Major Self-mutilation in the First Episode of Psychosis

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Major self-mutilation (MSM) is a rare but catastrophic complication of severe mental illness. Most people who inflict MSM have a psychotic disorder, usually a schizophrenia spectrum psychosis. It is not known when in the course of psychotic illness, MSM is most likely to occur. In this study, the proportion of patients in first episode of psychosis (FEP) who inflicted MSM was assessed using the results of a systematic review of published case reports. Histories of patients who had removed an eye or a testicle, severed their penis, or amputated a portion of a limb and were diagnosed with a schizophrenia spectrum psychosis were included. A psychotic illness was documented in 143 of 189 cases (75.6%) of MSM, of whom 119 of 143 (83.2%) were diagnosed with a schizophrenia spectrum psychosis. The treatment status of a schizophrenia spectrum psychosis could be ascertained in 101 of the case reports, of which 54 were in the FEP (53.5%, 95% confidence interval 43.7%–63.2%). Patients who inflicted MSM in FEP exhibited similar symptoms to those who inflicted MSM later in their illness. Acute psychosis, in particular first-episode schizophrenia, appears to be the major cause of MSM. Although MSM is extremely uncommon, earlier treatment of psychotic illness may reduce the incidence of MSM.

Key words: self-mutilation/schizophrenia/first-episode psychosis

Introduction

Self-mutilation has been defined as the direct and deliberate self-destruction of a part of a person’s own body without the intention of suicide.1 Minor self-mutilation is quite common, does not usually cause significant disability, and may even be part of recognized cultural practices. In contrast, major self-mutilation (MSM) is rare, usually only occurs in association with serious mental illness and often results in permanent loss of an organ or its function.2 The 3 main forms of MSM are ocular, genital, and limb mutilation. Patients who have removed an eye or cut off a limb are almost always psychotic, as are three quarters of patients who severely injure their genitals.3

The published accounts of MSM are almost all either single-case histories or small case series, from which it is difficult to make valid causal inferences. Even quite recent publications sometimes explain MSM in terms of the patients’ reaction to passages in religious texts or unconscious sexual conflicts. The few authors who have reviewed more than a small number of cases have attributed MSM to the direct effects of psychotic illness.3–7 In a recent review of self-inflicted eye injuries, we found that almost all cases of serious self-inflicted eye injuries result from schizophrenia spectrum psychosis and half the injuries that caused permanent loss of vision occurred in the first episode of psychosis (FEP).7

Studies that have examined prior treatment at the time of another uncommon and catastrophic complication of psychosis, homicide, generally report that between 30% and 50% occur during the FEP,8 usually in response to frightening symptoms.9 These studies also provide sufficient information to demonstrate that the risk of homicide in FEP may be as much as 20 times the subsequent annual risk after treatment.10 The hypothesis for this study was that MSM is similar to homicide in psychosis and may also be more likely to occur during the FEP.

We aimed to use published case histories to estimate what proportion of the MSM that is associated with schizophrenia spectrum psychosis occurs in the FEP. Our hypothesis was that there may be a greater proportion of MSM in the FEP than would be expected by chance, assuming that the risk of MSM is equal in FEP and previously treated psychosis (PTP). We included case reports of patients who had a diagnosis of schizophrenia spectrum psychosis and who had completely removed an eye or a testicle or severed their penis or a limb. The findings were used to examine the extent to which psychosis meets epidemiological criteria for causation of MSM.
Methods

Search Terms

The [Medline], [Embase], and [Psychlit] databases were searched from 1960 to February 2008 using the terms psychosis OR schizophrenia OR mental disorder AND self mutilation OR self-enucleation OR enucleation OR self-inflicted eye injuries OR eye injuries OR oedipism OR castration OR emasculation OR orchidectomy OR penile amputation OR penile injury OR amputation OR limb amputation OR mutilation OR self-mutilation OR deliberate self-harm. The references of the located articles were hand searched for further cases.

Definition of MSM

We included published case histories of patients with a schizophrenia-related diagnosis in which prior treatment status could be ascertained and who had completely removed (1) an eye (2) or a testicle or severed (3) their penis (proximal to the glans) or (4) a limb (proximal to the hand or foot). We also collected data on cases that met our severity criteria for MSM but were not diagnosed with a schizophrenia spectrum disorder or did not provide sufficient information to establish prior treatment status. The latter 2 groups were excluded from the main analysis. Cases of patients with mutilation that did not meet our injury severity criteria or removed other body parts were not considered. Case histories of people who required an amputation or enucleation for medical purposes after self-injury and cases of fatal self-amputation were also excluded.

A narrow definition of MSM was used to ensure that all the cases were of similar severity. There was great variation in the severity of other forms of mutilation, and in some cases, the severity was difficult to assess. Moreover, many cases of limb, neck, chest, abdomen, and penetrating orbital injuries occurred in failed suicide attempts. The other forms of self-injury inflicted by the patients included in this study, in addition to their MSM, were broadly defined as any self-inflicted physical injury that did not meet our definition of MSM.

Definitions of Psychosis and Treatment Status

The diagnoses used by the authors of the case histories were accepted at face value, and no attempt was made to standardize the diagnostic criteria. Psychosis was defined as any diagnosis characterized by hallucinations, delusions, or thought disorder and included psychosis secondary to medical conditions or substance use, affective psychosis, and schizophrenia spectrum psychosis. The category of schizophrenia spectrum psychosis included schizophrenia, delusional disorder, schizoaffective disorder, schizoaffective disorder, brief psychotic disorder, or psychosis not otherwise specified (NOS). Cases diagnosed with affective psychosis or psychosis secondary to substance abuse were not included in the main analysis, even if it appeared that the patient had persisting hallucinations or delusional beliefs.

We classified a case as FEP if the case history stated that the patients had never received antipsychotic treatment or if they had only recently commenced treatment for the first time, had not had a remission from symptoms and were still in hospital. Few of the case histories provided sufficient information to use the preferred definition of the FEP, which is the period from the onset of psychotic symptoms until the patient had a remission after receiving an adequate trial of antipsychotic treatment. Hence, patients were also classified as being in the FEP if an otherwise comprehensive history did not report previous treatment or if the case history clearly stated that the diagnosis of psychosis was not made until after the injury.

Few case histories documented the dosages and duration of prior treatment. Patients were classified as having PTP if the case history reported that the patient had been treated before, if the diagnosis had been made before the injury, or if the patient was described in the history as having chronic schizophrenia. Patients who had received prior treatment were included in the PTP group even if they had not been adherent to antipsychotic medication for many years. Cases in which the treatment status was not described or could not be determined and borderline cases of patients who had received 3 or 4 weeks of antipsychotic treatment were excluded from the analysis of treatment status.

We relied on the information provided by the authors for almost all the data points, including the presence of particular symptoms. If a paper failed to mention a specific psychotic symptom, the symptom was rated as not having been present. In cases that provided little information beyond the diagnosis and previous treatment status, absent data points were left blank. Some inferences about psychomotor agitation and the patient’s indifference to the injury were made on the basis of the descriptions of the patient’s behavior.

Statistics

The mean and 95% confidence intervals (CIs) for the proportion of case histories documenting each characteristic were calculated for the whole group. The secondary hypothesis of possible clinical difference between FEP and PTP groups was examined with a chi-square test for categorical variables or a Fisher’s exact test if N was less than 5 in any cell. A 2-tailed Student’s t-test was used to compare age in FEP and PTP groups.

Female cases were excluded for penile amputation and castration. A Bonferroni correction was calculated to be 0.0024 for the 21 comparisons that were independent of treatment status. Age and current treatment with antipsychotics were not regarded as independent of treatment.
status. SPSS version 15.0 was used for all statistical analysis.

Estimation of the Incidence of MSM

An attempt to estimate the incidence of MSM and proportion of cases that were the subject of a published report was made using acquaintance chains, in a method that was loosely adapted from that described by Stanley Milgram. The authors initially contacted 8 senior clinicians in key positions in mental health, forensic psychiatry, rehabilitation, and hospital-based ophthalmic services in New South Wales (NSW), a state of Australia with a current population of just under 7 million. Each clinician was asked about their knowledge of patients with MSM and was also asked to nominate other clinicians who may know of cases. The nominated clinicians were then contacted in a sequence that continued until no new names were suggested.

Results

Results of the Searches and Case Selection

One hundred and ninety six publications were examined after the exclusion of case histories describing superficial mutilation, finger amputation, cases of MSM of the tongue, breasts, or nose and 4 histories of fatal amputation. These publications contained a total of 305 case histories of probable MSM, including 189 case histories in which there was complete amputation or removal of an organ. The case histories were then coded according to whether (1) injuries met inclusion criteria, (2) the psychiatric diagnosis, and (3) if there was an adequate account of previous treatment.

First, case reports of patients who had self-inflicted injuries that were not reported in association with a diagnosed psychotic illness were excluded. This included 39 cases of less severe eye or genital injuries and 44 cases of major genital injury. The major genital injury cases included 25 patients who had amputated one or both testicles, 13 patients who had amputated their penis, and 6 cases of complete genital amputation. Seventeen of the 44 genital mutilation cases were reported to have gender identity disorder, 6 alcohol dependence, 12 other nonpsychotic disorders, and there were 9 cases in specialist surgical journals that only reported the presence of psychiatric disorder without providing any further details. There were several cases of men from Asian backgrounds who believed that penile amputation would result in death and who were regarded by the authors to be suicidal but not necessarily psychotic. There were 2 cases of upper limb self-amputation in patients who were not diagnosed with a psychotic illness but there were no case histories of nonpsychotic patients who had performed an enucleation or a lower limb amputation. A total of 42 publications were excluded on this basis.

Second, cases of patients with psychosis whose self-inflicted injuries did not meet the inclusion criteria were excluded. These consisted of 53 case histories of penetrating, superficial, and blunt force self-inflicted injury to the eye, 19 reports of genital self-injury that fell short of amputation, 2 reports of genital mutilation by females, and 3 reports of incomplete upper limb amputation by psychotic patients. As a result, a further 38 publications were excluded.

Third, 24 case histories of patients with a nonschizophrenia spectrum psychosis who had inflicted MSM were excluded. These consisted of 5 self-enucleating patients with psychosis that were secondary to various medical conditions, 12 cases in which the diagnosis was affective psychosis, and 7 cases of psychosis reported to be secondary to substance use. Exclusion of these cases resulted in the exclusion of a further 22 publications.

Finally, we excluded case histories of 18 patients with a schizophrenia spectrum psychosis who had amputated or removed an eye, limb, or genital part but did not provide sufficient information to establish if the patient had been previously treated. These comprised 18 cases in 15 publications and included 2 patients who inflicted MSM after several weeks of hospital treatment with antipsychotic medication but who remained unwell.

One hundred and one cases of MSM from 79 publications were included. There were 42 cases that met our criteria for MSM and psychosis but did not document the treatment status or the presence of a schizophrenia spectrum psychosis and were excluded from the main analysis.

Reliability of Data Collection

M.L. and N.B. independently examined the cases for inclusion or exclusion. There were two disagreements about the inclusion of cases, 1 due to accidental double counting by one author and the second as a result of multiple publications about the same patient.

M.L. and N.B. also independently collected clinical data using spreadsheets of data points versus case histories. There were no disagreements about the injuries, the psychiatric diagnosis, demographic details, or the setting of the injury. There was 1 disagreement about the rating of previous treatment in a history that was subsequently excluded because of the uncertainty on this point. Disagreement about 5% of other data points was resolved by a further review of the cases.

Results of Examination of the Cases

We located 189 cases of patients who had removed an eye or 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 schizophobia spectrum psychosis. Treatment status could be ascertained in 101 of 119 (84.9%) schizophrenia spectrum
psychosis cases, of which 54 were classified as FEP (53.5%, 95% CI = 43.7%–63.2%).

Table 1 is a summary of the characteristics of the included and excluded cases.

The sample has a predominance of younger men, most of whom were in the FEP. Most of the cases were of genital amputation or enucleation, with comparatively few cases of limb amputation. The excluded cases were similar in characteristics to the included cases. However, cases that were excluded on the grounds of diagnosis were significantly more likely to be in FEP (17 of 19 in FEP, chi-square = 8.582, P = 0.003).

Eighty-nine of 101 cases were diagnosed with schizophrenia, 4 schizophreniform psychosis, 2 schizoaffective disorder, 2 with a brief psychotic disorder, 1 with delusional disorder, and 3 were considered to have psychosis NOS.

Eighty-seven of 101 histories made a specific mention of at least 1 psychotic symptom and 82 described the presence of a delusional belief. The most common delusions involved a false belief about the amputated organ including that the organ was evil (43%), that the organ had a special, usually threatening supernatural powers such as the ability to spread evil (28%), or that it needed to be sacrificed in order to save the patient or others (20%).

The FEP and PTP groups and the sample of excluded patients had high proportion of patients with religious delusions, disorganized thinking, and behavior, and many patients were indifferent to their injuries (Tables 1 and 2). A third of PTP patients were reported to be taking antipsychotic medication at the time of MSM.

Table 2 compares those with FEP to those with PTP. Patients in their FEP were, as expected, younger and less likely to be taking antipsychotic medication. Habitual substance use was more common in the FEP group, and more patients in the PTP group reported command hallucinations, but neither finding was significant after a Bonferroni correction. It may be that FEP patients were less able to identify voices as hallucinations.

Excluded Cases

The psychosis cases that were excluded because their eye injuries were not severe enough to meet our inclusion criteria, were those with affective psychosis, substance abuse, and in 18 cases a schizophrenia spectrum psychosis with undocumented treatment status.

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#### Table 1. Characteristics of Patients With Psychosis and Self-enucleation, Limb, or Genital Amputation

<table>
<thead>
<tr>
<th>Age (mean)</th>
<th>32.7</th>
<th>95% CI</th>
<th>34.0</th>
</tr>
</thead>
<tbody>
<tr>
<td>Frequency</td>
<td>90</td>
<td>89.1</td>
<td>90.2</td>
</tr>
<tr>
<td>Male</td>
<td>54</td>
<td>53.5</td>
<td>37.5</td>
</tr>
<tr>
<td>Enucleation</td>
<td>40</td>
<td>39.6</td>
<td>9.5</td>
</tr>
<tr>
<td>Limb amputation</td>
<td>9</td>
<td>8.9</td>
<td>6.5</td>
</tr>
<tr>
<td>Penis amputation</td>
<td>40/90</td>
<td>44.4</td>
<td>52.6</td>
</tr>
<tr>
<td>Castration</td>
<td>30/90</td>
<td>33.3</td>
<td>18.4</td>
</tr>
<tr>
<td>On antipsychotics</td>
<td>20</td>
<td>19.8</td>
<td>5.7</td>
</tr>
<tr>
<td>In hospital, jail, or care facility</td>
<td>21</td>
<td>20.8</td>
<td>5.7</td>
</tr>
<tr>
<td>Cases with more complete data</td>
<td>N = 89</td>
<td>N = 24</td>
<td></td>
</tr>
<tr>
<td>Any delusion</td>
<td>82</td>
<td>92.1</td>
<td>100</td>
</tr>
<tr>
<td>Delusion about the amputated organ</td>
<td>62</td>
<td>69.7</td>
<td>62.5</td>
</tr>
<tr>
<td>Religious delusions</td>
<td>61</td>
<td>68.5</td>
<td>62.5</td>
</tr>
<tr>
<td>Reference to a religious text</td>
<td>30</td>
<td>33.7</td>
<td>28.0</td>
</tr>
<tr>
<td>Command hallucinations to remove an organ</td>
<td>28</td>
<td>31.5</td>
<td>29.2</td>
</tr>
<tr>
<td>Visual hallucinations</td>
<td>10</td>
<td>11.2</td>
<td>10.7</td>
</tr>
<tr>
<td>Thought disorder</td>
<td>41</td>
<td>46.1</td>
<td>24.0</td>
</tr>
<tr>
<td>Behavior indicating indifference to injury</td>
<td>44</td>
<td>49.4</td>
<td>41.7</td>
</tr>
<tr>
<td>Psychomotor agitation</td>
<td>35</td>
<td>39.3</td>
<td>33.3</td>
</tr>
<tr>
<td>Habitual substance abuse</td>
<td>20</td>
<td>22.5</td>
<td>36.1</td>
</tr>
<tr>
<td>Intoxicated at the time</td>
<td>7</td>
<td>7.9</td>
<td>16.7</td>
</tr>
<tr>
<td>Previous stimulant or hallucinogen use</td>
<td>20</td>
<td>22.5</td>
<td>33.3</td>
</tr>
<tr>
<td>Violence toward others</td>
<td>10</td>
<td>11.2</td>
<td>8.0</td>
</tr>
<tr>
<td>Suicidal thoughts or acts</td>
<td>18</td>
<td>20.2</td>
<td>32.1</td>
</tr>
</tbody>
</table>

*5 cases of psychosis secondary to a medical condition, 12 cases had affective psychosis, 7 cases of psychosis secondary to substance abuse, and in 18 cases a schizophrenia spectrum psychosis with undocumented treatment status.

*Proportion in FEP based on an N of 19 cases only.
criteria are reported elsewhere. A lower proportion of the patients with schizophrenia spectrum psychosis and less serious genital or limb injuries were thought to be in FEP, but the histories were less detailed than the case histories of more severe MSM.

There were 42 cases that met inclusion criteria for MSM, but were excluded because they were not diagnosed with a schizophrenia spectrum psychosis (24 cases), because treatment status was not documented (18 cases) or for both of these reasons (5 cases). The non-schizophrenia spectrum psychosis cases included 5 cases of psychosis secondary to a medical condition (including systemic lupus erythematosus, hypothyroidism, and epilepsy), 12 cases diagnosed with an affective psychosis, and 7 cases of psychosis secondary to substance abuse. Overall, the excluded patients had symptoms that were similar to the included cases. Patients reported to have affective psychosis were just as likely to have a bizarre organ-specific delusion but were more likely to have amputated part of the genitals, were older, with a mean age of 40 years, and 9 of 12 were considered to have pathological guilt.

Table 2. Comparison of Previously Treated Psychosis and First-Episode Psychosis Patients

<table>
<thead>
<tr>
<th></th>
<th>PTP</th>
<th>FEP</th>
<th>df</th>
<th>Chi-Square</th>
<th>P</th>
</tr>
</thead>
<tbody>
<tr>
<td>Demographics and circumstances of the injury, n = 101</td>
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<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>N</td>
<td>47</td>
<td>54</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Age</td>
<td>36.2</td>
<td>29.7</td>
<td>99</td>
<td>0.006</td>
<td></td>
</tr>
<tr>
<td>95% confidence interval</td>
<td>33.2–39.3</td>
<td>27.4–32.0</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Range</td>
<td>21–67</td>
<td>16–53</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>40</td>
<td>50</td>
<td>1</td>
<td>0.338</td>
<td></td>
</tr>
<tr>
<td>Enucleation</td>
<td>21</td>
<td>19</td>
<td>1</td>
<td>0.947</td>
<td>0.330</td>
</tr>
<tr>
<td>Limb amputation</td>
<td>3</td>
<td>6</td>
<td>1</td>
<td>0.498</td>
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</tr>
<tr>
<td>Penis amputation</td>
<td>18 (40)c</td>
<td>22 (50)c</td>
<td>1</td>
<td>0.099</td>
<td>0.924</td>
</tr>
<tr>
<td>Castration</td>
<td>14 (40)c</td>
<td>16 (50)c</td>
<td>1</td>
<td>0.090</td>
<td>0.764</td>
</tr>
<tr>
<td>Other self-injury</td>
<td>12</td>
<td>15</td>
<td>1</td>
<td>0.065</td>
<td>0.799</td>
</tr>
<tr>
<td>On antipsychotics</td>
<td>16</td>
<td>4</td>
<td>1</td>
<td>0.001b</td>
<td></td>
</tr>
<tr>
<td>In hospital, jail, or care facility</td>
<td>9</td>
<td>12</td>
<td>1</td>
<td>0.144</td>
<td>0.704</td>
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<td>Psychotic symptoms, substance use, and evidence of dangerousness, n = 89</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>N</td>
<td>39</td>
<td>50</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Any delusion</td>
<td>37</td>
<td>45</td>
<td>1</td>
<td>0.461</td>
<td></td>
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<tr>
<td>Delusion about the amputated organ</td>
<td>29</td>
<td>33</td>
<td>1</td>
<td>0.724</td>
<td>0.395</td>
</tr>
<tr>
<td>Religious delusions</td>
<td>29</td>
<td>32</td>
<td>1</td>
<td>1.090</td>
<td>0.296</td>
</tr>
<tr>
<td>Referred to religious textd</td>
<td>14</td>
<td>16</td>
<td>1</td>
<td>0.149</td>
<td>0.700</td>
</tr>
<tr>
<td>Command hallucinations to remove an organ</td>
<td>17</td>
<td>11</td>
<td>1</td>
<td>4.736</td>
<td>0.029</td>
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<tr>
<td>Visual hallucinations</td>
<td>5</td>
<td>5</td>
<td>1</td>
<td>0.175</td>
<td>0.676</td>
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<td>Thought disorder</td>
<td>18</td>
<td>23</td>
<td>1</td>
<td>0.000</td>
<td>0.989</td>
</tr>
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<td>Behavior indicating indifference to injury</td>
<td>19</td>
<td>25</td>
<td>1</td>
<td>0.014</td>
<td>0.904</td>
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<tr>
<td>Psychomotor agitation</td>
<td>15</td>
<td>20</td>
<td>1</td>
<td>0.022</td>
<td>0.883</td>
</tr>
<tr>
<td>Habitual substance abuse</td>
<td>5</td>
<td>15</td>
<td>1</td>
<td>3.712</td>
<td>0.054</td>
</tr>
<tr>
<td>Intoxicated at the time</td>
<td>2</td>
<td>5</td>
<td>1</td>
<td>0.001b</td>
<td></td>
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<td>Previous stimulant or hallucinogen use</td>
<td>8</td>
<td>12</td>
<td>1</td>
<td>0.153</td>
<td>0.696</td>
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<tr>
<td>Violence toward others</td>
<td>5</td>
<td>5</td>
<td>1</td>
<td>0.175</td>
<td>0.676</td>
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<tr>
<td>Suicidal thoughts or acts</td>
<td>7</td>
<td>11</td>
<td>1</td>
<td>0.223</td>
<td>0.637</td>
</tr>
</tbody>
</table>

*a*Unpaired 2-tailed *t*-test, *t* = 2.8.
*b*Fisher’s exact test.
*c*Reduced n in brackets.
*d*No patient was reported to have mentioned any religious text other than the gospel of Matthew.

The acquaintance chain method enabled us to locate long-serving clinicians in every mental health service in NSW after only 3 steps, and after 5 steps, no new doctors were suggested. A total of 38 clinicians or administrators were contacted directly by telephone or email. Many of the clinicians we contacted spoke to colleagues before responding, but no new cases were identified from secondary contacts.

We were able to obtain corroborated accounts of 6 enucleations and 3 completed upper limb amputations in NSW between 1990 and 2007. Cases of genital amputation were not remembered in as much detail as the ocular and limb cases, but there were at least 11 cases, all but one of which was in association with a psychotic illness. Of the 13 cases in which treatment status was clearly remembered, 6 were thought to have occurred in the FEP and several occurred soon after the first admission to hospital. The clinicians also described a number of severe cases that fell short of amputation, including 3 almost...
complete hand amputations, 4 self-inflicted eye injuries that resulted in blindness, and a self-amputation of a breast. A number of less severe genital cases, several failed enucleations, a case of psychotically motivated nonsuicidal self-evisceration, 3 additional cases of penetrating injury to the brain via the orbit or nose, and numerous cases of finger amputation were excluded. In total, we found 28 cases of MSM resulting in significant disability in NSW in the 17 years, including 20 cases that would have met the inclusion criteria for our study.

Discussion

Based on reports of 28 cases in NSW between 1990 and 2007, we estimated the incidence of very extreme severe forms of MSM to be at least 1 case per 4 million of population per year during that period. The true figure may have been greater because it is likely that we did not have information about every case, and we did not include cases of less severe MSM. Only 2 of these cases were the subject of a published case report.19 Hence, despite its rarity and confronting nature, published case reports of MSM are only a limited and semirandom sample of MSM events, and the results of this review of published cases should be treated with some caution, because biases and some types of error are possible.

Possible Sources of Error and Bias

The predominance of ocular and genital MSM in published cases may be because these injuries are easier to perform, that the genitals and the eyes have special psychological significance, or because of a publication bias as these injuries may be seen as being of greater interest. Some of the cases of penile and limb amputation were reported because the surgical replantation methods and some ocular cases concerned ophthalmic complications. Hence, although we found a similar ratio of injury types in our informal survey in NSW to the ratio in the published cases, the ratio of MSM injuries in publications may not accurately reflect their incidence in the community.

A more important consideration is whether diagnostic error, the methods used to define FEP, or publication bias influenced the proportion of cases that we counted as FEP.

The number of FEP cases may have been overestimated if the authors of some case histories were unaware of previous treatment. However, 44 of 54 (81%) of the first-episode case histories included a psychiatric and personal history, and the remaining 10 histories clearly stated that the patient had not been diagnosed or treated prior to the injury. On the other hand, the proportion of PTP patients may have been overestimated because 8 cases were classified as PTP on the basis of a diagnosis of chronic schizophrenia, and 3 further patients were classified as PTP because of a preexisting diagnosis of schizophrenia. Previous treatment was assumed but not documented in these 11 cases.

We may also have underestimated the proportion of patients in FEP if some cases were wrongly classified as nonpsychotic. Firstly, there were men who castrated themselves for religious reasons,91,92 a teenage student who amputated his penis so he could concentrate on his studies,93 a man who castrated himself to treat alopecia, although his doctors thought he did not suffer from this condition,94 and a traumatized woman with gustatory hallucinations who amputated her hand because she believed it had done “bad things.”95 Second, all but one of the 7 cases of drug-induced psychosis with known treatment status was in a FEP and several had persistent symptoms, which suggested the correct diagnosis was schizophrenia. Third, few of patients who were given a diagnosis of affective psychosis had received prior treatment but many had bizarre delusions that are more typical of a schizophrenia spectrum psychosis. If the inclusion criteria had included all psychotic illnesses, 71 of 120 (59.2%) cases would have been classified as FEP.

If the 18 case histories of schizophrenia spectrum psychosis that did not document treatment status were assumed to have received treatment, 54 of 119 (45.4%) of the cases could have been classified as FEP. However, some of these cases were almost certainly in the FEP because 6 patients were in their teens or early 20s and as a group they had a mean age that was slightly younger than the mean age of the included cases.

The number of FEP cases did not appear to have been overestimated by reporting or publication bias. Cases in published series of 2 or more patients, which it can be assumed included all the cases known to the author, had a similar chance of being in the FEP (29 of 56, 52%) than cases reported as a single case history (25 of 45, 55%).

Although patients who enucleate both eyes in FEP cannot do so later in their illness, it is unlikely that the proportion of patients in FEP was increased by a survivor effect because patients with MSM make up a tiny proportion of all cases of schizophrenia. Furthermore, only a few FEP patients, including those who underwent limb, penile, or testicular replantation surgery, inflicted further MSM.

The Relative Risk of MSM in FEP and PTP

We found that half the cases of MSM and a schizophrenia spectrum psychosis had never received antipsychotic treatment and more than half were in the FEP. This is a notable finding because schizophrenia is a lifelong condition that usually begins in early adult life.

The majority of published studies of the incidence of schizophrenia report that between 0.01% and 0.03% of the population will develop schizophrenia in any given year and about 0.5% have a preexisting schizophrenic illness.96 If one assumes an incidence of 0.02% per annum...
and a prevalence of 0.5%, there are 25 times more previously treated schizophrenia spectrum patients than first-episode patients in a typical population in any given year. If the risks of MSM were evenly spread over the course of the illness, then the ratio of PTP to FEP patients with MSM would be about 25 to 1. In fact, previously treated patients and those in FEP were found in about equal in numbers, indicating that that the risk of MSM in FEP is as much as 25 times greater than the subsequent annual risk after initial treatment.

The finding supports the hypothesis that the risk of MSM is significantly greater in the FEP compared with subsequent episodes of psychosis, a finding that is similar to recently published studies of homicide in psychosis.

**Psychosis and Causation of MSM**

The authors of some case histories have attributed the MSM to unconscious sexual conflicts or knowledge of the Bible. More recently, it has been suggested that limb self-amputation may be the result of a nonpsychotic disorder provisionally named body integrity identity disorder, and there is also a recent series of patients with nonpsychotic self-emasculation. These 2 series must be viewed with some caution because they are based on contact via the Internet and telephone-based assessment rather than personal interview. However, they do raise the question of how the causation of MSM should be considered.

We used the criteria for epidemiological causation as set down by Austin Bradford Hill to guide a consideration of whether psychosis can be considered to be a cause of MSM. Hill’s criteria (in italics) and our conclusions are

- **Strength of association:** There is a very strong association between psychosis and MSM because at least 143 of 180 (79.4%) patients (excluding 9 cases with no with no mention of the persons mental state) had a psychotic illness, despite chronic psychotic illness affecting less than 1% of the population.
- **Consistency of association:** Published reports of MSM in association with psychosis have come from all parts of the world with increasing frequency since the first case was reported in the 19th century.
- **Specificity of association:** MSM is specifically reported in association with schizophrenia spectrum psychosis. Reports of MSM in association with substance abuse and affective psychosis are rare, but the characteristics of these patients are similar to those with a schizophrenia spectrum psychosis.
- **Temporality:** We only found 2 cases in which MSM may have preceded over psychosis, although it is also possible that some of the cases in which psychosis was not diagnosed were in the prodromal phase of psychotic illness.
- **Biological gradient:** The cases examined in this study had severe psychotic illness with numerous acute symptoms. Many were so ill that they were indifferent to pain and to the loss of their organs. Most had religious delusions, which may be a marker of the severity of psychosis. Plausibility: A reaction to a bizarre delusion leading the patient to remove an organ that they believed was threatening or an experience of hallucinations directing them to remove their organs are plausible links between the psychosis and MSM. Coherence: The presence of MSM among differing types of psychosis suggests a coherent link between psychosis and MSM. Experiment: The lower risk of MSM in treated psychosis represents what Hill described as a “semiexperiment” in which a modification of the cause reduces the effect. Analogy: The risk in first-episode psychosis, the threatening psychotic symptoms, and a predominance of male patients with schizophrenia with an average age in the early 30s are features that MSM patients have in common with psychotic patients who commit homicide.

**Reasons for the Decline in MSM After Treatment**

Although psychosis, particularly first-episode schizophrenia, seems to meet epidemiological criteria for causation of MSM, the reason for the decline in MSM after a period of initial treatment is uncertain.

The lower incidence of MSM in PTP patients may be partly explained by ongoing treatment with antipsychotic medication. However, this is unlikely to be the sole explanation because adherence to medication by patients with schizophrenia is known to be poor and about a third of the PTP patients in this review were reported to be adherent to treatment.

The most obvious explanation is that once patients have experienced a remission and have received a medical explanation of their symptoms, they are less likely to act in such a drastic way when the symptoms return, regardless of the severity of their symptoms or their apparent loss of insight. Other possible explanations are that bizarre organ-specific delusions are less common in PTP or that the intensity of delusional beliefs declines later in the illness. The differences may even be due to changes in the brain after treatment with antipsychotic medication. Other factors such as an ongoing relationship with a treating team or awareness of the patient’s illness by family and friends may result in some protective measures such as encouraging treatment when the patient becomes unwell. Although we found no major clinical differences between those who self-mutilated in FEP and those who did so in PTP, it would have been of interest to compare possible protective factors such as engagement with a treating team and the level of insight of MSM and non-MSM patients in FEP and PTP.

**Risk and Management**

Males in a first episode of a schizophrenic illness that is characterized by delusions associated with a body part or religious delusions are at the greatest risk for MSM.
However, MSM of this severity is so rare that it cannot be predicted accurately unless there has been a previous attempt at self-injury or the patient has spoken about wanting to remove or injure an organ.

Threatened ocular mutilation deserves special mention because it may occur in a hospital setting, and the case histories suggest that one to one nursing is not always sufficient to prevent enucleation. All MSM requires urgent medical attention because genital and limb MSM may cause exsanguination, and subarachnoid hemorrhage, meningitis, and pituitary failure are potentially fatal complications of self-enucleation. Replantation of amputated genitals and limbs can be performed with some return of function, and testicular replantation may avoid the requirement for long-term testosterone therapy. Repeated MSM has been reported but seems to be uncommon. Hence, emergency limb and genital replantation can be justified in the presence of severe psychosis that renders the patient incapable of informed consent. Patients with MSM warrant a detailed assessment for the presence of psychosis and even patients who are not initially forthcoming about the reason for MSM should be regarded as suffering from a psychosis until proved otherwise.

Conclusions

The importance and limitations of published case histories are highlighted by this study. Research about MSM requires new methods because case histories are inevitably subject to publication bias, and valid inferences cannot be made from the observation of small numbers of cases. The reliance on memory and retrospective ways of case finding, such as we used to make an estimate of the incidence of MSM, are also unsatisfactory. Ideally, state health authorities should collected data about MSM events. An alternative is for networks of clinicians to gain ethical consensus. Patients with MSM warrant a detailed assessment locating the publications and the many clinicians in NSW who shared their knowledge of cases of MSM. Declaration of interests: None.

References


