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Original Article

Radiology Section

Clinical Presentation and Short-term Outcomes of Endovascular Onyx Embolisation in Dural Arteriovenous Fistula: A Prospective Observational Study

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ABSTRACT

Introduction: Dural Arteriovenous Fistulas (dAVFs) are abnormal vascular channels formed between venous sinuses or cortical veins and dural arteries, with idiopathic or multi-factorial causations, varied clinical presentations, and complex radiological angio-architecture. However, intra-arterial embolisation is the preferred management.

Aim: To study the clinical presentation and short-term outcomes of endovascular onyx embolisation for dAVFs.

Materials and Methods: The present prospective observational study was conducted at a tertiary care centre in Maharashtra, India from September 2016 to August 2017. Adults presented to an Emergency department with the symptoms of headache, convulsions, vomiting, or stroke and subsequently underwent Digital Subtraction Angiography (DSA) were screened. Patients diagnosed with dAVFs and treated with endovascular embolisation under general anaesthesia via the transfemoral route using a liquid embolic agent (onyx) were studied. Clinical presentation and angio architecture of dAVF have been reported along with the post embolisation complications.

Results: A total of 10 (8 males) patients were studied. Patients had an age range of 21-45 years. Convulsions 5 (50%) neurodeficit, and headache 4 (40%) each) were the most common presenting complaints. Left transverse sinus was the prominent dAVF location observed 4 (33.33%). A total of 12 arteries were embolised, (10 (83.33%) arteries showed no angiographic residue after a single session of embolisation, and 2 (16.67%) patients had minimal residue after two sessions of trans-arterial onyx embolisation).

Conclusion: Endovascular onyx embolisation should be the treatment of choice for dAVFs. Onyx in appropriate concentration and a finely-honed technique achieves better cure rates.

Keywords: Femoral arterial access, General anaesthesia, Honed technique, Scalp necrosis

INTRODUCTION

The dAVFs are anomalous vascular communications that form between venous sinuses or cortical veins and dural arteries. dAVFs comprise 10-15% of intracranial Arteriovenous Malformations (AVMs), however, their detection rate is a low 0.16 per 100,000 per year among adults [1]. While most dAVFs are acquired without any predisposing cause, studies report a significant positive association of sinus thrombosis with the evolution of dAVF. Consequently, thrombophilic states are considered high risk for dAVFs [2]. Intracranial complications of sinusitis, trauma, surgical interventions, and tumours affecting cortical veins or dural sinuses, such as meningiomas, are other reported possible causes. Common clinical presentations include severe pulsatile tinnitus, headache, seizures, neurological deficits (associated with intraparenchymal haemorrhage), exophthalmos, chemosis or progressive dementia [2,3].

dAVFs can arise anywhere within the dura however the common locations include transverse, sigmoid and cavernous sinuses. Widely used classification of dAVFs is Cognard classification [Table/Fig-1], based upon the anatomy of venous drainage, which predicts the clinical outcome [3,4].

Lesion type	Definition
I	Drains into dural venous sinus with antegrade flow
IIa	Drains into dural venous sinus with retrograde flow
IIb	Drains into dural venous sinus with antegrade flow+CVD
IIa+b	Drains into dural venous sinus with retrograde flow+CVD
III	Drains directly into subarachnoid veins (CVD only)
IV	Drains directly into subarachnoid veins with ectasia of the draining vein
V	Drains directly into spinal peri-medullary veins

[Table/Fig-1]: Cognard Classification of dAVFs.

Neuroradiological advances in last few decades have resulted in deeper understanding of these lesions. This with simultaneous advances in endovascular, microsurgical and radio-surgical techniques have radically modified the treatment strategies for dAVFs [5-7]. Since dAVFs are infrequent, large series publications and population-based long-term outcome studies after treatment in such cases have been scarce [6,7]. This study Presented angiographic evaluation and endovascular treatment of 10 patients with intracranial dAVFs with demographic data, clinical presentation, angiographic architecture, technique, and short-term outcome of onyx embolisation as descriptors.

MATERIALS AND METHODS

The present prospective observational study was conducted in the study was conducted at Division of Interventional Neuroradiology, Department of Radiology of Seth G. S. Medical College and KEM Hospital, Mumbai, Maharashtra, India, from September 2016 to August 2017. Informed written consent was obtained from the eligible participants.

Inclusion and Exclusion criteria: Adults (either gender) presented to an Emergency department with the symptoms of headache, convulsions, vomiting, or strokes were screened. Those with aggressive clinical or imaging signs (neuro-deficit, intracranial hemorrhage, reflux into cortical vein, direct leptomeningeal drainage), those with severe benign symptoms (bruit or intolerable headache),

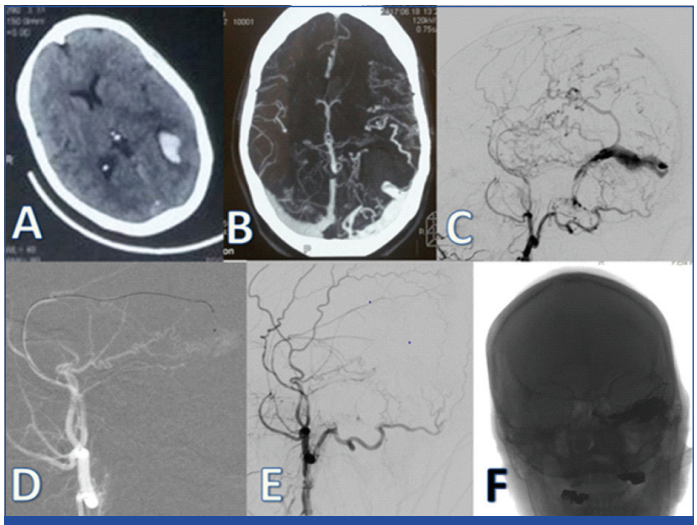
diagnosed with dAVF, and willing to participate were included. Patients with age less than 18 years and uncorrectable coagulopathy were excluded from the study. We enrolled 10 patients and studied 12 fistulas (two patients had dual fistulas).

Study Procedure

General demographic information was recorded. Clinical assessments were performed. All of them underwent a six vessel DSA to assess the precise angioarchitecture of the dAVF and to formulate further treatment plan. Endovascular embolisation was done under GA via transfemoral route and using onyx (ethylene vinyl alcohol copolymer) as embolising material.

Endovascular technique: Angiography and embolisation was performed under general anaesthesia after systemic heparinisation, on Artis Zee, Siemens Medical Systems (biplane angiographic unit). ECA, ICA, and Vertebral Artery (VA) catheterisation were done using a 6F guiding catheter. Super-selective cannulation of dominant feeding arteries was done with Dimethyl Sulfoxide (DMSO) compatible micro-catheters (Marathon, ev3). Mirage or X-Pedion (ev3) was the micro-guidewires used. After positioning the microcatheter as near as possible to the draining sinus or vein within the fistula, angiogram was performed to confirm dAVF architecture and catheter positioning. A 10 mL normal saline, followed by 0.25 mL of DMSO was slowly injected over 40 seconds, and was sequentially used to flush the catheter dead space

The onyx vial was agitated on a mixer for 20 minutes prior to injection. An initial slow injection of 0.2 mL was performed without image guidance. Subsequent injections were carried out under biplane roadmap guidance and continued until angiographic cure was achieved [Table/Fig-2]. Anticoagulation was done only in patients with significant cortical venous stasis post-embolisation.



[Table/Fig-2]: a) Axial non-contrast CT showing acute intraparenchymal haematoma in left occipital lobe; b) Contrast-enhanced axial CT showing multiple convergent vessels on left transverse sinus suggestive of DAVF; c) Lateral left ECA angiogram showing dAVF involving left transverse sinus; d) Final micro-catheter position close to fistula; e) Post-procedure angiogram showing complete exclusion of fistula; f) Spot image showing onyx cast.

STATISTICAL ANALYSIS

Descriptive statistics were used to summarise the characteristics of the study population. Categorical variables were presented as frequencies and corresponding percentages. All data were compiled and analysed using Microsoft Excel 365.

RESULTS

A total of ten (8 males) patients with dAVF were studied. Patients had an age range of 21-45 years. Convulsions (50%), neuro-deficit, and headache (40% each) were the most common presenting complaints.

- A total of 12 arteries were embolised in ten patients: Eight middle meningeal artery and four superficial temporal artery. Etiology of dAVF was venous thrombosis in 6 (60%) patients, trauma in 2 (20%) patients (both of whom developed osteo-frontal dural vein dAVF), and no identifiable cause could be found in the remaining 2 (20%) patients.
- Clinical presentation:

Left transverse sinus was the prominent location observed [Table/Fig-3]. Patients with superior sagittal sinus involvement had a sudden onset of diminished vision [Table/Fig-4].

No.	Location	Frequency (%)
1	Superior sagittal sinus and left transverse sinus	2 (16.67%)
2	Bilateral transverse-sigmoid sinuses and Torcula	2 (16.67%)
3	Left transverse sinus	4 (33.33%)
4	Left occipital cortical vein	2 (16.67%)
5	Left frontal osteo-dural vein	2 (16.67%)
	Total	12 (100%)

[Table/Fig-3]: Distribution of dAVF location.

No.	Clinical presentation	Frequency
1	Sudden onset diminution of vision	2 (16.67%)
2	Convulsions	6 (50%)
3	Altered mental status (GCS 5/15)	2 (16.67%)
4	Headache	4 (33.33%)
5	Post-traumatic proptosis of left eye globe	2 (16.67%)
6	Intracranial (cerebral) haemorrhage	6 (50%)
7	Progressive neuro-deficit	4 (33.33%)

[Table/Fig-4]: Distribution of clinical presentation in dAVF.

Angio-architecture of dAVF: The locations of the fistulae were: left transverse sigmoid (4), bilateral transverse-sigmoid (2), superior sagittal sinus (2), frontal osteo-dural (2) and cortical venous (2) [Table/Fig-5].

Outcomes and complications post-embolisation: In 08/10 cases, complete fistula closure (cure) was seen after a single session through single pedicle injection. In two patients (dAVF with multiple feeders of superior sagittal and transverse sinuses), slow-flow residue, despite two sessions of separate arterial pedicle embolisation, was seen. Seven patients were extubated postoperatively, observed in the Intensive Care Unit (ICU) for 24 hours, and remained stable. A total of 3/10 (30%) patients suffered post-interventional complications with mortality in 2 (16.67%) cases [Table/Fig-6].

- **Follow-up at one month:** Clinical follow-up after one month and an MRI at three months was done. Post follow-up eight patients had resolution of symptoms during short-term follow-up (including the cases with residual fistula).

DISCUSSION

Dural AVFs are rare entities formed by abnormal shunts between dural venous sinuses or leptomeningeal veins and meningeal arteries, which have varied symptomatology [5,6]. Presence of cortical venous reflux has been shown to correlate with aggressive symptoms. Hence, the therapeutic goal has to be complete shunt closure, which is done by occlusion of affected vein or sinus, if it is not draining the brain. Embolisation, radio-surgery and surgery are the available therapeutic options [5,6]. While initially multidisciplinary strategy was applied for complex dAVFs [7,8], developments in endovascular methods, such as high flexibility hydrophilic catheters, wires, and newer embolic agents, have resulted in dramatic improvement in their outcome, compared to the mixed results seen using surgery and radiosurgery despite technical advancements in all fields.

Nogueira RG et al., conducted a study in which 17 procedures on 12 patients were performed using transarterial embolisation with onyx for treatment of dAVF [9]. The age range of patients was 44-68 years.

Type*	Location (N)	No. of fistulae	Reflux	Venous rerouting	Outflow stenosis	Venous ectasia
Sinus (4)	Left transverse sinus (4)	Multiple (2)	Absent	Absent	Absent	Present
		Single (2)	Absent	Absent	Absent	Absent
Cortical veins (4)	Left frontal osteo-dural vein (2)	Single (2)	Absent	Absent	Absent	Absent
	Left occipital cortical vein (2)	Multiple (2)	Present	Absent	Absent	Absent
Sinus and cortical venous (4)	Superior sagittal sinus and left transverse sinus (2)	Multiple (2)	Present	Absent	Present in middle 1/3rd	Absent
	Bilateral transverse-sigmoid sinuses and Torcula (2)	Multiple (2)	Present	Present	Absent	Absent

[Table/Fig-5]: Distribution of angio-architecture of dAVF.
*It represents whether the fistula is on dural venous sinus or cerebral cortical vein

No.	Complications	Frequency	Location of DAVF	Management	Outcome
1	Scalp necrosis	01 (8.3%)	Superior sagittal sinus and left transverse sinus	Local wound care	Complete healing
2	Ventilator Associated Pneumonia (VAP)	01 (8.3%)	Bilateral transverse-sigmoid sinuses	Ventilator support and critical care	Succumbed to VAP after 12 days
3	Intra-cranial haemorrhage (ICH)	01 (8.3%)	Bilateral transverse-sigmoid sinuses	Ventilator support and critical care	Succumbed to VAP after 10 days

[Table/Fig-6]: Distribution of post-embolisation complications.

Two patients had minimal residual shunting after embolisation. There was one angiographic recurrence on follow-up. There was no significant morbidity or mortality. In the present study, the age range of patients was 21-45 years. We also had two patients with minimal residual fistula even after two sessions of embolisation. This was attributed to the presence of multiple shunts. In contrast to the study of Nogueira RG et al., there were two mortalities in our study. The mortality was attributed to long-term ICU stay.

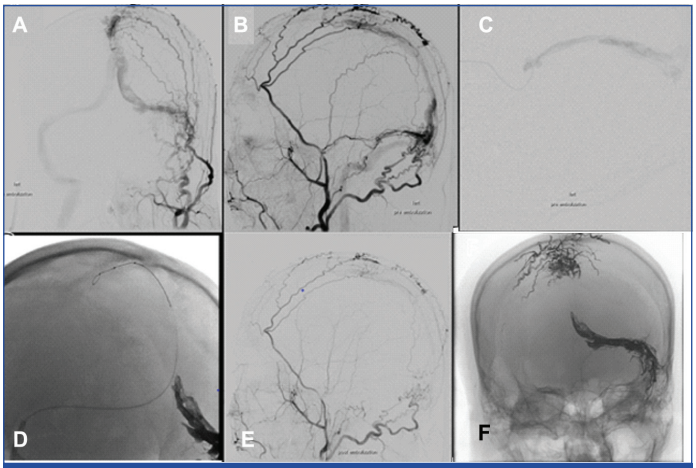
In a retrospective multi-institutional study by Li Y et al., the mean follow-up after transarterial embolisation treatment for dAVF was 29 months [10]. They had major complications in six patients. Presence of flow symptoms, age over 65, presence of an occipital artery feeder, and preprocedural home anticoagulation use were predictive of non-obliteration. The transverse-sigmoid sinus junction location was associated with fewer complications, whereas the tentorial location was predictive of poor functional outcomes. In the present study, follow-up was of short term, i.e., six months. The present study also showed fewer or no complications in patients with a transverse-sigmoid sinus junction located dAVF. We did not have any patient with tentorial located dAVF.

Hu YC et al., analysed 63 dAVF in 50 patients treated with transarterial onyx embolisation with angiographic cure in 79% [11]. Middle meningeal artery was conduit for embolisation. One patient had a permanent complication of unimproved cranial nerve palsy. In our study, all procedures were transarterial only, and we did not use trans-venous approach. The main conduit of the present study for embolisation was similar to Hu YC et al., i.e., middle meningeal artery. None of our patients had cranial nerve palsy as seen in the study of Hu YC et al., This may be attributed to the small sample size in our study as compared to the study by Hu YC et al., [11].

Singla A et al., reported a case of dAVF treated by transarterial onyx embolisation via occipital artery [12]. The patient came after three months with small pieces of onyx cast extruded from the scalp with areas of scalp necrosis. In the current study, we also encountered one patient with scalp necrosis one month after treatment.

Endovascular treatment of dAVFs has been drastically changed by onyx (ev3) due to its unique physical characteristics. It is a non-adhesive liquid solution of ethylene vinyl alcohol copolymer in DMSO, with tantalum for radio-opacity. This facilitates longer duration and higher volumes of injection, with a higher range in the rate of injection from a single pedicle [13-15].

Excellent endovascular handling reduces complications by allowing complete dAVF occlusion, without residual reflux. Additionally, during onyx injections, interruption of the procedure for assessment using guiding catheter contrast angiography is possible, which cannot be done using NBCA. In midline fistulas, the occlusion balloon used on the venous side can avoid superior sagittal sinus occlusion [Table/Fig-7]. Lower concentration has better distal penetration due to reduced viscosity. In addition, pre-heating the onyx vial for 5-10 min at 70 C, compounds the penetration and flow of the polymer. Permanent or long-term cure with minimal complications using onyx can be expected since it has been proven not to result in any significant inflammatory changes in animal studies [16,17].



[Table/Fig-7]: a&b) Frontal and lateral left ECA angiogram showing dAVF involving superior sagittal sinus and left transverse sinus; c) Final position of microcatheter; d) Balloon catheter in superior sagittal sinus; e) Post-procedure angiogram showing exclusion of fistula; f) Frontal fluoro-spot showing final onyx cast.

Limitation(s)

The limitations of the present study were small sample size, short follow-up period and lack of comparison with alternative treatment.

CONCLUSION(S)

Endovascular onyx embolisation shows good outcomes in the treatment of intracranial dural arteriovenous fistulae. Physical properties specific to onyx make slow better controlled, injections possible, which gives a more predictable penetration. Endovascular onyx embolisation should be the treatment of choice for dAVFs.

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