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CASE REPORT

Genital self-mutilation following cannabis-induced psychosis: Klingsor syndrome – a case report

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Penile amputation is a severe injury to the penis associated with significant morbidity. It may be therapeutic, especially in patients with penile cancer, or traumatic, which could be accidental, assault, animal attack, industrial injury, self-inflicted, or circumcision-related. The most common aetiology of penile amputation in adults is self-inflicted mutilation, especially during an episode of psychosis (Klingsor syndrome). As with all trauma, initial management focuses on the assessment and resuscitation of the patient. When available, hypothermic preservation of the detached penis should be undertaken. When micro-replantation cannot be performed, older corporal reattachment techniques may be offered. We aim to report the first case of self-inflicted penoscrotal amputation following cannabis-induced psychosis in Nigeria.

Keywords: cannabis, psychosis, penile amputation, Klingsor

Background

Genital self-mutilations in psychiatric patients, also known as Klingsor syndrome, are a rare urologic trauma. Men with religious conflicts, low self-esteem, unresolved transsexual issues, and feelings of guilt are the most vulnerable.¹

Penile amputation is a severe injury to the penis associated with significant morbidity. It may be therapeutic, especially in penile cancer, or traumatic, which could be accidental, assault, animal attack, industrial injury, self-inflicted, or circumcision-related. The most common aetiology of penile amputation in adults is self-inflicted mutilation, especially during an episode of psychosis, while circumcision-related penile amputation is the main aetiology in children.^{2,3}

It is classified as either phallicide when the intention is to commit suicide or Klingsor syndrome when the patient does not intend to commit suicide. Anatomically, penile amputation can be peri-pubic, a proximal part of the penis, or at the level of the glans. Traumatic amputation of both the penis and scrotum is a rare surgical emergency. A systematic review of 80 cases from 1996 to 2007 reported only 37.5% of cases undergoing a successful replantation.

In the 1970s, an epidemic of penile amputations was reported in Thailand where women amputated their husbands' genitalia for infidelity. That case series of 18 patients remains the largest to date.⁵

As with all trauma, initial management focuses on the assessment and resuscitation of the patient. When available, hypothermic preservation of the detached penis should be undertaken.² When phallic reconstruction is required, a microsurgical forearm free flap phalloplasty may be performed to restore the patient with an acceptable cosmetic and functional phallus.² When micro-

replantation cannot be performed, older corporal reattachment techniques may be offered.²

Male genital self-mutilation from a psychiatric disorder or substance-induced psychosis has been reported; however, the exact prevalence of these conditions is unknown. Some reports state about 100 cases within the past two decades.^{6,7} Few cases of self-amputation of the penis in cannabis-induced psychosis have been reported.^{8,9} Scrotums were spared in the reported cases of cannabis-induced self-inflicted amputation, but penoscrotal amputation following assault has been reported.¹⁰

We aim to report the first case of self-inflicted penoscrotal amputation following cannabis-induced psychosis in Nigeria.

Case presentation

A 22-year-old artisan presented to our clinic 11 days after the self-inflicted amputation of his external genitalia following cannabis-induced psychosis. He was introduced to cannabis by his friends and was smoking about 1–2 improvised sticks (wrapping some quantity of cannabis in paper) almost daily for about three years. He usually had auditory and visual hallucinations with occasional penile erections following cannabis use. On the day of the incident, he smoked an improvised stick of cannabis and had a penile erection, which was cut off along with the scrotum using a kitchen knife. The amputated genitalia were thrown in the dustbin following an auditory hallucinatory command to do so.

The patient had no history of use of other hard drugs or a history suggestive of psychiatric symptoms before the commencement of cannabis smoking. The psychotic symptoms resolved after stopping cannabis use following the external genital mutilation. He had no family history of psychiatric disorders and all other history are unremarkable. He was taken to the peripheral hospital when he was noticed to be bleeding. He had haemostasis secured and resuscitation with alternate day dressing for 11 days before



Figure 1: After catheterisation

presenting to our facility. He was voiding satisfactorily through the perineal wound.

Examination showed a calm, coherent, and oriented young man. A remarkable finding was the perineal wound with penoscrotal absence. Testes were not palpable. The urethral opening was patent and adequate in the perineal wound (Figure 1). There were no other remarkable findings on examination. Baseline investigations were essentially normal. He had wound debridement, eversion of the edges of the urethral stump, and urethral catheterisation.

The patient is presently on local wound care while undergoing psychiatric evaluation and phallic reconstruction is planned. He is voiding satisfactorily via a urethral stump after catheter removal making perineal urethrostomy unnecessary. He was not placed on any antipsychotics because the psychiatric symptoms resolved after discontinuing cannabis smoking. No symptoms suggest schizophrenia. He is been followed up and rehabilitated.

Discussion

Traumatic amputation of the penis is a rare surgical emergency. A systematic review of 80 cases from 1996 to 2007 reported only 37.5% of cases undergoing a successful replantation.³

Occasionally, patients attempt self-mutilation under the influence of an ordering hallucination (Van Gogh syndrome).^{11,12} This is similar to what happened to our patient. The eponym of the Klingsor syndrome is applied to the occurrence of autocastration as a consequence of religious delusions, which has been further expanded to include all delusions associated with genital self-mutilation.¹¹⁻¹³

Three general patient groups in cases of autocastration have been identified: psychotic individuals, non-psychotic individuals with significant character pathology, and individuals influenced by socio-cultural factors and religious beliefs.^{14,15} Our patient had a

psychotic influence. Four common premorbid characteristics have been identified: delusions (particularly religious delusions), themes of guilt and sexual conflict, a history of depression (often with past suicidal attempts), and severe deprivation in childhood with major personality deviation in adult life.^{14,16} This could not be identified in our patient as the identified behavioural changes were related to cannabis use.

The principles of management following self-penile amputation are saving life, restoration of function, and management of the underlying psychiatric condition. To Following the advanced trauma life support (ATLS) protocol to save life, the options for definitive surgical management are replantation, stump-plasty, and total phallic reconstruction.

Replantation offers the best results with a majority having restoration of micturition and sexual function. Microscopic re-anastomosis of the venous sinusoids and nerves has better outcomes than macroscopic re-plantation. ^{18,19} Requirements for re-plantation are early presentation, proper preservation of the amputated organ, minimal contamination, and the availability of prerequisite skills and equipment. In addition, stabilisation of the predisposing psychiatric condition is paramount to minimise chances of repeat occurrence. Complications following replantation include urethral stricture, urethral fistula, skin necrosis, venous congestion, and diminished sexual function. ²⁰ Our patient could not benefit from replantation as the amputated genitalia were thrown into the dustbin 11 days before presentation.

In recent decades, cannabis, dried flowers and leaves of Cannabis sativa have been widely used.²¹ Cannabis exhibits many adverse effects, including impaired judgement. With heavy use, paranoia and psychosis may be expected.^{22,23} Male genital self-mutilation from a psychiatric disorder or substance-induced psychosis has been reported; however, the exact prevalence of these conditions is unknown. Some reports stated about 100 cases within the past two decades.^{6,7} Nonetheless, self-amputation of the penis in cannabis-induced psychosis has rarely been reported.⁸

Jengsuebsant et al.⁹ reported a case of cannabis-induced psychosis with painful penile erection in less than four hours leading to self-penile amputation in a 23-year-old man. This is similar to our patient who was 22 years old when he amputated his erect penis along with the scrotum following cannabis smoking. In this patient, the diagnosis of substance-induced psychotic disorder was mainly clinical as his symptoms began after cannabis use without evidence of other substance abuse, as well as from the resolution of psychotic symptoms after abstinence.²⁴

The routes of cannabis exposure are ingestion (73.9%), inhalation (22.5%), topical/parenteral/rectal (0.8%), and unknown/other (2.8%). Most of the patients were male, and two-thirds of the overall age group were intentional use.²⁵ The route of exposure in our patient was inhalation. Cannabis use is reported to increase the risk of psychosis, loss of insight, and thought disorders leading to unexpected behaviour, including patients with no previous psychiatric disorders, as seen in our patient.²⁶⁻²⁸

The severity of psychosis depends on the amount of tetrahydrocannabinol (THC).²⁹ THC, consumed by smoking cannabis, is one of the primary cannabinoids that produce psychoactive effects through the dopaminergic pathway.³⁰ We could not quantify the amount of cannabis used by our patient because the quantity wrapped in paper for smoking varied without measurement.

In an animal model, a dopamine receptor agonist increases central oxytocinergic neurotransmission and facilitates penile erection. 31-34 Therefore, cannabinoid use promoting the dopaminergic pathway might play a role in penile erection. 33 Moreover, cannabinoids block the thoracolumbar sympathetic pathway, which could result in the penis being unable to detumescence and increases the risk of priapism. 31-35 THC interacts with a cannabinoid type 1 (CB1) receptor in the central nervous system (CNS), peripheral nervous system, and vasculature. Consequently, cannabinoids might potentiate vascular effects and lead to penile erection and priapism. 32,35,36

A previous case report suggested a relationship between cannabis use alone and priapism. However, the patient smoked cannabis for the previous six months and no self-harm or psychosis occurred.³¹ Although priapism is a painful event, self-amputation is rare in a patient with normal judgement. Our patient could not be said to have priapism. Acute cannabis exposure has the following effects: CNS excitation (38.3%), CNS depression (24.4%), cardiac problems (14.6%), nausea and vomiting (9.5%), unusual/unexpected subjective sensation (strange, weird, bizarre) (3.6%), abdominal pain (2.4%), and psychosis (1.6%).²⁵

Our patient had an erection that could not fulfil the criteria for priapism, as there was no significant pain and he had penoscrotal amputation in less than four hours of the erection. We believe that the reason for self-genital amputation was cannabis-induced psychosis as the patient obeyed the auditory hallucinatory command to cut off his external genitalia.

Conclusion

Cannabis-induced psychosis is an adverse effect of cannabis, which may lead to impaired judgement and unexpected self-harm. A multidisciplinary team approach, including a primary care physician, an emergency physician urologist, and a psychiatrist is essential when dealing with a patient with cannabis-induced psychosis and a urogenital injury.

Conflict of interest

The authors declare no conflict of interest.

Ethical approval

The informed consent of the patient was obtained and the case report was conducted in compliance with the guidelines of the Helsinki Declaration on biomedical research in human subjects. Confidentiality of the patient and personal health information was maintained.

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