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Case Report

Surgical management of a nasal AVM in a pediatric patient: A case report

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ABSTRACT

Arteriovenous malformations (AVMs) are a type of high-flow vascular malformation that are characterized by abnormal capillary communications between the arterial and venous systems. While they are most commonly located in the head and neck region, their appearance in the nose is considerably rare, resulting in a paucity of literature regarding the surgical management of these lesions. We present the case of a 13-year-old male with a 6.5 × 6 cm AVM of the nose with a history of frequent nosebleeds since early childhood, often requiring aggressive measures, such as silver nitrate cauterization for control. Use of nasal decongestants and aminocaproic acid provided only transient improvement. After determination of arterial supply, AVM was approached with a combination of preoperative selective embolization and surgical excision with subsequent forehead flap defect coverage. Due to the size and complexity of this AVM, extra precautions were taken to avoid severe intraoperative bleeding, and femoral sheaths were placed prior to excision. The patient tolerated the procedure well, and with subsequent debulking surgery and Laser Hair Removal achieved an acceptable cosmetic outcome.

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Introduction

Arteriovenous malformations (AVMs) are a type of high-flow vascular malformation that are characterized by abnormal capillary communications between the arterial and venous systems. Although typically the result of a congenital malformation, in most cases, AVMs are not clinically relevant until later in life, when infection or trauma can lead to pain, bleeding or ulceration of the lesion.¹ They are diagnosed at a mean age of 19 years and show no sex-related associations.² Even though AVMs can arise in any type of tissue of the body, they are most commonly located in the head and neck region.³ Their appearance in the nose is considerably rare, resulting in a paucity of literature regarding the surgical management of these lesions.⁴ This study presents the workup and management of a nasal AVM in a pediatric patient.

Case report

A 13-year-old male presented with a 6.5 × 6 cm arteriovenous malformation of the nose, with glabella involvement and no intracranial extension (Figure 1, left). This lesion, present from birth and initially incorrectly diagnosed as a hemangioma or capillary malformation, was initially treated with multiple sessions of Pulsed Dye Laser, starting at age 2, with only slight improvement in color. In addition to the cosmetic disturbance, the patient experienced progressively worsening episodes of nosebleeds, lasting up to 60 minutes, resulting in multiple trips to the Emergency Department throughout childhood. In episodes where conservative measures were not sufficient to stop bleeding, silver nitrate cauterization was used. Nasal decongestants (oxymetazoline) and aminocaproic acid use provided only transient improvement. Quality of life was negatively impacted by the repeated episodes of epistaxis and associated systemic symptoms.

After evaluation at the Vascular Anomalies Center at The Children's Hospital Colorado, a diagnostic cerebral angiogram defined the lesion as an AVM involving the anterior nasal cavity, tip of the nose and nasal bridge with arterial supply from bilateral facial arteries and the right ophthalmic artery. In addition, a 5.3 mm flow-related aneurysm of the left ophthalmic artery involving the paranasal sinuses was found (Figure 2). It was decided that the aneurysm needed to be addressed prior to any further interventions concerning the AVM nidus, and a successful coil embolization was performed.

Thereafter, attention was directed toward the main nidus of the AVM, and surgical resection was planned. Forty-eight hours prior to resection, the patient underwent coil embolization of bilateral facial



Figure 1. Pre-operative appearance of nasal AVM. Left: gross look prior to any intervention. Right: post-embolization status.

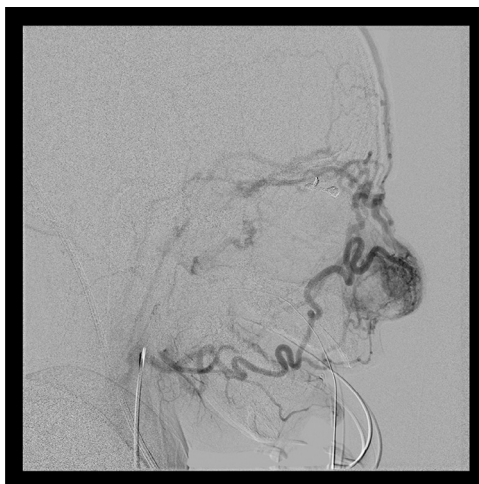


Figure 2. Diagnostic cerebral angiogram. Pre-operative planning for AVM resection.

arteries and Onyx 18 (Medtronic, Fridley, MN) embolization of left ophthalmic artery, resulting in apparent near-complete resolution of AVM opacification (Figure 1, right). However, during pre-incision preparation, a Doppler exam revealed turbid, tumultuous flow, with bruits and thrills throughout the lesion, despite embolization. Surgery was postponed until access to potential interventional radiology with embolization capabilities could be guaranteed, due to the high risk of severe intraoperative bleeding.

The patient was taken into the Hybrid OR again 3 days later, and bilateral femoral sheaths were placed by the Endovascular Neurosurgery service. Nasal endoscopy demonstrated no evidence of full thickness defect of the mucosa. The rhinectomy was designed as bilateral incisions from medial cheeks up to the glabella, sparing both alar subunits as well as the septum and nasal lining. Embolized vessels were resected as completely as possible, leaving underlying cartilage intact (Figure 3, left). This allowed reconstruction to be performed with a paramedian forehead flap without the additional need of a potentially anticipated fasciocutaneous radial free flap for lining. First, bilateral cheek advancement flaps were performed to place the cheeks along the pyriform aperture, correcting the bilateral cheek defects. This was accomplished by anchoring the undermined soft tissue to drill holes placed at the midpoint of the pyriform apertures, camouflaging the scar lines. Attention was then shifted to repairing the now 6.5×4.5 cm defect of the nose, and thus a forehead flap was designed, using the Doppler to identify the right supratrochlear and supraorbital arteries which would provide axial blood supply. Due to the patient's relatively short forehead, the flap did not extend into the hairline. The flap was raised in the subgaleal plane, rotated medially, and taken all the way down to the superior columella. After inset, the back wall of the pedicle was grafted with a 3×2 cm allograft to limit drainage (Community Tissue Services, Dayton, OH). Finally, the forehead donor site was extensively undermined and closed primarily (Figure 3, right). The flap was well perfused and was divided and inset three weeks later. Six sessions of 755 nm Alexandrite laser for hair reduction resulted in an acceptable cosmetic outcome (Figure 4). Postoperative angiography performed 3 months postoperatively revealed no evidence of recurrence.

Discussion

AVMs are vascular lesions in which there is a structural anomaly in the connection between arteries and veins. The lack of a proper capillary bed to disperse the high-pressure nature of arterial blood flow is thought to be one of the factors driving the progressive nature of AVMs. Even though



Figure 3. Intra-operative images showing simultaneous AVM resection and defect reconstruction. Left: defect after resection. Right: forehead flap prior to division.



Figure 4. Post-operative outcomes following forehead flap division and six sessions of Laser Hair Removal.

most are present from birth, they may not be symptomatic or grossly notable until undergoing a phase of exponential growth that may be linked to hormonal influence during puberty or pregnancy, or other insults such as trauma (including attempted resection) or infection, especially during childhood/adolescence.⁵ There is no consensus on the treatment of choice for AVMs, and some authors think, due to the high rate of recurrence, treatment should solely focus on selective embolization, even if only symptomatic control could be achieved.^{6,7} For others, including the authors of this paper, selective embolization followed by surgical excision with immediate reconstruction within 48–72 hours is the best way to provide long term control and acceptable aesthetic results. If necessary, a combination of flaps can be used for functional and cosmetic goals.^{4,5,8–10}

Conflict of interest

The authors have no conflicts to disclose.

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