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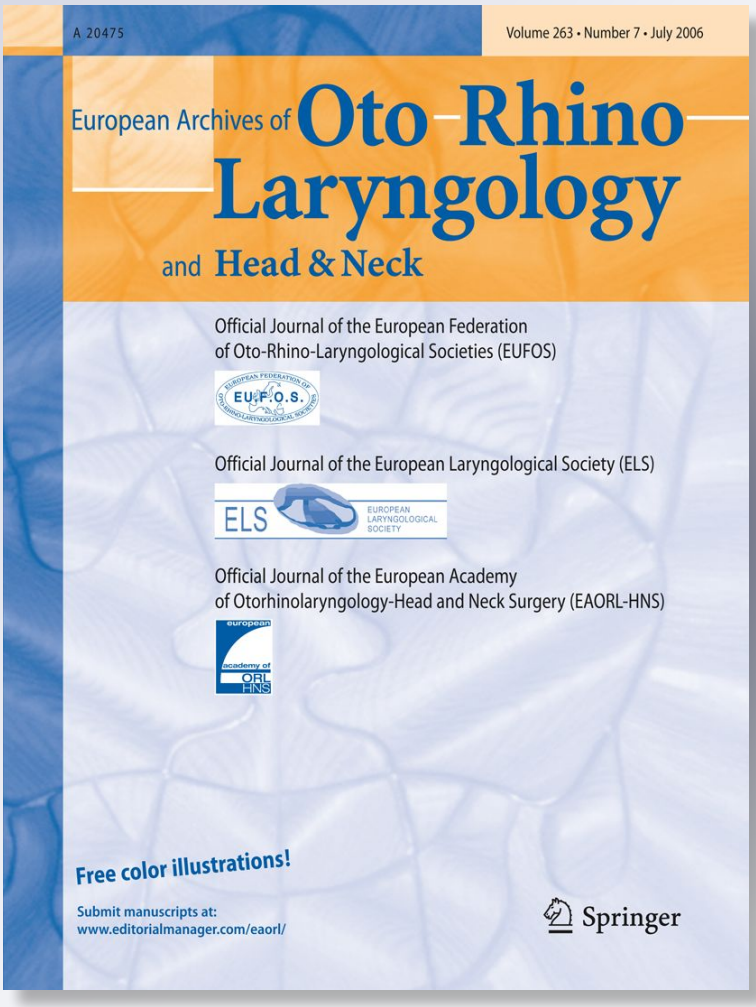

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CASE REPORT

The use of internal carotid artery stenting in management of bilateral carotid body tumors

Masaya Konishi · Paolo Piazza · Seung-Ho Shin · Shailendra Sivalingam · Mario Sanna

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Introduction

Carotid body tumors (CBT) are paragangliomas that arise from chemoreceptors at the carotid bifurcation with surgical removal indicated in the majority of cases[1, 2]. Surgical resection remains complicated by bleeding from the tumor, possible internal carotid artery (ICA) injury, cranial nerve injuries and cerebral ischemic events. In Shamblin type III tumors, the likelihood of vascular injury increases [1]. Therefore facilities for vascular repair, reconstruction, on table balloon occlusion and rapid transfer to the neuroradiology suite should be available for all CBT. ICA stenting is a good option to reduce intraoperative injury and we have utilized this technique for complex tympanojugular paragangliomas without postoperative complications [3–5]. We report a case of large bilateral carotid body tumors where bilateral uncomplicated surgical removal was performed in a staged fashion with the use of uncovered ICA stents.

Case report

A 35-year-old woman with bilateral neck swellings, dysphonia and a family history of paragangliomas was referred to us. Fiberoptic laryngoscopy revealed a narrow oropharynx with bilateral fullness and paresis of the left vocal cord. MRI showed bilateral Shamblin type III tumors completely encasing the internal and external carotid arteries measuring 3.0 × 3.0 × 4.5 cm on the right side and 3.5 × 3.5 × 4.5 cm on the left side (Fig. 1).

Bilateral carotid arteriograms showed intense tumor blush at the level of bifurcation (Figs. 2a; 3a). The left tumor was removed first. A self expanding stainless-steel stent (Carotid Wall Stent® Boston Scientific company, Natick, Massachussets, USA) was inserted into the left ICA with the aims of reinforcing the arterial wall at 3 months before operation (Figs. 2b, c). Antiplatelet therapy using a combination of clopidogrel (75 mg/day) and aspirin (100 mg/day) was commenced 5 days before the stent insertion and continued for 1 month after stenting and then reduced to single-drug treatment with aspirin only. Antiplatelet agents were stopped and low molecular weight heparin (LMWH) commenced 5 days before surgery. Antiplatelet agents were introduced 2 days after surgery and LMWH was stopped 3 days after surgery.

After formation of a stabilized neointimal lining on the luminal surface of the stent, the excision of the left CBT was performed via a retroauricular-transcervical approach. It was difficult to demarcate between the tumor and the ICA due to tumor encasement. The External Carotid Artery together with its branches was ligated and divided in order to mobilize and rotate tumor adherent to the ICA. During tumor dissection, the stent-reinforced arterial wall allowed subadventitial dissection with little risk for the ICA injury and the bloodless surgical field enabled cranial nerve preservation. Ten months after the first stage surgery, the same stent was deployed in the right ICA (Figs. 3b, c). Then three months after the stent deployment, the right CBT was successfully excised through an identical approach. The operation was performed using the same subadventitial technique used in the previous surgery. The branches of the right ECA were also ligated and divided to achieve complete removal of the tumor.

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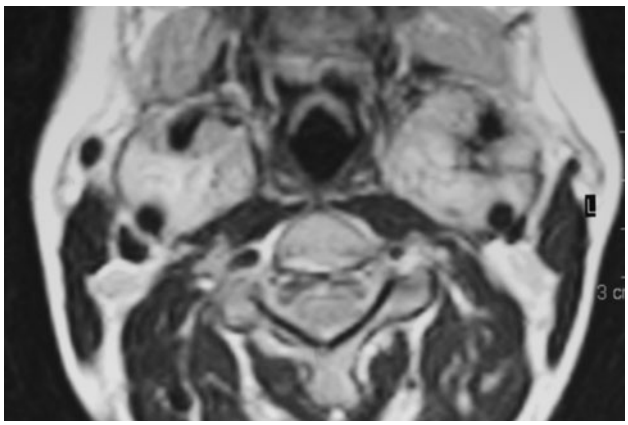

Fig. 1 Bilateral tumors are seen on MRI at the level of carotid bifurcation with medialization of the oropharyngeal walls. The tumors encase the internal carotid arteries

Postoperatively she showed no functional deficits including the lower cranial nerve palsy. However, during the immediate postoperative period, the blood pressure rose to 160/97 mmHg and the pulse rate was maintained at an average of 120 per minute. The tachycardia and the blood pressure gradually stabilized after 1 week without medication. At discharge, the blood pressure was 130/80, but pulse rate ranged from 100 to 110 per minute. One year after surgery, the blood pressure had recovered to normal and the pulse rates ranged from 80 to 90 per minute. High resolution CT at 6 and 18 months after surgery demonstrated bilateral patency of the ICA (Fig. 4). On the left side stent shortening was seen at 6 months after surgery, but the shortening was not proceeding afterwards. Contrast-enhanced MRI at 6 and 18 months after surgery demonstrated no tumor recurrence. The patient has thus far been free from disease for 3 years.

Discussion

For excision of bilateral tumors, the surgery should be staged to avoid the possible risk of lower cranial nerve injury complicating the initial procedure and if there are severe lower cranial nerve deficits after the initial surgery, the second procedure is contraindicated.

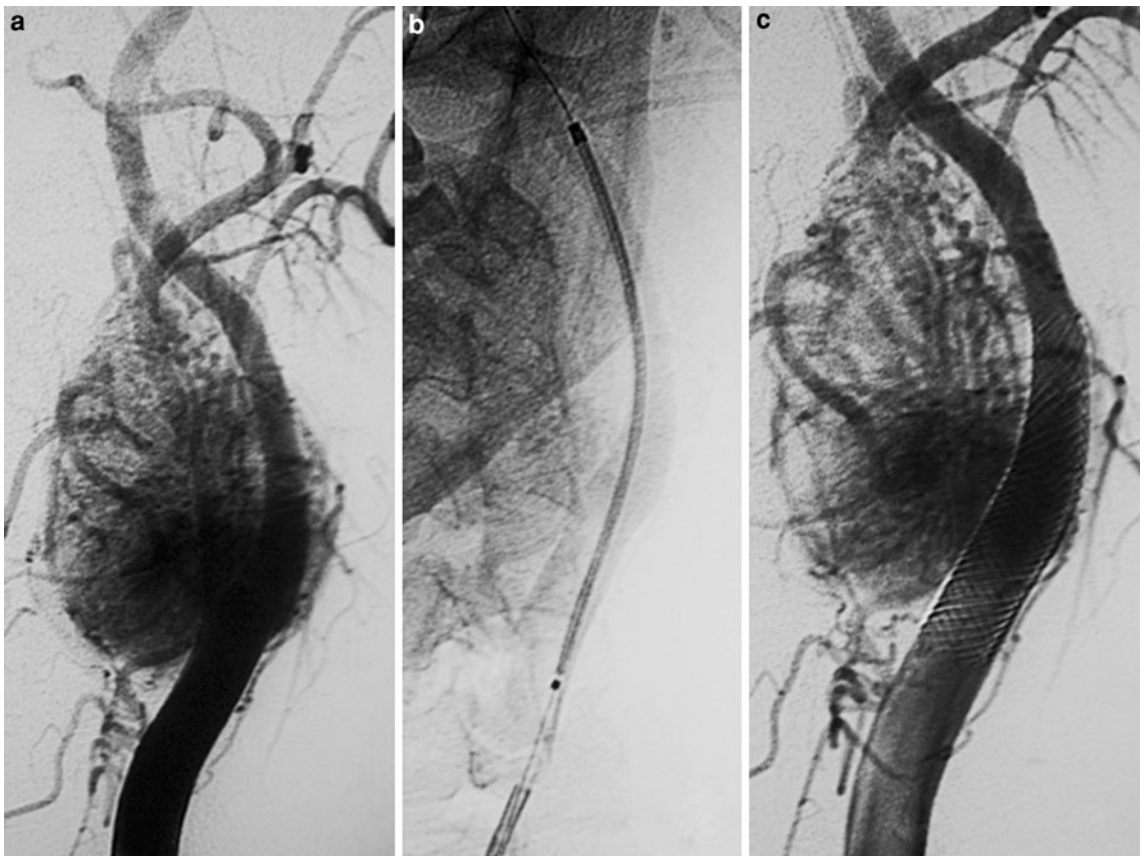

Fig. 2 Angiogram showing tumor blush at the level of left carotid bifircation (**a**) and the self-expanding stent within the left internal carotid artery covering the bifurcation (**b** before deployment, **c** after deployment)

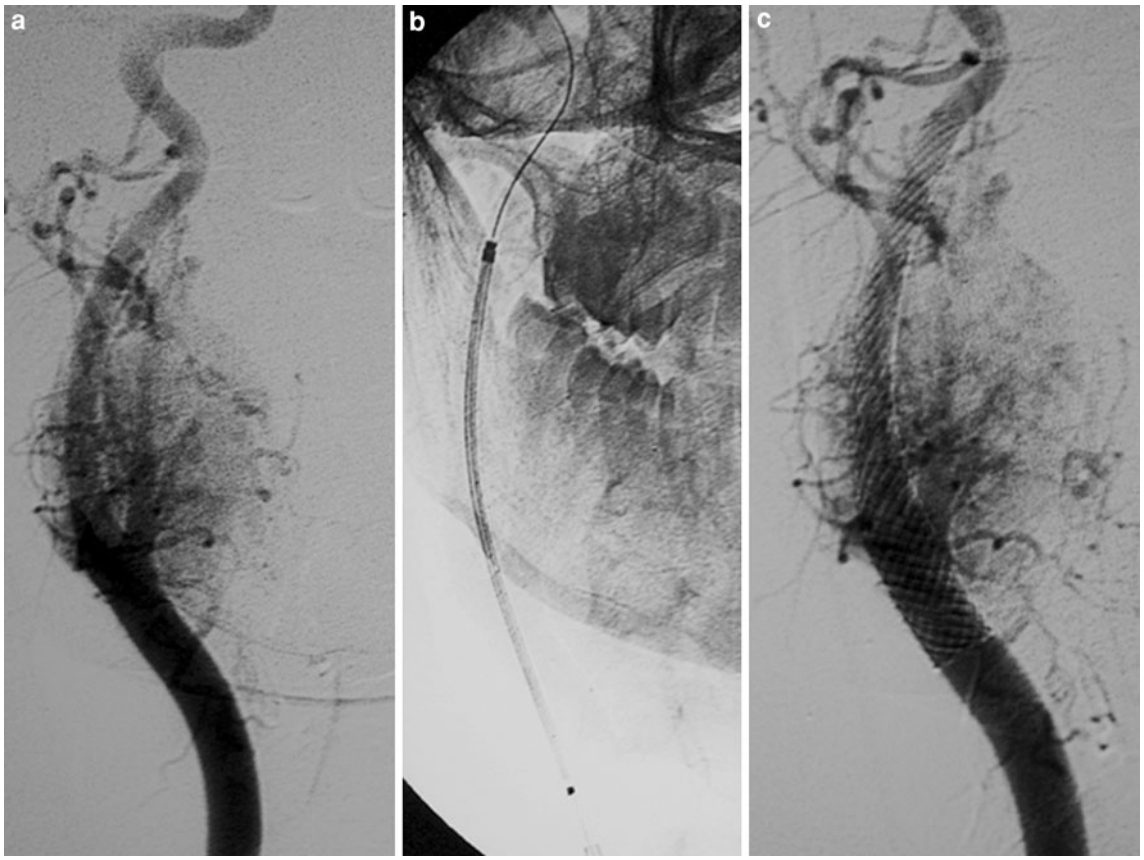

Fig. 3 Angiogram showing tumor blush at the level of right carotid bifircation (**a**) and the self-expanding stent within the right internal carotid artery covering the bifurcation (**b** before deployment, **c** after deployment)

The tumors originating from chemoreceptors can be best excised by dissection in the subadventitial plane. Often, this method can result in baroreceptor failure [6] with significant tachycardia and hypertension, which are the result of baroreceptor or carotid sinus nerve ablation [7]. However, it has been reported that severe hypertension and tachycardia are less frequent long-term complications after removal of bilateral carotid body tumors [8]. In this patient the immediate postoperative hypertension and tachycardia subsided 1 year later.

Vascular injury and cranial nerve injuries are major morbidities related to removal of carotid body tumors. However, there is a vast difference in the reported morbidity rates [9] ranging from 0 to 40%, [10–12] which are linked to the size and the anatomic location of the tumors [11]. A higher risk of vascular injury is associated with Shamblin type III tumors. While some authors report that preoperative embolization of feeding vessels from the carotid artery minimizes complications especially intraoperative bleeding, [13] recent reports also indicate that preoperative selective embolization does not lead to significant reduction in intraoperative blood loss, [14] and the risks of cerebrovascular accidents still exist secondary to embolization [15]. Therefore the role of these adjuvant treatments still remains controversial.

In our institute, stents have been utilized for management of the ICA in paraganglioma surgery with the primary aim to reinforce the arterial wall and not for devascularisation of the tumor [16]. It provides a robust dissection plane allowing the surgeon to perform a safe subadventitial dissection with easy manipulation of the artery [3]. Self-expanding stents are less likely to undergo plastic deformation and are able to track when advanced through tortuous areas after the carotid bifurcation [17].

The main risks associated with stenting are distal embolization, thrombosis [18] and the potential risks associated with lifelong antiplatelet therapy. ICA stenting requires regular follow up to detect stenosis in stent as well as to identify recurrence and metastasis of the tumors. Since 1988 we have performed 19 cases of stenting in patients with tympano-jugular and vagal paragangliomas of the head and neck, and stenosis has not been seen in any patient at 5 years follow-up via duplex ultrasonography (US). In this case, we have not detected any sign of stenosis in the bilateral stents. Stent shortening was seen on the left side, but the shortening is not proceeding and we speculate

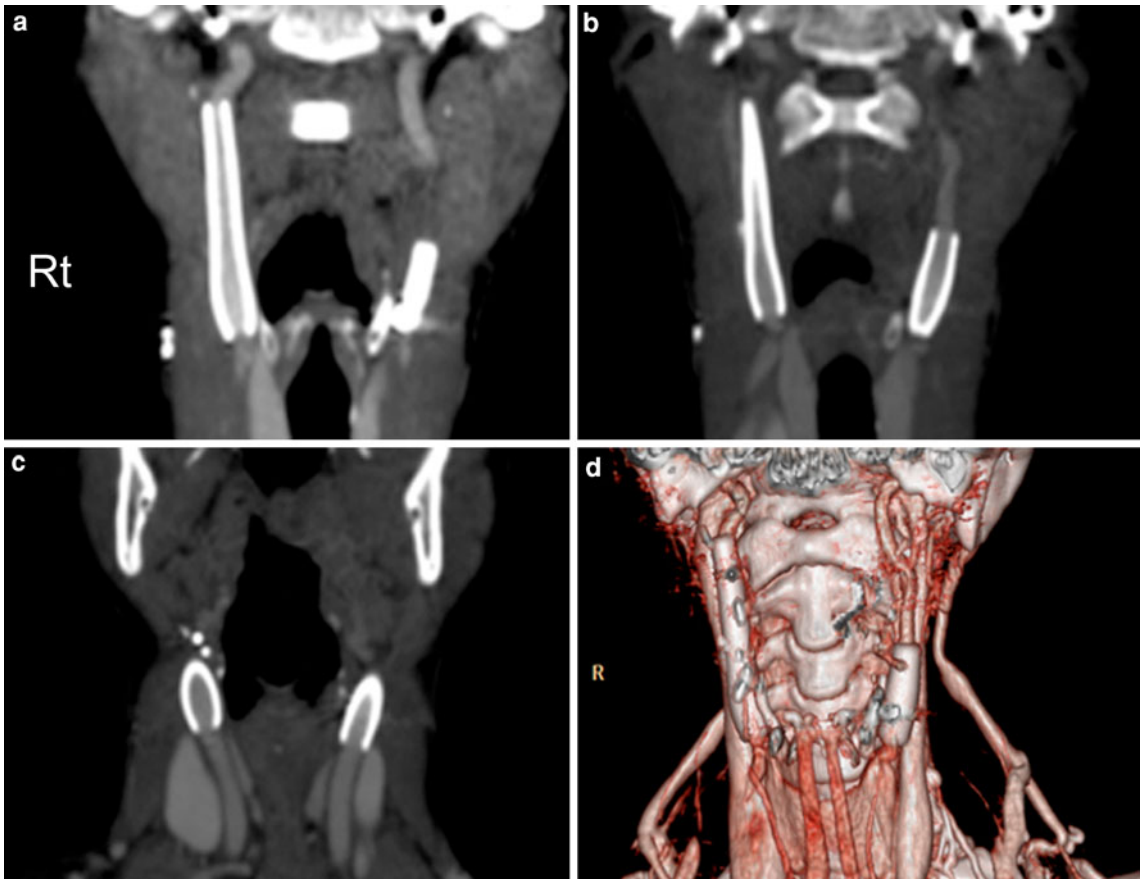

Fig. 4 Postoperative high resolution CT scan shows bilateral stable deployment of the inserted stents (**a–c** coronal view, **d** 3-dimensional reconstruction)

that it was done during the surgery. Any complication due to stent shortening is not seen so far, but careful observation is also required. In cases where the stent cannot be inserted into the ICA due to its tortuosity or stenosis by tumor, permanent balloon occlusion and bypass should be considered [3].

Conclusion

In cases of Shamblin type III CBT with involvement of the intimal layer, with the artificial neo-vessel consisting of a struts armed neointima which allows to preserve cerebral blood flow and continuity of ICA from surgical injuries, surgery can be performed without complications. In bilateral CBT cases as we have reported, stenting is an extremely useful adjunct to surgery facilitating staged surgical excision.

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Conflict of interest None.

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