Aortic Coartation Associated With Multiple Cerebral Aneurysms

ERDENER TİMURKAYNAK, FERRUH GEZEN, YUSUF ERŞAHİN, NACİ SEBER

Department of Neurosurgery, Gülhane Medical School, Ankara, Türkiye

Abstract : A/21-year-old man presented with headache, left hemiparesis and hypertension. Xantochromic cerebrospinal fluid was obtained by lumbar puncture. Aortic coarctation and two aneurysms in the bifurcation of the left internal carotid artery and one in the bifurcation of the right internal carotid artery were detected on angiograms. The patients was operated and three cerebral aneurysms were clipped at the one-stage operation. Coarctation was repaired 5 months later. Literature regarding this combination of anomalies, which is very rare, is reviewed.

Key Words : Subarachnoid haemorrhage; Coarctation of the Aorta; Cerebral aneurysms.

INTRODUCTION

Eppinger first described the association of aortic coarctation(AC) and cerebral aneurysms in 1871 (2). Waltman and Shelden (1927), Abbott (1928), Lichtenberg and Gallaher (1933) and Reifenstein et al. (1947) reported fatal cases of AC in which deaths were due to rupture of cerebral aneurysms or intracerebral haemorrhage (1,5,7). The outcome of surgical treatment of these patients with this combination of anomalies had previously been fatal. Recent advances such as safer anaesthesia, microneurosurgical techniques and modern thoracic surgery have improved the prognosis. We present a patient with three cerebral aneurysms associated with AC who was successfully treated.

CASE REPORT

The patient was a 21-year-old male admitted to the department of internal medicine for the treatment of hypertension. He had been complaining of headache for two weeks. He was transferred to our department after developing left-sided weakness. On physical examination, he was alert but slightly apathetic. Blood pressure in the upper limbs was 170/100 mmHg, the femoral pulses were weak. Neurological examination revealed slight nuchal rigidity and mild left hemiparesis and left hemihypoaesthesia. CT scan was unremarkable. Xantochromic cerebrospinal fluid was obtained by lumbar puncture. Transfemoral digital subtraction angiography (DSA) was not successful because of AC. Bilateral percutaneous carotid angiograms displayed two aneurysms in the bifurcation of the left internal carotid artery (ICA) and one in the bifurcation of the right (Fig. 1). Bilateral fronto-temporal craniotomies were done and three aneurysms were clipped at the one-stage operation three days after the patient had been transferred to our department (Fig. 2). The postoperative course was uneventful. He was discharged from hospital without neurological deficit on the tenth postoperative day. The AC was repaired in the department of cardiovascular surgery five months later (Fig. 3). The patient is still alive and neurologically intact four years after surgery.

DISCUSSION

Adult AC is found once in every 3000 or 4000 autopsies; the incidence is exceedingly low in

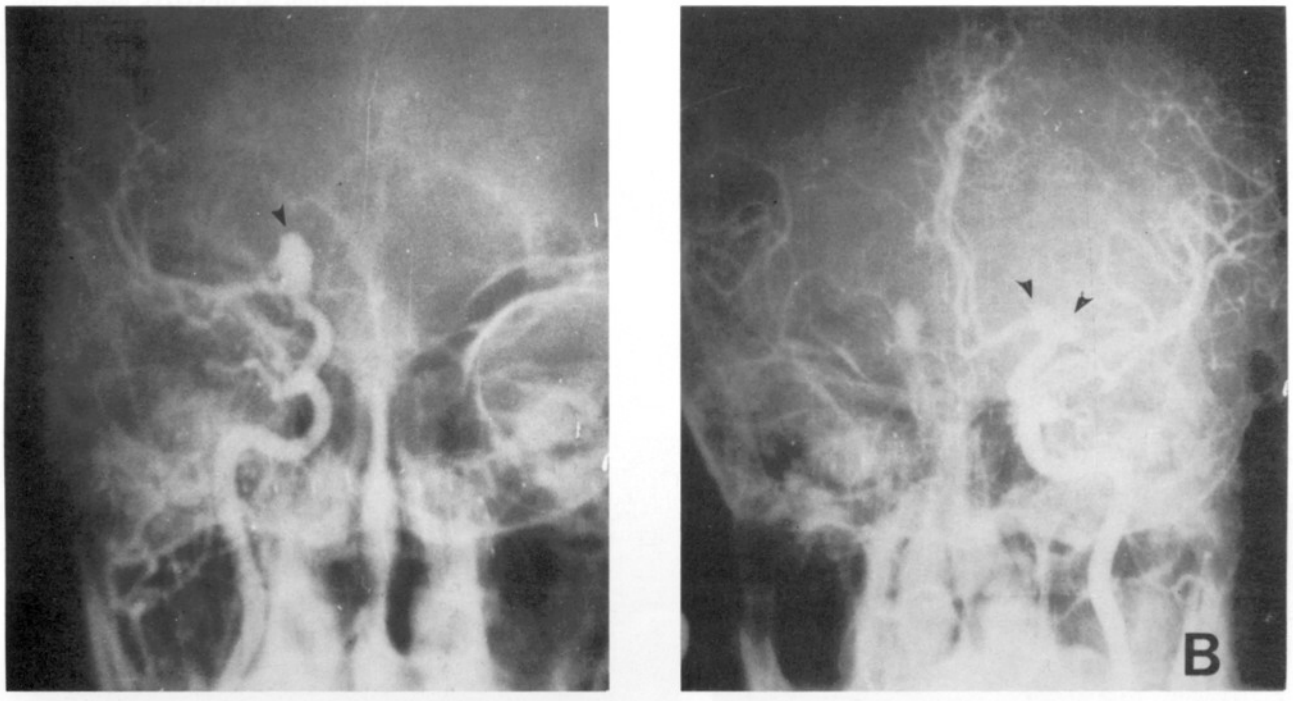

*Fig. 1 : Percutaneous carotid angiograms showing (A) an aneurysm **(arrow head)** in the right ICA bifurcation and (B) two aneurysms **(arrow heads)** in the left ICA bifurcation.*

Table 1(*) : Multiple Cerebral Aneurysm Cases Associated With Ac

CASE	DATA	AUTHORS	ANEURYSMS	SIDE	AGE	SEX	SURGERY	RESULT
1	1871	EPPINGER	ACeAA	RIGHT	17	MALE	–	DEATH
			ACeAA	LEFT				
2	1926	PARKER	pcaIICAA	LEFT	20	MALE	–	DEATH
			ACoAA	MIDLINE				
3	1927	WOLTAMIN	pcaICAA	LEFT	20	MALE	–	DEATH
		SHELDEN	ACoAA	MIDLINE				
4	1928	GREEN	ACeAA	RIGHT	21	MALE	–	DEATH
			ACEAA	RIGHT				
			scaBAA	(?)				
5	1943	O'REILLY	achaICAA	LEFT	13	MALE	–	DEATH
		CHAPMAN	ACoAA	MIDLINE				
6	1947	DABSS	bICAA	RIGHT	(?)	MALE	–	DEATH
		DESFORGER	AIAA	RIGHT				
			AIAA	LEFT				
			ACoAA	MIDLINE				
7	1954	KING	MCAA	RIGHT	27	MALE	–	DEATH
		et al.	MCAA	LEFT				
8	1959	HIRANO	pcaIICAA	RIGHT	11	FEMALE	–	DEATH
		et al.	bICAA	LEFT				
9	1960	SCHWARTS	bICAA	LEFT	14	MALE	–	DEATH
		BARONOFSKY	PCeAA	(?)				
10	1962	afBJRKESTERN	ACeAA	RIGHT	26	MALE	–	GOOD
		TROUPP	ACeAA	LEFT				
11	1967	ROBINSON	bICAA	LEFT	19	MALE	–	DEATH
			ACEAA	MIDLINE				
12	1969	ISHERWOOD	bCAA	LEFT	37	FEMALE	CLP	(?)
		DULTON	MCAA	RIGHT				
13	1974	SCHWARTS	MCAA	RIGHT	16	MALE	CLP	GOOD
		SCHARFETTER	MCAA	RIGHT			SYN	
14	1992	TİMURKAYNAK	bICAA	RIGHT	21	MALE	CLP	GOOD
		et al.	bICAA	LEFT			CLP	
			bICAA	LEFT			CLP	

ACeAA: anterior cerebral artery aneurysm, pcaICAA: Internal carotid aneurysm at posterior communicating artery take off. ACoAA0 Anterior communicating artery aneurysm, scaBAA: Basilar artery aneurysm at superior cerebellar artery take off, achaICAA: Internal carotid artery aneurysm at anterior choroidal artery take off, bICAA: Internal carotid artery aneurysm at bifurcation, AIAA: Aneurysm of Al segment of anteror cerebral artery, MCAA: Middle cerebral aneurysm, PCeAA: Posterior cerebral artery aneurysm, SYN: Synthetic wrapping. (?) : Insufficient information. (*) : Data in reference 3 was used in the preparation of this table.

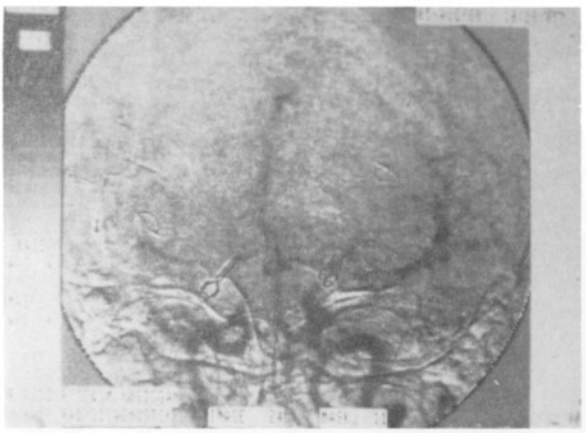

Fig. 2 : Postoperative IV. DSA.

should be kept in mind that an infective aneurysm might lead to intracranial or subarachnoid haemorrhage.

Cerebral aneurysm should be operated in a patient with subarachnoid haemorrhage before repair of the coarctation, since a ruptured aneurysm is a major threat to life.

This paper was presented in part at the meeting of the 9th Congress Neurosurgery 23-28 June 1991, Moscow, USSR

Correspondence : Dr. Erdener Timurkaynak
Department of Neurosurgery
Gülhane Medical School
Etlik, Ankara 06018

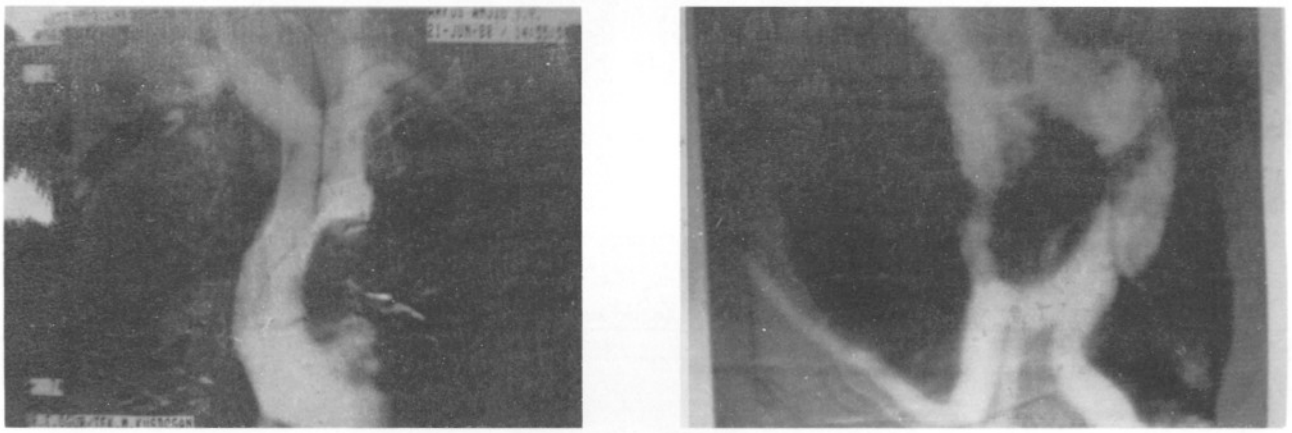

Fig. 3 : Aortic angiograms showing aortic coarctation (A) before the repair, (B) after grafting.

patients with cerebral aneurysms (6). Fox gleaned 59 cases from the literature from about 5000 case reports of aneurysm incluiding 13 multiple cerebral aneurysms (Table I) two of which were treated surgically in which there was an association with AC (1.2%) (3). Fukuda et al. reported a case of anterior communicating artery aneurysm associated with AC out of 154 aneurysm cases (0.65%) (4). To our knowledge, ours is the 61st case which has this combination of anomalies. The location of aneurysms associated with AC is similar to that of intracranial aneurysms in general; 79% in the anterior and 21% in the posterior circulation. In 61 cases, 47 patients were male (77%) and 14 female (23%) (3,4). The multiplicity of aneurysms of patients with AC 30% have multiple aneurysms. Ten to 12% of patients with AC die of intracranial haemorrhage. The relationship between cerebral aneurysm and AC may be one of high blood pressure and blood flow patterns stressing intracranial arteries. Bacterial endocarditis is a frequent complication of AC, and although there has been no instance of bacterial aneurysm to date, it

REFERENCES

1. Abbott ME. Coarctation of the aorta of adult type II. A statistical study and historical retrospect of 200 recorded cases with autopsy of stenosis or obliteration of the descending arch in subjects above the age of 2 years. Am Heart J 3: 574-618, 1928.
2. Eppinger H. Stenosis aortae congenita seu isthmus persistans. Vjschr Parkt Heilk 112: 31-67; 1871.
3. Fox JL. Inracranial aneurysms. New York, Spirgner-Verlag, 1983 vol 3.
4. Fukuda H, Sako K, Yonemasu Y. Coarctation of descending aorta with aneurysm of anterior communicating artery. Surg Neurol 1985.
5. Lichtenberg HH, Gallager H. Coarctation of the aorta, anomaly of the great vessels of the neck and intermittent leakage of cerebral aneurysms, diagnosed during life. Am J Dis Child 1933; 45; 1253.
6. Reifenstein H, Levine SA, Gross RE . Coarctation of the aorta. A review of 104 autopsied cases of adult type, 2 years of age or older. Am Heart J, 33: 146-168; 1947.
7. Woltman HW, Shelden WD. Neurologic complications associated with congenital stenosis of the isthmus of the aorta: A case of cerebral aneurysms with rupture and a case of intermittent lameness presumably related to stenosis of the isthmus. Arch Neurol Psychiatry 17: 303-316; 1927.