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Case Report

# A case report of misdiagnosed iatrogenic arteriovenous fistula as an arteriovenous malformation during infancy: diagnostic challenges and treatment options

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## ABSTRACT

**Introduction:** Arteriovenous malformations (AVMs) are the most frequent vascular abnormality during childhood. However, it is essential to differentiate it from other less common conditions, such as iatrogenic arteriovenous fistula (AVF), due to different diagnostic and treatment modalities.

**Case Presentation:** Here, we reported a four-year-old boy who was followed up with a diagnosis of AVM from the age of two. In our center, a diagnosis of peripheral AVF was made for the patient in his right axillary region based on computed tomography angiography (CTA) findings. During open surgery, vein ligation and repair of the brachial artery were considered.

**Conclusion:** It is recommended that all patients with a diagnosis of AVM during infancy, especially with a history of numerous hospitalizations and catheterization, when follow-up till the age of four, have a confirmatory test are a viable option.

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## Introduction

Any repetitive vascular manipulations make patients susceptible to iatrogenic arteriovenous fistula (AVF). Iatrogenic arteriovenous fistulas of the vessels of the thorax and upper limbs are clearly outlined in adults, especially after cardiac device implantations[1-3]. However, it is rarely highlighted during childhood. Moreover, due to their nonspecific presentation, besides being rare, AVFs in this region pose a diagnostic challenge. This is why the definitive diagnosis and differentiating it from other vascular malformations, such as arteriovenous malformations (AVM) and hemangioma, are critical before treatment due to different treatment modalities. Here, we reported a boy referred to our department for treatment of a peripheral AVM in his right axillary region, whose final diagnosis was the brachio-brachial AVF.

## Case

A four-year-old boy was referred to our department for assessment of edema of the right upper limb. The boy was the know case of galactosemia leading to recurrent hospitalizations. He had been followed up with a diagnosis of AVM in his right axillary region from the age of two. The physical examination revealed engorgement of the thoracic and arm veins, with a thrill and bruit in the antecubital region, accompanied by numerous collateral veins in both the upper and lower thoracic regions (Figure 1). Doppler ultrasound study showed dilated and tortoise vessels of axillary artery (diameter 12.5 mm) and vein (diameter 8.5 mm), and also the brachial artery (diameter 8.7 mm) and vein (diameter 7.1 mm), besides turbulent arterial flow of aforementioned arteries and arterialized blood flow in axillary and brachial veins, suggesting the presence of AVM and or hemangioma. However, to make a definitive diagnosis, computed tomography angiography (CTA) of the head, neck and right upper limb was requested. CTA showed multiple serpiginous and vermiform arteries in the subcutaneous region of the right shoulder, right hemi-chest wall, and right arm. An abnormal AV connection between the brachial artery and vein, and also between the subclavian artery and vein, was detected. Moreover, a stenosis at the subclavian vein just before its connection to the internal jugular vein was recorded. When the medical documents of patients were reviewed retrospectively, a significant history of multiple vascular manipulations and cauterizations of the brachial vein was recorded. Therefore, with a definitive diagnosis of the brachio-brachial AVF, patients underwent open surgery and primary repair of the brachial artery. An intraoperative Doppler ultrasound detected the location of AVF was visualized, and using an excision of the antecubital area, the fistula between the brachial artery and the deep brachial vein was exposed (Figure 2A). Therefore, vein ligation and repair of the brachial artery were performed (Figure 2B). After surgery, there was no bruit and thrill. Two weeks after surgery, there was no sign of oedema and subcutaneous collateral veins.

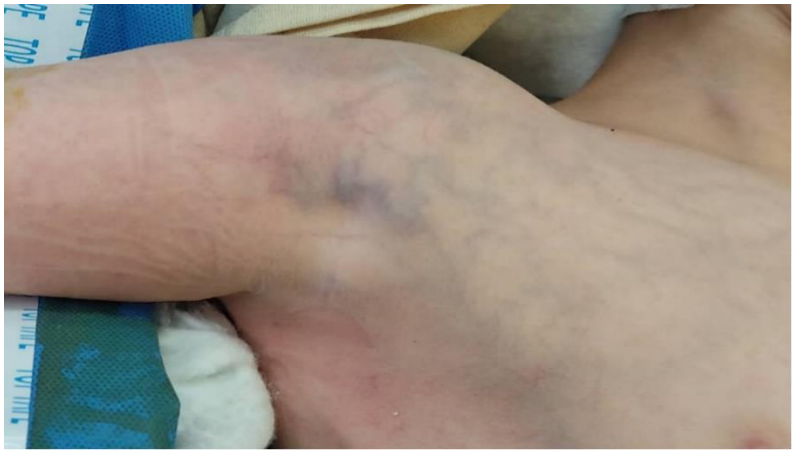

**Figure 1**: Primary presentation of patients with oedema of the right upper limb engorgements of the thoracic and arm veins

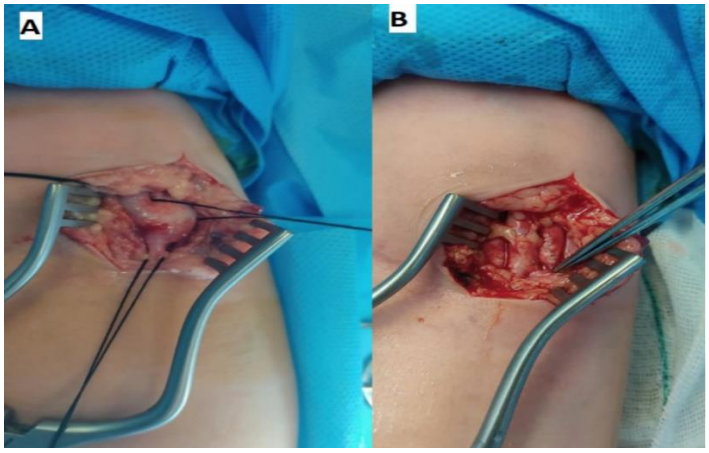


**Figure 2:** During the surgery, a 0000 was observed (Figure 2A) and therefore vein ligation and repair of the brachial artery were performed (Figure 2B).

## Discussion

Iatrogenic AVFs during childhood and infancy are only reported as case reports[4-8]. It is hypothesized that recurrent endothelial damage following frequent catheterizations of vessels may lead to the development of a hematoma. And the healing processes cause fibrosis and adhesions between the artery and vein. Subsequent punctures may lead to the formation of a fistula.

Not only is it the rarity that makes the diagnosis of iatrogenic AFV challenging, but also the nonspecific findings on physical exam, which mimic other medical conditions, exacerbate it. Other vascular malformations, such as hemangioma and AVM, or trauma, are the main differential diagnoses. In our patient, a palpable thrill and an audible bruit in the antecubital area were the two main signs associated with AVF. The presence of collateral veins in the upper thoracic regions is not a common presentation in brachio-brachial AVF/AVM and might have resulted from the obstruction of the ipsilateral subclavian vein opening. A detailed history and examination, besides imaging investigations including colour-coded Doppler ultrasound, CTA or magnetic resonance angiography (MRA), play a critical role in the diagnosis[9-11]. Doppler ultrasound, as a non-invasive method of assessment, is usually the first assessment showing multiple dilated tortuous vessels arising from the posterior auricular artery[12]. The preliminary diagnosis of the patient presented in our care report was AVM or hemangioma based on Doppler ultrasound findings. However, considering the history of frequent peripheral venous catheterization on the antecubital area, a possibility of AVF arose, and further investigations, including CTA, were performed, showing an abnormal brachio-brachial connection in favor of AVF (Figure 3). Although the challenges of performing either MRA or CTA, including but not limited to the need for sedation, and the exposure of the child to ionizing radiation in children should be weighed against its clinical benefits. Moreover, it is worth mentioning that intraoperative Doppler ultrasound helped surgeons to pinpoint the location of AFV before the surgery. Therefore, this modality of investigation has its unique role in the management of AVF. It should be taken into consideration that AFV-induced hypertrophy, especially in high-flow AFVs, can be preventable in case of receiving appropriate treatments in, timely manner. Although in AVMs, a longer follow-up time might be justifiable to allow the vascular malformation to grow to an operable volume.

There is no established standard of care for managing AVFs during childhood due to the scarcity of pediatric evidence. Therefore, most treatment options, including surgical ligation, trans-catheter embolization, ultrasound-guided compression, or exclusion with a covered stent, are recommended based on studies that were done on adults[6, 13]. In our case, we adopted vein ligation in addition to the primary repair of the brachial artery approach, which resulted in the complete resolution of patient signs and symptoms during postoperative follow-up visits within 6 months.

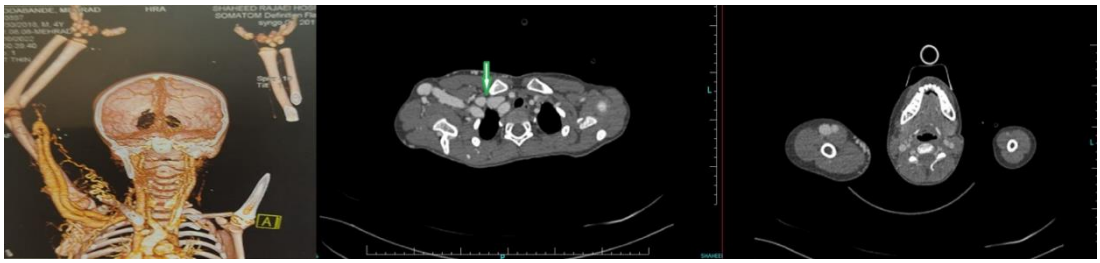

**Figure 3:** CTA of head, neck and right upper limb showing multiple serpiginous and vermiform arteries in the subcutaneous region of the right shoulder, right hemi-chest wall and right arm were noted. An abnormal AV connection between the brachial artery and vein, and also between the subclavian artery and vein, was detected. Moreover, a stenosis at the subclavian vein just before its connection to the internal jugular vein was recorded (green arrow).

## Conclusion

Iatrogenic AVF is an uncommon diagnosis during infancy due to its rarity. However, it should be considered in patients with a history of frequent hospital admissions, especially in common locations used for peripheral venous access. Therefore, taking a detailed history and reviewing all of the previous medical documents are essential when admitting patients with peripheral vascular abnormalities.

### Consent for Publication

Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

### Ethics Approval and Consent to Participate

The study protocol was approved by the Research Ethics Committee of Tehran University of Medical Sciences and a written informed consent form was obtained from the patient and his legal guardian.

### Authors' contributions

Study concept and design: J.S.; acquisition of data: A.Sh & A.S, drafting of the manuscript: A.Sh & A.S; critical revision of the manuscript for important intellectual content: A.S.; statistical analysis: Not applicable

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### Data Availability Statement

Data from the presented case report cannot be shared with other investigators based on the request of the child's legal guarantor.

### Conflicts of Interest

The authors report no conflicts of interest.

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