Letters to the editor

The potential role of electroconvulsive therapy in the 'Iron Triangle' of pediatric catatonia, autism, and psychosis

Shorter and Wachtel recently proposed that childhood catatonia, autism, and psychosis do not always occur as separate illnesses, but may constitute a distinctive subpopulation within the larger autism pool, representing a single underlying neuropsychiatric disturbance, or 'Iron Triangle' with a wide symptom constellation. The historical concomitance of symptoms from all three sides of the triangle—psychosis, catatonia, and autism—has been reviewed in the pediatric literature of the past 150 years, raising the question as to how this repeatedly documented hybrid presentation was largely ignored in favor of rigid diagnostic divisions between autism, catatonia, and schizophrenia, and associated distinct treatment paradigms. Two patient conundrums are presented as representative of the Iron Triangle, with the authors' suggestion that the common pathology of the three illnesses merits further study, and might offer improved treatment options for these most challenging cases (1).

Recently, the authors have successfully used electroconvulsive therapy as the primary treatment modality in a 13-year-old female whose psychiatric presentation is pathognomonic for the Iron Triangle, suggesting that ECT may have significant therapeutic potential in these distinctive cases.

Z. is a African-American female diagnosed with autism at age 3 who received early intervention services and subsequent special educational programming in a Western state without incident until age 9, when she developed sudden behavioral outbursts consisting of unprovoked aggression toward others, largely slapping faces of family members, teachers, and peers. Functional behavioral assessments did not identify any environmental trigger for this aggression. Within 1 year, she had started repetitively slapping her own face, to the point that both eyes were chronically bruised with diffuse ocular and facial edema, discoloration, and disfiguration. Headbanging on hard surfaces ensued a few months after the start of hand–head slapping, and Z. succeeded in putting her head through both drywall and a car window, necessitating placement of a protective sports helmet. At one point, she banged her head so intensely on the floor that she cracked her helmet in half. Episodes of self-injury were noted to occur across time and setting, again without any clear operant function.

Between the ages of 10–12, Z. had nine in-patient psychiatric hospitalizations for extreme self-injury and aggression. Aggression included slapping, kicking, biting, scratching, pinching, pushing, grabbing, head-butting, and hair-pulling and had led to multiple family and staff injuries. Self-injury consisted of hitting her face, head, or neck with an open hand or closed fist, as well as headbanging on hard surfaces and body slamming to the floor from a seated or standing position. Mother described these episodes 'like an internal explosion', which could occur

several times daily, and require up to 45 min of physical restraint to prevent injury. Z. was also noted to have mood instability and to typically display hyperactivity, inappropriate giggling and laughter prior to behavioral episodes, although these could also be preceded by staring. Primary and secondary insomnia was prominent since age 3, at times waking every 30 min and engaging in self-slapping episodes. By age 12, Z. was also noted to begin mumbling to herself and engaging in new-onset self-talk, as well as demonstrating outward distress while pressing her hands over her ears as if hearing something and tracking visual stimuli not evident to others. She was also noted to have episodes of staring and unresponsiveness.

We note that all three components of the Iron Triangle are clearly present in this history: her longstanding autism, the repetitive self-injurious behaviors recognized within catatonia (plus staring, posturing and psychomotor agitation), and her attentiveness to what were apparently aural hallucinations.

Multiple unsuccessful psychotropic trials over 3 years included methylphenidate and dextroamphetamine formulations, clonidine, atomoxetine, buspirone, naltrexone, valproic acid, oxcarbazepine, quetiapine, olanzapine, risperidone, aripiprazole, haloperidol, and lorazepam. Olanzapine and risperidone had both led to dystonic reactions.

Due to ongoing escalating behavioral episodes, acute risk of ongoing bodily harm and growing concern for concomitant psychosis, Z. was transferred to an intensive in-patient neurobehavioral unit at age 13. Admission medications included valproic acid 250 mg in the morning and afternoon and 500 mg at bedtime, haloperidol 1 mg TID, clonidine 0.1 mg QHS, and benztropine 0.25 mg BID. Z. presented as a very thin African-American female wearing a pink protective helmet who demonstrated prominent psychomotor agitation and irritable affect. She was observed to vigorously wag her finger at unseen visual stimuli, yelling as if accosting someone directly in front of her, screaming 'stop it' with various expletives, and then engaging in sudden bursts of repetitive hand–head slapping or headbanging. This alternated with minutes of stupor, unresponsiveness and staring associated with frank rigid posturing of the arms and hands, as well as echopraxia of both the upper and lower extremities as if rowing a boat. Please refer to "Video Clip S1:SB Classic Iron Triangle Example." Negativism was present from the first day of admission in the form of food refusal. Mutism was present with the exception of the aforementioned cursing and yelling and echolalia for greetings and simple questions. Presence of multiple catatonic symptoms of posturing, rigidity, staring, echophenomena, mutism, negativism, and behavioral agitation led to prompt discontinuation of haloperidol to determine whether the antipsychotic was causing or exacerbating the catatonic symptoms. Valproic acid was weaned concomitantly due to reported lack of efficacy, and during the first several days of the admission, the aforementioned symptoms gradually decreased, although some

sudden SIB bursts occurred leading to chin lacerations in areas of already-friable skin. However, after 5 weeks without antipsychotic therapy, Z. presented again with the same constellation of physical agitation including repetitive self-injury and aggression toward staff, yelling, cursing, posturing, boat-rowing echopraxia, and intermittent staring as well as both verbal and physical aggression to unseen visual stimuli. She required isolation in a padded treatment room, 24 h usage of a protective helmet, as well as usage of protective equipment by all staff.

Given the floridly psychotic presentation, a trial of fluphenazine was pursued starting at 0.5 mg thrice daily with vigilant monitoring for any worsening of catatonic symptoms, including thrice daily vital signs and daily complete blood counts and creatinine phosphokinase for the first 2 weeks of fluphenazine titration. Fluphenazine was chosen over other antipsychotics based on literature regarding possible benefits of combined D1 and D2 antagonism in self-injurious behaviors (3). Psychotic and catatonic symptoms, including self-injury, abated as fluphenazine were titrated upwards to 3 mg thrice daily, and Z. was successfully discharged home on fluphenazine and benztropine with a complementary behavioral protocol for leisure and demands situations.

Despite medication compliance, Z. relapsed a few weeks later with the same psychotic and catatonic symptoms; rehospitalization was prompted due to concern for renewed head injury from SIB. A course of electroconvulsive therapy was chosen based on the growing literature of its safety and efficacy of such in pediatric affective, psychotic, and catatonic illness, including individuals with autism and repetitive self-injury as an alternate symptom of agitated catatonia (4–6). At time of admission, Z. demonstrated ongoing SIB toward her head, and Bush-Francis Catatonia Rating Scale (BFCRS) score was 19. Ophthalmological examination revealed bilateral operculated and atrophic retinal holes, but without acute tear or detachment. An index course of 12 bilateral ECT resulted in drastic reduction in self-injurious behavior, as well as marked affective improvement and increased verbal output, including direct response to questions and spontaneous initiation of conversation, a communication skill previously not seen for years. Although fluphenazine was tapered prior to ECT start due to propensity for antipsychotics to worsen catatonia, intermittent evidence of psychosis in the form of cursing, agitation, and aggression in response to internal stimuli led to reinitiation of fluphenazine 2.5 mg TID with excellent result. BFCRS scale at time of discharge was five, and Z. returned home with plan for maintenance ECT.

Z.'s case is demonstrative of the Iron Triangle and accordingly invites a reassessment of the autism spectrum disorders concept. Autism may not in fact represent a spectrum but a cluster of separate diagnoses having social withdrawal as their common symptom. One of the entities in that cluster may be the Iron Triangle, describing autistic children who also are psychotic and catatonic, and unlike other autistic children in other subpopulations in the autism pool. Z. responded remarkably well to ECT, with prominent reduction in SIB and other catatonic symptoms, as well as amelioration of psychosis and marked improvement in communication deficits, which may have been fueled by all three symptom clusters of the Triangle. Individuating diagnoses in this manner is important because children in the Iron Triangle subpopulation may well respond distinctively to such anticatatonic remedies as ECT in a way that other autistic children do not. The benefit of ECT may be substantial for these patients; not only did Z. experience acute symptom resolution, particularly of repetitive self-injury that had already caused retinal damage and is documented to have led to blindness in similar patients (5), she was able to return home after eleven in-patient admissions in 3 years and is expected to resume developmentally appropriate school, community, and family activities.

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Supporting information

Additional Supporting information may be found in the online version of this article:

Video Clip S1. SB classic iron triangle example.