Primary osteolytic intraosseous meningioma of the frontal bone

Pierwotny śródkostny oponiak osteolityczny kości czołowej

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Abstract

Almost 1-2% of meningiomas are lesions described as ectopic or extradural meningiomas. Primary intraosseous meningiomas are a rare form of intra-bone tumours that account for approximately 67% of extradural meningiomas.
A 41-year-old male patient presented with a headache and a bulge at the right frontal region. Cranial computed tomography displayed a hyperostotic lesion. Magnetic resonance imaging showed enhancement of the bone lesion after injection of gadolinium. A burr hole was drilled at the thickest section of the lesion, and pathological examination of the bone dust extracted from this site was performed. The pathological study indicated the presence of a meningioma. Right frontal craniectomy was performed and the hyperostotic bone was resected. No invasion was observed at the dura. A calvarial defect was reconstructed during the same session with methyl methacrylate cranioplasty.

Key words: intraosseous meningioma, osteolytic, resection.

Streszczenie

Oponiaki ektopowe lub zewnątrzoponowe stanowią 1–2% wszystkich oponiaków. Pierwotne śródkostne oponiaki są rzadką postacią guzów śródkostnych i stanowią ok. 67% wszystkich oponiaków zewnątrzoponowych.
Czterdziestojednoletni mężczyzna zgłosił się z powodu bólu głowy i uwypuklenia w okolicy czołowej prawej. W tomografii komputerowej uwidoczniono zmianę powodującą hiperostozę. W badaniu za pomocą rezonansu magnetycznego stwierdzono wzmocnienie sygnału w obrębie zmiany po wstrzyknięciu gadolinu. W miejscu, w którym zmiana była najszersza, wywiercono otwór trepanacyjny, a pył kostny poddano badaniu histopatologicznemu. Po stwierdzeniu obecności oponiaka wykonano prawostronną kraniektomię czołową i wycięto zmienioną kość. Nie stwierdzono naciekania opony twardej. Podczas tej samej operacji wykonano kranioplastykę ubytku kostnego za pomocą metakrylanu metylu.

Słowa kluczowe: oponiak śródkostny, osteolityczny, wycięcie.

Introduction

Meningiomas commonly originate from the arachnoid cap cell bundles located on the external layer of the arachnoid membrane. Almost 1-2% of meningiomas are lesions described as ectopic or extradural meningiomas, which have no association with the surface of the arachnoid membrane [1]. Primary intraosseous meningiomas are a rare form of meningiomas that account for approximately 67% of extradural meningiomas. Most of these tumours are calvarial meningiomas [2] rather than the osteolytic form of primary intraosseous meningiomas, which is the least common.

In this report, we describe a case of primary osteolytic intraosseous meningioma of the frontal bone.

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Case report

A 41-year-old male patient presented with a headache and a bulge at the frontal region. Neurological examination was normal but cranial computed tomography (CT) displayed a hyperostotic lesion within the frontal bone. Cranial magnetic resonance imaging (MRI) indicated a lesion of 6 × 6 cm, with the thickest part measuring 16 mm. After intravenous gadolinium infusion, contrast retention was observed at the bone lesion (Fig. 1).

A burr hole was drilled at the thickest section of the lesion, and a pathological study of the bone dust extracted from this site was performed. The pathological study indicated the presence of a meningioma. Right frontal craniectomy was performed and the hyperostotic bone was resected. No invasion was observed at the dura. A calvarial defect was reconstructed during the same session with methyl methacrylate cranioplasty.

The pathological study assessed the tumour as an intraosseous osteolytic meningioma. We observed scattered neoplastic infiltration in the tumour cross-sections, which formed whorl structures consisting of diffuse atypical meningothelial cells between the bone lamellae. Cellularity was considerably high and 1-2 psammoma bodies were observed at mitosis in 10 high-powered field (HPF). Morphological findings revealed osteolytic diffusion in the bone structure that was assessed as meningotheliomatous meningioma (grade 1 according to WHO 2007) (Fig. 2). No post-operative complications were observed.

Discussion

Meningiomas that are not found on the arachnoid surface are recognized as ectopic or extradural meningiomas. A large percentage of meningiomas are primarily intradural lesions located at a subdural distance. Nevertheless, extradural meningiomas account for 1-2% of all meningiomas and can localize themselves in the bone, skin, nasopharynx and neck [1].

Primary osseous meningiomas are rarely localized inside the bone. These meningiomas account for 2/3 of

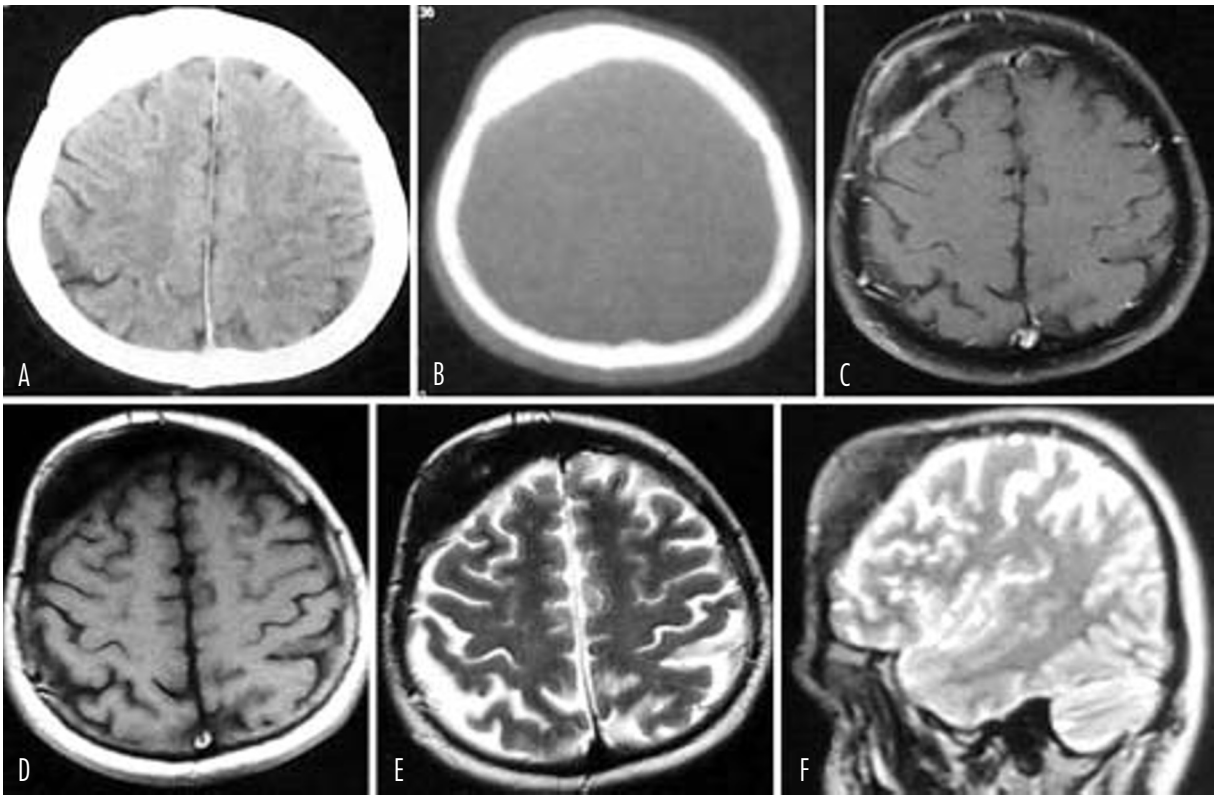

Fig. 1. CT scan with bone window (A, B) demonstrates a right-sided frontal hyperostotic mass expanding the calvaria. Axial post-contrast T1-weighted (C), T1-weighted (D), T2-weighted (E) and sagittal T2-weighted (F) MR images show the hypointense right frontal intracalvarial mass lesion. After intravenous gadolinium infusion, contrast retention is observed at the bone lesion (C)

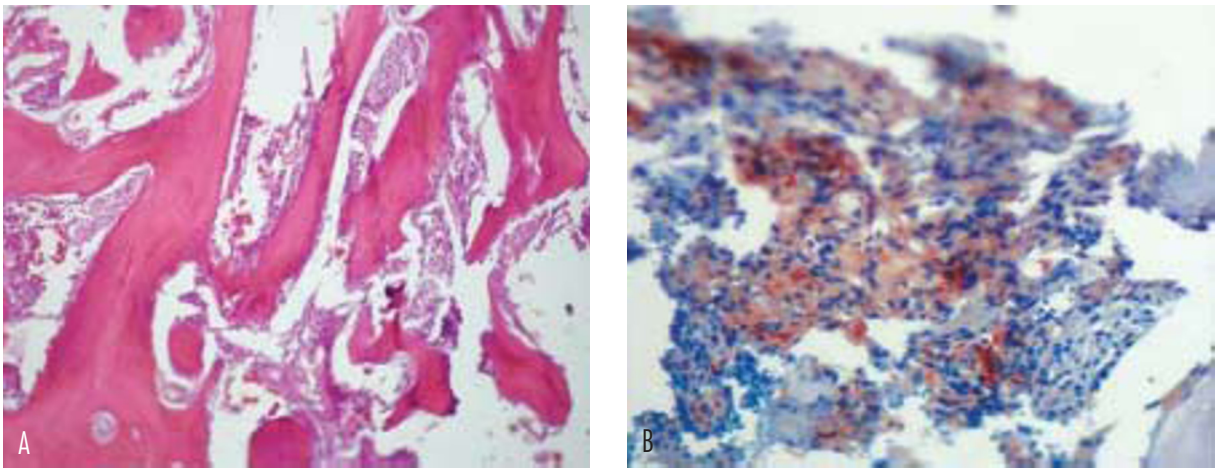

Fig. 2. (A) Photomicrographs showing islands of osteolytic meningotheliomatous tumour cells organized in clusters among thick trabecular bony lamellae (HE 100×); (B) immunohistological study (+) staining was observed with epithelial membrane antigen (EMA 100×)

all extradural meningiomas, and the majority of these tumours are located at the calvarium [2,3]. In our case, the tumour was detected as a regional puffiness located at the right frontal zone in the calvarium.

There are several theories concerning the pathogenesis of primary intraosseous meningiomas. Arachnoid cells accompany cranial nerves and may change their location towards the calvarial foramina. These cells can convert to meningioma cells. Arachnoid cells may also change their location by means of arterial sheat that feed the periosteum and calvarium [4]. In our case, the tumour probably arose from resting intraosseous arachnoid cells because there was neither an overt history of trauma nor proximity to the cranial sutures. It is possible, however, that this intraosseous tumour might have emerged from the adjacent dura because the subjacent inner table of the skull and the dura were disrupted at the central portion of the lesion.

Lang proposed a general classification of primary extradural meningiomas [3]. Accordingly, the tumour is classified as type I (purely extracalvarial), type II (purely calvarial) or type III (calvarial lesion extending beyond the calvaria). Each type is further divided into subgroups, recognized as B for the skull base and C for convexity. Using this classification, intraosseous meningiomas were classified as type II or type III due to the presence of an extracalvarial extension. In the present case, the tumour was classified as a type II C tumour.

Intraosseous meningiomas are divided into subgroups known as osteoblastic (hyperostotic), osteolytic and mixed. A focal hyperdense lesion can be observed in the CT of the osteoblastic subgroup. Therefore, meningioma en plague, osteoma, osteosarcoma, Paget's disease and fibrous dysplasia, Brown tumour, multiple myeloma, plasmacytoma, giant cell tumour, aneurismal bone cyst, eosinophilic granuloma and metastatic cancer should be considered in differential diagnosis [3-5].

Primary intraosseous meningiomas rarely appear as osteolytic calvarial lesions [1,4-17]. Nevertheless, these lesions may display a hypodense appearance similar to other primary lytic calvarial lesions observed in radiographs. Osteolytic lesions may appear in CT as separated and widened internal and external layers of the calvarium rather than a thickened sclerotic bone. In this case, a compact piece of bone was detected and outlined by a hypodense border zone. These features are rather atypical for other tumours included in the differential diagnosis [4]. However, the present case involves an osteolytic-type meningioma, and a focal hyperdense appearance was present in CT [2,7]. In our case, MRI showed contrast enhancement of the lesion after gadolinium injection.

If intraosseous meningiomas are amenable to surgical removal, then a wide incision should be used. Ideally, cranial reconstruction should be performed during the same session. In lesions of the skull base, where total resection may not be possible, the therapeutic goal should consist in decompression of vital neural structures [2,5]. Surgical intervention was undertaken in our case, and the lesion located at the frontal bone was totally resected by craniectomy. The calvarial defect was reconstructed with methyl methacrylate. Adjuvant therapy must be initiated in patients with malignancy as well as in those with non-resected tumours and lesions that

cause neurological deficits. Adjuvant therapy may include Gamma knife, chemotherapy and diphosphonate treatment [2,3].

Disclosure

Authors report no conflict of interest.

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