CASE REPORT

The known unknowns of perioperative stroke during carotid body tumour resection

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SUMMARY

Carotid body tumour (CBT) surgery has a risk of stroke. A 57-year-old lady presented with lethargy and generalised limb pain secondary to a right parathyroid adenoma. CT scan demonstrated as an incidental finding of a left CBT. Intraoperatively the CBT was infiltrating the vessel wall. The carotid bifurcation was resected and reconstructed using a reversed saphenous vein graft. Postoperatively the patient developed right arm weakness. A CT scan showed a left hemispheric watershed infarct. On discharge (day 8 postoperatively) she had no functional deficit. Reconstruction of the internal carotid artery in patients with CBT is associated with an increased risk of stroke. This index case and literature research highlight the knowns and unknowns on stroke associated with CBT resection.

BACKGROUND

The first case report of a carotid body tumour (CBT) was by Von Haller¹ in the year 1743. The incidence is very rare and the aetiology is unknown. Rarer still is the occurrence of both a CBT and parathyroid adenoma (PA) with few reported in the literature. The difficulty of surgical resection is highly documented with complications of carotid artery injury, stroke and cranial nerve dysfunction being the most serious.² CBT may be classified according to the Shamblin classification. Type I tumours are well localised and do not surround any vessels, type II tumours are adherent or partly surround the vessel and finally type III tumours are large and encase the vessel.³ Our case reports the synchronous resection of a CBT and PA. Postoperatively the patient developed right arm weakness. A CT scan showed a left hemispheric watershed infarct. There has been a very little focus in the literature on the aetiology and prevention of stroke associated with the resection of CBT. The purpose of this report is to review the literature and provide recommendations on stroke prevention in CBT surgery.

CASE PRESENTATION

A 57-year-old housewife presented with symptoms of lethargy and chronic upper and lower limb pain for several months. Her medical history included type II diabetes and hypothyroidism. Her mother had primary breast cancer and her father had primary liver cancer in their later years. On examination at initial presentation she was clinically obese with a palpable swelling in the right thyroid region. Results of clinical examination including a

detailed neurological examination were otherwise unremarkable.

INVESTIGATIONS

Neck contrast CT scan showed an enhancing structure in the inferior pole of the thyroid gland consistent with a PA. It also showed a tissue mass (16×15 mm) with increased vascularity encasing the left carotid bifurcation, with splaying of the internal carotid artery (ICA) and the external carotid artery (ECA). These findings are suggestive of a CBT. The right carotid bifurcation was normal.

DIFFERENTIAL DIAGNOSIS

The differential diagnosis of CBTs include superficial lipoma, enlarged lymph nodes, thyroid tumour, parathyroid tumour, metastatic cancer of a distant origin, branchial cyst and carotid artery aneurysms. A detailed history and physical examination can eliminate most of these causes.

TREATMENT

Synchronous resection of both the parathyroid gland and CBT was performed. Through a collar incision the right inferior parathyroid gland was excised. Subsequently, the incision was extended into a hockey stick anteriorly to the sternocleidomastoid, leaving a small skin bridge. The CBT was infiltrating the vessel wall with no subadventitial plane present for complete excision (Shamblin III) (figure 1). The carotid bifurcation was resected and reconstructed using a reversed saphenous vein interposition graft (figure 2). Anastomosis of the ECA and the ICA was performed over a Pruitt-Inahara shunt. Ischaemic carotid clamp time totalled 3 min with no hypotension. Histology confirmed the CBT to be infiltrating the adventitia. Postoperatively the patient developed right arm weakness. A CT scan showed a small left hemispheric watershed infarct. A postoperative T2 diffusion weighted MRI showed absence of the posterior communicating cerebral artery as well as a cortical border zone infarction and perifocal oedema measuring 2.6×2.8 cm between the vascular territories of the middle cerebral arteries and the anterior cerebral arteries (figure 3).

OUTCOME AND FOLLOW-UP

On discharge (day 8 postoperatively) she had no functional deficit. At a 2-month follow-up she was fully recovered. She has had no further symptoms for the last 2 years.

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Figure 1 An operative photograph showing how CBT was infiltrating the vessel wall with no subadventitial plane present for complete resection.

DISCUSSION

CBT and PA are neuroectodermal in origin indicating their rarity. It remains unclear whether their concurrent presentation is related or coincidental. Owing to the hereditary predisposition of CBT, screening was offered for CBT and multiple endocrine neoplasia to our patients' family but they declined.

The risks of stroke in CBT surgery has declined with earlier studies having higher risks than more recent ones. Hallet *et al*⁴ compared perioperative stroke rates in CBT resection from time period I (1935–1965) to time period III (1976–1986) where a decrease in stroke was found from 23% to 2.7%. Variable risks of stroke have been shown in the last 10 years from 0% to 25%. Huang *et al*⁵ in 2001 with 30 patients showed 0% stroke rates, unfortunately the Shamblin classification is unknown in these patients. Gwon *et al*⁶ showed a 25% risk of stroke with 15 patients, 75% of the strokes were Shamblin III. Plukker *et al*⁷ showed an 8% stroke risk, all strokes were Shamblin III.

Unfortunately not all groups use the Shamblin classification system. Of the studies reviewed, the higher the Shamblin classification, the higher the risks of stroke.^{6,7} A review of 15 patients with Shamblin III found higher risks of postoperative neurovascular complications.⁸ Morbidity and death rates associated with CBT resection without sacrifice of the ICA range from 0% to 5%.^{9,10} Hallet *et al*⁴ reviewed 153 tumours and found higher rates of stroke in patients requiring arterial repair (20% vs 4.5%); fewer strokes were found during preservation of the ICA blood flow. Risk factors for stroke during CBT surgery are large tumours and any ICA reconstruction, ligation or repair.⁶

There may be several factors that reduce stroke risks in patients. Preoperative planning by cross-sectional vascular imaging in the form of CT and MRI is required.¹¹ In a recent

Figure 3 A postoperative T2 diffusion weighted MRI shows a cortical border zone infarction and perifocal oedema between the vascular territories of the middle cerebral arteries and anterior cerebral arteries.

study where preoperative embolisation was performed in 21 patients 48 h prior to surgery, the median blood loss, operation time and hospital stay for these patients were significantly reduced compared with those without embolisation.¹² Reported benefits of perioperative embolisation also include a reduction in tumour size and improved visualisation theoretically reducing neurological morbidity by lessening the risk of stroke and damage to cranial nerves. These new advances should certainly be considered preoperatively. However, others have reported procedure-related stroke during preoperative embolisation even in experienced operators, thus this technique is not currently a standard that is widely accepted.¹³ Intraoperative electroencephalographic monitoring can be useful.¹⁴ Surgical technique is important; early proximal and distal control of the carotids reduce stroke rates.⁴ As in our case study selective shunting of the ICA during carotid reconstruction can reduce stroke risks.⁵ Early ECA division in patients with Shamblin class II and III tumours may minimise stroke risk.⁶

The cause of stroke was unclear in our patient. A watershed infarct was shown in the left hemisphere with CT. The carotid clamp time was only 3 min with no period of hypotension perioperatively and minimal blood loss. As CBTs are highly vascular there is a high risk of major blood loss. The amount of blood loss should be reported as a potential mechanism that may contribute to the development of a watershed infarct. The MRI postoperatively did show an incomplete circle of Willis with the absence of the posterior communicating artery which may have been a contributing factor. A lack of potential collaterals in the case of ongoing ischaemia may have led to an infarct. There were no signs of pre-existing intracranial atherosclerosis seen on MRI and the patient had no history of transient ischaemic attacks or stroke. There was no evidence of narrowing or occlusion of any intracerebral arteries on CT or MRI that could have explained an underlying stenotic lesion which may have provoked brain ischaemia. Advanced vascular neuroimaging such as CT angiogram or MR angiogram is lacking. Careful planning preoperatively with vascular imaging of the cerebral vasculature

Figure 2 An operative photograph showing the carotid bifurcation was resected and reconstructed using a reversed saphenous vein graft.

should be performed to provide the patient with a risk stratification of the procedure. So far there are no reports on the type of stroke that complicates the procedure. This information is important since it will contribute to the development of strategies that may minimise the risk of stroke.

Learning points

- ▶ Carotid body tumours (CBTs) are rare. The difficulty of surgical resection is highly documented with complications of carotid artery injury, stroke and cranial nerve dysfunction being the most serious.
- ▶ Careful planning with appropriate investigation prior to surgery is essential.
- ▶ Reconstruction of the internal carotid artery in patients with CBT is associated with an increased risk of stroke.
- ▶ In spite of all precautions being taken such as early proximal and distal control of the CBT, the use of a carotid shunt and minimising the cerebral ischaemic time, perioperative cerebrovascular events may still occur.
- ▶ More research is needed to establish the aetiology and ways to minimise cerebrovascular events.

Competing interests None.

Patient consent Obtained.

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