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MAJOR REVIEW

Schizotypal Disorder in Children—A Neglected Diagnosis

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Disabling psychotic-like perceptions, thoughts, and behavior have long been recognized in children. These symptoms have an adverse impact on child and family and are a developmental predictor of Schizophrenia Spectrum Disorders (SSD). Attempts to classify this phenomenon separately and within the Diagnostic and Statistical Manual of Mental Disorders (DSM) and International Classification of Diseases (ICD) systems have been unsuccessful until the DSM-5 and ICD-11. The categorization of Schizotypal Disorder within the SSDs in DSM-5 and ICD-11, and recognition that it is manifest in childhood, has established Schizotypal Disorder in Childhood (SDC) as a focus for clinical attention and research. This article aims to increase the awareness of this debilitating disorder by describing 3 case studies (ages 6, 8, and 9), which illustrate and refine the clinical presentation and cognitive profile of SDC. Biopsychosocial risk factors, comorbid disorders, and features that differentiate it from Autism Spectrum Disorder (ASD) are discussed. A comprehensive understanding of SDC will improve the accuracy and validity of the diagnostic process and pave the way for further research into its etiology, developmental pathway, and treatment.

Key words: childhood/schizotypal/psychosis/schizophrenia/Autism Spectrum Disorder/fantasy preoccupation/perceptual disturbance/paranoid ideas/odd behavior and speech/neurodevelopmental disorders

The Problem

For 60 years, there has been growing recognition that some children have a cluster of distressing and unusual social, communication, perceptual, thought, and behavioral difficulties and preoccupations with bizarre fantasies that have a detrimental impact on mental health and well-being.[1–6] We argue that it is timely, necessary, and sufficient to diagnose this neurodevelopmental psychopathology as Schizotypal Disorder (SD)[7,8] occurring in Childhood (SDC).[1,2,5,9]

There are disparate historical approaches to the recognition of these psychotic-like perturbations in children. Some do not recognize this disturbance as a disorder because "the character and personality of children is fluid and not persistent,"[10,11] or at most a manifestation of childhood reactive emotional states such as depression. Others have conceptualized the diagnosis of these children within either the Schizophrenia or Autism spectra (figure 1) in the Diagnostic and Statistical Manual of Mental Disorders (DSM) and International Classification of Diseases (ICD) systems by early terms such as "Schizophrenia, childhood type" (DSM-I, DSM-II, ICD-8),[12–14] "Pervasive Developmental Disorder NOS (PDD-NOS)" (DSM-III, DSM-IV-TR),[15,16] or "atypical autism" (ICD-10).[17] These diagnoses remained poorly defined and have not been retained in the current editions (DSM-5, ICD-11).[7,8] Another approach to categorize children with dysfunction in multiple emotional, behavioral and social domains, on the boundary between

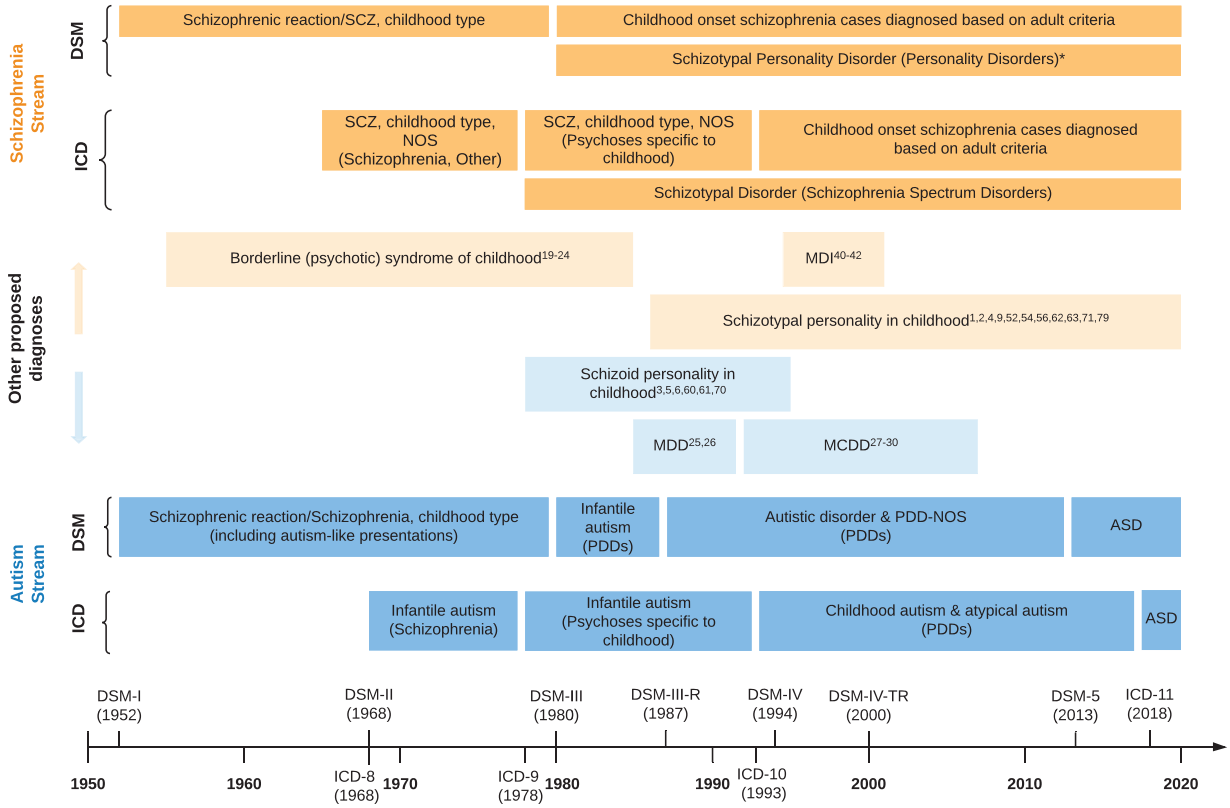

Fig. 1. The 70-year history of the diagnoses of psychosis, schizophrenia, and autism in childhood, included in, or excluded from, the DSM and ICD classification systems. Abbreviations: DSM, Diagnostic and Statistical Manual of Mental Disorders; ICD, International Classification of Diseases; SCZ, schizophrenia; NOS, Not Otherwise Specified; MDI, multidimensionally impaired; MDD, Multiplex Developmental Disorder; MCDD, Multiple Complex Developmental Disorder; PDDs, Pervasive Developmental Disorders; ASD, Autism Spectrum Disorder. *DSM-5 states that "the features of a Personality Disorder usually become recognizable during adolescence or early adult life," but also recognizes that "schizotypal personality disorder may be first apparent in childhood and adolescence."[7(p657)]

Schizophrenia and Autism,[18] was to group them under the term "borderline (psychotic) syndrome," a precursor to the distinct DSM-III adult categories of borderline and schizotypal personality disorders. This failed to yield greater diagnostic application in children.[19–24] Conversely, in the Autism/PDD literature a similar group of children were described with Multiplex Developmental Disorder or Multiple Complex Developmental Disorder.[25–30] However, these constructs were overinclusive and were not recognized by the DSM and the ICD, which continued with the unsatisfactory category of PDD-NOS.[31]

In the early schizophrenia literature, longitudinal research of psychopathology in children, including high-risk offspring of parents/mothers with schizophrenia, described children with odd behavior, rejection by peers, perceptual and thought disorder, and antisocial behavior. They were also more likely to develop schizophrenia in adolescence/early adulthood.[32–39] A landmark NIMH longitudinal study of 160 children who screened positive for schizophrenia symptoms described a group of "multidimensionally impaired" children. They presented with transient psychotic symptoms, emotional reactivity, preoccupation with fantasies, difficulties when interacting socially, and cognitive deficits with information processing and visuospatial skills.[40–42] The similarity of these children with the current criteria for SD within the schizophrenia spectrum is apparent.[43] They belong to a spectrum of recognizable schizotypal characteristics, influenced by neurodevelopmental, genetic, and psychosocial determinants[44–50] that vary from subtle impairments in functioning, to the fluctuant but persistent clinical disturbance of SD, and at its extremity to the decompensation of schizophrenia.[37,51,52]

SD is a complex condition, including cognitive-perceptual impairments, oddness, disorganization, and interpersonal difficulties.[53–55] In the ICD-11,[8(6A22)] SD is included within the group of Schizophrenia or other primary psychotic disorders. The DSM-5 describes SD in the cluster A group of Personality Disorders,[7(p655–659)] but also includes it within the Schizophrenia Spectrum Disorders (SSD)[7(p90)] given their overlap at the behavioral, neural, and cognitive levels.[56–59] It is characterized

by the presence of 5 or more of the following culturally abnormal symptoms: ideas of reference, odd beliefs and magical thinking (in children bizarre preoccupying fantasies), unusual perceptual experiences including bodily illusions, odd thinking and speech, suspiciousness or paranoid ideation, inappropriate or restricted affect, odd or eccentric behavior or appearance, lack of close friends, and social anxiety beyond reassurance associated with paranoid fears.

SD is usually diagnosed from young adulthood, but "may be first apparent in childhood"[7(p657)] and can be reliably and validly diagnosed in children aged 5–12 years.[1,2,6,9,56,60–63] The prevalence of SDC is not yet established. Population studies in a UK cohort of 2127, 12-year-old twins[64] and 27 000, 11- to 12-year-old Australian children[65] indicated a prevalence of definite psychotic symptoms of 5.9% and 10.2%, respectively, but attribution of symptoms to other causes such as sleep phenomena were not excluded. In a large birth cohort study (Avon longitudinal study of parents and children, ALSPAC), 38.9% of 6455 children aged 11.4–14.3 years (mean 12.9) were reported with one or more psychotic symptoms.[66] Following a structured clinical assessment (Diagnostic Interview Schedule for Children Version IV; DISC-IV),[67] 5.6% were identified with "definite psychotic-like symptoms" and 2.6% remained after eliminating those whose symptoms could be attributed to sleep, fever, organic causes or substance use. Given that the DISC-IV[67] does not specifically identify SDC, it is possible that this 2.6% would meet criteria for SDC.

Potential risk factors for SDC also require investigation, particularly those known to be associated with the development of psychotic symptoms, including genetics,[36,52] psychosocial and environmental risk factors,[68] childhood psychopathological disorders,[65] and childhood trauma.[52,69] The maturational pathway of SDC into adulthood and its salience to the onset of psychosis is unclear. Small longitudinal studies suggest that SDC symptoms persist and may develop into schizophrenia or other SSD.[1,4,32–34,56,61,70,71] There is some evidence for the relative stability of schizotypal traits in children,[52] but given the dynamic nature of brain plasticity and development in children, longitudinal studies are required to discover if SDC may resolve. For example, we reported that brain structural connectivity abnormalities observed in siblings of those with childhood schizophrenia normalized by mid-adolescence.[72] Further research on the developmental course and outcome of SDC is necessary, particularly regarding resilience as well as risk factors for psychosis.

What is the Symptomatology of SDC?

In addition to the DSM-5[7] and ICD-11[8] criteria for SD (box 1 and table 1), clinical features of SDC might include neurodevelopmental delays and deficits in motor functioning, including clumsiness, poor fine graphomotor skills, and limited motor planning.[1,6,9,60,70] Individual children might have a scatter of intellectual functioning and specific neurocognitive impairments similar to

Box 1. Diagnostic Formulation for Case Studies 1, 2 and 3

Schizotypal disorder: A "pattern of social and interpersonal deficits and cognitive/perceptual distortions and eccentric behavior" (p.655, DSM-5)[7] which begins in childhood, interferes with social and family life, attention, learning, and school participation, and causes emotional and behavioral disturbance; indicated by:

1. Ideas of reference
2. Bizarre fantasies and preoccupations
3. Perceptual disturbance (auditory, visual)
4. Odd thinking and speech
5. Suspicious (paranoid) ideation
6. Disturbed affect
7. Odd behavior
8. Disturbed ability to make and keep friends
9. Anxiety (and mood disturbance with periods of depression, irritability, and difficulty regulating emotions) associated with fearful paranoid preoccupying ideas and perceptions

Not in the course of:

1. Schizophrenia, as perceptual disturbance and paranoia fluctuate with periods of more normal mental state in the absence of negative symptoms.
2. ASD, as they are capable of periods of more normal social interaction, communication, and empathy. They are preoccupied with elaborative inner fantasies that distort their social interactions and play rather than having a focus on detail in the external world and the rigid, concrete interests and rituals apparent in ASD.

In the context of varied cognitive profiles:

Case Study 1 (MARK): wide scatter of average to very low cognitive skills, with relatively better verbal skills and greater difficulty with visuo-motor tasks. Performance compromised by his preoccupation with fantasies impairing his concentration on external tasks and social interactions.

Case study 2 (RUTH): low average-borderline ability compromised by inattention due to distraction by fantasies.

Case Study 3 (TOM): wide scatter of cognitive abilities with high average verbal and perceptual reasoning skills but significant difficulty with working memory and visuo-motor deficits impairing his handwriting. His cognitive performance was inhibited by difficulty shifting attention from his preoccupying thoughts.

Table 1. The Diagnostic Differentiation of Schizotypal Disorder in Childhood (SDC) from Autism Spectrum Disorder (ASD) Based on DSM-5[7] and ICD-11[8] Criteria

Diagnostic Features of Schizotypal Disorder in Childhood (SDC)	
Shared with ASD	Eccentric behavior, inappropriate affect and odd thinking and speech (in SDC, associated with fantasies, paranoid ideas and perceptual disturbances). Social and interpersonal deficits (in SDC, interaction is sought but distorted by fantasies, paranoid ideas and perceptual disturbance).
Specific to SDC	Preoccupation with odd beliefs, bizarre fantasies and magical thinking. Suspicious and paranoid ideas and ideas of reference. Perceptual disturbances including auditory hallucinations and bodily illusions. Social anxiety associated with paranoid fears.

Note: ASD, Autism Spectrum Disorder; SDC, Schizotypal Disorder in Childhood; DSM, Diagnostic and Statistical Manual of Mental Disorders; ICD, International Classification of Diseases.

those observed in adolescents with SD, including impaired processing speed, working memory and executive functioning.[1,6,73–75] Impairments in attentional self-control and set-shifting abilities might underlie the child's difficulties shifting from their preoccupation with unusual fantasies and perceptual disturbances in order to attend to external tasks.[54]

Can SDC be Differentiated From Other Neurodevelopmental Disorders?

SDC symptoms fluctuate in severity and are aggravated by stress and anxiety. In contrast, Schizophrenia in childhood presents with progressive, pervasive and disabling negative symptoms, disorganization, and progressively developing hallucinations and delusions.[56] SDC clearly shares some diagnostic criteria with DSM-5[7] and ICD-11[8] Autism Spectrum Disorder (ASD), including abnormalities of social communication and interaction, restricted fixated interests, and idiosyncratic behaviors. These well-described similarities[3,5,6,76] have led to diagnostic ambiguity as well as prognostic and therapeutic challenges, resulting in diagnostic overshadowing or substitution of SDC by ASD. This is further complicated by the allocation of funding for educational and disability services for ASD rather than for other diagnoses.[77,78] Nevertheless, there are key diagnostic criteria for SDC which differentiate it from ASD (table 1). In particular, SDC is defined by perceptual disturbances, a preoccupation with fantasies and paranoid thinking causing inattention, disturbed speech,[1,6] social difficulties, and anxiety.[1,61] A recent structural and functional brain imaging study suggests that the difficulty children with SDC have in shifting from internally focused attention differentiates them from neurotypical children and those with ASD, and might contribute to their abnormal perceptions and reality testing and inattention to external tasks.[79]

A major constraint on clinicians and researchers assessing children with schizotypal symptoms is that current classification systems state that SD "does not occur exclusively during the course of … ASD" (DSM-5)[7(p657)] or "excludes ASD" (ICD-11).[8(6A22)] Two international guidelines for the diagnosis of ASD, the UK National Institute for Health and Care Excellence (NICE) Clinical Guidelines[80,81] and the American Academy of Pediatrics (AAP) Practice Guidelines,[82,83] together with a leading structured diagnostic assessment instrument, the Autism Diagnostic Observational Schedule—Second Edition (ADOS),[84] do not acknowledge SDC as either a possible differential or a comorbid diagnosis. A diagnosis of ASD made in preference to that of SDC or in ignorance of comorbid SDC potentially disqualifies identification of schizotypal symptoms, with implications for etiology, treatment, and outcome. This is despite the evidence suggesting that a comorbid diagnosis of SDC can be identified in around 40% of children with ASD.[9,54,85] To address this issue, we developed a novel structured clinical diagnostic tool for SDC, the Melbourne Assessment of Schizotypy in Kids (MASK),[9,54] that facilitates the recognition of potential comorbidity of SDC with other neurodevelopmental conditions.

Case Studies

The neglect of SDC and the apparent barriers encountered by clinicians and researchers in identifying these children has potential adverse long-term consequences by facilitating misdiagnosis and a lack of research into outcome and treatment. The aim of this paper is to give these children a voice by providing detail of 3 de-identified case studies (with informed parental consent) that illuminate the clinical presentation of SDC. These children were independently diagnosed with DSM-5[7] criteria for SD by the authors B.J.T. and A.V.B. or R.T. in the process of the development of the MASK.[9] All met the DSM-5[7] diagnostic formulation for SDC presented in Box 1. Diagnostic features demonstrated by these children are flagged in brackets for each case. The history of biopsychosocial developmental risk factors for each child is presented in table 3, and their longer-term outcome is presented in table 4.

Case Study 1

MARK (5 y) was referred by a Pediatrician requesting an assessment for ASD and Attention-deficit/hyperactivity disorder (ADHD) because of his disruptive behavior at preschool.

Table 2. Neurocognitive Profiles

A. MARK

WPPSI-IV Index and *Subtests*[a]	Composite Scores	Percentile	Performance Range for Age
Verbal Comprehension:	85	16	Low Average:
Information		***16***	***Low average***
Similarities		***25***	***Average***
Visual Spatial:	79	8	Borderline-Low Average
Block Design		***<1***	***Very Low***
Object Assembly		*50*	*Average*
Fluid Reasoning:	79	8	Borderline-Low Average
Matrix Reasoning		***75***	***Average***
Picture Concepts		*<1*	*Very Low*
Working Memory	88	16	Low Average
Picture Memory		***16***	***Low average***
Zoo locations		*25*	*Average*
Processing Speed	79	8	Borderline-Low Average
Bug Search		***25***	***Average***
Cancellation		*5*	Borderline
Full-Scale IQ	**71**	**3**	**Very Low-Borderline** (This score is not interpretable due to significant discrepancy between the WM and PR indices and VC and PR indices)

B. TOM

WISC-IV Index	Standard Score	Percentile	Performance Range for Age
Verbal Comprehension (VC)	104	61	Average
Perceptual Reasoning (PR)	117	87	High Average
Working Memory*	80	9	Borderline
Processing Speed*	91	27	Low Average
Full-Scale IQ	(101)	(53)	(This score is not interpretable due to significant discrepancy between the WM and PR indices and VC and PR indices).
WIAT-II	Standard Score	Percentile	Performance Range
Word Reading	97	42	Average
Pseudoword Decoding	107	68	Average
Spelling*	87	19	Low Average
Numerical Operations**	132	98	Superior

Note: WPPSI-IV, Wechsler Preschool and Primary Scale of Intelligence—Fourth Edition[87]; WISC-IV, Wechsler Intelligence Scale for Children-Fourth Edition[89]; IQ, Intelligence Quotient; WIAT-II, Wechsler Individual Achievement Test-Second Edition. For Case Study 2 (RUTH), WISC-IV assessment was attempted but incomplete because of inattention due to preoccupying thoughts.
[a]WPPSI-IV Subtests in **bold** are used to determine **Full-Scale IQ.**
*Significant weakness (critical values) at the .05 level. **Significant strength (critical values) at the .05 level.

Assessment included an interview with mother, school reports, and a school observation. Mark's teacher described him as "at times delightful, caring and sociable but also feared by children and often excluded from class due to aggression, particularly when he is acting as another character." His mother described Mark as "mostly a lovely and kind boy with lots of personality" but who had difficulty playing with peers because he often has "characters from films and who he makes up in his mind" that make him angry and disruptive. She was "at her wit's end" because her son "suddenly switched into a real world for him of angry characters, then you can't reason with him." His preoccupation with his characters made him "excitable, aggressive towards children and inattentive in class activities."

Preschool Observation. Mark insists on wearing a T-shirt with a graphic image of a skull to preschool every day (odd behavior). Mark was observed participating and conversing happily in a cooperative and imaginative game of blocks with 3 children when suddenly he rushed to another group of children making threatening punching movements, yelling at them to stop talking about him because he was a "superhero" (ideas of reference/paranoia, odd beliefs, social difficulty). Following intervention by the teacher he became aloof, muttering aggressively whilst scribbling on paper. He suddenly ran across the room threatening to "stab" another teacher with play scissors, yelling "*Stop talking….it's not me…. Evil Mark will kill you.*" Later during a group garden walk he wandered off by himself, acting oddly, and talking as if he could see, hear and was being directed by a character called "Mario" (inappropriate affect, paranoid ideation, unusual perceptions with anxiety, social difficulties).

Psychiatric Mental State Examination (Mother Sitting Quietly in the Background). Mark was wearing his skull T-shirt. He was initially shy but soon made appropriate eye contact and chatted happily as he initiated an interactive and imaginative game with a toy train set. Suddenly he pointed at a red crayon mark on the wooden tracks, became agitated and started making punching movements shouting, "*that's evil blood….evil Mark….punch you in the face*" (odd behavior, perceptual disturbance, odd fantasies). He became incoherent and distressed and his mother came over and tried to calm him.

Later, he was unable to focus on visual mood scales (happy-sad, anxious-calm, angry-peaceful) but stabbed the sad-happy scale with his finger while saying incoherently, "*more bad red…..fighting him…I'll punch him real fast…..,*" then mumbled and appeared anxiously distressed (odd fantasies, paranoia, anxiety).

When invited to draw a dream (DAD task)[86] (figure 2), his affect changed from flat to agitated as he rapidly drew

Table 3. History of Biopsychosocial Risk for Case Studies 1, 2, and 3

	MARK (5 y 10 mo)	RUTH (8 y)	TOM (9 y)
Pregnancy Perinatal	Normal. Breastfed well. Described as a "cuddly baby".	Normal. Fetal distress-vacuum suction extraction. Normal Apgar scores (9). Breastfeeding ceased at 3 mo due to mastitis. Formula-fed well.	Normal. No pre- or post-natal concerns reported. Breastfed and slept well. Settled infant.
Early Development	Language normal. Motor delays. Remains clumsy.	Delayed language but used gestures well. Sentence speech at 3 y but "difficulty following her thoughts" made her "angry". Delayed gross motor skills, clumsy.	Language normal. Delayed gross motor skills. Remains "clumsy".
Early Trauma	Father described as aggressive and substance abusing. *18 mo old*: father left, no further contact. Stopped talking for 3 mo. *2 y old*: Mother in new stable relationship. Resumes phrase conversation. *3 y old*: speech difficult to understand at times. Relates better to babies/adults than peers. Disturbed sleep.	Stable parental relationship and care.	*18 mo old*: parents separated, amicable financial/access arrangements. Emotion regulation problems, which continue. *3 y old*: often "kind and friendly" fluctuated with "moody withdrawal", daydreams, paranoia, tantrums/aggression. *5 y old*: fluctuating mood dysregulation with times of "anxiety", low self-esteem, and "muddled thinking".
Family History	*Mother*: anxiety/mood disorder in adolescence, post-natal depression, depression (Mark 4 y). SSRI effective. *Paternal history*: Grandfather "psychosis", 2nd cousin ID. *Maternal history*: grandmother "anxious/odd".	*Mother*: fluctuating "depression" since adolescence. *Maternal history*: grandfather, grandmother, and aunts—"depression". *Paternal history*: grandfather—"schizophrenia", Uncle described as "strange recluse".	*Mother*: "depression" in adolescence. Post-natal depression/agoraphobia. Inpatient with "Bipolar Disorder" (Tom 30 mo old), then fluctuating mental health. Two more hospital admissions (Tom 8–9 y old). Mental health difficulties adversely affected the "care of her children". *Maternal history*: grandmother, Uncle, Great Aunt, "psychosis, bipolar".

Note: SSRI, selective serotonin reuptake inhibitor; ID, intellectual disability.

a dream from the left of the page which demonstrated paranoid ideas, perceptual (auditory/visual) and bodily disturbance, bizarre fantasies, fragmented speech and thoughts, and anxious distress.

He said, *"Darkness, I'm afraid, there's this guy wants to kill you." (*Clinician comments *(C): Kill you?)*

He wrote M on the smaller figure of himself. *"With a sword."* He wrote the word weapon. *(C: A weapon?)*

"Throws a bomb…. he says I'm bad….makes me a robot."

Mark then drew a picture of himself as "A robot" on the right with a "bomb" on his head. *(C: What are you doing?)*

"Punch him real fast," further muttering….as if in an internal conversation (*C: Can you hear someone*?)
"Yes, talking growling." (C: Talking?)

"Saying Yoshi you'll die." [Yoshi is his soft toy]. *(C: Is it your imagination?)*

"No, a real traff tragon." (C: A tragon?)

He rapidly did 3 more fragmented drawings with disjointed incoherent comments referring to *"little helfs," "evil school things," "scorpions,"* and *"Yoshi died."* He was very agitated and internally preoccupied. He took some minutes to calm down with mother's help. Mark turned his attention to a cooking game with his mother, chatting to her and with a smile, shared some "eggs on toast" with his mother and the clinician.

Neurocognitive and Behavioral Assessment and Other Investigations. A Wechsler Preschool and Primary Scale of Intelligence for Children-Fourth Edition

Table 4. Outcome and Follow-up for Case Studies 1, 2, and 3

	MARK (5 y 10 mo)	RUTH (8 y)	TOM (9 y)
Initial Management	Preschool consultation. Individual learning plan. Psychological treatment (anxiety, behavior). Parent education program.[90] Risperidone 0.5 mg/d.	School individual learning plan. Parent education program.[90] Psychological treatment (anxiety). Fluoxetine, Risperidone, Olanzapine, Aripiprazole tried but all ceased due to side effects.	School consultation and Counsellor. Parent education program.[90] Mental Health Nurse (family support). Risperidone to 1 mg/d.
6 mo	Risperidone increased to 1.5 mg/d. Improvement in learning, socialization, play, sleep.	Haloperidol 0.5 mg/d. Improved mental state and adjustment.	Improved mental state, socialization, learning. Risperidone ceased.
1 y	Psychosocial therapy continued. Risperidone ceased.	Psychosocial therapy continued. Haloperidol continued. Dose varied from 0.5 to 2 mg/d in response to stressors.	Deterioration with family stress and house move. Psychosocial treatment and Risperidone 1 mg/d resumed. Mental state improved.
2 y	Deterioration with transition to new class. Psychosocial treatment reinforced. Risperidone up to 1.5 mg/d gradually tailed off with improved mental state.	Family moved home. New teacher and class. Haloperidol continued. Dose adjusted in response to stressors.	Remains well. Risperidone 1 mg/d continued.
3 y	Deterioration with change of school. Risperidone to 2 mg/d but drowsiness side effects. Switched to Aripiprazole 5 mg/d. Improved mental state and behavior.	Onset of puberty (aged 11 y). Haloperidol 1 mg/d with some emotion regulation difficulty.	Deterioration with onset of puberty. Mother mentally unwell (inpatient). Unreliable medication compliance. Developed comorbid Major Depression. Psychological treatment continued. Father assumes primary care. Mental Health Nurse for family support. Modified school program. Risperidone 2.5 mg/d. Fluoxetine 10 mg/d.
4 y and beyond	Deterioration after mother away with work. Aripiprazole increased to 10 mg/d. Psychosocial treatment continued. Settled, with rich fantasies but able to separate fantasy from reality and attend to external world (last assessed aged 10 y).	Deterioration, withdrawn from school. Haloperidol increased to 2 mg/d. Onset of menses (aged 13 y). Haloperidol ceased (dystonia, mood lability). Risperidone 0.5 mg/d. Returns to school and modified educational program. Improved mental state/adjustment.	Improved mental state/euthymic. Fluoxetine ceased. Risperidone tailed off.
5 y	Deterioration, schizotypal symptoms re-emerged in context of social stressors (mother mentally unwell, breakup with his girlfriend). Psychological treatment and Aripiprazole 5 mg/d. Improved mental health (last assessed aged 15 y).		

(WPPSI-IV)[87] assessment suggested he was "probably in the borderline-low average range with a wide scatter of abilities" (table 2, A). He was "difficult to assess due to poor concentration and non-compliance due to daydreaming and preoccupation with imaginary cartoon characters." The Developmental Behaviour Checklist (DBC)[88] completed by mother and teacher revealed a total score of emotional and behavioral problems in the very high clinical range (96th percentile) and a positive screen on the "Psychosis" scale. Neurological and laboratory investigations were normal.

Case Study 2

RUTH (8 y) was referred by a Pediatrician who reported a 2-year history of Ruth unpredictably withdrawing into a fantasy world, laughing and talking to herself and becoming angry, and frightening other children (preoccupations with fantasies, social problems).

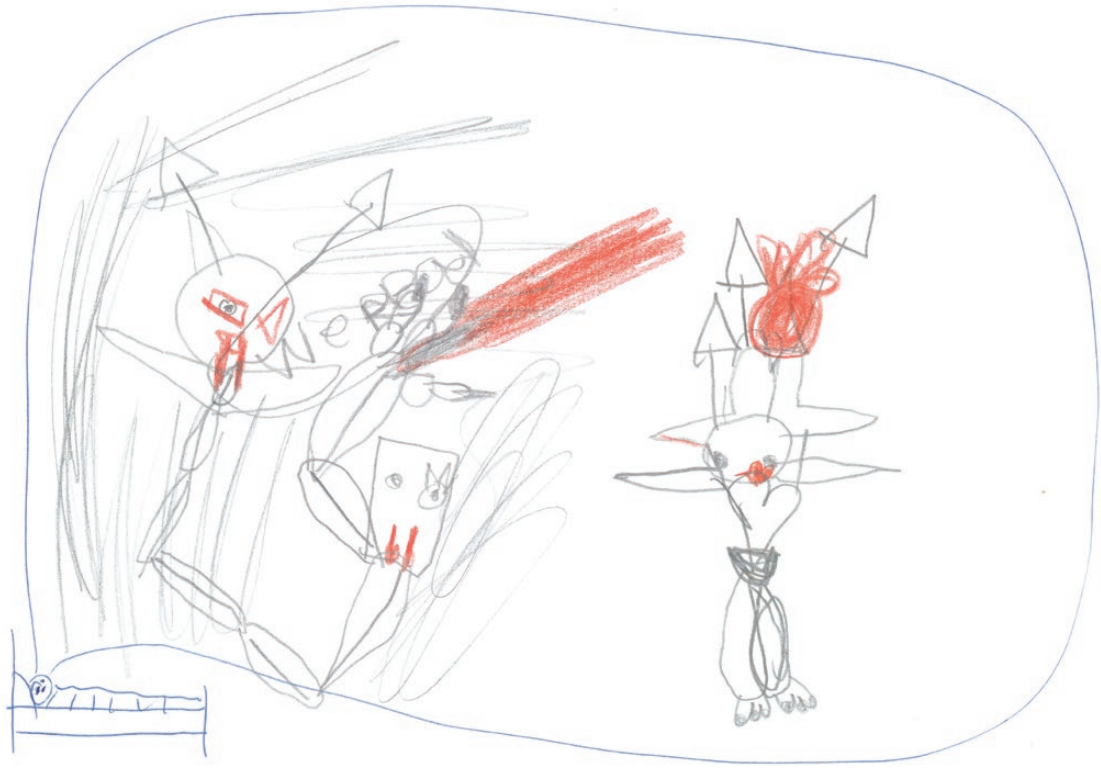

Fig. 2. MARK Draw a Dream Task. Mark is afraid that a "guy" wants to kill him ("M") with a sword ("weapon") and throw a "bomb" on the head of Mark, who is now a "robot."

The Pediatrician reported that it was "difficult to follow her conversation, which was rambling and off the point." The teacher reported that Ruth "often talked to herself using different voices as if in a conversation with others and required continual reassurance at school" (fantasy preoccupation, anxiety). Ruth's parents described her as often "anxious" believing that other children were hostile towards her (ideas of reference). She had a persistent fear of leopards but insisted on going to school dressed as one (odd behaviors and anxiety). Her mother and teacher described how Ruth "talked nonsense" and was inattentive "in a world of her own" (odd speech, fantasy preoccupation). She had been suspended from school for aggressive behavior. A DSM-IV[16] PDD-NOS diagnosis was made at age 4 due to "abnormal social interactions, irrelevant conversation, and a preoccupation with odd fantasies such as thinking she is an animal."

Psychiatric Mental State Examination (Both Parents Present). Ruth was dressed oddly in frayed leopard skin patterned clothes and on arrival crawled about, growling, *"I'm a leopard"* (odd behavior and beliefs). Her eye contact fluctuated, and her affect varied from appropriate to incongruous (inappropriate affect). She engaged in reciprocal conversation to direct questions about home and school, but during free play activities, her thinking became circumstantial and fragmented with self-referenced suspicious ideas and perceptual disturbance. For example, when asked what she liked to play with friends, she responded with animation, *"We play cartwheels and splits." (C: With your friends?)*

"Yes, outside with A and B." (C: With A and B?)

She became self-preoccupied. *"Outside….this girl I loved the most….like 15….she's mixed up….mean."* Looking upward as if listening. *"You're bad. I know where you live." (C: Who is bad?)*

She appeared frightened. *"She keeps looking at me." (C: Is she looking at you now?)*

"She's just behind me saying creepy things." (C: Saying creepy things?)
"Stop talking. I want you to be happy once."

She became agitated and curled up crying on a chair. It took some minutes for her to settle when comforted by her mother. Ruth then played with the doll's house in conversation with the clinician. When asked what she enjoyed playing at home, she responded warmly:

"Leopards and popstars." (C: How do you play Leopards and popstars?)

She became detached and agitated, as if preoccupied with her thoughts. With evidence of paranoid bizarre fantasy, perceptual disturbance, and fragmented thinking she said:

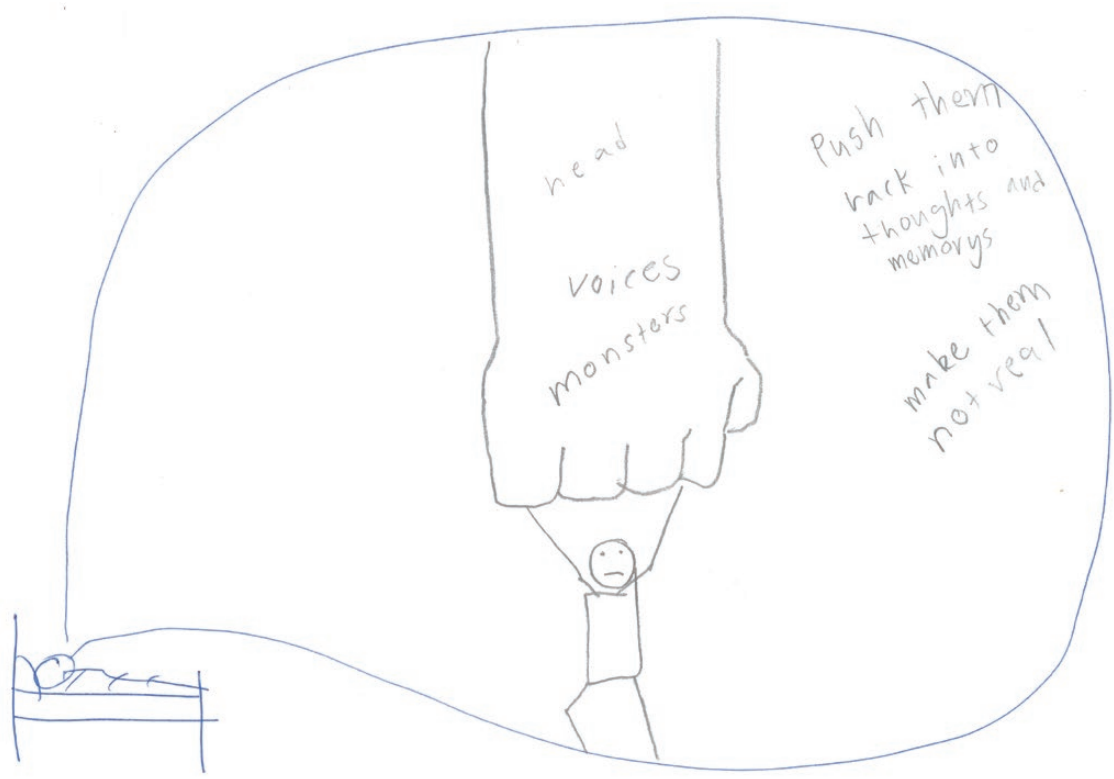

Fig. 3. RUTH draw a dream task. Ruth wants to recover from the perceptual disturbances and paranoid fear she suffers from in her "head."

"Something is banging on the door." Ruth looked fearfully towards the closed office door although all was quiet outside.
"It's creeping me out….a red eye.." (C: A red eye?)
"Yes. A red eye coming down." (C: Is it real? Where is it coming down?)

She appeared startled, wide-eyed and fearful.

"It is really coming down outside. There are people on the traffic island outside talking to a different girl."

Her mother comforted her telling her they would buy her an ice-cream on the way home, which helped settle Ruth.

Ruth responded happily when asked to do a drawing of a whole person. She drew a picture of herself with a heart-shaped body, lips and head, and repeated oval shapes for legs, but no arms. She added 2 arrow shapes on the head, which she called "spears." Three visual mood scales were drawn, and Ruth was asked to make a mark on the happy-sad, anxious-calm, and grumpy-peaceful scales to indicate where she had mostly felt over the past week.

On the happy-sad scale, she drew a picture of herself stating, *"I was so sad playing a game with Sue because he told me to be rude to her."* (social difficulties) *(C: Who told you to be rude?)*

"The angry voice." (C: A voice in your imagination?)
"No, he really told me." (*C: You really heard him tell you*?)

She said "yes" as she drew an angry face. *"He's angry in here"* (pointing to the face that she was drawing). *(C: Who is this you're drawing?)*

"…That's me." She cried tearfully. *"I'm sorry what I'm saying to him."* (perceptual disturbance).

Ruth settled when she was asked to show her responses to the worried and grumpy mood scales. Ruth scribbled with a red pencil over the eyes on the mood scale faces saying she was *"really worried (and grumpy) for what I'm saying about the angry voice."*

She readily responded to the invitation to draw a dream (DAD task),[86] rapidly becoming absorbed in drawing a "Monster" and commenting:

"He's a monster stopping me….a scary guy." Then loudly, *"Oh hahahah" (C: Is it your imagination?)*
"Oh no….he's real I can see him from inside my eyes." (*C: Is he talking to you*?)

She appeared fearful. *"He's saying I'm going to destroy you."* (perceptual disturbance, paranoia)

She became increasingly agitated, hitting the chair arm and her head with her fist. *(C: Is there any way I can help you Ruth?)*

She drew another dream (DAD task)[86] (figure 3) of distressing perceptions of "voices and monsters in her head." She asked the clinician to help by writing on the

picture *"push them back into thoughts and memories"* and *"make them not real"* (acute discomfort associated with paranoid fears).

Neurocognitive and Behavioral Assessment and Other Investigations. A Wechsler Intelligence Scale for Children-Fourth Edition (WISC-IV) assessment[89] at age 7 was attempted but incomplete because of "inattention due to preoccupation with irrelevant thoughts." Adaptive behavior, assessed by the teacher, was in the low average range. Total score of emotional and behavioral problems on the DBC[88] completed by mother and teacher was in the very high clinical range (99.8th percentile), with a positive screen on the "Psychosis" scale. Metabolic and chromosome investigations, electroencephalography results and brain scans were normal.

Case Study 3

TOM (9 y) was referred by the family doctor for erratic, impulsive and disruptive behavior, inattention, mood swings, and school refusal.

Mother reported that over the previous 6 months, Tom's mood had been unstable with irritability, angry outbursts, jealousy, and aggression towards his 2 older brothers. He was often preoccupied with "daydreams," had separation anxiety about being alone at home and at bedtime, and was fearful and anxious about falling asleep by himself. At school, Tom's irritability was alienating children, including his long-standing group of friends. He was inattentive, forgetful and disorganized, and had regressed in his learning, particularly in math. He tired more easily at sports and physical activities. Tom's motivation to attend school fluctuated, with some school refusal, distress, and tearfulness.

Psychiatric Mental State Examination. Tom was first seen together with his mother. He was initially shy and withdrawn with a flat affect but greeted the clinician politely and with appropriate eye contact. Throughout the session, he had a small teddy bear, which he took in and out of his pocket and talked to (odd behavior and fantasy). As his mother was reporting how irritable he could be towards family and friends, Tom became more animated with an incongruous smile, commenting that he was justified being angry because

> *"Everyone, even Mum is out to get me." (C: Why would that be?)*
> *"To ruin my day." (C: Would this just be at home or elsewhere?)*
> *"Everywhere." (C: You mean there are others in the community and at school trying to ruin your day?)*
> *"Yes, it's obvious. Zombies are everywhere." (C: How do you know this?)*
> *"I can see through things." (Ideas of reference, odd beliefs, and incongruous affect*).

Later when seen by himself, Tom was more engaged but fidgety. Initially, he related well and conversed about his friends and activities he enjoyed. He had a "secret" friend, which only he could "see and talk with and who helped him work out his worries" (perceptual disturbances). His mother reported that Tom would become distressed and scream in the car unless she buckled the car seat for his imaginary friend. Tom said that he had fearful, paranoid ideas of "*feeling the presence of scary zombies who touch me and talk to me*" (paranoid ideas, bodily illusions, auditory hallucinations). He struggled to sleep because at night, "the zombies visited" and could harm him. When asked about the "zombies" he claimed to "*feel their presence and ghosts as well*," and at times he could "*feel people touching me*" (bodily illusions). He believed these experiences were "*real and not just imaginary*." *(C: Do these zombies, ghosts or people touching you talk to you or can you see them?)*

> *"No, I can't hear anything but my brain tells me that they are real and to do things." (C: Is it actually your brain that tells you to do things?)* (perceptual disturbances, ideas of reference)
> *"I'm not sure but I think it's the zombies." (C: What sort of things are you told to do?)*
> *"Like drown under the water when I'm swimming." (C: Where do you go swimming?)*.

Tom talked about swimming with his friends but then became distracted, staring at the ceiling, and his speech became vague, disjointed and incomprehensible (odd thinking, inappropriate behavior and affect).

When asked to complete some visual mood scales, Tom had a fixed, silly smile and marked the scales indicating that he had widely fluctuating emotions of anxiety, depression and irritability. He denied suicidal ideas, including ideas of drowning himself when swimming because *"That is just my brain or ghosts telling me."* These fluctuating emotions related to *"kids at school having it in for me"* or when he didn't know where his mother *"might be at night"* or *"panic if my brothers are walking behind me"* (anxiety related to paranoid perceptions). His picture of a bad dream was of "*a ghost chasing me to eat me*" and his mother who "*couldn't help because she was sad*." (A probable reference to the impact of stress of his mother's mental illness on him).

Neurocognitive and Behavioral Assessment. During 2 assessment sessions, Tom was inattentive and distracted by preoccupying thoughts. His attention to tasks improved when prompted or given the opportunity to discuss what he was thinking about. Basic receptive and expressive language was age-appropriate, but in conversation, his train of thought was pressured and tangential, jumping to topics dominated by his preoccupying anxious thoughts. He fatigued quickly and needed frequent breaks.

WISC-IV assessment[89] revealed a wide scatter of abilities (table 2, B). He had excellent visual reasoning and construction skills but struggled with tasks of social comprehension and "working" and "long-term" memory, due to executive functioning difficulty in inhibiting and shifting his attention from his preoccupying thoughts. Academically, his spelling was adversely affected by this inattention and working memory difficulty.

Total score on the DBC[88] completed by mother was in the high clinical range (78th percentile), with a positive screen on the "Psychosis" scale.

Discussion of the Case Studies

The Assessment Process. The clinical assessment of the 3 children included several elements. First, a parent/teacher psychopathology screening tool—the Developmental Behaviour Checklist (DBC, DBC-2)[88,91]—indicated the possibility of clinically significant emotional and behavioral problems, including flag items for "Psychosis" and a screen for Autism.[92] There are a number of informant behavior checklists that also include questions regarding hallucinations, delusions, odd behaviors, speech/thought disorder, and ideas of reference.[43,93] These include the Behavior Assessment System for Children 2nd Edition, "atypicality scale" (BASC-2)[94]; the DSM oriented Psychotic Symptoms Scale[95]; the Child Behavior Checklist "thought problems" scale[96]; the Screening Questions for Psychotic Symptoms[66]; and the Schizotypal Personality Questionnaire for Children (SPQ-C),[52] which supports a 3-factor structure (cognitive/perceptual, interpersonal, disorganized, plus a general factor). The SPQ-C also has construct validity with respect to gene (family history) environment (childhood abuse) interaction.[52] However, these checklists might have low levels of agreement across multi-informants,[94] do not exclude symptoms attributed to sleep, fever, and substance use, and do not reliably confirm an SDC diagnosis.[52,66] At best, informant questionnaires indicate the likelihood of SDC requiring further clinical assessment.

Secondly, the diagnosis of SDC was determined by a comprehensive clinical assessment informed by structured interviews—the Schedule for Affective Disorders and Schizophrenia for School-Age Children (K-SADS-PL-DSM-5)[97] including the "Psychosis Supplement," and the Melbourne Assessment of Schizotypy in Kids (MASK)[9]—enhanced by mental state examination and observation conducted by an experienced clinician. Although the K-SADS and other robust structured clinical assessment tools such as the NIMH DISC-IV[67] have diagnostic algorithms for "Psychosis" and Schizophrenia, they do not specify an algorithm for SD. Therefore, with these tools, if the criteria for the diagnosis of Schizophrenia are not met, then the identified psychotic symptoms remain undiagnosed. To date, the MASK[9] is probably the only structured diagnostic interview that specifically elucidates a clinical diagnosis of SDC. In order to improve the recognition of SDC, clinicians should be alert to "atypical thought problems and psychotic symptoms" flagged in parent/teacher behavior checklists or indicated by structured clinical assessment tools, and follow-up with a focused clinical and mental state assessment to confirm that these symptoms meet criteria for SDC.

Comorbidity. All 3 cases presented a range of associated emotional and behavioral difficulties in addition to their SDC diagnosis. While some of these symptoms can clearly be attributed to the effects on emotions and behavior of the paranoid fears and perceptual and thought disturbances, others might also be regarded as comorbid disorders. For example, they all suffered from generalized anxiety, as well as their anxiety-related schizotypal fears. In a similar way, anxiety is also both inherent and comorbid with other neurodevelopmental disorders such as intellectual disability (ID), ADHD, and ASD.[98] Fluctuating depression (Dysthymia) was experienced by Ruth and Tom. Tom also had a later episode of Major Depression. Developmentally excessive inattention and distractibility were also present in all 3 children associated with their internal preoccupations, thus meeting criteria for ADHD of the Inattentive type. Significant problems with emotion regulation were apparent in all 3 children. Difficulties with the control of emotions are also a feature of other neurodevelopmental disorders such as ASD, ADHD, and ID.[98] A study of 700 children (6–12.9 y) attending mental health outpatient clinics in the United States[99] found 7% screened positive for "Psychosis" using the K-SADS.[100] These children were further identified as suffering a mixture of Major Depression, Bipolar Spectrum Disorder, ADHD, Post-traumatic Stress Disorder, and ASD but not Schizophrenia. Given that the K-SADS does not have an algorithm to identify SDC, and the Psychosis items of the K-SADS are congruent with the symptoms of SDC, then it might be likely that at least some of the 7% identified with "Psychosis," had SDC. The other conditions identified in any of these children would therefore be comorbid disorders. The longer-term association between SDC and comorbid conditions requires further study.

Cognitive Profile. All 3 children had a wide scatter of cognitive skills, with Ruth and Mark in the intellectually disabled to low average range and Tom in the borderline to high average range. The greatest impairments were identified in the active processing of information in working memory and self-directed attentional control; skills most vulnerable to disruption by preoccupation with internal fantasies. The ALSPAC study found that children (mean age 12.9 y) with low IQ (less than 79–89)

were at the highest risk of having "definite psychosis symptoms."[66] Those children with "higher than average IQ" also had a somewhat increased risk of psychosis.

Inherited Risk. There was a history of mental illness (psychosis, affective disorder) in the families of the 3 children, emphasizing the potential relevance of genetic influences that require further study.[36] There is evidence that family history might predicate the dysregulation aspects of SDC symptomatology and perhaps its attenuation with development.[52,55]

Trauma and Stress. Psychosocial stress and traumatic early life events were experienced at least by Mark and Tom. All 3 children deteriorated in their mental health at times of stressful life events, indicating that the stress response might be both a risk and perpetuating factor of salience to episodes of psychotic decompensation and perhaps predictive of later development of schizophrenia and psychosis.[65,68,69] Longitudinal studies are imperative to explore the evidence that suggests that childhood trauma increases the risk of SDC, particularly cognitive/perceptual disturbance, and provide further insights on gene-environment interactions.[52,101]

Why Diagnose SDC?

When making a diagnosis of SDC, care must be taken to ensure that the symptoms are not transient or a part of "normal developmental variation,"[43] such as vivid imaginary play and daydreaming. The symptoms must be a cause of distress, disability, and impaired adaptation and cannot be attributed to another cause such as a sleep phenomenon, fever, organic disorder, or substance use. The 3 cases clearly demonstrate that SD can be diagnosed in children and is likely to be a neurodevelopmental disorder. Its symptoms, although fluctuant and reactive to stress, are pervasive, persistent and cause considerable developmental disruption, emotional and behavioral problems and distress to the child and family.

Conclusion

Consistent with evidence that the brain has developmental continuity from childhood,[102] we contend that SDC is a Neurodevelopmental Disorder rather than emerging de novo in later adolescence as a first-episode psychosis. Because SDC is largely unrecognized and under-researched, a number of critical questions remain unanswered regarding phenomenology, epidemiology, etiology, and treatment. Our understanding of diagnostic criteria and comorbidity requires refinement; the developmental trajectory including prevalence and psychosis risk is unclear, although symptoms have been described to persist for 20–30 years[56,70,71]; the influence of culture and environment is uncertain; potential etiological factors such as birth trauma,[1,103] child abuse,[52,104,105] psychosis family history, and genetics[36,52,106] require further study; and specific features of brain function require confirmation.[79] Neurocognitive assessment, which demonstrates difficulty shifting attention from internal preoccupations and perceptual disturbances, can inform the management of educational, social, and behavioral difficulties and assist with differential diagnosis from other conditions such as ASD, where attention is externally focused.[54] Treatment and management remain speculative but include education for learning and motor difficulties, psychological treatment for stress and anxiety, ecological interventions for family interactions and parenting style, and biological treatments such as the use of neuroleptics.[107] Better recognition of SDC might result in effective early intervention to prevent psychosis in youth. It is critical not to leave children disabled with SDC to languish unrecognized.

Ethics Approval Statement

The project was approved by Monash University Research Ethics Committee (CF08/1420 - 2008000688: Investigation of unusual personality traits in children presenting to a Learning Difficulties Clinic) and Western/Melbourne Health Research Ethics Committee (2009.650: A pilot study investigating brain functioning in children with unusual thinking and atypical personality traits).

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