# **Surgical Neurology International**

SNI: Cerebrovascular, a supplement to Surgical Neurology International

OPEN ACCESS

For entire Editorial Board visit : http://www.surgicalneurologyint.com Editor:

James I. Ausman, MD, PhD University of California, Los Angeles, CA, USA

## Intracranial capillary hemangioma in an elderly patient

Ai Okamoto, Ichiro Nakagawa, Ryosuke Matsuda, Fumihiko Nishimura, Yasushi Motoyama, Young-Su Park, Mitsutoshi Nakamura, Hiroyuki Nakase

Department of Neurosurgery, Nara Medical University, Nara, Japan

E-mail: Ai Okamoto - himawari.aiai@gmail.com; \*Ichiro Nakagawa - nakagawa@naramed-u.ac.jp; Ryosuke Matsuda - cak93500@pop02.odn.ne.jp; Fumihiko Nishimura - fnishi@naramed-u.ac.jp; Yasushi Motoyama - myasushi@naramed-u.ac.jp; Young-Su Park - park-y-s@naramed-u.ac.jp; Mitsutoshi Nakamura - mnaka@grandsoul.co.jp; Hiroyuki Nakase - nakasehi@naramed-u.ac.jp \*Corresponding author

Received: 05 May 15 Accepted: 16 September 15 Published: 23 October 15

### **Abstract**

**Background:** Capillary hemangiomas are neoplasms involving skin and soft tissue in infants. These lesions rarely involved an intracranial space and reported age distribution ranges from infancy to middle age. We report an extremely rare case of rapidly rising intracranial capillary hemangioma in an elderly woman.

Case Description: The 82-year-old woman presented with vomiting, reduced level of consciousness, and worsening mental state. Computed tomography showed a contrast-enhanced extra-axial lesion in the left frontal operculum, although no intracranial mass lesion was identifiable from magnetic resonance imaging taken 2 years earlier. Complete surgical excision was performed and histopathological examination diagnosed benign capillary hemangioma consisting of a variety of dilated capillary blood vessels lined by endothelial cells.

**Conclusion:** This is the first description of rapid growth of an intracranial capillary hemangioma in an elderly woman. These lesions are exceedingly rare in the elderly population, but still show the capacity for rapid growth. Complete excision would prevent further recurrence.

Key Words: Capillary hemangioma, elderly, intracranial

# Access this article online Website: www.surgicalneurologyint.com DOI: 10.4103/2152-7806.168066 Quick Response Code:

### INTRODUCTION

Capillary hemangiomas are benign vascular tumors that involve skin and soft tissues. Although usually located on the face or scalp, they may appear anywhere in the body. These lesions often occur at birth or in early infancy. When located in the intracranial space, they usually manifest in infancy to childhood in males or adolescence to adulthood in females. [3] We report an extremely rare case of intracranial capillary hemangioma in an elderly woman and discuss with reference to the literature.

### **CLINICAL REPORT**

An 82-year-old woman presented with vomiting, reduced level of consciousness, and worsening mental state.

Mini-mental state examination (MMSE) score was 4/30, and the revised Hasegawa's Dementia Scale (HDS-R) score was 2/30 on admission to our department. The HDS-R stresses the weight of memory and verbal fluency more than the MMSE and is superior to the MMSE for cognitive screening of early Alzheimer's disease.

This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

How to cite this article: Okamoto A, Nakagawa I, Matsuda R, Nishimura F, Motoyama Y, Park YS, et al. Intracranial capillary hemangioma in an elderly patient. Surg Neurol Int 2015;6:S539-42.

http://surgicalneurologyint.com/Intracranial-capillary-hemangioma-in-an-elderly-patient/

Computed tomography (CT) showed an iso-dense mass lesion without calcification or hemorrhagic changes, and contrast enhancement demonstrated a lesion about 40 mm in diameter in the left frontal operculum. On magnetic resonance imaging (MRI), the lesion appeared hypointense on Tl-weighted imaging [Figure 1a], hyperintense with surrounding vasogenic edema on T2-weighted imaging [Figure 1b], and clear contrast enhancement with attachment to the dura mater (dural with homogeneous enhancement sign) Tl-weighted imaging [Figure 1c]. Two years before this admission, she received screening MRI because she suspected transient ischemic attack. Axial T2-weighted imaging had not demonstrated any lesions in the left frontal operculum [Figure 1d]. Blood examination showed no clear abnormalities, including tumor marker levels. Differential diagnoses were extra-axial tumors such as meningioma, hemangiopericytoma or solitary fibrous tumor, or metastatic tumor to the dura. The patient was advised to undergo surgery to resect and diagnose the lesion, as well as prevent further progression.

reddish, Intraoperatively, the tumor appeared presented with a rubbery consistency, and was highly vascularized [Figure 2]. Complete surgical excision was performed without any vascular injury to tissues surrounding the tumor. Pathological examination with hematoxlyin and eosin revealed no nuclear atypia, significant mitotic activity in greatly dilated capillary vessels lined by endothelial cells, and no necrosis [Figure 3a]. With positive findings for vascular antigens CD31 [Figure 3b], CD34, smooth muscle antigen, CD163, and capillary hemangioma was diagnosed. Ki-67 was about 7% [Figure 3c]. Epithelial

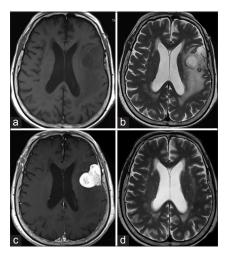


Figure 1: On magnetic resonance imaging, the lesion appears hypointense on T1-weighted imaging (a) and hyperintense with surrounding perifocal edema on T2-weighted imaging (b). Homogeneous enhancement with dural tail sign is seen on T1-weighted imaging (c). Axial T2-weighted imaging 2 years before admission did not suggest any mass lesion in the left frontal operculum (d)

membrane antigen and glial fibrillary acidic protein yielded negative results. The diagnosis was capillary hemangioma.

Postoperative CT showed no hemorrhage [Figure 4a] and the patient did well postoperatively without neurological deficits. Mental status improved moderately, with the MMSE score of 14/30 and the HDS-R score of 7/30 at 1 month postoperatively. No adjuvant treatment was performed after surgery. Disappearance of perifocal edema was shown [Figure 4b], and no recurrence was demonstrated on gadolinium-enhanced MRI [Figure 4c] at 2 months after the operation.

### **DISCUSSION**

This is the first report of intracranial capillary hemangioma developing rapidly in an elderly woman. Capillary hemangioma is considered to be a hamartomatous lesion, that is, commonly found in the skin and soft tissues of infants.<sup>[4]</sup> Capillary hemangiomas are benign neoplasms but display a tendency to grow rapidly. [2] These lesions are rarely found in the intracranial space and are usually identified infancy to childhood in males or adolescence to adulthood in females.[3] In the literature, no reports have described intracranial capillary hemangioma in elderly patients. Phi et al. reported a significant difference in age at diagnosis between sexes.[3] The median age for male patients was 4.8 years (range, 6 weeks to 20 years) and the median age for female patients was 22.5 years (range, 4 months to 44 years). Pregnancy and hormonal changes have been observed to be related to periods of rapid growth in capillary hemangiomas. This may be explained by an increase in tumor size caused by increased cardiac output and fluid retention during pregnancy because another study has shown that capillary hemangioma lacks estrogen and progesterone receptors. [1,4] In the present case, intracranial capillary hemangioma showed rapid

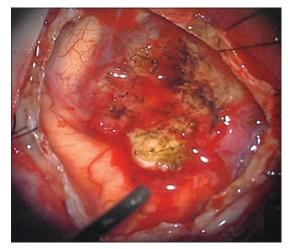


Figure 2:An intraoperative photograph shows an extra-axial mass located frontal operculum

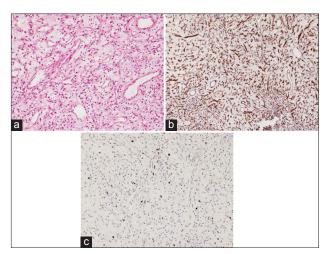


Figure 3: Pathological examination with H and E reveals no nuclear atypia, significant mitotic activity in many capillary vessels and no necrosis (a). Positive results for vascular antigens CD31 (b). CD34 and smooth muscle antigen, and CD163 led to the diagnosis of capillary hemangioma. Ki-67 was about 7% (c). Magnification, ×400

growth within 2 years in an elderly woman. Furthermore, no evidence of head trauma, central nervous infection, or hormonal abnormality were seen during this period. One possibility is that a very tiny occult lesion present from a young age may have undergone rapid development due to triggers such as minor bleeding, cytokines, inflammatory materials, and hormones.

Radiologically, capillary hemangiomas present circumscribed hypervascular lesions that meningioma, hemangiopericytoma, highly vascular metastatic lesions. Meningiomas show some radiological features in common with intracranial capillary hemangiomas. Both present as low-intensity areas on T1-weighted imaging and high-intensity areas on T2-weighted imaging, with intense gadolinium enhancement, whereas multiple flow-voids, intralesional hemorrhage, and the absence of a dural tail or bone changes are described as typical findings in capillary hemangiomas.[2] In this case, however, dural tail sign was demonstrated, and none of the typical features of capillary hemangioma applied. Prolonged retention of contrast in capillary hemangioma as an angiographic appearance would suggest the correct diagnosis to the radiologist.

The diagnosis of intracranial capillary hemangiomas is based on histochemical staining or in combination with immunostaining for vascular antigen CD34, CD31, F-γ-related antigen, vascular endothelial growth factor, and smooth muscle antigen. Negative immunostaining for epithelial and melanocytic markers excludes metastases, and immunostaining for estrogen and progesterone receptors also yield negative results. In this case, testing was conducted for CD34, CD31, and

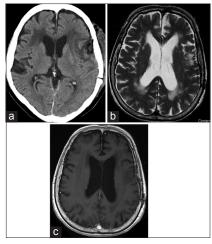


Figure 4: No hemorrhagic or ischemic changes are evident on postoperative computed tomography (a). At 2 months postoperatively, perifocal edema has disappeared (b) and no recurrence of any gadolinium-enhanced mass lesion is evident (c)

smooth muscle antigen. Strong positive results were seen for CD34 and smooth muscle antigen, identifying vascular endothelial cells. Numerous capillary vessels were found and showed oval cells. CD163 also displayed positive results, so almost of the oval cells may have been histiocytes.

The majority of common cutaneous capillary hemangiomas spontaneously regress around school age, so treatment can be reserved for large, disfiguring, or symptomatic lesions. In intracranial capillary hemangiomas, complete resection is central to the treatment of symptomatic lesions, and use of adjuvant radiotherapy should be considered for unresectable or incompletely resected lesions. [4] In cases where the lesion has been totally resected, outcomes have been uniformly excellent. No recurrences were observed after complete resection while lesions progressed within 3–6 months if incompletely resected. [3] Long-term, event-free, and progression-free survival may thus be achieved with complete resection alone.

### **CONCLUSIONS**

This is the first description of rapid growth of an intracranial capillary hemangioma in an elderly woman. These lesions are exceedingly rare in the elderly population, but still show the capacity for rapid growth. Complete excision seems most likely to prevent further recurrence.

Financial support and sponsorship Nil.

### **Conflicts of interest**

There are no conflicts of interest.

### **REFERENCES**

- Mirza B, Shi WY, Phadke R, Holton JL, Turner C, Plant GT, et al. Strawberries on the brain – Intracranial capillary hemangioma: Two case reports and systematic literature review in children and adults. World Neurosurg 2013;80:900.e13-21.
- 2. Morace R, Marongiu A, Vangelista T, Galasso V, Colonnese C, Giangaspero F, et al.
- Intracranial capillary hemangioma: A description of four cases. World Neurosurg 2012;78:191.E15-21.
- Phi JH, Kim SK, Cho A, Kim DG, Paek SH, Park SH, et al. Intracranial capillary hemangioma: Extra-axial tumorous lesions closely mimicking meningioma. J Neurooncol 2012;109:177-85.
- Simon SL, Moonis G, Judkins AR, Scobie J, Burnett MG, Riina HA, et al. Intracranial capillary hemangioma: Case report and review of the literature. Surg Neurol 2005;64:154-9.