CASE REPORT

Near fatal haemorrhage 35 years after radiation for laryngeal cancer: emergency embolisation of a vertebral artery aneurysm

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The authors report a rare case of a 75 year old man who presented 35 years after radiotherapy for carcinoma of the larynx, with catastrophic bleeding through the mouth. Both the time and mode of presentation were unusual. The patient required emergency sacrifice of the left vertebral artery to stop the bleeding. The literature has been reviewed.

Osteoradionecrosis is a known complication of radiotherapy to the larynx and pharynx. It normally presents within the first year post-radiation, although there have been reports of presentations up to 50 years after treatment.¹ When present, radionecrosis usually occurs in the temporal, mandibular bones, and the larynx. It is known to present as haemorrhage intracranially,²⁻³ but not as often in the case of laryngeal and pharyngeal carcinomas. However, some authors have reported haemorrhage from the internal carotid artery⁴⁻⁷ in post-radiotherapy patients in this group.

Osteoradionecrosis is described as diffuse or localised depending on the extent of bone involvement. In the case of focused temporal bone osteoradionecrosis, the treatment of choice is conservative with regular aural toilet when the ear is affected and treatment for infections until the sequestrum separates on its own.⁷ If the involvement is diffuse, surgical procedures such as radical mastoidectomy or partial petrosectomy are recommended.⁵ The most common complication is infection and the most serious documented one is rupture of a pseudoaneurysm in the internal carotid artery.

Aneurysms of the intracranial vertebral artery are increasingly treated with endovascular interventions.⁸⁻¹⁰ This treatment option seems to be increasing in popularity and appears to be effective. It offers a minimally invasive option, which is relatively safe and can be used early in the presentation, thus reducing the risk of rebleeding as compared to conservative management. The main disadvantages are infarct of the brain distal to the occlusion and the risk of rebleed.¹⁰

We report an unusual case of haemorrhage from the extracranial course of the vertebral artery as a result of osteonecrosis of the upper cervical vertebra and skull base. This interesting case of late radiotherapy complications highlights the effectiveness of emergency endovascular parent artery occlusion as a life saving treatment modality.

CASE REPORT

A 75 year old man presented with a two week history of a left posterior pharyngeal wall ulcer and hoarseness of his voice. He had also developed pain over the left side of his face and neck, halitosis, and bad taste in the mouth. He reported to have lost two stones in weight during this period. He had left tonsillar carcinoma excised 35 years earlier and received

postoperative radiotherapy. Six years later he had a local recurrence of tumour. He, therefore, had a further course of radiotherapy. Although he had some soft tissue fibrosis there was no morbidity.

On examination he had an ulcer in the left posterior oropharyngeal wall, which measured 1.5×1 cm and swelling of the base of the tongue. Flexible laryngoscopy showed decreased mobility of the left vocal cord compared to the right one.

During the admission he had recurrent episodes of spontaneous and short lasting witnessed bleeding through the mouth, thought to be originating from the left tonsillar bed. He was treated expectantly and a CT scan was performed to assess the anatomy of the ulcer (fig 1). This showed asymmetry of the parapharyngeal soft tissues with an ill defined, air-containing mass within the paravertebral tissues. There was partial destruction of the vertebral body, including the foramen transversarium where pathological tissue was seen to encase the left vertebral artery. The contour of the artery was abnormal due to the presence of a false aneurysm.

As the bleeding through the mouth ulcer persisted, an angiogram was performed. The left common carotid artery was found to be occluded at the level of the aortic arch. The

Figure 1 Contrast enhanced CT shows an ill defined, air-containing low density mass within the left paravertebral tissues. There is partial destruction of the vertebral body, including the foramen transversarium where pathological tissue is seen to encase an enlarged irregular segment (false aneurysm) of the left vertebral artery.

Figure 2 A total of 20 platinum microcoils have been used to sacrifice the left vertebral artery immediately proximal to the false aneurysm. Note the radio-opaque markers within the oral pack.

brain was perfused by the right carotid artery and the vertebro-basilar circulation. A convincing bleeding point could not be identified on the angiogram as selective catheterisation of the left vertebral artery was not performed at this time.

The patient later developed significant haemorrhage through his mouth and was kept stable with fluids and blood. At this point the decision was taken to perform an examination under general anaesthesia as an emergency. On the way to the operating theatre, he suffered a hypoxic pulseless electrical activity arrest caused by upper airway obstruction from a blood clot. He had a rapid sequence induction and the examination was carried out. He was found to have a 3.5×1.5 cm ulcer on the left pharyngeal wall, which was 1.5 cm deep. The bleeding was arrested with diathermy and packing. The patient went to the high dependency unit for the next six days as he developed paroxysmal atrial fibrillation and aspiration pneumonia.

Five days later he underwent further angiography and embolisation of the false aneurysm that could be identified on the original CT study. A microcatheter was placed within the vertebral artery immediately proximal to the aneurysm and the parent vessel was successfully occluded using 20 detachable microcoils (fig 2). Following sacrifice of the artery complete perfusion of the cerebellum was observed via retrograde flow in the intracranial segment of the left vertebral artery (fig 3). Two days later, however, he developed a bulbar palsy, presenting as silent aspiration and speech impairment. A MR scan showed an acute posterior inferior cerebellar artery infarct but there was no radiological evidence of a medullary infarct (fig 4).

He had extensive rehabilitation and was eventually discharged home 10 weeks after his admission. He has been followed up over the last 30 months and his ulcer has

Figure 3 A right vertebral angiogram, performed after occlusion of the left vertebral artery, shows collateral flow to the intracranial territory of the left vertebral artery.

Figure 4 T2 weighted MR shows an acute non-haemorrhagic infarct of the cerebellum within the territory of the left posterior inferior cerebellar artery.

resolved completely. Moreover, his bulbar palsy symptoms have resolved as well. He has had repeat CT scans of his neck, which show no significant changes from previous ones. At present his main complaint is intractable left sided neck pain likely to be secondary to radiotherapy.

DISCUSSION

Radiotherapy is known to cause severe adverse effects, some of which present early and others which can present as late as 50 years after treatment.¹ Late presentations almost always constitute fibrosis and radionecrosis.¹¹ In the case of osteo-radionecrosis a study showed that its incidence was 8%.¹¹ All of the studies found of osteoradionecrosis after pharyngeal

Summary

Already known

- Osteoradionecrosis can present late.
- Erosion of the internal carotid has been documented.
- Endovascular stenting/embolisation of the intracranial vertebral artery is a recognised treatment.

Unique to this case

- Osteoradionecrosis rarely presents 35 years after radiotherapy.
- Erosion of the vertebral artery has not been documented.
- Embolisation of the extracranial vertebral artery has not been documented.
- Unusually our patient had a full recovery from infarct in the distribution of the posterior inferior cerebellar artery.

irradiation involved the internal carotid artery.^{4-7 11} The mortality of these cases is high. However, haemorrhage from erosion of the extracranial vertebral artery and its successful management with embolisation has never been described.

Bleeding from the mouth, as a presenting symptom of osteoradionecrosis, was also unusual and catastrophic bleeding such as our patient had is even more so. He had extensive fibrosis and a recurrence of a mass, which could not be differentiated between benign or malignant. This is known to be difficult and neither CT nor MRI scans can reliably differentiate these two diagnoses.¹² Biopsies and long term follow up have demonstrated that this mass is necrotic tissue.

The easy access to the posterior pharyngeal wall would imply that the bleeding would be amenable to surgical treatment. That is the reason this patient had surgery as a first line of treatment. However, during surgery the bleeding point was inaccessible, as it was later found to be on the vertebral artery at the skull base. Hence, the need for an endovascular approach. This line of treatment was successful, although not without complications. This patient suffered ischaemia of the posterior inferior cerebellar artery distribution. This is not uncommon,¹⁰ but what was surprising was the full recovery made in this case.

Endovascular embolisation of parent artery is a recognised treatment for dissection of intracranial vertebral artery. Traditionally, haemorrhage has been treated surgically. This paper illustrates further the advances of endovascular radiology, as well as an unusual case of successful management of vertebral artery haemorrhage on the background of delayed osteoradionecrosis.

The algorithm to follow in bleeding from the extracranial vertebral artery and, more commonly, the internal carotid artery advocated by two of the reviewed articles^{5 6} and followed by our team is: fluid resuscitation of the patient, packing of the bleeding cavity followed by an angiogram to establish the source of bleeding, and the perfusion of the brain. Once this is established the options are to embolise or ligate the vessel proximal to the aneurysm/rupture. If the brain cross circulation is inadequate after a balloon occlusion test, an ipsilateral cerebral revascularisation procedure is advisable before embolisation/ligation.⁵

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