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Simple Bone Cysts of the Jaws: Case Series and Literature Review

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Abstract **Original Research Article**

Simple bone cysts (SBCs) of the jaws are benign intraosseous cavities empty or filled with serous, serohematic or blood fluid. They are characterized by the absence of an epithelial lining. ***Aim*:** To describe the clinical, radiographic and surgical findings of SBC through a series of 4 clinical cases and a literature review of previously published cases. ***Methods*:** A series of 4 case reports of our department were reported with presentation of the clinical, radiological and surgical investigations performed. In addition, a literature search of SBCs was conducted using the PubMed database for a period of 10 years. ***Results*:** 19 papers were returned; A total of 34 patients (including our 4 patients) with a total of 40 lesions were analyzed. SBC are generally observed around the second decade of life (68%) without gender predominance. They are preferentially located in the mandible and are frequently asymptomatic and of fortuitous discovery (82%). Radiologically, SBCs can be unilocular or multilocular with a characteristic scalloped appearance between the roots of the teeth. The management of these pseudocysts is based on surgical treatment. ***Discussion*:** Knowledge of these different characteristics of SBCs is essential to make the right diagnosis and to provide the best management, especially as these lesions may have similarities with other lesions considered more aggressive.

Keywords: Simple bone cyst, empty cavity, surgical intervention.

INTRODUCTION

Simple bone cysts (SBC) are intraosseous cavities empty or filled with serous, serohematic or blood fluid without epithelial lining [1].

These lesions, classified by the WHO 2017 as intraosseous pseudo-cysts [2], can be found in the jawbones with a percentage of 1% of all jaw cysts [3]. Usually occurring during the second decade of life, they preferentially affect the lateral parts of the mandibular body and exceptionally the maxillae [4]. The etiopathogenesis of SBC remains poorly understood [4]. Usually asymptomatic, these cysts are discovered accidentally during a routine X-ray examination [3]. Nonetheless, pain, hypoaesthesia of the chin or even a deformity can be revealing signs. Of different clinical and radiological presentation, the conventional management of these SBCs consists of surgical exploration of the lesions, but other combined treatments can be proposed.

The aim of this study was to describe, through a series of four clinical cases, the clinical, radiographic

and surgical results of solitary bone cysts and to highlight through a literature review of recent publications the SBC features and the latest proposed treatment modalities.

METHODS

A series of four clinical cases of SBCs (Three males and one female aged respectively 18, 22, 21 and 17) followed and treated at the Department of Dental Medicine of the Military Principal Hospital of Instructions of Tunis, Tunisia, were reviewed.

We retrospectively identified these 4 patients with a suspected diagnosis of SBCs based on clinical and radiological analysis.

To be eligible, the diagnosis of SBCs in these patients was confirmed following surgical exploration of the lesions, which showed the presence of an empty endosseous cavity devoided of epithelial lining.

The folders of these patients were reviewed and the various clinical, radiological and treatment data as well as the follow-up period and the presence of a possible recurrence were collected.

Patient's medical and family histories were non-contributory. Two of these patients were initially referred by a colleague for the evaluation of a radiolucent image discovered fortuitously during a routine radiological examination and the two others patients consulted us for the extraction of a lower wisdom tooth which presented episodes of recurrent pericoronitis and radiolucencies were discovered incidentally on the panoramic.

Additionally, a literature search of articles was conducted using the PubMed database for a period of 10 years going from 2012 to 2021. The following keywords; “simple bone cyst”, “solitary bone cyst”, “idiopathic bone cavities”, “traumatic bone cyst”, “unicameral bone cyst” were used in combination with “Jaws”.

The number of cases collected from the literature review plus our 4 clinical cases determined the sample size.

These different parameters were collected and examined for the total number of patients: number of lesions, epidemiological data (age and gender), chief complaint at presentation, location of lesions, history of trauma, vitality of the involved teeth, radiological Data, Surgical management, outcomes of treatment and follow-up.

RESULTS

The PubMed research led to the analyze of 19 papers. Including our 4 cases, a total of 34 patients were analyzed.

For the epidemiological data, SBC were mainly observed in Young Subjects, around the second decade of life (68%) followed by the 3rd decade (21%). The first, fourth and fifth decades were in the minority, representing one, one and two cases respectively (Table I).

No gender predominance was noted (17 females against 16 male). The gender was not reported in one patient (Table I).

Regarding clinical features, SBC are usually asymptomatic (82%, including our 4 cases), but slight swelling (6%) or mild pain (12%) may present the chief complaint of the patients (Table I) (Figures 1, 2, 3, 4).

Previous history of trauma was noted in only 6 patients (Table I). Pulp vitality tests were positifs in almost all the involved teeth (Table I).

A total of 40 lesions were included in the study. Six patients had two SBCs in two different locations, which may occur simultaneously or asynchronously (Table II).

SBC preferentially affect the mandible and exceptionally the maxilla. In fact, all SBCs of our sample were of mandibular location with the posterior mandibular body as the most common location (50%) followed by the symphyseal region (17.5%), the ramus (15%), the ramus and the posterior mandibular body (12.5%), the ramus and the condyle (5%) (Table II).

SBCs occurred in the posterior mandibular body in our two first patients, in the symphyseal region in case 3 and in the ramus in case 4 noted on radiographs (Figures 5, 6, 7).

The most common radiological features (panoramic radiograph and mandibular CT scan) found were, a well limited radiolucency, no root displacement or resorption, expansion and thinning of cortices generally without perforation and a specific scalloped aspect between the roots of teeth (Figures 8, 9, 10, 11).

SBCs may be unilocular or multilocular with a predominance of unilocular ones (Table II). The multilocular presentation was noted in our case 4 (Figure 11).

The management of SBC was surgical for all lesions which showed the presence of an empty cavity without epithelial lining (Figures 12, 13, 14, 15) (Table III).

The follow-up periods ranged from 3months to 6 years and the recurrence was noted in only one case (Table III) (Figure 16).

Fig 1: Patient 1: Absence of intraoral swelling

Fig 2: Patient 2: slight buccal expansion opposite the 36-37 covered by a normal-looking mucosa

Fig 3: Patient 3: absence of swelling

Fig 4: Patient 4: a slight vestibular swelling

Fig 5: Patient 1 panoramic radiograph showing an Osteolytic image well limited by a radiopaque border in the right horizontal branch

Fig 6: Patient 2 panoramic radiograph showing an Osteolytic image well limited in the left horizontal branch

Fig 7: Patient 4 panoramic radiograph showing a clearly limited multilocular radiolucency located in the right ramus

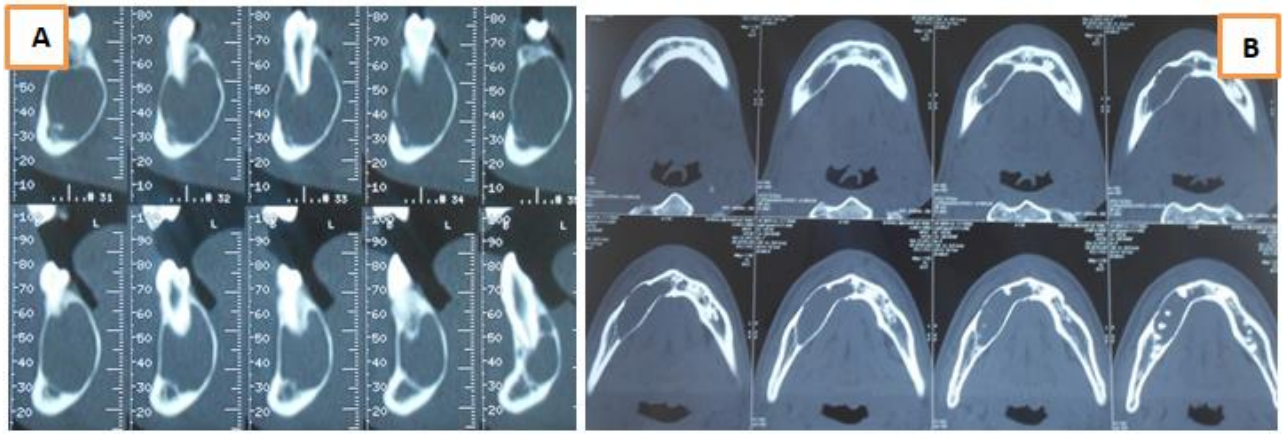

Fig 8: Patient 1: Mandibular CT scan showing integrity of the bony cortices and repression of the mandibular canal. A: oblique coronal sections, B: Axial sections

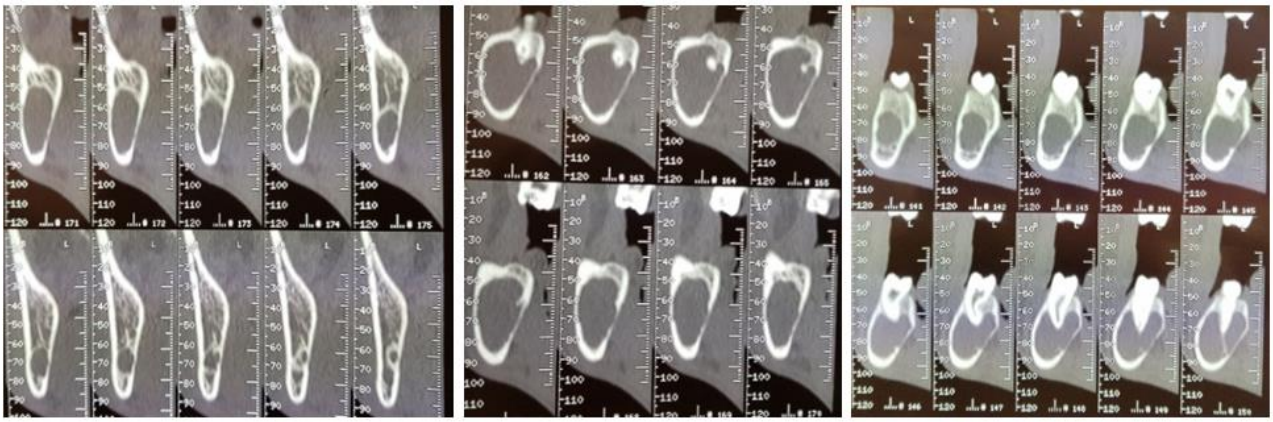

Fig 9: Patient 2: CT examination of the mandible: hypodensity filling the entire horizontal branch with repression of the mandibular canal towards the lingual cortex. The dental roots appear to be directly embedded in the lesion

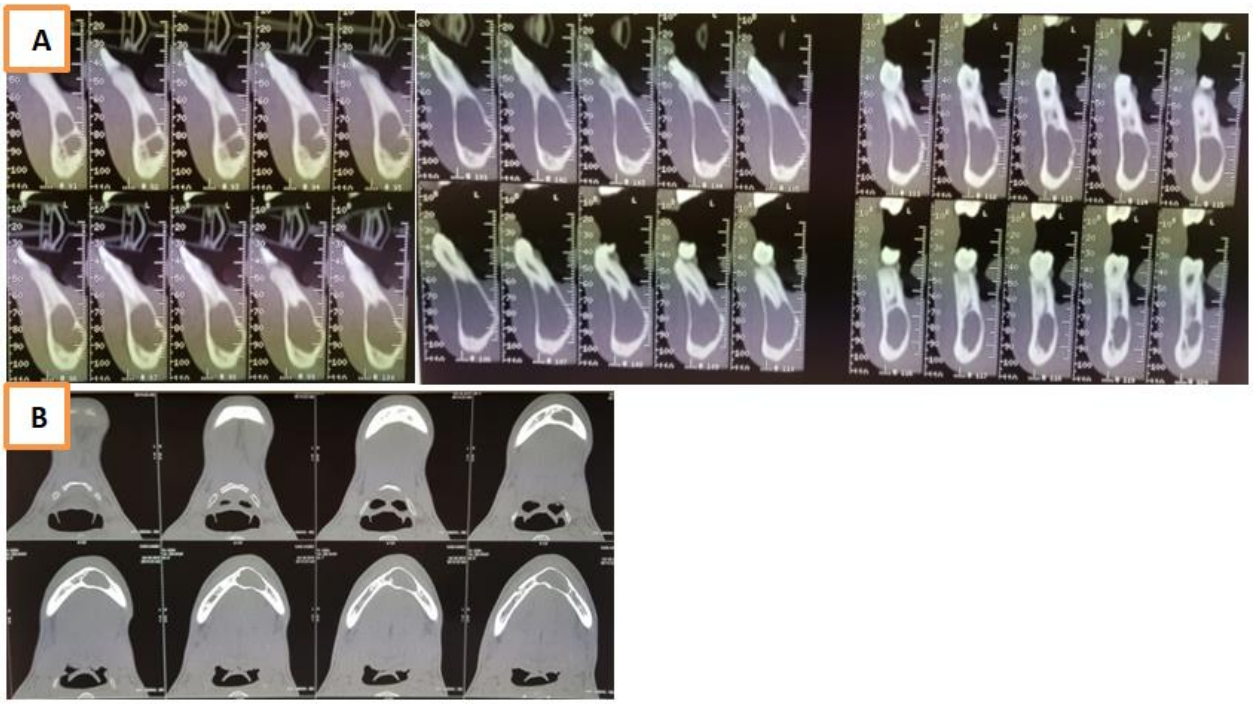

Fig 10: Patient 3 mandibular CT scan. A: Oblique coronal sections. B: Axial sections Well limited radiolucency located in the symphyseal region. Absence of cortical deformation

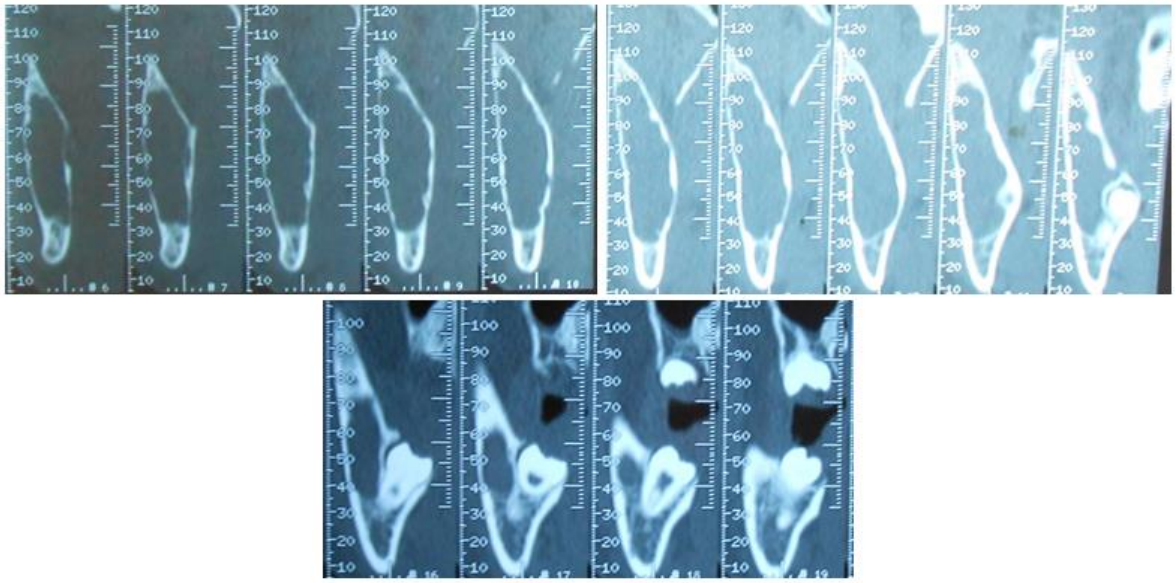

Fig 11: Patient 4 Mandibular CT: a well circumscribed multilocular radiolucency. Note the lack of continuity with the crown of the tooth 48

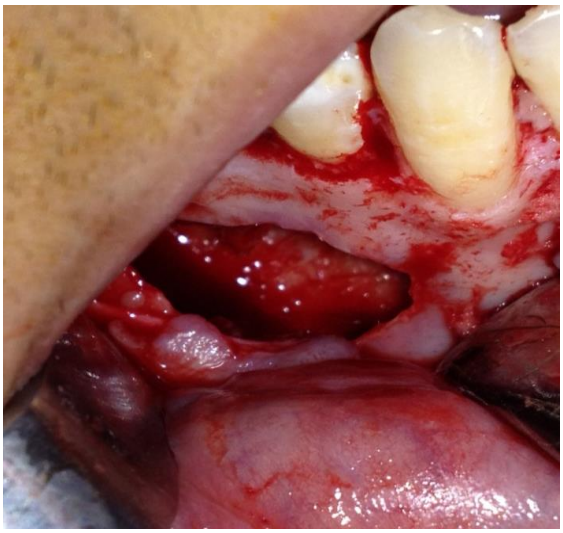

Fig 12: Patient 1 surgical management: empty cavity + absence of cystic walls

Fig 13: Patient 2 surgical management: empty cavity + absence of cystic walls

Fig 14: Patient 3 surgical managment: empty cavity

Fig 15: Patient 4: Intraoperative aspect: empty cavity

Fig 16: Patient 4: Control at 6 months: beginning of bone formation

Table I: Summary of clinical features. Literature revue including our 4 cases

	Author/ year	Age	Sex	Chief complaint	History of trauma	Viatlity of the involved teeth
1	DINCER *et al*., 2012 [6]	16	M	RR	+	-
2	MATHEW *et al*., 2012 [7]	15	F	Pain +swelling	-	NM
3	DE SOUZA NORONHA *et al*., 2012 [8]	17	F	RR	NM	+
4	HATAKEYAMA *et al*., 2012 [9]	6	M	RR	-	NM
5	SABINO-BEZERRA *et al*., 2013 [10]	16	M	RR	NM	+
6	SABINO-BEZERRA *et al*., 2013 [10]	19	F	RR	NM	No contact with the teeth
7	SABINO-BEZERRA *et al*., 2013 [10]	15	M	RR	NM	+
8	SABINO-BEZERRA *et al*., 2013 [10]	13	M	RR	NM	+
9	SABINO-BEZERRA *et al*., 2013 [10]	45	F	RR	NM	+
10	SABINO-BEZERRA *et al*., 2013 [10]	37	F	RR	NM	NM
11	SHAH *et al*., 2013 [11]	18	NM	RR	+	+
12	MADIRAJU *et al*., 2014 [3]	12	M	RR	-	+
13	SATISH *et al*., 2014 [12]	15	M	pain	NM	NM
14	SATISH *et al*., 2014 [12]	21	M	pain	NM	+
15	AN *et al*., 2014 [13]	11	F	RR	-	+
16	AN *et al*., 2014 [13]	17	M	RR	-	+
17	AN *et al*., 2014 [13]	18	M	RR	-	+ except 45
18	NAGORI *et al*., 2014 [14]	22	F	RR	NM	+
19	HORNE *et al*., 2014 [15]	41	F	RR	NM	+
20	CHELL *et al*., 2015 [16]	16	M	Fractured mandible	+	NM
21	PROVOST *et al*., 2016 [17]	17	M	Pain	NM	No contact with the teeth
22	PROVOST *et al*., 2016 [17]	16	F	RR	NM	NM
23	ZHANG *et al*., 2017 [18]	16	M	RR	-	No contact With the teeth
24	AOKI *et al*., 2018 [19]	22	F	RR	NM	No contact with the teeth
25	RAZMARA *et al*., 2019 [20]	13	F	RR	NM	+
26	RAZMARA *et al*., 2019 [20]	14	F	pain	+	+
27	RAZMARA *et al*., 2019 [20]	23	F	RR	NM	NOT RELIABLE RESPONSE
28	EMERICH *et al*., 2019 [21]	14	F	RR	+	NM
29	PÉREZ-IGLESIAS *et al*., 2021 [22]	16	M	RR	-	No contact with the teeth
30	BERNABEU-MIRA *et al*., 2021 [23]	22	F	RR	+	+
31	Our case 1	18	M	RR	NM	+
32	Our case 2	22	M	RR	NM	+
33	Our case 3	21	M	RR	NM	+
34	Our case 4	17	F	RR	NM	No contact with teeth

RR: routine radiograph, NM: not mentioned, M: male, F: female

Table II: Characteristics of SBCs. Literature revue including our four cases

Case No	Author/year	No lesions	Location of lesions	Radiographic findings
1	DINCER *et al*., 2012 [5]	1	PMB	Unilocular
2	MATHEW *et al*., 2012 [6]	2	1 : PMB+R 2 : PMB	Expansion, thinning, scalloping
3	DE SOUZA NORONHA *et al*., 2012 [7]	2 (asynchronous)	1 : PMB 2 : R	Unilocular, scalloping
4	HATAKEYAMA *et al*., 2012 [8]	1	R+C	Unilocular
5	SABINO-BEZERRA *et al*., 2013 [9]	1	PMB+R	Scalloping, expansion, perforation of the lingual cortex
6	SABINO-BEZERRA *et al*., 2013 [9]	1	R	Unilocular, scalloping
7	SABINO-BEZERRA *et al*., 2013 [9]	1	PMB+R	Multilocular, expansion
8	SABINO-BEZERRA *et al*., 2013 [9]	1	PMB	Unilocular, scalloping
9	SABINO-BEZERRA *et al*., 2013 [9]	1	PMB	Multilocular, expansion
10	SABINO-BEZERRA *et al*., 2013 [9]	1	PMB	Unilocular
11	SHAH *et al*., 2013 [10]	1	SR	Unilocular, scalloping
12	MADIRAJU *et al*., 2014 [3]	1	SR	Unilocular, scalloping
13	SATISH *et al*., 2014 [11]	1	PMB	Multilocular, expansion, scalloping, thinning
14	SATISH *et al*., 2014 [11]	1	PMB	scalloping
15	AN *et al*., 2014 [12]	2	1: PMB 2: SR	Unilocular, thinning, expansion
16	AN *et al*., 2014 [12]	2	1: PMB 2 : PMB	Unilocular, Thinning
17	AN *et al*., 2014 [12]	2	1: SR 2: PMB	Root resorption, expansion
18	NAGORI *et al*., 2014 [13]	1	PMB	Multilocular, Scalloping, expansion
19	HORNE *et al*., 2014 [14]	2	1 : PMB 2: PMB	Expansion
20	CHELL *et al*., 2015 [15]	1	PMB	Unilocular
21	PROVOST *et al*., 2016 [16]	1	R	Multilocular, expansion, thinning
22	PROVOST *et al*., 2016 [16]	1	PMB+R	Multilocular, scalloping, thinning
23	ZHANG *et al*., 2017 [17]	1	R+C	Unilocular , scalloping, thin mandibular cortex
24	AOKI *et al*., 2018 [18]	1	PMB	Unilocular , expansion
25	RAZMARA *et al*., 2019 [4]	1	SR	Unilocular, expansion
26	RAZMARA *et al*., 2019 [4]	1	PMB+R	Unilocular, expansion, thinning, scalloping
27	RAZMARA *et al*., 2019 [4]	1	PMB	Unilocular
28	EMERICH *et al*., 2019 [19]	1	R	Unilocular , cortical bone thinner than normal
29	PÉREZ-IGLESIAS *et al*., 2021 [1]	1	R	Unilocular, expansion
30	BERNABEU-MIRA *et al*., 2021 [20]	1	SR	Unilocular, Scalloping
31	Our case 1	1	PMB	Unilocular, scalloping
32	Our case 2	1	PMB	Unilocular, thinning, Scalloping
33	Our case 3	1	SR	Unilocular, scalloping
34	Our case 4	1	R	multilocular, thinning, expansion

PMB: posterior mandibular body, SR: symphyseal region, R: Ramus, C: Condyle.

Table III: Management and follow-up. Literature revue including our four cases

Case No	Author/year	Treatment	Follow-up period	recurrence
1	DINCER *et al*., 2012 [5]	Surgical intervention	5 months	no
2	MATHEW *et al*., 2012 [6]	Surgical intervention	3 months	no
3	DE SOUZA NORONHA *et al*., 2012 [7]	Surgical intervention	5 years	no
4	HATAKEYAMA *et al*., 2012 [8]	Surgical intervention	2 years	no
5	SABINO-BEZERRA *et al*., 2013 [9]	Surgical intervention	1 year	no
6	SABINO-BEZERRA *et al*., 2013 [9]	Surgical intervention	2 years	no
7	SABINO-BEZERRA *et al*., 2013 [9]	Surgical intervention	3 years	no
8	SABINO-BEZERRA *et al*., 2013 [9]	Surgical intervention	2 years	no
9	SABINO-BEZERRA *et al*., 2013 [9]	Surgical intervention	6 months	no
10	SABINO-BEZERRA *et al*., 2013 [9]	Surgical intervention	6months	no
11	SHAH *et al*., 2013 [10]	Surgical intervention	NM	NM
12	MADIRAJU *et al*., 2014 [3]	Surgical intervention	24 months	no
13	SATISH *et al*., 2014 [11]	Surgical intervention	NM	NM
14	SATISH *et al*., 2014 [11]	Surgical intervention	NM	NM

Case No	Author/year	Treatment	Follow-up period	recurrence
15	AN *et al*., 2014 [12]	Surgical intervention	2 years	no
16	AN *et al*., 2014 [12]	Surgical intervention	NM	NM
17	AN *et al*., 2014 [12]	Surgical intervention	3 months	no
18	NAGORI *et al*., 2014 [13]	Surgical intervention	1 year	no
19	HORNE *et al*., 2014 [14]	Surgical intervention	6 years	yes
20	CHELL *et al*., 2015 [15]	Surgical intervention	6 months	no
21	PROVOST *et al*., 2016 [16]	Surgical intervention	1 year	no
22	PROVOST *et al*., 2016 [16]	Surgical intervention	36 months	no
23	ZHANG *et al*., 2017 [17]	Surgical intervention	18 months	no
24	AOKI *et al*., 2018 [18]	Surgical intervention	NM	NM
25	RAZMARA *et al*., 2019 [4]	Surgical intervention	6 months	no
26	RAZMARA *et al*., 2019 [4]	Surgical intervention	6 months	no
27	RAZMARA et al. /2019 [4]	Surgical intervention	6 months	no
28	EMERICH *et al*., 2019 [19]	Surgical intervention	6 months	no
29	PÉREZ-IGLESIAS *et al*., 2021 [1]	Surgical intervention	6 months	no
30	BERNABEU-MIRA *et al*., 2021 [20]	Surgical intervention + filling the defect with A-PRF+ membranes	6 months	no
31	Our case 1	Surgical intervention	Patient lost to follow-up	NM
32	Our case 2	Surgical intervention	Patient lost to follow-up	NM
33	Our case 3	Surgical intervention	6 months	No
34	Our case 4	Surgical intervention	6 months	No

NM: not mentioned

DISCUSSION

The simple bone cyst is a relatively common lesion of the jawbones (a percentage of 1% of all jaw cysts [3]) the working group reviewed the cases of SBCs published in the last 10 years and reported on a series of 4 clinical cases managed and followed up in our department with the aim of synthesizing the various epidemiological, clinical and radiological data as well as how to do when faced with these lesions.

This entity has been referred to by several names such as: simple bone cyst, solitary bone cyst, idiopathic bone cavities, traumatic bone cyst, unicameral bone cyst; which were used in our literature search strategy. This variation in naming mostly reflects the uncertainty about the etiology of these lesions [6, 16].

Traumatic etiology has long been proposed to explain the occurrence of these injuries. This is not an etiology of certainty since the notion of a previous trauma was reported in only 6 patients from our review. Other theories have been suggested in the literature such as tumour degeneration, bone ischaemia, and bone growth abnormalities [16]. Further researchs must be carried out to try to explain the etiopathogeny of SBCs, which remains poorly understood [4].

SBCs usually occur in the 2nd decade of life without gender predominance (16 males against 17 females), which is consistent with the findings in the literature (Table 1).

They are often located in the mandible (75% of cases) [4]. The horizontal branch of the mandible is the most affected area in our sample (20 lesions). The Maxilla is rarely affected; only 3 cases of SBCs of the maxillae in the series of 51 patients of Matsuma *et al*., [21].

Involved teeth often respond positively to pulp vitality tests in the absence of tooth decay.

According to the Rhushton criteria established in 1946 [22], the diagnosis of SBC is essentially based on the clinical aspect during surgery. Indeed, an empty or serum-filled endo-osseous cavity surrounded by hard bone walls associated with the absence of an epithelial lining are characteristics of SBC [19]; which means that we generally do not have a surgical specimen to carry out an anatomopathological examination.

The discovery of SBC is most often made following a routine radiological examination. These pseudo cysts are generally asymptomatic (82% our sample). However, a slight expansion (6%) and mild pain (12%) may be the chief complaint of some patients (Table 1).

SBCs are generally single in presentation. However, Seo-Young An *et al*., [12] reports a case series and a literature review of multiple SBC. Multiple SBCs are generally synchronous although asynchronous SBCs have been reported [7].

The most common radiographic finding is scalloping between the roots of involved teeth. Expansion, thinning of the cortices and unilocular radiolucent can be present. [6]

SABINO-BEZERRA *et al*., [9] discussed atypical presentations of simple bone cysts of the mandible especially the multilocular aspect of SBCs which gives confusion with other lesions of a multilocular nature such as ameloblastoma, odontogenic keratocyst, myxoma or central giant cell granuloma, as was reported in our fourth case report (Figure 4).

The management of SBCs remains simple; surgical intervention is the gold standard consisting of a bone trepanation giving access to the lesion, curettage of the bone walls causing bleeding which will favour the formation of a blood clot and the healing of the bone [1].

Some authors suggest that spontaneous resolution may occur without any intervention in cases where the patient refuses surgery. In this case, periodic radiological follow-up should be instituted [12].

CHELL *et al*., [15] reports a case of An Unusual Finding of a Solitary Bone Cyst in a Patient with a Fractured Mandible. This advocate surgical intervention, as soon as the diagnosis is made, to avoid such complications.

You *et al*., [24] Recommends associating allogenic bone graft or collagen plug to the conventional surgical exploration of SBCs. They suggests that allogenic bone graft in SBCs has many advantages such as early bone healing, prevention of soft tissue ingrowth, and easily identified recurrence.

Juan Carlos Bernabeu-Mira *et al*., [20] describes a conservative approach that consists of creating an access to the SBC through submarginal incision, osteotomy with ultrasound, filling with APRF+ membranes and repositioning of cortical bone which ensures optimal bone healing and avoids gingival recessions at six months follow-up, showing to be a good alternative for satisfactory defect resolution in similar clinical cases.

HORNE *et al*., [14] showed that the average time needed to observe complete radiographic healing or recurrence was 3 years and 5 months and that cases involving multiple SBCs were much more likely to recur than those with solitary lesions. Hence the importance of long-term follow-up, especially for patients treated for multiple SBC because of the high probability of recurrence.

CONCLUSION

SBCs are lesions that usually occur in the second decade of life without gender predominance. The dentist must be able to suspect this type of lesion when faced with a radiolucency diagnosed through a routine radiological examination since these lesions are most often asymptomatic and may go unnoticed.

Conflicts of Interest

The authors declare that there is no conflict of interest regarding the publication of this article.

Authors' Contributions

All authors have read and agreed to the final version of this manuscript and have equally contributed to its content and to the management of the cases.

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