

CASE REPORT

Surgical Management of a Patient with an Internal Carotid Artery Stenosis, Eagle Syndrome, and Internal Carotid Artery Tortuosity: A Case of Four Pathologies of the Carotid Arteries

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Introduction: In 1937, W.W. Eagle first described two clinical cases of elongated styloid process causing compression of adjacent anatomical structures. A case of left internal carotid artery (ICA) stenosis, Eagle syndrome (bilateral), ICA tortuosity, and occlusion of the right carotid arteries is presented.

Report: A 67 year old man was referred following ischaemic stroke two months previously. Computed tomography (CT) revealed the pathologies described. Intervention was performed under general anaesthesia. The digastric muscle was transected, and the styloid process was resected. Carotid endarterectomy with end to end anastomosis between the crossed ends of the ICA was carried out using a temporary shunt due to occlusion of the contralateral carotid arteries. The patient was discharged on the third post-operative day.

Discussion: The case described shows that one stage surgical treatment of ICA stenosis, coiling, and Eagle syndrome gives good results.

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INTRODUCTION

Eagle syndrome is the rarest of the following four pathologies: left internal carotid artery (ICA) stenosis, Eagle syndrome (bilateral), ICA tortuosity, and occlusion of the right carotid arteries. In 1937, W.W. Eagle first described two clinical cases of an elongated styloid process causing compression of the adjacent anatomical structures.¹ Eagle syndrome is a clinical condition in which there is abnormal ossification of the styloid process, the stylohyoid ligament, and the lesser cornua of the hyoid bone. Ossification of these elements leads to an increase in the thickness and length of the styloid process, which then presses on adjacent structures resulting in various pressure symptoms. Pain in the face and throat is more common, while hypersalivation and dysphagia are relatively rare.² This article reports a case of left ICA stenosis, Eagle syndrome, ICA tortuosity, and occlusion of the right common internal and external carotid arteries. Thrombosis of the right carotid arteries could have been caused by the Eagle syndrome, although atherosclerotic disease may also have been a factor.

CASE REPORT

A 67 year old man, who had had a stroke with dysarthria two months previously, was referred to the clinic. The dysarthria disappeared over two days. Risk factors included hypertension, hyperlipidaemia, and tobacco abuse, but he had been well previously on no medication. Following his initial symptoms he was prescribed a statin (rosuvastatin 40 mg daily), antithrombotic (acetylsalicylic acid 100 mg daily), and antihypertensive (calcium channel blocker 10 mg daily). At the time of admission, there were no neurological sequelae. Computed tomography (CT) revealed a minor ischaemic stroke at the level of the semi-oval centre on the right side, left ICA stenosis (60%), Eagle syndrome (bilateral), ICA tortuosity, and occlusion of the right common internal and external carotid arteries (Fig. 1A). No stroke foci were detected in the left half of the brain.

Intervention was performed under general anaesthesia. The tortuosity of the ICA located in the retroparotid region required a left pre-sternocleidomastoid cervicotomy extended distally. The posterior belly of the digastric muscle was transected, exposing the structures around the elongated styloid process (Fig. 1B). Resection of the styloid process was performed. After clamping of the carotid arteries, a longitudinal proximal ICA arteriotomy and resection of carotid tortuosity in the retroparotid region was performed. Carotid endarterectomy and formation of end to end anastomosis between the crossed ends of the ICA was carried out with the help of a temporary shunt due to

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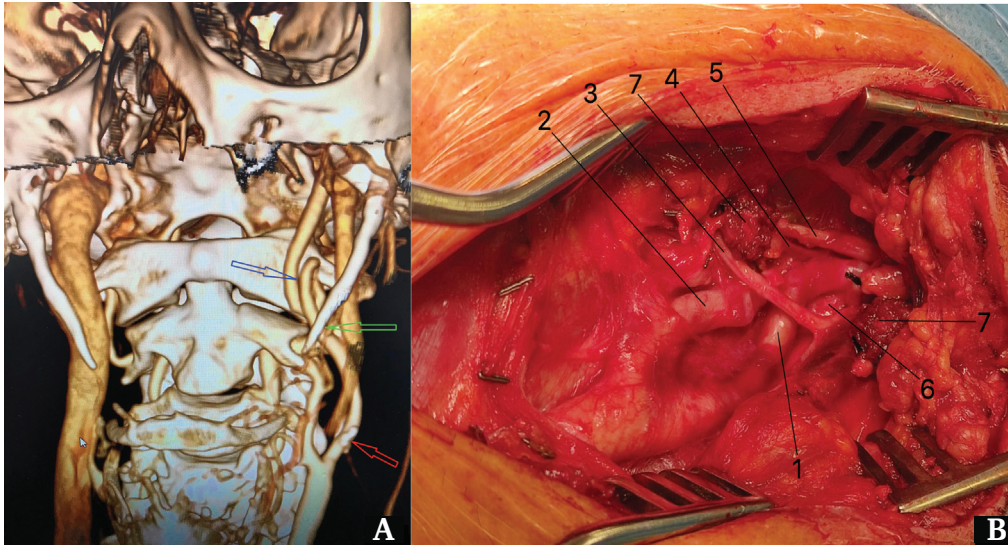


Figure 1. (A) Pre-operative computed tomography angiography: red arrow, stenosis (60%) of left internal carotid artery (ICA); green arrow, elongated styloid process; blue arrow, ICA tortuosity. (B) (1) Left ICA; (2) left external carotid artery; (3) hypoglossal nerve; (4) glossopharyngeal nerve; (5) elongated styloid process; (6) ICA coiling; (7) sectioned digastric muscle.

occlusion of the contralateral carotid arteries (Fig. 2). The arteriotomy was closed with patch plasty. The operation lasted two hours.

Low molecular weight heparin therapy was started on post-operative day one and continued for one week. Acetylsalicylic acid 100 mg daily was used as permanent antiplatelet therapy. The patient was discharged on post-operative day three and was prescribed clopidogrel 75 mg, acetylsalicylic acid 100 mg, rosuvastatin 40 mg, and a calcium channel blocker (amlodipine) 10 mg daily. At the three month follow up the patient's condition was normal. There had been no episodes of transient ischaemic attack. Duplex scanning revealed patent left carotid arteries.

Histopathological examination of the excised segment of ICA revealed fibrous tissue.

DISCUSSION

Some researchers have suggested that the mechanical impact of the elongated styloid process on the ICA is one of the causes of carotid dissection.³ Experience has shown that any kind of mechanical compression of the ICA can cause carotid obstruction and hence stroke.^{4,5} Surgical treatment of Eagle syndrome involves resection of the styloid process by either a transcervical or an intra-oral approach. After a failed intra-oral attempt, which results in an elongated residual stump, it is advisable to use a transcervical approach.⁶ Radak et al. presented a case in which bilateral Eagle syndrome was associated with significant left ICA stenosis and kinking of the right ICA. The patient experienced stroke with right sided weakness and underwent carotid endarterectomy and resection of the styloid process. The post-operative course was uneventful.⁷ The literature also describes conservative treatment of secondary ischaemic stroke in a background of Eagle syndrome. A patient with rheumatoid arthritis was referred with an ischaemic stroke. Diagnostic studies revealed occlusion of the middle cerebral artery and dissection of the left ICA. Surgical intervention for the elongated styloid process was not recommended. Follow up imaging after three months' antiplatelet therapy revealed recanalisation of the left middle cerebral artery.⁸

In the case described here, it would have been possible to use alternative techniques such as eversion endarterectomy or bypass with the great saphenous vein. In both cases, however, there would be difficulty using a temporary shunt. Because the patient had occlusion of the contralateral carotid arteries, and it was considered necessary to use a temporary shunt, classical endarterectomy and resection of ICA tortuosity were chosen. Some authors have noted that routine use of intravascular shunting for a stenosed carotid artery with contralateral occlusion may not be

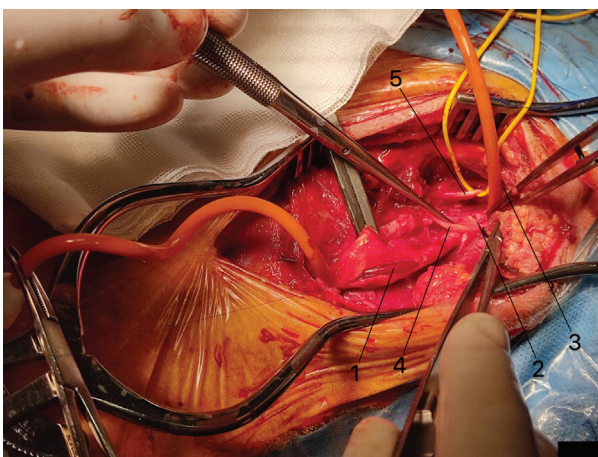


Figure 2. (1) Temporary carotid shunt; (2) the process of anastomosis formation between the crossed ends of internal carotid artery after tortuosity resection; (3) stump of the styloid process; (4) hypoglossal nerve; (5) glossopharyngeal nerve.

necessary and only the presence of pre-operative cerebral infarction increases the risk of stroke.⁹ Another study has shown that surgeons who place shunts selectively during carotid endarterectomy have higher rates of stroke/death in patients with contralateral occlusion.¹⁰ In this case, it was decided to use a temporary shunt because the patient had suffered an ischaemic stroke and retrograde blood flow was weak intra-operatively.

Conclusion

The case described here shows that one stage surgical treatment of ICA stenosis, tortuosity, and Eagle syndrome gives good results.

REFERENCES

- 1 Eagle WW. Elongated styloid processes: report of two cases. *Arch Otolaryngol* 1937;**47**:584–7.
- 2 Politi M, Toro C, Tenani G. A rare cause for cervical pain: Eagle's syndrome. *Int J Dentistry* 2009;**34**:68–71.
- 3 Farhat HI, Elhammady MS, Ziayee H, Aziz-Sultan Ma, Heros RC. Eagle syndrome as a cause of transient ischemic attacks. *J Neurosurg* 2009;**110**:90–3.
- 4 Keshelava G, Robakidze Z. Cervical vagal schwannoma causing asymptomatic internal carotid artery compression. *Ann Vasc Surg* 2020;**63**:460. e9–e11.
- 5 Keshelava G, Nachkopia M, Arabidze G, Janashia G, Beselia K. Unusual positional compression of the internal carotid artery causes carotid thrombosis and cerebral ischemia. *Ann Vasc Surg* 2012;**26**:572. e15–17.
- 6 Papadiochos I, Papadiochou S, Sarivalasis ES, Goutzanis L, Petsinis V. Treatment of Eagle syndrome with transcervical approach secondary to failed intraoral attempt: surgical technique and literature review. *J Stomatol Oral Maxillofac Surg* 2017;**118**:353–8.
- 7 Radak D, Tanaskovic S, Kecmanovic V, Babic S, Popov P, Gajin P. Bilateral Eagle syndrome with associated internal carotid artery kinking and significant stenosis. *Ann Vasc Surg* 2016;**34**:271. e15–18.
- 8 Qureshi S, Farooq M, Gorelick PB. Ischemic stroke secondary to stylocarotid variant of Eagle syndrome. *Neurohospitalist* 2019;**9**:105–8.
- 9 Cinar B, Goksel OS, Karatepe S, Kut S, Aidogan H, Filizcan U, et al. Is routine intravascular shunting necessary for carotid endarterectomy in patients with contralateral occlusion? A review of 5-year experience of carotid endarterectomy with local anaesthesia. *Eur J Vasc Endovasc Surg* 2004;**28**:494–9.
- 10 Goodney PP, Wallaert JB, Scali ST, Stone DH, Patel V, Shaw P, et al. Impact of practice patterns in shunt use during carotid endarterectomy with contralateral carotid occlusion. *J Vasc Surg* 2012;**55**:61–71.