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Successful Surgical Repair of Extracranial Internal Carotid Artery Aneurysm in A Patient with Behcet's Disease: A Case Report

Mohammad. A. Al-doud[1*], Bashar K. Anakrih[1], Khaldoon O. Alwreikat[1], Husam I. Al-khawaldeh[1], Fuad I. Khamis[1], Thair M. Al-Tarabsheh[1], Muhammad A. Al-Rawashdeh[1]

[1]Department of Vascular Surgery, Jordanian Royal Medical Services (JRMS), Amman, Jordan

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***Corresponding author:** Mohammad. A. Al-doud
Department of Vascular Surgery, Jordanian Royal Medical Services (JRMS), Amman, Jordan

Abstract **Case Report**

Introduction and importance: Behçet's disease (BP) is a chronic, recurrent inflammatory multi-systemic vasculitis of unknown aetiology that initially presented with a triad of recurrent oral ulcers, genital ulcers, and uveitis. Vascular involvement, especially arterial involvement, is one of the significant causes of morbidity and mortality. ***Case presentation:*** A 25-year-old man presented with a sore throat, dysphagia, weight loss (8 kg in 2 months), and swelling on the left side of his neck. He was referred to our vascular surgery department for assessment of an expanding pulsatile mass in the left side of his neck, which was associated with numbness and weakness in the right side of his body. Neck CTA revealed a 43 x 42.3 x 35.8 mm pseudoaneurysm in the left internal carotid artery. The pseudoaneurysm was excluded. Reconstruction was performed with a bypass from the CCA to the ICA using a reversed great saphenous vein graft interposition. ***Clinical discussion:*** Extracranial carotid artery aneurysms (ECAAs) are rare and distinct from the more common atherosclerotic occlusive disease in the same location. This rarity underscores the unique and complex nature of the case. The primary objective of treating ECAAs is to prevent permanent neurologic deficits that may arise from atheroembolism and thromboembolism. The main goal is achieved by excluding the aneurysm from the arterial circulation and restoring antegrade flow. ***Conclusion:*** It is crucial to note that extracranial carotid artery aneurysms (ECAAs) and pseudoaneurysms, while rare, can have significant consequences. Their asymptomatic nature and the variability of their symptoms according to location, size, and aetiology underscore the urgency and seriousness of prompt treatment.

Keywords: Behçet's disease (BD), Extracranial Carotid Artery Aneurysms (ECAAs), Pseudoaneurysms (PSA), Open surgical repair.

INTRODUCTION AND IMPORTANCE

Behçet's disease (BP) is a chronic, recurrent inflammatory multi-systemic vasculitis of unknown aetiology that initially presented with a triad of recurrent oral ulcers, genital ulcers, and uveitis. Behçet's disease, with its chronic (long-term) course and recurrent unpredictable inflammatory attacks, is a matter of grave concern due to its severe consequences and periods of relapse. However, relapses and remissions are characteristic of the condition [1].

Neurological manifestations are one of the most serious complications of Behçet disease due to its severe prognosis [2]. Patients may experience neurological disturbance as the heralding symptom, but most neurological complications develop 3–6 years after the onset of systemic features [3].

Vascular manifestations of Behçet's disease, known as vasculo-Behçet's disease (v-BD), were seen in up to 50% of patients. Various clinical presentations, such as venous thrombosis, arterial occlusion and arterial aneurysm formation, characterise Behçet's disease. According to Hamza's classification, there are five types of vasculo-Behçet's disease (v-BD) [See Table 1] [2, 3].

Table 1. Hamza classification of vascular involvement in Behcet disease.

Type	Vascular Involvement
I	Venous thrombosis
II	Arterial thrombosis
III	Arterial aneurysm
IV	Arterial thrombosis and aneurysm
V	Venous thrombosis and arterial lesion

The venous system is the most commonly affected site, and superficial or deep vein thrombosis is the most frequent vascular issue [4]. In Behçet's disease (BP), thrombus adheres to the vessel wall, and thrombosis may progress despite anticoagulation. Arterial complications are less common than venous complications, occurring in about 1% to 7% of patients. However, vascular involvement is a significant cause of morbidity and mortality in Behçet's disease [5,6]. Aneurysms are common among arterial lesions and affect various arteries. The aorta is the most common site of aneurysm formation, with pulmonary, femoral, popliteal, brachial, and iliac arteries also reported in descending order of frequency [6].

Extracranial carotid artery aneurysms (ECAAs) are generally sporadic, accounting for less than 2% of carotid surgery. This rarity adds a unique and complex dimension to the case. A significant part of the ECAAs will remain clinically silent. However, ECAAs may lead to neurologic presentations, including transient ischemic attacks (TIAs) or ischemic stroke. Other presentations include pulsatile neck mass and cranial nerve dysfunction. The morbidity and mortality rates of extracranial carotid aneurysms are alarmingly high if the condition remains untreated, highlighting the potential risks associated with this condition and the need for timely intervention. Conventional surgical treatment, currently the preferred treatment for symptomatic or growing aneurysms, consists of open resection of the entire aneurysm with or without artery replacement using an interposition graft [7].

We report our experience at the Vascular Surgery Department with a 25-year-old male with active vasculo-Behçet's disease who presented with a large pseudoaneurysm of the left internal carotid artery, which was treated with pseudoaneurysm exclusion. Using a reversed great saphenous vein graft interposition. It's important to note that the patient consented to publish his case report, demonstrating our respect for patient privacy and ethical conduct. This case report has been reported in line with the SCARE 2020 Criteria [8].

Case Presentation

A 25-year-old man presented with a sore throat, dysphagia, weight loss (8 kg in 2 months), and swelling on the left side of his neck. He was referred to our vascular surgery department for assessment of an expanding pulsatile mass in the left side of his neck, which was associated with numbness and weakness in the right side of his body. He currently smokes smoking one pack per day for the last ten years. No allergy to any drug has been reported. The patient had never undergone any surgical intervention.

A detailed medical history revealed that the patient had recurrent oral and genital aphthous ulcerations. There was no history of trauma, local surgery, or irradiation in his neck. He denied any headache, visual loss, speech difficulty, weakness, or sensory loss in other parts of the body. He had no fever, chills, palpitations, dyspnea, abdominal pain, or urinary symptoms.

One month before the presentation, he developed right-sided lower extremity deep vein thrombosis (DVT). He was initially treated with low-molecular-weight heparin (LMWH) and later discharged home on warfarin after a period of overlap.

On admission, the patient was hemodynamically stable and afebrile. His vital signs on arrival were: heart rate 76/minute, respiratory rate 14/minute, blood pressure 110/70 mmHg, and O2 saturation on room air was 100%.

Clinical examination revealed a left-sided neck mass that was pulsatile and tender on palpation. Neurological examination revealed decreased sensation in the right side of the body. The ophthalmic assessment was performed without significant pathological findings. Numerous scars of old ulcers were evident on the scrotal skin.

Arterial and Venous Doppler Ultrasound of the right lower extremity showed non-occlusive deep vein thrombosis (DVT) in the right common femoral vein (CFV) with acceptable filling and compressible right femoral vein till mid-level, where it became partially compressible with acceptable filling. The right confluence of the superficial inguinal veins and the great saphenous vein (GSV) were thrombosed. The right common femoral artery (CFA) and the proximal right superficial femoral artery (SFA) were patentable with triphasic signals. Doppler examination of the left extremity was unremarkable.

Doppler assessment of the neck revealed a 35 x 35 mm pseudoaneurysm with an intraluminal thrombus arising from the proximal left internal carotid artery (ICA), which has a wide neck. However, the distal left ICA was patent, with a normal Doppler Signal measuring proximally 4.2 mm and distally 6 mm [Figure.1].

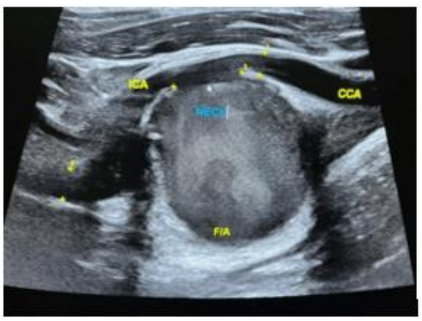


Fig. 1. A Color Doppler Ultrasound showed about 35 x 35 mm pseudoaneurysm with intraluminal thrombus arising from proximal left internal carotid artery (ICA) with wide neck.

In preparation for surgery, a computed tomography angiography (CTA) of the neck, chest, abdomen and lower extremities was performed to exclude aneurysmal lesions. Neck CTA revealed a 43 x 42.3 x 35.8 mm pseudoaneurysm in the left internal carotid artery [Figure 2, 3, 4, 5]. A magnetic resonance imaging (MRI) of the brain showed multiple acute infarctions involving the left posterior temporal, left frontal and left parietal lobes with no intracranial haemorrhage.

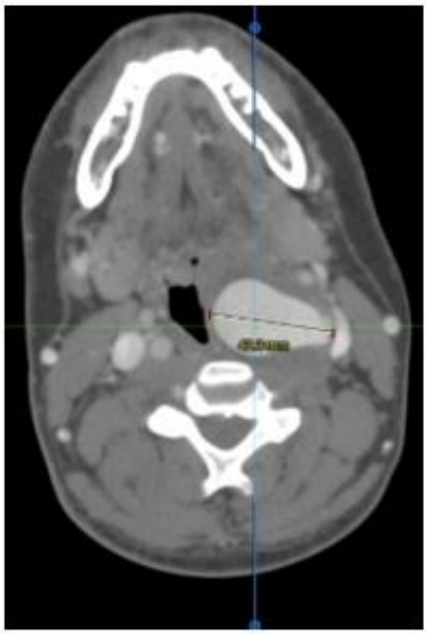


Fig. 2. A CT angiography of the neck: Axial view demonstrating a pseudoaneurysm of the left ICA with a maximal diameter of 42.3 mm.

Fig. 3. A CT angiography of the neck: Coronal view demonstrating a pseudoaneurysm of the left ICA.

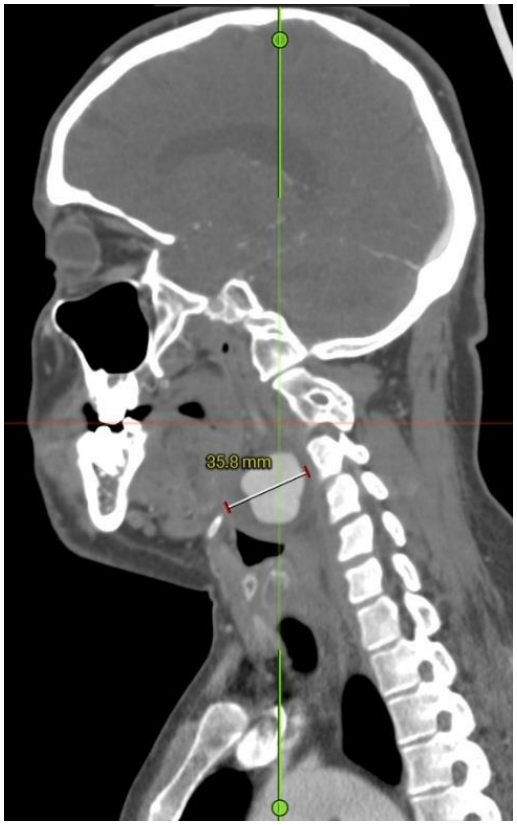


Fig. 4. A CT angiography of the neck: Sagittal view demonstrating a pseudoaneurysm of the left ICA.

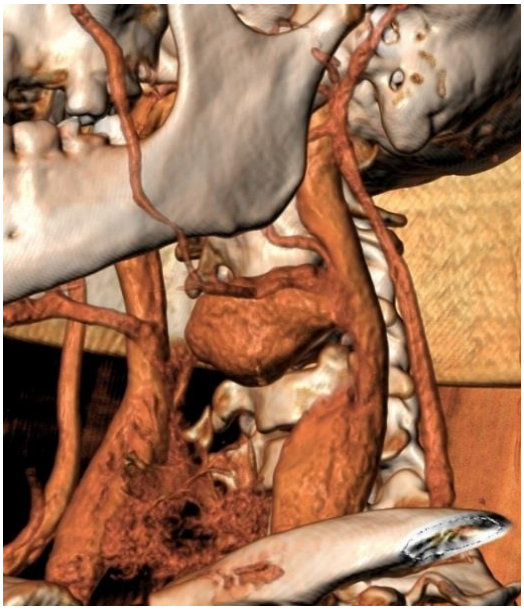


Fig. 5. A CT angiography with 3D reconstruction of the neck demonstrating a pseudoaneurysm of the left ICA

Laboratory studies revealed a severe inflammatory reaction response; WBC was 18,000 /µL; the erythrocyte sedimentation rate (ESR) was 100 mm/1st h (normal range <25), and the C-reactive protein (CRP) was 66 mg/L (normal range 0.0–5.0). A complete blood count (CBC) revealed normal hematocrit and platelet count. The coagulation profile: Prothrombin time (PT) was 16.5 sec; Activated Partial Thromboplastin

Clotting Time (aPTT) was 38.5 sec; and an international normalised ratio (INR) was 1.9.

Test results for autoimmune antibodies: serum antinuclear antibody (ANA), antineutrophil cytoplasmic antibody (ANCA), and Anticardiolipin (aCL) IgG and IgM antibodies were negative. The HLA-B51 was negative. Tests of infectious diseases, including syphilis, tuberculosis, and human immunodeficiency virus, were all negative. The pathergy test was positive.

After consultation with a rheumatologist, the patient was started on pulse steroids (methylprednisolone 1000 mg intravenously for five days) and cyclophosphamide 500 mg intravenously. Urgent surgery was deemed necessary to prevent fatal complications of extracranial carotid artery aneurysms and because most neurologic events are secondary to the embolization of thrombotic material from within the aneurysm wall.

After having a confirmed consent, the preoperative assessment included an imaging review and planning of proximal and distal anastomosis points. In addition, a venous examination of the left lower limb was also done to select a suitable autologous vein for use as a graft for arterial reconstruction. Preoperative Doppler ultrasound was performed to assess the condition of the left great saphenous vein (GSV) and to rule out deep vein thrombosis (DVT). The results revealed a patent left GSV with good calibre without evidence of DVT.

General anaesthesia was performed by endotracheal intubation. Arterial and central venous lines were inserted. The patient was positioned supine for a carotid endarterectomy. The left lower extremity was prepped and draped for saphenous vein harvesting. A skin incision was made in the neck along the anterior border of the sternocleidomastoid muscle. Proximal and distal control of the left common, internal and external carotid arteries were performed.

An incision along the pre-marked left great saphenous vein (GSV) was made, followed by harvesting the vein to be used in a reversed technique to repair the internal carotid artery after excluding a pseudoaneurysm.

After administering heparin, the common carotid artery was clamped proximal to the carotid bifurcation, excluding the pseudoaneurysm. Due to the large arterial wall defect and resection of damaged tissue, the gap between the internal and common carotid arteries was too long. Therefore, we performed a bypass between the internal and common carotid arteries using a reversed great saphenous vein graft interposition. A Javid shunt was inserted during surgery to decrease the risk of preoperative stroke by allowing continued perfusion during the procedure [Figure. 6, 7].

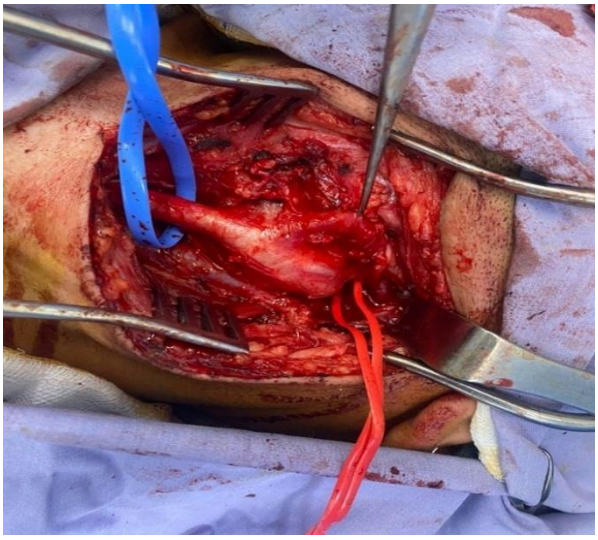


Fig. 6. An intraoperative photo showing a surgical exposure of a pseudoaneurysm of the left ICA with control of the distal segment of CCA and proximal segment of the left internal carotid artery (ICA).

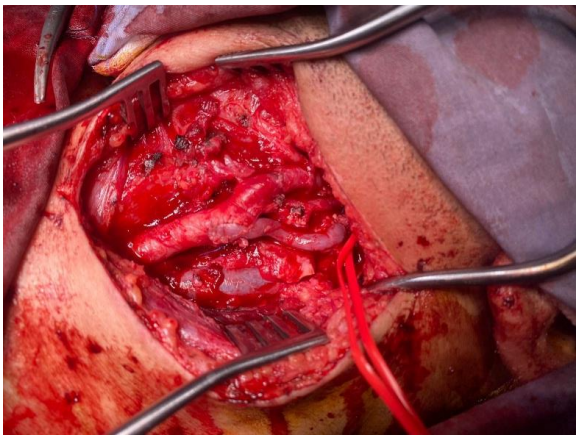


Fig. 7. An intraoperative image after interposition of the reversed saphenous vein between the internal and common carotid arteries by end-to-end anastomoses.

Postoperatively, the patient had an uneventful recovery; pain and swelling subsided immediately. During his hospitalisation, the patient received oral antimicrobial treatment and oral calcium plus vitamin D. Oral prednisolone and intravenous cyclophosphamide were continued postoperatively and adjusted according to the disease activity and rheumatologic assessment.

The patient was discharged from the hospital on the 5th postoperative day. He was placed on warfarin 5 mg daily, clopidogrel (Plavix®) 75 mg daily, atorvastatin 10 mg at bedtime, and oral steroids (prednisolone). Infliximab was added as maintenance therapy for the patient. Pre-discharge instructions were given for strict smoking cessation advice and regular follow-up in the clinic.

After two weeks, the patient returned to the clinic in good condition without complications. At his three-month follow-up visit, his systemic symptoms

improved with normal ESR and CRP values. In addition, four months postoperatively, a follow-up arterial Doppler ultrasound showed a patent vein bypass graft and the absence of recurrences [Figure. 8].

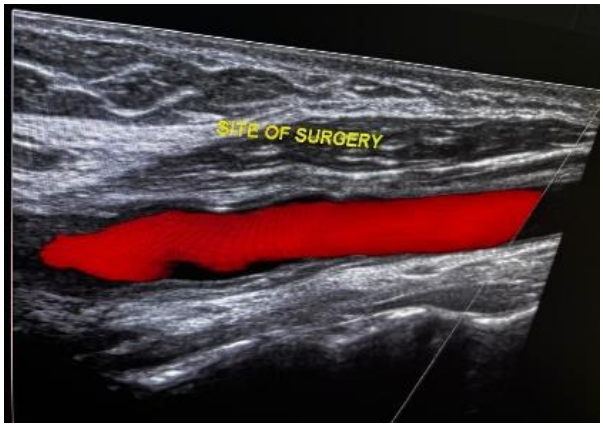


Fig. 8. Four months postoperatively, follow-up arterial Doppler ultrasound showed patent vein bypass graft, and the absence of recurrences.

Clinical Discussion

Behçet's disease (BD), a scarce condition first described by the Turkish dermatologist Hulusi Behçet in 1937, is a multisystem vasculitis typically presenting with oral and genital aphthosis and uveitis. Behçet's disease primarily affects young men between the ages of 20 and 40. Given its extreme rarity and frequent occurrence along the historic Silk Road trade routes, the Mediterranean, and the Middle and Far East, it is an area of study of utmost importance [9].

Behçet's disease is a complex condition characterized by different symptoms and varying degrees of severity. Its diagnostic process is complicated, lacking a definitive test or biomarker. Diagnosing Behçet's disease is difficult and depends on clinical criteria consisting of clusters of manifestations. This complexity underscores the need for further research and the development of more precise diagnostic tools [10].

Venous thrombosis occurs in 30%. It is much more common in men. Most have thrombosis of a leg vein (87%), thrombosis in a deep vein of the arm (2%), the vena cavae (8% each), cerebral venous sinus thrombosis (7%), the hepatic veins (3%) and the pulmonary arteries (2%) is less common and intracardiac thrombosis may occur [11, 12, 13]. Relapse occurs in 30% with a cumulative risk over 5 years of 38.4% [11]. Thromboembolism is rare; the thrombus adheres tightly to the vessel wall, and the veins become obliterated by organisation and fibrosis [12, 13].

Our research has revealed that the most common arterial manifestation in patients with Vasculo-Behçet's disease is the development of pseudoaneurysms, with a much higher frequency than true aneurysm formation. A pseudoaneurysm is a collection of blood that forms between the two outer layers of an artery, usually due to trauma or injury to the vessel wall. The underlying pathology of the affected artery is primarily active vasculitis, which initially occurs around the perivascular vasa vasorum and is a key factor in the development of pseudoaneurysms, leading to transmural necrosis, progressive thickening of the vessel wall and aneurysmal dilatation. Finally, perforation of the vessel wall and formation of a pseudoaneurysm. These findings have the potential to impact patient care significantly, guiding future research and treatment strategies to improve outcomes for patients with Behçet's disease [14, 15].

Managing vascular complications in Behçet's disease requires the expertise of professionals from various disciplines, including internal medicine, neurology, ophthalmology, gastroenterology, dermatology, rheumatology and vascular surgery. This multidisciplinary approach is crucial for effective patient care and emphasizes the disease's complex nature. It also highlights the necessity of collaboration in the medical field [16].

Immunosuppressive therapy, such as cyclophosphamide or azathioprine, is the mainstay treatment for Vasculo-Behçet's disease. Immunosuppressive therapy should always be considered to achieve complete remission, prevent recurrences, and reduce the risk of postoperative complications [17]. The significant reduction in postoperative complications in patients receiving immunosuppressants should instil confidence in their use, improving the overall prognosis [18].

Anticoagulants may be added to prevent the recurrence of venous thrombosis, post-thrombotic syndrome, and recurrent arterial occlusive events. [16].

The current options for managing pseudoaneurysms require considering multiple factors, including the availability of vascular intervention facilities, aetiology, size, location, and accessibility. Therefore, depending on the case, conservative management by ultrasound-guided compression and thrombin injection, surgery, and endovascular interventions are different approaches [20].

Open surgical options should be emphasized as the first-line option in patients with vasculo-Behçet's disease (v-BD) unless the cardiopulmonary risk is very high. The choice between open and endovascular interventions, which largely depends on the size and location of the pseudoaneurysm, is greatly influenced by the surgeon's experience. This underscores the crucial role of the surgeon in the decision-making process and the need for a personalized approach to treatment [20].

Endovascular stents have the advantage of reducing the incidence of postoperative complications by

limiting endothelial injury to the already fragile arterial wall. However, anatomical limitations and aneurysm formation commonly occur at arterial puncture sites or along the edge of a stent graft. Covered-stent grafts have recently been used to exclude pseudoaneurysms in some selected cases, preserving patients from the potential risks of mortality and morbidity due to the invasive nature of surgery [21].

Extracranial carotid artery aneurysms (ECAA) are rare, especially compared to atherosclerotic occlusive disease in the same location. An ECAA is a fusiform or saccular aneurysm between the origin of the common carotid artery (CCA) at the aortic arch and the internal carotid artery (ICA) at the skull base. The true incidence of ECAA is unknown, but it represents only 0.6% to 3.8% of procedures performed for extracranial cerebrovascular disease and is probably <2% of all carotid diseases. This rarity underscores the need for wariness and thoroughness in diagnosis and treatment [19].

An aneurysm is a pathological localized permanent dilatation of an artery, having at least a 50% increase in diameter compared with the expected normal diameter of the artery. However, the normal carotid bifurcation is typically 40% greater in diameter than the more distal ICA. Therefore, ECAA can be defined as bulb dilation >200% of the ICA diameter or >150% of the common carotid artery diameter. This definition has been internationally accepted and applied in the surgical literature [22].

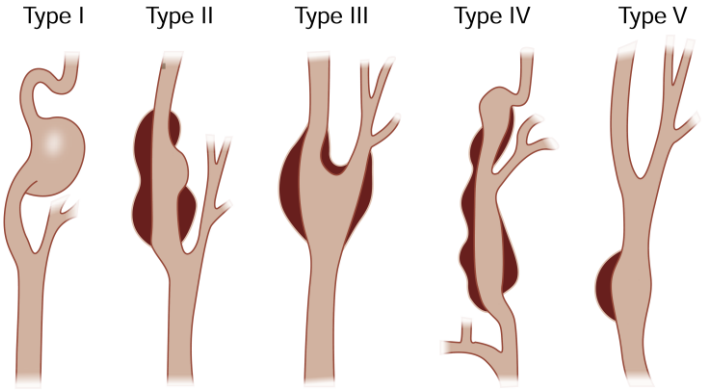
The presentation of extracranial carotid artery aneurysms varies depending on their location, size, and aetiology. The most common symptom is a painless, pulsatile neck mass, the initial symptom in 93% of patients in the series reported by Zhou et al. Imaging techniques such as computed tomography angiography (CTA), magnetic resonance angiography (MRA), or catheter-based angiography play a crucial role in diagnosis, as they are nearly always diagnostic when an aneurysm is present [19].

ECAAs can cause neurologic symptoms, with transient ischemic attacks occurring more frequently than completed strokes. Zhou et al. reported that 14% of patients presented with amaurosis fugax, transient ischemic attack (TIA), stroke, or Horner syndrome. Most neurologic events are secondary to the embolization of thrombotic material from within the aneurysm wall, potentially related to decreased flow and internal carotid artery compression from the mass effect of the large aneurysm. This emphasizes the need for continuous monitoring and vigilance in management [23].

ECAAs can present with a variety of symptoms related to cranial nerve dysfunction. Distal ICA aneurysms are more frequently associated with cranial nerve dysfunction than proximal aneurysms, but nerve injury or compression can also occur with large proximal carotid aneurysms. Common carotid artery aneurysms can cause hoarseness due to compression of the vagus nerve or the recurrent laryngeal nerve. Compression of the facial nerve can cause severe facial pain. Occasionally, the mass of a large aneurysm can cause difficulty swallowing and is discovered during evaluation for dysphagia. Protrusion of the aneurysm into the pharyngeal constrictor muscles can produce dysphagia. Difficulty swallowing may also be caused by nerve compression. Fortunately, rupture and haemorrhage are infrequent manifestations of carotid artery aneurysms. However, it's crucial to remember that mycotic aneurysms are especially susceptible to rupture and bleeding, underscoring the potential complications of ECAA and the need for vigilance in their management [19].

Attigah et al. performed a retrospective review of 57 patients who underwent 64 reconstructions for extracranial carotid artery aneurysms (ECAAs). They identified five different types of ECAA based on the anatomy of the aneurysm. This classification system, which has proven helpful in the surgical literature for describing the morphology and the basis for selecting a particular surgical approach, underscores the value of such systems in the practice of medical professionals, particularly surgeons and vascular specialists [See Table 2] - [See Figure.10] [24].

Attigah Classification of Extracranial Carotid Aneurysms (ECAAs)	
I	isolated, short aneurysms of the internal carotid artery above the carotid bulb.
II	long aneurysms of the ICA, ranging from the carotid bulb up to the line of Blaisdell, which is the line between the mastoid process and the angle of the mandible.
III	aneurysms of the proximal ICA and the carotid bifurcation.
IV	extensive aneurysms involving the CCA and ICA or concomitant separate CCA and ICA aneurysms.
V	isolated aneurysms of the CCA



The primary objective of treating extracranial carotid artery aneurysms (ECAA) is to prevent permanent neurologic deficits that may arise from atheroembolism and thromboembolism. The main goal is achieved by excluding the aneurysm from the arterial circulation and restoring antegrade flow. The choice of therapy should be individualised for each patient based on the location, size, underlying pathology, and comorbidities. With their skill and experience, modern vascular surgeons possess a vast array of open surgical and endovascular options, which have proven highly effective. Their expertise is invaluable in achieving the best possible outcomes for patients [19].

Indications for surgical interventions in patients with extracranial carotid artery aneurysms (ECAA) are (1) History of neurologic symptoms or cerebrovascular accident *(TIA/CVA) attributable to aneurysm, (2) Prevention of TIA/CVA, (3) Relief of compressive symptoms, (4) Erosion into adjacent structures, (5) Infection, (6) The aneurysm reaching 2 cm or more in diameter, (7)* Progressive expansion of the aneurysm [19].

Resection of the aneurysm with restoration of antegrade flow is the conventional treatment of ECAAs in contemporary practice. Autologous or prosthetic interposition grafts can be used with excellent results. Autologous grafts are preferred if infectious aetiology is suspected. Endovascular management of ECAAs, a less invasive approach, offers the advantages of avoiding a difficult dissection and eliminating the need for high cervical exposure, thus reducing the risk for cranial nerve injuries and other procedure-related complications [23].

In our case presentation, a young patient presented with extracranial carotid artery aneurysms (ECAA), a factor that significantly influenced our treatment decision. We opted for open surgical exclusion and repair, considering the available resources. We preferred autologous vein graft reconstruction over synthetic materials to minimize the risk of graft infection and occlusion. Surgical prosthetic infection is a significant cause of morbidity and mortality.

CONCLUSION

Emphasizing the importance of regular follow-up and appropriate treatment of Behçet's disease (BD) is crucial. This approach is mandatory to prevent irreversible organ damage, particularly in the early, active stages of Behçet's disease. It's important to note that one of the life-threatening manifestations of Behçet's disease is its vascular complications, further underscoring the importance of this issue.

It's crucial to remember that extracranial carotid artery aneurysms (ECAAs) and pseudoaneurysms are relatively rare. However, they have disastrous consequences when they occur. They are usually asymptomatic, and their symptoms vary according to location, size, and aetiology. The most common presentation is a painless pulsatile neck mass. Hemispheric neurologic events may also be the initial symptom of extracranial carotid artery aneurysms.

The treatment of ECAAs in patients with Behçet disease is a complex process, often fraught with complications and challenges. Reconstruction must be performed in healthy arterial segments, necessitating an extremely meticulous technique and immunosuppressive therapy.

The primary objective of treating aneurysms of the extracranial carotid artery (ECAAs) is to prevent permanent neurologic deficits that may arise from thromboembolism and aeroembolism and avoid catastrophic consequences such as disabling stroke or even death. In contemporary practice, resection of the aneurysm with restoration of antegrade flow is the conventional treatment of ECAAs.

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Ethical approval

Ethical approval has been taken from The Ethical Committee at King Hussein Medical Center, Jordanian Royal Medical Services (JRMS), Amman, Jordan. The reference number is 9/2025–50. The date of approval by the ethics committee 15/7/2025.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Mohammad. A. Al-doud, Bashar K. Anakrih, Khaldoon O. Alwreikat, Husam I. Al-khawaldeh, Fuad I. Khamis, Thair M. Al-Tarabsheh, Muhammad A. Al-Rawashdeh

Authorship contribution statement

- Interpretation of the CT images: • Mohammad A. Al-Doud, Bashar K. Anakrih.
- Pre-Operative care: • Khaldoon O. Alwreikat, Husam I. Al-khawaldeh, Fuad I. Khamis, Thair M. Al-Tarabsheh.
  Operation:
- Main Surgeon: Muhammad A. Al-Rawashdeh, Mohammad A. Al-doud.
- Assistants Surgeon: • Bashar K. Anakrih, Khaldoon O. Alwreikat, Husam I. Al-khawaldeh, Fuad I. Khamis.
- Post-Operative Care: • Bashar K. Anakrih, Khaldoon O. Alwreikat, Husam I. Al-khawaldeh, Fuad I. Khamis, Thair M. Al-Tarabsheh
- Drafting the manuscript: • Mohammad A. Al-doud.
- Revising the manuscript critically for important intellectual content: • Mohammad. A. Al-doud, Bashar K. Anakrih
- Approval of the version of the manuscript to be published (the names of all authors must be listed): • Mohammad. A. Al-doud, Bashar K. Anakrih, Khaldoon O. Alwreikat, Husam I. Al-khawaldeh, Fuad I. Khamis, Thair M. Al-Tarabsheh, Muhammad A. Al-Rawashdeh

Guarantor

Dr. Mohammad A. Al-Doud.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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