



Can vestibular caloric stimulation be used to treat apotemnophilia? ☆

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Summary Apotemnophilia, or body integrity image disorder (BIID), is characterised by a feeling of mismatch between the internal feeling of how one's body should be and the physical reality of how it actually is. Patients with this condition have an often overwhelming desire for an amputation- of a specific limb at a specific level. Such patients are not psychotic or delusional, however, they do express an inexplicable emotional abhorrence to the limb they wish removed. It is also known that such patients show a left-sided preponderance for their desired amputation. Often they take drastic action to be rid of the offending limb. Given the left-sided bias, emotional rejection and specificity of desired amputation, we suggest that there are clear similarities to be drawn between BIID and somatoparaphrenia. In this rare condition, which follows a right parietal stroke, the patient rejects (usually) his left arm as "alien". We go on to hypothesis that a dysfunction of the right parietal lobe is also the cause of BIID. We suggest that this leads to an uncoupling of the construct of one's body image in the right parietal lobe from how one's body physically is. This hypothesis would be amenable to testing by response to cold-water vestibular caloric stimulation, which is known to temporarily treat somatoparaphrenia. It could also be investigated using functional brain imaging and skin conductance response. If correct our hypothesis not only suggests why BIID arises, but also, in caloric stimulation a therapeutic avenue for this chronic and essentially untreatable condition.

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Background

Individuals with apotemnophilia, also called body integrity image disorder (BIID), feel that the internal identity of how their bodies should be does not match how their bodies physically are. These individuals have an, often, overwhelming desire for an amputation of one or more limbs. This is more than

merely a general wish to be an amputee; rather they want removal of a specific limb at a specific level [1]. Indeed many can draw a line along the precise level of amputation that they desire.

They describe this longing for an amputation as a feeling that, although it is perfectly healthy, the limb simply should not be there and clashes with the body image constructed internally by their brain. They frequently express an emotional discomfort towards the limb and say they wish rid of it in order to feel "complete" about themselves. Usually they recall these feelings as having existed since early childhood [1]. These individuals are

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neither psychotic nor delusional; they know that these feelings are not normal. Indeed in other respects they are mentally and neurologically normal.

Such patients often become obsessed with getting rid of the limb and as a result may be very depressed. Some BIID sufferers will request the help of a surgeon. Clearly, as few surgeons will amputate a healthy limb, this request is usually refused. Sometimes these patients may then take matters into their own hands, by, for instance, freezing the limb with dry ice. They do so with the hope that they will damage it to such an extent that an amputation is then mandatory.

It is noteworthy that it has been shown [1] that most patients with BIID desire amputation of either a left-sided limb (55%) or bilaterally (18%). It is the minority (27%) who solely want removal of a right-sided limb.

Given the highly specific level of the desired amputation, the left sided preponderance and the emotion tinged description of a feeling that the limb simply does not belong; there are clear parallels between BIID and somatoparaphrenia. In this bizarre condition, which can occur after a stroke in the right parietal region, the patient denies ownership of a limb on the left side of his body- typically the arm [2–4]. Often these patients may describe the limb as “alien”, report that it belongs to someone else and develop a strong emotional abhorrence towards it.

The parietal lobes have direct connections with the limbic system [5], which is intimately involved in emotion. This may explain why in somatoparaphrenia the rejection of the limb may be so emotionally tinged. That this phenomenon can occur after right parietal damage suggests an important role for the right parietal lobe in constructing body image.

Somatoparaphrenia is itself closely- but not obligatorily- tied in with anosognosia (denial), which is also associated with right parietal lobe damage. Whereas in the former the patient denies ownership of the arm, in the latter he may deny that there is anything wrong with his completely paralysed left side. Both of these conditions have been treated successfully, if temporarily, with cold-water vestibular caloric stimulation, which involves the infusion of cold-water into the external canal of a patient’s ear [6,7]. When this is performed into the left ear, it is known that it causes activation of the right parietal lobe, along with several other areas [8].

The “standard model” in neurology has been the notion of neural connections being largely fixed at birth but in the last decade this has been

replaced by the view that the brain is a dynamic evolving system that is in equilibrium with inputs from the environment and body [3,9,10]. For example phantom pain and stroke hemiparesis can be relieved, in some cases, by the simple device of using visual feedback from a mirror (MVF) [11,12]. We recently examined two patients with severe post-stroke thalamic pain (Dejerine-Roussy syndrome), each of more than 10 years duration. In both patients caloric irrigation caused an immediate relief from pain that lasted several weeks. Placebo procedures such as tepid water irrigation or ice on the ear had no effect on pain. This is the first known case in clinical neurology of a chronic disorder being relieved by a simple non-invasive procedure. Once again, it demonstrates the extraordinary malleability of the brain especially in relation to pain and body image.

The hypothesis

We postulate that a functional disturbance of the right parietal cortex lies at the root of BIID. We propose that BIID is caused by an uncoupling of the genetically based scaffolding of ones body image, in the right parietal region, from the actual physical body parts that this area normally represents. The fact that most BIID patients date their symptoms back to early childhood would be in keeping with a genetic basis for this uncoupling.

Testing the hypothesis

We suggest two ways in which our hypothesis can be tested. The first is with cold caloric irrigation, which, as stated above, temporarily ameliorates the symptoms of somatoparaphrenia. If our conjecture is correct, one might expect cold-water caloric irrigation to, at least temporarily, alleviate these patients’ intense desire for an amputation. Such a reduction of symptom intensity in BIID sufferers post irrigation would be suggestive of a similar aetiology.

Perhaps with repeated irrigations BIID patients might come to accept the rejected limb into their body image; a novel form of hydrotherapy. We are presently initiating such a study. The hypothesis could also be tested by the use of functional brain imaging, to look for abnormalities of the right parietal lobe.

Given the notion that the desire for amputation might result from a functional derangement of the parietal lobe and parieto-limbic connections; one

could measure the skin conduction response (SCR) to the left versus right sides. With the postulated uncoupling from body image one might expect changes in SCR between the two sides.

Finally, our mirror visual feedback technique (MVF) technique could be used to convey the visual illusion to the patient that his arm has been amputated or is missing. This might provide a sort of "dress-rehearsal" for the amputation and may de-sensitise and eliminate the desire. (Although this should be done only under medical supervision, to weigh potential benefits against risks.)

Conclusions

If correct, our hypothesis will offer both a neurological explanation of, and a potential therapy for, a long neglected, chronic and currently untreatable condition. Additionally, if it is an uncoupling between one's internal body image and one's actual physical self that leads to BIID, then a clearer understanding of how this happens could cast light on how the normal brain constructs body image.

Conflict of interest

None.

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