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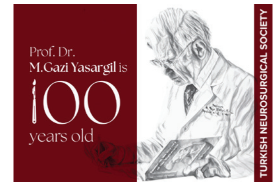

Endovascular Occlusion of Intracranial Pial Arteriovenous Fistula: Technical Aspects

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ABSTRACT

AIM: To evaluate pial arteriovenous fistulas (AVFs), focusing on the radio-anatomic architecture and contemporary endovascular devices and techniques.

MATERIAL and METHODS: Sixteen patients with congenital pial AVFs who underwent endovascular treatment between 2002 and 2023 at a single institution were included in this review. This retrospective study was approved by the Institutional Review Board. The study was descriptive and involved no statistical comparisons.

RESULTS: The study included 16 patients (6 female patients, 10 male patients) with a mean age of 19.93 ± 21.1 years (range: 1–63 years). Nine (56.25%) were pediatric patients, six (37.5%) of whom were younger than 5 years. Five patients (31.25%) had more than one feeding artery, whereas 11 (68.75%) had a single feeding artery. One patient had two separate fistulas. All fistulas were successfully occluded without complications. Four patients (25%) were treated with glue alone, four (25%) with coils alone, five (31.25%) with a non-adhesive liquid agent alone, and three (18.75%) with a combination of coils and a non-adhesive liquid agent. Venous sinus thrombosis occurred in two patients (12.5%) in the early postoperative period; both cases resolved without permanent deficits.

CONCLUSION: Pial AVF is a rare intracranial vascular malformation. Endovascular treatment using liquid embolic agents, coils, or a combination of these techniques is effective.

KEYWORDS: Glue, Coils, Non-adhesive liquid agent, Arteriovenous fistula, Pial AVF

ABBREVIATIONS: AVF: Arteriovenous fistula, **CT:** Computed tomography, **MR:** Magnetic resonance, **AVM:** Arteriovenous malformation, **SAH:** Subarachnoid hemorrhage, **EVOH:** Ethylene-vinyl alcohol, **DMSO:** Dimethyl sulfoxide

INTRODUCTION

A pial arteriovenous fistula (AVF) is a rare vascular disorder, accounting for approximately 1.6% of all intracranial vascular malformations. Pial AVFs differ from both pial and dural arteriovenous malformations (AVMs); the latter are characterized by vascular nidi (tangled vascular networks) in the parenchyma and meningeal layers, respectively. Although pial AVFs share some radiological features with pial AVMs, they are now recognized as distinct vascular anomalies (8). The defining characteristics of a pial AVF include pial arterial feeders, a direct arteriovenous fistula without an intervening nidus, and an aneurysmal varix with lobulated, serpiginous venous drainage.

We reviewed a series of patients with pial AVFs, focusing on the radio-anatomic architectures and the utility of contemporary endovascular devices and techniques.

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MATERIAL and METHODS

This retrospective study was approved by Ege University Faculty of Medicine, Institutional Review Board (protocol number 24-6.1T/37). The Board waived the requirement for informed patient consent due to the retrospective and observational nature of the study. All patient details were anonymized during data collection and analysis to ensure confidentiality.

Between 2002 and 2023, 16 patients with pial AVFs were treated using endovascular methods. Patient demographics, clinical presentations, endovascular management strategies, follow-up data, and outcomes were recorded. All patients underwent computed tomography (CT)/CT angiography and magnetic resonance (MR) imaging/MR angiography, which revealed the characteristics and anatomy of the vascular lesions. MR angiography was generally preferred for follow-up. Clinical outcomes were defined as "excellent" when neurological findings returned to normal and as "good" when no additional neurological deficit was observed postoperatively. Modified Rankin scores from the most recent neurological examinations were also recorded.

No statistical comparisons were performed in this descriptive study. Continuous variables are presented as medians with standard deviations (SDs) and ranges; categorical variables are presented as frequencies with percentages.

RESULTS

There were 10 male (62.5%) and 6 female patients (37.5%) of mean age 19.93 ± 21.1 years and median age 8.5 years (range 1 to 63 years). Nine patients (56.25%) were pediatric, six (37.5%) of whom were younger than 5 years. Patient data are summarized in Table I. Six patients (37.5%) presented with acute ictus, and imaging suggested pial AVF hemorrhages. Five patients (31.25%) had more than one feeding artery, whereas the remainder had single feeding arteries. One patient (case #11) had two distinct fistulas with separate drainage routes; all others had single fistulas and drainage routes. Nine patients (56.25%) exhibited aneurysmal venous varices associated with the pial AVFs; five (31.25%) of these patients were pediatric. Thrombosed varicose aneurysms were observed in three of these nine patients (18.75%).

All embolization procedures were successful, and no complications occurred. Four fistulas (25%) were occluded using glue alone, four (25%) with coils alone, five (31.25%) with a non-adhesive liquid agent alone, and the remaining three (18.75%) with combinations of coils and the non-adhesive liquid agent. Simple embolization (either coils or a liquid agent) was used in 13 patients (81.25%). The double microcatheter technique was utilized in two patients (12.5%) (Figures 1-4), and the flow arrest technique with a double microcatheter was used in another two patients (12.5%) (Figures 5 and 6).

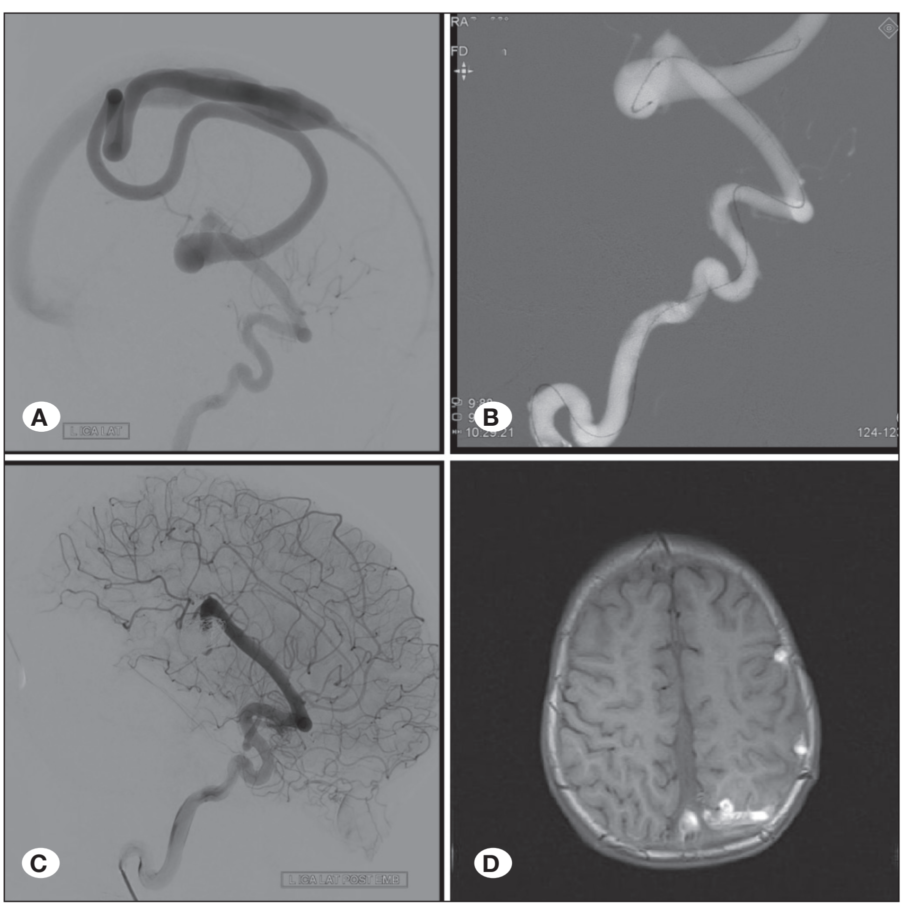

Figure 1: A 1-year-old male patient presented with macrocrania and hydrocephalus. **A)** Left internal carotid angiography lateral projection. There is a single-hole high-flow fistula fed by the M2 branch of left middle cerebral artery which drains to a superficial serpiginous dilated vein, then superior sagittal sinus. **B)** Although the proximal venous part of the fistula site has relatively dilated, deploying coils without migration would be difficult. This anatomy is ideal for using the double-microcatheter technique. **C)** Control angiography shows total occlusion with coils only. **D)** MR imaging 15 days later demonstrates subacute thrombosis in the fistula's serpiginous vein which also extends up to the superior sagittal sinus.

Table I: Summary of 16 Patients with Pial AVF

Case No/ Sex/Age (year)	Presentation	Bleeding	Feeding Artery	Draining Sinus	Venous Varix	Embolization Material / Technique	Follow-up	Clinical Outcome (mRS)
1/M/1	Macrocrania, Hydrocephalus		R AICA R PICA R SCA	Occipital	Yes (thrombosed)	Glue /Simple	6 months	Good (0)
2/M/55	Headache		L PCA L MCA	Superior sagittal	Yes	Glue /Simple	6 months	Excellent (0)
3/F/35	Headache		L MCA L PCA	Transverse	Yes	Glue /Simple	2 years	Excellent (0)
4/M/24	Syncope	Hematoma	R PCA	Transverse	No	Glue /Simple	6 months	Excellent (0)
5/F/2	Incidental (PHACE Syndrome)		R ACA	Transverse	No	Coil/ Simple	2 years	Excellent (0)
6/M/28	Headache		R MCA	Superior sagittal	Yes (thrombosed)	Coil /Simple	15 years	Excellent (0)
7/F/1	Incidental (PHACE syndrome)		L PCA	Rectus	Yes	Coil/ Simple	6 years	Excellent (0)
8/M/1	Macrocrania, Hydrocephalus		L MCA	Superior sagittal	Yes	Coil /Double catheter	6 months	Good (1)
9/M/32	Syncope	Hematoma	L PCA	Transverse	No	Onyx /Simple	2 years	Excellent (0)
10/M/5	Syncope, Paresia	Hematoma	R MCA	Superior sagittal	No	Onyx /Simple	2 years	Good (1)
11/F/1	Seizure	SAH	R PCA R ACA	Superior sagittal	Yes (thrombosed)	Onyx /Simple	4 years	Good (1)
12/M/63	Incidental		R MCA	Superior sagittal	No	Onyx /Simple	2 years	Excellent (0)
13/M/9	Headache	SAH	R ACA	Rectus	No	Onyx /Simple	6 months	Excellent (0)
14/M/8	Headache		R PCA	Rectus	No	Coil + Onyx/ Double-catheter	4 years	Excellent (0)
15/F/47	Headache Seizure		L ACA	Superior sagittal	Yes	Coil + Onyx/ Double-catheter flow arrest	2 years	Excellent (0)
16/F/7	Syncope	SAH	R, L PICA	Marginal	Yes	Coil + Onyx/ Double-catheter flow arrest	3 months	Excellent (0)

mRS: *Modified Rankin Scale,* ***M:*** *Male,* ***F:*** *Female,* ***R:*** *Right,* ***L:*** *Left.* ***AICA:*** *Anterior Inferior Cerebellar Artery,* ***PICA:*** *Posterior Inferior Cerebellar Artery,* ***SCA:*** *Superior Cerebellar Artery,* ***MCA:*** *Middle Cerebral Artery,* ***PCA:*** *Posterior Cerebral Artery,* ***ACA:*** *Anterior Cerebral Artery,* ***SAH:*** *Subarachnoid Hemorrhage.*

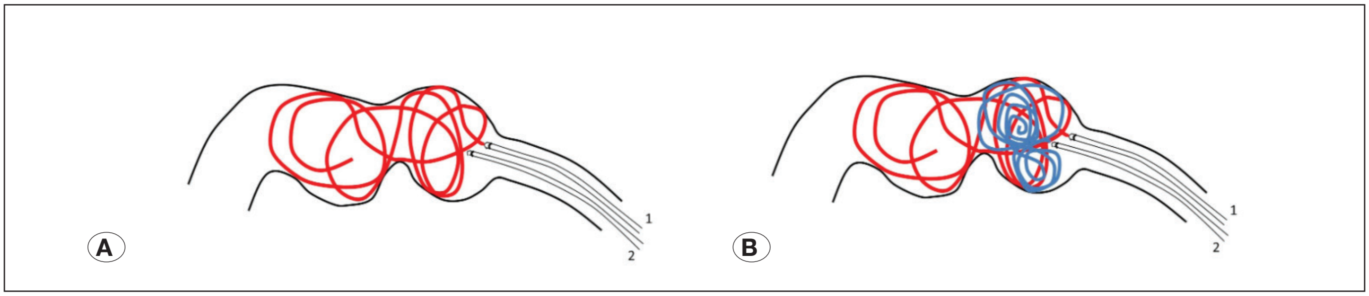

Figure 2: Schematic representation of coiling with the double-microcatheter technique. **A)** Two microcatheters are placed in the fistula, extending to the very proximal part of the venous route. The oversized first coil was deployed through the first microcatheter. **B)** Subsequent smaller coils were deployed in the nest of first coil through the second microcatheter, while the first coil remains undetached.

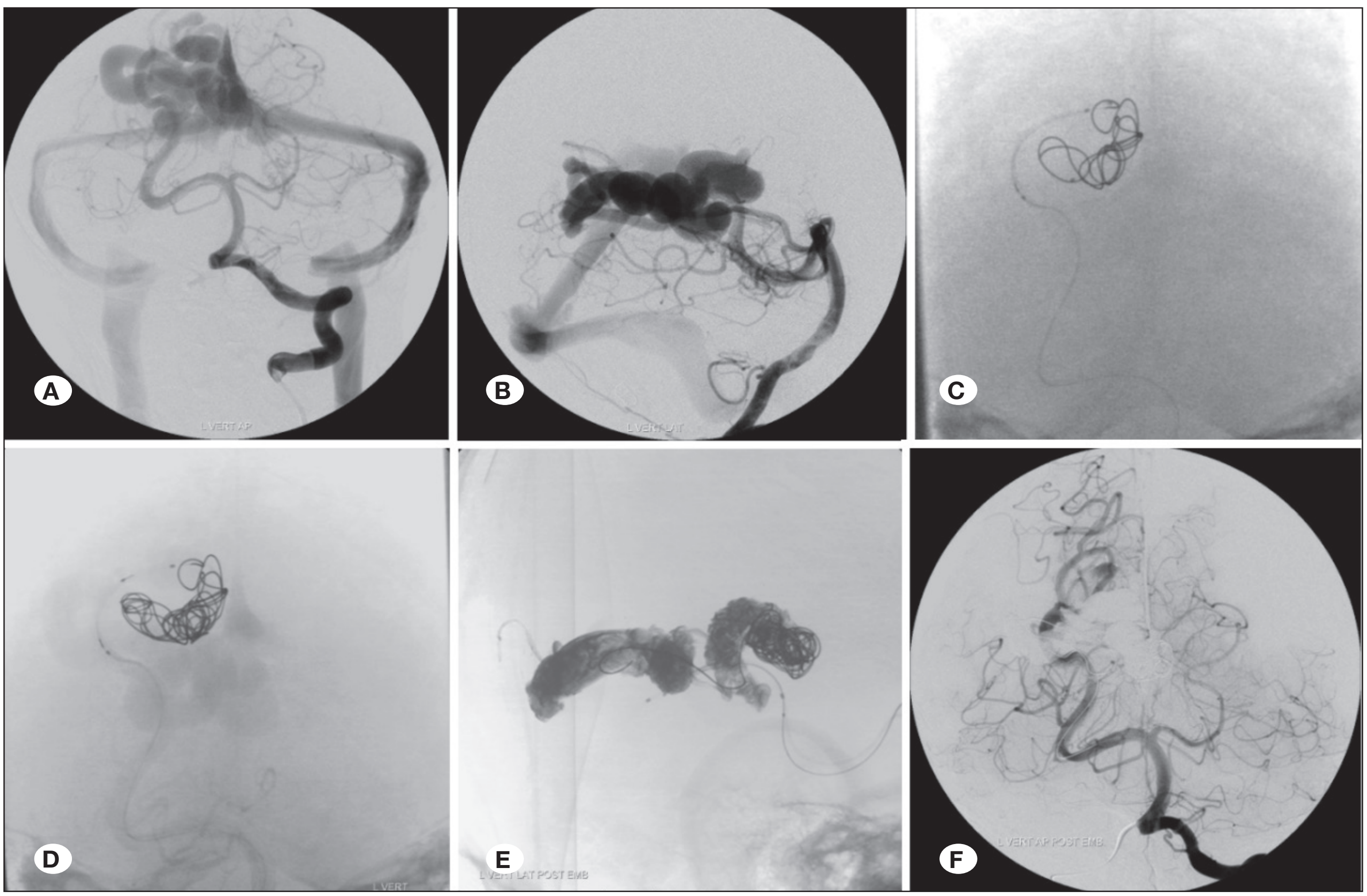

Figure 3: An 8-year-old male patient presented with headaches. Left vertebral artery angiography, frontal **(A)** and lateral **(B)** projection. There is a single hole high-flow fistula fed by the right posterior cerebral artery which drains to superficial serpiginous dilated vein and then ipsilateral Rosenthal vein, and sinus rectus. **C)** First coil is deployed through the first microcatheter and kept undetached. **D)** The second coil is deployed in the nest of the first coil through the second microcatheter. This is an example of the double-microcatheter technique. **E)** Lateral image. After blood flow is markedly reduced by coil packing, liquid is injected in front of the coil pack to achieve total occlusion of the fistula. Note the unintentional migration of coil pack in the distal venous segment. Also, note that coil nest successfully holds the proximal liquid cast without fragmentation. **F)** Control angiography frontal projection reveals total occlusion of the fistula.

In the early postoperative period, lethargy and drowsiness occurred in a 1-year-old patient (case #8), and an adult (case #15) experienced worsening headaches. MR imaging revealed the development of venous and/or dural sinus thrombosis in both cases, and both patients responded well to appropriate medical treatment. MR venography performed 6 months later showed complete recanalization of the thrombosed sinuses in case #8. Endovascular treatment and shunt surgery for hydrocephalus were performed simultaneously in two pediatric patients (cases #1 and #8). In a third patient (case #11), hydrocephalus developed later, attributed to a subarachnoid hemorrhage, and a shunt was placed after endovascular treatment. Aside from early thrombotic complications in two patients, the remaining 14 (87.5%) experienced no worsening of their clinical conditions. Symptoms improved over time; all hemorrhages were reabsorbed, and hydrocephalus resolved after shunt placement.

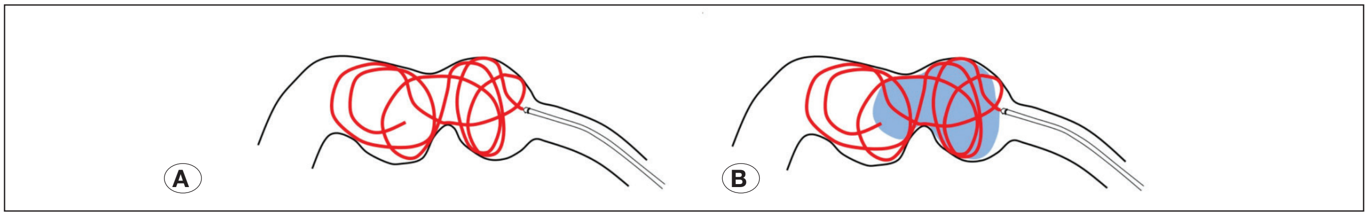

Figure 4: Schematic representation of the combined usage of coils and non-adhesive liquid embolization agent. **A)** Appropriate coil packing is initially achieved. **B)** To obtain stable and absolute occlusion, the spaces within the coil nest are filled with a liquid embolization agent using the same microcatheter.

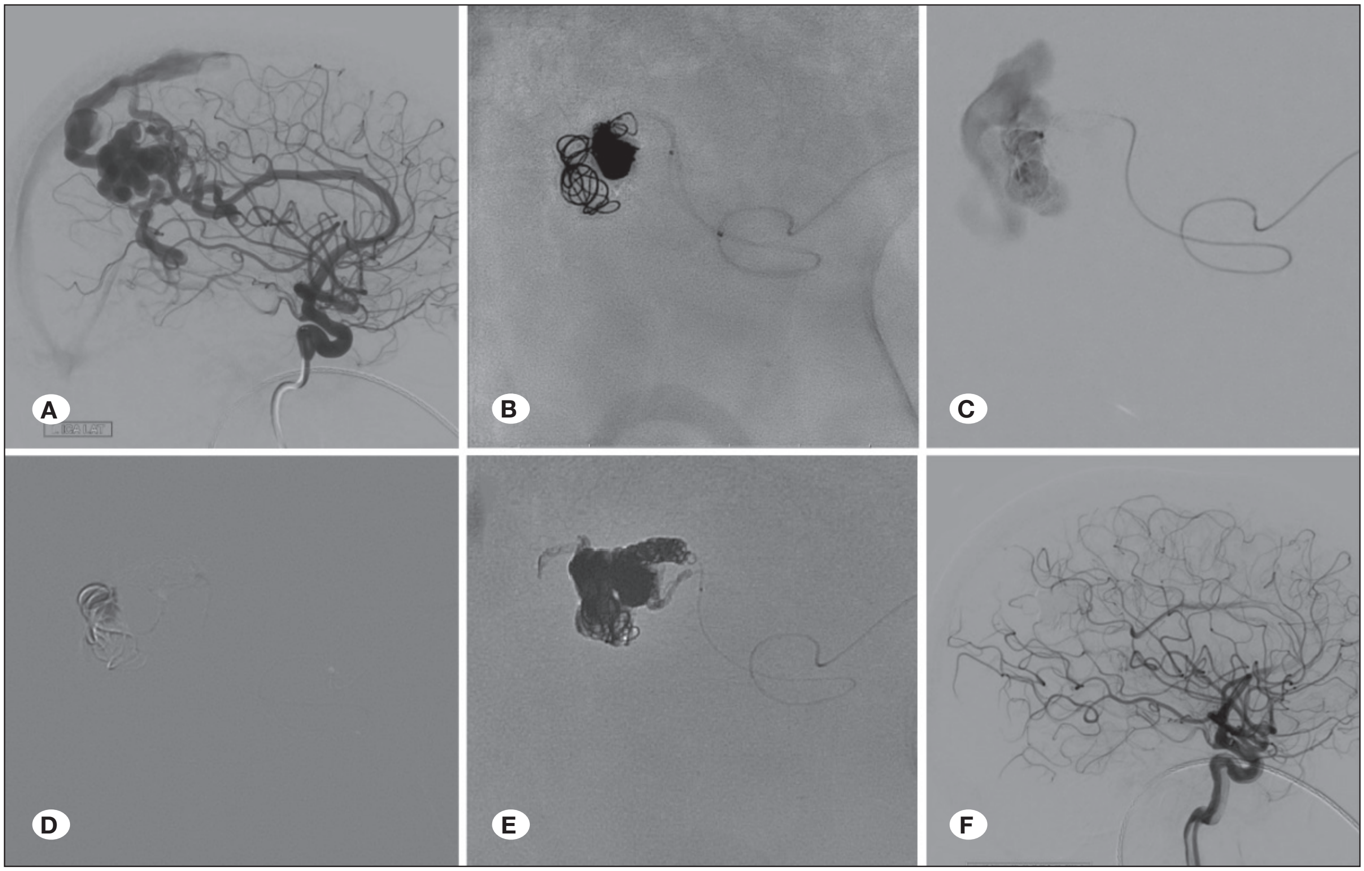

Figure 5: An 8-year-old male patient presented with headaches. **A)** Left internal carotid angiography lateral projection. There is a single-hole high-flow fistula fed by the left anterior cerebral artery, which drains to a superficial serpiginous dilated vein with multiple varicose aneurysms and then sinus rectus and superior sagittal sinus. **B)** Double-microcatheter flow-arrest technique. While one microcatheter is placed at the fistula point, the second microcatheter is placed 1-2 cm distal. Coils are deployed through the first microcatheter. **C)** Contrast injection through the second microcatheter reveals markedly diminished but still flowing blood. **D)** Slow non-adhesive liquid embolization injection through the second microcatheter on the roadmap image. **E)** Final embolic cast ensuring firm occlusion, consisting of coils and liquid. **F)** Final angiography confirming total occlusion of the fistula.

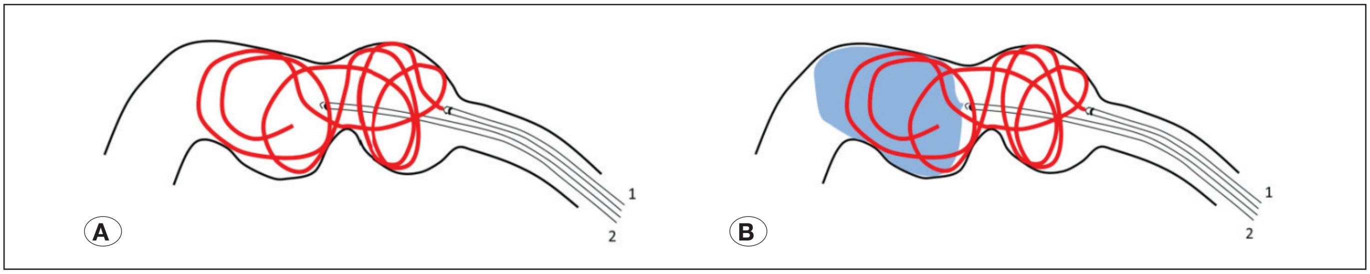

Figure 6: Schematic representation of double-microcatheter flow-arrest technique, combining the usage of coils and non-adhesive liquid embolization agent. **A)** Appropriate coil packing is achieved through the first microcatheter. The coil package diminishes markedly dragging force of blood flow. **B)** To obtain stable and

DISCUSSION

This study demonstrated that endovascular treatment is effective for pial AVF; all 16 patients underwent successful procedures without immediate complications. Aneurysmal venous varices were present in 56.25% of patients, particularly in the pediatric group. Multiple arterial feeders were observed in 31.25% of patients, and one patient had two distinct fistulas. Simple embolization was sufficient in most cases; complex techniques, such as the double microcatheter approach, rarely were required. Postoperative thrombotic complications occurred in two patients but resolved with medical management. Favorable clinical outcomes were achieved in 87.5% of cases.

Pial AVFs are generally characterized by single-hole arteriovenous connections with single feeding arteries and ectatic, serpiginous venous drainage; no nidus is present. A venous aneurysmal varix is another distinctive feature, especially in patients with high-flow pial AVFs. Low-flow pial AVFs may lack this varix, which can lead to confusion with pial AVMs. The typical subpial location, near the cortex, helps to differentiate pial AVFs from pial AVMs. Multiple feeding arteries with several venous routes (i.e., multiple fistulas) and multiple feeding arteries with single venous routes are rare in pial AVFs.

As of September 2024, 231 pediatric cases of pial AVF have been reported in the literature (16). Most pial AVF patients are diagnosed in childhood, typically before the age of 5 years (10,13,16,18). In these patients, the primary presenting symptoms include hydrocephalus, developmental delay, enlarged head circumference, cardiac failure, and seizures. Coubes et al. reported a potential association between pial AVF and Rendu-Osler-Weber disease, a rare autosomal-dominant disorder (3). In older children and adults, headaches, seizures, and neurological deficits are more common, often due to mass effects from venous varices and hemorrhages (9,10,13). Hydrocephalus was observed in three of our pediatric patients, all of whom required shunt placement. However, hydrocephalus may improve after endovascular treatment alone (11). Spontaneous thrombosis of a pial AVF has also been reported; three of our 16 patients displayed acute thrombosis of venous aneurysmal varices. Some patients may present with intracranial bleeding; three of our patients exhibited intraparenchymal hemorrhages, and two presented with subarachnoid hemorrhages at diagnosis.

The endovascular treatment strategy for pial AVF is similar to that for other intracranial fistulas: the fistula should be occluded, extending to the proximal drainage vein when possible. Adhesive agents (cyanoacrylates), non-adhesive liquids (12), and metallic endovascular implants (coils, stents) are used to achieve this occlusion.

Cyanoacrylate monomers undergo exothermic polymerization upon contact with blood, ensuring rapid adhesion and immediate occlusion of the injected vessel. Various intravascular acrylic agents are available, differing in both molecular structure and specific additives. Common N-butyl cyanoacrylate-based agents include Histoacryl (B. Braun, Melsungen, Germany), Glubran 2 (GEM SRL, Viareggio, Italy), and TruFill (Cerenovus, Irvine, CA, USA). Magic Glue (Balt, Montmorency, France) is an N-hexyl cyanoacrylate-based agent, whereas Fuaile (Beijing, China) is a combination of N-butyl and 2-octyl cyanoacrylates. Cyanoacrylates are typically mixed with Lipiodol (Guerbet, Villepinte, France) to slow polymerization and adhesion in a concentration-dependent manner (17).

Neurointerventional non-adhesive liquid embolization agents include Onyx (Medtronic, Irvine, CA, USA), Squid (Balt, Montmorency, France), and PHIL (MicroVention, Tustin, CA, USA) (17). These agents are permanent, non-absorbable, and non-adhesive. Onyx, one of the most widely used liquids, is an elastic polymer (ethylene-vinyl alcohol [EVOH]) dissolved in dimethyl sulfoxide (DMSO); micronized tantalum powder provides radiopacity. Squid is also based on EVOH/DMSO; its low-viscosity variant enhances distal penetration. PHIL contains an iodinated material bound to the copolymer, ensuring radiopacity and preventing CT artifacts caused by tantalum (17).

Since Guglielmi et al. first used detachable coils in 1991 (5), many manufacturers have developed coils with various distinct features; notably, the basic operating principle remains unchanged. Coils are both easy to use and effective for endovascular occlusion; they have numerous neurovascular applications. Considering the extensive literature, the specific types and features of detachable coils will not be discussed in detail here.

Despite advances in endovascular devices and techniques, neurointerventionalists still face two key challenges. First, it can be difficult to ensure that occlusive implants remain at a fistula site, given the relatively dilated and serpiginous vascular bed associated with high-flow blood. High-concentration glue (Lipiodol: cyanoacrylate ratio of 1:2 or a cyanoacrylate concentration above 65% v/v) has been successfully used to treat high-flow fistulas with techniques similar to those utilized for Galenic fistulas (10,18). In the present study, four patients were successfully treated with high-concentration glue alone. However, the use of glue has decreased with the development of alternative embolization agents and techniques; fewer operators today have sufficient experience with this method. Detachable coils offer more controlled embolization relative to that achieved using glue. Four of our patients were treated with coils alone. The enlarged venous side of a pial AVF contains vascular structures folded over each other, often exhibiting sharp bends and/or venous varices. An experienced neurointerventionalist can anchor detachable coils within these anatomic structures, ultimately achieving total occlusion of the fistula.

Non-adhesive liquid embolization agents have been in use for approximately 15 years. Compared with glue, they are easier to handle and less thrombogenic (17). However, they are soft, pliable, and persistently remain in fluid form, making them unsuitable for treating AVFs, except in cases of low-flow AVFs.

Using the standard endovascular techniques described above, most pial AVFs can be treated successfully. In more complex cases, where challenging anatomy or high-flow blood is present, advanced endovascular techniques may be necessary.

Coiling Using Double Microcatheters

The 6-Fr access systems routinely used today allow for the simultaneous placement of two microcatheters within a fistula. If ideal coil anchor sites are absent, an oversized initial coil can be placed without detachment, creating a barrier. Subsequent coils are then delivered into the nest of the first coil via the second microcatheter (Figures 1-3). This technique has been used to coil large-necked intracranial aneurysms (15). Additionally, the Solitaire endovascular device (Medtronic, Irvine, CA, USA), a detachable stent, may serve as an initial nest (19).

Combinations of Coils and Non-adhesive Liquid Embolization Agents

Coiling alone may not always achieve fistula occlusion, particularly in high-flow cases. Sometimes, the anatomy prevents dense coil packing. In such situations, complete occlusion can be achieved by filling the spaces between the coils with a liquid embolization agent using the same microcatheter. Non-adhesive liquid agents, rather than glue, should be preferred. These agents can be safely injected into an already-placed coil pack, even in high-flow fistulas. Controlled, fractionated, slow injection under roadmap guidance is essential when using non-adhesive liquids for embolization. Coil and liquid combinations have been employed for transvenous obliteration of cerebral and dural AVFs. This approach enhances venous sinus obliteration, as the liquid fills the entire vascular bed (1), and can even penetrate the arterial fistula. The already-placed coil pack acts as a nest for the liquid, preventing fragmentation, while liquid accumulation within the coil nest ensures stable occlusion within minutes (Figures 3 and 4). More experienced operators can combine coils with glues (Lipiodol: cyanoacrylate ratios of 4:1 to 1:1, cyanoacrylate at 20-50% v/v). Operators must be aware that the catheter may adhere to the coil pack during retrieval, potentially pulling it back. When performed correctly, glue injection ensures immediate fistula occlusion.

Flow Arrest Techniques

These techniques prevent undesirable distal embolization that may occur if a liquid agent is dragged distally by proximal blood flow. Either a double-microcatheter or a dedicated double-lumen balloon catheter is required. In the double-microcatheter flow-arrest approach, one microcatheter is positioned at the fistula site, and the second is placed at the same level or 1–2 cm proximally. When coils are deployed through the second microcatheter, the blood flow in the arterial feeder is largely or completely stopped. This stopped flow reduces the blood's dragging force, allowing the liquid agent to be safely injected through the first catheter to achieve permanent occlusion of the feeding artery (Figures 5 and 6). In the dual-lumen balloon technique, a balloon is inflated via one lumen near the fistula, arresting flow in the feeding artery. The liquid agent is then safely injected through the second lumen of the balloon catheter. Currently, four double-lumen balloon catheters from three manufacturers are available: Ascent (Codman Neurovascular, Raynham, MA, USA), Scepter (MicroVention Terumo), Copernic 2L (Balt Extrusion), and Eclipse (Balt Extrusion). These intracranial dual-lumen balloon catheters may not be compatible with Lipiodol (i.e., glue) (14); therefore, non-adhesive liquid embolization agents should be used. To ensure stable occlusion, the fistula and proximal vein must be completely filled, and the balloon should remain inflated for some time (Figure 7).

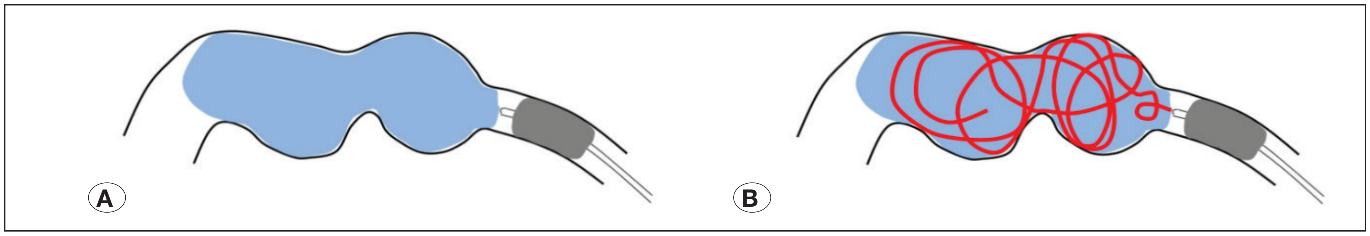

Figure 7: Schematic representation of dual-lumen balloon flow-arrest technique. **A)** After inflating the balloon to arrest flow in the feeding artery, a non-adhesive liquid agent is injected to occlude the fistula site and the proximal venous part. **B)** Coils and liquid can also be injected sequentially under balloon flow-arrest.

Coil-Liquid Embolization with Flow-arrest by a Dual-lumen Balloon

This is a variant of the "flow arrest by dual-lumen balloon" technique. All modern dual-lumen balloon catheters are DMSO-compatible and accommodate a 0.014-inch guidewire (2). After the balloon is inflated in an appropriate location and flow arrest is achieved, both coil placement and non-adhesive fluid injection can be safely performed, either separately or sequentially, through the second lumen (Figure 7). The safest approach during balloon flow arrest, even in high-flow pial AVFs, is to first create a coil pack, followed by gap-filling with a liquid agent (4).

Another challenge is the potential for hemorrhagic complications following successful obliteration of pial AVFs. The risk of new intracranial bleeding after surgical or endovascular treatment is 10–15% (10). The clinical phenomenon known as "normal perfusion pressure breakthrough hemorrhage" may occur due to the sudden occlusion of a high-flow fistula. Acute thrombosis, resulting from stagnant blood flow in the ectatic, serpiginous venous outflow or even in major dural sinuses, is not uncommon after successful embolization (9,13). Some patients with successfully occluded pial AVFs develop dural AVFs during follow-up, which may be related to post-embolization acute venous thrombosis (6,9). Previous dural sinus thrombosis is known to trigger acquired cranial dural AVF (7). However, objective evidence of "pressure breakthrough" is lacking. Nonetheless, it is reasonable to suggest that hemorrhage occurring after pial AVF occlusion is secondary to acute thrombosis of cerebral veins and/or sinuses. In our study, two of 16 patients experienced symptomatic acute thrombosis without hemorrhagic complications; one thrombosis was confined to the serpiginous draining vein of the fistula, whereas the other extended into the dural sinuses (Figure 1). Thus, post-embolization heparin therapy is strongly recommended for patients with high-flow fistulas and varicose veins (13).

The primary limitations of this study were its retrospective design and small sample size, both of which limit the discriminatory power.

CONCLUSION

Congenital pial AVF is rare. However, endovascular treatments using modern materials and techniques are highly effective.

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Declarations

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Availability of data and materials: The datasets generated and/or analyzed during the current study are available from the corresponding author by reasonable request.

Disclosure: The authors declare no competing interests.

AUTHORSHIP CONTRIBUTION

Study conception and design: CC, MK, EO, AE, IO
Data collection: CC, IO
Analysis and interpretation of results: AE, IO
Draft manuscript preparation: CC, AE, IO
Critical revision of the article: CC, IO
Other (study supervision, fundings, materials, etc...): CC, IO
All authors (CC, MK, AE, EO, IO) reviewed the results and approved the final version of the manuscript.

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