REPORTE DE CASO CLÍNICO

Cambios en la personalidad posterior a un infarto en el núcleo subtalámico izquierdo: reporte de caso

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Resumen

El núcleo subtalámico (NST) forma parte de los circuitos córtico-subcorticales motores y no motores, y por lo tanto se encuentra involucrado en diversas funciones límbicas y asociativas. En su mayoría, la literatura relacionada al NST únicamente describe y analiza su rol en funciones motoras. Se presenta el caso de una mujer de 37 años, en el que se describen múltiples cambios cognitivos, conductuales y de personalidad que ocurrieron posterior al clipaje de un aneurisma roto de la arteria cerebral media izquierda que desencadenó un infarto en el NST izquierdo; no se halló ninguna otra lesión estructural cortical o subcortical. Posterior al incidente, la paciente demostró cambios cognitivos y conductuales, pero destacan los cambios en sus patrones de interacción socioafectivos (i.e. personalidad), los cuales pudieron estar influidos por experiencias negativas en etapas tempranas de su vida. Una evaluación neuropsicológica demostró CI normal y deficiencias atencionales y ejecutivas leves; los cambios de personalidad se documentaron usando una entrevista semiestructurada. Ésta reveló que los patrones de interacción pasaron de caracterizarse por una tendencia disfuncional por ser emocionalmente distante, poco comunicativa, malhumorada e incluso físicamente agresiva, a presentar un patrón de interacción pueril, afectivo, de preocupación emocional por los demás y sociable, lo que resultó en una mejoría en sus relaciones familiares. Los cambios cognitivos y afectivos asociados a lesiones del NST han sido explicados por su función inhibitoria y de selección de la acción dentro de los circuitos frontoestriatales. Pocos estudios describen cambios de personalidad, en su mayoría negativos, en contraste con el caso que aquí se presenta.

Palabras claves: núcleo subtalámico, personalidad, circuitos frontoestriatales, caso único

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CLINICAL CASE REPORT

Personality changes following left subthalamic nucleus infarct: a case report

Abstract

The subthalamic nucleus (STN) is part of the cortico-subcortical motor and non-motor circuits and is, thus, involved in multiple associative and limbic functions. Most literature regarding the STN describes and analyzes only its roll in motor function. We present the case of a 37-year-old woman and describe multiple cognitive, behavioral, and personality changes following surgical clipping for a ruptured left-middle cerebral artery aneurysm and a subsequent left STN infarct; no other cortical or subcortical structural lesions were found. After the incident, the patient presented with cognitive and behavioral changes, although more relevant changes were observed in her social and affective interaction patterns (i.e., personality), which were presumably influenced by negative early-life experiences. A neuropsychological assessment demonstrated normal IQ with mild attentional and executive deficiencies; personality changes were documented using a semi-structured interview. Her interaction patterns changed from a dysfunctional tendency to be emotionally distant, uncommunicative, bad-tempered, and physically aggressive to a puerile, affectionate, caring, and sociable pattern of interaction, which resulted in improved family relations. Cognitive and affective changes related to STN lesions and stimulation have, in part, been explained by the inhibitory and action-selection functions within fronto-striatal circuits, although few studies describe and analyze personality changes; most report negative changes, in contrast to the case presented here.

Keywords: subthalamic nucleus, personality, fronto-striatal circuits, single-case

Introduction

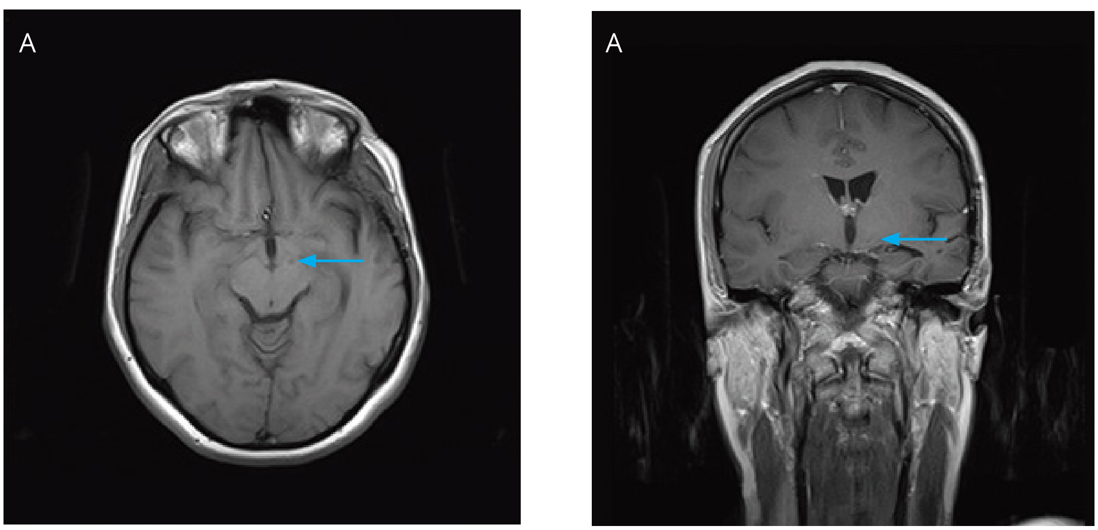
The subthalamic nucleus (STN) plays a major role in the cortico-basal circuitry due to its participation in the indirect and hyperdirect pathways, resulting in increased tonic inhibition of the thalamus and decreased activation of the cortex[1]. Thus, it has a key role in the avoidance of unwanted responses, including motor, behavioral, affective, and cognitive. Until recently, cortico-basal circuits were considered to only regulate motor activity, but it is now clear that they are also involved in associative and limbic functions[2]. Nonetheless, its participation in a complex construct such as personality through its´ implication in the inhibition or disinhibition of certain behavioral and affective patterns has not been described. Personality is a complex construct that involves dispositional traits, characteristic adaptations, and integrative life stories within a time and cultural context; it encompasses individual differences, including those in values, attitudes, personal memories, social relationships, habits, and skills, which tend to become more stable throughout one´s life[3]. Although personality is influenced by several factors, including temperament, genes, neurobiology, early life, and social experiences, evidence suggests that the increasing personality stability over the human lifespan predominantly results from environmental mechanisms[4], which, in turn, reflect learning mechanisms at a neurobiological level.

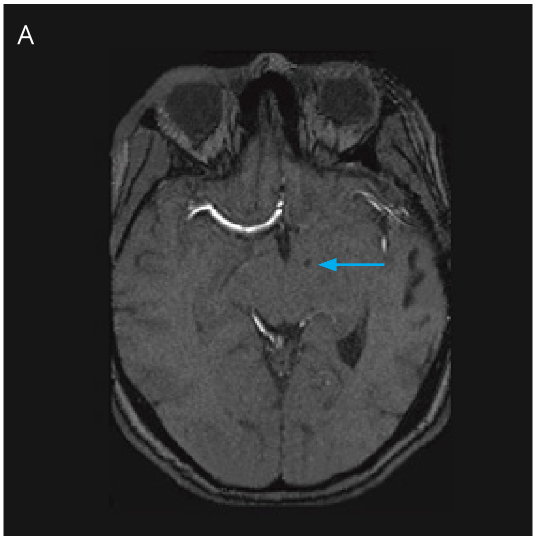
We present the case of a woman who demonstrated multiple cognitive, behavioral, and personality changes following a left STN infarct. Personality changes observed in the patient were described as positive by the family and were not explained by mild cognitive deficits. Our literature search did not render similar cases, and we think this case is important because it suggests an important role of the STN in the neurobiological basis of personality traits, which, in turn, determine the patterns of social interaction.

Case report

Ms. C., a 37-year-old divorced office employee with a high-school education, was referred by a neurologist to the Neuropsychology Service at the Specialty´s Hospital in Centro Médico Nacional Siglo XXI in Mexico City for evaluation 18 months after the sudden onset of incoherent language, personality changes, behavioral and sexual disinhibition, and right-side hemiballismus following surgical clipping for a ruptured left-middle cerebral artery aneurysm and subsequent left STN infarct (*figure 1*).

Figure 1. MRI study showing (**A**) STN lesion following surgical clipping for a ruptured left-middle cerebral artery and (**B**) FLAIR sequence showing the absence of white-matter lesions.
MRI: magnetic resonance imaging; STN: subthalamic nucleus

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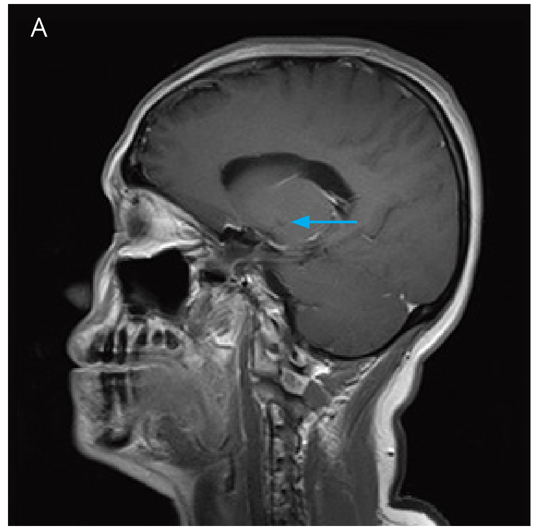
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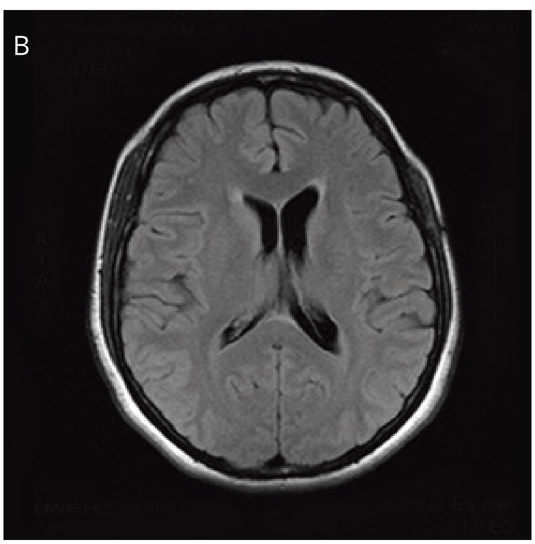
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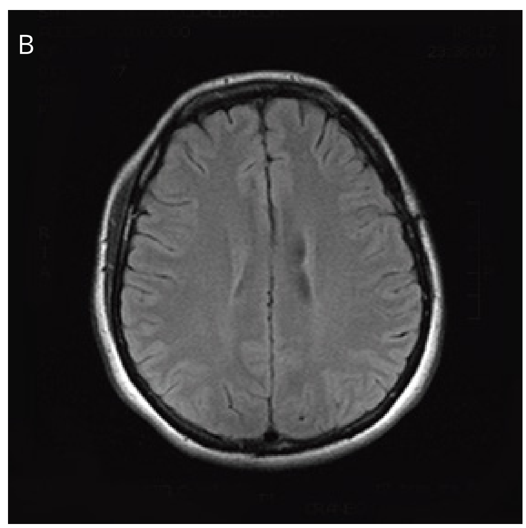
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Magnetic resonance imaging, reviewed by a trained neurologist, showed no other brain lesions. During the initial neuropsychological consultation, Ms. C. complained of attentional and memory problems, but the family (mother and sisters with whom she lived) especially reported personality and behavioral changes after the surgery. A semi-structured interview was conducted to document such changes. During the interview, the mother explained that the patient had been subjected to sexual molestation and emotional abuse by her father throughout her childhood and adolescence, which may have influenced the development of dysfunctional personality traits.

After development of the STN lesion, both the patient and her family reported a sudden change in the patient's personality (i.e., attitudes and socio-affective interaction patterns). She also demonstrated compulsive behaviors.

Additionally, a neuropsychological battery was administered to document cognitive changes. *Table 1* compares cognitive, behavioral, and personality characteristics pre and post-STN lesion. Although the cognitive profile (*table 2*) showed deficiencies, these were mild and found mainly in the attentional/executive domain, and they were not related to and did not explain the striking behavioral and personality change. The neuropsychiatric inventory showed no evidence of depression or psychotic symptoms.

Due to the theoretical importance of the case, we asked the patient and her family for informed consent for publication.

Table 1. Patient´s cognitive, behavioral, and personality changes docu-mented by a semi-structured clinical interview and neuropsychological assessment performed 18 months post-STN lesion.

	Pre-STN lesion	Post-STN lesion
Cognition	Presumed normal; able to sustain a full-time job as an office employee	→ Normal IQ (Full-scale IQ= 92) → Normal language function → Mild and isolated attentional and executive deficiencies: • Reduced backward digit span • d2 Test omission errors • 5-Digits Test counting and switching errors
Behavior	No hyperactivity or compulsive behaviors reported	Hyperactivity and compulsive behaviors including: → Picking up cigarette butts from the floor → Compulsive shopping → Hyperphagia and polydipsia
Personality	→ Emotionally distant → Uncommunicative → Self-interested → Bad-tempered → Physically aggressiveas a reaction to social stress	→ Affectionate and caring → Sociable → Puerile → Docile during all social interactions

Table 2. Results of the neuropsychological assessment performed on Ms. C. 18 months after surgical clipping for a ruptured left middle cerebral artery aneurysm and subsequent left STN infarct

Domain		Test	Scale / Subtest		Pc	T	SS	IS
Intelligence		WAIS-IV	Verbal Comprehension Index (VCI)					88
			Perceptual Reasoning Index (PRI)					100
			Working Memory Index (WMI)					85
			Processing Speed Index (PSI)					104
			Full Scale IQ					92
Memory		WMS-III	Text I	Total recall score	25		8	
			Text II	Total recall score	25		8	
			Word list I	**Total recall score**	**5**		**5***	
			Word list II	Total recall score	37		9	
				Total recognition score	84		13	
Attention		WAIS-IV	**Digit span forward**				**5***	
		D2 Test of Attention	Total characters processed		97			
			Total accurate targets		65			
			Errors of omission		**1***			
		5 Digit Test	Reading time		45			
			Reading errors		>99			
			Counting time		55			
			Counting errors		**1-3***			
Executive function	Working memory	WAIS- IV	**Digit span backwards**				4*	
	Planning	Tower of London	Total move score		37			
			Total initiation time		49			
			Total time		50			
			Total rule violation		66			
	Cognitive flexibility	WCST	Error %		27	44		
			Perseverative responses %		18	41		
			Perseverative error %		16	40		
		5 Digit Test	Flexibility		96			
	Inhibition	Stroop	Word-color		60			
			Interference		40			
		D2 Test of attention	Errors of commission		75			
			Concentration performance (CONC)		65			
		5 Digit Test	Choosing time		80			
			Choosing errors		>16			
			Switching time		85			
			Switching errors		**4-15***			
			Inhibition		90			

Pc, percentile scores; T, T scores; SS, scaled scores; IS, index scores; WAIS-IV, Wechsler Adult Intelligence Scale Fourth Edition; WMS-III, Wechsler Memory Scale Third Edition. *Scores considered below normal range.

Discussion

STN stimulation and lesions have been associated with diverse motor, cognitive, and behavioral changes in both animal models and diverse human clinical reports on hemiballismus[5] as well as with executive, attention, and memory impairment and depression, dysthymia, and mood changes[1]; hyperphagia[6]; and even modulation of obsessive-compulsive ideas and behaviors[7]. These changes have in part been explained by the inhibitory and action-selection functions of the STN[8]. In the case described here, the STN infarct resulted in not only behavioral and mild cognitive changes but also sudden and dramatic changes in the patient's social interaction and socio-affective patterns. These changes could be described as personality changes when compared to the personality the patient had developed, as consistently described by the patient's family, presumably associated with the negative early-life experiences. Few studies describe and analyze personality changes following STN lesions or stimulation. Most report maladaptive changes. Kumar, et al.[9] describe personality changes and disinhibition in one patient following STN deep brain stimulation, although no detailed description and discussion were provided. Increased anxiety and social maladjustment have also been described following STN deep brain stimulation for Parkinson´s disease. In addition, an animal model suggests that STN is involved in social behaviors. Specifically, STN-lesioned rats demonstrate fewer social contact and social exploration behaviors but also display less frequent attacking and both submissive and dominant aggressive behaviours[10]. Although the role of STN in social behaviors and its contribution to personality are not yet clear, our patient exhibited a shift in her social interaction patterns from emotionally distant, uncommunicative, bad-tempered, and physically aggressive to more affectionate, hyperactive, and less defensive and hostile. The modification of the behavior and affective patterns constituted a personality change that, in this case, led to improvements in family relations and may have resulted from STN-mediated disinhibition and a decrease in social aggressiveness.

Although personality changes have been classically described as part of orbitofrontal syndrome (OFS) (e.g., the case of Phineas Gage), and, therefore, both notions are not mutually exclusive, there are several reasons why we do not discuss this as a case of OFS. 1) OFS, as described by Fuster[11], is characterized neuropsychologically by severe alteration of attentional inhibition and presence of perseverative responses; in contrast, Ms. C´s neuropsychological assessment unequivocally demonstrates that domains classically altered in these cases are spared in her case: absence of Tower of London rule violations and D2 errors of commission and normal Stroop interference index. Additionally, our patient has normal scores regarding WCST perseverative responses and 5-Digit Test flexibility index. 2) In Ms. C´s case, we documented quite specific changes in her socio-affective interaction patterns with respect to her response tendency patterns (i.e., personality traits) previous to the neurological event in the absence of other maladaptive changes classically described in OFS, including euphoria accompanied by irritability and a contentious, paranoid stance, disregard for ethical principles, and even sociopathy.11 3) Ms. C´s personality changes presented along with other signs classically associated to STN lesions, such as hemiballismus; also, the compulsive behaviors described (hyperphagia, polydipsia, and cigarette-butts collecting) have commonly been described in patients with conditions associated to dopamine and basal ganglia dysregulation, such as punding and Parkinson´s disease-related impulse control disorders[12] and, in such cases, are not deemed to indicate OFS. The aforementioned points suggest a bottom-up dysregulation of circuits related to regulation of affect and social conduct.

The present case illustrates the importance of STN function in non-motor regulation and raises awareness of the neurobiological basis of the complex construct of personality.

Author´s Declarations

1. Financing: Nothing to declare
2. Conflicts of interest: Nothing to declare

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