

# **HYPERSEXUALITY FOLLOWING BILATERAL THALAMIC INFARCTION**

## **Case report**

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**ABSTRACT** - Hypersexuality is a rare but well recognized condition following brain injury. It has been described secondarily to dysfunction in the hypothalamus, the temporal and frontal lobes. We report a 63 year-old man that developed neuropsychological disturbances with hypersexuality as a prominent feature, disinhibition and moderate memory loss, hypersomnia and irritability after a bilateral paramedian thalamic infarction. A SPECT showed frontal hypoperfusion. We believe that these findings are expression of frontal-subcortical circuits dysfunction, particularly the orbitofrontal circuit, secondary to dorso medial thalamic infarction which probably plays a role in the determination of human sexual behavior. This case favors a thalamic modulation of frontal function.

**KEY WORDS:** hypersexuality, disinhibition, behavior, paramedian thalamic infarction, frontal-subcortical circuits.

### **Hipersexualidade após infarto talâmico bilateral: relato de caso**

**RESUMO** - Hipersexualidade é uma condição rara mas bem reconhecida após lesão do sistema nervoso central. A literatura medica registra casos secundários a disfunção do hipotálamo, do lobo temporal e do lobo frontal. Relatamos o caso de um homem de 63 anos de idade que desenvolveu alterações neuropsicológicas com hipersexualidade como característica mais proeminente, desinibição, moderada perda de memória, hipersonia e irritabilidade após infarto talâmico paramediano bilateral. O SPECT evidenciou hipoperfusão frontal. Nós acreditamos que esses achados sejam expressão da disfunção de circuitos córtico-subcorticais frontais, particularmente do circuito órbito-frontal, secundária ao infarto dorsomedial do tálamo, que provavelmente desempenha papel relevante na determinação do comportamento sexual humano. Este caso favorece uma possível função moduladora do tálamo sobre os circuitos córtico-subcorticais frontais.

**PALAVRAS-CHAVE:** hipersexualidade, desinibição, comportamento, infarto talâmico paramediano, circuitos córtico-subcorticais.

Hypersexuality and alteration of sexual preference have been recognized as rare consequences of brain injury<sup>1</sup>. Their occurrence provides important clues for understanding the anatomy and physiology of human sexual behavior. Lesions in the structures such as the amygdala, the hypothalamus, the temporal and frontal lobes have been described in association to these symptoms<sup>1</sup>. Lesion in the frontal lobe has been referred as an anatomical site that produces true hypersexuality while the other structures are more related to changes of sexual preference<sup>1,2</sup>. Frontal-subcortical circuits are implicated in the determi-

nation of many aspects of human behavior<sup>3</sup> thus raising the question whether sexuality could be one of them. Such circuits involve several structures such as striatum, globus pallidus, substance nigra and thalamus. Evidence that frontal-subcortical circuits mediate a given behavior is provided by the observation that lesions in circuit related structures alter such behavior<sup>3</sup>.

We report a patient with bilateral thalamic lesion due to ischemic stroke presenting with neuropsychological disturbances represented mainly by hypersexuality, associated to frontal hypoperfusion.

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Received 19 May 2005, received in final form 3 August 2005. Accepted 27 September 2005.

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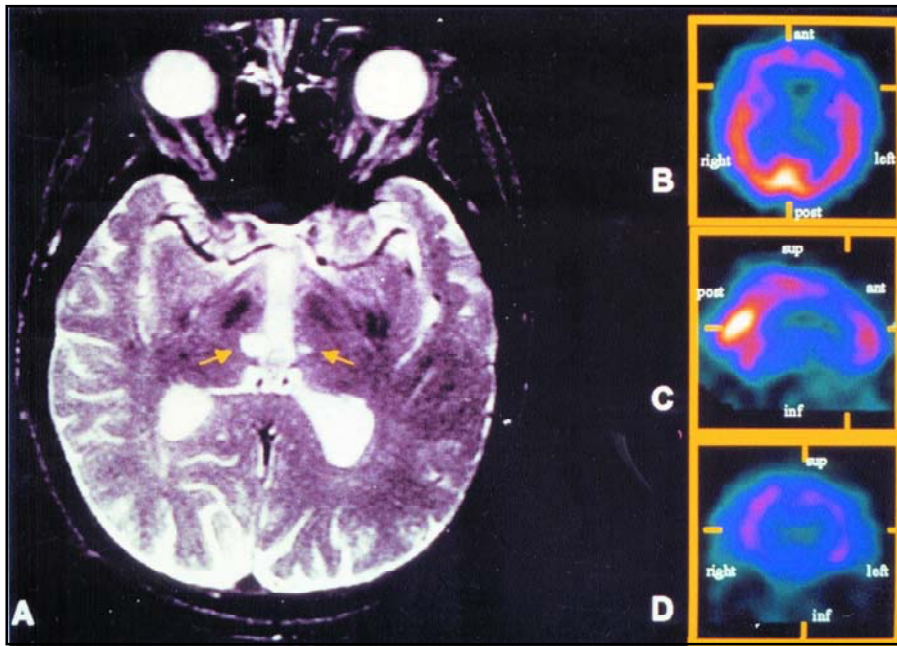


Fig 1. A) MRI with T2 weighted images, with arrows pointing to the infarcted areas corresponding to the thalamic paramedian territory; B), C) and D) SPECT showing bilateral frontal hypoperfusion (transverse, sagittal and coronal sections, respectively).

## CASE

A 63-year-old right handed man presented with a sudden loss of consciousness. His previous history was remarkable for arterial hypertension, myocardial infarction and the placement of a coronary artery bypass graft. His behavior was unremarkable, he was circumspect. Upon examination in the same day he was drowsy and a mild left hemiparesis was noted. Magnetic resonance imaging (MRI) revealed infarction in the territory of paramedian thalamic artery (Fig 1A). In the following weeks he recovered slowly and a behavior disorder became evident, characterized by hypersexuality, disinhibition and anterograde amnesia. He requested intercourse with his wife many times a day and started propositioning other women thus embarrassing his family. No change in sexual preference was present. Abnormal irritability and hypersomnia were also noted. A neuropsychologic testing revealed a moderate memory loss, mainly for recent events (Wechsler memory scale was 52 - delay recall 52, visual 73, verbal 61)<sup>4</sup>. There were no disturbances in language, executive functions, motor programming or constructional abilities (normal Wisconsin Card Sorting Test, Benton Test, Benton and Hamsher Multilingual Aphasia Examination and Raven's Coloured Progressive Matrices)<sup>4</sup>. A single-photon emission computed tomography (SPECT) showed bilateral frontal hypoperfusion (Fig 1B, C and D). The hypersexuality did not respond to neuroleptic drugs or carbamazepine. We have never tried testosterone inhibitors because of the risk of thrombosis. The patient behavior remained unmodified for the following 9 years, but his memory improved.

## DISCUSSION

Herein we present a case of a bilateral thalamic infarction. The corresponding ischemic area comprises the paramedian territory (retromamillary or thal-

amo-perforate pedicle)<sup>5</sup>. This area usually includes the paramedian part of upper midbrain and thalamus, including most of the dorsomedial nucleus, intralaminar nuclear group, mamillothalamic tract and the superior part of red nucleus<sup>6</sup>. In some cases one single unilateral pedicle on one side may supply the paramedian territory on both sides and the corresponding infarct may be bilateral, as observed in our case. The typical form of presentations a sudden loss of consciousness, confusion, vertical gaze disorders, sometimes associated to blepharospasm, hemiparesis and hemiataxia. Variable neuropsychological disturbances can be found such as memory impairment, dysphasia, hemineglect, visuospatial processing disturbances and behavioral disorders<sup>5-7</sup>. The latter consists of a mixture of aggressiveness with apathy, but disinhibition and mania may also be found<sup>8,9</sup>. Utilization behavior, usually seen in frontal lobe lesions, was also described<sup>10</sup>. Gentilini et al.<sup>9</sup> reported eight cases of paramedian infarction. Among them, four patients presented with bulimia. One of them showed a behavioral disturbance described as inappropriate, alternating "periods of fretfulness with periods of silly cheerfulness, when she ostentatiously kissed and hugged her husband in the presence of other people and indulged in coarse and inappropriate jokes about the physicians". This finding could be interpreted as a sign of hypersexual behavior. Nevertheless, to the present date, no case of hypersexuality as a prominent feature has been reported.

The frontal hypoperfusion observed in our case

is similar to several cases of paramedian infarcts described<sup>8,11,12</sup>, and support the frontal dysfunction forward by thalamic lesion. Moreover, some of the neuropsychological disturbances were similar to those present in frontal syndromes. These findings contributed to support current theories about the functioning of frontal-subcortical circuits<sup>3,13-17</sup>. It raises the question about whether the hypersexuality in our patient could or not correspond to a dysfunction in one of these circuits.

Cummings<sup>3</sup> proposed that frontal-subcortical circuits are implicated in mediating behavioral alterations when (1) lesions in several circuit-related structures produce a similar behavioral disorder, (2) the behavioral syndrome is not commonly seen with lesions in other brain regions and (3) simultaneous lesions in several circuit structures produce analogous rather than additive effects<sup>18</sup>. Indeed, improper sexual remarks or gestures and other antisocial acts may be part of an orbitofrontal syndrome, although overt sexual aggression is rare<sup>13-15</sup>. Cases of hypersexuality have been described as a consequence of medial basal frontal structures<sup>1</sup>. However, hypersexuality has been found in other than frontal lesions<sup>1</sup>, mainly in the temporal lobe<sup>16</sup> and other limbic structures such as the hypothalamus. It is well known that the medial dorsal nucleus has large connections with limbic structures parallel to frontal connections. The thalamus is poised at the interface of the medial-temporal circuit and frontal-subcortical circuits, causing difficulties in establishing which of the circuits is affected in a given thalamic lesion<sup>14</sup>. The frontal hypoperfusion present in our patient could be only an unrelated finding, not necessarily indicating association between frontal-subcortical dysfunction and hypersexuality.

However, some evidences in the literature lead us to conclude that, in our case, frontal-subcortical circuits are more likely to be implicated in the pathogenesis of hypersexuality: (1) PET studies<sup>1</sup> have demonstrated that medial thalamic infarctions show occipital and frontal hypometabolism (as in this case) while temporal hypometabolism is seen in posterior thalamic infarcts (absent here). (2) There are descriptions of sexual disturbances in patients with Parkinson's disease who start receiving levodopa therapy<sup>1,17</sup> suggesting a participation of striatal connections in these symptoms. Other diseases related to cortical-subcortical dysfunction may also show hypersexual behavior such as Huntington's disease and Gilles de La Tourette syndrome<sup>1</sup>. A case of capsulo-lenticular hematoma with hypersexuality was also described<sup>18</sup>.

(3) Limbic structures have been described associated to alterations of sexual preferences (homosexuality, pedophilia, fetishism) rather than to the true hypersexuality found in frontal lesions<sup>1,19,20</sup>. All these facts, in addition to the evidences present in our case suggests that the thalamus plays an important role in the control of human sexual behavior mediated by frontal-subcortical circuits.

**Acknowledgement** – We thank C. Buchpiguel, MD, A. Osawa, MD and C.R. Ono, MD for the help in processing the images.

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