

Manuscript Number: COMPRPSYCHIATRY-D-13-00375R1

Title: Cenesthopathy in adolescence: an appraisal of diagnostic overlaps along the anxiety-hypochondriasis-psychosis spectrum

Article Type: Review Article

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Abstract: Objective: To discuss the diagnostic validity of unusual bodily perceptions along the spectrum from age-specific, often transitory and normal, to pathological phenomena in adolescence to hypochondriasis and finally to psychosis.

Methods: Critical literature review of the cornerstone diagnostic groups along the spectrum embracing anxiety and cenesthopathy in adolescence, hypochondriasis, and cenesthopathy and psychosis, followed by a discussion of the diagnostic overlaps along this spectrum.

Results: The review highlights significant overlaps between the diagnostic cornerstones. It is apparent that adolescents with unusual bodily perceptions may conceptually qualify for more than one diagnostic group along the spectrum. To determine whether cenesthopathies in adolescence mirror emerging psychosis, a number of issues need to be considered, i.e. age and mode of onset, gender, level of functioning and drug use. The role of overvalued ideas at the border between hypochondriasis and psychosis must be considered.

Conclusion: As unusual bodily symptoms may in some instances meet formal psychosis risk criteria, a narrow understanding of these symptoms may lead to both inappropriate application of the new DSM-5 attenuated psychosis syndrome and of treatment selection. On the other hand, the possibility of a psychotic dimension of unusual bodily symptoms in adolescents must always be considered as most severe expression of the cenesthopathy spectrum.

Bruderholz, 15th February 2014

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Dear Editor,

We would like to thank you for giving us the opportunity to resubmit a revised version of the manuscript Ref.No. COMPRPSYCHIATRY-D-13-00375. We have carefully studied the recommendations and issues raised by the two reviewers and tried to address them adequately.

In the following we would like to address all points raised by the reviewers in more detail:

Answers to comments of Reviewer 1:

Although the paper contains interesting information including historical annotations on disturbances of the bodily self, the focus and method of the study is not very clear. The authors restrict their literature review on topics related to anxiety, hypochondriasis, and psychosis, and do not explicate how the literature search was performed. Was it a systematic review of the literature (how many papers reviewed? selection criteria?) or selective? If it was selective, why not discuss the concept of somatization, Briquet's syndrome, or somatic delusions?

We would like to thank the reviewer for raising this important point. As we did not follow specific guidelines 'Preferred Reporting Items for Systematic Reviews and Meta-Analyses' (PRISMA) guidelines (Moher et al., 2009), we agree with your comment that this is a selective and not systematic review. Therefore, we now removed the term "systematic" and clearly state that this is a "critical" review. Furthermore, we have rephrased the methods in more detail on page 4, line 21, to page 5, line 9.

We also acknowledge that including a discussion of somatization is an important aspect. This now features on page 9, lines 3 to 19. We did not include a discussion of the Briquet's syndrome. This is a specific somatization disorder with multiple symptoms, and our above mentioned paragraph includes the general concept of somatization, thus also Briquet's syndrome. We however now included the concept of somatic delusions in our section on "cenesthopathy and psychosis" that we have restructured substantially (see page 12, lines 13 to 24). We now also reference somatic delusions on page 16, line 7 (references 52 and 53).

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The paper is very long and repetitive. The authors should make efforts to shorten it. Also, I am not convinced that the three case-vignettes add any value to the paper. The sections on mode of onset, primary or secondary cenesthopathy, border of hypochondriasis/psychosis/overvalued ideas, should be shorter and more poignant.

These points are well-taken. We have now omitted all three case vignettes. We also omitted the entire paragraph on the discussion whether cenesthopathies are primary or secondary. We also shortened the sections on mode of onset, border of hypochondriasis/psychosis/overvalued ideas, level of functioning and on gender, rephrased parts of it more poignant and omitted any repetitions. The manuscript is now substantially shorter.

Answers to comments of Reviewer 2:

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In respect of psychotic prodromal symptoms (one area covered in this review) the literature review does not seem to have captured all relevant developments, i.e. some key publications from Parnas and Stanghellini, outlining new assessment tools re identification of abnormal bodily sensations prone/closer to developing psychosis: (other on cenesthesias and cenesthopathic schizophrenia: Bräuning et al. 2000); Priebe & Röhrlich 2002) Parnas (2005) re EASE instrument Stanghellini, G. (2009) Embodiment and schizophrenia. World Psychiatry, 8, 56-59. Stanghellini et al. (2012) Abnormal bodily experiences may be a marker of early schizophrenia? Current pharmaceutical design . 01/2012; 18(4):392-8.

We acknowledge that an inclusion and discussion of this additional literature is important. We now have substantially restructured the section "Cenesthopathy and psychosis" and have extended our discussion of this section, including the literature mentioned by the reviewer plus additional references (see page 12, lines 1 to 24; references 36 to 43).

We would once more like to thank for the thoughtful and detailed reviews that we hope have helped us to improve the paper substantially.

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Draft February 15th 2014

Cenesthopathy in adolescence: an appraisal of diagnostic overlaps along the anxiety-hypochondriasis-psychosis spectrum

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Abstract

Objective: To discuss the diagnostic validity of unusual bodily perceptions along the spectrum from age-specific, often transitory and normal, to pathological phenomena in adolescence to hypochondriasis and finally to psychosis.

Methods: Critical literature review of the cornerstone diagnostic groups along the spectrum embracing anxiety and cenesthopathy in adolescence, hypochondriasis, and cenesthopathy and psychosis, followed by a discussion of the diagnostic overlaps along this spectrum.

Results: The review highlights significant overlaps between the diagnostic cornerstones. It is apparent that adolescents with unusual bodily perceptions may conceptually qualify for more than one diagnostic group along the spectrum. To determine whether cenesthopathies in adolescence mirror emerging psychosis, a number of issues need to be considered, i.e. age and mode of onset, gender, level of functioning and drug use. The role of overvalued ideas at the border between hypochondriasis and psychosis must be considered.

Conclusion: As unusual bodily symptoms may in some instances meet formal psychosis risk criteria, a narrow understanding of these symptoms may lead to both inappropriate application of the new DSM-5 attenuated psychosis syndrome and of treatment selection. On the other hand, the possibility of a psychotic dimension of unusual bodily symptoms in adolescents must always be considered as most severe expression of the cenesthopathy spectrum.

Key words:

adolescent psychology; hallucination of body sensation; hypochondriasis; psychotic disorders; nosology

Introduction

Over the past two decades, the early recognition and intervention of psychotic disorders has developed to one of the most vigorously studied fields in psychiatry with innumerable mental health services around the globe now providing early psychosis programs [1, 2]. As a result, Section III of DSM-5 [3], has implemented the 'attenuated psychosis syndrome' as a new 'condition for further study', although not yet recommended for clinical use. The rising public awareness of both the availability of these services and the potential to improve illness outcome via early intervention has contributed to a larger diagnostic spectrum being assessed today in early psychosis services compared to pioneering days. This phenomenon is reflected by findings of significantly higher non-transition rates to psychosis [4] and considerable remission rates [5] in more recent studies of patients with psychosis risk states in comparison to earlier studies. This observation, however, is not surprising: symptoms that formally meet criteria for psychosis risk states may not always necessarily mirror an actual increased risk for psychosis, but may occur as epiphenomena of other underlying psychiatric disorders. Thus, whilst early psychosis services primarily set out to identify patients at risk for psychosis as early as possible in the disease course, they now more commonly face the additional task of disentangling genuine psychotic risk states from other overlapping psychiatric diagnoses.

This task is all the more challenging as not only psychosis, but also most other mental illnesses begin in adolescence [6]. Furthermore, as adolescence is a period of life characterized by multitudinous variants in behaviour, developing diversity of contextual thinking, and frequent emotional turmoil, patients that are referred to early psychosis services for risk assessment may simply experience symptoms that belong to the large scope of phenomena inhering in adolescence. Thus, formal psychosis risk symptoms may lie anywhere on the continuous spectrum reaching from

prototypal and generally transient adolescent phenomena to epiphenomena of other underlying mental disorders to genuine evolving psychosis.

Undoubtedly, one of the most challenging spectra that warrant accurate consideration as to whether adolescents are about to develop psychosis bears on the appearance of unusual bodily perceptions. Adolescence is also the life period with dramatic physical change and development, leading to a greater awareness of these young people of their physical appearance to a greater potential for concern as to their personal physical health and well-being.

In the present review, we discuss the diagnostic validity of unusual bodily perceptions in adolescence. As we explore the spectrum embracing age-specific normal phenomena to psychosis, we highlight the phenomenology of body related anxiety in adolescence, then move further along the spectrum to summarize the characteristics of hypochondriasis, to finally reach the “psychotic” end of the spectrum.

Aims of the study

To discuss the diagnostic validity of unusual bodily perceptions along the spectrum from age-specific, often transitory and normal, to pathological phenomena in adolescence to hypochondriasis and finally to psychosis.

Material and methods

We first critically review the terms that are essential to our theme and that refer to bodily perceptions. We then provide a literature review of the diagnosis constituting the cornerstones along the above-mentioned spectrum, i.e. anxiety and cenesthopathy in adolescence, hypochondriasis, and cenesthopathy and psychosis. In the final section, we expand our review to discuss the diagnostic overlaps along the investigated spectrum.

Specifically, electronic searches were performed in the PUBMED database by combining the following two sets of keywords: (1) 'cenesthesia', 'cenesthopathy', 'hallucination of body sensation', 'somatic delusion'; and (2) 'anxiety', 'hypochondriasis', 'psychosis', 'psychotic', 'psychotic disorder', 'prepsychosis', 'pre-psychoisis', 'pre-psychotic'.

We reviewed the database and carefully searched the reference lists of the included articles identified in the original search. We included all papers published in peer-reviewed journals until January 2014, without any language restriction though the vast majority of papers were in English.

Results

Cenesthesia and cenesthopathy

The general awareness of one's sense of bodily existence and the general feeling of well-being or malaise was coined in 1794, well over two centuries ago, with the term *cenesthesia* in a doctoral thesis of a student of the German psychiatrist and physician Johann Christian Reil [7]. Reil's discovery had a great impact on the development of neuroanatomical knowledge (i.e. the insular cortex) contributing significantly to a changed view of the human body and of mental processes [8]. Cenesthesia was defined as 'the means of which the soul is informed of the state of its body, which occurs by means of the nerves generally distributed throughout the body'. The term was equivalent to the German *Gemeingefühl* or *Leibgefühl*, for which the French equivalent became *cénésthésie* or *sensibilité générale* [9]. Cenesthesia expresses the general sense of bodily experience and was seen as the integrative denominator for *Meinhaftigkeit* or *I-ness*. Reil attributed changes in cenesthesia to a number of general disorders, but he also described idiopathic disorders of the cenesthesia with limitation to the nervous system. He postulated that a distortion sent

a misleading message to the brain about the body's condition, giving rise to a bodily illusion and, subsequently, to an aberrant belief in a dangerous disease.

It was, however, only a century later that French psychiatrists Dupre and Camus [10] introduced the term *cenesthopathy* for states of disordered cenesthesia, i.e. pathological bodily perceptions, and pointed out that cenesthopathies are likely to be mistaken for neurasthenic, melancholic or hypochondriacal states. They are characterized by prevailing abnormal and often bizarre sensations that usually lead to great concern in patients. Dupre [11] later alluded to the large familiarity of psychiatrists with cenesthopathy as they commonly see such states in their patients. In some analogy to Reil's cenesthesia, Wernicke [12] described the concept of *Somatopsyche* and *vital feelings*, and Jaspers described comparable sensations in the section of *Awareness of Body* in *General Psychopathology* [13]. While Wernicke [12] associated disruptions of vital feelings more commonly to affective psychoses, Jaspers [13] described these phenomena to particularly occur in schizophrenic patients, emphasizing Bleuler's [14] observation that among the most common and important symptoms of schizophrenia are those involving bodily feelings. It was not before 1957 that *cenesthetic schizophrenia* was introduced by Huber as a specific type of schizophrenia with characteristic cenesthopathies [15]. Besides German and French literature the concept of cenesthopathy has been described in Russian [16] and Japanese [17,18] literature, however, its existence is only elusively mentioned in English literature and not acknowledged at all in major American textbooks on psychiatry [19].

Anxiety and cenesthopathy in adolescence

In no other period of life do physical changes occur as dramatically as in adolescence. These changes come along with a marked increase of adolescents'

general sense of their bodily existence [20]. Thus, adolescence as a developmental 'milestone' is inevitably and intrinsically tied to the evolving awareness of the potential vulnerability of physical integrity and health. While these young individuals may grow accustomed to minor incidences such as sporting injuries of the muscular or skeletal system or transient infectious diseases long before they reach adolescence, and while any subjective symptom experience within the range of such circumstances would not be conceived as cenesthopathies, severe health concern may emerge in association with unexpected and previously unexperienced symptoms, notably of organs that may be attributed with higher fragility or vital importance, such as the eyes, the heart, or the brain. In such cases, young individuals experience symptoms that affect an entirely 'new' span of organs and that are well beyond the range of previously witnessed and thus non-disconcerting phenomena. For these, the term *cenesthopathic* may be more appropriate.

It is not unusual that the experience of 'new' bodily perceptions is preceded by a sudden episode of intense anxiety. The relationship of the latter with depersonalization is well documented in literature [21, 22], occasionally following cannabis use [23]. Importantly, one of the symptom dimensions of depersonalization disorder that emerged from two recent factor analytical studies was *anomalous body experience* [24] or *body distortion* [25], respectively. Further, cenesthopathy can co-occur with depersonalization and feelings of insufficiency to form a 'triangle' symptomatology for which the term *adolescent cenesthopathy* was proposed [18].

Adolescent cenesthopathy is more common in males than in females [18]. It is noteworthy that both patients with depersonalization disorder and adolescent cenesthopathy are described as being more anxiety prone, socially insecure and shy [18, 21, 22]. These patients may show disturbed narcissistic regulation [22] and tend to split their ego into an ego that observes the experiencing ego, a phenomenon that

has been named 'autoscopy' [26], i.e. it is now the patient himself who is under the 'coverslip of the microscope'.

Thus, in summary, following an initial episode of intense anxiety, anxiety prone adolescents with disturbed narcissistic regulation may react with a catastrophic appraisal of normally transient symptoms, not only of depersonalization, but also of bodily experiences, such as accelerated heart beat, blurred vision, or headache [27].

Hypochondriasis

The essential part of any definition of hypochondriasis is a morbid preoccupation with one's body or state of health, either mental or physical [28]. Although DSM-IV [29] hypochondriasis has been replaced as official diagnosis in DSM-5 by *illness anxiety disorder*, we shall use the term hypochondriasis throughout this review, as the latter term has uniformly been used in literature. DSM-IV [29] defines hypochondriasis as 'a preoccupation with fears of having, or the idea that one has, a serious disease based on a misinterpretation of one or more bodily signs or symptoms'. Despite vast medical evaluation that does not identify any general medical condition that fully accounts for the person's concerns about disease or for the physical signs or symptoms, these patients continue to experience an unwarranted fear or idea of having a disease. According to DSM-IV, the belief, however, is *not of delusional intensity* (i.e. the person can acknowledge the possibility that she or she may be exaggerating the extent of the feared disease, or that there may be no disease at all), but still causes significant distress or impairment in social, occupational, or other important areas of functioning. The preoccupation may be with bodily functions such as heartbeat or with vague physical sensations, and these concerns may involve several body systems or a specific organ or a single disease, not uncommonly a fear of having cardiac disease. Consequently, patients often present their medical history

in often meticulous detail, and deterioration of doctor-patient relationships and 'doctor-shopping' are common.

A common challenge in clinical practice is to differentiate hypochondriasis from somatization disorder, as the latter is a common disorder in primary and secondary health care [30]. Although both patients with somatization disorder and with hypochondriasis fear suffering from a serious disease and thus frequently seek medical help, the concern of patients with somatization disorder is more directed towards receiving symptom releasing treatment, whereas the main goal of patients with hypochondriasis is to having ruled out serious diagnoses. This distinction between these two types of disorders was more readily applicable with the DSM-IV [29] than with the DSM-5 [3] definitions. In DSM-IV, a pivotal diagnostic criterion of somatization disorder was that physical complaints lead to treatment being sought. In DSM-5, somatization disorder has been reconceptualised as somatic symptom disorder with the treatment-seeking behaviour being dropped, however now including "a persistently high level of anxiety about symptoms" as one of the main diagnostic hallmarks. Thus, as DSM-IV hypochondriasis has been reformulated as illness anxiety disorder in DSM-5, the differentiation between somatization and hypochondriasis remains a challenging task when relying on the current DSM definitions.

DSM-IV [29] provides a specifier for hypochondriasis which is used if, for most of the time during the current episode, the individual does not recognize that the concern about having a serious illness is excessive or unreasonable. DSM-5 also provides a specifier, distinguishing between frequently versus rarely used medical care [3]. In general, hypochondriasis carries a poor prognosis, the duration usually being measured in decades rather than years, and the response to treatments is uniformly unsatisfactory [28]. The disorder neither remits spontaneously nor progresses to

further delusional elaboration. Although the disorder can begin at any age, its most common age at onset allegedly is in early adulthood, with male and lower social classes being more prone than others [28]. As in adolescent cenesthopathy, there seems to be a predominance of shy and sensitive individuals with disturbed narcissistic regulation among hypochondriac individuals [28].

The prevalence of hypochondriasis in primary care settings has varied from 4% to 9% [29]. However, there may be a tendency to underestimate the prevalence of hypochondriasis in the general population, due to the relative isolation of many sufferers and the tendency to self-medicate. Indeed, many of these patients come to the attention of the psychiatric services only by referral from colleagues on other specialities [31].

Cenesthopathy and psychosis

Observing a high similarity with the cases previously described by Dupre and Camus [10], Huber [15] was the first to describe patients with *cenesthetic schizophrenia* as a subtype of schizophrenia that was characterized by peculiar disturbances of bodily perceptions, but remained often unidentified as psychotic disorder due to its hypochondriacal characteristic. Cenesthetic schizophrenia has never been incorporated in DSM, while it appears undefined in ICD10 [32] among 'other schizophrenia' without having been identified in previous editions. In contrast to Anglo-American literature, a rich body of publications on cenesthopathic forms of schizophrenia exists in Russian and Japanese psychiatry [16, 17, 18]. Huber [15] described several cenesthetic symptoms (*Prägnanztypen*), e.g. sensations of numbness, wandering sensations within the body, electrifying and thermic sensations, or sensations of movement, which are published in the *Bonn Scale for the Assessment of Basic Symptoms* (BSABS) [34]. Klosterkötter et al. [35] used the

BSABS [34] several decades later to study the power of cenesthopathies to predict schizophrenia, however, none of the BSABS cenestopathies featured among the ten basic symptoms reported to determine prediction. Huber [15] differentiated three 'developmental levels' of disease with progression from uncharacteristic hypochondriacal symptoms to qualitatively bizarre cenesthesias and finally to typical schizophrenic symptoms such as first rank Schneiderian symptoms, i.e. somatic passivity phenomena/bodily hallucinations, with schizophrenia diagnosis only permitted if the third level is reached. The individual patient can experience transition from the first to second and to third level and back to first level again. Uncharacteristic hypochondriac first level symptoms can prevail initially and during the later course of illness. Thus, cenesthetic schizophrenia can often only be diagnosed after substantial longitudinal observation, as cenesthopathies precede the onset of first psychotic episode of cenesthetic schizophrenia by several years, and as in comparison to other schizophrenia subtypes, the prodromal phase preceding psychosis onset is particularly long [33,36]. Given the particularities of the disease course, Huber suggested that this was a type of schizophrenia that comes to a standstill at its beginning or develops into pure residual syndromes after one or a few short psychotic episodes, and he thus claimed a close resemblance to Bleuler's *latent schizophrenia* [14]. However, Huber observed an acute onset of in about one quarter of his patient sample with cenesthetic schizophrenia, characterised by dysesthetic crises with vegetative symptoms and an elementary fear of dying [33].

In cenesthetic schizophrenia proposed by Huber [15], the classic schizophrenia symptoms are limited to psychotic exacerbations, while the entire disease course is characterized by these abnormal bodily sensations that generally occur in rapid changes in paroxysms and commonly co-occur with vital discomfort, fatigue and exhaustion. However, Huber [15] recognized that cenesthetic disturbances also

occurred in a large percentage (64%) of other schizophrenia subtypes. Already French psychopathologists in the early 20th century thought that hebephrenia was characterized by impaired nervous systems moderating cenesthesia [37]. Phenomenologically, the essential feature of schizophrenic existence is disembodiment, i.e. an increasing disruption between subjectivity and bodily experience where the schizophrenic person behaves like a soulless body, leading to initially *normal* cenesthasias being lived in hyperreflexive awareness and diminished self-awareness [37]. In studies using a scale for the phenomenological *Examination of Anomalous Self-Experience* (EASE) [38] that include *cenesthetic experiences* overlapping with Huber's cenesthopathies, self-disorders were more prevalent in adolescents meeting at-risk criteria for psychosis than in their non-psychotic help-seeking peers [39] and aggregated selectively in the schizophrenia spectrum [40]; however, prevalence of cenesthetic experiences was not reported specifically. The phenomenological view warrants that abnormal bodily experiences should be included as diagnostic hallmarks for schizophrenia [37]. Support stems from more recent studies that have demonstrated considerable prevalence rates of abnormal bodily experiences in the early onset of schizophrenia [17,41,42]. These findings underline that cenesthopathies are not restricted to Huber's cenesthetic schizophrenia where they generally emerge after many years. Further, the studies by Röhrich & Priebe [41] and by Stanghellini et al. [42] show a significant relation between cenesthopathies and disturbances of body concept, suggesting that cenesthopathic phenomena may be classified as delusional perception rather than perceptual aberration, i.e. misinterpretation of internal perception, and thus supporting the concept of somatic delusions [43].

An appraisal of diagnostic overlaps along the anxiety-hypochondriasis-psychosis spectrum

The comprehension and diagnostic validity of unusual and unexpected bodily perceptions in adolescence is a major challenge. Provided that any organic and/or substance induced aetiologies can be precluded, such symptoms warrant a thorough appraisal of a broad spectrum of overlapping disorders. Patients may fall neatly into established categories, while others remain enigmatic and present disease pictures that are fluid at the edges, flowing together easily with other diagnoses. When adolescents experience cenesthopathies, both a number of diagnostic and phenomenological particularities and commonalities can thus be distinguished. Latter need specific consideration to determine whether cenesthopathies in adolescence mirror emerging psychosis.

Mode of onset

Unusual and unexpected bodily perceptions may follow sudden episodes of anxiety. In some instances, e.g. following cannabis use, unexpected bodily perceptions may instantly precede and thus cause sudden anxiety. In both scenarios, the onset is acute, and any persisting bodily perceptions may be attributed to an underlying anxiety disorder, while it is debatable if in these cases bodily symptoms may be considered as cenesthopathies in a more narrow sense. Acute onset may occur in hypochondriasis, too, and then is a favourable prognostic indicator.

Thus, at a first glance, it would seem an easy task to exclude a psychotic dimension in the phenomenology of these acute onset symptoms. However, even if cenesthetic schizophrenia are described to develop progressively along an extended prodromal period, Huber noted that in one quarter of all cases, psychosis onset was acute and occurred as dysesthetic crises [15, 33]. Further, up to 70% of patients that develop

schizophrenia report to have experienced episodic or sustained symptoms of anxiety during the prodromal phases [44].

The border between hypochondriasis and psychosis and the role of overvalued ideas

Huber emphasized that the qualitatively peculiar disturbances of bodily perceptions, and not a delusional hypochondriasis, are the central and primary symptom formation in cenesthetic schizophrenia. This discriminator may facilitate distinguishing between hypochondriasis and cenesthopathic schizophrenia as defined by Huber [15]. However, Huber [33] underlined the frequent difficulty in distinguishing cenesthetic schizophrenia from hypochondriasis. Other authors emphasized since long that the variety of peculiar somatic sensation in psychosis had often been described under the umbrella term hypochondriasis [45], and Reil [7] was aware that not in all instances patients with cenesthopathies could correct their false impression. Indeed, in clinical practice, patients may be occupied by their bodily perceptions and be convinced about their reality to the extent that the conceptual distinction between a hypochondriac and a psychotic disorder becomes an immense challenge, reaching far beyond the distinction between hypochondriasis and the cenesthopathic schizophrenia type of psychosis.

The distinction between a hypochondriac and psychotic dimension in cenesthopathic phenomena cannot be discussed without a particular appraisal of the *overvalued ideas*, a concept that was established by Wernicke [12] corresponding to the French concept of 'idée fixe', but that is only referred to in sparse amount in British textbooks and is widely ignored in American psychiatry [46]. Wernicke noted that overvalued ideas appeared in a variety of clinical settings and are difficult to reconcile with the usual aetiological divisions in psychiatry. In Appendix C of DSM-IV [29] as well as in Appendix on 'Glossary of Technical Terms' of DSM-5 [3], an overvalued idea is

described as an unreasonable and sustained belief that is maintained with less than delusional intensity (i.e. the person is able to acknowledge the possibility that the belief may not be true). The available literature [12,13,46] summarizes overvalued ideas by following features: they are held strongly, but with less than delusional intensity; they usually preoccupy the individual's mental life; compared to most obsessions, they are ego-syntonic; their content is usually regarded as abnormal compared to the general population, but not bizarre as some delusions; they cause disturbed functioning or distress to the patient and others; and compared to many delusions, they are more likely to lead to repeated action which is considered as justified. The overvalued idea thus describes an isolated, preoccupying belief, neither delusional nor obsessional in nature, which comes to dominate the sufferer's life, often indefinitely.

Importantly, literature refers to hypochondriasis as prototypical disorder featuring overvalued ideas [12,47]. If hypochondriasis is the prototype for a disorder with overvalued ideas, then – following above summarized definitions - we would expect hypochondriac patients to be able to maintain the facility to put into question their belief to some extent. However, in clinical practice, not few hypochondriac patients maintain a strong conviction as to the reality of their bodily experiences. Accordingly, DSM-IV [29] - but not DSM-5 [3] - provides a specifier differentiating those patients with poor insight from those with good insight. It is this strongly held conviction that stands at the origin of the determined and repeated action and the high degree of affect that are further core characteristics of the definitions of both overvalued ideas and hypochondriasis. The criterion referring to a less than delusional intensity in maintaining a belief is thus of limited value in numerous cases.

In summary, the distinction between hypochondriac and psychotic quality in bodily perceptions is an ambitious diagnostic task. Numerous terms have thus been

proposed for the hypochondriasis-psychosis overlap. These include *progressive somatopsychosis* [48], *hypochondriacal hebephrenia* [49], *hypochondriac paraphrenia* [50], or *hypochondriacal psychosis* [51]. In schizophrenia, it is not unusual for bodily complaints to be considered first as simple hypochondriacal complaints, only to be later assessed as cenesthopathic disturbances, particularly when the bodily complaints consist of bizarre or delusional alterations in bodily perceptions [28,43,52,53]. Bleuler may have been the first to emphasize the clinical importance of bodily complaints among schizophrenics. He stated that the majority of (treatment-resistant) hypochondriacs are schizophrenics [14]. Interestingly, he suggested that idiopathic hypochondriasis is essentially masked schizophrenia or schizophrenia which stagnated at the initial stage of the disease process [54].

Level of functioning

It is of pivotal importance to emphasize that all diagnostic entities that are here discussed along the anxiety-induced cenesthopathy-hypochondriasis-psychosis spectrum are in most cases characterized by significant impairment in functioning. Anxiety-induced cenesthopathy can lead to adolescents suffering considerable social and vocational disintegration [23]. Similarly, hypochondriasis is commonly associated with a high degree of affect that impact negatively on social and vocational functioning [28]. Impaired functioning has been described both in adolescent cenesthopathy [18] as well as in the majority of cenesthetic schizophrenia [15, 33]. Glatzel & Huber [55] have described an *endogenous juvenile-asthenic malfunctioning syndrome* which is associated with three symptom groups, i.e. cenesthopathy, depersonalization symptoms, and disturbed thought or cognition, and often evolves before the age of 20 years. The authors pointed to the similarities with low-symptom schizophrenia and chronic prodromal states of schizophrenia. It is interesting here to

note that criteria for prodromal states of schizophrenia were listed in DSM-III-R [56], but due to concerns such as whether these criteria give a valid description of the initial prodromal period were dropped from the DSM-IV [29]; however, in DSM-IV, the very same criteria were listed for schizotypal disorder, a diagnosis for which ICD-10 [32] allows an interchangeable use of the term *latent schizophrenia*. Latter term, as already mentioned, was introduced by Bleuler [14] and was considered by Huber [15] to show close resemblance to cenesthetic schizophrenia.

This overview of similar and almost interchangeable concepts mirrors the often enigmatic task to disentangle psychotic from non-psychotic processes; a task that is all the more challenged by the finding that impaired level of both social and vocational functioning is one of the earliest phenomena in evolving psychosis [44].

Gender

There is some evidence that female adolescents' self-esteem depends on their bodily appearance [57], while physical performance is more relevant to self-esteem in male adolescents [58]. Also female adolescents are earlier exposed and grow more readily accustomed to more dramatic physical and physiological changes such as the menarche [57]. Such findings compare favourably to a higher prevalence of male gender in adolescent cenesthopathy [18], in hypochondriasis [28], in cenesthopathic schizophrenia [33], as well as in the *endogenous juvenile-asthenic malfunctioning syndrome* [55], while male preponderance is not found in body dysmorphic disorder [59].

Discussion

Minor degrees of health concern are a common phenomenon throughout the entire lifespan. However, they may, spontaneously or in the presence of real disease,

become exaggerated in some individuals. Such individuals become over-concerned with their health and are convinced that they are seriously ill, noticing various abnormal perceptions and recombine these in elaborate schemes that are incomprehensible to anyone else. Such patients not infrequently present as diagnostic conundrums. As shown in our review, they may bring into question the possibility of mixed and overlapping illness states. Although this approach is at odds with the traditional concept of classifying mental health disorders into single categories, our review provides a prototypical example that some symptoms may not be assigned to one specific diagnostic category, but instead tap a number of diagnostic categories that overlap in terms of symptoms and thus must be considered against the background of this spectrum [60]. This is the case specifically in adolescents who commonly present phenomena that theoretically not only span a large diagnostic spectrum, but often lie on a continuum from normal adolescent to actual pathological states, including attenuated or established psychosis.

A constricted understanding of these symptoms may lead to diagnosing psychosis risk and indicate treatment that may fall wide off the mark, and instead of conferring symptom relief may increase risk of stigmatizing these young individuals. Over the past two decades and in innumerable mental health services around the world [1], the potential at-risk state for psychosis has been assessed with psychometric scales in help-seeking individuals [2]. Findings using these psychometric scales have provided the basis for the definition of the new DSM-5 attenuated psychosis syndrome [3]. As shown in our review, cenesthesias may occur as phenotypical expression of emerging psychosis, as it also may mirror a vast array of other underlying mental states and disorders. Thus, a purely psychometric approach to understand the origin of cenesthopathies or abnormal bodily sensations is likely to be a restrictive approach. Even though DSM-5 underlines that the attenuated psychosis syndrome is

not for clinical use, individuals may thus more promptly be assigned to this single diagnostic category. These potential caveats need to be considered in any assessment of potential psychosis risk symptoms. On the other hand, the possibility of a psychotic dimension of unusual bodily perceptions in adolescents must always be considered as most severe expression of cenesthopathy.

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