A 65 year old previously healthy farmer was brought to the emergency room 18 hours after the acute onset of symptoms. There was no family history of movement disorders, or of neuroleptic-induced extrapyramidal reactions. Four days before admission, he experienced mild epigastric distress and abdominal fullness, for which 2 days later, he took one tablet, 5 mg of oral metoclopramide, dextromethorphan, and Gasgel (simethicone, aluminum magnesium hydrate and magnesium oxide). Some 30 hours after the first dose, and 2 hours after the second dose, he developed irregular, intermittent muscle jerking, most obviously in the face, mouth, and limbs (fig. a). The jerks could be initiated when the patient began to speak or to move a limb, but were not affected by external stimuli such as pinprick, touch, vibration, light, or sound. The jerks disturbed speech and swallowing. There was no change in the level of consciousness. Obvious irregular independent flapping tremors were observed in both outstretched hands when the wrists and fingers were extended (fig. b). There was involuntary hyperextension of the neck and forceful opening of the mouth. Routine laboratory studies were all normal including liver function tests. A chest radiograph was normal. EEG (awake) revealed no abnormality. The asterixis, myoclonus, and acute dystonic reactions cleared within 12 hours. On follow-up 6 months later the patient was normal.

Myoclonus has been described in association with the acute hyperkinetic syndrome

in neuroleptic agents.^{4,5} It has not been recorded in the extrapyramidal side effects of metoclopramide. Asterixis is a common sign in diffuse encephalopathies in various metabolic disorders as well as a rare manifestation of focal lesions in the parietal lobe, thalamus, midbrain and pons.⁶⁻⁸ Asterixis and myoclonus were the dominant features in our patient with a relatively mild acute dystonic reaction. Both sets of manifestations disappeared at about the same time. Neuroleptic-induced acute dystonic reactions may be due to enhanced dopamine release on supersensitive postsynaptic receptors.⁹ Whether the asterixis and myoclonus can also be attributed to a similar (hypothesised) transient increase of dopamine acting on postsynaptic receptors, is an open question.

Both myoclonus (using stronger stimulation) and asterixis can be produced by electrical stimulation of the human motor cortex through the intact scalp.¹⁰ The possibility of a metoclopramide-induced encephalopathy responsible for both the acute manifestations of asterixis as well as myoclonus might be considered in our particular patient.

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References

- Robinson OPW. Metoclopramide: side effects and safety. *Postgrad Med J* 1973;**49** (suppl 4): 77-80.
- Grimes JD, Hassan MN, Preston DN. Adverse neurologic effects of metoclopramide. *Can Med Assoc J* 1982;**126**:23-5.
- Bateman DN, Rawlins MD, Simpson JM. Extrapyramidal reactions with metoclopramide. *Br Med J* 1985;**291**:930-2.
- Rupniak NMJ, Jenner P, Marsden CD. Acute dystonia induced by neuroleptic drugs. *Psychopharmacology* 1986;**88**:403-19.
- Marsden CD, Tarsy D, Baldessarini RJ. Spontaneous and drug-induced movement disorders in psychotic patients. In Benson DF, Blumer D (eds) *Psychiatric Aspects of Neurologic Disease*. New York, Grune & Stratton, 1975:219-66.
- Degos JD, Verroust J, Bouchareine A, Serdaru M, Barbizet J. Asterixis in focal brain lesions. *Arch Neurol* 1979;**36**:705-7.
- Donat JR. Unilateral asterixis due to thalamic hemorrhage. *Neurology* 1980;**30**:83-4.
- Kudo Y, Fukai M, Yamadori A. Asterixis due to pontine haemorrhage. *J Neurol Neurosurg Psychiatry* 1985;**48**:487-9.
- Kolbe H, Clow A, Jenner P, Marsden CD. Neuroleptic-induced acute dystonic reactions may be due to enhanced dopamine release on to supersensitive postsynaptic receptors. *Neurology* 1981;**31**:434-9.
- Marsden CD, Merton PA, Morton HB. Is the human stretch reflex cortical rather than spinal? *Lancet* 1973;i:759-61.

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Bilateral spontaneous carotid-cavernous fistulas, associated with systemic hypertension and generalised arteriosclerosis: a case report

Sir: Spontaneous carotico-cavernous fistulas have been repeatedly reported as a result of improved angiographic technique and better clinical recognition. They are usually indirect. Of the 21 cases with spontaneous carotico-cavernous fistulas observed by Nukui *et al*¹ only one was direct; all 20 cases of direct fistulas reported by Peeters and Kröger² and Peeters,³ and all, but one, of the 54 cases reported by Debrun *et al*⁴ had history of trauma; thus direct spontaneous fistulas are very rare. Even rarer is the occurrence of bilateral direct fistulas whether spontaneous or traumatic. With the exception of one patient in the 54 patients series reported by Debrun *et al*, all the direct fistulas reported above were unilateral.

We report a patient with bilateral direct spontaneous carotid-cavernous fistulas associated with hypertension and intracranial arteriosclerosis.

A 70 year old woman noticed one morning, on waking up, a swelling of the right eye lids and redness of the right eye, accompanied by a pulsating whizzing noise all over the head. She also experienced transient diplopia on looking to the right with horizontally shifted images, and a clumsy feeling of the left arm. Her complaints were not preceded by episodes of headache or fever. There was no history of head injury. Acuity of her vision was not altered; she had had poor vision of the left eye ever since the delivery of a child more than 30 years previously. Apart from systemic hypertension which was well controlled on betablockers and diuretics she had no other major illnesses. During the next 2 months, her clinical symptoms steadily improved; the whizzing noise got less, the eye lids were less swollen and diplopia diminished. Because of her symptoms she consulted an ophthalmologist who referred her to the neurology

Fig Surface EMG recordings. a: simultaneous contractions of triceps (Ti) and biceps (Bi) indicating myoclonic jerks in the resting right arm. b: Intermittent lapse (↓) of finger extensors (FE) in the outstretched right hand with hyperextended wrist and fingers.

clinic with the provisional diagnosis of a carotico-cavernous fistula. Examination 2 months after onset of symptoms revealed swelling of the right eye lids, slight proptosis of the right eye with engorgement and tortuosity of the episcleral veins, blurring and congestion of the right fundus and slight pallor of the left fundus. The visual fields in both eyes were normal. There was slight central paresis of the left facial nerve and some abduction weakness of the right eye. A systolic high frequency murmur was heard all over the head and over both internal carotid arteries. No other abnormalities in the physical examination were seen.

Carotid angiography showed *double sided direct* carotid-cavernous fistulas. Catheterisation of the left side showed a rapid filling of a grossly dilated right superior ophthalmic vein. The fistula also emptied into the ipsilateral inferior petrosal vein and internal jugular vein. Angiography of the right internal carotid artery showed passage of the contrast-medium to the left side veins except to the superior ophthalmic veins. The fistula emptied into the right superior ophthalmic vein, as well as in the ipsilateral inferior petrosal vein and the internal jugular vein (fig). Atherosclerotic changes were observed in both internal carotid arteries: there was a haemodynamically significant stenosis of the bifurcation of the middle cerebral artery on the left; a stenosis of approximately 50% was visible in the supraclinoid part of the internal carotid artery on the right. There were no signs of an aneurysm.

Because of the small size of the fistulas, the benign clinical course before consultation and because of the presence of generalised intracranial atherosclerosis with signs of cerebral ischaemia, the patient was managed conservatively with antihypertensive therapy. She continued to improve on this regime; her diplopia disappeared and she was discharged. On subsequent outpatient follow up 6 months later she showed no deterioration. Angiography was not repeated.

Spontaneous carotico-cavernous fistulas are mainly *unilateral* and *indirect* and consist of dural arteriovenous shunts involving the cavernous sinus and branches of the external carotid artery, the internal carotid artery or both. The *bilaterality* of the *direct* fistulas in our patient may be considered rare. Direct fistulas formed by a tear in the cavernous portion of the internal carotid artery are predominantly of traumatic origin,^{1,5} but atherosclerosis with or without aneurysm formation can be contributory to vascular weakness, leading to a direct fistula.⁶

In our patient, atherosclerosis as a

sequella of hypertension and old age may have been responsible for weakness of the carotid siphons, resulting in bilateral fistulas. As demonstrated by our case, headache is not always a presenting symptom, and even with bilateral fistulas eye symp-

toms may remain unilateral.^{1,5,7} The abnormality in the left eye of our patient that had originated 30 years previously after childbirth, might have induced a reduction of blood flow from the left orbit to such a degree that the fistulas could not drain into

Fig 1 (a) Right carotid artery angiogram (lateral view): early arterial phase with rapid filling of the cavernous sinus (arrow). (b) Right carotid artery angiogram: venous phase showing filling of both the cavernous sinus, the homolateral superior ophthalmic vein and on both sides the internal jugular veins (AP view).

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the left ophthalmic vein.

The presence of cerebral ischaemia in our patient as evidenced by a central paresis of the left facial nerve and clumsiness of the left hand, and the presence of a small calibre fistula caused reluctance to attempt invasive therapy consisting of the occlusion of the fistulas with a detachable balloon via an arterial (internal carotid artery) or venous (superior ophthalmic vein, inferior petrosal vein) route.^{4-7,8} The patient improved, however, on conservative therapy despite the direct type of the fistula that mostly needs interventional therapy.

In our patient a traumatic cause could not be established. There was an emptying of the left side fistula through anastomatic vessels into the right side cavernous sinus and superior ophthalmic vein; a preexisting lesion of the left eye, possibly of thrombotic origin, may have been contributory to this overflow. The clinical course in this patient showed spontaneous improvement, thus precluding the necessity of invasive therapeutic interventions.

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References

- 1 Nukui H, Shibasaki T, Kaneko M, Sasaki H, Mitsuka S. Long-term observation in cases with spontaneous carotid-cavernous fistulas. *Surg Neurol* 1984;21:543-52.
- 2 Peeters FLM, Kröger R. Dural and direct cavernous sinus fistulas. *AJR* 1979;132:599-606.
- 3 Peeters FLM. Treatment of 20 direct carotid-cavernous fistulas. *Diagn Imag Clin Med* 1983;52:127-36.
- 4 Debrun G, Lacer P, Vinuela F, Fox A, Drake ChG, Cason JP. Treatment of 54 traumatic carotid-cavernous fistulas. *J Neurosurg* 1981;55:678-92.
- 5 Barrow DL, Spector RH, Braun IF, Landman JA, Tindall SC, Tindall GT. Classification and treatment of spontaneous carotid-cavernous sinus fistulas. *J Neurosurg* 1985;62:248.
- 6 Dandy WE, Folliis RH JR. On the pathology of carotid-cavernous aneurysm (pulsating exophthalmos). *Am J Ophthalmol* 1941;24:365-85.
- 7 Yamamoto I, Watabe T, Shinoda M, Hasegawa Y, Sato O. Bilateral spontaneous carotid-cavernous fistulas treated by the detachable balloon technique: Case report. *Neurosurgery* 1985;17:469-73.
- 8 Mullan S. Experiences with surgical thrombosis of intracranial berry aneurysms and carotid-cavernous fistulas. *J Neurosurg* 1974;41:657-70.

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Subdural and intraventricular tension pneumocephalus after transsphenoidal operation

Sir: Cerebrospinal fluid (CSF) rhinorrhoea develops in one to three per cent of patients¹ after a transsphenoidal pituitary operation; it is often followed by simple pneumocephalus which usually resolves spontaneously. Tension pneumocephalus is extremely rare,² and results in brain compression with rapid clinical deterioration. We describe a case and propose an explanation for its development.

For the previous 4 years, this 28 year old man had noticed progressive growth, coarsening of facial features and enlargement of extremities. He had classical acromegalic deformities and a computed tomographic (CT) scan demonstrated a large non-homogeneous pituitary tumour extending superiorly to the third ventricle. After radical removal of the tumour by a transsphenoidal approach, the neurosurgeon noticed a 3 mm opening in the diaphragma sellae through which clear CSF oozed. The sella was packed with synthetic fibrin sponge, sealed with glue and an external lumbar subarachnoid drain was inserted. Histological studies showed that the tumour was a mixed growth hormone and prolactin-secreting adenoma.

CT scan confirmed radical removal of tumour from the sella, which instead was filled with air. The CSF rhinorrhoea ceased 2 weeks after operation. Eighteen days after operation, the patient was found to be drowsy. He had a fever and intense neck rigidity. CT showed a large air collection in the ventricular and subarachnoid spaces with a mass effect on the frontal lobes. The lumbar CSF drain was removed and the patient slowly recovered during the subsequent month. He was discharged taking hormonal replacement therapy and resumed his previous occupation as a high school mathematics teacher.

Simple pneumocephalus most frequently arises as a complication of a head injury in which a compound basal skull fracture with tearing of the meninges allows entry of air into the cranial cavity. It can also follow a neurosurgical operation.³ Tension pneumocephalus is rare. Among the mechanisms that may be responsible is the combination of a fistula at the base of the skull and a CSF shunt as in a patient with hydrocephalus.⁴ The complication has been rarely encountered after transsphenoidal hypophysectomy.² In our patient the placement of a lumbar subarachnoid drain may have precipitated the development of tension pneumocephalus through a simpler mechanism. We propose that loss of CSF as a result of both rhinorrhoea and the lumbar catheter caused a drop in CSF pressure until the latter reached an equilibrium with atmospheric pressure. If the loss of CSF exceeds the production rate an equal volume of air enters through the fistula into the subarachnoid space.⁵ The sudden spontaneous resolution of rhinorrhoea in our patient several days before discovery of pneumocephalus is consistent with this explanation. The collection of air in the ventricles prevented a significant shift of the midline structures and for this reason, an urgent decompressive operation was not considered to be necessary. Frequent radiographs after removal of the lumbar CSF drain showed progressive re-absorption of the intracranial air.

Direct repair of the fistula is considered standard treatment of CSF leaks after a transsphenoidal operation. An alternative to further operation is the use of a lumbar subarachnoid drain.⁶ In the latter cases, meticulous care of the drainage system is important to avoid excessive loss of CSF and the risk of tension pneumocephalus.

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References

- 1 Landolt AM. Cerebrospinal fluid rhinorrhoea: a complication of therapy for invasive prolactinomas. *Neurosurgery* 1982;11:395-401.
- 2 Shields CB, Valdes-Rodriguez AG. Tension pneumocephalus after trans-sphenoidal hypophysectomy: case report. *Neurosurgery* 1982;11:687-9.