

SOCIAL COMMUNICATION DISORDER IN AN ADOLESCENT WITH ADHD AND TOURETTE'S DISORDER TREATED WITH ARIPIPRAZOLE

Lee-Hou Tsai¹, Jeng-Wen Lin^{2,3} & Meng-Chih Lee^{4,5}

¹Department of Psychiatry, Taichung Hospital, Ministry of Health and Welfare, Taichung, Taiwan

²Department of Civil Engineering, Feng Chia University, Taichung Taiwan

³Research Center for Information Technology Innovation, Academia Sinica, Nankang, Taipei, Taiwan

⁴Professor, Department of Family Medicine, Taichung Hospital, Ministry of Health and Welfare, Taichung, Taiwan

⁵Institute of Population Health Sciences, National Health Research Institutes, Miaoli, Taiwan

received: 7.6.2021;

revised: 23.9.2021;

accepted: 6.10.2021

* * * * *

INTRODUCTION

Before the advent of the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5) (American Psychiatric Association 2013), child psychiatrists in Taiwan tended to diagnose children with normal or even high intelligence quotient (IQ) social disorders such as Asperger's disease, high-functioning autism, and atypical autism by phenotype only, which ignored the nature of the disease and the etiology of the corresponding brain block. The symptoms of mental illness may have different causes that lead to similar manifestations. This case report describes a child's cognitive and psychological aspects and disease appearance following drug treatment.

Although the DSM-5 (American Psychiatric Association 2013) defines the diagnostic criteria of Tourette's disorder (TD) as the presence of motor and vocal tics for more than one year, TD may be misdiagnosed as its symptoms tend to vary in severity over time and present differently (Cavanna & Seri 2013). TD also has comorbidities such as attention-deficit/hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD)/trait, autism, and learning disorder (Robertson 2015, Cravedi et al. 2017). The stereotyped or repetitive behaviors of autism and tics can be confusing. Motor tics can be mistaken as a sign of autism. Unless the patient has obvious blinking, facial tics, vocal tics, etc., the probability of misdiagnosis is quite high. Hence, we propose this case as a living example.

The definitions of autistic disorder, Asperger syndrome (AS), and pervasive developmental disorder, not otherwise specified (PDD-NOS) were replaced by one term, "autism spectrum disorder" (ASD), in the DSM-5 (American Psychiatric Association 2013). For atypical autism cases, it is necessary to consider whether social communication disorder (SCD) better describes the psychopathology of an individual case (Tsai & Lin 2020).

METHODS AND CASE DESCRIPTION

We describe a case of a 17-year old adolescent who began speech therapy at three years of age due to slow language development. He did not like being touched and was sensitive to sound (tactile and auditory defenses) in early childhood. He was diagnosed with mild intellectual disability and autism. In elementary school, he was easily distracted and appeared absent-minded when talking, was hyperactive and restless, and sat on the left edge of his seat, although he was more attentive and obtained good grades in subjects that interested him, such as mathematics. However, he had poor concentration and academic performance in Chinese Mandarin and English classes. In terms of interpersonal relationships, he did not know how to interpret observations, had poor social skills, and had no friends. He even had been teased by classmates without knowing it and often negatively interpreted interpersonal perceptions, resulting in insufficient emotional control and problem-solving capabilities. At the same time, he presented with symptoms of OCD, requiring a fixed outfit every day and insisting that he could not go out on unlucky days. His emotional stability deteriorated.

In his first year of high school (August 2016), because of the diagnosis of ADHD, he began receiving short-acting methylphenidate (Ritalin, Novartis Pharmaceuticals Corporation, New Jersey, USA) and later switched to long-acting methylphenidate (18 mg, Janssen Pharmaceuticals, Inc., Beerse, Belgium) capsule daily because of his habit of forgetting to take his medicine and then having the side effect of poor appetite at noon. His symptom of being distracted improved in his second year of high school but resulted in more sensitive and suspicious interpersonal relationships, referential ideation, and recurrence of motor and vocal tics. Dogmatyl (50 mg/d, Sanofi-Aventis, Paris, France) was added in September 2017. Due to hallucinations and delusions lasting for many months, in April 2018,

dogmatyl was replaced with aripiprazole (5 mg/d) and combined with methylphenidate (18 mg/d) and biperiden (2 mg/d) to prevent extrapyramidal side effects such as akathisia. By November 2018, his concentration and mood had improved significantly. Thereafter, he was treated with methylphenidate (20 mg/d) and aripiprazole (5 mg/d) (December 2018 to July 2019) and treated only with methylphenidate (20 mg/d) to maintain attention quality (August to September 2019). While taking ADHD medications, intermittent vocal tics (cough/clearing throat) and multiple motor tics such as facial tics (squeezing and blowing) and intermittent involuntary body movement lasted for more than a year before TD was diagnosed.

RESULTS AND DISCUSSION

The patient received a psychological assessment without medication in August 2016. The results are shown in Table 1. His overall IQ was 77; his worst performance was a verbal comprehension index (VCI) of 65, followed by a working memory index (WMI) of 69. He performed better on the perceptual reasoning index (PRI=100) and processing speed index (PSI=99). The psychological evaluation report indicated that the

patient had barriers to language comprehension and abstract thinking, highly selective attention, poor empathy, limited fixations, autism characteristics such as habits, and inadequate social skills.

From August 24, 2016, to July 1, 2019, during nearly three years of outpatient drug treatment with ADHD (methylphenidate 15 mg/d to 28 mg/d) and TD (dogmatyl 50 mg/d to aripiprazole 5 mg/d) drugs, the patient gradually developed an awareness of his ADHD and TD and increased his medication compliance. The effect, especially from methylphenidate 18 mg/d with aripiprazole 5 mg/d (taken before bedtime), was particularly good. The patient's language comprehension ability, attention, and emotional control ability improved greatly, his medical treatment was more regular, and his overall intelligence score improved by 21 points (Full Scale Intelligence Quotient [FSIQ] (Wechsler 2014) 77 to 98 points), indicating that the correct medication would improve his overall functioning. However, the patient had a short period of hallucination and referential ideation and thought insertion while taking methylphenidate. This was most likely due to stress response to the self-improvement and eventually improved with aripiprazole.

Table 1. Psychological tests completed by the patient

Date/age/diagnosis	Psychological test items	Result
2016/8/12 Age: 15 y 5 m ADHD/Autism	Wechsler Intelligence Scale for Children-IV (WISC-IV) (Sadeghi et al. 2011) Swanson, Nolan and Pelham, Version IV Scale for ADHD (SNAP-IV) (Hall et al. 2020) Autism Behavior Checklist- Taiwan version (ABCT) (Marteleto & Pedromónico 2005) Child behavior Checklist (CBC) (Achenbach & Ruffle 2000) Gorden diagnostic system (GDS) (Dickerson Mayes et al. 2001)	FIQ = 77, VCI = 65, PRI = 100, WMI = 69, PSI = 99 Mother's report: inattention subset (+) T value >60 Father's report: both inattention and hyperactivity/impulsivity subsets (+) T value >70 Opposition/defiance subset (+) T value >70 Teacher's report: no significant anomalies Mother's report: >99% shows autism DSM-oriented scale: depression and anxiety reach clinical significance; syndrome group shows withdraw/depression, anxiety/depression, thinking problem reaches clinical significance Borderline range in vigilance task, other tasks: normal
2018/08/14 Age: 17 y 5 m TD/ADHD and AS	Wechsler Adult Intelligence Scale-IV (WAIS-IV) (Drozdick et al. 2018) Autism-spectrum quotient (AQ) (Baron-Cohen et al. 2001) Attention-Deficit/Hyperactivity Disorder Test (ADHDT) (Perdue 2016) Continuous performance test-III (CPT 3) (Keith Connors et al. 2018)	FIQ = 98, VCI = 82, WMI = 89, PRI = 126, PSI = 99 Mother's answer: total score 40 Significance difference in all domains (Z score > 2.5), except for the domain of attention to detail (Z = 0.60) Mother's report: average Normal

AS: Asperger syndrome; FSIQ: Full-scale intelligence quotient; VCI: Verbal comprehension indeks; PRI: Perceptual reasoning indeks; WMI: Working memory indeks; PSI: Processing speed index

After a period of drug treatment, the patient's empathy also substantially improved, as evidenced by his initial attention to his status only with later concern for the appearance of his parents. Reevaluation indicated that the patient should not be diagnosed with autism but rather SCD caused by the combined effects of ADHD and TD. Psychological evaluations before and after drug treatment showed that this case had TD with ADHD, learning disorder, and SCD, rather than ASD (Table 1).

Aripiprazole, the newest antipsychotic medication in TD treatment, has a unique mechanism as a dopamine D2 and serotonin 5-hydroxytryptamine (5-HT) 1A receptor partial agonist and 5-HT2A receptor antagonist (Janik & Szejko 2018). One study described the use of aripiprazole (10.0±4.8 mg/day) in a consecutive group of 28 patients with primary diagnoses of TD and comorbid ADHD, using the Yale Global Tic Severity Scale (YGTSS) (Scahill 2013) and ADHD-rating scale (ADHD-RS-IV) (George et al. 1998) for outcome assessment. Both significantly improved after treatment (Masi et al. 2012).

However, in an analysis of the adverse drug reaction (ADR) database among children and adolescents (<18 years) at the Danish Medicines Agency, Jakobsen et al. (2016), reported that the adverse drug effects of aripiprazole in non-psychotic children and adolescents (such as TD, ADHD, autism, OCD) included chronic insomnia, Parkinsonism, behavioral changes, psychoses, and weight gain while those in the psychotic group were predominantly anxiety, convulsions, and neuroleptic malignant syndrome. Suicidal behavior, perhaps due to the side effect of akathisia, was reported to health authorities. Serafini et al. (2019) presented a case of aripiprazole-induced hiccup in a young inpatient at his first psychiatric admission along with a review of the literature. They also discussed the possible etiology underlying the emergence of hiccups together with the clinical implications of this adverse event.

CONCLUSION

This case differs from studies applying clinical-rated instruments (YGTSS, Clinical Global Impression Scale (Jeon et al. 2013), etc.) to evaluate efficacy. Although to our knowledge, this is the only such case reported, it is useful to understand its various psychological aspects. In cases of children and adolescents with ADHD and TD, aripiprazole and ADHD drugs can be considered in a two-pronged approach to allow the dopamine system in the brain to rebalance to improve patient attention, concentration, and motivation while also reducing obsessive-compulsive symptoms and negative thinking. We recommend that children and adolescents start with the lowest dose based on the "start low, going low" principle. This report has limitations. In psychiatric nomenclature, current diagnostic systems, be it DSM-5 (American Psychiatric Association 2013) or International Classification of Diseases, 10th edition (ICD-10) (World Health Organization 2019), are atheoretical. Thus, diag-

nostic changes are based on observed phenomenology. In addition, Tourette's Disorder/syndrome is a gradually evolving condition. In such cases, associated ADHD symptoms (mostly hyperactivity and impulsivity) may diminish by adolescence, making it difficult to say that these changes occurred only due to medication.

Acknowledgements:

The work described in this paper was part of research funded by Academia Sinica, Taiwan (Contract No. 108-2-1-11-23).

Conflict of interest: None to declare.

Contribution of individual authors:

Lee-Hou Tsai: design of the study, interpretation of data, manuscript writing.

Jeng-Wen Lin: literature searches and analyses, statistical analyses, manuscript revision.

Meng-Chih Lee: manuscript revision, comments on the concept of article.

References

1. Achenbach TM & Ruffle TM: *The Child Behavior Checklist and related forms for assessing behavioral/emotional problems and competencies*. *Pediatr Rev* 2000; 21:265-271
2. American Psychiatric Association: *Diagnostic and Statistical Manual of Mental Disorders (DSM-5®), Fifth Edition*. 2013; [cited 23 September 2020]. Available from: https://www.appi.org/Diagnostic_and_Statistical_Manual_of_Mental_Disorders_DSM-5_Fifth_Edition
3. Baron-Cohen S, Wheelwright S, Skinner R, Martin J, Clubley E: *The autism-spectrum quotient (AQ): evidence from Asperger syndrome/high-functioning autism, males and females, scientists and mathematicians*. *J Autism Dev Disord* 2001; 31:5-17
4. Cavanna AE & Seri S: *Tourette's syndrome*. *BMJ* 2013; 347: f4964. doi: 10.1136/bmj.f4964
5. Cravedi E, Deniau E, Giannitelli M, Xavier J, Hartmann A, Cohen D: *Tourette syndrome and other neurodevelopmental disorders: a comprehensive review*. *Child Adolesc Psychiatry Ment Health* 2017; 11:59. doi: 10.1186/s13034-017-0196-x
6. Dickerson Mayes S, Calhoun SL, Crowell EW: *Clinical validity and interpretation of the Gordon Diagnostic System in ADHD assessments*. *Child Neuropsychol*. 2001; 7:32-41
7. Drozdick LW, Raiford SE, Wahlstrom D, Weiss LG: *The Wechsler Adult Intelligence Scale - Fourth Edition and the Wechsler Memory Scale - Fourth Edition*. In *Contemporary intellectual assessment: Theories, tests, and issues, 4th ed*, 2018; 486-511. New York, NY, US: The Guilford Press
8. George JD, Thomas JP, Arthur DA, and Robert R: *ADHD Rating Scale-IV: Checklists, Norms, and Clinical Interpretation*. ©1998 Reprinted with permission from The Guilford Press, New York. 1998; [cited 24 September 2020]. Available from: https://pcptoolkit.beaconhealthoptions.com/wp-content/uploads/2016/01/cms-quality-child_adhd_rating_scale_screener.pdf

9. Hall CL, Guo B, Valentine AZ, Groom MJ, Daley D, Sayal K, Hollis C: The validity of the SNAP-IV in children displaying ADHD symptoms. *Assessment* 2020; 27:1258–1271
10. Jakobsen KD, Bruhn CH, Pagsberg AK, Fink-Jensen A, Nielsen J: Neurological, metabolic, and psychiatric adverse events in children and adolescents treated with aripiprazole: *Journal of Clinical Psychopharmacology* 2016; 36:496–499
11. Janik P & Szejko N: Aripiprazole in treatment of Gilles de la Tourette syndrome - New therapeutic option. *Neurol Neurochir Pol* 2018; 52:84–87
12. Jeon S, Walkup JT, Woods DW, Peterson A, Piacentini J, Wilhelm S, Katsovich L, McGuire JF, Dziura J, Scahill L: Detecting a clinically meaningful change in tic severity in Tourette syndrome: a comparison of three methods. *Contemp Clin Trials* 2013; 36:414–420
13. Keith Connors C, Sitarenios G, Ayearst LE: *Conners' Continuous Performance Test Third Edition*. In *Encyclopedia of Clinical Neuropsychology*, Kreutzer JS, DeLuca J, Caplan B (eds). 2018; 929–933. Cham: Springer International Publishing
14. Marteleto MRF & Pedromônico MRM: Validity of autism behavior checklist (ABC): preliminary study. *Brazilian Journal of Psychiatry* 2005; 27:295–301
15. Masi G, Gagliano A, Siracusano R, Berloffo S, Calarese T, Ilardo G, Pfanner C, Magazù A, Cedro C: Aripiprazole in children with Tourette's disorder and co-morbid attention-deficit/hyperactivity disorder: a 12-week, open-label, preliminary study. *J Child Adolesc Psychopharmacol* 2012; 22:120–125
16. Perdue EA: Test Review: Gilliam, J. E. (2015). *Attention-Deficit/Hyperactivity Disorder Test (2nd ed)*. *Journal of Psychoeducational Assessment* 2016; 34:819–822
17. Robertson MM: A personal 35 year perspective on Gilles de la Tourette syndrome: prevalence, phenomenology, comorbidities, and coexistent psychopathologies. *Lancet Psychiatry* 2015; 2:68–87
18. Sadeghi A, Rabiee M, Abedi MR: Validation and reliability of the Wechsler Intelligence Scale for Children-IV. *Developmental Psychology: Journal of Iranian Psychologists* 2011; 7:377–386
19. Scahill LD: Yale Global Tic Severity Scale. In: Volkmar F.R. (eds) *Encyclopedia of Autism Spectrum Disorders*. Springer, New York, NY, 2013; https://doi.org/10.1007/978-1-4419-1698-3_1279
20. Serafini G, Piccinini G, Visimberga S, Cervetti A, Belvederi Murri M, Monacelli F, Pompili M, Amore M: Aripiprazole-induced persistent hiccup: a case report and review of the literature. *Psychiatr Danub* 2019; 31:26–31
21. Tsai LH & Lin JW: Adaptation of diagnosis from autism spectrum disorder to social communication disorder in adolescents with ADHD. *J Autism Dev Disord* 2020; 50:685–687
22. Wechsler D: *Wechsler Intelligence Scale for Children (WISC®-V), Fifth Edition*. 2014; [cited 24 September 2020]. Available from: <https://www.pearsonassessments.com/store/usassessments/en/Store/Professional-Assessments/Cognition-%26-Neuro/Gifted-%26-Talented/Wechsler-Intelligence-Scale-for-Children-%7C-Fifth-Edition-/p/100000771.html>
23. World Health Organization: *ICD-10 online versions - WHO* 2019; [cited 25 September 2020]. Available from: <https://www.who.int/classifications/icd/icdonlineversions/en/>

Correspondence:

Professor Jeng-Wen Lin, PhD
Department of Civil Engineering, Feng Chia University
No. 100 Wenhwa Road, Seatwen, Taichung 40724, Taiwan
E-mail: jwlin@fcu.edu.tw