

# Out on a Limb: The Ethical Management of Body Integrity Identity Disorder

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**Abstract** Body integrity identity disorder (BIID), previously called apotemnophilia, is an extremely rare condition where sufferers desire the amputation of a healthy limb because of distress associated with its presence. This paper reviews the medical and philosophical literature on BIID. It proposes an evidenced based and ethically informed approach to its management. Amputation of a healthy limb is an ethically defensible treatment option in BIID and should be offered in some circumstances, but only after clarification of the diagnosis and consideration of other treatment options.

**Keywords** Body integrity identity disorder · Apotemnophilia · Ethics · Therapy · Amputation

A 28-year-old gentleman presents to a surgeon requesting that his left leg be amputated below the knee. He recognises that the leg is perfectly normal, but he feels, and has always felt, that it is not truly part of him. When, in 2000, it became clear that a Scottish surgeon had removed a patient's healthy limb, under just these circumstances, his hospital banned further amputations, a Scottish politician sought legal prohibition and medical ethicist Arthur Caplan was quoted as saying,

“It's absolute, utter lunacy to go along with a request to maim somebody” [1, 2].

Body integrity identity disorder (BIID) is an extremely rare condition whereby an able-bodied and apparently normal individual describes a long standing desire to have a limb amputated. Sufferers report that their motivation for wanting an amputation is a perceived mismatch between their actual body and their internal image of their body. When faced with such a person doctors come upon poorly charted ethical ground. What is the best way to respond to such a person's request? Is it ethically permissible to arrange the amputation of a perfectly normal limb?

This paper seeks to plot an ethically informed course for the management of body integrity identity disorder. To do this, I will review the medical literature on BIID and describe the condition for a philosophical audience. I will argue that, once the nature of the condition is understood, automatic responses to the possibility of amputation like Caplan's are not justified. I will then review the small existing literature around the ethical management of BIID and will present a range of arguments that support the elective amputation of a healthy limb in defined circumstances.

Although BIID causes an enormous amount of moral concern whenever it is seen, the literature contains only three previous attempts to formally examine the ethics of its management. In their 2002 paper, Johnston and Elliot argue, on several grounds,

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that amputation of a healthy limb should not be considered a treatment option [3]. In contrast, in 2005, Bayne and Levy [4] argued that there was a *prima facie* case for allowing access to amputation, and in 2006, Savulescu [5], reviewing the whole area of controversial choices in health, proposed that “not only might amputation be permissible in some situations, it might be desirable”. This paper adds a clinical perspective to the debate and attempts to set down clear evidence-based, ethically informed guidelines for the management of people presenting with BIID. It supports amputation in carefully defined circumstances.

### The Medical Literature Around BIID

There is very little known about BIID and the literature surrounding it is scant. Our knowledge of the condition derives from only three sources—case reports, reports in the media and one methodologically weak survey.

There are only four formal case reports of patients who almost certainly suffered BIID [6–9].<sup>1</sup> The literature surrounding BIID often refers to a number of other cases from other papers, but in these, the patients described either suffered other disorders or insufficient information is presented to allow confidence about the diagnosis [10–16]. There are several news reports, magazine articles and narrative accounts from people who may have had the disorder but their informal nature makes interpretation of their relevance very difficult [1, 2, 17–20].

In the literature’s one survey, respondents were recruited anonymously via the internet and then interviewed over the telephone [6]. This design admits the strong possibility that many of the 52 respondents did not suffer from BIID, but from one of its several differential diagnoses.

Probably as a result of this dearth of scientific literature, there is currently no consensus on what constitutes BIID, nor even that BIID exists as an independent entity. Even if one assumes its existence, there is no good information on its frequency in the community, its clinical features, its underlying cause, or the effectiveness of any possible treatment options.

<sup>1</sup> I have recently seen a fifth case which will appear in the literature in due course.

### What is Body Integrity Identity Disorder?

Almost all psychiatric syndromes are defined by the symptoms that their sufferers typically report and the signs that they typically display. Almost every entry in the much vaunted Diagnostic and Statistical Manual of Mental Disorders (DSM) is of this form.

No formal definition of BIID appears in the literature. Below is a tentative definition structured as diagnostic criteria, of the form typically found in the DSM.

- A. A strong persistent desire for the amputation of a limb.
- B. The primary motivation for the desire is the feeling that being an amputee is one’s true and proper identity.
- C. The disturbance causes clinically significant distress or impairment in social, occupational, or other important areas of functioning.
- D. The disorder is not better explained by another medical or psychiatric syndrome such as somatoparaphrenia, a psychotic disorder or body dysmorphic disorder.

Criteria A–C address the core features of the syndrome as usually described.

The literature describes a related syndrome—apotemnophilia—where people desire amputation primarily for sexual gratification. Criterion B would differentiate apotemnophilia from BIID. First’s survey suggested that there is considerable overlap between the two conditions with 67% of respondents reporting sexual feelings as an important motivator. However, of those people who self identified as wanting an amputation, only 15% reported “feeling sexually excited or aroused” as their primary motivation. Under the criteria above, that 15% would not suffer BIID.

Criterion D looks to weed out other conditions that might be mistaken for BIID. Most importantly for a discussion on the ethics of its treatment, criterion D asserts that BIID should not be regarded as, or confused with, a psychotic disorder. This aspect will be carefully argued below.

### Is the Desire for Amputation in BIID Based in a Psychotic Illness?

Psychotic illnesses are those manifest by delusions or hallucinations. The most common response of the lay

public, and of some ethicists, on first hearing of BIID, is to confidently retort that a person who wants to have a healthy limb amputated must be severely mentally disturbed and that therefore his request should obviously be ignored.

The assumption that sufferers must be psychotic seems to underline Caplan's opposition to the possibility of amputation—"it's about whether (sufferers) are competent to make a decision when they're running around saying 'Chop my leg off'" [1]. Similarly, Wesley J. Smith, commenting on Bayne and Levy, appeals to common sense and claims that obviously no one "but a severely mentally disturbed person would want a healthy leg, arm, hand, or foot cut off" and that "such people need treatment, not amputation" [21].

If sufferers of BIID were delusional these relatively unprocessed responses might be reasonable. We tend to automatically regard those who suffer delusions as incompetent to make decisions that directly concern the contents of those delusions. A person with schizophrenia who believes himself able to fly, will be regarded as incompetent to request defenestration, without any real debate. If people who suffer BIID are suffering from a psychotic illness and their beliefs about body image are delusions, then there is a strong *prima facie* case that their requests are not made competently and that they should be ignored.

If, however, sufferers of BIID do not suffer a recognised psychotic illness and if their beliefs are not easily characterised as delusions, the *prima facie* element of the argument collapses, and their requests for amputation will, at least, need to be taken seriously. Differentiating BIID from psychotic illnesses will take some careful analysis, not least because, of all people described in the medical literature, who have amputated their own limbs, the majority did not suffer BIID, but *were* likely suffering from a psychotic illness when they did so [22, 23].

A simple text book definition of delusions sees them as false beliefs, not ordinarily accepted by members of the person's culture, based on incorrect inferences about external reality. They are firmly sustained despite what almost everyone else believes and despite what constitutes incontrovertible and obvious proof of evidence to the contrary [24].

Sufferers of BIID hold that one or more of their limbs do not form part of their internal body image. Three aspects of the presentation of BIID sufferers

suggest that they do not suffer a recognised psychotic illness and that their belief cannot easily be regarded as a delusion.

First, the BIID sufferers' unusual beliefs about their limbs are typically the only unusual things about them. Delusional beliefs rarely arise in otherwise normal individuals. They are usually just one aspect of a cluster of clinical features that are typically found in a range of psychotic disorders. People with schizophrenia, for example, rarely have just one delusion, and usually have a host of associated clinical features such as hallucinations, disorganised thinking, abnormalities in the way they experience emotions, and difficulties initiating tasks. Differentiating people with schizophrenia and most other psychotic disorders from BIID will present little nosological or diagnostic challenge.

Though psychotic illnesses *usually* manifest with more than just one delusion, monosymptomatic delusional disorders classically present as a single delusion in an otherwise normal individual. The monosymptomatic delusional disorders are much beloved of philosophers and include: Capgras syndrome, the delusion that one's loved ones have been replaced by imposters; Cotard's syndrome, the delusion that one is dead; and Ekbom's syndrome, the delusion that one is infested with insects or bugs [25–27]. If BIID were just another variety of monosymptomatic delusional disorder the *prima facie* argument from ignoring the psychotically ill would hold. However, a careful examination of the features of BIID suggests it should not be seen as a monosymptomatic delusional disorder.

In the real world, the classical forms of these illnesses are incredibly rare. Most sufferers of Cotard's syndrome, for example, also suffer the features of severe depression and these features predate their odd and potentially distressing belief in their own demise [28]. Capgras syndrome generally occurs in the context of other well recognised psychiatric disorders or a range of medical illnesses such as Alzheimer's disease and stroke [25]. Pure monosymptomatic delusions do occur, but they are extremely rare. If BIID were a monosymptomatic delusional disorder and the beliefs of sufferers were delusions, then the syndrome would be extraordinarily unusual because, unlike other monosymptomatic delusions disorders, it is always apparently truly monosymptomatic.

The second challenge to seeing the beliefs of BIID as delusions is that it is generally regarded as a core feature of a delusional belief that it be false. People with Cotard's syndrome believe they are dead, when patently they are not. Capgras' sufferers are mistaken in believing their relatives are imposters and people with Ekbom's continue to believe they are infested despite ample evidence this is not the case. It is not absolutely necessary that the content of a delusional belief be false—someone with Ekbom's syndrome may coincidentally suffer scabies for example. However, as a general rule delusional beliefs are objectively untrue.

Currently there is no way of determining whether or not a BIID sufferer's claim that his leg does not feel like part of his body is true or not. There is no external benchmark against which to compare his claim. The sufferer is simply reporting a mental state and we really only have his word to go on. Leaving aside the possibility, for the moment, that he is deliberately misleading us, it does not seem unreasonable to suppose that if the patient says that his leg does not feel like a part of his body, then that statement is true. After all, we are inclined to believe most other unverifiable reports of mental states, unless we have good evidence that the report must be false. There is no obvious reason to set a higher standard for BIID sufferers' reports about body their image.

The third reason that the beliefs of BIID should not be regarded as delusions or part of psychotic illness is again phenomenological. People with BIID are typically reluctant to speak much of their desires. They do not tell others of their beliefs because they recognise that others will generally see their beliefs as abnormal [6, 9]. The ability to stand outside one's belief and take a third person perspective on it is very unusual in delusions. Even when it does occur in a delusional person, it usually manifests as only a brief acknowledgement that others would see their belief as odd or impossible, but this fleetingly held acknowledgement does not usually provoke any action beyond a grudging agreement of possibility. Most delusional people will happily reveal their delusions as long as their interlocutor is not a subject of their beliefs. In First's survey 32% of sufferers in a relationship had kept their desire a secret even from their partner.

At this point our working definition of a delusion remains phenomenological. With such a definition it will be impossible to state categorically that the

beliefs central to BIID are not delusions, however it *is* possible to say they have few of the features of the beliefs that are usually categorised that way. It is also clear that any ethical argument that depends on BIID being unthinkingly characterised as a psychotic illness, will fall flat. BIID has few of the features that we are accustomed to seeing in psychotic illnesses, if an ethical argument against amputation is to depend upon this categorisation, the proposer of that argument has a lot of work to do. Though the beliefs of sufferers of BIID are very unusual, there is nothing obviously crazy about BIID.

### **Do Other Diagnostic Labels Better Account for the Phenomenology of BIID?**

While the question of whether or not BIID is a psychotic disorder will impact on whether or not a sufferer is capable of rationally consenting to treatment offered, the possibility that BIID might be a manifestation of another disorder will impact on the type of treatments offered. There are a number of candidate syndromes. If BIID were actually a subset of one of these candidate syndromes, then treatments that work for them might also work for BIID.

Body dysmorphic disorder (BDD), is a relatively common condition where sufferers believe that some part of their body is extremely ugly or deformed [29]. It has been estimated that BDD sufferers may account for 7–15% of people seeking cosmetic surgery [30]. About 50% of sufferers appear to hold their belief about their appearance with delusional intensity. The remainder are better described as “preoccupied” rather than “deluded”. In this sense being “preoccupied” pretty much maps with the lay sense of the term. People who are preoccupied with an unusual belief, have it their minds much of the time and attach a deal of emotional importance to it. However, unlike people with delusions, they are not maniacally concerned with the belief, and are not implacably opposed to attempts to challenge the veracity of the belief. People with BIID are probably best described as preoccupied with their unusual belief. Like people with BIID, those suffering BDD often present late for assistance because they believe that others will regard their concerns as crazy.

There is evidence that people with BDD can be helped with the prescription of selective serotonin

reuptake inhibitors (SSRIs) [31]. SSRIs include medications such as fluoxetine (Prozac®) that are usually used in depression. When used in BDD, SSRIs are given at doses higher than those usually used in depression and improvement may take longer to manifest. If BIID were a subset of BDD, the use of SSRIs might make the desire for amputation fade away.

In First's survey most subjects identifying themselves as suffering BIID, reported that their perception of the limb, which they wanted to have amputated, did not differ from that of their other limbs. Only one is reported as claiming that the limb was ugly and this was not the primary reason for wanting an amputation. As a belief in bodily deformity seems a key feature of BDD, its apparent absence in BIID, makes it unlikely that the two disorders are one, however this does not mean that the treatment of one may not work for the other. Obsessive compulsive disorder, another disorder where preoccupations, rather than delusions, seem central, also responds to high dose SSRIs [32].

Factitious disorder is a rare condition whereby sufferers deliberately feign symptoms or fake signs in order to convince medical professionals that they are suffering from a condition that they do not have [33]. Their dissembling is not motivated by the pursuit of financial or other obvious gain, as it is when a patient malingers; rather they seem to be feigning symptoms for some more cryptic motivation. Often it is felt that their aim is simply to achieve the status of a patient. On the face of it, BIID might be a variety of factitious disorder. Perhaps so-called sufferers are simply lying about their desire to want their limb amputated, so that they may become amputees, for no other reason than to achieve the status of an amputee. There is currently no way of differentiating a true statement about one's mental states from a false one. It is therefore possible that all sufferers of BIID, actually suffer from factitious disorder, but on balance this seems unlikely. As with other psychiatric syndromes, the symptoms of factitious disorder tend to occur in typical symptom complexes. In severe cases, sufferers move peripatetically from place to place. They are rarely in a committed relationship. They often present with a range of other feigned complaints and frequently lie about all aspects of their lives. First's survey suggests that none of these features are typical in BIID. Baubet et al. [10] describe a woman who eventually had her leg amputated whom they believed had factitious disorder. Though the authors of this

paper are BIID sceptics, they are clear that their patient did not have BIID because she did not have the cardinal features of BIID described above.

### What Causes BIID?

Having defined what we mean and don't mean by BIID, we are now in a position to ask about its aetiology. Obviously if we had a clear understanding of its cause, that would assist in its treatment. Not surprisingly, given the number of cases reported, there is little evidence upon which to base any hard conclusion.

The literature on BIID grew out of the literature on apotemnophilia, and one aetiological theory suggests that BIID, even if it is different from apotemnophilia, still finds its root cause in erotic attraction to amputees. Lawrence, for example, draws a parallel with theories on the aetiology of transsexualism, and suggests that BIID arises from an "erotic target location error" where sufferers are attracted to amputees but incorrectly target their own body in their libidinal strivings [34]. She goes on to predict, that if this theory were true, then sufferers would almost always be attracted to amputees and would display an increased prevalence of gender identity problems.

Other theorists have noted similarities between somatoparaphrenia and BIID. Somatoparaphrenia is a rare condition, usually following a stroke, that causes damage to the parietal lobe of the brain. As patients recover they typically deny ownership of one of their limbs, often experiencing it as "alien" or belonging to someone else [35]. Ramachandran and McGeoch [36] theorise that BIID might result from malfunction of that part of the brain damaged in somatoparaphrenia and have predicted that BIID sufferers may display functional derangement of the parietal cortex and its connections with the limbic system. A forthcoming paper from the group will provide some evidence supporting their hypothesis (Paul McGeoch, personal communication).

In summary therefore, although brain abnormality theories appear promising, it is still too early to be clear about the cause of BIID.

Finally we are in a position to examine our original dilemma. How should a doctor respond to a request from a person who would like their healthy limb amputated?

The first task is to take a thorough history and examination so as to assure one's self that the patient's desire is a symptom of BIID. This done, the question may be recast as, what is an ethical approach to the management of BIID?

### Available Treatment Options in BIID

It might be expected that patients presenting with the features of BIID will want their doctor to organise an amputation. There is some evidence that amputation may improve the life circumstances of those with BIID, but this data is scant. The best evidence comes from First's survey, where "six subjects had a major limb amputation at their desired site and reported that following the amputation they no longer had any desire for an amputation and that they felt better than they have ever felt" [6]. Concerningly though, in two of the literature's four case reports, patients who received one amputation, later wanted or organised another one [7, 8].

If there was good evidence of another less dramatic, irreversible and debilitating therapeutic manoeuvre reducing the suffering associated with BIID, this would be a strong reason for resisting the patient's request for amputation and counselling the use of this other measure instead. The two approaches often held out as alternatives are psychotherapy and pharmacotherapy. Unfortunately, at this point, neither of these has good evidence of efficacy either.

Psychotherapy has apparently been frequently tried. In First's survey, a majority of the subjects (65%,  $n=34$ ) had been in psychotherapy at some time in their lives, though none reported that it had reduced the desire for amputation [it is worth noting however that almost half ( $n=16$ ) never told their therapists about their desire for amputation, which would undoubtedly have proved a barrier to therapeutic efficacy]. Braam et al. [9] describe in considerable detail their experience with a patient with BIID, who at the time of their writing had undergone thirty sessions of cognitive behavioural therapy. Though they report that their patient exhibited a persistently lower level of distress over the course of therapy, it seemed that much of this was related to an improved ability to share the burden of his condition with others and there seemed little evidence that the underlying desire for amputation was much improved.

In First's survey, 40% ( $n=21$ ) of the subjects had taken psychotropic medication at some point in their lives, with the majority taking either a selective serotonin reuptake inhibitor (SSRI) or clomipramine (an antidepressant which also inhibits serotonin reuptake). Few could recall the prescribed dosage. None of these subjects reported any appreciable effect from the medication on the desire for amputation although many reported that their mood improved [6]. Braam also found that the addition of an SSRI (paroxetine 20 mg) only seemed to lessen the distress and not the desire [9]. Berger et al. [7] though reported some loss of desire in their patient, who was placed on a relatively high dose of another SSRI (fluoxetine 60 mg). There is no evidence of efficacy of antipsychotic medication in BIID in the literature.

Ramachandran and McGeoch [36] have postulated that vestibular caloric stimulation (flushing the ear canal with cold water) might present a possible treatment for BIID, based on its temporary effect on somatoparaphrenia. At this point however this is no more than speculation.

### Choosing Between Alternatives

Let us assume for a moment that there is no reason to prohibit the amputation of a healthy limb and that a doctor faced with a patient with BIID must choose between one of the treatment options above. This decision between competing treatments involves comparing them via a complicated but familiar algorithm that weighs four factors:

1. The probability that a treatment will successfully relieve the patient's suffering and the degree to which the suffering might be relieved.
2. The probability that a treatment might cause some harm to the patient and the degree of harm that might be involved.
3. The cost of the treatment to society (though this may be offset by patients funding the treatment themselves).
4. The patient's preference for a particular treatment.

At this point the treatment option with the most evidence of efficacy is amputation; however this evidence is anything but robust. Even if the amputation relieves the symptoms of BIID, it will also cause some harm to patients by leaving them disabled.

Amputation is also costly, not only in terms of the operation and rehabilitation which could conceivably be covered by the patient, but also in terms of lost productivity through disability. While there is even less evidence for the efficacy of selective serotonin uptake inhibitors (SSRIs), there are theoretical reasons to suspect they may be effective if prescribed in higher doses than have generally been reported in the literature to date. SSRIs are relatively cheap, carry few side effects and can be trialled over a period of six to twelve weeks. Unlike amputation, if it is clear that they are not helping after a period, they may simply be withdrawn and their effect reversed.

Vestibular caloric stimulation, the speculative treatment cited by Ramachandran and McGeoch is cheap, without side effects and its effects (or lack of them) should be become apparent immediately.

At this point, even if a patient were to strongly prefer amputation and were to offer to pay for it, it would be hard to justify meeting his request without at least a trial of vestibular caloric stimulation and then a trial of SSRIs at the higher doses that would be prescribed for obsessive-compulsive disorder.

If both of these were unsuccessful however, the next course of action is less clear. Though it is possible that one or other form of psychotherapy may prove useful, there is nothing in the literature to give any confidence that it might. In addition, most psychotherapeutic endeavours will be costly and their effects (or lack of them) may take months or years to become manifest. It seems, though, that many patients with BIID are willing to take up this option, and if this were the patient's preference, then a trial of psychotherapy might be useful, even if only to relieve distress rather than the underlying feeling that one's leg is not one's own. There is no published evidence supporting the efficacy of antipsychotics in BIID. However, it is possible that they might be efficacious, especially if there is a lingering possibility that a particular patient's belief might be best classified as a delusion. Like the SSRIs, antipsychotics are cheap, reversible, carry few side effects and can be trialled over a definite and relatively short period. Antipsychotics might be a treatment option depending on patient preference and a detailed review of the clinical picture.

What though, if our trials of caloric stimulation and SSRIs have failed, and trials of psychotherapy or antipsychotics are either refused or thought unlikely to be successful?

Just because something can be done, it does not follow that it should be done. An obvious, but underappreciated, treatment option at this point, is to do nothing. It seems that most patients with BIID wait years before seeking help. It may be many sufferers, though unhappy with their extra limb, would also be unhappy with an amputation. Some sufferers might prefer to wait for possible future developments, such as some variant of caloric stimulation, or to accept treatment aimed at assisting with their distress rather than (what they see as) their underlying problem. At least some patients, though, will not be content with conservative management. Some patients suffer enormously because of their BIID, so much so that, on occasion they are driven to suicide or to injure their limb to force its removal [6–8]. Unless there were good reasons to prohibit amputation in these circumstances, then, weighing the customary factors involved in making treatment decisions and assuming the patient understood the risks, benefits and uncertainty surrounding the procedure, the patient should be offered amputation and subsequent rehabilitation.

### **Are There Reasons for Prohibiting Amputation in BIID?**

In the section above we assumed that there were no reasons for prohibiting the amputation of a healthy limb. A number of arguments could be, and have been, made for prohibiting amputation in these circumstances. I have reviewed and countered each of them below.

#### **Primum Non Nocere**

Though an obviously weak argument, this Latin phrase has a strong grounding in the medical psyche and this is likely to be the first argument ventured by any physician initially uncomfortable with this proposal. The argument suggests that a doctor has a duty to “first do no harm” and that to amputate a healthy limb contravenes this duty. Both Wesley J Smith and Arthur Caplan have advanced this argument in relation to amputation in BIID [1, 21]. The principle itself is vague and despite its popularity, it is often not clear exactly how it is supposed to be applied. Before removing an inflamed appendix, a surgeon must make an incision through a perfectly normal abdominal

wall, but this of course raises no ethical dilemma, because of the obvious trade off in benefit achieved by this comparatively minor harm. If the principle is to have any worth, it must be as an injunction against physicians causing their patients more harm than benefit. In the case under consideration though, the harm, in terms of the loss of limb, would obviously be traded against the hope of a significant benefit, in terms of improved psychological well being.

There are already numerous examples in medical practice where healthy body parts are removed for perceived benefit, without serious ethical dissent. Living organ donors may give up healthy kidneys so as to benefit others and to derive a psychological benefit from having done so. Women at high risk of breast cancer regularly undergo removal of their healthy breasts or ovaries, so as to diminish their risk of developing cancer and to reduce the anxiety associated with living with their high cancer risk. In the closest parallel to BIID, those suffering gender identity disorder are offered sex reassignment surgery; so that their bodily appearance might better match the sex that they believe represents their true self.

#### The Possible Illegality of Amputation of a Healthy Limb

Johnston and Elliot [3] spend a considerable portion of their paper trying to establish that the amputation of a healthy limb is, or may be, illegal and that therefore it should not be conducted. Surgeons should not, as a general rule, break the law, but legal questions should not be confused with ethical questions. If amputation of a healthy limb were illegal, but in some circumstances the most ethical treatment approach, then efforts should be made to have the law changed.

As it happens however, the authors' arguments for illegality are not strong. They rest largely on the supposition that the courts would not "consider amputation of a healthy limb to be 'proper medical treatment' without evidence of some kind of therapeutic benefit" and that therefore performing such amputations would risk a charge of assault. As has been demonstrated above, however, there *is* some evidence of therapeutic benefit and the hope of achieving this would be the motivation for surgery. Perhaps more likely than being charged with assault, is the possibility that a surgeon undertaking such a procedure might be at risk of an action for negligence

if the amputation failed to help the patient. However, the hope of benefit without clear alternative, together with documentation that the consenting patient was realistically informed of the risks, benefits and uncertainties surrounding the procedure, would go a long way in providing a defence to that sort of action.

#### The Paucity of Knowledge About Healthy Limb Amputation

In a related argument Johnston and Elliot suggest that it would be unethical to "embark on...[a] surgical treatment for a psychiatric condition without first subjecting it to the rigorous standards of research... that have come to characterise sound scientific medicine". There are two problems with this argument. First, the authors seem to have an unrealistic view of the way medicine proceeds when dealing with rare or unusual disorders. While it is obvious that rigorously controlled studies into treatment efficacy are to be preferred, such studies are only available when illnesses are sufficiently common to make such studies possible. When illnesses occur infrequently, treatment decisions and even clinical guidelines are often based merely on small case series and published case reports, very similar to those I am outlining for BIID. For example there is very little good scientific data on the optimal treatment of Capras', Cotard's or Ekblom's syndromes mentioned earlier. When faced with these syndromes, doctors base their treatment decisions on the results of published case reports and case series and on analogy with other similar conditions for which stronger evidence exists. Doctors cannot afford to wait for rigorous research when deciding how to treat many rare illnesses, because if they did, the sufferers of these illnesses would wait forever. Even without good evidence, treatment decisions must be made, and even to decide to do nothing is to make a decision.

The argument also fails because it raises a *Catch-22*. If surgery is to be prohibited on the grounds of a paucity of data of its efficacy, then there is no possibility of gathering the data that would be needed to allow it. A lack of available evidence is an argument for therapeutic caution and for careful observation of the results of therapeutic interventions (see below), but it cannot be an argument for prohibition of a therapeutic endeavour, otherwise all new therapeutic endeavours would have to be banned.

### That We Should Err on the Side of Caution

Although no author explicitly posits this argument it is easily derivable from the argument from the paucity of current knowledge. Rather than embarking on this sort of radical and irreversible treatment, we should stay our hand and hope that something better comes along. In this context, this argument would not merely urge that the patient be advised that it might be better to await future developments, it would insist that, regardless of the patient's views, the surgery should be prohibited. If posited, the argument would need to be strong enough to trump the patient's own considered determinations of future risks and benefits.

Two arguments would counter this stance. First, at this point there is no particular reason to expect a breakthrough in the treatment of BIID anytime soon. Its aetiology is still unknown, and the promising advances toward discovery of its cause, suggest that it may be a congenital brain disorder. Assuming vestibular caloric stimulation comes to nothing, there is no other obvious treatment modality upon the horizon. Second, it is difficult to underestimate the degree of suffering that at least some sufferers of BIID seem to endure. It takes enormous load of desperation to drive otherwise normal individuals to traumatically amputate their own limbs. A call for an err toward caution appears to unreasonably minimise the consequences of doing nothing, and the inevitable suffering that this will entail for some BIID sufferers.

### The Possibility that Formal Recognition of the Disorder Will See its Prevalence Grow

Johnson and Elliot also argue against allowing amputation on the grounds that if we start "classifying the desire for amputation as a psychiatric disorder [this] may eventually encourage a much broader range of people to see their own psychic distress as a problem that can be relieved only by amputation.... Once the desire for amputation is recognised as a formal psychiatric disorder,...linguistic and institutional structures may also help nurture and shape an emerging social identity". [3].

The authors seem to be trying to establish a slippery slope argument. Even if amputation were morally acceptable for genuine sufferers of BIID, once that door is opened, others, not current sufferers of BIID, will begin to want their limbs off too. The

legitimisation of BIID within a diagnostic lexicon, will somehow lead to either the creation of new actual sufferers or will persuade people who do not have BIID, that they would like an amputation anyway. To support their argument Johnston and Elliot cite an anecdotally reported increase in the prevalence of gender identity disorder (GID) and the desire for sex reassignment surgery, in the years after GID become officially recognised [37]. Bayne and Levy [4] are also troubled by this argument. Although they eventually dismiss it, they cite, in its support, the anecdotally apparent increase in dissociative identity disorder (formerly multiple personality disorder) after its official sanction as a diagnosis.

The argument would only have force if it entails more than simply facilitating the access of current sufferers of BIID to treatment. Official recognition of BIID would almost certainly lead to more people who currently see themselves as sufferers announcing themselves to a medical profession that finally appears to be taking their concerns seriously. If amputation were to prove to be an effective treatment for the suffering of BIID, an increase in actual sufferers accessing it would be a good thing, not a bad thing.

If formal recognition of the diagnosis *were* to lead to some people, who would otherwise not have been affected by BIID, to now seek amputation, then there would be a *prima facie* argument for prohibiting the amputation of healthy limbs on utilitarian grounds. However, there are several reasons to doubt that this would occur.

First, it is not clear whether the apparent increase in the prevalence of these new psychiatric disorders after their recognition, represents a real increase in occurrence or simply an increase both in the number of sufferers coming forward and improved recognition of their condition by their doctors. The only way one could answer this question would be by reference to community symptom prevalence surveys conducted before and after a new syndrome was legitimised. At this point this sort of data is not available for the syndromes cited as likely prior examples of disorders that became more prevalent after they were formally recognised.

Second, it is likely that many people, who might come to identify themselves as having the new syndrome, still suffered symptoms before its recognition, but would now come to see their symptoms as

part of the new diagnostic entity. Another new disease, “Morgellons”, provides a good example of this. Morgellons is an unexplained and debilitating condition manifest by a “range of cutaneous symptoms including crawling, biting and stinging sensations; and granules, threads or black speck-like materials on or beneath the skin. Some sufferers also report systemic manifestations such as fatigue, mental confusion, short term memory loss, joint pain, and changes in vision” [38]. The Center for Disease Control has recently launched an investigation into Morgellons, because of concerns of an apparent increase in its prevalence. If prevailing medical opinion is to be believed, we may be fairly confident that no one suffered from Morgellons before the term was coined and widely publicised in 2002. This is because prevailing medical opinion holds that the disease does not really exist. Most doctors believe that the only factor that truly groups sufferers of Morgellons, is that they mistakenly believe they have Morgellons. Assuming this is true, it is quite likely that many, and possibly most people who identify themselves as sufferers of Morgellons, actually suffer from a range of other medical conditions. Many probably suffer from Ekblom’s syndrome [39]. The invention of Morgellons may not have led to any more suffering, it may simply have changed the way that people badge the suffering they already had. Some have even argued that the invention of the new disease has provided a new avenue for sufferers of Ekblom’s to access help [40].

Third, even if it is the case, as has been argued for multiple personality disorder, that the legitimisation of a disease may cause some vulnerable souls to actually develop it, this seems incredibly unlikely in the case of BIID. The vast majority of people would be devastated if they were to ever lose a limb. It is our acknowledgement of, and empathy with, this feeling of devastation that makes BIID so difficult for the rest of us to understand. This is what has made many assume that sufferers must be mad. It defies belief that legitimisation of BIID would somehow convert normal people into BIID sufferers or persuade non-sufferers, swept up in a new cultural paradigm, that they should amputate a healthy limb [41].

Finally, even if such transformations were to occur, these new “wannabe” amputees would still be excluded from access to amputation under the form of recognition envisaged here. Our draft diagnostic

criteria require the person to have experienced a strong *persistent* desire for amputation. Exactly what constitutes ‘persistent’ is not made explicit in the draft criteria, but in the case of gender identity disorder there is a general expectation that the desire for a sex change should have been present for at least two years and one might expect a similar time frame to be observed with BIID.

In short, the argument that invokes the possibility that formalisation of BIID as a diagnosis might see its prevalence grow, or demand for amputations increase, seems to rest on nothing more than fanciful speculation.

### Arguments Directly Supporting Amputation for BIID

The most obvious argument in support of amputation in BIID is that, once reasonable alternatives have been exhausted, it offers the best chance of relieving the suffering of those with the condition [4]. This argument is implied in most of the above. Two other arguments are also worth brief mention.

#### Autonomy

The argument from autonomy is the strongest of the arguments that could be mounted in opposition to an attempt to ban amputation for BIID. It has already been aired as part of the four factor model for choosing between treatment options above and is very familiar from a range of other contexts. Basically it is the notion that an individual’s informed conception of his or her good should be respected in the context of medical decision-making.

Autonomy is at the heart of Savulescu’s treatment of this issue. In ‘Autonomy, the Good Life and Controversial Choices’ he looks at all health related choices that seem either irrational or against the chooser’s best interest [5]. Amputation in BIID is just one example; others include, requests for euthanasia, refusals of blood transfusions and the decision to go bungee jumping. While a full recounting of his thinking is beyond the scope of this paper, he focuses particularly on a Kantian interpretation of autonomy, arguing that choices should be respected if they are informed and made rationally, with due consideration of all relevant values. Good normative decisions should be respected if they improve the person’s

overall well-being, even if they apparently harm the person's health. On the specific issue at hand, Savulescu criticises Bayne and Levy's conclusions as "rather timid", arguing that "not only might amputation be permissible in some situations, it might be desirable" and that "[w]e must be open to such radical possibilities".

The argument from autonomy would fail if sufferers of BIID were in some way incompetent to request the amputation of their limb, or in Savulescu's conception, irrational. The analysis of the first half of this paper reveals little reason to think that this applies to sufferers of BIID.

### Harm Minimisation

The current lack of ethical direction in the literature is of concern not only to clinicians and to the patients they find before them, but also to the unknown number of BIID sufferers who are yet to see a clinician. As is the case for many rare disorders, the internet provides sufferers of BIID with unprecedented opportunities to support each other and share information. Numerous websites proffer information on the disorder and academic papers are added to these resources as quickly as they are published. It is probably more likely that you, dear reader, have downloaded this paper via a BIID site than you have from the site of the original journal.

The lack of a consistent ethically informed approach to BIID will be noted by its sufferers and they will not be able to have confidence that their distress will be taken seriously, let alone acted upon. Unfortunately too, much of the information available even in the academic sphere is wrong. Several authors have echoed Smith's unreferenced and misleading statement that "at present it seems that elective surgery is not an option in the west...[and that]...true sufferers are therefore committed to self injury as the only way to achieve their requirements" [42]. While it is clear that there is no ethical consensus regarding amputation, there is simply no evidence that elective surgery is not available in the West. No western jurisdiction explicitly forbids such surgery and while it is certainly true that any surgeon would be cautious before embarking on elective surgery, this hardly translates into total unavailability.

Unfortunately though, sufferers, reading such statements and finding no authoritative counters to them,

are likely to come to believe that self injury (or surgeons from the third world) *are* their only options, thus exposing themselves to the numerous risks associated with this [17].

Were the medical profession or legislators to adopt a clear prohibition of amputation in BIID, then it is quite likely some sufferers of BIID would take matters into their own hands and suffer as a consequence [4]. Of course, the possibility that sufferers may harm themselves if we prohibit access to amputation, should not mean that we should allow ourselves to be strongarmed into providing access to an unethical treatment. If, however, as I have argued, amputation is not unethical, then the prospect that denying access to it will cause harm, provides further support for it being made available on simple utilitarian grounds.

For the time being though, such a clear prohibition is a long way off, and I would argue, should not be adopted at all.

### Other Ethical Issues

Three further issues warrant some discussion.

#### A Duty to Perform Amputation or to Refer On

Despite all argument above, many doctors will still find themselves unable to agree that the amputation of a healthy limb is ethically sound, even if all the caveats outlined are met. As is the case in termination of pregnancy, dissenting doctors should be under no obligation to proceed with an amputation in these circumstances, but are under an obligation to refer the patient to another doctor whom they believe might proceed with the amputation if all the caveats are met [43].

#### A Duty to Report the Results of BIID Treatment

While the lack of scientific knowledge about the treatment of BIID, does not, as Johnston and Elliot had argued, represent a reason to prohibit treatment, it does present a duty to those who embark on treatment strategies to report the results of their efforts to the broader medical community. There is no realistic possibility that there will ever be a properly controlled trial of interventions in BIID—it is just too rare. As a

result most of our knowledge of what will help to relieve the suffering occasioned by the disorder will come from case reports and small case series. In effect, all attempts at treatment should be considered experimental and when possible all cases should be written up, whether the trialled intervention is successful or not.

Unfortunately the small numbers of BIID sufferers complicates this duty somewhat. When most medical cases are written up, the patients described can usually be assured that their identities will be obscured and their confidentiality preserved. In BIID however, the community of sufferers is small, but Internet-savvy and new case reports will likely be picked up quickly and posted on web sites. In these circumstances, attempts to mask the identity of particular patients may prove impossible. These concerns about confidentiality need to be weighed against the duty to report treatment results and patients should be fully informed of the risks to confidentiality as part of gaining consent to write up cases.

#### The Possibility of Transplant

Bizarre as it may sound, it is worth briefly reviewing the possibility that in future sufferers of BIID, who are allowed access to upper limb amputation, might become donors to those who have lost their own hands and arms. Such is the quality of prosthetic legs, that those who lose a leg are able to regain function with the prosthesis that is similar to that that they had before their loss. Prosthetic hands though are a different story. The loss of a hand, particularly of both hands, represents an enormous disability that prosthetics can only begin to cover.

As of September 2004, 11 unilateral and six bilateral hand or forearm transplants had been performed around the globe [44]. The 17 recipients all received intensive physiotherapy and one year after the transplant all had at least some function in their new hands, including the ability to grasp a glass, shave and attend to personal hygiene.

Difficulties in securing donor kidneys and livers have meant that increasingly transplant services have enlisted live donors to augment supplies. In 2005, 55% of all Australian kidney donors were living [45]. Live donors allow timely matching of donor to recipient and the possibility of extremely healthy body parts. Unless good arguments can be found to counter the notion, it would seem sensible to allow

future sufferers of BIID who wanted amputation to become limb donors, if they wished.

#### Conclusion

When faced with a patient requesting the amputation of a healthy limb, clinicians should make a careful diagnostic assessment. If the patient is found to have body integrity identity disorder, amputation of the healthy limb may be appropriate after a trial of selective serotonin reuptake inhibitors and after careful consideration of the risks, benefits and unknowns of all possible treatment alternatives. The results of treatment trials should be published to allow growth in our knowledge of the condition.

Sufferers of BIID might be relieved to know that members of the medical profession will take their concerns seriously and that, after careful deliberation, elective amputation of their troubling limb is a real possibility.

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