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CASE REPORT

ECT for self-injury in an autistic boy

Lee E. Wachtel
Stephanie A. Contrucci-Kuhn
Merrie Griffin
Ainsley Thompson
Dirk M. Dhossche
Irving M. Reti

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L.E. Wachtel, MD (✉)
S.A. Contrucci-Kuhn, PhD
A. Thompson, MA
Kennedy Krieger Institute
Johns Hopkins School of Medicine
707 North Broadway St., Rm 232
Baltimore, MD 21205, USA
E-Mail: wachtel@kennedykrieger.org

M. Griffin, CRNP
Department of Anesthesiology
Johns Hopkins School of Medicine
600 N. Wolfe St.
Baltimore, MD 21205, USA

D.M. Dhossche, MD, PhD
Department of Psychiatry
University of Mississippi Medical Center
2500 North State St.
Jackson, MS 39216, USA

I.M. Reti, MBBS
Department of Psychiatry
Johns Hopkins School of Medicine
600 N. Wolfe St., Meyer 3-181
Baltimore, MD 21205, USA

Abstract *Objective* Self-injurious behavior presents a significant challenge in autism, and first-line psychopharmacological and behavioral interventions have limited efficacy in some patients. These intractable cases may be responsive to electroconvulsive therapy. *Clinical picture* This article presents an eight-year-old boy with autism, mental retardation, prominent mood lability and a five-year history of extreme self-injurious behavior towards his head, averaging 109 self-injurious attempts hourly. The patient was at high risk for serious head trauma, and required usage of bilateral arm restraints and protective equipment (i.e., padding on shoulders, arms, and legs). All areas of daily functioning were profoundly impacted by dangerous self-injury. *Treatment* Fifteen bilateral ECT treatments resulted in excellent mood stabilization and reduction of self-injury to 19 attempts hourly, and maintenance ECT was pursued. The patient was able to return to developmentally-appropriate educational and social activities. *Conclusion* ECT should be considered in the treatment algorithm of refractory cases of severe self-injury in autism.

Key words self-injury – autism – ECT – mental retardation – catatonia

Introduction

Classically defined as *any act directed towards oneself that results in tissue damage* [38], self-injury occurs regularly in individuals with autism and other developmental disabilities, with prevalence rates ranging from 5 to 66% [10, 36]. Self-injury varies widely in frequency and intensity, and has the potential to cause crippling and life-threatening bodily injury and death, as well as impair interpersonal, social, educational and occupational functioning with increased risk for institutional placement [10, 27, 31].

Multiple biological [1, 11, 36] and psychological [2, 19] theories have been postulated to explain self-injury in autism and other developmental disabilities, and proposed treatments for self-injury are accordingly myriad.

Psychopharmacological interventions reported to have efficacy in some cases include antipsychotics, antidepressants, anxiolytics, anticonvulsants, opioid antagonists and antihypertensives [1, 11, 29]. Cur-

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rently, risperidone is the only drug with a US Federal Drug Administration indication for self-injury in autism, approved after a multi-site clinical trial in autistic children conducted by the Research Units on Pediatric Psychopharmacology [30]. When self-injury is considered as part of the catatonic syndrome, benzodiazepines and/or electroconvulsive therapy (ECT) may offer reduction of this harmful, repetitive motor activity [18, 19].

Although psychopharmacological agents are commonly tried for self-injury, medication interventions in autism are often limited by a high rate of adverse effects [2], and some suggest that psychotropic interventions for self-injury may produce effects simply through sedation and chemical restraint, leading to excessive medication prescription and polypharmacy [1, 29, 41]. Outcome measures reported for behavioral interventions for self-injury suggest a high degree of effectiveness [24], yet treatment failures may well be underreported in the literature [23], and clearly not all patients are responsive to behavioral interventions.

Thus, self-injury may remain resistant to psychotropic and behavioral interventions. The following case demonstrates that ECT may have a role in the resolution of intractable self-injury.

Case report

We report an eight-year-old male with autism and mental retardation who presented with a five-year history of self-injury towards his head. Self-injury included slapping and punching his head as well as banging his head on his knees and shoulders, with daily rates averaging 109.3 attempts hourly based on 24-h data collection. On daily mental status examination, D. presented as an adorable little boy without any evident dysmorphology who chronically demonstrated multiple areas of erythema, edema and callous formation on his forehead and cheeks. While wearing bilateral arm restraints as well as arm and leg sports padding, D. repeatedly would strike his straightened arm to his head, strike his knees to his head, and hit his head and ears onto his shoulders. Without arm restraints or protective equipment, D. would immediately commence striking his forehead, cheeks and nose with a closed fist and the resounding crack of bone hitting bone.

D.'s self-injury severely impacted all areas of daily functioning. D. had never been able to participate in a structured learning environment at school or home due to ongoing self-injury, and his learning potential was completely unknown. Play and social activities were similarly interrupted by self-injury, and D.'s family functioning was sharply impacted. D. was also unable to undertake any self-care alone, even requiring hand-over-hand shadowing during all meals to block head blows while self-feeding.

D. had undergone extensive applied behavioral assessments along with behavioral and medication trials for 3 years without any sustained reduction in self-injury. Psychotropic trials included sertraline, fluoxetine, clomipramine, valproic acid, lithium carbonate, carbamazepine, oxcarbazepine, gabapentin, aripiprazole, quetiapine, risperidone, olanzapine, haloperidol, fluphenazine, propranolol, lorazepam and clozapine. Both sertraline and fluoxetine led to prominent agitation and behavioral exacerbation, clomipramine and fluphenazine led to transient, small reductions in self-injury, and the remaining medications had no impact on behavioral rates. An adequate lorazepam challenge for potential catatonia was unable to be completed due to development of disinhibition and associated increased irritability and rates of self-injury at a dosage of 1 mg thrice daily. D. participated in outpatient and inpatient behavioral assessments and interventions including, but not limited to multiple functional analyses, antecedent analyses, preference assessments, reinforcement based interventions (i.e., functional communication training, differential reinforcement procedures, noncontingent reinforcement), response reduction procedures (i.e., brief physical holds, contingent application of helmet), and bilateral arm restraints and protective equipment (i.e., padding). Although several interventions initially resulted in behavioral reductions, the effects were not long-lasting.

D. also exhibited significant mood instability characterized by irritability, tantrumming, alternating laughing and crying episodes as well as intermittent insomnia and anorexia. Negative affect was correlated with increased rates of self-injury. Medical history was noncontributory, although family history was positive for paternal grandfather requiring ECT for severe depression. D. did not evince any classic signs of catatonia, including posturing, echophenomena, rigidity, waxy flexibility, mutism (D. had never been verbal) or autonomic instability. However, he clearly demonstrated ongoing, stereotyped repetitive movements in the form of self-injury, as well as the above-noted periods of agitation.

Continuous self-injury of the head had required 24-h placement of D. in bilateral arm restraints with rigid metal stays, a cervical immobilizer as well as upper and lower extremity padding with protective sports equipment. Attempts at self-injury continued despite this highly restrictive situation, and a course of ECT was pursued.

Methods

Consent for ECT was obtained from D.'s parents after review by two child psychiatrists not involved with his care. A pediatric neurologist was also consulted as part of routine work-up and to review his seizure response. Modified ECT was supervised by Dr IR and administered with a MECTA Spectrum 5000Q unit. Anesthesia was induced by methohexital 50 mg iv, and succinylcholine 10 mg iv was administered for muscle relaxation. We opted for bitemporal electrode placement in light of prior case reports of both children and developmentally delayed adults with self-injury being successfully treated with bitemporal placement. Seizure activity was monitored clinically and by bifrontal EEG. Recorded seizure length was ascertained by EEG. Seizure threshold was estimated at 56 millicoloumbs (mC) and 15 treatments were administered over 5 weeks on a thrice weekly schedule during the acute course before tapering was commenced. Mean charge administered over the acute course was 168mC and average seizure length was 145 s. On three occasions seizures were terminated by propofol 15 mg iv when they exceeded 180 s.

Results

A profound reduction in rates of self-injury was observed in D. after the first ECT treatment. Upon waking, D.'s arm restraints were not immediately replaced and for a full ninety minutes D. was observed to not engage in a single episode of self-injury. He sat calmly in his bed, smiled and looked around his room, then rose to run about the unit with outward laughing. This result was astounding, in that prior to ECT, D. had consistently engaged in high-frequency and high-intensity hand-to-head SIB within seconds of arm restraint removal, leading to rapid facial edema and erythema, as well as frequent nose bleeds.

A consistent significant reduction in SIB was observed over the next 5 weeks while D. continued to receive bilateral ECT thrice weekly. Specifically, prior to the initiation of ECT, rates of self-injury were 100.6 h while wearing restraints and 109.3 h without restraint. During the reported phase of the first 5 weeks of ECT, rates of self-injury were reduced to 6.5 h with restraints and 19.4 h without restraints. Restraints after ECT consisted *only* of empty canvas sleeves (i.e. without the prior rigid metal stays in the sleeves, cervical immobilizer and full limb padding). See Fig. 1.

In addition to the reduction in SIB, D. was also able for the first time in his life to reliably engage in both

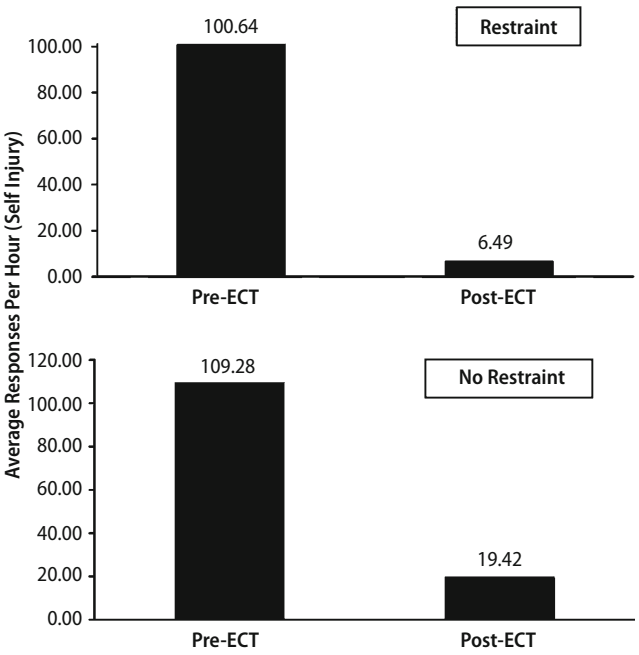

Fig. 1 Rates of self-injury before and after ECT

play and educational activities. D. was able to consistently work on daily structured academic tasks, thus affording his first opportunity to work on developmentally-appropriate educational goals with much potential for improved future functioning. He was also able to engage in meaningful family activities to the profound astonishment of his parents.

Due to the unknown effects of ECT on the brain of a nine-year-old, a repeat brain MRI post-ECT was obtained. It showed no changes compared with the pre-ECT MRI.

Discussion

As far as we are aware, this is the first published case report of intractable self-injury in a disabled prepubertal child being treated successfully with ECT. Only three English-language reports exist of ECT being used expressly for self-injury, one in an adolescent and two in an adult. A 14-year-old male with mental retardation, persistent headbanging and self-scratching had reduction of both topographies with bilateral ECT [18], a 25-year-old adult male with severe mental retardation and a several year history of head self-injury also showed remission with bilateral ECT [6], and a 32-year-old adult male with normal cognition, bipolar psychotic depression and multiple topographies of self-mutilation demonstrated excellent response to ECT [4]. There is also additional literature of successful ECT usage in children and

adolescents with pediatric psychotic or catatonic affective illnesses that were complicated by additional symptoms of self-injury. Carr et al. [13.] present a 12-year-old girl with bipolar affective disorder and concomitant body slamming self-injury, Black et al. [9] report an 11-year-old boy with depression, headbanging and self-biting, Cizadlo and Wheaton [15] discuss an eight-year-old girl with depression, catatonia and hand-to-head self-injury, Wachtel et al. [42] present an 18-year-old girl with autism, catatonia and severe hand-head self-injury resulting in nearly complete blindness from retinal detachment and Chung & Varghese [14] report an 11-year-old girl with psychosis, catatonia, headbanging and self-scratching. Over half century ago, Stauder also reported young adults with lethal catatonia who further demonstrated self-injury [37], and Leonhard vividly described self-injury in his writings on childhood catatonia [28]. Although the above reports present self-injury as an additional symptom along with catatonia, a more comprehensive conceptualization might consider self-injury as an integral part of the catatonic constellation.

Indeed, the question of catatonia as a possible etiology for D.'s extreme rates of self-injury is intriguing. Current thought supports the existence of catatonia as its own entity, with a myriad of psychiatric, neurological, medical and drug-related causative factors [3, 19]. Comprehensive catatonia rating scales such as the Bush-Francis Catatonia Rating Scale include symptoms such as excitement and motor unrest, stereotypy, mannerisms, impulsivity and sudden engagement in inappropriate behavior, as well as combativeness with potential for harm [12]. We believe that self-injury such as that demonstrated by D. would clearly meet such criteria.

Furthermore, catatonia is known to occur most commonly in bipolar affective illness [19], and D.'s ongoing mood lability, irritability, sleep disturbance and family history supported a potential bipolar diagnosis. Additionally, catatonia has been discovered to occur in autistic individuals like D. at increased rates of 12–17%, with ECT included in the current treatment guidelines if lorazepam fails [8, 20, 46].

Self-injury may also be explained in some cases by monoamine dysfunction, which can conceivably be reversed by ECT. For example, ECT has been shown to increase GABAergic transmission [34] which may ameliorate lack of GABA inhibition associated with SIB [36]. Similarly, decreased dopamine and serotonin function associated with SIB [36] may be rectified by ECT [47]. Additionally, theories of ECT's overall effectiveness discuss the hypothalamic release of neurohumors previously lacking in the patient's brain, as well as an overall normalization of the hypothalamic-pituitary-adrenal (HPA) axis [18]. This may be particularly salient in D.'s case, and we question whether ECT may also cause the release of a neurohumor or neurohumoral cascade with beneficial effects on self-injury. Finally, there is evidence in both primates and humans with developmental disabilities that HPA-axis dysfunction, specifically the uncoupling of proopiomelanocortin (POMC)-derived stress hormones, is a correlate of self-injury [26, 35, 40]. Possibly, peripheral uncoupling may reflect central dysregulation of the HPA axis which is sensitive to the therapeutic effects of ECT [48].

Although ECT in adolescents has proven to be safe and effective [43, 44], its use in prepubertal children has been controversial. D.'s case was no exception, with many concerns raised regarding the usage of ECT in a developmentally disabled eight-year-old, the unknown later effects on his developing brain and possible legal issues. Much of this may have been fueled by anti-ECT prejudice which continues to plague psychiatry, often preventing prompt access to life-saving treatment. Ironically, the efficacious and safe usage of ECT in children with affective and psychotic illness has been documented since the 1940s [7, 22]. Modern ECT case reports in children aged 6–12 offer further support for its resolution of pediatric psychotic or catatonic affective illness [13, 15, 17, 33, 45].

The literature also supports the safe and successful usage of ECT in the mentally retarded population. Multiple case reports review the usage of ECT for affective, psychotic and catatonic illness in mentally retarded adults [5, 32, 39]. There also exists a growing body of literature on ECT usage in mentally retarded adolescents, particularly autistic adolescents with catatonic deterioration [16, 20, 21, 25, 42, 49]. We believe that the scientific support of ECT in normally-developing pediatric, adolescent and adult patients, as well as adolescent and adult mentally retarded individuals should be extended to the pre-pubertal mentally retarded population, particularly in cases of dire need such as D.

We believe as well that the potential benefits of ECT in this case significantly outweighed the standard ECT and anesthesia risks as well as the "element of the unknown" in terms of longterm effects of ECT on D.'s developing brain. Having already failed years of behavioral and pharmacological therapy, D. was otherwise destined to (a) remain immobilized in full-body restraint, or (b) continue to engage in extreme rates of self-injury towards his head, with obvious long-term risk for intracranial damage. ECT proved to be truly life-saving in this case, affording a severely disabled child the opportunity to resume his developmental trajectory and reintegrate into life outside a locked inpatient unit.

Conclusions

Self-injury is a dangerous and sharply impairing condition that afflicts many individuals with developmental disabilities. Psychiatric, behavioral, neurochemical and neuroendocrine conditions have all been proposed as potentially explanatory. Additionally, self-injury may also represent a symptom of catatonia as a repetitive and purposeless motoric activity that may be associated with other catatonic, affective or psychotic symptomology. Recognition of this option should invoke a lorazepam trial at adequate dosages, followed by consideration of ECT if the former is ineffective. Judicious usage of ECT in the developmentally-disabled population with severe self-injury may afford a significant opportunity for recovery. Further research into the concomitance of self-injury and catatonia is warranted.

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