Rupture of An Anterior Communicating Artery Aneursym and Concomitant Coarctation of The Aorta In A Child Follow Up of The Secondary Vasospasm By Transcranial Doppler

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Abstract : A 10 year-old child presenting with subarachnoid haemorrhage secondary to the rupture of an anterior communicating artery aneurysm and concomitant coarctation of the aorta followed up by periodic transcranial doppler is reported and literature on the subject is briefly reviewed.

Key Words : Aneurysm. Subarachnoid haemorrhage. Transcranial Doppler

INTRODUCTION

Subarachnoid haemorrhage (SAH) secondary to aneurysm rupture is unusual in children. The occurrence of intracranial aneurysm and coartation of the aorta has been reported, but to our knowledge no case followed up by transcranial doppler (TCD) has been reported. TCD provides useful information about the vasospasm which follows SAH and helps neurosurgeons to determine the optimal period for operative treatment.

CASE REPORT

A 10 year-old boy presented with headache of sudden onset on October 3, 1992. He was treated for an infectious pathology with analgesics and antibiotics as findings of the gross neurological examination were thought to be normal. On october 10, he presented with a seizure and became lethargic following an acute headache and was referred to the department of paediatrics. An examination he was found to be comatose with a stiff neck. There was no response to pain stimuli. Episodic chewing was noted. The systemic blood pressure was 160 mm Hg systolic and 120 mm Hg diastolic. Lumbar puncture was suggestive of SAH. A CT scan without and with contrast enhancement confirmed the presence of widespread subarachnoid blood in the basal cisterns and identified an area of enhancement in the region of the suprachiasmatic cistern (Fig 1).

He was admitted to the neurosurgery department and four hours later consciousness improved and he answered questions although drowsy without any long-tract motor or sensory signs or cranial nerve findings. Antihypertensive treatment with Glycerol trinitrate at a rate of 21 cc/h was begun. When episodically the systemic blood pressure reached 180 and 120 mm Hg for sytolic and diastolic values sublingual Nifedipine (10 mg) and diuretics (Furosemide 40 mg) were added.

TCD examination showed a beginning vasospasm in the left middle cerebral artery (MCA) area: at 50 mm depth mean velocity (Cm) was 110 cm/sec and pulsatility index (P.I.) 0.71 (Fig 2A). On the right side at 65 mm depth the Vm was over normal values at 78 cm/sec and especially the P.I. was high at 1.14 (Fig 2D).

On October 12, the systemic blood pressure was maintained at 150-160 mm Hg for sytolic and 100-110 mm Hg for diastolic values; the patient suffered severe headache and the TCD showed an increase in the left side vasospasm: at 55 mm depth Vm became 132 cm)

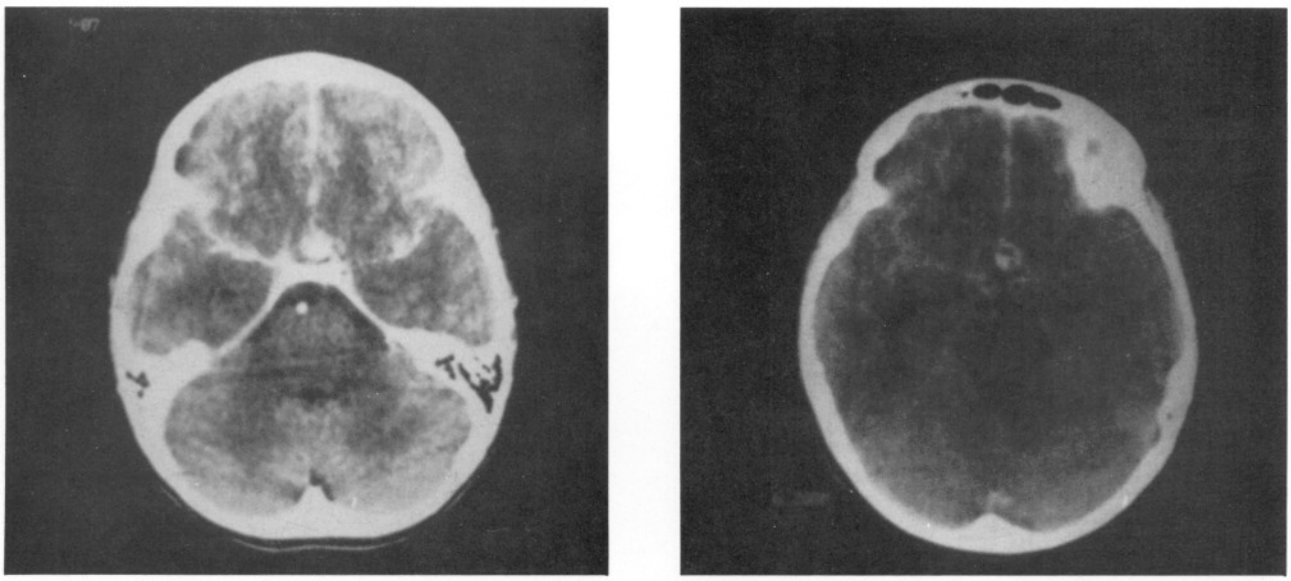

Fig. 1 : CT scan of the patient, with (A) without (B) contrast enhancement confirming the SAH and showing an enhancement in the region of the suprachiasmatic cistern.

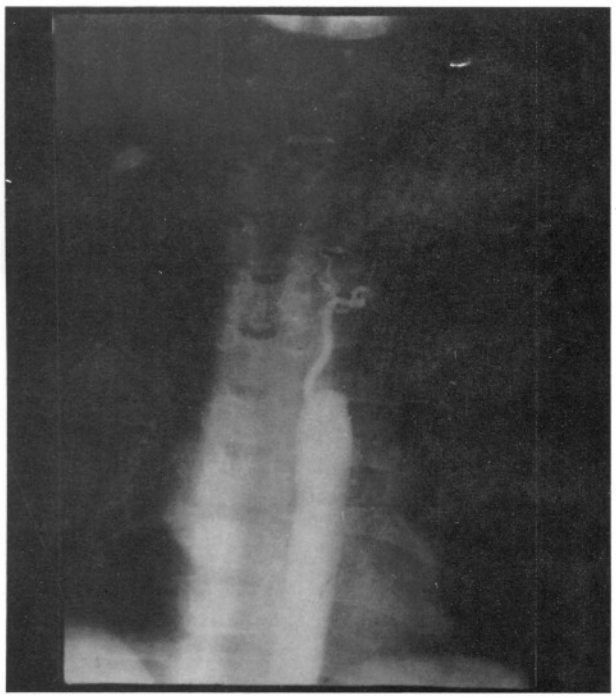

Fig. 3 : Aortography by femoral route showing coarctation of the aorta.

sec and P.I: 0.79. At this moment the right side began to be affected by the vasospasm: at 60 mm depth Vm was 92 cm/sec and P.I: 0.88 (Fig 2E).

Two days later a left side hemiparesis was noted, the reason was a right side vasospasm involving the M2 segment of the right MCA: at 45 mm depth Vm 52 cm/sec (P.I: 501), but at 55 mm depth 106 cm/sec (P.I. 0.56) (Fig 2F).

The right Al segment of the anterior cerebral artery was intact: at 70 mm depth reversed flow velocity was 46 cm/sec (P.I:0.67). At this moment the vasospasm of the left side MCA reached its peak level: at 65 mm depth Vm 174 cm/sec and P.I: 0.24 (Fig 2C).

The day after the patient became hemiplegic on the left side and developed a sixth nerve paresis on the right side. Consciousness was not altered. Systemic blood pressure again rose to 160-180 mm Hg systolic and 100-120 mm Hg diastolic. Glycerol trinitrate was substituted by Sodium nitroprusside at a rate of 5 cc/h and Nifedipine was administered twice a day.

In 5 days the left-sided hemiplegia gradually began to improve at the left upper limb which again became paretic. TCD revealed decreased velocity on both sides. On the left at 60 mm depth Vm was 118 cm/sec (P.I. 0.48).

On October 21, the patient underwent femoral angiography which had been postponed because of vasospasm. A coarctation of the aorta was discovered (Fig 3), but the intracranial arterial system could not be observed. Antihypertensive treatment was stopped as the systemic blood pressure became normal.

On October 28, headache and neck stiffness disappeared and the left side hemiparesis gradually recovered. A second angiography by cannulation of the left axillary artery was performed: postoroinferiorly-oriented a saccular aneurysm implanted at the junction of left A1, A2 and anterior communicating artery

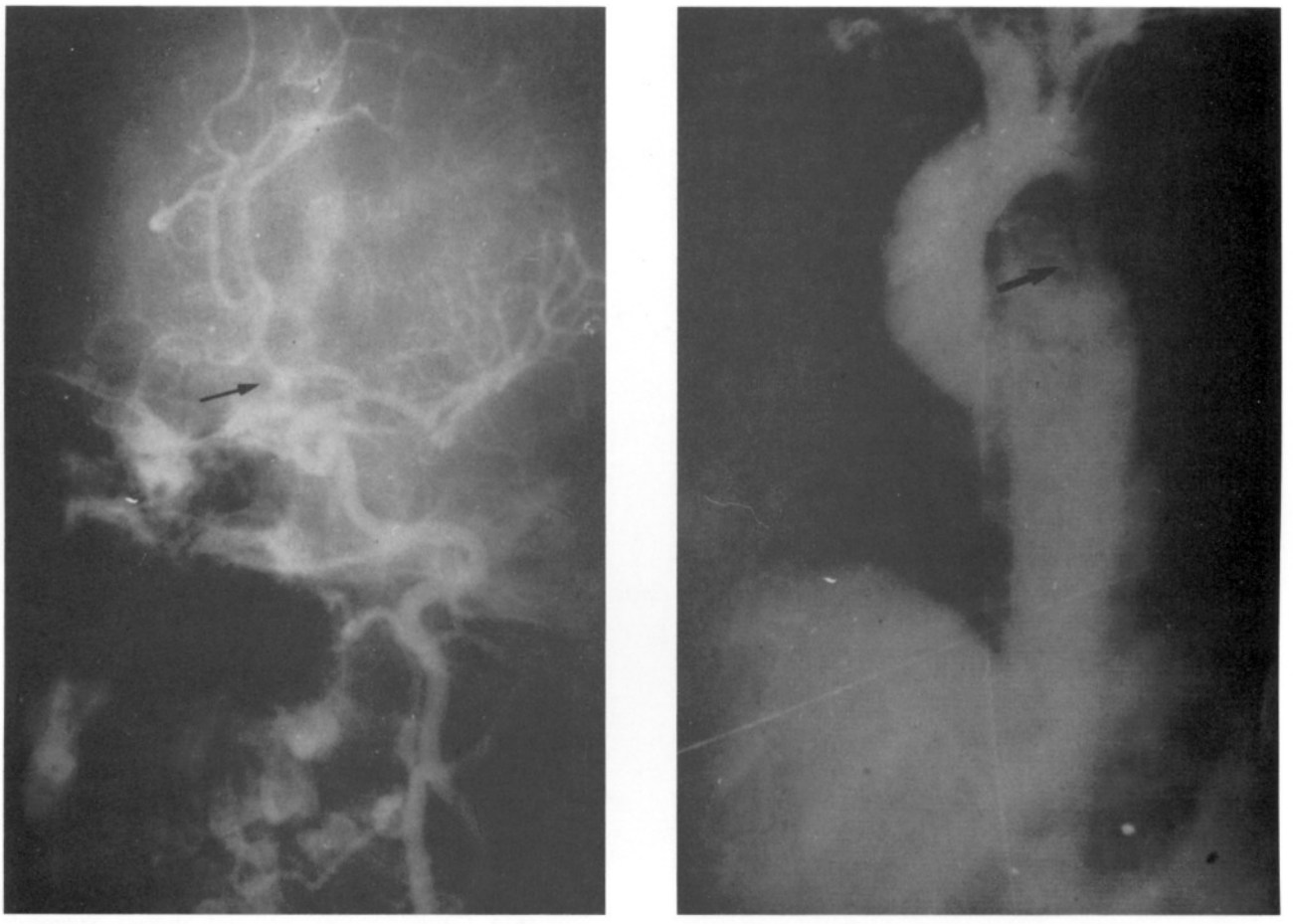

Fig. 4 : a) Cerebral angiography showing the aneurysm. b) Proximal portion of the arcus aorta.

was observed (Fig 4) and the proximal portion of the arcus aorta was visualisied. The ultimate preoperative check of the cerebral arteries by TCD was within normal limits.

A left pterional craniotomy was performed and a Yaşargil clip was placed at the neck of the aneurysm which was opened and severed. The postoperative course was uneventful, the patient was discharged on the 9th postoperative day with a prophylactic antiepileptic prescription.

Two months later the right sixth nerve paresis had completely recovered. There was no headache nor any sign of left hemiparesis. He was in excellent physical condition. He was referred to the Cardiovascular Surgery and Research Hospital of Haydarpaşa where a coarctectomy and an end-to-end anastomosis of the aorta was performed. He died two days later following profuse bleeding in the thorax due to loosening of the anastomosis.

DISCUSSION

Eppinger in 1871 (8, 9) first reported rupture of an anterior cerebral artery aneurysm in a thirteen year-old boy who also showed stenosis of the isthmus of the aorta at autopsy.

Intracranial aneurysms in the paediatric age group are rare neurosurgical lesions, occurring at a frequency of approximately 0.5 % to 4.6% in large aneurysm series (15).

In the Cooperative Study only 2.5 percent of aneurysms occurred in patients under 20 years of age (41 aneurysms in 6343 patients) (10). In Yaşargil's series (21) only 4 out of 1012 operated cases were under or 10 years of age (0.4%) and none had clinically obvious concomitant disease.

There was a number of clinical series and isolated case reports permitting a review of some 300 patients with aneurysms presenting as diagnostic and

therapeutic problems during the first two decades of life and only 15% of aneurysms occurring in the paediatric population were seen in the first decade of life (19).

Richardson (16) found 74 cases of childhood aneurysms in 3900 cases (1.9%); 7 patients had coarctation of the aorta and 2 polycystic kidney disease. The ovarall incidence of hypertension was only 15 percent. Vascular spasm was only seen at angiography in 9/74 (12%) of cases. In a clinic where more than 700 patients undergo surgery for coarctation of the aorta, Matson encountered only 15 children with intracranial aneurysms and only 3 of them had coarctation of the aorta (14).

A current theme in the history of those patients who presented initially with SAH was the general failure of the attending physician to consider an aneurysm in the initial differential diagnosis, leading to a second and more severe or fatal bleeding (15, 19). First developed by Aaslid in 1982 (1), TCD has been widely used in our department since 1991 in the detection and follow-up of intracranial haemodynmic phenomena because it is atraumatic, practical and repeatable. The accuracy of interpretation of TCD data depends upon the knowledge, skill and experience of the technician performing the procedure (6).

Nevertheless the utility of TCD in detecting and quantifying vasospasm after SAH has been extensively studied and the results have been correlated with angiographic findings. In patients with moderate to large volume SAH, flow velocities in basal cerebral arteries increase in the 3 rd to 10 th day of bleeding and reach a maximum between days 11 and 20 paralleling the development of aniographic vasospasm (6). Time averaged mean flow velocities in the MCA over 90 cm per second predict narrowing on angiography (5). Velocities are often asymmetrically increased on the side of the ruptured aneurysm and the degree of change correlates roughly with the location, volume and thickness of clot visible on CT. TCD is a noninvasive method for following patients with SAH for the development of vasospasm, perhaps allowing prevention of delayed ischaemic deficits (2, 3, 4, 6, 7, 11, 12, 13, 17, 18, 20).

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