

## Extracranial carotid artery aneurysm and surgical treatment

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### ABSTRACT

**Objectives:** This study aims to investigate clinical, laboratory, and neurological data and operations of the patients in patients with an extracranial carotid artery aneurysm (ECCA).

**Patients and methods:** A total of 14 patients (11 males, 3 females; mean age: 61.6±6.3 years; range, 48 to 70 years) who were operated for an ECCA in our clinic between January 2011 and January 2021 were retrospectively analyzed. Color Doppler ultrasound (CDUS) and computed tomography angiography (CTA) were used to evaluate the patients. Data regarding patient characteristics, strokes, mortality, obstructions, and other surgical complications, as well as preoperative atherosclerotic risk factors, were recorded.

**Results:** The surgically operated lesions were located on one side of the neck in all patients; particularly, they were located on the left side in three (22%) patients and on the right in 11 (78%) patients. The most common type of ECCA in all patients was Type 1 (saccular), and aneurysms were reported to be present only in the internal carotid artery. Primary end-to-end repair was performed in 11 (78%), saphenous vein graft interposition in two (14%), and synthetic graft in one (8%) patient. The mean length of stay in the intensive care unit was 2.1±0.4 days, the mean length of the hospital stay was 87±0.64 days, and the mean amount of drainage in the first two postoperative days was 24.4±10.8 mL. Bleeding at the wound site occurred in one patient due to the skin incision and was eliminated via suturing. A hematoma at the wound site was observed in one patient, and redness at the wound site was observed in one patient and disappeared prior to discharge with the treatments applied. The neurological examination revealed a temporary facial shift in one patient that resolved before discharge.

**Conclusion:** The natural course of ECCAs is still not well understood, and there is no definitive consensus about their treatment. The main goal of managing ECCAs is to prevent aneurysms. They are rare entities and may occur with or without various symptoms. Early and long-term outcomes of invasive treatment of ECCAs are favorable.

**Keywords:** Carotid artery, carotid artery aneurysm, saccular aneurysm.

Extracranial carotid artery aneurysm (ECCA) is a very rare clinical disease that may occur as a result of atherosclerosis, degeneration, fibromuscular dysplasia, or traumatic injury. Carotid artery aneurysm is defined as a localized increase of more than 50% in vascular calibration compared to the reference values. Rarely, it may also occur as a complication following a carotid endarterectomy (CEA) procedure. The signs of the disease include a pulsatile mass in the neck, headache and neck pain, a retro-orbital pressure sensation, ear pain spreading to the occipital region due to

glossopharyngeal compression, dysphagia, and cranial nerve compressions in aneurysms reaching near the carotid canal. Surgical intervention is recommended to prevent possible complications due to the risk of rupture, as well as neurological sequelae caused by cerebral atheroembolism.<sup>[1]</sup> The goal is to resect the aneurysm and repair the vessel while maintaining the cerebral blood flow.<sup>[2]</sup> Carotid aneurysms are very rare vascular pathologies in clinical practice.

All carotid artery surgeries with ECCAs have an incidence of 0.2 to 5%.<sup>[3]</sup> The most common aneurysm

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sites in the carotid arteries are bifurcations (tending to be fusiform) and the mid-distal internal carotid artery (ICA) (tending to be saccular).<sup>[4]</sup> Cervical lymphadenopathies, cervical abscesses, Castleman's disease (giant lymph node hyperplasia), and glomus tumors should be taken into consideration in the differential diagnosis. Many clinics only see one or two cases per year. It is important that the physician recognizes the disease and its complications and plans the treatment given the potential for serious complications.<sup>[5]</sup> The ideal surgical treatment is resection and, then, arterial continuity, as well as preventive treatment against cranial nerve damage and embolic complications. End-to-end anastomosis, synthetic vein grafting, or saphenous vein grafting can be interposed after resection. The protection of adjacent structures in the operative repair of CAA and the presence of intense inflammation in the surrounding tissues are other issues to be considered for surgeons.

In the present study, we aimed to examine the clinical, laboratory, and neurological data and operations of the patients with ECCA and to evaluate the success of the surgical intervention.

## PATIENTS AND METHODS

This single-center, retrospective study was conducted at Erzurum City Hospital, Department of Cardiovascular Surgery between January 2011 and January 2021. A total of 14 patients (11 males, 3 females; mean age:  $61.6 \pm 6.3$  years; range, 48 to 70 years) who were operated for ECCA in our clinic were included. A written informed consent was obtained from each patient. The study protocol was approved by the Erzurum City Hospital Ethics Committee (No: 2021/05-103). The study was conducted in accordance with the principles of the Declaration of Helsinki.

The patients initially applied to cardiology and neurology clinics with complaints of neck pain, swelling, and high blood pressure. The patients who were diagnosed with carotid artery aneurysms radiologically were referred to our clinic with advanced examinations and treatment plans. Carotid artery aneurysm was defined as a localized increase of more than 50% in vascular calibration relative to reference values. Saccular carotid artery aneurysm was defined as a bubble-shaped pathological enlargement in the carotid artery wall. Color Doppler ultrasound (CDUS) and computed tomography angiography (CTA) were used to

evaluate the patients. Possible difficult access and surgical planning were requested prior to operation for the surgical intervention to be performed after evaluating with CTA.

Surgical procedures were performed under general anesthesia. No intraoperative shunts were used in the patients, and systemic heparinization was not neutralized. Data including intraoperative bleeding, new acute ischemic stroke, and death were recorded. Postoperatively, the patients were followed and treated in the cardiovascular surgery intensive care unit (ICU). Drainage follow-ups and first neurological examinations after extubation were routinely performed. The patients were discharged from the hospital with 100 mg of acetylsalicylic acid to be taken once per day with appropriate antibiotherapy and daily wound dressing recommendations. The patients were scheduled for follow-up at specified intervals for a year to confirm the patency of the operation. Outpatient follow-ups were performed in the first 10 days after discharge. Patency was evaluated by performing CDUS imaging on Day 10, at three and six months and at one year after discharge. Data regarding patient characteristics, strokes, mortality, obstructions, and other surgical complications with preoperative atherosclerotic risk factors were obtained from the hospital records.

### Operation technique

General anesthesia was used in all patients. Intravenous antibiotics were routinely administered during induction. The carotid artery was reached by an incision along the anterior edge of the sternocleidomastoid muscle. The hypoglossal nerve, vagus nerve, and ansa cervicalis were preserved. Carotid arteries were found and clamped. Unfractionated heparin was routinely administered intravenously at a dose of 5,000 IU/mL before clamping, and the aneurysmatic sac was removed and repaired primarily end-to-end using synthetic or autogenous grafts. Bleeding control was performed, drains were placed with the layers closed in accordance with the procedure, and the operation was terminated. The patients were transferred to cardiovascular surgery ICU while intubated to continue their follow-up and treatment.

### Statistical analysis

Statistical analysis was performed using the IBM SPSS version 20.0 software (IBM Corp., Armonk, NY, USA). Descriptive data were expressed in mean  $\pm$  standard deviation (SD), median (min-max)

or number and frequency. The Kolmogorov-Smirnov test and Shapiro-Wilk test were used to check the normal distribution of variables. The Student's t-test was used for the data with normal distribution and the Mann-Whitney U test was used for the data without normal distribution. The chi-square test was performed to compare these data. A  $p$  value  $<0.05$  was considered statistically significant.

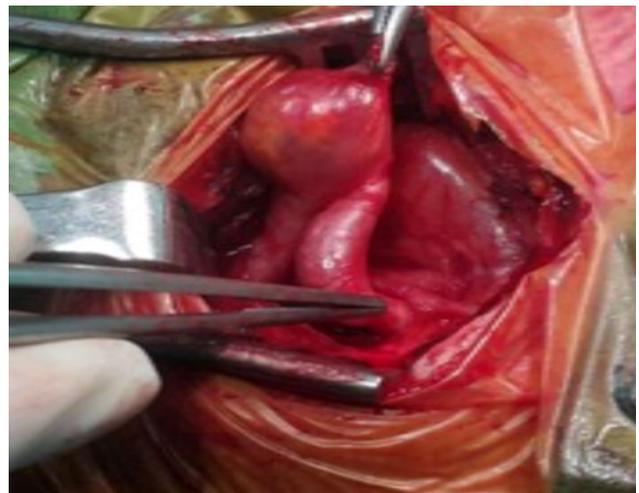
## RESULTS

The surgically operated lesions were on one side of the neck in all patients, on the left in three (22%) patients and on the right in 11 (78%) patients. The mean age of the male and female patients was  $61.5 \pm 6.5$  (range, 48 to 70) years and  $62 \pm 3$  (range, 59 to 64) years, respectively. Smoking, which is one of the risk factors for peripheral arterial disease, was found to be absent in female patients ( $n=0$ ), occurring in male patients at a rate of 45% ( $n=5$ ), and occurring at a rate of 35% ( $n=5$ ) in total. Hypertension was detected in two of the female patients (66%) and three of the male patients (21%) with a total of five patients (35%). Diabetes mellitus (DM) was detected in one of the male patients (7%) and in two of the female patients (66%) with a total of three patients (21.4%). The mean low-density lipoprotein (LDL) cholesterol level was  $170 \pm 35.8$  (range, 134 to 234) mg/dL. The most common atherosclerotic risk factors were found to be smoking and hypertension in male patients and hypertension and DM in female patients (Table 1). The most common type of ECCA in all patients was recorded as Type 1 (saccular), and aneurysms were reported to be present only in the ICA (Figure 1, 2).



**Figure 1.** A preoperative computed tomography angiography image of a patient.

ICA: Internal carotid artery; ECA: External carotid artery; CCA: Common carotid artery.



**Figure 2.** Intraoperative images (aneurysm sac and its image after removal).

**Table 1. Demographic and clinical characteristics of patients**

Patient characteristics	n	%	Mean $\pm$ SD	Range
Age (year)			61.6 $\pm$ 6.3	48-70
Sex				
Male	11	78		
Female	3	22		
Side				
Right	11	82		
Left	3	22		
LDL cholesterol (mg/dL)			170 $\pm$ 35.8	
Smoking	5	35		
Hypertension				
Male	3	21		
Female	2	14		
Diabetes mellitus				
Male	14	7		
Female	2	14		

SD: Standard deviation; LDL: Low-density lipoprotein.

**Table 2. Procedural data**

Operation type	n		%	
	Male	Female	Male	Female
Primary end-to-end anastomosis	9	2	64	36
Saphenous vein interposition	2	-	100	-
Synthetic graft interposition	-	1	-	100

**Table 3. Postoperative data**

Results	n	%	Mean±SD
Intensive care hospitalization time (2-3 days)			2.1±0.4
Hospitalization time (3-5 days)			3.9±0.6
Drainage amount (0-40 mL) (first 2 days)			24.4±10.8
Nerve damage (percentage shift)	1	7	
Obstruction (within the first 1 year)	0	0	
Patency (within the first 1 year)	14	100	
Reoperation	0	0	
Bleeding	1	7	
Hematoma	1	7	
Redness on scar	1	7	
Facial shift	1	7	
Visual impairment	0	0	
Death	0	0	
Stroke	0	0	

SD: Standard deviation.

Primary end-to-end repair was performed in 11 patients (78%), saphenous vein graft interposition was performed in two patients (14%), and synthetic graft was performed in one patient (8%) (Table 2). The mean length of stay in the ICU was 2.1±0.4 days, the mean length of hospital stay was 3.9±0.6 days, and the mean amount of drainage in the first two postoperative days was 24.4±10.8 mL. Moreover, bleeding at the wound site occurred in one patient due to the skin incision and was eliminated by suturing. A hematoma at the wound site (self-limiting without the need for surgical intervention) was seen in one patient, and redness at the wound site was seen in one patient and disappeared before discharge with the treatments applied. The neurological examination revealed a temporary facial shift in one patient that disappeared before discharge. No patients had visual impairments or other neurological complications. None of our patients were re-operated. Additionally, no mortality or postoperative strokes were observed in any of the patients (Table 3). The carotid artery that underwent surgical intervention was patent during the post-discharge follow-ups of the patients. The

patients were assessed throughout the study period, and they were followed closely for one year after the operation. During this period, no pathology was detected in the neurological tests of the patients. Neurological dysfunction was not detected in the later outpatient follow-ups of the patients. The patients were discharged with appropriate anticoagulant and statin treatments, and they were scheduled for follow-ups at prespecified time points. The CDUS was used as the imaging method during follow-up.

## DISCUSSION

Despite the development and increasing number of non-invasive treatment methods and the small number of patients, the surgical method is still effective in correcting and treating the pathology in carotid artery aneurysms. Complications that may occur would be minimal in experienced centers and competent hands. Extracranial carotid artery aneurysms are rare, critical health problems that may lead to stroke, death, and major neurological complications. Moreover, they represent vascular pathologies, accounting for approximately 4% of peripheral aneurysms. The male-to-female ratio is 2:1 in patients with ECCAs, and they are more common in males. The affected population has a wide age range.<sup>[6]</sup> There are many reasons for ECCAs, such as fibromuscular dysplasia, trauma, infection, congenital defects, radiation arteritis, and Behçet's disease; however, the most common cause is atherosclerosis.<sup>[7]</sup> The most common sites of aneurysms in the carotid arteries are carotid artery bifurcations and the mid-distal ICA (tending to be saccular).<sup>[4]</sup> The natural course of ECCA is chronic growth; therefore, it has the potential for rupture, distal embolization, and local compression. Signs of the disease include a pulsatile mass in the neck, headache and neck pain, a retro-orbital pressure sensation, ear pain spreading to the occipital region due to glossopharyngeal compression, dysphagia, and cranial nerve compressions in aneurysms reaching near the carotid canal.<sup>[8]</sup> The most common clinical result of our patients' complaints was a pulsatile mass in the neck. The CDUS, CTA, and magnetic resonance angiography (MRA) are commonly used radiological imaging methods in the diagnosis of ECCA, as well as in the planning of surgical intervention. The CTA and MRA, which are helpful in visualization of the anatomy in detail, are frequently preferred while planning the surgical treatment.

Extracranial carotid artery aneurysms have an important potential clinical effect due to the potential for cerebral ischemia, despite its low frequency. Thromboembolic pathologies and neurological complications may develop in patients with untreated ECCA.<sup>[9]</sup> The choice of treatment options often depends on the anatomy of the aneurysm, and the serious potential complications associated with ECCA require a surgical intervention.<sup>[1]</sup> It is reported that the number of strokes is low with a surgical intervention, which supports the finding that invasive treatment can prevent strokes.<sup>[10]</sup> If anticoagulant therapy fails, progressive expansion and neurological symptoms develop, and surgical intervention is required. The main goal of surgical treatment is resection and ensuring arterial continuity. The most suitable cases for this are common carotid artery and 1/3 proximal ICA lesions. While small saccular aneurysms can be repaired with primary closure or patching following aneurysmectomy, such local repair is not possible in wide neck saccular and fusiform aneurysms. After resection, arterial continuity can be achieved via end-to-end anastomosis, external carotid artery anastomosis to the ICA, or interposition with a synthetic or saphenous vein graft. Although the results of prosthetic and autologous saphenous vein grafts are good and equal, saphenous vein grafts should be preferred in case of infection. Surgical treatment for ECCA has been advocated due to the high risk of mortality in non-surgical cases.<sup>[11]</sup> The purpose of any surgical intervention is to eliminate the risk of thromboembolic complications or the rupture of an aneurysm. Srivastava et al.<sup>[12]</sup> reported a primary patency of over 90% at 30 months in their 10-year follow-up after surgical intervention. Preferred surgical techniques include end-to-end primary anastomosis, as well as autologous or synthetic graft interposition with resection of the sac. Currently, cases advocating an endovascular approach to treating ECCA have also reported favorable procedure results.<sup>[10]</sup>

The natural course of ECCAs has not been understood yet, and there is no definitive consensus about its treatment. No treatment guidelines or expert consensus has been developed for the management of ECCA thus far, as natural course is required to balance the benefit of any intervention. Probably, the main goal of managing ECCAs is to prevent aneurysms. These aneurysms are rare and may occur with or without various symptoms. All segments of the carotid artery are sensitive. The safety and long-term reliability of appropriate surgical treatments guarantee a wide surgical indication due to the risk of embolization

and rupture of these aneurysms. The early and long-term outcomes of invasive treatment in ECCA are favorable. Recently, non-invasive treatment methods for ECCAs have also been described in several studies. In particular, for the treatment applied by stenting, the number of studies on the numerous applications from many centers has been increasing day by day. Some authors have advocated that the comparison is made with the surgical method and that the results are similar. However, the rarity of carotid artery aneurysms, the fact that the publications are not yet multi-center, and the fact that the complications related to radiation, opaque material, and catheterization have not yet been clearly investigated indicate that the surgical treatment method in carotid artery aneurysms is still valuable, compared to other treatment methods.

The main limitations of this study include the fact that the study was conducted in a single center with a retrospective design, the cases investigated were rare, the sample size was low, and there is no large-scale case series on this issue in the literature.

In conclusion, in the present study, we examined patients who underwent surgical intervention for ECCA, which is a rare disease, in our clinic. We observed that eight patients were successfully operated on due to ECCA and recovered without experiencing any of its potential risks. No stenosis was detected in the aneurysms operated during the outpatient follow-ups in the post-discharge period. Based on these findings, we achieved successful results in our series. We, therefore, believe that surgical intervention is still the most effective method in the treatment of ECCAs, as it is a reliable treatment method that has been applied for a long time and has almost no complications when performed meticulously.

#### **Declaration of conflicting interests**

The authors declared no conflicts of interest with respect to the authorship and/or publication of this article.

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