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# Multifocal Traumatic Bone Cysts: Case Report and Current Thoughts on Etiology

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The traumatic bone cyst was first described by Lucas[1] in 1929 and later defined by Rushton[2] as a single cyst that has no epithelial lining, has an intact bony wall, is fluid filled, and has no evidence of acute or chronic inflammation. The term *traumatic bone cyst* has been recognized as a misnomer in that the incidence of prior trauma in patients with this entity is the same as in the general population.[3] A variety of other terms have been used by different authors to describe the traumatic bone cyst. These include solitary bone cyst,[2] simple bone cyst,[4] hemorrhagic bone cyst,[5] progressive bone cyst,[6] idiopathic bone cyst,[7] and unicameral bone cyst.[8] Overall, more than 95% of these cases involve the long bones such as the proximal humerus and femur.[9] Several hypotheses for the pathogenesis of this lesion have been postulated. Cohen[10] has proposed that the cyst develops because of a lack of collateral lymphatic drainage of venous sinusoids. This apparent blockage then results in the entrapment of interstitial fluid causing resorption of the bony trabeculae and cyst development. Alternatively, Mirra et al[11] proposed that traumatic bone cysts are synovial cysts, developing as a result of a developmental anomaly whereby synovial tissue is incorporated intraosseously.

Traumatic bone cysts are typically found as solitary lesions. Interestingly, in a review of the literature, multiple synchronous lesions were reported to occur in about 11% of cases.[3] This case report details an unusual presentation of multiple traumatic bone cysts.

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## Report of a Case

A 32-year-old white woman saw her general dentist for a routine annual visit when a large radiolucency of the left mandibular body was noted on her panoramic radiograph. Endodontic treatment of the lower left first molar tooth was begun for a presumed diagnosis of a radicular cyst. There was no change in the size of the lesion over the following 6-month period, and she was referred to the Department of Oral and Maxillofacial Surgery, Emory University (Atlanta, GA). The patient's medical history was only significant for a right cerebrovascular accident 8 years earlier that resulted in right-sided deafness and hypoesthesia of all divisions of the right trigeminal nerve. The patient also reported seasonal allergies for which she periodically took antihistamine medication. She reported occasional alcohol use but denied any use of tobacco products. She recalled no history of trauma.

Clinical examination was unremarkable with no evidence of lymphadenopathy, swelling, or asymmetry. Intraoral examination did not show any soft tissue abnormality or bony expansion. The periodontium was noted to be healthy with no evidence of gingivitis, periodontal pocketing, or tooth mobility. There were no carious lesions. The vitality of all teeth, with the exception of the lower left first molar, was confirmed with the application of a cold stimulus. The panoramic radiograph showed a scalloped unilocular radiolucency in the left body area and multiple unilocular periapical radiolucencies in the mandibular symphyseal region (Fig 1). Computed axial scanning further confirmed the presence of a unicystic lesion within the left mandibular body and multiple unicystic lesions within the mandibular symphysis (Figs 2-4).

The symphyseal lesions were thought to most likely be periapical cemental dysplasia. The left mandibular lesion was thought to be a traumatic bone cyst, radicular cyst, keratocyst, or unicystic ameloblastoma. Endodontic treatment was completed before the surgical procedure. The proposed treatment included exploration and excisional biopsy of the left mandibular body lesion with apicoectomy and retrograde filling of the lower left first molar. An excisional biopsy of one of the symphyseal lesions was also proposed. A complete blood count and serum electrolyte levels, including calcium, were assessed before the surgical procedure, and all indices were within normal limits. With the patient under general anesthesia, both lesions were approached with gingival crevicular incisions with anterior vertical releases. Subperiosteal dissection exposed the overlying bone, which was noted to be normal. A drill was then used to remove the buccal cortical plate overlying the lesions. In both locations the lesions were noted to be empty with no evidence of a lining or fluid content. In the

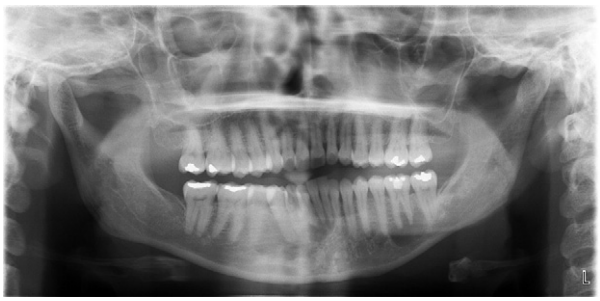

**FIGURE 1.** Initial panoramic radiograph with unicystic lesions of symphysis and left body of mandible.

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anterior lesion, dental branches of the incisal neurovascular bundle were noted traversing the cavity. Both lesions were curetted in an attempt to obtain tissue for histopathology. Despite several attempts, no soft tissue lining was encountered and, accordingly, no tissue could be obtained for histopathologic analysis. Minimal bleeding was present in both cavities as a result of the curettage. Apicoectomy and retrograde filling with super ethoxy benzoic acid were then performed on the mesial root of the lower left first molar tooth. The postoperative course was uneventful. A follow-up panoramic radiograph at 12 months has shown good bony filling of all lesions including the symphyseal lesions that were not surgically explored (Fig 5).

## Discussion

This case report describes a female adult patient with multiple unilocular lesions of the mandibular body and symphysis that are consistent with multiple traumatic bone cysts. As reported by Kaugars and Cale,[3] traumatic bone cysts have an equal prevalence in both genders, present at a mean age of 18 years, are

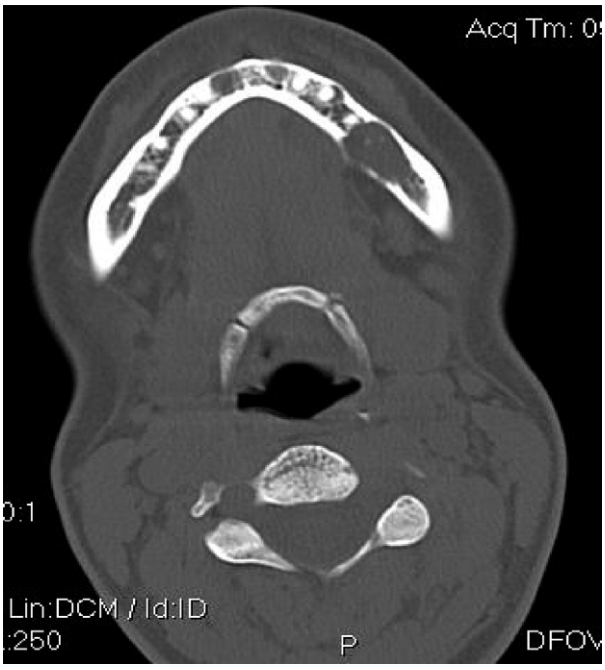


**FIGURE 2.** Axial computed tomography image of symphyseal and body lesions.

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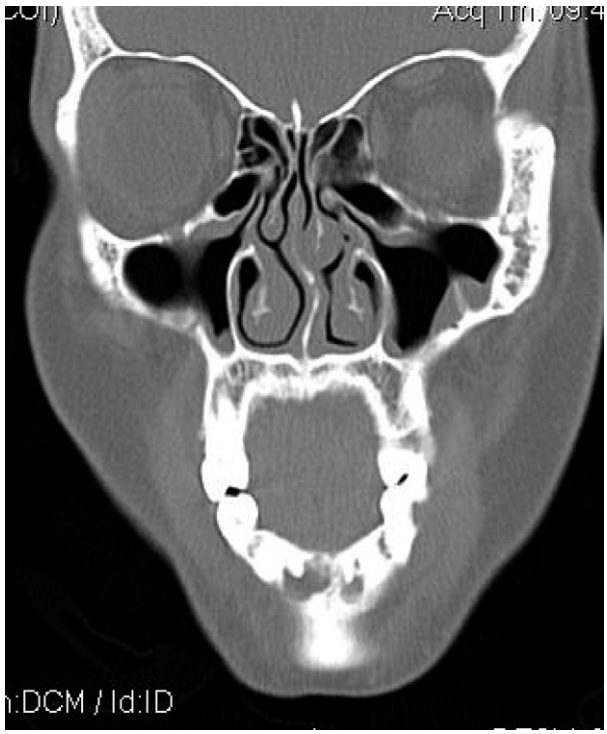


**FIGURE 3.** Coronal computed tomography image of symphyseal lesions.

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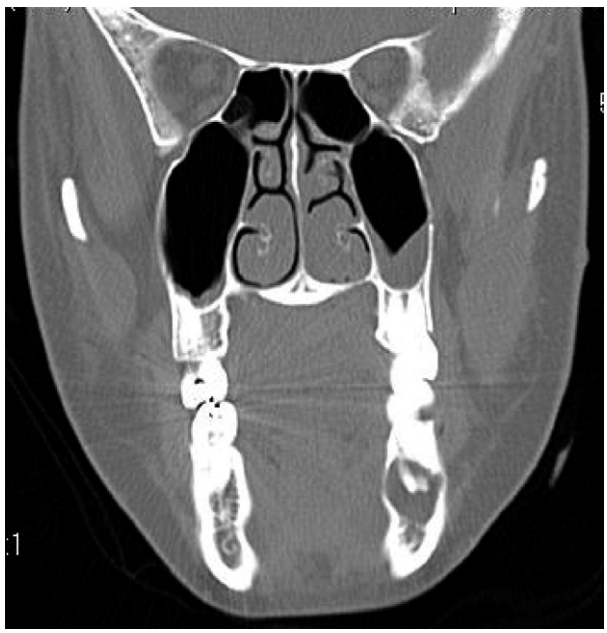

**FIGURE 4.** Coronal computed tomography image of left body lesion.

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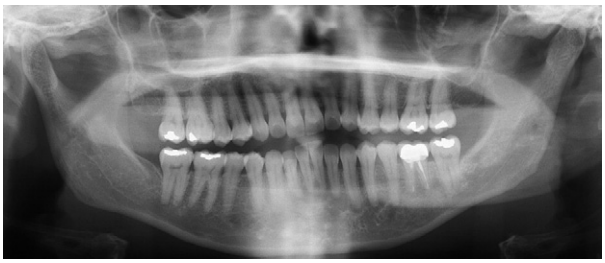

**FIGURE 5.** Postoperative panoramic radiograph at 12 months showing good bone deposition.

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most prevalent among white persons, and most often affect the posterior mandible. Our patient was unique in that she was an older adult with multiple cysts. Other cases of multiple mandibular traumatic bone cysts have also been reported in the literature.[6,7,12-15] The typical location for traumatic bone cysts is the mandibular body, whereas maxillary lesions tend to be uncommon, although the reasons for this are unclear. It is possible that the maxillary sinus makes radiographic visualization of maxillary lesions inherently more difficult.[14] In our case the initial presentation of the symphyseal lesions was so typical of periapical cemental dysplasia that a diagnosis of traumatic bone cysts was not considered.

Various hypotheses have been proposed for the pathogenesis of the traumatic bone cyst. The myriad of different proposed mechanisms provides some insight into the lack of understanding of this unusual entity. The most frequently proposed theory for the development of these lesions involves a traumatic event inciting medullary hemorrhage and a subsequent failure of the hematoma to organize and be replaced with tissue.[16] Many authors have questioned this mechanism, given that often there is no history of trauma and, furthermore, the incidence of trauma in patients with traumatic bone cysts is no greater than in the general population.[3] Although the mean age at presentation is 18 years, when it could be hypothesized that trauma to the jaws is more likely, there is no difference in the prevalence between males and females despite a higher incidence of trauma in males. No history of trauma could be elicited from our patient.

Cohen[10] proposed that the formation and existence of the traumatic bone cyst are due to a blockage of the normal draining of interstitial fluid. Because the normal hemodynamic pressures of the area are low, the expansion of the cyst would require only a small increase in the hydrodynamic pressure within the cyst.[10] Unfortunately, as in our case, many traumatic bone cysts are found to be empty at surgery with no evidence of cyst fluid. This would seem to mitigate against this proposal. Furthermore, if the cyst developed because of a blockage of draining interstitial fluid, one might expect that these lesions would develop with a more equal frequency in all locations within the facial skeleton rather than occur with a higher frequency in the posterior mandible as has been documented.[3]

The most recently proposed mechanism considers these lesions to be synovial cysts arising from a developmental juxtaepiphyseal error with the intraosseous incorporation of synovial tissue.[11] Mirra et al[11] proposed that a small nest of synovium becomes trapped intraosseously during fetal or early infant development and that this tissue may retain some secretory function, resulting in the development of a cyst. Furthermore, they hypothesized that the fibrous tissue and osteoid and giant cells often found at the periphery of the traumatic bone cyst would be from a host-bone reaction.[11] This theory may explain the greater occurrence in adolescents when developmental anomalies often first present. Similarly, traumatic bone cysts of the long bones are often discovered at young ages, although this is most often a result of pathologic fracture. Our case is not consistent with the synovial cyst theory for several reasons. First, our patient presented at an older age. Second, neither of the surgical cavities entered showed any fluid content or evidence of a synovial lining. Third, if synovial tissue exists within the cystic cavity, localized curettage to promote bleeding should not remove all of this tissue and cyst recurrence would seem likely. Recurrence of a traumatic bone cyst after localized curettage is rare, however.

The etiology of the traumatic bone cyst remains unclear. It appears to be developmental in nature, and the predilection for the posterior mandible is consistent with this, given the frequency of developmental odontogenic cysts and tumors in this location. Whether fluid accumulation plays a role in the initial development of a traumatic bone cyst is unclear, but it may explain the bony resorption and cavity development. The potential source of the fluid is unknown, with no real evidence to support or refute interstitial or synovial fluid. It is interesting that in our patient all symphyseal lesions resolved although only one was surgically entered. It would therefore seem likely that all of the symphyseal lesions were in communication, perhaps through very small sinusoids or channels.

The diagnosis and treatment of this interesting lesion may help to elucidate the pathophysiology. The diagnosis of traumatic bone cyst relies on clinical, radiographic, and ultimately, surgical findings. This applies to lesions located within the oral and maxillofacial skeleton as well as the appendicular or axial skeleton. One unique difference with the diagnosis of traumatic bone cyst in the orthopedic literature ex-

ists. Approximately 80% of orthopedic traumatic bone cysts are discovered because of pathologic fracture,[9] in stark contrast to maxillofacial lesions that are noted incidentally on panoramic radiographs. The diagnosis of traumatic bone cyst in the maxillofacial region related to a pathologic fracture has not been previously reported. Asymptomatic long bones in children do not routinely undergo radiography, and the true prevalence of orthopedic traumatic bone cysts may therefore be grossly underestimated. Annual dental screenings are much more likely to identify asymptomatic lesions. The diagnosis of orthopedic traumatic bone cysts relies on a typical radiographic appearance and aspiration of straw-colored fluid at surgery.[17-19] It is possible that many traumatic bone cysts that are discovered as a result of pathologic fracture accumulate fluid after the fracture. The diagnosis of maxillofacial traumatic bone cysts also relies on a typical radiographic appearance and the more common identification of an empty cavity at surgery. However, the original description of the traumatic bone cyst in the maxillofacial literature identified this cyst by the presence of clear cystic fluid at surgery. The reasons for this apparent dichotomy are unclear, although this may simply represent different stages in the development of the same lesion. Hansen reported on the surgical evaluation of 66 traumatic bone cysts of the jaws with only 30 of the lesions being empty.[20] The identification of an empty air-filled cavity on aspiration serves as a valuable diagnostic tool. Conversely, aspirating fluid or the failure to aspirate anything from a jaw lesion would mandate a surgical approach to the lesion for definitive biopsy, given the large number of odontogenic cysts and tumors that may occur. Further evaluation of a suspicious radiographic lesion on panoramic radiography may be performed with computed tomography. The latter will usually allow distinction between solid/fluid-filled lesions and air-filled cavities.

Within the orthopedic literature, intralesional injection of methylprednisolone has been described as a treatment modality for traumatic bone cysts in the long bones.[9,21] It has been proposed that the healing is not solely a response to the corticosteroid but results from a mechanical disruption of the cavity.[9,21] In a randomized multicenter clinical trial intralesional injection of methylprednisolone produced superior rates of healing compared with intralesional injection of bone marrow.[17] This trial evaluated lesions of the upper or lower extremity, and the diagnosis was confirmed with the aspiration of clear or straw-colored fluid.[17] Another study comparing operative treatment and steroid injection in 57 patients with lower and upper extremity lesions resulted in a 38% recurrence rate after surgical intervention compared with 5% after steroid treatment.[22] It was, however, noted that in the steroid group, multiple injections were required for 7 of the 20 patients. Overall, the authors concluded that the steroid method may have equal efficacy but less morbidity compared with operative treatment. The efficacy of the corticosteroid may be the result of influencing the cellular physiology of the lesion. The mechanism of action of corticosteroid is complex, with both anti-inflammatory properties and significant attenuation of cellular metabolism via modulation of nuclear transcription. Methylprednisolone has been shown to influence synovial cells to secrete less prostaglandin, resulting in a decrease in bone resorption while allowing other cells to rapidly reproduce.[23] The use of corticosteroid injection after the diagnosis of traumatic bone cyst is initially made may warrant further evaluation for lesions of the jaws. This could greatly simplify our current approach to this interesting lesion by necessitating only aspiration and injection of the lesion when an empty cavity is encountered. Further studies would be beneficial to help clarify the etiology and management of these curious lesions.

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# Diffuse Chronic Sclerosing Osteomyelitis of the Mandible With Synovitis, Acne, Pustulosis, Hyperostosis, and Osteitis: Report of a Long-Term Follow-Up Case

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Mandibular osteomyelitis is one of the most common infectious diseases and is usually odontogenic or traumatic in origin. Meanwhile, mandibular osteomyelitis caused by a process of unknown etiology is known to develop during the clinical course. In 1987, Chamot et al[1] described a syndrome associated with synovitis, acne, pustulosis, hyperostosis, and osteitis (SAPHO syndrome), which is characterized by osteoarticular and dermatologic symptoms.[2] The most prevalent site of bone lesions is the anterior chest wall with involvement of other locations including the sternum, clavicles, ribs, spine, and peripheral long and flat bones.[1-4] Bone lesions in SAPHO syndrome demonstrate clinical and radiologic features similar to diffuse sclerosing osteomyelitis.[5] Clinical diagnosis of SAPHO syndrome is defined as the presence of any one of the following: *1)* multifocal osteitis with or without skin manifestations; *2)* sterile acute or chronic joint inflammation associated with pustules or psoriasis of palms and soles, or acne, or hidradenitis; or *3)* sterile osteitis in the presence of one of the skin manifestations.[6] Other investigators have suggested that early diagnosis of this condition is crucial to avoid repeated examinations and invasive procedures; however, the etiology of SAPHO syndrome remains unknown.[1,2,5-7] Treatment has therefore been difficult and focuses on symptoms only.[3,5,7]

This report presents the long-term follow-up of a case of SAPHO syndrome in the mandible of a patient who received nonsteroid anti-inflammatory drugs (NSAIDs) and long-term administration of macrolides in combination with surgical procedures.

## Report of a Case

A 51-year-old woman was referred to the Department of Oral and Maxillofacial Surgery, Nagasaki University Graduate School of Biomedical Sciences (Nagasaki, Japan) in November 1998 because of a painful swelling of the right

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