A Choreoathetosis due to Subarachnoid Hemorrhage in a Togolese

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Abstract

The hemichorea is the most common movement disorder after stroke. This is a very rare complication of Subarachnoid Hemorrhage (SAH) of which only six cases have been reported to date before ours. We report the case of a 65-year-old woman who was admitted for choreoathetosis with a language disorder that had been evolving for three days ago. The neurological examination noted in addition a meningeal syndrome. Brain CT scan found intraventricular hemorrhage with a small hematoma of the corpus callosum. An aneurysm of the anterior communicating artery was visualized on the angiography. The abnormal movements regressed correctly with neuroleptic (Tiapride).

Keywords: Choreoathetosis; Subarachnoid hemorrhage; Togo; Africa

Introduction

Movement disorders can occur as primary or genetic disease, as a manifestation of an underlying neurodegenerative disorder, or secondary to a wide range of neurological or systemic diseases. Cerebrovascular diseases represent up to 22% of secondary movement disorders, and involuntary movements develop after 1% to 4% of strokes [1]. Some studies report hemichorea as the most frequent post-stroke movement disorder [2,3]. However, abnormal movements following Subarachnoid Hemorrhage (SAH) are poorly reported [4]. The first case of movement disorder after SAH was reported in Japan in 1991 [5]. To date, in the present state of our knowledge and after the review of the literature, there is no reported case in Africa. We report a case of a 65-years-old woman in whom choreoathetosis was the revealing manifestation of its SAH and do more review for this presentation.

Case Presentation

A 65-year-old woman, right-handed was admitted on September 21st, 2018 at the emergency of Teaching Hospital of Kara for language disorder and appearance of abnormal movements. Symptoms began three days earlier, by a sudden loss of consciousness of short duration, without fever or clonic and tonic manifestation. On the return of consciousness, she reported headaches, and her daughter noted a speech disorder. The persistence of symptoms and the appearance of abnormal movements 48 h later motivate his admission to the hospital for better care. There is no comorbidity in her medical history. She had no family history of degenerative neurological disease, including dementia, movement disorders, or psychiatric illness. The first examination noted an alteration of the general condition, a temperature at 37.4ºC and a blood pressure of 160/90 mmHg at both arms. The consciousness was altered with a Glasgow Coma Scale of 13/15. The patient was able to answer and execute simple commands which made it possible to note a Broca’s aphasia. She had no motor deficit. Deep tendon reflexes were normal associated with a bilateral sign of Babinski. There were abnormal limb movements made of wave sweeping movements from the roots of their members to end sometimes with a deformation of the trunk. These movements were mainly triggered by a change of position or passive flexion of the neck in search of meningeal stiffness. They disappeared during sleep. There was no disturbance of sensitive functions. The examination of the cranial nerves was normal. The neck was stiff with a positive sign of Brudzinski. Examination of the cardiovascular and pulmonary apparatus was normal. Abnormal movements and non-febrile meningeal syndrome have been suggestive of hemorrhage of the basal ganglia with intraventricular extension. A CT scan was
performed and revealed a small interhemispheric hematoma located in the corpus callosum associated with intraventricular hemorrhage (Figure 1). This CT scan was supplemented by a cerebral angiography scan that revealed aneurysm of the anterior communicating artery (Figure 2). A complete blood count, biological functions of the liver and kidney were normal. Serological tests for HIV and hepatitis B and C were negative as well as syphilis serology. There was no hemostasis disorder. The electrocardiogram was also normal. Conventional treatment of meningeal hemorrhage was done and included resuscitation measures, administration of analgesic, an inhibitor of angiotensin converting enzyme, and the prevention of vasospasm with Nimodipine 60 mg every 6 h by nasogastric tube. Tiapride 100 mg/12 h is used for abnormal movements. Oral medications were administered by the nasogastric tube. Consciousness improved on day 4 and choreoathetoid movements regressed on day 5. Tremors completely regressed at end of 13 days and the patient was released October 17th, 2018.

**Discussion**

Stroke related movement disorders are uncommon (3.6%) and are very rare in SAH cases. Chorea, tremor, dystonia, Parkinsonism, and myoclonus have all been associated with cerebral infarcts and hemorrhaging [6]. Movement disorders, which represent part of the clinical spectrum at the acute stage of stroke, may also be of late onset [4]. In our patient, the abnormal movements appeared at the acute stage, 48 h after the onset of symptoms. This mode of onset of abnormal movements in the acute phase of SAH was reported in Japan by Sakai in 1991 and Morigaki in 2008. They respectively found 8 and 2 days as the delay of onset of abnormal movements after the beginning of symptoms [5,7]. Chorea is characterized by involuntary, abrupt movements, literally exploding, brief, rapid, and unpredictable and of variable amplitude [8]. Athetosis is characterized by slow, sinuous, continuous, repetitive, irregular movements, which are often increased by the activation of another part of the body, stress or sensory stimuli, disappearing during sleep. Athetosis can be very slow approaching dystonia or sometimes faster bringing it closer to chorea. With the latter, it forms a continuum with, at one end, the slowest movements representing chorea; intermediate movements being defined as choreoathetosis [8]. The abnormal movement presented by our patient corresponded to a choreoathetosis. According to the literature review, abnormal motions after a SAH predominate in women [4] just as the case we report. Indeed, Pereira in 2011 claimed that by analyzing the literature data on movement disorders after a SAH, he observed a total of six patients (including his reported case). Of these six patients, five were female and one was male, with a mean age of 58.6 years (SD 21.6, ranging from 17 to 74 years). Chorea or choreoathetosis was observed in three of these cases (50%); tremor was observed in the other three cases. One patient in the tremor group exhibited associated dystonia [4]. In 2013, Diaz-Maroto also reinforced this assertion by saying that only 3 cases of choreoathetosis associated with SAH have been reported in the literature [9]. The case of Pereira was very atypical because he is the youngest patient ever described and was the only male to present with SAH-related involuntary movement [4]. The most common involuntary abnormal movement after a stroke is chorea. It occurs in the form of hemichorea and is usually associated with contralateral lesions involving the basal ganglia, the internal capsule, the diencephalon or midbrain [9]. The precise mechanism underlying movement disorders associated with stroke in the basal ganglia and circuitry is not fully understood [10]. Several hypotheses are therefore emitted. It may be a direct attack on the motor circuits involving the basal ganglia or a decrease in cerebral blood flow during cerebrovascular episodes [1,2,10]. In the case of an SAH the mechanism explaining the abnormal movements as in our patient is even more obscure. These abnormal movements after SAH are very uncommon the core mechanism is difficult to explain. In our patient, the brain CT scan showed a small hematoma in the corpus callosum. The same point was noted by Morigaki who reported the case of a 72-year-old woman who submitted hyperkinetic movements after SAH [7]. This author attributed the symptoms to the hematoma of the corpus callosum and according to him; involuntary hyperkinetic movements would be due to the interruption of the cortico-striato-pallido-thalamo-cortical feedback loop. Cortico-striatal fibers cross to the contralateral striatum; therefore, interruption of crossing fibers at the corpus callosum is an alternative explanation for her choreoathetosis [7]. The regression of the abnormal movements was obtained in our patient after administration of an atypical neuroleptic (Tiapride) simultaneously with the measures of prevention of vasospasm with nimodipine. Haloperidol was the neuroleptic used.
by Pereira et al. [4] to obtain a favorable evolution in 4 days. In the review of the literature, the efficacy of neuroleptics, like that of haloperidol, is well documented. The alternatives are the neuroleptics atypical, as the clozapine which allows avoiding the appearance of dyskinesia later [8].

**Conclusion**

Choreoathetosis is a rare complication of SAH. The pathophysiology of this abnormal movement after a SAH is unclear until this day. It is important to have in mind the diagnosis of SAH in patient with chorea or choreoathetosis especially when this sign is associated with meningeal syndrome.

**References**