Summary: Patients with body integrity dysphoria (BID) express a desire to amputate seemingly healthy parts of their body. A new study reveals that this condition is linked to altered functional connectivity in a distributed neural network underlying body image.

Our body is at the centre of our sense of self and personal identity. The neural mechanisms underlying our conscious experience of our body, however, remain incompletely understood. Anaesthesia of body parts or of the entire body does not lead to the experience of the body having vanished, but leads to continued experience of the form and posture of the body, even if the experienced posture may not match true posture [1]. This shows that the conscious experience of the body relies on a central representation of body form, the body image, rather than being driven by immediate sensory inputs. This body image can be clearly seen in the case of phantom limbs following amputation, in which amputees report vivid experiences of the limb continuing to exist [2]. Though the physical limb has been destroyed, the central body image remains unaffected, leading to continued subjective experience of the limb. Individuals with body integrity dysphoria (BID; sometimes called
Body integrity identity disorder, xenomelia, or apotemnophilia) show an intriguing inversion of phantom limbs, insisting that a seemingly healthy part of the body doesn’t belong and that they would feel more complete without it [3,4]. In many cases, these individuals desire to have the disowned body part surgically amputated, which they feel would make them more complete. BID raises important questions for biomedical ethics and offers a fascinating window into the nature of the body image. In a recent paper in *Current Biology*, Saetta and colleagues [5] use a combination of structural and functional magnetic resonance imaging (MRI) to show that BID is linked to a combination of structural atrophy in brain regions known to be involved in constructing the body image as well as altered functional connectivity of a larger-scale neural network.

Existing research has implicated a number of brain regions in constructing the conscious body image, notably including the superior parietal lobule (SPL; especially in the right cerebral hemisphere) and the ventral premotor cortex (vPMC). Brain lesions affecting the SPL have been reported to induce a range of disruptions of bodily experience. Critchley [6], for example, reported that some patients with right SPL lesions claimed that the entire left side of their body had disappeared. Other studies have similarly linked SPL lesions with perceived disappearance of individual body parts [7], denial of ownership of body parts [8], and lack of attention to the limbs [9]. A range of sensations related to phantom limbs have also been linked to changes in SPL processing, including referred tactile sensations and telescoping of the phantom limb [10] and there are some reports that SPL lesions can suppress the experience of phantom limbs entirely [11]. Similarly, both lesion studies of neurological patients [12] and neuroimaging studies of healthy participants [13] have implicated the vPMC in producing the conscious body image.
Neuroimaging studies investigating brain structure and function in BID have implicated both the SPL and vPMC, but have also produced conflicting findings. Whereas one study using magnetoencephalography (MEG) found reduced responses in right SPL to tactile stimuli delivered to the disowned limb [14], another study using functional MRI found increased responses in the same area [15], but reductions in the premotor cortex. Other studies using structural MRI have found reduced density or thickness of cortical grey matter in both the SPL [16] and vPMC [17]. Overall, this literature has been limited by small sample sizes given the comparative rarity of BID, as well as the heterogeneity of cases. For example, many studies have included individuals with disownership of different body parts, making direct comparisons difficult.

In the present study, Saetta and colleagues [5] tested sixteen people who desired amputation of their left leg, recruited and tested at separate sites in Italy and Switzerland. This is the largest neuroimaging study of BID to date, and critically includes a largely homogenous groups of individuals with disownership of the same body part, allowing more meaningful aggregation of data across individuals than in previous studies. In order to determine which brain areas show atrophic structure in BID, the authors used a structural MRI method called voxel-based morphometry (VBM) to compare the density of cortical grey-matter in individuals with BID versus controls. Consistent with previous reports, individuals with BID showed reduced grey-matter density in both the SPL in the right cerebral hemisphere and the vPMC in the left hemisphere. Moreover, the magnitude of this reduction of grey-matter density in the right SPL was correlated across participants with both their self-reported desire for amputation and the extent to which they pretend to be an amputee through use of crutches or a wheelchair.
More importantly, Saetta and colleagues also used analyses of resting-state functional-connectivity of functional MRI data to investigate alterations in the connectivity of larger-scale cortical networks in BID. This approach looks at the correlation between the time-series of activations of different voxels in the brain in the absence of any task, quantifying the intrinsic functional relationship between brain areas. Specifically, they used a measure called the intrinsic connectivity contrast (ICC), which quantifies for each voxel in the brain its overall level of functional connectivity to the rest of the brain. Compared to controls, individuals with BID showed reduced ICC in both the right SPL and left vPMC, as well as the right paracentral lobule containing the primary somatosensory cortex.

These results have important implications for understanding the nature of BID and its underlying neural basis. The functional connectivity results suggest that rather than emerging from atrophy in specific brain regions, BID may involve more wide-spread changes in broader neural networks. The reduced ICC in the paracentral lobule is particularly intriguing. The lack of any apparent atrophy in this region is consistent with the fact that the disowned body part in BID appears to have normal sensory function and motor control. Nevertheless, the present results indicate that sensory signals from the body may be integrated with higher-level body representations in a reduced or abnormal way. This in turn has important implications for understanding the nature of body image more generally. Studies using cutaneous anaesthesia to temporarily remove sensory inputs show rapid alteration of perceived body part size [18], showing that immediate sensory signals play an important role in shaping the body image. The current finding of disrupted functional connectivity of sensorimotor regions in BID complements such previous findings, indicating that integration of basic somatosensory processing with higher-order representations may be key to the formation and maintenance of the body image.
The present results add to a growing literature showing that many neurological and psychiatric disorders involve altered connectivity in widespread brain networks, and cannot be understood in terms of altered processing in single brain regions [19]. Altered patterns of resting-state functional connectivity have been reported in, for example, schizophrenia, autism, Alzheimer’s disease, epilepsy, and depression. BID is an intriguing addition to this list, and the parallels between BID and other disorders provides further support for the classification of BID as a genuine medical disorder [4] and its inclusion in the upcoming revision of the International Classification of Diseases (ICD-11).

Although this study is a significant advance in our understanding of BID, it also has some limitations. Despite having tested more people than any previous neuroimaging study of BID, in absolute terms the sample size remains fairly modest. This is an intrinsic difficulty in investigating conditions that, like BID, are comparatively rare and heterogeneous. The present study’s use of multiple testing sites in two countries shows the value of multi-site international collaborations, an approach which hopefully can be scaled up even further in subsequent research. Another limitation of the study is that the measure of functional connectivity used, the ICC, is relatively non-specific, showing which brain areas show reduced overall connectivity, not which specific areas they are less connected to. It will be important in future research to understand the specific neural networks involved in BID (and presumably in constructing the body image in everyone) and the factors which modulate their functioning.

One hundred and thirty years ago, William James [20] noted that although it is natural to talk about the body as something we own, our body isn’t ours, it’s us. By showing that specific parts of our body can be excluded from our sense of self, BID is an exception that proves the rule. Though the desire to amputate a limb seems intensely foreign to most
of us, the study of BID provides a revealing window into the way all of our brains generate our conscious experience of embodiment.
References


