Extracranial internal carotid artery aneurysm presenting as symptomatic hypoglossal and glossopharyngeal nerve paralysis

L. J. WILDING, M.R.C.P., D. C. HOWLETT, F.R.C.R., H. J. ANDERSON, F.R.C.R., P. D. SANGLE, F.R.C.S.*, N. VIOLARIS, F.R.C.S.*, G. H. EVANS, F.R.C.S.[†]

Abstract

Aneurysms of the extracranial portion of the internal carotid artery are rare, particularly in young patients. They usually develop following trauma, or secondary to infection involving the parapharyngeal space that extends to the vessel wall. This is a case of an internal carotid artery aneurysm presenting acutely following chiropractic neck manipulation with hypoglossal and glossopharyngeal nerve palsy. The imaging findings and subsequent operative management are described.

Key words: Aneurysm; Carotid Artery Diseases; Cranial Nerve Injuries; Hypoglossal Nerve; Glossopharyngeal Nerve

Case report

A 48-year-old woman presented to the Ear, Nose and Throat department with a one-day history of a painful swelling beneath the right side of her jaw. One month prior to presentation she had undergone three sessions of chiropractic neck manipulation. Two weeks after this she had experienced several episodes of dizziness associated with turning her head to the left. On examination she was found to have a tender swelling behind the angle of her jaw on the right and some small tender right cervical lymph nodes. Flexible nasoendoscopy at this time showed no abnormality in the posterior nasal space, larynx or hypopharynx. A provisional diagnosis of lymphadenitis was made and she was discharged with a course of antibiotics. Two days later she re-presented after experiencing further attacks of nausea and light-headedness. The pain in the right side of her neck had worsened and she had developed loss of hearing in the right ear. In addition her speech had become indistinct and she had noticed an odd sensation on swallowing, with some alteration in taste sensation. Examination at this time revealed marked tongue deviation to the right side with a painful swelling on the right side of the neck. An ultrasound of the neck was performed in the first instance but the right carotid bifurcation was not well visualized and the patient therefore proceeded to magnetic resonance (MR) imaging of the brain and neck.

MR demonstrated a large lobulated mass in the region of the carotid bifurcation, displacing the adjacent carotid vessels (Figure 1(a),(b)). The mass measured 5 cm in diameter with a thin peripheral wall that was well defined apart from medially where the lesion abutted the internal carotid artery (ICA). The centre of the mass was of low signal on T1 and T2 weighting, consistent with flowing blood, with a high signal periphery due to adjacent thrombus. These appearances were consistent with a large right ICA aneurysm. Selective right carotid angio-



FIG. 1(a)

Coronal T1-weighted MR sequence (TR/TE 616/15 msec) demonstrates the aneurysm situated at the right carotid bifurcation. This contains high signal thrombus peripherally (arrow) with the internal carotid artery displaced medially (small arrows).

From the Departments of Radiology, Otolaryngology* and Cardiology† Eastbourne District General Hospital, UK. Accepted for publication: 15 November 2003.



FIG. 1(b)

Axial T2-weighted MR sequence (TR/TE 7656/112 msec) through the aneurysm again demonstrates peripheral high signal thrombus with low signal medially consistent with slow flowing blood in the aneurysm lumen. The lumen abuts the internal carotid artery (arrow).

graphy confirmed the presence of a right internal carotid artery aneurysm (Figure 2). Left carotid angiography demonstrated normal cross filling from left to right via a patent anterior communicating artery.

The patient underwent exploratory surgery of the neck. This revealed a large ICA aneurysm extending to the skull base with the hypoglossal nerve stretched tightly across the aneurysm. The glossopharyngeal nerve was not visible within the surgical field. The surgeons were unable to get above the aneurysm to resect it and a decision to ligate the vessel was therefore made.

The common carotid artery (CCA) was exposed and punctured and a 4 French multipurpose angiography catheter inserted. Under fluoroscopic control a 0.35G hydrophilic guidewire was negotiated into the ICA and past the aneurysm neck into the carotid siphon. A carotid stent was then sited across the aneurysm neck. Following this a shunt was introduced. The distal limb was advanced over the guidewire/catheter and the proximal (CCA) limb introduced surgically. The shunt connected the lower CCA to the ICA above the aneurysm, providing a temporary intra-carotid, extra-aneurysmal bypass. The intracranial left internal carotid artery pressure was measured via the shunt and was within normal limits, allowing vessel ligation. The distal right ICA was ligated beyond the aneurysm sac and the aneurysm opened and resected. The aneurysm was saccular and measured 5.5 cm in length by 3.5 cm in width. A small hole was present within the vessel wall, which was surrounded by extensive thrombus. There was no evidence of local sepsis. Appearances were therefore consistent with a false aneurysm. The carotid shunt was removed following ICA ligation and the patient had an uncomplicated post-operative recovery.

During one year follow up the patient recovered her hearing and taste sensation and now has normal tongue movement, indicating recovery of hypoglossal nerve function.



Fig. 2

A selective right common carotid angiogram (catheter in the right CCA) demonstrates splaying of the carotid bifurcation with displacement of the external carotid artery laterally by the aneurysm (arrow). The internal carotid artery lies medially (curved open arrow) and contrast opacifies the aneurysm lumen (arrow head).

Discussion

Aneurysms of the ICA are relatively uncommon. One series documented 1118 aneurysms of peripheral arteries presenting over a 30-year period. Forty-one of these arose from the extra-cranial carotid system amongst which only four were saccular aneurysms of the ICA.¹

The major causes of extracranial carotid aneurysms are cervical injuries and atherosclerosis; followed by infection involving the parapharyngeal space, congenital anomalies (dysplasia) and mycotic infections.² Of all ICA aneurysms reported in the literature until 1925, 61 per cent were either pseudoaneurysms related to previous trauma or due to arterial 'erosion' from middle-ear infection or tonsilli-

tis.³ Extreme rotation or hyperextension of the neck can cause localized trauma to the ICA at its point of fixation at the skull base and subsequent aneurysm formation.

- This is a case report of an extracranial aneurysm of the internal carotid artery
- The case presented with progressive cranial nerve palsies
- The patient had undergone manipulation of the cervical spine by a chiropracter and a post-traumatic cause is suggested by the authors but is not proven

Such aneurysms may remain asymptomatic until their expansion causes mechanical compression of adjacent structures, in particular the hypoglossal and glossopharyngeal nerves, with resultant nerve palsy. The hypoglossal nerve emerges from the anterior condylar canal in the skull base and passes downwards between the internal carotid artery and jugular vein. Any lesion affecting the nerve results in ipsilateral paralysis (as in this case) and eventually wasting of the muscles of the tongue. The glossopharyngeal nerve leaves the skull via the jugular foramen and again courses downwards between the internal carotid artery and internal jugular vein. Complete section of this nerve results in sensory loss in the pharynx, loss of taste sensation over the posterior one third of the tongue and pharyngeal weakness.

Hypoglossal nerve paralysis secondary to ICA pseudoaneurysm⁴ and fibromuscular dysplasia resulting in ICA aneurysm and glossopharyngeal nerve compression⁵ has been reported. ICA aneurysm has also been reported as presenting with pharyngeal pain radiating to the ipsilateral ear (as in this case), due to neural irritation of the afferent pain fibres of the glossopharyngeal nerve and with syncopal episodes.⁶ However, ICA dissection,⁷ trauma and infection of the parapharyngeal space are all commoner aetiological factors in cranial nerve palsy than carotid artery aneurysm. The incidence of hypoglossal nerve palsy with ICA dissection is rare and has been reported at five per cent.⁸

Aneurysms occurring following traumatic damage to the ICA are usually false aneurysms. Cervical spine injury is a recognized cause of localized trauma to the ICA and chiropractic manipulation resulting in ICA dissection with complications such as cerebral infarction with hemiparesis and rarely death has been reported.⁹

Doppler ultrasound is the initial imaging modality of choice for the cervical carotid vessels, as this is readily available and involves no ionizing radiation. However the technique is operator dependent and further limitations include the fact that the mandible may obscure the bifurcation and hence the lower extent of an aneurysm (as in this case). Computed tomography (CT) and three dimensional time of flight MR angiography are further non-invasive methods of assessing the cervical carotid vessels where there is diagnostic difficulty. The gold standard, however, remains conventional selective carotid angiography prior to therapeutic intervention.

ICA ligation is an alternative to surgical repair, provided the patency of the circle of Willis has been established through selective angiography. In this case the shunt deployed allowed control of the aneurysm sac without the risk of cerebral embolism. ICA ligation has reduced the overall mortality from carotid aneurysms when compared with conservative management. There remains a risk of infarction following ligation due to propagation of clot or embolism with a two to four per cent stroke rate following ipsilateral ligation.¹⁰

Conclusion

Although rare, ICA aneurysms should be considered in the differential diagnosis of a mass in the anterior triangle of the neck and their presence confirmed with ultrasound supplemented by MR/conventional angiography. In this case an ICA pseudoaneurysm presented following a history of chiropractic manipulation, indicating a possible post-traumatic cause. Pressure effects may result in such aneurysms presenting as cranial nerve palsies, particularly involving the hypoglossal and glossopharyneal nerves. Creation of an intra-operative shunt (as in this case) both maintains brain perfusion during surgical excision and confirms patency of the circle of Willis. In aneurysms not amenable to surgical excision, ICA ligation is the accepted treatment and can prevent permanent nerve damage.

Acknowledgement

We would like to thank Mr G. Manjaly for his input with the preparation of this paper.

References

- 1 Welling RE, Taha A, Goel T, Cranley J, Krause R, Hafner C, *et al.* Extracranial carotid artery aneurysms. *Surgery* 1983;**93**:319–23
- 2 Webb RC, Barker WF. Aneurysms of the extracranial internal carotid artery. *Arch Surg* 1969;**99**:501–5
- 3 Winslow H. Extracranial aneurysm of the internal carotid artery: history and analysis of the cases registered up to August 1, 1925. Arch Surg 1926;**13**:689–729
- 4 Nusynowitz RN, Stricof DD. Pseudoaneurysm of the cervical internal carotid artery with associated hypoglossal nerve paralysis. Demonstration by CT and angiography. *Neuroradiology* 1990;**32**:229–31
- 5 Lane RJ, Weisman RA, Savino PJ, Schatz NJ. Aneurysm of the internal carotid artery at the base of the skull: an unusual cause of the cranial neuropathies. *Otolaryngol Head Neck Surg* 1980;**88**:230–2
- 6 Lim YM, Lee SA, Kim DK, Kim GE. Aneurysm of the extracranial internal carotid artery presenting as the syndrome of glossopharyngeal pain and syncope. *J Neurol Neurosurg Psychiatr* 2002;**73**:87–8
- 7 Struzzeneger M, Huber P. Cranial nerve palsies in spontaneous carotid artery dissection. *J Neurol Neurosurg Psychiatry* 1993;**56**:1191–9
- 8 Ursekar MA, Singhal BS, Konin BL. Hypoglossal nerve palsy due to spontaneous dissection of the internal carotid artery. *Clin Radiol* 2000;978–9
- 9 Peters M, Bohl J, Thomke F, Kallen KJ, Mahlzahn K, Wandel E, *et al.* Dissection of the internal carotid artery after chiropractic manipulation of the neck. *Neurology* 1995;**45**:2284–6
- 10 Nicolls SC, Kohler TR, Bergelin RO, Primozich JF, Lawrence RL, Strandness DE Jr. Carotid artery occlusion: Natural history. J Vasc Surg 1986;4:479–85

Address for correspondence: Dr Lucy Wilding, 5 Gordon Road, Surbiton, Surrey KT5 9AR, UK.

E-mail: graemelucy@supanet.com

D C Howlett takes responsibility for the integrity of the content of the paper. Competing interests: None declared