



Limb amputation and other disability desires as a medical condition

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See Online for appendix

Some people have a profound dissatisfaction with what is considered an able-bodied state by most others. These individuals desire to be disabled, by conventional standards. In this Review, we integrate research findings about the desire for a major limb amputation or paralysis (xenomelia). Neuropsychological and neuroimaging explorations of xenomelia show functional and structural abnormalities in predominantly right hemisphere cortical circuits of higher-order bodily representation, including affective and sexual aspects of corporeal awareness. These neural underpinnings of xenomelia do not necessarily imply a neurological cause, and a full understanding of the condition requires consideration of the interface between neural and social contributions to the bodily self and the concept of disability. Irrespective of cause, disability desires are accompanied by a disabling bodily dysphoria, in many respects similar to gender dysphoria, and we suggest that they should be considered a mental disorder.

Introduction

This Review integrates research findings concerning a condition that is not yet recognised as a mental disorder, which still needs a generally accepted medical label, and whose conceptual basis is still under construction. The condition involves a person's dissatisfaction with having an able-bodied state. It is referred to as body integrity identity disorder (BIID) in the psychiatric literature, xenomelia in neurologically oriented studies, and transability (transableism) in approaches to disability from a primarily sociological perspective (appendix, for the terminology). In this Review, we use the term disability desires with the aim to remain descriptive and avoid controversial concepts of identity or too-narrow notions of focal brain damage. Although deafness, blindness, or the status of eunuch can be the target of disability desires, the focus of our Review is on the desire for limb amputation or paraplegia. These forms of profound body modification are the most frequently reported type of BIID in case reports and surveys. Also, empirical data beyond those collected by questionnaire studies are available only about individuals who wish to get rid of one or more of their limbs.

Core features of disability desires

People with a disability desire describe a profound mismatch between their actual and their desired body, with respect to its shape or functionality. A frequently used word in their complaints is overcompleteness. In the most common variant—the desire for amputation—people suffer from having four limbs, because their felt identity is that of an amputee, and their inner body does not match their physical appearance (panel 1). In a case of desired hearing loss, normal hearing was experienced as a source of stress and fear in the form of hyperacusis and misophonia,¹¹ and the longing for blindness could be described as a suffering from regular light conditions. Commonly, the desire for a specific disability is not stronger than the desire to compensate for it by use of prosthetics, such as crutches, a wheelchair, or hearing aids. In fact,

pretending to already have the desired disability and to be dependent on the respective aids is an important, almost required, aspect of the condition (panels 1, 2). An onset of disability desires in childhood or early adolescence, between the ages of 5 and 15 years, is required for diagnosis.²⁴

Prevalence and demographics

The overall prevalence of disability desires in the general population is unknown. It is higher in western cultures that promote societal individualism,²⁵ but just a few cases in a single report have been described in Asian countries.²⁶ A growing number of virtual communities devoted to disability desires exist on the internet, but prevalence estimates cannot be derived from this resource. An overwhelming majority of cases occur in men. This gender difference is greater in individuals with the desire for limb amputation than those who wish for paraplegia,²⁷ but possibly smaller for disability desires targeting sight and hearing.²⁸ The prevalence of non-heterosexuality among people with disability desires is controversial (panel 2, table 1). In the three largest surveys, the incidences were 38% (n=52),¹ 39% (n=72),¹⁷ and 44.5% (n=54),⁸ which are far greater than the prevalence of non-heterosexuals in the general population (about 2%);²⁹ however, the reasons for the association between disability desires and sexual orientation are unknown.

Proposed diagnostic criteria and differential diagnosis

Disability desires are not currently listed in DSM-5 or ICD-10. The diagnostic criteria proposed by advocates of its future classification as a mental disorder under the label of BIID is presented in the appendix.¹⁶ Briefly, the desire to become disabled must be longstanding and intense, must produce persistent discomfort, and attempts to reach a disabled state might lead to self-inflicted harm.^{4,30,31} Onset of the disorder must be before adulthood.

The desire for major body modifications must not result from a psychotic disorder, in which self-amputations and

mutilation fantasies are occasionally observed. Diagnosis of acute brain damage should also be excluded, because cerebral lesions can be accompanied by an estrangement from one's own limbs. Frequently discussed in the context of the desire for limb amputation are somatoparaphrenia (ie, the denial of ownership over functionally impaired limbs) and misoplegia (ie, the hatred of own body parts).³² Although these and other neurological syndromes are superficially linked to disability desires (appendix), none of them captures the essence of disability desires—ie, the non-delusional fantasy, in the absence of sensory misperceptions, that some body modification would enable the person to realise their true identity. All these syndromes have a sudden onset (most typically they follow a cerebrovascular incident), whereas disability desires are a developmental disorder in the experienced unity of body and self. A one-to-one correspondence in phenomenology must not, therefore, be expected. Body dysmorphic disorder is sometimes confounded with disability desires.^{3,33,34} The two conditions can share a preoccupation with a specific part of the body, but body dysmorphic disorder is typically concerned with the visual aspects of that part, which is never the case in disability desires. Also, the concerns in body dysmorphic disorders usually focus on facial parts, not on major limbs.

The desire for healthy limb amputation and gender dysphoria have close conceptual links.^{14,25} In fact, the term BIID was modelled after that of gender identity disorder (GID).¹ Table 2 lists some of the commonalities between the conditions. They both comprise an early onset, a predominance in the male sex, and the frequent simulation of the desired state. An elevated prevalence of non-right-handedness points to abnormalities at early neurodevelopmental stages, but the relevant neural structures have not yet been identified. Proposed cerebral loci and circuits appear to increase with an increasing number of studies both for GID³⁸ and for BIID.

Before being conceptualised as an identity disorder, the desire for amputation was considered a paraphilia. Apotemnophilia⁴ denotes the sexual arousal by the fantasy of being an amputee and is, more often than not, accompanied by acrotomophilia,³⁹ the sexual arousal by others' disabilities (notably an amputation). However, apotemnophiles who were lobbying for an inclusion of amputation desires in the DSM argued that they do not have a paraphilic origin. Bridy⁴⁰ noted that "apotemnophiles and their advocates today are at pains to emphasise that apotemnophilia is a broad-based identity disorder and not one that is narrowly sexually determined". With the proposal that BIID, like GID, should be an identity disorder,¹ disability desires were released from the too-narrow focus on sexuality; however, this was at the price of downplaying the sexual aspects. In his seminal study of 52 people with disability desires,¹ the American psychiatrist First emphasised that only 15% of the participants indicated sexual arousal as the primary

Panel 1: Illustrative first-hand descriptions of the desire for disabilities

Limb amputation

- "I feel myself complete without my left leg...I'm over-complete with it."¹
- "I started out as a devotee but the need to see and be with an amputee became so strong that I knew the answer to my problem was to have my own stump."²
- "The soul feels as though it belongs to a body with only one leg. The body does not correspond to this inner reality."³
- "Since my 13th year, my conscious life has been absorbed, with varying intensity, in a bizarre and prepotent obsessive wish, need, desire to have my leg amputated above the knee."⁴
- "I want to wake up with a bandaged stump with two crutches by my bed which I wish to depend on for the rest of my life."⁵
- "I was 4 years old when I first knew that I wanted my leg cut off. I have no idea how I got this 'need' or where it came from."⁶

Paraplegia

- "I have needed to be paralysed from the belly-button downwards since I was a very young child. [...] The only thing that has helped me feeling some peace of mind is to use a wheelchair. In my mid to late 20s, I started living 'full-time'—That is, I used a wheelchair in my day-to-day life, all day, every day."⁷
- "I am using a wheelchair 'full-time' when I'm in public. I walk at home. This is the only way how to remain somewhat functional."⁸

Orchidectomy

- "[My] testicles seemed unnatural; a growth that shouldn't be there."⁹

Sensory impairment

- "I want to be deaf but I'd also use hearing aids to restore my hearing."¹⁰
- "When it's pitch dark, I come closest to life as it should be for me." (PB, unpublished)

Panel 2: Characteristic features of disability desires and the persons affected

Established

- Marked suffering
- Mostly men affected
- Typically high education status
- Largely unremarkable personality profile
- First manifestation in childhood or early adolescence
- Pretending to have the desired disability is almost universally present
- Most common form is the desire for limb amputation (xenomelia), which is more often for legs than arms, and more often for left-sided than right-sided limbs

Controversial

- Emasculation desire part of body identity integrity disorder? Yes;^{9,12} no.¹³
- Paraphilic component required? Yes;^{14,15} no.^{1,16}
- Association with non-heterosexual orientation? Yes;^{8,17} no.^{18,19}
- Key experience as a trigger? Rather yes;^{1,19} rather no.¹⁶
- Psychotherapy a successful treatment option? Yes;²⁰ no.²¹
- Surgery a successful treatment option? Yes;^{1,8,21} no.²²

motivation for their desire. But in later studies,^{15,33} another 52% indicated sexual arousal at least as their secondary motivation. Also, 45 of the original 52 participants'

reported being sexually attracted to amputees—thus 87% had paraphilic desires on top of an affliction purely of body identity disorder. More recent surveys have produced similar results; one study recruited 54 people with a disability desire on the internet and found that the proportion who reported specific sexual desires when imagining a disabled state was 90.7%.⁸ Even in the desire for hearing loss, the power of the sexual dimension was emphasised.²⁸ In a planned study of the brain morphometric correlates of disability desires, we intended to recruit participants with an exclusively asexually based amputation desire, but the recruitment (via the internet) showed similar average ratings for sexual versus non-sexual desires for amputation.⁴¹ Thus, as far as disability desires are concerned, sexual undertones are often part of the bigger picture of corporeal identity. Just how important the sexual aspect of disability desires will be, or will be reported to be, in each case, could depend on the criteria that define disability desires. Because of the inseparability of sexuality and identity, we suggest that the proposed diagnostic criteria for disability desires (appendix) should not exclude a sexual motivation.

The two latest efforts in the terminology of disability desires (appendix) tried to circumvent allusion to either the paraphilias or the concept of identity. Transableism⁴² is a sociological construct that attempts to demedicalise

disability desires and views them as a healthy person's challenge to the stigma of disability that is created by social norms. Xenomelia⁴³ can be positioned at the opposite end of a scale from social and biological determinants; it conceives of disability desires as resulting from developmental brain damage. The concept might have limitations (appendix), but work on xenomelia has produced a large body of empirical data.

Xenomelia: a neurological perspective on disability desires

Xenomelia (from Greek *ξενος* [xeno] meaning foreign; and *μελος* [melos] meaning limb) denotes estrangement from one's own limbs. The term was introduced by McGeoch and colleagues⁴³ to counter previous, theory-laden terms, particularly apotemnophilia and BIID. The authors suggested that disability desires represent a neurological disorder, specifically a focal syndrome of the right parietal lobe. Clinical neurology identifies many syndromes of various misperceptions of body parts after damage to this site of the brain, ranging from a total neglect of the left side of the body to illusory reduplications and the loss of agency and ownership, to an active aversion or hatred of left-sided limbs (appendix).⁴⁴

Several clinical observations support the concept of a neurological origin of disability desires that concern

| | Method of investigation | Size of study population (gender) | Age distribution | Age at onset | Legs:arms | Left:right: bilateral | Heterosexual: homosexual: bisexual or asexual | Right handed: non-right handed |
|--|--|---|-------------------------------|---|---|-------------------------------|---|--------------------------------|
| First, 2005 ¹ | Telephone interviews | n=52 (four women, one intersex) | Mean 48.6 years (range 23–77) | 65% before 8 years, 94% before 16 years (mean not reported) | 76%:24%* | 55%:27%:18%† | 61%:31%:7% | Not reported |
| Blanke and colleagues, 2009 ¹⁸ | Telephone interviews | n=20 (three women) | Mean 48.4 years (range 29–72) | 65% of cohort aged 3–9 years (mean 11.6) | 80%:20% | 35%:20%:45% | 90%:10%:0% | 90%:10% |
| Kasten, 2009 ³ | Postal survey using standardised personality inventories | n=9 men (one man desired paraplegia) | From early 30s to early 70s | 67% at or before 8 years (mean 8; range 4–12) | 100%:0% | 60%:20%:20% | 33%:56%:11% | Not reported |
| Johnson and colleagues, 2011 ¹⁷ | Internet questionnaire‡ | n=72 (eight women, three other) | Mean 46 years (SD 16) | Not reported | 81%:10% (n=8 arm–leg combinations) | 42%:28%:30%§ | 60%:25%:14% | 78%:22% |
| Blom and colleagues, 2012 ⁸ | Internet-administered standardised psychiatric inventories | n=54 (79.6% men; includes two with desire for blindness and two with desires for other physical disabilities) | Range 18–76 years | Mean 6.7 years (range 3–15) | 90%:7% (n=1 with desire for tetramelia) | 37%:30%:33%¶ | 56%:28%:17%¶¶ | 73%:27%¶¶ |
| Noll and Kasten, 2014 ²¹ | Internet questionnaire | n=18 (three women); all achieved the desired amputation | Range 27–73 years | Not reported | 86%:6% (n=1 arm–leg combination) | 50%:22%:28% | 76%:10%:14% | Not reported |
| Oddo and colleagues, 2014 ²³ | Individual examination, standardised personality inventories | n=15 men (data from one woman and two men not analysed; one amputated, one with insufficient data) | Mean 50 years (range 32–68) | Not reported | 100%:0% | Not reported (80% unilateral) | 40%:40%** | Not reported |

*Proportion of 50 individuals who wished for a major limb amputation (not just fingers or toes). †Proportion of 44 individuals who specified laterality. ‡Describes two surveys (total n=97, but some individuals responded to both); numbers here refer to larger sample of survey 2. §100%=all cases with an amputation desire (major limb or minor parts), but not to some cases with paraplegia desire. ¶¶Proportion of 30 individuals with a limb amputation desire (rather than paralysis desire). ||Three participants with non-amputation desires are not considered. **Bisexuality and asexuality not reported.

Table 1: Seven questionnaire studies of characteristics of the desire for amputation

limbs. First, the type of a desired disability is commonly circumscribed and developmentally stable. Thus, a person longing for a bilateral leg amputation usually abhors the idea of becoming paraplegic (and vice versa); although some cases have been reported of a more fluid symptomatology, both across body-space and time. Furthermore, left-sided limbs are far more frequently the target of an amputation desire than right-sided limbs (table 1). This left-sided preference is in line with the leading role of the right cerebral hemisphere in the representation of the bodily self and its disorders.^{44,45} The right parietal cortex also codes for left-sided and right-sided limbs, which explains why the rare switches of an amputation desire to the contralateral limb⁴⁶ do not invalidate the neurological hypothesis; similarly, a change in the type of a disability desire (eg, from amputation to paraplegia) is not necessarily evidence against a neurological cause. Commonly, a demarcation line on the limb more or less precisely indicating the site of a desired amputation is identified by the patient.

Some behavioural experiments have exploited this clear separation between what the patient considers as their own body and what is felt to be foreign. The experiments showed that tactile stimulation of the regions felt to be foreign are accompanied by an elevated response of the autonomic nervous system^{47,48} and a prioritisation in the judgment of temporal order,⁴⁹ which is indicative of a hyperattention towards incoming stimuli. Other experiments have compared the behaviour of people with xenomelia with that of patients with somatoparaphrenia. In both conditions, Romano and colleagues^{48,50} found a reduced anticipation of pain, which was specific to the disowned or non-accepted limb. Similarities (and differences) between the two conditions were also shown in an illusion paradigm that requires the integration of vision, touch, and proprioception.^{51,52} Caloric vestibular stimulation, known to transiently abolish somatoparaphrenic delusions,⁵³ did not affect the desire for amputation as assessed by questionnaire.⁵⁴ Somatoparaphrenia and xenomelia might differ along explicit variables, but share commonalities in attentional and autonomic nervous system functions. Using a conceptually distinct approach, a study of emotional processing in people with xenomelia showed a selectively reduced disgust sensitivity in response to pictures of body violations, which was interpreted as compatible with insular dysfunction.⁵⁵ The report highlights the importance of considering aspects of body representation beyond those of body schema in the narrow sense.

Neuroimaging studies have addressed brain-behaviour relationships in xenomelia more directly. One experiment examined the neural response to tactile stimulation in four individuals with a desire for leg amputation and four healthy controls.⁴³ Stimuli were taps delivered to the feet and anterior thighs, above the individual's demarcation line (and to mirrored locations

| | Disability desires | Gender dysphoria |
|---|---|---|
| Marked distress due to body morphology | Yes | Yes |
| Typical age of onset | Early (before adolescence) | Early in FtM, both early and late in MtF |
| Sex ratio (biological) | More common in men than women | More common in men than women |
| Elevated prevalence of non-right-handedness | Yes | Yes ⁵³ |
| Simulation of desired state | Frequent (pretending) | Frequent (crossdressing) |
| Sexual arousal associated with simulation | Frequent | Frequent in gynephilic MtF ¹⁴ |
| Psychotherapy an effective treatment? | No | No |
| Surgery an effective treatment? | Yes (illegally available in some non-Western countries) | Yes (legally available in many countries) |
| Secondary psychiatric disorders | Frequent | Frequent |
| Co-occurrence described? | Yes ^{1,16} | Yes ^{1,16} |

MtF=male-to-female transsexuals. FtM=female-to-male transsexuals.

Table 2: Comparison of characteristics of disability desires and gender dysphoria

on the healthy leg and on both legs of the healthy patient). Magnetoencephalographic signals to taps on the non-accepted leg compared with those on the accepted leg were reduced in the right superior parietal lobule of people with xenomelia, a reduction seen also when compared with the signals in people without xenomelia (figure 1A and B). Crucially, the diminished response of the right superior parietal lobule was independent of whether the amputation desire was targeting the left or the right leg, supporting the known bilateral body representations specifically of the right hemisphere.⁵⁶ A functional MRI experiment with five people who desired a leg amputation (three of the right leg, and two of the left), found an increased neural response to tactile stimulation on the lower leg compared with the response in healthy control participants.⁵⁷ Areas of increased activity were found in a widespread network comprising parietofrontal and occipitotemporal cortex bilaterally (stronger in the right hemisphere) and the right insula (figure 1D). Notably, this enhanced response was independent of whether the affected or the non-affected leg was stimulated. However, the difference in the response between the two legs was associated with the study groups: for xenomelia participants, but not for controls, touch on the non-accepted leg triggered a smaller premotor cortex response than that stimulated by touching the accepted legs (figure 1C). Together, these two findings indicate that xenomelia is associated with a generally higher responsivity to tactile stimulation, which might be due to an elevated attention to tactile stimuli. Ownership feelings might not primarily depend on activity in parietoinsular networks (as implied by research on somatoparaphrenia^{58,59}) but also on the premotor cortex, which has been shown to have a role in mediating ownership over body parts.⁶⁰

Structural brain correlates of xenomelia were investigated in 13 men with the desire for leg amputation

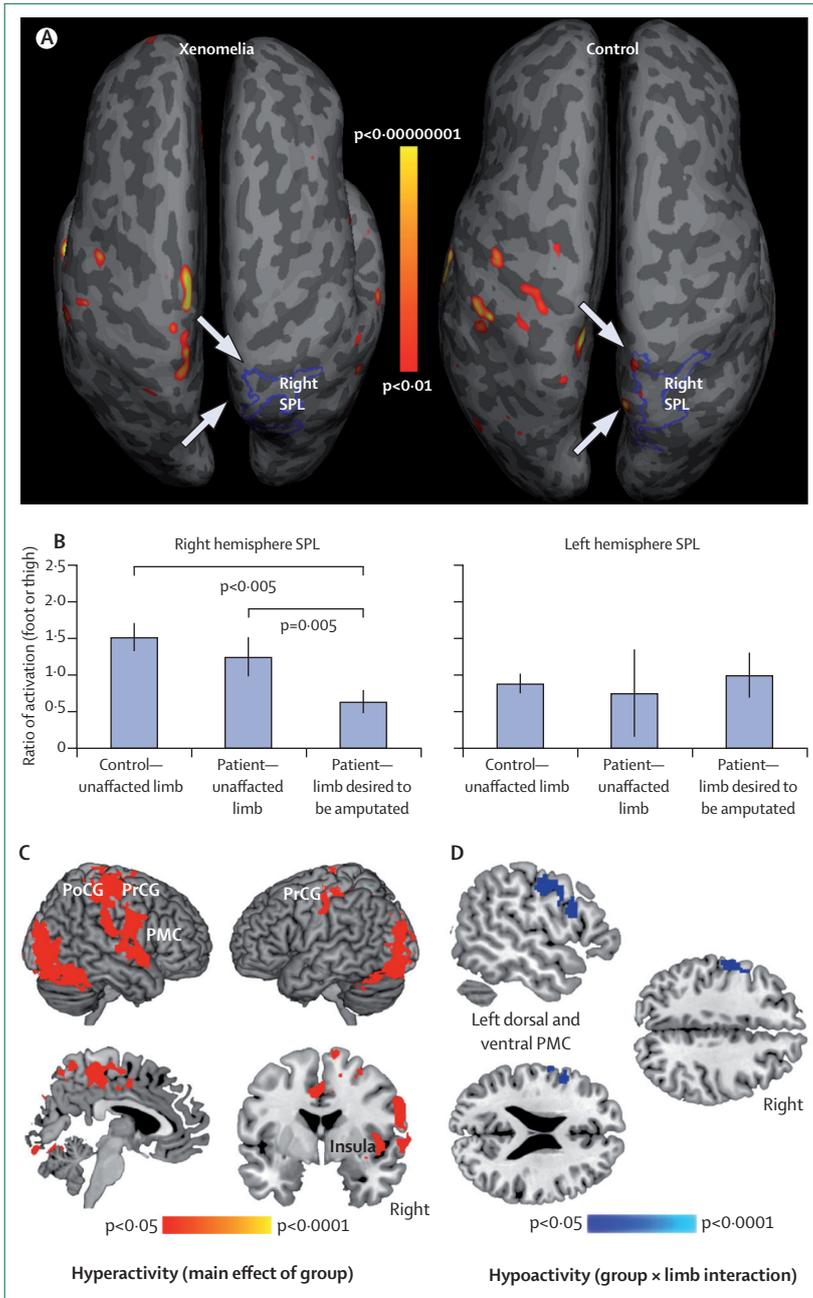


Figure 1: Functional brain correlates of xenomelia
 (A) MEG scan and (B) bar graph showing reduced responsiveness of the right SPL (blue outline) on inflated surface models of two representative participants in (A) to tactile stimulation of non-accepted parts of a leg in people with xenomelia. Bar graphs in (B) show mean and SD of the ratio between foot or thigh magnetoencephalographic activity of the SPL in four participants with xenomelia and four matched controls. (A) and (B) are adapted from McGeoch and colleagues with their permission.⁴³ (C, D) Functional MRI blood oxygen level-dependent signal to tactile stimulation of the leg in five participants with an amputation desire for one leg. (C) and (D) are reproduced from van Dijk and colleagues by permission of Public Library of Science.⁵⁷ (C) Red clusters indicate hyperactive regions in the participants with xenomelia compared with controls irrespective of the stimulated leg and comprise the right PoCG, bilateral PrCG, right PMC, and right insula. (D) Blue clusters represent hypoactive PMC in response to tactile stimulation (affected vs unaffected leg in xenomelia compared with the corresponding limbs of the controls). Colour bars show the error probability. PMC=premotor cortex. PoCG=postcentral gyrus. PrCG=precentral gyrus. SPL=superior parietal lobule.

(eight for the left leg, two for the right, three for both legs).^{61,62} Surface-based morphometry based on T1-weighted MRI showed a reduced cortical thickness of the superior parietal lobule in people with amputation desire compared with the 13 matched controls, corresponding topographically to the functional impairments previously published, and likewise it was confined to the right hemisphere.⁴³ A reduced cortical surface area was described for the right inferior parietal lobule, primary and secondary somatosensory cortex, and the right anterior insula in the men with amputation desires⁶¹ (figure 2A, left). The strength of an individual's amputation desire was inversely related to the cortical surface area in the right inferior parietal cluster (figure 2A, right). Shape analyses⁶² uncovered tissue displacements (local thinning or thickening) in the men with disability desires in several subcortical structures bilaterally, and specifically in subregions known to represent the body in a somatotopic manner or project to areas of the motor cortex (figure 2B–D).

Behavioural and neuroimaging findings in people with xenomelia have refined knowledge of the condition. The neural correlates of xenomelia involve a network of cortical and subcortical regions, comprising three key regions; the parietal lobes as the classical site of corporeal awareness,⁴⁴ the insula, known for its crucial role in the integration of body and mind,⁶³ and the premotor cortex, previously implicated in the formation of the unity of body and self.^{60,64} This network is strongly lateralised to the right hemisphere, which is compatible with the preference for left-sided body parts as targets for amputation desires and with an elevated incidence of non-right-handedness in people with disability desires (tables 1, 2).

However, the shortcomings of a conceptualisation of disability desires that are too narrowly based on physiology and neuropsychology are evident. On the one hand, all evidence for a focal dysfunction in higher-order sensorimotor circuits for limb representation has been obtained in experiments with people with an amputation desire. The present-day neuro-logical account of disability desires might thus be valid for the desires for limb amputation, but not necessarily for the desire to become paralysed, blind, or deaf. Furthermore, like the traditional concept of body integrity, xenomelia does not capture all aspects of disability desires, notably the sexual dimension. The notion of disability desires as a parietal lobe syndrome has once more exposed the gap between people adhering to an individualistic view of embodiment, and those focusing on sociological notions of a body in the world. Research on disability desires could diminish this divide between brain-based and mind-based world views by integrating neurological and sociological work on bodily representations.

Towards a social neuroscience of disability desires

To be human means to be a biosocial creature that requires description in both biological and sociological

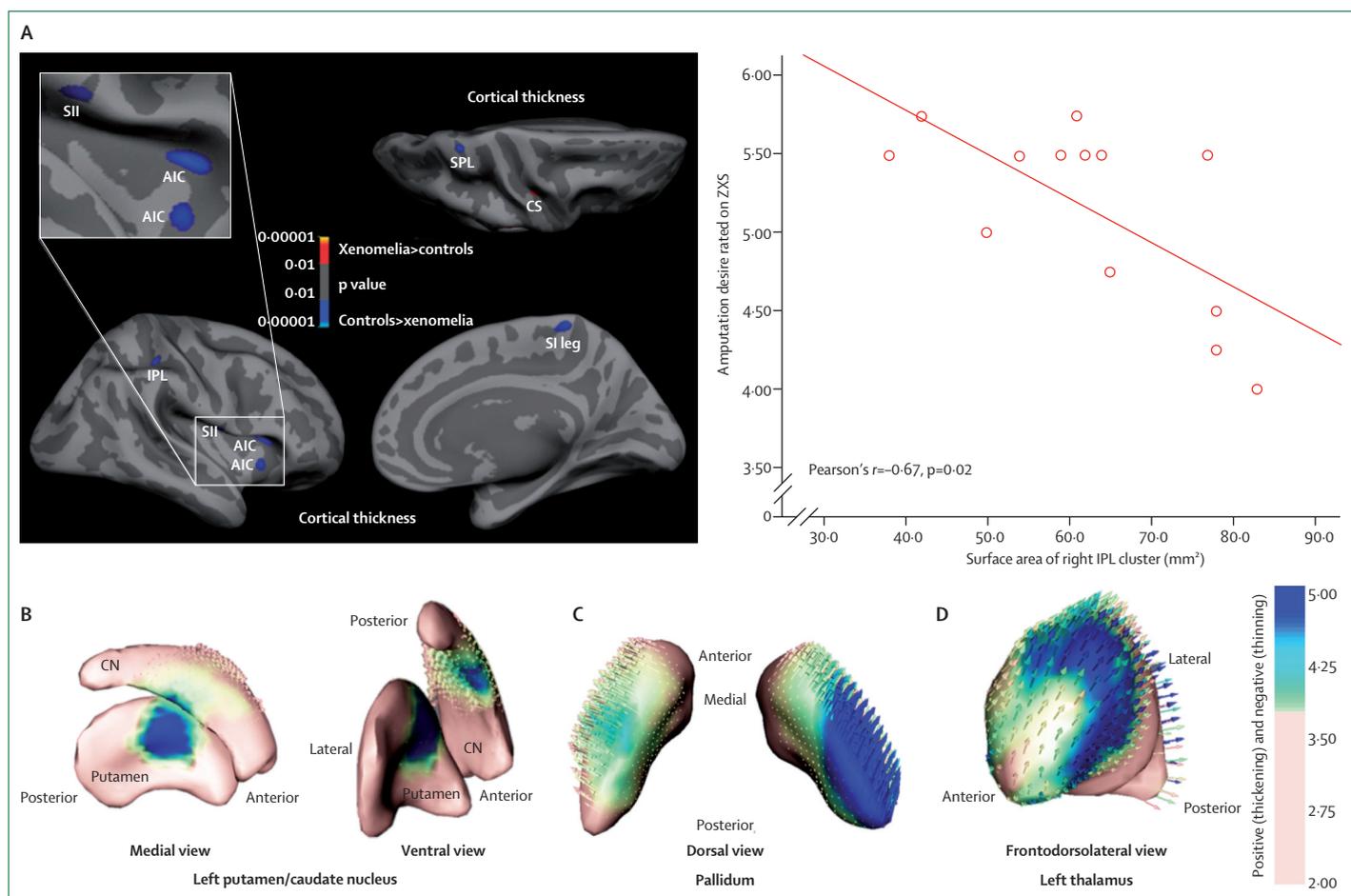


Figure 2: Structural brain correlates of xenomelia

(A) Surface-based morphometry in participants with a desire for leg amputation ($n=13$) and matched controls ($n=13$).⁶¹ Regions of reduced cortical thickness and surface area included the right SPL, the right IPL, SI, and SII, as well as the right AIC (left panel). The strength of an individual's amputation desire as rated on the ZXS⁴⁹ was inversely related to the cortical surface area within the right IPL cluster (right panel). Reproduced from Hilti and colleagues by permission of Oxford University Press. (B–D) Negative and positive tissue displacements shown by shape analyses of subcortical structures in the same participant population. Reproduced from Hänggi and colleagues by permission from Elsevier.⁶² (B) Local thinning (blue clusters) in xenomelia is evident in the left dorsomedial putamen and left ventral CN. (C) Local thickening (blue clusters) is evident mainly in the right lateral pallidum. (D) Thickening of the left anterior lateral thalamus (blue clusters). Tissue displacements in all these subcortical structures are mainly localised in subregions housing a somatotopic representation of the extremities or projecting to sensorimotor cortical regions. The colour bar represents error probability in (A) and the statistical values of the multivariate shape analysis in (B–D). The direction of the effects (thinning or thickening) is coded in the displacement vectors (small arrows; not visible in [B] due to inward direction). AIC=anterior insular cortex. CN=caudate nucleus. IPL=inferior parietal lobule. SI=primary somatosensory cortex. SII=secondary somatosensory cortex. SPL=superior parietal lobule. ZXS=Zurich Xenomelia Scale.

terms. A full understanding of disability desires will require consideration of causative forces beyond grey and white matter, and consider social matters as well. Schilder's visionary concept⁶⁵ of a neural representation of the human body as a medium between self and others allowed him to recognise that body image is a social phenomenon. He thus anticipated the now popular views of social neuroscience, such as that the ways we feel and experience our own body should be considered avenues to an understanding of how we communicate with others who, for the most part, have similar bodies.

Body image represents our corporeal awareness on the most abstract level, which allows us to assess our own appearance against aesthetic standards and cultural

norms.⁶⁶ This type of body representation is, at the same time, the most difficult to pinpoint in terms of cerebral localisation. However, work over the past two decades has demonstrated interactions between self and others at much lower levels of perceptual–motor integration. In fact, the neural prerequisites of the human mirror system reside in premotor and somatosensory cortices.⁶⁷ An experimental report about neonates' spontaneous imitation behaviour⁶⁸ has been supported by electrophysiological recordings in very young infants,⁶⁹ which showed that visually observed hand and foot actions are automatically matched in a somatotopic way to the observer's own somatosensory and motor cortices. Such hierarchically and developmentally early processing of other individuals' body shapes and movements is

important for an understanding of some xenomelia-associated behaviours. Approximately half of people with xenomelia claim that their desire for amputation was triggered by the sight of an amputee.¹⁹ Although such introspective reports are reminiscent of the concept of maternal impression⁷⁰ and might be considered secondary rationalisations, a neuropsychological basis should not be dismissed on such grounds. We have speculated⁶¹ that a hyperempathic response might predispose individuals to integration of observed bodily defects into their own body schema. This suggestion was based on the phenomenon of mirror-touch synaesthesia, observed in a minority of healthy people, indicating the feeling of touch on visual observation of others being touched.⁷¹ This form of synaesthesia is linked to psychological empathy⁷² and is especially frequent in amputees.⁷³ In the rare cases of phantom limbs in people with limb aplasia, a trigger function of the visual observation of others' limb movements has been documented.⁷⁴ The conceptualisation of xenomelia as a mirror image of aplasic phantom limbs⁷⁵ is perhaps more than a metaphor; if being born without limbs means that seeing a limb in motion can elicit the feeling of a corresponding own limb, in xenomelia, observing the absence of another person's limb could unmask a congenital underrepresentation of the person's own present limb—in the words of Robert Smith,⁷⁶ the sight of an amputee “seems to awaken an internal identity that had previously been unrecognised”.

The undeniable paraphilic component of the desire for amputation¹⁵ could be explained by the architecture of low-level body representations in the brain. When explaining the four-times greater incidence of legs over arms in amputation desires and the stronger erotic connotations

of disability desires with leg than with arm amputees, the fact that the foot and leg representations are adjacent to those of the genital organs in the postcentral gyrus might be more than a coincidence.⁷⁷ The homuncular vicinity is probably determined during foetal development, induced by the frequency of genital self-touch with the feet due to the typical position of the unborn child.⁷⁸ This positioning is the physiological explanation for the fact that touch to the face, but never to the genital region, can elicit phantom sensations in hand amputees, whereas the converse is true for foot amputees.⁷⁹ A large body of empirical work exists on the specificity of these referred sensations.⁸⁰ Furthermore, the insula as a hub for the integration of body and mind⁸³ and, more specifically, of somatosensation and sexual arousal,⁸¹ is adjacent to the secondary somatosensory cortex for leg representation. This arrangement might explain its repeated inclusion among the hotspots of functional⁴⁷ and structural⁶¹ cerebral abnormalities in people with xenomelia. De Preester¹⁵ localised a sexual schema in conceptual proximity to an insularparietofrontal system that merges somatosensation, erotic arousal, and visual observation of conspecifics. Specifically, she considered it conceivable that the observation of an amputee during early development could lead to a dramatic explication of the sexual schema into the body image.¹⁵ Clearly, prospective research is needed to substantiate or refute such proposals. Paraphilic components are largely neglected by both the advocates of brain-based approaches and the supporters of mind-based approaches to disability desires. These paraphilic components should be incorporated into future empirical research (panel 3).

The thoughts presented in this section are speculations, and they might inform people who work on the cognitive neuroscience of bodily self-consciousness as it relates to aspects of social life.^{45,83} However, the speculations are not directly helpful to the people with disability desires or to the clinicians who aim to treat them. The question arises: do we need to treat disability desires at all? Are they pathological desires or do they merely reflect a normal urge of a minority for a continual testing of the flexibility of their bodily self?⁶⁵

Disability desires: divergence or disorder?

Detailed diagnostic criteria for disability desires have been catalogued by First^{9,16} (appendix), who coined the term BIID and is hopeful that the condition will eventually be included in a future edition of the DSM. This opinion is shared by the British surgeon Smith⁷⁶ who has operated on several people with an amputation desire, and he proposed similar criteria before.²⁴ In stark opposition to this attempt to provide disability desires an official medical status, are the individuals who consider them “a new way to be mad”,⁸⁴ “a contemporary frame for psychological suffering”,⁸⁵ or “the first psychiatric disease manufactured in cyberspace”.⁸⁶ People with these views argue that the internet is a

Panel 3: Future directions

Empirical research

- Should expand to include variants of disability desires apart from xenomelia (eg, include people who desire sensory impairments)
- Should expand to include people with gender dysphoria, anorexia, and associated disturbances of bodily experience
- Should move on from investigations of an individual's body schema to probing higher-order processes of body representation that link a person to society (eg, mimicry, sexuality, and empathy)
- Should plan crosscultural studies
- Should consider data sharing in the case of neuroimaging investigations akin to requirements proposed for clinical trials⁸²

Nosology

- Should consider labelling disability desires body dysphoria and further examine the conceptual vicinity to gender dysphoria and the bodily dysphorias associated with eating disorders

| | Arguments in favour of elective disability | Arguments against elective disability |
|--|--|---|
| <p>Respect for autonomy: Decision-making capacities of autonomous people should be respected; individuals should be allowed to make reasoned informed choices Major issue: are the conditions for respecting the autonomy of the patient met?</p> | <p>The conditions for respecting the autonomy of the patients are fulfilled in at least some patients:</p> <ul style="list-style-type: none"> • The decision is informed and the patients have a reasonable understanding of what it is like to have the desired condition (expressed, for example, by pretending) • Patients have decision-making capacity; it's not the decision that needs to be rational but the thought process that cumulates in the decision • Degree of pressure from non-rational considerations (looping effect) is similar to pressure (advertising, gender norms) in other types of interventions like plastic surgery in which autonomy is granted to the patient • The choice is among the medically reasonable alternatives <p>Patients who fulfil these conditions can rely on the principle of respecting autonomy; this would be the case particularly for certain cases of limb amputation^{76,90,91}</p> | <p>Patients can be referred on the principle of respecting autonomy if the conditions in the left column are met, but these arguments question fulfilment of those conditions:</p> <ul style="list-style-type: none"> • Patients don't have crucial knowledge on how having the desired disability would be and so their consent cannot be considered informed • Having an irrational wish puts into question that the patients have decision-making capacity with respect to this desire • Indications that a disability desire is a psychiatric disease manufactured in cyberspace exist • Alternatives (psychotherapy, medication) exist that are much less invasive; research should be advanced instead of allowing an irreversible intervention <p>The main aim of autonomy is to protect patients from unwanted interventions, particularly in cases in which the surgeon could face criminal liability.⁹²⁻⁹⁴ Even if the principle of respecting autonomy is granted, this does not justify requesting an intervention</p> |
| <p>Non-maleficence: Causing harm to the patient should be avoided (also with respect to other involved people); harm should not outweigh the benefits of treatment Major issue: how severe is the harm caused by disability?</p> | <p>The (psychological) harm of suffering from a disability desire is worse than the (physiological) harm of having a disability. Physical disability need not result in substantially lower life satisfaction, even for people who did not choose to become disabled. Elective disability will prevent people with disability desires from taking matters into their own hands, with potentially disastrous consequences, particularly in cases of amputation^{33,95}</p> | <p>Disabilities are paradigmatic cases of harm, particularly cases of sensory disability (blindness or deafness). For example, amputations have great risks and often have severe consequences such as infection, thrombosis, paralysis, necrosis, or phantom pain. The history of medicine is filled with surgical treatments for psychiatric problems; many of which have been misguided. Only in extreme cases with high risk of self injury should elective amputations be an option^{93,94}</p> |
| <p>Beneficence: The aim should be the benefit of the patient; positive steps should be taken to prevent and remove harm to the patient Major issue: how credible are reports that elective disabilities increase the wellbeing of patients?</p> | <p>The desire for a disability in some patients is a set of stable values that are crucial for the person; to approve of the person's decision is to respect his or her bodily integrity. With regards to amputation, patients who have amputations because of ischaemia can gain enormous relief from their symptoms, and an amputation in a patient with a disability desire is equally relevant for their wellbeing^{76,96}</p> | <p>The scientific evidence gained so far is questionable with respect to the effectiveness of amputations for the wellbeing of patients with disability desire. The number of cases is too low and mechanisms of self-deception could be expected in those rare cases. After having invested enormous emotional resources in getting a procedure that is not only irreversible, but also something that they have always seen as the only possible solution to their problems, some people might find it difficult to admit that it has been a mistake^{93,94}</p> |
| <p>Justice: Patients in similar positions should be treated in a similar manner (individual level); benefits, risks, and costs should be distributed fairly (societal level) Major issue: how plausible are analogies with respect to fairness on the individual and societal level?</p> | <p>At the individual level, an analogy can be made to sex reassignment surgery, which involves irreversibly losing one's capacity for reproduction. This surgery has been accepted as appropriate for treating gender identity disorder. At the societal level, those who achieve amputation become more productive, happy, and contributing members of the community and no longer consume psychiatric resources. This improvement counteracts the resources needed for administering interventions and the support requirements that a functional handicap involves^{76,97}</p> | <p>Analogies of disability desires are made with other examples in which demanded (or refused) interventions lead to body modifications or even harm are not sufficiently supported—ie, the harm caused by amputation exceeds the harm of other types of body modifications. Societal costs (eg, medical treatment, rehabilitation, and early retirement) are high. Generally, the empirical data for assessing the potential economic benefit of elective disability is poor and unlikely to be positive. Justice considerations with respect to resource allocation should focus on research of the causes of disability desires to find alternative, less invasive treatment options^{94,98}</p> |

Table 3: Pros and cons in the ethical debate about elective disability

platform that popularises transient mental illnesses,⁸⁷ whose particular symptom constellation is in steady flux. This constellation is shaped by a continuous exchange between people with these desires who share their experiences in organised chats and the policymakers who compile and classify the reported symptoms.

Both DSM and ICD are endeavours in taxonomy that, far from categorising illnesses in an objective and neutral way, actively create, shape, and eliminate specific conditions. A case in point is homosexuality, which was described as a “sociopathic personality disturbance” in DSM-I, a “sexual deviation” in DSM-II, and then no longer a disorder from DSM-III onwards.⁸⁸ People with disability desires are increasingly lobbying that their condition should be listed, partly to promote the availability of surgical treatment. Given the massive distress accompanying disability desires, one might

wonder whether BIID would not better be read as BD, for body dysphoria (panel 3). Those who feel an urge to use a wheelchair or crutches or to wear special lenses that prevent light penetrating their eyes could do so, but their behaviour would not be considered pathological unless accompanied by considerable distress.

Another question asks whether conformance to disability desires can ever be ethically justified. This discussion has unfolded in connection with elective amputations offered to people with xenomelia in some countries. The framework of bioethical principlism⁸⁹ indicates that the controversy is mainly about empirical issues; for instance, whether the conditions for claiming respect for autonomy are fulfilled in patients with disability desires or whether evidence for therapeutic success of elective amputations is sufficient (table 3). Successful psychotherapy of disability desires is rarely reported²⁰ and probably reduces associated

symptoms such as depression, but not disability desires per se.⁹⁹ By contrast, reports about amputations in the case of xenomelia describe an immediate and lasting alleviation of chronic suffering,^{1,21,100} although exceptions have also been reported.²² The largest case series of surgically remedied disability desires comprises 21 people, 18 having had a major amputation during the previous 1–16 years.²¹ In all cases, quality of life was rated to be substantially increased, and no new disability desire emerged after surgery. Furthermore, evidence shows that competence for autonomous decision making cannot be denied for people with xenomelia.⁷⁶ In summary, considerable support exists for the view that elective amputations can be ethically justified in some cases, even if long-term effects of the intervention still need to be assessed. A consensus paper is needed that is authored by bioethicists, psychiatrists, and surgeons. Smith⁷⁶ specifically asks for regulations that guarantee protection from legal sanctions for surgeons who agree to undertake these procedures.

Conclusion

Disability desires represent a person's dissatisfaction, since early childhood, with what most people consider an able-bodied state. This dissatisfaction can be so disabling that conceptualisation of the condition as a mental disorder is justified. In the case of desired limb amputation, there is evidence for an altered cerebral architecture accompanying the disability desires. Yet, biological mediation does not imply biological cause.¹⁰¹ Bodily self-consciousness is subject to powerful sociocultural influences, which also does not imply that disability desires have primarily societal roots.⁸³ Future research should explore the intersections between neural and psychological levels of analysis and promote an ethnologically, sociologically, and neuropsychologically informed perspective on disability desires. The arguments about the ethical concerns of elective amputations yield to strong counterargument. However, whether cutting the body will ever cure the mind¹⁰² will remain uncertain in the discourse about disability desires and related conditions of body dysphoria.

Search strategy and selection criteria

This Review is based on material from the authors' personal files and the references in the contained articles. A combination of the search terms "body integrity", "identity", "disability", "amputation", "paraphilia", "xenomelia" and the terms listed in table 1, appendix, produced more items with the aid of PubMed, the Web of Science, and Scopus (last accessed May 17, 2016). Selection of the references finally included was based on an article's quality and originality and its usefulness to aid argumentation. We considered articles in English, French, Italian, Dutch, and German.

Contributors

All authors contributed to the compilation and selection of work included in this Review. PB drafted a first version of the Review, and all authors contributed to editing the text and preparing figures, tables, and panels. All authors have approved the final version.

Declaration of interests

We declare no competing interests.

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