Brief Report: Cases for an Association Between Tourette Syndrome, Autistic Disorder, and Schizophrenia-Like Disorder¹

Jeffrey Sverd² and Gerardo Montero

Sagamore Children's Psychiatric Center, Office of Mental Health, State of New York

Nina Gurevich Private practice

Despite evidence supporting discontinuity between autistic disorder and schizophrenia (Green et al., 1984; Kolvin, 1971; Rumsey, Rapoport, & Sceery, 1985; Rutter, 1972; Volkmar & Cohen, 1991) increasing numbers of patients with coexisting autistic disorder and schizophrenia-like psychosis have been described (Cantor, Evans, Pearce, & Pezzot-Pearce, 1982; Clarke, Littlejohns, Corbett, & Joseph, 1988; Comings & Comings, 1991; Petty, Ornitz, Michelman, & Zimmerman, 1984; Realmuto & August, 1991; Szatmari, Bartolucci, Finlayson, & Krames, 1986; Volkmar, Cohen, Hoshino, Rende, & Paul, 1988; Wolff & Chick, 1980) and some investigators have reported an association between past history of autistic symptoms and present diagnosis of schizophrenia (Russell, Bott, & Sammons, 1989; Waterhouse, Fein, Nath, & Snyder, 1987; Watkins, Asarnow, & Tanguay, 1988).

The following is a report of two children who were diagnosed as having co-occurring autistic disorder, schizophrenia-like psychosis, and Tourette syndrome (TS). Two additional autistic adults are described. Both of the latter patients showed tics and experienced episodes of schizophrenia-like psychosis. Evidence is reviewed to suggest that there exists a subgroup of autistic children who are at risk for the development of schizophrenia-like symptoms and that TS may underlie the coexistence of the disorders in some patients.

¹This work was supported in part by the Edith Wertheim Tourette-Autism Research Fund. ²Address all correspondence to Jeffrey Sverd, Sagamore Children's Psychiatric Center, 197 Half Hollow Road, Dix Hills, New York 11746.

CASE REPORTS

Case 1

D was an 11-year-old boy referred for treatment of psychosis. He evidenced auditory and visual hallucinations, intense fears, nightmares, sexual and religious preoccupations, masturbated in public, and made inappropriate sexual comments to his mother. During the course of illness, he heard voices telling him to destroy his toys, kick his siblings, kill his therapist and his baby brother. Voices of God and angels "called him to heaven so that evil could not get" to him. He talked to deceased relatives whom he had never met and on one occasion, he was observed flapping his arms and reported that he was hearing voices telling him he could fly. As an infant, he showed muscle hypotonia and an abnormal cry. He did not respond to the environment, resisted cuddling and being held, did not babble or coo, and failed to make eve contact until about 4 years of age. He did not respond to touch, pain, noise, or his name and was felt to be deaf and autistic by age 2 to 3 years. He did not utter words until 2 years and at age 5 years had limited speech which consisted of gibberish and echolalia. Play was nonrepresentational and consisted of the lining up of toy soldiers and matchbox cars. As a younger child, he enjoyed spinning and watching spinning objects. His scores on the WISC-R were Full-scale IQ 51, Verbal IQ 54, and Performance IQ 57.

At age $3^{1}/_{2}$ years, D was diagnosed by a neurologist as having TS. He showed thigh slapping, eye blinking, throat clearing, coprolalia, and involuntary giggling. He has since showed a fluctuating course of motor and vocal tics: head jerks, shoulder shrugs, facial grimaces, object smelling, copropraxia, blowing tics, and growling noises. D's maternal and paternal families showed evidence for pleiotropic manifestations of the TS gene. Natural father spoke rapidly, had the habit of shaking his leg, tapping his fingers, and biting his nails and cuticles. He needed little sleep, was a workaholic, a compulsive cigarette smoker (two packs per day), and had checking behaviors. His grandparents were psychiatrically institutionalized. D's mother had eye blinking, leg shaking, and lip biting, was a poor sleeper and felt vulnerable to abusing alcohol. Her father was alcoholic and her mother had lip biting and anxiety disorder. Diagnoses of TS, childhood schizophrenia, and autistic disorder were made.

Case 2

W, a 15-year-old male, was psychiatrically hospitalized because of chronic agitation, aggressivity, exhibitionism, bizarre thinking (he believed

Autism-Schizophrenia and Tourette Syndrome

other children had sexual relations with his mother), excessive, sometimes public masturbation, and threats to run from his residential treatment facility. Behavior disorder throughout childhood consisted of hyperactivity, wandering into neighbors' homes, encopresis, fecal smearing, enuresis, urinating in closets, and cruelty to pets.

W was adopted at 18 months from foster placement and except for a report that his natural mother was a drug abuser, information about natural parents was unavailable. Autistic disorder was apparent to foster and adoptive parents. He stared vacantly while he lay in his crib and showed unprovoked episodes of screaming, crying, and yelling. He did not tolerate being held, did not respond to his name until $2^{1/2}$ years, make eye contact until 3 years, or show interest in people until 4 years. Speech was not present until $2^{1}/_{2}$ years, was characterized by echolalic utterances and monotonous, staccato delivery. Play consisted of collecting toy cars and trucks which he enjoyed lining up and perseverative preoccupations from which he could not be distracted (watching objects spin or flicking objects). From age 3 to 5 years he was described as delayed in speech and language, unresponsive to adults and children, engaged in solitary play, and prone to temper outbursts and making noises. He was sensitive to loud noises resulting in extreme startle and covering of his ears, and was fascinated by anything colored green. His scores on the WISC-R were Full-scale IO 80, Verbal IO 77, and Performance IQ 86. During the index hospitalization, he evidenced the emergence of intense eye blinking, head jerks, lip licking, mouth tics, pulling at clothing tics, throat clearing and high-pitched shrieks, and reported he heard voices calling his name. Because he believed other patients talked about him, he engaged in frequent episodes of aggression. Presence of tics were noted in early childhood prior to the use of neuroleptics.

W also reported that when younger he heard voices calling his name and telling him "to do bad things" and he hallucinated a tree standing in the hallway of his home.

Head jerks, shoulder jerks, eye blinking, and chronic lip picking were evidence of a continuing tic disorder. Diagnoses of autistic disorder, TS, and schizophrenia-like psychosis were made and W was maintained on thiothixene 12 milligrams daily.

Case 3

V was a 34-year-old autistic man who attended a day treatment program. As an infant and child, V was not affectionate, resisted being held, seemed in his own world, showed disinterest in other children, did not communicate, and speech was perseverative. His play consisted of listening to the same music over and over and preoccupation with blowing up balloons. He was hyperactive, showed frequent rocking, and had temper outbursts which consisted of hand biting and thigh slapping. As an adult he evidenced obsessive-compulsive behaviors and was prone to temper tantrums when changes in his routine were imposed and his speech was monotonous, staccato in delivery, and mildly echolalic and literal. His score on the Stanford-Binet Intelligence Scale was IQ 75. V showed mouth, ankle, and wrist turning and face-touching tics.

V related episodes of hallucinating trees growing from his head and from the heads of staff members and "trees swaying back and forth in the wind." At other times he reported he heard trees telling him to hit people. On one occasion, V insisted that the toilet overflowed and became severely aggressive when staff tried to convince him that this was not so and that he did not need to mop the floor. Parents did not acknowledge family history of tic or psychiatric disorder, however, father who was a professional was perfectionistic and both parents showed repetitive finger tic movements. V's hallucinatory experience and aggressive outbursts were eliminated by treatment with fluphenazine 7.5 milligrams daily.

Case 4

E was a 37-year-old man who attended a day treatment program for autistic adults. As an infant he cried considerably, lacked affection for and interest in people, and showed absence of anticipatory behavior. As a child, he spoke little, showed poor eye contact, preferred being alone, lacked symbolic play and language, was echolalic, sensitive to smells, lined up buttons, and picked repetitively at the linoleum floor. As an older child, he talked to himself, rocked and flapped his hands, stared vacantly, and was preoccupied with trains and train schedules and he invented names for people. At age 10, he showed facial grimaces. His scores on the WAIS were Full-Scale IQ 84, Verbal IQ 79, Performance IQ 90.

As an adult E reported that he heard "good" and "bad" voices. The good voice told him he was doing well and the bad voice that he was bad, was not a good person and that he "should go to hell." He threw objects in response to the bad voice.

È's affect was blunted, constricted, and inappropriate. Speech was monotonous and staccato-like and conversation was characterized by failure to appreciate the listener's frame of reference. Purposeless and repetitive face-touching tics and tongue protrusion tics were evident. Treatment with mesoridazine resulted in decreased episodes of hallucination and aggressive behavior.

DISCUSSION

The cases of coexisting autistic disorder and schizophrenia-like psychosis reported here combined with other reports of patients with coexistence of the disorders provide support for the position that there exists a subgroup of autistic individuals who are at risk for the development of schizophrenia-like disorder. Issues involving diagnosis and ascertainment of subjects may partially explain the divergent opinions in studies that have examined the relationship between autistic disorder and schizophrenia. In a study that compared DSM-III autistic, atypical autistic, and schizophreniform children (Volkmar et al., 1988) evidence suggested that some children who would have met DSM-III-R criteria for autistic disorder may actually have experienced schizophrenia-like disturbance. In addition, autistic children who develop schizophrenia-like psychosis may be more likely to be patients whose psychosis predominates and whose autistic process has ameliorated and gone undetected. This group of patients may be more likely to seek treatment from psychiatric facilities (Watkins et al., 1988) and therefore may not be sampled in studies whose subjects are drawn from autism and developmental disorder centers (Rumsey, Andreason, & Rapoport, 1986). The subjects sampled from autism centers are more likely to comprise "uncomplicated" cases of autism.

Our current understanding of the possible mode of inheritance of TS and the wide array of neuropsychiatric disturbances which can result when individuals are carriers of the TS gene (Comings, 1990; Comings & Comings, 1991; Sverd, 1989, 1991) may explain some cases of coexistence of the disorders. TS is a common hereditary tic disorder (Comings, 1990; Pauls & Leckman, 1986; Shapiro, Shapiro, Young, & Feinberg, 1988) whose manifestations include hallucinations, paranoid ideation, schizotypal thinking, and schizophrenia-like psychosis (Comings, 1990; Comings & Comings, 1987, 1991; Kerbeshian & Burd, 1985, 1988; Sverd, 1991; Takeuchi et al., 1986). It has been proposed that autistic disorder and TS are genetically related (Comings & Comings, 1991; Sverd, 1991). Both children described in this report had TS. The significant association between TS, autistic disorder, and schizophrenia-like disorder in these patients can be appreciated if it is considered that the chance co-occurrence of the three disorders would result in only 1 case per 100 million children given prevalence rates for TS of 1 in 100 boys (Comings, 1990), 10 cases of autistic disorder per 10,000 (Sverd, 1991), and 1 case of schizophrenia per 1,000 children (Burd & Kerbeshian, 1987; Kydd & Werry, 1982; Petty et al., 1984). In support of the hypothesis that the TS gene is partially responsible for the presence of the three disturbances is the presence of pleiotropic manifestations of the TS gene in the patients' maternal and paternal families in Cases 1 and 3. In Case 2, mother was reported to have been a drug abuser and the patient was adopted. Adoption is not an uncommon feature of a subgroup of TS patients and may be indicative of impulse control problems in the biological parents (Comings, 1990; Comings & Comings, 1991; Sverd, 1989). Compatible with the theory of a significant role for TS in the pathogenesis of autistic disorder and coexisting schizophrenic-like disorder is the report of the presence of tics in one patient with schizophrenia and autism described by Watkins et al. (1988), possible tics in some patients studied by Waterhouse et al. (1987), and spitting, a recognized TS behavior (Comings, 1990; Shapiro et al., 1988) in one of the cases described by Petty et al. (1984).

Although the two adults with coexisting autistic disorder and schizophrenic-like symptoms may represent chance co-occurrence of the disorders (Volkmar & Cohen, 1991) the presence of tics in both patients argues against an independent association and for a significant association between the three disorders. If one accepts a high prevalence for chronic motor tic disorder of 1 case per 100 adults, 10 cases of autistic disorder per 10,000, and 1 case of schizophrenia per 100, the probability of all three disturbances occurring in one patient would be 1 in 10 million.

REFERENCES

- Burd, L., & Kerbeshian, J. (1987). A North Dakota prevalence study of schizophrenia presenting in childhood. Journal of the American Academy of Child and Adolescent Psychiatry, 26, 347-350.
- Cantor, S., Evans, J., Pearce, J., & Pezzot-Pearce, T. (1982). Childhood schizophrenia: Present but not accounted for. American Journal of Psychiatry, 139, 758-762.
- Clarke, D. J., Littlejohns, C. S., Corbett, J. A., & Joseph, S. (1989). Pervasive developmental disorders and psychosis in adult life. British Journal of Psychiatry, 155, 692-699.
- Comings, D. E. (1990). Tourette Syndrome and human behavior. Duarte, CA. Hope.
- Comings, D. E., & Comings, B. G. (1987). A controlled study of Tourette syndrome, IV Obsessions, compulsions and schizoid behaviors. American Journal of Human Genetics, 41, 782-803.
- Comings, D. E., & Comings, B. G. (1991). Clinical and genetic relationships between autismpervasive developmental disorder and Tourette syndrome: A study of 19 cases. American Journal of Medical Genetics, 39, 180-191.
- Green, W. H., Campbell, M., Hardesty, A. S., Grega, D. M., Padron-Gayol, M., Shell, J., & Erlenmeyer-Kimling, L. (1984). A comparison of schizophrenic and autistic children. *Journal of the American Academy of Child Psychiatry*, 23, 399-409.
- Kerbeshian, J., & Burd, L. (1985). Auditory hallucinations and atypical tic disorder: Case reports. Journal of Clinical Psychiatry, 46, 398-399.
- Kerbeshian, J., & Burd, L. (1988). Tourette disorder and schizophrenia in children. Neuroscience and Biobehavioral Reviews, 12, 267-270.
- Kolvin, I. (1971). Studies in childhood psychoses: I. Diagnostic criteria and classification. British Journal of Psychiatry, 118, 381-384.
- Kydd, R. R., & Werry, J. S. (1982). Schizophrenia in children under 16 years. Journal of Autism and Developmental Disorders, 12, 343-357.

Autism-Schizophrenia and Tourette Syndrome

- Pauls, D. L., & Leckman, J. F. (1986). The inheritance of Gilles de la Tourette's syndrome and associated behaviors. Evidence for autosomal dominant transmission. New England Journal of Medicine, 315, 993-997.
- Petty, L. K., Ornitz, E. M., Michelman, J. D., & Zimmerman, E. G. (1984). Autistic children who become schizophrenic. Archives of General Psychiatry, 41, 129-135.
- Realmuto, G. M., & August, G. J. (1991). Catatonia in autistic disorders: A sign of comorbidity or variable expression? *Journal of Autism and Developmental Disorders*, 21, 517-528.
- Rumsey, J. M., Andreason, N. C., & Rapoport, J. L. (1986). Thought, language and affective flattening in autistic adults. Archives of General Psychiatry, 43, 771-777.
 Rumsey, J. M., Rapoport, J. L., & Sceery, W. R. (1985). Autistic children as adults: Psychiatric,
- Rumsey, J. M., Rapoport, J. L., & Sceery, W. R. (1985). Autistic children as adults: Psychiatric, social and behavioral outcomes. *Journal of the American Academy of Child Psychiatry*, 24, 465-473.
- Russell, A. T., Bott, L., & Sammons, C. (1989). The phenomenology of schizophrenia occurring in childhood. Journal of the American Academy of Child and Adolescent Psychiatry, 28, 399-407.
- Rutter, M. (1972). Childhood schizophrenia reconsidered. Journal of Autism and Childhood Schizophrenia, 2, 315-337.
- Shapiro, A. K., Shapiro, E. S., Young, J. G., & Feinberg, T. E. (1988). Gilles de la Tourette syndrome (2nd ed.). New York: Raven.
- Sverd, J. (1989). Clinical presentations of the Tourette syndrome diathesis. Journal of the Multihandicapped Person, 2, 311-326.
- Sverd, J. (1991). Tourette syndrome and autistic disorder: A significant relationship. American Journal of Medical Genetics, 39, 173-179.
- Szatmari, P., Bartolucci, G., Finlayson, A., & and Krames, L. (1986) . A vote for Asperger's syndrome. Journal of Autism and Developmental Disorders, 16, 515-517.
- Takeuchi, K., Yamashita, M., Morikyo, M., Takeda, N., Morita, K., Tamura, T., & Karya, H. (1986). Gilles de la Tourette's syndrome and schizophrenia. *Journal of Nervous and Mental Diseases*, 174, 247-248.
- Volkmar, F. R., & Cohen, D. J. (1991). Comorbid association of autism and schizophrenia. American Journal of Psychiatry, 148, 1705-1707.
- Volkmar, F. R., Cohen, D. J., Hoshino, Y., Rende, R. D., & Paul, R. (1988). Phenomenology and classification of the childhood psychoses. *Psychological Medicine*, 18, 191-201.
- Waterhouse, L., Fein, D., Nath, J., & Snyder, D. (1987). Pervasive developmental disorders and schizophrenia occurring in childhood: A review of critical commentary. In G. L. Tischler (Ed.), *Diagnosis and classification in psychiatry. A critical appraisal of DSM-III* (pp. 335-368). Cambridge: Cambridge University Press.
- Watkins, J. M., Asarnow, R. F., & Tanguay, P. E. (1988). Symptom development in childhood onset schizophrenia. Journal of Child Psychology and Psychiatry, 29, 865-878.
- Wolff, S., & Chick, J. (1980). Schizoid personality in childhood: A controlled follow-up study: Psychological Medicine, 10, 85-100.