Restricted Left Gyrus Rectus Hemorrhage with Unusual Frontal Syndrome Presentation

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Dear Sir,

The study of localized and well-circumscribed brain lesions has contributed greatly to our understanding of the function of single brain areas. The gyrus rectus (GR) is part of Brodmann's area (BA) 11, which together with BA 10 and BA 47 form the orbito-frontal cortex. Lesions of this region are classically associated with disinhibition syndromes even if some reports described very few or no behavioral symptoms after such lesions [1–3]. Here we report the case of a 59-year-old woman who suffered a subarachnoid hemorrhage associated with a restricted lesion of her left GR, with an unusual frontal syndrome presentation.

Case Report

A 59-year-old right-handed woman with a history of fibromyalgia, intermittent asthma and bilateral glaucoma, who was undergoing substitutive post-menopausal therapy, suffered an explosive, unusual and paroxysmal headache while taking a shower. Although these symptoms persisted, albeit with lower intensity, she did not seek medical advice until 4 days later when she suffered syncope while coming down a flight of stairs. She was transferred to the emergency room where the clinical examination showed apathy and meningism, in an otherwise oriented

and collaborating patient with a Glasgow Coma Scale of 14/15 and no neurological deficit or lateralizing signs. The brain CT scan showed a left inferior frontal subarachnoid hemorrhage secondary to an aneurysm rupture from the distal left carotid artery bifurcation (M1; grade: Fischer IV, HH3, WFNS2), spread to both sylvian fissures and in the perimesencephalic cistern, which was associated with a left GR parenchymatous hemorrhage. She was treated by cerebral arteriography with embolization of the aneurysm by placement of 3 Guglielmi detachable coils, with an excellent result. There were no complications following this procedure.

A bedside neuropsychological assessment performed 6 days after symptom onset showed a preserved orientation to time, place, and person, fluent and informative spontaneous oral expression, very slight word-finding difficulties (Boston Naming Test 15/34) and impairment of verbal category-specific and literal fluency, maximum name of animals (5) and a word beginning with M (3) in 1 min, but preserved comprehension (complex command 4 out of 4 correctly executed). There were also slight verbal memory deficits, and a major behavioral slowdown with lack of initiative in action and in language that impacted on other cognitive functions as oral calculation (prolonged thinking time, loss of her train of thought in the course of calcu-

lation), constructive praxis and executive functions (frise de Luria, gestural sequences). Finally, a circumstantial and post-traumatic (10-min) amnesia was noted.

She was then transferred to the neurorehabilitation unit, where the clinical examination was unchanged. She was independent for the activities of daily living but suffered a persistent psychomotor slowing down, associated with executive (Trail Making Test part A: 133 s, part B: 265 s, 5 alternating errors), memory, and attentional dysfunction. Furthermore, she presented with elements of an athymhormic syndrome as described by Guiraud [4]: loss of interest for anterior preoccupations (anhormia), loss of subjective feeling when dealing with a pleasant or unpleasant situation (athymia) and loss of spontaneity. Progress was favorable, but at the time of discharge we noted the persistence of elements of this athymhormic syndrome.

During outpatient follow-up visits, we noted a few improvements of these symptoms, along with dulling of affection, and short-sentence answers to our questions. According to her husband, she also had significant difficulty starting the day. On the behavioral side, there was emotional dulling, severe apathy (Lille Apathy Rating Scale [5]: patient: 12/36, husband: 4/36), as well as a significant loss of interest and pleasure without suffering or sadness. The

Fig. 1. Sagittal (**a** left hemisphere, **b** right hemisphere), horizontal (**c**), and coronal (**d**) T₁-weighted MRI sections through the GR. Note the hypointensity is almost entirely restricted to the left GR.

patient was aware of her general lack of interest and indifference towards the outside world or even towards her close relatives; for example, she realized she was not affected by her daughter or husband telling her about their problems. She also described having been witness to a suicide without being affected by this event when her daughter, who was also present, was horrified. She criticized her behavior and was able to analyze it as inappropriate. In this context, the symptoms seemed closer to an athymhormic syndrome rather than that of depression. A psychiatric evaluation ruled out a diagnosis of depression or schizophrenia. A control MRI 3 months after symptom onset showed hemorrhagic sequelae of the left GR with a discrete extension to the orbital gyrus (fig. 1). The symptoms only partially responded to a selective serotonin reuptake inhibitor (SSRI) (citalopram) progressively increased to a dose of 60 mg/day. With this treatment, the patient could wake up earlier and manage to go outside for shopping without help.

Discussion

The hemorrhagic sequelae, almost entirely restricted to the left GR, suggest a precise function associated with this brain region. This region has mainly been associated with an inhibition function as a part of the orbito-frontal-frontosubcortical circuit, as described by Cummings and co-workers [6–8].

Interestingly, the patient was not at all disinhibited, but, on the contrary, she presented elements of an athymhormic syndrome that only responded partially to the SSRI treatment. To our knowledge, this syndrome has never been described in a small, restricted, unilateral GR lesion. In fact, this syndrome has been described in bilateral basal ganglia or cingulate gyrus lesions.

On the one hand, as is often seen in other cognitive dysfunctions (aphasia, apraxia, agnosia), the uncommon clinical radiological presentation may be due to an unusual brain circuitry for this function in this patient. Interestingly, frontal gray

matter changes have been described in fibromyalgia [9]. On the other hand, as very few reports of circumscribed lesions of the GR [10, 11] exist, the functional significance of this region might be underevaluated. For example, a case of unilateral GR cystic changes presented with Tourette syndrome [10]. However, a series of patients operated for an anterior communicating artery aneurysm, with possible iatrogenic lesions of the GR, showed very few, if any, behavioral disturbances [1, 2].

Large stroke databases allowing collection of similar cases of restricted lesions are still necessary to improve our understanding of localized brain functions.

Disclosure Statement

The authors have no financial disclosures to report.

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