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Case Challenge
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The Case of a Subtle Expansion of the Mandible

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The following Case Challenge is provided in conjunction with the American Academy of Oral and Maxillofacial Pathology.

Case Summary

A healthy 25-year old Caucasian man presented for a routine dental examination. He was unaware of a subtle expansion involving the right posterior mandible, which was bony hard and nontender. Radiographic findings prompted an immediate referral of the patient to an oral and maxillofacial surgeon for diagnosis and management of the jaw lesion.

After you have finished reviewing the available diagnostic information, make the diagnosis.

Diagnostic Information

Medical History

This apparently healthy patient had seasonal allergies that were managed by pseudoephedrine when symptoms would arise. The patient suffered a fracture of the left fibula as the result of a motor vehicle accident in 1988. No other organ system abnormalities were noted. Significant social history included a 5-year history of cigarette smoking, and he admitted to the continued use of tobacco products.

Clinical Examination Findings

The oral and maxillofacial surgeon palpated a slightly expansile area of the right mandible in the medio-lateral dimension. All mandibular surfaces were bony hard and without evidence of crepitus. The inferior border of the mandible was also intact and without obvious expansion. There were bilateral, < 1 cm, movable and non-tender lymph nodes in the submandibular region. Sensory neural examination revealed intact tactile function of all areas served by the inferior alveolar nerve. Cranial nerves II – XII were evaluated and assessed as being within the range of normal. All teeth tested vital to electrical stimulation. Occlusion was normal, without evidence of tooth movement. (Figure 1) All mucosal surfaces were intact, although some expansion of the right buccal plate was noted. (Figure 2) Minimal facial asymmetry was appreciated, when evaluating the frontal view of the face (Figure 3), but the changes were not obvious in the profile view. (Figure 4)

Preliminary Radiographic Findings

A cropped pantomographic radiograph revealed a well-defined, 5.0 x 2.0 cm radiolucent lesion. (Figure 5) Focal areas of the radiographic border of the lesion were somewhat scalloped, including

Figure 1. Occlusion and view of buccal vestibule.

Figure 2. Right side vestibule showing some expansion.

Figure 3. Preoperative photograph of full face with slight asymmetry noted.

Figure 4. Preoperative photograph showing the facial profile with no asymmetry noted from this view.

Figure 5. Pantomographic radiograph; evidence of multilocularity is seen.

Figure 6. Pantomographic radiograph showing scalloping between the roots and along the inferior border of the mandible.

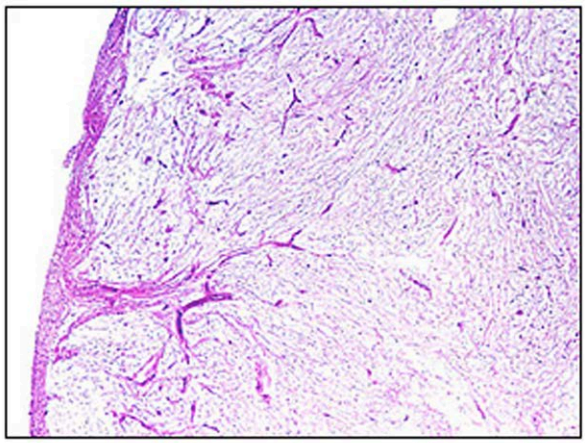

Figure 7. Low power photomicrograph showing a lesion that is dominated by loose, myxoid, and fibrocollagenous tissue that is hypocellular (Hematoxylin and eosin, original magnification 40x).

areas between the roots of the teeth. (Figure 6) The inferior cortex was focally thinned, but was not displaced. In addition, the inferior alveolar canal remained discernible without evidence of disruption or displacement. Several circular areas displayed increased radiolucent changes consistent with a multilocular pattern and a more aggressive process. These lytic changes were most obvious in the pericoronal area of the third molar and extended to the distal of the second premolar. Although the lesion was well demarcated, the margins varied from being corticated to non-corticated.

Incisional Biopsy and Microscopic Findings

Due to the significant size of the lesion, an initial incisional biopsy was performed under local anesthesia and conscious sedation in order to establish a definitive diagnosis. Unfortunately, the initial biopsy was inconclusive and the patient was referred to Eisenhower Army Medical Center, Ft. Gordon, GA, for an additional biopsy and treatment. During that procedure, abundant gelatinous material was obtained and the specimen was submitted in formalin for routine histopathologic examination. The gross specimen consisted of three fragments of soft tissue, which ranged from 1.5 x 1.5 x 0.4 cm to 0.6 x 0.4 x 0.3 cm.

The low power photomicrograph displayed variably dense fibrocollagenous connective tissue along the edge. The lesion itself dominated the image and was characterized by loose myxoid fibrocollagenous connective tissue that was hypocellular. (Figure 7) The medium power photomicrograph displayed loose and myxoid fibrous connective tissue with some small vascular channels interspersed as well as some extravasated erythrocytes. The

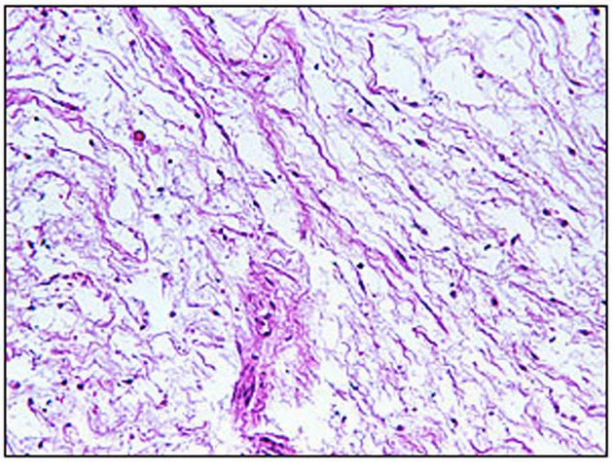

Figure 8. Medium power photomicrograph displaying generally loose and myxoid fibrous connective tissue with some small vascular channels. (Hematoxylin and eosin, original magnification 100x).

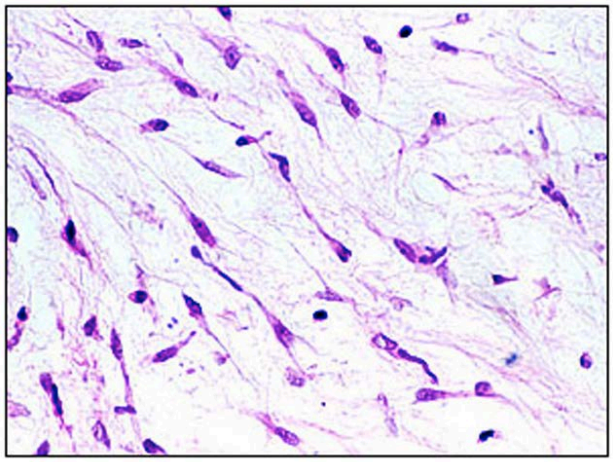

Figure 9. High power photomicrograph showing uniform nuclei within the myxoid stroma. These nuclei are spindle-shaped to stellate and evenly distributed across the field. (Hematoxylin and eosin, original magnification 400x).

Figure 10. This axial CT displays the multilocular nature of the lesion and explains some of the circular lytic areas seen on the pantomograph.

nuclei were spindle-shaped to stellate and evenly dispersed throughout the specimen. Although the nuclei were somewhat hyperchromatic, they were uniform in appearance and without evidence of mitotic activity. (Figure 8) The high power photomicrograph showed similar findings. (Figure 9)

Advanced Imaging

Computer-assisted tomographic images (CTs) were ordered, and both axial and coronal cuts were reviewed. The axial CT displayed the multilocular nature of the lesion and explained the circular lytic areas that were seen on the pantomograph. (Figure 10) In addition, the axial CT demonstrated the mandibular expansion in addition to the thinning and scalloping of the cortices. (Figure 11) Similarly, the coronal CT displayed the mandibular expansion of the lesion from another view. (Figure 12)

Figure 11. This axial CT displays the mandibular expansion, thinning of cortices, and scalloping of the cortices.

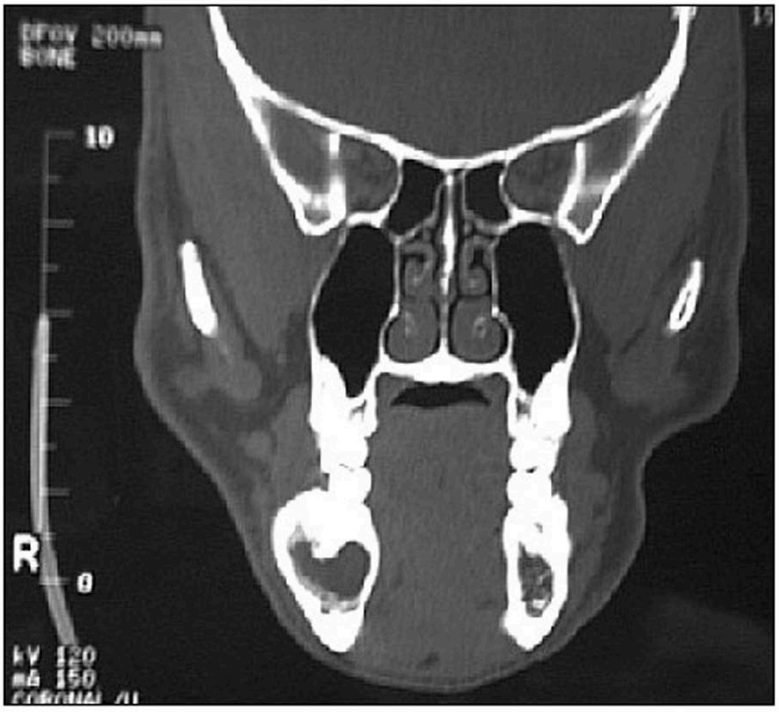

Figure 12. This coronal CT displays the mandibular expansion from another view.

Can you make the diagnosis?

A healthy 25-year old Caucasian man presented for a routine dental examination. He was unaware of a subtle expansion involving the right posterior mandible, which was bony hard and nontender.

Select the Correct Diagnosis

A. Odontogenic Keratocyst
B. Simple Bone Cyst
C. Ameloblastoma
D. Aneurysmal Bone Cyst
E. Odontogenic Myxoma

Odontogenic Keratocyst

Choice A. Sorry, this is not the correct diagnosis.

However, radiographically the odontogenic keratocyst (OKC) should be in your differential diagnosis. The OKC presents as a well-defined unilocular or multilocular radiolucency with scalloped borders. In addition, the location in the posterior mandible and the age is consistent with the clinical features of an OKC. Corticated and non-corticated margins, as seen in the present case, are also possible.[1-3]

Intraoperative findings of the OKC often include the presence of abundant “cheesy” keratinaceous debris. In addition, the cystic nature of this entity would be appreciated. Aspiration of the OKC may, or may not, yield thick keratin or a straw-colored fluid.

Please re-evaluate the information about this case.

Simple Bone Cyst

Choice B. Sorry, this is not the correct diagnosis.

The simple bone cyst is also known as a traumatic bone cyst or idiopathic bone cavity. The radiographic feature of a posterior mandibular radiolucency, scalloping between tooth roots, may allow the simple bone cyst to be included in the differential diagnosis.[4] Expansion and thinning of the cortices is indeed possible with this entity. However, with close examination of radiographic features in the present case, the likelihood of a simple bone cyst is diminished. In the simple bone cyst, "scalloping" is usually present only around the tooth roots. In other areas, the radiolucency should be uniformly rounded without scalloping.[5] Although not a characteristic finding, there are rare examples of the simple bone cyst exhibiting multiloculation with cortical expansion and slow growth rate. Intraoperatively, the simple bone cyst is described as a hollow cavity with minimal to no soft tissue contents. Small amounts of serosanguineous fluid are found in the majority of these lesions.

Please re-evaluate the information about this case.

Ameloblastoma

Choice C. Sorry, this is not the correct diagnosis.

However, radiographically the ameloblastoma should be in your differential diagnosis. The radiographic appearance of the ameloblastoma can be identical to the features seen in the present case. The ameloblastoma can be unilocular or multilocular with scalloped borders. In addition, the location in the posterior mandible and the age of the patient is consistent with the presentation of an ameloblastoma. Corticated and non-corticated areas as seen in this case are also possible.[6]

Intraoperative findings of the ameloblastoma may include either cystic or solid neoplastic tissue. Aspiration of cystic ameloblastomas may or may not yield fluid.[7-9]

Please re-evaluate the information about this case.

Aneurysmal Bone Cyst

Choice D. Sorry, this is not the correct diagnosis.

However, radiographically the aneurysmal bone cyst (ABC) may be in your differential diagnosis.[10] The radiographic appearance of the ABC can be identical to the features seen in the present case. The ABC can be unilocular or multilocular with scalloped borders and corticated and non-corticated margins. Some examples demonstrate a ballooning distention of the contour of the affected bone. A characteristic clinical feature of the ABC is the rapid onset of the swelling that is frequently painful. Usually this entity occurs under the age of 20, but a wide age range has been reported.[11] Intraoperative findings of the ABC include the presence of reddish brown tissue and dark venous blood.[11]

Please re-evaluate the information about this case.

Odontogenic Myxoma

Choice E. Congratulations! You are correct!

This case represents a typical presentation for an odontogenic myxoma. The average age for myxomas is the 15-30 year-old age group with a slight predilection for the mandible.[12-14] In addition, although small unilocular lesions may be encountered, the scalloping multilocular radiolucent features seen in the present example are characteristic of medium to large lesions.[12,13,15]

The typical odontogenic myxoma remains a sometimes controversial lesion from a therapeutic aspect.[16-19] This is due to its locally persistent nature. The lesion usually expands by infiltration between and through the medullary spaces.[20] Therefore, the radiographic evidence of the lesion may not be entirely indicative of the actual extent of the lesion.[12] Because of the recurrence rate, treatment options range from simple enucleation with curettage, to enucleation with peripheral ostectomy, to resection beyond the radiographic evidence of the lesion.[14,21,22] Some authors recommend magnetic resonance imaging (MRI) to evaluate the extension of the soft tissue component of these intrabony tumors.[15,23]

Adequacy of resection is a major concern, with recurrence of tumor following enucleation and curettage being reported in the range of 25%.[17,24] Morbidity associated with maxillary tumors is usually more significant than for mandibular lesions.[17,19] Regardless of which treatment modality is used and which jaw is affected, close clinical and radiograph follow-up is essential.

The gross pathologic examination of an odontogenic myxoma is often very telling. Centrally, the gelatinous yellowish material, which is generally an intraoperative feature of any highly myxoid lesion, is usually evident. (Figure 13) In addition to the classic myxoid appearance, the odontogenic myxoma may have scattered dystrophic mineralizations or scattered odontogenic epithelial rests.[16,25] Features of other myxoid neoplastic processes must be absent.[14,26]

Treatment

The patient underwent a local discontinuity resection of the right mandible via intraoral incision with 1-1.5cm surgical margins taken beyond the radiographic evidence of the lesion. (Figures 14, 15) Adequacy of the resection was evaluated by plane film radiography intraoperatively and by histopathologic sections following demineralization. A Synthes threadlock reconstruction plate (Synthes Maxillofacial Paoli, PA) was affixed to proximal and distal ends of the mandible and a prefabricated heat cured acrylic space maintainer was placed between the proximal and distal segment in an effort to maintain a soft tissue plane to facilitate future reconstruction. (Figures 16, 17)

The patient experienced wound dehiscence within several days of surgery, and approximately 2 weeks later had the space maintainer

Figure 13. The gross photograph seen here displays a cross-section of the resected mandible. Centrally, the gelatinous yellowish material is a diagnostic feature of a highly myxoid lesion.

Figure 14. Preoperative patient profile, showing general external orientation marking.

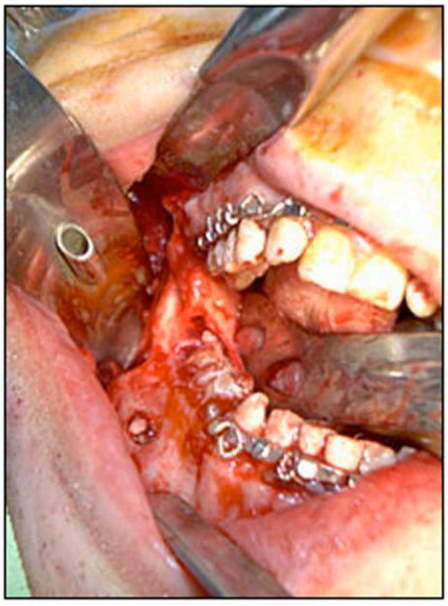

Figure 15. Intraoperative view just prior to vertical cuts.

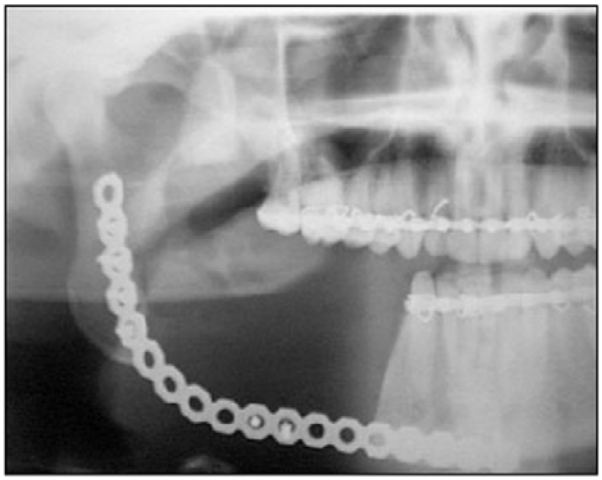

Figure 16. Post resection pantomograph with reconstruction bone plate.

Figure 17. Post resection skull film showing reconstruction plate.

removed, recontoured, and the primary closure reperformed. Over the course of one week, the wound was again noted to have a dehiscence with post-surgical signs of infection in the area. Gram stains showed gram-negative rods, gram-positive cocci, and rare gram-negative diplococci. The wound was cultured and displayed a mixed infection including heavy growths of *Escherichia coli*, *Streptococcus viridans*, and group G beta *streptococci*.

Five months following tumor resection, the patient underwent reconstruction of the mandible with a right posterior iliac crest graft, as well as a right fifth rib graft, via a submandibular incision. The Synthes reconstruction plate remained in place

and the space maintainer was removed. Ten months after the initial resection, four Replace Select implants (Nobel Biocare, Yorba Linda, CA) were placed and tibial bone, as well as allogenic tissue, was used to augment implant placement. Synthes DBX allogenic bone material (Synthes Maxillofacial Paoli, PA) in the putty form was utilized. The mucosa has healed well and implants appear integrated. (Figures 18,19) The final phase of treatment will include a combined fixed and removable prosthetic reconstruction, which is planned within the next few months. A potential fibrous union at the point of the proximal resection remains a concern in this patient, who is otherwise progressing well.[27]

Figure 18. Post reconstruction radiograph with bone graft.

Figure 19. Radiograph of bone graft with implants.

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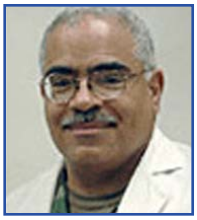

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