The Egyptian Journal of Neurology, Psychiatry and Neurosurgery

RESEARCH Open Access

Endovascular treatment of vein of Galen aneurysmal malformation: hospital-based case series in two tertiary centers

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Abstract

Background Vein of Galen aneurysmal malformation (VGAM) is considered a rare vascular malformation, constituting over 1% of all arteriovenous shunts. This malformation represents around 30% of congenital vascular malformations in the pediatric population. The focus of our study is to examine both the clinical and radiological outcomes following endovascular embolization in pediatric patients with Galenic arteriovenous shunts. Our aim is to evaluate the safety and efficacy of endovascular management. This research aims to contribute to understanding the management and outcomes of VGAM in pediatric patients, particularly in the context of endovascular embolization.

Method This study included 19 patients with vein of Galen malformation evaluated clinically and radiologically and we treated them with endovascular embolization with aim to assess safety and efficacy of endovascular treatment of VGA in the period from May 2019 to December 2022.

Results We evaluated 12 male patients 63.2% and 7 females 36.8% in pediatric age group with vein of Galen malformation. Patients were followed for 1 year. Our study included 3 neonates (15.8%), 10 infants (52.6%) and 6 children (2–10 years) (31.6%). Increase SC (skull circumference) was the most common presenting symptoms 8 (42.1%). We have reached total occlusion in 15 patients (78.9%) while 4 patients had residual (21.1%). 11 patients improved (57.9%) and 2 patients died (10.5%).

Conclusion VGAM are not very rare and should be kept in mind in children with refractory heart failure. Introduction of endovascular embolization as the primary therapy has significantly improved prognosis. Good selection of cases based on their score and good timing of treatment has impact on prognosis with less morbidities.

Keywords Vein of Galen, Pediatric, Embolization

Background

Vein of Galen malformation is rare congenital intracranial vascular malformation. It is believed that this condition develops in intrauterine life in the period between 6 and 11 weeks of gestation. Primitive choroidal arteries and the median prosencephalic vein are connected leading to different types based on angiographic pattern [1].

There are two widely used classifications done by Yasargil and Lasjaunias.

In 1988, Yasargil classified VGAMs into four categories:

- Type I: pure fistula with a direct arteriovenous shunt established between the arterial feeders and the collecting vein which is usually seen in the neonates with cardiorespiratory failure; the arterial feeders are typically choroidal branches of the pericallosal and posterior cerebral arteries.

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- Type II: an arteriovenous malformation (AVM) of transmesencephalic and transdiencephalic thalamoperforator arteries with branches to normal brain tissue. With its transmesencephalic thalamoperforators feeding a complex arterio-arterial network interposed between the feeding arteries and the venous collector, is usually associated with a late presentation.
- Type III: mixed type I and type II. Seen in the intermediate group of patients with delayed presentation.
- Type IV: corresponds to proximal AVMs that have draining veins that drain into the vein of Galen [2].

Lylyk P. et al. stressed that in type IV, arteriovenous shunting is intraparenchymal and not in the wall of the varix, in contrary to other types [3].

This classification was simplified by Berenstein and Lajaunias in 1987 who proposed separating malformations into mural and choroidal types based on their angioarchitecture and depending on the location of the fistula [4].

The mural type (39%) has a small number of direct arterial connections into the wall of the median prosencephalic vein of Markowski. Mural VGAMs are often supplied by a few feeders, including callosal and posterior choroidal arteries, and supply is usually unilateral [5].

Clinically, the mural types of VGAMs usually present in infants with macrocrania or developmental delay and may be associated with mild cardiac failure or asymptomatic cardiomegaly [6].

The mural type is much better tolerated than the choroidal type, and therefore is found in infants who do not experience severe cardiac symptoms and have better clinical prognoses [7].

The choroidal type (61%) has multiple choroidal feeders including choroidal, thalamoperforating and pericallosal arteries that form a nidal network that drains into the median prosencephalic vein of Markowski. The choroidal type is usually characterized by much higher flow and tends to present early with cardiac failure [8].

Based on the experience of the research group of Lasjaunias with over 300 patients with VGAM they introduce a scoring system named Bicètre neonatal evaluation score [9].

A 21-point scale factors in cardiac, cerebral, hepatic, respiratory, and renal function:

- Scores below 8 typically signify that the patient is in a bad clinical condition to an extent that does not allow treatment.
- Scores ranging from 8 to 12 indicate a need for urgent endovascular management.
- Scores exceeding 12 point to a robustly healthy newborn, prompting efforts to delay endovascular procedures through medical means.

It is important to note that initial numerical scores, assessed during the early days of life, may fluctuate based on the response to treatment over subsequent days. Reflecting on endovascular treatments for newborns with VGAMs, Lasjaunias et al. deduced that the prime timing for angiography and treatment, optimizing embolization effectiveness while minimizing the risk of cerebral maturation delay, is around 5 months [10].

Methods

This is a prospective hospital-based case series study carried out through the period from May 2019 to December 2022 conducted in 2 tertiary centers (endovascular unit in Assiut university hospital and endovascular unit in Ain Shams university).

The current study included 19 patients treated by endovascular embolization for VGM in pediatric group.

All patients underwent a complete admission history, physical examination, and standard screening laboratory work. Admitting data collection included calculation of Glasgow Coma Scale (GCS) score. All patients underwent diagnostic CT angiography or cerebral angiography before endovascular intervention, unless they had been transferred from another hospital and already had adequate studies.

All patients were admitted to hospital before the procedure and operated within 48 h of admission.

All procedures were done under general anesthesia for all patients.

The principal approaches to treatment involved attempts to eliminate the high flow through the vascular lesions by arterial embolization with a liquid adhesive agent.

Endovascular approach

All patients in our study were treated via transarterial approach and we used liquid acrylic agents.

Our option for evaluation of the newborn with VGAM is based on the Bicètre score to determine potential treatment options.

First the puncture and catheterization of the femoral artery was performed; 4F sheath catheter under constant X-ray control was placed and total selective angiography with diagnostic 4–5F catheter was performed. Internal carotid artery with its branches was studied first and then vertebrobasilar arteries were studied. After differentiation of afferent vessels of the arteriovenous (AV) shunt, the catheterization was performed directly within their confluence to the vein of Galen.

We used Histoacryl (n-butyl-2 cyanoacrylate [NBCA]) for embolization in all cases, polymerization time could be modified by the addition of oil-based contrast agents like Lipiodol with tantalum powder.

Flushing of the microcatheter was done with glucose 5% before injection.

All patients would be followed up clinically and radiologically immediate postoperative and one year postoperative from the last session of embolization for those patients who needed multiple sessions.

The outcome was assessed by the neurological and developmental findings over a scale from 4 to zero grade 4 representing patients with normal development, grade 3 represent mild level of impairment while grade 2 represent children having intermediate level of impairment affecting daily achievements but in low need for medications and grade 1 representing patients having a severe form of neurological deficits requiring aggressive care and medical treatment and finally grade 0 reflecting died cases [11].

As shown in Figs. 1, 2, 3, 4, 5, 6.

Results

Demographic data of our patients showed 7 females (36.8%) and 12 males (63.2%) with mean ± SD 1.81 ± 1.1 and median 1.2 (0.1–7).

We had 3 neonates (15.8%), 10 infants (52.6%) and 6 children (31.6%) as shown in Table 1.

The most common presenting symptoms in our study is increase in skull circumference (42.1%) followed by congestive heart failure (CHF) and seizures by (15.8%). Developmental delay (DD) and proptosis were presented in two cases for each (10.5%) and finally one case presented melting brain syndrome (5.3%) as shown in Table 2.

We had 13 cases of mural type and 6 cases of the choroidal type. We could reach total occlusion in 15 patients (78.9%) and only 4 patients had residual after occlusion (21.1%).

From the 15 cases who reached total occlusion, we accomplished that in one setting in 12 patients and only 3 patients needed further sessions.

All the patients who had total occlusion in one setting were of mural type and only one patient with mural type needed 2 sessions. We reached total occlusion in 2 patients with choroidal type through 7 sessions.

We had improvement in 11 patients (57.9%) in our study, while 4 patients discharged on score 3 (21%), and two patients discharged on score 2. Two patients died during the embolization procedure, one from glue embolization to the lung and the other had intraprocedural arterial perforation and were of choroidal type as shown in Tables 3, 4.

Discussion

VGAM, constituting less than 1% of all brain vascular malformations, is indeed a rare brain arteriovenous shunt in the pediatric population [1].

During the neonatal period, a significant portion of brain arteriovenous shunts (BAVS) present as VGAM, with many cases being associated with congestive heart failure or hydrodynamic disorders [12, 13].

An important meta-analysis on VGAM embolization, encompassing 35 studies with 307 participants, evaluated the outcomes post-endovascular embolization for VGAM. The follow-up duration averaged around 42 months, with the analysis reflecting a distribution of 42% neonates (less than 1 month), 45% infants (1 month to 2 years), and 13% children (above 2 years) among the participants [14].

The findings from this additional meta-analysis are equally insightful. This study incorporated 27 series with a larger cohort of 578 patients, emphasizing the wide age distribution within the VGAM patient population. The median age of patients was notably low at 0.1 month. Among the patients with available age data (547 patients), the distribution was as follows: 229 neonates (41.9%), 246 infants (45.0%), and 72 children (13.2%).

Furthermore, sex data, available for 252 patients, showcased a notable male predominance, with 173 patients (68.7%) being male. These statistics further emphasize the prevalence of VGAM in early infancy and the male predominance in this specific patient group. Understanding these demographics is crucial in tailoring treatments and management strategies for VGAM patients [15].

The study conducted by Rajeev et al. offers a comprehensive view of 26 patients with VGAM. Among these patients, there were 17 males (65.3%) and 9 females (33.3%), indicating a slightly higher male predominance. The age range spanned from 1 day to 18 years, encompassing a diverse group of patients.

The age distribution within this cohort was as follows: one neonate (3.7%), 16 in the infantile age group (59.2%), eight children (29.6%), and one adult (more than 16 years) (3.7%). This study highlights the occurrence of VGAM across a wide age spectrum, emphasizing its presence not just in infants and children, but also in adults, albeit less frequently. Understanding its occurrence across age groups aids in developing nuanced treatment approaches and care strategies for patients with VGAM [16].

Kartik et al., in their study over 48 patients, 31patients were boys [64.6%] and 17 patients were girls [35.4%] [17].

In our study, we had 19 patients with VGAM 3 patients of them were neonates (15.8%) and 10 patients were infants (52.6%) while 6 patients were in early childhood (31.6%) with no patients presented with age above 10 years.

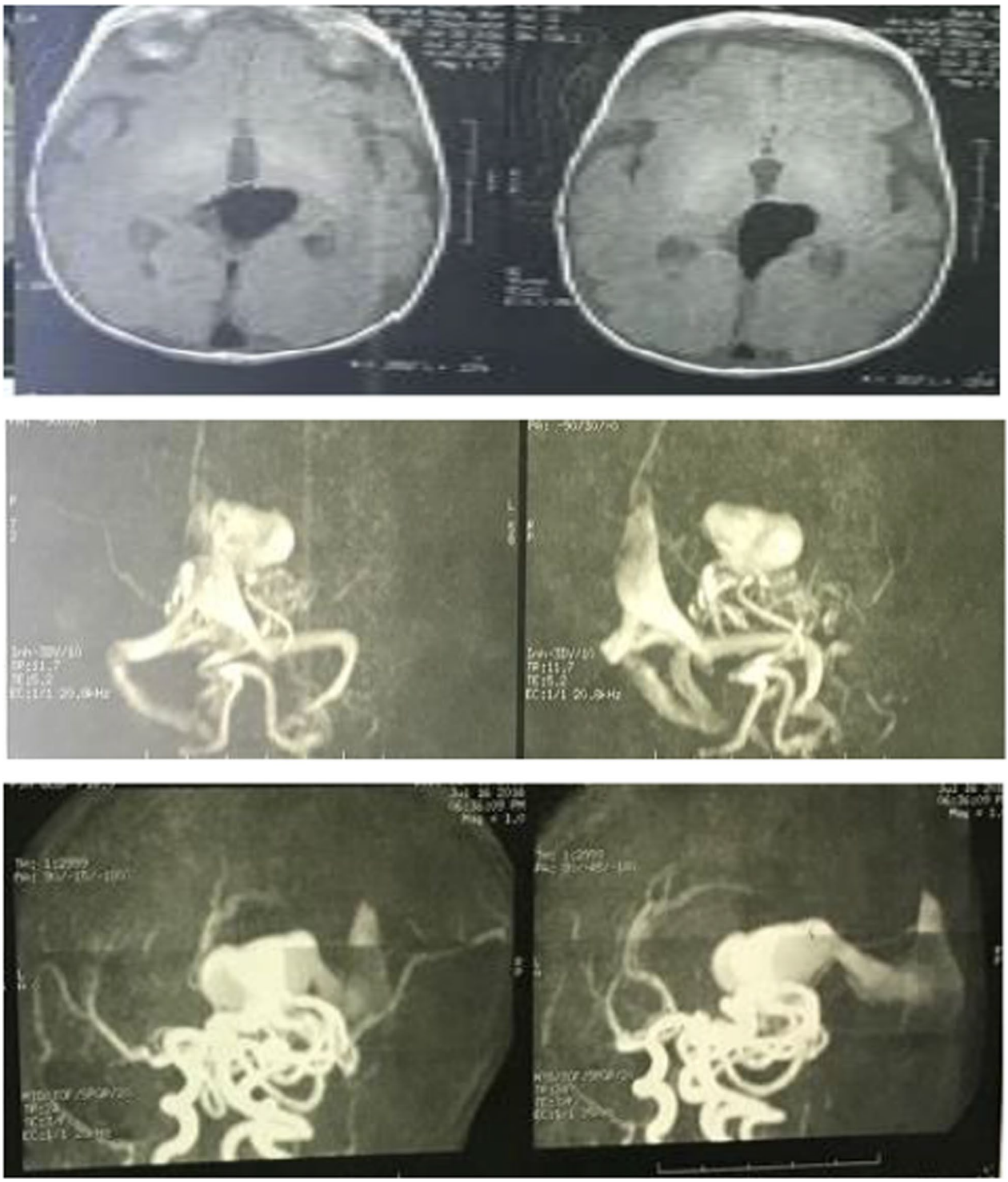

Fig. 1 MRI brain with MRA and MRV showing mural type VGAM

Regarding sex distribution, 12 patients were males (63.2%) while 7 patients were females (36.8%) from our 19 patients with VGAM.

This is in line with the previous studies that VGAM is mainly presented in neonatal and infantile pediatric age group except that neonatal age group is much more than

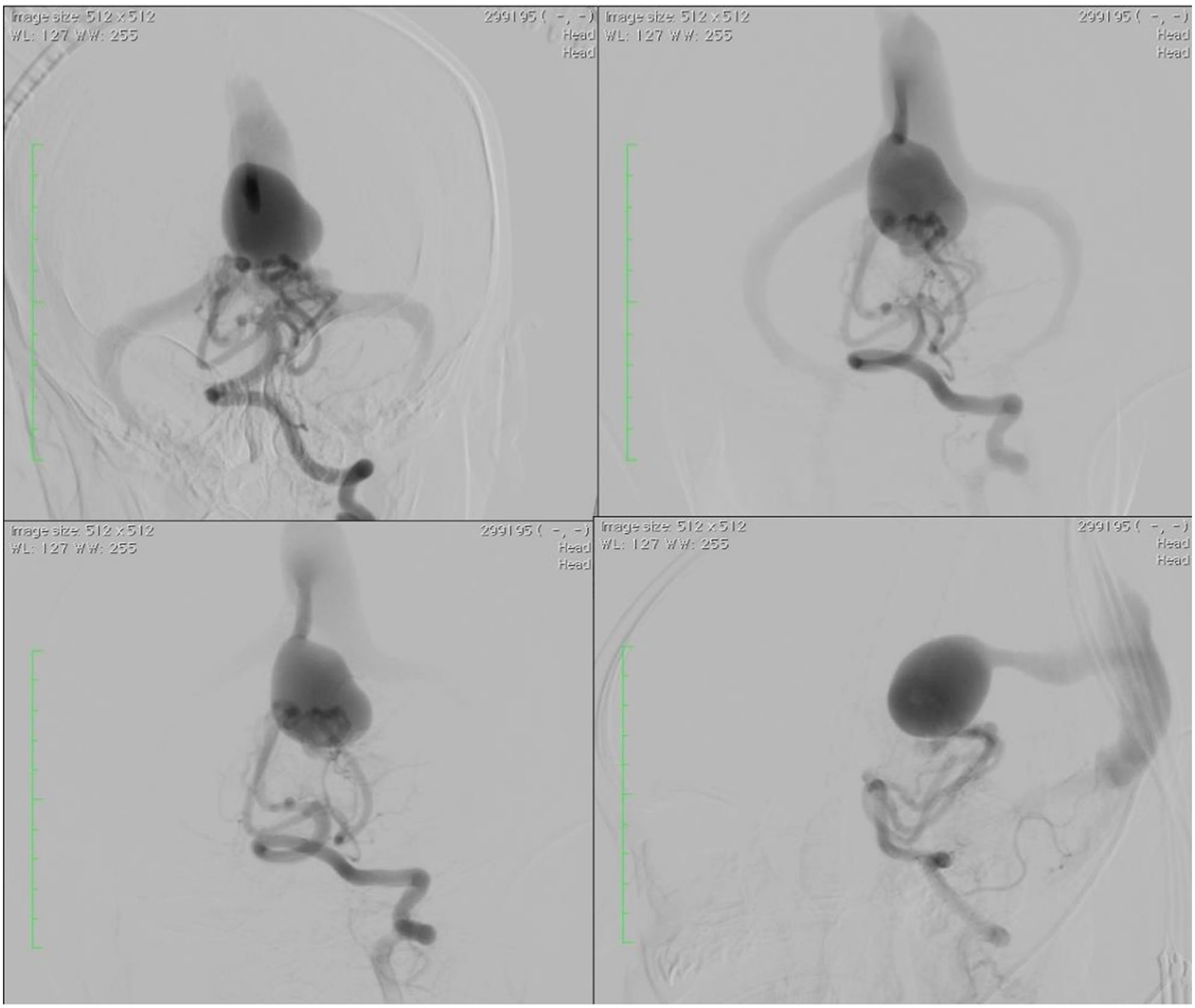

Fig. 2 Intraoperative angiographic picture of the VGAM

infantile one in Hale et al., and Brinjikji et al., studies, while in Rajeev et al. study it was as our study.

All the studies demonstrated that male sex is more predominant than females in cases of VGM which was also demonstrated in our study.

The studies by Rajeev et al. and Brinjikji et al. shed light on distinct clinical presentations of VGAM across different age groups.

Rajeev et al.'s study revealed that among neonates, congestive heart failure (CHF) was the sole presenting symptom. In the infantile age group, the most common presentation was macrocrania in 11 out of 16 patients (68.75%), followed by developmental delay in 5 out of 16 patients (31.25%) 0.16.

Similarly, Brinjikji et al. found that in neonates, CHF was the most frequent presentation, observed in 88.2% of cases. In contrast, in infants and children, increased head circumference was more prevalent, occurring in 53.3% and 37.5% of cases, respectively. Understanding these age-specific clinical manifestations is crucial in diagnosing and managing VGAM in pediatric patients, allowing for tailored treatment approaches based on the presenting symptoms in different age groups [15].

The meta-analysis conducted by Jun Yan et al. revealed that within the 34 studies evaluated, neonates represented 44% of the sample. The predominant presentation in this neonatal group was congestive heart failure (CHF). This aligns with previous findings emphasizing CHF as a primary presentation among neonates with VGAM [18].

Additionally, Vignesh et al.'s study focusing on 31 patients older than one month highlighted that macrocrania is the most common presentation among the infantile age group. These consistent findings across

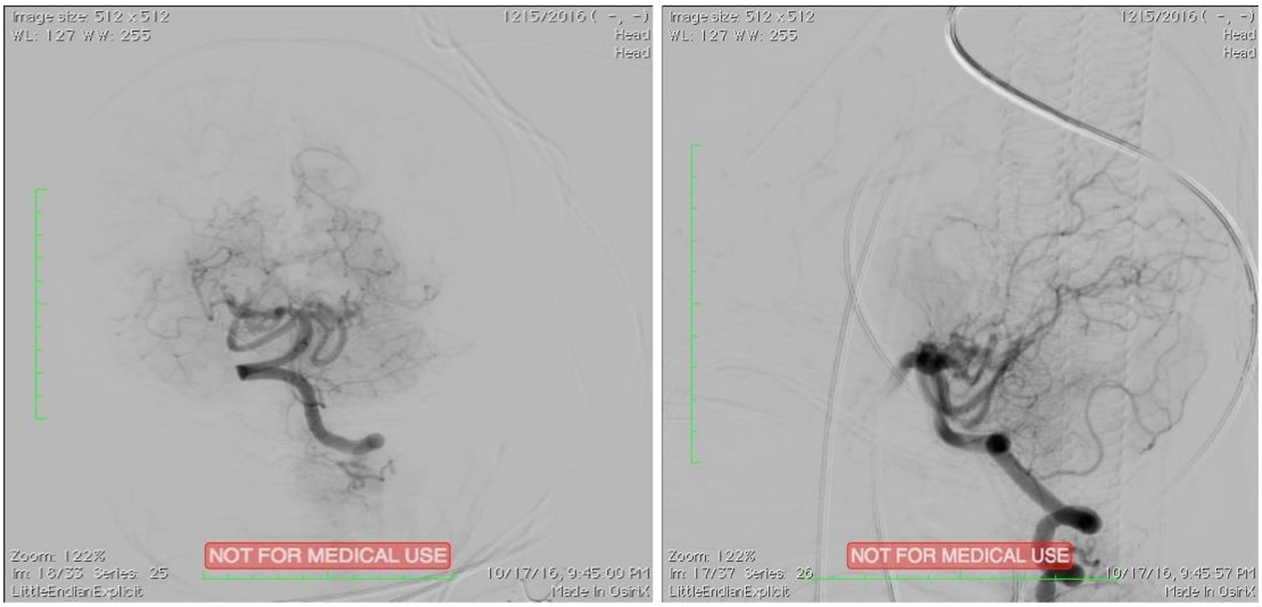

Fig. 3 Intraoperative angiographic picture showing complete occlusion of the VGM

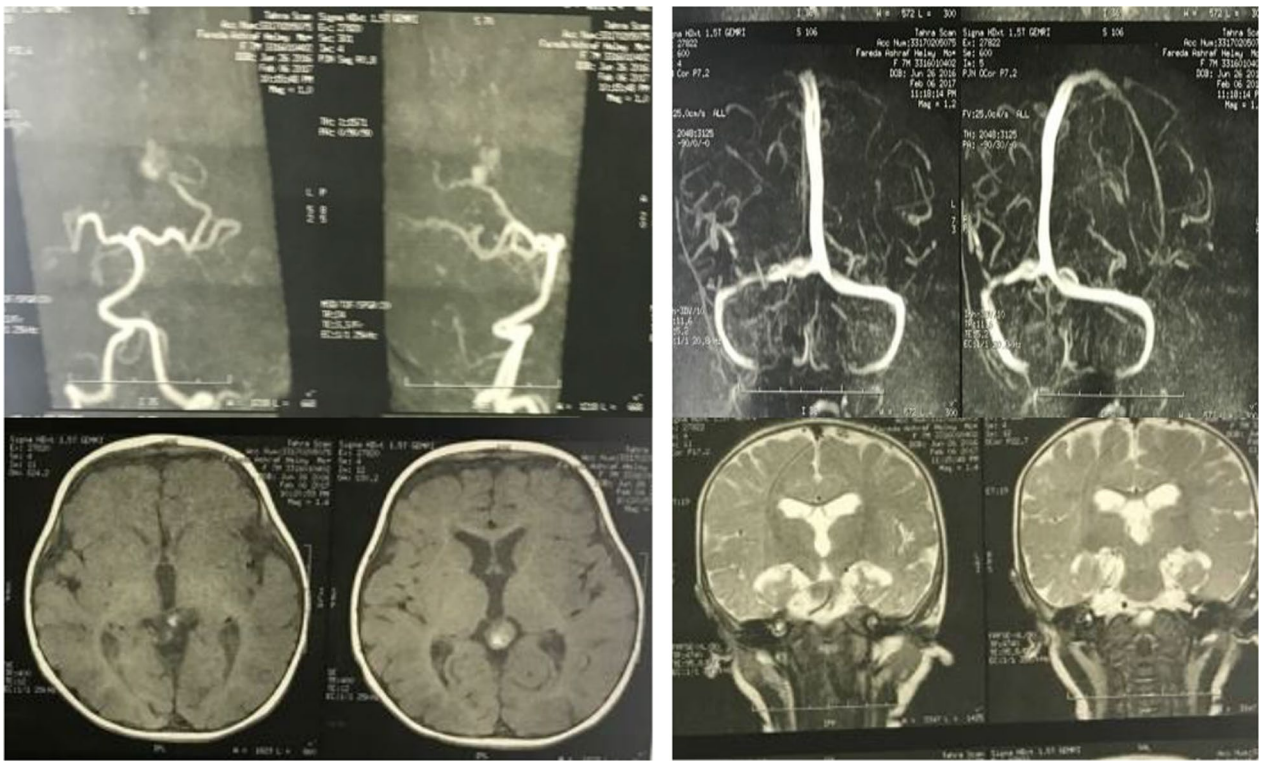

Fig. 4 Postoperative MRI brain with MRA and MRV showing complete occlusion of the VGM

studies underscore the age-specific symptomatology of VGAM, aiding in the identification and treatment of this condition within different pediatric age brackets [19].

Savage et al., in their study explained that the most common three symptoms were heart failure (68%; 142/210), hydrocephalus (15%; 31/210), and increasing head circumference (6%; 13/210) [20].

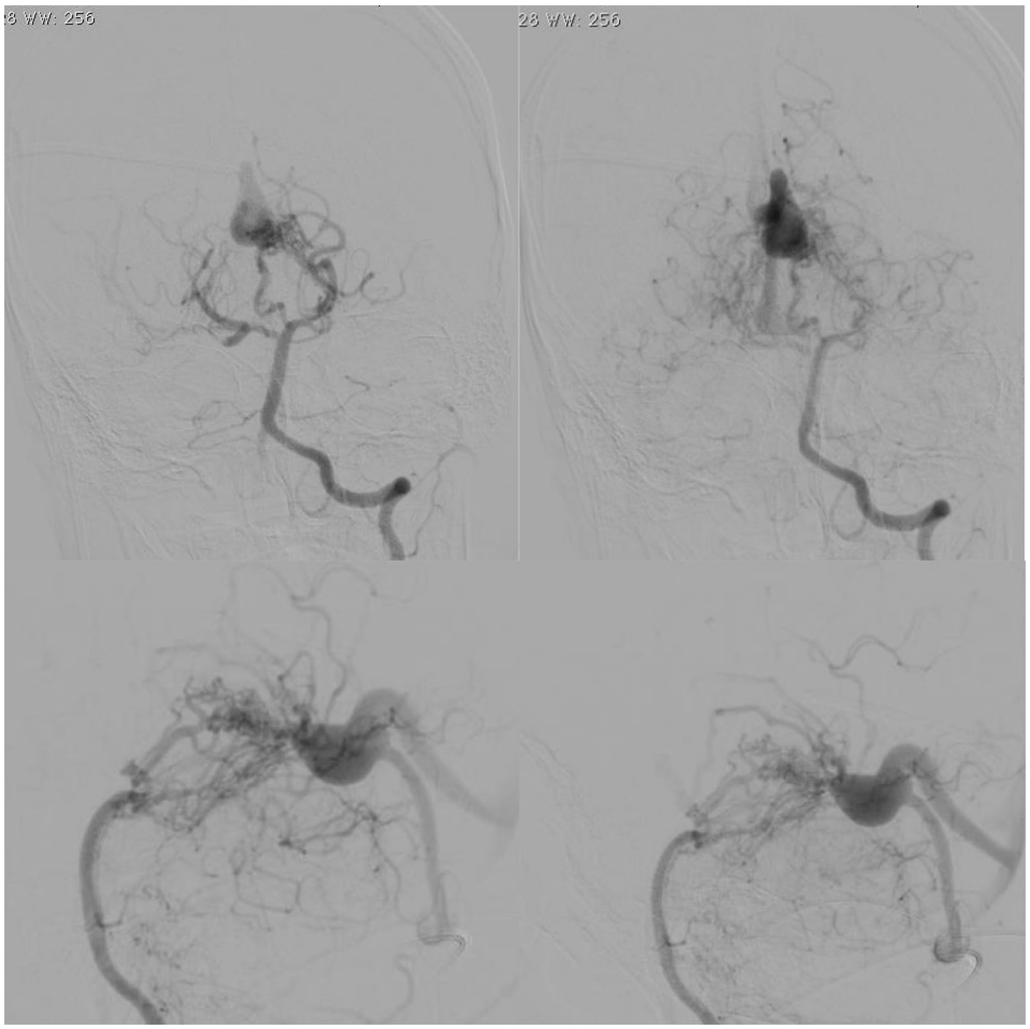

Fig. 5 Intraoperative angiographic picture of choroidal type VGAM

All these studies are in line with our study, as in our study we had 19 patients with VGAM 3 patients of them were neonates and 2 of them presented with CHF and the other one had brain melting syndrome (hydrodynamic disorder), we had 10 patients in infantile age group with 8 out of these 10 patients presented with macrocrania.

In our study in patients with VGAM 8 patients (42.1%) had macrocrania, 3 patients (15.8%) had CHF, another 3 patients (15.8%) presented with seizures and 2 patients (10.5%) presented with developmental delay.

As mentioned before, high-flow AV shunts and the physiological condition of the neonates and infants induce a specific symptoms according to the shunt type, there are other characteristic features of BAVS in young children (2–10 years), where new presentation of VGAM is rare and PAVF and AVM are seen more frequently. VGAM is rarely seen in older children with headache, seizures and ICH or incidentally [21].

Currently, endovascular therapy remains the preferred first-line treatment for VGAM, with the lowest mortality and complication rates and favorable clinical outcomes.

A meta-analysis conducted by Jun Yan et al. revealed that the mortality rate without treatment reaches 47%, whereas endovascular intervention reduces it to 12% [22].

Berenstein in his study on 45 patients with VGM, had 4.4% mortality rate, while Kartik in his study on 48 patients with VOGM had mortality rate reaching 31% [23].

The study done by Chingiz Nurimanov declared a mortality rate of 9.1% (two cases): one patient died the day postoperatively due to severe cardiac decompensation, and the other patient passed away after one month from pneumonia related to pulmonary hypertension [24].

In a study done by FB Nuñez on 30 cases with newly diagnosed VGM, 30 percent of all patients (n = 30) were discharged without any further complications. 95% of all

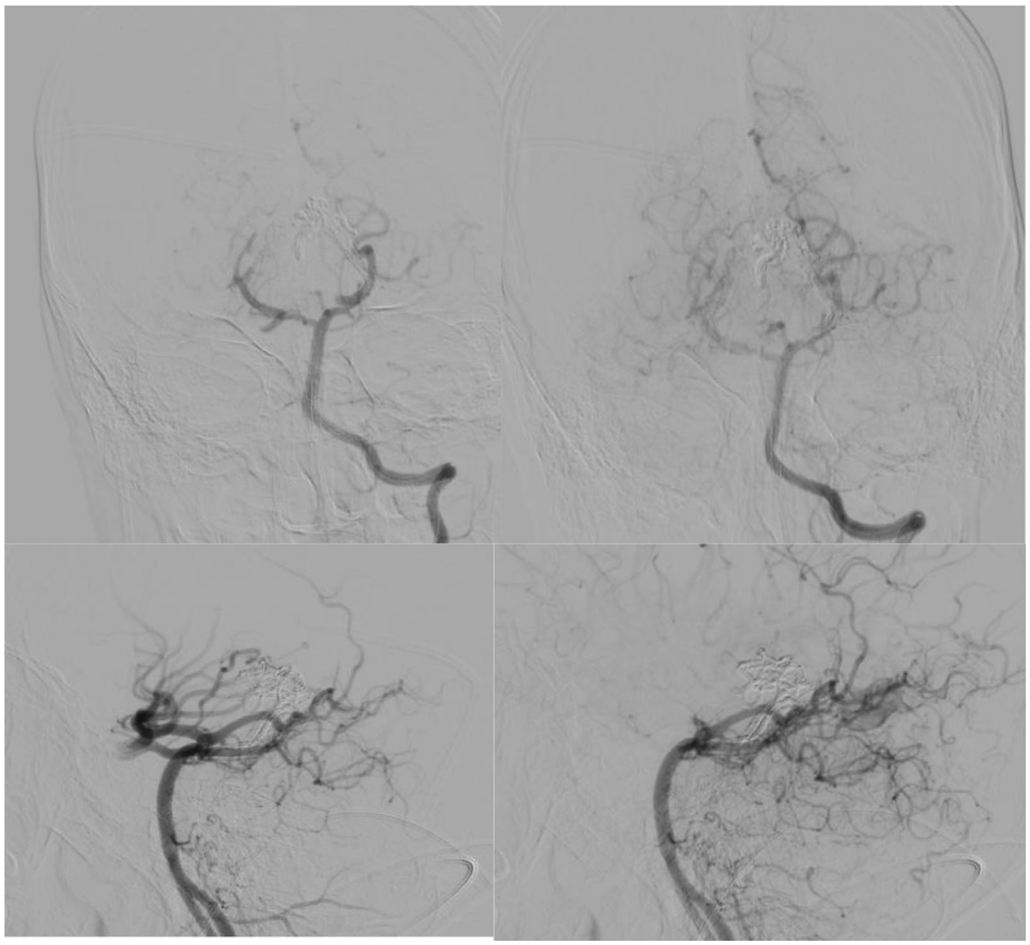

Fig. 6 Intraoperative angiographic picture showing complete occlusion of the choroidal type VGM

Table 1 Demographic data of the patients

	VGAM (II) (n = 19)
Age/year	
• Mean ± SD	1.81 ± 1.1
• Median (range)	1.2 (0.1–7)
Age group	
• Neonate	3 (15.8%)
• Infant	10 (52.6%)
• Child (2–10 y)	6 (31.6%)
• Child (10–18 y)	0 (0%)
Sex	
• Female	7 (36.8%)
• Male	12 (63.2%)

Table 2 Different clinical presentation of the patients

	VGAM (II) (n = 19)
Presentation	
• CHF	3 (15.8%)
• DD	2 (10.5%)
• Headache	0 (0%)
• Hydrocephalus	0 (0%)
• ICH	0 (0%)
• Increased SC	8 (42.1%)
• Melting brain syndrome	1 (5.3%)
• Neurological deficit	0 (0%)
• Proptosis	2 (10.5%)
• Seizures	3 (15.8%)

patients with endovascular treatment survived until discharge, but only 41% of these patients were discharged home without any complications [23].

Chingiz's study reported total occlusion rate (36.3%) and partial occlusion rate (63.7%) with good outcome in 72.7% and poor outcome in 27.3%. in comparison to

Table 3 Result and outcome

Result	VGAM (n = 19)
• Residual	4 (21.1%)
• Total Occ	15 (78.9%)
Outcome score	
• (4)	11 (57.9%)
• (3)	4 (21%)
• (2)	2 (10.5%)
• (1)	0 (0%)
• (0)	2 (10.5%)

Table 4 Shows the relation between the type of VGM and the occlusion rate and number of sessions needed

Type of VGM	Total occlusion	Residual	Number of sessions
Mural	13	0	14
Choroidal	2	4	19

Berenstein study who had total of 66.6% cases are neurologically and developmentally intact with outcome score 4, 20% had outcome score of 3, and 8.9% had outcome score of 2. He also reported reaching total occlusion in 82% of his cases. [24].

Arthur Hosmann had an experience on 18 consecutive patients 10 with choroidal and 8 with mural VGMs. Total occlusion was achieved in 87.5% of mural VGMs and 11.1% of choroidal VGMs. Good outcome was achieved in 53.8% and poor outcome in 46.2%, with an overall mortality of 16.7% [25].

In comparison to our study, we had 13 cases of mural type and 6 cases of the choroidal type. We could reach total occlusion in 15 patients (78.9%) and only 4 patients had residual after occlusion (21.1%).

From the 15 cases who reached total occlusion, we accomplished that in one setting in 12 patients and only 3 patients needed further sessions.

We had improvement in 11 patients (57.9%) in our study, while 4 patients discharged on score 3 (21%), and two patients discharged on score 2.

In our study, we had mortality rate 10.5% (two cases). One died during the embolization procedure, the other from glue embolization to the lung and the other had intraprocedural arterial perforation.

Conclusions

Introduction of endovascular embolization as the primary therapy has significantly improved prognosis.

Good selection of cases based on their score and good timing of treatment have a good prognostic factor with less morbidities.

Trans-arterial embolization is the primary choice of treatment. It is important to occlude the fistula sites for effective embolization.

Aiming to have complete closure of the shunt via endovascular embolization lower the incidence of complications and less exposure to other modalities.

Staged embolization should be considered for complicated multiple fistulas in order to minimize treatment risk.

Endovascular embolization of BAVS at appropriate time is safe and efficient method of treatment, understanding the pathological type and anatomical site of the fistula lead to successful and safe treatment with low rates of morbidity and mortality.

Abbreviations

VGAM	Vein of Galen aneurysmal malformation
SC	Skull circumference
AVM	Arteriovenous malformation
PAVF	Pial arteriovenous fistula
GCS	Glasgow Coma Scale
AV	Arteriovenous
mRS	Modified Rankin scale
CHF	Congestive heart failure
DD	Developmental delay
BAVS	Brain arteriovenous shunts

Acknowledgements

Not applicable.

Author contributions

Ass.Prof: Wael contributed in paper writing and data analysis. Prof: Radwan contributed in paper revision and design of the work. Dr. Osama contributed in design of the work and paper writing. Prof. Mohamed Alaa contributed in analysis and interpretation of data of the work.

Funding

This study was not supported by any sponsor or funder.

Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate

Study approval statement: This study protocol was reviewed and approved by Ethical committee of faculty of medicine Assiut university, approval number 17200365.

Informed consent

Written consent was obtained from the parents of the participants.

Competing interests

The authors have no conflicts of interest to declare.

Received: 15 November 2023 Accepted: 9 March 2024
Published online: 27 March 2024

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