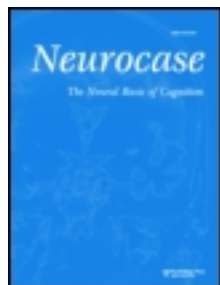


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Pathological generosity: An atypical impulse control disorder after a left subcortical stroke

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Pathological generosity: An atypical impulse control disorder after a left subcortical stroke

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Changes in socio-emotional behavior and conduct, which are characteristic symptoms of frontal lobe damage, have less often been described in patients with focal subcortical injuries. We report on a case of pathological generosity secondary to a left lenticulocapsular stroke with hypoperfusion of several anatomically intact cortical areas. A 49-year-old man developed excessive and persistent generosity as he recovered from a left lenticulocapsular hematoma. His symptoms resembled an impulse control disorder. ^{99m}Tc-HMPAO SPECT demonstrated hypoperfusion mostly in the ipsilateral striatum, dorsolateral, and orbitofrontal cortex. This case study adds pathological generosity to the range of behavioral changes that may result from discrete unilateral lesions of the lenticular nucleus and nearby pathways. In our particular case, post-stroke pathological generosity was not ascribable to disinhibition, apathy, mania, or depression. Because pathological generosity may lead to significant distress and financial burden upon patients and their families, it may warrant further consideration as a potential type of impulse control disorder.

Keywords: Frontal lobe syndrome; Basal ganglia; Stroke; Generosity; Impulse control disorder.

Generosity refers to an individual's capacity to sacrifice material means to benefit another individual or group even when no material profit or gain in reputation is expected in return (Lewis, 1960). Functional neuroimaging studies have shown that typical acts of generous behavior, such as anonymous charitable donations, recruit the mesolimbic reward system, especially the ventral striatum and adjoining septal nuclei, the ventral tegmental area, and the subgenual cingulate cortex (Moll et al., 2006). Pathological generosity (PGE) is a classical manifestation of mania, where it is

usually described as "prodigalism" (Leonhard, 1995). It has also been sporadically noted in cases of dementia (Dorey et al., 2008), neurosyphilis (Vallejo-Nágera, 1944), Parkinson's disease treated with dopaminergic drugs (O'Sullivan, Evans, Quinn, Lawrence, & Lees, 2010), and in the temporal variant of frontotemporal dementia (Mendez, Chen, Shapira, Lu, & Miller, 2006).

Because it is often difficult to disclose from the available reports if PGE was not but one of the symptoms of a manic state secondary to brain damage (Starkstein et al., 1990), it remains to be shown

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that PGE can occur as a symptom of brain damage in its own, especially outside the context of a manic state. PGE is in many respects the opposite of pathological hoarding (Vallejo-Nágera, 1944) and the selfish inclinations of acquired sociopathy (Barrash et al., 2011). Theoretically, PGE could also exist as a standalone symptom, in which case it might resemble an impulse control disorder (ICD). Therefore, the study of acquired PGE due to brain damage may provide novel insights into the neural substrates mediating the delicate balance between altruism and egoism, which make up one of the pillars of ordinary social motivation and decision-making.

The changes in personality traditionally observed in patients with prefrontal cortical lesions have also been reported on individuals with discrete lesions of subcortical structures, such as the globus pallidus (Laplaine et al., 1989; Strub, 1989). Nevertheless, we are not aware of any case of an isolated PGE as a main symptom of a focal subcortical lesion. In this article, we report on the case of a patient that developed PGE after a left lenticulocapsular stroke.

CASE REPORT

Mr. A, a right-handed 49-year-old man, presented with an inability to manage his financial life, which was first noticed one year previously as he recovered from an intraparenchymal hypertensive hemorrhage of the left lenticulocapsular region. His managerial problems were part of a broader change in personality, which was reported by his wife and teenage son over several interviews. Previously, Mr. A ran one of the departments of a large corporation where he was a distinguished and reliable employee. He was cautious and responsible, with a good relationship with his family and a few close friends. He spoke little, but enjoyed discussing his points of view, sometimes hotly. Always conciliatory, “he would never initiate an argument, but would reply strongly when challenged.”

After the stroke, personality changes, including PGE, hindered Mr. A from resuming his job and coping financially with his family. He would spend his money with street kids, buying them soda, candies, and junk food. There was no evidence of pedophilia, as emphasized by his wife, brother-in-law, and friends, who were always with him when he was out. Were it not for the close attention of Mr. A’s wife, he would be constantly in debt. At one point, Mr. A and his brother-in-law started a

small business of homemade fries, which eventually failed because he did not worry about debtors and because he gave away most of his product. If confronted by his mother or wife, he would make a joke and evade discussion. If pressed further, he sometimes would become angry and then leave the house for a while.

When first seen by our clinic, Mr. A was oriented to time and place, though appearing to be extremely fatigued owing to insomnia, as he was sleeping less than 4 hours a day. His answers were appropriate, but he had little emotional expression and his speech and gestures were slow and brief. Mr. A reported feeling depressed, without sexual initiative and interest in life. He complained of inattention, forgetfulness, and of missing some appointments. Of note, he denied weight/appetite changes, worthlessness, excessive or inappropriate guilt, and suicidal thoughts and/or behaviors. When asked if he intended to resume his former occupation, he replied that he had already worked enough and that it was now time “to enjoy life, which is too short.”

In addition to the above-mentioned depressive symptoms, Mr. A also exhibited some frontal lobe behavioral features, including blunted affect, lack of persistence and planning, puerility, and impaired judgment. He had partial insight into his behavioral and personality changes, often claiming, “I saw death from up-close, now I want to be in high spirits.” No features of behavioral disinhibition were present, other than the PGE behavior. Nevertheless, indecisiveness and tactlessness were absent. Moreover, there was no evidence of a mixed state (i.e., manic symptoms), including abnormal and persistently elevated, expansive, or irritable mood, nor inflated self-esteem or grandiosity, talkativeness, flight of ideas, or racing thoughts. The patient did not meet the criteria for dementia (see neuropsychological evaluation below).

Mr. A had the MRC grade 4 right hemiparesis, which interfered little or not at all with his balance, gait, and manual skills. His right knee jerk reaction was brisk, but his plantar responses were flexor bilaterally. There were no sensory or sensitive deficits. Neuropsychological examination revealed normal visual and tactile perception and recognition, constructional abilities, verbal fluency, abstraction, and memory. On the Wisconsin Card Sorting Task, Mr. A completed 6 categories, without failures to maintain set and taking 12 trials to complete the first category. MRI revealed a left subcortical lesion extending from the lenticular nucleus into the white matter that borders

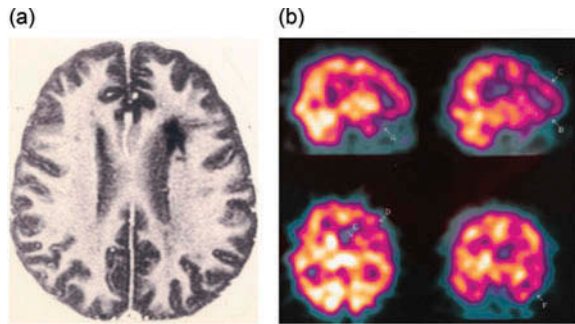


Figure 1. (a) MRI showing the left lenticulocapsular hypointensity and (b) ^{99m}Tc -HMPAO SPECT showing hypoperfusion of the ipsilateral caudate and lenticular nuclei (A) and normal perfusion of the ipsilateral thalamus (B). Hypoperfusion also evident in the dorsolateral (A) and orbitomedial (D, E) surfaces of the left frontal lobe and contralateral orbitofrontal cortex (F). [To view this figure in color, please see the online version of this Journal].

the angle of the frontal horn, which was focally enlarged (Figure 1(a)). ^{99m}Tc -HMPAO SPECT showed hypoperfusion in the projection of the anatomical lesions and in both cerebral hemispheres, mainly the ipsilesional (Figure 1(b)).

His medical history included hypertension and a major depressive episode with suicidal ideation 5 years before the stroke, from which he recovered after being treated with amitriptyline. His father had distinct episodes of hypomania and depression, and one of Mr. A's brothers committed suicide by hanging. Mr. A's treatment was followed for up to 4 years.

Initially, because of his depressive symptoms, he was re-prescribed amitriptyline, 100mg/day (plasma levels = 70,2 ng/ml; therapeutic range = 50–150 ng/ml). Core depressive symptoms (sadness and anhedonia), along with loss of interest in life and sexual initiative, fatigue, insomnia, psychomotor retardation, and inattention all remitted within a few weeks. He commented that he was “cured and as happy as before.” PGE did not noticeably change regardless of whether he was depressed or not. After almost 2 years of treatment, amitriptyline was gradually withdrawn and Mr. A was lost to follow-up.

DISCUSSION

This study reports on the case of a 49-year-old man who developed PGE subsequent to a left lenticulocapsular hemorrhage with hypoperfusion of both hemispheres, mainly the ipsilateral. PGE was pathological because it interfered with job

resumption, family finances, and the family business. Of note, post-stroke PGE was not ascribable to current disinhibition, apathy, mania, or depression, as indicated by the clinical details that exclude these disorders. Despite a severe major depressive episode 5 years before the stroke, careful clinical examination and extensive interviews with his relatives disclosed no evidence of a manic/hypomanic episode during his lifetime. The challenges posed by this case include psychopathological concepts, clinicoanatomical inferences, and putatively disturbed neural processes involved in the genesis of Mr. A's change in behavior.

It is difficult to fit isolated PGE into current classificatory systems. Classical descriptions conceptualized PGE as a loss of the “instinct of property”—the opposite of hoarding (Vallejo-Nágera, 1944). Clearly, PGE has clinical similarities with compulsive shopping and pathological gambling, which are two typical instances of ICD (Schreiber, Odlaug, & Grant, 2011). Mr. A usually optimistically downplayed the implications to his PGE (e.g., “I saw death from up-close, now I want to be in high spirits”), and also became quite upset when confronted about his change in personality. Arguably, one could suggest that the purposeful, proactive, and enjoyable characteristics of Mr. A's PGE may indicate a goal-directed, reward-dependent behavior that resembles an ICD. This hypothesis is supported by neuroimaging studies of anonymous charitable donations in healthy adults, whereby the activation of the subgenual cingulate cortex and closely related “reward system” structures, such as the ventral striatum, has been a robust finding (Moll et al., 2006).

Mr. A's case suggests that PGE should be included in the range of acquired behavioral changes that result from subcortical lesions, which have included impulsive behaviors such as pyromania (Bosshart & Capek, 2011), compulsive behaviors such as checking and counting (Daniele et al., 1997; Laplane et al., 1989), and impulsive-compulsive behaviors such as hoarding (Slama, Amrani, Leboyer, & Houenou, 2012). Other common changes include psychomotor inertia, apathy, autoactivation deficits, and motor stereotypies (Dubois, Funkiewiez, & Pillon, 2005). The widespread, albeit not generalized, hemispheric hypoperfusion in our case suggests that subcortico-cortical projections likely played a significant role in the determination of Mr. A's behavioral symptoms.

We can only speculate on how such a complex anatomic lesion might ultimately generate the

equally complex and persistent personality change of our patient. Since Mr. A's lesion predominantly involved the associative sector of the lenticular nucleus, hemorrhage is likely to have started in one of the penetrating branches of the medial lenticulostratial arteries (Feekes & Cassell, 2006). However, in contrast to the effects of unilateral or bilateral lesions of the cognitive sector of the lenticular nucleus (Bhatia & Marsden, 1994), the primary symptoms exhibited by Mr. A were mostly behavioral. Therefore, we believe that the enduring personality change shown by Mr. A (as well as his depressive symptoms) resulted mainly from the injury of white matter pathways, in particular of the heterogeneous fiber systems of the external and extreme capsules, as well as in the fronto-occipital fasciculus, which runs over the head and body of the caudate nucleus, in the vicinity of the frontal horn (Aralasmak et al., 2006).

The observation of hypoperfusion of anatomically intact regions—at a distance from the lesion focus, but connected with it by well-known anatomic fiber pathways—supports this hypothesis. The cortical deafferentation produced by discrete white matter damage in our case might have disrupted the interplay of neural systems that underpin key dimensions of personality, such as extroversion–introversion (Mendez et al., 2006). Part of Mr. A's personality change may be explained by disruption of ascending dopaminergic pathways from the ventral tegmental area, which has been directly linked to the regulation of extroversion (Depue & Collins, 1999); for example, extraversion has aroused as an adverse behavioral feature of the response to certain dopaminergic drugs (O'Sullivan et al., 2010).

It is difficult to speculate on the role of laterality in a case such as ours. For instance, whereas left-sided lesions have been associated both with post-stroke depression (Terroni et al., 2011) and with impulse control disorders including kleptomania (Aizer, Lowengrub, & Dannon, 2004; Kaplan, 2007), pyromania (Bosshart & Capek, 2011), and risk-taking behaviors (Floden, Alexander, Kubu, Katz, & Stuss, 2008), the right inferior frontal cortex has also been implicated in impulsivity (Chamberlain & Sahakian, 2007). Tranel and colleagues (Tranel, Damasio, Denburg, & Bechara, 2005) have suggested that laterality does not determine impulsivity *per se*, but interacts with gender in this regard.

It might be argued that depressive passivity produced Mr. A's lack of restraint in giving things away, since depression may aggravate both

compulsive (Viswanath et al., 2012) and impulsive behaviors (Romer Thomsen, Callesen, Linnet, Kringelbach, & Moller, 2009). However, we do not believe that this explains Mr. A's PGE, because his pre-stroke depressive episode was not associated with PGE and also because his PGE did not noticeably change after the successful treatment of his post-stroke depression.

Taking into account the limitations of single-case reports, this case adds PGE to the range of personality changes that might result from discrete unilateral lesions of the lenticular nucleus and associated neural pathways. Because PGE may lead to significant distress and financial burden upon patients and their families, it may warrant further consideration as a potential novel type of ICD.

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