

# A Case of Klingsor Syndrome: When There is no Longer Psychosis

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## ABSTRACT

The following case report describes an act of genital self mutilation. An employed, unmarried male suffering from schizophrenia paranoid type, autocastrated his genitalia during a period of illness when his psychotic symptoms were absent. Sufficient attention may not have been paid to his depressive symptomatology which may be primary as a core feature or secondary, in what can be called post-psychotic depression. The vulnerability of committing such an act increases when the person appears to be symptom-free and regaining insight. After a review of the available literature, it is considered that this case best fits the description for Klingsor Syndrome.

## INTRODUCTION

Self mutilation has been defined as the deliberate destruction or alteration of body tissue without conscious suicidal intent (1). It has been performed by individuals of all races, religions and cultures. The first report in English medical literature of genital self mutilation (GSM) was in 1901 by Strock (2). GSM may involve injuring, or even partial or total removal of the external genitalia, and is mentioned in Greek and Roman mythology (3, 4). Reported cases of GSM are rare. Stunnel et al. found only 122 reported cases and that most occur during psychosis (5). The associated psychiatric illnesses differ across gender. In females, GSM is mostly associated with personality disorders (predominantly Borderline type), whereas in males, psychosis is present in up to 80% of cases (2). Aboseif

et al. estimated that only 65% of patients had psychotic illness and that 31% of them made repeated attempts of GSM (6). Others concluded that psychosis was not present in one-third of cases (7). Greilsheimer and Groves identified three general patient groupings: psychotic individuals, nonpsychotic individuals with significant personality disorders, and individuals influenced by sociocultural factors and religious beliefs (8). Religious delusions, themes of guilt and sexual conflict, a history of depression with past suicide attempts and having been abused in childhood are associated with increased risk for committing GSM (4, 9). The causes, antecedents and associations for GSM are multidimensional (10). These range from transsexuals trying to reassign their gender on their own, secondary to alcohol or drug abuse like amphetamine, suicide attempts, etc. (11-13). Waugh et al. suggested that GSM most commonly occurs in men with chronic delusions or with guilt for sexual wrongdoing (14).

Self-mutilation of the external genitals in psychiatric patients is also known as Klingsor Syndrome which includes self-inflicted castration because of religious delusions. Schweitzer proposed that the syndrome should also include cases which involve genital self mutilation associated with all delusional syndromes (4). The name "Klingsor" was based on a fictitious character in Wagner's opera where Klingsor was a magician who castrated himself in an unsuccessful attempt to gain acceptance from the Knights of the Grail.

## CASE VIGNETTE

S.B., a 31-year-old Hindu, unmarried male residing in central Kolkata, India, attended the psychiatry outpatient department (OPD), and was brought by his older brother in January, 2008 with the following chief complaints:

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1. Hearing voices for one year of an unknown female asking him to marry her.
2. Frightening male voices threatening to kill him for ten months.
3. Suspiciousness that people have been conspiring against him for ten months.

His problem had started one year before while living in a rented house with his brother and mother after the death of his father in a traffic accident. He started hearing voices of an unknown woman when nobody was around, asking that he marry her. At the beginning it was pleasant but later these experiences became distressing due to the fact that the woman was trying to excite him sexually as if she was touching his private parts. It was then he started hearing male voices threatening that he must obey the commands of the female voice otherwise they would kill him. He ran away from home once in the middle of the night and was found in a small abandoned police outpost in his locality. He was then brought to the psychiatric outpatient department and was diagnosed as suffering from schizophrenia, paranoid type. He was prescribed olanzapine 10 mg at bedtime and asked to return in four weeks. On subsequent follow up, the psychotic features improved and auditory hallucinations completely disappeared within three months. He disclosed his hatred of marriage following a broken love affair one year earlier. He started visiting a temple for an hour each day. He was regular in follow up and attended the outpatient department at two week intervals. The family members ensured regular compliance and showed empty strips of the tablet olanzapine (10 mg) on each visit. The three specialists independently agreed that this was the optimum dose with best therapeutic benefit and with least chance of producing extra-pyramidal side effects. He responded well with this regimen as evident from reduction of PANSS and BPRS scores. Unfortunately at the fourth month of his symptom-free period (seventh month of his illness) he presented to the emergency department with severe self-inflicted injuries to his scrotal sac and total amputation of the penis. It was revealed that he had castrated his genitalia with a razor and a sharp knife without applying any anesthesia. He was brought to the emergency operation theater and the revision of the penis with closure of the scrotal wound was performed by the attending urologist. Microsurgical reimplantation, the most widely practiced approach by surgeons in these cases, could not be performed as the patient was brought to the hospital over 18 hours after the incident when most of the tissues were not viable (15).

He had never required psychiatric consultation in the past. He did not show any body image disturbances anytime during his period of illness. He was able to perform his job as a clerk in the postal department. There was no history of substance abuse or any contact history. He had no relationship with other women nor had he dated women other than his ex-girlfriend at any time. The family history was non-contributory. He used to be a jovial, energetic, charming and well-to-do person who loved listening to music. He used to mix with people easily and was always very popular in his peer group because of his helping attitude. No definite traits or personality disorder were diagnosed from his premorbid personality.

During mental status examination at the time of admission with GSM, he was alert, conscious but uncooperative, eye contact was poor, speech was inaudible and detailed examination could not be done at that point. He was guarded initially, did not appear to be overtly psychotic, and his trust of the staff gradually increased. After 24 hours, rapport could be established with difficulty. Speech was relevant and coherent; affect was sad, mood irritable; no formal thought disorder and perceptual disturbances were noted. There was no suicidal intent. Tests of higher cognitive functions were satisfactory. He acknowledged he was sick but attributed his illness to an unknown factor (Grade 4 insight).

Apart from mild pallor, local examination revealed the penis was totally amputated, catheterized with oozing of blood and raised temperature. Other systemic examinations and routine laboratory tests were within normal limits. CT scan of brain showed mild ventricular dilatation.

On query why he had performed such a drastic act, the patient disclosed that he was feeling intense guilt and sadness for his past psychotic illness which had caused significant emotional and financial burden on his family members. He further expressed that he had not slept well for the past few nights and could not recall what was his state of mind just before this act. The act was preceded by feelings of hopelessness and social avoidance. The family members revealed retrospectively that for the last week he had been staying home and not going to work. The patient had told his brother that he had taken leave for a week due for his working overtime in the past.

Extensive clinical interviews did not reveal any clear psychotic processes. The Wechsler Adult Intelligence Scale (WAIS) showed full-scale IQ was 98. The BPRS and PANSS scores were borderline (40 and 55 respec-

tively) to overtly psychotic. However, the HAMD-17 score was 28, which fell in the severe depression category. The diagnosis was revised as post-psychotic depression (F20.4) as per ICD-10 criteria and other possible diagnoses such as affective disorders, substance use disorders, personality disorders and obsessive compulsive disorders were ruled out. Some schizophrenic symptoms in the form of negative symptoms and change in personal behavior, lack of interest, social withdrawal were still present in absence of delusion or hallucination which supports the diagnosis.

## DISCUSSION

Predicting GSM is exceedingly difficult. The best predictor of future behavior is probably past behavior. GSM is often a one-off act but in some cases there are examples of prior self harm behavior. With psychotic patients it can be very difficult in much the same way as suicide is more usually unpredictable in schizophrenia.

GSM is not necessarily associated with psychosis as reported in the previous case reports. In all follow up visits, our patient did not show any psychotic symptoms and possibly was able to mask his depressive symptoms for long periods until he carried out such a drastic act. He denied that the autocastration was an attempt to atone for past sins or an attempt to change his sex.

Unlike previous published reports, this case report is interesting in formulating a hypothesis that a combination of antecedent factors can lead to the act of GSM. Remaining unmarried at the age of 31 is unusual for his ethnic background (16). The patient had an unsuccessful love affair and witnessed the unexpected death of his father and could not cope with these two significant life stressors. One of the symptoms associated with GSM is the presence of religious psychotic experiences (4). Here the patient could have been trying to attain a pure form of existence by amputating his penis without any features of psychosis at the time of commission of this act, as mentioned in previous case reports (17). The increased visit to temples in comparison to his premorbid period could be an indicator of his intense guilt, a mode of confession and an attempt to attain purity. The sexual hatred probably was due to overgeneralization and other cognitive errors.

The current case of autocastration by a man recovering from a psychotic episode with possible sexual guilt, religiosity and intense hatred towards women is one of the rare antecedents of reported acts of GSM and a care-

ful search for possible post-psychotic depression is warranted in order to prevent this act of self harm. Duggal et al. offer an interesting inversion of the commonly accepted relationship between psychosis and autocastration: they speculate that psychosis can be an effect rather than a cause (18). It is essential to identify patients at risk for GSM so that the act can be prevented. Psychosis with delusions of sexual guilt is an obvious warning sign in the causation of GSM. But at the same time during the recovery from psychosis, when the patient is regaining his insight and in the phase of post-psychotic depression he is vulnerable to commit such an act. Due to the rarity of the event, however, more precise identification of individuals at risk remains difficult.

Previous case reports on autocastration have identified individuals as having significant dysfunction of ego integrity and the occurrence of such an act is more common in men (19). A few reports described initial denial by the patients of having committed such act themselves. Some patients cut or maim their genitalia by violent methods, while others performed the act very meticulously. It is sometimes difficult to assess whether it was a failed suicide or successful male self-amputation. Even a case of autophagia of the amputated penis has been published. One patient himself had attempted surgically to reconstruct a foreskin (20). The reports described the influence of media in a susceptible individual with borderline intellectual functioning to undertake this act (21). Auto-aggressive actions can be prevented by adequate pharmacotherapy and psychotherapy. In schizophrenia six possible risk factors for GSM have been mentioned (22): 1) psychotic experiences, 2) presence of personality disorder, 3) past history of GSM, 4) alcohol or drug dependence, 5) sexual guilt feeling and 6) early loss of father. Depressive symptoms complicate the course of schizophrenia in as many as 25% cases and post-psychotic depression is complicated by an overlap with the negative symptoms of schizophrenia.

The post-psychotic depression can be early (within six months of an acute episode) or late (persists more than six months after an acute episode). In early cases depression resolves more slowly than other symptoms of acute episode but in late cases depressive symptoms develop in the aftermath of an acute psychotic episode or persist for more than six months (23). The present case is compatible with the diagnosis of late post-psychotic depression which warrants antidepressant treatment, preferably a SSRI. It seems that post-psychotic depression could be one of the warning signs for committing an act of GSM.

## INFERENCES

It appears that the number of reports of genital self-mutilation is on the rise. Whether this is due to a true increase in the incidence is not known. The high rate of repeated mutilation is due to the fact that patients do not come under the scrutiny of psychiatric services. A general awareness of GSM should be promulgated among medical practitioners so that it can be prevented and treated effectively. The careful assessment during follow up and family education to recognize early warning signs could be two important interventions to prevent such dreadful acts.

This case is unique as at the time of the autocastration the psychotic features were well under control. This case raises the question of whether the act of autocastration, in the absence of clear psychotic symptoms, justifies labeling an individual as psychotic. Perhaps future reports will help to elucidate the complex relationship between the GSM and associated psychopathology.

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