

Case Report

A Case Report of Autoneucleation and Psychosis

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Abstract

Autoenucleation is considered the most catastrophic of self-inflicted eye injuries. This is a case of a patient with unspecified psychosis and a history of self-enucleation of one eye. Autoenucleation is considered major self-mutilation [MSM] and a rare but devastating complication of severe mental illness. Acute psychosis, in particular first-episode schizophrenia, appears to be the primary cause of MSM. There are no set guidelines on how to prevent MSM, conclusive evidence on risk for future MSM, or how to prevent future MSM.

Keywords

- Autoneucleation
- Self-enucleation
- Major self-mutilation
- First episode psychosis
- Psychosis; Schizophrenia

INTRODUCTION

Self-mutilation, is described as the purposeful destruction or alteration of body tissue without conscious suicidal intent, and presents in a range of psychiatric disorders [1]. This is a case of a patient with unspecified psychosis and a history of self-enucleation of one eye. Autoenucleation is considered major self-mutilation (MSM) and a rare but devastating complication of severe mental illness [2]. This case report is consistent with many cases in the medical and psychiatric literature, which demonstrate a psychotic illness underlying acts of autoenucleation [3]. Acute psychosis, in particular first-episode schizophrenia, appears to be the primary cause of MSM [2]. Self-inflicted eye injuries among psychiatric patients are rare but important group of ophthalmic conditions that require coordination between different specialties to ensure optimum care [1].

BACKGROUND

There have been various psychiatric theories that try to explain the pathogenesis of self-inflicted behavior. The most notable ones include religious and sexual ideation, symbolism, guilt, and displacement [4]. Biological theories include disorders of serotonergic, dopaminergic, and opiate neurotransmitters [4]. Minor self-mutilation is common and does not often cause significant disability, and may even be part of certain cultural practices [5]. In contrast, MSM is rare, usually seen in people with serious mental illness and often results in permanent loss of an organ or its function [5,6]. The 3 main forms of MSM are ocular, genital, and limb mutilation [2]. Autoenucleation is considered the most catastrophic of self-inflicted eye injuries [2]. MSM is predominantly seen in young-to-early middle-aged men [4]. It was first described in 1846 by Bergman and subsequently termed "Oedipism" by Blonel 1906 [6]. This term refers to the character Oedipus in Sophocles' play Oedipus Rex, who autoenucleated both eyes upon discovering that he had unknowingly murdered his father and married his mother [2].

CASE REPORT DESCRIPTION

Mr. T is a 33 year-old Bangladeshi man, single, domiciled with his older brother and sister, Christian, with a past psychiatric history of psychosis, most likely suggestive of schizophrenia, left ocular prosthesis status post left eye autoenucleation in 2008 while experiencing command auditory hallucinations, one inpatient admission in 2008 after the autoenucleation, no prior suicide attempts or history of violent behavior towards others was brought in by his older siblings to the Psychiatric Emergency Department (ED) for complaints of not eating, not talking, bizarre behavior, and being totally withdrawn from his family and daily activities for 7 days.

On initial examination of the patient in the Psychiatric ED, he displayed negativism, mutism, catatonic posturing, would stare for prolonged periods of time and not verbally communicate with health care providers who evaluated him, would often smile inappropriately, and when prompted to speak he would either smile or ignore the interviewer. A history could not be elicited from him therefore it was obtained from his older siblings. The patient's siblings expressed their concern about his behavior because his symptoms were nearly identical to when he autoenucleated his left eye in 2008. Mr. T's older siblings indicated in 2008 a gradual deterioration was observed by family members prior to his autoenucleation, and were alarmed with the similarity of his current presentation in comparison to when he autoenucleated his left eye, and were worried he would engage in further self-injurious behavior. Mr. T emigrated from Bangladesh to New York 1 month prior to this Psychiatric ED visit to live with his older siblings. The patient's siblings reported that he was until recently taking Clozapine 25mg PO daily and Procyclidine 5mg PO daily that had been prescribed by an outpatient psychiatrist from Bangladesh but were not sure if he was continuing to take his medications since moving to the United States. The patient's siblings reported that beyond patient's left ocular prosthesis,

he had no past medical history. Mr. T's siblings reported that he had expressed his excitement to move in with them and on his arrival was at baseline functioning, not displaying negative symptoms, and communicating with them verbally. The patient's siblings remarked that he had done well on Clozapine based on communications they had with him prior to his move to the United States.

The patient's siblings reported that for the past 7 days prior to coming to the Psychiatric ED, he abruptly stopped attending to his activities of daily living, was lying down on the floor, acting bizarre, not eating, and being totally withdrawn. In addition, during this time he was mainly non-verbal, but would intermittently blurt out bizarre statements such as "my words are being twisted, my yes means a no, and my no means a yes!" and "the US Army made me say it!" In addition to the patient's bizarre behavior, his siblings noticed that since moving in with them he had increased his consumption of beer from 1 beer/day to 3-4 beers/day. The patient's siblings indicated that his behavior did not come across as if he were intoxicated, but caused them some concern.

While in the Psychiatric ED, diagnostic work up included Brain CT without contrast, CBC, basic metabolic panel, blood alcohol level, EKG, urinalysis and urine toxicology. The patient did not provide a urine sample and a result urinalysis and urine toxicology could not be performed. As per the patient's siblings, he has no history of illicit drug use. Table 1 below summarizes the key findings:

While in the Psychiatric ED, he was placed on 1:1 observation to reduce the risk for further self-injurious behavior and started on Haldol 5mg PO BID for psychosis and Benztropine 1mg PO BID for extra pyramidal symptoms.

The patient was admitted to the inpatient psychiatric unit and continued to exhibit similar behavior as was seen in the Psychiatric ED. On admission to inpatient psychiatric unit he refused physical examination and diagnostic work up included syphilis IgG with reflex RPR/TPPA, TSH, CBC, basic metabolic panel, lipid panel, hepatic function panel, and Quantiferon TB. Table 2 summarizes the significant findings:

On hospital day 1 a staff member who spoke Bengali attempted to interview the patient, but the patient would not respond verbally. On hospital day 2, Mr. T was offered interpreter services over the telephone and refused to participate in the interview. The patient's siblings on this day came in for a family meeting and indicated that he refused interpreter services because of fear that the interpreter was from his home country. The patient's siblings reported that when Mr. T was a teenager he was involved in a dispute with a neighbor regarding land property, but also felt discriminated by the neighbor as it was brought up during this argument that the patient is Christian and does not practice the predominant faith of Bangladesh. The siblings reported that since this incident the patient became wary of speaking to people he was not familiar with from his native country to avoid having another similar experience.

The patient was selectively mute, and would intermittently speak short sentences which did not make sense. In light of Mr. T not being able to provide a meaningful history, further collateral

information was obtained from the patient's siblings, indicating that the patient had a normal developmental history, was in regular education as a child, learned some English, and grew up in a tight knit community. In 2002 prior to his first psychotic break he met a female cousin who lived in India and believed her to be an angel. After meeting this cousin, the patient became obsessed with the idea of going to India. The patient's siblings reported that Mr. T would also talk about his "fantasy woman" and her purity. The patient's siblings stated that Mr. T also commented on the purity of the Russian people and that he perceived the international tennis star, Anna Kournikova, as his ideal "fantasy woman". As per the patient's siblings, he had never been sexually active and to their knowledge the patient has no history of being the victim or perpetrator of physical or sexual abuse.

In 2002 Mr. T had his first psychotic break. At this time the patient had a similar presentation as his current Psychiatric ED visit which self-resolved in 4 days. However, 1 month later he stopped speaking for 2 years. He endorsed that he did not want to speak because he felt his words were twisted and was religiously preoccupied where he perceived that those of Muslim faith lacked principle and discipline. The patient's siblings indicated that initially they thought the patient's behavior was due to having a similar encounter with someone as he did with his neighbor when he was a teenager. However, it became more clear to the family that some sort of psychotic process was going on and resulted in

Diagnostic Studies Performed in the Psychiatric Emergency Department	Result
Brain CT without contrast	No acute intracranial abnormality, left ocular prosthesis (see Figure 1)
CBC	WBC $14.5^{10^9}/L$ (elevated), MCV 87.3 (mildly elevated), Neut % 77.4 (elevated), Lymp % 12.4 (low), Neut # 11.2 [elevated], Mono % 9.5 (normal), Mono # 1.4 (mildly elevated)
Basic Metabolic Panel	Glucose 114mg/dL (mildly elevated)
Blood Alcohol Level	<3 mg/dL
Urinalysis and Urine Toxicology	Urine sample not provided
EKG	Normal sinus rhythm

Diagnostic Studies Performed in Inpatient Unit	Results
Syphilis IgG with reflex to RPR/TPPA	Non-reactive
TSH	2.234 uIU/mL [normal]
CBC	WBC $11.8^{10^9}/L$ [elevated, trending down]
Basic Metabolic Panel	Within normal limits
Lipid Panel	Chol 202 mg/dL (slightly elevated), LDL Calc 145mg/dL (elevated)
Hepatic Function Panel (AST, ALT, Alk P, T Bil, D Bil, TProt, Alb)	Within normal limits
Quantiferon TB	Negative

the patient not being able to finish college. During the 2 years the patient did not speak, he would have episodes of rage and anger, breaking glasses, windows, and mirrors, as well as engage in self-injurious behavior by burning his fingertips with matches.

In early 2008 the patient moved to Dhaka to work in a factory. As per the patient's siblings, this was short lived as he could not cope with living on his own and managing the stresses of working in a factory. The patient moved back home, and few months thereafter began his second psychotic break where he pulled out 2 of his teeth. Shortly after this incident the patient kept insisting that his mother bring him to India, and although a trip there was planned, he was frustrated at the delay of the travel arrangements, and subsequently autoenucleated his left eye using his finger. The patient's autoenucleation was not witnessed but his mother and fraternal twin brother were the first to find him after this incident MSM. Neither the patient's fraternal twin brother nor any other family member had a past psychiatric history. The patient was admitted to a psychiatric inpatient unit in Bangladesh, the details of which is not known and is unclear what medications he was treated with during this hospitalization. The patient was brought to see a psychiatrist in Dhaka who managed him with Oxcarbazepine 300mg daily, Clozapine 100mg daily, and began a course of electroconvulsive therapy. The patient's older siblings endorsed that his level of functioning progressively improved, noting that by 2014 he was more engaged with various family members.

On hospital day 3 he was started on Lorazepam 1mg PO BID to treat his catatonia and mutism and Haldol was increased to 10mg PO BID for psychosis. The treatment team wanted to restart the patient on Clozapine but as he was an undocumented immigrant and did not have a social security number it took a few days for the Clozapine registry to generate one for him for the purpose of providing this patient with access to Clozapine. During this interim time an echocardiogram was obtained to rule out underlying cardiac problems before resuming Clozapine, this diagnostic study was found to be normal. On hospital day 7, he was started on Clozapine 25mg PO daily. At this time Haldol was reduced to 5mg PO daily and Benztrapine to 0.5mg PO BID.

The patient showed mild improvement but remained for the most part silent, with the exception of blurting out a few words or short sentence when interviewed. In the initial part of Mr. T's inpatient hospitalization he was non-verbal, isolated to self but would interact with his family when they visited him, often smiling inappropriately, but was adherent with medications, did not act out, or engage in self-injurious behavior. This gradually improved and Mr. T was discharged on hospital day 40. At the time of discharge, he was provided with prescriptions for Clozapine 275mg PO daily, Haldol 5mg PO daily, Benztrapine 0.5mg PO BID. He was referred for outpatient psychiatric follow up and weekly CBC lab checks (Figure 1).

DISCUSSION

This case shares many similarities to other reported cases of autoenucleation. Most reported cases have described patients diagnosed with a psychotic illness, most commonly schizophrenia [3]. Autoenucleation resulting from substance-induced psychosis, bipolar mania, obsessive-compulsive "neuroses", posttraumatic



Figure 1

stress disorder, and major depressive disorder have also been reported. Cases of autoenucleation have also been reported with medical conditions, such as neurosyphilis, Lesch-Nyhan syndrome, Down Syndrome, and structural brain lesions [5,7]. The psychiatric literature on autoenucleation indicates that it is commonly associated with religious and sexual delusions [4,8-10]. These patients often refer to concepts of sin, evil, guilt, and atonement as their motives for self-harm. However, it is not known when in the course of psychotic illness MSM is most likely to occur [3,5,7].

Large et. al, reviewed published case histories to estimate what proportion of the MSM that is associated with schizophrenia spectrum psychosis occurs in the first episode of psychosis (FEP) [2]. They located 189 cases of patients who had removed an eye, testicle, or had severed their penis or limb. A psychotic illness was diagnosed in 143 of 180 (79.4%) cases of MSM in which a specific psychiatric diagnosis was mentioned. Of these 119 [83.2%] were diagnosed with a schizophrenia spectrum psychosis. Their findings supported the hypothesis that the risk of MSM is significantly greater in FEP compared with subsequent episodes of psychosis. Although psychosis, particularly first-episode schizophrenia, seemed to meet epidemiological criteria for causation of MSM, the reason for the decline in MSM after a period of initial treatment is uncertain [2].

In comparison, other medical literature suggests that autoenucleation patients are at greater risk for further self-harm. Witherspoon et. al, reported that patients who had enucleated one eye are at an increased risk of enucleating the other eye, even under the care of hospital staff [8]. Large et al, described that threatened ocular mutilation may occur in a hospital setting, and the case histories suggest that one to one nursing is not always sufficient to prevent enucleation [2]. The patient described in this case report engaged in self-injurious behavior during his second psychotic episode by extracting two teeth in early 2008 and 2 months later Mr. T autoenucleated his left eye in 2008. During his current inpatient admission there was concern that Mr. T might engage in further MSM. Fortunately, there was no adverse event during his current admission and Mr. T was able to communicate to the treatment team and family feeling better with Clozapine. Thus it is unclear whether patients with MSM

pose further danger to themselves after they have engaged in self-injurious behavior. However, these reports do emphasize the need for aggressive treatment of the underlying illness and close observation of autoenucleation patients to prevent further self-harm [3]. The patient in this case report was kept on constant observation throughout his hospitalization due to the severity of his MSM in his prior psychotic break and to ensure he would not engage in further self-injurious behavior.

CONCLUSION

There are no set guidelines on how to prevent MSM, conclusive evidence on risk for future MSM, or how to prevent future MSM. Research about MSM requires new approaches because case reports are subject to publication bias and valid inferences cannot be made [3]. Part of the solution to this may include having state health agencies compile data about MSM events [3]. Alternatively, an electronic registry of rare psychiatric cases could be developed via clinician network [3]. Although MSM is extremely uncommon, earlier treatment of psychotic illness may reduce the incidence of MSM. The risk of serious ocular self-mutilation is highest in first episode psychosis. Very rare events such as self-enucleation cannot be reliably predicted. There is no conclusive evidence that the risk of MSM increases over time if treatment is delayed. However, there is convincing argument that earlier intervention in psychosis may reduce the chance of MSM because adequate antipsychotic treatment appears to be protective.

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