Body ownership

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TITLE: Body ownership: When feeling and knowing diverge.

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RUNNING TITLE: Pain anticipation in BIID

Abstract

Individuals with the peculiar disturbance of '*overcompleteness*' experience an intense desire to amputate one of their healthy limbs, describing a sense of disownership for it (Body Integrity Identity Disorder - BIID). This condition is similar to somatoparaphrenia, the acquired delusion that one's own limb belongs to someone else. In ten individuals with BIID, we measured skin conductance response to noxious stimuli, delivered to the accepted and non-accepted limb, touching the body part or simulating the contact (stimuli approach the body without contacting it), hypothesizing that these individuals have responses like somatoparaphrenic patients, who previously showed reduced pain anticipation, when the threat was directed to the disowned limb. We found reduced anticipatory response to stimuli approaching, but not contacting, the unwanted limb. Conversely, stimuli contacting the non-accepted body-part, induced stronger SCR than those contacting the healthy parts, suggesting that feeling of ownership is critically related to a proper processing of incoming threats.

1.Introduction

Body representation is our body mapped in the brain (Head & Holmes, 1911). There are several distinct body representations, some of them responsible for processing primary sensory inputs, some others for controlling motor outputs (Zeharia, Hertz, Flash, & Amedi, 2012). Beyond this first level of processing, various supplementary representations of a higher cognitive order have been proposed, supposedly involved in complex behaviours (de Vignemont, 2011). The sense of ownership corresponds to the awareness of one's body as belonging to one's self, and the feeling that a given body part belongs to one's own body (de Vignemont, 2011). This sense of ownership is seemingly derived from a specific type of body representation. Ownership over body parts constitutes one of the prerequisites for the kind of embodied self-consciousness ordinary humans tacitly take for granted.

Individuals with Body Integrity Identity Disorder − BIID − report a highly disturbed sense of ownership for one or more of their limbs (Sedda, 2011). Paradoxically, they experience themselves as "incomplete", as long as they continuously feel the presence of the limb they do not accept as a part of their bodily self. Consequently, these people develop an intense desire to get rid of the particular limb, either physically ("desire for amputation", First, 2005) or functionally ("desire for paraplegia", Giummarra, Bradshaw, Hilti, Nicholls, & Brugger, 2012). This peculiar condition was originally ascribed to paraphilias and labelled as apotemnophilia ("love for amputation" Money, Jobaris, & Furth, 1977). In 2005, the first survey on a large group of individuals desiring amputation (n=42) lead to a change in nomenclature from apotemnophilia, which implied a sexual connotation, to the term BIID and the concept of body identity. The new focus on "identity" rather than sexuality was clearly inspired by work on gender dysphoria, then labelled "gender identity disorder", GID. Still more recently, the nosologically more neutral label "xenomelia" (McGeoch et al., 2011) was proposed, emphasizing the feelings of alienation, or disownership mentioned by individuals with BIID. This semantic path highlights the shift from considering only the psychiatric components of the condition to the inclusion of neurological correlates more related to the cerebral

representation of the body. In this paper, we will adopt the more descriptive label "amputation-desire" to avoid biasing the reader toward one particular etiological direction. A dysfunctional activity of the right parietal lobe has been proposed as a neural correlate for the amputation-desire (McGeoch et al., 2011). The authors examined three individuals with xenomelia and found a reduced responsivity of the right superior parietal lobule (SPL) for tactile stimulation of the affected limb, as compared to its healthy counterpart or the limb of control participants. Interestingly, another locus of altered brain activity was the insula, an area traditionally associated with higher-order body representations (Berlucchi & Aglioti, 2010). In accordance with these functional correlates of amputation-desire, Hilti et al. (2013) recently demonstrated structural differences in the right SPL, the right primary and secondary somatosensory cortices, and the anterior insula when comparing individuals with amputation-desire with control subjects (Hilti et al., 2013). Another recent study, adopting a functional paradigm, found an involvement of the premotor cortex in individuals desiring amputation (van Dijk et al., 2013).

One's sense of ownership is critically related to pain anticipation and the processing of incipient threat (Ehrsson, Wiech, Weiskopf, Dolan, & Passingham, 2007), which is relevant for adaptive purposes, as the recognition and avoidance of danger. It has been show that experiencing a sense of ownership toward an alien hand is related to the emotional reaction when that hand is threatened both in healthy participants under bodily illusions (Armel & Ramachandran 2003; Ehrsson, et al., 2007), and in patients experiencing pathological embodiment for alien hands (Garbarini, Fornia, Fossataro, Pia, Gindri, & Berti, 2014a). Early research in anosognosia, the non-recognition of one's own illness, revealed a basic disturbance in the "circuits for danger recognition" suggesting that being aware of one's own body is a critical function for adaptive behaviour (Vocat & Vuilleumier, 2010). "Personification anosognosia" (Critchley, 1955) is a particular form of anosognosia, relevant in the context of the sense of ownership, where a patient with hemiplegia, unaware of the paralysis, claims that the affected limb belongs to another person. This disorder, not uncommon in the initial phases of a right hemisphere stroke, is now better known as "somatoparaphrenia" (Gerstmann,

1942; Invernizzi et al., 2012; Romano, Gandola, Bottini, & Maravita, 2014a; Vallar & Ronchi, 2009).

Recently, it has been shown that Skin Conductance Responses (SCR) to sensory threats approaching the body are reduced in somatoparaphrenic patients, supporting the idea that a detachment of the affected body part from the patient's body representation also modulates pain anticipation (Romano et al., 2014a). While a preliminary exploration of pain perception in two individuals with amputation-desire showed increased SCRs for noxious stimuli contacting the unwanted limb (Brang, McGeoch, & Ramachandran, 2008), no data on pain anticipation are available yet. A conceptual similarity between somatoparaphrenia and the disturbance of 'overcompleteness' has been previously proposed based on the disownership sensations associated with both conditions (Berti, 2013; Brang et al., 2008; Lenggenhager, Hilti, Palla, Macauda, & Brugger, 2014). However, there is also a fundamental theoretical difference between the two disorders. De Vignemont recently proposed that the ownership experience could be divided in the feeling of ownership and the judgment of ownership. The hypothesis is that our judgments of ownership, which we have on our biological body parts under normal conditions, are based on a primitive non-conceptual feeling of ownership (de Vignemont 2011). We propose that a critical difference is that individuals with amputation-desire lack the *feeling* of ownership but not the *cognitive appreciation* that these are their own limbs.[1] On the other side, in somatoparaphrenia, patients are delusional and lack both the ownership feeling and the ownership judgment. Amputation-desire is also reminiscent of another peculiar disturbance of body awareness, namely misoplegia. This latter condition is defined as a morbid dislike or hatred toward paralyzed limbs in patients with hemiplegia (Critchley, 1955; 1974) that typically presents with somatoparaphrenia, even if double dissociations are on record (Loetscher, Regard, & Brugger, 2006). Despite amputation-desire and misoplegia share aggressive desires toward one's own limb, an important

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[1] In the original 2011 paper de Vignemont proposed that the desire for amputation was a matter of judgment of disownership with preserved feeling of disownership, the opposite of our view expressed here. However, she herself recently revised this distinction, entertaining an interpretation of the disorder in terms of an absent ownership feeling with preserved ownership judgments (de Vignemont, Australasian Journal of Philosophy, 2014 [e-pub ahead-of-print]).

difference should be acknowledged. First, only a few cases of misoplegia are well documented in the literature (Loetscher, et al. 2006). Thus, there is a profound need for more experimental data to precisely determine behavioural manifestations and neural underpinnings of misoplegia. However it is possible to observe that individuals with amputation-desire typically present with very precise characteristics of the body part to amputee, including a sharp line from where they do not experience the limb as a part of their body desiring its removal. Less is known for individuals with misoplegia, where aggressive behaviour toward the impaired limb seem to be more generalized ranging from verbal aggression to physical actions generally targeting the impaired limb as a whole. Moreover, these behaviors seem to be related to a general impairment of emotional processing such as anosodiaphoria, impulsive behavior, and other emotional disturbance (Loetscher, et al. 2006).

Thus amputation-desire shares some features with acquired impairments of body awareness, but is at the same time unique in that it reflects a lack of feeling of ownership accompanied by a normal judgment of ownership.

In this study we compared SCR to either noxious or neutral stimuli in individuals with desire for a limb amputation, testing whether the denial of ownership, occurring at the primitive level of the feeling of ownership, is so deep to impact autonomic responses to incoming threatening stimuli. Crucially, stimuli were delivered in two conditions: in the real contact condition stimuli touched the body part; in the simulated contact condition, stimuli only approached the body segment, avoiding a proper contact. This latter condition assesses the anticipatory response uncontaminated by tactile-sensory processing, and we hypothesized that, determined by a body representation dysfunction, we would observe a SCR reduction for the unwanted limb, mirroring previous findings in patients with somatoparaphrenia (Romano et al., 2014a). Contrarily, in the real contact condition we expected an increased SCR for painful stimuli on the limb sought for amputation (Brang et al., 2008). Importantly it has been shown that the processing of noxious stimuli does not change in psychiatric conditions, as measured for pain threshold in obsessive compulsive disorder (Greenspan et al., 2008) and in schizophrenia, where patients undergoing skin conductance coupled with a fear

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conditioning paradigm show no differences from healthy controls in the electrodermal signal, even though at brain level insular reactivity was diminished (Linnman, Coombs, Goff, & Holt, 2013). Thus, the hypothesized selective reduction for anticipation coupled with an increased reaction for real painful stimuli would add more evidence in favour of a selective impairment in body representation and a differentiation with known psychiatric conditions.

2.Materials and Methods

2.1.Subjects

Ten individuals with amputation-desire (mean age=42 ±9.6(StDev), 1 female; see Tab.1) were recruited to participate in the study. Participants were selected based on a preliminary face-to-face interview with one of the authors in Zurich. During this first contact an extensive psychiatric assessment, including a structured interview and validated self-report questionnaires (Hilti et al., 2013) excluded the presence of axis I disorders. In each person, BIID characteristics were confirmed (Ryan, Shaw, & Harris, 2010) and different dimensions were further quantified as in Aoyama, Krummenacher, Palla, Hilti, & Brugger (2012). All individuals of the Zurich study were made aware of the present experiment in Milan, and those volunteering to travel there were administered a semi-structured interview modelled after the SCID-II preliminary questionnaire (First, Gibbon, Spitzer, Williams, & Benjamin, 1994). This interview contains questions concerning family (i.e., whether parents and relatives know about the condition), psychotherapy treatment, mood disorders, and obsessions concerning the desire and sexual fantasies (the same instrument was previously used in Bottini, Brugger, & Sedda, 2014).

Most of our volunteers presented with dysphoria specifically related to the amputation desire. None of the participants attributed their desire to a primary sexual cause, even though some presented with a sexual attraction toward amputees. All subjects reported their amputation desire as a constant component of their life, manifesting since childhood (men reported the desire starting around age 6–7, the female individual at age 3–4). On average all of them spend most of the day thinking about

the amputation-desire. Only two participants are under drug treatment, and only one is under psychotherapy (see Tab.1).

Ownership of the unwanted body parts was investigated through the semi-structured interview performed in Milan. All participants reported that they felt the limb desired for amputation as not being part of their ideal body or body representation. Testing took place at the Cognitive Neuropsychology Centre at the Niguarda Ca' Granda Hospital in Milan, whose ethical committee had approved the study. The experiment was conducted according to the principles of the Declaration of Helsinki and all subjects gave informed written consent before participating.

[insert tab 1 about here]

2.2.Stimuli

Sixty-four mechanical stimuli were delivered in a single session, and the SCR was recorded simultaneously. Two types of stimuli were used: noxious (needle with a blunt end) and neutral (cotton swab) (Cheng et al., 2007; Romano et al., 2014a; Romano & Maravita, 2014b). All participants flawlessly distinguished the needle from the cotton swab by both visual and tactile inspection. The entire session took around 30 minutes.

Participants precisely indicated the line of the desired amputation. This allowed us to determine two sites of stimulation, i.e. one well below that line and one at a symmetric location on the other, accepted limb. The two patients desiring arm amputation indicated the elbow, thus stimuli were delivered on the dorsal surface of the hands. The other eight individuals, seeking a leg amputation, indicated a line at the level of the knee or above. In these cases we stimulated the lateral surface of the calves.

In the "real" condition the examiner touched the body either with the needle and the cotton swab for about 0.5 seconds. In the simulated condition the needle or cotton swab only approached the body remaining at a distance of approximately half a centimetre from the skin for about half a second before being retracted (Romano et al., 2014a; Romano & Maravita, 2014b).

Stimuli were delivered to the right and left limb. Overall, the stimulation conditions were: Painful Real Right, Painful Real Left, Painful Simulated Right, Painful Simulated Left, Neutral Real Right, Neutral Real Left, Neutral Simulated Right, Neutral Simulated Left. The stimuli were divided into eight independent blocks of eight stimuli each (one stimulus per condition in each block), the sequence of the stimuli in each block was randomized. Thus stimulus presentation was balanced in such a way that habituation, typically occurring with SCR paradigms, could not affect differences between conditions.

2.3.Setting and procedure

Participants were comfortably reclined on a medical bed (in the case of desired leg amputation) or sitting on a chair in front of a table (in the case of desired arm amputation). They were asked to relax remaining as stationary as possible, breathing regularly, while gazing at the point where the stimuli emerged (either from below the bed or from below the table surface).

On each trial a stimulus was presented by the experimenter, who was trained to use the same trajectory on each stimulation, unpredictably approaching one of the subject's limbs. Participants were instructed to fixate the stimulus for the whole trajectory.

In order to check whether stimuli were perceived correctly on both sides, and to be sure that needle stimulation was critically evaluated as painful, we collected an evaluation of tactile and painful sensations at the end of the experiment. We used a verbal scale ranging from 1 (not painful) to 10 (worst pain experienced). All participants were asked about the unpleasantness of the stimulation for both stimuli and sides. All the volunteers reported a value of 1 for the q-type touch on both limbs and values larger than 3 for the needle (unwanted limb: 3.8 (average) ±1.3 (St.Dev); healthy limb: 4.1 ±1.4; see Tab.1 for individual scores), suggesting that the stimulation was perceived as different from the cotton swab touch, and critically as a painful contact. We did not collect a trial by trial evaluation of pain in order to reduce artefacts on the SCR signal that usually occur because of deep breathing, speaking, and movements.

2.4. SCR apparatus

SCR was recorded through a SC-2701 biosignal amplifier (Bioderm, UFI, Morro Bay, California) connected to a PC through a serial port. The gain parameter was set at 10 μmho/V; the signal was sampled at 10 Hz. The signal was acquired by means of two silver electrodes (1081 FG Skin Conductance Electrode) placed on the first phalanx of the index and ring fingers of the right hand for five volunteers and of the left hand for the remaining five. A saline conductive paste was applied to the electrodes to improve signal-to-noise ratio. Data were digitalized using the SC-2701 software with a resolution of 12 bits.

2.5. Data pre-processing

The peak-to-base measure was computed for each trial as the difference between the maximum value detected in 6-second post-stimulus and the baseline calculated as the average value of a 0.3 seconds pre-stimulus (Romano et al., 2014a).

Triggers coding for the stimulus type were manually sent to the SCR trace through the computer keyboard at the moment when the stimulus became visible to the participants.

The peak-to-base measures were then normalized within subject and converted to Z-scores (Rhudy, McCabe, & Williams, 2007; Romano, Pfeiffer, Maravita, & Blanke, 2014c), to reduce the effect of the inter-subject variability of SCR, which is commonly large and also to reduce the effect of stimulations on different body districts in different participants.

2.6. Data analysis

Data were analyzed with SPSS 21 (IBM® SPSS® Chicago, Illinois). A 2*2*2 repeated measures ANOVA was used on SCR data, factoring: Stimulus (painful/neutral), Contact (real/simulated), and Side (unwanted limb/healthy limb) as within subject factors. Achieved power and effect size, measured with the partial eta squared (η^2) were computed with G*Power 3.1 (http://www.psycho.uni-duesseldorf.de/abteilungen/aap/gpower3/). Significant interactions were

explored by looking at the confidence intervals (CIs) - i.e., average ± standard error of the mean (SEM) * t-critic (t-distribution value for the level of confidence set) (Cohen, 1990; Cumming, 2011; Romano et al., 2014a) - setting at 90% the confidence level. It is worth noting that CIs show the range of probability in which data are distributed in a given condition representing a reliable method to graphically explore significant interactions (Cumming, 2011, 2014; Masson & Loftus, 2003; Romano et al., 2014a).

3.Results

The ANOVA showed a significant main effect of Stimulus ($F_{(1,9)}$= 51.432 $p< 0.001$, η^2= 0.851, power> 0.999; painful= 0.358 (average z-transformed SCR) ±0.05 (Standard Error), neutral= -0.358 ±0.05), and Contact ($F_{(1,9)}$= $p\leq 0.05$, η^2= 0.386, power= 0.565; real= 0.155 ±0.06, simulated= -0.155 ±0.06). The analysis also revealed a significant interaction between Stimulus and Contact ($F_{(1,9)}$= 10.135 $p\leq 0.01$, η^2= 0.530, power= 0.99; painful real= 0.66 ±0.11, painful simulated= 0.06 ±0.12, neutral real= -0.345 ±0.08, neutral simulated= -0.371 ±0.05) and, critical to our purpose, an interaction between Contact and Side ($F_{(1,9)}$= 5.132 $p\leq 0.05$, η^2= 0.363, power= 0.878; real to-be-removed= 0.226 ±0.08, real healthy= 0.084 ±0.81, simulated to-be-removed= -0.233 ±0.07, simulated healthy= -0.077 ±0.08). There was nor a significant main effect of Side ($F_{(1,9)}$= 0.115 $p= 0.905$, η^2= 0.002, power=0.051), neither any other interaction (stimulus*side: F=0.091, p=0.77, η^2= 0.010, power= 0.058; stimulus*contact*side: F= 3.603, p=0.09, η^2= 0.286, power= 0.396).

CIs of the Stimulus by Contact interaction showed stronger SCR for the painful real condition [lower limit at 90% level= 0.399; upper limit at 90% level= 0.911], reflecting global pain experience, than for the painful simulated condition [-0.200; 0.323], which assessed pain anticipation. Both painful real and painful simulated stimulations showed stronger responses than neutral real [-0.516; -0.173] and neutral simulated [-0.493; -0.250] stimulations.

The observation of CIs on the critical significant interaction Side by Contact showed that simulated conditions induced larger SCR on the healthy [-0.233; 0.079] than on the unwanted limb [-0.361; -0.105]. Conversely, when the stimuli actually touched the limb, the SCR was stronger on the unwanted [0.87; 0.364] than on the healthy side [-0.064; 0.232] (Fig.1).

[Insert Fig.1 about here]

4.Discussion

The desire for amputation is an apparently paradoxical condition where a person's body representation seems 'overcomplete' (Brugger, Lenggenhager, & Giummarra, 2013), such as one's true bodily self appears to require amputation (or functional impairment).

We explored whether the feeling of ownership is relevant for the fundamental adaptive behaviour of pain anticipation in ten individuals with an amputation-desire of one single own limb. Our hypothesis was that individuals with amputation-desire should show similar responses to those described for patients with somatoparaphrenia, as both conditions feature an underrepresentation of a body part (Berti, 2013). More specifically, we predicted that a compromised feeling of ownership would be detectable both in individuals with amputation-desire and somatoparaphrenic patients. A critical difference between these two conditions, however, is that individuals with amputation-desire lack the feeling but not the judgment of ownership, while somatoparaphrenic patients lack both the ownership feeling and the ownership judgment. Our experiment exploring SCR responses in individuals with amputation-desire contributes to the understanding of the level of body awareness related to pain anticipation: the primitive level of the feeling, or, at the more cognitive level, that of judgment. Importantly, the same paradigm has been previously applied to patients with somatoparaphrenia showing a lack of anticipatory responses only when noxious stimuli approached the disowned limb (Romano et al., 2014a). In contrast, patients affected by a "pure" psychiatric condition showed normal pain threshold and normal skin conductance responses for anticipatory responses (Linnman et al., 2013). Therefore, we predicted that individuals who seek the amputation of a limb would likewise show a reduced anticipatory response to threatening stimuli approaching

the unwanted limb if the condition is caused by a selective impairment of body representation paralleling somatoparaphrenia. The confirmation of such prediction suggests that what is critically related to the anticipation of incoming noxious stimulation is the primitive feeling of ownership, rather than the explicit judgment of ownership.

In accordance with this prediction we found a decreased SCR to stimuli approaching, but not contacting, the unwanted limb as compared to stimuli directed toward the healthy limb. Conversely, when stimuli actually touched the underrepresented limb, the SCR was stronger than those for stimuli contacting the healthy counterpart. Such increased arousal by contact was previously observed and generally interpreted as an altered processing of sensory stimuli (Brang et al., 2008). An alternative hypothesis assumes an increased attentional state, reflected by higher SCR, directed toward the unwanted limb. This hypothesis was supported by findings from an experiment on spatio-temporal integration judgements, which showed that individuals with amputation-desire have an exaggerated "tactile attention" toward the unwanted body part (Aoyama et al., 2012). However, such an explanation appears contradicted by the observed absence of pain anticipation, as one would predict higher SCR also in the case of simulated touch. Reasoning in terms of a disruption of higher-order body representations, still another alternative hypothesis is the following: the desire for limb amputation is not related to an elementary deficit of somatosensory perception. It seems that only to the point of the tactile/painful stimulus a solicitation of the arousal system occurs. This observation suggests that the limb desired for amputation is not properly inscribed into the central representation of the body as a whole. If this is the case, such an under-representation might induce a scarce attention for any signal coming from the environment directed to the limb felt as outside from the body representation, even in an experimental setting where participants are explicitly invited to take care of the incoming stimuli. Thus, individuals with amputation-desire might not properly anticipate pain on the unwanted side, however they could still perceive it once the noxious stimulation contacts the limb. This would turn into an unexpected stimulation that is known to

induce stronger pain sensation and physiological reactions than the expected stimulations (Brown, Seymour, Boyle, El-Deredy, & Jones 2008a; Brown, Seymour, El-Deredy, & Jones 2008b).

The primary feeling of ownership has been defined as primitive, exclusive sensation for biological body parts on which judgments of ownership are typically based (de Vignemont, 2011).

Our results suggest that the amputation-desire is associated with a selective disruption of the primitive biological component. Albeit involving only a specific body part, whose primary sensory and motor functions are preserved, this disruption is so profound as to inhibit the anticipatory physiological responses to incoming threats. This is not the case in complex psychiatric conditions, such as schizophrenia, where electrodermal signals evoked by pain anticipation are preserved (Linnman et al., 2013).

Our results support the parallelism between amputation-desire and somatoparaphrenia. However, it is worth to highlight that in contrast to patients with somatoparaphrenia, individuals with amputation-desire do acknowledge that the unwanted limb is a part of their body. It is the gap between this conscious, reflective judgement and the overwhelming feeling of extraneousness toward it that elicits a desire for amputation, which is experienced as irrational, even for the person with this condition. Following de Vignemont's distinction between feeling of ownership and judgments of ownership (de Vignemont, 2011), our data support the idea that in individuals with amputation-desire the judgment of ownership is still preserved although the feeling of possession of the unwanted limb has gone, while somatoparaphrenic patients have lost both, feeling and judgment of ownership. Our findings support also the idea that the feeling of ownership might be very relevant in anticipating incoming sensory stimuli, suggesting that the emotional component of nociception is critical in this adaptive behaviour. Converging evidence from different pathological ownership conditions supports this hypothesis. Not only are the aforementioned data from somatoparaphrenic patients in line with it, but findings from individuals with acquired pathological embodiment, also known as E+ (Garbarini, Pia, Piedimonte, Rabuffetti, Gindri, & Berti, 2013; Garbarini et al., 2014b), suggest a similar interpretation. E+ patients are characterized by a peculiar

disturbance: after a stroke they recognize every arm that would appear to the contralesional side of their body in an anatomically plausible position as their own arm (Garbarini et al., 2013; Garbarini et al., 2014b). These patients never attribute more than one arm per side to themselves, and they typically select the one closer to the midline on the contralesional side and their own on the healthy side. Critical to our hypothesis, E+ patients have equally strong SCR when a needle approaches their healthy limb or the embodied alien limb, while SCR decrease if alien limb is in a location where it cannot be incorporated (Garbarini et al., 2014a).

Additionally, in healthy individuals the sense of ownership for one's own body, mostly investigated in the frame of the rubber hand illusion (RHI) (Botvinick & Cohen, 1998), showed that the emotional reaction following the threatening of a body part is related to the sense of ownership felt for that body part, and our findings are reminiscent of such results. These findings are further compatible with the notion that the sense of ownership for a limb tunes analgesic effects when a noxious stimulus is delivered to that body part (Longo, Betti, Aglioti, & Haggard, 2009; Romano et al., 2014c). Despite the interesting results from bodily illusion paradigms in healthy individuals, it is unquestionable that the sense of limb ownership disrupted by brain damage is incomparably more disturbed. While in healthy subjects artificial embodiment sensation for external objects is more variable (de Vignemont, 2011), body disownership in patients appears to be more constant.

Our findings may have consequences for clinical work within the spectrum of amputation-desire. This condition, that could be considered the most extreme condition of degradation of body representation, raises relevant ethical issues on the concept of ownership (Müller, 2009; Sedda, 2011) and is not yet included in the DSM-5, nor in the ICD classification, as it is still considered a fairly obscure condition. There are suggestions that there might be different subcategories within this condition, eliciting several problems of classification (First, 2005; McGeoch et al., 2011; Sedda & Bottini, 2014; Sedda, 2011). Paradigms such as the one introduced here could be useful for an assessment of the disorder that is more objective than questionnaire approaches, embedding also implicit aspects of distorted body representations. Further, such a paradigm would be useful to

discriminate between xenomelia and psychiatric conditions mimicking or absorbing this peculiar symptomatology.

To conclude with a caveat: although our results are indicative of a body representation dysfunction in individuals with an amputation-desire, we cannot rule out the possibility that this dysfunction could be the consequence rather than the cause of the disorder (Sedda & Bottini, 2014). Further studies are needed before any definite conclusion regarding the etiology of this condition can be reached.

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Figure Captions

Fig.1: The 'cat-eyes' graph represents the 90% confidence intervals for each specific condition. The cat-eye shape is composed of two attached symmetrical normal distributions, thus it graphically represents the true probability of data distribution - i.e. the larger section correspond to the average value and represent the most probable value for that condition as compared to values in the cues that are less probable to be observed.

Tables

Tab.1 Demographic and clinical features of the 10 individuals presenting with amputation-desire. Age and Education are reported in years. Pain ratings were expressed on a verbal scale ranging from 1 (not painful) to 10 (worst pain experienced).

	Participant	P1	P2	P3	P4	P5	P6	P7	P8	P9	P10
Demographic Details	**Gender**	Male	Male	Male	Female	Male	Male	Male	Male	Male	Male
	Age	42	29	36	43	36	52	49	61	34	37
	Education	13	18	13	18	13	18	15	18	18	19
	Handedness	Right	Right	Left	Right	Right	Ambidextrous	Right	Right	Right	Right
Amputation Desire	**Limb**	Leg	Leg	Leg	Arm	Leg	Leg	Leg	Arm	Leg	Leg
	Side	Left	Right	Left	Left	Left	Right	Left	Left	Left	Left
	Location	10 cm above the knee	10 cm above the knee	10 cm above the knee	Elbow	10 cm above the knee	Just above the knee	Just above the knee	Elbow	Half of the thigh	Just below the knee
	Sexual desire (apotemnophilla)	yes	yes	no	no	no	no	no	no	yes	no
	Onset (age in years)	6-7	7-8	6	3-4	6-7	6-7	6	5	6-7	6-7
	Dysphoria related to BIID	yes	yes	yes	yes	yes	yes	yes	no	-	yes
	Time spent thinking on BIID/24 hours	24	2	4	24	3	20	2	24	1	24
	Time spent on BIID forums/ per day	-	30 minutes	-	60 minutes	60 minutes	-	-	-	-	-
Therapy	**Drug treatment**	yes (remeron)	no	no	yes (citalopram)	no	no	no	no	no	no
	Psychotherapy	no	no	no	yes	no	no	no	no	no	No
Pain Evaluation	**Unwanted Limb (cotton swab/needle)**	1/3	1/3	1/3	1/3	1/4	1/4	1/7	1/3	1/3	1/5
	Healthy Limb (cotton swab/needle)	1/6	1/3	1/7	1/3	1/4	1/3	1/3	1/3	1/4	1/5