Asperger's Syndrome in a Clinical Sample: Reasons for Referral and Comorbidity

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Abstract: Asperger's Syndrome (AS) is an autism spectrum disorder without mental retardation and language delay. AS often remains unrecognized until these children fail to adapt to school or kindergarten. The comorbid psychiatric disorders, achieving clinical significance, were considered as another pathway to diagnosis. This study is aimed to elucidate the reasons for referral, the frequency and the kinds of comorbidities in a clinical sample of consecutive cases of children and adolescents with AS. To this objective, clinical records of children and adolescents, who have received a DSM-IV diagnosis of AS after multidisciplinary assessment in a given time period were reviewed. After excluding 3 cases due to insufficient information, 24 cases of children and adolescents with Asperger's Syndrome (23 boys and one girl) were identified. The mean age at the time of assessment and receiving diagnosis was 9.6 yrs. (age range 4 to 17 years). In twenty-one (87%) of the cases the reason for referral was an episode of disorganized behavior following an attempt to enroll the child at school or kindergarten, and more rare referral occurred within the significant school transition period. In the remaining 3 cases, the reason for referral was a comorbid condition. Comorbid conditions identified at the moment of assessment include: ADHD documented in 4 cases, tics in 3 cases, obsessive-compulsive behaviors in 4 cases, Stereotypic Movement Disorder or Trichotilomania in 4 of the cases. Within the clinical sample, a priori expected to include relatively severe cases, a higher frequency of comorbidity was found as compared to the rates in the general population. Adjustment reactions and comorbidities occasioned the referral, while AS was diagnosed only after specialized multidisciplinary assessment.

Keywords: Asperger's Syndrome, reason for referral, comorbidity.

INTRODUCTION

Asperger's syndrome or Asperger's disorder is a neurodevelopmental disorder, belonging to autism spectrum disorders and characterized by cognitive, perceptive, linguistic, sensory and motor deviances and deficits. The general intellectual functioning of children with AS is formally within normal range. Asperger's children differ from those with high functioning autism (autism without mental retardation) in their spoken language skills being closer to normal or even adultomorphic, together with their language developmental milestones situated within the expected range. Single words are attained by the age of two and two-word communicative utterances by the age of three [1]. In neuropsychological assessment, Asperger's children score higher on verbal, compared to nonverbal subscales of tests [2]. Restricted, repetitive, and intense interests and preoccupations are usually present.

Children with Asperger's syndrome exhibit difficulties to understand the code of social conduct, to understand and follow the unwritten rules of social interaction and to distinguish metaphorical and idiomatic from literal speech. As a consequence, they may appear like a stranger in a totally different culture, feeling in social interactions like "an anthropologist on Mars" [3].

They have more marked neuropsychological, language and psychomotor deficits than schizoid children. In addition, they may be actively seeking social contact while making it bizarrely. They appear to be rather "active but odd", then isolated in social relationships. Language-impaired children fitting the picture of Semantic-pragmatic disorder differ from Asperger's children in that they typically present with delayed language development and evident comprehension problems, and have a marked IQ discrepancy in favour of performance IQ. Asperger's children meet the three groups of DSM-IV-TR criteria for Pervasive Developmental Disorders, while children with PDD-NOS, one other group that can include autistic-like individuals with intellectual functioning within the norm, only two [1].

In a typical case, the main parental concern for the preschool autistic child is lack of expressive language. By this time, the AS child is verbally fluent, frequently knowing letters, even being able to read or write and usually displaying scientific-like interests, sounding like a "little professor" or "walking dictionary". His parents may inaccurately perceive him as rather gifted than developmentally impaired. The crisis usually occurs

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when this child has to enter the social world of the kindergarten or the primary school.

This disorder is named after H. Asperger, the Austrian physician, who described a group of boys with marked social problems but a rather good language and cognitive skills [4]. With the influential for the English speaking professionals works of L. Wing, and later with the inclusion of Asperger's disorder in both DSM-IV and ICD-10, clinical use of the term has increased dramatically [5-7].

Individuals with AS enter different pathways to diagnosis as compared to those with autistic disorder. It is obvious that children and adults with AS may be an object of clinical attention, assessment, and treatment prior to the recognition of the disorder, due to associated or secondary for the AS problems and without recognition of AS. This seems more to be the common case than an exception.

The association of AS with other mental disorders is not unexpected [6-8]. According to T. Attwood, the association exists with ADHD and tics, and after adolescence – with depression and suicide attempts [7, 8].

There is a great need for in-depth research into the area of comorbidity in the field of autism, and even more now when DSM 5 do not rule out comorbid conditions such as ADHD in individuals diagnosed (according to DSM-5 criteria) with Autism Spectrum Disorder [9, 10]. Even before DSM-5, however, there were opinions that the exclusion of the diagnosis of comorbid conditions in ASD may have to be revised [8].

It is widely accepted that the early detection of developmental disorders, including AS, is of primary importance. The timely diagnosis makes it possible to set up parent counseling and psychoeducation, and enroll children in appropriate school-based and social welfare programs. In addition to being an "entrance ticket" to the systems of care, an early diagnosis may clear the way for working on the parental emotional process of accepting the lifelong disability of their child.

In our clinical practice, however, AS children appear first with disruptive behaviors in social settings or are rarely referred due to comorbid disorders, while AS may be recognized later by a small circle of professionals.

This observation motivated us to study referral reasons and comorbidity in a clinical sample of Asperger's children and adolescents, supposing that this may be helpful for psychiatrists, neurologists, general practitioners and pedagogues working with children.

Here we call "comorbid" those disorders, conditions and behaviors, which are not obligatory for AS but may be frequently associated.

METHODS

The medical charts of all children and adolescents diagnosed with AS within the Day-care department of the Clinic of child psychiatry to the University Hospital Alexandrovska in Sofia between January 2014 and December 2016 were reviewed. Out of the 27 cases, who received a diagnosis of AS, 24 (23 boys, one girl) were retained for the study. The mean age of the subjects at the first contact was 9.6 years (range, 4-17 years). The exclusion of the remaining three cases was due to lack of targeted information in clinical charts.

As part of the clinic's evaluation program for children and adolescents with autism spectrum disorders, clinical charts were completed in line with the following assessment protocol:

Each child or adolescent suspected for AS was assessed by an experienced child psychiatrist and a clinical child psychologist. The child psychiatric examination included: at least two interviews with both parents (except in single parent cases where only one parent or the only parent and other significant adult were interviewed) and two clinical interviews with the child or the adolescent. Australian Scale for Asperger's Syndrome (A.S.A.S.) was completed by the child psychiatrist, using information from parents and data from the interview with the child or the adolescent. The psychological assessment consisted of minimum four diagnostic sessions. The following instruments were used in psychological assessment- WISC-III, the "Strange Stories" Test- F. Happé, Draw-a-Person test. Each case assessment included a meeting of the assessment team, followed by one or preferably more final meetings with the parents. The duration of the assessment period was at least one week.

The clinical diagnosis of AS was made according to DSM-IV-TR criteria. Fulfillment of both sets of criteria was required. In addition, the consensus of both professionals in the assessment team was needed for the diagnosis.

By rule, the medical records of cases included data with definite statement about existence of comorbid

conditions and their type using DSM-IV-TR nomenclature and criteria for psychiatric disorders. The reason and source for referral were documented in charts by rule, as well as supposed disorder or problem stated by a referral agent.

RESULTS

Reasons for referral are shown in Table **1**. School or kindergarten-related behavior problems to the point of requiring referral were exhibited by the majority of children (87%, n=24). In almost all of the cases, referral was initiated by the kindergarten or the school. In no case the diagnosis of AS was considered prior to the assessment.

Fourteen (58%, n=24) children had an episode of disorganized behavior (adjustment reaction) at the period of the first attempt for enrollment (primarily in the elementary school and less commonly in the kindergarten) – which served as the main reason for referral. According to school's and kindergarten's formulations, the referral was related to unacceptable and disruptive behavior with disorganized quality, while the children themselves were described as "aggressive, auto-aggressive, leaving out the seats and constantly speaking in class, some of them telling obscenities or writing obscene words or symbols on the board". One of the children reacted to school enrollment with a somatic problem - alopecia.

Table 1: Age, Sex and Reason for Referrals/Pathway to Diagnosis

Age at first assessment			**9.6 yrs.**
Age range at first assessment			**4-17 yrs.**
		n	**%**
Sex		24	
	Boys	23	
	Girls	1	
Reason for referral			
School-related behaviors problems		21	87
	Early Adjustment Reaction to enrollment	15	58
	Late Adjustment Reaction	6	25
Comorbid condition being reason for referral		3	12
	OCD	2	8
	Depressive Mood	1	4

In six of the cases, also referred primarily through the school (25%, n=24), such temporary episode of disturbing behavior is related to school adaptation was not observed to the point of warranting referral. Despite permanently difficult behavior and other problems or recess periods in class (not working in class, answering questions without permission, being easily aggressive, or chronically subjected to or in risk of bullying) these children and the situation around them were tolerated. However, at some later point and namely in the transition to secondary school classes, problematic behavior became a reason for referral.

Referral was initiated, due to comorbidities only in three (12%, n=24) subjects, all of them adolescents. Even described as "difficult" or "strange", up to that point they were passed through elementary and primary school, without being considered for diagnostic referral.

The type and rates of comorbidity are displayed in Table **2**.

Table 2: Type and Rate of Comorbidity

(Lifetime) Comorbidity at assessment		**n**	**%**
DSM-IV Disorders	ADHD	4	17
	Tic Disorders	3	12
	OCD	4	17
	Stereotypic Movement Disorder and Trichotillomania	3	12
	Other speech dysfluencies (in a previous period)	3	12
	Depressive Mood	1	4

Attention-Deficit/Hyperactivity Disorder

The DSM-IV A to D criteria for ADHD were met in four (17%) of the children. All of them also met DSM 5 criteria for ADHD Two were monozygotic twins, showing markedly hyperactive and impulsive behavior at early school age, imposing enrollment at individual tuition program. At this moment and in the course of the following years they were not recognized as having AS.

Tic Disorders

Tics were observed in three (12%) children. In two of them, Tourette's Disorder was diagnosed, in the third – Chronic Motor Tics (CMVT).

In two children copropraxic gestures were observed, such as repetitive touching the genitalia-seemingly corresponding to copropraxic complex motor tics

observed in some cases of Tourette's Disorder. Here the copropraxic acts were situationally bound – occurring at school. Upon request, children were able to stop these acts temporarily. In the absence of evidence and history of tics and tics being more frequent and intense at home, not at school, these copropraxic movements were not documented as complex tics.

Obsessive-Compulsive Disorder and Features

Obsessive-compulsive behaviors were found in four (17%) children – manifesting in 3 of the cases in adolescence and in 1 – at the age of eleven. In three of these children, meeting criteria for Obsessive-Compulsive Disorder, hand washing compulsions were observed and in one child - repetitive time-consuming and ego-dystonic compulsive mental calculations using the number seven.

Two other children (the monozygotic twins, mentioned earlier) exhibited watching television all over the day. They reacted with intense protest even at night to any attempt of parents to make them stop watching. Here we qualified this behavior rather as a repetitive and pleasurable than (OCD-like) compulsive.

Speech Dysfluency

As an early manifestation seen in preschool age and imposing consultation with a speech and language pathologist, without recognition of Asperger Disorder, speech fluency was noted in three of the children. By the time of assessment and diagnosing AS however, this dysfluency had disappeared.

Stereotypic/Movements Disorder and Trichotillomania

Chronic repetitive nonfunctional movements with self-inflicted skin damage were noted in two of the children, respectively with a wound on the lower lip and damage to the skin around the ungles. In another child trichotillomania was observed.

Depressive Mood

Only in one of the cases, a child aged thirteen, depressive mood together with suicidal ideation (not to the extent of fulfilling the criteria for Major Depressive Episode) were observed. In this case, depressive mood was understood as a reaction to the appraisal of the real situation and the expected life perspective.

DISCUSSION

Some points related to AS need to be stressed prior to the discussion of findings. The first one is the well-known problem with the reliability of AS diagnosis. The removal of the subtypes of PDDs (including AS) in DSM-5 holds this problem as one of its premises [11]. It is obvious that a child diagnosed with AS in one institution may not receive the same diagnosis in another. On the other hand, children with ASD are different, as all children are, in terms of deficits, resources and individuality, and categorically or dimensionally these differences were, are (DSM-5 uses specifiers to match with this phenotypic heterogeneity) and will be taken into account. Officially accepted or not, however, similarities within the groups of children with ASD do exist and in everyday practice we acknowledge this automatically in expressions such as "X. resembles Y, N is just like them – verbose but awkward". The brain is wired to notice not only differences but similarities as well. Is such grouping, apart from clinical and practical occurrences, needed for research in the underlying neurobiological domain? Quite agreeing with King *et al*. that biology does not respect our classification categories and genetic findings do not support even the distinction of ASD and ADHD [11], we prefer to answer the above question positively. According to the current research findings on genotypes, they do not match with phenotypes, but "genetic factors" are not only given genes or combinations. Genes interact with the environment especially in sensitive periods and as a result altering their effects. Many genetic pathways may lead to one condition and a given gene may be implicated in different disorders - processes called genetic heterogeneity and pleiotropism [12]. By taking this into account we must not expect to find a simple gene or definite genes to correlate with a given phenotype! Thus, a genetic confirmation is not needed to retain diagnostic categories, at least for the moment.

Children with AS in our sample may be diagnosed according to DSM-5 as ASD children without gross language impairment (apart from the pragmatic one) and without intellectual deficiency in the formal aspects of intelligence. It is obvious that these relatively less impaired children in the group of ASD will have different pathways to diagnosis, (this difference will include also the time before diagnosis and respectively the child's age at the time of diagnostic assessment). After receiving diagnosis they will have a different life trajectory with different challenges, needs and opportunities. They will require distinctive forms of

support as compared to typical autistic children or to those with associated pronounced intellectual deficiency. At least for internal use professional teams will continue to use "Asperger's syndrome", or "Asperger-like" as a "label" for such a group of relatively better functioning ASD children with no early language delay.

According to our experience typical children with AS do not present frequently for assessment even in a specialized unit like ours. Less than 30 children and adolescents were diagnosed within a 2-year period (on the average, children receiving diagnosis of ASD within a 2-year period in the unit are about 600). This may reflect also our caution in assigning this controversial diagnosis as well as an inclination to choose a larger diagnostic "umbrella".

In our sample of AS children and adolescents the most common reason for referral for diagnostic assessment was seriously disruptive behavior in school or (in some cases) in kindergarten. Before that time, in a family or a close friends' circle the given child's behavior had not appeared disruptive. Developmental deviations were interpreted by parents rather as child's assets than as signs of disturbance. For example, the indiscriminative easiness of contact with strangers was usually viewed as a trait of sociability and adult-like and pedantic speech in a preschooler - as a marker of maturity.

Most of the children failed to adapt at their first enrollment at school, thus entering the well-known main "pathway" to diagnosis AS [6, 7]. Being "dropped" in the much more complex situation at school with many rules, some of them unstated, and with different behaviors of classmates towards rules, children with AS express their frustration, confusion and anxiety through disruptive behavior. In the majority of cases, these behaviors included situational motor and verbal overactivity of the child; these were not stable ADHD characteristics. Often chaotic and repetitive, such overactivity sometimes included aggressive, coprolalic and other unacceptable verbal, gestural or graphic productions. These children scared teachers, appearing "very ill" or "crazy" and their behaviors led the school staff to decide immediately to pressure the parents in consulting a mental health professional. As expected, remaining at home, children gradually calmed down.

Less commonly, referral to specialized diagnostic agency, though also initiated by the school, appeared later - after a period of more than one year of less disruptive chronic behavior problems, not to the point of warranting immediate action. In the prototypical case, "adjustment failure" occurred in 5th grade – when the child was expected to cope with the replacement of the mother-like figure of a single teacher with new and multiple ones. Children provoking "maternal" (rather than "predatory" instinct [6] had been specially protected by female teachers at elementary school or kindergarten. Losing this maternal kind of attention and protection, later on, subjects these children to shock-like experiences with breakdown of adaptation. In addition, by this time of preadolescence, social relations become relatively more complex, and school rules less valued and readily ignored by typical schoolmates. This is not understandable and anxiety provoking for Asperger's children enlarging the gap between them and the others.

Only in three of the children with AS and relatively late for the diagnosis (in adolescence) the reason for referral was a comorbid condition. Despite the small percentage of these cases, we consider this pathway to diagnosis to be important. No adequate therapeutic plan can be made without considering AS in these cases.

The association of AS with other mental disorders is not unexpected. M. Ghaziuddin *et al*. found symptoms of comorbid psychiatric conditions in 23 out of 35 patients with AS with a mean age of 15.1 years [9]. According to T. Attwood an association in AS exists with ADHD and tics, and after adolescence – with depressions and suicide attempts [6, 7].

Comorbidities in our sample comprised disorders and conditions which were not consequent of child's inclusion in out-of-home environment but were co-existing. They included different disorders and syndromes, given the fact that a "syndrome mix" of neuropsychiatric conditions runs in families [13]. Noted during the assessment DSM-IV comorbid disorders consisted of: ADHD, tics, obsessive-compulsive disorder or behaviors, Stereotypy/movement Disorder, history of impairments in speech fluency – stuttering or stuttering-like disrupted speech fluency.

It is evident that within the studied sample of children and adolescents with AS the percentage of comorbidity was higher than expected, compared to the usually stated prevalence of disorders in the general population. The interpretation of this fact, however, must take into consideration two points. The first one is

that an elevated rate of comorbidity might be associated also with other neurodevelopmental and neurobehavioral disorders, such as ADHD, being not specific for AS. The second point is whether, due to referral selection of cases, the registered comorbidity in these cases is additionally elevated. Given the relatively high prevalence of AS in the epidemiological study of Ehlers and Gillberg (1993) - 0.3 % - we may suppose that only a small proportion of Asperger's individuals and probably those with higher rates of comorbidity, come to clinical attention, while the predominating milder cases remain unrecognized [14]. It is probable that in milder cases the rate of comorbidity is lower.

Even if not documented as tics, some behaviors displayed by the children may superficially appear to be complex motor (i.e. copropraxic) tics. Other behaviors that may be initially pleasurable for the children, but due to their highly overwhelming quality (as in the case of the twins not being able to stop watching television) they may be seen as rather compulsive and ego-dystonic.

Comorbidity with ADHD may be expected to facilitate early school adaptational failure and in our sample this comorbidity is lower than expected. When considering ADHD as comorbidity with AS, it should be taken into consideration that under stress children with AS may appear hyperactive and even remain overactive for a given time without having impulsivity-hyperactivity as a trait. Besides, when confronted with a task out of interest, an Asperger's child may demonstrate inattentive and hyperactive behavior. In such cases, a comorbid diagnosis of ADHD might be given only after careful consideration.

In the case of female patient, assessment was undergone at the age of fifteen, because of inhibited behavior and being victim of bullying at school. Chronic motor tics, mentioned earlier, were associated with AS in this case.

In view of AS's complications and comorbidities, it is necessary to hold a lifetime perspective. Probably, the existence and severity of some comorbid disorders are related to the individual's age. For example, Obsessive-Compulsive Disorder may manifest or achieve clinical significance in adolescence. Reports and observations that after the adolescence many people with AS show symptoms of depression and even make suicide attempts do exist. In addition, AS tends to or may be associated with other problems and disorders, such as offending and probably alcoholism, also not mentioned here, because of their supposed appearance later in life [8].

This shows the necessity for lifelong follow-up of children and adolescents with AS as a distinctive group within ASD in order to obtain a more accurate knowledge of comorbid conditions as well as of the real needs of those people for the purpose of professional interventions and support throughout the different periods of their life.

A continuum between below threshold personality characteristics and "full-blown" AS may be presumed. Within this continuum some closer to norm AS individuals may remain undiagnosed. Some of them may be referred later because of comorbid condition and thus AS may be recognized or suspected. In this regard, what deserves attention is the fact that in part of the perpetrators of violent crimes in Sweden diagnosed with ASD (constituting about 1% of all perpetrators of violent crimes) a diagnosis of ASD was made after the crime [15]. In our opinion, within this group of individuals with an "aftermath diagnosis," some of them probably have AS or close to AS characteristics, which were not supposed by authors who linked the longer period prior to the diagnosis with a lack of timely interventions and the worsened prognosis as a result.

REFERANCES

[1] American Psychiatric Association, Diagnostic and Statistical Manual of Mental Disorders, 4th, text revision (DSM-IV-TR). Washington, DC: American Psychiatric Association 2000.

[2] Koyama T, Tachimori H, Osada H, Takeda T, Kurita H. Cognitive and symptom profiles in Asperger's syndrome and high-functioning autism. Psychiatry and Clinical Neurosciences 2007; 61: 99-104. https://doi.org/10.1111/j.1440-1819.2007.01617.x

[3] Sacks O. An Anthropologist on Mars: Seven Paradoxical Tales. New York, Vintage Books, 1995.

[4] Asperger H. Die "autistichen Psychopathen" im Kindersalter. Archive fur Psychiatrie und Nervenkrankheiten 1944; 117: 76-136. https://doi.org/10.1007/BF01837709

[5] Wing L. Asperger's Syndrome: A Clinical Account, Psychological Medicine 1981; 11: 115-129. https://doi.org/10.1017/S0033291700053332

[6] Attwood T. Asperger's Syndrome: A Guide for Parents and Professionals. Jessica Kingsley, London 1997.

[7] Attwood T. The Complete Guide to Asperger's Syndrome. Jessika Kingsley, London 2006.

[8] Gillberg C, Billstedt E. Autism and Asperger syndrome: coexistence with other clinical disorders. Acta Psychiatr Scand 2000; 102(5): 321-330. https://doi.org/10.1034/j.1600-0447.2000.102005321.x

[9] American Psychiatric Association. Diagnostic and Statistical Manual of Mental Disorders. 5th ed. (DSM 5). Washington DC: American Psychiatric Publishing 2013.

[10] Achkova M, Manolova H. Diagnosis "Autism" - from Kanner and Asperger to DSM-5. J Intel Disability-Diagnosis and Treatment 2014; 2: 112-18.

[11] King B, Weenstra-Vander Weele J, Lord C. DSM-5 and Autism: Kicking the Tires and Making the Grade. J Am Acad Child Adol Psychiatry 2013; 52: 454-456. https://doi.org/10.1016/j.jaac.2013.02.009

[12] Mandy W, Skuse D, Charman T, Frazier T. In Defense of Lumping (and Splitting) Letter to the editor. J Am Acad Child Adol Psychiatry 2012; 51: 441-442. https://doi.org/10.1016/j.jaac.2012.02.004

[13] Kutschner ML, Attwood T, Wolff RR. Kids in the Syndrome Mix of ADHD, LD, Asperger's, Tourette's, Bipolar, And More!: The One Stop Guide for Parents, Teachers, and Other Professionals, London, 2007.

[14] Ehlers S, Gillberg C. The epidemiology of Asperger syndrome: A total population study. Journal of Child Psychology and Psychiatry 1993; 34: 1327-1350. https://doi.org/10.1111/j.1469-7610.1993.tb02094.x

[15] Heeramun R, Magnusson C, Gumpert CH, *et al.* Autism and Convictions for Violent Crimes: Population-Based Cohort Study in Sweden. J Am Acad Child Adolesc Psychiatry 2017; 56(6): 491-497. https://doi.org/10.1016/j.jaac.2017.03.011

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