CASE REPORT

Primary Extracranial Meningioma of the Maxillary Antrum

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ABSTRACT—A case of primary extracranial meningioma of the maxillary antrum is reported. A 45-year-old male presented with symptoms of chronic sinusitis. Imaging studies showed a soft tissue mass with calcification, filling the maxillary antrum. The mass was removed surgically, and pathological studies revealed a fibroblastic meningioma. The maxillary antrum is an uncommon location of primary extracranial meningioma, and our case is the sixth to be reported in that location.

Meningioma is the most common nonglial intracranial neoplasm,¹ arising from meningocytes (arachnoid cap cells) of arachnoid granulation tissue and constitutes 15 to 20% of all intracranial tumors.² Approximately 6–17% of all meningiomas can be found in extracranial or extraspinal sites.^{3,4} The head and neck is the most frequent location of extraneuraxial meningiomas, commonly seen as an extension of primary intracranial tumor (secondary extracranial meningioma). Less than 2% of all meningiomas arise at a primary ectopic site (primary extracranial meningioma).⁵ Even more unusual is the metastasis of an intracranial meningioma to an extracranial location.

Extracranial meningiomas of the sinonasal tract have been reported most frequently to involve the frontal sinus⁴; however, review of the available literature revealed five reported cases of primary extracranial meningioma of the maxillary antrum.^{6–10}

placed medially by a submucosal mass. No other abnormalities were detectable. His past medical history was unremarkable. Specifically, there was no diagnosis of a prior meningioma.

Computed tomographic (CT) scan of the lesion revealed a mass filling and expanding the left maxillary sinus with focal areas of calcification. The mass did not extend outside the confines of the maxillary antrum (Fig. 1). CT scan of the brain was normal.

An intranasal biopsy of the lesion revealed a “benign spindle neoplasm with extensive microcalcification” consistent with “extradural fibroblastic meningioma.”

The patient underwent a medial maxillectomy with complete removal of the tumor via a sublabial incision (Fig. 2). The postoperative course was uneventful, and follow-up imaging studies showed no evidence of residual tumor.

CASE REPORT

A 45-year-old white male presented with a 6-month history of left-sided nasal obstruction, maxillary pain, and dental pain. He was treated for chronic sinusitis without relief of his symptoms. Nasal examination revealed complete obstruction of the left side of the nasal cavity. The medial wall of the maxilla was dis-

PATHOLOGICAL FINDINGS

Gross Pathology

The tumor was a 5.5 × 5.0 × 4.6 cm spherical, firm mass. The external surface was nodular and completely invested by a thin delicate silver-gray fibrous

Figure 1. (A, B) Coronal CT image showing an enhancing mass filling the left maxillary antrum with areas of calcification. There is no communication with an intracranial mass.

capsule. On hemisectioning, the mass appeared to be diffusely pale, yellow-white, and showed thin delicate silver-gray striations coursing irregularly throughout the tumor. No necrosis, excessive hemorrhage, or cartilage formation was noted (Fig. 3).

Histology

The tumor was composed of interlacing bundles of spindle cells with ovoid to elongated, hyperchromatic nuclei and pink to amphophilic cytoplasm. Numerous, randomly distributed calcific deposits were apparent throughout the lesion, some of which were round and laminated, and consistent with psammoma bodies.

Mitoses and necrosis were not observed. There was no evidence of an integral component of bony trabeculae or tumor-forming osteoid (Fig. 4).

Figure 2. A medial maxillectomy via a sublabial incision was used to remove the tumor mass (arrows).

Electron Microscopy

Examination revealed spindle cells with a moderate amount of euchromatin with peripheral heterochromatin. The cytoplasm contained abundant rough endoplasmic reticulum, often with cystic dilation. Rare interdigitating cytoplasmic processes were identified, some with poorly formed intercellular junctions. The extracellular spaces were filled with abundant collagen material.

Immunohistochemistry

The tumor cells were strongly positive for vimentin, focally positive for epithelial membrane antigen (EMA), and negative for S-100 protein.

Figure 3. Tumor mass, external and cut surface.

The above pathological findings, when combined, were consistent with the diagnosis of “extradural fibroblastic meningioma.”

DISCUSSION

Meningiomas are considered to be one of the most common tumors of the central nervous system. These tumors are rarely found extracranially, primarily in the head and neck region. The majority of reported extracranial meningiomas have demonstrable connection with an intracranial tumor. Farr et al³ reviewed 405 meningiomas and found that the most frequent location

of extracranial meningiomas associated with an intracranial mass was the orbital cavity. Extracranial meningiomas have also been found in the ear, nose, paranasal sinuses, parotid, neck, pharynx, mandible, and skin.¹¹

Primary extracranial meningiomas can be defined as those not connected with an underlying meningioma of the neuraxis. Due to their rarity, one must exclude an extension from a central meningioma.¹² The pathogenesis of primary extracranial meningioma is not completely established. It is postulated that meningocytes may become entrapped during closure of midline structures (fusion line inclusion), similar to the development of meningoceles.¹³ Meningocytes can also be found

Figure 4. Interlacing bundles of bland-appearing spindle cells associated with calcific deposit. Hematoxylin and eosin. $\times 200$.

around foramina of the skull or along perineural sheaths of cranial nerves and were found to have a predilection for areas of dural penetration by cranial nerves I, III, VII, IX, X, XI, and XII.¹² In sites that are distant from cranial nerves and fusion lines, meningiomas might arise from metaplasia of undifferentiated mesenchymal cells.¹⁴

Histologically, meningiomas are divided into different histological subtypes according to the predominant cellular morphology, such as meningiothelial (syncytial), fibrous (fibroblastic), psammomatous, angiomatous (angioblastic), and transitional (mixed).¹⁵ The angioblastic type, which is relatively more aggressive, may have a mesenchymal nonmeningothelial origin and is best regarded as a hemangiopericytoma of the meninges.¹⁶ The majority of extracranial meningiomas of the paranasal sinuses have been syncytial.¹⁷ Our case is the third fibroblastic primary extracranial meningioma of the maxillary sinus to be reported. The other three cases were all syncytial.

Immunohistochemical studies are indispensable in the differential diagnosis of primary extracranial meningiomas from other benign tumors of peripheral nerve origin. The positive expression of EMA and vimentin antigen demonstrates the dual epithelial and mesenchymal characteristics of the cells found in these tumors and agrees with previous studies on antigen expression in meningioma.¹⁸

Treatment of primary extracranial meningioma is surgical excision because meningiomas are known to be

radioresistant and recurrence is very rare following adequate surgical resection.¹⁹

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