Letter to the Editor

Effect of Etidronate on Intraosseous Meningioma

To the Editor;

Intraosseous meningioma is a rare disorder that develops outside in the meninges while usual meningiomas occur in the intradural regions [1]. We here present the case of 60-year-old Japanese woman who developed intraosseous meningioma localized in the left parieto-occipital region. The patient complained of deformity of head and continuous tinnitus. Skull roentgenogram exhibited hyperosteotic changes in the left parieto-occipital area. Bone scintigraphy using technetium-99m diphosphonate exhibited a specific high uptake in the parietal region in whole body scanning (Fig. 1A), but neither thallium nor gallium was significantly accumulated in the same area of the skull. Serum alkaline phosphatase, calcium, and inorganic phosphate levels were within normal ranges, and the levels of serum intact parathyroid hormone and other bone resorption markers in urine were also normal. Cranial magnetic resonance imaging (MRI) showed an expanded and thickened skull in the left parietal area with local enhancement by gadolinium (Fig. 1B). Bone biopsy from the skull lesion exhibited the proliferation of meningoceles with calcified psammoma bodies and the increased number of cement lines and trabecular plates, which was pathologically compatible with intraosseous meningioma and secondary hyperostosis (Fig. 1C).

Meningiomas account for approximately 15% of primary intracranial tumors and occur as intradural tumors of the syncytial, fibroblastic or transitional type. On rare occasions, meningiomas also occur in extradural lesions including calvarium, paranasal sinuses, parotid glands, neck and skin [1, 2]. In these cases ectopic meningiomas seem likely to arise outside of meninges from nests of arachnoid cells along the lines of fusion of the embryonic skull or otherwise from multipotential mesenchymal cells [2].

The symptomatic cases of intraosseous meningioma should be surgically treated by excising the tumor and by reconstructing the bones as required [3]. In the present case, however, we could not attempt the surgical removal of meningioma and osteoplasty since the meningioma was widely spread across the sagittal sinus in the parieto-occipital skull. Therefore, in order to suppress the reactive hyperostosis, etidronate disodium 200 mg/day was administered daily for 6 months. After

![Fig. 1](image-url) (A) Bone scintigraphy in the whole body. (B) Cranial MRI finding. (C) Microscopic finding of the skull biopsy (hematoxylin-eosin staining, ×100). (D) Changes of bone scintigraphy of the skull before and 6-month after the treatment with etidronate disodium.
a series of the etidronate therapy, her tinnitus was clearly ameliorated and the bone scintigraphy showed apparent reduction of the tracer accumulation in the skull lesion (Fig. 1D). Among 39 reported cases of intraosseous meningioma, the most common site of involvement is the orbit followed by the fronto-parietal region and anterior cranial fossa [1–3]. Hyperostosis is the most common radiographic abnormality, and is found in 60% of the cases. The origins of the primary intraosseous meningiomas are postulated to be arachnoidal cells that accompany the vessels, existing nerves or periosteum attached to the sutures. Although intraosseous meningioma is, in general, clinically benign and usually asymptomatic, it may cause localized swellings and develop into local compressive symptoms such as hearing loss, tinnitus, visual disturbance, headache and ill-defined sensations in the skull with its enlargement [4]. Surgical resection is the only curative treatment if the location is suitable for extensive operation.

The clinical features of intraosseous meningiomas including bone scintigraphy and roentgenogram were quite similar to those of skull Paget’s disease [1], which is a more common condition than intraosseous meningioma. The increase of serum alkaline phosphatase level or the biochemical signs of bone-remodeling excess can be a good hallmark to differentiate Paget’s disease from other hyperosteotic disorders even without pathological diagnosis. The most common sites of Paget’s disease are the lumbar spine, pelvis, skull, femur and tibia, in which multiple regions are often involved [5]. Bisphosphonate administration is a very effective therapy for Paget’s disease [5]; however, it is also considerable therapy for reducing the symptoms due to skull compression caused by reactive hyperostosis in intraosseous meningiomas unless the tumor is surgically removable.

References


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