Abstract. The present study reported a nearly asymptomatic case of intracranial capillary hemangioma (ICHs), which are rare benign vascular tumors or tumor-like lesions. A 33-year-old female came to the hospital with a complaint of a slight but recurring morning headache concentrated in the left posterior occipital area. These headaches spontaneously resolved without any treatment. Computed tomography and magnetic resonance imaging revealed a mass inside the left occipital lobe. The patient refused to undergo conservative observation at home and insisted on radical therapy. Prior to surgery, an atypical meningioma or astrocytoma was suspected. A navigation-guided brain-mass resection was performed under general anesthesia and a solid mass closely associated with the tentorium cerebelli was completely resected. Histopathological analysis confirmed diagnosis of an ICH. The patient recovered well and experienced no major neurological defects, apart from an issue with the right visual field. The present study also conducted a retrospective literature review of papers published in English describing cases of intracranial capillary hemangiomas. A PubMed search identified 19 articles comprising 29 cases. The clinical symptoms of ICH are diverse and all reported cases in the literature were symptomatic. Previous studies demonstrated that diagnoses of intracranial capillary hemangioma are usually made during surgical resection by histopathological examination. Treatment for ICH remains empirical and surgery is the most common method of treatment. Patient prognosis is generally good-the majority of patients achieve long-term, event- and progression-free survival.

Introduction

Capillary hemangiomas (CHs) are benign vascular tumors or tumor-like lesions that usually occur at birth or in early infancy. Typically, the lesions involve skin and soft tissue, and are usually found on the face, scalp, chest, or back (1,2). They have been reported to occur in 1.1 to 2.6% of full-term neonates, with an estimated frequency of 10 to 12% within the first year of life (2). They are characterized by a proliferative phase followed by an involutionary phase and patients often experience spontaneous regression with age (2,3). Lesions can occur in adults, occur more often in women than men and undergo changes in size relative to hormonal cycles and pregnancy (2).

CHs involving the nervous system are rare (4) and primarily occur in the spinal nerve roots or cauda equina (5,6). Intracranial capillary hemangiomas (ICHs) are even rarer; only 29 cases have been reported so far, however the intracranial location of these lesions varies. All reported cases were confirmed histologically however no patients were diagnosed prior to pathological examination. Clinical experience of this disease is poor and the lesion is usually considered to be a meningioma prior to surgery (7). Due to the small number of reported cases, the true prevalence of ICHs may be underestimated as they may remain asymptomatic or undergo spontaneous regression (3).

The current study reported a case of ICH in a patient admitted to the Department of Neurosurgery, PLA Army General Hospital (Beijing, China). Furthermore, previous studies investigating the diagnosis, differential diagnosis with other brain tumors such as meningiomas, and treatment for ICHs were reviewed.

Case report

A 33-year-old woman attended the Department of Neurosurgery, PLA Army General Hospital in December 2014 with the complaint of a minor morning headache for 7 days that was concentrated in the left posterior occipital area. The headache was alleviated following rest. The patient had suffered from mild hypertension for nearly 5 years since her last pregnancy but her blood pressure was well controlled with oral medication of amlodipine benzenesulfonate (Pfizer, Inc.,
A solid mass closely associated with the tentorium cerebelli was completely resected. Intraoperative bleeding was kept under control and no substantial bleeding was encountered; however, an infratentorial epidural hematoma formed following surgery. This was identified 24 h after surgery in the routine CT exam, requiring a second operation of hematoma evacuation (Fig. 4). The patient recovered well with no major neurological defects apart from a half visual field defect (right temporal side). An MRI scan performed ~2 months later revealed no recurrence of the tumor (Fig. 5); however, the right visual field defect persisted and the patient is being observed at the outpatient clinic every 3 months.

Histopathological analysis. Histopathological analysis confirmed diagnosis of ICH. On visual inspection, the completely resected tumor was similar to a dark-red mulberry lesion that was primarily supplied by the meningohypophyseal trunk; however, no evident draining vein was identified (Fig. 3). A navigation-guided brain-mass resection was performed under general anesthesia. Sufentanil (IDT Biologika GmbH, Dessau-Rossla, Germany; 25 µg), propofol (AstraZeneca, Cambridge, UK; 100 mg) and rocuronium bromide (N.V. Organon; Merck KGaA, Darmstadt, Germany; 50 mg) were used to induce anesthesia. Propofol (AstraZeneca, 200 mg/h) and remifentanil (Yichang Humanwell Pharmaceutical Co., Ltd., Yichang, China; 1,000 µg/h) were used to maintain anesthesia. High intracranial pressure was detected during surgery. At first, entry was attempted through the tentorium cerebelli space to reduce brain damage but this failed. Eventually the occipital cortex was perforated to get to the tumor directly.

A solid mass closely associated with the tentorium cerebelli was completely resected. Intraoperative bleeding was kept under control and no substantial bleeding was encountered; however, an infratentorial epidural hematoma formed following surgery. This was identified 24 h after surgery in the routine CT exam, requiring a second operation of hematoma evacuation (Fig. 4). The patient recovered well with no major neurological defects apart from a half visual field defect (right temporal side). An MRI scan performed ~2 months later revealed no recurrence of the tumor (Fig. 5); however, the right visual field defect persisted and the patient is being observed in the outpatient clinic every 3 months.
neuronal nuclei and Oligo-2 (all Abcam) exhibited negative results, while immunostaining with cluster of differentiation 31 (CD 31), CD 34 and vimentin (all Abcam exhibited strong positive results, which implied vascular proliferation (Fig. 6C and D). The vascular nature of the lesion was confirmed and a diagnosis of meningioma was ruled out.

**Discussion**

CHs are relatively common benign vascular tumors that occur in 1.0-2.6% of live births and are generally located in the skin and soft tissues (8). These tumors are more common in females than males and may undergo changes in size according to hormonal fluctuations and pregnancy (2). CHs involving the central nervous system are rare. The majority of documented cases have been identified in the spinal roots and the cauda equina. ICHs are even more rare. In the current study, a search was conducted using the query ‘intracranial and capillary hemangioma’ on Pubmed (https://www.ncbi.nlm.nih.gov/pubmed/) and 19 articles were identified reporting 29 cases of ICH that were histologically confirmed (Table I).

The 29 cases included 14 male and 15 female patients with ICH ranging in age from 2 weeks to 69 years. The available data were limited, however there were no significant differences in cases based on sex. Data regarding age demonstrated that the majority of patients were infants (7) and young adults <30 years old. Furthermore, only one case of ICH was reported prior to 2000 (9). One possible reason for this is that similar cases may have been described using other terms of pathological diagnosis (4). Due to greater in-depth knowledge of ICH gleaned from pathology reports, an increasing number of cases have been identified more recently. However, accurately diagnosing all cases of ICH is nearly impossible due to the use of non-standardized nomenclature to describe intracranial vascular lesions. One case with mixed capillary and cavernous hemangioma histology was included in the current study (10), however other similar cases may not have been correctly diagnosed and therefore were not correctly reported. Four cases of intraosseous CH were not included (11-14).

Despite the fact that ICH exhibits consistent pathological features, the clinical characteristics of the disease exhibit a high degree of heterogeneity. ICH can grow in the cerebral lobe (4,9), cavernous sinus (15), lumen of the superior sagittal
sinus (16), cerebellum (4), anterior choroidal artery (17), fourth ventricle (8) and sellar region (18). Following analysis of the 29 cases, no predilection for a specific site of ICH growth was apparent and in a number of cases, only a single lesion was detected but there were multiple tumors (4 of 29 reported cases) (4, 19, 20). The clinical symptoms of ICH are very diverse and differ according to tumor location and size. All reported cases in the literature were symptomatic and more common symptoms include headaches, cranial nerve palsy and/or seizures. In 4 of the previously reported cases, ICH was associated with pregnancy (2, 21, 22). By contrast, the patient in the current study was nearly asymptomatic and visited the hospital due to a slight but recurring morning headache that increased in intensity. However, these headaches spontaneously resolved without any treatment; therefore, the patient experienced no marked symptoms associated with ICH, aside from anxiety, prior to initial surgery. An intracranial lesion was detected by neuroimaging examination, however it could not be confirmed that the headaches were associated with the lesion.

The patient had last been pregnant 5 years ago, which resulted in mild hypertension and the patient's blood pressure was controlled by oral medication. The tumor may have been associated with this pregnancy and may have therefore been present for several years. The tumor that was resected during surgery was solid and small and may have been in an involution phase by the time it was detected. All these factors further support the hypothesis that the incidence of ICH may be underreported, particularly if the tumor regresses in size over time and remains asymptomatic.

Multiple differential diagnoses were noted in previous studies but there was limited information available to guide preoperative diagnosis. Due to differences in tumor location and size, differential diagnoses made in one article could not be directly applied to other cases. Capillary hemangioma was diagnosed by pathological observations in all cases. However, ICH was not considered in preoperative differential diagnoses. In the present case, a tentorium-based lesion mimicking a meningioma was identified, which was preoperatively diagnosed as an atypical meningioma. Astrocytoma was also considered, with no expectation of the final result. Therefore, preoperatively diagnosing ICHs is difficult.

Treatment for ICH is empirical (22). Different modalities, including surgery, stereotactic radiotherapy, or treatment with corticosteroids or interferon have been used; however, the rationale and indications for different treatments have not been clearly stated in the literature. The vast majority of patients (28 of 29) underwent surgery, among which there were 24 resections and 4 biopsies. A capacity for rapid growth of the lesion was reported and the tumor recurred after a short time in three cases (2, 7, 18). In symptomatic cases, performing surgery is a less controversial option, however performing surgery in asymptomatic cases should be considered more carefully. Theoretically, conservative observations should be conducted on a regular basis. Taking the final histopathological results into account, it may have been more appropriate for the current patient to undergo conservative observation; however, a tumor that is suspected to be serious requires a more radical approach, such as surgery. In the current case report, the patient refused conservative observation and requested radical surgery as soon as possible. The outcomes of the 29 patients included in the literature review were generally satisfactory, with only one case of mortality and 3 definite recurrences and no more severe neurological defects. More details are shown in Table I.

In conclusion, the present study reported a nearly asymptomatic case of ICH. ICHs are rare entities defined by highly
Table I. Previous cases of intracranial capillary hemangiomas.

| Case no. | Author, year | Patient age, sex | Intracranial tumor location | Symptoms                                      | Surgery                     | Other treatment for ICH | ICH recurrence | Other tumors | Result | Relationship to pregnancy |
|----------|--------------|------------------|-----------------------------|-----------------------------------------------|----------------------------|--------------------------|-----------------|--------------|--------|--------------------------|
| 1        | Willing et al, 1993 | 17 months, male | Right temporal lobe, dural based | Seizures                                     | Total resection            | No                       | No              | NR           | NR     |                          |
| 2        | Tsao et al, 2003  | 15 years, female | Right cavernous sinus       | Right-sided VI, III cranial nerve palsy       | Biopsy sampling and partial removal | Stereotactic fractionated radiation therapy | No              | NR           | Nearly complete resolution of tumor, persistent cranial nerve palsy |
| 3        | Tsao et al, 2003  | 19 years, female | Left cavernous sinus        | Left-sided VI cranial nerve palsy            | Biopsy sampling            | Stereotactic fractionated radiation therapy | No              | NR           | Nearly complete resolution of tumor, persistent cranial nerve palsy |
| 4        | Abe et al, 2004   | 20 years, male   | Multiple, frontoparietal lobes | Headache, seizures                           | Partial resection          | Systemic administration of corticosteroid drugs | No              | NR           | RW     | (4)                          |
| 5        | Abe et al, 2004   | 16 years, female | Multiple, cerebrum and cerebellum | Diplopia                                      | Partial resection          | Systemic administration of corticosteroid drugs followed by IFNα | No              | NR           | RW     | (4)                          |
| 6        | Abe et al, 2004   | 8 years, male    | Left temporal lobe          | Headache, nausea                             | Total resection            | No                       | No              | NR           | RW     | (4)                          |
| 7        | Simon et al, 2005 | 31 years, female | Left occipital lobe and left cerebellar, hemispheric tentorium based: Anterior chonoidal artery | Severe headaches, nausea and vomiting | Total resection (3 times, partial resection 2 times, gross total resection last time) | No | Yes, NR twice | RW | During pregnancy |
| 8        | Le Bihannc et al, 2005 | 6 weeks, male | Anterior chonoidal artery | Vomiting and disturbance of consciousness, left hemiparesis | No                         | No | Hemangiomas found on the eyelid and thigh | Succumbed | (17) |
| 9        | Broetchi et al, 2005 | 10 years, female | Lumen of the superior sagittal sinus | Intracranial hypertension | Partial resection | No | NR | RW | (16) |
| 10       | Karikari et al, 2006 | 3 months, male | Fourth ventricle            | Central hypotonia                            | Total resection            | No | NR | RW | (8) |
| 11       | Smith et al, 2007  | 26 years, female | Left petrous temporal region | Severe frontal headaches associated with one episode of vomiting | Total resection            | No | NR | RW | During pregnancy |
| Case no. | Author, year | Patient age, sex | Intracranial tumor location | Symptoms | Surgery | Other treatment for ICH | ICH recurrence | Other tumors | Result | Relationship to pregnancy (Refs.) |
|---------|--------------|------------------|-----------------------------|----------|---------|------------------------|----------------|-------------|--------|---------------------------------|
| 12      | Uyama et al, 2008 | 4 months, female | Left cerebellar hemisphere | Hydrocephalus | Total resection | Corticosteroids | No | Hemangiomas distributed widely throughout body. | NR | (3) |
| 13      | Daenekindt et al, 2008 | 7 weeks, male | Right temporal fossa | Head enlargement | Total resection | No | No | Not found | RW | (1) |
| 14      | Maurer et al, 2010 | 44 years, female | Multiple, left temporal lobe | POEMS syndrome | Resection 3 times | No | No | Not found | NR | (19) |
| 15      | Lee et al, 2010 | 59 years, female | Pituitary stalk and infundibular recess | Severe headache | Endoscopic biopsy | NR | NR | NR | NR | (23) |
| 16      | Younas et al, 2011 | 69 years, male | Multiple, subcortical regions in bilateral hemispheres | Transient cerebrovascular insufficiency | Biopsy sampling | No | NR | Not found | NR | (20) |
| 17      | Phi et al, 2012 | 8 years, male | Right occipital lobe involving the right tentorium and transverse sinus | Intracranial hypertension and decreased visual acuity | Nearly total resection | No | No | NR | RW | (7) |
| 18      | Phi et al, 2012 | 13 years, male | Right temporo-occipital area | Worsening headache | Partial resection | Required, but no details | Yes | NR | Under close observation for potential further treatment | (7) |
| 19      | Phi et al, 2012 | 30 years, female | Posterior fossa, attached to the tentorium | Worsening headache, vomiting, and vertigo | Total resection | No | No | NR | RW | (7) |
| 20      | Phi et al, 2012 | 44 years, female | Ethmoid and sphenoid sinuses | Progressive visual loss and field cut in the right eye | Partial resection | Radiation therapy (5,400 cGy to the mass) | No | NR | RW | (7) |
| 21      | Morace et al, 2012 | 26 years, female | Sellar lesion extending into the right cavernous sinus and anterior temporal region | Galactorrhea and irregular menstrual cycles as well as high serum prolactin levels | Partial resection | Radiotherapy | No | NR | RW | (18) |
| Case no. | Author, year | Patient age, sex | Intracranial tumor location | Symptoms | Surgery | Other treatment for ICH | ICH recurrence | Other tumors | Result | Relationship to pregnancy (Refs.) |
|---------|--------------|-----------------|-----------------------------|----------|---------|------------------------|---------------|-------------|--------|--------------------------------|
| 22      | Morace et al, 2012 | 61 years, female | Sellar lesion extending into the left cavernous sinus | Left eye visual impairment and a left second trigeminal branch sensory deficit | Partial resection | Stereotactic radiotherapy with Cyberknife | No | NR | RW | (18) |
| 23      | Morace et al, 2012 | 14 years, male | Left middle cranial fossa, extending into the cerebello-pontine angle and infratemporal fossa | Left VII cranial nerve palsy, left deafness, dysphagia, and vertigo | Partial resection | Radiotherapy after recurrence | Yes | NR | RW | (18) |
| 24      | Morace et al, 2012 | 42 years, male | Left temporoparietal area | Dysphasia | Total resection | No | No | NR | RW | (18) |
| 25      | John et al, 2012 | 59 years, male | Right temporoparietal region | Transient focal neurological deficits and behavioral abnormalities mimicking Ganser’s syndrome | Total resection | No | No | NR | RW | (24) |
| 26      | Zheng et al, 2012 | 3 years, male | Right middle cranial fossa | Progressively enlarged subcutaneous, mass somnolence, vomiting and ptosis of the right eyelid | Total resection | No | No | NR | RW | (25) |
| 27      | Mirza et al, 2013 | 28 years, female | Right temporal region adjacent to the transverse sinus | Seizures | Total resection | No | No | NR | RW | Symptoms started during pregnancy (22) |
| 28      | Mirza et al, 2013 | 41 years, female | Occipital region | Progressive visual disturbance | Total resection | No | No | NR | RW | Symptoms started during pregnancy (22) |
| 29      | Jalloh et al, 2014 | 2 weeks, male | Left middle fossa | Intracranial hypertension | Biopsy followed by total resection | No | No | NR | RW | (10) |

NR, not reported; IFN, interferon; RW, recovered well.
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