Case Report

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Kleptomania and other Disinhibition Linked with Pseudobulbar Affect Resulting from TBI; Treatment Remission from Dextromethorphan/Quinidine 39 Years Post-Injury

Isabel Snee*, Catherine A Mazzola and Dr Jonathan Fellus

The University of Notre Dame, Notre Dame, Indiana, United States of America

***Corresponding authors:**
Isabel Snee

The University of Notre Dame, Notre Dame, Indiana, United States of America

isnee@nd.edu

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Abstract

A fifty-one-year-old man sustained a severe Traumatic Brain Injury (TBI) at the age of nine. He suffered from a twelve-day coma, biparietal fractures, underlying contusions, hypothalamic damage leading to growth hormone deficiency, growth retardation, and impulsive eating-induced obesity. Magnetic Resonance Imaging (MRI) thirty years post injury revealed encephalomalacic changes in left frontal, temporal, and parietal lobes with mild left ventricular dilatation. Soon after injury, he developed a compulsion to steal, originally only food but eventually extending to other unneeded items, even though he did not lack access to financial resources and support. Ten years after the injury, the victim was arrested for shoplifting and kleptomania for food or other items (sometimes in duplicate) and had developed Pseudo Bulbar Affect (PBA) in the form of mildly excessive/inappropriate laughter. After his psychiatrist died, he came under neurological care thirty-nine years post injury, and was treated with DM/Q (Nuedexta® 20 mg/10 mg), reaching a maintenance dosage of one capsule twice a day at week two. Reduced impulsivity and inappropriate laughter were seen by two weeks with complete remission after four weeks.

Keywords: Traumatic brain injury; kleptomania; Compulsive/Disinhibited behavior; Dextromethorphan/Quinidine; Pseudobulbar affect

Abbreviations: DXM/: Dextromethorphan/quinidine; FDA: Food a nd Drug Administration; M RI: Magnetic Resonance Im aging; PBA: Pseudo-Bulbar Affect; PTSD: Post-Traumatic Stress Disorder; TBI: Traumatic Brain Injury

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Introduction

There are an estimated annual 2.53 million deaths, hospitalizations, and emergency department visits due to Traumatic Brain Injury (TBI) related incidents, with approximately 837,000 occurring among children [1]. Within this group, most TBIs result from motor vehicle and traffic collisions. Due to the diverse, unpredictable, cognitive and developmental sequelae, clinicians have nicknamed TBI the “silent epidemic” [2]. Thus, accurately identifying TBI’s spectrum of disabilities remains essential to optimizing quality of life; yet clinical overlap with other conditions poses a significant challenge [3]. Given that an estimated 40%-62% of TBI patients suffer from depression its symptoms- reduced cognitive functioning anxiety and/or aggression decreased functional ability disturbances in appetite and weight change and disinhibited behavior may be confused with other, less common conditions and thus treated incorrectly [4-9]. For instance, inappropriate emotional displays caused by Pseudo-Bulbar Affect (PBA) a syndrome sometimes more accurately described as “affective incontinence” afflict those with TBI over 50%.

Understandably, such mood-incongruent affect is often confused with depression or other mood disorders such as Post-Traumatic Stress Disorder (PTSD) and bipolar disorder, and thus either goes misdiagnosed or undiagnosed. Since the only Food and Drug Administration’s (FDA) approved drug to treat PBA was not approved until 201012 and made commercially available until 2011, many patients suffering from this disorder were undertreated and suffered for decades with sub-optimal pharmacological options. We report an illustrative case of an individual who was successfully treated for PBA and disinhibited or impulsive behavior with Nuedexta® thirty-nine years after his

accident [10].

Case Report

A fifty-one-year-old male suffered a severe traumatic brain injury at nine years old when struck by an automobile after sledding into the road. At the time of the accident, he remained comatose for 12 days. His neuroimaging at the time revealed biparietal fractures, underlying contusions mostly of the left hemisphere, and deep hypothalamic and pituitary areas [11]. He initially suffered from respiratory arrest and had resultant hypothalamic damage. Associated neurological damage stunted his physical growth and neurophysiological development. Diagnosed with growth hormone deficiency, growth retardation, as well as hypothalamic-induced binge eating disorder, the patient reached a peak height of 5'3" and a weight of 487 lbs, but after undergoing a gastric stapling procedure, stabilized to 210 lbs. Magnetic Resonance Imaging (MRI) thirty years after injury showed prominent encephalomalacic changes in left frontal, temporal, and parietal lobes as well as mild left ventricular dilatation.

Concerning his behavioral changes, the patient began stealing food shortly after his TBI. Soon, he impulsively acquired other items, even though they went unused and he had sufficient funds to purchase these materials himself. His tendencies worsened, and ten years after injury he was arrested for shoplifting of food and other items, though charges were dismissed on medical grounds. Despite these incidents, he repeatedly over-purchased facial tissues and paper towels and continued to hoard food. Some thirty years after his TBI, while living in a supervised group home, he started compulsively withdrawing credit card cash advances only to redeposit the money in savings for the enjoyment of watching money "fall into the tray and watch his account grow." This behavior led to an excess of $55,000 in cash-advance and interest fees in addition to untallied expenditures on almost-daily taxi rides to and from the bank.

Upon his current physician's evaluation, the subject was diagnosed with PBA, as evidenced by his mildly excessive and inappropriate laughter. Moreover, he demonstrated cognitive slowing, with a documented intelligence quotient of 89, worked part-time, attended a day program, and walked with mildly abnormal and antalgic gait. A chart review for drugs aimed at controlling his impulsivity revealed his failure of over twelve different medications- most notably sibutramine, lamotrigine, topiramate, phenobarbital, valproic acid, carbamazepine, phenytoin, acetazolamide, pemoline, and fluvoxamine.

Based on the FDA-approved indication of DM/Q (Nuedexta® 20 mg/10 mg) in treating PBA, as well as the clinician's observation of control of post-TBI impulsive shopping behavior pharmacotherapy was initiated, reaching maintenance dosing of one capsule twice daily at week two. There was a noticeable decrease in impulsive behavior after two weeks and a complete remission after four weeks. There remained, however, a suggested remnant of impulsive tendency as he was reported to continually be overgenerous when paying for others' meals at restaurant outings [12].

Discussion

Traumatic brain injuries continue to destroy millions of lives, causing most victims- largely young children between the ages of 0-141 to suffer from severe physical and neurological damage. The secondary effect on these victims' families is literally incalculable. In addition to the immediate and concerning sequelae, these victims tend to have lingering cognitive and developmental issues that may not be initially attributed to their apparent injuries, causing many to not receive accurate or adequate treatment for their disorders. Failure to correctly diagnose and treat these disorders leads to chronic neurological suffering, affecting their daily activities, abilities, and quality of life.

However, correctly assessing, diagnosing, and treating these disorders remains challenging due to the substantial clinical overlap between TBI symptoms and other common conditions. Attempted treatment of cognitive, mood, and behavioral impairments across such conditions as dementia, depression, PTSD, bipolar disorder, and anxiety may sometimes improve symptomatology. But such efforts may also interfere with proper treatment for less recognized conditions, such as PBA, which shares similar phenomenology. The essence of PBA is the clinical identification of mood-incongruent affect. To screen for these episodes, the author has developed the probing question, "Do you find that you spend time and/or energy trying to keep your emotions in 'check'?" To distinguish PBA from depression or mania and other disorders with overlapping symptoms, the following diagnostic criteria must be considered: involuntary, unpredictable, exaggerated, often stereotyped, and inappropriate episodes that occur suddenly and with disproportionate or little provocation [13,14].

In addition to being undiagnosed or misdiagnosed as this case illustrates, an FDA-approved drug for this condition did not exist on the market until 2011, leading to a trial and error, off-label approach. While the pathophysiology of PBA remains to be completely understood, some theories propose that PBA may occur from damage to neural pathways that regulate the brain's emotional responses. Such connections between the frontal lobes and the cerebellum regulate the final output pathway to the brainstem's control of facial affective motor displays [15]. Since the proposed neurotransmitters involved in PBA, serotonin and glutamate, are also therapeutic targets for depression disorders, clinicians have prescribed drugs that modulate and attempt to normalize neuroreceptor functioning in the brain [16,17]. These types of treatments include Tri-Cyclic Antidepressants (TCAs), Selective Serotonin Reuptake Inhibitors (SSRIs), among other medications, many of which have unfavorable side-effect profiles compared to DXM

Regarding this case study, no FDA-approved drug had been available to effectively treat his PBA and disinhibited behavior for almost four decades after the initial onset of these symptoms. To control these symptoms, the patient had been placed on over twelve different medications, yet none succeeded in controlling his neurological issues. Without a proper treatment plan to alleviate these long-term symptoms caused by his TBI, the patient and his loved ones were forced to live with his compulsive/disinhibited tendencies, such as hoarding and stealing items as well as incurring a massive debt from withdrawing and depositing his funds in an unhealthy manner. In addition, he was rendered unable to control his excessive laughing episodes precipitated by PBA.

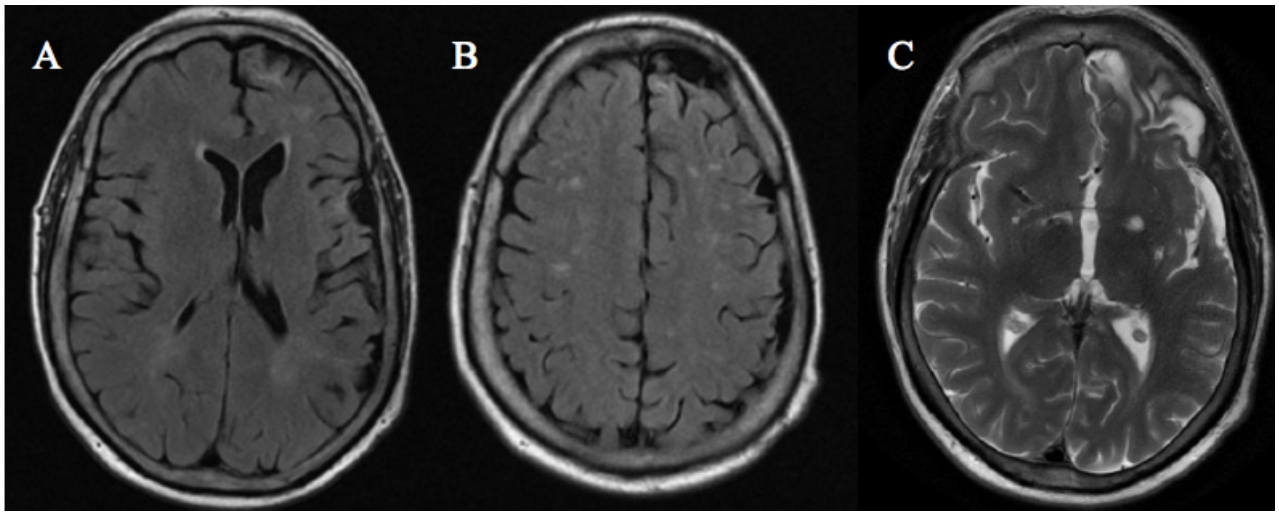

Figure 1 MRI imaging of patient done thirty years post injury. Panels A and C demonstrate significant left frontal atrophy. Panel B shows evidence of sub-cortical white matter residual lesions resultant from the TBI. In combination, these three panels represent the diffuse atrophy of the brain via encephalomalacic changes in left frontal, temporal, and parietal lobes as well as mild left ventricular dilatation.

By acting as an uncompetitive antagonist of the NMDA-sensitive ionotropic glutamate receptors and as a sigma-1 receptor agonist, DM/Q's mechanism of action more closely aligns with the most widely acknowledged pathophysiology of PBA [18,19]. While adverse effects may occur, such as falls, weakness, nausea, headaches, dizziness, diarrhea, and urinary tract infection this individual experienced none of these, and has remained on maintenance therapy for more than six years [20]. The remarkable treatment efficacy achieved within two weeks after nearly four decades of such consequentially dysfunctional behavior is further confirmed by his repeated recidivism when the drug was inadvertently discontinued. With each reintroduction of DM/Q, he rapidly gained control of his impulsive desires and urges to hoard, steal food or other items, withdraw and deposit money irrationally, participate in binge eating, or display inappropriate or unprovoked laughing episodes. However, had the patient been accurately diagnosed earlier or had access to the correct medications, these symptoms would never have perpetuated such psycho-social disruption **(Figure 1).**

Conclusion

From this case study, it becomes apparent that when one type of disinhibited behavior is observed or identified, it is clinically fruitful to probe for similar or associated impulsive behaviors. By recognizing the presence of PBA in addition to the patient's compulsions, the clinician effectively treated and stabilized most persistent issues. Thus, we recommend that when evaluating those for brain injury, clinicians should screen for PBA, and if detected, should perform further testing for other disinhibited behaviors. While these behaviors may vary from patient to patient, some may include shopping, binge-eating, or even suicidal ideation. Accordingly, this case study emphasizes the need to properly diagnose underlying conditions that may arise from TBIs, the proper treatment of these ancillary conditions, and the affirmation of the efficacy of drugs on the market to alleviate compulsive behaviors and PBA symptoms.

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