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The Dural Lucent Line:
Characteristic Sign of Hyperostosing Meningioma en plaque

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Hyperostosis of the skull associated with en plaque form of meningioma may present a diagnostic challenge, since the intracranial part of the tumor is not visualized by skull radiography, computed tomography (CT), or other neuroradiologic methods. The authors report four cases of hyperostosing meningioma en plaque demonstrating a characteristic feature: a subdural layer of ossification along the hyperostotic bone with a dural lucent interface. Polytomography or high-resolution CT at bone window settings is necessary to identify the dural lucent line. The absence of this sign does not exclude meningioma en plaque.

Hyperostosis is the most common skull change associated with meningioma, occurring in 23%–44% of cases [1–3]. The recognition of hyperostosing meningioma is not difficult in the globular (en masse) form, in which the intracranial part of the tumor may be detected readily by computed tomography (CT) or even on plain skull radiographs, if it is calcified. However, hyperostosis of the skull associated with the flat (en plaque) form of meningioma may present a diagnostic challenge, since the intracranial part of the tumor is not visualized by plain skull radiography or CT. Furthermore, cerebral angiography may be normal in this form of the entity. The radiographic diagnosis relies on the bony changes of the skull; however, differentiation from osteoma, fibrous dysplasia, osteoblastic metastasis, and various other hyperostosing entities is difficult. We report four cases of hyperostosing meningioma en plaque, two surgically verified, demonstrating a radiographic feature that we believe is characteristic of this entity.

Case Reports

Case 1

A 47-year-old man had a gradually increasing lump in the left frontoparietal region of the skull for several years. In recent months, the patient had had episodic loss of vocabulary and tingling in his right hand and arm.

Plain skull films showed an area of dense hyperostosis in the left frontoparietal region (fig. 1A). CT showed no intradural mass, but there was edema in the left frontal lobe. A polytomogram in anteroposterior view showed the sclerotic hyperostosing lesion to be homogeneous; the inner, middle, and outer tables were indistinct. There was a thin layer of calcium density along the inner table of the hyperostotic bone with a lucent interface (fig. 1B). Cerebral angiography showed hypertrophic changes of the meningeal artery in the area of the hyperostotic bone.

The hyperostotic bone was surgically removed en bloc. The central part of its inner surface was irregular. The underlying dura was friable in the center and was noted to be hardened in this region on palpation. The dura was opened, revealing a whitish gray hard plaque between its inner surface and the arachnoid. The dural-subdural hard plaque complex, which adhered to the underlying brain in some areas where the arachnoid was thickened, was excised. On microscopic examination, the subdural hard plaque was found to be an ossified layer containing meningioma cells (figs. 1C and 1D). Meningioma cells were found also within the hyperostotic bone.

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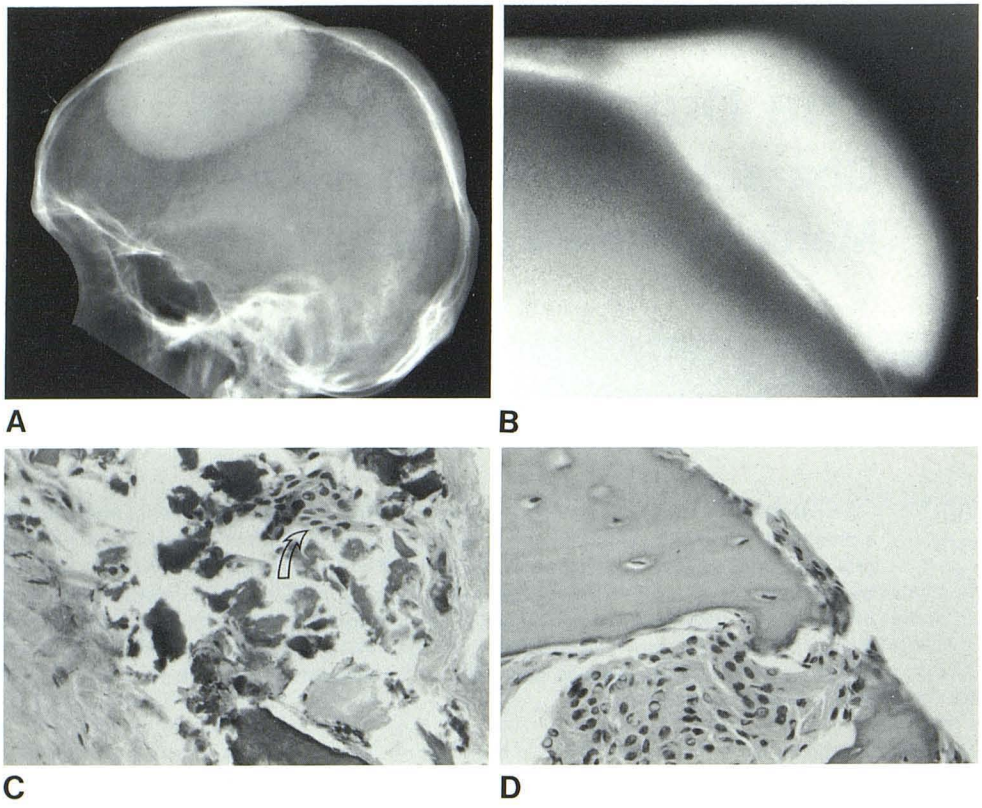

Fig. 1.—Case 1. **A**, Plain skull radiograph. Well defined area of dense hyperostosis in left frontoparietal region. **B**, Anteroposterior polytomogram. Inner, middle, and outer tables are indistinct. Thin layer of calcium or bone density along inner table of hyperostotic bone with interfacial dural lucent line. Photomicrographs of excised dural-subdural hard plaque before (**C**) and after (**D**) decalcification show ossification and meningioma cells (*arrow*) along inner layer of dura.

Case 2

A 35-year-old woman had blurred vision in her right eye for 2 months. Her neurologic examination was normal except for decreased vision in the right eye.

Plain skull films showed hyperostosis and pneumatization of the right sphenoidal lesser wing. Polytomograms in the anteroposterior (fig. 2A) and lateral views showed a calcium plaque along the inner table of the hyperostotic bone with a lucent interface. High-resolution CT at bone window settings in coronal and axial views again demonstrated a calcium plaque along the inner table of the hyperostotic bone and separated from it by a lucent line (figs. 2B and 2C). The right optic canal appeared narrowed by bony overgrowth. There was no intracranial mass or abnormality in the brain. The cerebral angiogram was normal.

At surgery the dura around the right anterior clinoid was found embedded between the hyperostotic bone and an intradural bony plaque. No soft-tissue tumor was identified. The optic foramen proved to be narrowed by the bony overgrowth and was unroofed. Microscopic examination showed the subdural bony growth to be ossification containing meningioma cells, which were noted also within the hyperostotic bone.

Case 3

A 90-year-old woman was admitted because of an episode of blackout resulting in numbness of the right arm, cheek, and lip. She had had congestive heart failure for over 10 years. The neurologic examination was normal.

Plain skull films showed a large area of dense hyperostosis in the right frontoparietal region (fig. 3A). CT showed no intracranial mass

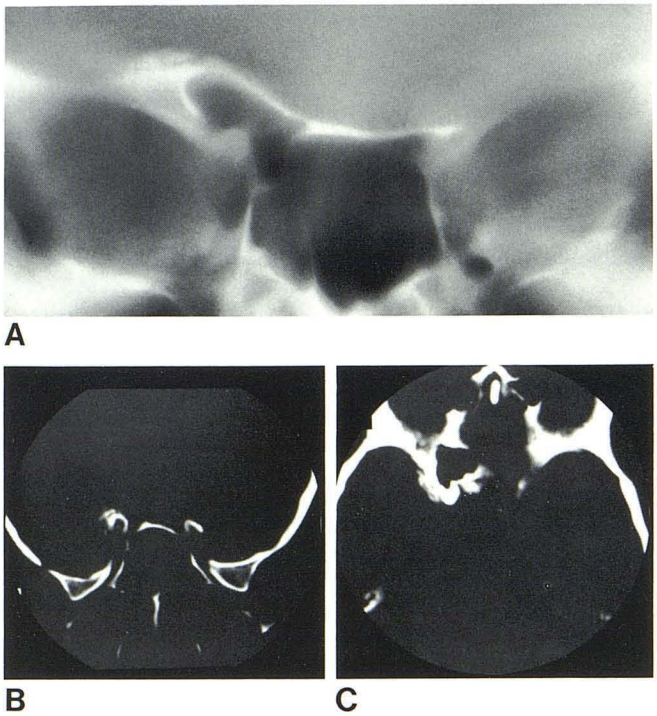

Fig. 2.—Case 2. Anteroposterior polytomogram (**A**) and high-resolution CT scans at bone window settings in coronal (**B**) and axial (**C**) views. Dense hyperostosis and pneumatization of right sphenoidal lesser wing. Subdural calcium or bone plaque along hyperostotic bone with dural lucent interface.

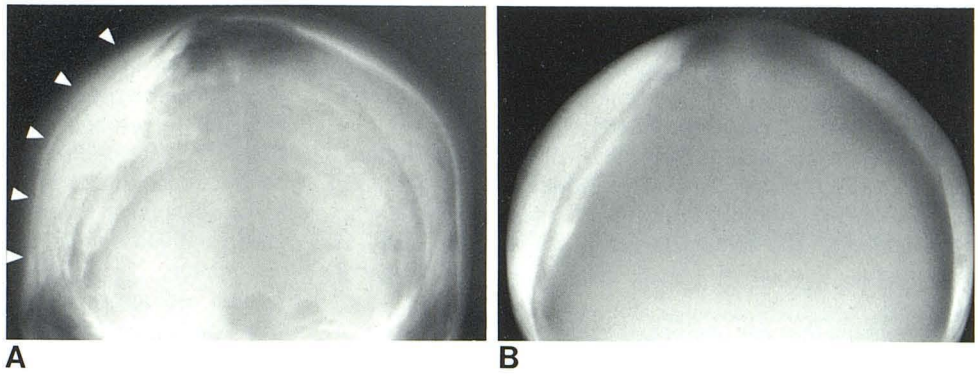

Fig. 3.—Case 3. **A**, Plain skull radiograph. Dense hyperostosis in right frontoparietal region (*arrowheads*, outer aspect of hyperostotic bone). Calcium or bone density along inner table of hyperostotic bone with interfacial dural lucent line. **B**, Anteroposterior polytomogram. Inner, middle, and outer tables of hyperostotic bone are indistinct. Subdural layer of calcification or ossification and the dural lucent interface.

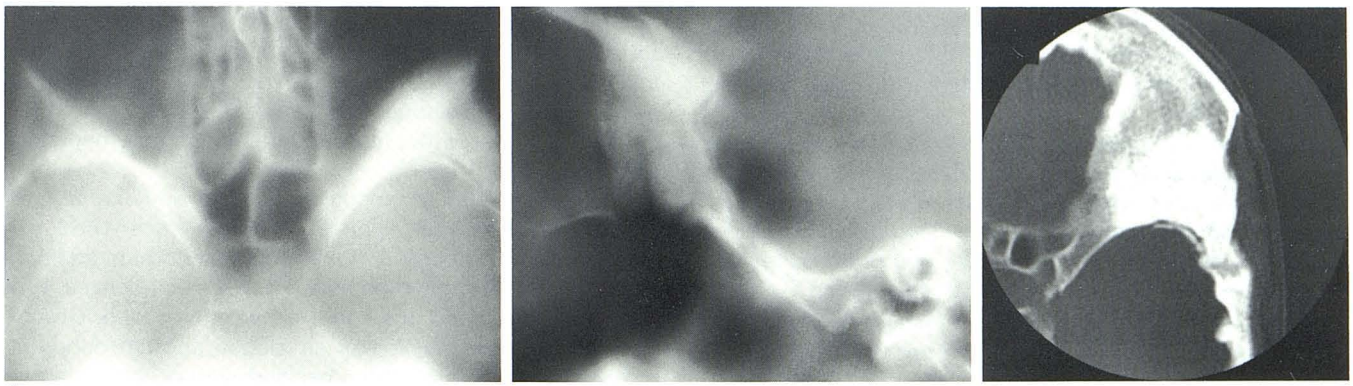

Fig. 4.—Case 4. Basal (**A**) and lateral (**B**) polytomograms. Diffuse, dense hyperostosis of left sphenoidal wings. Thin subdural layer of calcium or bone density along hyperostotic bone with dural lucent interface. **C**, Axial high-resolution CT scan at bone window setting. Thin subdural layer of calcium or ossification separated from hyperostotic bone by dural lucent line.

or abnormality in the brain. A polytomogram in anteroposterior view showed the sclerotic hyperostosis to be homogeneous with indistinct inner, middle, and outer tables. There was a layer of calcium or bony density along the inner table of the hyperostotic bone with a dural lucent interface (fig. 3B). The patient was discharged with a diagnosis of hyperostosing meningioma en plaque.

Case 4

A 60-year-old woman had mild unilateral exophthalmos in the left eye. Her vision was unaffected and the neurologic examination was normal.

Plain skull films showed dense hyperostosis in the greater and lesser wings of the left sphenoid bone. Polytomograms in the basal and lateral views showed a thin layer of calcium density along the inner table of the hyperostotic sphenoid wing, separated from it by a lucent line (figs. 4A and 4B). High-resolution CT at bone window settings in axial view again showed a thin subdural layer of calcium density along the inner table of the hyperostotic bone with a dural lucent interface (fig. 4C). There was no intracranial mass or abnormality in the brain. The cerebral angiogram was normal. The patient was discharged with a diagnosis of sphenoid wing meningioma en plaque; follow-up examination was recommended.

Discussion

Hyperostosis associated with meningioma generally has been attributed to invasion of the adjacent bone by tumor cells. The affected bone frequently contains tumor cells in the diploe or Haversian canals [4–6]. The precise mechanism of hyperostosis is not yet established. Cushing [2] and other investigators [6–8] thought meningioma cells stimulated the normal osteoblasts to produce new bone without acting directly as osteoblasts themselves. Freedman and Forster [9], on the contrary, believed that bone growth in cranial hyperostosis associated with meningioma was a function of tumor cells, since the tumor cells of meningioma could produce fibroblasts, osteoblasts, and osteoclasts. This theory has not received wide support.

In some cases hyperostosis has occurred at a considerable distance from the tumor, especially in the en plaque type in the sphenoid ridge [4, 10]. The reason for this reaction is poorly understood. Kolodny [11] suggested that hyperostosis was related primarily to osteoblastic stimulation resulting from vascular dilatation and stasis, rather than to invading tumor. Rowbotham [10] believed the structure

of hyperostosis depended not only on the nature of the invading tumor, but also on the properties of the affected bone. The route of invasion generally is believed to be via tumor emboli through vascular connections or direct extension through the dura [7, 9, 10, 12].

Meningioma en plaque is characterized by sheetlike subdural spread along the plane of the meninges with the major connection being to the dura and minimum attachment to the arachnoid. The tumor covers a considerable area in some cases. Vascularity usually is minimal in this type of tumor [4].

For reasons that are unclear, meningiomas en plaque are more likely to provoke adjacent bony hyperostosis from tumor invasion than are the larger globular tumors [2, 5, 9]. The amount of hyperostosis often is disproportionate to the relatively small tumor [2]. It is this bony change that produces the clinical symptoms and signs by pressing against the adjacent structures [2, 13]. Meningioma en plaque arises commonly in the sphenoid ridge and the convexity [2, 4].

Hyperostosis associated with this type of meningioma is almost always sclerotic [13–15]. Early changes may be limited to the inner table of the skull [16]; however, in most cases the sclerotic hyperostosis becomes homogeneous and diffuse so that the inner, middle, and outer tables are not easily distinguished at the time of diagnosis [14].

The difficulty in diagnosing hyperostosing meningioma en plaque stems from the fact that the intracranial part of the tumor is hardly recognizable radiographically. (The globular form, in contrast, is identified readily by CT and other neuroradiologic methods.) The common feature in our four cases was a thin layer of calcium or bony density along the inner table of the hyperostosing bone with a lucent interface. On surgical inspection and microscopic examination, the layer of calcium or bone density seen radiographically proved to be a subdural plaque of ossification containing meningioma cells (cases 1 and 2; figs. 1C and 1D).

Some meningiomas are known to be ossified within their substance [1, 2]; it is possible that ossification may have developed within the en plaque meningioma itself. However, the occurrence of ossification within the meningioma is reportedly very rare [1, 7, 10]. Another, perhaps more plausible theory is that meningioma en plaque with its diffuse subdural infiltration may elicit metaplastic transformation of the inner layer of the dura mater, resulting in a subdural layer of bone formation. The inner layer of the dura mater, being mesothelial in origin, is capable of undergoing metaplastic change and bone formation [17], and ossification in the falx and tentorium of normal individuals is well known [18].

The lucent interface between the hyperostosing bone and the opposing subdural layer of ossification represents the dura mater, whose soft-tissue density remains unchanged. The affected dura mater may be either thickened to varying degrees or thinned or fragmented in the central portion [2, 6, 7, 13]. The dural lucent layer is observed also between the hyperostotic bone and the intradural calcified mass in cases of hyperostosing and calcified meningioma en masse.

The combination of hyperostotic bone and opposing subdural ossification with an interfacial dural lucent line appears to be a characteristic feature of a meningioma en plaque. Although histologically not proven, the diagnosis of hyperostosing meningioma en plaque in cases 3 and 4 is justified by the presence of this unique feature. This feature may not be present if (1) a subdural ossified plaque is absent or (2) the interfacial dura is destroyed by invading tumor or ossified both in its inner and outer layers. Osteoma, fibrous dysplasia, osteoblastic metastasis, or other hyperostosing entities should not give rise to a subdural layer of ossification.

Skull radiographs demonstrated the dural lucent line in only one of our four cases (case 3). The dural lucent line was not identified in two cases (cases 1 and 3) when an older-generation scanner was used. However, the dural lucent line was visualized in both cases 2 and 4 by high-resolution CT at bone window settings, and in all four cases by polytomography. Polytomography and high-resolution CT appear to be the most reliable methods for detecting this thin lucent interface between the hyperostotic bone and the subdural ossified plaque. The plane of section should be tangential to that of the meninges underlying the hyperostotic bone.

In reviewing 100 consecutive cases of intracranial meningioma seen between April 1977 and November 1982, we found three proven cases of the en plaque form in addition to the four cases presented here. Of these three cases, two were sphenoidal and one had a convexity location. One of the sphenoidal meningiomas en plaque showed subdural ossification, but no subdural ossified plaque was noted in the other two cases. The dural lucent line was not demonstrated in the case with subdural ossification, possibly because an older-generation CT scanner was used and polytomograms were not obtained. Thus, five of seven cases (including the four cases presented here) had a subdural plaque of ossification, and four of these five demonstrated the dural lucent line.

Our cases are too few to draw meaningful conclusions about the incidence of the subdural ossified plaque and the reliability of the dural lucent line as a sign of hyperostosing meningioma en plaque. However, the fact that the four cases presented here have been encountered at our institution (a 1,000 bed general hospital) within a 2 year period suggests that the occurrence of subdural ossified plaque may not be rare and that the dural lucent line is a useful radiographic sign of its presence.

Meningioma en plaque should be suspected in patients with hyperostosis in the sphenoid wing or the convexity of the skull. Polytomography or high-resolution CT at bone window settings is recommended to identify a subdural plaque of ossification along the inner aspect of the hyperostotic bone. The subsequent finding of a dural lucent line between the hyperostotic bone and the subdural ossification is believed to be a characteristic radiographic sign of meningioma en plaque. However, the absence of this sign does not exclude meningioma en plaque.

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