Ossified Intracranial Meningiomas: Description of the First Series of Cases and Review of the Literature

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BACKGROUND: **Metaplastic meningiomas are characterized by mesenchymal differentiation with formation of bone, cartilage, fat, or xanthomatous elements. However the presence of extensive areas of ossification is rare in meningiomas. In addition, intracranial location of ossified meningiomas is uncommon. Surgical management represents the optimal treatment for ossified meningiomas, but ossification may interfere with surgery and condition outcome.**

METHODS: **By reviewing patient records and contacting patients, families, and referring physicians, the following information was gathered: age, sex, tumor location, clinical presentation, preoperative and postoperative functional status, and surgical data. Each surgical specimen had been formalin-fixed, paraffin-embedded, and cut into parallel 4-μm-thick sections for histological evaluation.**

RESULTS: **Our literature search identified 8 cases, all of whom had undergone surgical treatment. Histopathological analysis revealed the presence of disorganized bone spicules with solitary oval osteocytic nuclei and lined by osteoblasts, with clear evidence of bone tissue in at least 50% of tumor tissue.**

CONCLUSIONS: **Here we present the largest series, to our knowledge, of surgically treated intracranial ossified meningiomas reported to date. The mechanism of ossification in these meningiomas remains unclear, and various hypotheses have been formulated. Complete lesion removal, or subtotal debulking in those cases characterized by tenacious adherences to vascular structures and/or critical areas, may represent the optimal treatment.**

Meningiomas are benign extracerebral lesions and constitute 15%–30% of the intracranial tumors in adults.[1] Metaplastic meningioma is a rare subtype of meningioma classified as grade I by the World Health Organization. It is characterized by focal or widespread mesenchymal differentiation, with formation of osseous, cartaligineous, lipomatous or xantomatous tissues, single or in combination.[2] The wide spectrum of differentiation reflects the pluripotentiality of the arachnoidal cells from which meningiomas originate. They may differentiate along various mesenchymal cell lines or show epithelioid differentiation. Calcification, ossification, bone invasion, bone infiltration, and osteoblastosis can represent usual features associates with meningiomas.[2] In particular, meningiomas showing calcification and an irregular number of bone spicules, within the tumoral tissue, have been named osteoblastic or osteomatous meningiomas.[3] However, total and/or extensive ossified intracranial meningiomas (OIMs; i.e., with <50% bone spicules in the absence of any evident changes in the adjacent bone) are very uncommon and account for approximately 1% of all meningiomas.[4]

Surgical management in OIM can be complex and may affect outcomes. Hard-rock consistency, widespread adhesions to surrounding eloquent cerebral structures, and management of dural attachment represent consistent challenges for the surgeon.

Here we report the first main series of 8 cases of OIM and describe clinical, radiologic, histological, and surgical features. We also include a review of the pertinent literature.

Key Words
- Calcified meningiomas
- Meningiomas
- Metaplastic meningiomas
- Ossified meningiomas
- Osteoblastic meningiomas
- Osteomatous meningiomas

Abbreviations and Acronyms
CT: Computed tomography
H&E: Hematoxylin and eosin
MRI: Magnetic resonance imaging
OIM: Ossified intracranial meningiomas

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METHODS

Patient Population

We reviewed the records at the University of Messina to identify all patients affected by OIM treated between 2001 and 2013, and identified a total of 8 cases. These cases represent 1.5% of meningiomas treated surgically during the aforementioned period in our department. By reviewing patient records and contacting patients, families, and referring physicians, the following information was gathered: age, sex, tumor location, clinical presentation, preoperative and postoperative functional status, and surgical data.

Histopathological Analysis

The histopathological analyses were performed in the Neuropathology Laboratory of the Department of Human Pathology at the University of Messina. Each surgical specimen had been formalin-fixed for 48–72 hours at room temperature, paraffin-embedded at 56°C, and cut into parallel 4-μm-thick sections for histological evaluation with the HE stain. Preliminary decalcification of samples was performed with formic acid 5% or ethylenediaminetetraacetic acid 5% (pH 7.4), for a no longer than 24 hours.

RESULTS

The 8 study patients included 7 women and 1 man, with a mean age of 52.7 years (range, 37–84 years) (Table 1). OIMs were located in the frontal region in 2 cases, in the frontotemporal region in 3 cases (left convexity in 2 cases, left temporal in 1 case), and in the left frontotemporoparietal region, in the trigonal region, and at

Table 1. Summary of Nosologic, Clinical, and Neuroradiologic Data

Case	Age (years)/ Sex	Symptoms	Neurologic Findings	Neuroradiologic Findings and Tumor Location	Histopathological Findings	Simpson Grade and Operative Findings
1	28/F	Headache, diplopia	Left homonymous emianopsia, left troclear palsy, central type left facial nerve palsy	CT: Calcified mass within the trigonal region of the right lateral ventricle MRI: Hypointense lesion in the trigonal region; peritumoral edema	Presence (>80%) of bone spicules with solitary, osteocyte nuclei lined by osteoblasts; psammoma bodies and calcifications; cytologic atypia positivity	Simpson grade I; bilobular, rock-hard consistency lesion covered by vascularized ependyma
2	37/F	Headache, seizures, derangement	Unremarkable	CT: Left temporal calcified mass MRI: Extrassial, hypointense, frontotemporal mass	Presence (>85%) of disorganized bone spicules with osteocyte nuclei lined by osteoblasts; presence of psammoma bodies and calcifications	Simpson grade I; roundish, rock-hard consistency and scarcely vascularized lesion
3	83/F	Headache, neurologic delay	Right hemiparesis, right Babinski sign	CT: Left frontal calcified mass MRI: Dysomogeneous hypointense frontal mass with tail sign	Presence (>60%) of disorganized bone spicules, psammoma bodies, and calcifications	Simpson grade III; notable adherence to the carotid and middle cerebral arteries
4	42/F	Headache, visual loss	Bilateral papilledema	MRI: Left hypointense frontotemporoparietal mass	Presence of immature bone trabeculae (>70%), mineralized chondroid matrix, vessels, and osteoclasts	Simpson grade IV; notable adherence to carotid and middle cerebral arteries and optic nerve
5	62/F	Headache	Right hemiparesis, right Babinski sign	MRI: Densely hypointense clinoid mass; hyperintense central core	Presence (>70%) of bone spicules; psammoma bodies and calcifications	Simpson grade I; rock-hard consistency
6	47/F	Headache, visual loss	Bilateral papilledema	MRI: Left hypointense frontotemporal mass	Presence (>65%) of disorganized bone spicules with solitary, oval, osteocyte nuclei lined by osteoblasts; presence of calcifications	Simpson grade I; rock-hard consistency
7	73/F	Seizures	Unremarkable	MRI: Left hypointense frontotemporal mass	Presence (>60%) of bone spicules; presence of psammoma bodies and calcifications	Simpson grade I; rock-hard consistency
8	49/M	Headache	Unremarkable	CT: Frontal basal totally calcified mass MRI: Hypointense lesion of the sphenoethmoidal plane	Presence (>70%) of bone spicules; presence of psammoma bodies and calcifications	Simpson grade I; rock-hard consistency