Coexistence of Two Different Types of Intracerebral Hematomas Caused by Arteriovenous Malformation

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Spontaneous intracerebral hematomas usually produce the sudden onset of devastating neurological symptoms. We describe a case showing slowly progressive clinical symptoms followed by a sudden deterioration caused by two different types of intracerebral hematomas coexisting in the adjacent area. A 72-year-old female with a 2-year history of Alzheimer's disease and a 3-month history of occasional headache and vomiting was admitted after she experienced the sudden onset of right hemiparesis and a speech disturbance. Neuroradiological examinations demonstrated two different types of intracerebral hematomas coexisting in the left temporal lobe. The patient underwent a left frontotemporal craniotomy. A solid hematoma was found immediately below the cortex and a large hematoma cavity, which contained degraded bloody fluid, was found below the solid hematoma. Histological study demonstrated an arteriovenous malformation (AVM). The AVM may have been responsible for the pathogenesis of these two different types of intracerebral hematomas. Initially, a silent intracerebral hemorrhage from the AVM and liquefaction of the hematoma probably formed the cavity and repeated small subclinical hemorrhages into the cavity during a prolonged period of time may have caused the growth of the inner hematoma. The sudden rupture of the AVM most likely caused the outer solid hematoma, resulting in the abrupt onset of hemiparesis and speech disturbance. This is the first case to demonstrate two different types of intracerebral hematomas coexisting in the adjacent area. A possible mechanism for this rare condition is discussed.

Key Words: arteriovenous malformation, intracerebral hematoma, magnetic resonance imaging

Spontaneous intracerebral hematomas are usually secondary to systemic arterial hypertension and produce the sudden onset of devastating neurological symptoms. In contrast, another type of intracerebral hematoma producing stable or slowly progressive neurological deficits has been reported. [5,21] We present a case showing slowly progressive clinical symptoms, which went untreated, followed by a sudden deterioration. Neuroradiological studies demonstrated two different types of intracerebral hematomas coexisting in the left temporal lobe. A possible mechanism for this rare condition is discussed.

Case Report

This 72-year-old normotensive female had been cared for at a nursing home for two years following the diagnosis of Alzheimer's disease. After placement in the nursing home,

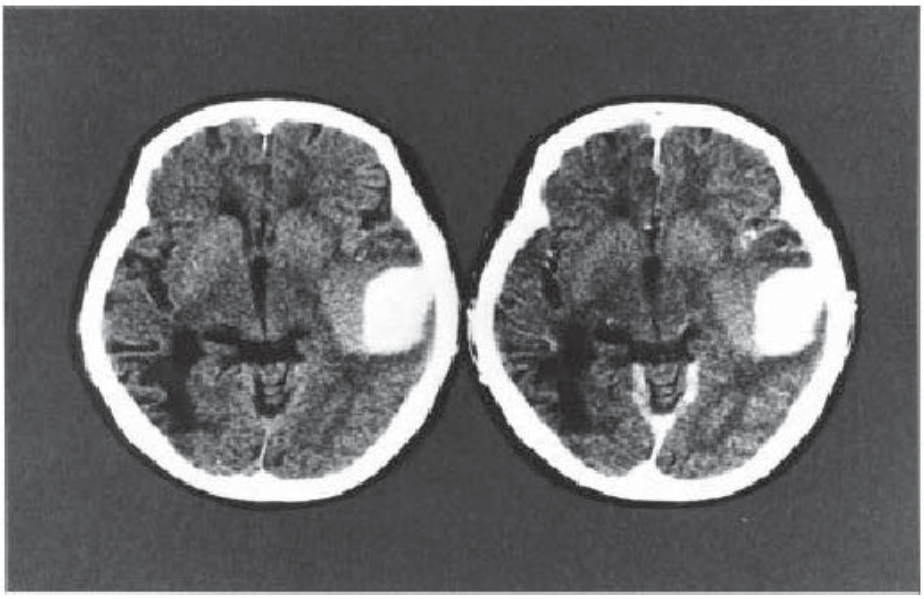

Figure 1. Computed tomography scans on admission showing intracerebral hematoma in the left temporal area without significant enhancement (left, plain image; right, postcontrast image).

caregivers sometimes found it difficult to communicate with her, but her talking ability was normal. Three months prior to admission to our hospital, she began to complain of occasional headaches and exhibited nausea and vomiting. Three weeks prior to admission, she began to have balance problems and fell easily, but her mood did not change. On the day of admission, a caregiver found that she experienced a sudden onset of right hemiparesis and a speech disturbance, and she was referred to our institution. A magnetic resonance imaging (MRI) had been performed 2 years earlier when she was diagnosed with Alzheimer's disease. This examination had demonstrated an old cerebral infarction on the right occipital lobe, but no vascular malformation was seen.

Upon admission, a neurological examination demonstrated right hemiparesis and a speech disturbance. Funduscopic examination showed blurring of the nasal margin of the disk. Computed tomography (CT) scan of her head obtained emergently demonstrated an intracerebral hematoma in the left temporal lobe. The hematoma had both blurred and dense components (**Figure 1**). Subsequent MRI her head clearly demonstrated the coexistence of two distinct types of intracerebral hematomas. T1-weighted imaging (TIWI) showed the inner hematoma as hypointense and the outer as isointense. T2-weighted (T2WI) sequence showed a fluid level within the inner hematoma cavity. The inner hematoma wall did not enhance after gadolinium administration (**Figure 2**). The MRI findings of the inner hematoma were comparable to those of a chronic subdural hematoma. The outer hematoma showed a slight postcontrast posterior peripheral enhancement. Therefore, we concluded that the lesion was composed of two different types of intracerebral hematomas. Both the clinical and radiological findings suggested that the inner hematoma had been present before the formation of the outer hematoma. Cerebral angiography was not performed.

The patient underwent an emergent left frontotemporal craniotomy on the day of admission. A solid hematoma was found immediately below the cortex. An abnormal vessel structure was found in the posterior wall of the outer hematoma, and was completely excised. A large cavity, which contained about 25 ml of degraded bloody fluid, was found below the outer hematoma. The wall of the inner hematoma cavity was also sampled. Histological examination of the excised lesion demonstrated an AVM (**Figures 3**). The wall of the inner hematoma cavity was normal brain tissue.

The postoperative course was uneventful, and her right hemiparesis gradually improved. Postoperative cerebral angiography did not demonstrate the residual nidus. The patient returned to the nursing home 2 weeks after surgery. MRI performed at the time of discharge showed no signs of vascular malformation.

Discussion

Intracerebral hematomas usually present with the sudden onset of focal neurological deficits. Maximal severity is usually reached immediately or within several hours of the initial clinical ictus. However, our case showed a slowly progressive clinical course until an abrupt deterioration occurred. Similar instances have been reported of intracerebral hematomas behaving like slowly expanding neoplasms, with progressive neurological deficits. [5,9,10,12,21] Pozzati, et al., reported 10 patients with chronic expanding intracerebral hematomas. They divided these chronic expanding intracerebral hematomas into two groups: liquefied collections and encapsulated lesions.[9] The inner hematoma in our case was not encapsulated, and the cavity was filled with liquefied blood.

AVMs are rarely associated with large cavities, with only 11 cases having been reported.[1,3,4,6,9,13,17,19] The content of these cysts was xanthochromic or liquefied blood, and past

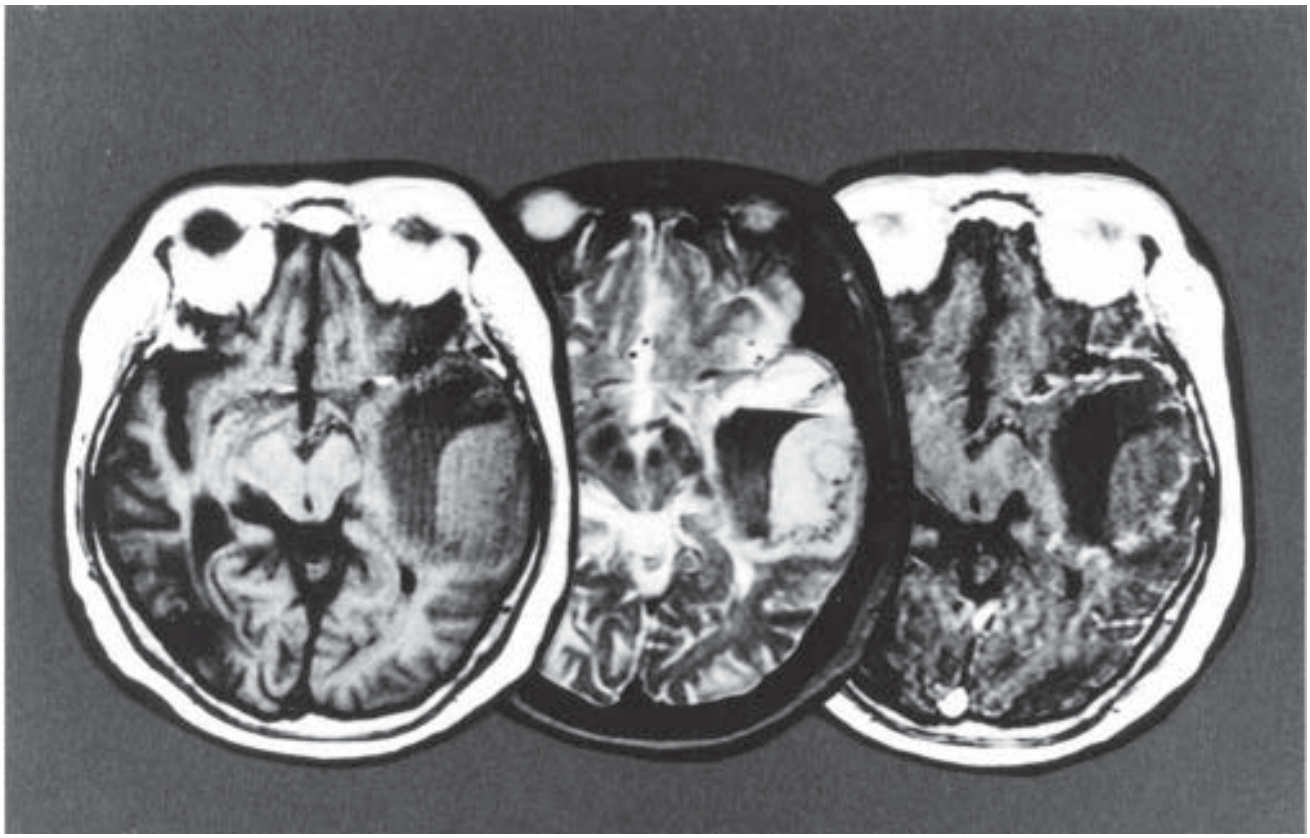

Figure 2. Magnetic resonance images on admission showing two different types of intracerebral hematomas in the left temporal lobe. T1-weighted image (left) shows the inner hematoma as hypointense and the outer hematoma as isointense. T2-weighted image (center) shows a fluid-fluid level within the inner hematoma cavity. Retrospective review identifies the flow void-like appearance (arrow). Gadolinium enhanced T1-weighted image (right) shows posterior peripheral enhancement of the outer hematoma.

reports have hypothesized the participation of hemorrhages, or an exudative process in formation of the cyst. [1,3,6,20] In this case a silent intracerebral hemorrhage from the AVM and liquefaction of the hematoma probably caused the initial cavity, and repeated small subclinical hemorrhages into the cavity during a prolonged period of time may have caused the growth of the inner hematoma. The patient's slowly progressive symptoms, which did not receive medical attention, were thought to be due to the expansion of the inner hematoma. Then the sudden rupture of the AVM most likely caused the outer solid hematoma, resulting in the abrupt onset of hemiparesis and a speech disturbance. On MRI, intracerebral hematomas start out hypointense for the first few days and then become markedly hyperintense on T1WIs. On T2WIs hematomas are initially almost indistinguishable from non-hemorrhagic edematous lesions, appearing somewhat brighter than the surrounding brain. Within 24 hours, they become markedly hypointense.[2] Thus these MR findings support the assumption that the inner hematoma had been present before the formation of the outer hematoma. Although hematomas of various ages are not rare, this is the first case to demonstrate two different types of intracerebral hematomas coexisting in the adjacent area. This case is also interesting in regards to the developmental process of AVMs. AVMs have been considered to be congenital malformations. However, clinical reports on the presence of enlarged AVMs and the recurrence of cerebral AVMs after normal findings on postoperative angiograms have fostered arguments against the assumption that AVMs are strictly congenital lesions. [7,8,11,15,18] Recently, enhanced vascular proliferation and neoangiogenesis were documented in cerebral AVMs.[14,16] These clinical data lead to the hypothesis that cerebral AVMs may not be just congenital lesions. In this case, the patient had an MRI examination 2 years prior to admission that was negative for AVM. A retrospective review of the MR examination performed upon admission identified a flow void-like appearance (**Figure 2**), but the first MR examination with the same high-field, 1.5-tesla imager (GE Signa Scanner; GE Medical Systems, Milwaukee, USA) using similar sequences did not demonstrate any vascular lesions. It is possible that the AVM was present, but was missed 2 years prior to this admission because cerebral angiography was not performed at that time. However, the possibility of de novo AVM formation may not be denied. At least this case suggests the possibility of behavioral changes in a silent AVM in the 7th decade of life.

In addition, the majority of patients with Alzheimer's disease are cared for by family members at home. Even in the nursing home setting, patients often receive inadequate

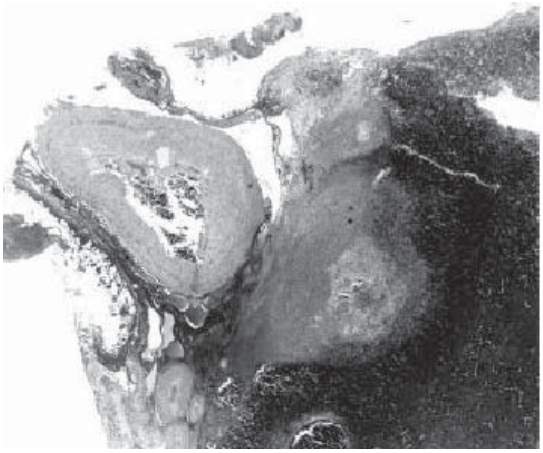

Figure 3. Photomicrographs of the solid hematoma wall demonstrating arteries of various sizes adjacent to the clot (Hematoxylin and Eosin stain , x 20).

management mainly because of communication difficulties. As a result of the shift towards care in the community, the informal caregivers occupy an increasingly central role in the care of these patients. Therefore, knowledge and recognition of slowly progressive diseases, such as the hematoma observed in this case, is essential for the management of patients with dementia.

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