
ORIGINAL ARTICLE

Single-stage Embolisation Followed by Excision for Vascular Malformations at the Head or Neck

JB Chiang¹, HS Fung¹, WL Poon¹, PCM Chan², MWY Leung², C Liu², EYM Li³, TCY Chan³, ACO Cheng³, KL Yuen³, KW Tang¹

¹Department of Radiology and Imaging, ²Department of Surgery, Queen Elizabeth Hospital, Jordan, Hong Kong; ³Hong Kong Eye Hospital, Ma Tau Wai, Hong Kong

ABSTRACT

Objective: To review the safety, efficacy, and outcome of single-stage embolisation with n-butyl cyanoacrylate (n-BCA) followed by surgical resection for head and neck vascular malformations.

Methods: Medical records of patients who underwent single-stage embolisation with n-BCA followed by surgical resection for vascular malformations at the head and neck region between October 2011 and April 2015 were retrospectively reviewed.

Results: A total of 10 men and 17 women (mean age, 36.7 years) was included. Symptoms included disfigurement (n = 27), haemorrhage (n = 4), and visual impairment (n = 2). The mean lesion size in the largest dimension was 2.9 cm. Lesions were located in the scalp (n = 4), orbit (n = 11), oral cavity (n = 3), lip (n = 3), face (n = 4), and neck (n = 2). The mean operating time was 201 minutes. Five patients required further embolisation during surgery; 18 patients had complete resection and nine had partial resection. The mean length of hospital stay was 4.7 days; 22 patients had uneventful recovery and the other five had minor complications. The mean follow-up period was 20.6 months. All patients had good cosmetic outcome and symptom control. One patient who underwent partial resection had recurrent symptoms but required no further treatment.

Conclusions: Single-stage embolisation with n-BCA followed by surgical excision for management of vascular malformations of the head and neck region is safe and effective with minimal complications.

Key Words: Embolization, therapeutic; Endovascular procedures; Interdisciplinary communication; Vascular malformations

中文摘要

單階段栓塞和切除頭頸部血管畸形

蔣碧茜、馮漢盛、潘偉麟、陳志滿、梁偉業、廖思維、李琬微、陳頌恩、鄭智安、袁國禮、鄧國穎

目的：回顧使用氰基丙烯酸正丁酯（n-BCA）進行單階段栓塞然後手術切除頭頸部血管畸形的安全性、有效性和結果。

Correspondence: Dr JB Chiang, Department of Radiology and Imaging, Queen Elizabeth Hospital, Jordan, Hong Kong.
Email: jbchian@gmail.com

Submitted: 12 Apr 2016; Accepted: 4 Jul 2016.

Disclosure of Conflicts of Interest: All authors have no conflicts of interest to disclose.

Funding/Support: This research received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

方法：回顧分析2011年10月至2015年4月接受單階段n-BCA栓塞然後手術切除頭頸部血管畸形患者的病歷資料。

結果：納入10名男性和17名女性（平均年齡36.7歲）。症狀包括毀容（n = 27）、出血（n = 4）和視力受損（n = 2）。平均病變最大直徑為2.9厘米。病變位於頭皮（n = 4）、眼眶（n = 11）、口腔（n = 3）、唇部（n = 3）、面部（n = 4）和頸部（n = 2）。平均手術時間為201分鐘。五名患者在手術期間需要進一步栓塞。18例患者病變完全切除，9例有部分切除。平均住院時間為4.7天。22例患者康復，5例有輕微併發症。平均隨訪期為20.6個月。所有患者有良好的美容效果和症狀控制。只有一名患者切除部分病變後有復發性症狀但毋須進一步治療。

結論：單階段n-BCA栓塞然後手術切除頭頸部血管畸形安全有效並將併發症減至最低。

INTRODUCTION

Vascular anomalies are difficult to treat. According to the International Society for the Study of Vascular Anomalies,^{1,2} vascular anomalies are classified into vascular tumours and vascular malformations. The latter are further classified based on characteristics of channel (lymphatic, venous, or arteriovenous) and flow (slow flow, combined flow, or fast flow). These lesions are usually present at birth and continue to expand over time,³ and result in physical disfigurement, profuse haemorrhage, and complications caused by the mass effect such as orbital or airway compression. Treatment options include sclerotherapy, laser photocoagulation, embolisation, and surgical resection. Sclerotherapy and laser photocoagulation have been successful for slow-flow vascular malformations, but require multiple procedures and may result in sclerosant-induced inflammation and swelling.³⁻⁵ Laser therapy has been successful in superficial venous malformations, but has a high rate of recurrence.⁴ Surgical resection is the most definitive treatment, but is technically demanding and dangerous due to the risk of profuse intra-operative bleeding,³ particularly for vascular malformations in the head and neck.

Embolisation with n-butyl cyanoacrylate (n-BCA) followed by surgical resection significantly reduces intra-operative blood loss. n-BCA forms a firm consistency within the vascular malformations and results in a clear demarcation between the vascular malformations and healthy tissues.⁶ Safety and efficacy of embolisation with n-BCA have been reported⁶⁻¹²; most studies have described embolisation followed by delayed (days to weeks) surgical resection, and one study described same-stage embolisation (at the interventional radiology suite) followed by surgical resection (at the operation theatre).⁶ In this article, we

review the safety, efficacy, and outcome of single-stage embolisation with n-BCA followed by surgical resection for head and neck vascular malformations.

METHODS

This study was approved by the ethics committee of the Queen Elizabeth Hospital and conducted in compliance with the Declaration of Helsinki. We retrospectively reviewed the medical records of patients who underwent single-stage embolisation with n-BCA followed by surgical resection for vascular malformations at the head and neck region between October 2011 and April 2015 at Queen Elizabeth Hospital. Patients were referred to the multidisciplinary team for evaluation by interventional radiologists, paediatricians, and head and neck surgeons. Patients with imaging diagnosis of vascular tumours (e.g. haemangioma), vascular lesions with unclear diagnosis, vascular lesions not at the head or neck location, or use of other embolisation medium (e.g. contour particles) were excluded.

The hybrid procedure was performed under general anaesthesia in the endovascular operating room (Figure 1) by the multidisciplinary team. Preoperative ultrasonography, computed tomography, and magnetic resonance imaging were performed (Figures 2 and 3). Access to the vascular malformations was either by direct puncture with a 21-gauge butterfly needle or intra-arterially by the Marathon Flow Directed Microcatheter (Covidien; Irvine [CA], USA) and Mirage 0.008 Guidewire (Covidien; Irvine [CA], USA), depending on lesion type, site, and vessel characteristics. Venous malformations were accessed by the direct approach, whereas arteriovenous malformations were preferably accessed by the intra-arterial approach, particularly when they were deep and had few supplying arteries. Catheter position was confirmed with digital subtraction



Figure 1. Endovascular operation room.

(Figure 4), and with free back flow of blood in the case of direct puncture. Pretreatment angiography further delineated the margins and flow pattern of the vascular malformations for subsequent embolisation. The n-BCA / lipiodol mixture was prepared at 25% to 50% concentration depending on lesion location, size, and presence of draining vessels. The mixture was then injected under fluoroscopic guidance until complete filling, with the needle or catheter removed on completion. External compression to the draining veins was given if necessary. Surgical resection was then performed in the same room without patient transfer. Dynamic computed tomography may be performed intra-operatively to guide resection.

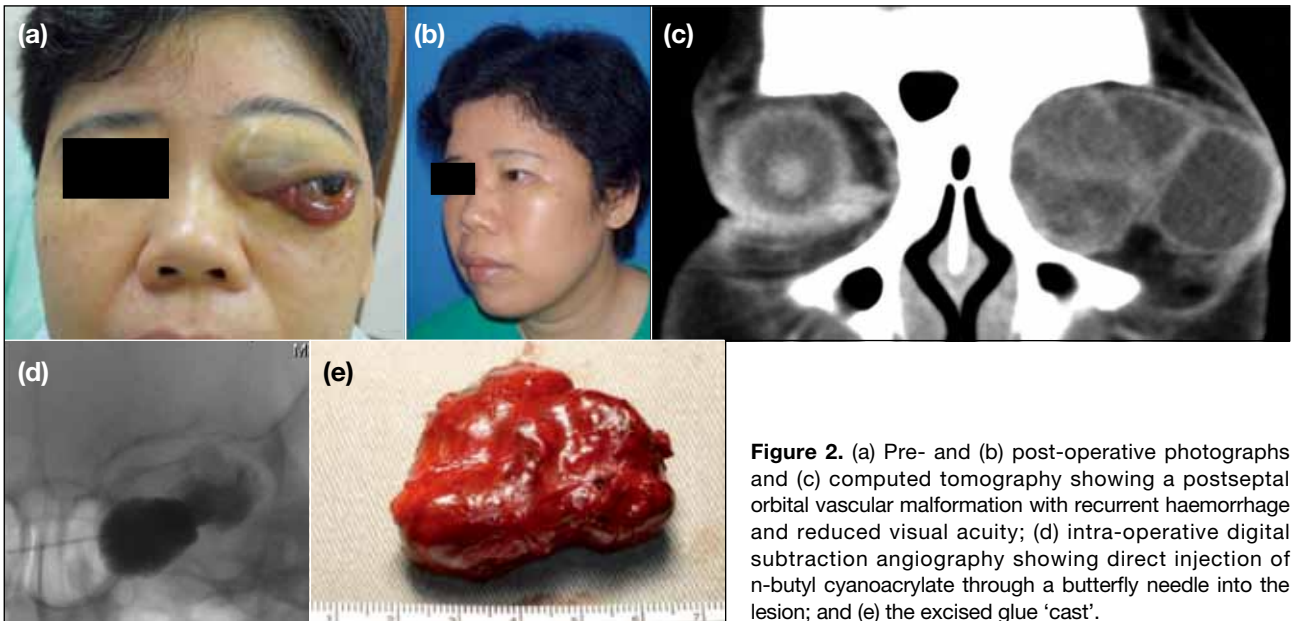


Figure 2. (a) Pre- and (b) post-operative photographs and (c) computed tomography showing a postseptal orbital vascular malformation with recurrent haemorrhage and reduced visual acuity; (d) intra-operative digital subtraction angiography showing direct injection of n-butyl cyanoacrylate through a butterfly needle into the lesion; and (e) the excised glue 'cast'.

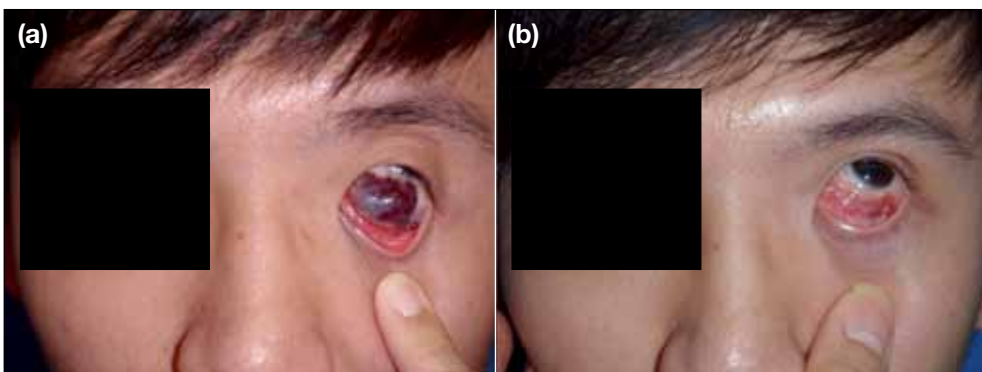


Figure 3. (a) Pre- and (b) post-operative photographs showing a preseptal venous malformation with recurrent haemorrhage.

RESULTS

A total of 10 male and 17 female patients were included (Table). Their mean age was 36.7 (range, 6-69) years and 20 patients had no prior treatment. Symptoms included disfigurement (n = 27, 100%), haemorrhage (n = 4, 15%), and visual impairment (n = 2, 7%). The mean

lesion size in the largest dimension was 2.9 (range, 1.4-6.0) cm. Lesions were located in the temporal (n = 3) or suboccipital (n = 1) scalp, preseptal (n = 7) or postseptal / intraconal (n = 2) orbit, tongue (n = 2), buccal (n = 1), lip (n = 3), chin (n = 1), forehead (n = 1), cheek (n = 1), neck (n = 2), eyelid (n = 2), and submandibular (n = 1).

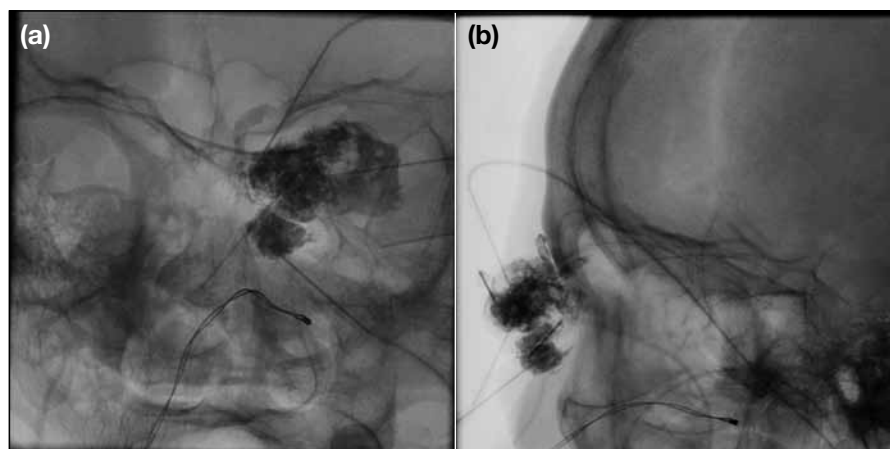


Figure 4. (a) Anteroposterior and (b) lateral digital subtraction angiography images after embolisation with n-butyl cyanoacrylate with butterfly needles in situ showing multiple compartments that may require repeat embolisations.

Table. Lesion characteristics and outcomes.

| Sex / age, y | Lesion location | Size, cm | Classification (pathological / radiological) | Operating time, min | Hospitalisation, d | Complication | Recurrence | Follow-up, mo |
|--------------|------------------------|----------|--|---------------------|--------------------|--|------------|---------------|
| M / 35 | Left orbit, preseptal | 2.2 | Slow-flow | 138 | 2 | – | – | 43 |
| F / 38 | Left orbit, preseptal | 4.2 | Slow-flow | 324 | 9 | Haematoma | – | 41 |
| M / 22 | Left orbit, preseptal | 2.6 | Lymphovascular | 161 | 3 | – | – | 42 |
| F / 69 | Left tongue | 1.9 | Venous | 82 | 3 | – | – | 40 |
| M / 51 | Right temporal | 3 | Arteriovenous | 264 | 5 | – | – | 38 |
| M / 61 | Right tongue | 2 | Slow-flow | 152 | 4 | – | – | 37 |
| F / 69 | Left orbit, preseptal | 2.1 | Venous | 171 | 5 | – | – | 32 |
| F / 49 | Right buccal | 2.1 | Venous | 140 | 5 | – | – | 30 |
| F / 62 | Suboccipital | 3.1 | Fast-flow | 277 | 4 | – | – | 29 |
| F / 23 | Left orbit, preseptal | 2.1 | Venous | 173 | 4 | Supraorbital nerve damage with mild paraesthesia | Local | 26 |
| M / 22 | Left temporal | 6 | Arteriovenous | 354 | 5 | – | – | 25 |
| F / 23 | Right chin | 2.5 | Slow-flow | 120 | 3 | – | – | 25 |
| M / 36 | Right temporal | 5.6 | Arteriovenous | 187 | 4 | Wound infection | – | 23 |
| F / 42 | Left neck | 1.5 | Slow-flow | 120 | 3 | – | – | 17 |
| M / 40 | Right orbit, preseptal | 3.7 | Arteriovenous | 330 | 7 | – | – | 17 |
| F / 18 | Back of neck | 2.8 | Venous | 140 | 4 | – | – | 16 |
| F / 17 | Left orbit, postseptal | 1.9 | Lymphatic | 300 | 9 | Diplopia | – | 13 |
| F / 39 | Right submandibular | 2.4 | Slow-flow | 145 | 3 | – | – | 11 |
| F / 6 | Left cheek | 3.7 | Slow-flow | 212 | 4 | – | – | 8 |
| F / 30 | Right lip | 4.6 | Slow-flow | 240 | 5 | – | – | 8 |
| F / 8 | Left lip | 3.5 | Slow-flow | 165 | 7 | – | – | 7 |
| M / 42 | Left lip | 2.6 | Arteriovenous | 166 | 3 | – | – | 7 |
| M / 33 | Left orbit, preseptal | 1.4 | Arteriovenous | 227 | 4 | – | – | 7 |
| F / 50 | Left intraconal | 4.9 | Slow-flow | 420 | 10 | Left ptosis | – | 6 |
| M / 21 | Right forehead | 1.7 | Arteriovenous | 155 | 4 | – | – | 4 |
| F / 40 | Left lower eyelid | 1.9 | Slow-flow | 99 | 5 | – | – | 3 |
| F / 45 | Left upper eyelid | 2.9 | Slow-flow | 179 | 3 | – | – | 1 |

The mean operating time was 201 (range, 82-420) minutes. Five (19%) patients required further embolisation during surgery, with negligible intra-operative blood loss (<10 ml). In all, 18 (67%) patients had complete resection and nine (33%) had partial resection.

The mean length of hospital stay was 4.7 (range, 2-10) days. In all, 22 patients had uneventful recovery. One patient had an orbital haematoma that later subsided. One patient had mild wound infection that resolved with regular dressing. One patient had intra-operative supraorbital nerve damage that was repaired in the same setting, with minimal paraesthesia. One patient had ptosis that improved on recovery. One patient developed diplopia when the vision was recovered after treatment for an intra-orbital lesion.

The mean follow-up period was 20.6 (range, 1-43) months. All patients had good cosmetic outcome and symptom control. Two patients with orbital lesions had improved visual acuity. No patient had imaging or clinical evidence of recurrence; only one patient who underwent partial resection had recurrent symptoms but required no further treatment.

DISCUSSION

The risk of profuse intra-operative haemorrhage owing to hypervascularity of lesions is potentially fatal and renders surgical resection difficult when the operative field is obscured. Preoperative embolisation reduces intra-operative blood loss and enables a bloodless operative field for resection and reconstruction. The use of n-BCA enables formation of a 'cast' and easy demarcation of the vascular malformations and normal tissue to facilitate surgical excision. In addition, n-BCA / lipiodol is compatible with intra-operative fluoroscopy or dynamic computed tomography. n-BCA causes minimal inflammation and is not associated with discolouration of skin or combustion with diathermy. In contrast, ethanol results in marked soft tissue inflammation and often precludes resection. Onyx (Medtronic; Minneapolis [MN], USA) is expensive and associated with bluish discolouration of the overlying skin,¹⁰ and intra-operative combustion when used with diathermy.¹³ In addition, Onyx has better permeation to smaller arterioles than n-BCA; its infiltrating nature may lead to increased complications.

The safety and efficacy of embolisation with n-BCA have been reported.⁶⁻¹² Embolisation with n-BCA

greatly reduces intra-operative blood loss and provides better visualisation of the surgical field.^{8,9} It enables delayed surgical resection of the cast within 10 to 15 days, as n-BCA results in intense soft tissue foreign body reaction that results in a pseudocapsule.⁷ Single-stage embolisation with n-BCA (in the interventional radiology suite) followed by surgical excision (in the operating theatre) provides the benefit of a single episode of anaesthesia.⁶ Acute inflammatory reaction related to n-BCA enables formation of a 'cast' and thus delayed surgery after soft tissue foreign body reaction does not seem to be necessary.⁶

Single-stage embolisation with n-BCA followed by surgical excision in the endovascular operating room allows multidisciplinary collaboration.¹⁴ Despite elaborate preoperative imaging and planning, incomplete infiltration of n-BCA within the vascular malformations is not uncommon. In one study, 22% of patients required multiple episodes of embolisation, each under general anaesthesia, before surgery.⁷ The endovascular operating room enables radiologists and surgeons to work simultaneously and allows multiple episodes of embolisation after surgical exposure. In our series, five (19%) patients required further embolisation during surgery. They usually had lesions that were larger and deeper with multiple compartments. Without multidisciplinary collaboration, these patients would have required several independent procedures, each under general anaesthesia, and possibly over a longer period. Digital subtraction angiography in the operating theatre enables intra-operative fluoroscopy and dynamic computed tomography to assess the residual glue cast and guide further resection.

CONCLUSIONS

Single-stage embolisation with n-BCA followed by surgical excision by a multidisciplinary team in the endovascular operating room for management of vascular malformations of the head and neck region is safe and effective with minimal complications.

REFERENCES

1. Enjolras O, Wassef M, Chapot R. Color Atlas of Vascular Tumours and Vascular Malformations Excerpt. New York: Cambridge University Press; 2007. [cross ref](#)
2. Mulliken JB, Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. *Plast Reconstr Surg.* 1982;69:412-22. [cross ref](#)
3. Richter GT, Friedman AB. Hemangiomas and vascular malformations: current theory and management. *Int J Pediatr.* 2012;2012:645678.

4. Burrows PE, Mason KP. Percutaneous treatment of low flow vascular malformations. *J Vasc Interv Radiol.* 2004;15:431-45. [crossref](#)
5. James CA, Braswell LE, Wright LB, Roberson PK, Moore MB, Waner M, et al. Preoperative sclerotherapy of facial venous malformations: impact on surgical parameters and long-term follow-up. *J Vasc Interv Radiol.* 2011;22:953-60. [crossref](#)
6. Tieu DD, Ghodke BV, Vo NJ, Perkins JA. Single-stage excision of localized head and neck venous malformations using preoperative glue embolization. *Otolaryngol Head Neck Surg.* 2013;148:678-84. [crossref](#)
7. Cil BE, Vargel I, Geyik S, Peynircioglu B, Cavusoglu T. Venous vascular malformations of the craniofacial region: pre-operative embolisation with direct percutaneous puncture and N-butyl cyanoacrylate. *Br J Radiol.* 2008;81:935-9. [crossref](#)
8. Lacey B, Rootman J, Marotta TR. Distensible venous malformations of the orbit: clinical and hemodynamic features and a new technique of management. *Ophthalmology.* 1999;106:1197-209. [crossref](#)
9. Couch SM, Garrity JA, Cameron JD, Cloft HJ. Embolization of orbital varices with N-butyl cyanoacrylate as an aid in surgical excision: results of 4 cases with histopathologic examination. *Am J Ophthalmol.* 2009;148:614-8. [crossref](#)
10. Arat A, Cil BE, Vargel I, Turkbey B, Canyigit M, Peynircioglu B, et al. Embolization of high-flow craniofacial vascular malformations with onyx. *AJNR Am J Neuroradiol.* 2007;28:1409-14. [crossref](#)
11. Han MH, Seong SO, Kim HD, Chang KH, Yeon KM, Han MC. Craniofacial arteriovenous malformation: preoperative embolization with direct puncture and injection of n-butyl cyanoacrylate. *Radiology.* 1999;211:661-6. [crossref](#)
12. Cheng AC, Li EY, Chan TC, Wong AC, Chan PC, Poon WW, et al. Hybrid procedure for orbital venous malformation in the endovascular operation room. *Eye (Lond).* 2015;29:1069-75. [crossref](#)
13. Smith SJ, Thomas A, Ashpole RD. Intra-operative combustion of Onyx embolic material. *Br J Neurosurg.* 2009;23:76-8. [crossref](#)
14. Kubiena H, Cejna M, Kreuzer S, Frey E, Schoder M, Frey M. Interdisciplinary management of craniofacial vascular malformations. *Eur J Past Surg.* 2007;30:81-6. [crossref](#)