# Solitary Bone Cyst of the Jaws: A Review of the Etiopathogenic Hypotheses

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# Solitary Bone Cyst of the Jaws: A Review of the Etiopathogenic Hypotheses

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Solitary bone cysts (SBCs) of the jaws are often polymorphic, show scalloped borders when located between the teeth roots, are devoid of an epithelial lining, and are usually empty or contain blood or a straw-colored fluid. The numerous synonyms referring to these lesions reflect their uncertain nature (eg, traumatic bone cyst, simple bone cyst). SBC, also found in other skeletal locations, is often suspected after epidemiologic and radiologic test results and confirmed at surgery. Histology usually shows fibrous connective tissue or only bone. The various etiologic elements responsible for SBC include tumor degeneration, trauma, or abnormalities during bone growth. The pathogenesis of the SBC is unknown, but it is widely accepted that it could be the result of a vascular dysfunction leading to a local posthemorrhagic ischemia, inducing an osseous aseptic necrosis. This article reviews likely but still-debated etiopathogenic hypotheses of lesions of the jaws and other, more frequent bony locations, such as the humeral and femoral metaphysis.

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Solitary bone cyst (SBC) of the jaws is uncommon, representing approximately 1% of all jaw cysts. The pathogenesis and etiology of these lesions remain unknown. The many different synonyms for SBC (eg, traumatic bone cyst, hemorrhagic bone cyst, idiopathic bone cyst, osteodystrophic cyst) reflect different etiopathogenic processes that remain difficult to elucidate, contributing to the confusion and lack of understanding when such terms such as "idiopathic cyst" and "essential cyst" are used. Classified as a jaw pseudocyst,[1] SBC is an osteolytic lesion forming a cavity with either a geodic or polymorphous shape. It may be empty or filled with blood, serum, or a serohematic liquid.

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## Epidemiology

The most common SBC locations are the long bones (90%), with a predominance in the metaphyseal region of the proximal ends of the humeral (65%) and femoral (25%) shafts.[2-5] The evolution of these lesions is a centrifugal extension toward diaphysis cotemporaneous of the bone growth. SBC of the jawbones appears to be far less frequent (10%). The body of the mandible is usually affected (75%) in the premolar and molar regions, with a possible, and sometimes important, posterior extension.[6,7]

In both cases, SBC is diagnosed predominately in the first 2 decades of life (75%). The maxillomandibular location is distributed almost equally between the genders, although there is a male predominance for the extrafacial variants.[8]

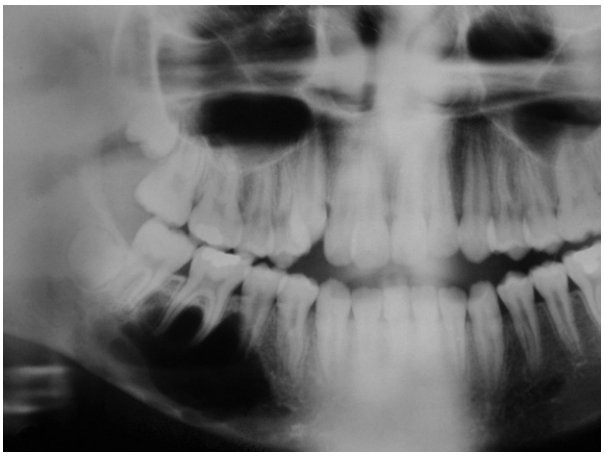

**FIGURE 1.** Panoramic radiograph showing a solitary bone cyst in the mandible of a 12-year-old girl.

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## Symptomatology

Most often, SBC is clinically asymptomatic, with no swelling or other functional signs. It is often discovered accidentally during examination of a panoramic radiograph.[9,10] SBC evolution in long bones is asymptomatic at the beginning, but eventually produces clinical signs, such as a sporadic limp in the case of the femoral location. Another feature is severe pain due to spontaneous fracture, occurring in 90% of the humeral SBC locations.[11,12]

## Radiographic Findings

The radiologic interpretation of mandibular SBC is often straightforward for typical lesions. Generally, images show a unilocular homogeneous osteolysis, surrounded by a narrow cone-shaped bony condensation with horizontal or vertical apex. The largest lesions demonstrate a more radiolucent polymorphic image, often with a scalloped appearance of the upper edge extending in between the roots, which are not affected by the lesion (Fig 1). In some cases, SBC appears multilocular with septum-like images, thus evoking other possible lesions.

SBCs of long bones show a radiolucent lacuna, usually located in the intramedullary metaphysis with no alteration at the epiphyseal cartilage plate. The bottom is generally heterogeneous with osseous spiculae, which are "fallen" bone fragments in the cyst after a spontaneous fracture. When present, these fallen fragments are considered pathognomonic of a solitary cyst. The lower border shows a characteristic "egg cup–like" appearance (Fig 2).

## Histopathogy

Gross examination shows a cystic wall composed of a thin connective membrane. This gray-yellowish tissue is very friable, hemorrhagic, and difficult to remove. Some foci of osseous aseptic necrosis are seen, more frequently in long-bone lesions. The cystic contents seem to change according to the SBC's evolution and location; the cyst can be filled with blood or serohematic or serous fluids and also can be empty, especially in mandibular lesions.

Microscopic examination typically shows the cystic wall as a connective tissue membrane with numerous collagen fibers, with no epithelial lining (Fig 3). Numerous fibroblasts and giant cell–like osteoclasts are sometimes visible, with some newly formed trabecular bone surrounded by numerous osteoblasts. Numerous congested capillaries and cholesterol crystals related to the osseous necrosis also may be present.[13,14]

## Etiopathogenesis

The etiopathogenesis of mandibular SBC has not been extensively investigated to date. Confusion of-

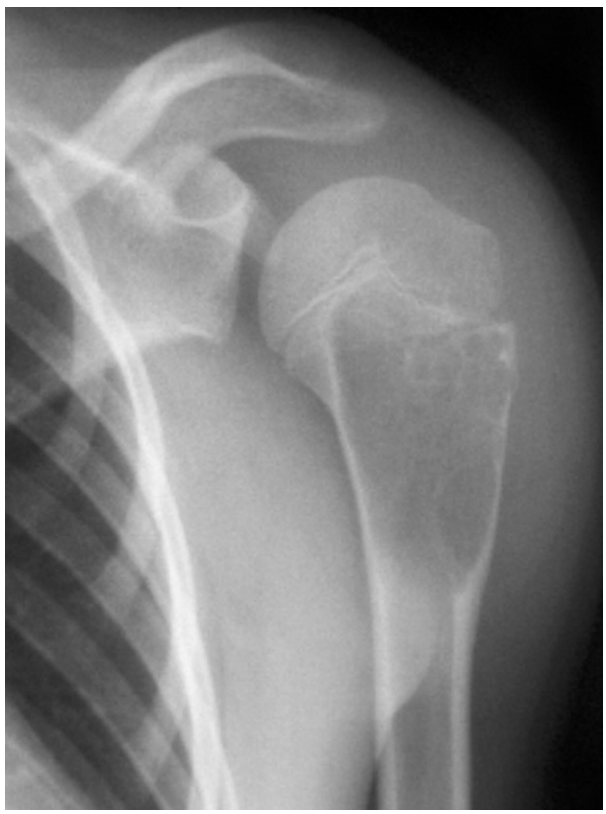

**FIGURE 2.** Roentgenogram showing a solitary bone cyst in the humeral metaphysis of a 10-year-old boy.

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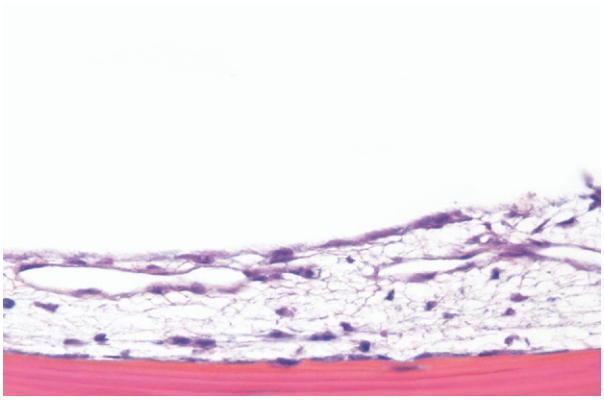

**FIGURE 3.** Bone covered by a thin layer of loose fibrous connective tissue. (Hematoxylin and eosin; original magnification ×40.)

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ten rises due to the unclear understanding of the interaction between the lesion's pathogenic process and etiologic nature. Although there seems to be some consensus about the pathogenesis among orthopedists, much controversy also exists as to the possible triggering factors.

Although microscopic examination can often be disappointing because commonly expected characteristic elements of the lesion may be missing, areas of osseous necrosis and bone remodeling in the cyst wall may be present. This aseptic osseous necrosis could be possible evidence supporting the vascular origin of SBC. The local ischemia seems to be due to an arterial dysfunction, along with altered intramedullary venous drainage, which might be interrupted.[15,16] Cyst growth then may be enhanced by the increased intramedullary pressure due to an overflow of intracystic fluid.[17] This fluid, which is very similar to blood serum, contains enzymatic elements from osseous remodeling areas present on the SBC wall, which contains significant vascularity.

Analysis of the cystic fluid seems to support the finding that this is a transudate devoid of microorganisms[18]; it shows significant concentrations of enzymatic factors, indicating osteoclastic activity.[11] The presence of metalloproteinases is of particular significance, because these substances are involved in osteogenesis and osteoclastic phenomena.[19] In mandibular SBC, the absence of cystic content, as commonly observed in extensive lesions, might indicate the end of its evolution.

## Etiologic Hypothesis

Considering the diversity of theories put forward in the literature, whatever the skeletal location of SBC, the etiology of SBC remains unclear. Among the many theories, 3 predominate: *1*) an abnormality of osseous growth, *2*) a degenerating tumoral process, and *3*) a particular factor triggering hemorrhagic trauma.

### ABNORMALITY OF BONE GROWTH

This theory arose from clinical observations, the time of diagnosis (preadolescence or adolescence), and the development within or near osseous remodeling areas. The earlier term "osteodystrophic cysts" supports this etiologic hypothesis; a local abnormality during the development and growth of bone might indeed explain the predominant locations of SBC near the cartilage growth plates, which could either act as a barrier against the cyst spreading toward the epiphysis or promote cyst expansion toward the diaphysis.[10,20,21]

Humeral and femoral metaphyses are also areas in which osseous remodeling and ossification spots multiply and reshape significantly during body growth. Thus, some authors suspect that SBC may represent an "out of control" remodeling osseous area.[22]

In the mandible, the primary ossification spot is located near the mental foramen. Because this area is the preferential zone of SBC occurrence, it is possible to consider the hypothesis of an abnormality in cellular differentiation during ossification and growth related to local environmental factors inducing mechanical constraints during osteogenesis and angiogenesis.[23] In fact, the mandible is a bone in which various complex ossification mechanisms interact. Developing a clear theory is difficult, however, because many diverse ossification processes (cartilaginous, membranous, and enchondral) occur at this location.

### TUMOR DEGENERATION

This theory of a tumoral degeneration process is based on clinical observations mostly reported in osteodystrophic pathological conditions, such as fibrous dysplasia and central giant cell granuloma. SBC may appear as a benign complication of fibrous dysplasia due to fibrous degeneration and microcystic formations.[24] This hypothesis should be discussed keeping in mind the prevalent fibrous dysplasia locations in diaphyseal areas and maxillary bone.

The liquefaction in the middle part of a central giant cell granuloma is purported to lead to SBC formation, and hence is likened to a healing process.[15,25] In this respect, the connection between the prevalence of central giant cell granulomas in the epiphyseal and symphyseal areas and the tumoral degeneration hypothesis remains a matter of debate.

### TRAUMATISM

This is the most widely accepted hypothesis, based on the occurrence of an intramedullary hemorrhage followed by a hematoma after trauma insufficient to fracture a healthy bone. The pressure from the hema-

toma causes venous stasis that leads to an area of bone marrow necrosis[2,15,26] and osteoclastic resorption attributable to decreased tissue pH.[27,28]

In the mandible, trauma also might provoke a thrombosis or a lingering spasm of a terminal artery with ischemia and aseptic necrosis, thereby leading to cyst formation. Vascular alterations are supposedly related to the resorption phenomena. The process by which osteoclasts differentiate remains unknown. Nevertheless, the traumatic etiology hypothesis is challenged by the fact that more than 50% of cases have no traumatic history. Moreover, the preferential sites of long-bone SBC are not systematically found at the most exposed areas.[29] Indeed, the humeral shaft is much less traumatized compared with the tibia, where the lesion is quite uncommon.[30] This theory could apply to the mandible due to numerous microtraumas to the teeth and alveolar process.

Based on current clinical knowledge, we have investigated the major etiologic hypothesis of SBC of the jaws and the much more frequent humeral and femoral metaphysis locations. Some consensus about the osteolytic pathogenesis seems to emerge; it might be due to an alteration of the vascular system causing a posthemorrhagic ischemia, responsible for aseptic bone necrosis as well as an intracystic transudate whose enzymatic factors contribute to the bone resorption. The major etiologic processes often quoted, but still being debated, are tumoral degeneration, traumatism, and defects linked to bone growth periods.

Although mandibular SBC and long-bone SBC have some similarities with respect to diagnosis and etiopathogenesis, they should not be approached in the same way, because their management and prognosis differ. For jawbone lesions, treatment is easy, consisting of curettage, frequently leading to complete healing with no recurrence. In long-bone SBC, where the benign nature may be debatable, the therapeutic approach is more difficult and quite different, involving various technical combinations and uncertain results.

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