



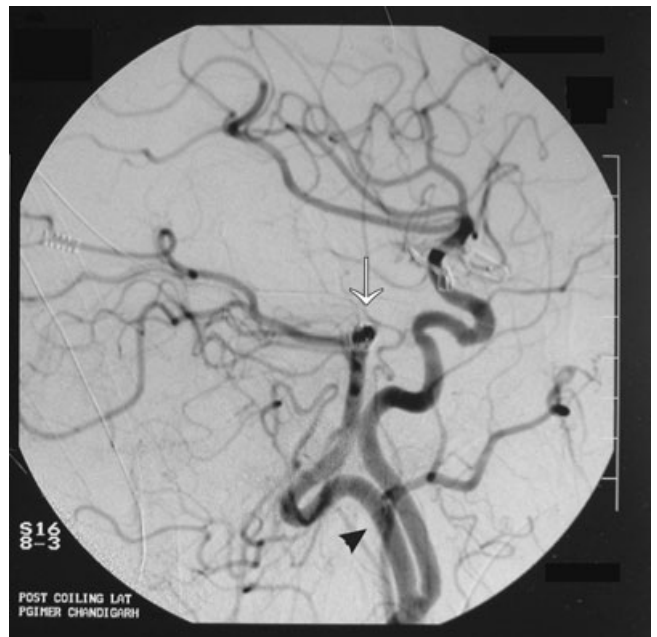
**Fig. 1.** Right internal carotid angiogram lateral view: catheter in the proximal internal carotid artery (arrow). The persistent hypoglossal artery (arrow head) leads to the basilar artery with the aneurysm at the basilar top (open arrow). Note the absent posterior communicating artery from the carotid siphon.



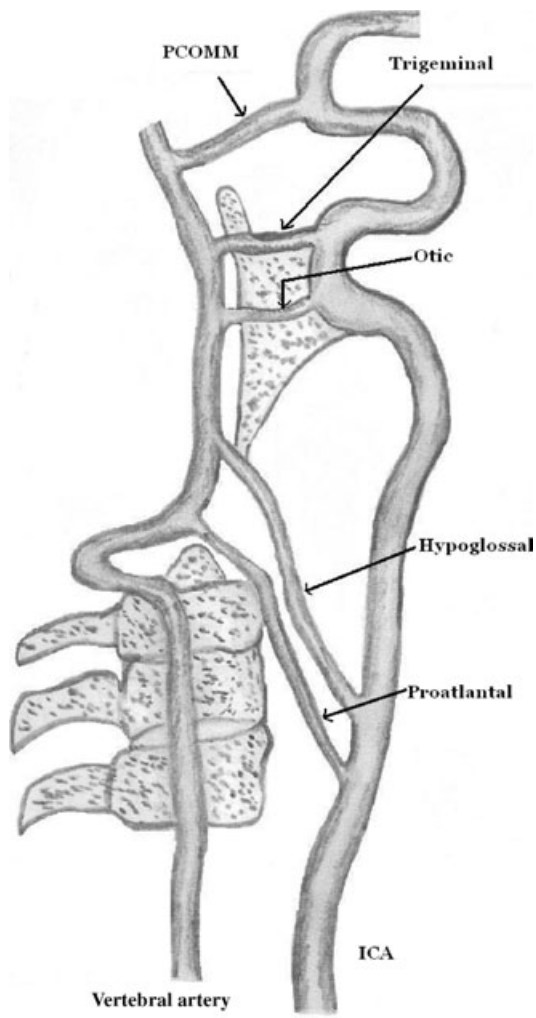
**Fig. 3.** Post GDC coiling right carotid angiogram, right anterior oblique view shows complete occlusion of the basilar top aneurysm (open arrow). GDC, Guglielmi detachable coils.



**Fig. 2.** Left anterior oblique view of the right internal carotid artery angiogram shows the basilar top aneurysm (open arrow). Overlap of the right middle cerebral artery and the posterior cerebral artery is present in this projection.



**Fig. 4.** Post GDC coiling right carotid angiogram, lateral view shows complete occlusion of the basilar top aneurysm (open arrow). GDC, Guglielmi detachable coils.



**Fig. 5.** Schematic diagram of persistent fetal carotid-vertebrobasilar anastomoses. ICA, internal carotid artery; PCOMM, posterior communicating artery.

# Basilar artery aneurysm treated with coil embolization via persistent primitive hypoglossal artery

JR Bapuraj,<sup>1,3</sup> V Ojili,<sup>1</sup> N Khandelwal,<sup>1</sup> AKP Shanbhogue<sup>1</sup> and SK Gupta<sup>2</sup>

Departments of <sup>1</sup>Radiodiagnosis and <sup>2</sup>Neurosurgery, Postgraduate Institute of Medical Education and Research (PGIMER), Chandigarh, India

<sup>3</sup>Division of Neuroradiology, Department of Radiology, University Michigan, Ann Arbor, MI, USA

## SUMMARY

A saccular aneurysm at the basilar artery bifurcation associated with a persistent primitive hypoglossal artery (PPHA) was successfully treated by endovascular occlusion with Guglielmi detachable coils. As both vertebral arteries were aplastic, a microcatheter was advanced via PPHA. To the best of our knowledge, this is the first case report describing the treatment of a basilar top aneurysm through the PPHA.

**Key words:** aneurysm; Guglielmi detachable coil; persistent primitive hypoglossal artery.

## INTRODUCTION

The primitive hypoglossal artery is the second most common persistent carotid–vertebrobasilar anastomosis with a reported incidence of 0.02–0.1%.<sup>1</sup> The most important clinical feature of persistent primitive hypoglossal artery (PPHA) is its association with intracranial vascular anomalies such as aneurysms.<sup>2–5</sup> Surgery has been the standard treatment for these aneurysms.<sup>2</sup> We present a rare case of ruptured basilar top aneurysm which was treated by endovascular occlusion with Guglielmi detachable coils (GDC) via the PPHA. To the best of our knowledge, this is the first case report describing the endovascular treatment of an aneurysm through the PPHA.

## CASE REPORT

A 60-year-old woman patient presented with sudden severe headache associated with nausea and vomiting. Neurological examination revealed impaired consciousness and marked neck rigidity. Her medical history and family history were unremarkable. She had undergone surgical clipping of an anterior communicating artery aneurysm 2 years back in another hospital. The exact details of the surgery and other investigations performed at that time were unknown.

Computed tomography showed subarachnoid haemorrhage which was most conspicuous in the interpeduncular cistern, suggesting an aneurysm of the basilar artery. Digital subtraction angiography showed aplastic vertebral artery bilaterally and a PPHA arising from the cervical part of the right internal carotid artery supplied the basilar artery. There was a 5-mm saccular aneurysm at the basilar artery bifurcation (Figs 1,2).

Under general anaesthesia, the aneurysm was treated by the GDC embolization. The procedure was performed via the right transfemoral route, first positioning a 6F guiding catheter (Fasguide, Boston Scientific Corporation, Fremont, CA, USA) in the right internal carotid artery, followed by coaxial introduction of an Excel 14 microcatheter (Boston Scientific Corporation, Fremont, CA, USA). The microcatheter was advanced to the aneurysm via the PPHA and then three GDC 10 coils (Boston Scientific Cor-

poration, Fremont, California, USA) were positioned within the aneurysm and electrolytically detached. An immediate post-embolization angiogram demonstrated complete occlusion of the aneurysm (Figs 3,4).

## DISCUSSION

Persistent primitive hypoglossal artery is the second most frequent carotid–basilar anastomoses after persistent trigeminal artery, which makes up the vast majority of persistent primitive connections, whereas the persistent otic and proatlantal intersegmental arteries are less frequent. The first case of PPHA was reported by Batujeff in an autopsy case in 1889. Lindgren in 1950 demonstrated the first case of PPHA by angiography. Since then, approximately 160 cases have been reported in the literature.<sup>6</sup>

The embryology and anatomy of the persistent fetal carotid–basilar anastomoses was described by Padget in 1948.<sup>7</sup> During the 4-mm embryo stage, the forebrain is supplied by the carotid system, and along the surface of the hindbrain are two parallel longitudinal neural arteries, which eventually fuse to form the basilar artery. The paired longitudinal neural arteries are supplied by the carotid arteries via four important arterial anastomoses, namely the trigeminal, otic, hypoglossal and proatlantal intersegmental arteries. During the 5–6-mm embryo stage, an anastomosis forms between the distal internal carotid artery and corresponding longitudinal neural artery, which becomes the posterior communicating artery. Subsequently, the presegmental arteries (i.e. the trigeminal, otic and hypoglossal arteries) and proatlantal intersegmental artery regress and obliterate. The first to obliterate is the otic artery, followed in order by the hypoglossal, trigeminal and proatlantal intersegmental arteries. Failure of obliteration of any of these arteries results in a persistent carotid–vertebrobasilar anastomosis (Fig. 5).

Lie has reported four criteria for the anatomic and angiographic definition of PPHA: (i) it arises from the cervical part of the internal carotid artery at the C1–C3 level; (ii) it enters the skull via the hypoglossal canal together with the XIIth cranial nerve; (iii) the basilar artery is filled only beyond the point where the

J Bapuraj MD, PDCC; V Ojili MD, DNB; N Khandelwal MD, DNB; AKP Shanbhogue MBBS; SK Gupta M.Ch.

Correspondence: Dr Jayapalli R. Bapuraj, Department of Radiology, Division of Neuroradiology, 1500 E. Medical Center Dr UHB2-A2Q9 Ann Arbor, MI 48109-5030, USA.

Email: jrajiv.bapuraj@gmail.com, jrj@med.umich.edu

Submitted 9 January 2007; accepted 9 April 2007.

doi: 10.1111/j.1440-1673.2007.01759.x

hypoglossal artery enters it; and (iv) the posterior communicating artery is absent and the ipsilateral vertebral artery is hypoplastic.<sup>8</sup>

As the presence of PPHA may be completely asymptomatic, it may appear as an incidental finding in cerebral angiogram performed for another diagnostic purpose. However, its identification is clinically important before carotid endarterectomy or skull base surgery. This is obviously related to the fact both the anterior and posterior cerebral circulation is dependent on the arterial supply of internal carotid artery. Persistent primitive hypoglossal artery has been reported to cause glossopharyngeal neuralgia and glossopharyngeal nerve paralysis. But the most important clinical feature of PPHA is its association with intracranial aneurysms.<sup>2-5</sup> It has been suggested that PPHA may be associated with anomalous structure of the vessel wall and exposes the basilar trunk to unusual haemodynamic stress predisposing to the onset of aneurysms.

A review of previously reported cases indicates no consistent symptoms or signs that would suggest the existence of PPHA and it is usually an incidental finding during angiography. There have been reports of associated aneurysms of the anterior cerebral artery, internal carotid artery, intracranial carotid bifurcation, basilar artery bifurcation and the PPHA itself at its junction with the basilar artery or posterior inferior cerebellar artery.<sup>2-5</sup>

Surgery has been the standard treatment for aneurysms associated with PPHA.<sup>2</sup> In surgically inaccessible aneurysms, proximal clipping or ligation of PPHA which is the parent artery of the aneurysm has been considered to be an alternative operative treatment. However, these operations should be avoided because the vertebral arteries are usually hypoplastic or aplastic on both sides and the posterior communicating arteries are functionally absent.

Basilar artery bifurcation aneurysms are difficult to treat surgically because they are located deep in front of the brainstem and close to important cranial nerves and perforating vessels. Indications for open surgery may differ among institutions and depend on the level of experience of the neurosurgeons. Indications for aneurysmal embolization in our institute include surgical inaccessibility, failed clipping, wide aneurysm neck and a medical condition precluding craniotomy. Bavinzski *et al.*<sup>9</sup> reported that the use of GDC coils led to excellent clinical and angiography results in the majority of 45 patients with basilar artery bifurcation aneurysms.

To the best of our knowledge, there has been no report of endovascular treatment of basilar top aneurysm via the PPHA. However, there is a single case report by Ikushima *et al.*<sup>10</sup> where a basilar artery aneurysm has been treated by endovascular occlusion with GDC via the persistent primitive trigeminal artery. In our case, as well as in that reported by Ikushima *et al.*,<sup>10</sup> both vertebral arteries were aplastic and the fetal carotid-vertebrobasilar anastomoses were the only route to approach the aneurysms and excellent results were obtained in both the cases.

## REFERENCES

1. Hahnel S, Hartmann M, Jansen O, Sartor K. Persistent hypoglossal artery: MRI, MRA and digital subtraction angiography. *Neuroradiology* 2001; **43**: 767-9.
2. Yokota N, Yokoyama T, Ryu H. Aneurysm of persistent primitive hypoglossal artery. *Br J Neurosurg* 1999; **13**: 608-10.
3. Huynh-Le P, Matsushima T, Muratani H, Hikita T, Hirokawa E. Persistent primitive hypoglossal artery associated with proximal poste-

rior inferior cerebellar artery aneurysm. *Surg Neurol* 2004; **62**: 546-51.

4. Sakai K, Tanaka Y, Tokushige K, Tanabe A, Kobayashi S. Basilar bifurcation aneurysms associated with persistent primitive hypoglossal artery. *Neurosurg Rev* 1998; **21**: 290-4.
5. Yeh H, Heiss JD, Tew JM Jr. Persistent hypoglossal artery associated with basilar artery aneurysm. *Neurochirurgia* 1987; **30**: 158-9.
6. Luh GY, Dean BL, Tomsick TA, Wallace RC. The persistent fetal carotid-vertebrobasilar anastomoses. *AJR* 1999; **172**: 1427-32.
7. Padgett DH. The development of the cranial arteries in the human embryo. *Contr Embryol* 1948; **32**: 205-61.
8. Lie TA. Primitive hypoglossal artery. In: *Congenital Anomalies of the Carotid Arteries*. Excerpta Medica Foundation, Amsterdam, 1968; 76-83.
9. Bavinzski G, Killer M, Gruber A, Reinprecht A, Gross CE, Riehling B. Treatment of basilar artery bifurcation aneurysms by using Guglielmi detachable coils: a 6-year experience. *J Neurosurg* 1999; **90**: 843-52.
10. Ikushima I, Arikawa S, Korogi Y, Uehara H, Komohara Y, Takahashi M. Basilar artery aneurysm treated with coil embolization via persistent primitive trigeminal artery. *Cardiovasc Intervent Radiol* 2002; **25**: 70-1.