

Introduction

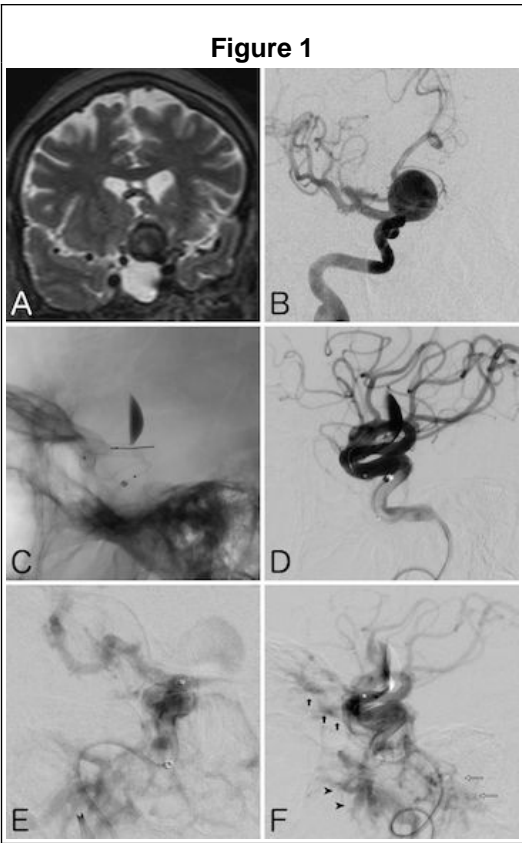
- Direct carotid cavernous fistulas (CCF) are rare complications of flow diversion treatment for cavernous carotid aneurysms (CCA).
- We present the first case of an intra-procedural direct CCF documented immediately after flow diversion for treatment of a symptomatic paraophthalmic right internal carotid artery (ICA) aneurysm.
- Our case was successfully treated with further flow diversion alone.

Case Presentation

- A 69-year-old woman with right-sided vision loss due to an unruptured paraophthalmic right ICA aneurysm, presented with worsening left-sided vision.
- On exam, no light perception in the right eye, 20/60 visual acuity in the left eye, right relative afferent pupillary defect with normal left pupillary response to light.
- MRI of the brain and digital subtraction angiogram (DSA) demonstrated a wide-necked right ICA aneurysm with mass effect on the optic chiasm.
- Due to the worsening left-sided vision, paraophthalmic location, and wide aneurysmal neck, flow-diversion was planned.

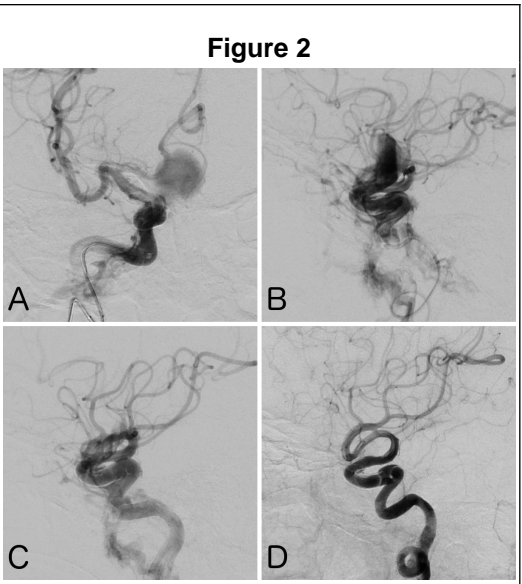
Endovascular Procedure

- 4.5mmx18mm pipeline embolization device (PED) was successfully deployed across the aneurysm neck.
- DSA showed early opacification of the right cavernous sinus, inf. petrosal sinus and sup. ophthalmic vein, consistent with a direct CCF.
- 4 additional PEDs telescoped across aneurysm neck and cavernous ICA, with decreased CCF flow.
- On post-intervention exam, conjunctiva were normal. No palpable thrill or bruit over either eye. Intraocular pressures measured 21mmHg in both eyes.
- On 2-month follow-up, she remained asymptomatic with DSA showing complete



(A) MRI brain, coronal T2-weighted image shows large paraclinoid right ICA aneurysm with mass effect on the optic chiasm. (B) DSA shows wide-necked aneurysm. (C) Unsubtracted lateral view immediately after PED #1 shows contrast stagnation in aneurysm, apposition of PED to parent ICA. (D) DSA of right ICA in lateral view immediately after PED #1 shows patent antegrade flow and no evidence of CCF. Subsequent DSA of right ICA in (E) frontal and (F) lateral views shows decreased antegrade flow and development of CCF with retrograde flow anteriorly through superior ophthalmic vein (arrows), inferiorly to pterygoid plexus (arrowheads) and clival plexus (open arrows).

Table 1: Reported Cases of Direct CCFs after Flow Diversion				
Study, year	Aneurysm Type	Aneurysm treatment	Interval to CCF formation	Treatment of CCF
Lin et al., 2015	Cavernous left ICA aneurysm	PED	6 weeks	Transvenous coil embolization of CS
Lin et al., 2015	Cavernous left ICA aneurysm	PED	1 month	Transvenous coil embolization of CS
Park et al., 2014	Traumatic carotid cave/cavernous ICA pseudoaneurysm	PED	Short term	Transvenous coil embolization
Bescke et al. (PUFS), 2013	Cavernous ICA aneurysm	PED	180 days	Unknown
Kulcsar et al., 2011	Cavernous left ICA aneurysm	Silk	3 days	Parent artery occlusion
Kulcsar et al., 2011	Cavernous right ICA aneurysm	Silk	110 days	Parent artery occlusion
Mustafa et al., 2010	Cavernous right ICA fusiform aneurysm	Silk	2 weeks	Transvenous coil embolization



DSA of right ICA in (A) frontal and (B) lateral views after deployment of four additional PEDs shows improved antegrade flow and decreased filling of CCF. (C) DSA on post-intervention day #1 of right ICA in lateral view shows continued improvement of antegrade flow and further decreased filling of CCF. Note decreased opacification of superior ophthalmic vein, pterygoid plexus and clival plexus. (D) 2-month follow-up DSA shows vessel reconstruction with resolution of CCF.

Table 2: Reported Cases of Direct CCFs Treated with Flow Diversion				
Study, Year	Etiology	Finding	Treatment	Outcome
Nossek et al., 2015	Aneurysm rupture	Ruptured left cavernous carotid aneurysm with fistulous outflow via the left SOV and into the pterygoid venous plexi bilaterally	PED X 3 in left ICA; Transarterial coiling of the aneurysm and left cavernous sinus	Complete symptomatic resolution; obliteration of the fistula with occlusion of the aneurysm at 1-yr follow-up DSA
Pradeep et al., 2015	Trauma	Direct CCF of the left ICA with anterior and posterior venous drainage	PED X 2 in left ICA; Transvenous coil and Onyx embolization of left cavernous sinus	Persistent left CN6 palsy, improved exophthalmos and left lateral field deficit at 3 months
Pradeep et al., 2015	Trauma	High flow, direct left CCF	PED in petro-cavernous left ICA; Transvenous Onyx embolization of left cavernous sinus; PED X2 in Transarterial coil embolization of right cavernous sinus and SOV; Silk tent in cavernous right ICA	Completely remodeled ICA and occlusion of the CCF at 7-month follow-up DSA
Iancu et al., 2015	Post-surgical	Right ICA injury with CCF refluxing into right SOV	Right cavernous sinus and SOV; Silk tent in cavernous right ICA	Oculomotor symptoms immediately improved. Normal angiogram on 1-yr follow-up
Nadarajah et al., 2011	Trauma	Transection of cavernous right ICA with CCF; partially thrombosed and	PED X 4 in cavernous right ICA	Complete resolution

Discussion

- Development of a direct (Barrow type-A) CCF after flow diversion has been reported in only 7 patients (Table 1), all occurring subacutely, 3-110 days after treatment. (1-5) Ours is the first reported direct Barrow type-A CCF to develop immediately after PED deployment.
- The incidence of delayed spontaneous aneurysm rupture after treatment with a flow diverter ranges from 0.6% to 1%. (2, 6)
- The 2 hypothesized pathomechanisms of delayed aneurysmal rupture after flow-diversion involve development of intra-aneurysmal thrombosis and acute changes in flow hemodynamics. (4)
- Traditional management of direct CCF's resulting from rupture of an untreated CCA consists of transarterial obliteration of the fistulous site, transvenous embolization, parent artery sacrifice, or surgical ligation.
- The use of flow diversion for the treatment of direct CCFs has been reported in five cases (Table 2). (7-10) In all but one of these cases, transarterial flow diversion was combined with concomitant transvenous embolization of the ipsilateral cavernous sinus.
- This is only the second reported case of a direct Barrow type-A CCF to be successfully treated using solely flow diversion, without additional transvenous intervention.

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