Characteristics and Outcomes of Pediatric Dural Arteriovenous Fistulas: A Systematic Review

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Abstract

Background

Dural arteriovenous fistulas (dAVF) are arteriovenous shunts in communication with the dural vasculature in the brain or spine. Apart from single-center series, risk factors and treatment outcomes for pediatric dAVFs are largely undescribed.

Methods

We performed a systematic literature review of pediatric (< 18 years at diagnosis) intracranial and spinal dAVF according to PRISMA guidelines. We queried PubMed, CINAHL, SCOPUS, and Embase databases without time/date restriction. Search strings included a variety of MeSH keywords relating to dural AV fistulas in combination with MeSH keywords related to pediatric cases (see Appendix). Manuscripts describing patients diagnosed with dural sinus malformations or pial AVF were excluded.

Results

We identified 61 studies describing 69 individual patients. Overall, dAVF were more common in males (55.1%) with a mean age of diagnosis (5.17 ± 4.42 years). Approximately 20.2% of patients presented with cardiovascular disease (CVD), and 31.9% were discovered incidentally on neuroimaging studies. Transverse-sigmoid junction was the most common location (17.3%). Ninety-three percent (64 patients) were treated, most commonly using endovascular embolization (68.1%) followed by surgery (8.7%) and radiosurgery (2.9%). Almost half (43.8%) of dAVFs were completely obliterated. Of the 64 procedures, there were 19 neurological complications (29.7%) of varying severity where 12.5% were considered transient (i.e., pseudomeningocele) and 17.2% permanent (i.e., mortality secondary to acute sinus thrombosis, etc.).

Conclusion

There is a paucity of information on pediatric dAVFs. This systematic review summarizes the published cases of dAVFs in the pediatric population. While the rate of missing data is high, there is publication bias, and precise details regarding complications are difficult to ascertain, this review serves as a descriptive summary of pediatric dAVFs.

Introduction

Dural arteriovenous fistulas (dAVF) are arteriovenous shunts between meningeal arteries and veins or dural venous sinuses that account for ~ 10% of all intracranial vascular malformations.[1,2] The first

cranial dAVF was described as an AVM involving the dura mater by Rizzoli in 1873; however, the incidence and prevalence of dAVF in pediatric patients is not known, making epidemiologic estimates in children difficult.[3] Similarly, the natural history of pediatric dAVFs has not been described. Our understanding of pediatric dAVF has been limited to single-center reports and through application of adult studies, treatment strategies, and outcomes to pediatric patients, with a few exceptions. Common presenting symptoms include congestive heart failure, cyanosis, cranial bruits, and facial vein prominence in neonates while older children more often present with sequalae from intracranial hypertension, which include headaches, focal neurological deficits, syncope, seizures, and tonsillar herniation.[3,4] In both instances, expeditious diagnosis and treatment is critical to prevent irreversible brain injury.[5,6] The etiology in many instances is unclear, but trauma, prior intracranial surgery/radiation, intracranial infection, venous stenosis, or sinus thrombosis have been hypothesized as etiologic substrates.[2,7] Classic clinical signs such as tinnitus, are not always possible to ascertain in young children. While substantially large dAVF in neonates may present with congestive heart failure in severe cases, many lesions are incidentally discovered.

Classification systems for adult dAVFs have been developed based upon the pattern of venous flow.[8,9] The two most commonly used classification systems were developed by Borden and Cognard in adult patients.[9] Table 1 details the characteristics for each of these classification systems. It is important to note that these classification systems have not undergone validity or reliability testing in the pediatric population.[9,10] Nonetheless, risk stratification and understanding the natural history of pediatric dAVF is essential, given the potentially catastrophic consequences of hemorrhage.

Table 1
– Summary of Borden and Cognard Classification.[3,10]

Borden Classification	Cognard Classification
Type I: Drainage into dural venous sinus or meningeal veins only	Type I: Drainage into dural venous sinus only, with normal anterograde flow
Type II: Drainage into dural venous sinus or meningeal vein + cortical venous reflux	Type IIa: Drainage into dural venous sinus only, with retrograde flow
Type III: Cortical venous reflux only	Type IIb: Drainage into dural venous sinus with anterograde flow + Cortical venous reflux
	Type IIa + b: Drainage into dural venous sinus with retrograde flow + Cortical venous reflux
	Type III: Cortical venous reflux only without venous ectasia
	Type IV: Cortical venous reflux with venous ectasia
	Type V: Drainage into spiral perimedullary veins

Risk of sequalae such as cerebral hemorrhage, venous infarction, and cardiovascular disease depend upon largely on venous drainage patterns, among other features.[9,10] Some patients may also present with imaging (CT/MRI) suggestive of Chiari malformation or a brain mass/tumor.[2,9,10] However, catheter angiography is required to make a definitive diagnosis. Initial management generally involves endovascular approaches; however, some locations (ethmoidal/tentorial dAVFs) are potentially more suitable for open surgical approach.[3,11] In addition, in recent years, stereotactic radiosurgery has been reported in patients with low-risk dAVFs without venous reflux.[3,12,13] This systematic review aims to summarize the published cases of dAVFs in the pediatric population and describe patient and fistula characteristics, treatment modality, treatment success, and complication rates.

Methods

Our systematic review adhered to the PRISMA guidelines,[14] and included articles from PubMed, CINAHL, SCOPUS, and Embase databases without time/date restriction. Search strings included a variety of MeSH keywords relating to dural AV fistulas in combination with MeSH keywords related to pediatric cases (see Appendix). Google Scholar was used to identify references in review articles identified by our initial search. Search results were screened and reviewed sequentially by title, abstract, and, finally, full content by two independent reviewers (P.D.M., C.S.) with any discrepancies reviewed by a third author (A.T.H.). We included only manuscripts describing (1) pediatric patients less than 18 years of age, (2) studies where outcomes, treatments, and factors could be identified in pediatric patients (if adult patients also included in the study), (3) studies that had a diagnosis of dAVF on cerebral angiogram, (4) and studies that were original research articles. The type of surgical intervention and underlying etiology of dAVF were not exclusionary criteria applied for study selection. Review articles, meta-analyses, case reports, editorials, non-human studies, conference papers, and abstracts without companion full text were excluded. Articles without English full-text translations or without details specific to pediatric patients were also excluded. Data fields included author, publication year, journal of publication, country, study design, sample size, and study limitations. The variables collected include PMID, year of publication, sex, mean age, presence or absence of cardiovascular disease and/or venous ectasia, anatomic location of dAVF, Borden/Cognard type, treatment administered, embolic agent used, treatment success rate, and details and rate of complications. Statistical analyses were largely descriptive in nature. The mode for each category was collected to find the most common correlations between the data collected.

Results

Our initial search criteria yielded 1072 results (435 in PubMed, 84 CINAHL, 87 SCOPUS, 241 Embase) prior to any screening. Initial screening of our search results identified 732 unique studies. Further review of abstracts left 121 studies remaining which met inclusion criteria. Full text analysis excluded a further 60 studies, leaving 61 total studies which met criteria for final inclusion. The literature review and study selection process are summarized in Fig. 1. Table 2 lists the characteristics and data available from all included studies.

Table 2
– Summary of included studies in this systematic review. Factors reported: a) age b) sex c) cardiovascular disease d) venous ectasia e) location of dAVF f) treatment g) embolic agent h) treatment success i) complication.

Study	Study Type	Number of Participants	Factors Reported
Kuo 1995[16]	Case Study	1	a, b, c, d, e, f, g, h, i
Kincaid 2001[5]	Case Series	7	a, b, c, d, e, f, g, h, i
Fernández 2008[17]	Case Study	1	a, b, c, d, e, f, g, h, i
Crowley 2009[18]	Case Study	1	a, b, c, d, e, f, g, h, i
Vassillyadi 2009[19]	Case Study	1	a, b, c, d, e, f, g, h, i
Reig 2010[20]	Case Study	1	a, b, c, d, f, g, h, i
Solarte 2010[21]	Case Study	1	a, b, c, d, e, f, g, h, i
Teranishi 2010[22]	Case Study	1	a, b, c, d, e, f, g, h, i
Ventura 2010[23]	Case Study	1	a, b, c, d, e, f, g, h, i
Abla 2011[24]	Case Study	1	a, b, c, d, e, f, g, h, i
Karanam 2011[25]	Case Study	1	a, b, c, d, e, f, g, h, i
Cohen 2013[26]	Case Study	1	a, b, c, d, e, f, g, h, i
Leu 2015[27]	Case Study	1	a, b, c, d, e, f, g, h, i
Zaidi 2015[28]	Case Series	11	a, b, c, d, e, g, h, i
Hayward 2016[29]	Case Study	1	a, b, c, d, e, f, g, h, i
Oshiro 2016[30]	Case Study	1	a, b, c, d, e, f, g, h, i
Hetts 2016[31]	Case Series	22	a, b, c, d, e, f, g, h, i
Brinjikji 2017[15]	Case Study	1	a, b, c, d, e, f, g, h, i
Soni 2017[32]	Case Study	1	a, b, c, d, e, f, g, h, i
Tomycz 2017[33]	Case Study	1	a, b, c, d, e, f, g, h, i
Koutsouras 2018[34]	Case Study	1	a, b, c, d, e, f, g, h, i
Agnoletto 2019[35]	Case Study	1	a, b, c, d, e, f, g, h, i

Study	Study Type	Number of Participants	Factors Reported
Gatto 2019[36]	Case Study	1	a, b, c, d, e, f, g, h, i
Guo 2019[37]	Case Study	1	a, b, c, d, e, f, g, h, i
Koduri 2019[38]	Case Study	1	a, b, c, d, e, f, g, h, i
Rajadurai 2020[39]	Case Study	1	a, b, c, d, e, f, g, h, i
Jordan 2021[40]	Case Study	1	a, b, c, d, e, f, g, h, i
Niimi 2021[41]	Case Study	1	a, b, c, d, e, f, g, h, i
Saenz 2021[42]	Case Study	1	a, b, c, d, e, f, g, h, i
Yadav 2021[43]	Case Study	1	a, b, c, d, e, g, h, i
Chu 2022[6]	Case Study	1	a, b, c, d, e, f, g, h, i
Roth 2022[44]	Case Study	1	a, b, c, d, e, f, g, h, i

Table 3
– Patient demographics and fistula characteristics Abbreviations include SD – Standard Deviation, CVD – Cardiovascular Disease, SSS – Superior Sagittal Sinus, MCF - Middle Cranial Fossa.

Variable	Value
Total no. of pts	69 (100%)
Male sex	38 (55%)
Mean age (SD), years	5.17 (4.42)
CVD	
Yes	14 (20%)
No	55 (80%)
Venous ectasia	
Yes	22 (32%)
No	47 (68%)
Location	
Transverse-sigmoid	12 (17%)
Tentorial	1 (1%)
Cavernous sinus	1 (1%)
Convexity/SSS	4 (8%)
Anterior cranial fossa	5 (7%)
Torcular	6 (9%)
Sylvian/MCF	4 (8%)
Other	13 (19%)
Missing	23 (33%)

Table 4
– Modalities used for patients undergoing treatment for pediatric dAVF.

Variable	Value
Total Number of Patients	69 (100%)
Untreated	1 (1%)
Treated	64 (93%)
Embolization alone	47 (68%)
Surgery alone	6 (9%)
Radiosurgery alone	2 (3%)
Total Multimodality therapy	
Embolization + Surgery	9 (13%)
Embolization + Radiosurgery	0 (0%)
Embolization + Surgery + Radiosurgery	0 (0%)
Missing	4 (6%)
Total	
Total Embolization	56 (81%)
Total Surgery	6 (9%)
Total Radiosurgery	2 (3%)

In total, 69 pediatric patients diagnosed with a dAVF were included. The majority of patients were male (55%). Mean age was 5.2 with a standard deviation of 4.4 years. Twenty percent of patients presented with cardiovascular disease (CVD), while 31% presented with venous ectasia. The most common location for dAVF was in the transverse-sigmoid junction at 17% (12 patients). We summarize patient and fistula characteristics in Table 2.

Overall, 64 patients (93%) underwent treatment with either endovascular techniques, open surgery, radiotherapy, or some combination thereof. Forty-seven patients (68%) were treated with embolization alone, 6 patients (9%) were treated with surgery alone, and 2 patients (3%) were treated with radiosurgery alone. Furthermore, 9 patients (13%) were treated using a combination of embolization and open surgical approaches. Treatment modality is further summarized in Table 2. The most common embolic agents used were liquid embolics (ethanol, N-butyl cyanoacrylate (NBCA) glue or EVOH) and liquid embolic agent plus coils at 12 uses (18%) each respectively. Itemized approach to embolization is summarized in Table 5. Of the 64 patients who underwent treatment, 44 patients (69%) had posttreatment cure and 11 patients (17%) had posttreatment residual lesion. Furthermore, 79% of the 56 embolizations has

posttreatment cure and 20% had posttreatment residual. One of the 6 surgical patients had posttreatment cure while the outcome of the remaining 5 patients was not available. Similarly, outcomes in the radiosurgery group were not available. These data are summarized in Table 6.

Table 5
– Embolic agent used for patients who underwent embolization for treatment of dAVF.

Variable	Value
Total	68 (100%)
Liquid Embolic agent	12 (18%)
Coils	8 (12%)
Glue	6 (9%)
Liquid Embolic agent + Coils	12 (17%)
Liquid Embolic agent + Glue	1 (1%)
Coils + Glue	2 (3%)
Missing	27 (40%)

Table 6
– Treatment success based upon treated patients

Variable	Value
Embolization	
Total Embolization	56 embolizations
Posttreatment Cure	44 (79%)
Posttreatment Residual	11 (20%)
Missing	1 (1%)
Surgery	
Total Surgery	6 surgeries
Posttreatment Cure	1 (17%)
Posttreatment Residual	0 (0%)
Missing	5 (83%)
Radiosurgery	
Total Radiosurgeries	2 radiosurgeries
Posttreatment Cure	0 (0%)
Posttreatment Residual	0 (0%)
Missing	2 (100%)
Total	
Total Procedures	64 procedures
Posttreatment Cure	44 (69%)
Posttreatment Residual	11 (17%)
Missing	9 (14%)

Complications were stratified by treatment modality as follows. Out of 56 embolizations, 12 patients (21%) of patients experienced a complication. One was transient while the other 11 were permanent in nature. Of the 6 surgical procedures, 5 patients (83%) experienced a temporary complication that later resolved. Of the 2 radiosurgeries, both patients experienced a temporary complication that subsequently resolved. Of all 64 procedures, there were 19 complications (30%). None of these complications were technical in nature, 8 (42%) were temporary and 11 (58%) were permanent. Temporary complications included pseudomeningocele, access-site femoral artery stenosis following embolization, postoperative infection following craniotomy, catheter-induced vasospasm, and nontarget coil embolization to the

lungs with subsequent endovascular coil recovery. Permanent complications included death secondary to acute sinus thrombosis, epidural hematoma, and cerebral malformation developed secondary to venous congestion from residual lesion. A summary of the complications is detailed in Table 7.

Table 7
– Complications based upon treated patients categorized by treatment modality.

Variable	Value
Embolization	
Total Embolization	56 (100%)
Total Complications	12 (21%)
Technical	0 (0%)
Temporary	1 (2%)
Permanent	11 (20%)
Surgery	
Total Surgery	6 surgeries
Total Complications	5 (83%)
Technical	0 (0%)
Temporary	5 (83%)
Permanent	0 (0%)
Radiosurgery	
Total Radiosurgeries	2 radiosurgeries
Total Complications	2 (100%)
Technical	0 (0%)
Temporary	2 (100%)
Permanent	0 (0%)
Total	
Total Procedures	64 procedures
Total Complications	19 (30%)
Technical	0 (0%)
Temporary	8 (13%)
Permanent	11 (17%)

Discussion

This systematic review summarizes the published literature of dAVFs in the pediatric population. We report patient and fistula characteristics, treatment modality, posttreatment cure rate, and complications. Most reports included both adult and pediatric patients. Due to variability in reporting, there is also a significant amount of missing data. However, general trends could be observed. There is a slight male predominance, age at presentation varies considerably, venous ectasia was the most common presentation, and nearly 20% of lesions involved the transverse-sigmoid junction. There is likely a significant publication bias in reported cases, and characteristics of patients treated with observation were not included. There was also a considerable rate of both transient and permanent complications, although precise details about each complication are not immediately obvious in the literature. Future prospective studies are needed to measure complications and compare against the natural history of pediatric dAVFs.

Most patients (93%) were treated using endovascular techniques, open surgery, radiosurgery, or a combination thereof. Embolization was the most common treatment modality at 68%, followed by surgery (9%), and radiosurgery (3%). Liquid embolic agents alone and in combination with coils were the most common agents used at 18% respectively. Individual patient characteristics and anatomy likely dictated preferred approach; however, additional granularity could not be collected from reported articles. The most common treatment result was dAVF posttreatment cure at a rate of 69%. The true extent and reasoning for the high complication rate was difficult to discern from published literature; however, some cases discuss complexity of the dAVF and significant venous congestion, technical difficulties related to small patient size, and lack of established approaches due to the rarity of the condition.[15]

Limitations to our study include a small number of included articles secondary to rarity of dAVFs, publication bias in setting of systematic review, and inability to assess patients who were not treated. To our knowledge, there is no rigorous natural history study of pediatric dAVF available. Individual lesional anatomy could also not be delineated owing to the nature of our study design. Future, prospective registry-based studies are needed to understand the natural history and more granular features of dAVF in the pediatric population.

Pediatric dAVFs are uncommon vascular pathologies that can be treated using endovascular embolization, surgical disconnection, radiotherapy, a combination thereof, or managed conservatively. There is a paucity of information regarding dAVFs in the pediatric population. Thus, there are no guidelines or standardized treatment recommendations for who, when, and how to intervene. This systematic review summarizes the published cases of dAVFs in the pediatric population and decipher patient and fistula characteristics, treatment modality, embolic agents used, treatment success, and complication rates based upon treatment modality. While this study is descriptive, it offers a summary of our current understanding of pediatric dAVFs. Additional prospective, multi-center studies are needed to increase our understanding of this disease.

Conclusions

There is a paucity of information on pediatric dAVFs. This systematic review summarizes the published cases of dAVFs in the pediatric population. While the rate of missing data is high, there is publication bias, and precise details regarding complications are difficult to ascertain, this review serves as a descriptive summary of pediatric dAVFs.

Abbreviations

dAVF – Dural arteriovenous fistulas

AVM – Arteriovenous malformation

CT – Computed Tomography

MRI – Magnetic Resonance Imaging

CVD – Cardiovascular disease

AVM – Arteriovenous malformation

PRISMA - Preferred Reporting Items for Systematic Reviews and Meta-Analyses

NBCA - N-butyl cyanoacrylate

EVOH - Ethylene Vinyl Alcohol Copolymer

Declarations

Ethics approval and consent to participate

Approved by all participating parties

Consent for publication

Not applicable

Availability of data and material

Datasets and search terms for this systematic review can be found in the appendix below

Competing interests

Not applicable

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Authors' contributions

All authors contributed to the study conception and design. Material preparation, data collection and analysis were performed by Pedram D. Maleknia and Andrew T. Hale. The first draft of the manuscript was written by Pedram D. Maleknia, and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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Figures

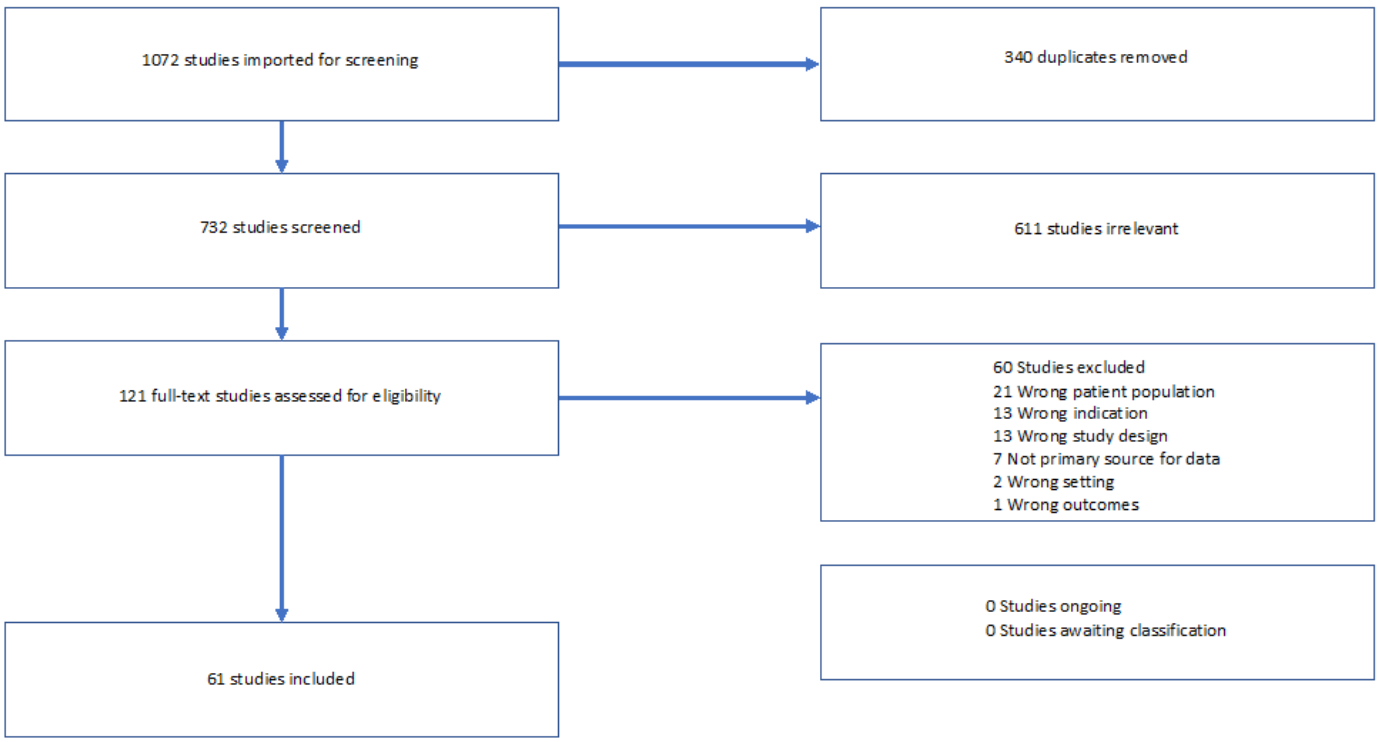

Figure 1

Flow diagram demonstrating meta-analysis inclusion

Supplementary Files

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