

Bilateral spontaneous dissection of extracranial vertebral arteries

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Summary. A previously healthy 35-year-old man suddenly developed vertebrobasilar ischaemia while playing tennis. Cerebral arteriography revealed a dissecting aneurysm of the cervical portion of both vertebral arteries. The neurological deficit and the angiographic features resolved with anti-coagulant treatment alone. Only 15 patients with non-traumatic extracranial vertebral artery dissection have been previously reported. It is necessary to recognize the condition very quickly, because the authors consider that anticoagulation is required as an emergency.

Key words: Vertebral artery – Vertebrobasilar ischaemia – Dissecting aneurysm

Introduction

Reports of spontaneous dissection of the internal carotid artery appear regularly in the medical literature, sometimes in large series, such as 36 cases of Mokri et al. [12] and 14 cases of Bogousslavsky et al. [3]. However, only 15 patients with spontaneous dissections limited to the extracranial part of the vertebral artery have been reported in the English, French and German literature: five patients by Caplan et al. [6], two by Bladin [2] and eight in single case reports [1, 4, 5, 9, 11, 16, 17]. They are quite different from intracranial vertebral artery dissections, which may be revealed by subarachnoid haemorrhage [15] as well as posterior fossa ischaemia [7, 8], and are not further discussed in this paper.

We report a 16th case, which presented some particular features: dissection was exclusively extracranial, was bilateral, occurred without apparent trauma, and partially resolved with anticoagulation.

Case report

This previously healthy 35-year-old man was referred on 10 June 1984 because of the sudden onset of vertigo, diplopia, right limb weakness and severe pain in the occiput and neck while making a passing shot in a game of tennis. He recalled no obvious trauma or sudden rotation of his head. On admission, blood pressure was normal, as were the findings of a general examination; neurological abnormalities included, on the right, limb weakness with Babinski's sign, and, on the left,

limb dysmetria with hypotonia, sixth cranial nerve involvement, meiosis, facial global hypoesthesia and coarse horizontal nystagmus. CT scan was normal. Cerebellar blood flow, measured with xenon-133, revealed decreased values in both cerebellar hemispheres. Bilateral vertebral angiography showed marked narrowing and irregular stenosis of the third segment of the left extracranial vertebral artery (Fig. 1a, b) and mild stenosis of the third portion of the right extracranial vertebral artery with a double barrel lumen in its fourth portion, just before entering the foramen magnum (Fig. 1c); bilateral carotid angiography was normal. Heparin anticoagulation was begun and clotting time was increased to 2 times normal.

Two months later clinical examination revealed only a persistent left meiosis with right hypoesthesia to temperature and pain which had appeared a few days after the onset; heparin was replaced by acetylsalicylic acid (500 mg daily).

In April 1986, the patient was normally active, and the results of clinical examination were unchanged. Bilateral vertebral angiograms revealed a dramatic improvement (Fig. 2): on the right side, the stenotic segment was no longer present, but there was a slight residual abrupt narrowing of the end of the third portion; the left vertebral artery was almost normal; on both sides, movement of the head did not cause any change in the arterial lumen.

This man had no history of previous illness, in particular diabetes, hypertension, or elastic tissue disease; he often played tennis and had even performed more than 200 parachute jumps up to 1982. He subsequently remembered that, 15 days before the onset of his symptoms, he had experienced sudden severe pain in his neck, when weight-lifting; the pain lasted for a few minutes.

Discussion

Diagnosis with certainty of vertebral artery dissection requires pathological evidence of an intramural clot separating the media and compromising the lumen [8]. In spite of the absence of pathological studies, the following features favour the diagnosis according to Caplan et al. [6]: (a) the prominence of pain, (b) the absence of obvious vascular disease, (c) the location of the vertebral lesions, posterior to the atlas, (d) the close angiographic resemblance to cases of traumatic dissection and (e) the improvement of the lesions in a second angiogram. Radiographic features in our case agreed with those previously described in extracranial vertebral artery dissection: the luminal stenosis is usually more irregular [10] and associated with the "string sign" or the "string of beads" configuration [10]; a re-

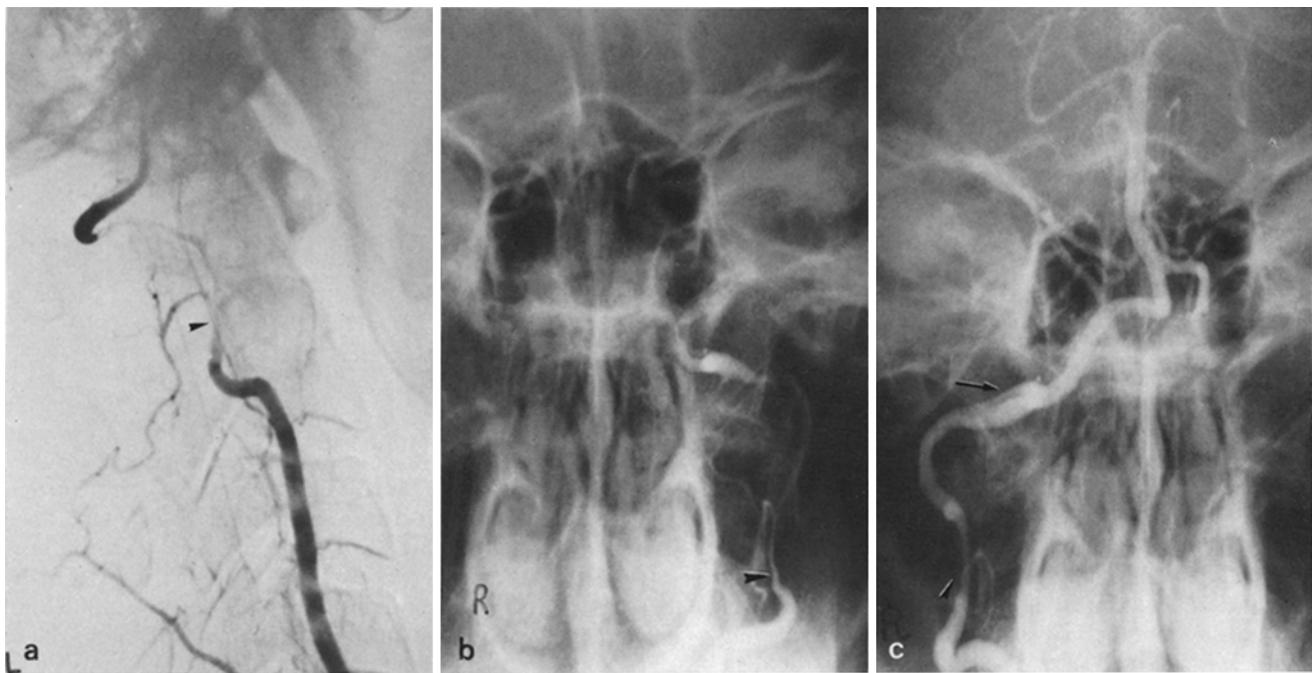


Fig. 1a–c. Vertebral angiography (June 1984). **a, b** Marked narrowing and irregular stenosis of the third portion of the left vertebral artery (►). **c** Mild stenosis (►) of the third portion of the right vertebral artery with double barrel lumen in its fourth portion (→)

constitution of the lumen has been described, as was seen on our second angiogram; our patient had marked narrowing of the lumen on both sides, but no intimal flap [5] or pseudoaneurysmal dilatation [12]. The double lumen sign, observed in his right vertebral artery, has been demonstrated rarely at the vertebral level, but is more frequent in internal carotid artery dissection [12]; it means that the dissection plane has re-established a direct communication with the true lumen distally, allowing blood and contrast to flow in both pathways simultaneously [12]; in our patient, a bilateral slow vertebral artery flow was seen, as has been previously reported [16], but there was no sign of distal embolism.

Extracranial vertebral artery dissection must be differentiated from false aneurysm without narrowing of the lumen [14] and from occlusion without dissection, which may follow abrupt changes in head position [13]. These occlusions are usually caused by a prolonged hyperextension and rotation of the head [13, 19], while traumatic dissections are more frequently caused by a sudden and brief rotation or extension of the head [17]. Our patient did not remember any abrupt head movement, but the possibility that such a movement may be made while playing tennis [17] or weight-lifting cannot be excluded, neither can the role of trivial trauma [12]. In reports on 15 other patients, the dissection occurred after trauma [12] or abrupt rotations of the head. In accord with the view of Caplan et al. [6] our patient's dissection has been termed "spontaneous" because no definite trauma was recalled. The same clinical and radiological features have been observed with both traumatic and spontaneous dissections [6].

The present and previous reports of spontaneous dissection of extracranial vertebral arteries include 16 patients, 9 women and 7 men, whose ages ranged from 15 to 56 years (average 35.25 years); our patient and 10 others had no medical history, but migraine [2, 5], oral contraceptive [6, 11] and fibromuscular dysplasia [18] have been associated with extra-

cranial vertebral artery dissection. In 3 reports [6, 9; present case], symptoms began during sports activities (tennis, softball) and in 1 case [6] while driving; in others it was possibly related to a special neck position during surgery [4] or resuscitation [6]. In our patient, it was possibly due to an exercise performed with expiration while the glottis is closed, as been reported previously for carotid dissections [20]. The neurological deficits developed rapidly in 13 of these 16 patients, but appeared to evolve gradually in the others [1, 6]. Pain was a prominent and early finding in our case and in 10 of the previous reports and was located in the occiput and posterior neck. Our patient showed signs of medullary infarct: in the literature, 11 have had features of brain-stem ischaemia, 8 in the medulla [2, 6, 9] and 3 in the pons [5, 7, 10]; 3 patients with associated internal carotid artery dissection had only features of hemisphere infarct [1, 11, 16] and 1 patient died suddenly without having experienced any neurological signs [4]. Vertebral artery dissection affected the right side in 2 of the 16 patients, the left side in 6 and both sides in 8 patients. The lesions usually began in the third segment of the artery at its exit portion from the transverse foramen of the axis [1, 2, 4–6, 11, 16] and commonly extended to the horizontal portion of this segment, as it travels over the posterior arch of the atlas. In 6 cases the lesion began at the level of C6, where it enters into the foramen of C6 [1, 2, 6, 10, 16, 17]; the first and third portions of the vertebral artery are movable, whereas the second and fourth segments are fixed by bony surroundings. This anatomy probably explains the predilection for the first and third portion for dissection as well as in traumatic lesions [13]. Moreover, according to Wilkinson [21], there are fewer elastic fibres in the wall of the third portion. In 24 dissected extracranial vertebral arteries from 15 patients reported in the literature, including our own, 20 were affected by an irregular stenosis and 4 by total occlusion. During 2 years' follow-up our patient did not develop new neurological symptoms after

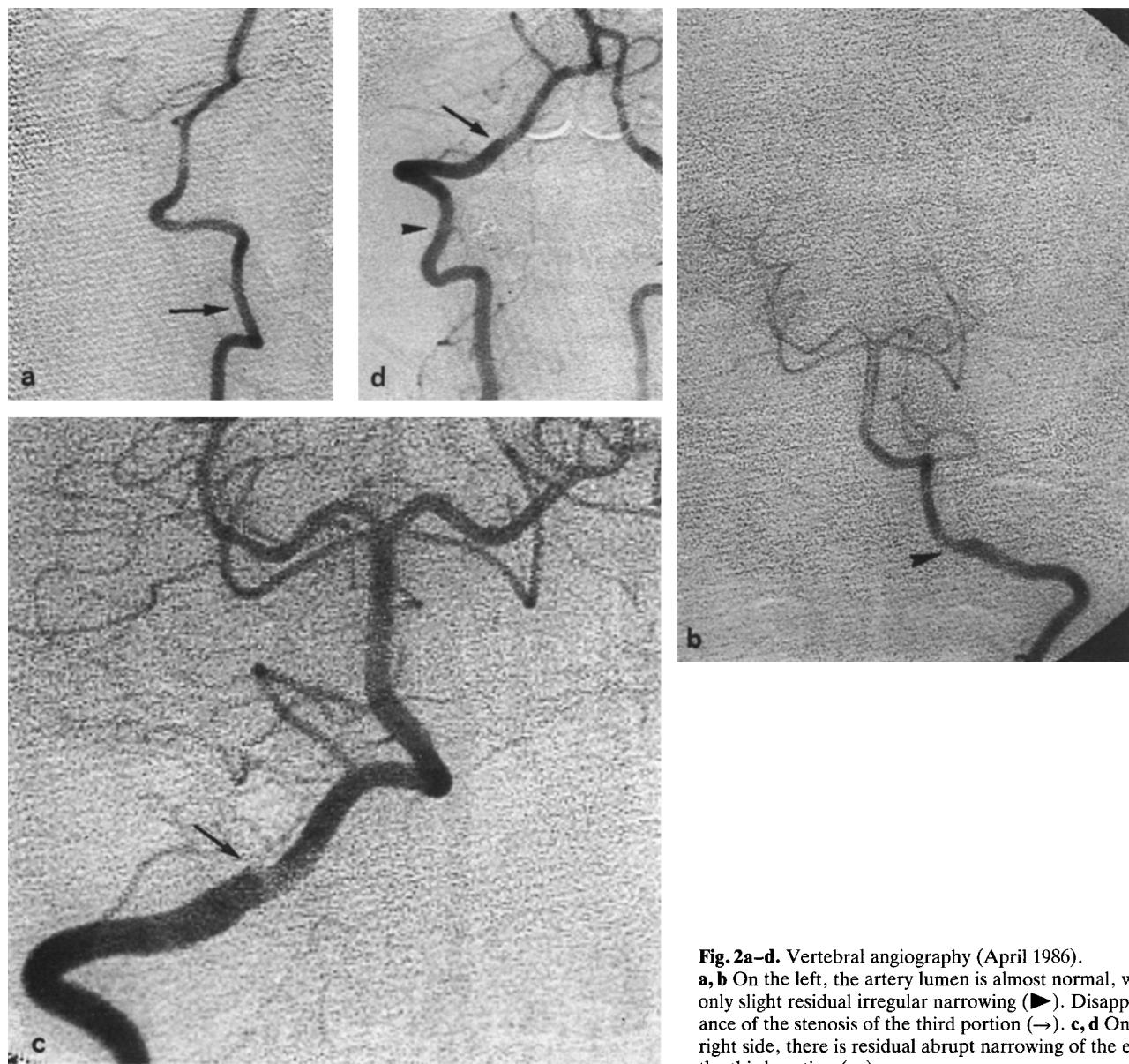


Fig. 2a-d. Vertebral angiography (April 1986). **a, b** On the left, the artery lumen is almost normal, with only slight residual irregular narrowing (►). Disappearance of the stenosis of the third portion (→). **c, d** On the right side, there is residual abrupt narrowing of the end of the third portion (→)

his original stroke; in most previous reports, signs have resolved satisfactorily [5, 6, 10, 11, 17] and only 2 patients died [4, 16]. Only a few cases, therefore, had confirmation of dissection by necropsy [4, 6] or surgery [6], but repeated angiographies showed definite improvement in most cases [5, 6, 10, 11].

Our patient was treated with heparin, because at the site of intimal and endothelial disruption platelet deposits can form in the lumen and reduced flow can potentiate the development of an intraluminal clot [12]. Three patients previously treated in this way had a total recovery [6, 10]. One patient [6] needed surgery because of deterioration and recovered too. In 11 cases in which the treatment was not clearly reported, 2 patients recovered [12, 19], 4 had minor sequelae [1, 5, 6], 2 died [6, 18], and in 3 the outcome was not stated [4, 9].

In conclusion, extracranial vertebral artery dissection should be especially considered in young patients with painful vertebrobasilar stroke occurring during exercise. The clinical and radiological features are less well known than those of

carotid artery dissection. However, an early diagnosis is necessary because we consider that anticoagulant treatment should be started at once to avoid the development of an intraluminal clot and distal embolization.

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