Neuroradiological Features of Intraosseous Cavernous Hemangioma
—Case Report—

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Abstract
A 40-year-old man presented to our hospital because of a painless lump on his right forehead. Radiography showed a radiolucent defect in the frontal bone. Bone window computed tomography demonstrated a lucent mass which expanded externally from the diploë destroying and passing through the outer plate. T₁-weighted magnetic resonance imaging revealed the lesion as non-homogeneously isointense, and T₂-weighted imaging as non-homogeneously hyperintense. The lesion was enhanced non-homogeneously after contrast administration. The lesion was subtotally removed. Histological examination suggested cavernous hemangioma. Preoperative examinations could not provide a definitive diagnosis, which was established by the operative finding. Because of the difficulty of diagnosis in the early stage, surgical treatment in the early stage is recommended for total removal and definitive diagnosis of intraosseous tumor.

Key words: cavernous hemangioma, frontal bone, intraosseous tumor

Introduction
Primary intraosseous hemangioma is a rare, benign, slow-growing neoplasm, and more than 50% of cases occur in the vertebrae or skull. Primary intraosseous hemangiomas of the cranium are rare benign tumors comprising only 0.2% of all osseous neoplasms, and account for 7% of all skull tumors. The parietal and frontal bones are the most common sites of involvement. These lesions are usually solitary and occur more frequently in females than males, in the ratio of three to one, and are typically found in adults, although persons of any age may be affected. The pathogenesis of these neoplasms is unknown, but the causes are believed to be either congenital or related to previous trauma.

The differential diagnosis of skull tumors is frequently difficult because of the variety. Therefore, the majority are identified only after surgery and biopsy. We present a typical case of intraosseous hemangioma arising from the frontal bone, and discuss the radiological characteristics, operative view, and the pathological analysis.

Case Report
A 40-year-old man consulted our hospital because of a painless lump on his right forehead which he had first noticed 4 years previously, but had not sought medical attention because of the absence of other complaints. The lump had grown slowly and gradually, so he consulted a hospital about 2 years ago. The lesion was suspected to be osteoma, although not investigated adequately, and was observed conservatively. Recently, the lump had begun to bulge further with stimulating pain of the scalp, so he presented to our hospital on September 19, 2000.

A lump of 2 cm diameter and 4 mm in height was recognized in the right forehead just above the eyebrow. The lump was bony hard, with a smooth surface and no mobility. The scalp over the lesion was normal without adhesion to the mass. Both laboratory data and neurological findings were normal. Radiography in axial and lateral views showed a 1.5 × 2.0 cm radiolucent defect in the frontal bone just superior to the supraorbital margin and lateral to the frontal sinus (Fig. 1). Soft x-ray radiography showed bony expansion. Computed tomography (CT) revealed a bone dense mass growing from the frontal bone by expansion of the outer table but not in-
Fig. 1 Radiograph in the lateral view showing a 1.5 × 2.0 cm radiolucent defect (arrows) in the frontal bone just superior to the supraorbital margin and lateral to the frontal sinus.

Fig. 2 The bone window image of computed tomography scan demonstrating the lucent mass as an irregular reticular pattern expanding externally from the diploë destroying and passing through the outer plate (arrow).

Fig. 3 T₁-weighted magnetic resonance (MR) image (A) showing the lesion as non-homogeneously isointense (arrow), and T₂-weighted MR image (B) showing the lesion as hyperintense (arrow). The lesion was enhanced non-homogeneously after contrast administration (C, arrow).

The lesion was enhanced non-homogeneously after administration of contrast medium (Fig. 3C). Thallium-201 chloride bone scintigraphy revealed accumulation of radioisotope in the lesion. Digital subtraction angiography showed no particular vascularization of the lesion.

Surgical resection was performed on October 19. A right arcade incision was performed 2 cm posterior to the hair line and the skin flap was

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reflected anteriorly. The tumor had developed from the frontal bone and partially adhered to the periosteum and subgaleal tissue, so the adherent area moderately bled during detachment. The surface of bulging area was macularly dark blue and distinctly separated from the surrounding normal bone (Fig. 4). The frontal bone was hollowed with a pneumatic drill, and the tumor was removed en bloc, but the part adjacent to the frontal sinus was left to avoid perforating the roof of the sinus. Moderate bleeding from this part was controlled with cauterization and bone wax. Cranioplasty was performed by filling resin into the bone defect. The section of the removed bone revealed a vascular separated space. Gross examination of the outer table was similar, but the inner table was intact. Histological examination revealed bone trabeculae widely separated by loose connective tissue enclosing multiple thin-wall vascular spaces lined by endothelial cells, suggesting cavernous hemangioma1) (Fig. 5).

Discussion

Radiography of the cranium is the most effective method to identify intraosseous hemangioma8) as an intradiploic, extensive, well-defined area of rarefaction with a honeycomb configuration on axial views and a classic sunray pattern of trabeculation on tangential views.9,13,14) There is usually no reactive sclerosis at the margins.14) This classical feature may be absent in many cases, like ours, which present only as lytic or dense bone expanding masses.3,6,14) Calvarial hemangiomas usually grow by expansion of the outer table and rarely grow intracranially.8,14) The characteristic calvarial radiographic sunray pattern of radiating trabecula with or without a thin peripheral sclerotic rim was not present in our case. Angiography is important in the preoperative surgical planning and embolization for reducing the vascularity of large tumors, but small tumors usually have poorly developed or unrecognizable vascularity. CT revealed an intradiploic lytic mass with rarefaction and a honeycomb pattern.4,10,12) CT is more helpful than MR imaging or other neuroimaging modalities in planning surgery, because it shows the site and extent of tumor better in bone windows.10)

T1-weighted MR imaging shows the lesion as non-homogeneously intense. The bulk of the lesion is isointense with gray matter, with both hyperintense and hypointense foci scattered throughout the lesion.11) Such signal intensity reflects the fatty contrast of these lesions.7) Therefore, the absence of hyperintense areas in our case may reflect a relatively low fat content of the lesion. High signal intensity on T2-weighted MR imaging may be caused by pooling of blood or slow-flowing blood.11,12) Rapid serial MR imaging with bolus injection of gadolinium pentetate dimeglumine demonstrated enhancement in focal areas in the early phase and spread into the entire lesion in the later phase.5) These features may be characteristic of intraosseous hemangioma.

The differential diagnosis of slow-growing intraosseous neoplasms includes osteoma, aneurysmal bone cyst, giant cell tumor, fibrous dysplasia,
Langerhans' cell histiocytosis, sarcoma (particularly rhabdoid myosarcoma), meningioma, metastatic disease, hematic cyst, and dermoid tumor. These tumors are not difficult to identify by epidemiological and neuroimaging methods, but the frequent atypical or small cases are not so easy to characterize. Classic radiological features can sometimes differentiate these tumors, but the final diagnosis is established in the majority of cases only after surgery and biopsy.

In our case, radiography revealed the radiolucent lesion, but not the classical features of honeycomb configuration or sunray pattern. Cavernous hemangioma was highly suspected based on the origin, location, expanding pattern, and CT, MR imaging, and scintigraphy findings. In particular, the MR imaging findings of isointensity on T1-weighted images, hyperintensity on T2-weighted images, and obvious enhancement after administration of contrast medium indicated this tumor. However, the definitive diagnosis was only established at operation with the recognition of cavernous hemangioma by the mottled appearance. Tumors of the skull base or orbit tend to become large before intervention, and thus are difficult to remove totally. In contrast, tumors of the calvarium tend to be detected at the early stage from the bulging feature. However, many types of intraosseous tumor grow slowly and are sometimes difficult to diagnose in the early stage, so are frequently observed conservatively until the late stage. When our patient presented at our hospital, the tumor had progressed to the orbital margin and upper wall of the frontal sinus.

Because of the difficulties in the diagnosis in the early stage and treatment in the late stage, early surgical treatment of intraosseous tumors is recommended for total removal and definitive diagnosis.

References

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