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CALCIFYING EPITHELIAL ODONTOGENIC TUMOR
OF THE MANDIBLE (PINDBORG TUMOR). RPC
(RADIOLOGIC-PATHOLOGIC CORRELATION) FROM
THE AFIP

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Armed Forces Institute of Pathology
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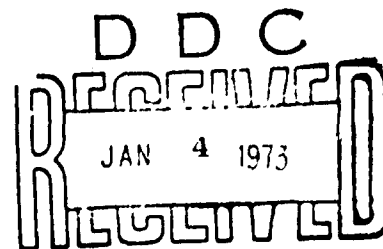
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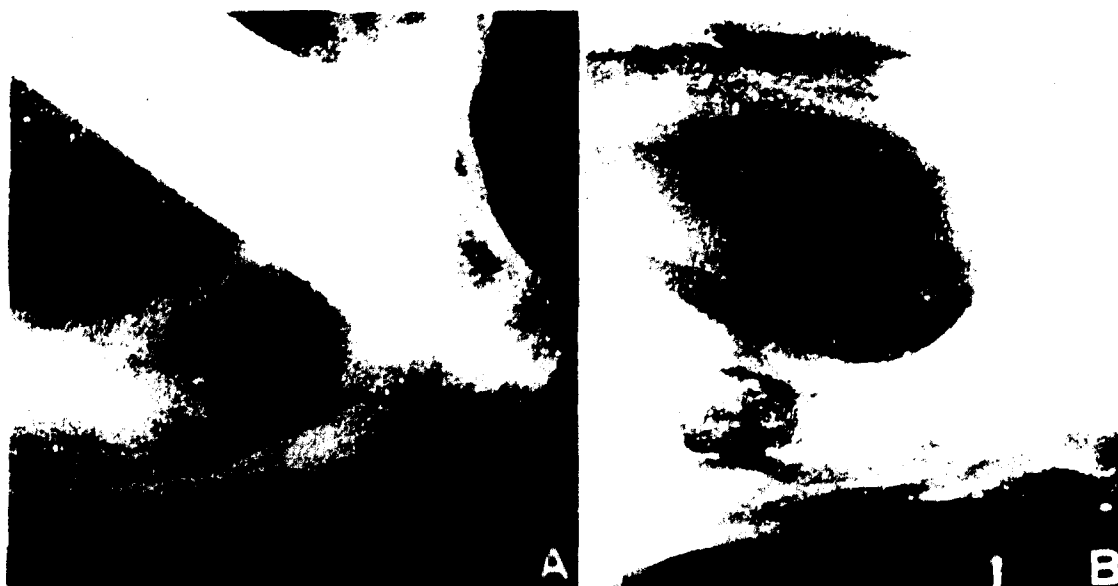
RPC

AD 753379



RPC¹ from the AFIP²

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This 21-year-old man presented with a lump in his left jaw, which was associated with pain when palpated. A lower left premolar had been extracted eight months previously. Since that time, expansion of the mandibular cortices had been noted by his oral surgeon.

From the radiographic features shown here, select the group below which includes the most appropriate possibilities for differential diagnosis:

- (a) Adenameloblastoma, odontoma, ossifying fibroma, calcifying epithelial odontogenic tumor, or osteomyelitis
- (b) Radicular cyst, abscess, ameloblastoma, histiocytosis X, or myeloma
- (c) Osteopetrosis, Garré's osteomyelitis, thalassemia, ossifying fibroma, or cementoma.

¹RPC - Radiologic Pathologic Correlation

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Fig. 1 (AFIP negatives 72-1363-2 and 68-5856). Solitary, somewhat oval, well-circumscribed lytic defect within the body of the mandible on the left side. There is no rim of sclerosis or periosteal reaction. Multiple areas of increased density may be seen within the lesion.

RADIOLOGIC DISCUSSION

A lateral view of the mandible and a Panorax view of the jaws reveal a solitary, somewhat oval, well-circumscribed lytic defect in the body of the mandible on the left side (Fig. 1). There is no rim of sclerosis, nor evidence of periosteal reaction; however, multiple areas of increased density within the lesion give it a mixed radiolucent and radiopaque appearance. The posterior teeth are missing on the left side, but the alveolar ridge appears to be intact and there is no suggestion of a soft-tissue mass. Other views revealed that the buccal and lingual cortices of the mandible were expanded but not destroyed.

The localized pattern of mixed destruction and repair favors a benign process. Moreover, the expansile quality of the lesion and the intact cortices (not illustrated) also make aggressive behavior less likely.

Answer *c* represents a range of possibilities which might be considered with an opaque lesion of the jaws. Although some of these lesions may present mixed appearances, e.g., ossifying fibroma or cementoma, osteopetrosis and Garré's osteomyelitis are typically opaque at all stages and make this selection untenable. The lesions listed in answer *b* are purely lytic and never show a mixed or opaque appearance in the jaw; hence they should also be excluded. Thus the most appropriate list of diagnostic possibilities for the lesion shown in Figure 1 is the group of mixed lucent and opaque lesions of the jaw included in answer *a*.

The range of lesions that are partly radiolucent and partly opaque spans several categories of bone

TABLE 1: LESIONS OF THE JAWS
(MIXED LUCENT AND OPAQUE)

Anomalies or neoplasms
Ossifying fibroma
Fibrous dysplasia
Cementoma (Stage II)
Cystic odontoma
Calcifying epithelial odontogenic tumor (Pindborg tumor)
Ameloblastic odontoma
Osteosarcoma
Chondrosarcoma
Metastatic carcinoma (breast, prostate)
Inflammation
Osteomyelitis
Trauma
Retained root
Metabolic, mechanical, or circulatory
Paget's disease

disease (TABLE 1). The pathogenesis involves (a) predominantly radiolucent lesions which do not produce a matrix, but in which bone repair becomes recognizable radiographically (e.g., osteomyelitis), (b) predominantly matrix-producing radiopaque lesions preceded by a radiolucent phase (e.g., cementoma), or (c) certain rare biphasic odontogenic tumors which contain both radiolucent and radiopaque elements (e.g., calcifying epithelial odontogenic tumor). The categories of disease consistent with this pattern of behavior and pathogenesis include developmental anomalies, benign neoplasms, inflammation, and trauma. Circulatory disorders, mechanical disturbances, and metabolic diseases would generally be distributed more diffusely, so that the solitary focal nature of the lesion discussed here makes these latter conditions unlikely; for example, Paget's disease, which was not included in any of the choices, can also show a mixed pattern in the jaw.

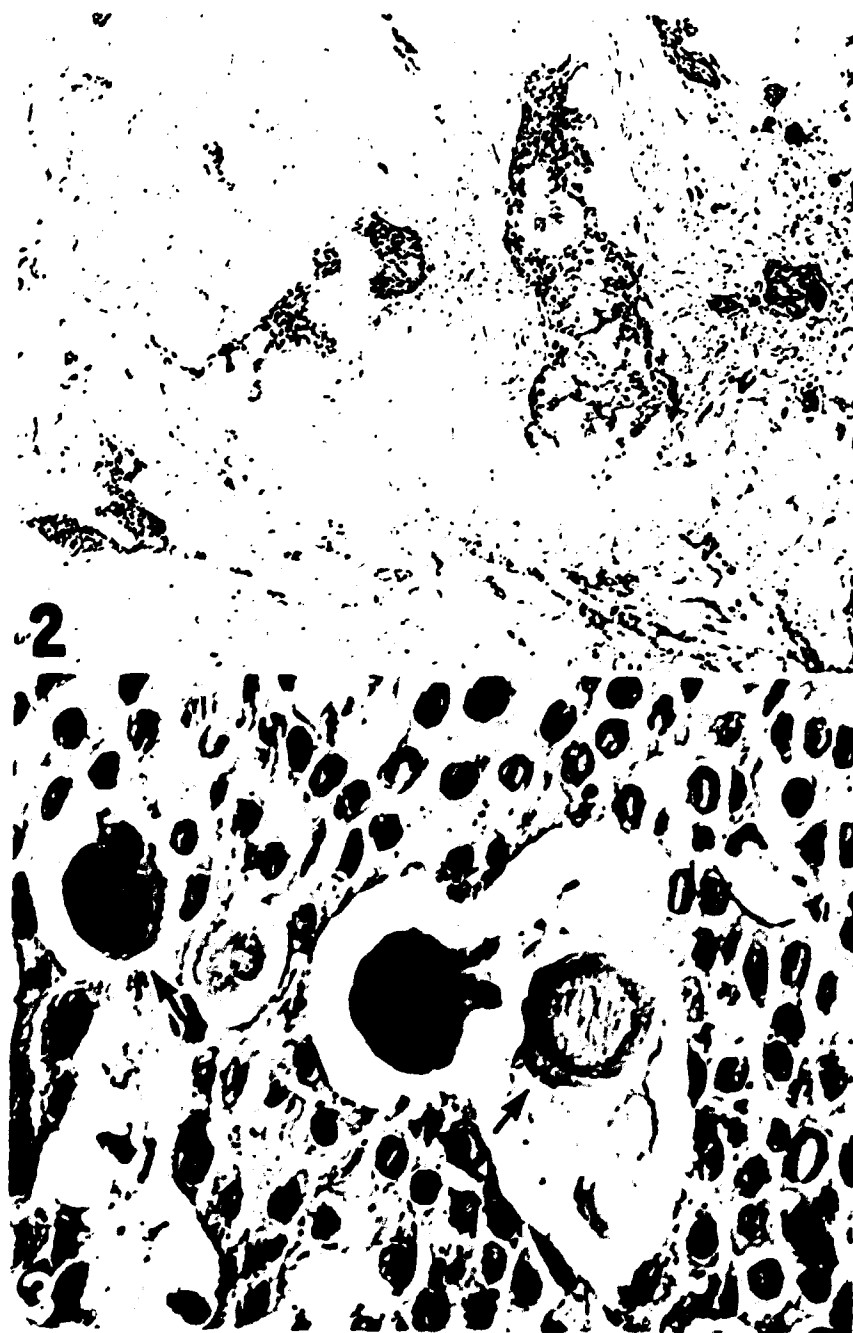


Fig. 2 (AFIP negative 71 9855). The characteristic histologic appearance consists of strands, nests, and clusters of epithelial cells with a slight amount of fibrous connective tissue and large areas of acellular homogeneous material representing amyloid (H & E $\times 55$).

Fig. 3 (AFIP negative 71 12394). The epithelial cells are polyhedral, with large nuclei and distinct nucleoli. Desmosomes may be seen between some cells. The calcifications are typically spherical, with Liesegang's rings (arrows) (H & E $\times 320$).

Malignant conditions such as osteosarcoma, chondrosarcoma, and metastases generally show more aggressive behavior and do not expand the jaw.

Adenoameloblastoma is one of three odontogenic tumors that can demonstrate extensive histological calcification. Such calcifications are generally minute and not visible radiographically.

The tumor consists chiefly of epithelial cells, with a scanty stroma of connective tissue. Furthermore, adenoameloblastoma is located more anteriorly and is often associated with an unerupted tooth. It occurs most often during the second decade in the cuspid area of the maxilla.

A cystic odontoma may have a radiolucent rim surrounding one or more radiopaque areas result

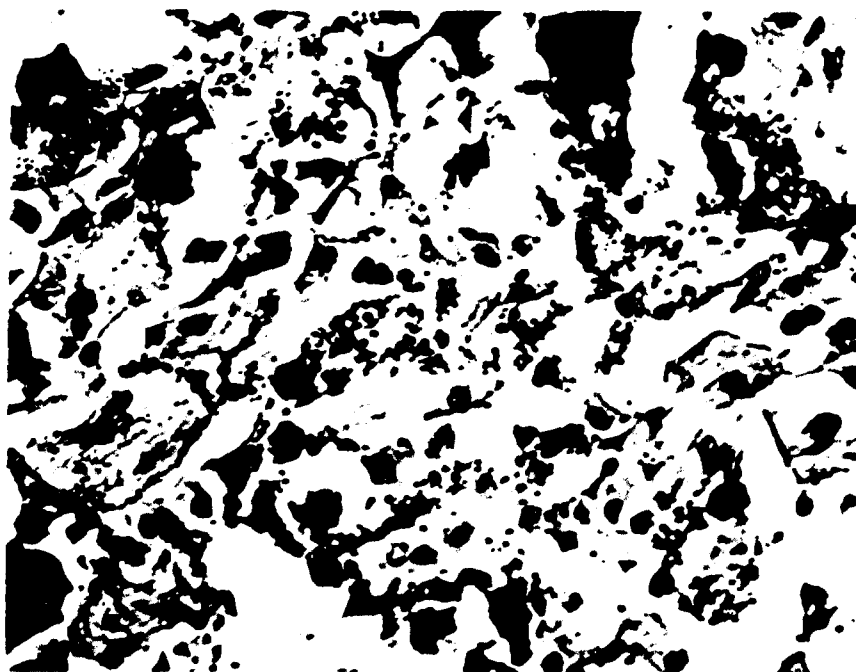


Fig. 1 (AFIP negative 71 0962). Calcifying epithelial odontogenic tumors occasionally have a more pleomorphic epithelial component and clear cells. Mineralization may take the form of tiny droplets or larger aggregates without Liesegang's rings (H & E X370).

ing from the presence of enamel, dentin, and cementum. If these hard deposits superficially resemble teeth, the name compound odontoma is applied; if there is only an irregular mass, the term complex odontoma is used. Most complex odontomas are found in the molar area, while compound odontomas occur in the anterior region of the jaw. The radiodense areas in Figure 1 do not resemble dentition, and in that respect the lesion could be consistent only with complex odontoma. Both types of odontomas are usually associated with impacted or imbedded teeth, do not expand the cortices, and are found more frequently in young patients.

Ossifying fibromas may appear as radiolucent, radiopaque, or mixed lesions, depending upon the amount of calcification present. Early lesions usually appear radiolucent. The tumor is generally well-circumscribed, relatively slow-growing, and produces deformity of the jaw. An ossifying fibroma could have an appearance such as that seen in Figure 1, but the clinical symptoms and the duration of the lesion in this particular case make this possibility highly unlikely.

Acute osteomyelitis first appears as a diffusely lytic, poorly demarcated lesion with indistinct trabeculae. If sequestra are formed, they retain their density and are surrounded by a radiolucent sea of pus. The sequestra may look similar to that seen in Figure 1, but the sharp demarcation of the

process rules out acute suppurative infection. Subacute or chronic osteomyelitis may resemble the acute phase of the disease, but the pain is less severe. Perforation of the cortex may occur in both types of osteomyelitis.

Focal sclerosing osteomyelitis is seen as a dense radiopaque area, most commonly associated with the first mandibular molar. It does not tend to become lytic, even when the tooth is extracted.

Calcifying epithelial odontogenic tumor (Pindborg tumor) was the histologic diagnosis of the lesion demonstrated in Figure 1. It is a rare tumor which may appear largely lytic or show a combination of lytic and productive areas. It is most commonly found in the premolar or molar region of the mandible. The tumor behaves similarly to ameloblastoma, grows slowly, and expands the cortices. It may be seen equally as often in men and women, and most cases occur in the third and fourth decades of life.

PATHOLOGIC DISCUSSION AND CORRELATION

Pindborg tumor is an odontogenic lesion containing nests and sheets of polyhedral epithelial cells in a connective tissue stroma, with varying amounts of calcification and amyloid (Fig. 2). The typical calcifications are closely associated with the epithelial cells and are spherical, with a laminated appearance (Liesegang's rings) (Fig. 3); however, they may also appear as tiny droplets,

irregular masses (Fig. 4), or large aggregates which dominate the histologic pattern. Since the degree of calcification is variable, it is understandable that the radiologic appearance can fluctuate from a completely lytic to a chiefly productive or radiopaque lesion. The tumor is always well-circumscribed, and expansion of the cortices is common. Penetration of the cortex may occur, but this is the exception rather than the rule. The mandible (especially the bicuspid and the molar area) is most frequently involved, and the tumor may be associated with an unerupted tooth.

Although all of the patients discussed in the initial report were men, the accumulation of additional cases has shown the sexes to be evenly affected. All age groups except the very young may be affected. Most patients are in the third or fourth decade.

Clinically, this tumor appears to behave in a benign but locally aggressive fashion, as ameloblastoma does. However, the distinctive histology of the epithelial cells, coupled with the presence of calcification and amyloid, define calcifying epithelial odontogenic tumor (Pindborg tumor) as a specific nosologic entity.

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Calcifying Epithelial Odontogenic Tumor of the Mandible (Pindborg Tumor)

RPC from the AFIP

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ABSTRACT—A radiographic picture of mixed bone destruction and osteosclerosis which is well-circumscribed and expansile is compatible with calcifying epithelial odontogenic tumor (Pindborg tumor). Because of the marked variation of calcification in the tumor, the radiologic appearance ranges from totally radiolucent to chiefly radiopaque. The size of the slowly-growing lesion corresponds to its duration. The differential diagnosis should include adenameloblastoma, odontoma, ossifying fibroma, and osteomyelitis.

INDEX TERMS: Armed Forces Institute of Pathology • Jaws, diseases • Jaws, neoplasms

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