Case Report

The Surgical Management and Histology of Symptomatic Large Extracranial Internal Carotid Artery Aneurysms

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Abstract

The aim of the present study is to describe the treatment strategy and histological findings in patients with large extracranial internal carotid artery (ICA) aneurysms. The resection of the aneurysm and end-to-end anastomosis of the ICA was performed in Case 1 because the patient displayed balloon test occlusion (BTO) intolerance and ICA coiling. Case 2 was treated with an extracranial to intracranial bypass and proximal ICA ligation without BTO. Endovascular embolization of the proximal ICA was performed in Case 3 because the patient displayed tolerance to BTO. The etiology of the aneurysm in Case 2 was determined to be vascular Behçet disease. The findings of these cases indicate that extracranial ICA aneurysms should be treated according to the anatomical, radiological and etiological factors of the aneurysms.

ABBREVIATIONS

BTO: Balloon Test Occlusion; CTA: Computed Tomography Angiography; ECA: External Carotid Artery; ICA: Internal Carotid Artery; MCA: Middle Cerebral Artery; MR: Magnetic Resonance; SPECT: Single-Photon Emission Computed Tomography; STA: Superficial Temporal Artery; T2WI: T2-weighted imaging

INTRODUCTION

Aneurysms of the extracranial carotid artery, which account for 0.09-2.0% of all carotid surgical procedures [1-3], are uncommon. Aneurysms arising from the internal carotid artery (ICA) are extremely rare. Various surgical techniques are used in the treatment of extracranial ICA aneurysms [2,4]. Recently, the advent of endovascular therapy has changed the treatment options [1,5]. The study of these aneurysms is complicated due to their low incidence, differences in etiology and their unknown natural history.

This report presents three cases of large extracranial ICA aneurysms that were treated by different procedures, either endovascularly or surgically. The histological findings of the cases are also described.

CASE PRESENTATION

Case 1

A 64-year-old woman presented with swelling to the right side of her neck and a progressive cervical pulsatile mass that had been present for 1 month. Cervical angiography demonstrated a 1.6 cm × 1.3 cm extracranial ICA aneurysm with coiling of the ICA (Figure 1A and 1B). The aneurysm was located at the C2-C3 spinal interspace. The patient underwent direct surgery by means of an internal shunt under general anesthesia, after she displayed intolerance to balloon test occlusion (BTO). Nasotracheal intubation was used. The posterior belly of the digastrics muscle was retracted and the hypoglossal nerve was mobilized rostrally after the descending hypoglossal nerve had been severed. An intraluminal shunt was used. The distal and proximal site of ICA and the aneurysm were totally exposed, which allowed for the adequate mobilization of the ICA and the aneurysm. The aneurysm was totally resected and end-to-end primary anastomosis was performed without grafting to reconstruct the...
coiling of the ICA (Figure 1D and E). A histological examination revealed myointimal hypertrophy with vascular proliferation (Figure 2A). Elastic van Gieson staining revealed the partial interruption of the internal elastic membrane (Figure 2B). The postoperative period was uneventful, and no neurological deficit was noted. A follow-up angiogram obtained at 1 month showed the good patency of the right ICA (Figure 1C), which remained unchanged on duplex ultrasonography during a 5-year follow-up after surgery.

Case 2

A 77-year-old woman was admitted with a right-sided cervical pulsatile mass and pain that had been present for 1 month. She had been diagnosed with vascular Behçet disease 6 years prior to this admission and a diagnosis of bilateral extracranial ICA aneurysms and right renal artery aneurysm had already been established 3 years prior to this admission. Conservative treatment had been selected because the patient did not have any symptoms or neurological deficits. Computed tomography angiography (Figure 3A and 3B) and cervical angiography (Figure 3C) showed multiple right extracranial ICA aneurysms (4.0 cm×3.0 cm, 0.9 cm×0.7 cm, 2.2 cm×2.0 cm) and a left extracranial ICA aneurysm (2.0 cm×2.1 cm). The aneurysm located in the proximal right ICA was partially thrombosed. Sufficient cross-flow from the anterior and posterior communicating arteries was obtained. Surgical treatment was planned for the right symptomatic ICA aneurysms because a balloon catheter was unable to be navigated through the tortuous carotid artery, preventing BTO from being performed. The left asymptomatic extracranial ICA aneurysm was managed conservatively. A bypass of the superficial temporal artery (STA) to the M2 segment of the right middle cerebral artery (MCA) was performed, and the proximal site of the aneurysm was occluded with silk sutures just distal to the cervical bifurcation (Figure 3D). The proximal ICA was partially removed. A histological examination revealed thickening of the intima and adventitia, collagen fiber deposition in the media and adventitia, and the proliferation of the vasa vasorum (Figure 3E and 3F). The patient was diagnosed with vascular Behçet disease based on HLA-B51 positivity, the presence of multiple aneurysms, and the pathological findings. The patient was discharged in an excellent condition. The magnetic resonance (MR) angiography findings obtained at a two-year follow-up examination showed the patency of the right-sided bypass and the disappearance of the right ICA aneurysms, while the size of the left ICA aneurysm remained unchanged.
Case 3

A 61-year-old woman was admitted with a right-sided cervical pulsatile mass and dysphasia that had been present for 1 year. MR imaging showed an aneurysm in the cervical portion of the right ICA, which compressed the trachea to the left (Figure 4A). Cervical angiography revealed a 5.0 cm × 3.2 cm right extracranial ICA aneurysm (Figure 4D). The right carotid artery bifurcation was located at the level of C2. Under local anesthesia, the ICA was endovascularly embolized just proximally to the lesion with Guglielmi detachable coils (GDC) and fibered coils (Figure 4E) because the patient displayed tolerance to BTO. Her symptoms gradually improved and the aneurysm decreased in size. MR imaging at 3 months after the procedure showed that the aneurysm was partially thrombosed (Figures 4B). It was found to have totally disappeared at a 6-year follow-up examination (Figure 4G).

DISCUSSION

Extracranial carotid artery aneurysms provide a challenge to the clinician because of their multiple causes, therapeutic difficulties, and their rarity. They account for only 0.4-1% of all extracranial aneurysms [6,7]. El-Sabrout et al. reported that 7 out of 23 (30.4%) extracranial carotid artery aneurysms occurred in the ICA [2]. The bilateral presentation of extracranial ICA aneurysms is exceptionally rare [8,9] and is only found in 3-8% of extracranial carotid artery aneurysms [2,10].

The causes of extracranial ICA aneurysms are reported to include atherosclerosis, previous carotid endarterectomy [2,4], dissection [11,12], congenital, infection, radiation, and connective tissue disease [9,13]. Extracranial carotid artery aneurysms are subdivided into true aneurysms and false (pseudoaneurysm) aneurysms. In meta-analysis description, most than 80% of extracranial carotid artery aneurysms were classified as pseudo aneurysms [14].

To our knowledge, among 40 reported case of extracranial carotid artery aneurysm and Behçet disease, only 13 cases (32.5%) were located in extracranial ICA aneurysms [11,15-19] (Table 1). Surgery has a role in the treatment of extracranial ICA aneurysms in patients with Behçet disease, because few cases have been treated endovascularly and due to the lack of long-term follow-up [16]. Tuzun et al. reported a case of bilateral fusiform aneurysms at the common carotid arteries in a patient with Behçet disease [20], in which the patient received medical treatment. To our knowledge, this is the first report of bilateral extracranial ICA aneurysms in a patient with Behçet disease.

In the current study, the histological examination of extracranial ICA aneurysms showed two distinctive categories: aneurysms after dissection and degenerative aneurysms [12]. Well eewerd et al. found degenerative aneurysms in 10 of 13 (73%) patients. The histological findings of Case 1 and Case 2 appear to show regression (Table 2).

Extracranial ICA aneurysms present as a cervical or parapharyngeal mass and have the potential to cause a transient ischemic attack or stroke. Patients can also present with symptoms caused by laryngeal or esophageal compression, and glossopharyngeal nerve or sympathetic nerve compression. Ruptures have rarely been reported [2]. Surgical intervention can be justified for symptomatic aneurysms related to cerebral ischemia or other types of local discomfort. The reported surgical thresholds, even in asymptomatic cases, are an aneurysm of more than 2-cm in diameter, or 150% larger than the diameter of the common carotid artery, 200% larger than the ICA, or 120% larger than the normal ipsilateral ICA [2,4,21,22].

The proximal and distal extent, outer diameter, etiology, and the status of the collateral flow [2,9] may affect the choices of treatment. The treatment options include aneurysm clipping, resection and end-to-end anastomosis, resection with an interposition graft, extracranial to intracranial bypass, and carotid artery ligation [1]. No recurrence was observed in 7 of 12 cases in which carotid ligation was performed in patients with extracranial carotid artery aneurysms [11]. However, trapping could theoretically prevent backflow into the aneurysm due to cross-circulation. If the patient tolerates a 20-minute test occlusion without neurological deficits and the regional perfusion on single-photon emission computed tomography (SPECT) remains normal, proximal occlusion of the ICA can be performed. When the patient manifests neurological signs during BTO or shows hypo perfusion in the ipsilateral ICA territory on SPECT, the surgical obliteration of the aneurysm should be performed in combination with the reconstruction of the ICA. In the current series, resection of the aneurysm and end-to-end anastomosis of the ICA was performed in Case 1 because the patient displayed intolerance to BTO and due to the coiling of the ICA. The indications for end-to-end anastomosis might therefore depend on accurately detecting the distal site of the ICA and exposing the aneurysm. In Case 2, STA-MCA bypass and proximal ICA ligation were conducted because the patient had good collateral flow from anterior and posterior communicating arteries. No neurological complications were observed in either of these cases, and the aneurysms completely disappeared on follow-up images. An external carotid artery (ECA) - saphenous vein-MCA bypass or ECA-radial artery-MCA bypass should be considered if the patient has insufficient collateral flow from the
### Table 1: The published cases of surgically treated Behçet’s extracranial ICA aneurysms.

<table>
<thead>
<tr>
<th>Case (year)</th>
<th>Age (years)/Sex</th>
<th>Location</th>
<th>Treatment</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Dhnobb et al. (1986)</td>
<td>44/F</td>
<td>ICA</td>
<td>Resection and end-to-end anastomosis</td>
<td>Good</td>
</tr>
<tr>
<td>Tacal et al. (1993)</td>
<td>35/M</td>
<td>ICA</td>
<td>Resection and tube grafting</td>
<td>Thrombosis</td>
</tr>
<tr>
<td>Canova et al. (1997)</td>
<td>32/M</td>
<td>ICA</td>
<td>Repair (ND)</td>
<td>Good</td>
</tr>
<tr>
<td>Bonnotte et al. (1999)</td>
<td>47/M</td>
<td>ICA</td>
<td>Bare stent and coil embolization</td>
<td>Good</td>
</tr>
<tr>
<td>Iscan et al. (2005)</td>
<td>43/M</td>
<td>ICA</td>
<td>PTFE bypass</td>
<td>Good</td>
</tr>
<tr>
<td>Sayed et al. (2010)</td>
<td>37/F</td>
<td>ICA</td>
<td>Ligation</td>
<td>Died from cardiomyopathy</td>
</tr>
<tr>
<td>25/M</td>
<td>ICA</td>
<td>Ligation</td>
<td>Good</td>
<td></td>
</tr>
<tr>
<td>35/M</td>
<td>ICA</td>
<td>Excision and vein grafting</td>
<td>Recurred and was ligated and resulted in a minor stroke</td>
<td></td>
</tr>
<tr>
<td>30/M</td>
<td>ICA</td>
<td>Ligation</td>
<td>Good</td>
<td></td>
</tr>
<tr>
<td>33/M</td>
<td>ICA</td>
<td>Ligation</td>
<td>Good</td>
<td></td>
</tr>
<tr>
<td>32/M</td>
<td>ICA</td>
<td>Ligation</td>
<td>Good</td>
<td></td>
</tr>
<tr>
<td>25/M</td>
<td>ICA</td>
<td>Ligation</td>
<td>Good</td>
<td></td>
</tr>
<tr>
<td>Takamiya et al. (2016)</td>
<td>40/M</td>
<td>ICA</td>
<td>Bare stent and coil embolization</td>
<td>Good</td>
</tr>
<tr>
<td>Present case (2017)</td>
<td>77/F</td>
<td>Bilateral ICAs</td>
<td>Rt side: proximal ligation and STA-MCA anastomosis, Lt side: conservation</td>
<td>Good</td>
</tr>
</tbody>
</table>

**Abbreviations:** F: Female; ICA: Internal Carotid Artery; M: Male; MCA: Middle Cerebral Artery; ND: Not Described; PTFE: Polytetrafluoroethylene; STA: Superficial Temporal Artery.

### Table 2: The characteristics of the patients with extracranial ICA aneurysms.

<table>
<thead>
<tr>
<th>Age (Years)/Sex</th>
<th>Location</th>
<th>Histology</th>
<th>BTO</th>
<th>Treatment</th>
<th>Outcome (years)</th>
</tr>
</thead>
<tbody>
<tr>
<td>64/F</td>
<td>Rt ICA</td>
<td>Degeneration</td>
<td>Intolerant</td>
<td>Resection and end-to-end anastomosis</td>
<td>Good (5)</td>
</tr>
<tr>
<td>77/F</td>
<td>Bilateral ICAs</td>
<td>Degeneration</td>
<td>Attempt</td>
<td>Rt side: proximal ligation and STA-MCA anastomosis, Lt side: conservation</td>
<td>Good (2)</td>
</tr>
<tr>
<td>61/F</td>
<td>Rt ICA</td>
<td>-</td>
<td>Tolerant</td>
<td>Proximal occlusion (endovascular treatment)</td>
<td>Good (10)</td>
</tr>
</tbody>
</table>

**Abbreviations:** BTO: Balloon Test Occlusion; F: Female; MCA: Middle Cerebral Artery; ICA: Internal Carotid Artery; Rt: Right; STA: Superficial Temporal Artery.

Recently, several case reports/series have advocated the use of an endovascular approach in the treatment of extracranial carotid artery aneurysms [1,5,7,23,24]. Some researchers found that in cases involving pseudoaneurysms, the long-term outcomes after endovascular procedures were superior in comparison to cases involving true aneurysms [24]. The use of a covered stent might decrease the incidence of postoperative endoleak, whereas bare self-expanding stents might easily pass through tortuous carotid arteries [7]. Cerebral protection devices were not necessary because the widely patent lumen of most extracranial carotid artery aneurysms implies a low risk of embolism [7]. In the current series, the endovascular embolization of the proximal ICA was successfully performed in Case 3. In true aneurysms, surgery is recommended because the aneurysms could have a significant amount of thrombus inside the sac and because they frequently cause cerebral embolic events during the endovascular procedures [25]. On the other hand, the surgical repair and total resection of giant aneurysms (>4.5 cm), combined with arterial reconstruction, are reported to be associated with a high incidence of cranial nerve injuries [21]. The endovascular placement of a stent can be an excellent alternative to surgical revascularization in patients with such lesions, especially in cases in which obtaining surgical access to the distal end of the aneurysm is difficult or complicated.

The present study is associated with some limitations. The study population was relatively small and the various etiologies of the patients might have limited the generalizability of the results. The results of this study and the conclusions that can be drawn only apply to patients undergoing surgical repair for extracranial ICA aneurysms.

**CONCLUSIONS**

Due to their low incidence, no optimal treatment modality has been established for extracranial ICA aneurysms. At present, they should be treated either surgically or endovascularly, based on the BTO results and according to the anatomical, radiological and etiological factors of the aneurysm.

**REFERENCES**