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Maintenance Electroconvulsive Therapy for Agitation and Self-Injurious Behaviors in Autism Spectrum Disorder

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SUMMARY

Self-injurious behaviors (SIBs) in autism spectrum disorder (ASD) are destructive symptoms that can lead to dangerous injuries and life-threatening risks. Agitation and SIBs may not respond to psychopharmacological and behavioral interventions. There are reports in the literature on improvement after electroconvulsive therapy (ECT) in cases not responding pharmacotherapy. However, data on the efficacy of the therapy on the benefiting patients, the course of the treatment and on the use of maintenance ECT (m-ECT) are very limited. This report presents the clinical features and the course of m-ECT on two cases under follow up for pharmacotherapy resistant ASD with significant agitation, mood disorder and SIBs that could cause severe head traumas.

The initial stage of therapy consisted of 7 sessions of ECT patients showed improvement after the 5th session. m-ECT were started since the agitation repeated one week after discharge despite ongoing pharmacotherapy. In the first case, m-ECT was continued once every two weeks for a total of 46 sessions; and in the second case a total of 18 weekly sessions were conducted. No significant side effects or complications were observed and the general state of well-being was preserved. Our paper is among the few that reported succesful treatment of agitation with m-ECT. m-ECT should be considered in treatment resistant cases.

Keywords: Autism spectrum disorder, self-injurious behavior, maintenance electroconvulsive therapy

INTRODUCTION

The maintenance electroconvulsive therapy (m-ECT) is one of the current treatment options when pharmacotherapy is insufficient in mainly unipolar depression, bipolar depression, bipolar mania, catatonic situations, resistant schizophrenia and schizoaffective disorders. ECT is also being used, although rarely, for autism spectrum disorder (ASD) and mental disability. Positive results have been reported in related recent studies even though m-ECT does not take place in the routine treatment protocols. Consequentially, some authors have argued that besides being taken as the last resort treatment approach, implementing m-ECT at an earlier stage could also be crucial for the clinical course (Ak 2018). Age limit for ECT use has not been stipulated. It is applicable comfortably during pregnancy and in the postpartum period even as the first option treatment.

Self-injurious behavior (SIB) is described as "a class of behavior which is generally repetitive and rhythmic and results in physical damage to the individual". SIB and agitation are destructive conditions that can lead to dangerous tissue injuries and severe psychological problems mostly seen in ASD (Wachtel et al. 2018). Prevalence of SIB among children and adolescents diagnosed with ASD is variable and mostly reported to be between 30% and 40% (Dominick et al. 2007, South et al. 2005). The deficiency of verbal and non-verbal communication skills in children and adolescents as compared to their peers causes increased behaviours of harming themselves and others (Matson et al. 2009). Another risk factor for SIB is the inefficiency of the sensory processing

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process. It has been speculated that the desensitization to heat, pressure and pain resulting from ASD is compensated by gaining bodily awareness through SIBs (Duerden et al. 2012). Age, the need for sameness, rituals and obsessions are also defined as risk factors for SIB (Duerden et al. 2012).

More frequent observation of SIB in individuals with ASD and mental disability, has not been adequately comprehended and has been difficult to treat with psychological and pharmacological methods. It has been proposed that ECT can be used to treat repetitive SIBs in ASD when pharmacological and behavioral interventions are ineffective (Wachtel et al. 2018). Given the adverse cardio-metabolic and endocrine side effects such as weight gain and hyperprolactineaemia and extrapyramidal symptoms, recent research has been focused on the efficiency of ECT in agitation (Ellawala 2015). It has been observed in the last decade that ECT has a quick and well-tolerated effect on these cases (Dhosssche et al. 2006).

Besides SIB, catatonia is also frequently comorbid with ASD and other developmental disorders (Wachtel et al. 2018). The high number of case reports evinces the effective use of ECT for catatonia in individuals diagnosed with ASD (Sajith et al. 2017, Wachtel et al. 2018). SIB can be considered as a stereotypic classic catatonia symptom which fully responds to ECT. Comorbidity of catatonia like symptoms frequently consisting of difficulty to start and complete movement, symptoms resembling those of Parkinson's disease, slowness in verbal response, inability to distinguish between day and night, excitability, aggression and repetitive behaviours (Wing and Shah 2000).

In this report the process of m-ECT use and its outcomes on two ASD patients with agitation, agressiveness and severe SIB not responding to pharmacotherapy will be presented.

Case 1

The 19-year old single female patient living in family together with her two siblings, had been diagnosed with ASD at the age of 3.5 years. After having had education until the age of 17 years, she was withdrawn from special education as her parents were convinced that she could not gain the necessary social skills. It was learned from her mother that, starting approximately by the age of 3.5 years, the patient started to turn around herself, watch and try to enter the washing machine, put sand and stones in her mouth, shake from side to side while standing at the same place with a fixed gaze for hours, posturing, smelling animal faeces or her intimate bodily areas. In the previous 6 months she had started banging her head against the wall, attempting to ingest the blood exuding from the wounding which necessitated consulting a healthcare center. She was put on, and used with regularity, valproic acid (400 mg/day), lorazepam (7.5 mg/day), haloperidol (5 mg/day), fluvoxamine (400 mg/day), aripiprazole (15 mg/day), risperidone (5 mg/day), olanzapine (20 mg/day), but had not distinctly benefited from the treatment.

The patient, second eldest of four siblings, was born in term with normal delivery at a hospital, weighing 2.7 kg. Her motor skill development was retarded compared to her peers, such that she was able to walk without any support after the age of 9 and that she still could not perform her personal care without the help of her mother. The patient was able to say her first word at the age of 2.5 and still could not make meaningful sentences during the interviews. Extreme interest in turning objects, turning around herself and shaking her head were stereotypic behaviours still continuing since the age of 3.5. We were informed that she also had complaints of insomnia, especially inadequate sleep and poor appetite since childhood. Her medical records showed that previous pediatric neurology examination had not detected traces cerebral palsy or epilepsy in the patient who still could not control her bladder and bowel functions. Her parents did not have blood kinship and she did not have a family history of psychiatric disorder.

In her mental examination, she was conscious but not cooperative; her orientation could not be evaluated and eye contact could not be formed. Her personal care was bad, her physical development was delayed for her age, her posture was slightly forward leaning. Her stereotypic rocking back and forth, catatonic symptoms of remaining in the same position were noteworthy. When left on her own, she hit her head against the wall or hard objects. Her communication was limited, and she seemed inattentive. She reacted extremely towards any physical approach. Her score was 6 on the agitation subscale of the Brief Psychiatric Rating Scale (BPRS) and 52 on the Childhood Autism Rating Scale (CARS). Results of the routine laboratory investigations including thyroid function tests, liver-kidney function tests and haemogram were within normal limits, but vitamin B12 and D values were low and vitamin replacement was started. Pathology was not detected in the cranial magnetic resonance imaging (MRI) except the iron suture made for an injury caused by hitting her head. The routine awake electroencephalography was within normal limits.

Informed consent was taken from the parents to conduct ECT using the MECTA Spectrum 5000Q unit (MECTA Corp, Lake Oswego, Ore). Anesthesia induction was performed with 130 mg intravenous methohexital and muscle relaxation was achieved by intravenous administration of 100 mg succinylcholine. In accordance with previously reported success in patients displaying aggression and self harm, the bitemporal electrode placement method was elected for the patient. The attack activity was monitored clinically and with bifrontal electroencephalography. Important complications were not observed after the sessions conducted once a week. The patient's SIB started to decrease after the fifth session.

Her score on the agitation subscale of the BPRS regressed to 2. After the seventh ECT, the patient's condition stabilized and the formerly prescribed 2 mg clonazepam was discontinued while therapy was maintained on valproic acid (1000 mg), fluvoxamine (200 mg), aripiprazole (10 mg), and olanzapine (15 mg). The patient's complaints repeated after a week of suspending the ECT, when it was decided to conduct m-ECT every two weeks. After the 46th ECT session, recurrence or undesired effects were no longer observed.

Case 2

The 23-year old single male patient living with his parents consulted our outpatient with complaints of aggressive behaviours, screaming, forcing himself to vomit, swinging back and forth, thumb sucking and biting his hand that had been continuing since the age of 1.5 years. He had previously been recommended treatment at another healthcare center with risperidone (4 mg/day) and sertraline (100 mg/day). Despite having following this treatment for 2 years on a regular basis, it was terminated as improvement was not observed in his condition. We were informed that the family had consulted a hospital for vomiting in early stages of life after milk intake but a diagnosis had not been made. When the patient was about 30 months old, he was diagnosed with phenylketonuria and since then he had been following the phenylketonuria diet and developed insomnia when not complying it. The patient received special education for 2 years but wasn't able to continue on problems of adaptation.

The patient was born with normal delivery, without any complications except the maternal bleeding history. The patient was breastfed for 1.5 years but had vomiting episodes after ingesting the milk. The patient started to walk at the age of seven and started to talk at the age of ten by saying "mum". The patient did not have proper bladder and bowel control till the age of ten and started to go to the toilet with his mother after the age of ten. It was learnt that the patient could not usually set any games, had an overwhelming interest in revolving objects, overreacted when the position of the objects in his room were changed, also reacted to noise by closing his ears, could not communicate with his friends and that he spent time on his own. His parents did not have blood kinship and his family history was uneventful.

In his mental examination he was conscious. His orientation could not be assessed as it was not possible to achieve cooperation or eye contact. The patient's personal care was poor, his physical development appeared retarded. He displayed stereotypic swinging. Bite scars on his hands and ecchymosis were observed in some areas. Mental deficiency with cognitive functions not in compliance with his age was observed. The patient made abnormal sounds and did not have meaningful speech. When touched, he responded by shouting.

Given the patient's symptoms of limited social communication, not sharing feelings, not making eye contact, extreme sensitivity to sounds, inattentiveness to his environment, keen interest in turning objects, swinging, and persistence in sameness, his condition was diagnosed with ASD in the foreground according to the criteria of the *Diagnostic and Statistical Manual for Mental Disorders* (DSM) (American Psychiatric Association 2013). Considering phenylketonuria comorbidity and the delay in developmental stages, it was decided that the patient also had moderate level of intellectual disability.

The patient was hospitalized for reasons of nervousness, uneasiness, aggressiveness and SIB. The routine laboratory investigation results were within normal limits and abnormalities were not observed in EEG and MRI. The patient's scores were 6 on the agitation subscale of the BPRS and 46 on the CARS. Treatment was started on lorazepam (2 mg/day), olanzapine (20 mg/day), carbamazepine (400 mg/day), escitalopram (20 mg/day) but the symptoms persisted for 3 weeks under this therapy and decision was taken to intervene with ECT under anesthesia after obtaining informed consent of the parents. The weekly ECT treatment protocol was the same as for Case 1. The patient's aggression decreased after the fifth ECT session and his score on the BPRS agitation subscale regressed to 3. As the patient's complaints repeated upon suspending the ECT after seven weekly sessions, continuation was planned with m-ECT. Once the patient's condition stabilized, ECT was performed every two weeks. There were not any complications. The patient, whose aggression and SIB regressed considerably after the 18th ECT, is currently under m-ECT treatment on an outpatient basis.

DISCUSSION

M-ECT is being used for diverse psychiatric disorders in adults. In 2008, the American Psychiatric Association (APA) recommended m-ECT use for cases with a periodically repetitive course that respond to ECT, pharmacotherapy intolerance, failure in preventing early flare ups and cases with m-ECT compatibility. It was stated in some publications that, apart from the cases depicted by the APA, m-ECT could also be applied in cases with a history of over 7 hospitalizations, in cases without response to treatment with minimally five psychotropic agents and in cases with a history of SIB by over-dose medication (Bilgi et al. 2010). In both of the cases presented here, m-ECT was conducted upon flare up of the symptoms after the 7th ECT session by also taking into consideration the cases reported in the relevant literature.

Agitation can present in diverse psychiatric disorders such as schizophrenia, delusional disorder, psychotic disorder, not otherwise specified psychotic disorder, bipolar disorder,

major depressive disorder, anxiety disorder, acute stress reaction, post-traumatic stress disorder, anti-social paranoid personality disorder, ASD, intellectual disability, attention deficient hyperactivity disorder, conduct disorder, delirium, dementia, intoxication/deprivation due to psychoactive substance use and akathisia (Yıldız 2003). The first aim in approaching agitation is to calm down rather than anesthetizing the patient. The four different approaches used in treating aggressive patients include environmental organisation, soothing, somatic restriction or isolation and pharmacological therapy. Guidelines recommend first step interventions of reducing aggression before using medication (Bilici et al. 2013). Antipsychotic drugs frequently used for the management of aggression have the Food and Drug Administration (FDA) warning for using one box, regarding the high mortality risk (Glass et al. 2017). Most patients with ASD diagnosis having symptoms of agitation and aggressive behavior may not respond to pharmacotherapy.

In the case of a 19-year old autistic patient with major depressive disorder, considerable regression was noted after receiving ECT in complaints like hitting his head against the wall, biting himself, swinging and taking posture (Wachtel et al. 2009). Significant reduction in SIB of an 11-year old child with bipolar affective disorder and ASD diagnoses was also reported after ECT (Wachtel et al. 2018). However, when ECT was terminated, recurrence of SIB and aggression was observed in many cases (Haq and Ghaziuddin 2014). In case 2 presented here, the prescribed medication was not sufficient to reverse permanently the display of behaviours accompanied by similar symptoms, and, ECT was considered as an alternative option that gave positive results.

In the case of an 18-year old male patient with congenital sensorineural deafness, low-level intellectual disability, ASD and catatonia, whose lack of appetite had persisted and aggression worsened after antipsychotic drug therapy for 5 years, remission in psychomotor retardation, stereotypic behaviors and catatonia were observed after twelve ECT sessions. Considerable healing was also noted after the sixth ECT session in the case of a 16-year old patient with SIB and moderate level of intellectual disability (Ghaziuddin et al. 2010). Haq and Ghaziuddin (2014) reported the effectiveness of m-ECT's on distinct agitation symptoms of two ASD patients 15 and 16 years of age. Regression in SIB and aggression after the 15th ECT session was observed in an 8-year old male with ASD, intellectual disability and a 5-year history of punching his head, hitting his head against his knees and shoulders (Wachtel et al. 2009). Remission after ECT was reported in a 23 year old with a history of 9 hospital admissions in the previous 3 months for SIBs, hair pulling and stereotypic movements and in the SIBs of a 21-year old with ASD, intellectual disability, hitting his head against the wall, aggressiveness and tendency to smash the glazing (Sajith et al. (2017).

Comorbidity of catatonia in ASD is mostly included in case reports in the literature and its prevalence in ASD patients above the age of 15 is declared to be 17% (Wing ve Shah 2006). Catatonic symptoms in individuals with ASD diagnosis mostly include extremely high level of psychomotor agitation, extreme slowness at targeted activities, prolonged immobility during a movement and difficulty in starting a movement appear at the forefront (Hare and Malone 2004, Breen and Hare 2017). Behaviours such as repetitive talks and actions, echolalia, avoiding response when addressed, grimacing, sudden interruption of movement and rigidity are accepted as indicants of catatonia or as catatonia-like symptoms in ASD (Mazzone et al. 2014, Dhosssche et al. 2006). Treatment of catatonia comorbidity in ASD with benzodiazepines and/or ECT are known to give positive results. Successful results were obtained after using bilateral ECT and lorazepam on a 14-year old male patient with ASD, mild intellectual disability and catatonia like symptoms such as stereotypic repetitive behaviours, posturing, grimacing, selective mutism and echolalia (Wachtel et al. 2008). Considering the stereotypic swinging, posturing, stupor and agitation observed in case 1 can be said to represent catatonia like symptoms which were significantly reduced by m-ECT.

Literature reports cited above and the two cases reported here evince that ECT is used effectively for psychotic symptoms, catatonia and mood disorders in children and adolescents. Although a clear consensus decision has not been made on the use of ECT in children and adolescents, it has been reported in the literature that using ECT at early stages of the cases can be beneficial for the course of the disorder (Wachtel et al. 2010).

Reports in the literature on ECT use in ASD predominantly concern effects on the catatonia like symptoms whereas ECT application on cases with agitation are relatively rare. Further reports ECT use for agitation in ASD will provide the information needed on the treatment procedures. To the best of our knowledge, our cases are one of the limited case series contributing to the literature on the use of m-ECT for agitation in ASD. Evaluating the BPRS results of our patients indicated the significant reduction after ECT in the scores on the agitation subscale representing the increased emotional tone, aggressiveness and reactivity in ASD. Intervention with ECT was attempted on our patients who did not respond to treatment with multiple medications. ECT was well tolerated without the development of any complications. Our patients underwent m-ECT treatment while continuing with their respective pharmacotherapies. This causes limitation in evaluating the specific effectiveness of medication and ECT separately.

Adverse side effects of agitation, caused especially by the therapeutic use of antipsychotic agents, are likely to occur and give rise to the argument whether aggression is indigenous to the disorder or the side effect of therapeutic drugs. Although positive results were obtained with m-ECT, remission of agitation in the long term and the longitudinal treatment results with m-ECT need to be investigated. Further case reports in this respect should contribute the knowledge on the use of m-ECT in agitation. M-ECT may be a good treatment option for ASD cases with aggression and comorbid catatonia. The purpose of this presentation is to increase the awareness on the availability of m-ECT as a potential treatment option for agitation and aggression in ASD resistant to pharmacotherapy, especially at the early stage of treatment.

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