

First reported single-surgeon transpalpebral hybrid approach for indirect cavernous carotid fistula: illustrative case

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BACKGROUND Failure to reach the cavernous sinus after multiple transvenous attempts, although rare, can be challenging for neurointerventionists. The authors sought to demonstrate technical considerations and nuances of the independent performance of a novel hybrid surgical and endovascular transpalpebral approach through the superior ophthalmic vein (SOV) for direct coil embolization of an indirect carotid cavernous fistula (CCF), and they review salient literature regarding the transpalpebral approach.

OBSERVATIONS An illustrative case, including patient history and presentation, was reviewed. PubMed, MEDLINE, and Embase databases were searched for articles published between January 1, 2000, and September 30, 2021, that reported ≥ 1 patient with a CCF treated endovascularly via the SOV approach. Data extracted included sample size, treatment modality, surgical technique, performing surgeon specialty, and procedure outcome. The authors' case illustration demonstrates the technique for the hybrid transpalpebral approach. For the review, 273 unique articles were identified; 14 containing 74 treated patients fulfilled the inclusion criteria. Oculoplastic surgery was the most commonly involved specialty (5 of 14 studies), followed by ophthalmology (3 of 14). Coiling alone was the treatment of choice in 12 studies, with adjunctive use of Onyx (Medtronic) in 2.

LESSONS The authors' technical case description, video, illustrations, and review provide endovascular neurosurgeons with a systematic guide to conduct the procedure independently.

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KEYWORDS coil; endovascular embolization; indirect carotid cavernous fistula; superior ophthalmic vein; transpalpebral

Carotid cavernous fistulas (CCFs), defined as abnormal connections between the carotid arteries and the cavernous sinus, are classified as either direct or indirect, depending on the feeding branches.^{1,2} The classification can also be based on hemodynamics (high flow or low flow) or etiology (traumatic or spontaneous).³ Type A fistulas are direct communications between the cavernous sinus and the internal carotid artery (ICA), making them high-flow, direct CCFs. Types B, C, and D fistulas are low-flow, indirect pathological connections between the cavernous sinus and branches of the ICA, external carotid artery, or both, respectively.^{3,4} Nevertheless, all

fistulas result in high intraocular pressure (IOP) due to increased blood flow causing intraorbital venous congestion and symptoms that are pathognomonic for CCF, such as proptosis, chemosis, blurred vision, and diplopia.⁵⁻⁷

Transarterial embolization using detachable coils or liquid embolic agents, such as *N*-butyl cyanoacrylate (NBCA) or Onyx (Medtronic), serve as curative treatment for type A fistulas due to direct and easy arterial access.^{4,7-9} In contrast, indirect CCFs, although known to occasionally resolve spontaneously, are treated via transvenous embolization.^{7,10,11} Conventionally, transvenous access to

ABBREVIATIONS CCA = common carotid artery; CCF = carotid cavernous fistula; ICA = internal carotid artery; IOP = intraocular pressure; IPS = inferior petrosal sinus; NBCA = *N*-butyl cyanoacrylate; PRISMA = Preferred Reporting Items for Systematic Reviews and Meta Analyses; SOV = superior ophthalmic vein; SPS = superior petrosal sinus.

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the fistula is easiest and shortest via the inferior petrosal sinus (IPS).^{12,13} When access through the IPS is not possible due to thrombosis of the sinus or the IPS cannot be visualized angiographically, the superior petrosal sinus (SPS) can serve as an alternative.^{12,13} In rare cases, the facial vein can also be used if both aforementioned approaches fail.^{14,15}

Despite better-suited, flexible guidewires and microcatheters for transarterial and transvenous access, rare cases require alternative orbital pathways, such as access to the cavernous sinus through the superior ophthalmic vein (SOV) when these methods fail.^{7,16} This could be achieved through direct percutaneous puncture and cannulation of the SOV, which carries an inherently high risk for retro-orbital bleeding and hematoma formation, or a transpalpebral approach, whereby an incision is made in the upper eyelid and the SOV is accessed. Through a case description accompanied by illustrative images and a video, we aimed to demonstrate this approach in detail with technical considerations and nuances explained from an endovascular neurosurgeon's point of view. We also aimed to demonstrate that this approach can provide endovascular neurosurgeons with the information necessary to perform the exposure and treatment independently, particularly when ophthalmology or oculoplastic surgery services are unavailable. Moreover, we conducted a literature review to highlight the substantial involvement of these specialists in performing this neurointervention and the current shortage of data from an endovascular neurosurgeon's perspective.

Illustrative Case

Clinical Presentation

This patient presented with diplopia, right retro-orbital pain, and pulsatile tinnitus over several weeks. An ophthalmologist conducted a thorough examination, and the patient was found to have right-sided proptosis (Fig. 1A). The patient underwent brain/orbit magnetic resonance imaging, which revealed an enlarged right SOV suggestive of a right CCF (Fig. 1B and C). The patient was then referred to neurosurgery for further evaluation. Diagnostic cerebral angiography demonstrated an indirect type D CCF, which is typically amenable to a transvenous endovascular or a direct surgical approach.³ Neither the IPS nor the SPS could be catheterized, making a routine transvenous endovascular embolization approach impossible. We noted a large, dilated, and arterialized SOV that was located very superficially. An ophthalmologist was unavailable to aid with the open approach; therefore, we performed a modified hybrid approach that was believed to be within the realm of the neurosurgical

skill set. A transpalpebral approach was undertaken, providing direct transvenous access to the cavernous sinus and, therefore, the fistula. From here, direct coil embolization (Target soft coils, Stryker Neurovascular) could be performed. The hybrid transpalpebral-endovascular approach resulted in obliteration of the fistula with resolution of the patient's symptoms. Of note, we had discussed the options of direct percutaneous puncture of the SOV versus a transpalpebral approach with the patient; after all the risks were discussed (including retro-orbital hematoma), the patient elected to proceed with the transpalpebral approach.

Operative Technique

The patient is brought into the operating room and intubated using general endotracheal intubation. Before preparation of the orbit, tetracaine drops (0.5%) are administered to the target eye for a local anesthetic. The affected orbit is prepared and draped using 5% Betadine solution (Aviro Health L.P.), because the 10% solution is toxic to the cornea. (Dilution with sterile saline is necessary if performed with stock 10% Betadine solution.) The orbit is then draped using blue towels, followed by a thyroid drape (Fig. 2A and B, Video 1). A midline palpebral incision is marked, and ultrasound imaging is used to confirm the presence and location of the dilated SOV. A skin incision is made using a 10-blade scalpel followed by monopolar cautery through the subcutaneous tissue, until the orbicularis muscle is encountered. For the initial dissection through the orbicularis oculi muscle and septum, we used monopolar cautery (15/15 blend setting). For hemostasis in the orbit, we used bipolar cautery (18-W setting). From here, a mix of blunt and sharp dissection is performed through the orbicularis down to the orbital septum, using Westcott scissors and cotton swabs, respectively. Once the periorbital adipose tissue is encountered, the swabs are further used to mobilize the adipose tissue to expose the dilated and arterialized SOV, which will typically present itself with little mobilization (Fig. 2A and B). Once the vein is encountered, it is mobilized from the surrounding tissue, and two vessel loupes are placed around it on either side using a right-angle technique.

VIDEO 1. Clip showing the performance of a hybrid endovascular-transpalpebral approach in a stepwise fashion. University at Buffalo Neurosurgery, Inc., February 2022. With permission. [Click here to view.](#)

The patient is then transported to the angiography suite, and a 5-French right radial sheath is placed for endovascular access using a modified Seldinger technique. A Simmons-2 diagnostic catheter (Merit

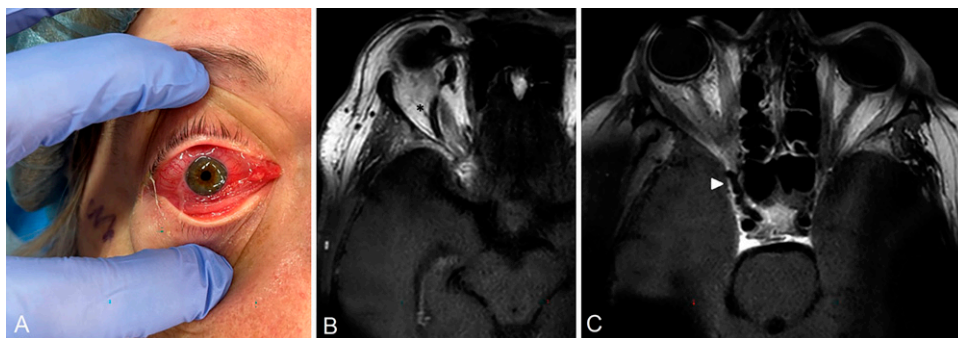


FIG. 1. Preoperative photograph (A) of the patient's right orbit showing proptosis and chemosis. Axial T2-weighted magnetic resonance images show an enlarged SOV (B, black asterisk) and CCF (C, arrowhead).

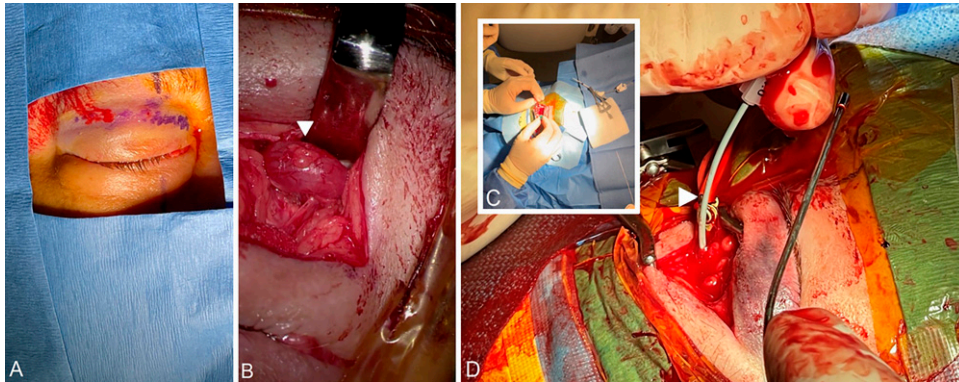


FIG. 2. Intraoperative photographs of the right orbit showing preparation and draping with palpebral incisional marking (A) and exposed SOV (B, arrowhead) from the underlying periorbital adipose tissue. Inset (C) shows direct micropuncture of the dilated SOV. Placement of the 5-French microdilator using a modified Seldinger technique (D, arrowhead).

Medical Systems) is then navigated to the common carotid artery (CCA) of the affected side while the patient is receiving a heparinized saline flush. For access into the SOV, the vein is punctured using a 21-gauge Merit Advance short radial micropuncture needle (Merit Medical Systems). A 0.014-inch Nitrex guidewire (Medtronic) is placed inside the radial access needle, and a modified Seldinger technique is used to subsequently place a 5-French microdilator sheath (Cook Medical). A CCA injection is performed for roadmap guidance during the venous phase. A microcatheter (Prowler Select LPES, Cerenovus) is then inserted over a Synchro-Select Support microwire (Stryker Neurovascular) to enter the SOV and approach the cavernous sinus in a retrograde fashion (Figs. 2C, 2D, 3A, and 3B). Once in the cavernous sinus, coils are placed at the level of the vein for complete occlusion

(Figs. 3A, 3B, and 4). We decided against using liquid embolic agents, such as Onyx, because their use would have resulted in potential loss of microcatheter access. A final run is performed demonstrating obliteration of the CCF. Back in the operating room, the incision was irrigated copiously using Ancef (SmithKline Beecham). Both ends of the SOV were ligated with 2-0 silk ties and then cauterized using bipolar cautery. The vessel loupes were removed. Once hemostasis was obtained and irrigation performed, a single-layer closure was performed using 6-0 Prolene (Ethicon) for skin. Bacitracin was applied to the incision for infection control, and the patient was discharged (Supplemental Fig. 1A and B).

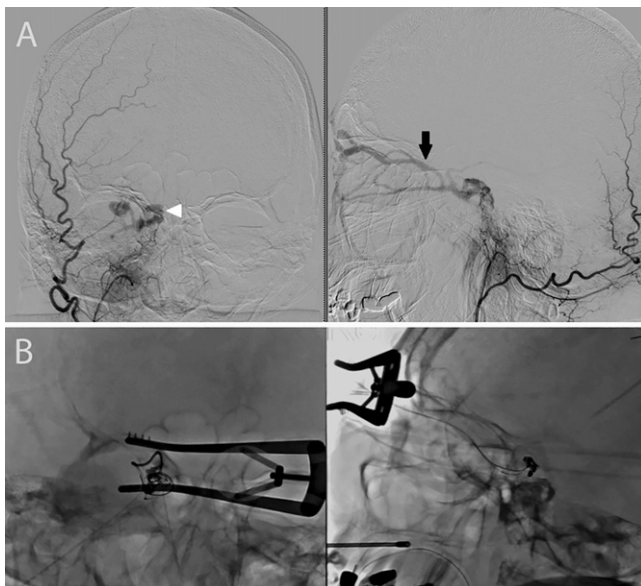


FIG. 3. Intraoperative diagnostic cerebral angiograms, right external carotid artery cranial injection (A; left, anteroposterior; right, lateral), showing a type D CCF (white arrowhead) with a dilated SOV (black arrow) and direct transvenous approach to the cavernous sinus via the SOV with coil embolization of the proximal SOV (B; left, anteroposterior; right, lateral).

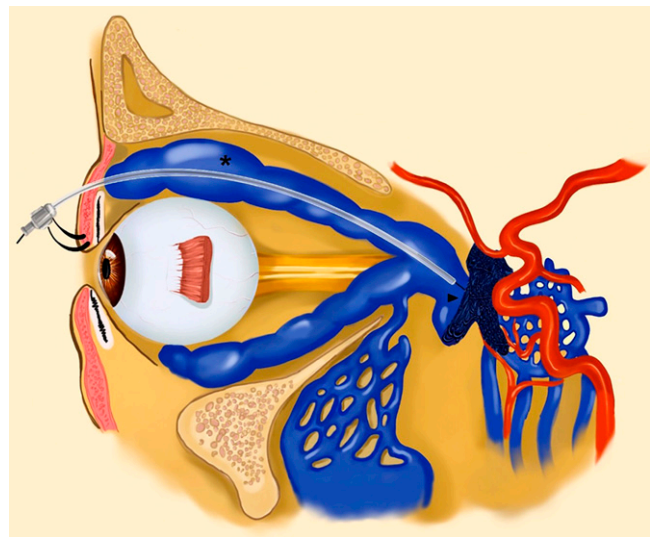


FIG. 4. Illustration created to demonstrate the anatomy of the SOV and the cavernous sinus, as well as the setup for the transpalpebral approach. A microcatheter is inserted over a microwire (black asterisk) to enter the SOV. Densely packed coils are seen in the fistula (black arrowhead). University at Buffalo Neurosurgery, Inc., February 2022. With permission.

TABLE 1. Studies included in the literature review reporting a transpalpebral approach to access the SOV

Authors & Year	Study Design	Disease Treated	Sample Size	Tx Modality	Performing Specialties	Outcome
Brenna et al., 2020 ¹⁸	Retro case report	Indirect CCF	1	Coiling	Ophthalmology & plastic surgery	Complete obliteration
Wolfe et al., 2010 ¹⁶	Retro case series	Indirect CCF	10	Coiling + Onyx	Not stated	Complete obliteration in 90% of cases
Daigle et al., 2021 ¹⁹	Retro case report	Indirect CCF	3	Coiling	Ophthalmology	Complete obliteration achieved in all 3 cases
Iglesias et al., 2021 ²²	Retro case report	Indirect CCF	1	Coiling	Oculoplastic surgery	Complete obliteration achieved
Baldauf et al., 2004 ¹⁷	Retro case report	Indirect CCF	2	Coiling	Not stated	Complete obliteration achieved
Klink et al., 2001 ²⁴	Retro case series	Indirect CCF	2	Coiling	Oculoplastic surgery	Complete obliteration
Park et al., 2021 ²⁷	Retro case series	Indirect CCF	2	Coiling	Oculoplastic surgery	Complete obliteration
Güven Yilmaz et al., 2013 ²⁰	Retro case series	Indirect CCF	4	Coiling	Ophthalmology	Complete obliteration
Sur et al., 2020 ²⁸	Retro case series	Direct & indirect CCF	8	Coiling	Not stated	Complete obliteration in 7 of 8 cases
Haider et al., 2017 ²¹	Retro case series	Indirect CCF	1	Coiling	Oculoplastic surgery	Complete obliteration
Lee et al., 2012 ²⁵	Retro case series	Indirect CCF	5	Coiling	Plastic surgery	Complete obliteration
Wajnberg et al., 2009 ²⁹	Retro case report	Indirect CCF	1	Coiling	Not stated	Complete obliteration
Leibovitch et al., 2006 ²⁶	Retro case series	Indirect CCF	25	Coiling	Oculoplastic Surgery	Complete obliteration in 19 of 25 cases
Jiang et al., 2011 ²³	Retro case series	Indirect CCF	9	Coiling & Onyx	Ophthalmology	Complete obliteration

Tx = treatment.

Literature Review

Search Strategy and Study Selection

A systematic search of the PubMed, MEDLINE, and Embase databases was conducted using the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines. The keywords of “transpalpebral,” “ophthalmic,” “carotid cavernous fistula,” “carotid-cavernous fistula,” and “superior ophthalmic vein” were used with Boolean operators to increase specificity and sensitivity. Two authors independently conducted screening and initial study selection for the review. Articles published in English between January 1, 2000, and September 30, 2021, that reported ≥ 1 patient with a CCF treated endovascularly with the SOV approach were included. Background articles and animal and cadaveric studies were excluded. Studies in which data specific to patients treated via the SOV approach could not be separately extracted were excluded. Full-text articles were assessed regarding study center and time period, and those with overlapping patient populations were excluded while retaining the most recent and/or complete study. Data extracted included sample size, treatment modality, surgical technique, performing surgeon’s specialty, and procedure outcome.

Results of the Literature Review

Our search resulted in 405 articles, of which 130 were duplicates. From the 273 unique articles, 253 were excluded for reasons listed in the PRISMA flowchart (Supplemental Fig. 2). The remaining 20 studies were assessed in full text, and 14 fulfilled the inclusion criteria. These 14 articles were included in our final analysis (Table 1).^{16–29}

Our review revealed oculoplastic surgery as the most commonly involved specialty during the procedure (5 of 14 studies; 35.7%).

Most commonly, the role of oculoplastic surgery was to perform the initial subbrow incision with dissection of the periorbital fat and exposure of the SOV. Assistance from ophthalmologists was sought in 3 studies (21.4%) and plastic surgery in 2 (14.3%). A total of 74 patients were treated in these studies with a median of 2.5 cases. Coiling alone was the treatment of choice in most studies (12 of 14; 85.7%), with adjunctive use of Onyx in the remaining 2 studies (14.3%) (Supplemental Table 1).

Discussion

Observations

In this study, we demonstrate the hybrid transpalpebral-endovascular approach for direct coil embolization through the SOV for treating indirect CCFs. We provide the technique description, video, and illustration as a systematic guide for performing the procedure. In addition, our review identified the prevalence of various specialties (oculoplastic surgery 35.7%), followed by ophthalmology (21.4%) and plastic surgery (14.3%). Coiling alone was the treatment of choice in 12 studies (85.7%), with adjunctive use of Onyx in 2 (14.3%).

Endovascular Embolization

Management of CCFs often mandates close coordination between ophthalmology, neurosurgery, and interventional neuroradiology.^{16–29} Endovascular intervention using transvenous embolization technique with detachable coils or liquid embolic agents has largely been accepted as the first-line treatment modality that has been proved to be safe and effective with high rates of complete obliteration and a concomitantly low complication rate.^{7,10–13} With more operator

experience and advances in neurointervention device technology, rates of complete obliteration have increased significantly, ranging from 70% to 90%, with the rate of complications reported as low as 3.6% of cases.^{30,31} In a multicenter study by Alexander et al.,³¹ long-term outcomes after endovascular treatment of indirect CCFs were investigated and an obliteration rate of nearly 90% was reported in patients who underwent embolization exclusively through the transvenous route. Permanent symptomatic complications were reported in only 0.5% of cases.³¹ Similarly, in a systematic review recently published by Texakalidis et al.,³² a direct comparison between transvenous and transarterial approaches for both direct and indirect CCFs was conducted. Fifty-seven studies comprising 1,575 patients were identified, of which 31 reported embolization through the transarterial route, and the remaining 26 reported outcomes after a transvenous embolization approach.³² Pooled data revealed complete obliteration in 425 (86%) of 494 cases of indirect CCFs treated with transvenous embolization technique.³² Importantly, embolization using a liquid embolic agent (NBCA, Onyx, glue, or polyvinyl alcohol) resulted in a slightly lower complete obliteration rate than transvenous coil embolization (83.3% vs. 87.5%).³² Interestingly, a subgroup of patients with a direct CCF underwent embolization transvenously, and an obliteration rate of 91.7% (33 of 36) was achieved.³² In addition, pooled obliteration rates were similar among the 5 studies directly comparing transvenous versus transarterial embolizations for indirect fistulas (odds ratio 0.62, 95% confidence interval 0.31–1.23, $I^2 = 0.0\%$).³² These results and several others have demonstrated the clear benefit and established efficacy of the transvenous approach for indirect fistulas.^{30,31} These studies also point to the expansion of indications for transvenous embolization to include direct fistulas that cannot be accessed transarterially.

Superior Ophthalmic Vein Route

Although a transvenous approach is generally successful for indirect fistulas, traditional access routes may not be amenable to endovascular intervention in some cases. The simplest and shortest venous route to the cavernous sinus via the IPS may not be angiographically visible due to thrombosis or severe vessel tortuosity.^{13,33} In those cases in which a transvenous route cannot be successfully cannulated for access to the cavernous sinus, various orbital approaches can serve as salvage therapies.^{7,17–19,22,34–43} Most of these last-resort therapies involve cannulating the SOV either percutaneously or through a surgical transpalpebral approach, as we illustrated above. In our systematic review, we identified studies that reported embolization via the SOV through the transpalpebral approach.^{17–19,22,34–43} More important, we identified the operating surgeons in each study to highlight the importance of explaining this technique from an endovascular neurosurgeon's point of view.^{17–19,22,34–43} Our systematic review revealed that among the 8 studies reporting the specialist involved in the transpalpebral approach, all except 1 reported performing the procedure with the help of an ophthalmologist or oculoplastic surgeon.^{18,19,22,35,38,39,41} This highlights the critical importance of explaining the technical details of the transpalpebral approach from an endovascular neurosurgeon's perspective so that proceduralists can use this as a guide and perform these procedures independently in cases where an ophthalmologist or oculoplastic surgeon is not available.

Alternate Orbital Approaches

In a systematic review conducted by Phan et al.,⁷ studies reporting various orbital approaches for the treatment of CCFs were reviewed in terms of embolization techniques used and outcomes reported. Thirty studies were included with a total of 140 patients who underwent treatment via an orbital approach. Transvenous embolization via the SOV was the primary orbital approach in 69 patients (49.3%).⁷ Direct percutaneous puncture of the SOV was performed in 15 cases (10.7%). Pooled analysis of patient data revealed an overall obliteration rate of 89.9%, with visual symptom resolution in 93.4% of cases. Notably, of the 30 studies included in their review, 17 that reported using a transvenous embolization technique via the SOV lacked homogeneity in terms of patient selection (previously treated fistulas), venous route selected (facial vein, intracanal approach), and embolic material used (platinum coils, Guglielmi detachable coils, Stryker Neurovascular; or Onyx).⁷ Moreover, endovascular technical details, such as catheter systems employed, use of microguidewires, and operative nuances, were not explained.⁷ This may be partly because most of these studies reported individual cases with limited descriptions and no video supplements to aid readers in understanding the technique in a stepwise fashion.

Other, less commonly used orbital approaches include direct percutaneous puncture of the SOV, access through the inferior ophthalmic vein, transorbital puncture into the SOV via the extraconal part, and direct puncture of the superior ophthalmic fissure into the cavernous sinus.^{17,34,36,37,40,42} Although direct percutaneous puncture has the advantage of not requiring open surgery, better cosmetic results, and reduced risk of infection, it does carry an inherent high risk of vein perforation and hematoma formation. In a study by Teng et al.,⁴⁴ patients who underwent direct SOV puncture developed subconjunctival hemorrhages, with 1 patient experiencing an eyelid hematoma. Better results can be expected through the direct approach if the SOV is sufficiently dilated and clearly identified on the angiogram.

Lessons

We demonstrate the transpalpebral approach for direct coil embolization through the SOV for treatment of an indirect CCF to provide endovascular neurosurgeons with information necessary to perform the exposure and treatment independently, particularly when an ophthalmologist is unavailable. Our technique description, video, and illustrations provide a systematic guide for independent performance of the procedure.

Our study has limitations, foremost of which is the retrospective and observational nature of the single case description. The systematic review also includes retrospective case reports that evaluated very small populations and may be affected by publication bias. However, we highlight some key points regarding the case description and a systematic guide for performing the procedure.

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Disclosures

Dr. Cappuzzo is a consultant for Cerenovus, Johnson & Johnson Medical Device Companies, Integra Lifesciences Corp., MIVI Neuroscience Inc., Penumbra Inc., and Stryker Neurovascular Corp. Dr. Levy is a consultant for Clarion, GLG Consulting, Guidepoint Global, Imperative Care, Medtronic, StimMed, Misionix, Mosiac, and IRRAS AB; has received payment or honoraria from Medtronic, Penumbra, Microvention, and Integra for lectures, presentations, speakers bureaus, manuscript writing, or educational events; has been reimbursed for travel and food for some meetings with the CNS and ABNS; is the national principal investigator for studies sponsored by

Medtronic; is the site principal investigator for studies sponsored by Microvention and Medtronic; holds a patent with Bone Scalpel; is the chief medical officer for Haniva Technology; serves on advisory boards for Stryker, NeXtGen Biologics, MEDX, Cognition Medical, and IRRAS AB; holds a leadership or fiduciary role with CNS, ABNS, and UBNS; and owns stock in NeXtGen Biologics, RAPID Medical, Claret Medical, Cognition Medical, Imperative Care, Rebound Therapeutics, StimMed, and Three Rivers Medical.

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Conception and design: Levy, Cappuzzo, Baig, Waqas. Acquisition of data: Levy, Cappuzzo, Baig, Metcalf-Doetsch, Waqas. Analysis and interpretation of data: Levy, Cappuzzo, Baig, Waqas. Drafting the article: Cappuzzo, Baig. Critically revising the article: Levy, Cappuzzo, Baig, Monteiro. Reviewed submitted version of manuscript: Levy, Cappuzzo, Metcalf-Doetsch. Approved the final version of the manuscript on behalf of all authors: Levy. Statistical analysis: Cappuzzo, Baig. Administrative/technical/material support: Cappuzzo, Monteiro. Study supervision: Monteiro.

Supplemental Information

Video

Video 1. <https://vimeo.com/690961855>.

Online-Only Content

Supplemental material is available with the online version of the article. *Supplemental Figures and Table.* <https://thejns.org/doi/suppl/10.3171/CASE22115>.

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