Neuropsychol Rev (2011) 21:334–336
DOI 10.1007/s11065-011-9186-6

REVIEW

Body Integrity Identity Disorder: From A Psychological to A Neurological Syndrome

Anna Sedda

Received: 9 October 2011 / Accepted: 2 November 2011 / Published online: 10 November 2011

Abstract Body Integrity Identity Disorder (BIID) is a condition in which individuals experience an intense desire for amputation of an healthy limb. Recently, McGeoch and colleagues provided the first direct evidence that this syndrome may be neurological rather than psychological in its origin. However, before including BIID in body ownership disorders, several concerns should be clarified, exploring other components of body representation and not only somatosensory perception.

Keywords Body Integrity Identity Disorder (BIID) · Apotemnophilia · Xenomelia · Right parietal lobe syndrome · Body representation disorders · Body image

Introduction

Since 1970, case reports have begun to describe individuals with an intense desire for amputation of an healthy limb (Everaerd 1983; Money et al. 1977). The discomfort for the limb is so strong that it interferes with everyday life functioning, and several individuals have asked a surgeon for an amputation of the "extraneous" extremity, or even performed it by themselves (Patrone 2009). This unusual behavioral condition was initially described as a paraphilia, implying that its core is a sexual disturbance of psychological origin (Everaerd 1983; Money et al. 1977). More recently a group study highlighted that the central core of the disturbance might be the need to remove the affected limb in order to "feel complete" rather than a paraphilia (First 2005). From the term "*apotemnophilia*" (Money et al. 1977) several researchers moved to "*Body Integrity Identity Disorder"* (BIID), first coined by First in 2005 (Berlucchi and Aglioti 2010; Patrone 2009; Oddo et al. 2009; First 2005).

Recently, McGeoch and colleagues provided the first direct evidence that the sense on incompleteness reported by BIID patients (First 2005) finds a correspondence in a dysfunctional activity of the right parietal lobe (McGeoch et al. 2011), strongly supporting the hypothesis that BIID in not simply a paraphilia but rather a neurological syndrome (Blanke et al. 2009; Ramachandran and McGeoch 2007; Aoyama et al. in press). McGeoch and colleagues propose that 'xenomelia', from the Greek terms 'foreign' and 'limb', would describe this new parietal lobe syndrome better than apotemnophilia or body integrity identity disorder (McGeoch et al. 2011).

In the study of McGeoch and colleagues, four BIID individuals, who desired the amputation of either the right or the left leg, with no psychiatric or neurologic diseases, and a matched control group have been recruited. Somatosensory evoked fields were recorded by means of magnetoencephalography while participants were touched: a) on the dorsum of each foot, b) on each anterior thigh above the line of desired amputation, and c) during the electrical stimulation of the median nerve over the volar aspect of each wrist (control condition). McGeoch and colleagues found a significant reduction in the activity of the right Superior Parietal Lobule (SPL) when comparing the BIID individuals somatosensory responses for the affected leg with that of the unaffected leg and that of the control participants. No other significant activity reductions were found in areas known to be involved in body representation, such as the insula (Berlucchi and Aglioti 2010).

A. Sedda (✉)
Psychology Department, University of Pavia,
Piazza Botta 11,
27100 Pavia, Italy
e-mail: sedda.anna@gmail.com

In line with findings from skin conductance experiments (Brang et al. 2008), these results encourage the idea that BIID may be a parietal lobe syndrome (Blanke et al. 2009; McGeoch et al. 2011; Brang et al. 2008), rather than being purely psychological in its origin (First 2005; Everaerd 1983; Money et al. 1977). McGeoch and colleagues suggest that BIID individuals are able to perceive the affected limb because visual and somatosensory cortices are intact, but they fail to incorporate it into their body image due to parietal lobe dysfunctions.

The notion that body image but not body schema may be damaged in BIID is implicit in this assumption. According to a classic dichotomy, body schema refers to a dynamic representation of body parts in space, continuously updated during movement, while body image refers to a description of the body (Berlucchi and Aglioti 2010; de Vignemont 2010). However, the study of McGeoch and colleagues only investigates tactile perception, that is usually ascribed to body schema rather than body image (Berlucchi and Aglioti 2010; Aoyama et al. in press). The absence of other tasks tackling diverse components of body representation may explain why other areas known to contribute to the sense of ownership, such as the insular cortex (Berlucchi and Aglioti 2010; de Vignemont 2010), did not show a differential activity. Body image and body schema of BIID patients should be assessed with more extensive neuropsychological batteries, containing among others naming and pointing of body parts and motor imagery tasks, evaluating all forms of body representation (de Vignemont 2010), before drawing conclusions in favor of a body image disorder. Furthermore, the dichotomy between body image and body schema has been repeatedly questioned, due its vagueness in explaining the great variety of neurological disorders that can affect body representation (Berlucchi and Aglioti 2010; de Vignemont 2010). The ideas that self recognition and awareness are mapped into one single area and that body schema and body representation are independent modules are too simplistic in view of findings on body representation (Berlucchi and Aglioti 2010; de Vignemont 2010). Similarly, it may be simplistic to consider BIID as a deficit in ownership emerging solely from a SPL dysfunction.

McGeoch and colleagues results are promising, but research on BIID as a neurological phenomenon affecting body representation has just begin, and several concerns have to be clarified before accepting this categorization. In fact, many differences can be found between BIID and classical body representation disorders, such as somatoparaphrenia (Bottini et al. 2009), even though parallelisms between the two syndromes have been repeatedly indicated (Brang et al. 2008; McGeoch et al. 2011).

BIID individuals can present with left, right or bilateral amputation desires (First 2005; McGeoch et al. 2011). On the contrary, somatoparaphrenic patients only present with a sense of disownership directed to one side of the body and never with bilateral symptoms (Vallar and Ronchi 2009; Bottini et al. 2009). Half of the participants of McGeoch and colleagues study wanted their left leg removed, the other participants targeted the right leg and one participant initially reported a bilateral desire. Studies enrolling greater groups of individuals classified by means of side of amputation desire would help in clarifying the role of laterality, if there is any, and whether bilateral and unilateral desires of amputation are really part of the same disorder or, conversely, only lateralized amputation desires are part of BIID.

Another crucial issue about BIID, that differentiates it from somatoparaphrenia, is the possibility of spontaneous recovery. In contrast with somatoparaphrenic patients (Vallar and Ronchi 2009; Bottini et al. 2009), BIID individuals do not spontaneously recover but rather show a lifelong desire of amputation (First 2005). One of the patients in the study of McGeoch et al. initially wanted a bilateral amputation, while at the time of the study he reported a decreased amputation desire for the right leg, that completely disappeared after 1 year (McGeoch et al. 2011). Even though the authors reflect that this may be indicative of brain plasticity and recovery and that this condition is reversible (McGeoch et al. 2011), it is not clear what mediated recovery, as previous studies report that none of the BIID individuals recovered spontaneously, or following drug treatment or psychotherapy, but only after amputation of the desired limb (First 2005). Very recently, successful treatment of BIID with psychotherapy has been reported (Thiel et al. 2011), although this is the only known case. Interestingly, the patient of Thiel and colleagues wanted both his legs amputated, similarly to the individual from McGeoch et al. study, who recovered from the amputation desire only for the right leg, corroborating the importance of understanding the role of laterality.

In conclusion, the study of McGeoch et al. is the first attempt to provide direct evidence of brain dysfunctions in BIID and to relate it to body representation disorders. Future studies will have to replicate and extend McGeoch et al. findings to conclusively prove that BIID is a neurological rather than a psychological syndrome. In particular, it is necessary to explore all features related to body representation, and not only tactile perception, and to understand the difference between bilateral and unilateral desire of amputation.

Beyond the theoretical implications of a psychological or neurological origin of BIID (Oddo et al. 2009) and its relevance in understanding the still mysterious body representation disorders (Berlucchi and Aglioti 2010), future neuroimaging studies may help in establishing a possible and efficient treatment for this debilitating

condition. If BIID is a condition arising from a brain dysfunction, similarly to other neurological diseases such as depression (Davidson 2010), a combination of psychotherapy and drug treatments should be carefully planned, basing on recent research findings. Moreover, there is a strong debate on the ethical, functional and emotional consequences of a surgery treatment to remove the "foreign" leg (Patrone 2009). If BIID is a neurological condition of parietal lobe dysfunction (McGeoch et al. 2011; Aoyama et al. in press) or disownership syndromes (Berlucchi and Aglioti 2010), there is further evidence that a surgical treatment should not be suggested, as the outcome of less invasive techniques (such as the mirror box paradigm or the caloric vestibular stimulation) (Ramachandran and McGeoch 2007) should be tested first.

References

Aoyama, A., Krummenacher, P., Palla, A., Hilti, L., & Brugger, P. (in press). Impaired spatial-temporal integration of touch in body integrity identity disorder (BIID). *Spat Cogn Comput*.

Berlucchi, G., & Aglioti, S. M. (2010). The body in the brain revisited. *Experimental Brain Research, 200*(1), 25–35. doi:10.1007/s00221-009-1970-7.

Blanke, O., Morgenthaler, F. D., Brugger, P., & Overney, L. S. (2009). Preliminary evidence for a fronto-parietal dysfunction in able-bodied participants with a desire for limb amputation. *Journal of Neuropsychology, 3*(Pt 2), 181–200. doi:10.1348/174866408X318653.

Bottini, G., Sedda, A., Ferre, E. R., Invernizzi, P., Gandola, M., & Paulesu, E. (2009). Productive symptoms in right brain damage. *Current Opinion in Neurology, 22*(6), 589–593. doi:10.1097/WCO.0b013e328332c71d.

Brang, D., McGeoch, P. D., & Ramachandran, V. S. (2008). Apotemnophilia: a neurological disorder. *Neuroreport, 19*(13), 1305–1306. doi:10.1097/WNR.0b013e32830abc4d00001756-200808270-00011.

Davidson, J. R. (2010). Major depressive disorder treatment guidelines in America and Europe. *Journal of Clinical Psychiatry, 71 Suppl E1*, e04, doi:10.4088/JCP.9058se1c.04gry.

de Vignemont, F. (2010). Body schema and body image–pros and cons. *Neuropsychologia, 48*(3), 669–680. doi:10.1016/j.neuropsychologia.2009.09.022.

Everaerd, W. (1983). A case of apotemnophilia: a handicap as sexual preference. *American Journal of Psychotherapy, 37*(2), 285–293.

First, M. B. (2005). Desire for amputation of a limb: paraphilia, psychosis, or a new type of identity disorder. *Psychological Medicine, 35*(6), 919–928.

McGeoch, P. D., Brang, D., Song, T., Lee, R. R., Huang, M., & Ramachandran, V. S. (2011). Xenomelia: a new right parietal lobe syndrome. *Journal of Neurology, Neurosurgery & Psychiatry*. doi:10.1136/jnnp-2011-300224.

Money, J., Jobaris, R., & Furth, G. (1977). Apotemnophilia: two cases of self-demand amputation as a paraphilia. *Journal of Sex Research, 13*, 115–125.

Oddo, S., Thiel, A., Skoruppa, S., Klinger, D., Steis, N., Markowitsch, H. J., et al. (2009). Neurobiological and psychological aspects of BIID - An integrative approach. In A. Stirn, A. Thiel, & S. Oddo (Eds.), *Body integrity identity disorder: psychological, neurobiological, ethical and legal aspects* (pp. 238–245). Lengerich: Pabst Science Publishers.

Patrone, D. (2009). Disfigured anatomies and imperfect analogies: body integrity identity disorder and the supposed right to self-demanded amputation of healthy body parts. *Journal of Medical Ethics, 35*(9), 541–545. doi:10.1136/jme.2009.029256.

Ramachandran, V. S., & McGeoch, P. (2007). Can vestibular caloric stimulation be used to treat apotemnophilia? *Medical Hypotheses, 69*(2), 250–252. doi:10.1016/j.mehy.2006.12.013.

Thiel, A., Ehni, F. J., Oddo, S., & Stirn, A. (2011). Body integrity identity disorder - first success in long-term psychotherapy. *Psychiatrische Praxis, 38*(5), 256–258. doi:10.1055/s-0030-1266128.

Vallar, G., & Ronchi, R. (2009). Somatoparaphrenia: a body delusion. A review of the neuropsychological literature. *Experimental Brain Research, 192*(3), 533–551. doi:10.1007/s00221-008-1562-y.