BIID: WHAT DO WE KNOW?

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Body Integrity Identity Disorder (BIID): What do we know?

Review Article

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Abstract

Time and time again and just when we think we know everything there is to know about everything, science proves that we are still vastly ignorant to the mysteries of life that lie right before our eyes. Medicine is, obviously, not a stand-alone science, but an entanglement of different sciences whose leaders (those with highest levels of knowledge on certain topics) collaborate in an attempt to increase the amount of knowledge we possess. This is done through conferences, workshops, etc. Leaders of the psychiatric, psychological and neurological divisions of health usually meet when discussing certain phenomena. BIID is a phenomenon which requires the presence of such leaders. So, what is BIID (Body Integrity Identity Disorder)?

Key words: BIID, psychology, psychiatry, neurology, rare diseases*, *Embodied emotions, Apotemnophilia, Xenomelia, Amputee identity disorder, Body incongruence disorder, infrequently studied disorders, Voluntary amputation/palsy

Introduction

BIID is a disease in which individuals feel the need to either paralyze or amputate a limb. Some have described the limb as "foreign". BIID was long believed to be a psychological disease. Recently, however, multiple studies have driven us to a conclusion that the disease might be linked to abnormalities within the brain structure. It is important that we denote that patients with BIID seem to show any psychiatric symptoms and are 100% aware of what their wish might sound like to others [1]. Here, we will attempt, to the best of our abilities, to explore this phenomenon and enlighten readers on what the most recent studies on the disease have concluded. Up and until 2012, BIID was not included in both the ISCD11 and the DSM IV [9]. This, as expected results in decreased awareness of the topic by surgeons, psychiatrists, psychologists, neurologists, etc. [9], what makes this even worse is that patients tend to avoid, rather than seek, healthcare and will often fulfill their desires individually [9,21-24] With that being said, we wish you an informative experience reading this article. As expected, some link the disease to supernatural causes which could be the reason why we weren't able to come across any statistical analyses of the disease in countries where certain religious/traditional practices prevent individuals from seeking medical advice. In this article we will be covering multiple aspects including, but not limited to,

1) **How do patients fulfill their desire(s)?**
2) **Coping with BIID**
3) **Epidemiology/awareness of BIID**
4) **What causes BIID?**

5) Desire for amputation?
6) Reports following amputation
7) Side effects of BIID
8) What have imaging studies of the brain shown?
9) Childhood and BIID
10) Is the disease neurological or psychiatric?
11) Treatment
12) Conclusion

Before we continue it is important that readers are aware that BIID and BDD (Body Dysmorphic Disorder) are not the same thing {1}. We will not be discussing BDD in this article whatsoever. In 2011, McGeoch and colleagues termed this syndrome "Xenomelia" {2}. It is never easy, next to impossible in fact, to pinpoint the exact time at which a disease (or any fact of life) made its first appearance. What is easy, however is identifying when we, as a species, identified a condition as a disease and gave it a name. Unfortunately, we were unable to come across conclusive, trustworthy facts that determine exactly when the first case was reported. Fortunately, though, we do have a better understanding of the disease now than ever before. This disease has resulted in quite a controversy amongst medical professionals as we shall see in upcoming paragraphs. In a 2012 study, Ramachandran implied that BIID patients present the opposite of patients who have suffered accidental amputations who sometimes report feeling a "phantom limb" {14}. With the focus being directed towards the amputation aspect of BIID and avoiding the desire for disabilities, the latter have been understudied, so strong in fact that as recently as 2012, they were never looked into {9}, we are not aware of any studies being held with that respect after 2012 as well. Keep in mind that some might still refer to the disease as *apotemnophilia*, which is the old name of the disease {12}. Also keep in mind that, in the future, there is a possibility that BIID will be re-termed *xenomelia* {12}.

How do patients fulfill their desire(s)?

As humans, we all have wishes and desires we wish to fulfill to the highest extent possible. Patients with BIID have the desire to be amputees/impaired. So (other than actually amputating their limbs), how do they fulfill these desires. They fulfill and stimulate this desire by "pretending" {1}, which is done by using a wheelchair or crutches {8}, we believe that these acts somehow further increase their "need" for an amputation/ paralyzing of their intended limb(s).

Coping with BIID

BIID is very difficult to deal with, especially since 1) patients are aware of how absurd their desire sounds to others and 2) low levels of public and professional awareness in respect to the topic. Previously, we have discussed methods which patients use in an attempt to fulfill their desire. We believe that by doing so, they increase their desire to actually become amputated /paralyzed which makes coping with the diseases even harder. Surgeons will usually reject performing such an operation, this sometimes leads to the patients choosing to self-amputate the involved limb (e.g. with a chainsaw) [1]. Discussions are currently taking place in order to determine whether or not allowing such operations could help [1].

Epidemiology/ awareness of BIID

It is extremely difficult, if not impossible, to accurately estimate a population's knowledge of a certain topic. Nevertheless, it is agreed upon that BIID is rare. In fact, in one study in which 680 people were chosen randomly, only 1 has been found to have the disease [1]. Sadly, awareness in respect to the disease was not much better. In a 2012 study, 7.9% of the general population were found to be, at least, vaguely aware of the topic and 2.1% have reported actually experiencing the desire multiple times (at least twice) [3]. Other studies proved our hypothesis that even medical professionals found difficulty in identifying, and thereby diagnosing BIID [4,27]. There is currently an ongoing debate in regards to whether or not the desire for chronic disease and loss of sense should be categorized under BIID [5,6,7]. In most studies, (e.g. [9]) patients were mostly Caucasian which could be a correct representation, but is, unfortunately, of no use to us since there are not enough worldwide studies on the topic to form a conclusion. Different studies (e.g. [1,9]) have shown that the disease had a higher count of homosexuals and bisexuals than the average population, Stirn [25] had proposed a higher incidence of homosexuals than was found, however their hypothesis that a higher percentage of homosexuals and bisexuals were present in BIID forums than the population remains true. Multiple studies (e.g. [11]) have proven that most patients are male, the etiology behind this is, as far as we can tell, still unknown.

What causes BIID?

The cause of BIID is thought to be a misrepresentation of the body in the brain [1,9], how and why that occurs, however, is a different discussion to which n true answer is yet available. It is also thought that certain childhood experiences can cause the disease [10]. It seems as if the right posterior insula is an important structure in patients who lack proper sensation of their limb/ ownership of said limbs of limb, some case-

studies have reported patients with alien hand syndrome and asomatognosia with single lesions in the corpus callosum or premotor cortex, respectively {13} which makes us believe that it is possible that more than 1 area of the brain is involved. McGeoch et. al. found absence of representation of the involved limb in the superior aspect of the parietal lobe {2}. BIID symptoms highly mimic those of somatoparaphrenia {9, 17-20}. This, accompanied with the early onset, leads us to believe that BIID could be congenital {9, 17-20}. A hypothesis, made by Rianne M. Blom et al in 2012 {9}, which we agree with is that BIID might be of multigenic origin.

Desire for amputation?

If one thing is for sure, it is that BIID has an early onset. Some studies suggest as early as 6-7 years in men and 3-4 years in women {12}. Patients usually find themselves unable to pinpoint the motive(s) behind their desire, instead they usually responded with "cloudy statements" {1}. Some patients reported feeling sexually aroused, thereby marking an erotic "fetish" towards BIID {1}. Strangely enough, a 2012 study by Rianne M. Blom et. al. showed that a change of preference to site of amputation may occur, this happened to 5 of their study participants {9}. The same study also showed that a vast majority of patients wish for the procedure to take place on the non-dominant side of their bodies {9}.

Reports following amputation

Now that the patients have satisfied their "need" to lose/paralyze a limb, a question arises to whether they feel satisfied or would like to repeat the process (i.e. lose another limb). In a 2014 study by Sarah Noll and Erich Kasten none of the participants involved regretted having the operation, reported being much happier with their bodies and were found to have significant psychiatric improvements (depression, anxiety, etc.), they also reported **not** feeling handicapped following the operation {1}. The patients also reported some disadvantages following the procedure {1}, but were minor resulting in an overall increased state of happiness. Müller {15} proclaimed, it would be reprehensible to amputate healthy body parts when there is an efficient alternative therapy. O. Sacks {16} reported a possible method of therapy which involved the use of music therapy to reach a state of acceptance towards his leg.

Side Effects of BIID

Other than those of the disability itself, BIID seemed to have no side effects after the procedure took place [1]. The enhancement of their quality of life following the procedure [1] is a sure indicator that the principle side effects were those that had an adverse effect on quality of life (depression [1,11], etc.) which further augments our desire for further research and acknowledgement on the topic.

What have imaging studies of the brain shown?

As we hypothesized, imaging studies have shown involvement (structural abnormalities) of the insular cortex [12]. Areas responsible for body representation in the brain, such as the insula and parietal lobe have been found to be impaired in individuals with BIID [12]. Different studies have shown the insula to play a role in body representation, namely awareness [28] as well as processing of disgust [29]. A 2016 study [13] concluded that other anatomical structures such as the premotor cortex and cerebellum play an important role in *"the experience of body ownership"*. The same study showed decreased amounts of grey matter volume in the LD and LV premotor cortices and an increase in grey matter volume in the cerebellum in individuals with BIID. We should be aware that similar cases can present in the hospital following a stroke which has effected the parietal cortex, in such cases patients may ask their attending physician (or nurse) to 'rid them' of the 'alien' leg.

Childhood and BIID

We have previously touched on the topic that BIID might be related to certain childhood experiences. In this division of our article, we shall focus on that point in hopes of achieving a better understanding of the topic. All of the articles we have reviewed, that touched on the topic, agreed that BIID has an early onset (childhood to adolescence), studies on BIID and youth are very few and far in between [10] as is the case, unfortunately, with BIID as a topic. Catharina Obernolte et. al. in a 2015 paper [10] remarked that parental malpractice and childhood trauma (such as emotional neglect and sexual abuse, respectively) was almost identical in both their control group and BIID patients. All in all, the study concluded that BIID patients and controls lead almost similar childhoods, thereby further endorsing a neurological origin of the disease rather than an aftermath of psychological experiences. What was remarkable though was that, the same study showed, patients with BIID had more vivid memories of severely-ill and amputees/ palsy patients. This study proved a hypothesis put forward by Stirn et. al. [26] in 2010 that stated a pre-pubertal genetic imprinting of the desire.

Is the disease psychiatric or neurological?

Unfortunately, we were unable to come at a conclusion to an answer to this question. The answer which we were able to feel contempt with is *both* (i.e. the disease is both neurological as well as psychiatric), this means that the experts from both fields should come together in order to arrive at a proper treatment for the disease. A 2015 study by Bottini, G [12] only increased to our confusion on classification by stating *"Importantly, emotional impairments have been found in psychiatric disorders, and a psychiatric aetiology is still a valid alternative to purely neurological accounts of BIID."*. All in all, it is not greatly significant how we classify the disease as long as we actually study it and work towards finding a treatment.

Treatment

As is the case, unfortunately, with many other diseases there is no one "golden" treatment for BIID. In individual studies, therapy (e.g. antidepressants, behavioral therapy, etc.) has been found to have a positive effect in reducing the strange urge experienced by these individuals, but were never found to be 100% successful in treating the disease [1,9]. We were unable to come across a widely-accepted treatment for BIID. Surgery is seen, by most, to be the only effective method of treatment [11]. No treatment, or effect of treatment can successfully take place without the examination of "successful wannabes" (individuals who have succeeded in becoming handicapped [1]. It is important that the medical community either finds a cure or legalizes surgeries worldwide in order to avoid patients deliberately injuring themselves in an attempt to lose/paralyze the involved limb. On a related note, therapy (psychotherapy, behavioral therapy, psychopharmacological medications, etc.) seemed to have little to no effect in oppressing the desire in patients with BIID [1]. Sarah Noll and Erich Kasten asked their participants if they would like to undergo another surgery (due to the argument against legalization that states that patients would want even more surgeries done), most participants said they were happy and desired no more surgeries [1].

Conclusion

It is, undoubtedly, truly saddening what these individuals are dealing with on a day-to-day basis. It is our responsibility, as medical personnel, researchers and even patient to act in order to stop the suffering these individuals are undergoing 24/7. We

hereby request all governments increase funding to not only the research of BIID, but all research aimed towards positively impacting human life. It is also extremely saddening that we were not able to come across any studies with respect to the topic in Africa, the Arab World and Eastern Asia. We hope that there are researches on the topic and that us not finding any data was merely our fault. We also want to use this opportunity to request different departments (ministries) of health around the world to help these individuals and for society to respect their wishes and desires so that they would be more open towards conversing the topic to others. With that being said, if you or someone you know suffer or think you might be suffering from this disease, seek help and tell you doctor exactly what you are feeling, what you think you might have and why you think that.

- Disclaimer: We did *not* cover the emotional aspect of the disease; we have an article (currently under publication) completely devoted to that topic.

Abbreviations

1) BIID = Body Integrity Identity Disorder
2) BDD= Body Dysmorphic Disorder
3) DSM= Diagnostic and Statistical Manual of disorders

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