

Atypical clinical presentation in an anterior cerebral artery territory infarction - case report

Apresentação clínica atípica em infarto no território de artéria cerebral anterior – relato de caso

Leopoldo Antônio Pires¹, Marcelo Maroco Cruzeiro², Thiago Cardoso Vale³, Luciana de Souza Nogueira⁴, Gláucio Mendes Franco⁵, Sérgio Franca de Souza⁶

RESUMO

Infarto no território da artéria cerebral anterior (ACA) ocorre em apenas 0.3-4.4% dos infartos cerebrais. Relata-se um infarto progressivo, prolongado e incomum da ACA em paciente de 58 anos que teve seu diagnóstico baseado em investigação neurorradiológica e na exclusão anátomo-patológica de desordem neoplásica.

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Neurology and Neurosurgery, Department of Internal Medicine at University Hospital of Federal University of Juiz de Fora, Juiz de Fora-MG, Brazil.

1.Msc (Universidade Federal de São Paulo - UNIFESP), Neurologist, MD. Head and Lecturer of Neurology in Department of Internal Medicine at University Hospital of Federal University of Juiz de Fora, Juiz de Fora-MG, Brazil.

2.PhD. (Fluminense Federal University - UFF), Neurologist and Neurophysiologist, MD. Assistant Lecturer in Department of Internal Medicine at University Hospital of Federal University of Juiz de Fora, Juiz de Fora-MG, Brazil.

3.Medical resident in Neurology at the Clinics Hospital, Federal University of Minas Gerais (UFMG), Belo Horizonte-MG, Brazil.

4.Neurologist, MD. Former Medical Resident in Department of Internal Medicine at University Hospital of Federal University of Juiz de Fora, Juiz de For a-MG, Brazil.

5.Msc (Rio de Janeiro Federal University - UFRJ); PhD (Fluminense Federal University - UFF). Neurologist and Neurophysiologist, MD. Former Head and Senior Lecturer of Neurology in Department of Internal Medicine at University Hospital of Federal University of Juiz de Fora, Juiz de For a-MG, Brazil.

6.Neurosurgeon, MD. Lecturer of Neurosurgery at University Hospital of Federal University of Juiz de Fora, Juiz de For a-MG, Brazil.

SUMMARY

Infarction of the anterior cerebral artery (ACA) territory accounts for only 0.3-4.4% of cerebral infarctions. We report an unusually prolonged progressing stroke of the ACA in a 58-year-old patient who had his diagnosis based on neuroimaging investigation and in anatomopathological exclusion of neoplastic disorder.

Keywords. Cerebral Infarction, Anterior Cerebral Artery, Thrombosis.

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Correspondence address:

Leopoldo A Pires

R. Dr José P Teixeira, 395

CEP 36021-540, Juiz de Fora-MG, Brazil

Email: leopoldo@artnet.com.br

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INTRODUCTION

Infarction of the anterior cerebral artery (ACA) territory accounts for only 0.3–4.4% of cerebral infarctions reported^{1–4}. In Kumral et al's large stroke registry⁵, patients with ACA infarction represented 1.3% of 3705 patients with ischemic stroke. According to Bogousslavsky et al⁶, 63% of ACA territory infarctions result from cardiogenic embolism or artery-to-artery embolism. Gac et al⁷ reported other possible causes: contralateral or ipsilateral occlusions of the internal carotid artery (ICA), distal extensions of ICA thrombosis, and local thrombosis caused by vasculitis.

In recent years, single case reports and case series have focused on specific symptoms and syndromes resulting from the involvement of different territories of the frontal lobe, thus broadening our knowledge of the clinical manifestations of ACA infarctions⁵. In a prospective magnetic resonance imaging (MRI) study conducted by Kumral et al⁵, the first MRI-based study on the clinical findings of ACA infarction, the main risk factors for ACA infarctions were hypertension in 58% of patients, diabetes mellitus in 29%, hypercholesterolemia in 25%, cigarette smoking in 19%, atrial fibrillation in 19%, and myocardial infarction in 6%. According to their clinico-radiologic analysis, there were three main clinical patterns depending on the lesion's side: left-sided infarction consisting of mutism, transcortical motor aphasia, and hemiparesis with lower limb predominance; right-sided infarction accompanied by acute confusional state, motor hemineglect and hemiparesis; bilateral infarction presenting with akinetic mutism, severe sphincter dysfunction and progressing to functional dependence⁵.

We report an unusually prolonged progressing stroke of the ACA in a 58-year-old patient who had his diagnosis based on neuroimaging investigation and in anatomopathological exclusion of neoplastic disorder.

CASE REPORT

JMS, a 58-year-old right-handed man (HUSAME-297918) with hypertension, depression, lower limb chronic arterial insufficiency, peptic ulcer complicated with perforation, erosive oesophagitis, gastroesophageal reflux disease, and various episodes of pneumonia was brought to the Emergency Room with a history of progressively worsening weakness of the limbs, behavioural disturbance, labile affect and apathy.

The first episode of weakness occurred three years before and was exclusively due to right lower limb paresis. A second similar episode occurred three months before admission and led to an ascending paresis over the following months with progressive involvement of the left lower limb. Mild behavioural and cognitive disturbances as well as impairment of the upper limbs were noticed in the following weeks before actual hospitalization. This impairment was characterized by an ideomotor apraxia, which enabled the patient to perform in-phase alternating hand movements well, but he had moderate difficulties performing out-of-phase alternating hand movements, particularly if vision of the hands was occluded.

Brain MRI investigation of the second episode showed a left frontal hypointense lesion on T1-weighted images. The lesion was hyperintense with contrast enhancement on T2-weighted and on fluid-attenuated inversion recovery (FLAIR) images. The subsequent workup included carotid and vertebral artery Doppler ultrasonography, electrocardiography, and transthoracic and transesophageal echocardiography with bubble study, none of which showed any source of embolism. Laboratory tests included normal levels of antithrombin III activity, protein C and S antigen, plasma homocysteine, antiphospholipid antibody panel, anti-hepatitis C virus antibodies, antinuclear antibody, anticardiolipin antibody IgG, anticardiolipin antibody IgM, vitamin B12, lupus anticoagulant, Factor V Leiden, prothrombin gene mutation, activated protein C resistance and partial thromboplastin time, as well as a normal complete blood count and routine blood chemistry. Tests for HIV I + II antibodies were negative. There was no family history of haematologic or neurologic disease.

A new brain MRI investigation was performed three months after the second episode of weakness and it showed progression of the previous lesions with involvement of the parietal lobe (Figure 1). Cerebral arteriography revealed partial occlusion of the left ACA (Figure 2). Conservative treatment was proposed by the neurosurgery staff after in-loco biopsy ruled out a neoplastic disorder.

At the time of admission, his physical examination was normal, except for Raynaud's phenomenon in his right lower limb. Neurologic examination revealed upper motor neuron disease signs affecting predominantly the right limbs. Neuropsychological standard instruments revealed moderate

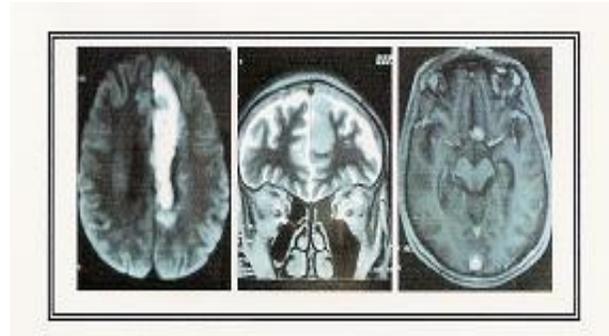


Figure 1. Brain MRI showing a left fronto-parietal lesion with progression over a two-year period.

cognitive impairment. The patient developed a dysexecutive syndrome characterized by impairments in planning and memory as well as a tendency to confabulate. Poor insights into his problems and an inability to adopt systematic strategies when attempting to solve problems were also observed.

Anticoagulant therapy was established and the patient remained cognitively impaired and with a right lower limb sequela. This case report was performed with the approval of the Ethics Committee of the University Hospital of the Federal University of Juiz de Fora (Minas Gerais), Brasil.

DISCUSSION

According to Ay et al⁴, clinical progression in patients with ischemic stroke may occur up to seven days after onset. It can be territorial, a gradual progression of symptoms attributable to the involvement of a single arterial territory, or non-territorial, a deterioration in consciousness or the appearance of new signs following the involvement of different vascular regions⁵. Territorial progression spanning a period of months is quite uncommon, particularly at the ACA territory.

The patient presented progressive right hemiparesis with restrictions of daily activities as well as bilateral lesions of frontal supplementary motor areas and adjacent medial frontal lobe structures such as the anterior cingulate gyrus. This case vignette is interesting because its clinical presentation is similar to tumoral lesion progression. Oddly enough, the patient did not develop urinary incontinence, despite having such a progressive and extensive territorial stroke.

Unusually prolonged infarction in the ACA territory, unrelated to subara-

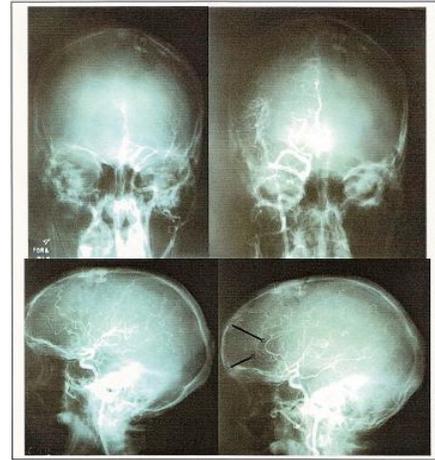


Figure 2: Cerebral conventional angiography yielding lack of perfusion in the left ACA territory (top and bottom left) and a normal perfusion in the right ACA territory (top and bottom right). The long and short arrows point out the pericallosal and cingulate sulcus arteries, respectively.

choid hemorrhage, surgery, or trauma, has been reported rarely. Ay et al⁴ have a very similar case of unusually prolonged progressing stroke, but without contralateral lobe impairment.

The current report shows that, in occasional patients, clinical worsening is not limited to a matter of hours or days, but may last weeks, which prompt a clinical suspicion of tumoral lesion. Reasons behind progressive strokes are not available in literature to date. Above and beyond all other consideration, timely recognition of patients under risk of territorial progression is essential. The use of markers of tissue injury and tissue perfusion might optimize future therapeutic efforts to halt progression in these patients.

CONCLUSION

This case vignette describes a patient whose symptoms gradually progressed over two years, highlighting this unusual manifestation of vascular thrombosis in the ACA territory. What makes it more intriguing is the absence of aneurysm, which would be responsible for the thrombosis. Medical treatment and regular follow-up were carried out with relative favorable outcome.

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REFERENCES

- 1.Orlandi G, Moretti P, Fioretti C, Puglioli M, Collavoli P, Murri L. Bilateral medial frontal infarction in a case of azygous anterior cerebral artery stenosis. *Ital J Neurol Sci* 1998;19:106-8.
- 2.Yamaguchi K, Uchino A, Sawada A, Takase Y, Kuroda Y, Kudo S. Bilateral Anterior Cerebral Artery Territory Infarction Associated with Unilateral Hypoplasia of the A1 Segment: Report of Two Cases. *Radiat Med* 2004;22:422-5.
- 3 .Klatka LA, Depper MH, Marini AM. Infarction in the territory of the anterior cerebral artery. *Neurology* 1999;52:1519.
- 4.Ay H, Uluc K, Arsava EM, Saatci I, Saribas O. Unusually prolonged progressing stroke: an expanding anterior cerebral artery infarction. *Cerebrovasc Dis* 2002;13:64-6.
- 5.Kumral E, Bayulkem G, Evyapan D, Yuntun N. Spectrum of anterior cerebral artery territory infarction: clinical and MRI findings. *Eur J Neurol* 2002;9:615-24.
- 6.Bogousslavsky J, Regli F. Anterior cerebral artery territory infarction in the Lausanne Stroke Registry. Clinical and etiologic patterns. *Arch Neurol* 1990;47:144-50.
- 7.Gacs G, Fox AJ, Barnett HJ, Vinuela F. Occurrence and mechanisms of occlusion of the anterior cerebral artery. *Stroke* 1983;14:952-9.