tic disorder diagnoses.

METHODS

Data acquisition

Data were obtained from the National Health Insurance Service (NHIS)-National Health Information Database, which was extracted through the National Health Insurance Sharing Service with approval from the NHIS (No. NHIS-2021-1-637). The diagnosis of tic disorders was defined as F95 (F95.0, F95.1, F95.2, F95.8, and F95.9) based on ICD-10, among which transient tic disorder (F95.0), chronic motor or vocal tic disorder (F95.1), and Tourette's syndrome (F95.2) were of main interest. The study participants were all individuals with a principal diagnosis of tic disorder made between 2018 and 2020. In addition to the diagnostic categories of tic disorders, information on the age, sex, type of insurance, and prescriptions of aripiprazole, risperidone, or haloperidol was obtained. This study was exempted from review by the Institutional Review Board of Seoul National University Hospital (E-2210-099-1371).

Diagnostic errors

According to the ICD-10, a diagnosis of transient tic disorder (F95.0) cannot be made after a diagnosis of chronic motor or vocal tic disorder (F95.1) or Tourette's syndrome (F95.2), and a diagnosis of chronic motor or vocal tic disorder (F95.1) cannot be made after a diagnosis of Tourette's syndrome (F95.2). We termed these violations in the diagnostic definition as type A error. In addition, a diagnosis of transient tic disorder (F95.0) could not be repeated after a period of >12 months. We termed this violation in the diagnostic definition as type B error. The number of diagnostic errors for each study participant was counted.

Statistical analyses

First, age was compared among the five categories of tic disorders using analysis of variance, and age range, sex, and type of insurance were compared using the chi-square test. Pairwise comparisons between each pair of groups were conducted using Student's t-tests for continuous variables (e.g., age) and chi-square tests for categorical variables (e.g., age range, sex, and type of insurance). Second, we aggregated the number of participants based on the frequency of each diagnostic error in a single participant using simple counting. Third, age, sex, type of insurance, and prescriptions of aripiprazole, risperidone, or haloperidol were compared according to the diagnostic error types using the Student's t-test and chi-square test for continuous and categorical variables, respectively. Initially, comparisons were made between individuals with either type A or B errors and those without any errors. Then, separate analyses were conducted based on the presence or absence of type A and B errors, respectively. Fourth, we aggregated whether errors of each type occurred within a single institution or across two different institutions using simple counting. Statistical tests were performed using R software (R Foundation for Statistical Computing, Vienna, Austria), and statistical significance was tested at an alpha value of 0.05.

RESULTS

Participant characteristics

Age, sex, and type of insurance of the 38084 participants with different tic disorders are presented in Table 1. The variables were collected based on when the principal diagnosis of tic disorder was first made between 2018 and 2020.

Among the three main diagnostic categories, participants with transient tic disorder (F95.0) had the youngest mean age (10.33 years) and those with chronic motor or vocal tic disorder (F95.1) had the oldest mean age (17.65 years). Interestingly, a higher proportion of adults was observed among

those diagnosed with chronic motor or vocal tic disorder (F95.1) (31.9%) than among those diagnosed with Tourette's syndrome (F95.2) (21.7%), although the total number of adults and youths with Tourette's syndrome (F95.2) was far greater. The proportion of males tended to increase in line with the diagnostic hierarchy, starting from transient tic disorder (F95.0) (71.9%) to chronic motor or vocal tic disorder (F95.1) (78.7%), followed by Tourette's syndrome (F95.2) (80.6%). The pairwise comparison results between each pair of groups are presented in Supplementary Table 1 (in the online-only Data Supplement).

Distribution of the diagnostic errors

The majority of participants (96.5%) were without errors in their diagnosis of tic disorders (Supplementary Table 2 in the online-only Data Supplement). Only 2.5% and 1.1% of the participants had type A and B errors, respectively. The number of participants with type A errors was comparable to the number of their type B error counterparts. For type A errors, most participants (n=849) entailed a single error. In contrast, for type B errors, most participants (n=255) entailed four or more errors.

Characteristics of the participants according to the error types

Participants with tic disorders whose diagnoses entailed either type A or B error included a higher proportion of males (p=0.005) and a higher proportion of prescriptions for antipsychotic medications (p<0.001 for all three medications) (Table 2). When each error type was examined separately, the higher proportion of males remained significant only for participants with error type A (p=0.039) (Table 3). In addition, a higher proportion of health insurance holders was detected only among participants with type A error (p=0.027) (Table 3). A higher proportion of prescriptions of antipsychotic medications remained significant in the participants with types A, B, or both errors (Tables 2-4).

Additional analyses

Additionally, we examined the patterns of errors made in a single institution and two different institutions. The majority of errors made in the same medical institution were type B (94.9%) (Supplementary Table 3 in the online-only Data Supplement). However, the majority of errors jointly made in two different medical institutions were type A (75.6%), among which the most common was the diagnosis of chronic motor

Table 1. Demographic characteristics of participants with tic disorders

Variable	F95.0 (n=5062)	F95.1 (n=5200)	F95.2 (n=14799)	F95.8 (n=2336)	F95.9 (n=10687)	p
Age (yr)*	10.33±10.84	17.65±12.42	15.32±9.73	16.54±16.15	16.00±15.70	<0.001
Age range†						<0.001
0–9 yr	3590 (70.9)	1440 (27.7)	4140 (28.0)	1104 (47.3)	5227 (48.9)	
10–19 yr	1017 (20.1)	2102 (40.4)	7437 (50.3)	641 (27.4)	2909 (27.2)	
20–29 yr	196 (3.9)	941 (18.1)	2120 (14.3)	235 (10.1)	1004 (9.4)	
≥30 yr	259 (5.1)	717 (13.8)	1102 (7.4)	356 (15.2)	1547 (14.5)	
Sex, male†	3640 (71.9)	4093 (78.7)	11930 (80.6)	1732 (74.1)	7767 (72.7)	<0.001
Insurance, health insurance†	4961 (98.0)	5002 (96.2)	14284 (96.5)	2240 (95.9)	10341 (96.8)	<0.001

Values are presented as mean±standard deviation or number (%). Insurance status was dichotomized into health insurance and medical aid. F95.0: Transient tic disorder. F95.1: Chronic motor or vocal tic disorder. F95.2: Tourette's syndrome. F95.8: Other tic disorders. F95.9: Tic disorder, unspecified. *analysis of variance; †chi-squared test

Table 2. Characteristics of participants with tic disorders whose diagnoses entailed error type A or B

Variable	Patients without error type A or B (n=36751)	Patients with error type A or B (n=1333)	p
Age (yr)*	15.24±12.84	15.28±10.60	0.902
Sex, male†	28098 (76.5)	1064 (79.8)	0.005
Insurance, health insurance†	35530 (96.7)	1298 (97.4)	0.186
Antipsychotics prescriptions, yes†			
Aripiprazole	11227 (30.5)	721 (54.1)	<0.001
Risperidone	4165 (11.3)	321 (24.1)	<0.001
Haloperidol	3660 (10.0)	276 (20.7)	<0.001

Values are presented as mean±standard deviation or number (%). Insurance status was dichotomized into health insurance and medical aid. Prescriptions of antipsychotics were dichotomized into yes and no. *Student's t-test; †chi-squared test

Table 3. Characteristics of participants with tic disorders whose diagnoses entailed error type A

Variable	Patients without error type A (n=37127)	Patients with error type A (n=957)	p
Age (yr)*	15.24±12.83	15.42±9.80	0.658
Sex, male†	28402 (76.5)	760 (79.4)	0.039
Insurance, health insurance†	35890 (96.7)	938 (98.0)	0.027
Antipsychotics prescriptions, yes†			
Aripiprazole	11364 (30.6)	584 (61.0)	<0.001
Risperidone	4229 (11.4)	257 (26.9)	<0.001
Haloperidol	3719 (10.0)	217 (22.7)	<0.001

Values are presented as mean±standard deviation or number (%). Insurance status was dichotomized into health insurance and medical aid. Prescriptions of antipsychotics were dichotomized into yes and no. *Student's t-test; †chi-squared test

Table 4. Characteristics of participants with tic disorders whose diagnoses entailed error type B

Variable	Patients without error type B (n=37667)	Patients with error type B (n=417)	p
Age (yr)*	15.24±12.77	14.84±12.24	0.519
Sex, male†	28828 (76.5)	334 (80.1)	0.099
Insurance, health insurance†	36428 (96.7)	400 (95.9)	0.449
Antipsychotics prescriptions, yes†			
Aripiprazole	11786 (31.3)	162 (38.8)	0.001
Risperidone	4409 (11.7)	77 (18.5)	<0.001
Haloperidol	3868 (10.3)	68 (16.3)	<0.001

Values are presented as mean±standard deviation or number (%). Insurance status was dichotomized into health insurance and medical aid. Prescriptions of antipsychotics were dichotomized into yes and no. *Student's t-test; †chi-squared test

or vocal tic disorder (F95.1) in the second institution after a diagnosis of Tourette's syndrome (F95.2) had been made in the first institution (55.3%).

DISCUSSION

A relatively small number of errors was observed that violated the diagnostic hierarchy of tic disorders. Type A errors mainly occurred when the patient changed medical institutions, whereas most type B errors occurred in a single institution.

These errors in the diagnostic hierarchy were more commonly encountered in males and those who were prescribed antipsychotic medications. Although some studies observed no differences in the severity between males and females with tic disorders [7,8], a recent study suggested that males often experience more severe tics than females, especially at younger ages [9]. We focused on aripiprazole, risperidone, and haloperidol, which are antipsychotic medications commonly prescribed for the treatment of tic disorders [10]. These medications are possibly prescribed more frequently to patients experiencing more severe symptoms [11]. This could indicate that individuals with more severe symptoms make more frequent visits to clinics, and the chance of diagnostic error increases with an increasing number of visits to the clinics and diagnostic events.

A significantly higher proportion of males and health insurance holders were observed only in participants with type A errors. However, regarding the higher proportion of males, the absence of statistical significance in those with type B errors may be partly attributed to a relatively smaller sample size, given that the actual proportion of males was greater in participants with type B error (80.1%) than in those with type A error (79.4%). In contrast, the actual proportion of health insurance holders tended to be smaller in participants with type B error than without type B error (95.9% and 96.7%, respectively), suggesting that a higher proportion of health insurance holders is exclusively related to type A error. Based on the additional observation that most of the type B errors were made in a single institution, it may be possible that medical aid beneficiaries who have a relatively limited choice of clinics [12] tend to visit a single institution longer and thus become more prone to encounter this type of error.

The finding that the errors made in the same medical institution mostly comprised type B errors (94.9%) possibly indicates that an initial diagnosis of transient tic disorder (F95.0) may be repeated for patients visiting the institution for >12 months. Paying more attention to the date of the first visit may effectively reduce this type of error. In contrast, type A errors occurred mainly when patients changed medical in-

stitutions. Specifically, the majority of errors were in the diagnosis of chronic motor or vocal tic disorder (F95.1) at the next institution after a diagnosis of Tourette's syndrome (F95.2) was established at the previous institution. As these two disorders have the same duration of tic symptoms (i.e., >12 months), paying more attention to the presence of either motor or vocal tics, or both, may be helpful in reducing this type of error.

The relatively small number of diagnostic errors in this study may be related to the reliability of the health insurance data. Choi et al. [13] reported the prevalence of various tic disorders in Korea using health insurance claims data. If there are many errors in the coding of each of these diagnoses, relying on the research findings may be challenging. In this study, contrary to our hypothesis, we found few instances in which the diagnostic codes entered by clinicians contradicted the hierarchy of tic disorder diagnostic criteria. Thus, the current findings support the reliability of tic disorder research conducted using Korean health insurance claims data. Conducting similar studies using health insurance data from other countries would be interesting to establish the reliability of the data.

Limitations

We defined errors based on the date of visit to a medical institution rather than the date of initial tic occurrence. Therefore, type B error was detected when a diagnosis of transient tic disorder (F95.0) was made again after a period of >12 months. However, because the date of initial tic occurrence was unavailable, diagnostic errors could have occurred earlier than detected. In addition, when we detected type A error, we were unable to confirm which one was erroneous among either the previous or following diagnosis, or both. Moreover, without detailed information about comorbidities, such as ADHD, obsessive-compulsive disorder, anxiety, or depression, it is uncertain whether patients with greater diagnostic errors experienced more comorbidities that are male predominant or that increase the chance of antipsychotic use [8,10,14,15], possibly having an interaction effect. Finally, although different doctors can provide medical care within a single institution, it was not possible to determine whether the diagnostic errors occurring within the same institution were committed by the same doctor or whether they occurred owing to a change in the responsible doctor. Alternatively, when a doctor changes his/her workplace, some patients may choose to follow their designated doctor and transfer to another medical institution. However, in this study, it was not possible to ascertain whether the diagnostic errors that occurred between different institutions could potentially be attributed to the same doctor.

CONCLUSION

Most tic diagnoses made in real-world clinical practice were without errors in terms of the diagnostic hierarchy. Only a small number of errors violated the diagnostic hierarchy for tic disorders. Overall, medical practitioners appeared to comply with the diagnostic hierarchy of tic disorders.

Supplementary Materials

The online-only Data Supplement is available with this article at https://doi.org/10.5765/jkacap.230034.

Availability of Data and Material

The study data are available from the National Health Insurance Service (NHIS).

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

Author Contributions

Conceptualization: Soon-Beom Hong. Data curation: Yeeji Sung, Soon-Beom Hong. Formal analysis: Yeeji Sung, Soon-Beom Hong. Funding acquisition: Soon-Beom Hong. Investigation: Yeeji Sung, Soon-Beom Hong. Methodology: Yeeji Sung, Soon-Beom Hong. Resource: Soon-Beom Hong. Supervision: Soon-Beom Hong. Writing—original draft: Yeeji Sung, Soon-Beom Hong. Writing—review & editing: Yeeji Sung, Soon-Beom Hong.

ORCID iDs

Yeeji Sung https://orcid.org/0009-0003-4166-8015
Soon-Beom Hong https://orcid.org/0000-0003-1030-0763

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