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Case report

Involuntary masturbation and hemiballismus after bilateral anterior cerebral artery infarction

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Abstract

Ischemia of the areas supplied by the anterior cerebral artery is relatively uncommon. In addition, combined hemiballismus and masturbation have rarely been reported in patients with cerebrovascular disease. We describe herein a 62-year-old right-handed man simultaneously exhibiting right side hemiballismus and involuntary masturbation with the left hand after bilateral infarction of the anterior cerebral artery territory. Right side hemiballismus was related to the disruption of afferent fibers from the left frontal lobe to the left subthalamic nucleus. Involuntary masturbation using the left hand was exclusively linked to a callosal type of alien hand syndrome secondary to infarction of the right side of the anterior corpus callosum. After 2 weeks, these abnormal behaviours were completely extinguished. This report stresses the wide diversity of clinical manifestations observed after infarction of the anterior cerebral artery territory. © 2007 Elsevier B.V. All rights reserved.

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1. Introduction

Infarction of the areas supplied by the anterior cerebral artery is relatively uncommon, accounting for approximately 4% of all ischemic strokes [1,2]. Clinical features vary according to the exact site of the occluded branches and the spread of the ischemic lesion, but the most frequent signs reported are contralateral crural monoplegia or hemiplegia with predominant lower extremity involvement, forced grasping or groping of the ipsilateral upper limb, ideomotor apraxia of the left arm, and dynamic aphasia, regardless of the side of the anterior cerebral artery occlusion [3].

Hemiballismus and masturbation have rarely been observed in patients with cerebrovascular disease. We report herein the case of a patient with bilateral lesions in the vascular territory of the anterior cerebral arteries who exhibited these two very rare symptoms simultaneously.

2. Case report

A 62-year-old right-handed man was admitted to our stroke unit because he had experienced difficulties in moving his left leg at night when he was trying to stand up. He had controlled hypertension, was a current smoker, and did not have a history of cerebrovascular accidents or alcohol abuse. He was married and had two children. He was a retired skilled worker. When he was examined 5 h after the onset of the symptoms, he was oriented. He had paresis of the left lower extremity, predominant in the proximal segment with less strength than antigravity, in contrast with only a slight deficit in the left upper extremity. Strength was normal on the right side. Deep tendon reflexes were normal, but plantar responses were extensor in both feet. Examination of sensory and cranial nerves did not reveal any abnormalities. He had

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neither cerebellar ataxia nor dysmetria, but he presented with intermittent abnormal movements of his right side. They were vigorous, rapidly executed, asynergic, complex and combined, and affected predominantly the proximal segments of the right upper and lower extremities. The face and neck were not involved. Right hemiballismus was evoked. In addition, his contralateral hand presented anarchic involuntary movements, leading to difficulties in bimanual tasks, such as eating or manipulating objects. Moreover, he exhibited repeated exposure of his genitals and masturbation with his left hand and affirmed that this behaviour was not under his control. He was very self-critical with regard to this activity, and according to his family, he had never experienced such behaviour in the past. Further examination revealed bilateral forced grasping and the smooth and complex courses of skilled movement were disturbed as were kinetic melodies. Neuropsychologic assessment revealed marked reduction in spontaneous speech with normal comprehension and repetition. There were difficulties in initiating talking, but oral denomination, writing, and reading were normal. The intelligence quotient, as evaluated by a seven-subtest short form of the Weschler Adult Intelligence Scale-Revised (WAIS-R) [4], was in the average range. The Mini-Mental State Examination [5] score was 28/30. Anterograde and retrograde memory skills were normal. The Frontal Assessment Battery (FAB) [6] total score was 12/18; he had difficulties in motor programming and executive control of action (score: 2/3), resistance to interference (score: 2/3), inhibitory control (score: 1/3), and environmental autonomy (score: 1/3), whereas conceptualization, abstract reasoning, and mental flexibility were normal. Left ideomotor dyspraxia was noted, whereas left agraphia, left visual, tactile anomia, and constructive apraxia were not observed. Both social cognition and the patient's judgment assessment did not reveal any impairment.

Magnetic resonance imaging was then performed and showed bilateral hyperintense signals in diffusion-weighted images in the anterior cerebral artery territories involving the right anterior corpus callosum, suggesting a hemodynamic mechanism (Fig. 1). Trans-oesophageal echocardiography was performed, but no cardio-embolic sources were detected. No rhythm disturbances were apparent either on the repeat EKG or on the Holter recording. Carotid and vertebral ultrasonography showed diffuse atheromas with no significant arterial stenosis. Blood tests were normal. The patient was admitted to the rehabilitation center after 2 weeks and his abnormal movements and masturbation did not persist.

3. Discussion

The most interesting point of our observation is the association of two symptoms that are rarely described in ischemia of the anterior cerebral artery: (1) hemiballismus and (2) involuntary masturbation.

Hemiballismus is a rarely reported hyperkinetic disorder secondary to cerebrovascular pathologies with an estimated annual incidence in the general population of 0.45/100,000 [7], accounting for 0.4% of all strokes [8]. The vascular lesion usually involves the contralateral subthalamic nucleus [9]. In the patient described herein, there were no lesions in this cerebral area on diffusion-weighted MRI. Nevertheless, hemiballismus has also been reported after lesions involving the frontal and parietal lobes, the thalamus, or the corona radiata [8,10,11]. From a pathophysiologic point of view, the disruption of the balance between the direct (cortex-caudate-internal pallidum) and the indirect (cortex-caudate-external pallidum-subthalamic nucleus-internal pallidum) pathways modulating the glutamatergic activator thalamocortical retroactive pathway by such lesions could be responsible for the induction of hemiballismus, explaining these clinical features in our patient [12].

The onset of involuntary masturbation following a cerebrovascular lesion is not common, whereas it is often observed in patients with temporal lobe epilepsy [13]. Hypersexuality has been reported after bilateral thalamic infarction associated with frontal hypoperfusion in SPECT imaging, suggesting dysfunction of frontal–subcortical circuits [14]. Moreover, cases of hypersexuality have been described as

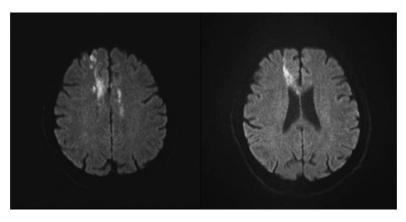


Fig. 1. Diffusion-weighted magnetic resonance imaging showing hyperintense signals over the right anterior corpus callosum and the two frontal lobes related to bilateral anterior cerebral artery infarction.

part of an orbitofrontal syndrome [15]. Nevertheless, whereas our patient exhibited symptoms, such as a disturbance of skilled movements, forced grasping, a marked reduction in spontaneous speech, or apathy, and had a pathologic FAB score, we could not attribute the occurrence of masturbation to a loss of control of social behaviour in the case of a frontal syndrome. Indeed, this right-handed patient always used his left hand to masturbate, and this act appeared to be independent of the patient's will. Moreover, he was selfcritical with regard to this involuntary compulsive behaviour and felt alienation, suggesting that it could be related to a motor alien hand syndrome. Two types of motor alien hand syndrome have been distinguished by Della Sala et al. [16]. The first and most frequent affects the dominant hand, the so-called "frontal alien hand syndrome," and is characterized by reflexive grasping, groping, and compulsive manipulation of tools, and results from damage to the supplementary motor area, the anterior cingulate gyrus, the medial prefrontal cortex, and the anterior corpus callosum of the dominant hemisphere. The second type, or "callosal type," is described in patients suffering from lesions involving the corpus callosum with or without associated frontal lesions of the non-dominant hemisphere, especially in the supplementary motor area. Clinically, intermanual conflict and apraxia of the non-dominant limb are observed. A unique case of involuntary masturbation associated with intermanual conflict and apraxia secondary to ischemia of the right medial frontal lobe and anterior corpus callosum on imaging similar to that observed in our patient has been reported [17]. Like that noted in our patient, this activity was limited to the left hand and consequently, it can be regarded as a manifestation of a callosal type of alien hand syndrome. Classically, other signs of callosal disconnection, such as left ideomotor apraxia, left agraphia, left visual, or tactile anomia can be found. We did not observe any such manifestations except for left ideomotor apraxia.

Infarction of the anterior cerebral artery territory is unusual, accounting for only 1.3–4% of all strokes [1,2,18] and bilateral infarction in this territory is very rare, affecting only 4% of all anterior cerebral artery infarcts [18]. Very few cases of bilateral infarction have been reported in the literature and neuropsychological evaluation has not been as extensive as the description of neurological signs. Hence, the classic picture including akinetic mutism, paraplegia, incontinence, bilateral grasp reflex, and amnesia with apathy [18,19]. Boccardi et al. reported the case of a patient suffering from bilateral infarction that damaged the supplementary motor areas and who presented severe utilization behaviour [20]. They considered this pathologic behaviour as a double anarchic hand due to damaged balance between the premotor cortices, responsive to environmental triggers, and the supplementary motor areas, which modulate actions and inhibit them. Some authors suggested that bilateral frontal lesions may cause an imbalance between impaired frontal and intact parietal lobes, and released activity of parietal lobes may yield to utilization behaviour and environmental-dependency

syndrome [21]. Bilateral lesions of the supplementary motor areas have also been associated with gait apraxia, suggesting that these regions are required for the complexity of the mostly automatic gait movements [22]. In a unique detailed neuropsychological observation, Bird et al. observed a dysexecutive syndrome, characterized by a severe planning and prospective memory impairment associated with confabulatory and amnestic syndrome, following bilateral medial frontal lobe infarction, but they failed to demonstrate that this region plays a critical role in the "Theory of Mind" network in the human brain [23]. Neither involuntary masturbation nor hemiballismus have been reported after bilateral anterior cerebral artery infarcts.

4. Conclusion

To our knowledge, our patient is the first case of bilateral lesions involving the anterior cerebral artery territory presenting with two rare symptoms simultaneously. The right side hemiballismus was related to disruption of afferent fibers of the left subthalamic nucleus from the left frontal lobe, whereas the involuntary masturbation with the left hand was linked to a callosal type of alien hand syndrome secondary to infarction of the right side of the anterior corpus callosum.

Conflict of interest

None.

References

- Bogousslavsky J, Van Melle G, Regli F. Stroke registry: analysis of 1000 consecutive patients with first stroke. Stroke 1988;19:1083–92.
- [2] Giroud M, Dumas R. Clinical and topographical range of callosal infarction: a clinical and radiological correlation study. J Neurol Neurosurg Psychiatry 1995;59:238–42.
- [3] Critchley M. The anterior cerebral artery and its syndromes. Brain 1930;53:120–65.
- [4] Weschler D. Weschler Adult Intelligence Scaled-Revised. New York: Psychological Corporation; 1981.
- [5] Folstein MF, Folstein SE, McHugh PR. "Mini-mental state". A practical method for grading the cognitive state of patients for the clinician. J Psychiatr Res 1975;12:189–98.
- [6] Dubois B, Slachevsky A, Litvan I, Pillon B. The FAB: a Frontal Assessment Battery at bedside. Neurology 2000;55:1621–6.
- [7] Pekmezović T, Svetel M, Ristić A, Raicević R, Ivanović N, Smiljković T, Kostić VS. Incidence of vascular hemiballism in the population of Belgrade. Mov Disord 2004;19:1469–72.
- [8] Ghika-Schmid F, Ghika J, Regli F, Bogousslavsky J. Hyperkinetic movement disorders during and after acute stroke: the Lausanne Stroke Registry. J Neurol Sci 1997;146:109–16.
- [9] Shannon KM. Hemiballismus. Clin Neuropharmacol 1990;13:413–25.
- [10] Martin JP. Hemichorea (hemiballismus) without lesions in the corpus Luys. Brain 1957;80:1–12.
- [11] Mitzushima N, Park-Matsumoto YC, Amakawa T, Hayashi H. A case of hemichorea–hemiballism associated with parietal lobe infarction. Eur Neurol 1997;37:65–6.

- [12] Rossetti AO, Ghika JA, Vingerhoets F, Novy J, Bogousslavsky J. Neurogenic pain and abnormal movements contralateral to an anterior parietal artery stroke. Arch Neurol 2003;60:1004–6.
- [13] Jacom DE, Risko MS. Absence status manifested by compulsive masturbation. Arch Neurol 1983;40:523–4.
- [14] Mutarelli EG, Omuro AM, Adoni T. Hypersexuality following bilateral thalamic infarction: case report. Arq Neuropsiquiatr 2006;64:146–8.
- [15] Monga TN, Monga M, Raina MS, Hardjasudarma M. Hypersexuality in stroke. Am J Phys Med Rehabil 1986;67:415–7.
- [16] Della Sala S, Marchetti C, Spinner H. Right-sided anarchic (alien) hand: a longitudinal study. Neuropsychologia 1991;29:1113–27.
- [17] Ong Hai BG, Odderson IR. Involuntary masturbation as a manifestation of stroke-related alien hand syndrome. Am J Phys Med Rehabil 2000;79:395–8.
- [18] Kumral E, Bayulkem G, Evyapan D, Yunten N. Spectrum of anterior cerebral artery territory infarction: clinical and MRI findings. Eur J Neurol 2002;9:615–24.

- [19] Bogousslavsky J, Regli F. Anterior cerebral artery territory infarction in the Lausanne Stroke Registry, clinical and etiologic patterns. Arch Neurol 1990;47:144–50.
- [20] Boccardi E, Della Sala S, Motto C, Spinnler H. Utilisation behaviour consequent to bilateral SMA softening. Cortex 2002;38:289– 308.
- [21] Lhermitte F, Pillon BI, Serdaru M. Human autonomy and the frontal lobes. Part I. Imitation and utilization behaviour: a neuropsychological study of 75 patients. Ann Neurol 1986;19:326– 34.
- [22] Della Sala S, Francescani A, Spinnler H. Gait apraxia after bilateral supplementary motor area lesion, 1. J Neurol Neurosurg Psychiatry 2002;72:77–85.
- [23] Bird CM, Castelli F, Malik O, Frith U, Husain M. The impact of extensive medial frontal lobe damage on 'Theory of Mind' and cognition. Brain 2004;12:914–28.