Body integrity identity disorder crosses culture: case reports in the Japanese and Chinese literature.

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Abstract

Body integrity identity disorder (BIID) is a condition in which people do not perceive a part of their body as their own, which results in a strong desire for amputation. The disorder is likely to be congenital due to its very early onset. The English literature only describes Western patients with BIID, suggesting that the disorder might be merely prevalent in the West. To scrutinize this assumption, and to extend our knowledge of the etiology of BIID, it is important to trace BIID cases in non-western populations. Our objective was to review Chinese and Japanese literature on BIID to learn about its presence in populations with a different genetic background. A systematic literature search was performed in databases containing Japanese and Chinese research, published in the respective languages. Five Japanese articles of BIID were identified which described two cases of BIID, whereas in Chinese databases only BIID related conditions were found. This paper reports some preliminary evidence that BIID is also present in non-western countries. However, the extremely low number of cases found hampers making general statements about the biological background of the disorder. This low number possibly resulted from the extreme secrecy associated with the disorder, perhaps even more so in Asian countries.

Introduction

Body Integrity Identity Disorder (BIID) involves the desire to have a limb amputated or to be paralyzed in order to correct the perceived mismatch between a person’s sense of body identity and his or her actual anatomy.\textsuperscript{1–3} The etiology and prevalence of the disorder are poorly studied. The 100 to 200 cases described in the literature are mostly males of the Caucasian origin.\textsuperscript{1,4,5} To extend the limited knowledge of BIID, in this report we try to retrieve BIID cases in Asian literature.

Originally, the disorder was seen as a paraphilia. According to this view, the comparison of an amputation stump with a phallus would lead to sexual arousal. The disorder was named apothemnophilia, which is ancient Greek for ‘love to cut away’.\textsuperscript{6,7} In the beginning of this century the incongruence between one’s experienced and assigned body thus became paramount to the sexual desire. Parallels –the ‘not feeling well in one’s body’- were made with Gender Dysphoria, and the condition was named ‘Body Integrity Identity Disorder’.\textsuperscript{1,5,8} To date, an alternative, neurobiological hypothesis about the aetiology of this rare condition has been proposed. It has been suggested that the feeling of body ownership is mediated by a frontal parietal network, including multisensory integration areas, such as the parietal cortex and premotor cortex, as well as the insula.\textsuperscript{9,10} Structural and functional abnormalities in this network recently have been reported in BIID patients.
Currently, some neuroscientists prefer to refer to the condition as xenomelia instead of BIID, as it is a more neutral term.\textsuperscript{11,12}

Generalization of BIID features and prevalence estimations are hampered by the absence of epidemiological studies. Drawing from the 100 to 200 BIID patients described in case-reports and small cohorts we have learned that approximately eighty percent of the BIID patients is male and most of the patients have received higher education.\textsuperscript{1,4,5,13} Besides their BIID, patients are otherwise healthy individuals with no major psychiatric comorbidity.\textsuperscript{1,5,14} Nevertheless, they experience a high disease burden and there is no pharmacological or otherwise therapeutic treatment available.\textsuperscript{1,13} BIID patients recognize the bizarre character of their wish to amputate or paralyze body parts, which results in being vigilant and reserved in sharing their feelings with others.\textsuperscript{1,14}

Historically leg amputation is the most common variant of BIID, but other desired disabilities such as arm amputation, paralyzation or even blindness are proposed to be a form of BIID as well.\textsuperscript{2,15} Unlike in factious disorder, BIID patients do not aim for disability in order to gain any variety of benefit (i.e. attention, nurturance, leniency, profit) other than feeling at ease with their own body.\textsuperscript{1,5} There is no motor or sensory dysfunction of the affected body part and there is no pain, disease, conversion, psychosis or imagined ugliness that could explain the desirability of amputation.\textsuperscript{1,5,16} In some reported BIID samples, BIID is associated with a higher prevalence of homosexuality (of about forty percent) and is related to gender dysphoria.\textsuperscript{1,2,5,17} However, these prevalences could be an overestimation due to selection biases of the reported samples.\textsuperscript{1,5} The onset of BIID lies in early childhood and the main motivation for preferred body modification is that the particular body part feels supernumerary.\textsuperscript{1,5} Although familial BIID has not yet been described, it has been hypothesized that there might be a genetic background.\textsuperscript{1,18} The main rationale for suggesting this genetic background is that the disorder is likely to be congenital. Most patients with BIID have feelings of disownership from as long as they can remember, and those feelings are fixed and do not seem to be malleable by any form of therapy.\textsuperscript{1,10,18} It is uncertain whether the absence of reported familial cases results from their actual non-existence or because many patients with BIID do not share their bizarre wish with their loved ones.\textsuperscript{1}

To the best of our knowledge, only cases in Western countries have thus far been described. Internet forums have even seen speculations that all individuals with BIID might share a German ancestry.\textsuperscript{19} However, this has not been confirmed. We aimed to investigate whether reports exist on BIID cases in Asian countries, in order to gain more knowledge of the origin of this rare condition. More specifically, our aim has been to learn whether BIID is influenced by cultural factors; to learn about its presence in populations with slightly different
genetic backgrounds and to compare phenotypic characteristics across different populations. Therefore, we
performed a literature search in local medical databases in China and Japan in an attempt to find BIID patients
with an Asian background.

Methods

Literature search

To find Japanese literature on BIID, a systematic search was performed in May 2013 in Ichushi Web, which is
the largest Japanese database of medical literature. First, publications were retrieved by a computerized search of
databases using the keywords 身体完全同一性障害. (身体 shintai = body or physical; 完全 kanzen = integrity,
perfect or complete; 同一性 douitsusei = identity; 障害 syougai = disorder). Five articles were found and all of
them described patients with BIID. After reading the full text of the papers, it became apparent that some of
them overlapped. Two cases described the same patient, doubly reported by some plastic surgeons and a
dermatologist who both treated the same patient. The other three papers described another case. This case is
reported by a group of plastic surgeons in three different papers (a conference report, a regular case report and in
a review). We thus found two original cases. Second, to retrieve more papers the search was extended using
BIID related keywords: “四肢 lims and 同一性障害 identity disorder”, “肢切断術 amputation and 自已 self”,
“四肢 lims and 自傷 self-injury”, “四肢 lims and 假麻痹 pseudo-paralysis”, “四肢切断愛者
crotonomophilia”, “四肢切断熱望者 apotemnophilia”, “四肢 lims and 切断者愛好 devotee”, “四肢 lims and
偽装 pretender”, “四肢 lims and 切望 wannabe”. The extended search using BIID-related keywords revealed
no new BIID articles, not even BIID related reports.

Furthermore, to discover Chinese papers about BIID, a systematic literature search was performed in the three
main databases of Chinese articles including China Academic Journals Full-text Database (1984-2013), Wan-
fang Database (1998-2013), VIP Database (1989-2013). Initially, a search was performed using “躯体完整性认
同障碍”or “截肢者认同障碍”(body integrity identity disorder); “肢体”and “认同障礙” (limbs and identity
disorder). Through this search no BIID articles were found. Later, more keywords from Western BIID articles were retrieved. These terms were used in a second search and included: “自行截肢”(self-amputation); “截肢”(amputation); “肢体自残”(limbs self-injury); “自残”(self-injury); “假性瘫痪”(pseudo-paralysis); “慕残”or“慕残者”(acrotomophilia, devotee);“扮残”or“扮残者”(apotemnophilia, pretender); “自残者”(wannabe).

In this second, extended search, some papers were retrieved. After reading the full texts, it turned out that the cases found mainly concerned self-injury, automutilation or amputation for other reasons. For example, there was a report of a man who amputated his arm after being crushed by a boiler for three days. Other papers described a man who cut his left palm and a woman who broke both her legs during a car accident. However, in these cases it was assumed that these measures were taken in order to claim money from medical insurance companies. In the end, therefore, we retrieved two cases of possible BIID in China.

Results

The two cases from the papers found in Japanese literature were translated from Japanese to English by a bilingual psychiatrist and are summarized below.

Case 1 is the report of a young female patient seen by the article’s authors (a dermatologist, a plastic surgeon and a consulting psychiatrist) after mutilation of her left leg. At age 16 she was diagnosed with major depressive and self-mutilating behavior such as wrist cutting and cigarette burning, for which she took antidepressants. She had a ‘feeling of wrongness’ relating to her left leg, and always had the feeling that she needed to cut it off herself. At the age of 18 she laid her leg in dry-ice for several hours, after which a skin graft operation was necessary. The operation wound never healed completely, and the woman continued harming the unhealed wound. At age 21, she developed osteomyelitis, which resulted in an operative amputation of her left leg to the level above her knee. “I wanted to cut my leg from the beginning. I have a leg. That is the problem”, she argued. No official diagnosis of BIID had been made.

The second case is a 21 year-old man with a request for the amputation of his left leg after putting it in dry ice for six hours. The case is reported by the plastic surgeons who treated the patient. This patient had been experiencing a ‘feeling of wrongness’, relating to his left leg since he was a child; he adored Captain Hook and other amputees. Later on he was diagnosed with BIID by a consulting psychiatrist. The plastic surgeons opposed
his request and performed reconstructive amputations, enabling him to walk, although with some difficulties. He still had a feeling of wrongness towards his leg, but he did not want to perform self-mutilation anymore. He was somehow satisfied because he felt his state was close to what he initially had in mind.

In the Chinese literature, two cases of possible BIID were retrieved. The first one was a letter from a psychologist who helped a woman on an internet forum. This 28-year old woman had pretended from childhood on to be disabled. She used medical supplies to enhance credibility. This pretending excited her. Being disabled granted her attention from others that she had always missed from her parents. The second letter is from a patient who asked why he is so obsessed with other people with disabilities. Seeing other amputees, especially girls, excited him and made him happy. Afterwards he felt remorse. He could not understand why he was so obsessed with disabilities.

Discussion
This paper describes four BIID related cases in China and Japan. We hypothesize the patient in the first Japanese case suffered from BIID as she claimed her leg to be superfluous; her thoughts about her leg not being her own were very persistent and pharmacological therapy did not influence these BIID thoughts. One might argue that burning her leg with dry-ice can be seen as automutulative behaviour, just as the patient performed wrist cutting in the past. However the wrist-cutting seems more of an impulse-control disorder; in contrast to the mutilation of the leg, which had been meticulously planned. The second case from Japan reports explicit BIID features: the early onset, the feeling of overcompleteness and the perseverance of this thought. The patient’s adoration of Captain Hook may be either jealousy (“that is how I would like to be”) or a form of sexual attraction toward other amputees, better known as acrotomophilia. Such feelings of jealousy and sexual attraction both have been described in BIID.

The Chinese cases are less clear. Patients faking accidents in order to get medical insurance fees are often described in the literature and these behaviours are known as simulation. However, due to the secrecy and shame that often accompanies BIID, some patients create accidents in order to get their leg amputated, without letting their relatives know the true reason of the accident. The woman who pretended to be disabled is not likely to suffer from BIID. Although pretending is a common symptom in BIID, her motivation to pretend was rooted in the attention it drew and not ‘to feel complete in her own body’. The patient that mentioned getting
excited by seeing other amputees might be suffering from acrotomophilia. This condition can exist alongside BIID, although it may exist independently as well. To summarize, after a search in Japanese literature one straightforward and one likely BIID case were found, both describing the amputation variant of BIID. In the Chinese literature, only BIID related conditions are described. The reports do not give sufficient information to reach any conclusions anything concerning the background of the symptoms. The fact that only few reports were found may have several causes. It is known that patients with BIID have difficulties in sharing their BIID-related feelings with people in their environment, because they are aware of the peculiarity of these feelings. In one of our other studies including 54 patients with BIID, just half of the patients shared their bizarre thoughts with spouses, friends or family. All the patients examined in this other study were from Western countries. Chinese and Japanese cultures are known to be more reserved when it comes to self-disclosure. Therefore, letting others in on BIID might be even more difficult. Likewise, in Japan and China it is assumed to be more difficult to live with an identity that most people would judge as unusual. For example, people with a homosexual identity or feelings of gender dysphoria are less publicly accepted than in some Western countries. Therefore, it might be more difficult for people with BIID to ‘come out of the closet’. Moreover, as literature is sparse, medical professionals might not be familiar with this condition. Therefore, individuals with BIID in these countries could easily be wrongly diagnosed. This might also explain why no paraplegia BIID cases have been reported, as this specific type has only recently been described in literature.

This paper reports some preliminary evidence BIID’s presence in non-western countries. The extremely low number of cases found hampers making general statements about the biological background of the disorder. However, BIID is a rare and extremely secretive condition, perhaps it is even more so in Asian countries, which poses a major obstacle estimating its prevalence. We hypothesize that the cases found are likely to be an underestimation of the presence of the disorder. On the other hand this might be a reflection of the extreme rarity of the disorder in China and Japan. Further research is needed to reveal the prevalence and phenomenological features of BIID in Asian and other non-western countries. We hope this paper will encourage the submission and publication of additional Chinese and Japanese papers on BIID and improve the screening of BIID in those countries. This should create more awareness of the existence of BIID among local healthcare workers. Awareness could subsequently lead to acknowledgment and respect for this unusual desire, which is always a first and crucial step in the treatment of BIID patients.
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