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Cavernous Angioma of the Middle Cranial Fossa. Report of Two Cases and a Review

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Intracranial extraparenchymal cavernous angiomas (cavernomas) are rare. A few such lesions involving the cerebellar tentorium[7,16] or the dura mater of the cerebral convexity[11] have been reported. In most of the reported cases of intracranial extracerebral cavernomas, however, the lesion has been found in the middle cranial fossa, particularly involving the wall of the cavernous sinus. We recently experienced two such cases.

Report of Cases

Patient 1: A 53-year-old woman was admitted with 3 months' history of intermittent severe pain in her left cheek. She had experienced two episodes of ptosis of her left lid 13 years and again 11 years before admission. On both occasions ptosis resolved spontaneously in a month.

Physical examinations on admission were normal except for mild hypesthesia in her left cheek and decreased left corneal reflex. Results of the laboratory examinations including hormonal pofile were within normal limits.

Plain radiograph of the skull showed erosion of the left anterior clinoid process and depression of the left sellar edge. Technetium brain scan revealed an intense accumulation of radioactivities in the left parasellar region (Fig. 1). Computed tomographic (CT) scan demonstrated a left parasellar isodense mass, which showed marked homogeneous enhancement following an administration of the contrast medium. The mass was cylindrical in shape, had its base on the left lateral surface of the body of the sphenoid bone, and pointed superolaterally (Fig. 2). Carotid angiography demonstrated superior displacement of the cavernous segment of the left internal carotid artery and elevation of the trunk of the left middle cerebral artery. A faint tumor stain was seen but no feeding artery was found. Cavernous sinography via the inferior petrosal sinuses showed obliteration of the left cavernous sinus.

The parasellar mass was reached intradurally via a frontotemporal craniotomy. The tumor arose from the left cavernous sinus and it was covered by the dura. The capsule (the dura) was incised, and the vascular tumor was subtotally removed. Bleeding from the tumor was rather severe but not pulsatile. Mild paresis of the left abducens nerve was noted postoperatively but

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Fig. 1. *Patient 1.* Technetium brain scan showing an intense radionuclide uptake in the left parasellar region (arrow).

it cleared in a few months. Pathological diagnosis was a typical cavernous angioma.

Patient 2: Two years prior to admission, this 43-year-old woman noticed paresthesia in her right cheek. One year later she began to complain of double vision.

Neurologic examination on admission found paresis of the right oculomotor and abducens nerves. Sensory and motor functions of the trigeminal nerve were objectively normal. The

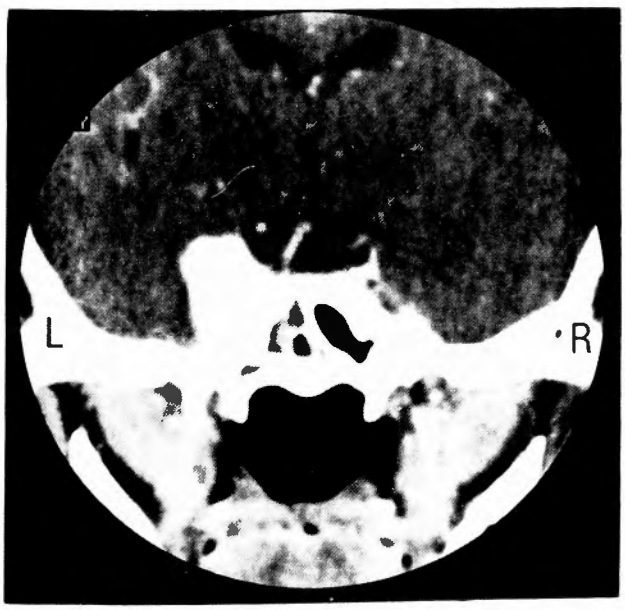

Fig. 2. *Patient 1.* Direct coronal computed tomogram after contrast medium injection, showing markedly enhanced left parasellar mass.

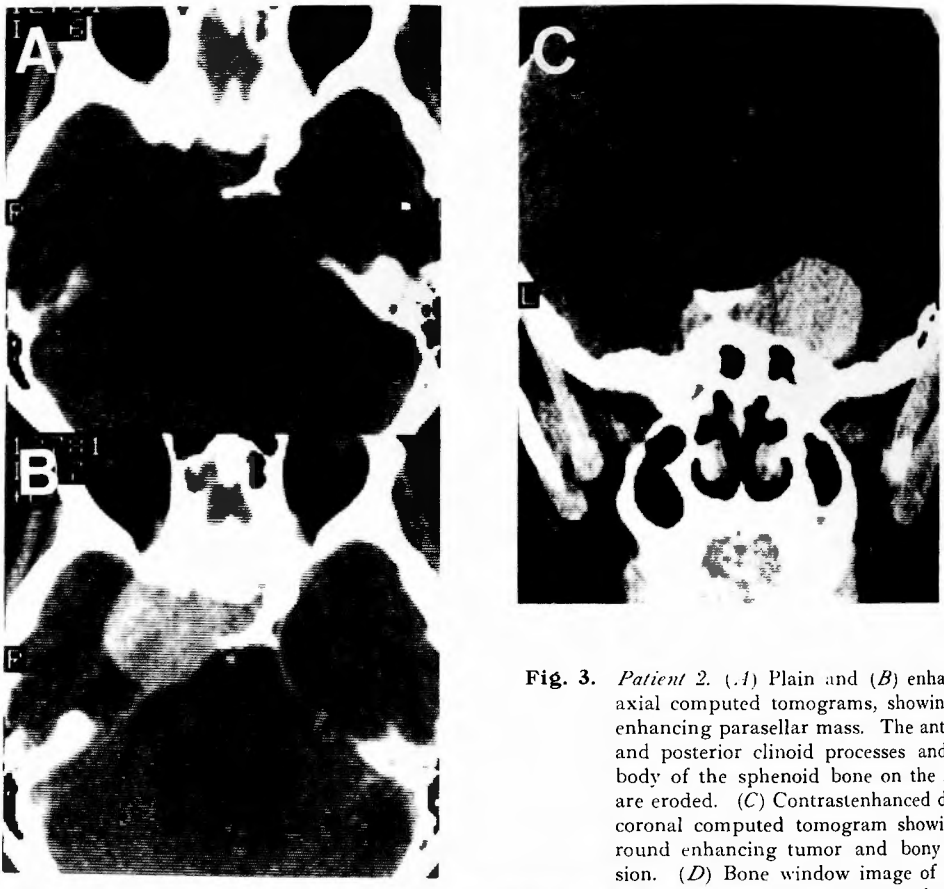

Fig. 3. *Patient 2.* (*A*) Plain and (*B*) enhanced axial computed tomograms, showing an enhancing parasellar mass. The anterior and posterior clinoid processes and the body of the sphenoid bone on the right are eroded. (*C*) Contrastenhanced direct coronal computed tomogram showing a round enhancing tumor and bony erosion. (*D*) Bone window image of axial computed tomogram, showing an erosion of the sphenoid bone.

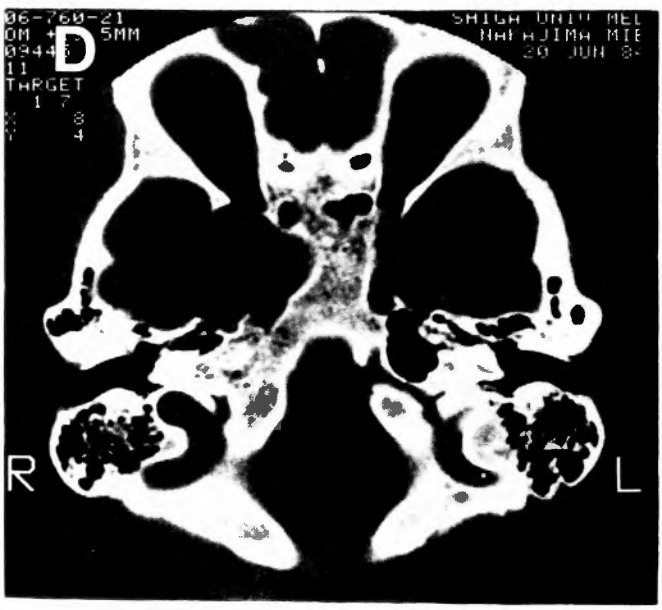

Fig. 3.

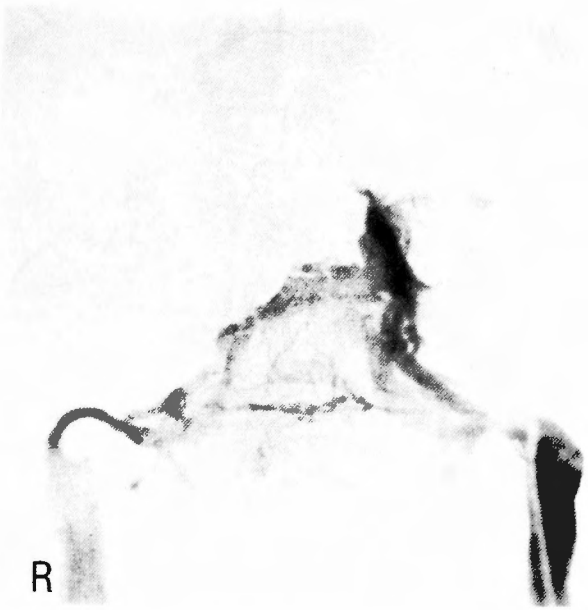

Fig. 4. *Patient 2.* Axial projection of cavernous sinography, showing an obliteration of the right cavernous sinus and the anterior intercavernous sinus.

results of routine laboratory examinations were within normal limits.

Plain radiograph of the skull showed erosion of the right anterior and posterior clinoid processes and excavation of the right half of the sella turcica. The right superior orbital fissure was enlarged. CT scan demonstrated an isodense round tumor in the right parasellar region, which was markedly and homogeneously enhanced after an administration of the contrast material (Fig. 3). The right cavernous sinus and the anterior intercavernous sinus were not opacified at cavernous sinography (Fig. 4). Right carotid angiography showed superior displacement of the cavernous and ganglionic segments of the right internal carotid artery. In addition, the inferior cavernous sinus artery and tiny tumor vessels were opacified (Fig. 5), and homogeneous tumor stain was seen on the subtraction films. Bilateral selective external carotid and left internal carotid angiograms were normal.

A right frontotemporal osteoplastic flap was raised and the parasellar region was reached intradurally. The dura mater of the medial one-third of the middle cranial fossa was elevated by an underlying compressible mass. A needle puncture of the mass resulted in profuse but non-pulsatile bleeding. The dura was incised and the spongy, highly vascular tumor was partially removed. First division of the trigeminal nerve was found to have penetrated the tumor. On removing the tumor, it became evident that the tumor arose from the cavernous sinus and extended between the two layers of the dura mater of the middle fossa. Paresthesia of her face disappeared but double vision persisted after the operation. Pathological diagnosis was a cavernous angioma.

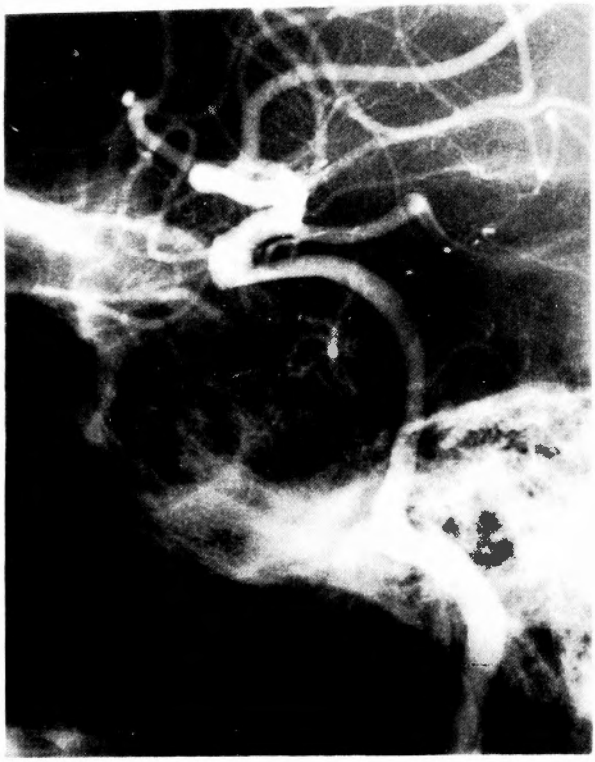

Fig. 5. *Patient 2.* Lateral projection of left carotid angiogram, showing a marked displacement and narrowing of the cavernous, ganglionic and petrous portions of the internal carotid artery. Small, irregular tumor vessels are opacified.

Discussion

Cavernous angiomas are relatively infrequent vascular anomalies involving the central nervous system. They usually occur in the parenchyma of the brain, and are characterized by an aggregation of sinusoidal blood vessels separated by fibrous septa without intervening brain tissue[15].

Intraparenchymal cavernous angiomas may occur in any age groups, and the sex distribution is equal. Of 164 such intraparenchymal lesions reported by VOIGT and YASARGIL[23], 125 (76.8%) were supratentorial and 34 (20.7%) were infratentorial in location. In the remaining 4 patients, multiple lesions were found both infra- and supratentorially. Clinical onset is usually acute or subacute, commonly with initial symptoms of epileptic seizures, acute headache and subarachnoid or intracerebral hemorrhage[4,23,24]. Conventional angiography most often reveals a hypovascular area without abnormal feeding vessels, although a subtle vascular stain, possibly associated with a few draining veins, may be detected. CT scan usually demonstrates a well demarcated collection of round densities with mild contrast enhancement and no significant mass effect. CT scan is highly sensitive diagnostic method, but its findings are by no means specific[22,24].

Extracerebral location of intracranial cavernomas are very rare. Although a few such lesions

involving the cerebellar tentorium[7,16] or the dura mater of the cerebral convexity[11] have been reported, a majority of intracranial extracerebral cavernomas in the previous papers were found in the middle cranial fossa. In a careful search of the literature, we could collect 23 such cases excluding the cases repeatedly quoted by various authors[1-3,5,6,8-10,12-14,17-22,25] Of particular interest is the fact that they often arose primarily from the cavernous sinus as was the case in our two patients[1,5,6,12,14,22,25], or at least secondarily involved it.

In sharp contrast to intraparenchymal counterparts, cavernomas of the middle cranial fossa affect predominantly the middle-aged woman. Namely, twenty-two out of 25 patients including 2 of ours were female and only 3 were male. Twelve out of 25 patients were in their fifth decade of life, although the youngest was 22 years old and the oldest was 72 years old. It seems interesting to note here that the symptoms may occur or be exacervated during pregnancy and disappear after delivery[1,25]. Unfortunately, however, sex hormone receptors in the tissue specimen have not been assayed yet. The tumor involved the left side in 10 patients and the right side in 10. In the remaining 5, laterality of the lesion was not specified or the mass occupied the midline position.

Onset of symptoms was acute in only 3 of 25 patients ; in one of them hemorrhage from the tumor was found at operation[6]. In 12, onset was insidious and the clinical course was chronic, with the interval between onset and admission ranging from 3 months to 12 years. No clinical details were given in the remaining 10 patients ; two of them died of unrelated causes and the cavernoma of the middle cranial fossa was an incidental finding at autopsy[19,21]. Most common symptoms were headaches and those of slowly progressing involvement of cranial nerves II through VII in various combinations.

Plain radiograph of the skull was normal in 2 of 16 patients, and it demonstrated destruction of the bone in the parasellar region and/or the base of the middle cranial fossa in 14. Neither abnormal calcification within the tumor nor hyperostotic changes have been described. Radionuclide brain scan was positive in all of 8 patients with large lesions. Angiography was performed in 21 out of 25 cases. In contrast to intraparenchymal cavernomas, tumor stain was present in 14 and absent in 7, and the hypertrophied feeding vessels were found at least in 5 patients. Cavernous sinography in a case of SAVOIARDO[22] and 2 of ours showed obstruction of the involved cavernous sinus. In all 9 patients in whom the CT scan was available, it demonstrated a highly and homogeneously enhancing mass with sharp margin and little or no peritumoral edema. Prior to an administration of the contrast medium, density of the mass was high in 4 patients[2,3,17,18] and similar to that of the brain in 4 (cases of HARADA et al.[5] and SAVOIARDO and PASSERINI[22] and 2 of ours). The remaining one patient showed a mixed high and low density lesion ; the low density area was proved to have corresponded to an area of previous hemorrhage[5]. Bony erosion was precisely shown on the CT images with high level and wide window settings. Thus, CT scanning obviously is the single most useful diagnostic method, but its findings are by no means specific to cavernous angiomas and the CT differentiation between meningiomas and cavernous angiomas particularly seem to be hardly possible.

Although the cavernoma of the middle cranial fossa is a well-defined extraparenchymal lesion,

a total surgical removal is difficult for two reasons. Twenty-three of 25 patients were operated upon but the mass could not be totally removed and at least 7 of them expired mainly due to massive bleeding. In the remaining 2 patients the tumor was an incidental autopsy finding[19,21]. In our experience and that of several others, however, bleeding from the lesion at operation is certainly severe but usually not pulsatile and controllable by compression and application of hemostatic agents such as oxidized cellulose. The mass is also characteristically compressible, and can be reduced in size by enucleating a part of it and applying a series of purse-string sutures with progressively large diameters on the capsule[1]. The use of surgical laser would potentially improve the operative results in the future. Another factor that often prohibits a total removal is an anatomical one that cavernomas of the middle cranaial fossa often arise from the cavernous sinus or secondarily involve it, incorporating the cranial nerves III through VI within it.

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和文抄録

中頭蓋窩の海綿状血管腫

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頭蓋内脳実質外の cavernoama は，殆どが中頭蓋窩にみられ，海綿静脈洞内やその近傍に発生して，同静脈洞を巻き込みながら徐々に拡大してゆく．この型の cavernoama は我々の2症例を加えて25例の文献報告があり，青壮年の日本人女性に多く，頭痛，第2～7脳神経の症状が妊娠によって悪化をみることがしばしば報告されている．放射線学的診断には，CT scan が最も確実で，中頭蓋窩底の骨融解像，明瞭な境界をもち強く均一な enhancement をうける dumb-bell型の腫瘍がみとめられる．しかし meningioma や転移性脳腫瘍との鑑別は必ずしも容易ではない．

外科的な全剔出は，高度に易出血性であることと，第3～6脳神経を腫瘍内に巻き込んでいることから，非常に困難である．今後，レーザーによる剔出術や，放射線治療の評価が必要である．