AJNR

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AJNR Am J Neuroradiol 2007, 28 (1) 129-131
http://www.ajnr.org/content/28/1/129

CASE REPORT

Primary Intraosseous Meningioma of the Mandible: CT and MR Imaging Features

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SUMMARY: We describe the rare entity of an intraosseous meningioma arising in the mandible. The meningioma was found incidentally in an asymptomatic adult patient on dental radiography, mimicking other cystic-appearing jaw masses. The CT and MR imaging features of mandibular meningioma are reviewed with reference to prior published descriptions of this unusual entity.

Primary intraosseous meningiomas are rare. To the best of our knowledge, only 3 cases involving the mandible have been reported; all were imaged with conventional radiography. We present the cross-sectional CT and MR imaging features that help to differentiate this tumor clearly from the more common cystic lesions of the mandible.

Case Report

A 40-year-old asymptomatic woman was referred to an oral surgeon because of an incidental finding of a lytic mandibular lesion. A dental film, obtained to assess the dental status, demonstrated a well-defined lucency in the right mandible. The patient's medical history was unremarkable. Clinical examination revealed a bulge of the right mandibular body. The patient denied having associated pain, anesthesia, or paresthesia. The radiographic features were thought to be consistent with a cyst of the mandible, and needle biopsy was performed. The results of histologic examination revealed a fibrous tumor. The patient was transferred to the department of maxillofacial surgery. Surgical biopsy was performed, and the patient was scheduled for wide resection and fibula free flap reconstruction of the mandible. CT and MR imaging were performed preoperatively.

CT

Multisection CT before and after injection of contrast material was performed to evaluate the lesion and to exclude cortical destruction and soft-tissue involvement. CT demonstrated a well-defined purely osteolytic lesion containing homogeneous soft-tissue material with a mean attenuation of 55 HU. After contrast material injection, a moderate, homogeneous enhancement pattern was found; the mean attenuation value was 80 HU (Fig 1). The lesion was expansile and the cortical bone was thinned. No interruption of the cortical margins was found except at the site of prior biopsy. The apices of the right premolars and the first and second molar were involved, as was the mandibular canal.

MR Imaging

MR imaging was performed on a 1.5T system to search for a possible primary lesion that might be the source of a metastatic mandibular deposit; however, no primary lesion could be found. The lesion in the right mandible demonstrated low-to-intermediate signal intensity on short T inversion recovery (STIR) images and intermediate signal intensity on T1-weighted images. After administration of contrast material, homogeneous enhancement was found. Multiplanar reformations of the 3D magnetization-prepared rapid acquisition of gradient echo (MPRAGE) sequence (Fig 2) demonstrated intact mandibular cortical margins analogous to CT.

Histopathologic and Immunohistochemical Findings

The histopathologic features consisted of a tumor composed of uniform spindle-shaped cells partly arranged in whorls and interconnecting fascicles (Fig 3*A*). Tumor cells demonstrated oval-shaped nuclei with pale "wavy" eosinophilic cytoplasm and were set in a background of fibrillar ground substance. No mitotic figures and no significant nuclear atypia were identified in either case. Some psammoma bodies were detectable (Fig 3*A*, insert).

Using avidin-biotin immunostaining techniques on paraffin-embedded sections, we evaluated the neoplasm for vimentin, epithelial membrane antigen (EMA), and S-100 protein. The immunohistochemical stains demonstrated strong cytoplasmic staining for vimentin and focal staining for EMA (Fig 3*B*). S-100 protein staining was negative in the tumor cells. All other immunohistochemical markers (epithelial cytokeratins, actin, and desmin) were negative. Appropriate positive and negative controls were used. Vimentin and EMA are markers for meningiomas in the absence of S-100 epithelial cytokeratins; S-100 positivity is typically found in nerve sheath tumors and neurogenic tumors.[1]

The tumor was classified as fibrous (fibroblastic) meningioma.

Discussion

Primary extradural meningiomas are rare, accounting for less than 2% of all meningiomas; most cases involve the skull. A review of 169 cases was published by Lang et al.[2] Only 3 cases of primary meningioma involving the mandible have been reported so far. The first case was by Landini and Kitano in 1992[3]; another 2 cases were by Jones and Freedman in 2001.[4] All patients, including ours, were women, and the radiographic presentation was a lytic lesion. Three cases of extracranial meningioma of the maxilla have been reported, excluding cases within the paranasal sinuses. A male patient presented with an oral lesion and a multilocular radiolucency on a periapical x-ray film.[5] In 2 female patients, dental x-ray demonstrated a mixed radiolucent-radiopaque mass[6] with ill-defined margins in 1 patient.[7]

To our knowledge, the appearance of mandibular meningioma in CT and MR imaging has not been previously reported.

The lesion in our patient appeared as a cystic lesion of the mandibular body on conventional radiographs. CT revealed a soft-tissue mass with a well-defined margin (narrow transitional zone; Fig 1*A*, -*B*) involving the mandibular nerve canal. The intraosseous mass was homogeneous on pre- (Fig 1*C*) and

Received November 8, 2005; accepted after revision November 25.

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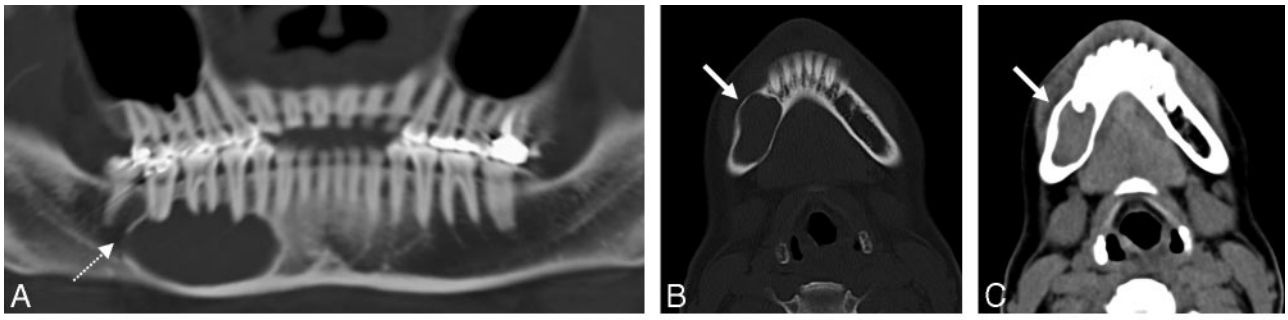

Fig 1. Panoramic view of the mandible (*A*) in bone window demonstrates involvement of the mandibular canal (*dotted arrow*) and dental apices (multisection spiral CT data: 64 × 0.6 mm, 120 kV, 200 mAs). CT before (*B* and *C*) and after contrast injection (100 mL; 350 mg of iodine per milliliter) (*D*). Expansile lytic lesion (*arrow*) is seen in the right mandible without cortical interruption with moderate enhancement (attenuation precontrast, 55 HU; postcontrast, 80 HU) after administration of contrast material.

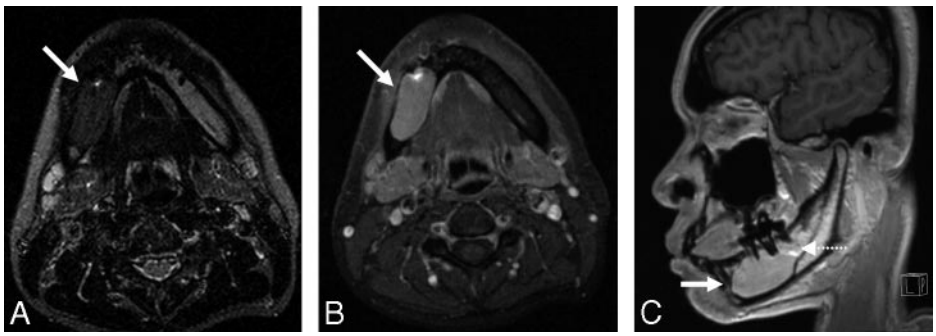

Fig 2. MR imaging shows a lesion with low signal intensity on STIR (*A*, TR/TE/TI, 4100/105/140 ms; section thickness, 4 mm) and homogeneous enhancement on postcontrast T1-weighted images (*B*, Fat-saturated T1-weighted turbo spin-echo: TR/TE, 560/17 ms; section thickness, 4 mm; *C*, MP-RAGE: TR/TE/TI, 1960/3.9/1100 ms; $\alpha = 15°$; SL, 1 mm) (*arrow*). Sagittal oblique image demonstrates involvement of dental apices and mandibular canal (*dotted arrow*).

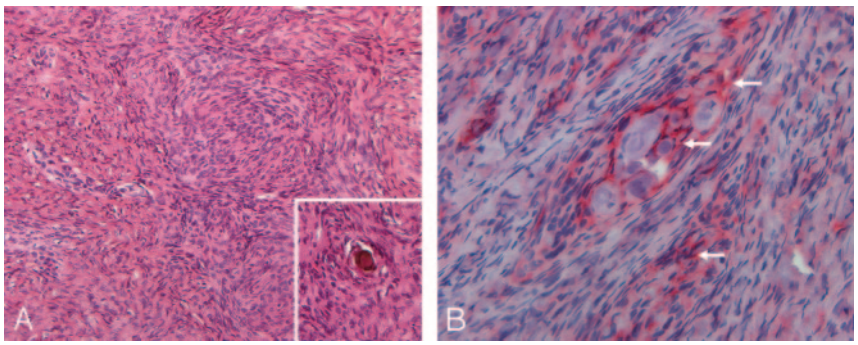

Fig 3. Photomicrograph of the meningioma (hematoxylin-eosin, original magnification ×90) shows spindle cells partly arranged in whorls and interconnection fascicles with little interlaying collagenous matrix (*A*). Psammoma bodies can be identified (*A*, insert). Photomicrograph of tumor cells show focal staining for EMA (*B*, original magnification ×180) (*arrows*).

postcontrast (Fig 1*D*) scans; contrast enhancement was moderate. No extraosseous soft-tissue mass and no calcifications within the tumor matrix were found. MR imaging demonstrated a homogeneous lesion with intermediate signal intensity on T1-weighted spin-echo images and low signal intensity on T2-weighted (STIR) images (Fig 2*A*). Enhancement after injection of contrast material was homogeneous on fat-suppressed T1-weighted spin-echo (Fig 2*B*) and MPRAGE (Fig 2*C*) images.

Tokgoz et al[8] described different imaging features for a primary intraosseous meningioma of the calvaria; CT demonstrated a lytic mass with cortical destruction and prominent enhancement. The lesion demonstrated high signal intensity on T2-weighted images and intense homogeneous enhancement. Marked hyperostosis and high signal intensity on T2-weighted and postcontrast T1-weighted images were also reported for meningiomas of the calvaria.[9,10] Signal intensities of the intracranial component of jugular foramen meningiomas have been reported to be significantly higher than those of the extracranial component on T1-, T2-, and postcontrast T1-weighted images, presumably because of different histologic composition and collagen content in particular.[11] The histologic composition with densely packed spindle-shaped tumor cells in a background of a fibrillar matrix could explain the low signal intensity on STIR images in our patient. The low signal intensity on T2-weighted images makes many differential diagnoses like hemangioma, epidermoid cyst, eosinophilic granuloma, and aneurysmal bone cyst unlikely. Ameloblastoma

often show foci of high signal intensity on T2-weighted images, but intermediate signal intensity can also be found as in metastatic cancer, myeloma, lymphoma, giant cell tumor, or fibrous dysplasia.[12] In the mandible, frequent findings are odontogenic and nonodontogenic cysts. All these lesions can present as a lucent area on panorex images, but CT and MR imaging can unequivocally differentiate cysts and solid tumors. An intact cortex and a sharp border of the lesion suggest a benign process; these findings can be best appreciated on high-resolution CT.

Many different hypotheses exist regarding the origin of primary intraosseous meningiomas. Meningiomas originate from meningothelial cells.[13] Misplacement and entrapment of meningothelial cells as a result of trauma have been speculated.[14] Meningiomas may develop from multipotential mesenchymal cell precursors, explaining the occurrence of meningiomas in uncommon locations.[15] Cutaneous meningiomas could be congenital in origin, in which case they arise from arachnoid cell rests located in the skin as a result of defective closure of the neural tube, wherein the meningeal tissue is pinched off into the surface.[16] The origin of extracranial meningiomas has also been attributed to proliferation of ectopic embryonal nests of arachnoidal cells, and the proliferation of perineural cells of peripheral nerves is another possibility, as a result of the structural and functional similarities of perineural cell and arachnoid cells.[3] The intimate relation to the mandibular nerve in our patient may favor this hypothesis. Rarely, ectopic meningiomas arise through metastatic spread, in the lung, liver, and bone.[17-19]

Recommended treatment for extradural meningioma is wide surgical resection followed by reconstruction. If only subtotal resection is possible because of the involvement of critical structures, the residual tumor should be followed up radiologically. Radiation therapy is an option for patients with symptomatic or progressive residual tumor.[20]

In conclusion, primary intraosseous meningioma, though rare, should be considered as a differential diagnosis in lytic lesions of the mandible. Although CT and MR imaging features are nonspecific, solid tumor is unequivocally differentiated from cystic lesions and the low signal intensity on T2-weighted images narrows the differential diagnosis. CT is best suited for excluding cortical destruction; MR imaging is the technique of choice to exclude soft-tissue extension as well as to detect intracranial meningioma as a potential site of metastatic spread.

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