Intraosseous Neurilemmoma of the Mandible

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Case Report

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Summary: We report a rare case of intraosseous neurilemmoma of the mandible, with an emphasis on radiographic findings. The tumor, located mainly in the premolar region, presented as an expansive, unilocular, well-defined, radiolucent lesion on plain radiography. No dilatation of the mandibular canal was identified. MR imaging helped to identify the solid nature of the tumor. A biopsy was necessary to make the final diagnosis because of the relatively nonspecific nature of the lesion.

The neurilemmoma, also called a schwannoma or neurinoma, is a benign neoplasm originating from the peripheral neural sheath. Although the head and neck region is one of the most common sites for benign nerve-sheath tumors, intraoral lesions are unusual, particularly in the intraosseous region of the jaw. Previous literature related to intraosseous neurilemmomas of the jaw only includes discussions concerning panoramic and dental radiography and CT examinations (1). We describe a rare case of intraosseous neurilemmoma of the mandible evaluated by use of dental CT with curved- and cross-sectional reformatted images and MR images.

Case Report

A 19-year-old woman complained of experiencing crepitation in the right temporomandibular joint for 2 years. Neurosensory examination was normal, and the patient reported no previous episodes of pain or sensory disturbances. A physical examination revealed normal soft tissue of the oral cavity, and no facial swelling, percussion pain, or mobility of the molar teeth in the right mandible was noted. Her past medical history was essentially noncontributory. The patient underwent dental radiography, and later a dental CT scan and 1.5-T superconductive MR imaging. Panoramic radiography identified a well-defined, expansive, osteolytic lesion with no septation in the right posterior mandible. The lesion was located in the supracanal mandibular body between the roots of the second premolar and the first molar teeth. There was no apparent resorption of roots of the premolar and molar teeth, although oblique orientation of the roots suggested a long-standing process (Fig 1A). A CT scan (Proseed Libra; GE-Yokogawa Medical Systems, Tokyo) was performed with the following parameters: 120 kV, 60 mA, and 1.0-mm slice thickness consecutive axial images. Then, reconstruction with curved- (parallel to the dental arch) and cross-sectional (perpendicular to the dental arch) planes was performed using a dental software algorithm (DentaScan, GE). Axial CT images revealed a well-delineated, expansive lesion with peripheral scalloping and buccolingual erosion of the cortex, mainly in the bone marrow of the premolar region (Fig 1B). On the reformatted images, the mandibular canal shifted slightly toward the lingual side with a defect in its upper cortex. The mass, approximately 30 × 20 mm, appeared to be contiguous with the mandibular canal. There was no apparent dilatation of the mandibular canal (Fig 1C). On spin-echo T1-weighted MR images (400/8 [TR/TE]; slice thickness, 5 mm), an almost homogeneously intermediate signal intensity was shown in the mandible (Fig 1D). On fast spin-echo T2-weighted images (3000/105), a bright signal intensity with foci of intermediate signal was identified (Fig 1E). After intravenous administration of gadopentetate dimeglumine (0.1 mmol/kg), intensive peripheral enhancement with a relatively poorly enhanced central portion was shown (Fig 1F). A biopsy was obtained under local anesthesia, and a preliminary diagnosis of neurilemmoma was made. Subsequently, under general anesthesia, the patient underwent total removal of the tumor with a bone graft. The tumor was easily removed, leaving the inferior alveolar neurovascular bundle intact. The lesion was edematous and solid with a grayish color and no apparent cystic cavities.

Microscopic examination of the hematoxylin-eosin–stained section of the biopsy specimen revealed foci of Antoni type-A tissue, including spindle cells arranged with palisading, ovoid, basophilic nuclei and acidophilic cytoplasm, and Antoni type-B tissue. Acellular, eosinophilic areas described as Verocay bodies were also noted (Fig 1G and H). No infiltration into the mandibular canal was identified. Immunohistochemical examination revealed positive anti-S-100 protein. The histologic diagnosis of benign neurilemmoma was confirmed by evaluation of the resected specimen. No recurrence was observed in the follow-up study at 6 months.

Discussion

Neurilemmoma is not commonly seen in the oral cavity, and the tongue is the most common site of neurilemmomas found there. Less than 1% of all neoplasms arising in the bone are intraosseous neurilemmomas (1); of these, the mandible is the most commonly affected area, particularly in the posterior segment of the body and ramus. Our survey of the English-language literature revealed only 34 proven cases of intraosseous neurilemmoma of the mandible (2–6).

Clinically, neurilemmoma is a slow-growing tumor that may be present for years before becoming symptomatic. Swelling is the most common symptom, but pain or paresthesia may be present in

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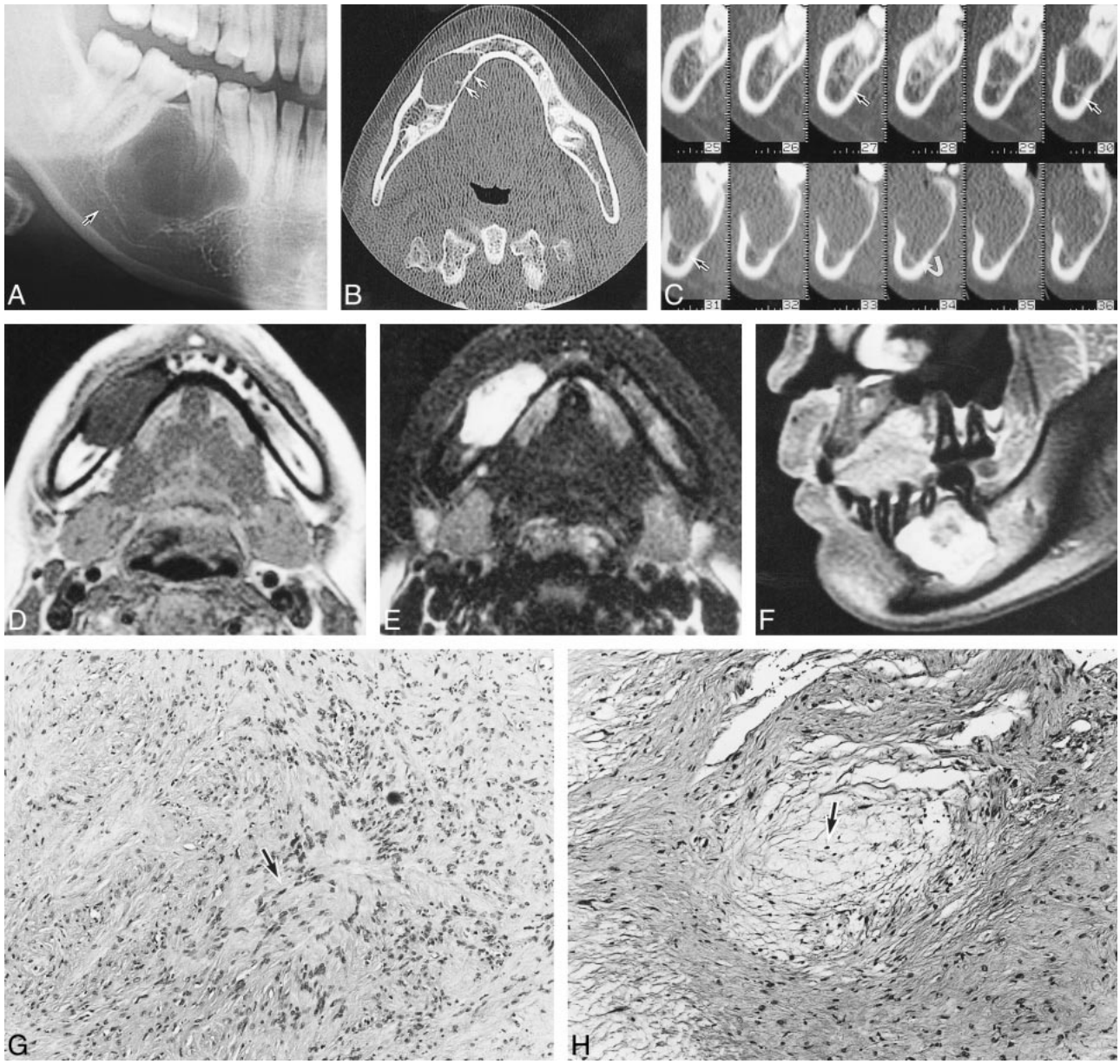

FIG 1. 19-year-old woman with intraosseous neurilemmoma of the right side of the mandibular body.

A, Panoramic radiograph shows a well-defined, unilocular, expansive lesion in the supracanal region. No evidence of dental root resorption or dilatation of the mandibular canal was noted (*arrow*).

B, Axial CT scan with 1-mm slice thickness shows an expansive lytic lesion associated with incomplete septae (*arrows*) and cortical thinning of the lingual plate.

C, Cross-sectional reformatted images from the second molar (image 25) to the second premolar (image 36) regions show the lesion located in the supracanal mandibular body associated with defect (*curved arrow*) of the upper cortical margin of the mandibular canal (*arrows*). Thinning and defect of the buccal cortical plate of the mandible are also seen.

D, Axial T1-weighted MR image shows a well-demarcated intermediate signal lesion in the mandibular body.

E, Axial T2-weighted MR image shows a lesion with bright signal intensity.

F, Contrast-enhanced sagittal T1-weighted image shows intensive enhancement.

G, Photomicrograph of a resected specimen (hematoxylin and eosin stains; original magnification ×100) shows Antoni type-A tissue (fascicular type) consisting of palisading arranged nuclei (*arrow*) and Verocay bodies (acellular zones).

H, Photomicrograph of a resected specimen (hematoxylin and eosin stains; original magnification ×100) reveals Antoni type-B tissue (reticular type) associated with irregularly arranged, dark-stained, atrophic nuclei and myxoid degeneration (*arrow*).

about 50% of cases (7). There is a female predilection, with a 1.6:1 female-to-male ratio. Seventy-seven percent of the patients were below the age of 50 at the time of diagnosis, and 46% were below the age of 30 (2).

Upon microscopic evaluation, Antoni-A and Antoni-B tissue are usually seen (8). In the Antoni-A region, homogeneous acellular zones known as Verocay bodies are sometimes noted. The Antoni-B region consists of a random arrangement of tis-

sue with microcysts. Neurilemmomas are typically well encapsulated, whereas encapsulation is seen in only 4% of neurofibromas (9). Nonetheless, Murphy and colleagues reported a rare case of nonencapsulated, atypical, intraosseous neurilemmoma of the mandible (2).

Radiographically, neurilemmomas are uniloculated or multiloculated, well-defined radiolucencies located in the posterior mandible, which are suggestive of a benign process such as odontogenic keratocyst, periodontal cyst, or ameloblastoma (4). Erosion of the adjacent roots is a common finding, and dystrophic calcification within the radiolucency has also been described (8). On CT scans, incomplete septae have been reported in the lesion. MR findings of intraosseous neurilemmoma of the mandible have not been reported to our knowledge, and it helps in differentiating solid from purely cystic lesions (eg, dentigerous cysts, periodontal cysts) (10). Minami et al reported strong gadolinium enhancement of the solid portions of the tumor in multicystic and unicystic ameloblastoma, including papillary projections, walls, and septa. Other hypervascular ameloblastomas and ameloblastohemangiomas may also reveal marked enhancement. CT features of ameloblastoma include shell-like bulging of the cortex and incomplete bone septae (11). These features of ameloblastoma on MR and CT images are similar to those of our case, and a definite diagnosis is difficult to make on the basis of radiography alone; a biopsy is usually necessary. Neurilemmomas arise rarely in the mandibular canal, and such lesions become rounded (12). On the other hand, neurofibromas tend to grow specifically in the canal, which typically become ovoid-shaped (13). The recurrence rate of a neurilemmoma is lower than that of a neurofibroma because of encapsulation (5). The differentiation between these two neoplasms is imperative because neurofibromas tend to recur frequently and have the potential for malignant transformation.

Conclusion

In conclusion, a rare case of intraosseous neurilemmoma of the mandible was reported. The radiographic features revealed a markedly enhanced solid mass, which may be distinguished from purely cystic odontogenic masses. It is difficult, however, to differentiate an intraosseous neurilemmoma from an ameloblastoma associated with a substantial solid component.

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