Delusion of oral parasitosis and thalamic pain syndrome: A case report

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Abstract

BACKGROUND: Delusional parasitosis is an uncommon psychiatric condition in which patients have the immutable conviction that small, living organisms, such as worms, insects, or larvae infest their skin or other organs. Objective/METHOD: The authors describe a case of an unusual association of delusional parasitosis and thalamic pain syndrome after left-posterior thalamic hemorrhage. The patient initially suffered from dysesthesia and burning pain typical of thalamic pain syndrome and subsequently developed delusional oral parasitosis ("worms" infesting her mouth). RESULTS: Sulpiride 100 mg/day administered in addition to amitriptyline gradually improved her delusions within 3 months. DISCUSSION: The authors speculate that this specific type of delusion can be elicited by the disruption of the somatosensory pathway and that the subsequent cortical sensory deafferentiation and reorganization arising from this disruption may contribute to the development of delusional parasitosis.

Delusional parasitosis (DP) is an uncommon psychiatric condition in which the patient has the immutable conviction, despite the complete lack of corroborating evidence, that small living organisms such as worms, insects, or larvae infest their skin or occasionally their intestinal organs.[1] An isolated form of DP is classified under monosymptomatic hypochondriacal psychoses (MHPs), which are delusional disorders of the somatic type.[2] Further, DP is often associated with a wide variety of illnesses, including physical conditions, psychiatric disorders, and substance abuse.[3] In contrast, thalamic pain syndrome or Dejerine-Roussy syndrome is a neurological disorder resulting from a specific lesion in the thalamus. It is clinically characterized by dysesthesia, allodynia, absent or decreased sensation in the contralateral half of the body, and transient hemiplegia and hemiataxia.[4, 5] Dysesthesia is a unpleasant abnormal sensation, either spontaneous or evoked, and allodynia is pain evoked by stimuli that are not normally painful. In thalamic pain syndrome, dysesthesia and allodynia are characterized by severe scalding, shooting, or burning pain. The development of DP following structural lesions in the brain is rare. To our knowledge, the association of DP and thalamic pain syndrome has not been previously described. We present a case of DP secondary to left posterior thalamic hemorrhage in which DP developed following the diagnosis of central post-stroke pain syndrome. We speculate that cortical somatosensory deafferentation and the subsequent reorganization associated with the disruption of the spino-thalamic-cortical pathway may have contributed to the development of DP in this patient.

Case Report

The patient was a 64-year-old right-handed housewife who had adequate spousal support. She had a history of hypertension and did not have a previous history of psychiatric illness or substance use. At the age of 62, she experienced her first episode of stroke, which resulted in gait ataxia. Magnetic resonance (MR) imaging of the brain revealed a small localized infarction in the right cerebellum. The patient was admitted to the hospital for rehabilitation. She was first referred to the psychiatric outpatient department because of emotional labiality. At presentation, she exhibited depressive mood and irritability. She was treated with paroxetine 20 mg/d; this provided relief from her depressive symptoms. The patient was discharged after 6 months. At this time, paroxetine administration was discontinued. At the age of 63, 1 year after the first

stroke episode, she experienced a sudden onset of numbness and right hemiparesis. A computed tomography (CT) scan of the brain revealed a massive hematoma in the left posterior thalamic region that extended to the internal capsule and the head of the left caudate nucleus (Fig. 1A). The patient was admitted to the hospital and was treated with intravenous glycerol therapy for 1 week. Laboratory work-up, including complete blood cell count, thyroid function tests, urinalysis, serum electrolyte levels, renal and liver function tests, glucose levels, syphilis serology, and electrocardiography, was unremarkable. After 2 weeks, the patient experienced a spontaneous burning sensation that was predominantly confined to her right hand and to the right half of her face. A serial brain CT scan revealed that the left-sided hemorrhage had started to resolve (Fig. 1B). She was alert and cooperative but slightly dysarthric. Mental-state examination did not reveal cognitive deficit. Neurological examination revealed right hemiparesis and ataxia. Sensory assessment revealed that hypesthesia and dysesthesia were present in the right extra-perioral, hand, and leg regions; the sense of vibration was also reduced. Spontaneous burning pain predominantly occurred in the distal portion of her right hand and right perioral region and was also easily elicited by vibration and cold-sense tests that were performed using a tuning fork. A diagnosis of thalamic pain syndrome (Dejerine-Roussy syndrome) was suspected. She was treated with amitriptyline 20 mg/d. The spontaneous burning sensation persisted for 8 weeks but gradually improved over time.

Three months after the second stroke, she complained that many live worms had infested her oral cavity and were lodged between her teeth. She often looked into her mouth in a mirror for several hours and extensively brushed her teeth to remove the worms. She insisted on being referred to a dentist for a check-up and for the extermination of the small worms. The dentist did not find any evidence of an infestation. She expressed displeasure at the dentist's assessment and insisted: "I am trying to crunch and swallow the worms, but they don't disappear. I can really feel the worms attached to the back of my teeth, and they sometimes migrate along both sides of my gums. Unfortunately, I can't find them with a mirror, but I'm sure they are playing hide-and-seek in my mouth." Neurological examination revealed right ataxic hemiparesis. Sensory assessment revealed that hypesthesia and reduced vibration sense persisted in the right side of her body. Dysesthesia was present in the right extra-perioral and hand regions, and its severity was reduced, while allodynia was

absent. She refused to believe that dysesthesia was responsible for her sense of having a worm infestation. Sulpiride 100 mg/d administered in addition to amitriptyline gradually improved her delusions within 3 months. She reported that there were fewer worms in her mouth, and that they had stopped playing hide-and-seek. Sulpiride administration at a dose of 100 mg/d was maintained thereafter. During the subsequent 1.5-year follow-up period, she was almost free of the symptoms of delusions.

Discussion

Our patient held an unwavering belief, despite overwhelming evidence to the contrary, that small worms inhabited her mouth; this indicated a diagnosis of DP. Furthermore, in our patient, an unusual association was observed between DP and thalamic pain syndrome secondary to posterior thalamic hemorrhage. The primary manifestation of sensory phenomena in our patient was the thalamic pain syndrome. Unexpectedly, within the subsequent 3 months, the dysesthesia in our patient was transformed into a worm-crawling sensation accompanied by delusional belief.

Dysesthesia and allodynia due to thalamic pain syndrome may begin immediately or shortly after a stroke. Occasionally, the pain may spontaneously subside, but more commonly, it becomes intractable. The pain experienced in thalamic pain syndrome is regarded as central pain since patients with this syndrome usually have ipsilateral sensory impairment and contralateral posterior thalamic lesions, which interrupt spinothalamoparietal projections.[5] The underlying neuromechanisms of central pain are unclear. A modern hypothesis proposes that central pain results from a lesion that is located within the lemniscal system and produces an imbalance between the old and new pain pathways in the spinothalamic tract. Moreover, it disinhibits the old pain pathway that projects to the anterior cingulate cortex.[6, 7] Another theory states that hyperactivity and plastic change in the deafferentiated parietal neurons induce spontaneous painful sensations.[8, 9] Our patient developed hypesthesia and dysesthesia in the right side of the body and subsequently a burning sensation in the right hand and right perioral region. Serial CT scanning revealed a lesion in the ventroposterior thalamic region; this lesion was thought to be crucial in the development of thalamic pain syndrome.[7] The chronological changes in the sensory symptoms in our patient suggest that cortical somatosensory deafferentation resulting from the disruption of the spino-thalamic-cortical pathway and the subsequent reorganization may have

contributed to the development of DP. There are 2 pathophysiological explanations for DP: (1) the delusions were the manifestations of a primary thought disorder or (2) the delusions were secondary to tactile hallucinations.[2, 3] In our case, the findings strongly supported the latter. Furthermore, these findings also suggest that disconnected cortical sensory systems possibly contribute to the development of tactile hallucinations.

The onset of DP symptoms following a stroke is usually delayed.[10, 11] The delayed onset of symptoms suggests that more complex mechanisms such as cortical plastic change or reorganization may be involved. Interestingly, Kato et al[12] reported the case of a woman who complained about the sensation of a "metal-like thing" in her oral cavity, 4 years after a right caudate stroke. They described a possible association between somatic delusional psychosis and cortical reorganization. Interestingly, a recent discovery related to phantom pain indicated that the phantom limb sensation in a nonexistent amputated limb possibly originated from cortical sensory systems, and that plastic changes in the sensory cortex played an important role in pathological sensation.[13]

The relationship between the location of a lesion and delusion is of particular interest. There have also been several reports of DP following structural brain lesions. However, the type of lesion involved varied, e.g., occipito-temporal, parietal, putaminal, and temporoparietal.[10,11,13,14] Delusional disorders can be induced by lesions in a variety of locations; most often however, lesions are found in the limbic and subcortical structures such as the basal ganglia and thalamus. These structures form basal ganglia-thalamocortical loops, and lesions that disrupt these loops can be expected to alter belief and emotional behaviors.[16]

Medication with sulpiride was well tolerated in our patient, and gradual improvement was seen. Antipsychotic medications are usually effective in controlling the symptoms of DP, and a relatively good long-term prognosis was reported.[1] As individuals with DP are frequently elderly, the selection of drugs whose adverse effect profiles are benign is considerably important.

In conclusion, in our patient, an unusual association of DP and thalamic pain syndrome secondary to a posterior thalamic lesion was seen. The chronological transition of sensory symptoms supports the sensorialist approach: tactile hallucinations secondary to elaborate delusions. We suggest that cortical somatosensory

deafferentation and the subsequent reorganization resulting from the disruption of the somatosensory system may contribute to the development of DP.

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Figure legend

Figure 1: A and B, Head CT scan of the patient. A. A massive hematoma in the left posterior thalamic region extending to the internal capsule and the head of the left caudate nucleus. B. Head CT performed 2 weeks later, showing a lesion in the left ventroposterior thalamic region.