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A Case of Moyamoya Syndrome with Subdural and Intracerebral Hematoma due to Different Bleeding Sources

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One case with a moyamoya syndrome is reported in which each of an intracerebral and subdural hematoma was caused by different bleeding sources. At first, an intracerebral hematoma was made by rupture of moyamoya vessels per se. Then, distortion of the brain would have resulted in tearing of an aneurysm which had been formed by a overloading to its wall at one of transdural anastomotic channels. Thus a subdural hematoma was developed.

INTRODUCTION

In the presentation of his Caldwell Lecture, 1968, Taveras [2] cited a case with a subdural hematoma developed after ventriculography with considerable subdural air. He pointed out an importance of shearing of enlarged transdural arterial channels as a significant source of bleeding in moyamoya syndrome.

We are reporting a case with acute subdural hematoma caused by rupture of an aneurysmal dilation of vessel located in one of a transdural meningo-cortical artery. The rupture itself would be triggered by distortion of the brain which resulted from bleeding of moyamoya vessels into the brain tissue, thus both an intracerebral and a subdural hematoma due to different source were observed.

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CASE REPORT

A 52-year-old housewife had experienced a sudden onset of unconsciousness on January 11, 1978. It lasted about ten minuted. Upon recovery she noted a slight weakness in the left upper extremity which continued stationary. There had been no prior neurologic symptoms, nor was any evidence of systemic disease elicited. Then, over a period of about two months prior to admission on March 22, she had recurrent attacks of muscle twitching of the left limbs of about 10–15 minutes in duration. At least 3 of these attacks were also accompanied by transient numbness of the left hand.

Upon admission, she was alert and no mental deterioration was noted. There was a slight weakness of the left arm and leg. The left tendon reflexes were accentuated, but no pathological reflexes were elicited. The rest of neurological examinations were normal and routine laboratory studies were all within normal limits.

An angiographic occlussion was noted in the right internal carotid artery at the level of the siphon with marked appearance of the basal and the ethomoidal moyamoya (Fig.1). Collaterals between the right posterior cerebral artery and the right middle and pericallosal arteries were noted (Fig.2). A external carotid angiogram also revealed marked develop-

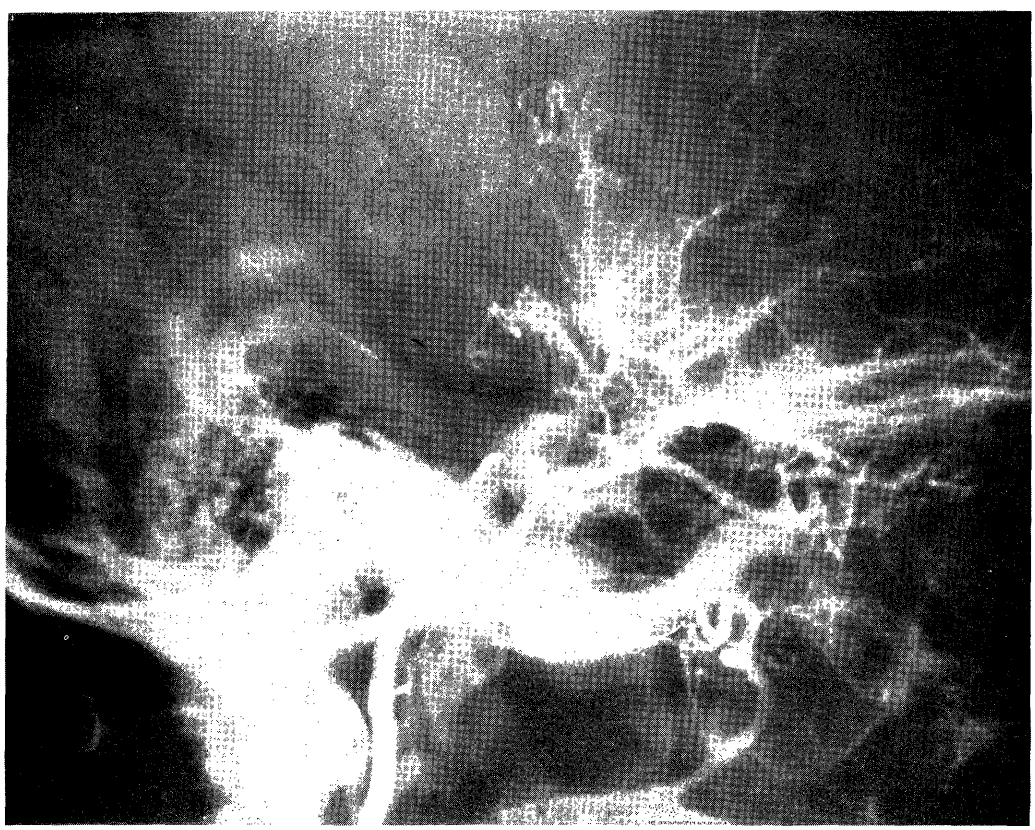


Fig. 1. Right common carotid angiogram shows severe stenosis of the internal carotid artery in its supraclinoid portion with enlarged opthalmic artery. A fine network of collateralis (moyamoya) is seen in the basal ganglia.

ment of transdural connections between the meningeal arteries of the dura mater and the cortical vessels (Fig.3).

The left carotid angiogram was normal.

CT scan on March 24, demonstrated an enlarged anterior horn of the right lateral ventricle and low absorption foci in the surrounding brain tissue. However, CT provided no information on the rich vascular networks observed in angiograms even after intravenous contrast enhancement (Fig.4).

Her hospital course was stable until on April 17, when she suddenly had a severe headache with recurrent vomiting. She complained of an numbness in the left arm. Moderate hemiparesis in the left limbs and the left facial palsy of the central type were noted. About three hours later, she became completely comatose and decerebrate. Emergency CT depicted a different picture as compared to the previous one indicating a presence of the right intracerebral bleeding with the ventricular compression and midline shift. In addition, a high absorption extracerebral space was also seen on CT (Figs. 5a, b).

A tentative diagnosis was an intracerebral hematoma due to rupture of moyamoya vessels and a part of the blood would penetrate into the subarachnoid and subdural spaces. There was no suspicion at that time about a presence of extravasated blood in the subdural

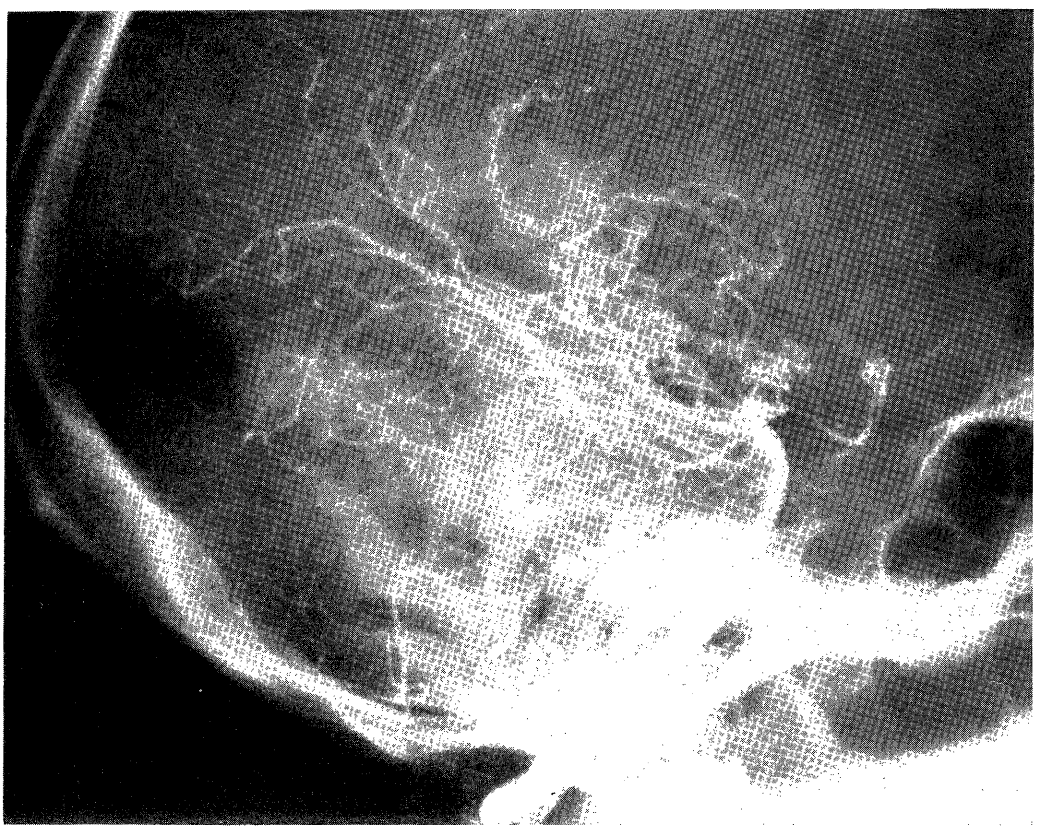


Fig. 2. Vertebral angiogram demonstrates abundant collaterals between the right posterior cerebral artery and the basal ganglia region.

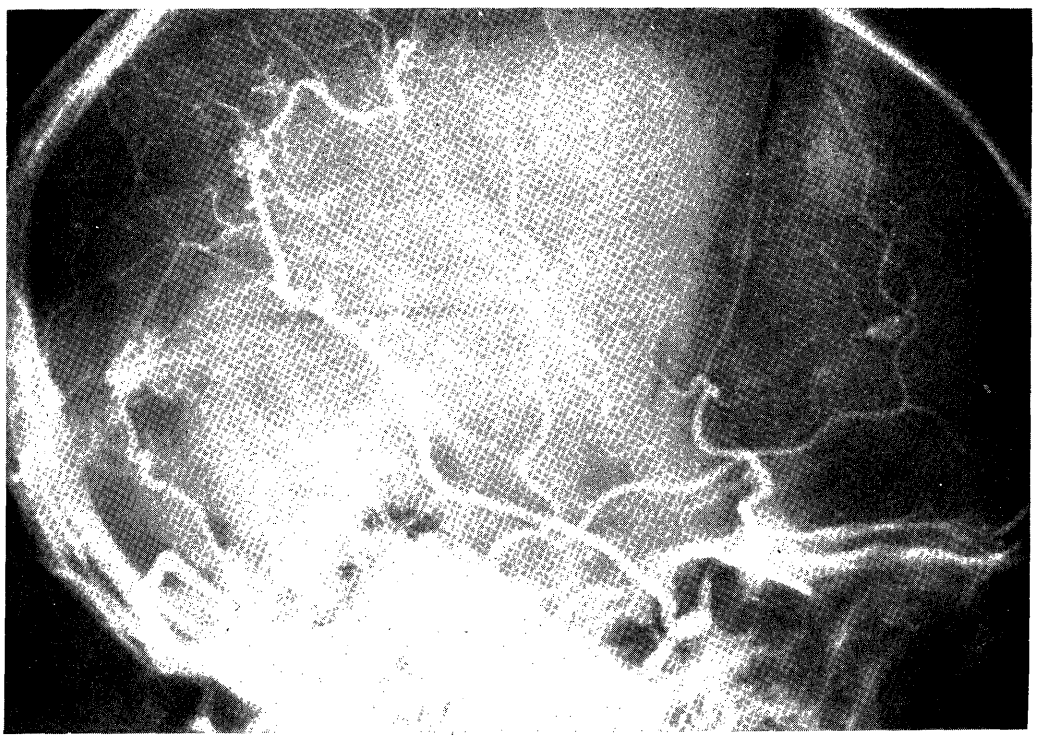


Fig. 3. Right external carotid angiogram showed transdural anastomotic channels by way of its menigeal branches.

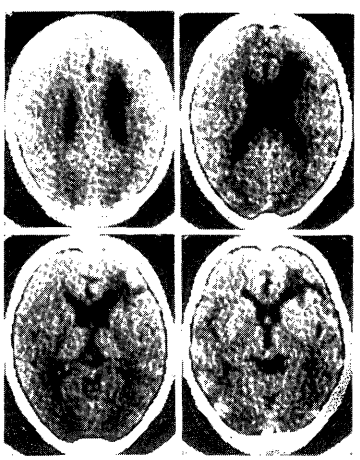
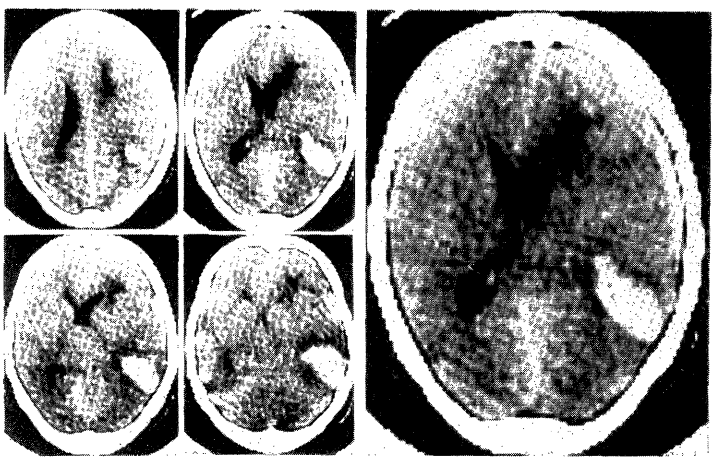


Fig. 4. Precontrast CT made on March 24, shows moderate ventricular dilatation with lucent foci in the right frontal region.



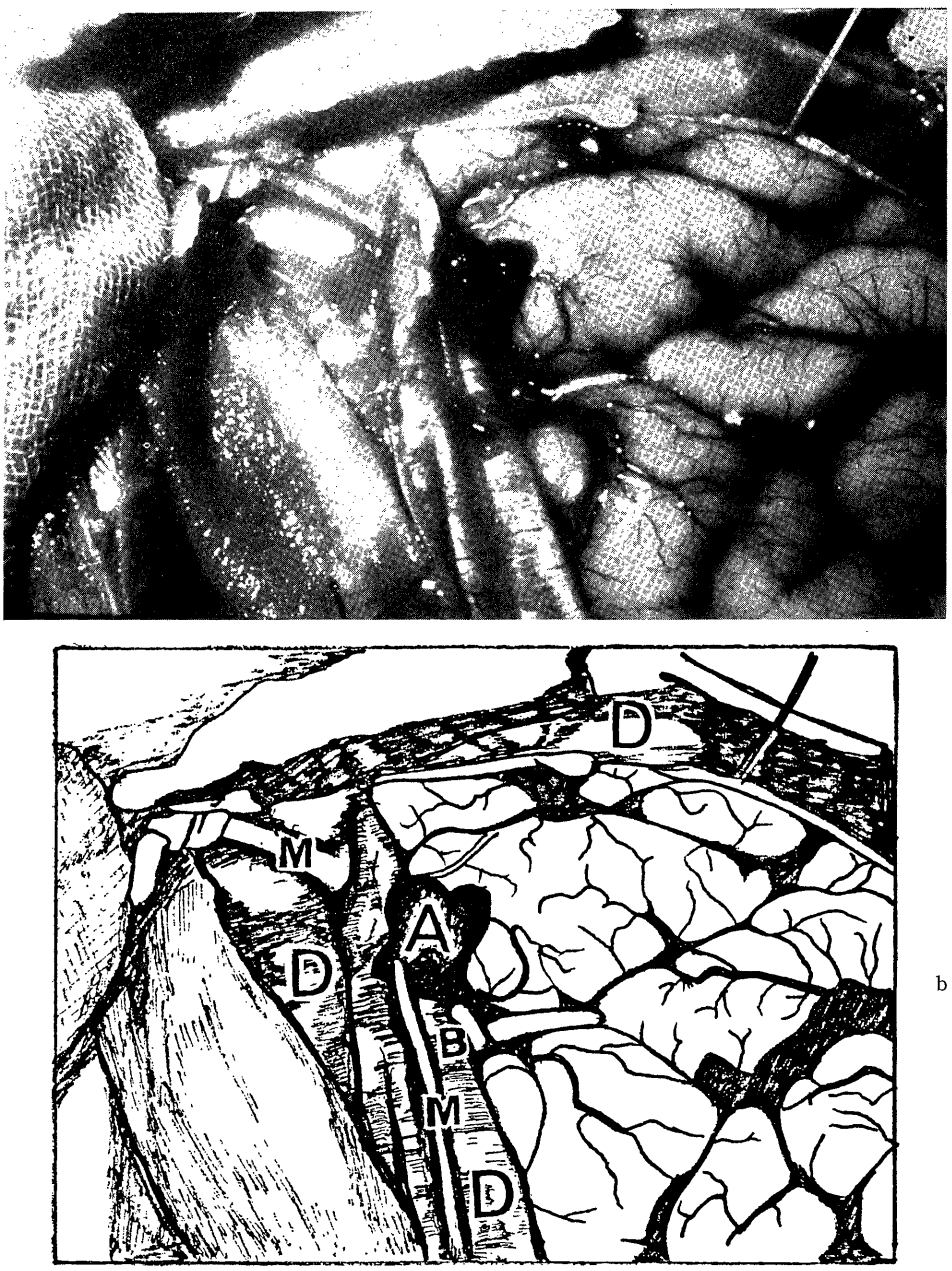
a b

Fig. 5. a anb b. Precontrast CT taken on Apr. 17, indicates an area of increased density with small peripheral rim of lucency suggesting an existence of intracerebral hematoma. An extracerebral lesion of blood density is also seen. Operation confirmed an acute subdural hematoma.

space due to the ruptured aneurysm of the transdural collateral channel.

OPERATION

After marking the right front temporal bone flap, the dural incision was made at an area surounded by the frontal and parietal asts of the middle meningeal artery which



a

b

Fig. 6. a and b. Operative photograph and sketch showing structures in photograph : ruptured aneurysm, A: meningocortical anastomotic vessel (bridging artery), B: dura mater, D: anterior branch of the right middle meningeal artery, M.

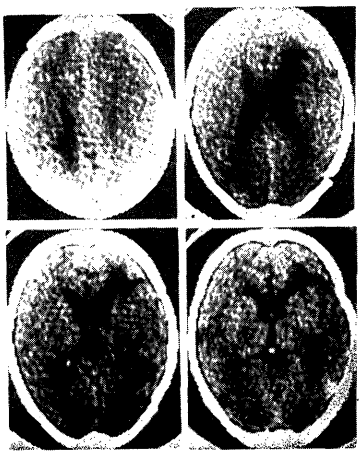


Fig. 7. CT about one month after operation.

had become elongated markedly to serve as collateral channels. Upon reflection of the dura, a collection of dark red blood was found to be overlying the cortex and removed by suction. It became clear that a connecting channel coming from the frontal ast of the middle meningeal artery formed a fusiform aneurysm of an indian been size at the point just it entered into the subdural space after penetrating the inner layer of the dura (Fig. 6). The aneurysm did not bleed spontaneously, but it began to bleed from a crack in its wall when the anastomotic channel was pulled tensely by such a way as to compress the brain with a supatula. The aneuysm was excised. The vascular spiders were found at the cortical surface, but no evidence was noted to indicate a leakage of intracerebral blood into the subarachnoid space. The intracerebral clot of about 30 ml was removed separately by making a small cortical incision. The detailed examination of the evacuated cavity wall could not be performed. Postoperatively, patient regained the consciousness. Although a follow-up CT taken about a month later showed remarkable improvement (Fig. 7), she is still a state of akinetic mute with moderate hemiparesis of her left limbs.

DISCUSSION

It is well know that moyamoya syndrome in adults is commonly manifested by the occurrence of the meningocerebral hemorrhage and the cause of the hemorrhage has been interpreted as direct rupture of moyamoya vessels mainly into the subarachnoid space. However, the bleeding source would not be so simple and it might be diveded into two main different ways according to whether the bleeding comes from the moyamoya vessels per se or due to rupture of collateral channels. In the former, intraparenchymal hematoma will occur in some cases, as it has been pointed out by Kodama and Suzuki [1] and the blood erupts into the ventricular cavity. In the later, rupture will result from high pressure on the vessel wall. In our case, the aneurysm was thought to be formed by overloading. It is not unlikely that rupture of the artery connecting between the meningens and the cortex (bridging artery) is a significant source of a subdural or a subarachnoid bleeding in cases with well developed transdural anastomosis. Slight distortion of the brain caused by various factors accelerate to ripping the preexisting thinwalled channels as in the case of Taveras [2] and ours. The present case is of special interest as it showed coexistence of two hematomas, intracerebral and subdural, with different bleeding sources.

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