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Investigation of the Syndrome of Apotemnophilia and Course of a Cognitive-Behavioural Therapy

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Key Words

Apotemnophilia · Body integrity identity disorder · Amputee · Behavioural therapy

Abstract

Background: The syndrome of apotemnophilia, body integrity or amputee identity disorder, is defined as the desire for amputation of a healthy limb, and may be accompanied by behaviour of pretending to be an amputee and sometimes, but not necessarily, by sexual arousal. Sampling and Methods: A case history is presented of a 35-year-old man who was referred because of his desire for amputation of his left leg, without sexual connotations. The course of a combined cognitive behavioural psychotherapy with SSRI treatment is described. Results: Symptoms showed considerable similarity with obsessive-compulsive disorder, and some similarity with body dysmorphic disorder according to DSM-IV, but the core symptom seemed to be strongly connected with a sense of identity. Treatment with a selective serotonin re-uptake inhibitor decreased levels of distress only. The effects of cognitive restructuring of the psychotherapy were limited, whereas the behavioural elements substantially reduced the behaviour of pretending to be an amputee. Conclusions: The rare syndrome of apotemnophilia raises unresolved questions of classification. Psychotic disorders should be ruled out carefully. The model designed in the current cognitive behavioural approach may serve as a starting point for further development of intervention protocols for this rare disorder.

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Introduction

The term apotemnophilia (from ancient Greek: $\alpha\pi$ o-, from; τέμνειν, to cut, and φιλία, love) has been introduced by Money and colleagues in 1977 [1]. It denotes a syndrome or symptom complex in which a person is preoccupied with the desire to amputate a healthy body part. In most instances, the preoccupation entails the amputation of one's own arm or leg, which is also the phenomenon of study in the present report. The preoccupation may be directed to other people who miss a limb: this 'allo-apotemnophilia' is also described as acrotomophilia (άκρον, extremity; τομή, a cutting; and φιλία, love) [2]. Since both types of the desire, directed to oneself or to another person, can be accompanied by erotic fantasies, the first descriptions of this phenomenon emerged in the literature on fetishism [3]. Nevertheless, apotemnophilia is also encountered without sexual connotations.

Bruno [4] describes three subtypes of presentation of apotemnophilia, for which he reserves the label 'factitious disability disorder'. The first subtype is represented by 'wannabes', those who long for a physical handicap, ac-

cording to their sense of identity, sometimes going to extraordinary lengths to have a limb amputated. Next, 'pretenders' are those who act as if they have a disability by using assistive devices (e.g. using a wheelchair or walking with crutches). Finally, 'devotees' are those who feel sexually attracted to people with disabilities and especially amputees. This last subtype matches, in fact, the phenomenon of acrotomophilia.

To date, several case reports appeared in the literature [1, 4–9]. Attempts are made to introduce a more comprehensive definition of the syndrome, described as 'body integrity identity disorder' [7] (website www.biid.org), suitable for the DSM-IV classification, instead of classifying the syndrome as a paraphilia or body dysmorphic disorder. With respect to the epidemiology of apotemnophilia, only some assumptions are available [7]: the condition is rare, and accurate numbers for the occurrence in the general population are undetermined. Males seem to be more likely to be affected than females, but the ratio is not defined.

We are presenting a case history of a 35-year-old man who was referred for diagnosis and treatment to our academic outpatient clinic because of a sustained wish to amputate his left leg, without substantial sexual connotation. In addition, we discuss the diagnostic considerations and the course of a combined cognitive behavioural psychotherapy with treatment with a selective serotonin reuptake inhibitor. The term 'apotemnophilia' may be regarded as old-fashioned. Although there are alternatives, such as 'factitious disability disorder', 'body identity disorder' or 'body integrity identity disorder', these terms do not appear in the literature. The only term that has been used more or less consistently in the scientific papers or reports so far is apotemnophilia, which is therefore also used by the authors of this contribution.

Case Report

Case History

The essential complaint of the patient concerns his permanent desire to amputate his left leg up to 15 cm above the level of the knee. He has no explanation for this desire, other than that his leg feels totally unnecessary. The patient compares his problem to transsexuality, in which a similar mind-body discontinuity is encountered. The role of being a disabled person is regarded by the patient as paying justice to his real self. There is hardly any fluctuation in the intensity of his desire, although his thoughts about it slightly diminish during intensive work. He remembers that his desire emerged when he was about 9 years of age. The only way he discovered to cope with his continuing thoughts about amputation is that he feels more comfortable at home when he doubles his left leg in

his trousers. He attaches his doubled leg with the aid of a sport bandage. In this way, he looks like an amputee, and he uses crutches to move. He wears the bandage as frequently as possible, although he is only able to do this inside his house or during distant holidays. He thinks that he would function better in daily life when he could walk with crutches the entire day. He expects his life after an amputation will be less stressful, without his preoccupations and that he will be able to concentrate better, perhaps start an additional education, spend more time on hobbies or work as a volunteer.

For the patient, there is no sexual correlate for his complaint. He characterises himself as a sober-minded and straightforward person and feels a bit embarrassed with the fact that many others who can be traced with apotemnophilia underline the sexual meaning of their condition. He considers his sexual life satisfying, and he neither recognises a homosexual preference nor any particular sexual interests, such as types of fetishism.

Through a television programme, and later via the internet, he discovered that there are other people who suffer from this problem, and he contacted some of them by e-mail abroad. Some of them had succeeded in obtaining surgery for deliberate amputation. Some years ago, he informed his wife about his desire and she was able, after some time, to accept his problem. He promised her that he will not attempt to harm himself or to have his leg amputated in a third world country.

He is reluctant to inform his friends and family on his desire, not only because of his worries that they will no longer accept him, but also because of feelings of shame and guilt towards those who are handicapped after an amputation. Especially the necessity to hide his personal secret, because of his feelings of shame and fear of negative reactions, is bothering him frequently. Altogether, these worries and the binding up of his leg are time-consuming, and he is less concentrated by the ruminations.

About a year ago, he informed his general practitioner, who referred him to a psychiatrist in another centre. This psychiatrist confirmed the diagnosis of the syndrome of apotemnophilia, classified the symptoms under the DSM-IV category of body dysmorphic disorder and referred the patient to our centre which is closer to the region where the patient lives. Apart from this contact, he had never had contact with mental health care services before, and the same holds for his family members. Nevertheless, he typifies himself as a nervous person. The patient sought medical advice to increase his understanding of his problem. Furthermore, although he can hardly imagine that he might get rid of his wish of amputation, he is willing to try psychotherapy.

Outline of Biographic Information and Current Social Situation

The patient was born in a family living in an urbanised region of the Netherlands. He has a 3-year younger sister. He had a fairly good relationship with his father, suddenly ending after the divorce of the parents when the patient was 12 years of age. For the patient, it was unacceptable that his father had been involved in an extramarital affair for over 3 years, and he refused to see his father any longer. During treatment he restored contact with his father and his father's new wife. He describes his mother as a loving, caring, but also vigorous and disciplined person. After the divorce, she had contact with her son as a confidant, which stopped 3 years later, when she started a new relationship. It took the patient some time to adapt to this, but eventually he got attached to his mother's new husband.

Basic education was without problems, except that the patient, when he was about 10 years of age, was teased by one boy for over a year: how he walked and how he looked like. However, no physical injury occurred, or any trauma related to amputation. The patient passed technical education at intermediate level, and later through study in the evenings at high level. He had good friends in this period. He found work in manufacturing industry and made further career as a manager.

When he was 19 years of age, he had his first relationship, which lasted 9 months. With this girl, he experienced his first sexual relationship. Six years later he met his wife, who is about 10 years older. He is very content about their relationship. He even felt more comfortable in his relationship after he told his wife about his struggle with the desire to have his leg amputated. Because of his complaint, the patient and his wife decided not to have children.

Mental State Examination (Summary)

The patient presents with an entirely normal appearance and makes contact adequately and friendly. He shows increased gesticulation and some restlessness when explaining his main problem. There is a pervasive sensation of depersonalisation with respect to one limb. The pattern of thinking is coherent and the speed of thinking undisturbed. The contents of thoughts are determined by the preoccupation with the desire to amputate the left leg and the delusion-like conviction that the left leg is superfluous. His mood is slightly anxious, whereas his affect is modulating normally. There seems to be some degree of social isolation. The patient reports compulsion-like behaviour of binding up his left leg with sporting tape and to walk with crutches, but he does not show this behaviour during examination or follow-up.

Somatic Evaluation

The somatic history contains no particularities or relevant complaints. Neurological examination did not reveal deviations regarding muscle tone or power, sensation, reflexes or co-ordination. His gait is symmetrical, but slightly wide based. He had no problems in initiating movements with his left leg, nor impaired sensory perception, nor decreased awareness of presence or function of his leg, as may be observed among patients with hemiplegia following a stroke. The patient gave up smoking (40 cigarettes daily) 2 years ago. He consumes about 14 alcoholic beverages weekly. He has never used other drugs or psychoactive substances.

Additional Examination

No anomalies were observed in a structural MRI scan of the brain. Particular attention has been paid to the right-sided sensorimotor cortex, parietal cortex and the internal capsule, but no lesions could be detected. Electromyographic examination revealed a lower amplitude in the left peroneal muscle compared with the right (0.3 vs. 3.2 μ V) and a slightly longer latention time, but the findings fall within normal ranges.

Diagnostic Considerations

The symptoms reported by this 35-year-old patient seem to be exclusively conferred to his feeling that his left leg is unnecessary. According to DSM-IV, diagnostic options to classify this sign are: bodydysmorphic disorder (BDD), delusional disorder (somatic type), obsessive-compulsive disorder (OCD), impulse regulation disorder not otherwise specified and paraphilia not otherwise specified.

With respect to BDD, there is indeed an obsession with physical appearance. Nevertheless, the patient's complaint is not about a defect in appearance, as he clearly states the opposite, namely to lack a defect in appearance. Another passing similarity with BDD concerns the feelings of social embarrassment. There is, however, no social embarrassment with his current physical appearance, but with his convictions about his physical identity as an amputee.

A somatic delusion is de facto not the case: the patient shows marked insight about his desire to amputation, which contrasts with general perceptions of the intact body. Moreover, he is very well aware of the values attached to that.

The phenomenology of the complaints and clinical impression come close to the diagnosis of OCD, both with a continuous obsessive (the desire for amputation) and repetitive compulsive component (to pretend to have an amputated leg). Especially the compulsive component is associated with feelings of anxiousness and dysphoric mood, when there is no opportunity for the pretending behaviour. Nevertheless, preoccupations with the body are ruled out for this diagnosis in the DSM-IV classification.

Longing for amputation might be classified as an impulse regulation disorder, with a violent, auto-mutilating content. Cases in the literature may fulfil the criteria, although the current case history does not.

Finally, the paraphilia may be an option for other patients with similar complaints but who experience in addition a sexual fascination with amputated extremities.

Summarising, we conclude that the complaint under question fits somewhere in between the OCD and BDD, but that the current DSM-IV classification does not exactly cover the syndrome of apotemnophilia.

With respect to axis II, there are no obvious signs for a personality disorder. Screening for neurological deficits did not provide any evidence of anatomic or functional abnormalities.

Biographically, there might be some overlap between the period of developing or discovering his wish for amputation on the one hand, and the hidden extramarital relationship of his father and after its disclosure, the subsequent loss of contact with his biological father on the other hand. The patient, however, does not feel that this could explain his problem, and further probing psychodynamic interpretations seems to be farfetched. Another biographical issue may be more relevant: being teased by a fellow schoolboy about his posture and gait. Nevertheless, the patient does not seem to suffer from these memories.

From a behavioural point of view, the patient developed a remarkable way to cope with his burden by his behaviour to pretend to be an amputee. This habit originated from the time he was a schoolboy in elementary education, when the patient was under some pressure by negative comments on his gait and appearance by another schoolboy. This might have interfered with the development of identity formation. Some of the themes listed above are also encountered in the course of the cognitive behavioural therapy.

Course of Pharmacotherapy

The anxiolytic drug oxazepam 10 mg 1–2 doses daily was started during the stressful period of help-seeking and disclosing to mental health workers, the personal preoccupation with the desire to amputate the leg. The patient felt some decrease of tension. Later on, briefly after the start of the cognitive behavioural therapy, a selective serotonin reuptake inhibitor, paroxetine 20 mg was start-

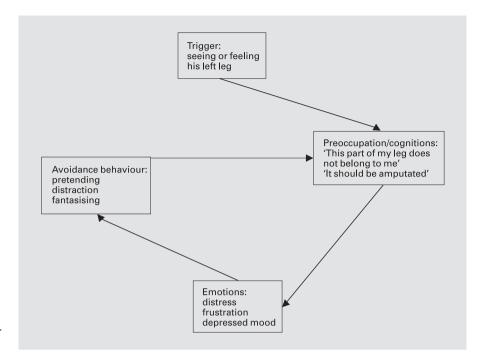


Fig. 1. A cognitive behavioural model for apotemnophilia.

ed because of ongoing feelings of distress, accompanied by restlessness, early awakening and dysphoric mood. The treatment was effective against these symptoms. Medication was prescribed by the general practitioner. Although the medication helped against stressrelated feelings, the intensity of the preoccupation with the desire to amputate the leg was unaffected. The patient stopped the medication for a period of half a year, but during al longer holiday, in which he had enough time to be preoccupied with an amputation, he became increasingly distressed and dysphoric. Therefore, he started the medication again in the same dosage. The distressed and dysphoric mood diminished, but the preoccupation with an amputation remained.

Course of Cognitive Behavioural Therapy

After consultation of several psychotherapists, nationally and internationally, who had at least to some degree experience with the syndrome of apotemnophilia, it was decided to offer the patient cognitive behavioural psychotherapy. Until now, no successful psychotherapy for apotemnophilia has been reported, but we tried to reformulate treatment models of OCD [10] and hypochondriasis [11, 12] to a treatment model for apotemnophilia. On the cognitive level, the ruminations about amputating the leg were considered as obsessive egosyntone ruminations, which cause distress, dysphoric mood and frustration. The distress diminished in the short term by active avoidance behaviour, like pretending to be an amputee, or fantasising about an amputation, or distraction by hard work or specific music (figure 1), but in the long term the preoccupation with the amputation increased.

The psychotherapy was conducted by a psychotherapist, who is experienced especially in the field of hypochondria and OCD. The patient was instructed about the principles of cognitive behavioural therapy and told that there were no documented treatment protocols with this type of complaints. The patient consented to this (experimental) treatment proposal. The frequency of sessions was once in 3 or 4 weeks. The first sessions especially concentrated on motivating the patient and reaching consensus on the treatment model and the treatment goals. For the sake of treatment, the patient agreed to consider his wish for amputation as obsessive ruminations, although he stressed that there were significant differences with obsessions and compared his wish of amputation with a transsexual's wish to have a gender change. The therapist and patient agreed that the goal of the treatment would be diminishing the distress caused by the ruminations, and not a preparation for amputation, which took the patient some effort to accept.

Until now the psychotherapy consisted of 30 sessions. At the beginning, a function analysis in which the triggers, cognitions, avoidance behaviour and consequences were formulated, was discussed with the patient.

Triggers. Most of the time, triggers for an increase of distress are vague or unclear to the patient. There is a relation to rest and relaxation. The patient has then sufficient time to ruminate about his unfulfilled wish for an amputation. This happens during holidays, but also in daily life, for instance during taking the dog out. There is also a relation to an increase in distress in other areas of his life. For instance, increased stress in his work resulted in a more dysphoric mood accompanied by an increase of frustration about not having an amputation. In general, the patient has relatively poor insight in direct triggers for his distress and ruminations.

Cognitions. The main element in this function analysis concerns the obsessive rumination that the leg is not a part of his identity and should be amputated. He has a standard number of automatic cognitions and questions related to this rumination: 'If I am amputated then I will be: (a) happier, (b) able to concentrate better and (c) able to do things I cannot do now like education, hobbies and

voluntary work'. And the recurring questions are: 'Why me? Why does nobody help me? When will be the moment?'

Emotion. The emotional reactions concern distress, frustration, dysphoric mood and disappointment.

Behaviour. The avoidance response is constituted by active avoidance behaviour of pretending to be an amputee, by binding his leg, walking with crutches and sitting in a wheelchair. Thinking through the possibility of never having an amputation is avoided. Avoidance by distraction of the preoccupation by working hard or listening to favourite music seems to have some short-term positive effects.

Consequences. The consequences of this behaviour are clear. In the short term, there is a reduction of distress or a short period without ruminations. The long-term consequences are intense. First of all, there was an increase in the preoccupation over the last years. This has consequences for his personal life, like not having children. He is not initiating activities like hobbies, education or voluntary jobs.

The cognitive element seems to be the most crucial in the above analysis. Therefore, the focus of treatment was on cognitive interventions first. During cognitive restructuring, the patient was asked to explore other ideas about his leg, such as the advantages of having and using it. According to the opinion of the patient, however, the advantages of his leg were very limited and the disadvantages of an amputation were fairly relative. Even a behavioural experiment of interviewing a friend's friend, who had an amputation after a car accident, did not alter his ideas about the disadvantages of an amputation. Alternative ways of seeing his leg as an undesired part of his body, and trying to experience grief and acceptation with that situation was met with his strong resistance. The prospect of the possibility of amputation seemed to provide hope and meaning in his life. Exploring cognitions about the future if an amputation would never take place, caused a lot of distress for the patient, and he refused to deeply explore the emotional consequences.

Meanwhile, on his own initiative, the patient reduced some of the negative consequences of his secret desire, and informed his close family, as well as some of his colleagues (living with his secret also caused a lot of distress). This resulted in comforting reactions, more than he had dared to expect, and lessened the feeling of isolation. Nevertheless, his pretending behaviour was not shown to others than his wife.

After a number of sessions with cognitive restructuring, more behavioural interventions were added. The principle of responseprevention was explained. Thereafter, the patient showed considerable effort in his exercises to reduce the amount of time spent to pretending he was an amputee. He discovered that the pretending behaviour could be diminished to only twice a week in the morning hours, before he set off to work, and could be omitted in the evening hours without an increase of distress. This created more leisure time, which was experienced as an advantage. Nevertheless, the ruminations that the leg did not belong to him were still pervasive. Strategies to diminish the ruminations were further explored. Being very busy with work and being absorbed by favourite music lessened the intensity of his ruminations considerably. Because the ruminations normally intensify when he is at home, this daily situation was not really keyed as a special situation of exposure. Other distracting strategies are now explored and also the possibility of mindfulness acceptance strategies: the ruminations are events in the mind and not facts. Currently, the treatment proceeds on a less frequent basis.

Evaluating these first 30 sessions, it is concluded that the obsessive ruminations that the leg did not belong to the identity were resistant against normal cognitive challenging. The patient was reluctant to explore a future life with a complete leg, without amputation, in order to work through a grief reaction. He agreed, however, with the problem definition, that he was suffering from the distressing preoccupations besides his wish for an amputation. The psychotherapy therefore focused on these recurring preoccupations and their consequences. Elaborating on the consequences of his behaviour provided more insight into the mechanism of distress reduction and cleared the way for exercises with response prevention, which yielded, so far, at least partial success. The current psychotherapy kept an experimental character, and was not carried out in a systematic way as is normally achieved in behavioural therapy. For example, the exercises were not guided by tape recording, and registration was only used until the functional analysis was formulated. The lower level of distress persisted at least over the 16 months since it had been achieved. The compelling desire to explore the possibility of amputation lessened to a certain extent, but the patient by no means ignores his condition, and seems to feel more free to communicate about his identity as an amputee with others.

Discussion

The syndrome of apotemnophilia was recognised in the current case report, especially by the obsessive thoughts, identity questions and distress-reducing behaviour of pretending to be an amputee. The phenomenology is close to that of OCD, and there is a passing similarity with BDD, but apart from this there is a connection with an identity disorder that is difficult to ignore. Researchers in the US advocate the introduction a new category of body integrity identity disorder. This may increase the sense of recognition by patients, which may also lead to more communication about this unsettling syndrome. It should be emphasised that in the current case report there was no sexual connotation with the state of being an amputee, and that there was no acrotemnophilia. Therefore, the debate on the overlap or comorbidity with other disorders will not end, at least as far as it concerns paraphilia not otherwise specified.

There may be disadvantages of a new diagnostic definition as well. One could imagine, for example, that patients who chronically suffer from a dysmorphophobic delusion or from imperative acoustic hallucinations with the content of cutting off a limb may feel attracted to the problem definition of an identity disorder. This may obstruct the opportunity of psychoeducation, and could lead to higher risk of self harm. Furthermore, the DSM classification is meant to be descriptive. Emphasising the identity component may lead to causal interpretations

which have not been proven either psychologically or biologically.

Across the boundaries of psychopathological categories, one may question whether apotemnophilia can be understood from a dimensional point of view. Both the compulsive behaviour and the compelling desire of amputation might fit into the 'OCD-spectrum disorder' model introduced by Hollander [13]. According to this model, a wide range of disorders is related to OCD, sharing the occurrence of repetitive behaviours with underlying serotonergic dysfunction. This range of disorders is marked by an impulsive, risk-seeking pole on the one hand, and a compulsive, harm-avoiding pole on the other.

A point of concern is the reaction of the therapist on the cruel and awesome desire of the patient. The patient in the current case report asked the first examiner not to fall from his chair, before he started to reveal his problem. The intuitively unnatural phenomenology requires respect, patience and sufficient distance. The formulation of a problem definition proved to be constructive in the collaboration, and was also a key in the course of the therapy.

Therapists may choose ways to facilitate acceptation with the identity problem, and a suggestion for future work is to adopt a paradoxical handicap model, namely, that the patient has to cope with a redundant limb, and in turn may experience disadvantages of, for example, time-consuming coping behaviour. The current report of the cognitive behavioural therapy may serve as a point of reference to develop similar or new psychotherapeutic interventions.

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