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Orbitofrontal syndrome after a cerebral aneurysm operation. A case report

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Introduction. Orbitofrontal syndrome is an organic mental disorder characterized by cognitive, affective and behavioral alterations, triggered by the suppression of the inhibiting cerebral mechanisms.

Clinical case. A 19 year-old woman developed a «change of personality» in the context of an aneurysm operation in the middle cerebral artery, with serious behavioral alterations during one year. After the drug treatment was established, there was significant reduction in the intensity of the psychopathology. Furthermore, the utility of psychotherapy in the improvement of the familiar management was affirmed.

Conclusions. Orbitofrontal syndrome implies damage to any of the structures and connections included in the orbitofrontal system. The psychopathological overlapping with some psychiatric disorders opens the possibility of greater understanding of the biological bases of the mental disease. Because of the deficit in the metacognitive capacity and high dependency on environmental stimuli, it seems that behavioral techniques such as psychotherapy would be the treatments of choice for this type of patient.

Key words:

Orbitofrontal syndrome. Change of personality. Hypersexuality. Cerebral aneurysm. Middle cerebral artery.

son desencadenadas por la supresión de los mecanismos cerebrales inhibitorios.

Caso clínico. Se presenta el caso de una mujer de 19 años que desarrolló un cambio de personalidad en el contexto de una operación por un aneurisma en la arteria cerebral media, con graves alteraciones conductuales de 1 año de evolución. Tras la instauración del tratamiento farmacológico se apreció una reducción significativa en la intensidad de la psicopatología; asimismo se constató la utilidad de la psicoterapia en la optimización del manejo familiar.

Conclusiones. El síndrome orbitofrontal implica un daño en cualquiera de las estructuras y conexiones que comprenden el sistema orbitofrontal. El solapamiento psicopatológico con algunos trastornos psiquiátricos abre la posibilidad de un mayor entendimiento de las bases biológicas en la patología mental. El déficit en la capacidad metacognitiva y la alta dependencia de los estímulos ambientales sitúan las técnicas conductuales como la psicoterapia de elección en esta clase de pacientes.

Palabras clave:

Síndrome orbitofrontal. Cambio de personalidad. Hipersexualidad. Aneurisma cerebral. Arteria cerebral media.

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Síndrome orbitofrontal tras una operación por aneurisma cerebral. A propósito de un caso

Introducción. El síndrome orbitofrontal es un trastorno mental orgánico caracterizado por una serie de alteraciones de índole cognitivo, afectivo y conductual que

INTRODUCTION

The orbitofrontal syndrome is a neuropsychiatric disorder originated by a deficit or impairment of the orbitofrontal system. It causes dysfunction in the self-regulatory capacity, preventing the suppression or modulation of impulses and emotion. The most characteristic symptoms are impulsivity, emotional lability, childishness, distractibility and anosognosia.

CLINICAL CASE

The case of a 19 year-old woman, who was an only child, born in Valencia, who lives with her mother and maternal grandparents, is presented. She is a trainee as a hairdresser. She

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was referred to the Addictive Behavior Unit (ABU) of Torrent from primary health care due to «Internet addiction» problem.

REASON FOR VISIT

According to that reported by the family, for almost one year, the patient has been going out with many boys she has met through chats to maintain «sexual relationships», sometimes for money. They refused to believe it when they found condoms in her room and when they saw her dressed in extremely «provocative» clothes when she went out. They manifested their fear that she could be moving in marginal settings, with the consequent potential risk for her physical integrity (e. g., violation or HIV) and drug abuse. In general, they observed a significant change regarding her previous behavior («she was more timid and unsure before»).

In regards to what the patient stated, she indicated that since approximately one year ago, she has been feeling «more active, mouthy, conceited and bossy», with a parallel increase in sexual desire, performance and satisfaction. She stated that she had sexual relationships with 15-20 new persons per week, «men and sometimes women», and that she performed acts in trios that were filmed.

PERSONAL BACKGROUND

The patient was born by normal delivery and had a post-delivery period without incidences. Her psychomotor and linguistic development was normal. No known adverse drug reaction or allergies. Regarding temperament, she was an «alert and happy» child. Since she was three years old and after the separation of her parents, she has lived together with her mother and maternal grandparents. Her childhood was not happy as her mother partially washed her hands of her. She generally cut up her mother's clothes «with scissors» when she observed that she was going to go out. Regarding her father, he distanced himself from the beginning from the family nucleus «and worked as a traveling salesman throughout the province». At the onset of her adolescence, the conflictivity between the patient and her mother increased when her mother presented her new partner to her daughter, even with physical aggression. During that period, she changed schools, and had a very difficult time adapting. At 14 years of age, she began to take cannabis and began with nighttime activities. At 15 years, she met a boy with whom she maintained a stable relationship until she was 18. She initially experienced the breakup with «anxiety» but returned to her social contacts later. She generally «cut school» and her academic performance was deficient, so that she dropped out of school, and took a course as a beautician. On the weekends, she went to discotheques, consuming cannabis and sometimes cocaine and ecstasy. Since then, she has smoked one pack of cigarettes a day. She had unsatisfactory sporadic sexual meetings

BACKGROUND

Familial

Organic

- Maternal grandmother: myocardial infarction and angina pectoris.
- Mother: lumbar and cervical herniations (invalidity benefits).

Psychiatric

- Maternal grandmother: dysthymia treated with drugs in primary health care.
- Mother: post-traumatic stress disorder (traffic accident). Treated with drugs in mental health center in Torrent.
- Father: cocaine dependence. Without treatment

Personal

Organic

- Mild congenital beta thalassemia and thalassemia hemoglobinopathy
- Facial traumatism (16 years) with sphenoid fissure (some transient facial ones).
- Aneurysm of 8×6 in middle cerebral artery (diagnosed at 17 years) (fig. 1). She underwent right pterional craniotomy in the area hospital at 18 years of age, suffering contralateral facial hemiparesis, that abated at two weeks after the operation. The family and pa-

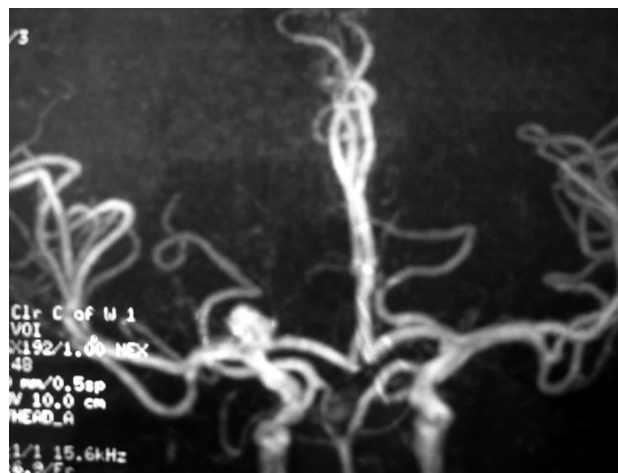


Figure 1 | Magnetic angioresonance that shows aneurysm in the middle cerebral artery (July 2005).

tient reported that the behavioral change began «after the operation».

- Genital herpes (19 years.)

CLINICAL EXAMINATION

During the interview, she was aware and alert. Careful hygiene and dress. Auto and allopsychically spatial-temporal oriented. Immediate, recent and remote memory conserved. Significant distractibility and hypermetamorphosis. Speech was fluid and spontaneous, coherent and structured. She quickly went from one subject to another, this being poor in content, was witty and acted childish, with a sexual nature and megalomaniac («I am the most beautiful»). There was no evidence of delusional ideation or sensory-perceptive disorders. On the affective level, labile and irritable mood stood out, alternating negativism with over-friendliness. At other times, her affect was inadequate and humorous. She had psychomotor restlessness, frequently getting up to walk. She manifested disinhibition, hypererosia and impulsiveness. She also reported hyporexia and inverted sleep-awake cycle. She had insight into the behavioral change and partial awareness of the consequences of her behavior. She did not attribute any pathological quality to her symptoms («I like what I do»).

COMPLEMENTARY TESTS

- Complete blood count: Hb: 11.6 g/dl; MCV: 69.3 fl; MCH: 22.6 pg; RDW: 16.6 %.
- Biochemistry: normal.
- HIV serology, hepatitis B and C: negative.
- Urine samples: positive for cocaine, heroin and cannabis.
- EEG: tracing within normality.
- Brain CT scan: porencephalia area in left caudate nucleus adjacent to metallic clip that generates retraction of frontal loop of homolateral ventricle (fig. 2)
- Neuropsychological assessment: mini mental state examination: 35; WAIS: digital symbol subtest: 10/30.

DIAGNOSIS

«Disinhibited-labile» type personality change due to injury to orbitofrontal system» F 07.0

COURSE AND TREATMENT

Because it was difficult for the family to manage her, she was admitted for two weeks to the Psychiatric Hospitalization Unit of the area. She then continued psychotherapy in the Addictive Behavior Unit and her drug treatment was supervised in the Mental Health Clinic of Torrent. The family



Figure 2 Brain CT scan that shows a metallic clip introduced during the operation in the adjacent porencephalia area (April 2007).

decided to initiate the steps to have her referred to a Brain Damage Unit at that time.

In regards to the drug treatment, topamax 50: 1-1-1, and zyprexa 5: 0-0-1, were prescribed initially to her. As the picture persisted, topamax 100: 1-1-1, and zyprexa 10: 1-0-1 were prescribed, there being a noticeable reduction of the impulsiveness-disinhibition, emotional lability and psychomotor restlessness at two months of treatment. Her childish behavior, humorous affect and distractibility underwent less promising decrease.

In regards to the psychological intervention, an attempt was made to decrease her disadaptive behaviors. The cognitive-behavioral techniques used were: psychoeducation, stimulus control (internet and cell phone), contingency contract, structuring of daily life activity (mean and sleep schedules) and masked sensitivity. With this, it was possible to achieve a significant improvement of the social-familial dynamics. She was partially toxic abstinent (cannabis).

CONCLUSIONS

The orbitofrontal syndrome is an organic mental disorder that can involve another type of injury or defect, not only in the orbitofrontal cortex but also in its varied projections towards the subcortical nuclei¹, especially the basal ganglia (caudate nucleus, putamen, globus pallidus) and limbic system (amygdala, hypothalamus, parts of the thalamus, septum, entorhinal cortex).

The psychopathological characteristics of the syndrome reflect a deficit action in the inhibitory and modulating brain centers², this disabling the biological «base» to develop empathy and reflexivity, milestones characteristic of phylogenetic and ontogenetic development of the human being. This causes severe deficits in socialization capacity³ and self-monitoring, with a tendency to childish, impulsive, erratic and egocentric behaviors^{4,5}. Regarding psychotherapy, due to her low metacognitive capacity and the high environmental stimulus dependence, use of the behavioral techniques over the cognitive ones is advisable.

In regards to its etiology, most of the personality changes due to injury in the orbitofrontal system are caused by severe cranioccephalic traumas⁶. Similarly, there is evidence of orbitofrontal syndromes caused by ruptured aneurysm of anterior communicating orbitofrontal artery. There is no evidence of its incidence after an operation due to middle cerebral artery aneurysm, in which only deterioration in the memory and attention has been observed⁷. This patient would be the first case in this context.

Regarding the differential diagnosis, different organic conditions have symptoms in common with orbitofrontal system injury-induced personality change (e.g., Kluver-Bucy syndrome, Klein Levine syndrome, frontotemporal dementia, etc.), and should be taken into account. On the other hand, some authors state that the borderline between organic and idiopathic mental disorders is more diffuse than the limit mediated by the lexicon used. With the advance of the modern neuroimaging techniques (e.g., PET), the possibility that there is a vulnerability common to orbitofrontal syndrome and some psychopathologically similar idiopathic mental disorders is suggested. In support of this thesis, reduction in the orbitofrontal cortex volume has sometimes been found in schizophrenia^{8,9}. In the same way, some studies have indicated that there is a decrease in cerebral flow in the orbitofrontal cortex in mania^{10,11}. On the contrary, hypoactivation of the orbitofrontal cortex is postulated in borderline and antisocial personality disorders^{12,13}. As a whole, these findings open up new pathways to understand the possible biological correlates of the mental condition, and should be considered in future investigations.

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