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CASE REPORT

Recurrence of simple bone cyst: a case report and brief review of literature

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Abstract:

Background: Simple bone cyst (SBC) is an uncommon intraosseous lesion, often incidentally discovered on routine radiological examination. The etiology remains unclear, and a definite diagnosis is confirmed during surgical exploration. Recurrence rates have been controversial. Case Report: This article presents a case of a 13-year-old boy with SBC in the posterior left region of the mandible and a brief review of previously published cases of recurring SBC. The patient denied a history of trauma. Surgical exploration revealed an empty cystic cavity and the clinical diagnosis of the SBC was established. A follow-up radiographic examination two years later revealed significant regression of the lesion. However, the lesion recurred after 4 years. A new surgical approach was performed and the diagnosis of recurrence of SBC was again established. In addition to this case, a review of 12 cases of recurring SBC is presented here. Conclusion: Clinicians should ensure a long-term follow-up of these patients. The patient is in follow-up for 10 years after the first surgical intervention without signs of recurrence.

Keywords: Recurrence; jaws; simple bone cyst; idiopathic bone cavity; traumatic bone cyst; solitary bone cyst.

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INTRODUCTION

Simple bone cyst (SBC), also commonly known as traumatic, hemorrhagic or solitary bone cyst, is an uncommon intraosseous lesion classified as a pseudocyst due to the absence of an epithelium lining. The cavity is commonly filled with serous or bloody fluid with no evidence of inflammation[1,2]. The etiology and pathogenesis of SBC still remain unclear; however, some studies suggested that a traumatic event could cause intramedullary hematoma or ischemic vascular phenomenon and osteolysis secondary to altered bone metabolism[1-3]. The lesion is often incidentally discovered on routine radiological examination as it is commonly asymptomatic. The highest prevalence of SBC is in the second decade of life and there is no associated gender predilection. The most commonly involved sites include the long bones, with only 10% of the cases found in the maxillofacial region. The body of the mandible is usually affected in the premolar and molar regions[1,4].

Radiological examination reveals an unilocular radiolucent area, well circumscribed with a sclerotic margin, and rarely expands the cortical plates[5]. It is possible to penetrate the interdental space, described as a scalloped image radiolucent lesion[3].

Differential diagnosis is extremely important as SBC does not show any exclusive clinical and radiological features and should include odontogenic keratocyst, ameloblastoma, odontogenic myxoma, aneurysmal bone cyst, central giant cell lesion, and central hemangioma[1,5]. Although the clinical and radiographic features can aid in the diagnosis, they are generally not decisive. In most cases, the definite diagnosis is established with surgical findings based on the presence of an empty cavity[1,2,6].

The treatment of choice for SBC is surgical exploration, in which the bony cavity is curetted till the onset of bleeding[7]. Other modalities of treatment have been proposed, such as packing the curetted cavity with autologous blood, autologous bone, and platelet-rich plasma (PRP). Additionally, previous studies have also shown cases of SBC with spontaneous resolution[6,8]. Clinical and radiological follow-up postoperatively is mandatory to ensure complete healing after 6-12 months. Rates and time to recurrence are variable; nevertheless, some cases have been documented recurring even within three years after surgery[4,8].

This article presents a case of SBC in the posterior mandible without any previous history of trauma, and with recurrence after four years of follow-up.

CASE REPORT

A 13-year-old boy was referred for an orthodontic evaluation and an extensive radiolucent area in the posterior mandible was observed during routine radiographic examination. The medical history was not contributory, and the patient denied history of any dentoalveolar trauma. However, the patient was a Muay Thai (Thai boxing) fighter. Extraoral examination revealed no facial asymmetry, no tenderness on palpation, and no lymphadenopathy.

On intraoral examination, the overlying mucosa in the mandibular posterior region appeared normal. There was no pain or any other jaw-related symptom, and the teeth adjacent to the lesion were all vital. A panoramic radiograph showed a large unilocular radiolucency with thin corticated borders in the posterior left side of the mandible, extending to the ascending ramus and angle. The margins of the lesion were slightly irregular and partly well-defined with radiopaque margin. Following this, a cone-beam computed tomography (CBCT) scan was done. Axial slices of the CBCT showed a hypodense lesion in the posterior mandible extending to the second molar with thin corticated borders and preservation of the cortices. The lesion was 4.2 × 3.8 × 1.6 centimeters in size and slight expansion of the buccal cortical plate was observed (Figure 1). In addition, the patient no had resorption or displacement of any tooth root. The differential diagnosis included odontogenic keratocystic, unicystic ameloblastoma, and SBC.

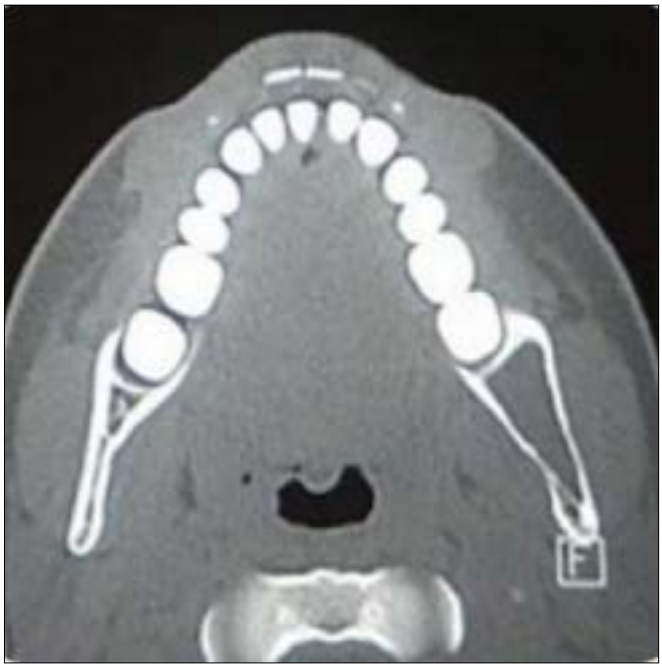

Figure 1. Axial view of the lesion in CBCT image. Note the hypodense lesion in the posterior mandible extending to the second molar with preservation of the cortices.

An exploratory surgery was performed under local anesthesia (mepivacaine hydrochloride with epinephrine) with inferior alveolar, buccal and lingual nerve block, in addition to infiltration in the mandibular branch. An aspiration attempt was made before the incision with negative punction. The bony cavity was completely empty of tissue or fluid (Figure 2) and a careful curettage walls was performed. These surgical findings were highly suggestive of SBC and the final diagnosis was established. Postoperative healing was uneventful. A follow-up panoramic radiograph was performed annually revealing significant regression of lesion after two years (Figure 3).

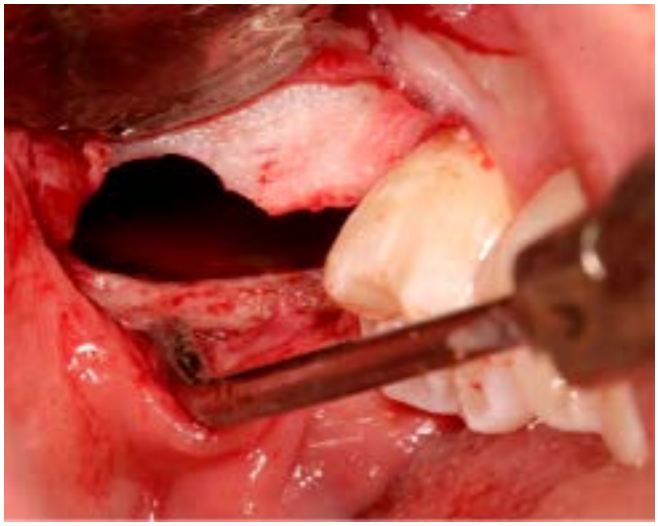

Figure 2. Clinical appearance after surgical exposure of the lesion exhibiting a voided cavity without a soft tissue lining

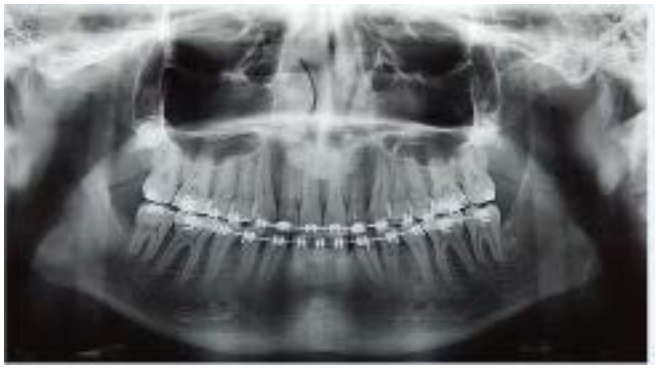

Figure 3. Panoramic radiograph showing resolution of the lesion with the bone formation two years after treatment.

However, four years later, follow-up radiographic examination revealed a unilocular lesion in the same region as previously seen on CBCT: a hypodense, well-defined lesion, sizing 4.3 × 3.5 × 1.3 centimeters approximately (Figure 4). There was no expansion of cortical bone or clinical signs, and no symptoms were reported by the patient. A new surgery with the same technique used in the first procedure was performed. An empty cavity was found again, confirming the diagnosis of recurrence of SBC. Bleeding was caused by bone perforations to ensure filling of the entire cavity with the fluid.

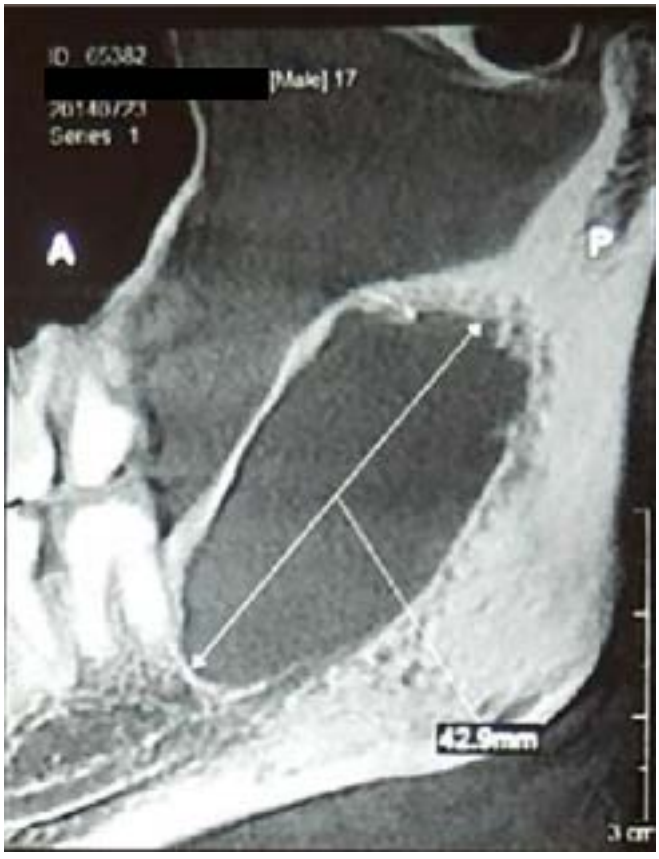

Figure 4. Recurrence of the lesion for 4 years after first operation.

Postoperative healing was uneventful, and follow-up panoramic radiograph indicated bone formation at approximately seven months postoperatively. A panoramic reconstruction of CBCT image ten years later of the first surgical exploration showed extensive bony filling in the mandible (Figure 5). In addition, the tooth 37 remained responsive to pulp vitality tests after the surgical treatments. The patient still remains under careful follow-up and till now there is no sign of recurrence.

Discussion and review of the literature

A literature search of articles published in English, using PubMed database was performed. The term "simple bone cyst" and its synonyms (traumatic bone cyst, hemorrhagic/hemorrhagic bone cyst, solitary bone cyst and idiopathic bone cyst/cavity) were searched in combination

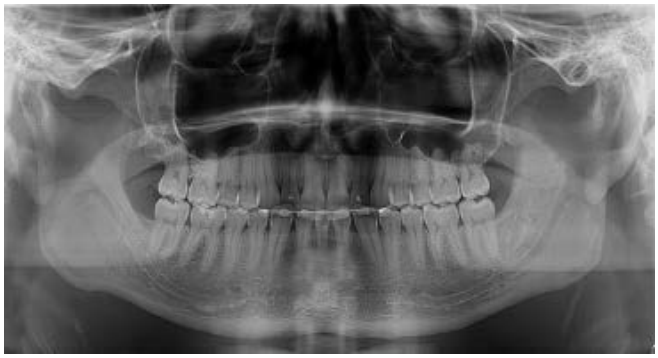

Figure 5. A panoramic reconstruction of CBCT image at 10 years later of the first surgical exploration showing remarkable bone repair.

with "mandible or jaws" and "recurrence." The relevant titles and abstracts were evaluated and the articles that were available for reading were read in full. We collected and examined the number of cases, patient demographic, clinical, and radiographic presentation, including age, sex, location, history of trauma, vitality of the involved teeth, treatment, healing (time), number of cases with recurrence, average time to recurrence, location, radiographic findings (scalloped margin, multilocular pattern and bony expansion). This search yielded 12 articles including case reports, case series and reviews, in which, a total of 1261 cases of SBC and 90 recurrences were presented[9-18].

This study aimed to report one case of SBC with recurrence after a long follow-up period and review previously published cases that reported recurrence of the lesion. The pathogenesis of SBC remains an enigma which explains the different names for the same lesion[4,19,20]. However, intraosseous hemorrhage due to trauma is the most widely discussed etiological factor[12,21-24]. Few cases from our literature review reported a history of trauma, as shown in Table 1. Although our patient had no history of trauma, this theory may be having been an important factor in the development of SBC in the present case as the patient was a martial arts practitioner and may have suffered some previous traumatic event in the maxillofacial region in this sport. The SBC can be detected at any age; however, most cases present in young patients, as seen in the present case. Moreover, the theory of trauma in the etiopathogenesis of SBC seems to explain this higher rate of occurrence in young patients, due to the greater probability of occurrence of trauma in this age group[13-21].

According to previous reports, SBCs have no gender predilection; however, a high female predilection was shown in our literature review (Table 1). This finding was in agreement with the research conducted by Suei et al. (2007) and Flores et al. (2017). Another aspect of the present case is the location of the lesion; the mandibular branch, one of the rare anatomical sites of involvement, as well as angle and the condylar process (Table 1)[13]. SBC is often seen in the premolar-molar area of the mandible. Given the asymptomatic nature of the lesion[22-27], it is often an incidental finding on radiographs performed for other reasons[20,24,25]. In symptomatic cases, pain is the most commonly reported finding. In addition, paresthesia, tooth sensitivity and jaw fractures have also been reported; however, the occurrence is rare[28]. In the present case, the radiographic examination of the patient was performed due to orthodontic evaluation, similar to other cases in the literature.

The SBC should be included in the differential diagnosis of radiolucent lesions of the jaws, such as, odontogenic keratocystic, ameloblastoma, odontogenic myxoma, and central giant cell lesions. In the present case, based on the clinical and radiographic features, including the age of the patient and anatomic site of the lesion, some diagnostic hypotheses were drawn[19,23]. Odontogenic keratocystic and unicystic ameloblastoma was considered as these lesions commonly manifest in young patients, have a male predilection, and can have similar radiographic features[1,5,12]. Because of the radiographic features on simple imaging, advanced imaging such as computed tomography is essential to establish an accurate diagnosis[26,27]. In our patient, the final diagnosis was established during exploratory surgery based on the presence of an empty cavity. Therefore, this approach was essential for diagnosis and to differentiate the entity from other lesions with similar presentation, as well as for choosing the appropriate course of treatment[24], since a diagnosis based only on clinical and radiographic findings may subsequently lead to more aggressive treatments and thus may result in further complications.

The choice of therapeutic treatment included surgical exploration with careful curettage of the bone walls and the filling of the cavity with blood[29]. According to Homem de Carvalho et al. (2010), the technique should be minimally invasive with access small enough to view the cavity and allow curettage. More aggressive procedures can promote more inflammatory reactions and longer repair times[20]. Because SBC may show spontaneous cure, there are some authors who do not recommend surgical intervention due to the associated morbidity[1] or recommend treatments involving only aspiration of the cavity or surgical exploration without curettage[4]. In addition, treatment options for SBC include filling of the cavity with bovine lyophilized bone or the introduction of autologous blood with bone from the patient or hydroxyapatite, especially in cases of

Table 1. Demographic, clinical, and radiographic presentation of selected SBC cases with emphasis in the recurrence.

Author (Year)	Number of cases	Location	Sex	Age	History of trauma	Vitality of teeth	Clinical presentation	Radiographic presentation	Treatment	Healing (time)	Number (or %) of cases with recurrence	Average time to recurrence
Baqain et al. (2005)	1	Posterior mandible bilaterally	F	50	No	Yes	Asymptomatic, swelling, buccal and lingual expansion	Scalloping, multilocular, mostly well-defined without a sclerotic margin.	Surgical intervention with curettage	7 months	1	2,5 years
Horne et al. (2014)	1	Mandible bilaterally	F	41	Not stated	Yes	Mild buccal expansion and mild tenderness to palpation	Unilocular radiolucent lesions	Surgical intervention	Radiolucencies near 19 and 27 never fully healed	1	4 years
Peacock et al. (2015)	20	Posterior mandible	F	Mean: 30	1 yes	Not stated	Asymptomatic	Unilocular lesions, scalloped borders	Surgical intervention with curettage	Not stated	1	3 years
You et al. (2017)	27 patients: 30 SBCs	Posterior mandible	F	Mean: 29.5	2 yes	Not stated	Asymptomatic	Scalloping and erosion of the mandibular border	Surgical intervention with curettage	Not stated	1	2 years
Suei et al. (2007)	Jaw bone: 881	Jaw bone and extracranial	No sex predilection	Not stated	Not stated	Not stated	Asymptomatic.	Solitary, cyst-like radiolucency	Surgical intervention with curettage of the bone wall	Not stated	2%	No stated
Suei et al. (2007)	132	Not stated	Not stated	Not stated	Not stated	Not stated	Not stated	Not stated	Surgical intervention, with curettage of the bone	12 to 17 months	34	Mean: 3 years
Suei et al. (2010)	139	Not stated	F	Mean:21.1	Not stated	Not stated	Not stated	One group: intact lamina dura, smooth margin and no/ smooth bone expansion Other group: absent lamina dura, root resorption, scalloped margin, nodular expansion, radiopaque mass or multiple lesions	Surgical intervention with curettage of the bone, followed by exploration of the cavity	Not stated	37	Not stated
An et al. (2014)	34	Posterior mandible	F	Mean:39.3	2 yes	20 yes 1 no 13 not stated	Asymptomatic	Scalloping	Surgical intervention	4 months to 5 years	3	2 to 3 years
Feinberg et al. (1984)	1	Right mandibular body	F	37	No	Yes	Asymptomatic	Multilocular	Surgical intervention More extensive surgical procedure Injection with venous blood	Not stated	3	29 months 6,2 years 8 years
Horner et al. (1988)	2	Right Mandible	F M	13 9	No	1 no 1:not stated	Asymptomatic	Large radiolucency	Surgical intervention	Not stated	2	18 months 3 years
Kuttenberger et al. (1992)	1	Right condyle	M	10	Yes	Not stated	Swelling in the preauricular and parotid area	Radiolucency	Surgical resection and immediate reconstruction	Not stated	1	2 years
MacDonald--Jankowski (1995)	20	Mandible	F	Mean 33.1	1 yes	9 yes	Asymptomatic	Well- defined unilocular oval-shaped radiolucencies	Surgical intervention	Not stated	6*	Not stated
Our case	1	Posterior mandible	M	13	No	Yes	Asymptomatic	Large unilocular radiolucency	Surgical intervention with curettage	24 months	1	4 years

SBC, simple bone cyst, * six recurrences in the only patient

failure of the conventional approach[22-24]. Nevertheless, according to a recent systematic review, curettage is the first choice of treatment for SBC[4]. In the present case, treatment by surgical exploration with curettage, and continuous follow-up radiographic evaluation indicated bone regeneration. However, recurrence occurred even with initial bone formation after surgery.

In our brief review of 12 papers, 90 cases of recurrence were reported, out of which, some even occurred several times in the same patient. We are adding one case to our literature review. Kuroi (1980) reported the recurrence rate of SBC to be less than 2%11; however, these rates appear to be higher in other studies[10]. The highest rate found (26%) was in the review performed by Suei et al. (2007), probably because those cases had multiple lesions like cemento-osseous dysplasia, which also has relatively high recurrence rates. The healing time of the lesion as described in Table 1 varies from months to years; however, an average of 3.5 years is estimated for recurrence, which may explain the low rates due to loss of follow-up of cases[10-13]. Our case of recurrence after four years of healing also supports this. Although our case did not demonstrate complete repair of the lesion until the time of recurrence, the case described by Horne et al. 2014 with multiple SBCs also showed this behavior. Recurrence was considered even in the areas that never fully healed, as they were extensive lesions. Thus, the authors suggest annual and long-term follow-up until complete healing has been confirmed by radiographic examination.

Finally, if SBC is diagnosed early and treated with curettage, it presents a good prognosis and lower morbidity. However, due to the possible variations in prognosis, rigorous and continuous clinical and radiographic follow-up is strongly recommended, mainly in multiple, large lesions with scalloped margins and nodular bone expansion. Some of these clinical features were observed in the present case, which could explain the recurrence of the lesion. Moreover, SBC associated with other diseases such as florid cemento-osseous dysplasia and benign fibro-osseous lesions, may contribute to lesion recurrence[12,24]. The adjacent teeth generally remain vital, as observed in this patient, therefore, non-vitality does not appear to be a cause for occurrence of the lesion[22].

CONCLUSION

The case reported contributes to dentist knowledge about an incidentally detected condition with emphasis on the recurrence of the lesion. In addition, the case still warns orthodontists who, during treatment planning, may encounter this lesion. Hence, the importance of radiographic examination and cooperation among professionals responsible for patient care. This study demonstrated that the recurrence rates of the simple bone cyst are very variable, and we emphasize that may also be attributed to bias in follow-up time. Due to, we strongly recommend long-term follow-up such as the case presented with 10 years of continuous monitoring.

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