

Clinical Case Discussion

Medically Unexplained Physical Symptoms

Masking (Cenesthopathic) Schizophrenia:

A Case Series

Case presentation:

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Case discussion:

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The authors present a case series involving four patients who were referred to a specialized somatoform disorders clinic because of chronic medically unexplained physical complaints. The patients were eventually diagnosed with cenesthopathic schizophrenia, a term that has been proposed for a type of schizophrenia in which tactile hallucinations and delusional somatosensory beliefs are prominent. Two of the four had definite responses to antipsychotic medication. All of the patients were immigrants to the United Kingdom, and cultural and language barriers were likely factors in delayed diagnoses. (*Journal of Psychiatric Practice* 2010;16:258–264)

KEY WORDS: schizophrenia, cenesthopathic schizophrenia, somatic symptoms, psychosis, differential diagnosis, cross-cultural issues

We have two presentations this month, and I am pleased to have two colleagues as guest commentators for this issue. The first article presents a case series by Drs. Röhricht and Gudi on somatic presentations of psychosis among immigrants to the United Kingdom. These authors introduce many of us to a term that is used internationally in the International Classification of Diseases (ICD-10),¹ but that is less familiar to American readers: cenesthopathic schizophrenia, related to marked, dominating bodily experiences that are delusional or hallucinatory in nature. The term is derived from cenesthesia, defined in the Merriam-Webster Unabridged Dictionary² as “the general feeling of inhabiting one’s body that arises from multiple stimuli from various bodily organs.” Dr. Lewis-Fernández, our guest commentator, is an authority on cross-cultural issues in psychiatry. He studies the way culture affects individuals’ experience of mental disorders and their help-seeking expectations, including how to explore this cultural variation during the psychiatric evaluation. Among his many activities, he serves on the Culture and Gender Study Group of the DSM-5. In his remarks, he reminds us that cultural context shapes the expression of symptoms, and that somatic psychosis may be interwoven with linguistic and socially determined references that call for special understanding.

David A. Kahn, MD

CASE PRESENTATIONS

Background

Medically unexplained physical symptoms account for a high number of primary care consultations, estimated to be between 15% and 65% of presentations.^{3–5} These symptoms are not sufficiently explained by any organic pathology; however, patients are often convinced that they are suffering from some kind of underlying, not yet detected physical disease. The classification and the differential diagnosis of these symptoms pose a particular challenge to clinicians. Descriptive diagnostic labels include such complex syndromes as chronic fatigue syndrome and fibromyalgia. Many of these patients also have comorbid psychiatric diagnoses,^{6,7} mainly anxiety and depressive disorders, and, to a lesser degree, personality disorders and, rarely, psychotic disorders.⁸

Little attention has been given to the specific differential diagnosis of schizophrenia in the context of med-

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ically unexplained symptoms. Abnormal bodily sensations are frequently reported symptoms in patients suffering from psychosis, even though they are often omitted from mental state examinations. Once a diagnosis of schizophreniform disorder is made, these sensations are mainly referred to as “somatic hallucinations” or “delusional perceptions,” despite their questionable psychopathological nature.^{9,10} During initial, prodromal states prior to the first episode of psychosis in schizophrenic illnesses, a subgroup of patients present predominantly with a range of different abnormal bodily sensations, for which the term “cenesthesias” has been used (for a review, see Jenkins and Röhrich 2007¹¹). These disturbances in body experience have been identified as relevant early warning signs of schizophrenic syndromes in children and adolescents^{12,13} as well as in adults.^{14,15}

While most patients with fully developed schizophrenia describe fleeting disturbed body experiences, some studies have identified a subgroup with marked and dominating bodily symptoms.^{10,16–18} A subtype called “cenesthopathic schizophrenia” is included within the category “undefined schizophrenia” (F20.8) in the ICD-10 classification.¹

In this article, we present a case series of four patients, who were referred to a specialized somatoform disorders clinic because of chronic medically unexplained physical complaints.

Patient 1

Patient 1, a 50-year-old woman from Gambia, was referred to psychiatric services by her general practitioner in 2001 with a working diagnosis of depression and because she felt that there was something seriously wrong with her, despite having been reassured about her physical health following numerous investigations. According to her clinical notes, she had been presenting with a variety of nonspecific complaints since 1997. Her previous diagnoses included irritable bowel syndrome and somatoform pelvic pain. She described sensations of “blacking out” since she was 6 years old as well as getting nervous in crowded places for no apparent reason. She also described a history of back and head pains and complained about frequently occurring “hot flushes” associated with anxiety and mild depressive symptoms. Due to these symptoms, she regularly presented at the general practitioner’s office and also went to the accident and emergency department on a number of occasions. Prior to being referred to our clinic, the

patient had experienced a series of adverse incidents, including the breakup of an abusive marriage in 1997, the death of her father in 2000, and her sister having an accident that resulted in her becoming paraplegic.

The patient was initially prescribed antidepressants and referred to psychological therapy services. Her condition, however, became worse and, in March 2002, she complained about a host of somatic symptoms, including pain in various parts of her body, episodes of facial swelling, and a sensation of heat from the base of her spine spreading through her body. She made repeated requests to be referred for a brain scan because she feared she had a serious disease. A computed tomography brain scan and all blood tests did not reveal any abnormalities. Due to the peculiar, abnormal nature of her physical complaints, the clinicians performing the psychiatric assessments at this stage considered the possibility that the condition might be psychotic. The patient described changes in tissue/body size and pointed toward spots on her body which she felt acted as pain triggers when she touched them. When directly questioned, she confirmed that she was hearing “rushing sounds,” a phenomenon she had not previously reported. These experiences were located in internal space and did not fulfill the criteria for classical auditory hallucinations. She described sensing fire ahead of her on the floor. Continued psychiatric reviews revealed that she had experienced urges to walk over the fire and had had sensations as if the fire was being trapped in her body. She frequently suggested spiritual explanations for her experiences. By September 2002, the patient began to express delusional thoughts of a persecutory and passive nature, namely, that she believed her ex-husband was performing witchcraft on her and was poisoning her with lead. She often asked to have the lead levels in her blood tested. She was now experiencing auditory hallucinations of hearing the names of various members of her family being called. Because she felt that her feet were getting very hot and would “explode,” she put them into cold water. She suffered from poor sleep with nightmares. She continued to see things when awake, including the fire on the floor which she felt was becoming trapped within her. She was given a diagnosis of cenesthopathic schizophrenia and was started on olanzapine, which was gradually increased to 10 mg/day. Her medication was later changed to risperidone 4 mg/day due to considerable weight gain. When her condition was reviewed in the autumn of 2003, her psychotic symptoms appeared to have resolved, her sleep pattern was back to normal,

and her pain sensations had decreased remarkably. She remained relatively stable for 2 years. In 2006, she was seen by a locum consultant. At that time, there were no psychotic symptoms present but she complained about pain symptoms and depression following her husband's death. Her antipsychotic medication was reduced and, soon after, she presented with a return of all her cenesthetic sensations. When risperidone was restarted, her symptoms improved but did not remit completely until she reached a high dose of 8 mg/day. When seen in 2009, she remained free of symptoms on a high dose of risperidone.

Patient 2

Patient 2, a 30-year-old woman from Lithuania, was referred to mental health services by her general practitioner in June 2009 for a psychiatric assessment due to constant complaints of a flickering sensation on one side of her face leading to pain around her eye and the sensation of her eye being "sucked in" along with her brain shrinking. She was terrified of these experiences and worried that she would lose her job as her concentration had been poor. The situation had been frightening her for the past 3 months and was gradually worsening.

When asked about her psychiatric history, the patient reported that her symptoms started in 2000, when she came to the United Kingdom after her divorce; she noted that she started to experience pain symptoms soon after this. She mainly described "bodily aches" which failed to respond to analgesics, a feeling of being hit constantly by tennis balls, and a pulling sensation, especially in her legs and genital area at night. Due to an inadequate command of English, the subtleties of her psychical experiences were merely reported as "aches" to her doctor. She felt that her problems were not being understood, and she continued to present over a period of 2 years with pain in different areas of her body, which she described as "electric shock-like sensations in the lower part of the body." On one occasion in 2002, the patient reported hearing someone calling out her name and feeling trapped in her own body and unable to call out for help. Following this frightening experience, she flew back to Lithuania for treatment. On further questioning, the patient reported that, while in Lithuania, she was started on a combination of antipsychotic and antidepressant drugs, namely sertindole and duloxetine, which relieved her symptoms.

When she was referred for a psychiatric assessment in 2009, she requested an increase in her antipsychotic

medication, which she usually obtained from Lithuania. Following the psychiatric assessment in 2009, a diagnosis of cenesthopathic schizophrenia was made and her antipsychotic medication was changed to olanzapine with a maximum dose of 15 mg/day. When the patient was seen for follow-up appointments, her symptoms were greatly reduced in intensity and frequency, allowing her to sleep well and concentrate on her work during the day and have a better quality of life. The patient continues to be followed in our psychiatric clinic.

Patient 3

Patient 3, a 40-year-old man from Somalia, was referred by the community mental health team to the specialized somatoform disorders clinic in January 2009 with a 15-year history of symptoms of discomfort and pain which had failed to respond to analgesics, antidepressants, and anxiolytics. A review of the patient's case notes revealed that a diagnosis of schizophrenia was made in Somalia in 1989, which was later confirmed in 1994 in the United Kingdom, at which time the patient was suffering mainly from somatic and auditory hallucinations (e.g., a feeling of pain being inflicted on his limbs, voices commenting on his actions and calling out his name). At that time, the patient was started on the antipsychotic trifluoperazine. His symptoms remitted completely and he was weaned off antipsychotic medication over the coming year and subsequently discharged from psychiatric services. After this, he only engaged with mental health services intermittently, continuing to complain about peculiar bodily symptoms, associated with feelings of depression and anxiety. Between 2000 and 2002, the patient remained stable without psychiatric medication, and only minor psychopathological symptoms were noted, mainly mild depressive symptoms. In 2005, he again presented to his doctor with "pain all over his body." The doctor treated the patient with various analgesics which failed to alleviate his distress. He was referred to a rheumatologist and then to an orthopedic surgeon for further investigations and pain relief, but no organic disorder was diagnosed. He had several physiotherapy sessions which gave him little relief. He was also started on antidepressants and anxiolytics with little benefit. At this point, the general practitioner referred the patient to mental health services for assessment. Despite knowing about the history of a diagnosis of schizophrenia, the consultant psychiatrist who saw the patient in 2005 explicitly concluded that the patient "...didn't really

suffer from any psychotic illness.” Instead, a diagnosis of anxiety disorder was made, and symptoms of obsessive-compulsive disorder were also noted. The patient was restarted on antidepressants and discharged back to his primary care doctor.

During his initial assessment in our somatoform disorders clinic, the patient was constantly stretching and turning from side to side. When directly questioned about this, he said that he was trying to obtain his “body balance.” He described an abnormal, uncomfortable sensation of left-sided tightness in his body and said that he had begun swimming to obtain relief from this tightness. The patient believed that this imbalance was somehow connected to his head. He said that sometimes he experienced the feeling that the left side of his head disappeared, which was something he said that only he could feel, while it was not visible on the outside. The psychiatric interview revealed delusional thoughts of a persecutory nature (he strongly believed that his ex-partner in Somalia performed black magic on him which he felt changed his body, head, and personality) and somatic passivity phenomena (ex-partner causing him direct physical discomfort). The patient had no history of drug or alcohol abuse nor was there any family history of mental illness. A diagnosis of cenesthopathic schizophrenia was made and he was referred to the local community mental health team, with a recommendation that he be started on antipsychotics. He was initially treated with aripiprazole but refused to take the medication due to potential side effects. He has since been treated with olanzapine, so far with only little improvement.

Patient 4

Patient 4, a 30-year-old man from Bangladesh, was seen in the somatoform disorders clinic following an overdose of tranquilizers and because he presented with a range of medically unexplained somatic symptoms, including headaches, tinnitus, ear aches, dizziness, and vertigo. He went to his general practitioner in December 2008 with complaints of “ringing” in his ears. His tinnitus was extensively investigated with no positive results (including a specialist otorhinological assessment). He also reported a 7-month history of insomnia to his doctor, for which he was prescribed tranquilizers which did not help. In a desperate attempt to overcome his insomnia, he took an overdose of pain killers and came in contact with psychiatric services in February 2009 when he was initially

referred to the day hospital for acute treatment. The patient was diagnosed as suffering from somatoform pain disorder. He was described as an anxious person and was prescribed antidepressants for his anxiety problems with no clinical efficacy noted. In June 2009, he was assessed in the somatoform disorders clinic, at which time he reported a 2-year history of having the sensation of ringing and hissing in his head which moved from ear to ear; he said that he could feel his whole body “swinging” when he went to sleep at night. He acknowledged that his body would be resting on the bed without any visible movement but that he could feel his body moving which scared him and hindered his sleep. Apparently, his symptoms started 2 years previously after he donated bone marrow to his uncle who was suffering from cancer. He described the procedure as physically painful and said that it had somehow interfered with his normal self and that he felt that this gradually brought on the changes and symptoms in his body. As a result of his distressing bodily symptoms, he had stopped working as a full time chef 2 years earlier. During the psychiatric assessment, he described a feeling as if a lump would be coming out of his neck or growing into his throat, he felt as if his ear was blocked, and he described feeling that he had “too much oxygen” in his body. Simultaneously, he noticed that his voice sometimes changed in tone and he was experiencing what he described as a “pulling sensation” on his skin. Asked how he explained these sensations, he proposed a rather complicated pseudoscientific explanation for them. He scored high on the “boundary loss” and “somatic depersonalization” subscales of the Body Distortion Questionnaire,¹⁹ as well as on the somatoform disorder screening instruments that were administered, the Patient Health Questionnaire (PHQ-15)²⁰ and the Screening for Somatoform Symptoms-7 (SOMS-7).²¹ Based on the peculiar nature of the patient’s abnormal bodily sensations, a diagnosis of schizophreniform disorder was considered, and the assessment concentrated on the presence of other psychotic phenomena. At this point, auditory hallucinations and paranoid delusions were elicited. The patient admitted hearing the voices of members of his family talking to him on and off even though they were clearly physically absent. He also reported that sometimes at night he sensed a presence “as though somebody might be there.” In August 2009, the patient was given a formal diagnosis of cenesthopathic schizophrenia. Risperidone 1 mg/day was prescribed which was sub-

sequently increased to 4 mg/day. There is some early indication of positive response to treatment.

Discussion

In this article, we described the cases of four patients who presented to primary care physicians with predominant bodily complaints, who were then referred to our specialized somatoform disorders clinic. For a considerable period of time, all four patients did not present with any signs or symptoms suggestive of a mental disorder other than those qualitatively abnormal body experiences. In the first case, symptoms started with hot flushes and abnormal thermic sensations; the patient then developed anxiety and depressive symptoms. In the second case, it must be acknowledged that a language barrier may have prevented the patient from expressing her concerns more fully and comprehensively, which could have been easily resolved by using interpreters for each consultation in order to obtain a full history. The peculiar nature of the patient's physical complaints could nevertheless have pointed towards a psychotic condition much earlier, but it took some years until she was referred to a psychiatrist. The symptoms described by the third patient were misdiagnosed mainly because his psychiatric history was not known when he presented to his general practitioner 5 years after his last psychotic episode, but also because he did not present with any specific first rank symptoms when he was seen by a psychiatrist. In the fourth case, the peculiar nature of the range of cenesthetic sensations with which the patient presented, as well as the decline in his overall functioning, led to a different focus of the mental state examination, which revealed a diagnosis of schizophrenia.

When assessing the quality/phenomenology of medically unexplained physical symptoms and taking a psychiatric history, cenesthetic sensations (cenesthesias) need to be kept in mind with regard to a differential diagnosis of prodromal schizophrenia and more specifically cenesthopathic schizophrenia.¹¹ When this is done, unnecessary investigations, misdiagnoses, inappropriate use of health services, and, most important, delay in appropriate treatment can in some cases be avoided. It has been well established that the duration of untreated psychosis has a clear impact on long-term prognosis. Even though pharmacological (antipsychotic) treatments are not currently recommended for patients in early prodromal states, a range of nonpharmacological interventions can be considered,

including novel therapies such as arts/non-verbal, body-oriented psychological therapies.²² It is hoped that this article will stimulate further research on the nature, course, and prognosis of cenesthesias in the development of psychosis as well as the clinical relevance of the subtype "cenesthopathic schizophrenia."

COMMENTARY by

Roberto Lewis-Fernández, MD

Differentiating between psychosis and other forms of psychopathology in a diagnostic interview can be notoriously difficult, particularly when the diagnostician and the patient are from different cultural backgrounds and when language barriers complicate the assessment. Subtle and protracted expressions of psychosis may be hard to distinguish from unfamiliar but nonpsychotic expressions of somatic distress, dissociative symptoms precipitated by traumatic exposure, or cultural idioms of distress that are nonspecifically associated with anxiety or depression. Culturally particular explanations of how the body, the mind, and/or the spirit interact with the environment to produce a symptom (the person's "explanatory model") can sound bizarre and even psychotic to clinicians accustomed to a biomedical model of illness. Even seasoned cultural consultants can be trumped, and time and substantial contextual information often provide the most useful diagnostic data.

These cases, presented by Röhricht and Gudi, illustrate this diagnostic conundrum in four immigrants to the United Kingdom from very diverse cultural backgrounds. All four are eventually diagnosed with cenesthopathic schizophrenia, an ICD-10 diagnosis not included in DSM-IV-TR. A major strength of the article is the careful documentation of the clinical course over several years, allowing the reader to witness the unfolding of various diagnostic and treatment decisions over time. By the end of each case, the authors are convinced that the eventual diagnosis of cenesthopathic schizophrenia is correct and that unnecessary delay, treatment error, and patient suffering could have been averted by reaching this conclusion earlier. The evidence for psychosis as a possible explanation for these patients' presentations is indeed compelling at different points in the case reports.

Without knowing more about the cultural perspective for each case, however, the conclusions are more tentative. It would be fascinating for us to sense the voices of the patient and his or her social network.

What was their understanding of the problem? What contextual or interpretive factors could help clarify whether or not psychosis was actually present, such as cultural prototypes for this type of presentation, or information about possible metaphoric meanings for specific attributions, such as sorcery? When presented, patients' views are taken somewhat uncritically as evidence of psychosis, such as the images used to describe their pain (e.g., "fire trapped in the body") or the attribution of a supernatural etiology. These, by themselves, are not unusual in many cultural groups. The differential in West African patients for somatic symptoms that may sound bizarre to a Western psychiatrist, but are nonetheless nonpsychotic, is famously broad, such as a range of heat and pain sensations, "water in the brain," or "worms crawling inside the body."²³ Reports of altered perceptions, such as transient auditory hallucinations, can likewise be mistaken for a psychotic disorder, whereas they usually indicate subclinical distress or nonpsychotic psychopathology. For example, approximately 10% of Latinos in the United States describe altered perceptions such as "hearing their name called" or "seeing visions" on a standardized diagnostic interview, but only 7% of positive endorsers of these symptoms meet Structured Clinical Interview for DSM-IV criteria²⁴ for a psychotic disorder.²⁵ Descriptions of pain experience can be protean, and the more severe and protracted they are, the harder the patient seems to struggle to convey them in language and to overcome the desperation they produce.²⁶ Differential diagnosis of all of these expressions requires careful parsing of patients' verbal reports, which is much harder when clinicians and patients do not share the same primary language or metaphoric schemas. Interpreters, cultural brokers, or other cross-cultural experts can play an extremely helpful role in cases such as these in validating our clinical diagnoses by Western standards, or opening us to the possibility of a different understanding within a cultural context.

Despite these limitations, the four cases raise important questions about the usefulness of a diagnostic category for chronic, somatically focused psychosis. The current DSM-5 process should review the overlap between the ICD-10 diagnosis of cenesthopathic schizophrenia and the related DSM-IV-TR category of delusional disorder, somatic type. Nevertheless, either diagnosis, or a consensus category, should be made in cross-cultural settings only after careful cultural evaluation. The DSM-IV Cultural Formulation, a systematic method for assessing cultural factors in a clinical

encounter, can be a useful guide to diagnosticians in conducting this type of assessment.^{27,28}

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