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Case Report

Successful treatment by coil embolization for pediatric brachial arteriovenous fistula: A case report ☆,☆☆

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ABSTRACT

Arteriovenous fistulae of the upper limbs are rare in the pediatric population. They can be caused by trauma, needle puncture, or other iatrogenic injuries. A 5-year-old boy presented with progressive swelling of the right hand, which was initially misinterpreted as an arteriovenous malformation based on his noninvasive diagnostic work-up. He was ultimately diagnosed with right brachiocephalic arteriovenous fistula by catheter angiography, and the fistula was then successfully treated with coil embolization. This article describes the relevant imaging findings and potential implications for treatment.

Introduction

Arteriovenous fistulae (AVFs) of the upper limbs are rarely encountered in children [1,2]. They can be caused by trauma, needle puncture, or other iatrogenic injuries [3–5]. The most common presenting symptom is a palpable lump with a pulsatile thrill, especially in patients with longstanding diseases [1,2]. Accurate diagnosis and localization of the fistulous point are important, and differential diagnosis with other vascular lesions, such as arteriovenous malformation (AVM) or hemangioma, is critical because the treatment methods and potential complications differ between these entities [6,7]. Conventionally, AVFs of the upper limbs are treated with surgical ligation, but endovascular embolization is also feasible in selected cases and is being increasingly performed to treat peripheral AVFs [2,4,8].

Case report

A 5-year-old boy had a history of preterm birth and a relatively large size of the right forearm since birth. He presented

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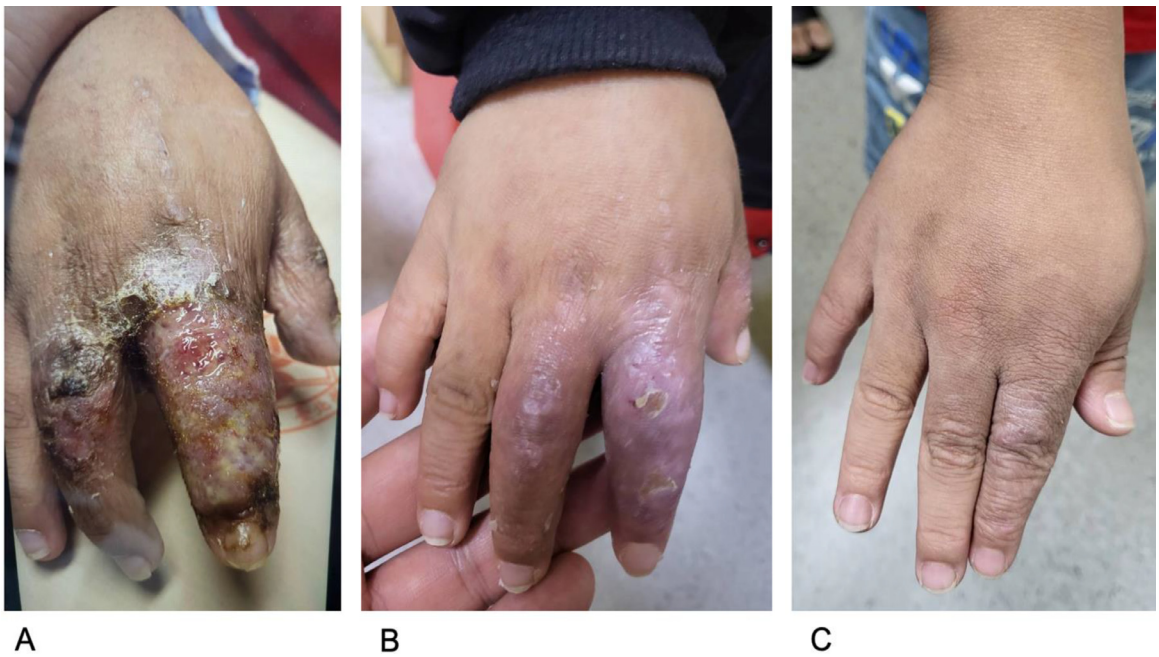

Fig. 1 – Patient's right hand before and after embolization. (A) The patient presented with marked swelling of his second and third fingers on the right hand, with erythema, skin erosions, and crusts. He was later found to have a brachiocephalic AVF. (B) Three weeks after AVF embolization, the wound was mostly healed, but mild swelling and erythema were still present. (C) Ten months after embolization, the swelling had almost completely resolved, and only increased skin pigmentation could be observed.

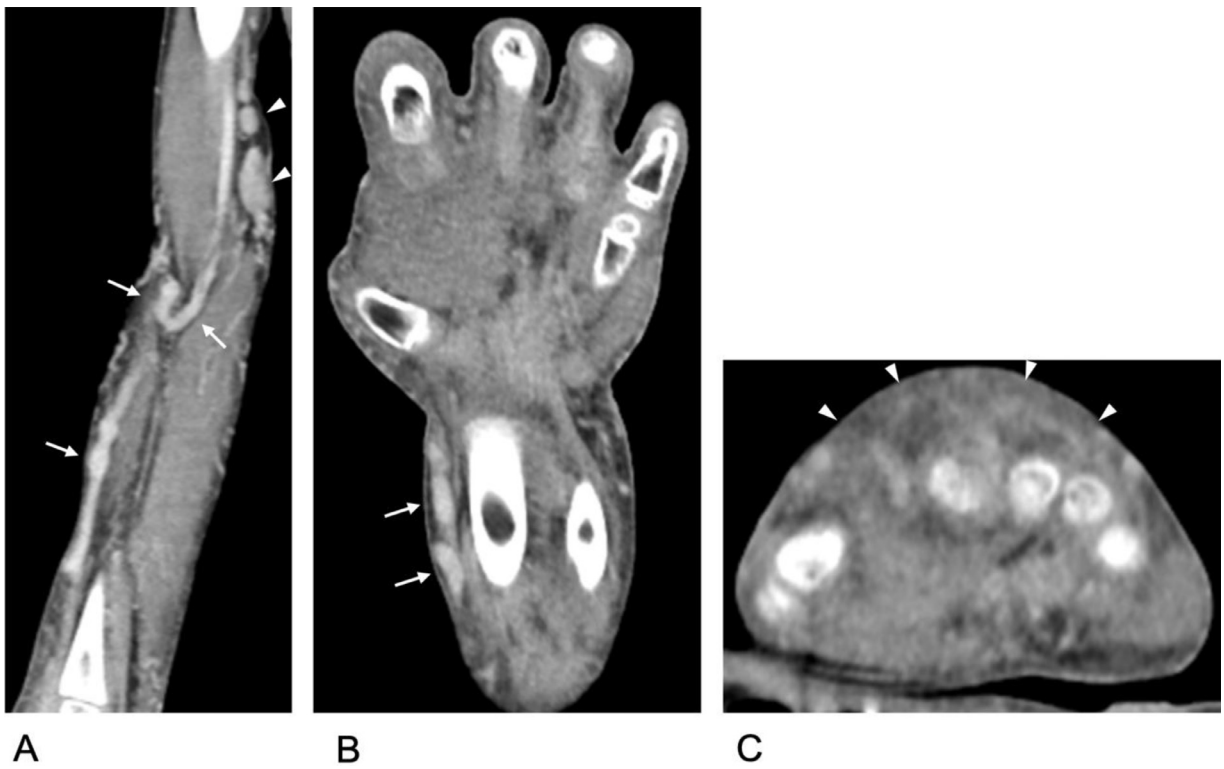

Fig. 2 – Contrast-enhanced CT of the right forearm and right hand in the venous phase. (A, B) Coronal images of the right arm (A) and right hand (B) showed enlarged vessels on the radial side of the patient's right forearm and right hand (arrows). Several enlarged lymph nodes on the medial side of the patient's right upper arm were observed (arrowheads in A). (C) Axial view of the right hand showed extensive soft tissue edema (arrowheads).

to our clinic with progressive swelling of his right hand after an insect bite 2 months previously. Upon inspection, his right second and third fingers were markedly swollen with erythema, skin erosions, and crusts (Fig. 1A). The patient was admitted, and antibiotic treatment was initiated for suspected infection, but his symptoms did not improve. Because of progressive swelling and poor wound healing, contrast-enhanced computed tomography (CT) of his right hand was performed to evaluate the extent of inflammation. CT revealed enlarged, serpiginous vascular structures on the radial side of his right forearm as well as soft tissue edema in the right hand (Fig. 2). Subsequently, contrast-enhanced magnetic resonance (MR) angiography was performed and revealed an enlarged and mildly tortuous vessel draining into many indistinct collateral veins; this vessel was presumed to be the radial artery (Fig. 3). Initially, the lesion was erroneously described as an AVM that was fed by the right radial artery, which drained into many indistinct vascular niduses. The patient did not have any symptoms or signs of high cardiac output heart failure. Therefore, interventional radiologists were consulted for catheter angiography and to determine further treatment.

The patient underwent diagnostic angiography under general anesthesia. Digital subtraction angiography (DSA) after brachial artery injection revealed similar results, as did contrast-enhanced MR angiography. However, DSA also revealed several bulbous protrusions along the dilated vessel, and the relationship of the enlarged vessel with the brachial artery and the normal-appearing radial artery were better delineated (Fig. 4A). After careful evaluation of the images, the lesion was diagnosed as a brachiocephalic AVF; the fistula drained retrogradely into an engorged cephalic vein (Figs. 3A and B, and 4A), which was incorrectly identified as an enlarged radial artery by CT and MR angiography, and the bulbous protrusions along the vessel were in fact venous valves of the engorged right cephalic vein.

After a discussion regarding the benefits and risks of surgical ligation and endovascular embolization, the patient's parents opted for embolization. Embolization was performed 1 week later with the patient under general anesthesia. The right common femoral artery was punctured, and access was secured using a 4-Fr introducer sheath (Terumo, Tokyo, Japan). We placed a 4-Fr JB1 catheter (Cordis, Miami, FL, USA) into

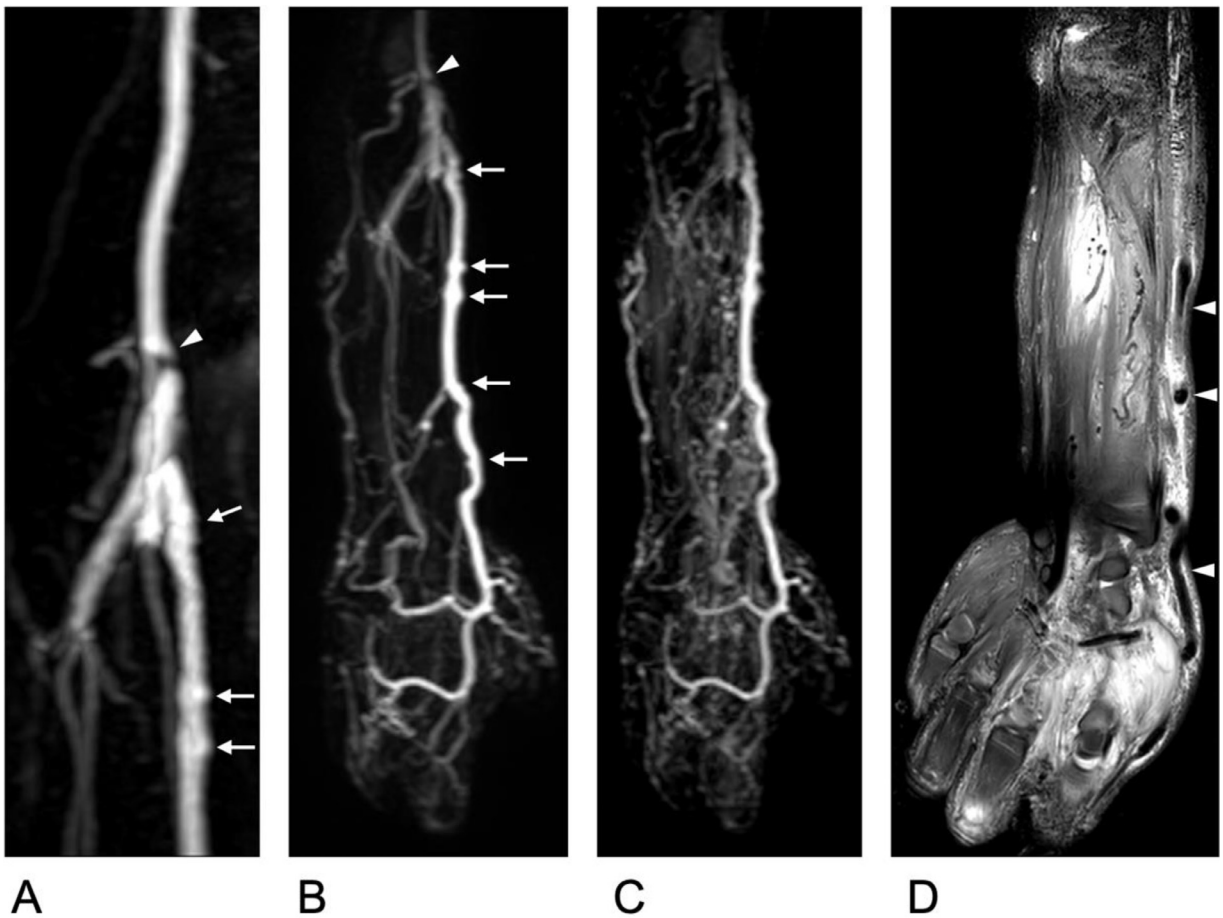

Fig. 3 – Contrast-enhanced MR angiography of the right hand. (A, B) Maximal intensity projections (MIPs) of contrast-enhanced MR angiography of the right elbow (A) and right forearm (B) revealed an enlarged and mildly tortuous vessel on the radial side of the right forearm in the early arterial phase. The brachiocephalic AVF (arrowhead) and the venous valves in the cephalic vein (arrows) could be identified retrospectively. (C) In the venous phase on this MIP image, multiple small, indistinct collateral veins were observed. (D) Coronal T2-weighted images with fat saturation showed extensive soft tissue edema in the right hand, with prominent flow-void artifact (arrowheads) on the radial side.

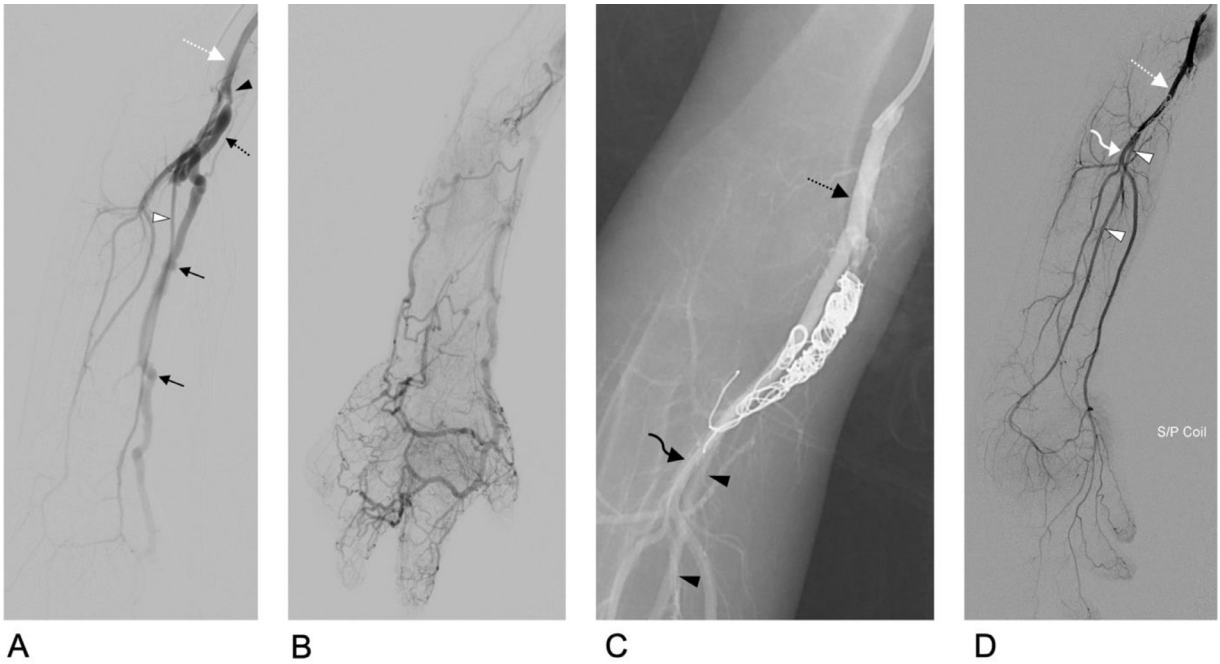

Fig. 4 – Catheter angiography and embolization. (A) Brachial angiogram revealed a fistula (black arrowhead) between the right brachial artery (dotted white arrow) and the right cephalic vein (dotted black arrow). The normal right radial artery could be clearly identified (white arrowhead). Several bulbous structures were present along the engorged cephalic vein (black arrows); these structures were venous valves that should not be present in a normal artery. (B) Delayed brachial angiogram showed retrograde contrast opacification of the venous arch of the right hand. (C) Stored fluoroscopic roadmap image during embolization showed several 0.018-inch microcoils deployed at the proximal segment of the drainage veinous limb of the AVF. (D) After embolization, control angiogram showed the complete occlusion of the AVF. The brachial artery (dotted arrow), radial artery (arrowheads), and ulnar artery (curved arrow) and their distal branches were patent after embolization.

the right brachial artery and then advanced a 1.98-Fr ASAHI Masters PARKWAY SOFT microcatheter (Asahi Intecc, Aichi, Japan) into the fistula. The AVF was embolized with several 0.018-inch microcoils placed at the proximal segment of the drainage venous limb (Fig. 4C). Post-embolization angiogram showed the complete occlusion of the fistula and patent distal arteries of the patient's right forearm (Fig. 4D). Subsequently, his symptoms gradually improved, and the wound healed after embolization at 3-weeks and 10-months follow-up (Figs. 1B and C).

Discussion

Brachial AVFs in the pediatric population are rare and seldom reported in the literature [1,2]. They can be caused by trauma, needle puncture, or other iatrogenic injury [3–5]. Most patients remain asymptomatic for several years until local swelling and a palpable lump with a pulsatile thrill become clinically noticeable. However, brachial AVFs can result in an abnormally strong cardiofugal (i.e. retrograde) venous flow, causing venous congestion and tissue ischemia, as in the present case; these AVFs with retrograde venous flow are contrary to the surgical AVFs created for hemodialysis access, which are created by vascular surgeons to increase cardiopetal (i.e. antegrade) blood flow for ease of needle puncture [9]. Furthermore, untreated brachial AVFs have been reported to cause high cardiac output heart failure, especially in very young patients such as newborns [10].

The exact cause of the brachiocephalic AVF in our patient was unclear. Given the patient's history of preterm birth, we speculated that the AVF was caused by repeated venipuncture during hospitalization for preterm birth, but it was not clinically apparent for several years. Moreover, the patient's parents recalled a slight asymmetry of the boy's both forearms since he was very young. We theorized that the aforementioned insect bite caused an infectious episode and the release of inflammatory cytokine, leading to vasodilation and increased vascular shunting [11]. The patient did not have any clinical signs of heart failure, and no evidence of syndromes associated with vascular malformation, such as Klippel-Trenaunay syndrome or hereditary hemorrhagic telangiectasia, was found in the diagnostic work-up.

The accurate diagnosis of and differentiation between an AVF and AVM are crucial because the treatment methods of these conditions differ and because inappropriate management may lead to undesirable complications. According to the classification system of the International Society for the Study of Vascular Anomalies (ISSVA), both conditions are defined as high-flow vascular lesions that shunt arterial blood directly to one or more drainage veins without an intervening capillary

bed [12]. AVMs are characterized by the presence of a vascular nidus, and most are believed to be congenital; by contrast, AVFs are direct communication between an artery and a vein, and they are often acquired [6,13]. According to another classification system for the trunk and extremity AVMs proposed by *Cho et al*, an AVF with 1 to 3 arteries that shunt to a single vein is defined as type 1 AVM based on the angiographic morphology; this classification system has been used by interventional radiologists to guide therapy and to compare treatment outcomes [14].

Ultrasound is the first-line tool for the diagnosis of peripheral vascular lesions because of its abilities to access vascular structures and to measure the velocity and direction of blood flow without ionizing radiation exposure. CT and MR imaging can provide high-resolution images with a large field of view of blood vessels and their surrounding structures, and multiplanar reconstruction can aid in the diagnosis of and treatment planning for vascular lesions.

The engorged drainage veins of AVFs and its venous valves may be inaccurately identified as enlarged feeding arteries in AVMs [8]. If an AVF is misdiagnosed as an AVM and is embolized distally into the drainage veins, theoretically, venous congestion may be aggravated, potentially leading to limb ischemia and tissue infarction. Therefore, the careful evaluation of angiographic images and the identification of key angioarchitecture are of paramount importance. In the present case, the fistulous point, the venous valves in the cephalic vein, and the normal radial artery were identified only through catheter angiography; they were difficult to identify even retrospectively by CT and MR angiography.

Conventionally, peripheral AVFs are treated with surgical ligation by vascular surgeons, but endovascular embolization is feasible in selected cases and is being increasingly performed for the treatment of peripheral AVFs [2,4,8]. For AVF ligation, surgery at the antecubital fossa carries a risk of inadvertent injury to the surrounding structures, such as the median nerve or normal branches of the brachial artery, during exploration, and the location of the fistula may be difficult to identify, especially in pediatric patients [15]. Although associated with ionizing radiation, which should be kept as low as reasonably possible, endovascular embolization is a minimally invasive approach involving only a small femoral puncture, and it enables the accurate localization of the fistulous point and is associated with minimal risk of injury to the surrounding structures.

Conclusion

Brachial AVFs are rare in the pediatric population. They can be caused by trauma, needle puncture, or other iatrogenic injuries. A palpable lump with a pulsatile thrill is the most common presenting symptom; however, these fistulae can also cause venous congestion or high cardiac output heart failure if left untreated. The engorged drainage vein in a chronic AVF may be incorrectly identified as an enlarged feeding artery of an AVM, and the presence of venous valves and the identification of normal arteries are crucial for accurate diagnosis. AVFs are conventionally treated with surgical ligation. However, an endovascular approach for embolization is a novel and minimally invasive strategy for the treatment of peripheral AVFs; this approach enables the accurate localization of the fistulous point and is associated with minimal risk of injury to the surrounding structures. In conclusion, endovascular embolization can be safely and effectively performed in selected patients with peripheral AVFs.

Patient consent

Written informed consent was obtained from the patient's mother.

REFERENCES

[1] Megremis SD, Christaki MA, Mourkoyiannis NK, Papadopoulos GS, et al. Iatrogenic brachial arteriovenous fistula in a child: color Doppler ultrasonographic evaluation. J Ultrasound Med 2006;25(6):809–12. doi:10.7863/jum.2006.25.6.809.

[2] Youn YJ, Kim CW, Park IIH, Byun CS, et al. Delayed treatment of iatrogenic brachial arteriovenous fistula. Korean J Thorac Cardiovasc Surg 2020;53(6):408–10. doi:10.5090/kjtcs.20.019.

[3] Norcross WA, Shackford SR. Arteriovenous fistula. A potential complication of venipuncture. Arch Intern Med 1988;148(8):1815–16. doi:10.1001/archinte.148.8.1815.

[4] Lin JX, Hill A. Delayed presentation of an iatrogenic, traumatic brachio-brachial fistula. Trauma Case Rep 2021;32:100438. doi:10.1016/j.tcr.2021.100438.

[5] Hashimoto S, Banshodani M, Marubayashi S, Shintaku S, Moriishi M, Tsuchiya S, et al. Iatrogenic arteriovenous fistula in the brachial artery after percutaneous coronary intervention in a patient with end stage kidney disease. J Vasc Access 2024;25(3):1011–14. doi:10.1177/11297298231158427.

[6] Ota Y. Vascular malformations and tumors: a review of classification and imaging features for cardiothoracic radiologists. Radiol Cardiothorac Imaging 2023;5(4):e220328. doi:10.1148/ryct.220328.

[7] Behr GG, Johnson C. Vascular anomalies: hemangiomas and beyond–part 1, Fast-flow lesions. AJR Am J Roentgenol 2013;200(2):414–22. doi:10.2214/ajr.11.7852.

[8] Yakes WF. Endovascular management of high-flow arteriovenous malformations. Semin Intervent Radiol 2004;21(1):49–58. doi:10.1055/s-2004-831405.

[9] Lok CE, Huber TS, Lee T, Shenoy S, Yevzlin AS, Abreo K, et al. KDOQI clinical practice guideline for vascular access: 2019 update. Am J Kidney Dis 2020;75(4):S1–s164. doi:10.1053/j.ajkd.2019.12.001. Suppl 2.

[10] Teomete U, Gugol RA, Neville H, Dandin O, Young ML, et al. High output cardiac failure resolving after repair of AV fistula in a six-month-old. Case Rep Vasc Med 2016;2016:8564081. doi:10.1155/2016/8564081.

[11] Vila E, Salaices M. Cytokines and vascular reactivity in resistance arteries. Am J Physiol-Heart Circulat Physiol 2005;288(3):H1016–21. doi:10.1152/ajpheart.00779.2004.

[12] ISSVA Classification of Vascular Anomalies. International Society for the Study of Vascular Anomalies. Updated May 2018. Accessed April 10th, 2024. https://www.issva.org/UserFiles/file/ISSVA-Classification-2018.pdf.

[13] Wang MX, Kamel S, Elsayes KM, Guillerman RP, Henget L, et al. Vascular anomaly syndromes in the ISSVA

classification system: imaging findings and role of interventional radiology in management. Radiographics 2022;42(6):1598–620. doi:10.1148/rg.210234.
[14] Cho SK, Do YS, Shin SW, Kim Dk, Kim YW, Park KB, et al. Arteriovenous malformations of the body and extremities: analysis of therapeutic outcomes and approaches according to a modified angiographic classification. J Endovasc Ther 2006;13(4):527–38. doi:10.1583/05-1769.1.
[15] Dukkipati R. Median nerve and ulnar nerve entrapment with cubital tunnel syndrome in a hemodialysis patient following creation of an arteriovenous fistula. Case Rep Nephrol Dial 2021;11(3):275–80. doi:10.1159/000518001.