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ABSTRACT

The authors report a case of cavernous hemangioma in the occipital region, which resembled sinus pericranii, protruded in the recumbent posture. A 28-year-old male was admitted with a chief complaint of an occipital fluctuating mass, 5cm in diameter, accompanied by slight pain. The skull X-P was normal. A direct puncture revealed that the lesion was a blood cyst. A cystogram by percutaneous needle puncture revealed paramedian blood pooling with some draining veins but did not show any transcranial communicating vessels. A T2 weighted MR image demonstrated a well demarcated high intensity lesion just beneath the corium. The subtotally removed specimen turned out to be a cavernous hemangioma.

We discerned a conceptual confusion of pseudosinus pericranii with scalp cavernous hemangioma, based on the literature review. And we propose that scalp cavernous hemangioma, even if it changes its size according to posture, should not be simply designated as sinus pericranii.

Key words: Cavernous hemangioma, Scalp, Sinus pericranii, Magnetic resonance imaging

Cavernous hemangioma is a common disease in the cutaneous region^{2,5,11)}, but is not so familiar in the head^{1,8,16)}, especially in the scalp region. We experienced a case of cavernous hemangioma in the occipital region which had no mass effect in an upright posture but protruded in a recumbent posture, and remarkably so with the Valsalva's maneuver. We are reporting this case especially with regard to the clinicopathological correlation of this disease with sinus pericranii and the diagnostic usefulness of MRI.

CASE REPORT

A 28-year-old male was admitted to the Department of Neurosurgery at Hiroshima University School of Medicine on October 25, 1990 with a chief complaint of occipital mass. He had not had a severe head injury. He had first noticed the lesion when he was 13 years old, but had not worried about it because of its small size and the lack of pain. From about two years ago, it gradually became larger and caused slight pain when lying in a recumbent posture.

On admission, he was in good health and had no skin lesions, except the occipital one. The neurological examination was also normal. The occipital mass could not be identified in the upright posture, but it appeared as a subcutaneous immovable, ovale, non-pulsatile, and fluctuant mass, 5cm in longer diameter in the recumbent posture, an protruded remarkably when he raised intrathoracic pressure (Valsalva's maneuver). It was accompanied by port wine skin stains in the rostral and caudal adjacent regions (Fig.1). Bruit was not audible. A needle aspiration disclosed that the mass was filled with venous blood of which the O concentration was 49.9mmHg, and the CO_2 concentration was 42.4mmHg. A plain craniogram showed no abnormality. Enhanced CT revealed a subcutaneous poorly differentiated enhanced mass, difficult to distinguish from the nuchal muscles. Neither an external nor internal carotid angiogram showed any feeding vessels or tumor stain. A cystogram by percutaneous needle puncture revealed paramedian suboccipital blood pooling with some draining veins, believed to drain mainly into the vertebral plexus. but did not show any communication with the intracranial dural sinuses (Fig.2). A T1 weighted MR image revealed a slightly low signal intensity lesion, which was partially enhanced by means of a gadolinium-DTPA injection. A T2 weighted MR image, most useful for ascertaining the real extent of the lesion, revealed a well demarcated high intensity lesion, 10cm in length (rostral to caudal), extending from just below the external occipital protuberance to just behind the C2 spinous process

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Fig. 1. A photograph of the patient's occiput.left: in upright posture.right: in recumbent posture.The occiput subcutaneous mass protruded in the recumbent posture.



left

right

Fig. 2. The cystogram through the direct needle puncture.left: antero-posterior view.right: lateral view.The cyst was filled with 5ml contrast media. No opacification of the intracranial venous system was seen.



left

middle

right

Fig. 3. left: T1 weighted MRI showed an indistinct lesion with slightly low signal intensity (white arrow). middle: The lesion was partially enhanced by gadolinium injection (arrow head). right: T2 weighted MRI clearly showed the lesion with high signal intensity (arrow).



Fig. 4. upper: Photomicrograph of the tumor. The tumor is composed of numerous spaces of various sizes and fibro-collagenous strands. In some places, these spaces are contiguous to the muscle fiber and peripheral nerve. (H.E. $\times 50$)

lower: Endothelial lining (arrow) and underlying scant smooth muscle fibers (arrow head) are shown. (H.E. $\times 400$)

(Fig.3).

An operation was performed under general anesthesia in the prone position. Using a suboccipital Z shaped skin incision, the subcutaneous soft tumor was exposed. The tumor lay just beneath the corium and extended into the suboccipital muscle group. The tumor had a tough and white capsule which was filled with blood, and it could be easily detached from the corium and the cranium. However, the separation from the muscles was relatively troublesome because of the adhesion. A few small feeding arteries crossing the margin of the tumor were manageable. Slow venous bleeding occurred when the capsule was torn, resulting in the transient collapse of the tumor. The tumor was pursued into the nuchal muscles, and was finally subtotally excised.

A histological examination revealed that the tumor was composed of a honeycomb of unequal numerous spaces, sometimes large, which were separated by fibrous strands (Fig.4a). In some places, the spaces adjoined striated muscles. The stroma consisted of fibro-collagenous tissue, which was lined with a single layer of endothelial cells. Under the endothelial lining, scant smooth muscle fibers could be seen (Fig.4b). The histological diagnosis was cavernous hemangioma.

DISCUSSION

This case involved scalp cavernous hemangioma, which protruded in the recumbent posture, and by Valsalva's maneuver. In this case, increased intrathoracic pressure might have cause the engorgement of the draining veins and the subsequent distention of the blood space in the angioma, resulting in the marked swelling of the lesion.

When we first came across this kind of scalp fluc-

tuant swelling which changes its size depending on posture, our first impression was that it was an encephalocele, posttraumatic leptomeningeal cyst, or sinus pericranii. According to the literature review, eosinophilic granuloma⁶⁾ must be added. However, in this case, a plain craniogram and CT scan easily ruled out the first two kinds of lesions.

Sinus pericranii is an epicranial blood cyst, which changes its size depending on posture and has communications with the intracranial dural sinuses¹³⁾. It is only a symptom complex, so etiologically it may contain a wide spectrum^{7,9,11)}, ranging from 2) $congenital^{7}$, to 3)neoplastic1)traumatic⁴) entities^{1,10,17}). The essence of a large part of reported cases of sinus pericranii is considered to be neoplasm^{1,10,17} (cavernous hemangioma or capillary hemangioma). According to the Volkmann's proposal¹⁵⁾, true sinus pericranii is a blood filled cyst which disappears completely with pressure on the tumor, and pseudosinus pericranii is a tumor consisting of angioma or cavernoma which does not disappear completely with external pressure. Poppel¹⁰⁾ et al daringly proposed that "Sinus pericranii is a specific type of cavernous hemangioma which may or may not communicate by emissary or adventitious vessels through abnormal foramina into the skull with the dural sinuses." In contrary. Vogelsang¹⁴⁾ et al designated three cases of vascular neoplasm, which had transcranial communications with the sagittal sinus, as "cavernous hemangioma". Thus there seems to be a conceptual overlap or confusion between sinus pericranii and scalp cavernous hemangioma. This relationship is presented in Fig. 5.

In the presented case, no communication with the intracranial venous system could be discerned, and blood was believed to drain mainly into the paravertebral venous plexus. So, it might be named "pseudosinus pericranii" or "sinus pericranii without

epicranial blood cyst which changes its size depending on posture



TCC: transcranial communication with intracranial venous system

Fig. 5. Conceptual figure of the epicranial blood cyst which changes its size depending on posture.

transcranial communication" based on the characteristic clinical feature, i.e. the epicranial blood cyst changes its size depending on posture. However, sinus pericranii is only a symptom complex. So, we would like to propose that scalp cavernous hemangioma, even if it changes its size or it has transcranial communication with the dural sinuses, should not be designated as sinus pericranii but as a cavernous hemangioma based on histopathological diagnosis.

In most cases of scalp cavernous hemangioma, the conventional carotid angiogram does not show a tumor stain, because the cavernous hemangioma are fed by very a few fine arteries and the intratumoral blood flow is very slow. On the other hand, the cystogram obtained by direct needle puncture provided essential information about the tumor. However, it may not show the real extent of the lesion, since it is hindered by the intratumoral septa.

Many authors report the superiority of MRI in diagnosing hepatic cavernous hemangioma^{3,12)}. They are composed of lakes of slowly flowing blood and have a long T2 relaxation time. So, on T2 weighted images they have a significantly greater signal intensity. On the other hand, T1 weighted images do not show the tissue specificity. Conventional gadolinium administration rarely enhances the lesions because of very slow blood circulation accompanied by large vessels in this lesion.

These characteristics of cavernous hemangioma on MRI were also appeared in our case, and the MRI was more useful than the cystogram in ascertaining the real extent of the lesion and was felt to be a very useful method for diagnosing scalp cavernous angioma.

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