Pseudocysts of Jaw Bones

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Abstract: Odontogenic cysts are epithelial-lined pathological cavities and surrounded by fibrous connective tissue that originate from odontogenic tissues that occur in tooth bearing regions of maxilla and mandible. Other non-odontogenic, non-epithelial lined cysts known as pseudocysts are not considered in the classification as separate entity. Only two pseudocysts—Aneurysmal bone cyst and the Simple bone cyst are considered under the broad term of Giant cell lesions and bone cysts according to the latest WHO 2022 classification. Hence the present review reveals the importance of including pseudocysts of jaw bones as an entirely separate entity under the heading of classification of cysts of orofacial region.

Keywords: Pseudocysts, Aneurysmal bone cyst, Simple bone cyst, Static bone cyst, Bone marrow defects

1. Introduction

Kramer (1974) has defined a cyst as ‘a pathological cavity having fluid, semifluid or gaseous contents and which is not created by the accumulation of pus’. Most cysts, but not all, are lined by epithelium. Pseudocyst of jaw is bone cavities without an epithelial lining. Pseudocysts of jaw bones as an entirely separate entity. ABC was described as脑海中清虚，可作为眼科手术的潜在研究目标。其他非牙源性、非上皮性囊肿被称为假性囊肿，不单独作为entity. Only two pseudocysts—Aneurysmal bone cyst and the Simple bone cyst are considered under the broad term of Giant cell lesions and bone cysts according to the latest WHO 2022 classification. Hence the present review reveals the importance of including pseudocysts of jaw bones as an entirely separate entity under the heading of classification of cysts of orofacial region.

The present review is compilation of all the relevant data regarding pseudocysts of the jaw bones and its clinical significance.

1) Aneurysmal bone cyst (ABC)

Synonym: Aneurysmal bone cavity

Before 1942 ABC was described as- Ossifying Hematomas, Hemorrhagic osteomyelitis, Osteitisfibrosacystica, Expansilehemangioma, Aneurysmal giant cell tumors. [3]

Definition: Intraosseous accumulation of variable sized blood filled spaces often admixed with trabeculae of reactive woven bone.

Jaffe and Lichtenstein:Intraosseousosteolytic lesion affecting metaphyseal regions of long bones.

Percentages: Long bones- 50 %, Vertebral column- 20 %, Non-odontogenic + non-epithelial cysts of Mn – 1.5 %. [4]

2 % of ABC lesions found in head and neck region due to low venous pressure of skull bones.

There are two distinct variations of ABC in terms of clinicopathology: the primary ABC and the secondary ABC, which originates from the secondary lesion. Congenital and acquired versions are other classifications for primary ABC lesions. Congenital ABC is determined by the growth, maturation, and arteriovenous malformation of the teeth throughout infancy. The correlation between trauma and the acquired subtype of primary ABC provides an explanation. Secondary ABC can be linked to the gradual degeneration of pre-existing lesions like a tumor, cyst, or fibro-osseous lesions like giant cell granuloma, ossifying fibroma, or solitary bone cyst.[4]

Etiology: There are multiple theories proposed. Few of them are as follows:

a) Lichtenstein proposed that persistent local alteration in hemodynamics leads to increased venous pressure leading to the development of engorged vascular bed following this resorption of bone occurs by giant cells which is then replaced by connective tissue, osteoid and new bone.

b) Steiner and Kanton proposed that ABC occurs as Primary and Secondary lesion associated with other bone diseases.

c) Bieseker proposed that primary bone lesion initiates an osseous arteriovenous fistula which then creates secondary reactive lesion due to changes in hemodynamic forces. This is one of the most accepted theories.

d) Shear et al gave the concept of Microcyst formation.

The concept of microcyst formation is facilitated by localized areas of necrosis. Following this the microcysts are lined by stromal connective tissue and multinucleated giant cells. This enlarges further with stromal breakdown and eventually coalesces with each other. Loss of stromal support leads to dilution and rupture of thin-walled cells which ultimately leads to hemorrhage into the stroma leading to microcyst formation.

Three stages are recognized in the formation of the ABC. First is the initial phase, with predominant osteolysis and...
non-characteristic appearance. Second is the growth phase, with the rapid growth of the tumor, marked bone destruction, and expansion of the bone. The tumor is not circumscribed, and bony septa are indistinct. Progressively, the first signs of a bony shell appear around the tumor and the last stage is the stabilization phase, with a well-defined unilocular or multilocular radiolucency with histological features of blood-filled sinusoidal spaces along with fibrocellular connective tissue stroma. [5]

Radiographic features: Ballooning growth pattern, Blown-out cortical expansion, Honey comb or soap bubble radiolucency with radiopaque borders.

Pathology:
Macroscopic: Blood soaked in sponge
Operation specimen is brownish to dark blue and soft in consistency. Upon entering the lesion there is excessive bleeding which is encountered. Blood is seen welling up from the tissue.

Microscopic: Fibrous Connective tissue stroma containing many cavernous or sinusoidal blood filled spaces. Young fibroblasts as well as multinucleated giant cells are also noted. Varying amounts of hemosiderin pigment is present along with new osteoid and bone formation. Blood filled spaces have elastic tissue or smooth muscle around them. Spaces of varying sizes containing unclotted blood surrounded by cellular fibroelastic tissue. [6]

ABC is classified as follows: [7]
 a) Conventional or vascular type – Shows rapidly growing, expansile as well as destructive lesion causing cortical plate perforation and soft tissue invasion.
b) Solid-type – Small asymptomatic lesion which is accidental radiographic finding.
c) Mixed type – Showing features of both types and may be due to transitory intermediate phase.

Differential diagnosis:
a) Central giant cell granulomas- ABCs may expand to a greater degree, and they are more common in the posterior parts of mandible compared with giant cell granulomas.
b) Ameloblastoma-Usually occurs in the older age group
 c) Cherubism-It is a multifocal bilateral disease.

Treatment:
Complete removal of the lesion with curettage and enucleation. It has an excellent prognosis.

2) Simple bone cyst (SBC)

Synonyms: Simple bone cavity, Solitary bone cyst, Hemorrhagic bone cyst, Extravasation bone cyst, Unicameral bone cyst, Trabecular bone cyst, Progressive bone cyst, Idiopathic bone cavities.[8]

Definition: Unilocular cavity or pseudocyst that occasionally occurs as a bilateral or even multifocal lesion. Close resemblance to unicameral bone cavity which is analogue to lesions of long bones.

RUSHTAN’s Criteria’s (1985) for diagnosis of SBC:
 a) Cyst should be simple, have no epithelial lining, and show no evidence of acute or prolonged infection.
b) It should principally contain fluid and not soft tissue.
c) Its walls should be hard bone which may thin in parts.
d) Pathologic and chemical findings do not exclude a diagnosis of SBC.

Pathogenesis:
The pathogenesis of SBC is unclear but could be multifactorial in origin. Olech et al proposed that trauma to bone could lead to intramedullary hemorrhage leading to failure of early organization of hamartoma. Fisher et al suggested that SBC could be cysts in fibrous- osseous lesions. Shafer –Neville et al proposed the most accepted the Trauma-hemorrhage theory which states that trauma to bone leads to hemorrhage that occurs within medullary spaces of bone. Further the healing in bone is by organization of clot and formation of connective tissue stroma and new bone.

Radiographic features: When teeth are involved along with their apices radiolucent defect often shows domelike projections. The cyst is localized above and in front of mandibular canal. Scalloping may be present giving an appearance of root apices hanging within SBC cavity. [10]

Pathology:
Macroscopic: During surgical resection of the lesion it may show empty cavity. Some of them may contain blood, serous or serosanguinous fluid. A thin membrane, granulation tissue or clot can be present.

Microscopic: Loose vascular fibrous tissue membrane of variable thickness with no epithelial lining is noted. Hemorrhage and hemosiderin pigment is also present. Also small multinucleated giant cells found. [11]

Donker and Punnia- Moorothy have subclassified SBC based upon the content in the cavity as follows:
a) Empty cavities- Idiopathic
b) Solid content- Designated according to histologic appearance of bulk of the solid
 c) Profile similar to serum- extravasation cysts

Matsumura et al have classified SBC as – Type A + Type B
Type A – Cavity membrane consisted of a thin CT. lining Type B - Presence of thickened myxofibromatous wall

Differential diagnosis: Lingual salivary gland depression of mandible is located below mandibular canal but simple bone cyst is located above mandibular canal.

Treatment and recurrence: Surgical resection is best treatment option. Chances of recurrence are less and have an excellent prognosis. Cautions needed during treatment of large lesions inorder to prevent severance of neurovascular bundles extending from inferior alveolar nerve to areas of vital bone.
3) Lingual mandibular bone depression (LMBD)
   a) Synonyms: Stafne’s cyst, Static bone cavity, Lateral bone cyst, Lingual cortical mandibular defect.
   b) Definition: Unusual form of slightly aberrant salivary gland tissue wherein a developmental inclusion of glandular tissue is found within or, more commonly, adjacent to lingual surface of body of mandible within a deep and well-circumscribed depression. [12]

It was first described by Sir Stafne (1942) as bone cavities situated near angle of mandible.

Pathogenesis: Multiple theories have been proposed as follows:[13]
- Stafne’s first proposed that radiolucencies represent areas of Merkel’s cartilage that failed to ossify. Later he proposed that the lesion was developmental in origin.
- Peterson et al explored areas surgically and found that the areas were devoid of content. Hence this supported the theory of congenital origin.
- Fordyce et al were first to report presence of salivary gland tissue in radiolucencies. Inclusions of salivary gland tissue within mandible were noted during ossification.
- Kay et al proposed the concept of abnormal vascular pressure in facial artery as it pursues its course over inferior border of mandible leading to necrosis and resorption of bone.
- Lello and Makek et al suggested that the defect was a result of ischemia due to combined effect of unfavorable hemodynamics of facial artery and degenerative arterial change in middle age.
- Present theory comments about LMBD to have its origin from the sublingual, submandibular and parotid glands. It leads to local atrophy and resorption in response to pressure.

Radiographic features: Well circumscribed lesion having sclerotic borders. Always inferior to mandibular canal

Anterior LMBD (ALMBD): This is usually superimposed on roots of incisors, canines and premolar hence mimicking periapical lesion.

Posterior LMBD (PLMBD) is a circular or ovoid with well-defined radiolucencies

In order to detect LMBD there need of at least 12 % reduction in volume of mineralized matrix which is needed to visualize defect on radiograph. Hence pressure resorption caused by salivary gland lobe adjacent to mandibular bone takes several years. And hence LMBD’s rarely diagnosed before age of 40 years. [14]

Microscopic: Normal salivary gland tissue usually submandibular salivary glandular tissue is noted. Defect is devoid of tissue or contains muscle, blood vessels, fat, CT., lymphoid tissue.

Differential diagnosis: Traumatic or hemorrhagic bone cyst – This is usually located almost invariably superior to inferior alveolar canal.

Treatment: No treatment needed in most of the cases. Conservative treatment and regular follow-ups are needed.

4) Focal OSTEOPOROTIC BONE MARROW CAVITY/Focal marrow-containing jaw cavity (FMJC)
   a) Synonyms: Hematopoietic defect of jaws, Osteoporotic marrow defect of jaw.
   b) Uncommon and entirely innocuous jaw conditions or cavity.
   c) Pathogenesis: Aberrant form of bone healing with focal formation of hematopoietic bone marrow. Following extraction of tooth the defect is replaced with hematopoietic marrow rather than trabecular bone normal for that area.
   d) Radiographic features: Irregular round or oval radiolucency, varying in size. According to Makek and Lello the anterior border is well delineated but the posterior border is poorly delineated. All mandibular are located above inferior alveolar canal. [15]
   e) Microscopic: Large lymphatic follicle containing an active germinal center. Presence of normal hematopoietic elements with the usual spectrum of blast and mature cells in erythrocytic, myelocytic, lymphocytic and megakaryocytic series is usually noted. [16]
   f) Treatment: No treatment needed. Need for routine follow-up. [17]

Summary about Pseudocysts of the jaw bones is tabulated in Table 1

2. Conclusion

Pseudocysts are defined as bone cavities without an epithelial lining. They are largely asymptomatic and are detected accidentally on radiographic examination. Pseudocysts need to be considered as a separate entity and have to be considered into the classifications for the cysts of the jaw bones. Management in most of the cases is complete removal of the pseudo cystic cavity which is followed by excellent prognosis.

References
Aneurysmal Bone Cyst

Simple bone cyst

Solitary bone cyst

Lingual mandibular bone depression (LMBD)

Focal osteoporotic bone marrow cavity/Focal marrow–containing jaw cavity (FMJC)

Table 1: Summary about Pseudocysts of the Jaw Bones

<table>
<thead>
<tr>
<th>Pseudocysts of jaw bones</th>
<th>Synonyms</th>
<th>Age</th>
<th>Gender</th>
<th>Site</th>
<th>Clinical features</th>
<th>Clinical presentation</th>
<th>Aspirate</th>
</tr>
</thead>
<tbody>
<tr>
<td>Aneurysmal Bone cyst</td>
<td>Aneurysmal bone cavity</td>
<td>8-55 years (Peak 2nd decade)</td>
<td>F:M – 1:2:1</td>
<td>Mandible -Posterior region – Premolar – Molar ramus area, Angle of Mandible Maxilla- Posterior region</td>
<td>Striking feature – Firm swelling develops rapidly</td>
<td>History of trauma preceding the lesion</td>
<td>Dark red or brownish hemorrhagic fluid</td>
</tr>
<tr>
<td>Simple bone cyst (SBC)</td>
<td>Simple bone cyst, Solitary bone cyst, Hemorrhagic bone cyst, Extravasation bone cyst, Unicameral bone cyst, Trabecular bone cyst, Progressive bone cyst, Idiopathic bone cysts.</td>
<td>2-75 years (Peak 2nd decade)</td>
<td>M:F- 1:1:2</td>
<td>Mandible – Premolar – Molar region</td>
<td>Patient might not give any H/O previous trauma since it might be of mild intensity and pt. might be unaware of it. Usually symptom free cyst like lesion</td>
<td>Patient might show the cystic cavity that is empty. Few cases might contain blood, serous or serosanguinous fluid</td>
<td></td>
</tr>
<tr>
<td>Lingual mandibular bone depression (LMBD)</td>
<td>Stafne’s cyst, Static bone cavity, Lateral bone cyst, Lingual cortical mandibular defect.</td>
<td>11-87 years</td>
<td>M:F – 6:1</td>
<td>Anterior LMBD – Lingual mandibular bone region near sublingual salivary gland Posterior LMBD – Between angle of mandible and first molar region below inferior alveolar canal</td>
<td>Asymptomatic, Detected accidentally on routine radiographic examination.</td>
<td>-</td>
<td></td>
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<tr>
<td>Focal osteoporotic bone marrow cavity/Focal marrow–containing jaw cavity (FMJC)</td>
<td>Hematopoietic defect of jaws, Osteoporotic marrow defect of jaw</td>
<td>Middle aged patients – 4th – 5th decade of life</td>
<td>Females&gt;&gt;&gt; Males (M:F ratio- 1:3.3 )</td>
<td>Mandible&gt;&gt;&gt; Maxilla (Mx: Mn – 1: 9.5) Mandible – Mandibular molar and ramus region</td>
<td>Asymptomatic</td>
<td>-</td>
<td></td>
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