Original Article

High Diagnostic And Therapeutic Value of Digital Subtraction Angiography in Direct Carotid-Cavernous Fistulas: A Retrospective Case Series

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Abstract

Aims: *To describe the diagnostic and therapeutic value of digital subtraction angiography (DSA) and endovascular treatment in patients with direct carotid cavernous fistula (CCF) over a period of three years.*

Settings: *Tertiary referral hospital, Chhattisgarh*

Design: *Retrospective, noncomparative case series.*

Material & Methods: *Medical records of consecutive patients who had direct CCF between January 2014 to January 2017 with minimum six months of follow-up were retrospectively analyzed. All patients were initially evaluated by non-invasive imaging technique followed by DSA for confirmation. Endovascular embolization was done in all cases. A clinical cure was considered when there was a complete resolution of signs and symptoms of direct CCF and when the fistula was not observed on repeat DSA after three months, which was labeled as anatomically cured patients.*

Results: *Total ten patients were analyzed; nine patients had unilateral CCF, only one had bilateral CCF. All patients had history of head trauma. Five (50%) patients were embolized with a coil, and the rest were embolized by a combination of coil and Onyx. In our study, carotid artery preservation was performed in 70% of patients. Latest clinical follow up showed good recovery Furthermore no procedure related complications were noted in any patients. There were no intraoperative or perioperative mortality noted.*

Conclusion: *With the observed results and low rate of complications and morbidity, endovascular therapy could be the primary treatment for patients with direct CCFs.*

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Introduction

Management of neuro-ophthalmic disorders has been dramatically revolutionalized by the advancements in neuroimaging and interventional techniques.[1] Digital subtraction angiography (DSA) is an interventional technique.

First developed in the 1970s and it is useful in the diagnosis and treatment of arterial and venous occlusions, arteriovenous malformations, and carotid-cavernous fistulas (CCFs).[2]

CCF is defined as an abnormal communication between the cavernous sinus and the carotid artery branches, which is categorized into direct and indirect types by Barrow. Type A (direct) fistulas involve a direct communication between the intracavernous portion of the internal carotid artery (ICA) and cavernous sinus, commonly due to trauma.[3] Direct CCFs are typically high flow type and usually requires active intervention because spontaneous resolution is rare.[4] The traditional management of CCFs is associated with mortality and several complications.[5] With advanced catheterization techniques and new embolizing agents, endovascular treatment is considered the treatment of choice for all CCFs. DSA determines the angioarchitecture of the injury and provides precise information about the size, location, and character of the fistula and its drainage pattern.[6,7]
This study aimed to evaluate the efficacy of DSA for the diagnosis and treatment of direct CCFs because untreated direct CCFs always results in poor visual prognosis, but can be managed promptly.

Subjects and Methods

The study was approved by the institutional ethical committee and adhered to the tenets of the Declaration of Helsinki. Informed consent was obtained from all patients before initiation of DSA and endovascular procedure. This was a retrospective observational study of patients with direct CCF visiting our department between January 2014 to January 2017. Case records of all ten patients with direct CCF during the study period who underwent angiographic evaluation and endovascular procedure were drawn from a medical record department. Data from patients more than eighteen years age with features of direct CCF and with a minimum follow up to six months were included for analysis. Patients with any prior ocular conditions that may influence visual acuity such as corneal opacity, glaucoma or retinal pathology were excluded from study. A retrospective analysis of all noninvasive imaging studies and angiographic reports was performed. The records were reviewed for demographic variables like age and sex, mode of trauma, time interval between injury and admission and comprehensive ophthalmic evaluation including best corrected visual acuity (BCVA) using Snellen's chart, intraocular pressure assessment using applanation tonometry, dilated fundus examination using slit lamp

biomicroscopy and indirect ophthalmoscope. BCVA was categorized into three categories- mild visual impairment (6/9 to 6/12), moderate visual impairment (6/18 to 6/36) and severe visual impairment (less than 6/60).

All patients were independently evaluated by an ophthalmologist and a radiologist before and after the treatment. A retrospective analysis of all non-invasive imaging studies and angiographic reports were done. All patients were first evaluated using noninvasive imaging techniques, followed by DSA for definitive diagnosis and management. Follow-up was scheduled at one, three, and six months of treatment. A clinical cure was considered when there was a complete resolution of signs and symptoms of direct CCFs, which was labeled as clinically cured patients, and when the fistula was not observed on repeat DSA after three months, which was labeled as anatomically cured patients. The primary outcome measure was resolution of signs and symptoms of direct CCF after endovascular treatment and secondary outcome measure was change in BCVA after treatment. The one-line improvement in Snellen's chart after procedure is considered as fair visual recovery, two-line improvement as good visual recovery and more than three-line improvement as a very good visual recovery.

Procedure of digital subtraction angiography

Written informed consent was obtained from all patients after undergoing pre-anesthetic checkup. The standard transfemoral Seldinger technique was used in all patients. After the selective injection of the ICA, ECA, and vertebral artery bilaterally, standard anteroposterior and lateral radiography and three-dimensional rotational angiography were performed. This provided precise information about the location of the fistula, collateral supply to the brain, and venous drainage pattern of the lesion. After complete assessment of the anatomy and architecture of CCFs, embolization treatment strategies were planned accordingly. Small carotid artery tears were treated with closure alone, while in cases with larger fistula trapping of the ICA, embolization using a coil or liquid embolus (Onyx) was considered. Hemostasis was achieved at the end of the procedure.[8] In the present study, we used coil and a liquid embolizing agent (i.e., Onyx).

Coils: They are made up of platinum or steel and hence easily visible on radiographic films. They induce clotting owing to the Dacron wool tails around the wire. They are significantly good for fast-flowing vessels because they immediately clot the vessel. Coils are better than balloon embolization because of the ease of access and availability of embolic devices of various sizes.[7] However, coils have a disadvantage in that a risk of incomplete fistula occlusion with loss of transarterial access is observed when using coils. Thus, some complications, such as thromboembolism, ICA compromise by protruding coils, and ICA dissection, can be observed.[4]

Liquid Embolic Agent (Onyx): It is a liquid embolic agent formed by a combination of ethylene vinyl alcohol copolymer, dimethyl sulfoxide, and tantalum powder. It comes in two separate vials that are mixed 20 min prior to injection. It forms a cast after coming in contact with the ionic material. It is better than glue because it has better control over the injection. Its only disadvantage is its higher cost compared to other agents.[9]

Results

Data from ten patients that satisfied the inclusion criteria were analyzed. The mean age of patients was 34.3yrs with range from 24-55 years and eight (80%) were males. There were five right sided CCFs, four left side CCFs and one bilateral CCF. All the patients of direct CCFs were preceded by head trauma. Seven of them had motor vehicle accident, two had accidental fall and one had sports injury. The time interval from injury to admission ranged from 20 days to six weeks (mean, 25.3 days). On ocular examination we found that seven patients had severe visual impairment, two patients had moderate visual impairment and only one patient was presented with mild visual impairment.

Ocular findings were evaluated- ten (100%) patients demonstrated the signs of chemosis and subconjunctival hemorrhage, eight (80%) presented with lid edema, and six (60%) had proptosis with bruit, raised intra ocular pressure and diplopia in five (50%) patients. The visual acuity was recorded with Snellen's chart- 4 patients had mild visual impairment, 5 patients had moderate visual impairment and only one patient presented with severe visual impairment. (Table 1)

Table 1: Tabulation of ocular finding details

Ocular Features	No of Patients presented with ocular finding
subconjunctival Hemorrhage chemosis	10
Lid edema	8
Pulsatile Proptosis with bruit	6
Increase IOP	5
Diplopia	5
Disc edema	2
Dilatation of retinal vessels only	6
Visual Acuity Mild Visual Impairment Moderate Visual Impaiment Severe visual Impairment	 4 5 1

On fundus examinations, disc edema was noted only in two (20%) patients and slight dilatation of retinal vessels in six (60%) patients Imaging evaluation of direct CCF by contrast-enhanced computed tomography (CT) and magnetic resonance angiography revealed a dilated superior ophthalmic vein in all patients. After noninvasive imaging, diagnostic DSA was performed in all cases, and endovascular embolization therapy was planned accordingly.

Five patients were treated with coil embolization of the fistula. In two patients, closure of the fistula was performed without occlusion of the parent artery (Figure 2). The

remaining three patients with large tears in the ICA and good collateral circulation from the contralateral ICA were treated with embolization of the fistula with trapping of the ICA (Figure 1). In the other five patients, embolization was performed using a combination of coils and Onyx. The patient with a bilateral CCF required two sittings of endovascular treatments, one for each side. The right-sided fistula closed first, followed by the left side at an interval of one month (Figure 3).

In repeat DSA at one month, nine out of ten patients showed complete closure of the fistulas. One patient required repeat embolization, which, on further follow-up after three months, showed complete obliteration. All patients demonstrated significant improvement in clinical symptoms with regression of chemosis, proptosis, and decrease or absence of bruit within 2–3 days after the procedure (Figure 4). Five patients showed good visual recovery, four patients had very good visual recovery and only one had fair visual recovery after embolization.

No signs of distal embolization, cerebral hemorrhage, cranial nerve palsy, or cerebral ischemia were observed in any of the patients. One of the patients complained of diminished

Figure 1: *Lateral DSA, preprocedure internal carotid artery (ICA) angiogram (image A) shows direct fistula (arrow) between cavernous sinus and ICA with enlarged draining Superior ophthalmic Vein. Post procedure angiogram (image B) after coiling of fistula with trapping of ICA (arrow) shows no flow in fistula*

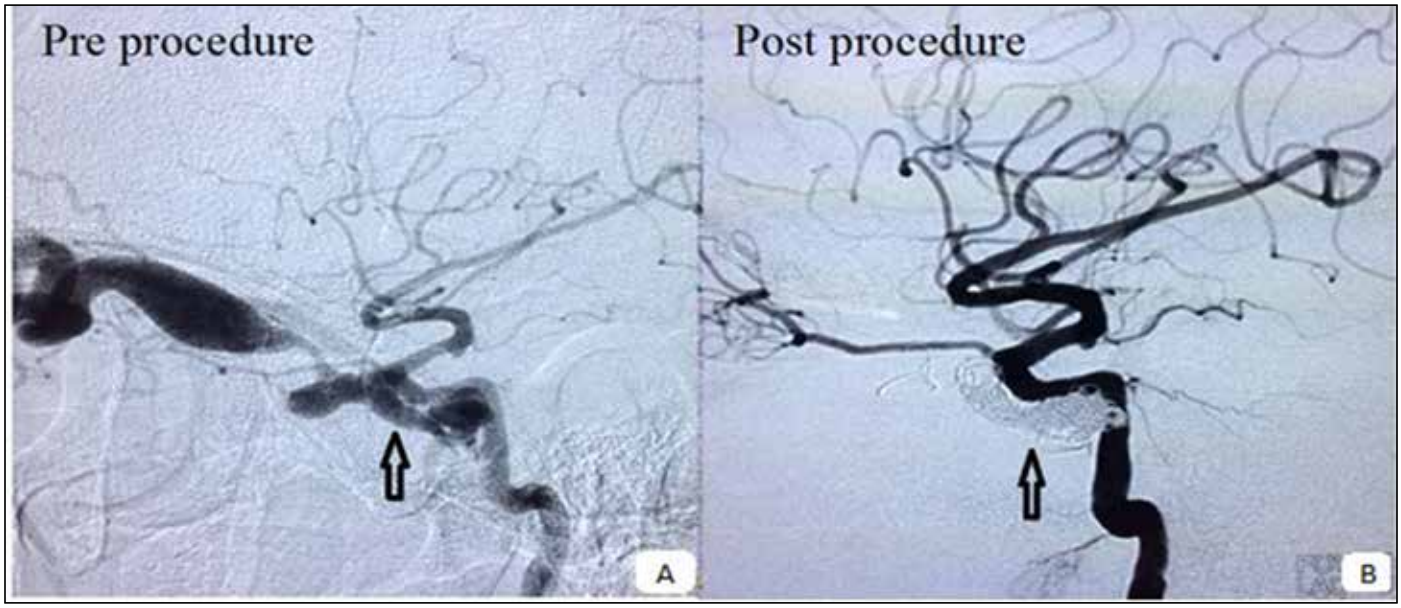

Figure 2: *Image A shows Direct CCF (arrow) with dilated superior ophthalmic vein (SOV) and post procedure (post-coiling) image (B) shows complete obliteration of fistula (arrow) with preservation of internal carotid artery (ICA)*

Figure 3: *Bilateral CCF, (A) coronal view, (B) lateral view, showing obliteration of right-side fistula with coiling, without trapping of internal carotid artery with left side dilated cavernous sinus*

Figure 4: *Pre-operative clinical picture (image A) and post-operative picture day 3 (image B)*

vision in the affected eye following the procedure but recovered spontaneously after 24 hours. None of the patients reported signs or symptoms of recurrence within six months of follow-up. Intraoperative or perioperative mortality was not observed.

Discussion

We found that timely management with endovascular techniques through DSA was associated with good visual recovery and resolution of all sign and symptoms of direct CCF. A direct CCF is a rare life-threatening condition and the most common cause of CCFs is trauma (76%).[10,11]

Other causes can be iatrogenic in origin, such as after transsphenoidal surgery for pituitary adenomas, treatment of trigeminal neuralgia, thrombectomy of the carotid artery, or presence of collagen disorders such as Ehlers-Danlos syndrome type IV.[12]

In the present study, all patients had experienced head trauma, similar to previous studies.[13,14] Cavernous sinus is prone to injury owing to its anatomical location, where the artery is completely surrounded by venous structures, and to the close proximity of the ICA to surrounding dura at skull base. Hence, it is immediately affected by shear force during trauma.[15]

Direct CCFs are commonly observed in men, as further confirmed by the current study.[15]

In present study, all cases of direct CCFs were abrupt in onset and progressed rapidly and presented with proptosis,

subconjunctival hemorrhage, chemosis, and diminished vision.[16,17] Transmission of high-pressure arterial blood into the cavernous sinus and draining veins led to venous hypertension, which is responsible for all the four mentioned signs and symptoms.[5]

Traumatic CCFs might be overlooked initially because of the critical condition of patients. Mild visual impairment is common in 60%–90% of patients with direct CCFs, but there could be progressive loss of vision or even total visual loss in approximately 25% of patients.[12] According to some studies, 89% of untreated cases could have visual impairment, but if closed completely, vision could improve in 94% of patients.[18] In present study, we also found visual acuity improvement in all ten patients but very good visual improvement was found in 50% of patients at the end of 1 month.

The current study demonstrated that noninvasive modalities such as contrast-enhanced CT and magnetic resonance angiography imaging can be considered in diagnosing CCFs, which typically show tortuous dilated superior ophthalmic veins (the hockey stick sign),[19] which can then be confirmed using the gold standard DSA.[12]

In the current study, all patients underwent active intervention, as majority of the subjects presented with moderate visual impairment. The first treatment for CCFs reported in 1809 was ligation of the common carotid artery and is still preferred in some selected cases.[5,20] Currently, endovascular therapy is considered the treatment of choice for all CCFs.

Endovascular treatment of CCFs in the 1970s was performed with detachable balloons, which were replaced by coils in the 1990s, followed by the era of liquid embolizing agents such as Onyx.[13,21,22]

In the current study, the transarterial approach was chosen for all patients. Five (50%) patients were embolized with a coil, and the rest were embolized by a combination of coil and Onyx. In our study, carotid artery preservation was performed in 70% of patients, whereas it was performed in 81% of patients in another similar study.[23] The 100% closure of fistula after treatment noted in this study was similar to that reported in other studies.[24,25,26]

The current study demonstrated adequate closure of Barrow type A fistula (direct CCF) without any adverse neurological complications, the only limitation being its small sample size.

Role of Ophthalmologist

Although all the interventions presented in present study were performed by interventional radiologists, but ophthalmologists definitely have an important role. The Ophthalmologists should be able to establish an initial presumptive diagnosis, as many patients with CCFs may initially present to ophthalmologists. They should be able to order appropriate diagnostic tests and be able to monitor patients. Timely referral to a neurosurgeon or neurointerventional radiologist is the key to successful closure of fistulas and could be performed by ophthalmologists.

Conclusion

Untreated direct CCF always results in poor visual prognosis, but can be managed promptly. With the observed results and low rate of complications and morbidity, endovascular therapy could be the primary treatment for patients with direct CCFs.

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