Traumatic Extracranial Vertebral Artery Aneurysm

A Case Report and Review of Literature

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SUMMARY:

A young female patient was referred to our Department of Vascular Surgery with a pulsatile cervico-occipital lump with neurological symptoms of three years duration. She underwent routine investigations followed by selective four vessel angiography which confirmed the presence of a very large sized Extracranial Vertebral Artery Aneurysm, Retrograde flow via the opposite Vertebral Artery was demonstrated. An aneurysmectomy was performed after proximal ligation of Vertebral Artery at source. The existing literature was reviewed and updated.

CASE REPORT:

A 25-year old lady (IB) was transferred from a District teaching General Hospital with a history of swelling in the cervico-occipital region on the left side, of three years duration. Having been previously asymptomatic, she had developed intermittent difficulty with swallowing and ataxia. The neck lump had also become painful in the preceding six months. The transient attacks of dysarthria and ataxia (lasting 1-2 hours) were occurring 2-3 times every day, at the time of her presentation. This had resulted in severe distress to the patient with nutritional disorders and recurrent respiratory infections. No disturbances of vision were reported and there was no history of loss of consciousness, given. Although she denied injury, the relatives were sure that she had indeed developed minor trauma to the neck prior to the onset of the lump three years earlier. Physical Examination revealed her to measure 5' 2" in height, 48 Kg in weight. Her pulse was regular at 82/min and the supine arm BP was 115/70 mm Hg. She was apyrexial. She was patchily disorientated in events but not in space or time. At the time of the examination she had no dysphasia but severe dysarthria, alexia, agraphia, dyscalculia, finger agnosia and right-left disorientation (Gerstmann's Syndrome). Cranial Nerves, Sensory and Motor systems were normal. No nystagmus was detected. Ankle clonus was present. Her reflexes were normal. Pupils were 3mm in diameter, equal and brisk. No meningeal signs were found. In the neck a 15 cm diameter, firm, circular, tender and pulsatile lump was present, extending from the left mastoid region to the cervico-occipital margin and across the midline well into the right side, (Figure 1). It had restricted extension of Cervical spine and attempts at extension caused increased pains in the lump. It was tense and a local bruit was audible although no radiation was detected in either Carotid. Doppler examination revealed this to be an arterial aneurysm which did no collapse on Carotid pressure. A small cavity surrounded by a much larger mass was detected.

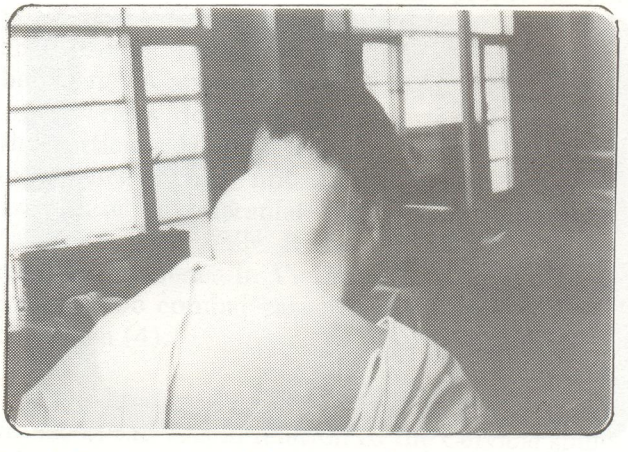

Fig. 1 Cervico-occipital left vertebral aneurysm

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INVESTIGATIONS:

Haemoglobin = 11g/dl
Bl, Urea = 29mg/dl
Urinalysis = Normal
Skull X-rays = Normal

ESR = 18mm 1st hr
KT = Negative
ECG = Normal
Cervical Spine X-rays = Normal

WCC = 5.2/mm^3
Sugar = 98mg/dl (random)
Chest X-ray = Normal

Arteriography, at South World NICVD Karachi via the femoral route showed the following features:

The right Carotid injection (Figure 2) showed normal Common Carotid Artery with a normal bifurcation. The Internal Carotid Artery was

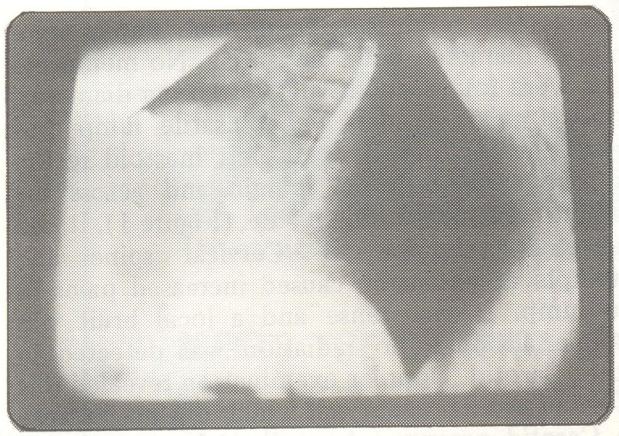

Fig. 2 (Right Carotid angiogram)

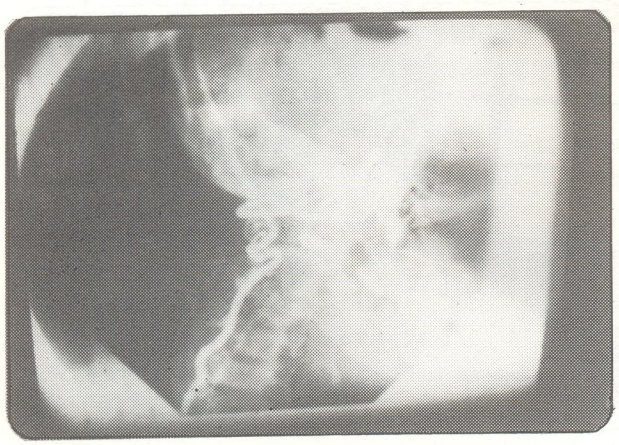

Fig. 3 (Right Vertebral angiogram showing refluxing left VA.

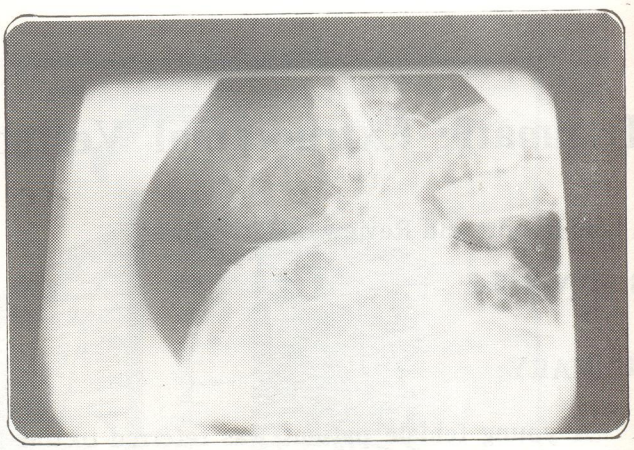

Fig. 4 (Left Carotid angiogram

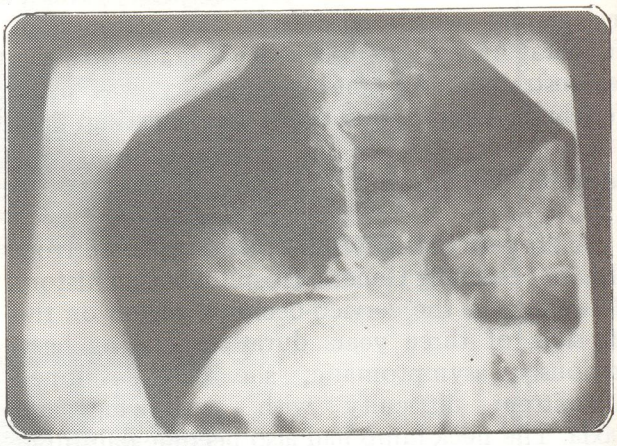

Fig. 5 (Left Vertebral angiogram and aneurysm)

normal and dye flowed across the midline. The right Vertebral Artery injection (Figure 3) showed the extracranial course of the Vertebral Artery to be normal. No aneurysm or stenosis was seen. The Basilar Artery was normal and its branches were outlined well, without any abnormalities but a quick reflux took place into the limb of the Left Vertebral Artery. The left Common Carotid injection (Figure 4) revealed a large Occipital arterial branch of the External Carotid Artery encircling the soft tissue mass of the neck vaguely outlined by the preliminary arch angiography. The rest of the Left Carotid tree was normal and a cross flow was again demonstrated. The left Vertebral Artery injection (Figure 5) revealed a normal course of the extracranial artery upto the C1 level. At this point the Artery deviated posterolaterally and through a small ostium discharged into a large Cavernous multiloculated Aneurysm. No additional outlets of the aneurysm were demonstrated. A careful review

of the Right Vertebral injection revealed spillover of blood via the reflux from the Left Vertebral Artery into the aneurysm. This was therefore a Left Vertebral Artery Aneurysm which was being bilaterally fed and which was causing her multiple posterior fossa TIAs.

OPERATION:

Through a supraclavicular approach the Left Vertebral Artery was dissected and was occluded with a soft clamp. The pulsation of the lump disppeared but there was no appreciable reduction in size. Digital compression did not collapse the aneurysm. The artery was ligated. With the patient lying now on her right side, a curvilinear incision was given at the cervico-occipital junction. Adipose tissue was separated and underneath the deep muscular plane a very large sized multi-oculated aneurysm was identified. The aneurysm was dissected free with considerable difficulty. Its outer layer was densely adherent to the superficial tissues. During the dissection a massive bleeding took place resulting in the loss of 0.5 litres in 1-2 minutes. The injury was repaired with Prolene under digital compression. The friability at this point, precluded any other method of haemostasis. It was then found that the internal cavity measured 5cm in diameter but the rest of the lump which consisted of thick adherent thrombosed multi-loculi, measured 15 cm x 8 cm. (Figure 6 and 7). The part of the aneurysm closest to the ostium was very friable and thin walled, confirming that the lesion was an acquired type of aneurysm. The feeding principal vessel was eventually isolated and ligated. The aneurysm was then removed. Because of the excessive friability, vein graft reconstruction was not considered safe. With

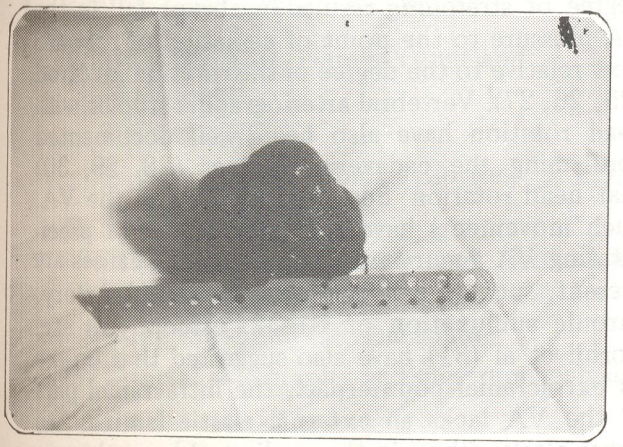

Fig. 6 'Excised aneurysm with its ostium'

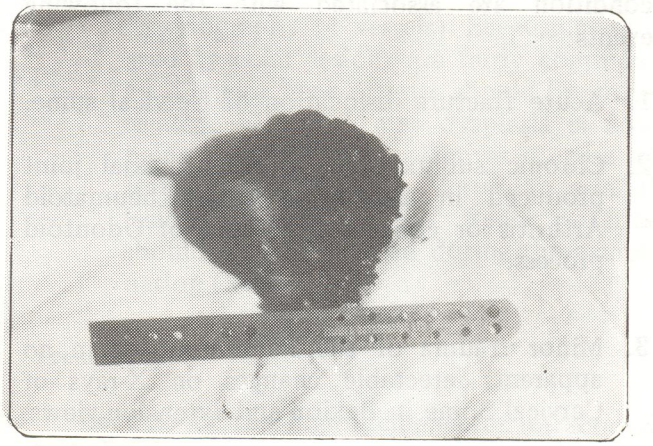

Fig. 7 'Multiloculated thrombosed aneurysm'

removal of redundant adipose tissue, a good skin cover was achieved.

REVIEW OF LITERATURE

Incidence and Pathogenesis

Vertebral Arteries are seemingly well protected from external trauma as they pass through transverse foramina of Cervical Vertebrae. However, where the cervical alignment is disturbed by injury or by disease, the artery is in danger of being stretched, compressed or twisted. Many traumatic arteriovenous fistulae of the Vertebral Artery have been reported in the literature but traumatic aneurysms have been described much less frequently. In 1893, Matas found mention of 19 VA aneurysms in the literature and included one of his own, in which he had successfully ligated the aneurysm in 1888 (1), Kister and Ranco (2) reported a case of their own and reported 30 recorded cases in the preceding 100 years of AV fistulae of which only 6 were purely aneurysmal. Lack of angiographic distinction between VA aneurysm and AVF of the Vertebral Artery casts doubt in the reported accuracy of the article. Hughes (3) in his Korean war experience could not find a single case of traumatic extracranial VA aneurysm in 212 casualties with AVF. Rich, outlining the entire military incidence in Vietnam found only 2 cases among 505 combat casualties with 558 AVF like condition (4).

Because of the unique, complex and variable course of the VAs in relation to the Cervical spine, injuries to these vessels resulting in an aneurysmal

condition are associated with the following events:

1. Acute fracture dislocation of Cervical spine.

2. Chronic subluxation of Atlanto-axial joint produced in association with Rheumatoid Arthritis or congenital absence of Odontoid process.

3. Minor trauma of the neck resulting in no apparent detectable changes on X-rays of Cervical Spine in flexion and extension views.

4. Radiation-induced lesion of medium sized vessels in the neck occurs rarely (5,6). The incidence of VA aneurysms, caused by radiation-exposures has not been scientifically documented.

5. Fibromuscular dysplasia of Carotid Artery is second only to the lesion of Renal Arteries in frequency (7). In Collin's series, less than 1% of Carotid Arteries showed fibromuscular dysplasia and of these, 98% were in females. It is highly probable that Vertebral Arteries are also susceptible to similar involvement (8, 9, 10).

6. Syphilis (11), Cystic Medical Necrosis (12) and congenital defects of internal elastic lamina (13) are rare causes of Vertebral Artery Aneurysms, Atherosclerosis generally causes TIAs and may also lead to a dissection of the intima but is not an important cause of VA aneurysms (14, 15, 16).

In 1947, Pratt et al (17) described 2 young adults who died suddenly following chiropractic manipulation of neck. The cause of death was arterial thrombosis and multiple brain stem and cerebular haemorrhagic infarcts, Kunkley et al (18) described the first survival in a patient with Wallenberg's syndrome and postulated that Vertebral Arteries are far less protected in their passage between C1 and Foramen magnum than at any other levels and are thus susceptible to external contusions. Ford and Clark (19) reported two cases of VA injuries resulting in vascular insufficiency following manipulation of neck. However, other forms of manipulations and injuries have also been reported requiring much less force. Such minor injuries have been reported as causing traumatic VA aneurysm as in painting ceilings (17,20), yoga (21, 22, 23) and gymnastic exercises (25). Injury to Vertebral Artery can result in surprisingly minor trauma of neck and this is usually a combination of rapid rotation with or without sharp extension. A clear history is usually not available. Delayed traumatic aneurysm formation has been well documented by a number of authors in Carotid Arteries but VA aneurysms has been much rarely reported. VA aneurysm, as a complication of Rheumatoid disease of Cervical Spine was reported by Fidel George in 1986 (24) where he described a case of a 63-year old man who had had conventinal anti-arthritic treatment with gold, penicillamine, azothiaprin, methotrexate and corticosteroids and then underwent C1-C2 fusion at which time a golf ball size pulseless posterior neck lump was detected. Eventually an angiographic diagnosis of VA aneurysm was made and at operation for its excision, a spicule of C2 was found to be causing the trauma. On reviewing the literature, he concluded that there were two main neurological complications of Atlanto-axial subluxation, Cervical compressive neuropathy and Vertebral Artery insufficiency.

It is surprising to note the rarity with which VA is manifestly injured with forced head movements when one considers that this artery has a devious course through six transverse foramina of the cervical vertebrae, passing through the groove of the arch of the Atlas, penetrating two strong membranes (atlanto-occipital membrane and cervical dura mater). Many cadaveric studies of flow through Vertebral Arteries have been done clearly showing that head rotation to the contralateral side with minor degree of stretching results in abrupt fall in the VA pressure to the point of cessation of forward flow relative to the degree of the rotation involved (25, 26, 27). Vertebral arteriographic studies with head rotation have also been well documented confirming the cadaveric findings (28, 29, 30) that head rotation does reduce flow in the VA. Such movements have a greater chance of precipitating VA aneurysms if a pre-existing causes are present, such as a hypoplastic Vertebral Artery, chronic subluxation of C1-C2, osteophytes etc. Yoshii et al (31) have also suggested that unlike its extracranial counterpart, the intracranial part of the VA lacks an external elastic lamina, has fewer elastic fibres in the media and has a thinner adventia (32). Cerebral perfusion is expected to

rise transiently during head movement (33) and with such increased demands and transiently reduced supplies, haemodynamic stretching occurs resulting in false aneurysm formation and dissection. Vertebro-vertebral arteriovenous fistula is more common in the second part of the extracranial Vertebral Artery because both the vessels are in close proximity within the canal.

Clinical Manifestations

1. Asymptomatic. This happens rarely when the patient is in a state of balance.
2. Vertigo and vomiting.
3. Slurring of speech.
4. Diplopia.
5. Ataxia.
6. Hypalgesia.
7. Local pains, if erosion of cervical vertebrae has taken place.
8. Stroke and sudden loss of consciousness.
9. Severe bilateral occipital pains.
10. Slight dysphagia.
11. Pulsatile mass.
12. Unresolving bruit with Carotid or Vertebral arterial compression.
13. Horner's syndrome. This occurs rarely.
14. Intermittent visual field defects.
15. Concomitant Carotid lesion is present in a minority of cases.

Investigations

1. Routine investigations, including:
   - Hb, FBC, ESR, Platelet count and morphology
   - RA and ANF
   - Blood glucose, lipids, Clotting profile.
   - Thyroid function tests.
   - EGG for dysarrythmia and ischaemic heart disease.
   - X-ray Chest and Cervical spine.
   - Doppler sonography and spectrum analysis.
   - Duplex vasoscans.
   - Digital Subtraction Angiogram (intra-arterial)
   - Arteriography (4 vessels). Because associated vascular lesions are fairly common, all 4 major neck arteries should be investigated. Fibromuscular dysplasia is now being detected more often as a cause of Renal, Carotid and Vertebral arterial lesions. Therefore the Renal Arteries should also be studied during the examination (34).

Treatment

Because of the high incidence of intracranial embolisation, early treatment is necessary. Incidence of TIAs is fairly high to recommend an aggressive approach.

Prior to the planning of the treatment, a full study of Circle of Willis is necessary to study crossflow, forward and atypical blood supply and to see if the Vertebral Artery can be safely ligated proximal and distal to the aneurysm.

An ideal treatment would be to isolate the aneurysm and to obliterate it whilst maintaining patency and continuity of forward flow in the Vertebral Artery.

1. Percutaneous balloon occlusion of the aneurysm should be tried (35, 36). During Vertebral angiography a latex balloon attached to a coaxial pair of catheters is passed. Balloon is positioned in the aneurysm and inflated with contrast and volume and pressure noted. If proximal injection of dye confirms total occlusion of the aneurysm then the dye is replaced with an equal volume of silicone monomers. These polymerize to form a solid silicone plug. The balloon is then detached with traction.

2. Aneurysmogrhaphy. This is done if replacement grafting is difficult to perform. It has been reported only twice in the literature (37, 38) and the results are still being followed up. It is a difficult and time-consuming procedure.

3. Excision of the aneurysm and proximal and distal ligatures. This is the commonest procedure performed due to its simplicity and practicality. An anterior approach is recommended for isolation and ligation of the corresponding Vertebral Artery. The aneurysm is then excised and the distal end of the Vertebral Artery is ligated. This

procedure should not be done if sufficient cross-(flow does not exist (39, 40, 41, 42, 43, 44, 45). Objections to this approach are raised by some authors suggesting that despite there being a retrograde flow present in the distal stump of the Vertebral Artery, embolic V-B disorders do occur and they suggest restoration of continuity (46, 47).

4. Excision of aneurysm and vein graft. Long Saphenous Vein can be interposed after excision of aneurysm but reported results are poor (48).

5. Excision of aneurysm and Subclavian-Vertebral bypass graft.

6. Excision of aneurysm and transposition of distal Vertebral Artery to the side of Common Carotid Artery or External Carotid (49, 50, 51).

7. Excision of the aneurysm and rotation graft of in situ Internal Mammary Artery to the VA.

8. In several cases where aneurysmectomy was considered difficult, simple ligation of the proximal Vertebral Artery has been suggested but with poor results (52, 53, 54, 1). Because of retrograde flow, thrombosis can occur subseequently.

9. In a vast number of cases, because of high surgical risks or lack of expertise, such aneurysms have been left in-situ, untreated, with uniform bad results (56, 57).

10. Supportive treatment whilst awaiting surgery is highly controversial but heparinisation and oral anticoagulation with Warfarin has been recommended by various authors for 12 weeks (55), Early post-operative use of steroids to reduce cerebral oedema is recommended by most clinicians, When neck injury has been suspected as having initiated the aneurysm, neck support should be provided.

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