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Research Paper

α Preliminary evidence for a fronto-parietal dysfunction in able-bodied participants with a desire for limb amputation

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Background. Reports of able-bodied participants with the persisting desire for limb amputation raise legal and ethical questions that are partly due to insufficient empirical knowledge about the condition. Here, we searched for potential neurological mechanisms in participants with desire for limb amputation in order to help develop adequate nosological classifications, diagnosis, and treatment.

Methods. Semi-structured interviews were carried out with 20 participants who self-identified themselves as able-bodied individuals desiring amputation of a limb.

Results. The results suggest that amputation desire is not unspecific, but in most cases specific for a circumscribed part of the body. Most frequently affected was the leg, mostly on the left, non-dominant side. Left-sidedness and limb specificity was associated with elementary and complex somatosensory disturbances of the affected limb akin to those reported by neurological patients. The most frequent neurological co-morbidity was migraine headache.

Conclusions. These results document the existence of an unusual condition in able-bodied participants characterized by a person's desire for the amputation of one or more particular limbs. Left-sidedness, limb specificity and somatosensory disturbances of the affected limb are suggestive of abnormal brain mechanisms in right fronto-parietal cortex. Based on this association we suggest that desire for limb amputation may be conceptualized as asomatognosia due to disturbed integration of multi-sensory information of the affected body parts into a coherent cerebral representation of the own body. This suggestion has to be regarded with caution as we did not perform any neurological examination.

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2 *Olaf Blanke et al.*

Most humans experience considerable distress when faced with the loss of a limb (Friedmann, 1978). Yet in 1997, a surgeon amputated a healthy, left leg on demand of an able-bodied patient. This patient was a male, married lecturer with a normal psychiatric examination (Bridy, 2004; Johnston & Elliott, 2002). Another person's fully functional limb was operated before the hospital's administration banned such operations. These participants suffered from what has been called 'body integrity identity disorder' (First, 2005; Furth & Smith, 2000), a rare condition in which able-bodied individuals report experiencing a lifelong desire for the amputation of one or several of their limbs. Desire for limb amputation is not known to most surgeons, psychiatrists, and neurologists, not included in the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR), and raises broad legal and ethical issues.

Empirical data on participants with the desire for limb amputation are sparse and largely based on anecdotal case reports suggesting serious negative consequences including lifelong distress, self-amputation attempts, and death in the course of 'back alley' amputations (Bridy, 2004). Prevalence, diagnostic classification, co-morbidities, and potential medical treatments of people with desire for limb amputation are unknown or controversial. A more systematic look at this desire thus seems mandatory. Following the initial report by Money, Jobaris, and Furth (1977), a few case reports have appeared in scientific journals (Bensler & Paauw, 2003; Braam, Visser, Cath, & Hoogendijk, 2006; Everaerd, 1983; Parsons, Brown, & Sirota, 1981; Storm & Weiss, 2003) and popular media (Dotinga, 2000; Dyer, 2000; Elliot, 2000; Horn, 2003). Preliminary evidence suggests that these individuals desire amputation because they wish to restore their true identity, feel sexually aroused, or claim feeling better if the body part they don't identify with were amputated (First, 2005). There is no agreement on how to classify this desire diagnostically with respect to known psychiatric diseases, perhaps due to the bizarre and uncommon symptoms and the fact that there is to date only one published group study in such individuals (First, 2005). Thus, it has been subsumed under the categories of body dysmorphic disorder (Dyer, 2000), psychosis (Scholzman, 1998), or paraphilia (Money *et al.*, 1977). More recently, based on analysis of several cases with desire for limb amputation, Furth and Smith (2000) as well as First (2005) proposed that it might be a separate psychiatric diagnostic entity related to gender identity disorder (Body Integrity Identity Disorder).

Based on phenomenological similarities with neurological symptoms associated with damage to parietal cortex several authors have mentioned possible neurological causes for the desire for limb amputation (Braam *et al.*, 2006; Bridy, 2004; First, 2005). These symptoms include asomatognosia (patients' experience that one of their limbs is absent or has been amputated) (Arzy, Overney, Landis, & Blanke, 2006; Gerstmann, 1942), somatoparaphrenia (patients' experience that one of their limbs belongs to somebody else) (Bottini, Bisiach, Sterzi, & Vallar, 2002; Gerstmann, 1942), and misoplegia (hatred of paralysed or functional limbs) (Loetscher, Regard, & Brugger, 2006), but also more elementary somatosensory symptoms such as paresthesia and hypoesthesia. Yet, to date no study has systematically investigated the phenomenology of desire for limb amputation with respect to the affected body part(s) and potentially associated neurological symptoms and diseases. It was the goal of the present interview study to search for potential neurological mechanisms in participants with desire for limb amputation in order to help develop adequate classification, diagnosis, and treatment.

Methods

Participants

The 20 participants of the study were recruited from the *Internet* over a period of 6 months and identified themselves as having the desire to have an amputation. An advertisement was placed on body integrity identity disorder websites and interested participants were asked to contact us. Following this first contact, the participants received a detailed description of the study, were asked to provide written informed consent and to fill out questionnaires about their condition. A date were then set for the telephone interview to take place.

Interview

The telephone interviews were conducted by two of the authors (OB, LSO) in the participant's preferred language (English, French, and German) after having received informed consent by e-mail. All interviews were conducted anonymously. Based on First's (2005) study a semi-structured interview was developed to facilitate consistent collection of information. The interview consisted of 80 questions, the majority being open-ended (see additional *Internet* material). Many questions were designed to gain more information about the condition's phenomenology and to test the authors' hypotheses about a potential neurological aetiology. Interviews lasted from 50 minutes to 2 hours, and all participants completed the entire interview. Participants were asked to complete the perceptual aberration scale, a 35-item true/false inventory for the assessment of schizotypy (Chapman, Chapman, & Raulin, 1978). Extensive notes were taken during the interviews. Topics such as demographics, specific questions about the amputation desire including the location of the desired amputation, its temporal evolution and precipitating factors were covered. The interview also addressed neurological symptoms such as paresis, hypoesthesia, paresthesia, dyssomatognosia, asomatognosia, somatoparaphrenia, and misoplegia (Hécaen & Albert, 1978). We employed these terms following the initial description of Gerstmann (1942): feeling of amputation or a positive sensation of a loss of awareness for a certain body part (asomatognosia), and the feeling that one's own hand belongs to someone else (somatoparaphrenia). There were also items asking about non-corporeal hallucinations and illusions. We finally inquired about the participant's general medical history and family history, with a special focus on neurological, neurosurgical, and psychiatric history. Handedness was determined by a 10-item-scale (Oldfield, 1971). Hand and foot dominance were tested by an eight item-scale (Coren, 1993).

Results

Participants

The majority of the 20 participants lived in France (30%, $N = 6$), Germany (30%, $N = 6$), and the USA (20%, $N = 4$). The remaining participants came from Belgium ($N = 1$), Austria ($N = 1$), Sweden ($N = 1$), and The Netherlands ($N = 1$). Most were men (85%; $N = 17$) aged between 29 and 72 years (mean age: 48.4 years) consistent with previous data (First, 2005).

All participants were highly educated and had at least 12 years of schooling. Eleven participants (55%) further pursued their education becoming a nurse, a team manager, or a pilot. Nine participants (45%) had a university degree (e.g. medical doctor, geologist, biomedical engineer, school teacher, and biologist; Supplementary Table 1).

4 *Olaf Blanke et al.*

Sixty-five percent ($N = 13$) of the participants were or had been married. Among them, nine were still married (45%), three were divorced (15%), and one was widowed (5%). One participant was in an ongoing relationship (5%). Six participants were single (30%). Contrary to what First (2005) had described, the sexual orientation of our population was heterosexual (95%; Supplementary Table 1).

Only one participant was left-handed (5%) and one ambidextrous, all others were right-handed (90%, $N = 18$). This pattern was confirmed for hand dominance using Coren scores (1993), according to which 18 participants had a dominant right hand (90%). The two participants who were classified as left-handed and ambidextrous following Oldfield (1971) were both classified as being dominant for their left hand using Coren scores. This was also reflected in foot dominance: 70% were right dominant; 20% without a dominant foot; and 10% left dominant. The Coren scores for hand and foot dominance in the present participants are comparable to those obtained in a large control population (Coren, 1993).

Amputation desire

Reasons for the amputation desire

Q1 When asked why they wanted to have an amputation (open question), all participants reported that one or several of their limbs did not belong to their body and that their physical body somehow did not match the 'image' or 'idea' that they have of 'their body' or 'themselves'. Some representative sample statements are given in Supplementary Table 2. When participants were asked to respond to several possible reasons why they wanted an amputation, they mostly stated that amputation of the affected body part would make them 'feel whole, complete, set right again' (85%; $N = 17$) or 'feel satisfied and elated inside' (70%; $N = 14$). 'Feeling sexually excited' was indicated by 65% ($N = 13$), but was rarely the main reason (First, 2005). It is interesting to note that participants clearly stated that it was not to draw attention and neither to be cared for by others. We further enquired about how they imagined their lives after amputation. All replied that although they knew they would be disabled, they would finally be able to go on with their lives without being constantly frustrated by their 'extra' limb(s). They all knew they would find solutions for their care (wheel chair, nurse, etc.).

Ninety percent of the participants ($N = 18$) strongly believed that only an amputation of the affected limb(s) would relieve them from their condition ('Operation is the only possibility'). Few were more critical about the potential outcome of the amputation (10%) stating that 'I cannot believe that amputation will solve my problem' or 'I wouldn't do anything to get an amputation, life is more important, but on the other hand, I don't want to live with this'. All participants were aware that their condition is particular and abnormal. Thus, despite the fact that all participants had amputation desire, opinions about the condition and the estimated success of an eventual amputation varied considerably.

Affected body part

Most participants had a localized amputation desire and wished for one or several specific body parts to be removed (85%, $N = 17$). In agreement with First (2005) all 20 participants wanted an amputation of a major limb as opposed to only fingers or toes (Figure 1; black bars). With respect to the affected body part, we found that 80%

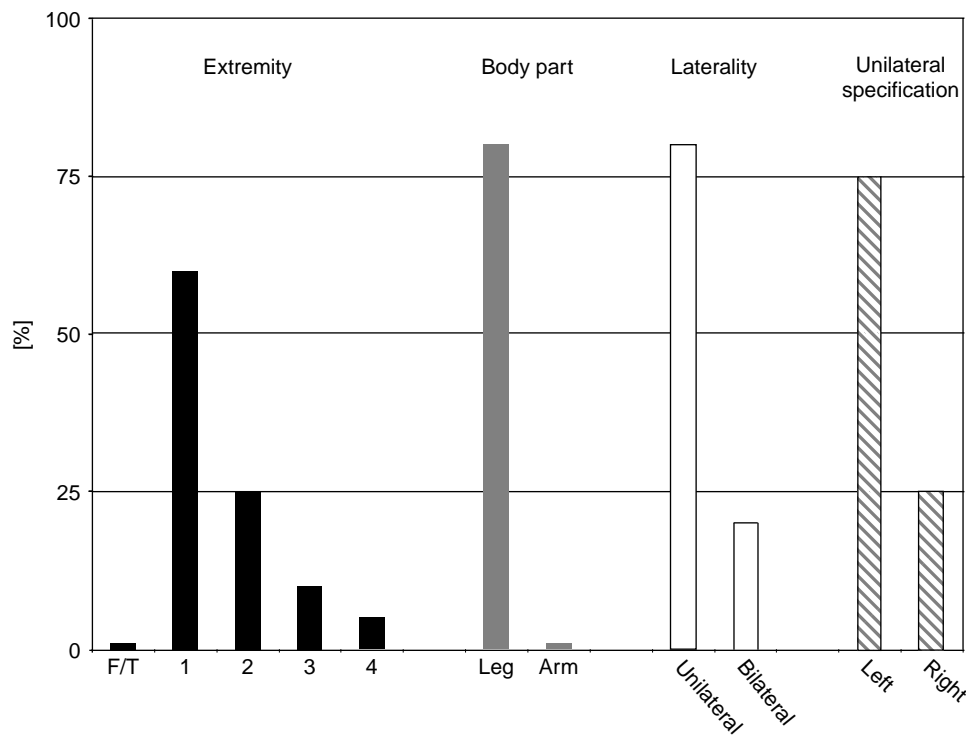


Figure 1. Amputation desire is leg biased and left lateralized. The majority of participants in the present study desired an amputation of one single limb (amputation of two or more extremities or fingers or toes only were less frequent) (black bars, left). This was also found by First (2005; percentage values are also given). In addition the leg was affected in the large majority of cases in the present study (grey bars) and the study by First (2005; percentage values). Moreover, the large majority (80%) of amputation desires were predominantly unilateral (white bars labelled 'Laterality'). Among them, 75% were left-sided and 25% were right-sided (striped bars labelled 'Unilateral specification', right).

($N = 16$) wished for specific leg amputation as opposed to arms (Figure 1; grey bars). Nobody had a specific amputation desire for the upper extremity (i.e. without leg amputation). The remaining participants wished for a combined leg and arm amputation (10%; $N = 2$) or fantasized about an additional arm amputation (10%; $N = 2$). The location of the amputation was specific in all participants. Most participants wanted an above-knee amputation (70%, $N = 14$) as opposed to a below-knee amputation (30%, $N = 6$). The two participants with amputation desire for an arm desired an operation below the elbow (10%; $N = 2$). For most of the participants, the location of the amputation had remained constant over time (80%; $N = 16$). Four participants indicated that the location of the desired amputation underwent some change, notably in a proximal-to-distal direction (e.g. from a location above the knee to a location below the knee). Three patients (15%) reported that they underwent professional or self-inflicted amputation (of mainly fingers and toes) mentioning that they did not dare at the moment to have larger parts of their body removed.

6 *Olaf Blanke et al.**Lateralization of affected body part*

For 11 participants (55%) the desire of amputation was unilateral, whereas the remaining participants wished for a bilateral amputation (45%; $N = 9$). Among the 11 participants who wanted a unilateral amputation, the majority (64%; $N = 7$) desired amputation for a limb on the left side of the body. In five participants (25%) with a desire for bilateral amputation this desire was nevertheless stronger for one limb. In other words, 16 participants (80%) had a unilateral ($N = 11$) or a bilateral, but predominantly unilateral ($N = 5$) amputation desire (Figure 1; white bars). Among these 16 participants, 12 (75%) reported a left-sided or bilateral, but left-predominant, amputation desire (Figure 1). Accordingly, a right-sided amputation desire or bilateral but right predominant amputation desire only affected 25% ($N = 4$).

Of the 16 participants with a dominant foot (right: $N = 14$; left: $N = 2$) (Coren, 1993), 15 desired unilateral (or predominantly unilateral) amputation and one participant desired bilateral amputation. Of these 15 participants, 73% had an amputation desire for their non-dominant foot.

Interestingly, among the 11 participants (55%) who wished for unilateral amputation, 10 were right-handed, and 6 of them (60%) wished for a left amputation. The left-handed participant wished for a left unilateral amputation. Among the same 11 participants who wished for a unilateral amputation, 8 were right-footed, and 6 of them (75%) wished for a left amputation. The three remaining participants were ambidextrous and two of them wished for a right amputation.

Gender, location, and lateralization of the affected body part

Of the three women, one wished for the amputation of all four limbs, one for both legs and an additional arm, and one for both legs. This is in contrast with the large majority of our 17 male participants, who had an amputation desire that affected a single limb (71%; $N = 12$). No male participant desired amputation of all four limbs. This interaction between gender and the number of affected limbs is in accordance with previous findings (First, 2005).

Sensations of the affected limbs and body

When asked whether they experienced any particular sensations in the affected limb(s) that were qualitatively different from the sensation of the rest of their body, 13 participants (65%) mentioned abnormal sensations of the affected body part. The remaining participants did not mention the presence of any abnormal sensations. For these 13 participants, the sensations most often included the feeling that the affected limb belongs to another person (somatoparaphrenia; 46%; $N = 6$) and that the affected limb was absent or already amputated (asomatognosia; 23%; $N = 3$). Both sensations were not delusional as participants had preserved insight into their unreality. Abnormal sensory experiences were also mentioned and consisted of paresthesia (38%; $N = 5$), hypoesthesia (38%; $N = 5$) and the feeling that the affected body part felt different than other parts of their body (23%; $N = 3$; Supplementary Table 2). All other experiences about which we inquired were rare (pain; microsomatognosia; kinaesthetic illusions) or absent (macrosomatognosia; illusory limb disconnection; supernumerary phantom limbs; illusory weight or size changes of the affected limb; misoplegia) (Hécaen & Albert, 1978). We further inquired whether these abnormal sensations were modified by movement, vision, or touch of the affected body part. Only one participant

with illusory leg movements reported that touching his leg would diminish the feeling. All sensations were specifically reported for the affected limb or limbs and never concerned the non-affected limb(s). We also asked for other abnormal sensations including auditory, visual, olfactory, gustatory, and vestibular sensations. These were negligible or absent altogether. It is important to note here that none of our *respondents* suffered from delusions concerning the affected body parts.

Evolution of the amputation desire

Most participants indicated that the onset of the amputation desire occurred in early childhood, that is, between 3 and 9 years (65%, $N = 13$) or at the beginning of teenage (12-16 years; 25%, $N = 5$). The mean age of onset was 11.6 years. In only 10% ($N = 4$) did the amputation desire start after the age of 16 years.

The evolution of the amputation desire over the years was described as chronic and of constant strength in 50% of the participants and as slowly progressive in 45% of them. Only for one participant was the strength of the amputation desire more variable over time.

Concomitant factors

Ninety percent ($N = 19$) of the participants reported that although the amputation desire was constantly present, several factors influenced its strength or level of awareness during daily life. Factors that diminished the amputation desire included professional activity in general, 'when busy', or when work requires a lot of attention (50%; $N = 10$). Other diminishing factors comprised physical activities (20%; $N = 4$), stress (15%; $N = 3$), and simulation of amputation (by bending the limb, wearing a prosthesis, or other mechanisms; 10%; $N = 2$). Most participants also mentioned that the amputation desire could be enhanced (70%; $N = 14$) in situations of loneliness, when seeing amputees, and during stress load.

Interest in another disability

None of the participants indicated ever having desired any other disability such as blindness, deafness, or being mute.

Medical history

Concerning medical history, six participants reported suffering from migraine (30%), and two additional participants reported having regular mild headaches (total headache: 40%). Four participants (20%) suffered from lumbar discal hernia treated by surgery. Two participants (10%) suffered from diabetes mellitus (type II) and one participant (5%) had a physical body abnormality (aplasia of left chest muscle). Four participants (20%) suffered from depression and had seen a psychiatrist. Three further participants (15%) consulted a psychiatrist for their amputation desire. Three participants were under psychiatric drug treatment, namely, antidepressants. One of these three participants also received treatment for schizophrenia (Haldol). However, these different treatments did not alleviate the amputation desire, but was judged as beneficial for depressive symptoms by patients. Three participants had a family history of vascular disease (15%), two of diabetes (10%), two of Parkinson's disease (10%), two of psychiatric disease (10%; bipolar disease, schizophrenia), and one each of brain tumor and cancer (5%).

Perceptual aberrations

The mean score on the perceptual aberration scale was 7.6 (6.9; *SD*), corresponding to a pre-schizotypy range (Chapman *et al.*, 1978). Norms on this test are 5.14 ± 5.4 . Twelve participants scored in the normal range (60%), six in the mid-range (30%), and two participant's (10%) scores were indicative of moderate-to-high schizotypy (above 19).

Discussion

The results of this survey further document the existence of an unusual condition in able-bodied participants that is characterized by a person's desire for the amputation of a particular limb or limbs and may lead to serious negative consequences (First, 2005; Furth & Smith, 2000)¹. Our demographic findings agree with previous results (First, 2005). The amputation desire was in the majority (62%) of the interviewed participants specific for one body part and mostly affected the left, non-dominant leg or both legs with a left predominance. Moreover, along with limb specificity, unilaterality, and left-sidedness of the amputation desire we also observed elementary and complex somatosensory disturbances of the affected limb. These findings are comparable to the association of symptoms in neurological patients suffering from right hemisphere brain damage, although this statement has to be regarded with great caution as we did not perform any neurological examination.

Previously published data are in accordance with our findings (Bensler & Paauw, 2003; Braam *et al.*, 2006; Everaerd, 1983; Money *et al.*, 1977; Parsons *et al.*, 1981; Storm & Weiss, 2003). In these eight previously reported participants, desire for limb amputation was limb-specific in all cases, unilateral in 71%, of which 80% were left-sided (Supplementary Table 3). Thus, as in First's (2005) and our study, the large majority of these previously reported participants with amputation desire also desired a unilateral leg amputation. In addition, even participants who wished for bilateral amputation (45%) also had predominance for left leg amputation. The lateralization of the amputation desire to the left leg is compatible with possible abnormal processing in right cerebral structures coding for the affected leg and other illusory perceptions present in our study and the left lateralization in neurological patients with body representation disorders (or body schema disorders; see below). Other factors such as leg dominance may also be important. The lower limbs, and especially the non-dominant leg, are important in supporting the human body. We therefore hypothesized that amputation desire could affect the non-dominant leg more often in order to avoid the function of the main body support. Yet, this was not supported by the present data. The functional role of the non-dominant leg in supporting the body therefore does not seem to play a major role in lateralization of amputation desire. If this were the case amputation desire should have been lateralized more often to the dominant leg. Yet, we found the opposite: in right-footed participants, the amputation desire affected the non-dominant, and thus supporting leg, in the majority of participants (75%). Based on the present interview data we cannot exclude that other factors related to leg dominance influence the laterality of the amputation desire. Most likely several factors are

¹ Although previous authors have proposed to call the desire for limb amputation body identity integrity disorder (First, 2005; Furth & Smith, 2000). We here propose to refer to it with a more descriptive term – desire for limb amputation – until empirical data allow to decide whether it is diagnostic category or rather a symptom of neurological or psychiatric disease.

involved in the predominant lateralization of the amputation desire to the left and non-dominant leg. Based on additional observations of the present study, we speculate that the frequent amputation desire for the left, non-dominant leg would suggest that amputation desire might be related to central mechanisms in fronto-parietal cortex, especially of the right hemisphere. This is further supported by the observations of an exaggerated malleability of left-sided limb representations as measured by a body part illusion (rubber hand illusion) that can be induced in healthy participants. Thus, the susceptibility to the rubber hand illusion for the left, but not right hand is correlated with the severity of body schema disturbances (Mussap & Salton, 2006).

This assumption is given further support by the presence of elementary and complex somatosensory manifestations of the affected limb. Our participants with amputation desire often reported paresthesia and hypoesthesia of the affected limb that are often associated with damage to fronto-parietal cortex (Victor & Rooper, 2005). Although these sensations may also follow damage to subcortical structures (Victor & Rooper, 2005), leg specificity in participants with amputation desire suggests interference with cortical rather than subcortical mechanisms. Alternatively, leg specific elementary somatosensory symptoms are also compatible with a peripheral origin of desire for limb amputation as in lumbar discal hernia (as reported in some of our participants) or diabetes (First, 2005). Yet, participants with desire for limb amputation frequently pretend amputation through bending and long-term usage of crutches and prostheses and the onset of the amputation desire is reported before the age of 15 years (First, 2005). This suggests that discal hernia and other peripheral abnormalities might rather be a consequence of the long-lasting desire for limb amputation due to the manipulations of the affected limbs (Bensler & Paauw, 2003; Storm & Weiss, 2003). Interference with right fronto-parietal mechanisms is also concordant with the presence of complex symptoms in the present participants that are not reported in patients with peripheral lesions. These complex symptoms are comparable with somatoparaphrenia (patients' experience that one of their limbs belongs to somebody else) (Bottini *et al.*, 2002; Gerstmann, 1942), asomatognosia (patients' experience that one of their limbs is absent or has been amputated) (Arzy *et al.*, 2006; Gerstmann, 1942). Our data did not reveal an association with misoplegia (hatred of paralysed or functional limbs) (Critchley, 1953; Loetscher *et al.*, 2006). One participant felt disgust about his affected limb and 25% of the participants stated to have performed 'aggressive' behaviour towards the affected limb as can also be seen in patients with misoplegia. All denied that this was done because they hated the targeted limb. We do not consider the leg bending as reported in several of our participants as misoplegic, but this needs to be evaluated more specifically in future studies and was not possible within the framework of the present study. We note however that in contrast to these mostly acute and severe manifestations of parietal lobe damage (somatoparaphrenia, asomatognosia, and misoplegia) that are often associated with delusional beliefs, the present participants with amputation desire were chronic and not delusional (Hécaen & Albert, 1978). The relationship between belief and underlying abnormal perception/sensation is complex and much debated in the literature on symptoms such as somatoparaphrenia or asomatognosia. We do not know for our participants whether abnormal experiences that show similarities with somatoparaphrenia and asomatognosia (as defined here) relate to abnormal experiences or non-perceptual mechanisms related to their wish or rationalization. Based on our interview we can only state that none of the participants suffered from delusions with respect to the affected body part or showed signs of delusions. Collectively, our data suggest that amputation desire might be conceptualized as chronic asomatognosia or a negative

form of the phantom limb phenomenon (Melzack, 1990) due to disturbed integration of multi-sensory information of the affected body parts into a coherent cerebral representation of the own body. This has to be regarded with caution as we did not perform a neurological examination and as 45% of the reported sample did not report any abnormal sensations. Moreover, next to unilaterality and left predominance, our data also suggests that many participants wanted both legs amputated. This is not necessarily compatible with a larger right hemisphere involvement in amputation desire, but rather suggests the presence of additional factors. Yet, there are reports that if illusory own body perceptions affect the lower limbs they tend to be often bilateral rather than unilateral (Blanke, Ortigue, Landis, & Seeck, 2002; Vuilleumier, Reverdin, & Landis, 1997). In addition, to our proposed neurological mechanisms, other neurological, dominance-related, psychiatric, or still other aetiologies need to be carefully evaluated and examined in future studies (see below).

We observed that migraine is often associated with amputation desire, although our participants did not report that the strength of the desire coincided with periods of migraine headaches. Migraine affecting the parietal lobes has been described and may be associated with asomatognosia and somatoparaphrenia as well as other, milder forms of bodily disturbances (Lippman, 1953; Podoll & Robinson, 2002). Further studies are needed to clarify this potential etiological link that is also concordant with the mildly elevated perceptual aberration scores we have found. Several practical conclusions can be drawn. A detailed neurological and neuropsychological, as well as a psychiatric examination should be performed in participants with amputation desire. Second, structural and functional abnormalities in fronto-parietal cortex including mesial primary sensorimotor cortex, premotor cortex, supplementary motor area, and Brodmann's area five should be examined by structural and functional MRI. These investigations should be complemented by somatosensory and motor evoked potentials.

With respect to the psychiatric nosological system, we realize that more psychiatric information would have been useful, but this was not possible beyond the questions asked in our interview. In the present short telephone interview our main interest was to gather additional information from a neurological perspective. Nonetheless, our data agree with the observation of several authors that desire for limb amputation does not represent a form of body dysmorphic disorder as the present participants did not perceive their affected limb as defective in any way, nor did they feel embarrassed or ashamed about its appearance (Braam *et al.*, 2006; First, 2005; Furth & Smith, 2000). Given the bizarre nature of the desire for limb amputation and the frequent mutilations and, more rarely occurring, self-inflicted amputations in individuals with desire for limb amputation, it might be suggested that desire for limb amputation is linked to schizophrenia and psychotic depression², as self-inflicted amputations have been regularly reported in these latter two conditions (Brenner, 1999; Demuth, Srain, & Lombard-Maher, 1983; Goldwyn, Cahill, & Grunebaum, 1967; Hall, Lawson, & Wilson, 1981; Jaffe, Earle, Fleegler, & Husni, 1975; Scholzman, 1998; Schweitzer, 1990; Stewart & Lowry, 1980). Yet, the present interview study as well as previous investigations did not find clinical signs of major psychosis in individuals with desire for limb amputation (Braam *et al.*, 2006; First, 2005; Money *et al.*, 1977). Further, the few psychiatric drug treatments (mostly antidepressants) administered to some of the participants

² Alternatively, self-mutilation attempts have also been reported in neurological patients with right parietal lesions for which the term *misoplegia* has been used.

were beneficial for depressive symptoms but did not alleviate the amputation desire, which suggests that the desire is probably related to other mechanisms. In addition, location and lateralization of the body part most often severed in self-inflicted amputation in schizophrenia and psychotic depression (right or bilateral hand in 77%) (Scholzman, 1998) differs from participants with desire for limb amputation. Finally, self-inflicted amputation is described as an acute phenomenon in schizophrenic patients, which is not the case in individuals with desire for limb amputation. Although individuals with desire for limb amputation may mention sexual arousal as an important reason for their amputation desire (Money *et al.*, 1977), only a small group of these participants considers this to be the main reason for their amputation desire (15%; paraphilia not otherwise specified (DSM-IV-TR)) (First, 2005) making the diagnosis of paraphilia not appropriate. Accordingly, it has been proposed that desire for limb amputation can be conceptualized as a separate psychiatric nosological entity (First, 2005; Furth & Smith, 2000). Resembling gender identity disorder, with which it shares several clinical features (First, 2005; Furth & Smith, 2000).

Due to several important limitations of the present study (small sample size; interviews were conducted by telephone; recruitment of participants via the *Internet*; absence of neurological examination; and separate rating by authors) and the current lack of clinical data in participants with desire for limb amputations, our data suggest that larger clinical studies will be needed to disentangle neurological from psychiatric and other causes. This seems mandatory also with respect to the broader legal and ethical issues raised by the desire for limb amputation. Courts are likely to consider amputation of a healthy limb as malpractice as it is not considered by a responsible body of medical opinion and data to be an appropriate and effective treatment of a medical condition (Johnston & Elliott, 2002). Surgery is only considered a lawful activity when it is reasonable, or when it constitutes proper medical treatment, and if it is performed with the patient's consent. Again, it is unlikely that courts consider amputation of a healthy limb proper medical treatment without any empirical knowledge about the underlying mechanisms and empirical evidence of some kind of therapeutic benefit (Johnston & Elliott, 2002). As stated by Johnston and Elliott, the patient's consent alone will not be enough to excuse the surgeon from criminal liability (Johnston & Elliott, 2002). The present data suggest that participants with amputation desire should consult with a team of health professionals including neurologists, neuropsychologists, and psychiatrists.

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12 Olaf Blanke et al.

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14 *Olaf Blanke et al.*

Author Queries

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- Q1** Please note that the term 'subject' has been change to 'participant' as per the journal style.
- Q2** Please note that the italicization in the sentence 'We therefore hypothesized that amputation desire could affect the non-dominant . . .' has been removed.
- Q3** Please provide the last accessed date for the reference Dotinga (2000).