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Diagnosis and Treatment of Intracranial Cavernous Hemangioma: a Report of 86 Cases

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OBJECTIVE To summarize and analyze the clinical manifestations features of imaging diagnosis, and therapeutic efficacy of surgical treatment for intracranial cavernous hemangioma (CH).
METHODS Data from 86 cases with intracranial CH from the Department of Neurosurgery of Tianjin Huanhu Hospital, Tianjin, China, during a period from 2000 to 2007, were retrospectively analyzed, and pertinent literature cited.
RESULTS Epilepsy, headache, dizziness, sensory disability and limb-kinetic apraxia were the most commonly seen clinical manifestations of the intracranial CH cases. MRI was one of the preferred ways to diagnose CH. All 86 patients were treated with microsurgery, among which neuronavigator-guided surgery was conducted in 16, a second surgical procedure was performed in 3 due to a postoperative intracranial hematoma, and death occurred in 1. All the other 85 patients had a good prognosis.
CONCLUSION MRI was the most sensitive diagnostic means for intracranial CH. Microsurgery was the main method to treat intracranial CH.

KEY WORDS: cavernous hemangioma, CT, MRI, microsurgery.

Introduction

Cavernous hemangioma (CH) is an occult vascular malformation that was given this name because of its sponge-like shape. It is an abnormal vascular clump composed of masses of thin-walled vessels, which is rarely seen clinically. In our study, data from the 86 CH patients who underwent microsurgery during an 8-year period, i.e. from 2000 to 2007, were retrospectively analyzed.

Materials and Methods

General data

In our study there were 46 male and 40 female patients, with an age range from 3 to 68 years. For the diseased region and clinical manifestations, see Tables 1 and 2.

Image analysis

CT scanning

CT was conducted in 82 cases, with a reinforced scanning in 10 of them. During the course of CT diagnosis, CH was found in 13 of the 82 cases. No other conclusive diagnoses were made in the remaining 72 cases, with no abnormality seen in 10.

Table 1. Diseased Regions.

Sites	Cases	%
Frontal lobe	19	22.1
Parietal lobe	11	12.8
Temporal lobe	20	23.3
Occipital lobes	2	2.3
Mesocerebrum	1	1.2
Bridge of varolius	6	7.0
Medulla oblongata	2	2.3
Cerebral ganglion	1	1.2
Basal ganglia	1	1.2
Cerebellum	13	15.1
Carvernous sinus	6	6.98
Multiple	4	4.7

Table 2. Clinical Signs.

Signs and Symptoms	Cases	%
Epilepsy	32	37.2
Headache	30	34.9
Dizziness	14	16.3
Nausea	14	16.3
Vomiting	14	16.3
Sensory disability	14	16.3
Limb-kinetic apraxia	10	11.6
Ambiopia	4	4.7
Conscious disturbance	3	3.5
Visual extinction	3	3.5
Smell hallucination	2	2.3
Language disorder	1	1.2
Hearing defect	1	1.2
Hypophasis	1	1.2

CT angiography (CTA)

CTA was performed in 9 cases, among which CH was found in 2, and no conclusive diagnosis was made in 3. In the other 4 of the 9 cases, 2 were misdiagnosed as a pituitary tumor and 2 as meningeoma.

MRI examination

MRI was conducted in 85 cases, with a reinforced examination in 19. CH was found in 71 and no conclusive diagnosis was made in 9 of the 85 cases. In the other 5 cases, 2 were misdiagnosed as a pituitary tumor and 3 as meningeoma.

Magnetic resonance angiography (MRA)

MRA examination was conducted in 6 cases, without noting abnormal findings.

Digital subtraction angiography (DSA)

DSA was conducted in 18 cases, among which 1 was diagnosed as CH, 3 were considered to have an aneurysm and 2 were reported as a chromosomal abnormality. No abnormality was seen in the rest of the 18 cases.

Pathological examination

All 86 cases were pathologically confirmed as CH, among them a concomitant hemorrhage was found in 15 cases, a concomitant hemorrhage and fibrosis in 14, an accompanying bleeding and calcification in 6, and a concomitant fibrous degeneration and calcification in 5.

Surgery

Micro-surgical treatment was conducted in all cases, of which neuronavigator-guided surgery was utilized in 16. Total resection was conducted in 80 cases with intracerebral CH, and 6 with extracerebral CH in the cavernous sinus. Total resection by stepwise operation was performed in 1 of the 6 cases.

Results

In the 86 patients undergoing microsurgery, a second surgical procedure was conducted in 3 due to a postoperative intracranial hematoma, with death occurring in 1 and a satisfactory prognosis in the others. After surgical operation, the symptoms of epilepsy were not significantly lessened in 2, and limb-kinetic apraxia was not apparently palliated in 1. There was an obvious improvement in the symptoms in 37 of the 86 patients, and a disappearance of the symptoms in 46, with their physical condition returning to normal.

Discussion

At present, the pathogenesis of intracranial CH remains unknown, however radiotherapy and other factors, such as vascular malformation, trauma, and heredity etc., may be involved. Most commonly the CH patients'age at onset was 20 to 50 years, with the CH patients accounting for approximate 5% to 13% of those with a vascular malformation of the brain[1]. In the cases of our group, the youngest patient was only 3 years old. Perhaps hereditary factors and radiotherapy might be responsible for this disease. In general, the incidence of supratentorial lesions is higher than that of subtentorial lesions, e.g., supratentorial CH accounted for 57% of the total CH cases, and the subtentorial made up only 43%, based on a report from overseas[2]. In our group, there were 73 supratentorial CH cases (84%), and 13 subtentorial cases (16%). There was a very close correlation between the clinical manifestations and the CH diseased region. Epilepsy and headache were the most frequently-seen CH clinical manifestations. Epilepsy was usually caused by frontal lobe and temporal lesions, and in our group 20 patients with frontal lobe CH showed epileptic symptoms.

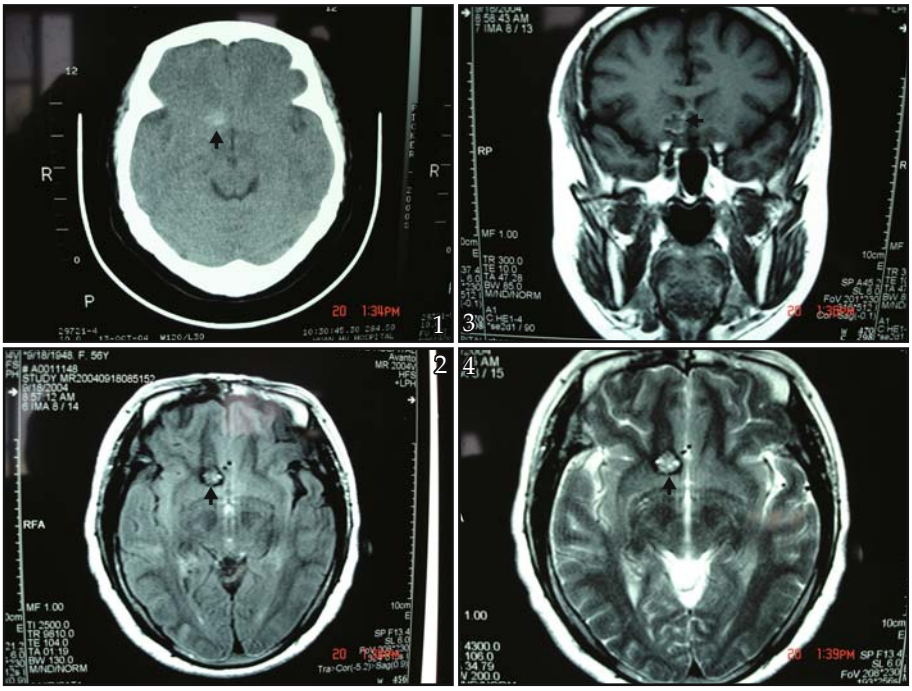

Fig.1. CT plain scan, with a round-like lesion of mixed density.
Fig.2. Mixed signals of the T1WI lesion on MRI, with a ring-shaped peripheral low signal.
Fig.3. Mixed signals of the T1WI lesion on MRI, with a semiring-shaped peripheral low signal.
Fig.4. Mixed signals of the T2WI lesion on MRI, with a round-like peripheral low signal.

A possible relationship between the conditions, such as headache, dizziness, sensory disability, hemiparalysis and conscious disturbance etc., and a spontaneous tiny hemorrhage of the intracerebral focus should be taken into account. Most of the lesions in the middle cranial fossa were extracerebral lesions, and the common reason for the clinical symptoms was a space-occupying oppression, that is, the lesion encroached the anterior cranial fossa if a tumor progressed forwards, the posterior cranial fossa if backwards, and it might invade the cavernous sinus, hypothalamus, pituitary body and optic nerve etc. If pressure was inward, related symptoms, such as smell hallucination, oculomotor paralysis, endocrine disturbance and visual extinction etc. could occur. A spontaneous tiny hemorrhage of the intracerebral focus was easily found in the CH brainstem. In a report from China[3], the hemorrhage rate of CH at the cerebral hemisphere was compared with that at the brainstem and the cerebellum, respectively, indicating that the CH bleeding at the brainstem is frequently seen, compared to CH at other sites, and the occurrence of its clinical symptoms is usually abrupt, with the clinical manifestations of headache, dizziness, nausea, as well as Grade II~VII cranial nerve disturbance.

CT offers a definite assistance in diagnosing CH, however its sensitivity and specificity are not very high. So it can be used for an auxiliary CH diagnosis. The typical CH appearance in CT imaging is a round or rounded-like image of mixed or high density (Fig. 1), presenting an obviously homogeneous reinforced CT figure after reinforcement. In our group, 82 patients received CT scanning, but CH was found in only 13, accounting for 15.9% of the 82 patients. CTA has been a popular diagnostic method for the past few years, which can be used for 3-dimensional reconstruction after angiography showing a better contrast between the focus and peripheral tissue. However, it lacks specificity for CH diagnosis.

In our group, a CTA examination was conducted in 9 cases, 2 of them were diagnosed as CH (22.2%). Moreover the CHs in the cavernous sinus and saddle area are easily misdiagnosed as pituitary tumor or meningeoma. In our cases, 2 receiving CTA were diagnosed as a pituitary tumor and meningeoma, respectively, and no manifest help was seen with MRA examinations. Six patients who received MRA showed no overt abnormalities.

DSA also lacks specificity for diagnosing CH. In our study, DSA was conducted in 18 cases, resulting in only 1 CH, 3 with a dubious aneurysm, and 2 with chromosomal aberrations, and no abnormalities were seen in the remaining cases. After an analysis of the poor CH specificity with a variety of angiography examinations, it was concluded that the contrast medium fails to develop an image after dilution because of a large vascular bed at the CH site, and a slow blood flow.

With the emergence of MRI, diagnostic sensitivity of CH has been considerably increased, and it is still the most sensitive and specific CH diagnostic method. MRI was conducted in 85 cases in our study, with a correct diagnosis of 71 (83.5%). The typical manifestation of intracerebral CH on MRI was that the T1 images presented a signal of equivalent resolution or mixed signals,

and in a few cases it showed a signal of low or high resolution. The T2 image presented a high-resolution signal, with a ring-shaped or semiorbicular low-resolution signal belt around the focus, which was formed owing to a hemosiderin deposition, known as a Hegar's sign. It presented an obviously homogeneous reinforced MRI figure after reinforcement, making it easy to provide a diagnosis based on the typical image appearance (Figs.2~4). However, since there was no low-resolution signal belt around the CH in the cavernous sinus, the CH was easily misdiagnosed as a meningeoma or pituitary tumor owing to the specific sites of the focus. In the 6 cases with a cavernous sinus CH in our group, 2 were diagnosed as a pituitary tumor, 3 a meningeoma, and only 1 as CH, showing a misdiagnostic rate of up to 83.3%. Reports from China[4-9] have presented misdiagnostic rates ranging from 8.3% to 77.27%. Although the cavernous sinus CH misdiagnostic rate was high, MRI imaging was still distinctive and helpful for differentiation between meningeoma and a pituitary tumor.

A summary of CH based on the related literature indicated that[7,8,10-12]: *i*) the lesion is solitary, with a big volume and a cross-section diameter of over 5 cm. The typical appearance of CH showed that the lesion is a cucurbit- or dumbbell-shaped lump, with a wide external part and a narrow internal part. Meningeoma usually presented as a bulbiform or irregular shape, and the coronal part of a pituitary tumor might assume a Girdling sign. The normal pituitary body can not be discriminated from the disease, while the compressed and deformed pituitary body can still be discriminated from CH in the cavernous sinus; *ii*) hemorrhage and calcification are rare, and bleeding is common with a pituitary tumor; *iii*) T1WI assumed an equivalent or low-resolution signal, and T2WI presented a signal of very high resolution. This is the distinctive appearance of CH in the cavernous sinus. For meningeoma and a pituitary tumor, T2Wi assumed the equivalent signal or the signal of a slightly high resolution; *iv*) an obviously homogeneous and reinforced MRI figure after the reinforcement was another distinctive appearance of the cavernous-sinus CH, which was similar to the T2WI signal of the cerebrospinal fluid. In diagnosing a meningeoma or pituitary tumor, the intensifying degree of the figure was lower compared to cavernous-sinus CH. As for the CH with an inward progression to the Turkish saddle, its signal of the increased pituitary body was obviously lower compared to the signal of the tumor on T1WI, and the signal of an increased pituitary body due to a pituitary tumor was in accordance with the signal of the lesion. Therefore a cavernous-sinus CH is highly suspected if there is the distinctive appearance of CH in an MRI examination, although the incidence rate of meningeoma and pituitary tumor in the middle cranial fossa is much higher compared to CH. Because the surgical risk with cavernous-sinus CH is higher than that with meningeoma or a pituitary tumor, preoperative preparations should be made.

Currently the viewpoint on treatment of intra-cerebral CH is basically concordant, i.e. the micro-surgical excision of the lesion is the optimal choice. The surgical mode of CH is various because the site of the lesions vary. Total resection of the intra-cephalic lesion should be conducted as far as possible, and the hemorrhagic focus should also be excised if is occurs. The removal of abnormal brain tissue around the lesion should be conducted if there are clinical manifestations of epilepsy, because the hemosiderin deposition, gliosis or calcification around the lesion may be the main cause of the epilepsy. A suboccipital approach is usually used for a subtentorial lesion. Either a suboccipital or a temporal approach can be used to treat the lesion in the brainstem, based on the specific symptoms and conditions. Total resection should be conducted as far as possible, since brainstem CH is usually found in a superficial site, with a well-circumscribed boundary. However, since the position of the brainstem is rather deep, the authors believe that the surgical procedure should be conducted by neuronavigator-guided localization, which is of more benefit for a total resection of the diseased region and injury diminution. Neuronavigator-guided microsurgery was conducted in 16 cases in our group, with a complete resection of the focus in all cases, producing a satisfactory prognosis. According to the reports from China[13-15], neuronavigator-guided microsurgery for intracranial CH has achieved an acceptable therapeutic effect.

Since the preoperative misdiagnosis of a cavernous-sinus CH is frequent, with a commonly seen ceaseless intra-operative hemorrhage, microsurgical treatment is a great challenge for the neurosurgeons. A gelatin-sponge packing method can be used for an effective hemostasis when there is profuse intra-operative blood loss following a partial resection, since there is the difficulty in CH preoperative diagnosis. After an electric coagulation in the cerebral dura mater, the opened cranium should be closed. Continuous excision can not be conducted as excessive intra-operative blood loss may result in death of the patient. In the 6 cavernous-sinus CH cases of our group, the rhinal sphenoid approach was conducted in 1, where the surgical procedure was stopped due to a failure of hemostasis. The pterion approach was used in 4 of the 6 cases, among which surgery was stopped in 1 because of excessive blood loss, a partial resection in 2 due to an excessive hemorrhage, and a total resection by a stepwise operation in 1. The malar arch pterion approach was conducted in another 1 of the 6 cases, after preoperative ligation of the external carotid artery, with a complete resection.

Based on an overseas report[16], the epidural approach was performed in 13 cavernous-sinus CH patients, with a total resection in 12, and a partial resection in 1. All 13 patients had a good prognosis, without a recurrence. It was reported in China that[6] an epidural approach

was conducted in 13 cavernous-sinus CH cases, with a total resection in 12, and a partial resection in 1. Postoperative amelioration of the symptoms occurred in all 13 cases, without a relapse. The subdural approach was conducted in the other 7 cases, with a partial resection in all, without a postoperative improvement of the symptoms.

It was also reported that[9] the subdural approach for separating the Sylvian cistern arachnoid mater could extensively expose the diseased region, cut open the parietal dura of the cavernous sinus, effectively expose and protect the cranial nerve, and cut off the feeding artery. The authors believe that there are various surgical routes, so the means for an excision of the lesion should be based on the nature of the lesion. The optimal surgical procedure should depend on the growth pattern of the lesion, and the micro-surgeon's own level of skillfulness.

Although CH is sensitive to radiotherapy, its therapeutic efficacy for CH remains unsure, and in addition radiation is sometimes one of the causative factors of CH. Therefore the authors believe that radiotherapy can be performed with a deep or minor lesion in patients who are unfit for surgery, or for those who have presented with related CH symptoms or have residual postoperative tissue from the lesion. Conservative treatment can be conducted if the CH is found by chance with no symptoms or the symptoms are slight, using regular follow-up.

CH is rare in clinical practice. There are miscellaneous clinical manifestations of CH due to different sites, with epilepsy and headache as the most commonly seen symptoms. MRI shows the optimal sensitivity and specificity for CH diagnosis. CT can be used as an auxiliary examination, but CTA, MRA and DSA show poor specificity. Moreover, a CH in the cavernous sinus is easily diagnosed as a pituitary tumor or meningeoma.

Microsurgical resection is the treatment of choice for treating intracerebral CH. Total resection should be conducted as far as possible in treating intra-cephalic lesions, while for treatment of extra-cerebral CH in a cavernous sinus, whether to perform the total resection will depend on the intra-operative condition. Neuronavigator-guided localization provides for the location, total resection and the diminution from injury of the lesion, especially for patients with a deep focus of the lesion, e.g. the CH in the brainstem. Radiotherapy can be used as an adjunctive therapy for those who are unfit for surgery or with a postoperative tumor residual.

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