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Bilateral Mandibular Simple Bone Cysts: An Unusual Case Report

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1. Abstract

1.1. Background

Simple bone cysts are intraosseous cavities that are surrounded by bony walls, and they are either empty or contain liquid and/or connective tissue. They have similar radiological findings to those of true cysts, but they do not have an epithelial lining and are thus regarded as pseudocysts. They are frequently asymptomatic, and they are most often an incidental finding during radiographic examination. The purpose of this case report is to present a rare case of bilateral occurrence of mandibular simple bone cysts.

1.2. Case Description

A 23-year-old female patient presented to the Department of Oral Diagnosis and Radiology of the School of Dentistry, University of Athens. Her chief complaint was tenderness and discomfort around the back of her mouth bilaterally. After taking medical history and conducting intraoral and extraoral examination, she was referred for panoramic radiography and Cone Beam Computer Tomography (CBCT).

1.3. Discussion

Upon examination of the panoramic radiograph, two large, well-defined and well-corticated radiolucent lesions with scalloping along the roots of teeth were detected on the body of the mandible bilaterally. CBCT showed a slight buccal–lingual expansion of the cortical plates, as well as thinning of the lingual cortical plate.

1.4. Conclusion

Bilateral simple bone cysts of the mandible are rare and only a few have been reported in the literature. The prognosis is good, and recurrences are rare. Knowledge of clinical and radiographic features is helpful in diagnosing this entity successfully.

2. Introduction

Simple bone cysts (SBC) are intraosseous cavities encased by bony structures, which may either be devoid of contents or filled with fluid or connective tissue. They have similar radiological findings to those of true cysts, but they do not have an epithelial lining and are thus regarded as pseudocysts. They are frequently asymptomatic and most often an incidental finding on radiographic examination. Synonyms of simple bone cysts are traumatic bone cysts, hemorrhagic bone cysts, solitary bone cysts, progressive bone cysts, extravasation cysts, and unicameral bone cysts [1].

In 1946, Rushton [2] developed early diagnostic criteria for SBCs, which are as follows: a solitary lesion, no epithelial lining, no evidence of acute or prolonged infection, a cavity surrounded by bony walls that may contain fluid and/or soft tissue, and no other pathologic or chemical findings that would exclude the diagnosis of SBC. In general, these criteria are still accepted, except that multiple cysts have also been reported, as well as cavities containing gas rather than fluid.

SBCs are rare. They have been reported more frequently in the long bones and, occasionally, in the jaws, comprising 1.25% of

jaw cysts. They mainly affect patients in their second decade of life, in men and women alike [3]. The etiology of these lesions is unclear. Despite the synonym “traumatic bone cyst”, trauma has been ruled out as a cause of simple bone cysts, because the incidence of these lesions in patients with prior trauma is the same as that in the general population [4]. According to that theory, trauma-induced intermedullary hematoma caused ischemia of the bone tissue, which subsequently led to osteolysis and, thus, the formation of a cavity. Since there is no gender predilection, the concept that trauma is the sole causative factor of SBC is discredited if one accepts the hypothesis that males experience traumatic injuries more often than females [5]. No more than half of the simple bone cysts currently reported in the literature are associated with a history of trauma. Other hypotheses include a blockage of lymphatic drainage from venous sinusoids, leading to resorption of bony trabeculae; developmental anomalies resulting in synovial fluid being incorporated intraosseously; and osteolysis secondary to altered bone metabolism [6, 7].

Radiographically, the simple bone cyst presents as a solitary radiolucent area with well-defined borders, with or without sclerotic lining across the periphery of the lesion. According to the literature, only 11% of SBCs present as multifocal lesions, with the first case reported by Hankey in 1947 [8]. In Cone Beam Computer Tomography (CBCT), slight bucco-lingual expansion of the cortical plates of the bone can be observed, as well as displacement of the inferior alveolar nerve canal. In general, SBCs tend to grow longitudinally in the bone, causing minimal expansion; however, expansion of the involved bone can occur, mostly buccally, and is more common with larger lesions. The most common radiographic finding of SBCs is the “scalloping effect,” where the cyst extends between the roots of the teeth [9]. It is also worth noting that this particular “scalloping effect” can also be seen in edentulous areas. Even though SBCs are usually an incidental finding in panoramic radiographs and CBCT examinations, there have been reports of swelling due to bone expansion (one fourth of the cases), pain (less than 10% of the cases), pathological fractures, and hypoesthesia of the inferior alveolar nerve [10]. SBCs are usually unilocular, and most are found in the mandible, especially in the posterior areas of its body, anywhere from the symphysis to the ramus. The uncommon occurrence within the maxilla (about one-third of them) may be better explained by its relatively lower bone marrow volume and vascularity. However, a simple bone cyst in the maxilla may resemble the floor of the maxillary sinus and may be overlooked [11, 12].

Rarely, more than one cyst may be found in the same patient; a few cases have been described, usually associated with florid cemento-osseous dysplasia (FCOD). Although mechanistic links remain speculative, the association between these two entities is still not understood. FCOD, usually seen in black females over 30 years of age, refers to the multifocal form of cemento-osseous dysplasia that is characterized by the presence of multiple lesions distributed throughout the jaw, especially in the posterior areas of the body of the mandible [13, 14]. It is not uncommon to find these extensive radiolucent lesions existing simultaneously in all four posterior quadrants of the jaw. In general, multiple radiolucencies in the jaw most frequently occur in the setting of endocrine or metabolic disorders, such as hyperparathyroidism, or in cases of multiple odontogenic keratocysts, which are associated with the Gorlin-Goltz syndrome [15]. In this article, a case report of a bilateral occurrence of simple bone cysts with no association with cemento-osseous dysplasia or other metabolic disorders will be presented.

3. Case Report

A 23-year-old white female patient presented to the Department of Oral Diagnosis and Radiology at the University of Athens School of Dentistry (Athens, Greece) in March 2024. Her chief complaint was tenderness and discomfort around the back of her mouth bilaterally, present for the past 2 months, as well as a gradual reduction in mouth opening. After taking the medical history and conducting intraoral and extraoral physical examinations, she was referred for panoramic radiography to assess the wisdom teeth. It is worth noting that no palpable lymph nodes were present, and the patient’s medical history was not contributory.

Upon examination of the panoramic radiograph, two large radiolucent lesions with well-defined borders were discovered incidentally on the body of the mandible bilaterally, which extend from the canines to the mesial root of the third molars, below their respective apexes (Figure 1) She was then referred for a Cone Beam Computer Tomography scan (CBCT) for further investigation, where the presence of the two well-corticated and well-defined radiolucent lesions was confirmed, with a size of 44mm x 13mm bilaterally. CBCT images were acquired with a Newtom VGi CBCT imaging unit (Cefla, Imola, Italy), using an 8cm x 8cm field of view for the maxilla and a 12cm x 8cm field of view for the mandible at 110kV, 2.69mA, with a 3.6s exposure time. A slight buccal –lingual expansion of the cortical plates was observed, as well as thinning of the lingual cortical plate. The inferior alveolar nerve had also been displaced from both sides, and no root resorption of the teeth was detected (Figure 2.) The present teeth were all vital.

Radiologic differential diagnosis included circumscribed odontogenic keratocyst (usually accompanied by root resorption in bigger lesions), SBC, circumscribed giant cell granuloma, cementoma (periapical cemento-osseous dysplasia) 1 st stage, and radicular cyst (pulp vitality test is required for final diagnosis). The patient was subsequently scheduled for wisdom teeth extractions (#18, #28, #38, #48) at the Maxillofacial Surgery Department.

In April 2024, a posterior superior alveolar nerve block was performed for tooth #28, along with an inferior alveolar nerve block for tooth #38. Infiltration anesthesia of the surrounding soft tissues was also administered, using articaine hydrochloride 40 mg/mL and adrenaline 0.01 mg/mL, EP (Septanest 1.7 mL N50

– Septodont Co., Saint-Maur-des-Fossés, France). After confirming anesthesia, each tooth and the surrounding tissues were isolated with sterile gauze. A syndesmotome was used circumferentially around the cervical margin of the teeth to detach the alveolar-dental ligaments. Gentle luxation was then performed using a straight dental elevator to mobilize the teeth within their respective sockets. Once sufficient mobility was achieved, each tooth was grasped firmly with extraction forceps and moved along the path of least resistance using controlled buccolingual motions. Care was taken to avoid unnecessary trauma to adjacent teeth and alveolar bone. After extraction, the sockets were examined for residual root fragments or debris and irrigated with sterile saline. Hemostasis was achieved by applying gentle pressure with sterile gauze. A single 3-0 resorbable polyglactin 910 suture (VicrylTM, Ethicon Inc., Somerville, NJ, USA) was placed in the gingiva medial to the extracted maxillary and mandibular third molar sockets.

Thereafter, an incision was performed that extended from the distal papilla of the mandibular left first molar around the cervical areas of the teeth to the mesial papilla of the left mandibular canine, where a vertical releasing incision was performed at the mesial aspect of the canine. A two-sided subperiosteal flap was reflected and retracted in order to expose the underlying cortical bone, while respecting the mental nerve and foramen. A small cortical window was created in the lateral aspect of the alveolar bone, between the roots of the second premolar and first molar, using a sterile round bur No. 8 (Komet Dental, Gebr. Brasseler GmbH & Co. KG, Lemgo, Germany) on a surgical handpiece, under copious saline irrigation. Upon entry into the lesion, the bony cavity exhibited a complete absence of tissue or fluid, with the walls lacking any lining except for an exceedingly thin layer of connective tissue present in certain areas. After meticulous curettage, small bone fragments and portions of the membrane were sent for microscopic analysis. The operative findings strongly indicated a diagnosis of simple bone cysts; consequently, no additional treatment was deemed appropriate beyond the curettage procedure. Following the formation of a stable blood clot within the residual bone cavity, the mucoperiosteal flap was repositioned and secured with resorbable 4-0 polyglactin 910 sutures (Vicryl™, Ethicon Inc., Somerville, NJ, USA). The patient was then prescribed niflumic acid 250mg up to three times per day PRN, methylprednisolone 16 mg every 12 hours for 2 days, and amoxicillin/clavulanic acid (875/125) mg every 12 hours for 5 days. Finally, the patient was scheduled for regular follow-up to monitor healing and radiographic bone repair.

The patient had a mild paresthesia of the left mental nerve that resolved over time. The post-surgical period was, otherwise, uneventful. Although the patient was subjected to a regular follow-up program, she refused to undergo surgery on the right side of the mandible and remove the impacted third right maxillary molar, for the time being. One year postoperatively, a panoramic radiograph was obtained (Figure 3), and it demonstrated a significant reduction in the size of the radiolucent lesion in the left body of the mandible, indicating satisfactory bone healing. Evidence of progressive bone regeneration was observed, with restoration of regular trabecular pattern and cortical integrity. There was no alteration in the size of the radiolucent lesion in the right side of the mandible.

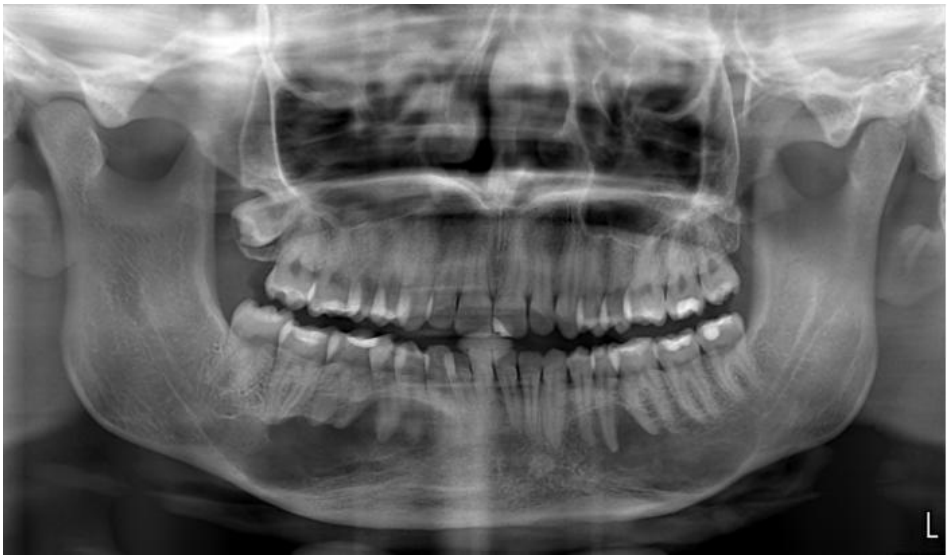

Figure 1: Preoperative panoramic radiograph, displaying two large radiolucent lesions with well-defined borders on the body of the mandible bilaterally, which extend from the canines to the mesial root of the third molars, below their respective apexes.

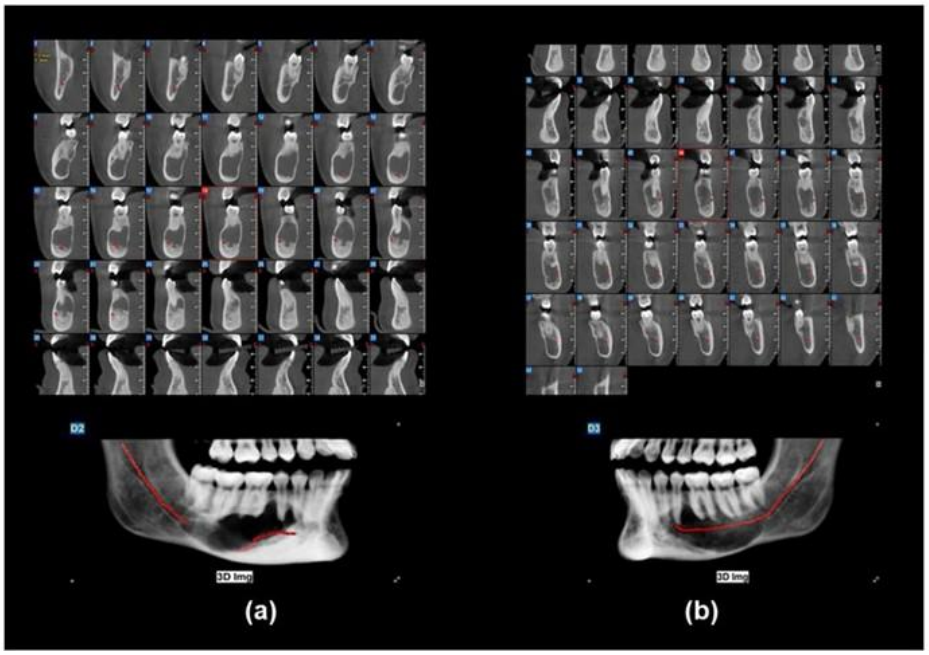

Figure 2: (a) right side (b) left side.

Cone Beam Computer Tomography scan (CBCT) confirmed the presence of two well-corticated and well-defined radiolucent lesions, with a size of 44mm x 13mm bilaterally. A slight buccal – lingual expansion of the cortical plates was observed, as well as thinning of the lingual cortical plate. The inferior alveolar nerve had also been displaced from both sides, and no root resorption of the teeth was detected.

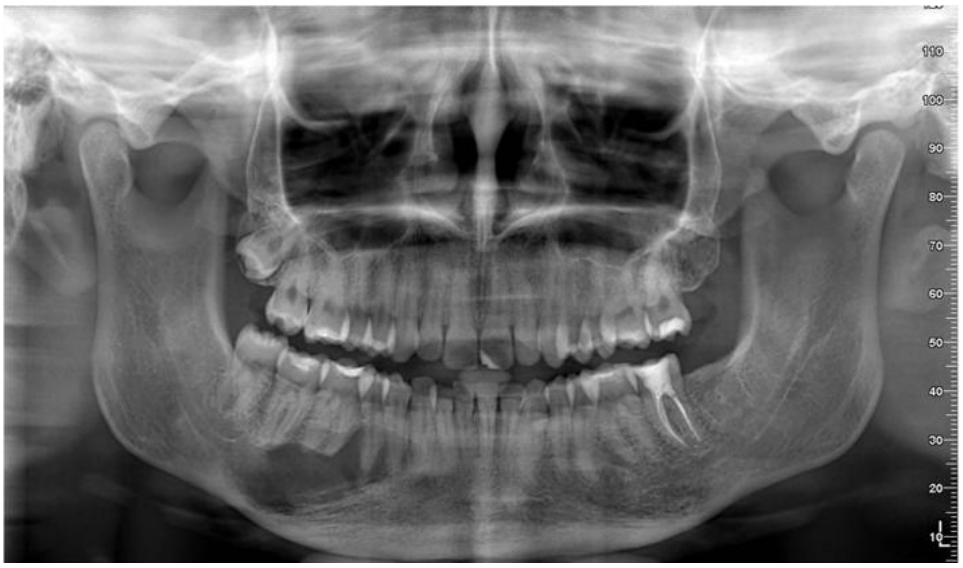

Figure 3: Postoperative radiograph, displaying a significant reduction in the size of the radiolucent lesion in the left body of the mandible, indicating satisfactory bone healing and ongoing regeneration. In contrast, the radiolucent lesion in the right body of the mandible remains untreated, as surgical intervention was deferred at the patient's request.

4. Discussion

This case report presents radiographic characteristics that may pose a challenge in the definitive diagnosis of the lesions. The bilateral localization of the cyst could potentially steer the clinician's mind further away from the diagnosis of SBCs, given that they are rare, only 0-1,25% of jaw cysts, and they are most frequently unilocular. [16] Based on this number, multiple SBCs comprise around 0.1375% of all cysts of the jaw, thus making them an exceptionally rare occurrence. This is the reason the final diagnosis of SBCs can be given only after thorough clinical, radiological, and histological examination. However, since material obtained from the histological examination may be scant or non-existent, it is often quite difficult for a definitive histologic diagnosis to be achieved. Based on the data obtained by the aforementioned methods, one can exclude the other entities in the differential diagnosis list and finalize the diagnosis. In this case report, the teeth were vital, and thus, the lesions could not be radicular cysts. Furthermore, the absence of root resorption excludes the diagnosis of odontogenic keratocysts, and the size of the lesions does not contribute to the diagnosis of 1st-stage cemento-fibrous dysplasia. A central giant cell granuloma will usually show evidence of internal bony septa, which the traumatic bone cyst generally lacks. Also, all true cysts have a more rounded, more "hydraulic" shape than do traumatic bone cysts.

It should, however, be noted that if a traumatic bone cyst is discovered accidentally while it is still small, it may be impossible to radiographically distinguish it from early forms of some of these lesions [17] In general, for asymptomatic radiolucent jaw lesions with an ambiguous clinical, laboratory, or imaging diagnosis, a biopsy is necessary to avoid misdiagnosing aggressive diseases as mere bone cysts.

In 2014, Seo-Young et al [18] reviewed the literature on multiple SBCs in the jaws and noted that, by that year, only 34 cases had been reported. The parameters they examined included the patient's age, sex, symptoms, history of trauma, vitality of the involved teeth, cavity contents, histology, treatment outcomes, number and location of lesions, and their radiographic appearance. Echoing Seo-Young et al.'s findings, our case also involves localization on the posterior mandibular body, female gender, and young age, characteristics shared by both multiple SBCs and unilocular lesions. The study revealed that these characteristics were consistently observed in all examined cases of multiple SBCs. In our case, the characteristic 'scalloping effect' was present, similarly to all previous 34 cases, serving as a distinct radiographic finding. Diverging from prior reports where almost half of the patients reportedly presented with fluid at aspiration, in our case, the cavity was completely empty.

Furthermore, in our case, no history of trauma was present, and the same applied to all those 34 cases. This could be interpreted by taking into consideration the fact that trauma rarely takes place bilaterally. The authors also stated that there may be a fundamental systemic factor in patients with multiple SBCs that predisposes them to vascular complications. However, no such thing was found in our patient's medical history. In all previously documented instances of multiple SBCs, the involved teeth were vital, and each patient exhibited only two SBCs. This observation was consistent in our case as well. A significant difference was observed in the reported frequency of bone expansion. In previous studies, bone expansion was noted in less than half of the patients and was typically associated with a multilocular radiographic appearance of the lesions. However, in all cases reported prior to 2012, the diagnosis of multiple SBCs relied primarily on 2D imaging techniques, such as panoramic and periapical radiographs, which have limited diagnostic accuracy, particularly in assessing bone expansion. This imaging limitation likely contributed to the underestimation of the frequency and extent of bone expansion in those cases. After CBCT, we identified a minor buccolingual expansion of the cortical plates, accompanied by thinning of the lingual cortical plate. However, the lesions did not display the previously mentioned multilocular radiographic presentation, thereby indicating that these findings are representative of a minority of cases.

In general, the lesion must be surgically explored to rule out the possibility that more dangerous lesions are not being addressed [19] Surgery reveals an empty bone cavity without an epithelial lining, and only normal bone or fibrous tissue can be curetted. Needle aspiration is usually unsuccessful, but when successful, only a few milliliters of straw-colored or serosanguinous fluid can be withdrawn, with minimal cellular components [20]. Usually, a gross specimen will be minimal in extent, although there may have been a relatively large cavity. Curettage of the bone walls is widely accepted as the gold standard for the management of SBCs of the jaws, as the goal is to promote the formation of a blood clot and subsequent bone repair. Furthermore, the use of methylprednisolone acetate, the application of Gelfoam (Pfizer, NY, USA), the grafting of allogenic bone combined with platelet-rich plasma, and intralesional injections that consist of a mixture of blood, porous hydroxyapatite, and bone fragments have been documented to yield favorable outcomes [21, 22]. Regardless of the selected therapeutic approach, follow-up for up to 3 years is recommended in order to ensure new bone formation and reduction of the bone cavity.

Recurrences are generally rare. According to Suei et al, [23]. Most recurrences occurred within 5 months of surgical treatment, and this group recommended that follow up should be continued until radiographic healing was confirmed within 3 years. It has been reported that recurrences were more common in cases of multiple SBCs than in solitary cases [24]. For example, in 1986, Campanacci et al. established that the frequency of recurrences was threefold higher in multilocular lesions than in unilocular lesions, and it was also twice as prevalent when the radiographic measurement exceeded 21 cm [25]. Large lesions with scalloping, as seen in this case report, may also hinder the formation of a blood clot sufficient for bone repair, thereby explaining their persistence or recurrence. Finally, recurrences are also reported to be more frequent in cases of lesions that are associated with cemento-osseous dysplasia [26].

5. Conclusion

In conclusion, SBCs are rare entities that are usually incidental radiological findings. Multiple SBCs are even rarer, and their presence may be associated with other conditions, such as cemento-osseous dysplasia. The etiology of SBCs is unclear, and they can remain asymptomatic for years. Their radiological features may vary; however, they should always be included in the differential diagnosis of radiolucent, unilocular, well-defined jaw lesions. The prognosis is good, and recurrences are rare.

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