Periorbital Inner Canthal Arteriovenous Malformations: Percutaneous Glue Embolization in Three Cases

Krishnan Nagarajan1, Arockia Sensan Babu Arul1 Sekar Sabarish1 Swamiappan Elango1 Krishna Ramesh Babu2 Sunil Kumar Saxena3

1Department of Radio-Diagnosis, Jawaharlal Institute of Postgraduate Medical Education & Research (JIPMER), Pondicherry, India
2Department of Ophthalmology, Jawaharlal Institute of Postgraduate Medical Education & Research (JIPMER), Pondicherry, India
3Department of Otorhinolaryngology, Jawaharlal Institute of Postgraduate Medical Education & Research (JIPMER), Pondicherry, India

Address for correspondence Krishnan Nagarajan, MD, DM, Department of Radio-Diagnosis, Jawaharlal Institute of Postgraduate Medical Education & Research (JIPMER), Pondicherry 600506, India (e-mail: lknagarajan1@gmail.com).

Abstract

Inner canthal or palpebral arteriovenous malformations (AVMs) are uncommon and difficult lesions to treat if they are of high-flow type. Though they may present with mainly cosmetic reasons, they derive feeders from the ophthalmic artery and are associated with dangerous anastomoses. Percutaneous liquid embolic agent has been used to treat various head and neck vascular malformations and tumors and, if done in meticulous attention to detail, can offer cure or control before surgical excision. We report three adults who presented with medial canthal swelling and on imaging diagnosed to have high-flow AVMs. They underwent percutaneous n-butyl cyanoacrylate (glue) embolization and subsequently operated to excise the embolized malformation without any blood loss or complications.

Keywords

► inner canthal AVM
► arteriovenous malformations
► liquid embolic agents
► glue embolization

Introduction

Arteriovenous malformations (AVMs) are described as developmental hamartomas consisting of an abnormal nidus of dysplastic vascular channels with feeding arteries and draining veins and absence of normal intervening capillary network.1 AVMs of the eyelid and canthus are uncommon with only few isolated case reports and small case series.2-5 Endovascular treatment of these highly vascular lesions prior to surgical removal by selective embolization of the arterial feeders has been documented.1 However, endovascular embolization in lid lesions is technically challenging, due to the complex nature of the arterial supply of the orbit and its adnexa with its rich anastomoses.6 Percutaneous direct puncture of the nidus and venous pouch is a more efficient alternative to transarterial embolization in these superficial lesions. In this report, we describe our experience and outcomes in treating three cases of superficial periorbital inner canthal AVMs using percutaneous direct nidus puncture embolization.

Case 1

A 55-year-old female presented with rapidly increasing painful, pulsatile swelling over the medial canthus of the right eye for 3 months. The swelling was pulsatile with throbbing pain. Computed tomography (CT) angiogram revealed compact nidus of 3 × 2 cm size with prominent bilateral ophthalmic arteries, superior ophthalmic veins, and frontal scalp arteries and veins (►Fig. 1A, B). Digital subtraction angiography (DSA) revealed nidus at the medial canthus of right eye with feeders from bilateral ophthalmic artery of internal carotid arteries, external carotid (ECA) branches of facial (bilateral), and right internal maxillary arteries. The venous drainage was noted into bilateral angular, superior ophthalmic, and scalp veins (►Fig. 1C, D). In the same sitting, percutaneous embolization was attempted after placing table-side lead protection sheet for radiation during fluoroscopic roadmap injection of glue. The nidus close to dilated venous sac
was punctured with a 20G scalp vein needle (►Fig. 2A). We used 20G to avoid glue clogging with a thinner gauge needle. Contrast agent was injected to confirm the position of the needle within the nidus and to look for any reflux into the arterial side. After direct confirmation of the anatomic details and dynamic status (drainage vein and flow), and flush with 5% dextrose to avoid interaction of glue and prior contrast injected, 50% solution of n-butyl cyanoacrylate (n-BCA, Endocryl, Endotech, India) glue in lipiodol was injected under fluoroscopy roadmap till glue filled the major part of the nidus. After injection of glue, the needle was removed and check angiogram was done, which showed partial (60%) obliteration of the nidus with normal filling of bilateral ophthalmic arteries (►Fig. 2B). The patient had relief of pain and reduction in pulsatility. On 1-month follow-up, the swelling was static with glue cast in CT (►Fig. 2C). Second session of percutaneous glue injection was done 3 months later again using similar technique with 50% n-BCA glue. The firm swelling was excised in toto without blood loss. Histopathology showed thick-walled blood vessels with inflammatory infiltrate, occasional giant cells, and refractive foreign material due to glue.

Case 2

A 45-year-old female presented with gradually increasing, pulsatile swelling over the right inner canthus close to the nasal bridge. CT angiogram showed compact nidus of 1 cm size over the medial canthus of the right eye with prominent right ophthalmic artery and vein (►Fig. 3A, B). DSA showed small high-flow nidus fed by right ophthalmic artery, ECA branches of superficial temporal, internal maxillary, and facial arteries, and venous outflow into superior ophthalmic and angular veins (►Fig. 3C, D). Using the same technique as the previous patient, 50% solution of n-BCA (Endocryl) glue in lipiodol solution was injected into the nidus percutaneously. Postembolization angiogram showed complete obliteration of the AVM nidus with preserved ophthalmic artery. Plain CT done after 2 days showed the glue cast within the nidus (►Fig. 3E). The patient underwent surgical excision after 3 weeks.

Case 3

A 37-year-old male presented with a pulsatile swelling of 3 × 1 cm size over the inner aspect of the left upper eyelid for 6 years. Eye motility and ophthalmologic examination were normal. CT angiogram showed intensely enhancing nidus with prominent left ophthalmic artery and vein (►Fig. 4A, B). DSA revealed moderate-flow nidus in upper eyelid with feeders from the left ophthalmic artery, left superficial temporal, and internal maxillary branches of ECA and minimally from the right ophthalmic artery (►Fig. 4C, D). Venous drainage was into the superior ophthalmic, angular, and scalp veins. Though the flow was slightly slower than previous patients, similar percutaneous technique was used and 50% solution of n-BCA (Endocryl) glue in lipiodol was injected under direct fluoroscopy till the entire nidus filled. Postembolization angiogram showed near-complete obliteration of the AVM nidus.
Fig. 3 Case 2. Plain (A) and contrast (B) computed tomography (CT) showing small soft tissue density lesion (arrows) along the right nasal bridge close to the inner canthus. Digital subtraction angiography (DSA) of the right internal carotid artery (ICA) (C) and external carotid artery (ECA) (D) in lateral projections showing small nidus (arrows) fed mainly by ophthalmic artery, facial, internal maxillary, and superficial temporal with venous drainage into the angular and superior ophthalmic veins. Postembolization CT axial section (E) showing glue cast (arrow) in the lesion.

(►Fig. 4E). CT done on the next day revealed glue cast with palpebral edema which resolved with topical and medical management (►Fig. 4F). There was a reduction in the swelling at 6 weeks’ follow-up and it was excised. -Table 1 summarizes the clinical and angiographic findings of all three patients.

Discussion

Head and neck region is a common location for vascular anomalies and tumors of various types. Of these, simple sporadic type (type IV) AVMs as per the International Society for the Study of Vascular Anomalies classification (revised 2018), are common type of high-flow malformations. These lesions are now more dealt in endovascular method of treatment in single sitting or staged procedure. Percutaneous direct puncture embolization is a simple and safe technique for the treatment of superficial AVMs in accessible locations compared with transarterial embolization.6 It is sometimes difficult to gain distal access and perform superselective embolization of feeders particularly the ophthalmic artery.7 Apart from the risk of incomplete embolization and retained microcatheter tip with transarterial embolization,2 inadvertent embolization or reflux of embolic agent into the central retinal artery can lead to severe visual deficits.8 Percutaneous approach to embolization of these lesions has proved to be equally efficacious and safe with immediate obliteration of vascularity and relief of symptoms.6,9

In their series of craniofacial AVMs, Han et al used percutaneous glue injection in 14 craniofacial AVMs including 9 lesions with feeders from ophthalmic artery and used
venous compression during the percutaneous glue injection.\textsuperscript{9} Phadke et al reported a case of medial canthal AVM treated using percutaneous glue embolization and recommended the use of occlusion of feeding arteries to reduce the arterial inflow and manual compression to promote venous stasis.\textsuperscript{2}

Ou et al\textsuperscript{4} described three (scalp: 2 and nasal bridge: 1) cases of AVMs fed by the ophthalmic artery in their series of four patients and achieved complete obliteration by percutaneous glue injection. Pekkola et al\textsuperscript{10} described 19 craniofacial AVMs treated with ethanol sclerotherapy including a periorbital/nasal lesion which recurred after previous particle embolization and local resection. Su et al\textsuperscript{11} described 16 cases of periorbital AVMs over a period of 4 years out of which 13 had ophthalmic artery feeders. They used absolute alcohol, either transarterial or percutaneously under general anesthesia, for embolization in 28 sittings. Decock et al described the occlusion of the draining vein by transvenous approach to prevent iatrogenic embolization of the superior ophthalmic vein and cavernous sinus.\textsuperscript{12} In our cases, we did not use any occlusive technique. As the nidi was distally away from the central retinal branch of the ophthalmic artery, it was decided to stop injection if there is any reflux back into the distal ophthalmic artery. Adequate compression may not be anatomically achievable in the inner canthus for all lesions, and in fact, may even cause inadvertent changes in the flow dynamics within the nidi or draining veins. We did not encounter any reflux of embolic agent into the arterial side or incomplete embolization though it can happen if glue concentration is inappropriate for the flow characteristics of the malformation. High-flow AVMs at the inner canthal region pose a considerable treatment challenge, requiring interdisciplinary approach for its management for acceptable cosmetic and functional outcome.

### Table 1

<table>
<thead>
<tr>
<th>S. No.</th>
<th>Demography</th>
<th>Lesion location</th>
<th>Onset and Schobinger grade</th>
<th>Arterial feeders</th>
<th>Venous drainage</th>
<th>No. of embolization sessions</th>
<th>Result of embolization</th>
<th>Surgical excision</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>55/F</td>
<td>Right medial canthus and nasal bridge</td>
<td>Spontaneous II</td>
<td>Bilateral ophthalmic A., facial A., and internal maxillary A.</td>
<td>Bilateral superior ophthalmic vein &amp; angular veins</td>
<td>2</td>
<td>Complete</td>
<td>Yes, excised in toto</td>
</tr>
<tr>
<td>2</td>
<td>45/F</td>
<td>Right medial canthus</td>
<td>Spontaneous I</td>
<td>Right ophthalmic A., superficial temporal A., facial A.</td>
<td>Right superior ophthalmic &amp; angular vein</td>
<td>1</td>
<td>Complete</td>
<td>Yes, excised in toto</td>
</tr>
<tr>
<td>3</td>
<td>37/M</td>
<td>Left inner canthus &amp; upper eyelid</td>
<td>Spontaneous II</td>
<td>Left ophthalmic A., superficial temporal A., internal maxillary A.</td>
<td>Left superior ophthalmic &amp; angular vein</td>
<td>1</td>
<td>Near-complete</td>
<td>Yes, excised in toto</td>
</tr>
</tbody>
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Abbreviation: A, artery.