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Clinical Records

Beware bleeding from the ear

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Abstract
We present the case of a 55-year-old man who presented with intermittent, profuse bleeding from the ear five years following radiotherapy for a nasopharyngeal carcinoma. He had developed osteoradionecrosis of the temporal bone, and the bleeding was shown to originate from an aneurysm of the internal carotid artery within his temporal bone. This has not previously been reported as a complication of radiotherapy for nasopharyngeal cancer.

Key words: Nasopharyngeal neoplasms; Osteoradionecrosis; Aneurysm.

Case history

A 55-year-old man was admitted with a two week history of spontaneous, intermittent, severe bleeding from his right ear. On admission he was shocked and required immediate fluid replacement. About five years previously he had received radiotherapy for a nasopharyngeal carcinoma (66 Gy by conventional fractionation). He had been followed up regularly in the oncology clinic, and there was no sign of recurrence of disease. Since the radiotherapy he had experienced intermittent, right otorrhoea and had received regular aural toilet from a private ENT surgeon.

After admission he continued to bleed intermittently in volumes of up to 500 ml at a time, and required repeated transfusions to maintain his haemoglobin level. Examination of his ear was difficult as the ear canal was filled with blood clot and necrotic tissue. It was feared that too vigorous cleaning would result in further, perhaps uncontrollable haemorrhage. However, no normal anatomy was apparent. No tympanic membrane or ossicles could be identified, and the medial wall of the middle ear seemed to be necrotic and mobile.

A clotting screen was normal. A CT scan showed only an opaque right mastoid air cell system 'compatible with infection'. A carotid angiogram showed a large aneurysm of the internal carotid artery (Fig. 1) with its lateral extent at the medial wall of the middle ear.

After discussion of the possible complications with the patient and his relatives, the lateral half of the aneurysm was embolized by the insertion of three platinum/dacron wool embolization coils (Cook Inc., Bloomington, USA) via the femoral artery. This procedure was halted when the patient developed a hemiplegia from which he recovered completely within 24 hours. It was apparently successful in stopping the bleeding, and the patient did not bleed for the next five days. Before the patient was discharged home his ear was tentatively examined once more. Embolization coils were visible, and were clearly in the process of extrusion (Fig. 2).

Following this discovery a further attempt was made to embolize the aneurysm. During this procedure it became apparent that the aneurysm was a trilobed structure. On introducing a further 10 coils, the lateral and middle lobes were apparently embolized completely. Again the procedure was halted when the patient developed a hemiplagia together with aphasia, from which he again recovered in some 24 hours. Though coils con-

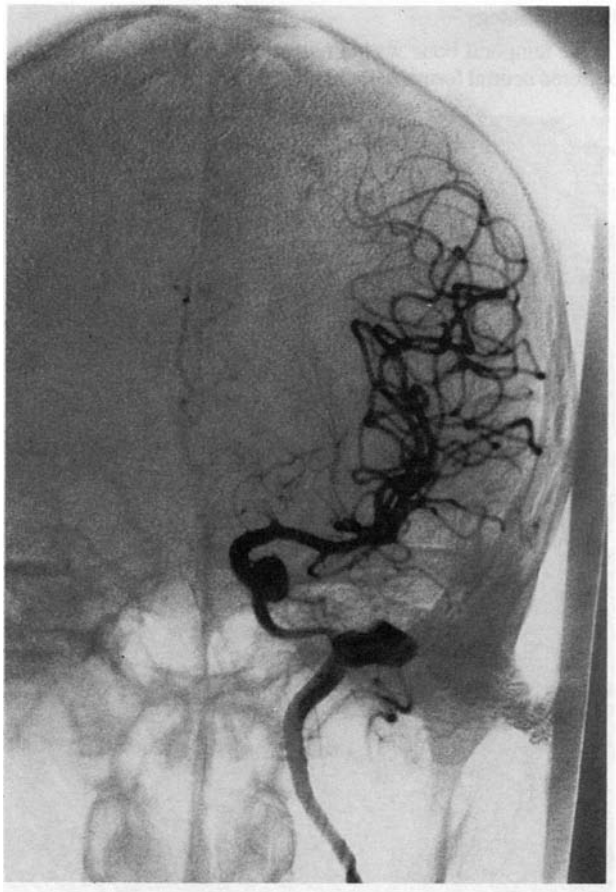


FIG. 1
Digital subtraction angiogram showing an aneurysm of the internal carotid artery.

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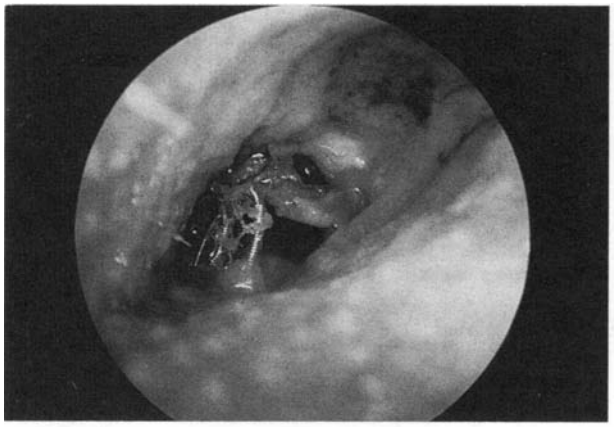


FIG. 2
Embolization coils being extruded into the ear canal.

tinued to be visible in his deep ear canal, no further bleeding occurred, and the patient was discharged home ten days after the second embolization with strict instructions to return if he experienced any further bleeding. He was seen every two days and all seemed well. However 13 days after discharge he experienced some minor leakage of blood from his ear, and did not seek medical attention. The next day he bled profusely and was admitted shocked and unconscious. He never regained consciousness and died three days later.

Histopathology

The temporal bone was removed at autopsy. It was fixed in buffered neutral formalin for two weeks followed by decalcification with formic acid for another two weeks (Prophet, 1992). After it was determined that decalcification was adequate, histological sections were processed by conventional methods. There was moderate atherosclerotic change in the arterial wall with destruction of the muscular wall and elastic tissue. Focal areas showed erosion of bone surrounding the aneurysm with fragmentation and increase in bone resorption (Fig. 3).

Discussion

Hong Kong has the highest incidence of nasopharyngeal cancer in the world (Muir *et al.*, 1987). The primary treatment of this malignancy is by radiotherapy, and the temporal bone inevitably receives a high dose of radiation during the course of this treatment. Bone is relatively resistant to radiation unless involved by tumour (Lederman, 1965), and osteoradionecrosis of the temporal bone is a relatively rare complication. So far as we are aware, severe haemorrhage from the ear as a complication of osteoradionecrosis and an internal carotid aneurysm has not been described previously.

Ramsden *et al.* (1975) described 29 cases of osteoradionecrosis of the temporal bone, and classified them into either 'localized' or 'diffuse' types. Patients in the localized group typically had an area of exposed, dead bone usually situated in the floor of the external auditory meatus. They complained of otitis externa-like symptoms, with mild otalgia and offensive otorrhoea. This was treated with regular aural toilet, and over several years this sequestrum gradually separated and the meatus healed with resolution of symptoms. Patients in the diffuse group suffered generalized ischaemic necrosis of the temporal bone. They experienced severe pain, and profuse, pulsatile, offensive otorrhoea. There was a much higher incidence of involvement of adjacent structures such as the brain, labyrinth, facial nerve, temporomandibular joint and parotid gland. Intracranial infection



FIG. 3
Photomicrograph of the aneurysm. There is erosion of the adjacent bone and destruction of the arterial wall. In the lumen is a thrombus (H&E, ×120).

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occurred in four of the 11 cases described. These patients with diffuse osteoradionecrosis had usually received a much higher dose of radiation to the temporal bone than those with the localized form.

The underlying change in osteoradionecrosis is aseptic, avascular necrosis (Schuknecht and Karmody, 1966). Infection accentuates the changes (Ewing, 1926). The temporal bone is unusual in that it communicates with the potentially infected upper respiratory tract via the eustachian tube. Characteristically the effects of irradiation osteitis may be delayed for years though the soft tissue effects (dermatitis, otitis media and labyrinthitis) are immediate (Schuknecht and Karmody, 1966).

Our patient clearly had severe osteoradionecrosis of the diffuse type with involvement of his carotid artery. Though it would have been an easy matter to tie off his carotid artery in the neck, this would have led to severe neurological sequelae in this patient. It was felt that the wall of the aneurysm would not be strong enough to bear a clip. After a full discussion with the patient and his family, the decision was therefore made to treat him by attempting to embolize the aneurysm. This procedure was complicated by temporary neurological changes, but was initially successful in controlling the haemorrhage. After ignoring some minor further bleeding from his ear, he then bled severely, presumably from an area of the aneurysm which had not been successfully embolized or after further extrusion of the embolization coils into the ear canal.

Osteoradionecrosis of the temporal bone is unusual, and to be further complicated by involvement of the internal carotid artery seems to be unique. Clinicians should regard any bleeding from the ear as potentially very serious.

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