Callosal mutism after AVM embolization in territory of distal anterior cerebral artery branches – a rare complication. Case report

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Abstract

Introduction: Callosum mutism (CM) is a rare complication of neurosurgical procedures. There is no previous report of its occurrence after interventional endovascular procedures for intracranial lesions. Material and method: We present a case of CM after embolization of anterior cerebral artery branches for treatment of a left frontal arterio-venous malformation (AVM). After reporting the particularities of clinical and radiological presentation of such case, we perform a critical literature review and discuss the clinical and etiological aspects of CM as well as its differences in relation to other genders of speech disturbances (like aphasias). Finally we provide a brief overview about present available methods of cortical function monitoring emphasizing their utility in therapeutic endovascular procedures in eloquent areas. Result and conclusion: In literature, there are no previous reports of CM as a complication of therapeutic angiographic procedures. Its exact pathophysiology is unknown.

It is usually believed that this syndrome may appear after lesion of any of the components of the dentate-thalamic-cortical pathway, as well as the supplementary motor area and meso-limbic regions involved in speech production. The diagnosis of CM must be considered after a clinical picture of speech deficit after therapeutic endovascular procedures in territory of distal branches of anterior cerebral artery, specially of frontopolar (Fpa branch) and anterior internal frontal (AIFA) branches.

Key words
Cerebral Angiography, AVM embolization, callosum mutism, endovascular treatment.

Sinopse

Mutismo caloso após embolização de malformação arteriovenosa em ramos distais da artéria cerebral anterior – relato de caso

Introdução: O mutismo caloso é uma complicação rara de procedimento neurocirúrgico, não havendo relato prévio na literatura de sua ocorrência após terapia endovascular de lesões intracerebrais. Materiais e Métodos: No presente artigo, relatamos um caso de mutismo caloso após embolização de ramos distais da artéria cerebral anterior para tratamento de uma malformação arteriovenosa (MAV) frontal no hemisfério
esquerdo. Após exposição das particularidades das apresentações clínica e radiológica do presente caso, realizamos uma revisão crítica da literatura atual discutindo a etiologia do mutismo caloso, bem como suas semelhanças e diferenças em relação a outros distúrbios da linguagem (como as afasias, por exemplo). Por fim realizamos um breve apontado acerca dos métodos atualmente disponíveis para a monitorização de funções corticais, enfatizando os benefícios de seu uso durante a realização de procedimentos terapêuticos endovascularmente em áreas eloquientes. **Conclusão:** O quadro clínico consiste em ausência transitoria da fala em um paciente com compreensão intacta e sem evidência de deterioração da atividade intelectual, lesão cortical em área primária da linguagem ou apraxia oral por lesão nervosa periférica. A fisiopatologia exata do mutismo caloso ainda permanece desconhecida. Acredita-se, no entanto, que tal quadro clínico possa surgir após lesão de qualquer dos componentes do sistema dentato-talâmico-cortical, bem como da área motora suplementar e regiões mesolimbícas adjacentes envolvidas na produção do discurso. O diagnóstico deve ser suspeitado diante de um quadro clínico de déficit de linguagem após realização de procedimento endovascular em ramos corticais da artéria cerebral anterior, em especial dos ramos frontoparietais (ramos FpA) e anteriores frontais internos (ramos AIFA).

**Palavras-chave**

Angiografia cerebral, embolização MAV, mutismo caloso, terapia endovascular.

**Introduction**

Embolication has a significant role in multimodality treatment of brain AVMs, by either enabling or facilitating subsequent microsurgical or radiosurgical treatment. Appropriately targeted embolization in otherwise untreatable AVMs represents a reasonable form of palliative treatment of either ameliorating the clinical condition of the patient, or reducing the potential risk of hemorrhage.

Some studies show that approximately 40% of patients with brain AVMs can be cured by embolization alone with mortality rate of 1.3%. Permanent morbidity related to pre-surgical embolization varied from 4% to 8.9%. The most common complication of AVM embolization is subarachnoid hemorrhage or intra-cerebral hemorrhage caused by the balloon bursting and dissection of the feeding vessel, or by a fistulized AVM.

Callosal mutism (CM), as complications of neurosurgical procedures, is a rare complication and its occurrence, due to therapeutic endovascular procedures, is not yet reported in literature.

**Case Report**

HVS, 24 years old man, presented with complex partial epilepsy crisis, was brought to the emergency of Santa Paula Hospital. He was put on phenotoin, and a CT scan was performed showing a left extensive frontal callosal lesion suggestive of an AVM (Figure 1). MRI showed a left sided AVM with high flow and deep venous drainage (Figure 2). He was submitted to an angiogram which showed arterial feeding from anterior cerebral artery (ACA) branches. A selective embolization with glue (Figure 3) of frontopolar (Fp) branch and anterior internal frontal (AIFA) branches of ACA, without embolization of middle internal frontal (MIFA) or distal pericallosal branches was performed. (see figures 4 and 5 for schematic representation of distal ACA branches). Three hours later, the patient became mute and presented a right hemiparesis, predominantly in the leg, without any cranial nerves deficits. The patient was conscious and conserved intact comprehension without evidence of oral apraxia or aphasia, meeting criteria for characterization of mutism. CT scan showed a hemorrhage inside the AVM perimeter with minimal deviation. He was submitted to corticosteroid therapy, with dexametasona 4mg IV 4/4hours; plasma 1 unit IV 8/8hours and platelets unit IV 8/8hours for 3 days in the Intensive Care Unit. Control CT scan did not show increase of the haematoma, and the patient recovered both speech and hemiparesis 48 hours after the embolization. Nine days after the first embolization, he was submitted to a second additional embolization, in order to occlude middle internal frontal (MIFA) branches of distal ACA.

**FIGURE 1.**

CT scan (non-contrast axial cut) – left frontal callosal lesion suggestive of an arterio-venous malformation (AVM).
FIGURE 2.
MRI (coronal cut)- high flow left sided frontal AVM with deep venous drainage.

FIGURE 3.
Angiogram - AVM feeding predominantly through branches of left anterior cerebral artery. A glue selective embolization of frontopolar artery and anterior internal frontal branch of callosum marginalis artery was proceeded.

FIGURE 4.
Schematic representation of distal ACA branches. A.I.F.A., anterior internal frontal artery; Cal., callosal; Cm., callosomarginal; Fp., frontopolar; I., inferior; Inf., inferior; M.I.F.A., middle internal frontal artery; Of., orbitofrontal; Par., parietal; Pce., paracentral; Perc., pericallosal; P.I.F.A., posterior internal frontal artery; Precal., precallosal; Rec., recurrent; S., superior; Sh., short.

FIGURE 5.
Vascular supply of distal ACA branches.
A.I.F.A., anterior internal frontal artery; Fp., frontopolar; I., inferior; M.I.F.A., middle internal frontal artery; Of., orbitofrontal; Par., parietal; Pce., paracentral; P.I.F.A., posterior internal frontal artery; S., superior.

with complete occlusion of AVM (Fig. 6). The patient was discharged from hospital 15 days after admission with no deficits and no speech disturbance. After 3 months, control MRI showed partial absorption of the haematoma, and absence of abnormal vascular flow inside the perimeter of previous AVM.

Discussion

CALLOSUM MUTISM

Mutism is generally defined as complete absence of speech in a conscious patient with intact comprehension and no evidence of oral apraxia. 2, 9 Mutism can be subdivided in motor aphasia, akinetic mutism, mutism following thalamotomy, pseudobulbar palsy due to diffuse bilateral hemispheric lesion, phonatory system lesion, 16 and transient mutism after transcallosal approach to ventricles13.

Some frontal cortices located in the internal (mesial) cerebral surface of the left hemisphere (including supplementary motor area and the anterior cingulate area) play an important role in the initiation and maintenance of speech. They also play a role in attention and emotion and thus, can influence many higher functions. Damage in these areas does not cause aphasia per se, but rather varied degrees of "akinesia", a difficulty with initiation of movement and "mutism", a complete absence of speech, which is rarely seen in true aphasia.
Current knowledge about mutism sustains that it is not caused by general intellectual deterioration, cortical lesion, or peripheral damage affecting speech production. The syndrome may result from severing inter-hemispheric connections in cases where both hemispheres are required for speech production.14

Mutism may be a result of division of the corpus callosum. Suppression of the limbic system caused by lesions in the anterior cingulate gyrus, septum pellucidum, and fornix are important causes of CM. Impairments of the supplementary motor cortex, thalamus and basal ganglia may also be factors reducing speech production. The mechanism of such transient mutism seems to be a complex of two or more of these factors, and their combinations may be different from one case to another.13. In the reported case, we believe that transient mutism was mainly due to haematoma mass effect over anterior cingulate gyrus and basal ganglia, specially the head of caudate nucleus.

Another type of mutism reported in literature is cerebellar mutism. It has been described as a benign complication after posterior fossa resection of midline huge cerebellar tumors, mainly in childhood. It is characterized by complete absence of speech without motor or nuclear long tract symptoms and signs. Cerebellar mutism is transient and in average takes 24-72 hours to clear up, similarly to our mentioned case. For this reason, we advocate that, as in cerebellar mutism, the dentate-thalamic-cortical pathway might be involved in callosal mutism, resulting in frontal cortex lesions nearby the corpus callosum.

CM is a rare complication of cerebral procedures and it is usually associated to callosotomy as treatment for drug-resistant epilepsy.5,15,17: other less frequent causes are the transcortical approach to the III ventricle,14 subarachnoid haemorrhage, infarction in the territory of the left anterior cerebral artery,6 and resection left frontal lobe tumors.11,14

### Monitoring Methods

There are several ways of monitoring brain activity. The most used during endovascular procedures are: Motor Evoked Potentials (MEP), Somato-sensory Evoked Potentials (SSEP) and scalp EEG.

There is also experimental research of new methods of monitoring brain’s electrical activity during angiographic procedures like the intracranial electro-encephalography (IAEEG). This method consists in intracranial monitoring of brain electrical activity by means of an intra-arterial guide wire as electrode. It is equivalent to using a semi-invasive electrode, but IAEEG recording can be performed simultaneously to the endovascular procedures by adapting it at the tip of angiographic catheter.

Literature about routine use of cortical monitoring methods during endovascular procedures is controversial. Although some authors consider it necessary in order to have dynamic information about brain functions, others argument that monitoring endovascular procedures would unnecessarily increase the cost of the treatment, bringing few, if any, practical results.5,9
Conclusions

Complications of AVM procedures are unpredictable and sometimes consist in rare syndromes, like the one previously reported. Therefore, methods of monitoring brain function are an important tool in order to make endovascular therapeutic procedures safer, once they are capable to detect early ischemic events during AVM embolization, enabling prediction, evaluation and prognosis of possible future deficits. In cases of onset of any functional deficit, AVM embolization must be stopped and new image exams must be performed. Although much care is required in the further management of such patients, a new embolization procedure is not contraindicated and can be proceeded once the clinical status of the patient is normalized.

The authors also emphasize the importance of theoretical knowledge of brain anatomy and its cognitive functions, as well as clinical features and differences among various types of speech deficits (like mutism and aphasia), in order to make the neurosurgeon able to precisely determine brain structures and cortical pathways which might be affected by ischemic events.

References


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