Endovascular Embolization and Direct Percutaneous Injection in Management of Craniofacial Arteriovenous Malformations

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Abstract

Background: Large craniofacial high-flow malformations are rare, and there is no consensus on their treatment. Embolization has become an integral part of the treatment of these malformations. Cure of these lesions may be attained by embolization alone or embolization followed by surgical removal [8].

Objective: To evaluate the safety and efficacy of endovascular embolization and direct percutaneous injection in management of craniofacial arteriovenous malformation.

Patients and Methods: Thirteen patients with craniofacial AVMs with different sites. The patients underwent both endovascular embolization and percutaneous direct injection using liquid embolic materials.

Results: Post embolization angiogram displayed eight lesions were completely devascularized. The other lesions, three of them were subtotal devascularized (> 75%) after two sessions and the other two was 65% devascularized after one session. Minor complications were encountered (alopecia and ischemic necrosis at the site of injection).

Conclusion: Mixed endovascular and direct-puncture embolization is an effective and safe technique for preoperative devascularization of craniofacial AVMs.

Key Words: Craniofacial AVM – Endovascular – Direct embolization.

Introduction

ARTERIOVENOUS malformations (AVMs) are vascular lesions that present as a direct communication between an artery and a vein without capillary connections but with a tangle of intervening vessels, known as the nidus [1,2]. Extra-cranial AVMs are most commonly found in the head and neck region, more specifically in the scalp, cheek, ear and nose [1,3]. The clinical presentations include cosmetic defects, pain, bleeding and ischemic ulceration [4]. The management of these lesions is challenging because of their unpredictable biologic behavior and high incidence of recurrence if not managed correctly. Trans-arterial endovascular embolization of high-flow AVM lesions with PVA particles is generally not effective because of frequent incomplete closure as well as recurrence of the AVM. However, embolization with n-BCA is more effective and permanent [5]. Direct-puncture is used for direct access into the vascular nidus or the draining vein to embolize a larger portion of the nidus [5,6,7]. In this study, we aimed to evaluate the safety and the efficacy of endovascular embolization and direct injection in management of ciroidal aneurysm of the scalp.

Patients and Methods

The study was conducted in the vascular and Interventional Unit-Radio-Diagnosis Department-Assiut University Hospitals during the period from August 2012 and August 2016, after approval of the institutional board of ethics. A written consent was obtained from each patient. A brief explanation of the technique was given to every patient.

Thirteen patients with craniofacial AVMs managed with trans-arterial and percutaneous (direct) embolization in this prospective study. They were 9 males and 4 females and their ages ranged from 14 to 35 years. Trauma was the reported a cause in 3 cases, and the other 10 patients had spontaneous lesions.

All patients presented with swelling. The main cause of management of our patients was cosmetic disfigurement and the second common cause was pain.
Different diagnostic modalities had been done for the patients before treatment. Informed consent was obtained from each patient before the procedure.

Our target was to gain total devascularization of the nidus as much as possible with no intended cure at the first session to avoid complications.

**Trans arterial embolization:**

We began by trans-arterial embolization through the main feeder, followed by direct puncture of the nidus if needed. The intervals between the sessions ranged from two to four weeks.

The procedure was started by doing right femoral puncture and insertion of 5F introducer sheath, followed by bilateral internal and external carotid angiogram using 5F catheter (Vertebral or Simmonds II curves) to obtain detailed anatomical and hemodynamic information. For doing embolization, we exchange the diagnostic catheter by a 5F guiding catheter (Guider soft tip, Boston Scientific) into the external carotid artery. A 1.5-1.8F microcatheter (Marathon, Ultra-flow, ev3, or Boston Scientific Renegade HI-FLO Micro-Catheter Kit 135cm x 10cm) was navigated under road mapping through the supplying artery and advanced as near as possible to the nidus of AVMs with the aid of blood flow. Super-selective angiography was performed through the micro-catheter to clarify the angioarchitecture and hemodynamics of the nidus. Trans-arterial embolization was performed using a mixture of n-butyl cyanoacrylate: NBCA (Histocryl; B Braun Aesclohp) and iodized oil (Lipiodol Ultrafluid; Andre Guerbet) n-BCA and Lipiodol Ultrafluid were mixed at the volume ratio of 1:3 or 1:4 according to the position of the catheter tip and flow speed. The micro-catheter was flushed with 5% dextrose solution, and then the mixture was injected under monitoring of the DSA images. When flow stasis of the feeding artery was obtained, and retrograde filling of the mixture toward the micro-catheter tip was observed, the injection was stopped and the micro-catheter was withdrawn. We embolized a single feeder per session, leaving the other supplying branches to visualize the nidus and to do roadmap for direct puncture.

**Direct puncture:**

We selected either the nidus itself or the supplying artery as near as possible to the nidus for direct puncture. A 20G short needle was introduced under guidance of angiographic roadmap or overlay techniques. We started injection of contrast medium before embolization to assess the needle position, filling of the nidus, and the speed of flow to the nidus and draining veins. If there was rapid filling of the venous side we chose to repeat the direct angiogram with manual compression of the rapidly filled veins. If the draining vein could not be occluded with compression, we sometimes used another needle for puncturing this vein and embolized it with high concentration NBCA (60%-80%) before embolization of the nidus. The nidus was then embolized using 20%-40% NBCA/Lipiodol mixture that was adjusted according to the flow rate throughout the nidus on the last direct angiogram. We stopped injection if there was flow to the venous side, extravasation or polymerization of the embolizing material, or after injection of 2ml of the mixture. We stopped further treatment when the nidus was totally devascularized, or when the residual lesion was too small to be accessed either by trans-arterial or direct embolization.

**Results**

The clinical characteristics of the patients and the outcome of the procedures are summarized in the (Table 1).

Thirteen patients were included; their age ranged from (10-48) years and the mean age was (28±11.3 years). They were 4 females (30.8%) and 9 males (69.2%) patients.

The main cause for management of craniofacial AVM was cosmetic. Three patients underwent embolization for other causes (headache and pain).

Most of our patients were scalp AVM lesions (69.3%) and the other lesions were cheek AVM.

Bilateral arterial supply was encountered in 3 patients bilateral STA and occipital artery in 3 patients. Ipsilateral arterial supply either by single or multiple branches was encountered in 10 patients.

Twenty-one sessions of embolization were done for thirteen patients with craniofacial AVMs. Eight patients (61.5%) were managed successfully by one session. Five patients needed more than one session due to large size of the nidus, avoidance of possible skin necrosis and presence of multiple arterial feeders necessitated more than one session of embolization.

The embolizing materials used were (n-Butyl cyanoacrylate) n-BCA that was injected in 12 patients and onyx was injected in one patient.

Total embolization had been obtained in 8 patients (61.5%), subtotal occlusion obtained in 3 patients (23%) and partial occlusion was detected in 2 patients (14.5%).
Causes of subtotal and partial embolization were the complete occlusion of the proximal part of the feeding pedicle with no definite other arterial pedicel for embolization and large sized nidus, avoidance of large area of skin necrosis and avoidance of embolization of the important blood supply.

Nine patients (69.2%) underwent both trans-catheter embolization and direct percutaneous embolization. Four patients (30.8%) underwent trans-catheter embolization.

No major complication had been noticed in any of our patients. Post procedure minor complications have been occurred in 4 patients (30.8%), 2 patients complaining of focal alopecia and wound necrosis and 1 patient complaining of focal alopecia and wound necrosis alone and 1 patient complaining of wound necrosis at the site of local embolization.

Skin necrosis healed by medical treatment without need for surgery after 3 weeks and normal hair growth regained within 6 months.

Most of the patients had some adverse effects after injection; pain, hotness and redness at the site of embolization and all of them needed non opioid analgesia and anti-inflammatory drugs and the adverse effects are completely disappeared.

Table (1): Description of all cases of craniofacial AVMs.

<table>
<thead>
<tr>
<th>Cases</th>
<th>Sex &amp; Age</th>
<th>Presentation</th>
<th>Feeding vessels</th>
<th>Embolic materials</th>
<th>Route of embolization</th>
<th>Degree of devascularization</th>
<th>No. of sessions</th>
<th>Complications</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>M/19Y</td>
<td>Right Parietal Painful pulsatile scalp swelling</td>
<td>Bilateral STA Bilateral OCA</td>
<td>n-BCA</td>
<td>Transarterial Direct</td>
<td>Total</td>
<td>4</td>
<td>Alopecia Skin necrosis</td>
<td>Complete disappeared</td>
</tr>
<tr>
<td>2</td>
<td>F/32Y</td>
<td>Right Parietal painless scalp swelling for cosmetic concern</td>
<td>Bilateral STA Bilateral OCA</td>
<td>n-BCA</td>
<td>Transarterial Direct</td>
<td>Total</td>
<td>2</td>
<td>No</td>
<td>Complete disappeared</td>
</tr>
<tr>
<td>3</td>
<td>M/26 Y</td>
<td>Left parietal painless scalp swelling for cosmetic concern</td>
<td>Bilateral STA Bilateral OCA</td>
<td>n-BCA</td>
<td>Transarterial Direct</td>
<td>Total</td>
<td>1</td>
<td>Alopecia</td>
<td>Reduced lesion size</td>
</tr>
<tr>
<td>4</td>
<td>M/30Y</td>
<td>Right temporo-parietal painless scalp swelling for cosmetic concern</td>
<td>RT STA RT IMA</td>
<td>n-BCA</td>
<td>Transarterial Direct</td>
<td>Total</td>
<td>2</td>
<td>No</td>
<td>Reduced lesion size</td>
</tr>
<tr>
<td>5</td>
<td>M/14Y</td>
<td>Right paramedian forehead painless scalp swelling for cosmetic concern</td>
<td>RT STA RT PAA</td>
<td>n-BCA</td>
<td>Transarterial Direct</td>
<td>Partial</td>
<td>1</td>
<td>Skin Necrosis Alopecia</td>
<td>Missed</td>
</tr>
<tr>
<td>6</td>
<td>M/33Y</td>
<td>Right occipital painless scalp swelling for cosmetic concern</td>
<td>RT STA RT PAA Rt. OCA</td>
<td>n-BCA</td>
<td>Transarterial direct</td>
<td>Total</td>
<td>1</td>
<td>Skin necrosis</td>
<td>Reduced lesion size</td>
</tr>
<tr>
<td>7</td>
<td>F/36Y</td>
<td>Right occipital pulsatile scalp swelling</td>
<td>RT OCA</td>
<td>n-BCA</td>
<td>Transarterial</td>
<td>Subtotal</td>
<td>1</td>
<td>No</td>
<td>Reduced lesion size and headache</td>
</tr>
<tr>
<td>8</td>
<td>M/32Y</td>
<td>Right chin painless swelling for cosmetic</td>
<td>RT FA</td>
<td>n-BCA</td>
<td>Transarterial</td>
<td>Total</td>
<td>2</td>
<td>No</td>
<td>Reduced lesion size</td>
</tr>
<tr>
<td>9</td>
<td>M/33Y</td>
<td>Right lip painless swelling cosmetic concern</td>
<td>RT FA</td>
<td>n-BCA</td>
<td>Transarterial direct</td>
<td>Subtotal</td>
<td>1</td>
<td>No</td>
<td>Surgical excision</td>
</tr>
<tr>
<td>10</td>
<td>F/26Y</td>
<td>Right cheek painless swelling for cosmetic concern</td>
<td>RT FA</td>
<td>Onyx</td>
<td>Transarterial direct</td>
<td>Subtotal</td>
<td>3</td>
<td>No</td>
<td>Surgical excision</td>
</tr>
<tr>
<td>11</td>
<td>F/17Y</td>
<td>Right temporal painless scalp swelling for cosmetic concerns</td>
<td>RT STA</td>
<td>n-BCA</td>
<td>Transarterial direct</td>
<td>Total</td>
<td>1</td>
<td>No</td>
<td>Reduced lesion size</td>
</tr>
<tr>
<td>12</td>
<td>M/10Y</td>
<td>Right Parietal painless scalp swelling(cosmetic concern)</td>
<td>RT STA, RT OCA</td>
<td>n-BCA</td>
<td>Transarterial</td>
<td>Partial</td>
<td>1</td>
<td>No</td>
<td>Surgical excision</td>
</tr>
<tr>
<td>13</td>
<td>M/48Y</td>
<td>Pulsatile painful sublingual swelling</td>
<td>RT lingual artery</td>
<td>n-BCA</td>
<td>Transarterial</td>
<td>Total</td>
<td>1</td>
<td>No</td>
<td>Surgical excision</td>
</tr>
</tbody>
</table>

STA = Superficial temporal artery. OCA = Occipital artery. FA = Facial artery. IM A = Internal maxillary artery. PAA = Posterior auricular artery. RT = Right. LT=left
Fig. (1): Male patient 32 years old complaining of right chain and lower lip painless swelling for cosmetic concern 
(a&b) Axial T2W and MSCT angiography displaying abnormal soft tissue lesion with high signal within the 
right submandibular and right lip region and vascular malformation involving the right facial artery and their 
branches (c&d). Show right external carotid angiogram right facial AVM and Post-procedural angiogram 
showing subtotal devascularization of the lesion.

Fig. (2): Male patient 21 years old presented with post traumatic painless pulsatile scalp swelling Image (a) photo shows patient 
before embolization, Images (b&c) CT shows parietal scalp swelling suggesting scalp AVM. Image (d) show carotid 
angiography conforms scalp AVM with complete AVM devascularization images (d&f) patients during percutaneous 
and trans-arterial embolization image.
Fig. (2): (g) Control angiogram shows total devascularization of the lesion, image (h) reformatted images post embolization shows complete devascularization of the scalp AVM. Image (i) photo of the patient after embolization.

Fig. (3): Female Patient 26 Years old presented by right cheek and chin painless pulstle swelling for cosmetic concern; Images (a&b) photos of the patients before embolization showing right cheek bluish swelling. Images (d&e) enhanced MSCT and CT angiography show right facial artery and its branches AVM. Images (c&f) pre and post angiography showing complete devascularization of the AVM after transcatheter and direct percutaneous embolization of the AVM. Images (g&h) 3D CT reformat images after post embolization show complete devascularization of the right cheek AVM.
Discussion

Craniofacial AVMs are especially difficult to be managed surgically because of disfigurement, functional impairment, and the associated risks of life-threatening and hemorrhage [1].

Before the technical advances in the field of endovascular therapy, the treatment of vascular malformations was primarily reliant on surgical excision or ligation of feeding arteries [2,3]. However, surgical excision alone poses several problems: (i) the risk of hemorrhage, which can be fatal; (ii) the difficulty in assessing the extent of the lesion resulting in extensive surgical intervention, as the entire fistula often has to be removed to avoid recurrence; (iii) the possibility of recurrence starting from the dormant shunts localized far from the central nidus; and (iv) the anatomic, functional and aesthetic restoration [4].

The majority of the craniofacial malformations observed at birth, have a tendency to grow during puberty and do not involute [6]. However, many patients may also show progression in response to pregnancy, surgery or trauma [8]. Thus the mean age of presentation is variable according to the severity of the lesion. In our current study, the age of the patients ranged from 10 to 48 years and the mean age was (28±SD 11.3) years and these results meet with the most published previous studies [5,6,7].

It was stated that craniofacial AVMs affect females more than males (1.5:1) ratio [9]. However, most of our patients were males (69.2%). This discrepancy could be due to the relatively small number of the patients in our study.

In the literature, the main cause for craniofacial AVMs management was aesthetic cause and the other causes include pain and hemorrhage [5,6,10]. This was correlated with our study patients were seeking treatment for aesthetic purpose (76.7%) and for other causes such as pain and headache (22.3%).

Our Data show that most of our cases had cirsoidal aneurysm of the scalp (69.3%) of our cases. This incidence is higher compared to other similar studies. Jeong et al. [1] reported that 50% of the cases in his study and Thiex et al. [7] reported that 2 cases from his 21 cases were cirsoidal aneurysm of the scalp.

The commonest branches supplying scalp AVMs were the superficial temporal artery (STA) followed by the occipital artery (OCA). These results were in agreement with Han et al. [11] who reported that the superficial temporal artery was the main feeder in his 14 cases of craniofacial AVMs and also with Dabus et al. [12] who managed 3 cases of scalp AVFs and reported that STA was the main arterial feeder. The facial artery was the main arterial feeder for the facial AVMs. This findings was in accordance with Han et al. [11].

Combined direct percutaneous and transcatheter embolization has become widely accepted techniques with surgery, for the treatment of craniofacial AVMs [3,13,14].

Trans-arterial approach is mandatory for diagnostic purpose, partial embolization of the nidus, providing roadmap or overlay guidance to the direct injection, and assessment of the degree of devascularization. It is better to direct transcatheter embolization at the AVM nidus to avoid angiogenesis.

It is often impossible to achieve a satisfactory therapeutic result in a single treatment session. Because of the rarity of the craniofacial AVMs and the relative lack of understanding of their pathogenesis, no standard treatment has been established. As a result, various interventional options and treatment algorithms have been proposed [9].

Our research show that the trans-arterial embolization either solely or combined with direct embolization was important for total or subtotal devascularization of the craniofacial AVM lesions. We found that (61.5%) of our patients were managed successfully by one session while 38.5% needed more than one session due to large size and complexity of the nidus for avoidance of possible skin necrosis and or the presence of multiple arterial feeders. This results were in agreement with Art et al. [5] and Thiex et al. [7] reported that combined transarterial and direct embolization was successfully managed craniofacial AVFs.

Different-types of embolic materials were described in the management of the craniofacial AVM lesions; poly vinyl alcohol (PVA) particles, ethanol, coils and liquid embolic materials (N-Butyl cyanoacrylate) n-BCA and Onyx. The advantages of n-BCA include deep intranidal penetration, high thrombogenicity, per-manent occlusion, and easy delivery via small atraumatic micro-catheters [18]. Other non-adhesive embolic agents, such as coils and PVA particles, tend to recanalization over time, preventing treatment of large AVMs with multistage embolization because n-BCA polymerization is a relatively quick and uncontrollable process. Onyx, is a less-adhesive and slower-polymerizing material,
has made significant improvements in eliminating small AVMs and reducing the size of larger AVMs for surgical or radio-surgical treatment [15,16].

It was stated that the liquid embolic materials were the most common embolic materials used in devascularization of the craniofacial AVMs lesions [5,7,17]. This was correlated with our study as we used n-BCA (N-butyl cyanoacrylate) in 12 cases and onyx used in one case. Because the onyx was not available in our center, it may cause bluish discolouration and it is much expensive than n-BCA.

Direct percutaneous embolization technique is quicker, cheaper and easier than trans-arterial embolization. It allowed devascularization of a larger portion of the nidus than did trans-arterial approach. Also, it allowed safe devascularization of the nidus portion supplied by dangerous arteries that carried high-risk if embolized. It reduced the risk and the cost by avoiding selective catheterization of each supplying branches with multiple micro-catheters to get complete nidal devascularization [11,18]. Thiex et al. [7] stated that combined trans-arterial and percutaneous approaches was effective and safe in management of craniofacial AVMs. Most of our patients (69.2%) underwent combined trans-arterial and direct percutaneous embolization and the rest of our patients (30.8%) underwent trans-arterial embolization alone.

Gomes [18] reported that the direct percutaneous approach alone is effective in the management of the craniofacial AVMs in special cases like previous surgery and proximal occlusion of the arterial feeders which prevented successful endovascular embolization of the arterial feeders and made the endovascular embolization difficult for management. In our study combined endovascular and direct percutaneous embolization was effective and safe in management of the craniofacial AVMs in the same session.

In our current study, total devascularization was obtained in (61.5%) and incomplete devascularization was achieved in (38.5%). These results are in accordance with Art et al. [8] reported that among 9 patients who underwent embolization, 6 patients showed complete devascularization and 3 patients showed unfavorable devascularization. Thiex et al. [7] also reported that among 22 patients who underwent embolization, four patients showed complete devascularization, eight patients showed near complete devascularization and ten patients showed partial devascularization.

In our study, we revealed that total devascularization was achieved in 7 patients out of 9 patients using combined endovascular and percutaneous direct injection 4 patients underwent trans-arterial embolization alone. Complete devascularization was obtained in 1 patient with incomplete devascularization in the other 3 patients. Thus, combined endovascular and direct percutaneous injection was more effective than endovascular embolization alone in management of extensive craniofacial AVM. These results are in concordance with Cil et al. [6] reported complete and near complete devascularization obtained in 8 cases among 13 patients and partial embolization obtained in 5 cases and Thiex et al. [7] reported that 12 patients out of 22 patients showed complete and near complete embolization.

No major complications were encountered in our current study. Post-procedure minor complications occurred in 4 patients (30.8%). Two patients developed focal alopecia and skin necrosis and 1 patient developed focal alopecia alone and 1 patient skin necrosis at the site of local embolization. Skin necrosis was healed by medical treatment without need for surgery after 3 weeks and normal hair growth was regained within 3 to 4 months. These results are agreeing with Jeong et al. [1] and Art et al. [5] who “both” reported no major complications in their studies.

Conclusion:

Trans-arterial approach is obligatory for diagnostic purpose, partial embolization of the nidus, providing roadmap or overlay guidance to the direct injection, and assessing the degree of devascularization. It is better to direct trans-catheter embolization at the AVM nidus to avoid angiogenesis. However, when we use combined trans-arterial and direct injection embolization approaches, the occlusion of the artery more proximal that slows the flow through the nidus and facilitates the total occlusion of the nidus by direct approach.

Direct embolization provides good devascularization of the larger portion of the nidus than did trans-arterial approach. It permitted safe devascularization of the nidus portion supplied by dangerous arteries that carries high-risk if embolized. The percutaneous direct puncture embolization technique can be used alone or in combination with surgical resection as an alternative approach for superficial craniofacial AVM with prominent venous pouch [12].

It was previously reported that endovascular workup does not reduce effective lesion size [19,10]. However, in our work we noted significant reduction of the lesion size few months after emboliza-
tion. This decreased the role of surgery in treating such lesions and protected it from excision of the recent or the post-embolization skin necrosis.

Mixed endovascular and direct-puncture embolization is an effective and safe technique for preoperative devascularization of craniofacial arteriovenous malformation.

References