

## CASE REPORTS

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## Bilateral medial frontal infarction in a case of azygous anterior cerebral artery stenosis

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**Abstract** We describe the unusual case of a 63-year-old woman with a history of arterial hypertension who presented a sudden weakness of the lower limbs followed by mutism, akinesia and dyspraxia. Magnetic resonance images showed a bilateral medial frontal infarction. Digital subtraction angiography documented a right azygous anterior cerebral artery with severe stenosis in its sub-callosal tract; the left anterior cerebral artery showed mild hypoplasia with only sub-frontal and fronto-polar branches. No embolic source was documented. Afterwards the patient presented a gradual and partial recovery of both motor and cognitive functions.

**Key words** Azygous anterior cerebral artery • Cerebral infarction • Digital subtraction angiography • Magnetic resonance imaging

### Introduction

Anterior cerebral artery (ACA) territory infarctions are uncommon. Indeed, large series of patients with ischemic stroke evaluated by means of computed tomography (CT) showed that the frequency of infarctions confined to the ACA territory ranges from 1% to 4.4% [1-4]. Moreover, angiographic studies documented isolated occlusion of the ACA in only 1%-3.7% of patients with cerebral ischemia [4-6]. Usually, ipsilateral severe carotid artery stenosis with hemodynamic or embolic pathogenesis accounts for these infarctions [2, 7]. On the contrary, ACA atherosclerotic stenosis is unusual and often occurs in the lower part of the curve around the genu of the corpus callosum [8]. An azygous or unpaired ACA is a common trunk that gives rise to branches with bihemispheric distribution; it is a rare anatomical variation documented in 1.1% of human autopsies and it is often associated with aneurysms [9].

In this report we describe an unusual case of bilateral frontal medial infarction caused by azygous ACA stenosis.

### Case report

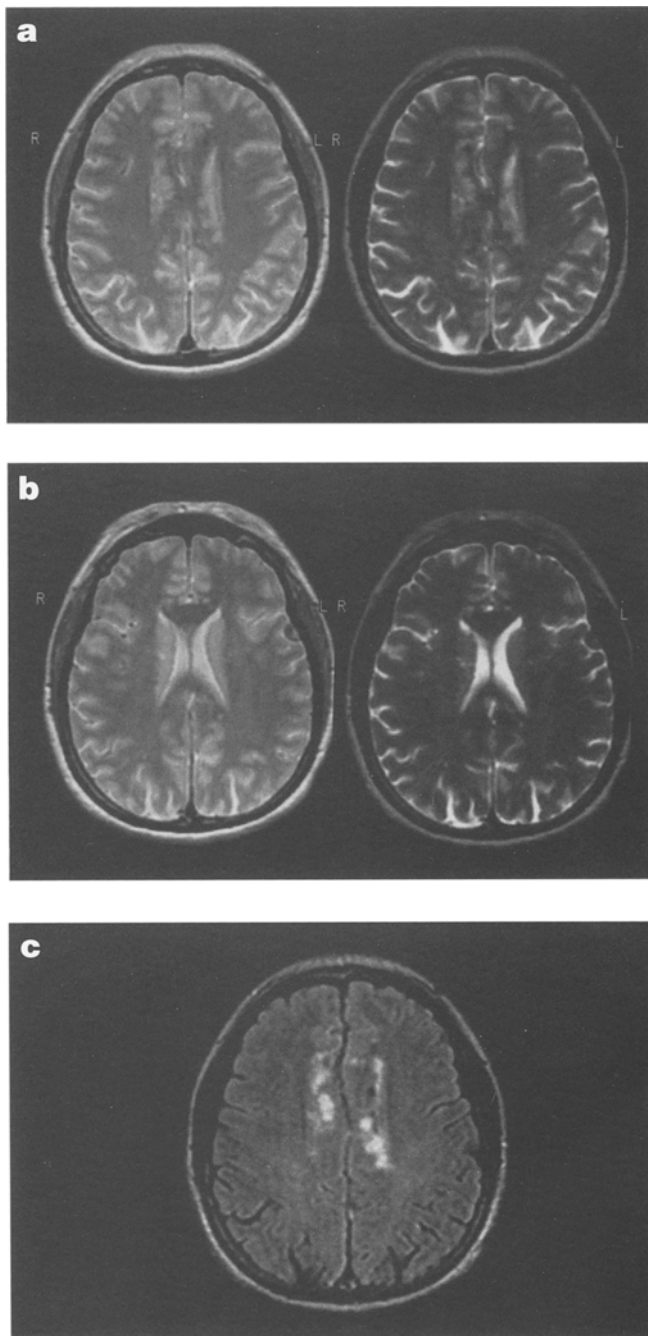
A 63-year-old woman with a history of arterial hypertension and with no other vascular risk factors presented a sudden weakness of the lower limbs, mostly affecting her left leg. Hypertension medication consisted of captopril (50 mg daily) and hydrochlorothiazide (25 mg daily).

Neurological examination at admission showed a fully oriented patient with normal speech and normal cranial nerves. No motor deficit was present in the upper limbs, whereas mild paraparesis was observed prevalently in the left leg. Sensory system was normal and no cerebellar impairment was documented. Deep Tendon Reflexes (DTRs) were brisk with a prevalence on the lower limbs; Babinski's sign was present bilaterally. CT examination

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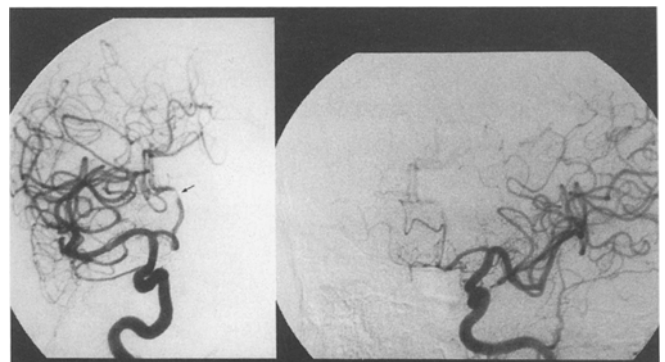
dence of recent lesions. Routine blood tests and ECG were normal. A few hours later paraparesis worsened, and the patient presented incontinence of urine and a marked reduction of spontaneous speech with difficulty in initiation of talking; afterwards she exhibited mutism. Moreover, the patient developed akinesia and forced grasping of both hands. Bilateral ideomotor apraxia was also observed.



**Fig. 1a-c.** a Hyperintense signal on PD and T2-weighted MR axial sections involving in a bilateral and symmetrical way the cortical medial frontal and b the pericallosal regions. c The lesion is more evident in the FIR sequence image

A wide plasmatic auto-antibody search was negative for anti-nuclear antibodies (FAN, FANHEP 2), anti-ENA antibodies, anti-double stranded DNA antibodies (dsDNA), anti-centromere antibodies (ACA), anti-smooth muscle cells antibodies (ASMA), anti-neutrophilous cytoplasm antibodies (ANCA) and anticardiolipin antibodies (ACLA). Coagulation tests including AT-III, protein C, protein S and APC resistance were within normal ranges. Cervical arteries duplex scanner and transcranial Doppler sonography were normal. Transthoracic and transesophageal echocardiography showed no evidence of embolic sources.

Magnetic resonance imaging (MRI) was performed with a 1.5 T system two days after the onset of symptoms. Sagittal T1-weighted images were obtained with spin echo (SE) sequences repetition time (TR), 500 ms; echo time (TE), 15 ms; 1 acquisition). Axial proton density (PD) and T2-weighted images were obtained with fast spin echo (FSE) repetition time (time repetition TR, 5000 ms; echo time TE, 18-19 ms; 1 acquisition) and with fast inversion recovery (FIR) (time repetition TR, 14,500 ms; echo time TE, 130 ms; echo time TE, 12 ms; time inversion TI, 2650 ms; 1 acquisition) sequences. The examination showed an area of hyperintense signal on PD and T2-weighted images involving the cortical medial frontal regions bilaterally and symmetrically, and suggesting an ischemic lesion (Fig. 1). A few days later, digital subtraction angiography (DSA) through a right femoral artery catheter with selective injection in the right and left internal carotid artery was performed. A dolicho-megalic appearance of both common carotid arteries and a bilaterally normal carotid bifurcation were observed. The right internal carotid artery showed kinking with mild stenosis in its middle extracranial tract, and kinking with coiling were present bilaterally in the internal carotid artery prepetrous tract. Intracranial images showed the right azygous anterior cerebral artery with severe stenosis in its sub-callosal tract. This common trunk gave rise to branches with a bihemispheric distribution (Fig. 2). Moreover, the left anterior cerebral artery showed mild hypoplasia and gave rise to only sub-frontal and fronto-polar branches (Fig. 2).



**Fig. 2.** Left Right azygous anterior cerebral artery with severe stenosis (arrow) in its sub-callosal tract. Right Left anterior cerebral artery giving rise to only sub-frontal and fronto-polar branches

The patient was discharged after 20 days with mild improvement of both motor and cognitive functions. A control three months later documented a further partial recovery.

## Discussion

The occurrence of a bilateral medial frontal infarction documented in this patient is due to the unusual presence of two rare conditions: an azygous ACA and isolated severe stenosis in its common trunk. Indeed, azygous ACA may be associated with the presence of aneurysms [9], although no thromboembolic mechanism involving such anatomical variation has been identified. Gacs et al. [2] reviewed CT exams from 413 consecutive patients with cerebral infarction and documented an ACA territory involvement in only 3% of cases. Only one of these patients presented a bilateral ACA territory infarction with bilateral ACA occlusion seen on angiography; no clear cause for this evidence was found. A pathogenetic mechanism identified in ACA territory infarctions is embolism in patients with increased flow through the anterior communicating artery because of ipsilateral carotid artery occlusion [2, 7]. Other mechanisms are spasm, embolism or propagating thrombosis associated with an anterior communicating aneurysm. Our case showed none of these occurrences, whereas we did document isolated atherosclerotic stenosis of the common trunk of ACA in which arterial hypertension was the only identified vascular risk factor. Indeed, no embolic source nor other cause of cerebral ischemia was identified.

The patient showed symptoms attributable to the lack of blood supply in the paracentral lobule and in the supplementary motor area [10]. The paracentral lobule impairment accounts for the crural motor deficit and the incontinence of urine. Sensory symptoms were not documented in our case. On this matter, in relation to the posterior extent of the ACA branches and the collaterals from the posterior cerebral artery, sensory symptoms caused by the involvement of the postcentral gyrus may be absent or too mild to be evident if patients have serious cognitive deficits. The supplementary motor area plays an important role in the initiation and regulation of voluntary movement and speech; therefore its impairment accounts for the mutism, akinesia and dyspraxia observed in our case. Also, the forced grasping of the hands is attributable to the involvement of the supplementary motor area [10]. The sudden occurrence of such symptoms may suggest an unusual ischemic lesion in both the ACA territories that may depend on the impairment of an azygous ACA.

**Sommario** *Descriviamo il caso di una donna di 63 anni affetta da ipertensione arteriosa che ha presentato un improvviso deficit di forza agli arti inferiori seguito da mutismo, acinesia e disprassia. La risonanza magnetica (RM) cranica ha evidenziato un infarto frontale mediale bilaterale. L'angiografia digitale ha documentato un'arteria cerebrale anteriore destra di tipo azygos con stenosi serrata nel tratto sotto-callosa; l'arteria cerebrale anteriore sinistra presentava una lieve ipoplasia e dava origine soltanto a rami sotto-frontali e fronto-polari. Successivamente la paziente ha presentato un graduale e parziale recupero sia del deficit motorio che cognitivo.*

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