

## Intracranial capillary hemangioma: extra-axial tumorous lesions closely mimicking meningioma

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**Abstract** Capillary hemangiomas are common tumorous lesions of the skin and soft tissue in infants. These lesions often involve internal organs and rarely develop in the intracranial space. Because of their rarity, clinical descriptions of intracranial capillary hemangioma have been anecdotal and have not provided a coherent understanding of these lesions. We report four cases of intracranial capillary hemangioma. Review of these 4 cases and 14 cases reported in the literature was undertaken to assess the influence of age, sex, location, clinical manifestation, treatment, and outcome. A significant difference was observed in age at diagnosis between sexes. 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). Approximately two-thirds of intracranial capillary hemangioma lesions develop in the vicinity of major venous sinuses, such as the cavernous/sphenoparietal sinus and the transverse sinus/torcular/superior sagittal sinus. This propensity for specific

locations appears to be responsible for the symptom manifestation. The majority of the lesions also seem to be extra-axial in imaging and operative findings. Complete surgical resection provided excellent outcome, but incomplete resection led to recurrence. Capillary hemangioma should be considered in the diagnosis of extra-axial, contrast-enhancing lesions, especially in children and adolescents.

**Keywords** Capillary hemangioma · Intracranial · Age · Location · Venous sinus

### Introduction

Capillary hemangioma is a benign vascular tumor or tumor-like lesion that usually develops in the skin and soft tissues [1]. Capillary hemangioma is sometimes found in the eyes [2], abdominal organs [3], cauda equina [4], and spinal cord [5]. Intracranial manifestation of capillary hemangioma is rare, and only 14 patients have been reported in the literature [6, 7]. The intracranial location of the lesions in these reports was diverse and included the supratentorial to infratentorial spaces. Although some authors have described the lesion as having a purely intra-axial location [8], the majority of authors have observed an extra-axial lesion adjacent to a venous sinus. The age of the reported patients also showed a wide distribution, ranging from infants to the middle-aged. Therefore, despite the seemingly uniform histology, clinical pictures of this disease remain diverse and unfocused. Here, we report four patients with intracranial capillary hemangioma. Interestingly, all of the patients had lesions that mimicked extra-axial tumors, such as meningioma. Based on our experience and review of the literature, we propose that a

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bimodal age/sex distribution exists for intracranial capillary hemangioma and that capillary hemangioma should be included in the differential diagnosis for meningioma-like tumors in male children.

## Patients and methods

Clinical databases at the Division of Pediatric Neurosurgery, Seoul National University Children's Hospital and the Department of Neurosurgery, Seoul National University Hospital were searched for intracranial capillary hemangioma diagnoses between 1998 and 2011. Four patients were diagnosed with intracranial capillary hemangioma during this period. Clinical records, radiological images, and pathological slides were retrospectively reviewed. For the literature review, relevant publications in the PubMed database (<http://www.ncbi.nlm.nih.gov/pubmed/>) published during the period from January 1990 through October 2011 were identified using the search terms "capillary hemangioma," "intracranial," "central nervous system," and "venous sinus." Only articles published in English were included for review. Articles that discussed capillary hemangioma arising from the spinal cord, spinal nerve root, or cauda equina were excluded. Serial reports involving the same patients were cautiously selected, and duplicated cases were discarded. We summarized the age, sex, symptoms and signs, lesion location, imaging characteristics, treatment, and outcome of the reported patients. This study protocol was approved by the Institutional Review Board of Seoul National University Hospital.

## Results

### Case I

An 8-year-old boy developed headache, nausea, vomiting, and decreased visual acuity. Brain computed tomography (CT) and magnetic resonance imaging (MRI) showed a lobular mass in the right occipital lobe involving the right tentorium and transverse sinus. Intralesional hemorrhage, multiple signal voids, and extensive perilesional edema were noted. The mass was hypometabolic on  $^{18}\text{F}$ -fluorodeoxyglucose positron emission tomography (FDG-PET) but was hypermetabolic in  $^{11}\text{C}$ -methionine PET (Fig. 1). Radical resection was performed. The mass appeared dark red in color and hypervascular. A well-developed arachnoid plane was present, but the mass was tightly attached to the transverse sinus. The mass was removed en bloc; however, the dural attachment to the transverse sinus was left but was meticulously coagulated. Pathological examination revealed a capillary hemangioma. The patient recovered without

neurological deficits. A follow-up MRI at postoperative month 24 showed no evidence of recurrence.

### Case II

A 13-year-old boy developed worsening headache in the morning. Brain CT and MRI revealed a huge mass in the right temporo-occipital area, occupying both supratentorial and infratentorial spaces (Fig. 2). A peritumoral cyst and extensive brain edema were noted. Angiography indicated that the mass was hypervascular and fed by ipsilateral middle meningeal and occipital arteries.

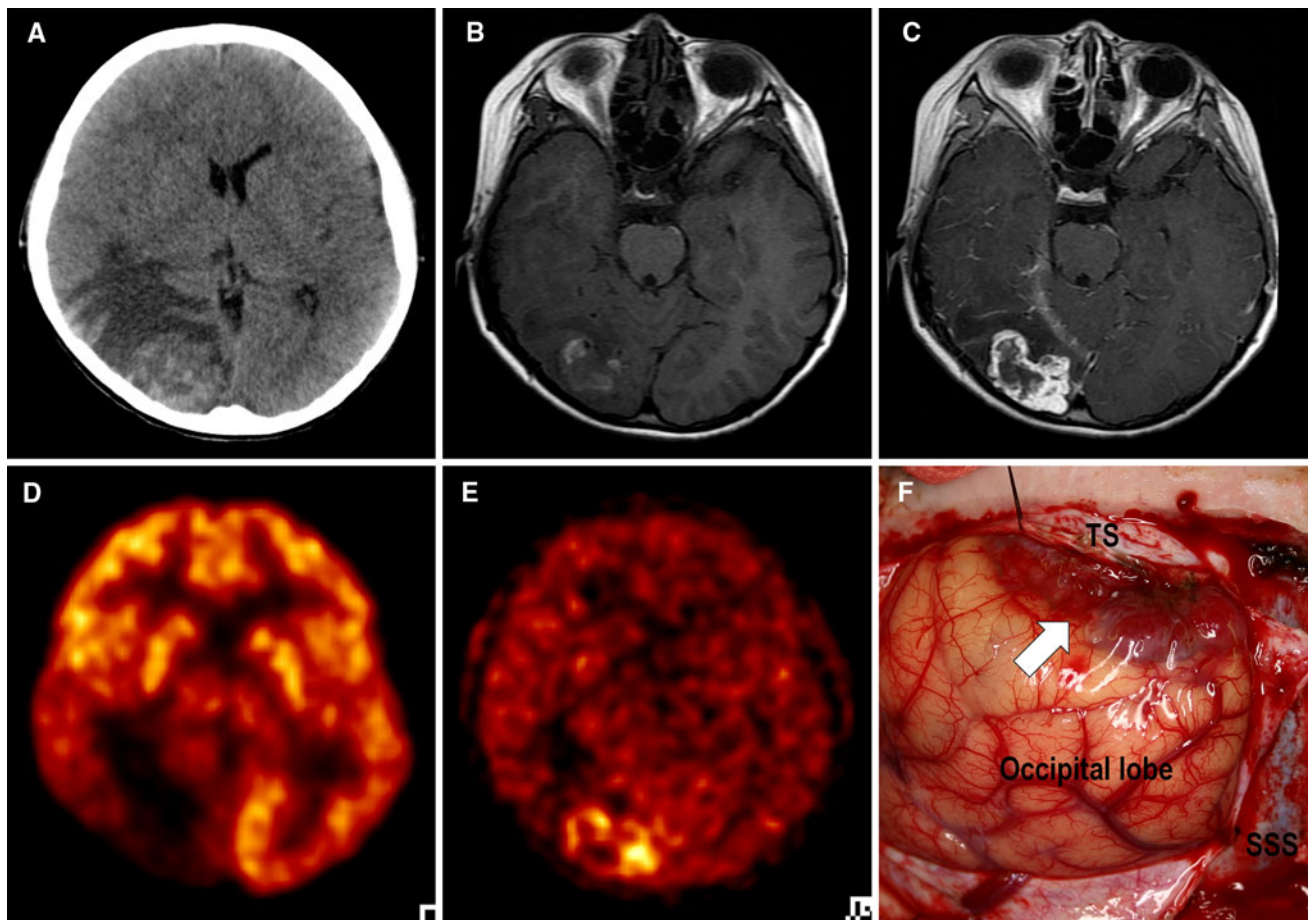
The mass was extra-axially located, attached to the transverse and superior petrosal sinuses, and crossed the tentorium into the posterior fossa. The mass was removed in pieces along with the tentorial attachment, and the involved sinus wall was meticulously coagulated. The patient displayed no postoperative neurological deficits. However, postoperative MRI showed a tiny enhancing nodule along the transverse sinus. Pathological examination confirmed the diagnosis of capillary hemangioma. After 3 months, the residual mass had increased slightly in size on MRI. The patient is under close observation for potential further treatment.

### Case III

A 30-year-old female developed worsening headache, vomiting, and vertigo. Brain MRI showed a lobular mass attached to the tentorium in the posterior fossa (Fig. 3). Strong contrast enhancement and edema in the cerebellum were observed. The tumor was surgically approached via the cerebellopontine angle. The mass was pinkish in color and extra-axial in origin. Due to massive bleeding, the mass was thoroughly coagulated and completely resected. The tentorial surface attached to the mass was also resected. The patient displayed no postoperative neurological deficits. Pathological examination confirmed the diagnosis of capillary hemangioma. The Ki-67 index was 2.0 %. A follow-up MRI performed 30 months later showed no evidence of recurrence.

### Case IV

A 44-year-old female developed progressive visual loss and field cut in the right eye. Only finger count was possible with her right eye, and three-fourths of the visual field was lost in the right eye. Brain MRI showed a huge contrast-enhancing mass in the ethmoid and sphenoid sinuses. The mass had extended into the right cavernous sinus, encasing the right internal carotid artery and compressing the right optic nerve and optic chiasm (Fig. 4). A small hemorrhagic focus was observed in the mass.



**Fig. 1** Images from an 8-year-old boy (case I). **a** Brain CT scan reveals a mass in the right occipital lobe and extensive surrounding brain edema. **b** Intraslesional hemorrhage is present in a nonenhanced T1-weighted axial image. **c** The lesion is well enhanced by

gadolinium. **d** The lesion is hypometabolic on FDG-PET, **e** but mildly hypermetabolic on  $^{11}\text{C}$ -methionine PET. **f** An intraoperative photograph. The mass (*arrow*) is dark red and extra-axially located. TS transverse sinus, SSS superior sagittal sinus

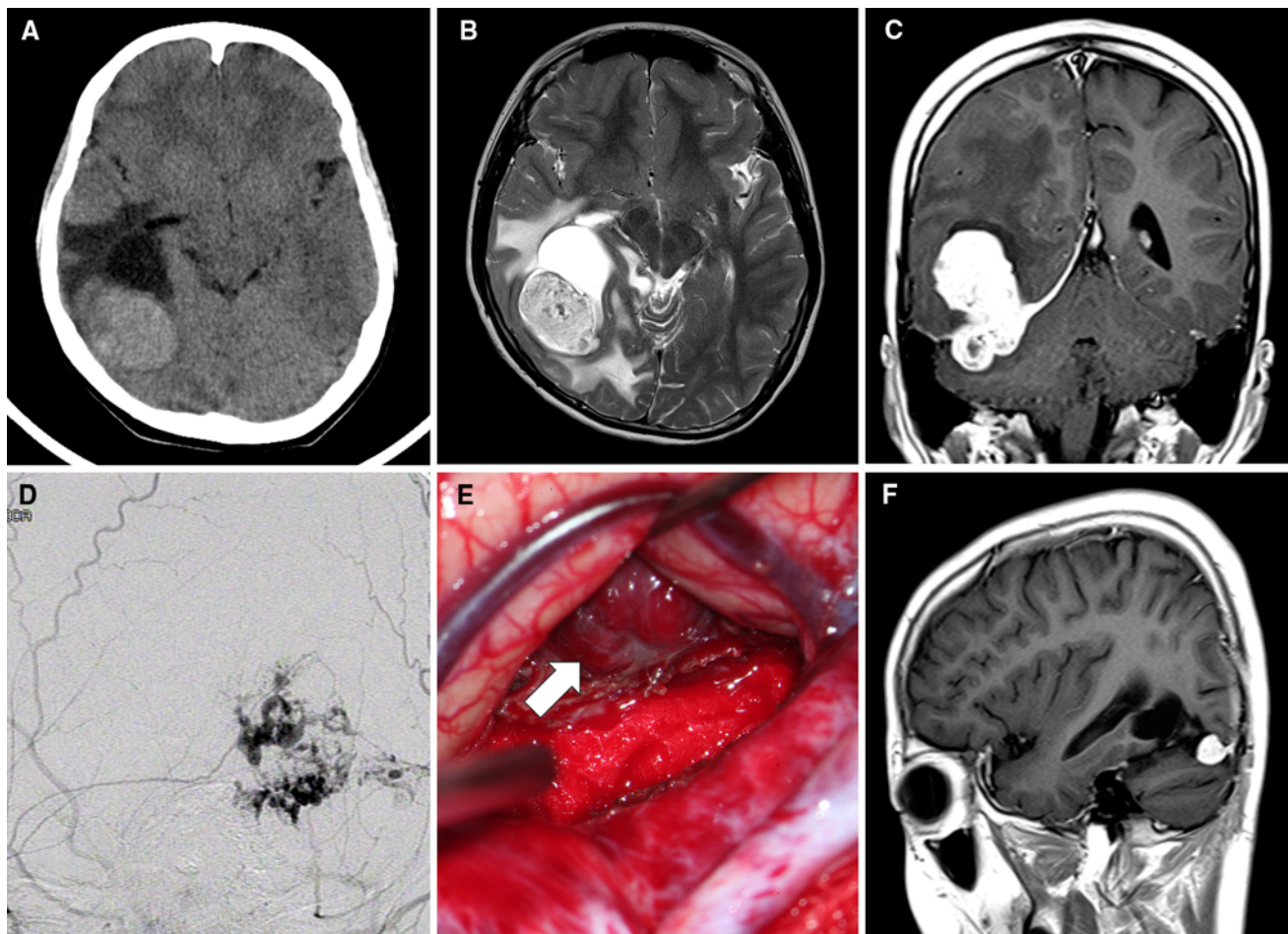
A transsphenoidal approach was attempted. The mass was hypervascular and bled profusely upon resection. Due to massive bleeding, the mass was only partially resected. The pathological diagnosis was capillary hemangioma with a high Ki-67 index (15.9 %). The patient received radiation therapy (5,400 cGy to the mass). A follow-up MRI performed 5 months after radiation therapy showed a marked decrease in the mass size. Her visual acuity of the right eye was 0.5, and the visual field examination was normal. Further shrinkage of the residual tumor was observed 23 months after radiation therapy.

#### Literature review

From the literature search, we found 11 articles dealing with intracranial capillary hemangioma, in which 14 cases were reported [6–16]. With the 4 new cases in the present study, literature review was performed in 18 patients (Table 1). The male-to-female ratio was 8:10. The age distribution was wide, ranging from infant (6 weeks) to

middle-aged (44 years). A significant difference was observed in age at diagnosis between the sexes ( $P = 0.023$ , Wilcoxon signed-rank test; Fig. 5). 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). Specifically, all male patients (except two) were under 10 years old, whereas all female patients (except one) were 10 years old or older.

The lesion location was diverse but could be categorized into the following three locations: (1) middle fossa–sphenoparietal/cavernous sinus–sellar region (six patients), (2) tentorium–transverse sinus/torcular/superior sagittal sinus (seven patients), and (3) miscellaneous (subarachnoid space, ventricle, cerebrum, and cerebellum) (five patients). The growth pattern could be dichotomized into intra-axial and extra-axial. However, whether the lesion was situated intra-axially or extra-axially was not clear in many reports. Therefore, if the description was ambiguous but images disclosed a lesion typical of an extra-axial mass, we classified the case as “extra-axial or cortical.” The other



**Fig. 2** Images from a 13-year-old boy (case II). **a** Brain CT scan reveals a mass in the right temporo-occipital lobes with cyst and brain edema. **b** Multiple signal voids are present in the lesion in a T2-weighted axial image. **c** The lesion is well enhanced by gadolinium. **d** The lesion is hypervascular and is supplied by branches of the

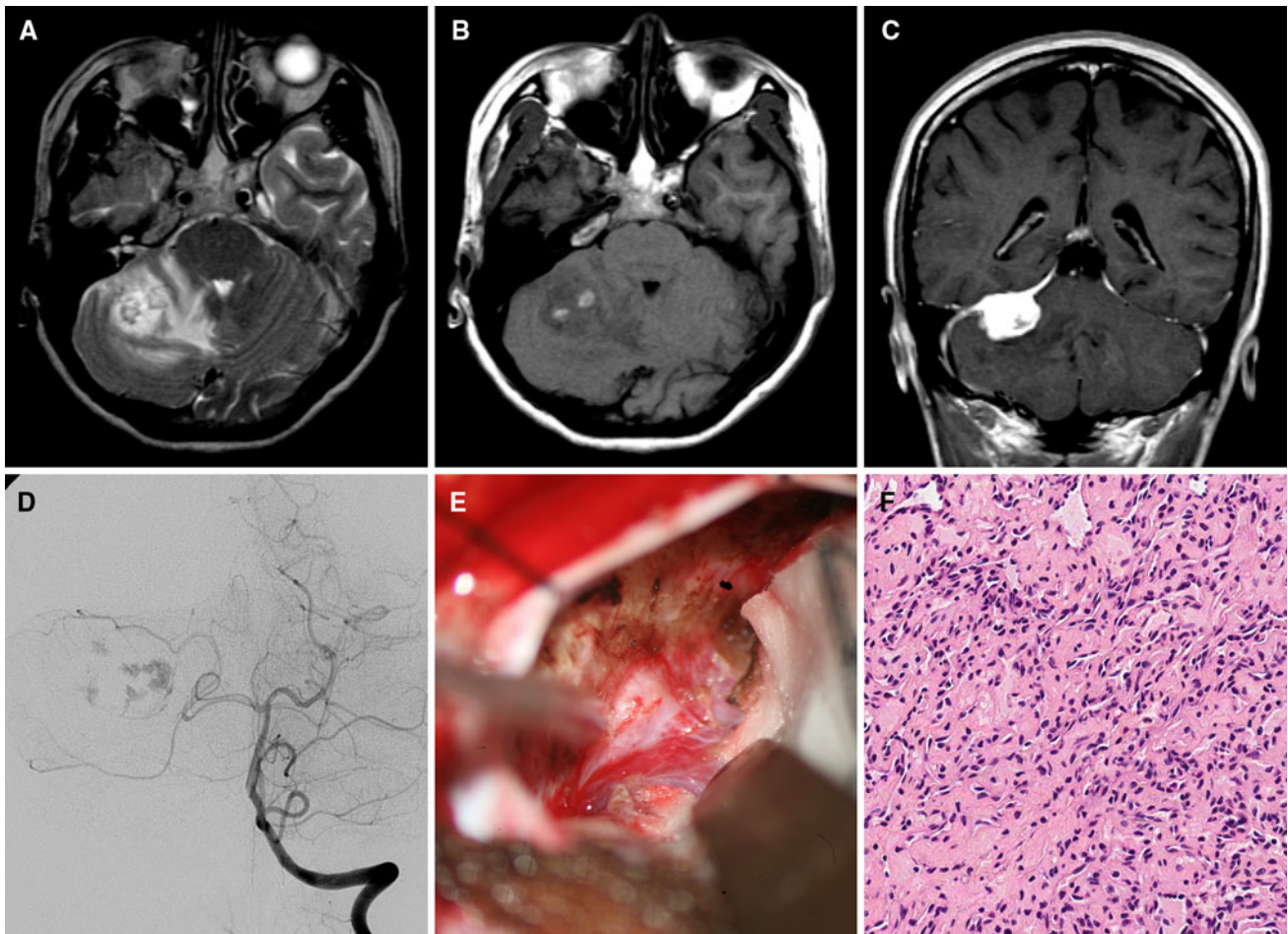
external carotid artery. **e** An intraoperative photograph shows an extra-axial mass (*arrow*) underneath the temporo-occipital lobes. The mass has been detached from the tentorium and superior petrosal sinus. **f** A growing residual mass on MRI taken at postoperative month 3

unambiguous cases were classified as either extra-axial or subcortical. The lesion was obviously subcortical in only two patients. All of the other cases were categorized as definite extra-axial (12 patients) or ambiguous (extra-axial or cortical; 4 patients). The majority of patients had symptoms or signs of increased intracranial pressure, such as headache, nausea, vomiting, or papilledema. Cranial nerve compression was found in five patients, and all patients with cavernous sinus involvement presented with cranial nerve palsies. The lesion was incidentally found in two patients. In imaging findings, intralesional bleeding, multiple signal voids, cyst formation, and cerebral edema were common. These findings were associated with presentation due to increased intracranial pressure. Thirteen patients received radical surgery. If the lesion was totally resected, the outcome was excellent. No recurrence was observed in 11 patients for a median of 2.5 years (range 4 months to 15 years) after complete resection. However,

the lesion progressed within 3–6 months if incompletely resected (two patients). Three patients with cavernous sinus involvement received biopsy/partial resection followed by radiation therapy (4,500–5,400 cGy). The outcome was also excellent with complete or partial response and no recurrence.

## Discussion

Capillary hemangioma is considered to be a hamartomatous lesion that is commonly found in the skin and soft tissues of infants [17]. Although the incidence is very low, capillary hemangioma can be found in the intracranial space. Intracranial capillary hemangioma behaves much like bona fide brain tumors, eliciting brain edema and cranial nerve compression which result in increased intracranial pressure and neurological deficits. Literature review



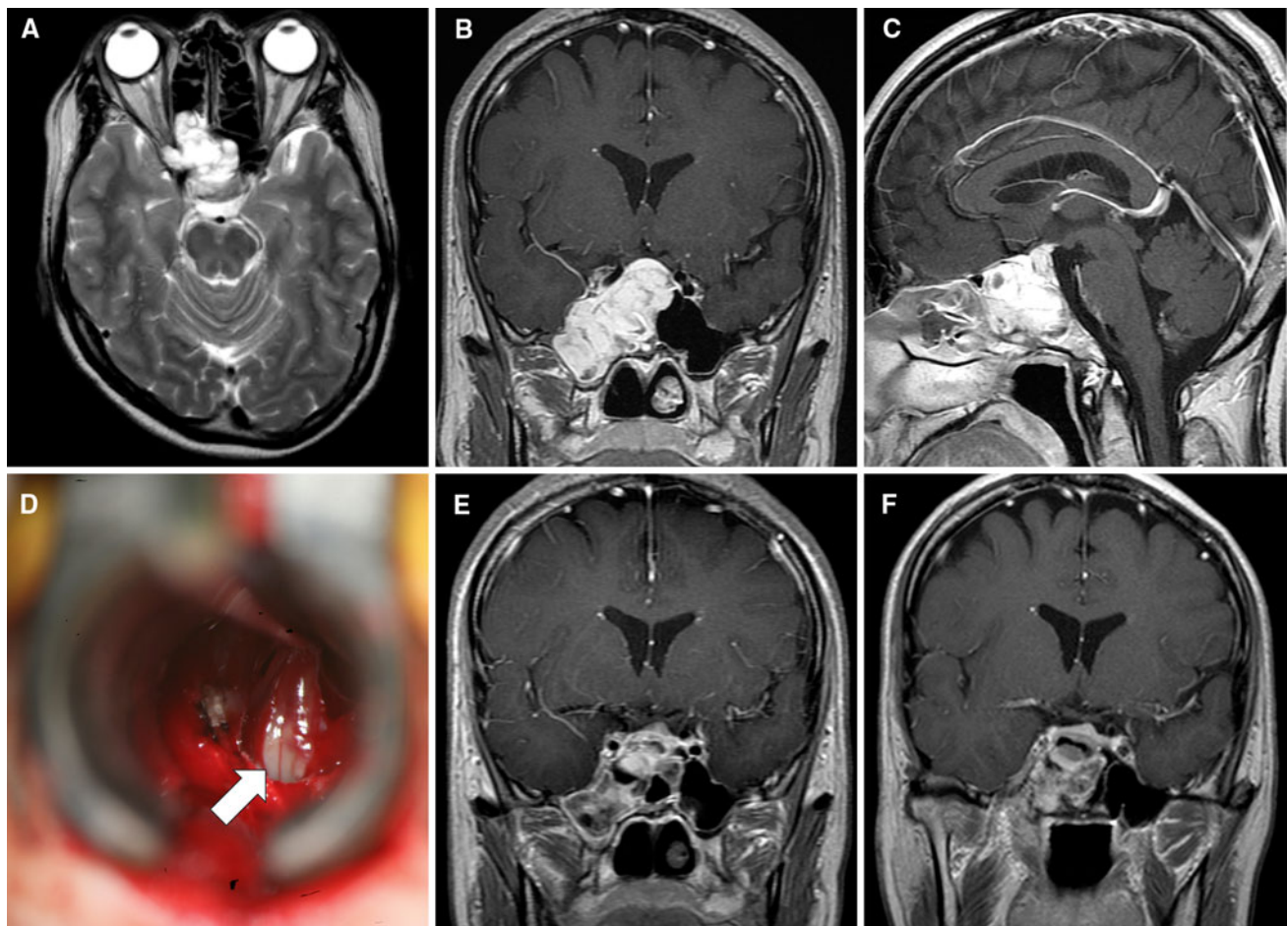
**Fig. 3** Images from a 30-year-old female (Case III). **a** A dural-based mass is found in the right cerebellopontine angle. The lesion is surrounded by brain edema. **b** Intralesional hemorrhage is observed in a nonenhanced T1-weighted image. **c** Dural tail sign is noted. **d** The lesion is hypervascular and is supplied by the superior cerebellar

artery. **e** An intraoperative photograph shows a *pinkish* mass attached to the tentorium. **f** The mass is composed of variably dilated capillary blood vessels which are lined by endothelial cells and pericytes [hematoxylin and eosin (H&E),  $\times 200$ ]

demonstrated that approximately two-thirds of capillary hemangiomas develop in the vicinity of major venous sinuses, such as the cavernous/sphenoparietal sinus and transverse/torcular/superior sagittal sinus. This propensity for specific locations appears to be responsible for the symptom manifestation in the patients. The majority of the lesions also seem to be extra-axial in imaging and operative findings. Therefore, these lesions can be easily mistaken for meningioma or hemangiopericytoma [11, 15]. The presence of multiple flow voids on MR images, intralesional hemorrhage, and the absence of a dural tail or bone changes are imaging characteristics of intracranial capillary hemangiomas that help differentiate the lesions from meningiomas [15]. Involvement of cavernous sinus or tentorial region is also rare for childhood meningiomas, which usually develop in convexity or ventricles [18]. However, these imaging features are not highly specific, and there are virtually no pathognomonic signs of

intracranial capillary hemangiomas [13, 15]. The four patients reported in the present study were also presumed to have meningioma or hemangiopericytoma preoperatively. Two of them had even dural tail signs. Only during the operation was other diagnosis strongly suspected because these lesions were less fibrous than usual meningiomas and because intraoperative frozen biopsies indicated vascular lesions rather than meningiomas or hemangiopericytomas. These suspicions led us to not resect the sinus walls opposing the lesions after complete removal of the extra-axial masses.

The most interesting finding revealed in the literature review was the different age distribution between sexes. The median age for male patients was significantly younger than that of female patients. Simply put, intracranial capillary hemangioma manifests itself mainly in male infancy to childhood and in female adolescence to adulthood. This finding is important in pediatric oncology because



**Fig. 4** Images from a 44-year-old female (case IV). **a** A mass of hyperintense signal intensity in T2-weighted images is situated in the ethmoid and sphenoid sinuses and encroaches into the right optic canal. **(b, c)** Gadolinium-enhanced T1-weighted images. The mass involves the right cavernous sinus and compresses the optic chiasm.

**d** An intraoperative photograph during a transsphenoidal approach. A *whitish* mass (*arrow*) is seen through the speculum. **e** MRI performed at 5 months after radiation therapy. **f** MRI performed at 23 months after radiation therapy

meningioma and hemangiopericytoma are also extremely rare in children, especially those under 10 years old. If clinicians detect a well-enhanced, dural-based, extra-axial mass on neuroimaging in children, they should consider the possibility of capillary hemangioma.

The majority of common cutaneous capillary hemangiomas exhibit a self-limited course and spontaneously regress around school age [1, 17]. Therefore, treatment can be reserved for large, disfiguring, or symptomatic lesions. Abe et al. [8] have reported the curious spontaneous regression of multiple intracranial capillary hemangiomas after resection of the largest or symptomatic lesions in two patients. However, all reported intracranial capillary hemangiomas were symptomatic and required urgent treatment, except for two young infants with lesions found incidentally before symptom development. A report even exists of fatal subarachnoid hemorrhage from a capillary hemangioma located in the cistern [10].

Two female patients were diagnosed with intracranial capillary hemangioma during pregnancy, raising questions regarding the hormonal influence on the pathogenesis of the lesion [11, 14]. Simon et al. [11] have attributed the coincidence of pregnancy and the symptomatic vascular lesion to increased vascular volume during pregnancy rather than to hormonal effects, because another study has shown that capillary hemangioma lacks estrogen and progesterone receptors [19]. Smith and Skelton [14] have also reported that immunostaining for progesterone receptor was almost negative in intracranial capillary hemangioma. However, the age difference between sexes and the increased incidence in females after adolescence indicate that some hormonal influences may contribute to the clinical manifestations of the disease.

To date, complete surgical resection has yielded excellent results. Two patients experienced progression of the lesion after incomplete surgical resection. The positive

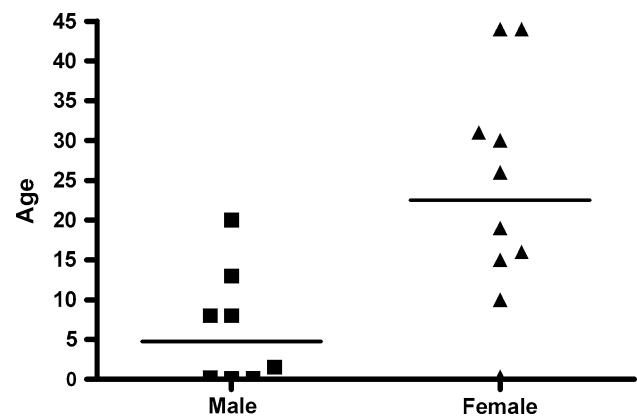
**Table 1** Summary of 18 patients with intracranial capillary hemangioma

Authors (year)	Case no.	Sex	Age	Location	Pattern	Pregnancy	IICP symptom and sign	Cranial nerve palsy	Bleeding	Cyst	Edema	Signal voids	Dural involve	Treatment	Outcome
Willing et al. [6]	1	M	17 m	Middle fossa	Extra-axial or cortical	-	-	-	-	-	-	-	+	GTR	ND
Tsao et al. [9]	2	F	15 y	Middle fossa-CS	Extra-axial	-	-	+	-	-	-	-	+	RT (4,500 cGy)	No recurrence (18 m)
	3	F	19 y	Middle fossa-CS	Extra-axial	-	-	+	-	-	-	-	+	RT (5,000 cGy)	No recurrence (21 m)
Abe et al. [8]	4	M	20 y	Multiple (F-P lobe)	Subcortical	-	+	-	-	+	-	-	ND	GTR of largest lesion	No recurrence and regression (15 y)
	5	F	16 y	Multiple (cerebrum, cerebellum)	Subcortical	-	+	-	+	ND	ND	ND	ND	GTR of parietal lesion	No recurrence and regression (14 y)
	6	M	8 y	Middle fossa	Extra-axial	-	+	-	-	-	+	-	+	GTR	No recurrence (4 yrs)
Le Bihannic et al. [10]	7	M	6 w	Circle of Willis	Extra-axial	-	+	-	+	-	-	-	-	Autopsy	Died
Simon et al. [11]	8	F	31 y	Tentorium-TS	Extra-axial	+	+	-	-	-	+	+	+	STR	Recurrence (6 m)
Brotchi et al. [12]	9	F	10 y	Torcular-SSS	Extra-axial	-	+	-	-	-	-	-	+	GTR	No recurrence (4 y)
Karikari et al. [13]	10	M	3 m	4th ventricle-CPA	Extra-axial	-	-	-	-	-	-	-	-	GTR	No recurrence (5 m)
Smith and Skelton [14]	11	F	26 y	Tentorium-SPS	Extra-axial or cortical	+	+	+	-	-	+	-	ND	GTR	No recurrence (4 m)
Daenekindt et al. [15]	12	M	7 w	Middle fossa	Extra-axial or cortical	-	+	-	-	-	-	+	-	GTR	No recurrence (6 m)
Uyama et al. [7]	13	F	4 m	Cerebellum	Extra-axial or cortical	-	+	-	-	+	-	-	-	GTR	No recurrence (ND)
Mauer et al. [16]	14	F	44 y	Multiple (tentorium, cerebrum)	Extra-axial	-	+	+	-	-	-	-	+	GTR	ND

Table 1 continued

Authors (year)	Case no.	Sex	Age	Location	Pattern	Pregnancy	IICP symptom and sign	Cranial nerve palsy	Bleeding	Cyst	Edema	Signal voids	Dural involve	Treatment	Outcome
Present study	15	M	8 y	Tentorium-TS	Extra-axial	-	+	-	+	-	+	+	+	GTR	No recurrence (24 m)
	16	M	13 y	Tentorium-TS, SPS	Extra-axial	-	+	-	-	+	+	+	+	STR	Recurrence (3 m)
	17	F	30 y	Tentorium	Extra-axial	-	+	-	+	-	+	+	+	GTR	No recurrence (30 m)
	18	F	44 y	CS-sellar region	Extra-axial	-	-	+	-	-	-	-	+	RT (5,400 cGy)	No recurrence (27 m)

M male, F female, w weeks, m months, y years, CS cavernous sinus, F-P frontoparietal, TS transverse sinus, SSS superior sagittal sinus, CPA cerebellopontine angle, SPS superior petrosal sinus, IICP increased intracranial pressure, ND not described, GTR gross total resection, STR subtotal resection, RT radiation therapy



**Fig. 5** Distribution of age at diagnosis in 18 patients according to sex. Horizontal bars represent median values ( $P = 0.023$ , Wilcoxon signed-rank test)

$^{11}\text{C}$ -methionine PET finding (case I) and high Ki-67 index (case IV) in our patients also raise concerns about the tumor-like aggressiveness of these lesions. Therefore, complete resection is the key for treatment of symptomatic lesions. Radiation therapy, despite limited use thus far, also provided favorable lesion control and should be considered for unresectable or incompletely resected lesions.

## Conclusions

The majority of intracranial capillary hemangiomas develop near major venous sinuses and are found in male infants/children and female adolescents/young adults. The early age of onset, lesion locations near the major venous sinuses, and atypical MR findings such as multiple signal voids or intralesional hemorrhage can be helpful in differentiating capillary hemangiomas from meningeal tumors. Capillary hemangioma should be considered in the preoperative diagnosis of extra-axial, contrast-enhancing mass lesions, especially in children and adolescents.

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**Conflict of interest** The authors have nothing to disclose and report no conflicts of interest.

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