Giant Intraparenchymal Meningioma in a Female Child: Case Report and Literature Review

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Case report

Keywords: Meningioma, fibrous, temporal lobe, Intraparenchymal

DOI: https://doi.org/10.21203/rs.3.rs-111294/v1

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Abstract

**Background:** Intraparenchymal meningiomas without dural attachment are extremely rare, especially in female children. To our knowledge, fibrous intraparenchymal meningioma located in the temporal lobe has never been reported in female children. The significance in the differential diagnosis of lesions in the temporal lobe should be emphasized.

**Case presentation:** A 12-year-old girl was admitted to our hospital, complaining of recurrent generalized seizures for 2 months. Magnetic resonance imaging demonstrated a solid lesion located in the temporal lobe. The lesion underwent gross total resection. Histopathological examination indicated that the lesion was a fibrous meningioma. Postoperative rehabilitation was uneventful.

**Conclusions:** This case report presents an extremely unusual intraparenchymal fibrous meningioma of the temporal lobe with peritumoral edema and reviewed 20 intraparenchymal meningiomas in children and to discuss the clinical presentation and treatment, differential diagnosis, and radiological features.

Background

Intraparenchymal meningiomas, considered the same type of subcortical meningiomas, are defined as meningiomas located in the brain parenchyma without dural attachment, even reaching the brain surface[1-3]. Intraparenchymal meningiomas are rare, in contrast to ordinary meningiomas, and are more frequent in children and adolescents than in adults[4]. To date, only 20 patients aged 0 to 18 years, including ours, have been reported in the English-language literature, which are summarized in Table 1[1-2, 4-20]. The gender distribution showed a male dominance in children and adolescents. To date, there are only 6 intraparenchymal meningiomas in female children, including ours, reported in the English-language literature, with most of them in the cerebral lobes.

Case Presentation

This 12-year-old girl presented with a history of generalized seizures that recurred six times in 2 months. MRI (Magnetic resonance imaging) revealed a solid mass, sized 5.0×6.5×6.0 cm, located in the temporal lobe. The tumor viewed with MRI showed isointensity on T1-weighted images and high intensity on T2-weighted images and homogenous enhancement on T1-weighted gadolinium enhancement, but no dural tail sign was noted (Fig. 1).

The tumor was located in the left temporal lobe, medium texture, grayish red, intact capsule, abundant blood flow, no definite base, branches of the temporal branch of the middle cerebral artery m2 segment were involved in the blood supply, and the boundary between the lesion and the brain tissue was clear. The lateral fissure vein shifted outward, the middle cerebral artery shifted medially, and the surrounding brain tissue was compressed. The tumor capsule was first cut and then resected with cusa. When the tumor volume was reduced and gradually freed along the envelope and the blood supply artery was broken, the tumor was then excised in blocks and ultimately underwent gross total resection under a microscope. M2 segments of the vein and middle cerebral artery were well protected. The tumor was diagnosed as a fibrous meningioma (Fig. 2). The patient's postoperative course was uneventful, and there was no evidence of recurrence on an MRI performed 3 months after the operation (Fig. 1).

Discussion And Conclusions

Primary intraparenchymal meningiomas are rare but are more frequent in males than in females in children. This difference may be associated with increased estrogen levels in adult women and increased susceptibility to meningiomas[21]. Female cases are extremely rare; to date, there are only 6 cases, including ours, and most of them have occurred in the cerebral lobes. The most common location was the frontal lobe (n=2, 33.3%) and temporal lobe (n=2, 33.3%), followed by the parietal lobe (n=1, 16.7%) and brainstem (n=1, 16.7%). Presenting symptoms depend on tumor location, and seizures (n=4) were most frequent. One case had hemiplegia because the tumor was located in the brainstem[51]. Another case was characterized by vomiting and microcephaly because the tumor was giant. The CT (Computed Tomography) scan of the head showed a 9.5-cm calcified cystic mass in the left frontal region[27].

In general, most meningiomas are grade I, accounting for 80%–90%, whereas 5%–15% are grade II and 1%–3% are grade III[22, 23]. The meningothelial type is the most common, followed by the fibrous type[22]. Among intraparenchymal meningiomas in children, the most common meningiomas are grade I (n=12, 66.7%), and the most common type is fibrous (including fibrous components) (n=8). There were 6 grade II and grade III cases, accounting for 33.7%. At present, the cases of female children are extremely rare. Only 4 cases had definite pathological results, which included fibrous (n=2), anaplastic (n=1), and clear cell (n=1). The present case is the first case of fibrous intraparenchymal meningiomas located in the temporal lobe in female children. Among 12 cases of children, homogeneous enhancement (n=6), fibrous (n=3), and meningothelial (n=1) were noted; none of the remaining cases had clear pathological reports. Heterogeneous enhancement (n=6), atypical (n=2), anaplastic (n=1), fibrous (n=1), clear cell (n=1), and transitional (n=1) were also reported. The present cases were fibrous and showed homogeneous enhancement, including ours; 4/5 fibrous intraparenchymal meningiomas showed homogenous enhancement.

Since intraparenchymal meningioma lacks dural attachment, characteristics such as cyst formation and the presence of peritumoral edema are relatively common in childhood. Moreover, it is often difficult to preoperatively distinguish it from gliomas, cavernous angiomas, metastatic tumors, malignant lymphomas, or sarcomatous lesions[24]. Among 20 cases, cysts were found in 6 cases, and peritumoral edema was found in 9 cases. Especially when cystic components are present, it is more difficult to correctly diagnose these lesions because cysts are often associated with gliomas or metastatic tumors and are rarer among meningiomas. Peritumoral edema of intraparenchymal meningioma surrounding the lesion develops strongly in the region where the lesion is attached to the surrounding brain tissue, whereas peritumoral edema of most gliomas or metastatic tumors tends to surround the entire tumor surface. Wada et al. believe that the feature of any peritumoral edema of intraparenchymal meningiomas can help differentiate between intraparenchymal meningiomas and...
high-grade gliomas or metastatic tumors. However, its characterization is not applicable to all intraparenchymal meningiomas. Peritumoral edema surrounding the entire tumor surface was found in 3 of 9 cases. Hence, the radiological features of most pediatric primary intraparenchymal meningiomas, such as cystic components, peritumoral edema surrounding the entire tumor, and no dural tail sign, make accurate preoperative diagnosis extremely difficult.

Almost all intraparenchymal meningiomas were located in the cerebrum and lacked dural attachment and no association with the dura of the skull base or the sinuses. Only 3 of 20 cases in children underwent subtotal resection. For lesions located in the hemisphere, one patient underwent STR (subtotal resection), but the reason was not stated. In one case, STR was performed because of the rich blood supply, tight adhesion with the ACA (anterior cerebral artery) and lower tolerance of blood loss in children. There was a 70% remnant in one patient because of the lesion in the brainstem. No recurrences occurred in any patients who underwent total tumor resection during the follow-up period (15 weeks-5 years). Postoperative radiotherapy is controversial for children. Some reports have shown the development of meningiomas after high-dose irradiation with long latency periods. Gosztonyi et al. believe that low-dose irradiation also causes the development of meningiomas with long latency periods in children. Some people treated patients with malignant meningiomas, and radiotherapy was used in the postoperative course. But, Ghim, TT et al. believe that high-dose radiotherapy should be avoided in the treatment of meningiomas in children. Some studies showed that the extent of initial surgical resection was the strongest independent prognostic factor for pediatric meningiomas and that upfront radiotherapy achieved no benefit. Hence, surgery is the best modality of treatment, and resection is recommended to achieve maximum extirpation.

Some authors thought that intraparenchymal meningiomas arise from arachnoid cells located within the pia mater and enter the surface of the brain or sulcus with perforating blood vessels. Several cases involved the MCA (middle cerebral artery) branches or had feeding arteries of the MCA in children. A patient's tumor is closely related to the perforating arteries of the ACA and middle cerebral artery and the recurrent artery of Heubner. In one case, several small arteries were found to enter into the lesion. These cases provide better support for the establishment of this theory. Therefore, for patients suspected to be intraparenchymal meningiomas and for whom the tumor is located in the blood supply range of the ICA (internal carotid artery), MRA (MR Angiography) or CTA (CT angiography) should be performed to evaluate the relationship between the tumor and blood vessels before the operation. During surgery, we should pay attention to the protection of blood vessels to avoid the neurological dysfunction caused by cerebral infarction after surgery, such as hemiplegia and aphasia. Our patient's preoperative CTA showed that the lesion appeared intimately connected to proximal MCA branches. During the operation, the temporal branch of the m2 segment of the middle cerebral artery was found to be involved in the blood supply of the tumor.

We present an extremely unusual case of intraparenchymal fibrous meningioma of the temporal lobe with peritumoral edema and review the pertinent literature. Imaging features such as cystic degeneration, peritumoral edema, and absence of meningeal tail signs make accurate preoperative diagnosis difficult. However, most patients can achieve total resection and a good prognosis.

**Abbreviations**

MRI: Magnetic resonance imaging, CT: Computerized Tomography, STR: subtotal resection, GTR: gross total resection, MAC: middle cerebral artery, ACA: anterior cerebral artery, MRA: MR Angiography, CTA: CT angiography, ICA: internal carotid artery

**Declarations**

**Acknowledgements**

The authors would also like to acknowledge the surgeons, nurses, and pathologists who helped to manage patients and provide pathological images.

**Funding**

This work was supported by grants from provincial or ministry level project of china (No. SB201902027).

**Availability of data and materials**

Not applicable.

**Authors’ contributions**

Yushe Wang, and Yong Wang designed the study. Zhen Ma, and Shuo Wen, collected the data. Huachao Guo wrote the manuscript.

All authors saw and approved the final version of the paper.

**Competing interests**

The authors declare that they have no competing interests.

**Consent for publication**

We have obtained consent for publication from parent and legal guardian.

**Ethics approval and consent to participate**
This study was approved by the Ethics Committee of Henan University People's Hospital, and informed consent was obtained from parent and legal guardian.

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| Study                  | Age| Location       | Size| Clinical presentation | MRI (solid part) | cystic | peritumoral edema | Surgery | feeding artery |
|-----------------------|----|----------------|-----|-----------------------|-----------------|--------|------------------|---------|----------------|
| **Female**            |    |                |     |                       |                 |        |                  |         |                |
| 1 Present case        | 12Y| Temporal       | 6.5cm| Seizure              | Homogeneous     | NO     | YES              | GTR     | MAC            |
| 2 Pinto et al. (2012) | 17Y| Temporal       | Ns  | Seizure              | Homogeneous     | NS     | YES              | NS      | NS            |
| 3 Teo et al. (1998)   | 1.8Y| brainstem      | 4.0 cm| Hemiparesis          | NS              | NS     | NS               | STR     | NS            |
| 4 Kohama et al. (1996) | 1.8Y| Frontal        | 2.5 cm| Seizure              | Homogeneous     | NS     | NO               | GTR     | MCA           |
| 5 Mamourian et al. (1991) | 2Y | Frontal        | 9.5 cm| Vomit, microcephaly  | NS              | Yes    | NS               | GTR     | NS            |
| 6 Morimoto et al. (1976) | 17Y| Parietal       | NS  | seizure              | Headache        | NS     | NS               | NS      | NS            |
| **Male**              |    |                |     |                       |                 |        |                  |         |                |
| 7 Liu X et al. (2018) | 8Y | Basal ganglia  | 4.4 cm| Headache, vomiting, left hemiparesis | Heterogeneous | Yes    | NS               | STR     | ACA           |
| 8 Nayil et al. (2015) | 3Y | Frontal        | NS  | Headache, vomiting   | Heterogeneous   | Yes    | Yes              | GTR     | NS            |
| 9 Werbrouck et al. (2014) | 13Y| Temporal       | 3.4 cm| Seizure              | Homogeneous     | NS     | Yes              | GTR     | NS            |
| 10 Jung and Song (2012) | 1.7Y| Frontoparietal | 4.0 cm| Seizure, hemiparesis | Heterogeneous   | NS     | Yes              | GTR     | NS            |
| 11 Shimbo et al. (2011) | 10Y| Frontal        | 2.2 cm| Seizure              | Homogeneous     | NS     | Yes              | GTR     | MCA           |
| 12 Zhang et al. (2007) | 16Y| Parietooccipital | 2.5 cm| Seizure              | Heterogeneous   | Yes    | Yes              | GTR     | Small Arteries |
| 13 Karadereler et al. (2004) | 14Y| Temporal       | 1.5 cm| Seizure, headache    | Heterogeneous   | NO     | Yes              | GTR     | NS            |
| 14 Starshak (1996)     | 6.8Y| Frontal        | NS  | Headache              | Heterogeneous   | Yes    | NS              | GTR     | NS            |
| Case | Authors | Age | Location | Maximum Diameter | Seizure | Location | Enhancement | Histology | Immunohistochemistry | Treatment | Vessels |
|------|---------|-----|----------|------------------|---------|----------|-------------|-----------|---------------------|-----------|---------|
| 15   | Perilongo et al. (1992) [16] | 2Y | Temporal | NS | NS | Homogeneous | NS | NS | GTR | NS |
| 16   | Schroeder et al. (1987) [1] | 7Y | Frontal | 3.0cm | Seizure | NS | NO | NO | GTR | NS |
| 17   | Sakaki et al. (1987)[17] | 0.9Y | Frontal | 3.0cm | Seizure | Homogeneous | NO | Yes | GTR | MCA |
| 18   | Kimura et al. (1987)[18] | 0.9Y | Frontal | 3.0cm | Seizure | NS | NS | NS | GTR | NS |
| 19   | Drake et al. (1986)[19] | 12Y | Temporal | 1.0cm | Seizure | NS | NS | NS | STR | NS |
| 20   | Legius et al. (1985)[20] | 1.2Y | Parietal | 2.0cm | Seizure | NS | NO | Yes | GTR | NS |

Table 1 Summary of cases involving primary intraparenchymal meningiomas in the literature

- Y: yes, MD: maximum diameter, CE: contrast enhancement, STR: subtotal resection, NS: not stated, MAC: middle cerebral artery, ACA: anterior cerebral artery, SA: Small Arteries, GTR: gross total resection, Rd: radiotherapy, ch: chemotherapy, W: week

**Figures**

**Figure 1**

a-f: Preoperative magnetic resonance imaging demonstrating a solid mass of temporal lobe. The tumor viewed with MRI showed isointensity on T1-weighted images and high intensity on T2-weighted images and homogeneous enhancement on T1-weighted gadolinium enhancement, but no dural tail sign was noted. CTA showed that the middle cerebral artery adhered closely to the tumor and shifted medially, g-f: Three-month after the operation, magnetic resonance imaging shows no evidence of disease recurrence.

**Figure 2**

Photomicrographs showing the histological and immunohistochemical features of the lesion. Histological: The tumor was diagnosed as a fibrous meningioma, WHO I. Immunohistochemical features: CD34(foci+), EMA(+), PR(-), SSTR2(+), STAT6(-), Vimentin(+), GFAP(-), Ki67(about1%+).