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Case Report

Endovascular Management of A Traumatic Carotid Cavernous Fistula Accompanied By A Pseudoaneurysm: A Case Report

Ayca OZKUL[1], Ali YILMAZ[2], Dong Seong SHIN[3], Bum-Tae KIM[3]

[1]Adnan Menderes University, Faculty of Medicine Neurology Department, Neurology, Aydin, Turkey [2]Adnan Menderes University Faculty of Medicine, Neurosurgery, Aydin, Turkey [3]Soonchunhyang University Bucheon Hospital, Neurosurgery, Seul, Korea, Republic Of

Abstract

Objective Carotid caverneous fistula (CCF) accompanied by a pseudoaneurysm is a rare life-threatening condition that requires emergency treatment. Herein, we report a case of traumatic CCF accompanied by a pseudoaneurysmal intracranial hemorrhage from the intradural internal carotid artery.
Case A man aged 29 years developed a traumatic intracranial hemorrhage after a motor vehicle accident. Cerebral computed tomography (CT) showed an acute subdural hematoma on the left frontal and temporal lobes. There was also an intracranial hemorrhage in the left inferior frontal lobe and a diffuse subarachnoid hemorrhage involving the prepontine cistern, basal cistern, bilateral sylvian fissure, and interhemispheric fissure. Decompressive craniectomy was immediately performed. In a follow-up CT, contrast extravasation in the left inferior frontal lobe with simultaneous venous filling in the left cavernous sinus was detected, concomitant with a high-flow CCF. Cerebral angiography showed that a CCF developed in the left internal carotid artery and a pseudoaneurysm was also detected above the CCF. The fistula was occluded through coil embolization and the pathologic lesions were covered with stents resulting in the disappearance of the fistula and pseudoaneurysm.
Conclusion Endovascular stent-assisted coil embolization can be successfully performed in this rare fatal disease without complications.

Keywords: Caroticocavernous fistula, endovascular treatment, pseudoaneurysm

Psödoanevrizmanın Eşlik Ettiği Travmatik Karotikokavernöz Fistülün Endovasküler Tedavisi: Bir Olgu Sunumu

Özet

Amaç Psödoanevrizmanın eşlik ettiği karotikokavernöz fistül (CCF) hayatı tehlikesi olan ve acil tedavi gerektiren nadir bir durumdur. Biz burada intradural internal karotis arterden kaynaklı psödoanevrizmal intrakranial kanamanın eşlik ettiği travmatik CCF olgusunu sunmaktayız.
Olgu 29 yaşında erkek hastada araçlı trafik kazası sonrası travmatik intrakranial hemoraji gelişmiş. Serebral bilgisayarlı tomografide (CT) sol frontal ve temporal loblarda akut subdural hematom izlendi. Aynı zamanda sol inferior frontal lobtaintrakranial hemoraji ve prepontin, bazal sistern, bilateral slyvian ve interhemisferik fissürleri etkileyen diffüz subaraknoid kanama mevcuttu. Dekompresif kraniektomi hemen uygulandı. Kontrol CTde sol kavernöz sinüste simultan venöz dolumla birlikte sol inferior frontal lobta kontrast ekstravazasyonu

görüldü. Serebral anjiografide sol internal karotid arterde CCF geliştiği izlendi ve CCF üzerinde psödoanevrizma saptandı. Fistülün koil embolizasyon ile oklüde edilmesi ve patolojik lezyonların stent ile kaplanması sonucunda fistül ve psödoanevrizma görünümünü kayboldu.
Sonuç Endovasküler stent destekli koil embolizasyonu, bu nadir ölümcül hastalık tedavisinde komplikasyonsuz başarıyla uygulanabilir.

Anahtar Kelimeler: Karotikokavernöz fistül, endovasküler tedavi, psödoanevrizma

INTRODUCTION

Carotid caverneous fistula (CCF) is an abnormal communication of the internal or external carotid arteries with the cavernous sinus that occurs through traumatic carotid artery injury. In some cases, a carotid artery pseudoaneurysm can occur with a traumatic internal carotid artery (ICA) laceration. However, it is rare for a CCF to be accompanied by a pseudoaneurysm. The definitive management of a CCF is obliteration of the fistulous connection with restoration of normal arterial and venous flow. This is usually achieved through an endovascular approach, preserving the parent ICA. CCF embolization using a detachable balloon or coil occlusion has been widely accepted as a therapeutic option (1-3). Here, we present a case of a traumatic CCF accompanied by a pseudoaneurysmal ruptured intracranial hemorrhage. Although it is a rare and life-threatening condition, endovascular treatment can be safely performed in such combined lesions.

CASE PRESENTATION

A man aged 29 years was admitted to our neurosurgery department after a traumatic head injury due to a motor vehicle accident. He was stuporous, and scored 7 on the Glasgow coma scale. Massive nasal bleeding and facial lacerations were noted on both nostrils and the peri-orbital area, and facial edema was present. The left pupil was dilated and non-responsive to light reflex. Brain computed tomography (CT) revealed multiple fractures of the bilateral frontal, left parietotemporal, and right temporal bones; ethmoid walls, left maxillary, and sphenoid sinuses; bilateral sphenoid wings; orbits; and nasal bones. Intracranial hemorrhage was present in the left inferior frontal lobe, and a diffuse subarachnoid hemorrhage involving the prepontine cistern, basal cistern, bilateral sylvian fissure, and interhemispheric fissure was also present with acute subdural hematomas on the left frontal and temporal lobes (Figure 1a, b). Decompressive craniectomy with hematoma evacuation was immediately performed. The follow-up brain CT and CT angiography (CTA) revealed contrast extravasation in the left inferior frontal lobe (Figure 2a). On the left ICA-selective angiography, a high-flow shunt was visualized from the left ICA paraclinoid lesion to the cavernous sinus, and shunting flow drainage was observed through the inferior petrosal sinus. A pseudoaneurysm located on the left paraclinoid ICA measuring 4.5 mm in diameter was also detected (Figure 2b).

Endovascular treatment was performed for the CCF and the pseudoaneurysm by using guide catheter irrigation device (4). A 6-Fr Chaperon guiding catheter (MicroVention, Tustin, CA, USA) was navigated into the left proximal ICA. A Prowler Selector Plus microcatheter (Codman, Raynham, Massachusetts, UA) was initially placed in the middle cerebral artery. A 90-degree preshaped Excelsior SL-10 microcatheter (Stryker Neurovascular, Fremont, CA, USA) with

Synchro microwire (Stryker Neurovascular, Fremont, CA, USA) was used for the coil embolization of the pseudoaneurysm; pseudoaneurysm obliteration was achieved with embolization using two coils. Although a 90-degree microcatheter was initially used for fistular selection, which was not effective, and a C-shaped catheter was subsequently used successfully. The fistula was occluded through embolization using four coils. After coil embolization, two Enterprise stents (Codman, Raynham, Massachusetts, USA) (4.5 mm x 22 mm and 4.5 mm x 28 mm) were delivered using a Prowler Selector Plus microcatheter, and the pathologic lesions were covered (Figure 2c, d, e, f), resulting in the disappearance of the fistula and the pseudoaneurysm. Follow-up angiography was performed 1 day post-operatively, which confirmed no evidence of recurrent fistula or regrowth of the pseudoaneurysm. There was also no evidence of intimal hyperplasia or thrombus formation within the segments of the stented ICA.

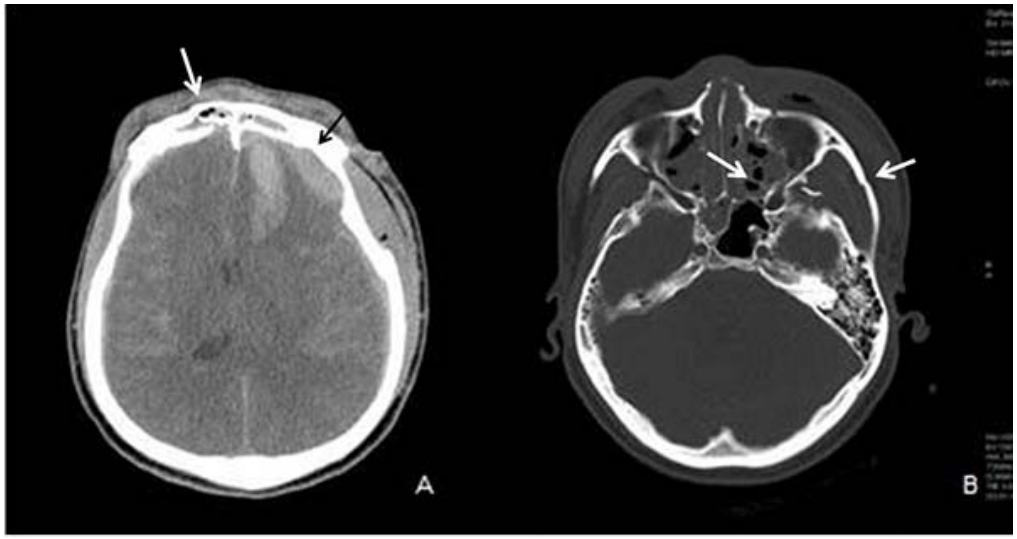

Figure 1: *Brain computed tomography showed intracranial hemorrhage involving the left inferior frontal lobe, causing midline shifting (black arrow) (A) and multiple fractures of the frontal and left parietotemporal bones and walls of the ethmoid sinus (white arrows)(B).*

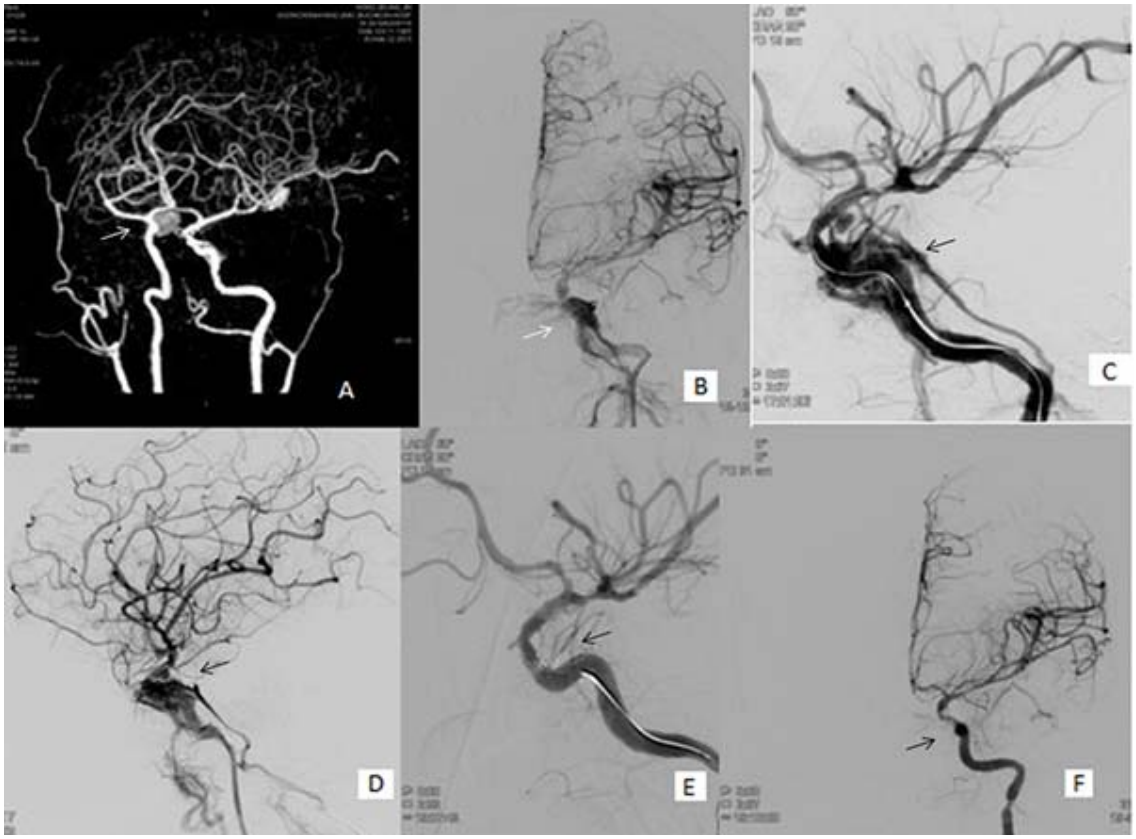

Figure 2: *In CTA and cerebral DSA, contrast extravasation in the left inferior frontal lobe concomitant with a CCF was detected (white arrows) (A,B). In cerebral DSA, drainage through the inferior petrosal sinus suggesting a CCF and a pseudoaneurysm in the left paraclinoid ICA was detected (black arrow) (C). The stent was placed before coil embolization to preserve the ICA (black arrow) (D) and stent-assisted coil embolization of the CCF and pseudoaneurysm was performed (black arrow)(E). Endovascular treatment preserving the patency of the ICA (black arrow) was performed (F).*
CTA: computed tomographic angiography, DSA: digital subtraction angiography, CCF: carotid cavernous fistula, ICA: internal carotid artery.

DISCUSSION

We presented a rare case of high-flow, type A CCF with a pseudoaneurysm. A traumatic CCF is a potentially life-threatening condition that requires urgent treatment, particularly in the presence of a pseudoaneurysm (1,5). An injury of the ICA occurs in 1 in 10,000 hospital admissions due to trauma (6). Traumatic intracranial pseudoaneurysms are also rare lesions, constituting only 0.15% to 0.40% of all intracranial aneurysms (3). Severe head trauma can damage the cavernous portion of the ICA and may result in a CCF, pseudoaneurysm, dissection of the artery or rupture and fatal hemorrhage (3). Early recognition of clinical symptoms of a CCF-like epistaxis, proptosis, and chemosis is essential for diagnosis and urgent treatment. The mechanism behind pseudoaneurysm formation can be described as blood flowing from the injured ICA directly into the bare perivenous spaces within the caverneous sinus, without having a shunt with one of the veins (6).

CCFs are angiographically classified into four types. Type A fistulas are a direct shunt between the ICA and the cavernous sinus, with high flow. Type B CCFs occur if the shunt is between the meningeal

branches of the ICA and cavernous sinus. In type C CCFs, the fistula is located between meningeal branches of the external carotid artery (ECA) and the cavernous sinus. Finally, type D CCFs are located between the meningeal branches of both the ECA and the ICA and the cavernous sinus. Most type A CCFs are high flow and traumatic in origin. Clinical symptoms are related to the size, duration, and location of the CCF; adequacy and route of venous drainage; and presence of arterial and venous collaterals (2). A CCF may present with increased intracranial pressure, rapidly progressive proptosis, diminished visual acuity, hemorrhage, and transient ischemic attacks. Cortical venous drainage from the CCF may also cause increased intracranial pressure with a high risk of intraparenchymal hemorrhage, which have poor prognosis (2). Patients presenting with ruptured pseudoaneurysms, massive epistaxis, and increased intracranial pressure have a high mortality rate (2). Identification of these high-risk features provides the basis for decision-making concerning appropriate treatment (3).

Our patient presented with a CCF accompanied by a pseudoaneurysm, with an interesting trauma mechanism. The patient had multiple facial fractures with massive epistaxis, and CT revealed multiple fractures of paraclinoid bony structures (ethmoid bone and orbital roof). Traumatic force was delivered to the paraclinoid ICA, which led to simultaneous intra- and extradural lesions on the ICA, resulting in an initial massive subarachnoid hemorrhage due to a pseudoaneurysm, and a CCF due to extradural ICA tearing. Additionally, the patient presented with severe head trauma as well as intraparenchymal hemorrhage with increased intracranial pressure that required urgent decompressive craniectomy. Brain CT and cerebral angiography also revealed drainage through the inferior petrosal sinus and a pseudosaccular aneurysm in the left paraclinoid ICA. Thus, our patient had a high risk of mortality and required immediate treatment.

It is difficult to diagnose a pseudoaneurysm and select an appropriate treatment strategy, because pseudoaneurysms do not have true walls. Surgical treatments include direct clipping, wrapping, trapping, and carotid artery ligation. However, these are difficult to perform because a pseudoaneurysm has a fragile fibrous wall with a broad neck, which results in a high risk of hemorrhage and ischemia during clipping. Therefore, some physicians prefer ligation of the parent artery (6). However, the hemodynamic effect of ICA occlusion should be evaluated initially to reduce mortality and morbidity after permanent carotid occlusion. Balloon test occlusion of the carotid artery is useful for managing patients before ligation of the ICA. In case of a limited cerebrovascular reserve, the surgeon should prepare for a bypass procedure. Nevertheless, endovascular strategies that preserve ICA patency should also be considered. Such vessel-preserving options include endovascular stents and coils, covered stents, and detachable balloon placement (1,2,7). Trans venous embolization of the shunt can also be performed (8).

The use of Onyx for the transarterial balloon-assisted embolization of traumatic CCFs has been shown to be feasible and effective (9). However, the safety and efficacy of this endovascular treatment requires further evaluation with respect to its use in cases of CCF combined with a pseudoaneurysm.

A traumatic pseudoaneurysm is a fragile sac surrounded by a blood clot that may disappear with fibrinolysis. Although a thin fibrin capsule may not be adequate to support the balloon in place over the

laceration in the carotid artery, it has been reported that detachable balloon occlusion of a pseudoaneurysm and fistula is a safe and efficient treatment choice. It is presumed that pseudoaneurysm sacs are very fragile along the line of bone fractures only (3). Coil placement is another method for the treatment of pseudoaneurysms (10); however, microcoils are too soft to tightly occlude the neck of pseudoaneurysms. In addition, intranasal extrusion of the endovascular coil after occluding the ICA for massive epistaxis has been reported in the literature (6). Overall, the use of endovascular microcoils should be considered with stent placement, depending on the weakness of the neck of the pseudoaneurysm.

Covered stent grafts can reconstruct injured vessels and have previously been used in the treatment of traumatic CCFs and pseudoaneurysms. The main disadvantage of this method is the poor navigability of the stents within intracranial vessels due to their rigidity. More recently, newly designed covered stents for use in intracranial vasculature have been employed successfully for the treatment of ICA pseudoaneurysms (9,11). However, high-dose anti-platelet and anti-coagulant therapy should be administered, which may cause hemorrhage (3).

Flow diverter stents have been developed to treat intracranial aneurysms. They are placed within the parent artery to reduce flow velocity in the aneurysm sac, resulting in gradual thrombosis of the aneurysm. Therefore, flow diverter stents can be used for untreatable, broad-necked, fusiform, or partially thrombosed intracranial aneurysms (5,10,12). However, clinical use in cases of a combined CCF with a pseudoaneurysm is limited. These novel devices may offer a simple and potentially safer vessel-sparing option in this rare condition. However, more clinical data are needed for its broader use, especially in cases where CCFs are complicated with pseudoaneurysms.

CONCLUSION

Traumatic CCF accompanied by a pseudoaneurysm is a life-threatening and rare condition that requires urgent treatment. These two lesions were successfully obliterated using endovascular stent-assisted coiling without complications in our patient.

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Correspondence to:
Bum Tae Kim
E-mail: bumtkim@gmail.com

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REFERENCES

1. *Celil G, Engin D, Orhan G, Barbaros C, Hakan K, Adil E. Intractable epistaxis related to cavernous carotid artery pseudoaneurysm: treatment of a case with covered stent. Auris Nasus Larynx 2004; 31: 275–278.*
2. *Huai RC, Yi CL, Ru LB, Chen G, Guo HH, Luo L. . Traumatic carotid cavernous fistula concomitant with pseudoaneurysm in the sphenoid sinus. Interv Neuroradiol 2008; 30: 59–68.*
3. *Isamat F. On posttraumatic intracavernous false aneurysms and arteriovenous fistulas. Acta Neurochir (Wien) 1987; 85: 148–153.*
4. *Ozkul A, Park JH, Shin DS, Yılmaz A, Kim BT.Invention of the Guide Catheter Irrigation Monitoring Device for Neuroendovascular Therapy. J Korean Neurosurg Soc. 2017;60:471-474.*
5. *Halbach VV, Hieshima GB, Higashida RT, Reicher M. Carotid cavernous fistulae: indications for urgent treatment. AJR Am J Roentgenol 1987; 149: 587–593.*
6. *Shin YS, Kim SY, Moon SK. Intranasal extrusion of the endovascular coil after occluding internal carotid artery for massive nasopharyngeal bleeding. Otolaryngol Head Neck Surg 2005; 133: 644*
7. *Li MH, Li YD, Gao BL, et al. A new covered stent designed for intracranial vasculature: application in the management of pseudoaneurysms of the cranial internal carotid artery. Am J Neuroradiol 2007; 28: 1579–1585.*
8. *Nomura M, Mori K, Tamase A, et al. Cavernous Sinus Dural Arteriovenous Fistula Patients Presenting With Headache as an Initial Symptom. J Clin Med Res. 2016 ; 8:342-345.*
9. *Yu Y, Huang Q, Xu Y, et al. Use of onyx for transarterial balloon-assisted embolization of traumatic carotid cavernous fistulas: a report of 23 cases. AJNR Am J Neuroradiol 2012; 33: 1305–1309.*
10. *Iancu D, Lum C, Ahmed ME, et al. Flow diversion in the treatment of carotid injury and carotid-cavernous fistula after transsphenoidal surgery. Interv Neuroradiol 2015;21: 346–350.*
11. *Madan A, Mujic A, Daniels K, Hunn A, Liddell J, Rosenfeld JV. . Traumatic carotid artery-cavernous sinus fistula treated with a covered stent. Report of two cases. J Neurosurg 2006; 104: 969–973.*
12. *Lin LM, Colby GP, Jiang B, Pero G, Boccardi E, Coon AL. . Transvenous approach for the treatment of direct carotid cavernous fistula following Pipeline embolization of cavernous carotid aneurysm: a report of two cases and review of the literature. BMJ Case Rep 2014;29: 2014.*