Endovascular Embolization to Control Life-Threatening Epistaxis Due to a Post Radiotherapy Pseudoaneurysm of the Maxillary Artery

P.Y. MARCY, N. MAGNÉ *, C. BAILET, G. POISSONNET **, R.J. BENSADOUN *, F. DEMARD **

Department of Radiology, * Department of Radiotherapy, **Department of Head and Neck Oncology, Centre Antoine Lacassagne, Nice Cedex 2, France

Key words: nasopharynx cancer, radiotherapy, complications

Summary

We report an unusual case (never described) of a false aneurysm of the maxillary artery following irradiation and biopsy that ruptured into the nasopharynx in an 34-year-old man who presented with severe epistaxis 15 months after radiation therapy of the nasopharynx (80 Gy). Angiography revealed a fusiform aneurysm of the second portion of the maxillary artery, which was successfully embolized but the patient died from a pseudomonas septicemia three months later. False aneurysms of the maxillary artery following irradiation are exceptional complications. Despite a successful endovascular treatment, the absence of collateral revascularization did not allow to complete healing of the radionecrotic pharyngeal area and led to a lethal septicemia.

Introduction

Radiation therapy is the standard treatment for nasopharyngeal carcinomas (NPC) and regional node metastases. Complications resulting from this treatment include radiation myelopathy, cranial and peripheral nerve paralysis, osteoradionecrosis of the temporal bone, fibrosis of the masticatory muscles, acute, early

and late delayed central nervous system reaction, multiple meningiomas and meningiosarcomas, and carotid artery disease². The incidence of accelerated atherosclerosis due to irradiation may be greater than in non irradiated patients. The most frequent vascular lesions related to radiation therapy are stenosis of the carotid artery⁵. Stenoses may become evident only two to three decades after radiation therapy, are often bilateral and involve a long segment. They occasionally may progress to unilateral or bilateral occlusion. Transmural necrosis of the vessel may also result in formation of a false aneurysm (FA). We present an unusual case of a FA of the maxillary artery, presenting as a life-threatening epistaxis fifteen months after high dose radiation therapy, that may have been precipitated by wound complication after surgical biopsy. To our knowledge, this is the first report of a post radiation FA of the maxillary artery in world literature.

Patients and Methods

A 34-year-old male presented with bilateral cervical lymph nodes in november 1997. Clinical examination disclosed a 30 mm sessile mass located at the left posterolateral area of the nasopharynx and the oropharynx. MRI revealed

Figure 1 Left carotid angiogram, under arterial pressure control. There is a tight stenosis below the 18 x 8 mm false aneurysm located at the second pterygoid segment, without any sign of contrast medium extravasation. No flow into the facial artery was seen at that time.

Figure 2 Left external carotid angiogram performed after coil embolization proximal to the false aneurysm. The arterial flow has ceased completely in the arterial mandibular segment of the internal maxillary artery, in the false aneurysm, and above. The coil is visible, at the level of the presumed origin of the middle meningeal artery. Note opacification of the facial artery that shows mid proximal and distal short stenosis.

an inhomogeneous hypervascularized lobulated mass, without any evidence of skull base erosion. Results of biopsy were undifferentiated carcinoma of nasopharyngeal type (UCNT), T4N1 (initial tumor-node-metastasis stage, International Union Against Cancer-American Joint Committee on Cancer, 1987). The patient subsequently underwent three courses of chemotherapy and 80 Gy radiation therapy with standard fractionation over a 14 week period (in observance of the treatment). In february 1999, the patient suffered from a trismus and repeated epistaxis. Clinical examination of the oral cavity and a flexible nasopharyngoscopy revealed a large necrosis of the lateral pharyngeal wall, in place of the previous tumour. Careful surgical biopsies were performed, showing no evidence of local recurrence. Conservative measures (antibiotics, analgesics and hyperbaric oxygenation) were promptly initiated, but the patient was admitted in emergency, six days later, for oropharyngeal haemorrhage, making him lose 6 g/dl of hemoglobin. Angiography, performed under arterial pressure control, revealed a 18 x 8 mm false aneurysm (FA) of the pterygoid segment

of the left internal maxillary artery (IMA), with a short stenosis upstream, without extravasation of contrast material (figure 1). After selective catheterization of the internal maxillary artery with a 2.6 F tracker catheter, percutaneous embolization of the false aneurysm was successfully performed with one 20 mm x 3 mm cone-shaped coil, proximal to the arterial stenosis (figure 2). This procedure completely obliterated the IMA excluding the false aneurysm. No rebleeding occurred after this emergency procedure but bacteriological samples revealed pseudomonas in the nasopharyngeal necrotic cavity.

The patient died three months later of *Pseudomonas* septicemia, with a large persistent radionecrotic cavity of the nasopharynx.

Discussion

Radiation therapy plays an important role in the therapeutic management of head and neck tumors. Side effects due to high dose irradiation of normal tissue may appear immediately or a few months after completion of the treatment. The most common vascular lesions relat-

ed to radiation therapy are stenosis of the carotid artery due to extensive fibrosis beneath the intima, scars in the vessel wall and inflammatory ulcers. They may progress to unilateral or bilateral occlusion. Occasionally, transmural necrosis of the vessel wall may also result in formation of a false aneurysm. Exceptionally, the pseudoaneurysm may be related to invasion of the arterial wall by the neoplasm itself⁶. A few cases of maxillary artery FA have been reported in the literature, but all without exception follow a trauma or a surgical procedure. Such abnormalities have been described in cases of blunt facial trauma, jaw fracture, maxillary gunshot injury, mandibular and maxillary Lefort I and Lefort III osteotomy, transsphenoidal surgery, or after functional endoscopic sinus surgery³. In the series reported by Mak et Al, three out of eight nasopharyngeal carcinoma patients presenting with intractable epistaxis, had an internal carotid artery aneurysm detected at angiography that was likely to be a result of irradiation⁴.

Among all the cases of FA and haemorrhage caused by carcinomas of the head and neck, different embolization materials were used such as gelfoam, polyvinyl alcohol, isobutyl-2 cyano-acrylate, detachable and non-detachable coils, detachable balloons, and more recently covered stents, in order to exclude the FA. The results of these techniques varied from a complete resolution of the symptoms to rebleeding in up to 37% of the cases^{4,6}.

In our case, the high irradiation dose received (greater than 60 Gy), the necrosis of the lateral pharyngeal mucosa and masticatory muscles (figure 3) and the presence of diffuse and multiple arterial stenosis in a young adult within the field of previous radiotherapy treatment, are positive arguments for a diagnosis of post-radiation FA. Subacute arterial rupture of the maxillary artery with life-threatening bleeding may have been precipitated by wound complications after performance of careful "blind" biopsies by the surgeon.

Internal maxillary artery ligation has been proved to be not so effective as percutaneous embolization in treating epistaxis and was not appropriate in this case. Many contributing factors may explain this difference such as incomplete ligation of vessels, alternative dominance of vessels, or reconstruction of flow through collaterals. Controversially, rebleeding has

Figure 3 CT scan of the infra-temporal fossa. There is a large aeric cavity of the left lateral wall of the pharynx, with necrosis of the lateral pterygoid muscle (aeric spot) and complete lysis of the left pterygoid process, thus causing the severe trismus. Note the Gianturco coil close to the left styloid process.

been described after endovascular embolization, in case of incomplete occlusion by insufficient or inadequate embolic agents, thus this technique should be performed by an experienced interventional radiologist who is aware of the vascular anatomy of the head and neck and technical tricks of the embolization. The optimal treatment should preserve IMA patency and exclude the flow from within the pseudoaneurysm; the expected result is thrombosis and reduction of the clinical worsening from the pharyngeal radionecrosis. That was not feasible because of the shape and location of the FA, and the technical difficulty and inherent danger of angioplasty (covered stent) in an already structurally compromised maxillary artery.

Yuen et Al stressed the importance of collateral revascularization from the peripheral tissue bed after percutaneous embolization in order to prevent necrosis⁷. The radiation-induced tissue fibrosis, the poor collateral network and the percutaneous permanent occlusion of the pterygoid, masseteric and buccal arterial branches (second portion of the IMA) may ex-

plain the worsening of the pharyngeal tissue necrosis and the absence of healing during follow-up, that led the patient to lethal septic meningitis¹.

Conclusions

FAs presenting with epistaxis are rare entities and are mainly the result of penetrating injury to the head and neck, occurring within three weeks up to years after the trauma. We report an exceptional case of FA subacute rupture of the IMA due to radiation therapy, probably facilitated by a wound injury during biop-

sy. The fibrosis and the absence of collateral circulation in the territory of the occluded IMA may have worsened the ischemia and induced a more severe necrosis in that case. Embolization prevents immediate death from haemorrhage, yet extension of the underlying radionecrosis and/or tumoral recurrence are key factors to later development of infectious complications.

Injury to branches of the maxillary artery should be suspected in patients who present with naso-oropharyngeal tumor and delayed epistaxis after radiation therapy of the nasopharynx and should lead to selective external carotid artery injections and superselective embolizations.

References

- 1 Allen WE, Kier EL, Rothman SLG: The maxillary artery: normal arteriographic anatomy. *Radiology* 118: 517-527, 1973.
- 2 Ballantyne AJ: Late sequelae of radiation therapy in cancer of the head and neck with particular reference to the nasopharynx. *Am J Surg* 130: 433-436, 1975.
- 3 Koh E, Frazzini VI, Kagetsu NJ: Epistaxis: vascular anatomy, origins and endovascular treatment. *Am J Radiol* 174: 845-851, 2000.
- 4 Mak WK, Chow TL, Kwok SP: Radionecrosis of internal carotid artery in nasopharyngeal carcinoma presenting as epistaxis. *Aust N Z J Surg* 70: 237-238, 2000.
- 5 Piedbois P, Becquemin JP et Al: Arterial occlusive disease after radiotherapy: a report of fourteen cases. *Radiother Oncol* 17: 133-140, 1990.
- 6 Wilner HI, Lazo A et Al: Embolization in cataclysmal haemorrhage caused by squamous cell carcinomas of the head and neck. *Radiology* 163: 759-762, 1987.
- 7 Yuen JC, Gray DJ: Endovascular treatment of a pseudoaneurysm of a recipient external carotid artery following radiation and free tissue transfer. *Ann Plast Surg* 44: 656-659, 2000.

Pierre-Yves Marcy, M.D.
Department of Radiology
Centre Antoine Lacassagne
33 Avenue de Valombrose
06189 Nice Cedex 2
France
e-mail: nicolas_magne@hotmail.com