CASE REPORT

Open Repair of Two Ipsilateral Extracranial Internal Carotid Artery Aneurysms

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Introduction: A patient with two ipsilateral extracranial internal carotid artery (ICA) aneurysms treated by open repair is reported, with an emphasis on pre-operative planning and decision making, and a review of literature.

Report: A 44 year old man was diagnosed with a right thyroid lobe nodule and two asymptomatic ipsilateral aneurysms of the right ICA. Diagnostic workup using three dimensional reconstruction and centre lumen line measurements on computed tomography revealed two aneurysms of the right ICA, both > 20 mm. Surgery was planned with intra-operative mandibular subluxation to maximise distal exposure. Neuromonitoring consisted of transcranial Doppler ultrasound and electroencephalography. After mandibular subluxation and complete dissection of the aneurysms and digastric muscle division, adequate exposure of the distal ICA was obtained, followed by resection of both aneurysms, and reconstruction with a reversed greater saphenous vein graft. No complications occurred and one year follow up showed a patent graft without signs of stenosis or anastomotic aneurysm.

Discussion: Open repair of two ipsilateral extracranial ICA aneurysms can be performed safely after careful pre-operative planning using visualisation of the vascular anatomy and distance measurements, and maximising exposure with digastric muscle division, styloidectomy, if necessary, and mandibular subluxation.

Keywords: Aneurysm, Carotid, Extracranial, Mandibular subluxation, Open surgery, Surgical planning

INTRODUCTION

Extracranial carotid artery aneurysm (ECAA) is a rare vascular condition and accounts for < 1% of all peripheral artery aneurysms.1 An ECAA is defined as a dilatation of the carotid artery > 150% compared with the diameter of the internal or common carotid artery.2 The aetiology of these aneurysms is diverse and includes atherosclerosis, infection, and trauma. Neurological symptoms can arise owing to cerebral embolism, resulting in transient ischaemic attack or stroke. Growth of the aneurysm can also cause cranial nerve compression or dysphagia. The risk of ECAA rupture is believed to be low.2

Several series and case reports of single ECAs have been published but reports on multiple synchronous ipsilateral carotid aneurysms are scarce. Currently, endovascular options are frequently sought owing to the expected limited exposure of the distal internal carotid artery (ICA). However endovascular treatment has its own risks: arterial dissection; embolism during manipulation and stent deployment; stent fracture; and in stent stenosis or occlusion. Open repair remains the gold standard, but careful preparation for optimal results is warranted.

Herein, the importance of pre-operative imaging analysis and intra-operative techniques to safely treat a patient with two extracranially located ipsilateral aneurysms of the right ICA are described. To the best of the authors’ knowledge, this is the first report to describe open surgical repair of two ipsilateral ICA aneurysms, using mandibular subluxation and reconstruction with a venous interposition graft.

CASE REPORT

A 44 year old man was referred with a mass in the right side of his neck. Complaints included hoarseness and mild dysphagia. The patient had a blank medical history. Physical examination revealed a painless mass, located in the right side of the neck. Ultrasound showed a 7 cm diameter thyroid nodule located in the right thyroid lobe. In addition, a focal dilation of the right ICA was seen. Computed tomographic (CT) angiography (CTA) revealed two isolated fusiform true aneurysms of the right ICA, without intraluminal thrombus (Fig. 1A). Fine needle aspiration of the thyroid nodule concluded Bethesda category V. Laryngoscopy observed no vocal cord disorders. Firstly, right
Hemithyroidectomy was performed. Histological examination revealed papillary thyroid carcinoma. Subsequently, total thyroidectomy was carried out.

After this, the patient was referred to the vascular department for treatment of the aneurysms. The diameters of the proximal and distal aneurysms were 2.2 cm and 2.0 cm, respectively. Indication for treatment was made based on the relatively young age of the patient, and the risk of future thrombo-embolic complications with the current aneurysm diameters. Growth of the aneurysms over the years would make treatment harder. Pre-operatively, the distances from the distal end of the aneurysm to the styloid process and from the carotid canal at the base of the skull were determined by centre lumen line measurements of the carotid artery with 3 Mensio Vascular imaging and three dimensional (3D) reconstructions of the ICA (Pie Medical Imaging BV, Maastricht, the Netherlands). Beside the narrow exposure field of the distal ICA because of the overlying mandibular condyle and ramus, the styloid process and the base of the skull are important limitations for adequate distal carotid artery access. As a result of the centre lumen line CT measurements it was assessed that enough access to the distal internal carotid artery could be obtained for open repair and that adjunctive actions such as styloid process resection and/or mandibular subluxation would be necessary. The distance from the end of the most distal aneurysm to the styloid process was 20 mm, and from to the carotid canal at the base of the skull was 40 mm. As can be seen in Fig. 1A, tortuosity could result in inadequate exposure of the distal ICA, as it is located medial to the aneurysm. It was concluded that mandibular subluxation would increase the width of the exposure at the base of the skull and facilitate the open repair, especially if shunting was necessary.

Therefore, the oral maxillofacial surgeon was consulted, and the patient was prepared for mandibular subluxation. Because of the ICA tortuosity, endovascular treatment was considered more risky than open repair. A hybrid approach was considered, but this has the drawbacks of both open surgery and endovascular treatment. Venous duplex examination showed a right greater saphenous vein with a diameter of 4 mm, which was sufficient to serve as an interposition graft.

Surgery was performed under general anaesthesia with nasotracheal intubation. Unilateral subluxation of the right temporomandibular joint was performed using an anterior contralateral pull. A unilateral subluxation was conducted from the right mandible to the left and fixed with two screws and a steel wire. After a standard lateral cervical approach, anterior to the sternocleidomastoid muscle, the aneurysms and normal parts of the ICA were completely dissected. As a result, the freely moveable artery and aneurysms could be pulled downwards to obtain a better exposure of the distal ICA. For additional access, the posterior belly of the digastric muscle was divided. Cranial nerves were identified, especially the glossopharyngeal nerve. Heparin (5000 IU) was administered and after three minutes the common, external, and internal carotid arteries were clamped. Mean arterial pressure was maintained above 90 mmHg during the clamping. Neuromonitoring, including transcranial Doppler ultrasound and electroencephalography, showed no changes and therefore intra-operative shunting was not deemed necessary. After resection of the aneurysms, a reversed great saphenous vein graft was interposed with an end to end anastomosis at each end of the ICA (Fig. 2A and B). The post-operative period was uncomplicated; the patient was discharged on...
the third post-operative day. There were no cerebrovascular complications, nerve damage, or mandibular joint dysfunction. Post-operative antiplatelet therapy consisted of clopidogrel. Histological studies of the aneurysm wall showed a degenerative aspect with discontinuation of the elastic layer without signs of inflammation or dissection. Clinical and DNA examinations by medical genetics showed no signs of connective tissue disease. Follow up by means of physical examination, duplex examination after three months, and CTA after one year showed no signs of restenosis or anastomotic aneurysms (Fig. 1B). The patient provided informed consent for publication.

DISCUSSION

Only four cases have been published describing several treatment strategies for multiple carotid artery aneurysms (Table 1). Reported endovascular approaches include covered stenting, intentional occlusion, and bare metal stenting combined with coiling of the aneurysm. One case of surgical repair was reported, but the technique used was transposition of the external to distal ICA, and thus varied from the present case.

In this era of increasing endovascular procedures, open repair remains the gold standard of treatment for true carotid artery aneurysms. However, adequate exposure of the distal ICA is mandatory to perform the procedure safely. Aneurysms of the ICA located above the line of Blaisdell (the retrostyloid region), or at the base of the skull, are known for limited exposure. In these cases additional exposure is essential or endovascular repair has to be considered. Previous studies of surgical access to the cervical ICA found that division of the posterior belly of the digastric muscle associated with mandibular subluxation provides 10–20 mm additional exposure, and styloidectomy provides another few millimetres extra. The most frequently encountered complications in open repair are cranial nerve injuries. To minimise the risk of cranial nerve injury, identification of the cranial nerves is essential. Mandibular subluxation increases the width of the exposure at the base of the skull and facilitates the identification of

![Figure 2](image)

Figure 2. (A) Intra-operative exposure and control of both internal carotid aneurysms. (B) Venous interposition graft with great saphenous vein.

Table 1. Overview of the literature on multiple ipsilateral internal carotid artery (ICA) aneurysms.

| Age, years/sex | Presenting symptoms | Anatomy | Maximum diameter, mm | Procedure | Procedure result |
|----------------|---------------------|---------|----------------------|-----------|------------------|
| Mase et al.    | 49/M                | Amaurosis fugax, motor weakness, urinary incontinence | Two aneurysms of the right ICA located at the cervical segment | NR | Endovascular (stenting) Persistent aneurysm exclusion, no stenosis/occlusion. Clinical outcome asymptomatic |
| Benndorf et al. | 28/F                | Migraines | Three aneurysms of the right ICA. Most distally located aneurysm arose from C5 portion | 18 (most distally) | Carotid occlusion, no further details provided Procedure was performed successfully. Clinical outcome recovering from migraine episodes |
| Han et al.     | 74/M                | Pulsatile neck mass with neck pain | Two synchronous fusiform aneurysms of the distal left ICA located at level C1 vertebrae and at the skull base | 20 (C1 vertebrae), 8 (skull base) | Endovascular (coil embolisation and stenting) Successfully performed without neurological deficits |
| Chedgy et al.  | 77/F                | Transient expressive dysphasia | Two synchronous fusiform and saccular aneurysms located to the left ICA origin | 10 (saccular) | Open surgery, internal to external carotid transposition |

M = male; NR = not reported; F = female.
the glossopharyngeal nerve in the superior part of the dissection. The technique is straightforward and does normally not induce cranial nerve injury or temporomandibular dysfunction. Alternative strategies for increased exposure, such as mandibular ramus osteotomy, ramus resection, or removal of part of the mastoid process, are associated with a higher morbidity rate.

An endovascular approach is favourable in patients with aneurysms that are located high up. An endovascular approach is attractive because it avoids concerns over anatomical exposure and cranial nerve injuries. The disadvantages of an endovascular approach are difficulty in introducing a stent delivery system in tortuous arteries, the risk of not eliminating compression effects of the aneurysm, the risk on further degeneration of an atherosclerotic aneurysm, and the risk of endovascular treatment in general, such as embolism during manipulation and stent deployment. Arterial loops or kinking may hinder endovascular stent placement. Hybrid approaches combining resection of the loop and endovascular treatment of the aneurysm have been suggested for these conditions. This approach should be restricted to those cases with actual limited exposure of a distally located ICA, as it has the drawbacks of both open surgery and endovascular treatment. A hybrid approach was considered in the present case if surgical access to distal ICA would have been insufficient. The kinking of the proximal ICA and the proximal aneurysm could have been resected and reconstructed with an interposition graft, followed by endovascular stent placement over the distal carotid artery aneurysm. Based on the 3D CT imaging and arterial distance measurements, the decision for open repair was made. The high quality 3D images of vascular structures, bony landmarks, and measurements of arterial distances facilitate the decision making process for open repair, endovascular repair, or a hybrid approach.

CONCLUSION

Herein, it has been shown that open surgical repair of challenging multiple ICA aneurysms can be done safely and successfully. Careful planning using pre-operative imaging techniques with visualisation of the vascular anatomy and measurements of distance, and maximising exposure with mandibular subluxation is mandatory.

CONFLICTS OF INTEREST

None.

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