

INVESTIGATIVE REPORT

Hypochondriasis Circumscripta: A Neglected Concept with Important Implications in Psychodermatology

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This article is devoted to a psychodermatological disorder with self-destructive behaviour – hypochondriasis circumscripta. Presented data are based on a clinical analysis of 22 consecutive cases (15 women; mean age 56.1 ± 12.6 years) observed in the dermatologic department of First Moscow State Medical University and managed in a multidisciplinary approach by dermatologists and a consultation-liaison psychiatrist. Psychopathology, clinical presentations, historical aspects and treatment options are discussed. The self-inflicted skin lesions result from a severe repetitive autodestruction of focal skin loci primarily affected with heterogeneous sensations associated with a hypochondrically over-valued idea. Hypochondriasis circumscripta is a serious diagnostic and treatment challenge and should be distinguished from dermatitis artefacta, skin picking disorder and delusional infestation. Key words: hypochondriasis circumscripta; autodestructive behaviour; self-destructive behaviour; dermatitis artefacta; factitial dermatitis; dermatitis factitia; psychodermatology.

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In dermatology, skin-focused autodestructive or self-destructive behaviour (1, 2) in general is attributed to dermatitis artefacta (factitial dermatitis, dermatitis factitia – L98.1, ICD-10 (3)) or neurotic excoriations (skin picking disorder – 698.4, DSM-5 (4)), which are classified as primarily psychiatric conditions (5) that cause self-inflicted skin lesions (6). Unconscious or semi-conscious motivating factors due to dissociation, e.g. la belle indifference (7), linked with assuming the role of the sick are typically mentioned as a psychological base for skin self-mutilation in dermatitis artefacta (5, 8, 9). Similarly, obsessive-compulsive and/or impulse-control disorders are considered as a mental cause for conscious self-destruction in neurotic excoriations (10–12). It is generally accepted that psychotic con-

ditions (e.g. delusional infestation (13), that may also present with skin lesions due to skin picking in a search for non-existing parasites) are excluded from dermatitis artefacta and skin picking disorder group and contribute to a separate clinical entity (delusional disorder, somatic type – F22, ICD-10 (3)). However, there is an evidence of casuistic cases of autodestructive skin lesions that do not correspond to above-mentioned conditions. They differ from dermatitis artefacta as being conscious; from skin-picking disorder as lacking compulsive/impulsive signs; and from delusional parasitosis as being non-delusional (no believe in parasite infestation). In these cases recalcitrant self-inflicted skin lesions (typically local ulcers) result from a conscious repetitive autodestruction of focal skin loci primarily affected with intensive and unbearable pain/dysaesthesia in order to relieve pathological sensations. There are anecdotal case reports in recent publications that designate these cases with different terms, e.g. “nummular headache” or “coin-shaped cephalgia” (only some cases, e.g. (14)) and “trigeminal trophic syndrome” (also only some cases, e.g. (15)) or contribute to atypical dermatitis artefacta, e.g. due to schizophrenia (16). However, there are definition (hypochondriasis circumscripta (17)) and psychopathological concepts (18–24) of the disorder that are generally neglected. Thus, the purpose of the present article is to summarize cases of hypochondriasis circumscripta observed in the dermatologic department of First Moscow State Medical University and to re-establish the clinical identity of the disorder.

METHODS

The methodological approach included precise dermatological, general somatic, neurologic, and psychiatric evaluation performed in an interdisciplinary paradigm. Psychiatric assessment was based on a phenomenological psychopathology of Karl Jaspers (25) and pointed on precise description of signs, symptoms, and syndromes, i.e. mental states (26), their interrelations and their transformation in time. The approach is typical for Russian clinical psychiatry, has been developed for decades, and has been adopted for consultation-liaison practice in general medicine and dermatology as well. The study methodology comprised initial examination of patients by a consultation-liaison psychiatrist (D.V.R.) with the following council of multidisciplinary case management team with supervising clinicians (A.B.S., A.N.L.) for the consensus diagnosis.

Study sample

The study sample included 22 subjects (15 women; mean age 56.1 ± 12.6 years) consecutively observed in the Dermatological Department of the I.M. Sechenov First Moscow State Medical University, consulted by a psychiatrists of Department of Psychiatry and Psychosomatics and diagnosed with hypochondriasis circumscripta in 2008–2014. Occasional frequency of hypochondriasis circumscripta (22 cases in a period of 6 years) in dermatology reflects as casuistic nature of the disorder, as associated diagnostic difficulties. Thus, patients with hypochondriasis circumscripta as a rule were referred to the Dermatological Department for another primary diagnosis (pyoderma ulcerosa chronica, actinomyces, lupus vulgaris, necrotizing vasculitis, necrotizing fasciitis, furunculosis, dermatitis artefacta etc.). See sociodemographic and clinical characteristics of the study sample in Table I.

RESULTS

According to obtained data psychopathological structure of hypochondriasis circumscripta in 22 examined subjects comprise basic/primary and secondary/derived components, that corresponds to conceptual binary model of psychodermatological syndromes (27, 28). “Basic” or primary symptoms are represented by different coenaesthesiopathic/sensory phenomena, while secondary, derived symptoms include corresponding cognitive psychopathological symptoms. The term “coenaesthesiopathic” is used here to define somatoperceptive disturbances in a form of medically unexplained pathological skin

Table I. Sociodemographic and clinical characteristics of patients diagnosed with hypochondriasis circumscripta (n = 22)

Characteristics	Values
Female, n (%)	15 (68.2)
Male, n (%)	7 (31.8)
Age, years, mean \pm SD	56.1 ± 12.6
Marital status, n (%)	
Divorced	8 (36.4)
Married	7 (31.8)
Single	4 (18.2)
Widowed	3 (13.6)
Employment, n (%)	
Retiree	10 (45.5)
Unemployed	7 (31.8)
Employed	5 (22.7)
Disease duration, years, mean \pm SD	4.7 ± 4.8
Median disease duration, years	3.0
Disease duration range	4 months–18 years
Unifocal lesions, n (%)	8 (36.4)
Head	4 (18.2)
Neck	1 (4.5)
Hand	1 (4.5)
Foot	1 (4.5)
Umbilical area	1 (4.5)
Multifocal lesions, n (%)	14 (63.6)
Head	8 (36.4)
Arms	7 (31.8)
Scalp	4 (18.2)
Trunk	4 (18.2)
Legs	3 (13.6)
Feet	3 (13.6)
Hands	1 (4.5)

sensations “Coenaesthesiopathy” (Greek: *coen* – common, shared; *aesthesia* – feeling, sensation, perception; *pathy* – disorder, disturbance) is a term derived from the proposed by J. Ch. Reil expression “coenaesthesia”. Coenaesthesia is the general feeling of one’s body that arises from multiple stimuli from various bodily organs.

The summary of the most important data on the psychopathological picture of hypochondriasis circumscripta is discussed below in a corresponding successive manner, i.e. summary description of basic sensory phenomena are followed by secondary cognitive symptoms.

Coenaesthesiopathic or basic sensory phenomena in hypochondriasis circumscripta are diverse and complex, but are circumscribed by a local skin area (or in some cases areas [Table I]). Sensations include (i) intradermal dysaesthesia, (ii) idiopathic pain, (iii) tactile illusions, and (iv) body fantasies.

Intradermal dysaesthesia (27) (Greek: “*dys*” – not-normal, “*aesthesia*” – sensation; i.e. “abnormal sensation”) is a kind of pathological skin sensation described metaphorically and characterized by penetrating pattern as piercing or prickling of skin (“like needles dig in”, “like insect stings”, “like under-skin nodules swelling and rupturing outside”). Sensations are presented with a nuance of violation of skin integrity, solution of continuity and homogeneity of skin, and sense of pervasion through the skin layer. Thus, the term “intradermal” (“intra” – inside, “derma” – skin, i.e. “inside the skin”) emphasizes spatial localization of sensations in a depth of skin.

Idiopathic pain is an extremely severe and intensive algopathic sensation that emerges abruptly, has a chronic course and insular (“circumscribed”) localization, and has features of possessive sensations (29, 30): perception of a corresponding dermal locus as an “alien part of skin” and an urge to discard from it. The sensation is characterized with unbearable affective pressure of pain, predominance in patient’s consciousness, inability to prescind from pain.

Tactile illusions are defined here as haptic sensations: minimal skin irregularities or inspissions on a skin surface are erroneously perceived as pathological lesions – “papules”, “tubercles”, “pustules”, “furuncles”, etc.

Body (or somatic) fantasies (31) are pathological sensations of fantastic character, described in terms of symbolic or vivid comparisons with a conditional “as if” modality and properties of uncommon configuration and location formations, situated in/under the skin. Body fantasies have florid, intrinsic for an objects descriptions, e.g. “branchy, under-the-skin ducts and tunnels” or growing out the skin “roots of pathological tissue”. Simultaneously, patients complain about volumetric, uncommon for human skin static formations or particles: “white fibres or creamy particles, resembling grit”, “capsules”, “vesicles with a colourless fluid”, “grains like a spawn”, “tiny balls connected in chaplets”). Those “particles” are

perceived primarily by patients in palpation and then after extraction of own tissue fragments are visualized like foreign bodies, i.e. represent “fantastic visual phenomena” (phantastischen Gesichterscheinungen (32). The phenomenon is in accordance to Jaspers estimations on disorders of perception that often correspond simultaneously to several sensory modalities (tactile, visual, visceral etc.), supplying each other, but not to a single “isolated” sensory modality.

At the same time, body fantasies lack the basic features of tactile hallucinations, observed in delusional parasitosis. Namely, lifeless “particles”, e.g. “little balls” (16), do not resolve into arthropods, and remain in patients’ perceptions just “pathological presentation of an undiagnosed disease”, but not a kind of a living creature. So there is no phenomenon of “animation” (28). The term “animation” is introduced by us to define a key clinical criterion of delusional parasitosis, that allows to differ it from circumscripita hypochondriasis: sense of living beings, which have “vital activities” (crawling, biting, etc.) and “die” after “appropriate measures are taken” (squashed by fingers or poisoned by insecticides).

As a secondary or derived from sensory (coenaesthesiopathic) phenomena hypochondriasis circumscripita comprise over-valued ideas (33, 34). Over-valued ideas content is determined by hypochondriacally over-valued possession phenomenon (35). Possession phenomenon here is not referred to delusions of possession (demonic or spirit (36)), but is based on experience of skin fragment or “particles” as alien ones and associated with desire to eliminate (“exorcise”) it from the skin.

Coenaesthesiopathic/sensory phenomena (intra-dermal dysaesthesia, idiopathic pain, tactile illusions, body fantasies) have a property of foreign, alien to own skin tissue sensations, associated with pathological activity, resembling paranoiac actions. The latter includes insistent, obstinate, unflinching, non-dissuasive urge to eliminate “necrotic”, “splinter-like” pathological skin loci or alien/foreign formations/objects/particles. At that, pieces of own epidermis and derma are perceived as alien/foreign/pathological formations/objects/particles. Patients use for their extraction not only nails, but also additional instruments, including needles, scissors, knives, pincers, scalpels. About one third of patients ($n=8$) apply

for surgery – indirect delegated self-harm. Herewith, they easily overcome pain, caused by self-destruction, that they perceive as milder than tormenting sensations (e.g. idiopathic pain), and pay no or minimal attention to cosmetic consequences of self-mutilation.

Self-injuries in hypochondriasis circumscripita are represented with artificial open, deep, ulcerous skin defects with sharp borders, surrounded by unaffected skin. At first sight they could resemble self-inflicted lesions in a skin picking disorder. In contrast, self-injuries are more focal, than scattered, and differ in distinctly circumscribed localization with a tendency to affect a single skin region with a solitary ulcer (left angle of an upper eyelid, central region of a right cheek, etc.) (Fig. 1). However, there are also cases with multifocal self-inflicted ulcers (Fig. 2). Typically injuries are deep and may affect also dermis and subdermal tissues. Persistent chronic ulcers with massive necrotic crusts, linear and oval deforming scars are typically observed.

The clinical course of hypochondriasis circumscripita is typically chronic. Median illness duration at the diagnosis establishment is 3.0 years, mean disease duration is 4.7 ± 4.8 years (see Table I). However the disease range in the study sample varies from 4 months to 18 years, that reflects a clinical course tendency. In most cases ($n=18$) the syndrome starts with a prodromal stage defined by intradermal dysaesthesia and idiopathic pain, associated with a skin picking behaviour. Tactile illusions and body fantasies with an over-valued hypochondriacal idea are supplemented later after months or even 1.5–2 years (gradual onset). In other cases ($n=4$) tactile illusions, body fantasies and over-valued hypochondriacal idea persist initially from the onset of disorder and become the most prominent presentations of the syndrome from the very beginning (acute onset). The latter patients could be diagnosed with hypochondriasis circumscripita even in first weeks of their illness. So the shortest disease duration in some patients just reflects quicker diagnosis establishment as a result of acute onset.

TREATMENT

Patients with hypochondriasis circumscripita were managed with a holistic interdisciplinary psychodermatologic approach by a dermatologists and a consultation-liaison psychiatrist (D.V.R.) in a naturalistic treatment manner. Psychotropic medications were combined with psychotherapy (cognitive behaviour therapy; CBT) and topical dermatologic medications. Treatment data were analysed for 17 subjects (77.3%) who gave their consent and took at least a single dose of psychotropic medication. Mean number of treatment courses per patient was 5.2, mean treatment duration (until



Fig. 1. Unifocal hypochondriasis circumscripita. Chronic solitary ulcer on a locus of idiopathic pain/dysaesthesia persisting for 3 years in a patient who refused psychiatric treatment.



Fig. 2. Multifocal hypochondriasis circumscripta (multiple self-inflicted ulcers are situated on forearms, neck and head).

discontinuation or lost to follow-up) was 20.8 weeks. Treatment course was defined as at least 4 weeks period of an unchanged psychotropic medication modality. After each period of efficiency and tolerability were assessed (clinically, as well as psychometrically with VAS-S). VAS-S is a visual analogue scale for skin sensations intensity (intra-dermal dysaesthesia, idiopathic pain, tactile illusions, body fantasies). It was chosen for the efficiency measure, as skin sensations had been considered as basic phenomena and it has been previously reported that reduction of skin sensations preceded improvement of cognitive (secondary) component (27, 29). So coenaesthesiopathic or basic sensory phenomena could be considered as a sensitive treatment efficiency marker. If acceptable efficiency (clinical improvement and mean VAS-S score reduction $> 50\%$) and good tolerability were achieved, treatment modality was continued (doses could be adjusted) and the course considered as successful. If there was no acceptable effect after 4 weeks or there were intolerable side effects (unsuccessful course), a new treatment course was started. The procedure was repeated till the effective course or lost for follow-up. Treatment results were modest: only 8 (47.2%) subjects achieved full remission (VAS-S=0), 5 (29.4%) improved (partial remission – $0 < \text{VAS-S} < 50\%$), and 4 (23.5%) were resistant to treatment (VAS-S $> 50\%$). Antipsychotics (haloperidol 7.5–15 mg, chlorpromazine 25–75 mg, risperidone 2–6 mg, sulpiride 400–600 mg) and antidepressant amitriptyline (50–100 mg) were psychotropic drugs associated with a better outcome as defined by assessment of course results (Fig. 3). Thus, hypochondriasis circumscripta seems to be a serious treatment challenge. Further treatment studies are required.

DISCUSSION

The interpretation of the term “hypochondriasis circumscripta” in psychiatry is ambiguous. The term was developed by Bonhoeffer’s colleague Schwarz (17). He considered hypochondriasis circumscripta as a psychiatric disorder with presentations circumscribed by skin. This is why the adjective “circumscripta” was used. However, initially Schwarz used the term to define delusional parasitosis. Later, Bonhoeffer gave another interpretation of the term, which became conventional

(19–24). Bonhoeffer defined circumscripta hypochondriasis as a syndrome peculiar for particular pathological body sensations, namely idiopathic pain. However, he observed projections of idiopathic pain not only in skin, but also in internal organs. Simultaneously, he has modified the meaning of the adjective “circumscripta”. Bonhoeffer implied “circumscribed or isolated locus of sensations”. As a key feature distinctive from other types of hypochondriasis, he identified a sense of foreignness or extraneity of pathological focus, contraposed by patients to other healthy tissues. He also noted that idiopathic pain had been associated with seeking for extraction of “pathological focus” with surgery (Operationsucht).

Our interpretation of the term “circumscripta” differs from the mentioned conventional one and shares characteristics of both concepts, i.e. proposed by Schwarz and Bonhoeffer. Thus, according to our data “circumscripta” hypochondriasis in dermatology includes as properties described by Schwarz, that are (i) exclusive skin involvement and (ii) persistence of sensations other than idiopathic pain (dysaesthesia, tactile illusions, body fantasies), as characteristics proposed by Bonhoeffer, i.e. (i) sense of foreignness of a sensation locus and (ii) seeking for its extraction that leads to self-destructive behaviour.

Thus, the autodestructive non-delusional psychodermatological disorder, described previously as

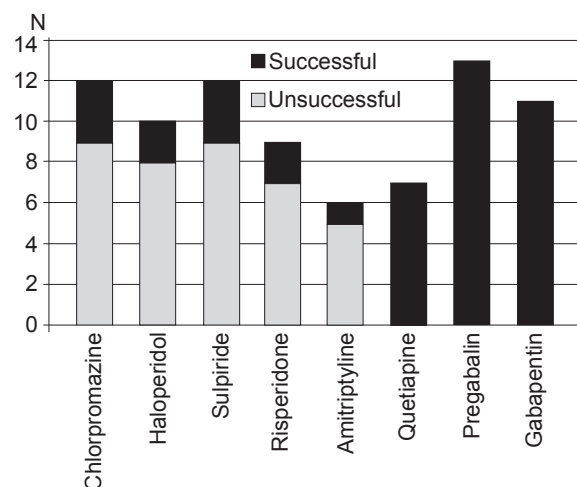


Fig. 3. “Successful” and “unsuccessful” courses of treatment in hypochondriasis circumscripta by medication (Y-axis N – absolute number of treatment courses).

“hypochondriasis circumscripta” is a syndrome with self-inflicted skin lesions (typically local ulcers) that result from a conscious repetitive autodestruction of focal skin loci primarily affected with intensive and unbearable sensations associated with a hypochondriacal over-valued idea. Hypochondriasis circumscripta is a serious treatment challenge. It is important to distinguish it from dermatitis artefacta, skin picking disorder and delusional infestation.

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