A Case Report of Endovascular Embolization for Giant Congenital Hemangioma with Arteriovenous Fistula in A Neonate

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AUTHORS' CONTRIBUTIONS

Liqi Zhang (First Author): Spearheaded the study by conceptualizing the research framework, performing the primary data curation and analysis, and drafting the initial manuscript.

Jiejun Xia (Second Author): Played a key role in the investigation process and secured critical resources essential for the research.

Lu Liu (Third Author): Was responsible for data validation and created the visual representations of the research findings.

Haibo Li (Corresponding Author): Provided the overarching conceptualization and supervision for the project, and undertook the critical tasks of reviewing, revising, and providing final approval for the manuscript.

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None.

DISCLOSURES

The authors declare no conflicts of interest.

CONSENT

Yes.

HUMAN AND ANIMAL RIGHTS

All procedures performed were in accordance with the ethical standards of the institutional and national research committee and with the 1964 Helsinki Declaration and its later amendments.

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ABSTRACT

Background: Congenital hemangioma (CH) is a vascular tumor that develops during fetal life. Although most CHs are benign, those associated with high-flow arteriovenous fistulas (AVFs) can lead to life-threatening complications such as high-output cardiac failure and severe pulmonary hypertension.

Case Presentation: We report a male preterm infant (birth weight: 2.8 kg) with a giant CH and AVF on the left forehead, confirmed by transcranial Doppler and magnetic resonance angiography (MRA). The patient rapidly developed high-output cardiac failure and respiratory failure. Despite maximal medical therapy, his condition deteriorated. A multidisciplinary team deemed surgical resection prohibitively high-risk, and transcatheter arterial embolization was selected as the primary intervention.

Intervention and Outcome: On day 11 of life, successful embolization was performed via a right femoral approach under general anesthesia. Despite a femoral artery diameter <1 mm, vascular access was achieved using a pediatric needle and Seldinger technique. Angiography confirmed a high-flow AV shunt, which was embolized using a combination of detachable and free coils. Post-procedurally, pulmonary artery pressure decreased from 77 mmHg to 42 mmHg, and the ductus arteriosus shunt converted from right-to-left to bidirectional. The patient was weaned from mechanical ventilation on day 8 post-procedure. Four months later, radical resection confirmed CH with AV malformation. At one-year follow-up, there was no neurological deficit or recurrence.

Conclusion: Transcatheter embolization is a life-saving treatment for neonates with CH and high-flow AVF when surgery is not feasible. This case highlights the feasibility of complex endovascular interventions in extremely low-weight infants and underscores the value of multidisciplinary management.

Keywords: Congenital hemangioma, Arteriovenous fistula, Embolization, Neonate, Cardiac failure

CASE REPORT

CASE PRESENTATION

A male neonate was born prematurely with a birth weight of 2.8 kg. On the first day of life, a soft, red mass (approx. 6.2×5.4×2.7 cm) was noted on the left frontoparietal region (Figures 1,2). Cranial ultrasound revealed a mixed echoic mass with internal flow signals, suggestive of a hemangioma. Subsequent magnetic resonance angiography (MRA) confirmed a large vascular space-occupying lesion (Figure 3).

Cardiac ultrasound demonstrated severe hemodynamic compromise, including pulmonary hypertension (77 mmHg), a patent ductus arteriosus (PDA) with right-to-left shunt, right heart enlargement, and moderate tricuspid regurgitation. Initial supportive care included antibiotics and milrinone.

On day 6, the infant's condition deteriorated abruptly, with severe hypoxemia (SpO_2 dropped to 40%), respiratory distress, and pulmonary hemorrhage, necessitating endotracheal intubation and mechanical ventilation. He was transferred emergently to our hospital.

Upon admission, inflammatory markers were elevated. Blood gas analysis indicated compensated respiratory acidosis. Chest X-ray showed bilateral pulmonary exudates and cardiomegaly. Despite aggressive medical management—including inotropic support, diuretics, inhaled nitric oxide, sirolimus, and corticosteroids—the hemodynamic status remained critical.

Given the patient's low weight, tenuous clinical status, and high surgical risk, a multidisciplinary team decided to proceed with transcatheter arterial embolization.

INTERVENTIONAL TREATMENT AND OUTCOME

On day 11 of life, the patient underwent embolization under general anesthesia. Using ultrasound guidance, the right femoral artery (diameter <1 mm) was cannulated via the Seldinger technique with a 4F vascular sheath. Selective angiography of the carotid arteries revealed a high-flow arteriovenous fistula fed by enlarged external carotid branches, with early venous drainage (Figure 4A).

A combination of detachable and free coils was deployed to embolize the nidus, achieving >80% devascularization on post-procedure angiography (Figure 4B). Liquid embolic agents were avoided to minimize risks of non-target embolization and inflammation.

Hemodynamic improvement was rapid. Pulmonary artery pressure decreased to 42 mmHg, and PDA flow became bidirectional. The patient was successfully weaned from mechanical ventilation on post-procedure day 8. The lesion size reduced markedly (Figure 5).

Four months later, the patient underwent uncomplicated surgical resection. Histopathology confirmed congenital hemangioma with arteriovenous malformation (Figure 6). One-year follow-up showed no residual tumor, normal neurological development, and sustained cardiac function.

DISCUSSION

This case illustrates the critical role of endovascular embolization in managing life-threatening neonatal vascular anomalies with high-output cardiac failure[1]. Several aspects warrant emphasis.

Comparison with Previous Cases

Compared to previously reported cases of neonatal CH with AVF, the present case was particularly challenging due to the extremely low birth weight (2.8 kg) and the femoral artery diameter of less than 1 mm. While there are reports of successful embolization in neonates, many involve larger infants or the use of liquid embolic agents like Onyx, which carry a higher risk of complications in the delicate craniofacial vasculature of a preterm newborn [1-3]. Our decision to use a 4F sheath and a combination of detachable and free coils, avoiding liquid embolics, represents a tailored approach that prioritized safety in a high-risk scenario. This strategy resulted in a successful outcome without neurological sequelae, underscoring that technical adaptability is as important as the choice of embolic agent. Furthermore, the rapid hemodynamic improvement post-embolization highlights that early intervention, before irreversible organ damage occurs, is a key prognostic factor, a principle that is consistent with but more critically demonstrated in this extreme-case setting.

Role of Embolization vs. Pharmacotherapy

While mTOR inhibitors like sirolimus have a role in managing vascular anomalies, their slow onset limits utility in acute heart failure [4]. In this case, only embolization could rapidly reverse the shunt-related pathophysiology.

Technical Considerations

The successful navigation of a sub-millimeter femoral artery underscores that vessel size alone should not preclude intervention. The use of a 4F sheath represented a balance between access safety and procedural feasibility [5]. The coil-based strategy provided controlled occlusion, avoiding risks associated with liquid embolics in neonates [6,7].

Multidisciplinary Collaboration

The integration of neonatology, interventional radiology, cardiology, and surgery was fundamental to success. Meticulous perioperative management of pulmonary hypertension and cardiac function enabled safe intervention.

CONCLUSION

This report confirms that transcatheter embolization is a viable, life-saving option for critically ill neonates with giant

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CH and AVF, even in extreme low-weight cases. Success hinges on early diagnosis, understanding of pathophysiology, technical precision, and multidisciplinary collaboration.

TEACHING POINT

1. Endovascular embolization should be considered as a primary therapeutic option in neonates with giant congenital hemangiomas complicated by arteriovenous shunting and life-threatening high-output heart failure, offering a minimally invasive alternative to surgery.

2. A multidisciplinary team comprising interventional radiology, neonatology, pediatric cardiology, and pediatric anesthesiology is essential for the safe and successful management of these complex neonatal cases.

3. Superselective angiography is critical for precisely defining the angioarchitecture of the lesion and the site of arteriovenous fistulization, which directly guides the choice of embolic agent and technique to ensure targeted therapy and minimize complications.

4. This case demonstrates that with appropriate expertise, endovascular intervention is feasible and effective in the neonatal period, challenging the notion that patient size or age precludes such advanced management and allowing for early, curative treatment.

QUESTIONS

Question 1: Which imaging modality is most useful for confirming a high-flow arteriovenous fistula in a neonatal hemangioma?

1. Plain X-ray
2. CT without contrast
3. Magnetic resonance angiography (applies)
4. Nuclear scintigraphy
5. PET-CT

Explanation: Magnetic resonance angiography (MRA) is the preferred non-invasive modality for confirming high-flow arteriovenous shunting in congenital hemangiomas. [MRA confirmed a large vascular space-occupying lesion.]

Question 2: What is the most common life-threatening complication of a high-flow arteriovenous fistula in a neonate?

1. Renal failure
2. High-output cardiac failure (applies)
3. Liver dysfunction
4. Sepsis
5. Seizure

Explanation: High-output cardiac failure is a well-documented complication due to significant left-to-right shunting. [Those associated with high-flow arteriovenous fistulas (AVFs) can lead to life-threatening complications such as high-output cardiac failure.]

Question 3: Which embolic material was avoided in this case to reduce the risk of non-target embolization?

1. Detachable coils
2. Free coils
3. Liquid embolic agents (applies)
4. Gelfoam
5. Stents

Explanation: Liquid embolic agents were avoided due to the risk of non-target embolization in the delicate neonatal craniofacial vasculature. [Liquid embolic agents were avoided to minimize risks of non-target embolization and inflammation.]

Question 4: What was the femoral artery diameter in this neonate?

1. <1 mm (applies)
2. 1–2 mm
3. 2–3 mm
4. 3–4 mm
5. 4 mm

Explanation: The femoral artery diameter was less than 1 mm, highlighting the technical challenges of vascular access in neonates. [Despite a femoral artery diameter <1 mm, vascular access was achieved using a pediatric needle and Seldinger technique.]

Question 5: Which of the following is a key factor for successful outcome in such cases?

1. Delayed intervention
2. Multidisciplinary collaboration (applies)
3. Use of liquid embolics only
4. Avoidance of imaging
5. Surgical resection as first-line

Explanation: Multidisciplinary collaboration is essential for managing complex neonatal vascular anomalies. [Success hinges on early diagnosis, understanding of pathophysiology, technical precision, and multidisciplinary collaboration.]

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FIGURES

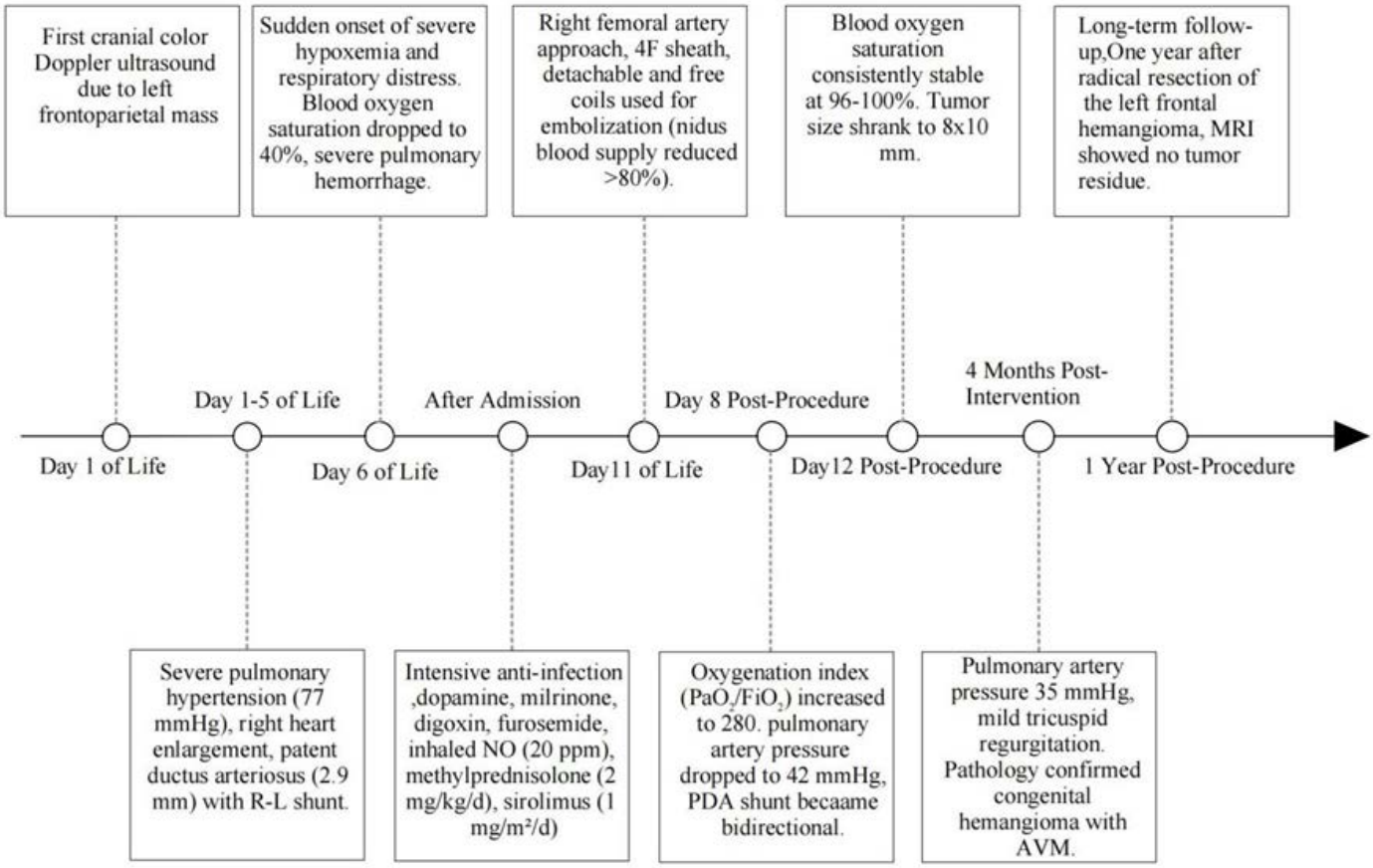

Figure 1: Timeline of patient's clinical course

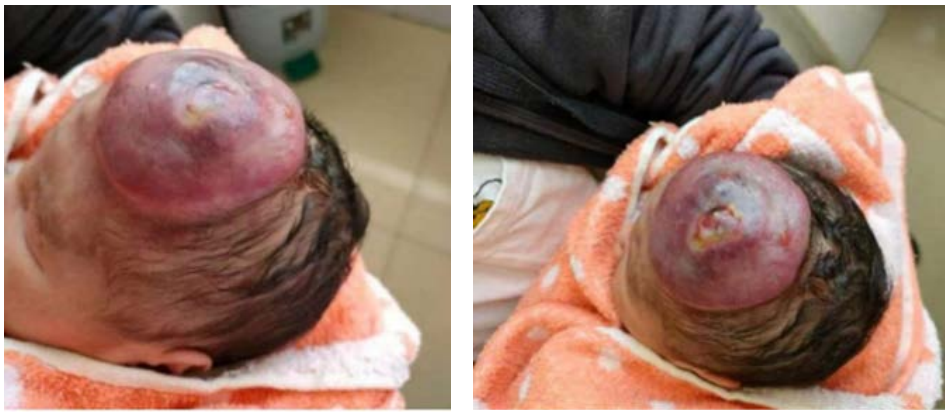

Figure 2: Preoperative clinical photograph. A large, dark red, soft-mass lesion is visible on the left frontal region of the infant. The overlying skin is thin, with areas of slight ulceration.

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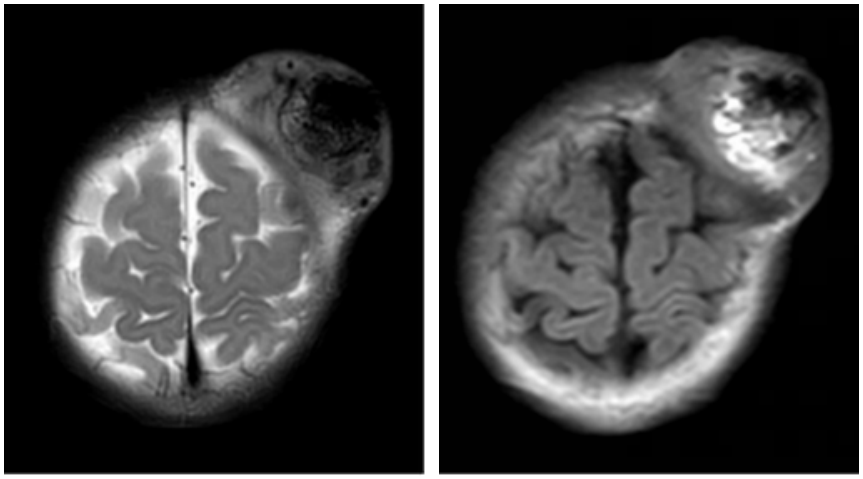
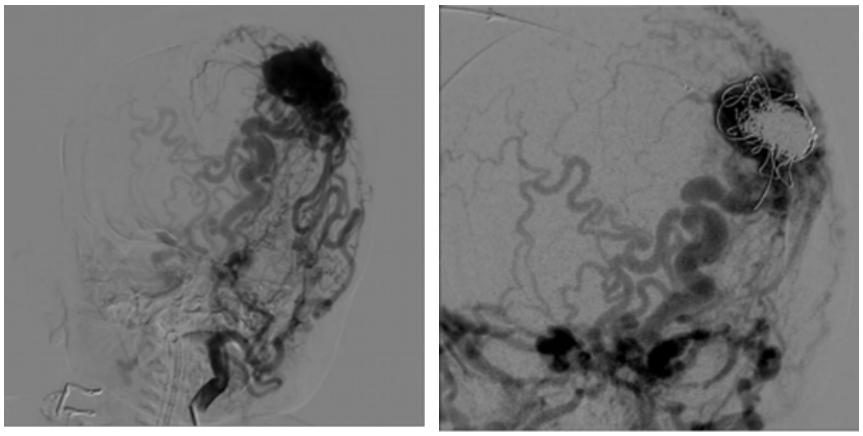


3A 3B

Figure 3: Magnetic Resonance Imaging (MRI), T1-weighted image(B) and T2-weighted image(A) show a huge abnormal signal mass in the left frontal region.



4A 4B

Figure 4: Digital Subtraction Angiography (DSA) images. (A) Pre-procedural image from selective left external carotid angiography shows enlarged arterial feeders and early opacification of draining veins, confirming a high-flow arteriovenous fistula. (B) Post-embolization angiography demonstrates a marked reduction in nidal staining and the absence of arteriovenous shunting.

5A 5B

Figure 5: Clinical appearance 7 days after interventional surgery. The mass in the left frontal region is significantly reduced in size.

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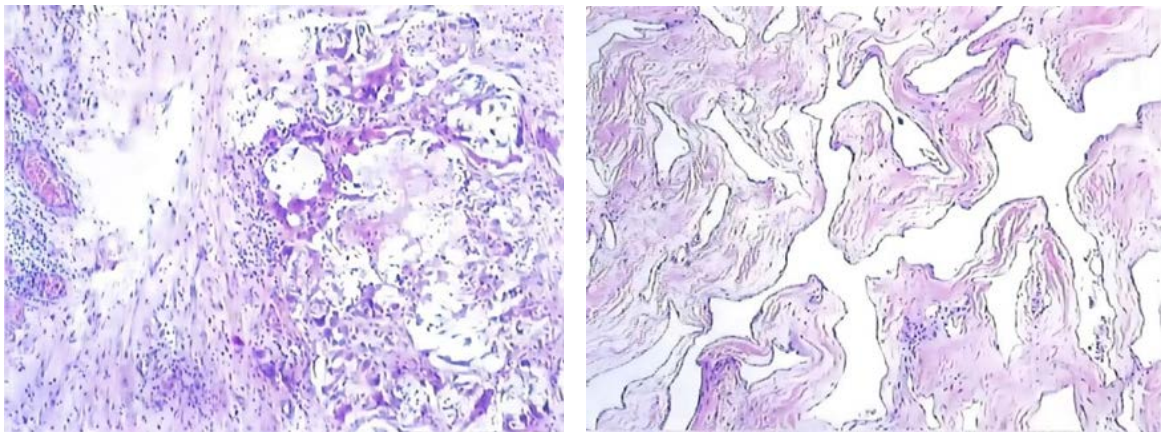

6A 6B

Figure 6: Histopathology of the forehead skin lesion (H&E stain) (A) Low-power view shows the dermis occupied by irregularly dilated, thin-walled vascular channels lined by flattened endothelial cells. (B) High-power view reveals a chronic inflammatory infiltrate, multinucleated giant cells, and focal calcifications within the stroma.

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Table 1: Summary of Giant Congenital Hemangioma with Arteriovenous Fistula in Neonates

Category	Details
Etiology	Abnormal vascular development during fetal life; the exact molecular mechanism is not fully elucidated but may involve somatic mutations.
Incidence	Rare. The overall incidence of congenital hemangioma is approximately 1%, with cases associated with significant AVF being even rarer.
Gender Ratio	Literature reports are variable.
Age Predilection	Congenital, present at birth.
Risk Factors	No clear prenatal or genetic risk factors have been identified.
Treatment	1. Emergency Care: Pharmacological management of high-output cardiac failure (inotropes, diuretics, pulmonary vasodilators). 2. Definitive Therapy: Endovascular embolization is the primary life-saving intervention to rapidly reverse hemodynamic compromise. 3. Adjunct/Subsequent Therapy: mTOR inhibitors (e.g., Sirolimus) have a slow onset and are often used for maintenance or non-emergent cases. Ultimate radical surgical resection is feasible.
Prognosis	Dependent on the timing of diagnosis and intervention. Untreated, the risk of mortality from cardiac failure is high. Prompt and successful embolization leads to rapid cardiac improvement, with a good long-term prognosis and no neurological sequelae or recurrence.
Findings on Imaging	See Table 2.

Table 2: Imaging Differential Diagnosis of Hypervascular Lesions in the Neonatal Head and Neck

Entity	Key Imaging Findings
Congenital Hemangioma with AVF	US/CT/MRI: Large, well-defined mass with prominent flow voids. Angio: High-flow arteriovenous shunt with early venous drainage.
Infantile Hemangioma	US: Solid, homogeneous, hypervascular mass. MRI: T2 hyperintense, intensely and homogeneously enhancing.
Arteriovenous Malformation	MRI: Tangled vessels with flow voids, no parenchymal mass. Angio: Direct high-flow shunt (nidus) with markedly enlarged draining veins.
Vascular Tumor (e.g., Sarcoma)	MRI: Infiltrative, destructive mass with heterogeneous enhancement and necrosis.
Meningoencephalocele	CT/MRI: Skull defect with herniation of CSF, meninges, or brain tissue. Typically non-enhancing or only mild peripheral enhancement.

KEYWORDS

Congenital hemangioma; Arteriovenous fistula; Embolization; Neonate; Cardiac failure

ABBREVIATIONS

CH = Congenital Hemangioma
AVF = Arteriovenous Fistula
MRA = Magnetic Resonance Angiography
PDA= Patent Ductus Arteriosus

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