

## Extracranial Vertebral Arteriovenous Fistula presenting as an Osteolytic Lesion of the Axis

- Case Report -

Sang Hoon Shin, M.D., Chun-Kee Chung, M.D., Hyun Jib Kim, M.D.,  
Moon Hee Han\*, M.D. and Dae Hee Han, M.D.

*Departments of Neurosurgery and Diagnostic Radiology\*, Seoul National University College  
of Medicine, Seoul, Korea*

*Arteriovenous fistulas of the extracranial vertebral artery are rare. The authors report a case of a spontaneous arteriovenous fistula of the extracranial vertebral artery presenting as an osteolytic lesion at the body of the axis. The patient presented with headache and posterior neck pain. The fistula was obliterated by an endovascular trapping.*

*Key Words : Arteriovenous fistula, Vertebral artery, Osteolytic lesion, Axis, Endovascular surgery*

### INTRODUCTION

Extracranial vertebral arteriovenous fistulas (AVF) are rare. There are three types; traumatic (including iatrogenic), spontaneous and congenital. (Rosenblum et al., 1986) Most of them are traumatic in origin, either accidental or iatrogenic. There are 45 cases of spontaneous vertebral AVFs reported in the literature including our case. (Goody et al., 1960; Faeth et al., 1961; Chou et al., 1967; Raskind et al., 1967; Erlich et al., 1968; Hiesiema et al., 1968; Verbiest, 1968; Geraci et al., 1969; Greenberg, 1970; Lawson et al., 1970; Bartal et al., 1972; Debrun et al., 1979; Reddy et al., 1981; Deans et al., 1982; Bahar et al., 1984; Reizine et al., 1985; Rosenblum et al., 1986) In these cases, the etiological factors leading to the formation of fistula are not clear. There has been no case presenting as an osteolytic lesion of the cervical vertebrae. A few cases are reported as congenital in origin. (Norman et al., 1950; Erlich et al., 1968; Geraci et al., 1969; Bartal et al.,

1972) The authors report a patient with a spontaneous AVF between the extracranial vertebral artery and epidural venous plexus presenting as an osteolytic lesion at the body of the axis.

### CASE REPORT

A 38-year-old man was referred for sudden headache and posterior neck pain in January, 1994. An osteolytic lesion at the body of the axis was noted on the radiological studies (Fig. 1, Fig. 2). Nine months previously he had been in a minor traffic accident. He had had posterior neck pain for 1 week and had recovered without any treatment.

#### Examination

On admission, the patient was neurologically intact except for a bruit in the left suboccipital region. His blood pressure was 120/80 torr and pulse rate 78/min. There was no change in the bruit or heart rate with carotid compression. Cerebral angiography demonstrated an abnormal vascular connection between the left extracranial vertebral artery and the epidural venous plexus at the level of the axis. (Fig. 3, A) There was an intracranial reflux from the right vertebral artery into the second portion of the left vertebral artery. The fistula was filled also from the right vertebral artery (Fig. 3, B).

**Address for correspondence :** Hyun Jib Kim, M.D., Departments of Neurosurgery, Seoul National University Hospital, 28 Yongun-Dong, Chongno-Go, Seoul 110-744, Korea. Tel : (02) 760-2358, Fax : (02) 744-8459

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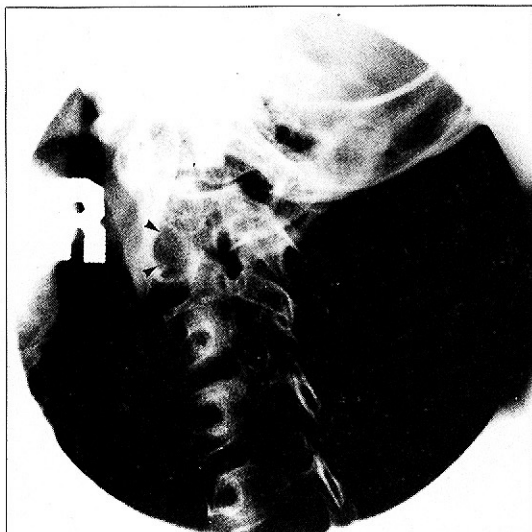


Fig. 1. Lateral radiograph of the cervical spine showing an osteolytic lesion at the C-2 vertebral body (arrows).

No abnormality was detected on selective bilateral internal carotid angiographs and there was no demonstrable "steal" from the intracranial circulation.

**Operation**

On June 13, 1994, the patient underwent a vertebral arterial trapping by detachable balloons. The left ver-

tebral angiography was performed by the femoral route and showed a fistula. We placed test balloons at the portions of the vertebral artery which was proximal and distal to the fistula. After five minutes of test occlusion his neurological status was unchanged and brain single photon emission computed tomography (SPECT) showed no perfusion defect (Fig. 4). The right vertebral arteriograms taken immediately after occlusion showed a retrograde filling of left posterior inferior cerebellar artery (Fig. 5). The repeated neurological examination and brain SPECT were performed and revealed no specific abnormality.

**Postoperative Course**

The postoperative course was uneventful. By the next morning the headache and posterior neck pain disappeared. Follow-up simple cervical spine views showed that the balloons were still inflated in the same position in which we had placed them. The patient left the hospital without bruit 6 days after trapping on June 18, 1994. He remained symptom-free at the 18-month postoperative follow-up evaluation.

**DISCUSSION**

This case represents an unique presentation of a direct AVF between the extracranial vertebral artery and the epidural venous plexus which presented as an

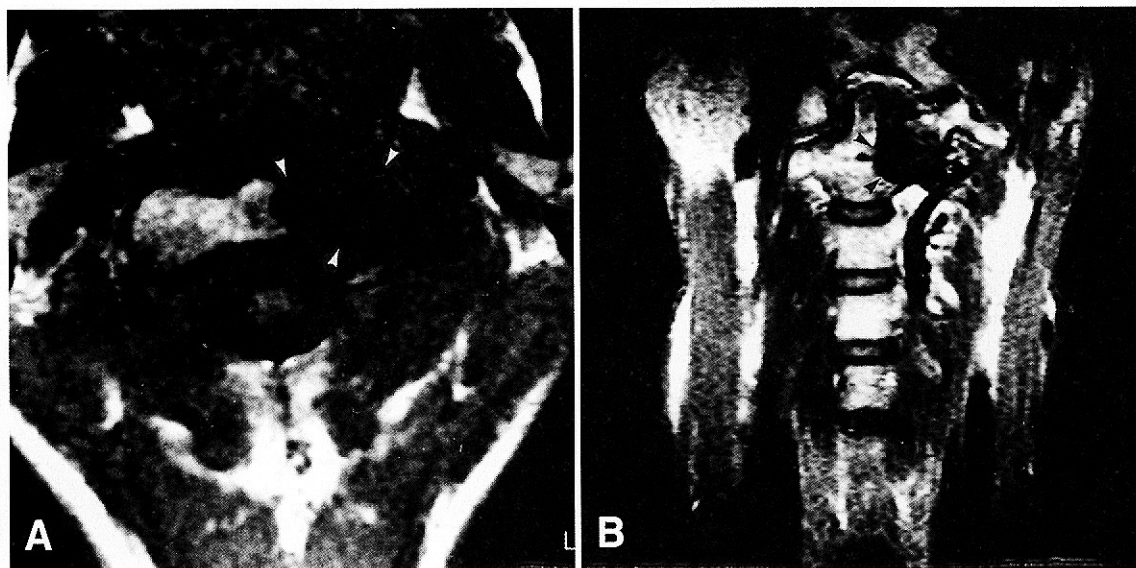


Fig. 2. Axial (A) and sagittal (B) views of Magnetic resonance images showing a signal-void mass at the C-2 vertebral body.

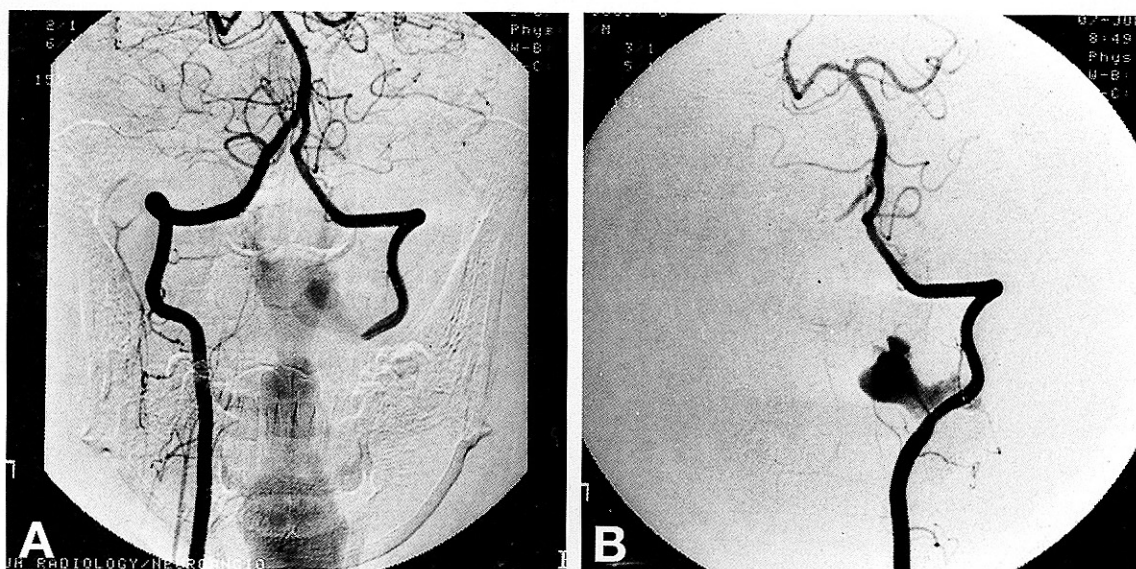


Fig. 3. Bilateral vertebral angiograms, anteroposterior view. A: demonstrating an abnormal vascular connection between the extracranial vertebral artery and the epidural venous plexus with C-2 vertebral body filling. B: showing the clear delineation of fistula feeding from the right vertebral artery.

osteolytic lesion at the body of the axis. Spontaneous extracranial vertebral AVF may arise as a solitary lesion from inflammatory processes (Hugenholtz *et al.*, 1982) or arterosclerotic changes. (Rosenblum *et al.*, 1986) Congenital lesions have been reported in patients with

a history of birth trauma or even without such evidence. (Norman *et al.*, 1950; Erlich *et al.*, 1968; Geraci *et al.*, 1969; Bartal *et al.*, 1972) In our case, the pulsating arterial pressure through high-flow AVF might give an expansile strength to basivertebral vein. This dilated



Fig. 4. Brain SPECT after five-minute occlusion of the left vertebral artery showing no perfusion defect.

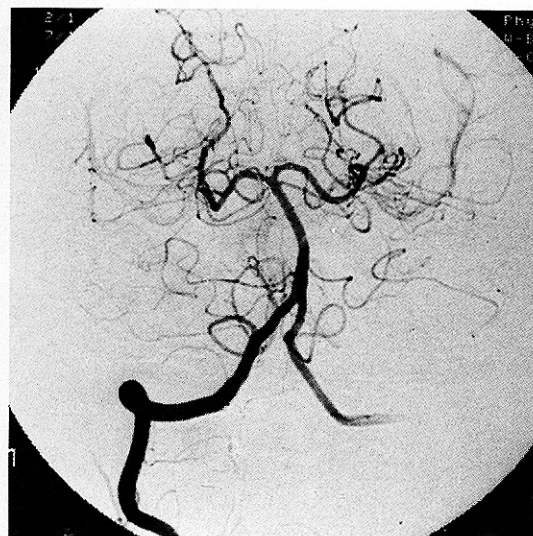


Fig. 5. Right vertebral angiogram after the occlusion of the left vertebral artery showing a retrograde filling of left posterior inferior cerebellar artery.

venous sac made an osteolytic lesion at the body of the axis.

Review of the 45 previously reported cases of spontaneous AVFs including our case revealed that the left side has been more commonly involved (27 cases, 60%) than the right (18 cases, 40%); there was no bilateral case. The second portion of the vertebral artery was most frequently involved (30 cases, 67%); the first portion was involved in 9 (20%) and the third portion in six (13%). The origin of these fistulas has been attributed to vertebral artery development from longitudinal anastomoses between six pairs of dorsal embryonic intersegmental arteries and its intimate association with draining epidural and muscular plexuses. (Padget, 1948; Lawson et al., 1970; Debrun et al., 1979) The 45 patients included 18 males and 27 females. Patients ranged in age from 6 months to 70 years. Tinnitus was the most common complaint (12 cases, 26.7%). The other symptoms were neck mass, headache, myelopathy and neck pain.

Extracranial vertebral AVF presenting as an osteolytic lesion must be differentiated from aneurysmal bone cysts, hemangiomas, hydatid cysts, fibrous dysplasia, osteosarcomas and metastatic lesions. Aneurysmal bone cyst frequently occurs in children and young adults and presents with a mild pain of several month duration. Expansile, trabeculated, lucent lesion primarily involves the posterior elements. Vertebral hemangioma is differentiated by its characteristic radiographic picture that shows no ballooning and does not involve neighboring vertebrae. Hydatid cysts which contain low density fluid are easily differentiated from vertebral AVF with computerized tomography. Fibrous dysplasia of the vertebra affects children and young adults, which usually involves more than one vertebra often with visible involvement of the skull, grows slowly until the skeletal growth ceases, and is painless. Osteosarcomas and metastatic lesions produce more pain, are seen in older age groups, and do not show any ballooning in the radiographs.

The ideal goal in treating vertebral AVF is selective elimination of the fistulous connection along with maintenance of normal patency and flow in the parent artery and draining vein. (Debrun et al., 1979; Reizine et al., 1985) We selected the balloon occlusion of the fistula because this technique seemed simpler, less invasive and anatomically more direct than a neck dissection aiming at eliminating the arterial input to the lesion. But we could not attain a selective occlusion of fistula due to a pin-point opening of the fistula. There are major

cerebrovascular risks involved with the indirect methods of treatment for vertebral AVF that sacrifice vertebral blood flow. So we had first occluded the left vertebral artery temporarily by test balloon for 5 minutes and performed brain SPECT to demonstrate the blood flow of the territory supplied by the left vertebral artery. This method may be helpful to anticipate the hemodynamic changes after sacrificing the parent artery.

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