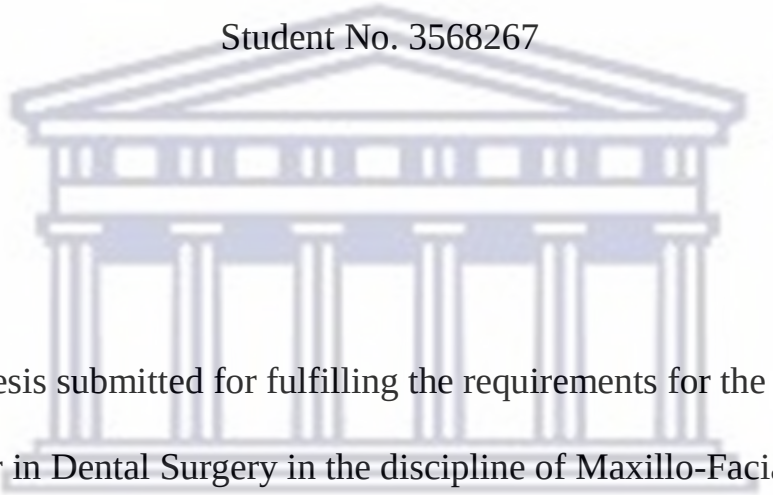
# NON-EPITHELIAL BONE CYSTS OF THE JAWS

Mahdi Dashti

Student No. 3568267



A mini-thesis submitted for fulfilling the requirements for the Degree of Master in Dental Surgery in the discipline of Maxillo-Facial and Oral Surgery, Faculty of Dentistry, University of the Western Cape.



Supervisor: Prof. J.A. Morkel

Co-Supervisor: Dr. D.A. Smit

January 2020

# Non-Epithelial Bone Cysts of the Jaws

Mahdi Dashti

**KEY WORDS**

Non-epithelial cyst

Simple bone cyst

Solitary bone cyst

Traumatic bone cyst

Unicameral bone cyst

Aneurysmal bone cyst

Pseudocysts of jaw

# ABSTRACT

## NON-EPITHELIAL BONE CYSTS OF THE JAWS

**Mahdi Dashti**

MDS Mini-thesis, Department of Maxillo-Facial and Oral Surgery, Faculty of Dentistry, University of the Western Cape.

Aneurysmal Bone Cysts (ABC) and Solitary Bone Cysts (SBC), both non-epithelial cysts of the jaws are defined as benign lesions of an unclear aetiology. There is limited literature available on these two primary non-epithelial cysts of the jaws, especially in African populations. This retrospective study focused on the clinical and radiographic features, as well as management of the non-epithelial cysts of the jaws presenting at the University of the Western Cape Oral Health Centre from 1970-2018.

The aim of this study was to describe the clinical and radiological features of non-epithelial cysts of the jaws that presented at the Departments of Maxillo-Facial and Oral Surgery and Diagnostics and Radiology at the University of the Western Cape Oral Health Centre as well as their management and recurrence patterns.

This was a cross-sectional analytic study of non-epithelial-lined cysts of the jaws. The sample for this study was selected by manually collecting all patient records available. All cases of ABC and SBC of the jaws included in this study were confirmed by radiological, histopathological and recorded clinical findings prior to the inclusion in this study. The data recorded included the patient's age, gender and ethnicity. The clinical

signs and symptoms of the lesion as well as the history were described. Radiographic features were documented and its effect on adjacent structures (adjacent dentition, mandibular canal and/or maxillary sinus) were noted. Management and follow-up was also documented. Data was analysed using Epi Info® 2000 by student's unpaired t-test to compare the findings and to correlate these findings with different parameters.

A total of 32 SBC and five ABC cases had complete demographic information and radiographs and were included in this study. The mean age of patients with SBC was 21 years (range 13-51 years) and 22 years for ABC (range 12-36 years). The majority of SBC patients were males (58.62%) while four out of five ABC patients were females (80.0%).

The majority of SBCs were discovered incidentally with only two cases being symptomatic (6.2%) while no history of trauma was noted for both pathologies. All SBCs presented in the mandible (96.8%) with exception of one case which occurred in the maxilla. All ABCs occurred in the mandible with the mandibular posterior regions (ramus and molar) the most commonly involved. Radiographically, the majority of SBCs (75%) appeared as radiolucent while all ABC lesions were of mixed density. The majority of SBC lesions (81.3%) appeared as unilocular with well-defined borders (71.8%). All ABCs had a honeycomb appearance.

Most SBCs were managed by surgical exploration (62.5%) and curettage (37.5%). In the majority of cases (62.5%), a fluid filled cavity was found. In some cases (30%), an empty cavity was found. None of the SBCs recurred following surgical management.

All ABCs were also managed with surgical exploration with the majority of lesions showing dark venous blood.

In conclusion, the majority of the features of ABC and SBC were similar to those reported in the literature. Exceptions included the larger size of SBC and ABC lesions in this sample compared to other studies. Surgical exploration and/or curettage have shown to be an acceptable treatment method for the management of these pseudocysts with no recurrence rate in this sample.

# DECLARATION

I declare ***NON-EPITHELIAL BONE CYSTS OF THE JAWS*** is my own work, that it has not been submitted for any degree or examination in any other university, and that all sources I have used or quoted have been indicated and acknowledged by complete references.



Mahdi Dashti 23 January 2020

Signed:

# ACKNOWLEDGEMENTS

This research project could not have been accomplished without the guidance and support of the following individuals:

- I would like to express my sincere gratitude to my study supervisor Prof. Jean Morkel (Head of Department of Maxillo-Facial and Oral Surgery) for all his support and valuable input throughout the study. His continuous encouragement and guidance has been very valuable for the completion of this study.

- I would like to thank Dr. Fadi Titinchi for his efforts, support and assistance to complete this mini-thesis with me. I appreciate his help and experience in this project.

- I would like to thank Dr. Dirk Smit for his guidance and assistance to start and finish this mini-thesis.

# DEDICATION

This thesis is dedicated to my parents, wife and teacher without whom it was almost impossible for me to complete this work. They have been with me through every step of the way, through good and bad times. Thank you all for the priceless love, guidance and unconditional support you have given me to succeed. Thank you for everything.

# CONTENTS

# LIST OF TABLES AND FIGURES

## List of Tables



## List of Figures

**Figure 9:** Pantomograph showing multilocular SBC with root resorption on lower left first molar.

**Figure 10:** Pantomograph showing SBC with expansion of cortex and resorption of the cortical line around the inferior alveolar canal (arrow).

**Figure 11:** Six-month post-operative pantomograph of same patient as in Figure 9 following curettage. Note new bone formation in the left posterior mandible.

**Figure 12:** Pantomograph showing multilocular ABC on the right posterior mandible preoperatively (arrow).

**Figure 13:** Pantomograph showing same patient as above following marginal resection of ABC on right posterior mandible (arrow).

**DEFINITIONS**

**Cyst:** a pathological cavity lined with an epithelial lining which is filled with fluid, semifluid, gaseous, or semisolid contents. The lesion grows through hydrostatic pressure by withdrawing fluid via osmosis (Kramer 1974).

**Non-epithelial cyst** "**Pseudocyst":** resembles a cyst cavity, but lacks the epithelial lining.

**Aneurysmal bone cyst (ABC):** An intraosseous accumulation of variable-sized, blood-filled spaces surrounded by cellular fibrous connective tissue and reactive bone with no epithelial lining.

**Solitary bone cyst (SBC):** An intraosseous cavity that is either fluid-filled or empty. It is mostly found in the proximal metaphyseal regions of the long bones in children and adolescents. Due to the lack of epithelial lining it is considered a pseudocyst. The uncertainty about this lesion is reflected in the many terms used for the same lesion.

**ABBREVIATIONS**

ABC: Aneurysmal bone cyst

SBC: Solitary bone cyst

UWC: University of Western Cape

USP6: Ubiquitin-specific protease 6

MRI: Magnetic resonance Imaging

MR: Magnetic resonance

CT: Computed tomography

SI: Signal intensity

COD: cemento-osseous dysplasia

IAN: Inferior alveolar nerve

# Chapter 1
# INTRODUCTION

The jaws are most commonly affected areas by cysts due to the various epithelial remnants present. Various lesions mimic the clinical features of a cyst and thus it is critical to distinguish between those lesions and true cysts. The classification of jaw cysts is an important tool to assist with the diagnosis. According to Neville and Damm (2016), cyst classification is divided into odontogenic and non-odontogenic cysts.

The 2005 WHO classification of non-epithelial cysts of the jaw included aneurysmal bone cysts (ABC), solitary bone cysts (SBC), and stafne cavity (Wright and Vered *2017*). However, according to Shear's classification of jaw cysts, stafne cavity is not regarded as a non-epithelial cyst of the jaw (Shear and Speight 2007). In the latest WHO classification which was updated in January 2017, the stafne cavity has been removed from the category of cystic lesions (Wright and Vered 2017). The 2017 WHO classification categorises the ABC and SBC under "Bone Tumours and Related Lesions". A Further sub-classification places the ABC and SBC under Giant Cell Lesions and Bone Cysts (Wright and Vered 2017).

Other sub-classifications under jaw cysts include; inflammatory cysts, developmental cysts, epithelial and non-epithelial cysts. This study focused on non-epithelial cysts. Such cysts derive from non-odontogenic tissues which are distinguished from other cysts mainly by the lack of an epithelial wall. The non-epithelial cysts of the jaw are not clearly understood in terms of aetiology, however, a few theories have been proposed.

ABCs are clinically more aggressive than SBC. The aetiology is uncertain, but there are few theories advocated of which the most well-known theory is related to an intraosseous haematoma. The latter can be caused by trauma, venous obstruction, irregularity of the bone growth, or malformations in the bone metabolism. The intraosseous clot then undergoes liquefaction and osteolysis.

ABC and SBC are both non-epithelial cysts of the jaw or so called "pseudocysts" which are benign bony lesions of vague aetiology. A pseudocyst has similar clinical and radiographic features as true cysts except that histopathologically they do not possess an epithelial lining. SBCs are rarely seen in the maxillofacial region (1-2% according to Hs *et al.* 2012 and Velasco *et al.* 2012). Ninety percent of SBCs are found in long bones. The ABC is encountered at the rate of 2% in the jaws, 20% in the spine, and 50% in the long bones (Bharadwaj *et al.* 2013; Grecchi *et al.* 2012).

# Chapter 2

# LITERATURE REVIEW

---

## 2.1. LITERATURE REVIEW OF ANEURYSMAL BONE CYST

### 2.1.1. Aetiology of ABC

The pathogenesis of ABC is unclear, although the lesion is mostly regarded as a reactive lesion. Trauma does not seem to play a significant role. Many authors support the theory that a traumatic event, vascular malformation, or neoplasm may have disrupted normal osseous hemodynamics leading to an enlarging area of haemorrhage and osteolysis (Neville and Damm 2016). However, 20-30% of reported cases are associated with other lesions. Flores *et al.* (2017) reported that in his 12 ABC samples, five cases had a previous history of trauma related to the region.

There is some cytogenetic evidence that primary ABC is neoplastic in nature. The majority of the primary lesions analysed showed recurrent translocations and consequent transcriptional up-regulation of the ubiquitin-specific protease 6 (USP6) (also known as Tre-2 or TRE 17) oncogene on chromosome 7p13. These USP6 translocations have been noted in very few craniofacial lesions so far. However, the mechanism is poorly understood (Oliveira *et al.* 2004).

### 2.1.2. Classification

The 2005 WHO classification of the non-epithelial cysts of the jaw; included ABC, SBC, and stafne cavity. In the 2017 WHO classification, the stafne cavity was removed from the classification.

The 2017 WHO classification included the ABC and SBC in the new classification under "Bone Tumours and Related Lesions" (Wright and Vered 2017). They were further sub-classified under giant cell lesions and simple bone cysts (Strong association with *CDH11* and/or *USP6* mutations is seen in primary ABC). Central giant cell granuloma (CGCG), peripheral giant cell granuloma (PGCG) and cherubism were also included in that sub-classification (Wright and Vered, 2017).

### 2.1.3. Epidemiology

Pseudocysts of the jaw are rare entities in the craniofacial region; however, it is relatively more common in the long bones and the spine. The only study that could be found in literature, which reviewed both ABC and SBC, was done by Flores *et al.* (2017). They reported 54 cases of pseudocysts after researching 20,456 records. The frequency of ABC was 2.39% of the total cysts in the jaws for their sample.

### 2.1.4. Clinical Features

#### 2.1.4.1. Frequency

ABC is regarded as a rare entity and of these, only 2% involve the jaws. A study conducted by Struthers and Shear in 1984, showed the incidence in South Africa to be 0.4%. Whereas a study in UK, noted an even lower incidence of 0.15% (Jones and

Franklin 2006). Sun *et al.* (2009) reported that there were only 92 cases of jaw ABCs reported in English literature involving 75 articles.

2.1.4.2. Age

ABC is primarily seen in patients younger than 30 years. Gnathic lesions are seen in the first three decades with a peak in the second decade, but may also occur in a broad age range. Flores *et al.* (2017) had a mean age of 18.6 years and age variation was 10-36 (nine cases appeared in the 2nd decade of life). Sun *et al.* (2009) reported 72.8% of ABC occurred in the first two decades.

2.1.4.3. Gender

Most authors report either no difference in gender or a slight female predilection. Kransdorf and Sweet (1995) reported a slightly higher predilection in females, while Motamedi and Stavropoulos (1997) found no difference. Flores *et al.* (2017) showed 10 females and two males in their 12 sample of cases, all of them were Caucasian.



2.1.4.4. Site

ABCs are primarily seen in long bones or vertebrae. Of those in the jaws, 62-68.5% of lesions are seen in the mandible of which 31.3% are noted in the ramus, 20.4% in the posterior mandibular region, and 20.5% in antrum of maxilla (Sun *et al.* 2009). Therefore, they are mostly seen in molar regions of both jaws. ABCs are rarely seen in condyle and coronoid process.

2.1.4.5. Clinical presentation

In the long bones, the majority of ABC present with a tendency for rapid growth and associated pain (Rosenberg *et al.* 2005). In the jaws, however, the clinical presentation may vary strongly. It can range from small asymptomatic lesions, to rapidly growing and destructive lesions (Motamedi and Yazdi 1994, Hardee *et al.* 1992). There may be a history of associated trauma in up to 28% of cases (Sun *et al.* 2009). Springiness or an egg-shell phenomenon is seen when the lesion perforates the cortex. No bruits have been detected. Maxillary lesions can bulge into the maxillary sinus and lead to nasal obstruction, bleeding, proptosis and diplopia (Devi *et al*. 2011).

2.1.4.6. Radiological features:

2.1.4.6.1. Conventional radiographs

ABC in long bones presents as a well-defined expansive radiolucent lesion surrounded by a thin overlying cortex (Mendenhall *et al.* 2006). In the jaw, it varies from unilocular or multilocular radiolucency with marked cortical expansion. Borders can be well-defined or ill-defined. Sun *et al.* (2009) reported that 93.8% of the lesions presented as a radiolucency, 4.2% were radiopaque while 2.1% were mixed. 69.4% of these lesions had a multilocular appearance.

Interestingly, in the literature, most studies report ABCs to be radiolucent on pantomographs. Motamedi and Yazdi (1994) reported their entire sample of 120 ABC lesions to be radiolucent while Flores *et al*. (2017) reported 11 of 12 ABC lesions to be radiolucent.

Frequently a “ballooning” or “blow-out” growth pattern is noted which appears as a radiolucent lesion with elevation of the periosteum. It forms an ovoid or fusiform bony expansion with the typical cortical bulging. Uncommonly, radiopaque foci are noted. It is thought to be small reactive trabeculae of bone (Kalantar 1998).

2.1.4.6.2. Advanced imaging

Authors noted that new subperiosteal osteogenesis might be visible on computed tomography (CT) and present as a thin continuous cortex, however, this might be subtle or invisible on plain radiographs (Kransdorf and Sweet, 1995). Lesions are often unilocular, but long-standing lesions may appear as a ‘soap-bubble’ due to calcifications (Kransdorf and Sweet, 1995). CT scans and magnetic resonance imaging (MRI) may outline this pathology in a more accurate way. Furthermore, it may show fluid levels in the lesion (Revel *et al.*, 1992). Teeth displacement and root resorption has been described (Hardee *et al.*, 1992). The latter may cause the radiographic differential diagnosis to be vaster and more difficult to differentiate.



According to Lenz 1993, Lenz 2000, and Lenz *et al.* 2000, MRI is a superior modality to view soft-tissue, especially using contrast and multiplanar facility. This makes MRI most useful in analysis of the lesion’s internal structures. However, only few studies were done on MR imaging of ABC (Revel *et al*. 1992, Gadre and Zubairy 2000, Hernandez *et al*. 1993).

Asaume *et al*. (2003) reported almost a homogenous intermediate signal intensity (SI), including a partial slight low-SI area on T1WI. On Gd-T1WI, it showed an intermediate SI area and apart from those areas, a slight low SI on non-enhanced T1WI were well-enhanced. This picture would give the ‘honeycomb’ appearance.

On the other hand, angiograms did not often show the specific feature of multiple anomalous vascular branches as reported by Pankey *et al*. (1984) and which then prevents this feature to be an indicator in the diagnosis of ABC (Ueno *et al*. 1982, Motamedi and Yazdi 1994). The giant cell tumour, the giant cell granuloma and benign fibrous histiocytoma may show CT features similar to ABC (Lee and Lum 1999, Khafif *et al*. 2000, Machiels *et al.* 1998, Dalley 1999). Fluid levels on MRI are not always seen in ABCs. Some lesions can show fluid levels e.g. telangiectatic osteosarcoma, giant cell tumour, and chondroblastoma (Davies and Wellings 1992, Tsai *et al*. 1990). Therefore fluid levels are non-specific for ABC.

Bone scintigraphy and radionuclide angiography may be able to differentiate ABC from other forms of tumour, especially hypervascularised tumour and central haemangioma. The doughnut-like or the ring-like appearance of the accumulated radioactivity corresponds to the expansile character of the lesion (Okuyama *et al*. 1985)

**2.1.5. Diagnosis**

ABCs have variable clinical and radiological presentations and should be considered in the differential diagnosis of any unilocular or multilocular radiolucency of the jaws and any mixed lesion. Presence of blood on aspiration during surgery is highly suggestive of ABCs while histopathological examination is the only definitive diagnostic method to identify this lesion (Urs *et al*. 2014).

### 2.1.6. Pathogenesis

The pathogenesis of ABC is controversial. Jaffe and Lichtenstein (1942) reported long bones cysts may show eccentric osseous expansion indicating a juxtacortical or a sub-periosteal lesion. Both juxtacortical and intramedullary location lesions can thus be the found. Kransdorf and Sweet (1995) concluded that a variety of causes could lead to ABCs as an end result of a pathophysiological process. They proposed that a juxtacortical lesion is due to traumatic origin and the intramedullary is caused by secondary change within a pre-existing lesion. Juxtacortical lesion was not reported in the craniofacial region. Kransdorf and Sweet (1995) suggested another theory that involved the cause of cysts by a sudden venous obstruction or formation of arteriovenous shunt.

Ewing (1940) supported the concept that ABC is a secondary phenomenon arising in pre-existing bone lesion. It was suggested that a giant cell tumour was modified by communication with a large blood vessel. Jaffe (1950) proposed that the cyst may result from other lesions when modified by haemorrhage.



Biesecker *et al.* (1970) showed evidence of associated osseous lesions in 21 of 66 ABC cases (32%). These include non-ossifying fibroma, chondroblastoma, giant cell tumour of bone, osteoblastoma, giant cell granuloma, fibrous dysplasia, myxofibroma, and SBC. They postulated a theory which suggested an arteriovenous malformation in the bone and that its hemodynamic forces caused the initial lesion to become ABC. Levy *et al.* (1975) suggested that an ABC maybe a primary lesion.

In an analysis of 19 cases, Struthers (1980) and Struthers and Shear (1984b) found smaller blood-filled spaces and numerous micro-cysts which were usually noted at the periphery of the large blood-filled spaces. These changes were particularly found in

central giant cell granuloma. Such micro-cysts were found in 28% of central giant cell granulomas. The same changes were observed in 8% of fibrous dysplasia, 4% of ossifying fibroma and 3% of cementifying fibroma. They suggested that microcystic changes are the initial transformation in the primary pathology. The propensity to form micro-cyst in the central giant cell granuloma is more likely to develop because of the loose, oedematous, fibrillar connective tissue stroma that contains numerous thin-walled blood vessels and extravasated erythrocytes (Shear and Speight 2007). It is thought that micro-cysts were the result of localised necrotic areas in the stroma. The necrosis resulted from stagnation and ischemia. The stromal connective tissue makes up the lining of the micro-cysts and multinucleate giant cells can be seen on their margins. Further stromal breakdown and coalesce result in enlargement of the micro-cyst. The loss of stromal support leads to the breakdown of the vessel walls that then leads to haemorrhage in the micro-cyst. The hemodynamic pressure is responsible for the growth of the lesion (Shear and Speight 2007).

It is thought that a connection is established between a vessel and the micro-cyst which causes hemodynamic pressure in the micro-cyst that increases the size. It is also proposed that the loose surrounding stroma and oedema reduces resistance to growth of the lesion. The spaces then coalesce to form a macro-cyst surrounded by compressed stroma. The pressure from these multiple expanding blood-filled cysts results in osteolysis. A ‘blow-out’ of the lesion occurs once the endosteal resorption breaches the cortical bone. This breach usually remains subperiosteal and new bone layer can be deposited to form a thin shell covering the ABC.

Struthers and Shear (1984a) had an opinion that malignant lesions are less likely to produce the classic clinicopathological picture of ABC because of the aggressive be-

haviour of malignant lesions. Nevertheless, some of the fibrosarcoma lesions they reviewed showed large blood filled spaces. Although rare, it is thought that a malignant ABC is associated with a malignant primary lesion. That would then possibly explain the occasional finding of a malignant cyst (Levy *et al.* 1975).

Tillman *et al.* (1968) studied 95 ABCs and reported that there was no evidence of precursor lesions in these cases. Ruiter *et al.* (1977) agreed with Tillman in the reported 105 cases. However, both groups of authors did admit that areas may have been present that resembled other lesions. Schajowicz (1981) found that areas similar to the ABC could be present in many bone lesion as a result of haemorrhage.

### 2.1.7. Pathology

ABCs typically have a thin shell of bone with a sound periosteum covering the lesion. Cortical bone can be perforated. Dark venous-like blood is seen once this thin shell is removed; the so called "blood-soaked sponge". This bleeding can be profuse and often difficult to stop until cyst removal is complete (Neville and Damm 2016).

The content of the cyst is a variable amount of soft tissue which consists of friable vascular tissue that subdivides the cavity into multiple blood-filled locules. Some solid tissue might be found in the lesion which may represent either areas of repair or remnant of a pre-existing lesion. Direct connection with any blood vessel cannot be seen intra-operatively (Neville and Damm 2016).

### 2.1.8. Histological features

The histology would entail numerous capillaries and blood-filled spaces of varying size lined by flat spindle cells and separated by delicate loose-textured fibrous tissue. These

blood-filled spaces lack an endothelial or epithelial lining. The majority of this lesion contains multinucleated giant cells and scattered trabeculae of osteoid and woven bone. In some areas it looks similar to Giant cell granuloma of the jaws as it contains in some of the solid areas sheets of vascular tissue, containing large numbers of multinucleated giant cells, fibroblasts, haemorrhage and haemosiderin. The giant cells in ABC exhibit an osteoclastic phenotype. The osteoid deposits may appear linear, nodular, or lace-like. Other solid areas may resemble fibrous dysplasia, ossifying fibroma and possibly other jaw tumours which can support the theory that ABC may present a secondary change in a precursor lesion (Neville and Damm 2016).

Clough and Price (1968) reported that there are no malignant cellular features unless the cyst has developed into a malignant tumour. Perroti *et al.* (2004) reported that occasionally the lesion can be solid with vascular fibrous tissue characteristic along with osteoid and woven bone without the blood-filled pseudocyst. Histologically such solid lesion is indistinguishable from giant cell granuloma so diagnosis is based on clinical and radiographic features.



Liu *et al.* (2003) compared the histopathology of 53 cases of giant cell lesions of the jaws, which included 34 central, six peripheral, seven cherubism and six ABCs. Histologically, they were all similar and contained multinucleated giant cells with similar morphology and distribution. However, 'cavernous or sinusoidal blood-filled spaces' were only found in ABC. The study supported the concept that ABC is primarily related to giant cell granuloma of the jaws which makes it a giant cell lesion. After the transudation and the stoppage of the haemorrhage, it is proposed that there will a gradual absorption of the serous fluid and a subsequent empty cavity.

### 2.1.9. Treatment and prognosis

ABCs of the jaws are usually treated by curettage or enucleation and might be supplemented by cryosurgery. En bloc resection is reserved for recurrent or extensive lesions. Some clinicians advocate embolization, however, the lesion is usually low-grade flow and does not require embolization. Sun *et al.* (2009) considered curettage to be suitable as treatment for ABCs.

Shear and Speight (2007) believed that ABC treatment must be determined by the nature of the associated lesions. El-Deeb *et al.* (1980) and Ginger *et al.* (1984) reported that curettage is the most common form of treatment with a recurrence rate of 9-26% in the jaws.

Clough and Price (1968) reported continued growth after careful curettage and bone grafting. They recommended complete excision provided this would not cause malfunction. Shear and Speight (2007) reported recurrence twice in one of their cases which was associated with ossifying fibroma after curettage. ABCs associated with central giant cell granuloma is less likely to recur after curettage. Shear and Speight (2007) noted that as most aneurysmal bone cysts of the jaws appear to involve central giant cell granulomas, the latter would explain the high success rate with conservative treatment.

Radiotherapy is not an option to treat ABC unless it is associated with a malignant lesion which is very rare. Patients should have periodic post-operative follow ups. Typically, the defect heals within six months to one year. Recurrence rates range from 8% to 70%. Overall, the long-term prognosis is favourable (Sun *et al.* 2009).

## 2.2. LITERATURE REVIEW OF SOLITARY BONE CYST

### 2.2.1. Aetiology of SBC

The pathogenesis of SBC is also is uncertain. The trauma-haemorrhage theory is supported by many advocates (hence the term 'traumatic bone cyst' is widely used) and it suggests that insufficient trauma to cause a fracture can lead to an intra-osseous haematoma. If the haematoma does not proceed to organisation and repair, it may liquefy and result in a pseudocystic defect. However, the history of trauma at the same location and the presence of haemorrhage are sometimes inconsistent.

Chadwick *et al.* (2011) reported three current hypotheses:

1-Local factors that can possibly disturb osteoblast differentiation during bone growth and development can result in the bone cavity.

2-A developing tumour in the bone can disintegrate via liquefactive degeneration and cause a cavity in the bone.

3-It is believed that traumatic/micro-traumatic events can result in bone fractures which can cause localised thrombosis that induce focal ischaemic event and aseptic necrosis of the bone. That is why it believed that the higher frequency of SBC in the mandible is due to the fact that the mandible is subjected to trauma more than the maxilla, especially in the premolar areas where SBC is mostly found (Harnet *et al.* 2008).

Chadwick reported in 2011 that the end result of the necrosis resulted in the formation of cavity that may be lined with fibrous connective tissue and contain serous fluid, and/or blood, or may be completely empty.

Other theories involve venous obstruction, local disturbance in bone growth, altered calcium metabolism, ischaemic marrow necrosis, aberrant synovial development and a bone tumour or cyst degeneration. Flores *et al.* (2017) reported 12 cases, out of 42, in association with a history of trauma on the affected area.

### 2.2.2. Clinical features

#### 2.2.2.1. Frequency

SBC is also a rare entity. The incidence ranges from 0.5-1% internationally. In South Africa, only 35 cases over a 46-year period have been documented (1% of jaw cysts) by Shear and Speight (2007). Hoffmeister and Härle (1985) reported an incidence of 0.6% (19 cases out of 3353). Jones and Franklin (2006 a, b) reported 36 cases out of 6869 in 30-year period (0.5%).



Howe *et al*. (1965) defined SBC as a single cyst with no epithelial lining and no evidence of acute or chronic infection. It should contain principally a fluid and no soft tissue with thin or thick bony walls. It was also reported that 87% of cemento-osseous dysplasia associated simple bone cysts are found in females in the fifth decades. On the other hand, SBC had no gender predilection in the 2nd decade of life (Chadwick *et al.* 2011).

2.2.2.2. Age

The age range for the presentation of SBC is between 2.5-75 years. More than 50% of SBCs are seen in the second decade of life (Howe 1965; Hansen *et al.* 1974). Killey *et al.* (1977) agreed on the 20s peak but did have one patient above 50 and another over 60. The mean age was 34 years according to Jones and Franklin (2006a).

2.2.2.3. Gender

The male:female ratio was 1.6:1 according to Howe (1965). Killey *et al.* (1977) reported 13 females and 10 males. Kaugars and Cale (1987) did a literature review and they reported no gender difference in the literature and their own 161 cases. Neville and Damm (2016) reported that there was no significant gender predilection in jaw lesions, but the extragnathic lesions exhibited a male predilection. Chadwick *et al.* (2011) also reported that solitary SBC has no gender significant, but cemento-osseous dysplasia (COD) associated SBCs are six times more found in females and it occurs more often in older age.



2.2.2.4. Site

SBCs have been reported in almost every bone of the body. Most cases involve the metaphyses of long bones with predilection for the proximal femur and humerus. In the jaws, the overwhelming majority are found in mandible. Kaugars and Cale (1987) reported that 95% occurred in mandible. Copete *et al.* (1998) reported 2% in posterior maxilla. Almost all maxillary cases were reported in the anterior part of the maxilla and the majority of the mandibular cases were reported in the symphyseal or parasymphyse-

al regions. Some authors reported cases in ramus and body of mandible. Very rarely the lesions are seen in the condyle (Persson 1985; Rubin and Murphy 1989). Chadwick et *al.* (2011) reported that the premolar, molar and symphyseal regions were mostly affected. Chadwick *et al.* (2011) reported that solitary SBCs were found in every anatomical site in the mandible. SBCs, which are associated with COD, were not found in the incisors area.

2.2.2.5. Clinical presentation

Unlike the extragnathic lesions, jaw lesions are found to be mostly asymptomatic. It is therefore safe to say that the actual frequency of the lesion exceeds the numbers reported in literature. It is noted that about 20% of the cases would experience a painless swelling. Paraesthesia and pain is seen infrequently (Fielding *et al*. 1999).

Howe (1965) reported that 60% of the reported cases were incidental findings on radiographs. 27% of the cases presented with swelling, 10% with pain, 2% with labial paraesthesia and 2% presented with both swelling and pain. More than half of the cases had a history of significant trauma to the affected area. The time-lapse between the injury and diagnosis ranged from a month to 20 years.

2.2.2.6. Radiological features

2.2.2.6.1 Conventional radiographs

The cyst appears as a well-delineated unilocular radiolucent lesion with irregular but definite borders and delicate cortication. Ill-defined and multilocular appearance has also been described. The size can range from 1-10 cm in diameter. The lesions may also be extensive (Suei *et al*. 1998).

Copete *et al*. (1998) reported that most SBCs (61%) were entirely radiolucent. Bone trabeculae were reported within SBCs in 32% of cases while 7% were diffusely radio-opaque. Flores *et al.* (2017) reported the majority of their SBC cases (82%) also to be completely radiolucent.

Some degree of marginal condensation is seen in 63% of cases but it is not as clear as in radicular cysts. Seventy-two percent of the posterior mandibular lesions scallop around the associated dental roots. According to Chaldwick *et al.* (2011), the scalloping pattern leaves the surrounding roots, lamina dura, periodontal ligament spaces or follicular sac undisturbed. Some or all of these features are considered pathognomonic of the SBC.

Chaldwick *et al.* (2011) reported that SBCs associated with COD lesions are more likely to feature thinning of the cortex, scalloping between the roots and cortical expansion. Solitary SBCs are less likely to have these classical features. Also, the loss of lamina dura is more commonly seen in COD associated SBCs. They also reported that 48.5% of solitary SBCs had identifiable corticated border and 54.4% featured scalloping around adjacent roots which was described as pathognomonic for SBC.



2.2.2.6.2. Advanced imaging

Suei *et al*. (1994) did a study to evaluate whether SBC contains gas levels in their cavity using CT images. Their results showed that CT can distinguish between air and liquid by analysing the difference in Hounsfield units between the gas and liquid. They concluded that operative findings of gas in SBC maybe an error in some cases.

Suei *et al*. (1998) compared CT images with surgical finding and reported that CT can show the existence of some fluid in the cavity. They suggested that in cases of empty

cavities, fluid may have escaped out by means of fenestration. Jacobs *et al*. (1955) reported that when SBC lesions were detected early, usually blood or serosanguineous liquid is present and later it disappears.

Matsuzaki *et al*. (2003) reported that MRI is the most useful modality to evaluate the internal structure of the lesion because of its superior soft tissue contrast and multiplanar views. This can distinguish SBCs from others. In a true cyst, MRI enhances the epithelial lining only (Minami *et al*. 1996). The contrast-enhanced MRI can differentiate a true cyst from SBC and ABC by the presence or the absence of epithelial lining. The content of SBC and ABC can assist to distinguish benign tumours, as ABC and SBC are generally filled with fluid while benign tumours are generally solid. By analysing the signal intensity of ABC and SBC on MRI, the radiologist can distinguish them from benign tumours.

One of the main features that makes SBC important to distinguish from other pathologies is that some studies showed spontaneous resolution, which makes surgical intervention unnecessary, unlike some other similar pathologies (Sapp and Stark 1990, Matsuzaki *et al*. 2003). However, when the SBC is unilocular in appearance it is difficult to distinguish it from a true cyst and hence surgical exploration is required (Hisatomi *et al*. 2003, Vergel De Dios *et al*. 1992).

### 2.2.3. Diagnosis

SBCs are mostly an incidental finding. There are many odontogenic and non-odontogenic lesions that resemble SBC and therefore surgical exploration is necessary to establish diagnosis. Diagnosis is primarily based on clinical, radiographic and intraoperative findings. One-third of cases have an empty cavity with smooth, shiny bony

walls. Two-thirds contain small amounts of serosanguineous fluid. The neurovascular bundle might be running free in the lesion. (Neville and Damm 2016)

### 2.2.4. Pathogenesis

The pathogenesis of SBC is unknown but there are a number of theories proposed. Olech *et al.* (1951) and Howe (1965) suggested a traumatic aetiology. Olech *et al.* (1951) postulated that following trauma, intramedullary haemorrhage occurs. Failure of early organisation of the haematoma in some of the marrow spaces and subsequent liquefaction of the clot can result in an SBC, hence the name traumatic bone cyst. However, a plausible explanation for the failure of organisation is a critical point in that theory. Shear and Speight (2007) proposed that the areas with spongy bone which contains haemopoietic marrow and enclosed by thick cortex, develop these cysts after trauma. Hence, the high frequency in sound individuals and in sites such as the metaphyses of long bones and in the mandible.

Although there is some debate about the accuracy of a trauma history in many cases, trauma can be considered as an initiating factor in some cases according to Shear and Speight (2007). As with trauma, some other stimuli can result in damage of thin-walled sinusoids and lead to intramedullary haemorrhage. Olech *et al.* (1951) argued that the primary haematoma will not organise if there is no contact with reactive and fibrous connective tissue.

Intra-operatively, the majority of SBCs are filled with only air or other gas. Some of them contain blood or serosanguineous fluid which support the concept of a broken down haematoma. The breakdown products that result from haemolysis lead to an increased osmotic pressure. Toller (1964) confirmed this via an experimental study that

osmotic tension of SBC fluid is greater than that of the patient's blood. This phenomenon leads to transudation into the cyst fluid with osteolytic activity and swelling.

Shear and Speight (2007) postulated that since the lesion is not seen in patients above 30 years, it would suggest that SBC is a self-limiting lesion. The defect normally heals after the space is filled with blood which can happen by surgical intervention or spontaneous.

Chadwick *et al.* (2011) and Flores *et al.* (2017) reported that SBCs at times might arise in association with cemento-osseous dysplasia and the fibro-osseous proliferations. Such cases have female predilection.

**2.2.5. Histological features**

SBC has no epithelial lining. Loose vascular fibrous tissue membrane of variable thickness or a thickened myxofibromatous proliferation with reactive bone is noted. Fragments of fibrin with enmeshed red cells may be seen scattered. Haemorrhage and haemosiderin pigments are usually present and small multinucleated cells are often found. Some cyst walls are more densely fibrous, possibly due to longer standing lesions. Osteoclastic resorption is noted in adjacent bone on the inner surface (Matsumura *et al*. 1998). Beasley (1976) reported haemorrhage associated with necrotic tissue or tissue showing myxoid degeneration. These occurred in cavities adjacent to bone resorption regions.

Occasionally giant cells and lace like dystrophic calcification is present. Some authors noted amorphous, cementum-like material, which represent osteoid (Tariq *et al*. 2014). Howship lacunae (resorptive areas of bone) maybe seen which indicates previous osteo-

clastic activity (Neville and Damm 2016). Hara *et al.* (1990), Melrose *et al.* (1976) and others described an uncertain association between SBC and fibro-osseous lesions including fibrous dysplasia and cemento-osseous dysplasia.

### 2.2.6. Treatment and prognosis

SBCs are normally treated as a part of the diagnostic process. During the biopsy of the cystic lesion, if an empty cavity is revealed, the cyst wall is curetted. This will result in an uneventful healing in the majority of the cases due to the granulation tissue formation and subsequently new bone formation. Recurrence is unusual but has been reported (Horner *et al.*, 1988; Kuttenberger *et al.*, 1992, Baqain *et al.*, 2005).

SBC in long bones are often treated aggressively with various combination of curettage, cryosurgery, decompression, intra-lesional steroid injections, bone substitute or autologous bone marrow injection, and bone grafting. Recurrence rates are relatively high (12-54%) (Neville and Damm 2016). In contrast, SBC of the jaws are managed by surgical exploration and curettage. Recurrence rate is about 1-2% and periodic radiograph is recommended till the lesion is resolved (Traub *et al*. 2016).

# Chapter 3

# AIM AND OBJECTIVES

---

## 3. AIMS AND OBJECTIVES

### 3.1. Rationale for study

There is lack of literature on ABC and SBC. As the incidence of both these entities is rare, a study to include both these sub-classification of non-epithelial cysts of the jaws was indicated as it could add value to the scientific literature.

### 3.2. Aim

The aim of this study was to describe the clinical and radiological features of non-epithelial cysts of the jaws that presented at the Departments of Maxillo-Facial and Oral Surgery and Diagnostics and Radiology at the University of the Western Cape Oral Health Centre as well as their management and recurrence patterns.



### 3.3. Objectives

1. To describe clinical data from the dental records.

2. To describe the demographic information of ABC and SBC of the jaws.

3. To describe the presenting radiographic features of ABC and SBC of the jaws.

4. To compare the demographic and radiological features of ABC and SBC with other cystic lesions of the jaws.

5. To describe treatment methods and recurrence rates.

# Chapter 4

# MATERIALS AND METHODS

---

## 4. METHODOLOGY

### 4.1. Study design

This was a cross-sectional analytic study of the non-epithelial-lined cysts of the jaws. The main aim was to analyse and describe the clinical data and radiographic features of archived cases involving these lesions as well as its management and recurrence patterns during a period of forty-eight years from 1970 to 2018.

### 4.2. Study population and size

The sample for this study was selected by manually collecting all patient records available at the Departments of Maxillo-Facial and Oral Surgery and Diagnostics and Radiology of the University of the Western Cape Oral Health Centre in Cape Town, South Africa. The sample was not a random sample. It is a convenient sample as the cases are all archived files and radiographs. All cases of ABC and SBC of the jaws included in this study were confirmed by radiological, histopathological and clinical findings prior to the inclusion in this study. All pathologic specimens were evaluated and diagnosed by specialist Oral and Maxillofacial pathologists.

### 4.3. Selection criteria

Inclusion criteria for this study included:

1. Patients' records with a histologically confirmed diagnosis of primary ABC or SBC.
2. Patients' records that were complete with all demographic and clinical data.
3. Presence of at least one pantomogram for each record.

Exclusion criteria for this study included:

1. Patients' records with unknown history or incomplete records.
2. Patients' records with inconclusive diagnosis, either due to an insufficient biopsy specimen or inadequate clinical data.
3. Patients' records with panoramic radiographs of poor or insufficient diagnostic quality.



### 4.4. Data collection

All data collected for this study was recorded on a Microsoft Excel spreadsheet (Appendix 1 and 2). The data spreadsheet was formulated based on the objectives set out for this study. The data recorded included the patient's age, gender and ethnicity. The clinical signs and symptoms of the lesion as well as the history were described. Radiographic features were documented and its effect on adjacent structures (adjacent denti-

tion, mandibular canal and/or maxillary sinus) were noted. Management and follow-up was also documented.

### 4.5. Validity and Reliability

All radiographs were examined by the same two pre-calibrated observers (principal investigator and study supervisor). Each image available for the study was examined independently by each observer followed by comparison of the results. In cases where there was a disagreement of the findings amongst the two observers, then a third observer was consulted and the final decision was taken by consensus. All radiographs examined in this study were observed on a bright and evenly illuminated light-reflecting radiograph viewing box in an enclosed room with no light entry. This was done to standardise the setting for analysing the radiographs.

The viewing box was positioned in a comfortable position for the investigator. Adjunctive tools such as magnifying glasses were utilised to allow for detailed examination of the radiographs.

The location of the lesion was categorised into different regions in the mandible and maxilla. The anterior region of the mandible extended from the left canine (33) to right canine (43) and in edentulous patients from the left to right mental foramina. The posterior region of the mandible extended from canine to the angle of the mandible, for both left and right sides. The anterior region of the maxilla extended from the left canine (23) to right canine (13) while the posterior region of the maxilla extended from canine to the maxillary tuberosity.

The size of the lesion was measured in centimetres along the widest diameter of the lesion from one border to the opposite border. Radio-density was classified as either radiolucent, radio-opaque and mixed (radiolucent and radio-opaque in appearance). Lesions were further classified as either unilocular in appearance whereby only one compartment is present or multilocular whereby the lesion appears to be formed of many adjacent compartments within the bone.

The dentition affected by the lesion was recorded to demonstrate the extent of the lesion and whether it crosses the midline. The effect of the lesion on the cortex of the mandible was also noted to determine the expansive nature of the lesion. Signs of root resorption were also documented to demonstrate the aggressive nature of the lesion.

**4.6. Data analysis**

Data were collected on Excel worksheets. Data were analysed using Epi Info® 2000 by student's unpaired t-test to compare the findings and to correlate these findings with different parameters such as age, gender, ethnicity, anatomical location of lesion, size of lesion, radiodensity, whether there were loculations and effects on adjacent structures.



**4.7. Ethics**

This was a retrospective case analysis of patient records from Departments of Maxillo-Facial and Oral Surgery and Diagnostics and Radiology, University of the Western Cape Oral Health Centre.

No identifiable patient data were recorded such as their name or date of birth. Only the patient’s record number was noted for reference purposes. All records were stored on a

password protected computer. Printed information was also stored in a locked office. All personal identifiers were hidden when the data is published.

The research protocol for this study was presented at the Faculty of Dentistry of the University of the Western Cape research committee and was approved by the Biomedical Science Research Ethics Committee (BM19/2/7) of the University of the Western Cape (Appendix 3). Permission to access records was obtained from the Dean, Faculty of Dentistry, University of the Western Cape (Appendix 4).

# Chapter 5

# RESULTS

A total 35 cases were diagnosed with Solitary bone cyst while seven cases were diagnosed with Aneurysmal bone cyst from 1970 to 2018 at the Departments of Maxillo-Facial and Oral Surgery and Diagnostics and Radiology, University of the Western Cape Oral Health Centre. Of these cases, only 32 Solitary bone cyst cases and five Aneurysmal bone cyst cases had complete demographic information and radiographs. The other cases were excluded from this study.

## 5.1. Demographic data

The ages of patients at time of diagnosis of SBC ranged from 13 to 51 years with a mean age of 21 years for this sample, while the age ranges for ABC were 12 to 36 years with a mean age of 22 years. The most affected age group with SBC was 11 years to 20 years group with 78.57% of patients. The majority of SBC patients were males (58.62%) while four out of five ABC patients were females (80.0%).

In regards with ethnicity, most SBC patients were of Caucasian and mixed race (66.67%) while black Africans were the least affected by this condition (33.33%). Mixed race patients were also most affected by ABC (80.0%).

### 5.2. Clinical presentation

The majority of SBCs in this sample were discovered incidentally on pantomographs during routine dental examination. Only two SBC cases were symptomatic (6.2%) while 18.75% presented with swelling. Surprisingly, no patients reported history of trauma to the jaws. Similarly, only two ABC patients were symptomatic and one case presented with swelling. No history of trauma was noted for all ABC patients.

### 5.3. Site

All cases of SBC presented in the mandible (96.8%) with exception of one case which occurred in the maxilla. The canine region was the most frequently involved area with SBCs (Figure 1). Eleven (34.4%) SBC cases crossed the midline (Figure 2) while only one case presented with bilateral SBCs (Figure 3). All SBC lesions were located above the mandibular canal. On the other hand, ABCs most commonly involved the mandibular posterior regions (ramus and molar).

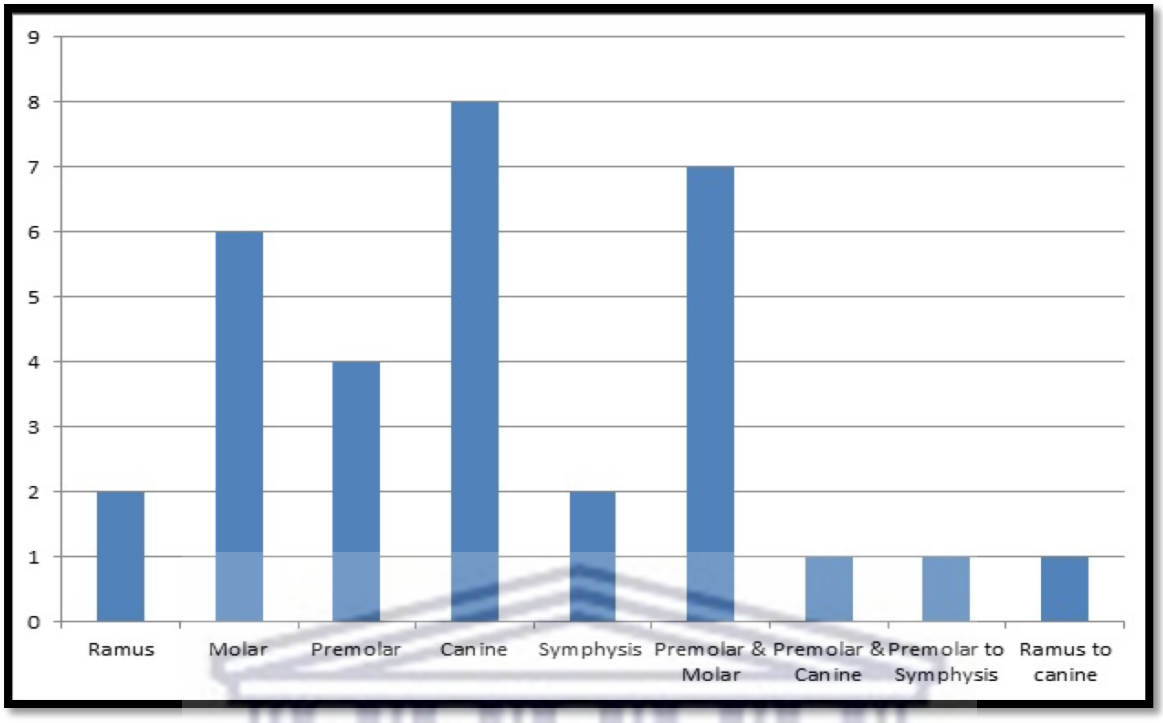



**Figure 1:** Graph showing regional distribution of SBCs in the jaws.

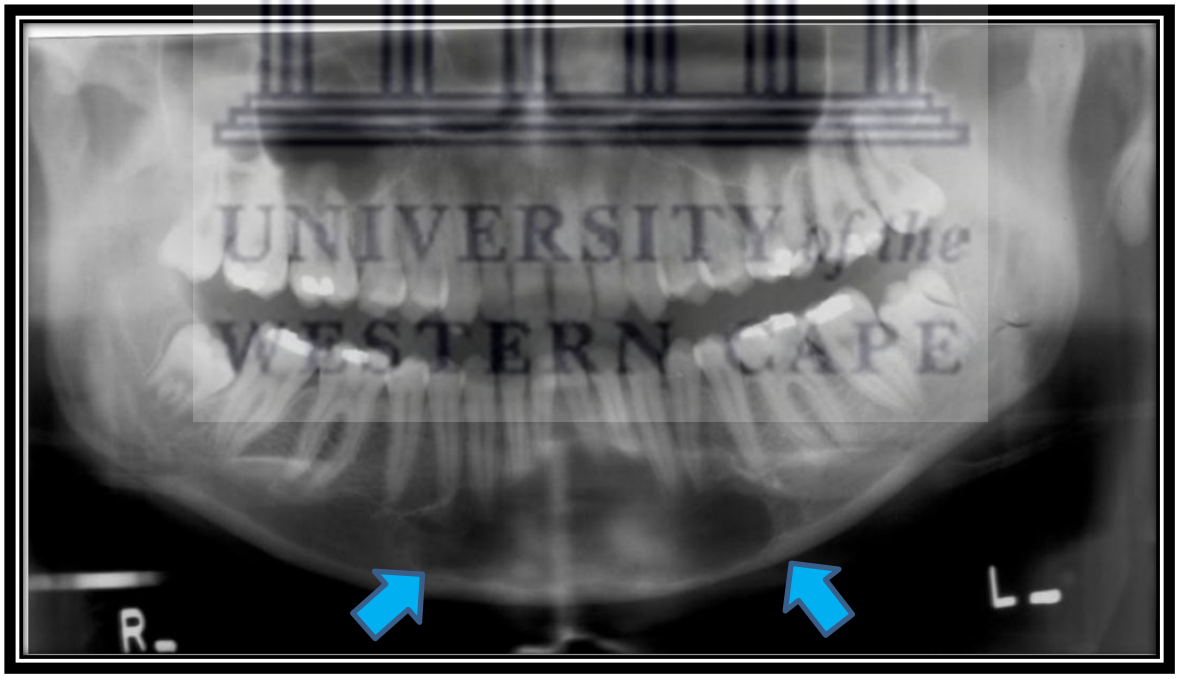



**Figure 2:** Pantomograph showing SBC in the anterior mandible region and crossing the midline.

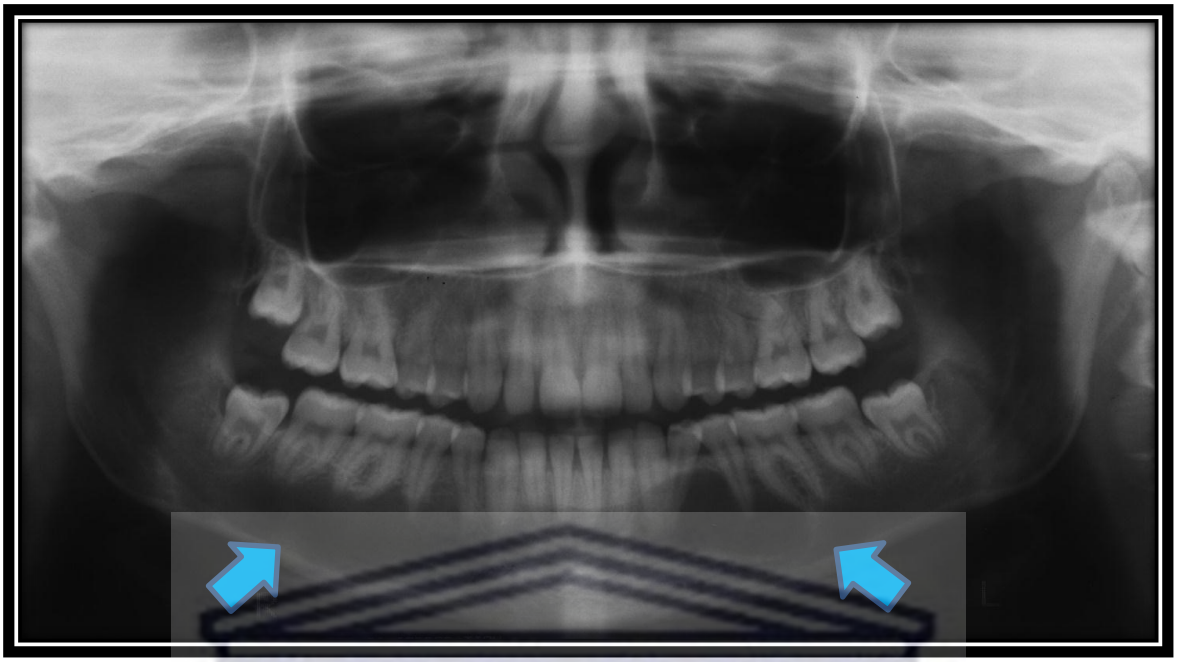


**Figure 3:** Pantomograph of patient with bilateral SBCs.

### 5.4. Radiographic features

#### 5.4.1. Dimension

SBCs have been described to have greater growth in an anterior-posterior dimension than superior-inferior direction. In this case series, 18 cases (56.3%) showed greater anterior-posterior growth on pantomographs.

The majority ABCs (80.0%) was not eccentrically located within the mandible while ballooning was present in four cases (80.0%). Four ABC lesions showed intra-lesional radio-opaque foci (Figure 4).

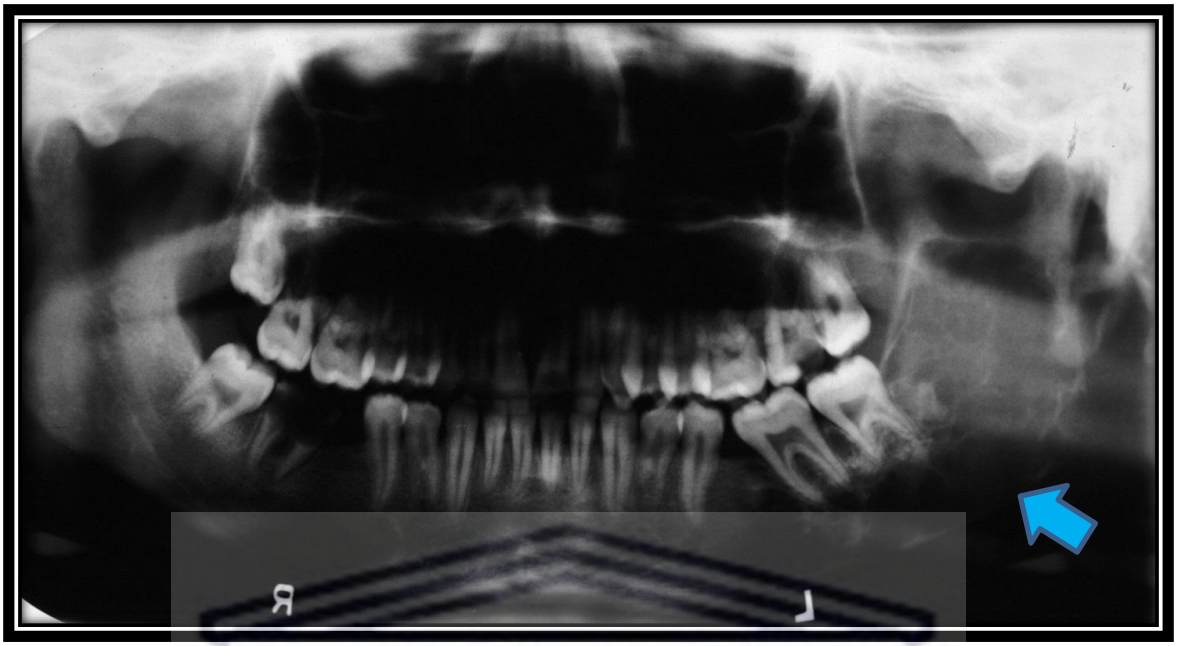


**Figure 4:** Pantomograph showing ABC in left posterior mandible with mixed density and intra-lesional radio-opaque foci.

### 5.4.2. Radio-density

The majority of SBCs (75%) in this study appeared as radiolucent on pantomographs (Figure 5). The rest of SBCs were mixed in density. All ABC lesions were of mixed density.

Twelve SBC lesions (37.5%) appeared over-exposed in the lesion on pantomographs. Only 5 (15.6%) SBC lesions showed signs of expansion of the cortex of the mandible.

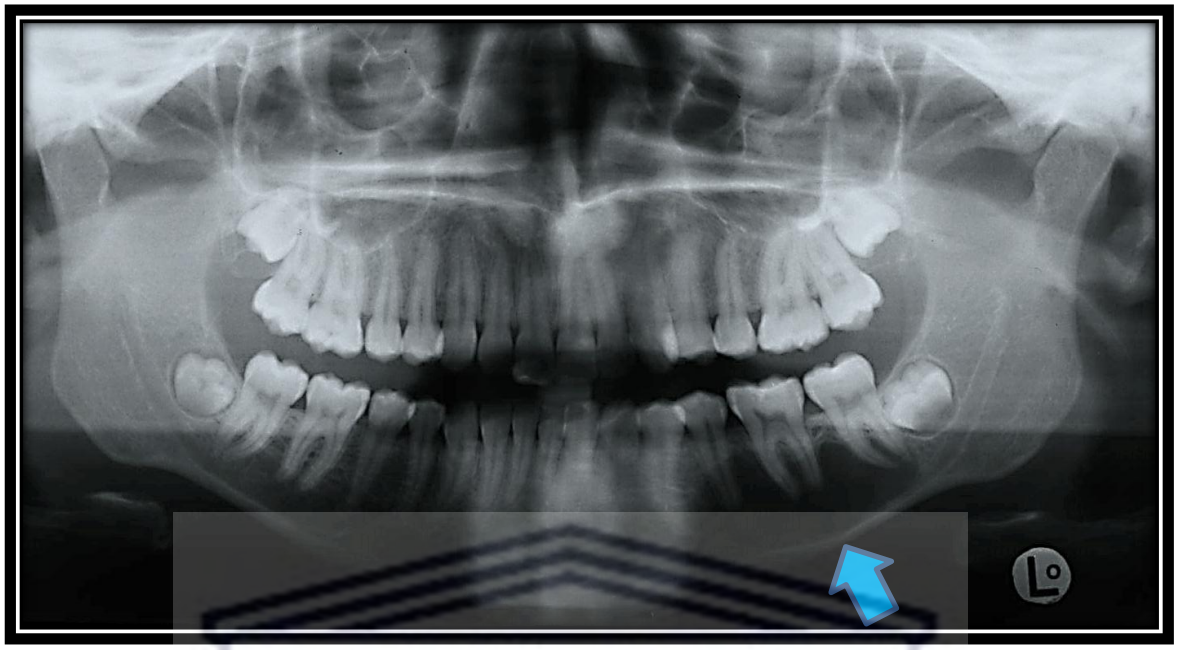


**Figure 5:** Pantomograph showing radiolucent SBC in left body of the mandible (arrow).

### 5.4.3. Shape

The shape of SBCs has been described as cone or V shaped, ovoid, round or irregular on radiographs. In this sample, a large number of SBCs appeared as cone/V-shaped or ovoid on pantomographs, however there were also a substantial number of round or irregular shaped SBCs (Table 1). Hence, the shape of SBCs in this sample differed widely and no pattern of presentation could be deduced.

**Table 1:** Summary of shape of SBC lesions in jaws.

| Shape | No of SBC | Percentage |
|---|---|---|
| Cone or V shaped | 9 | 28.1 |
| Irregular | 7 | 21.9 |

| | | |
|---|---|---|
| Ovoid | 9 | 28.1 |
| Round | 7 | 21.9 |
| Total | 32 | 100.0 |

### 5.4.4. Locularity

The majority of SBC lesions (81.3%) appeared unilocular on pantomographs (Figure 6) while only six cases (15.9%) appeared as multilocular lesions. All multilocular SBCs occurred in the posterior regions of the mandible. All multilocular SBCs presented in patients below 20 years of age (mean age= 17.5 years).

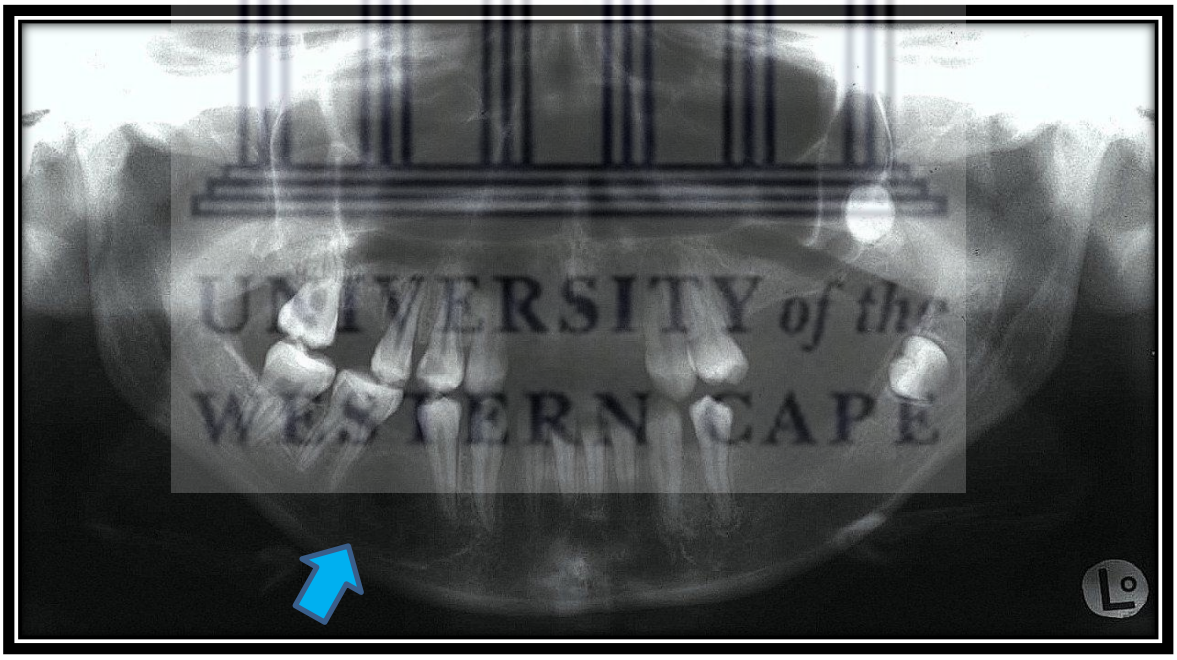



**Figure 6:** Pantomograph showing unilocular SBC in right premolar region of the mandible (arrow).

All ABCs showed honeycomb appearance on pantomographs while four lesions presented in the stabilizing phase (soap bubble) (Figure 7) and one case presented in the growth phase (bone destruction with internal septae).

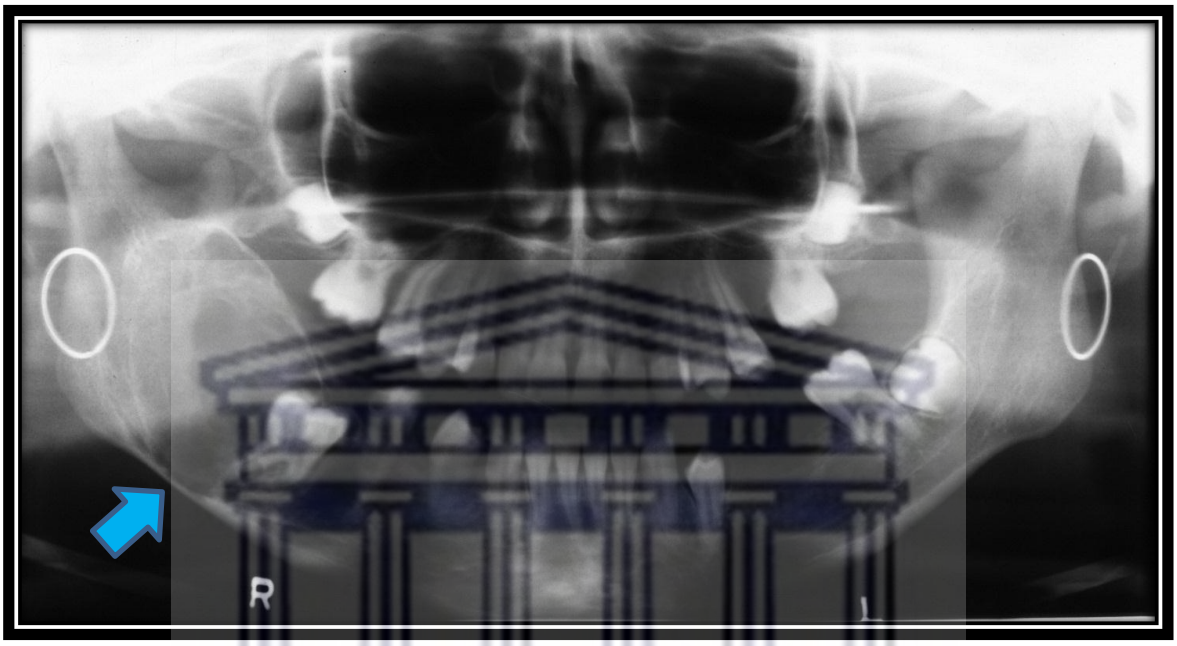



**Figure 7:** Pantomograph showing ABC in right angle and ramus regions of mandible with soap bubble appearance.



### 5.4.5. Margins of lesion

Most SBC lesions presented with well-defined borders (71.8%) and were easily identifiable from healthy surrounding bone. The presence of sclerotic rim was present in 31.2% (Table 2).

**Table 2:** Distribution of margins of SBCs.

| Cortication | No of SBCs | Percentage |
|---|---|---|
| Well corticated | 4 | 12.5 |

| | | |
|---|---|---|
| Delicate sclerotic rim | 10 | 31.25 |
| Lesser cortication | 9 | 28.12 |
| No cortication | 9 | 28.12 |
| Total | 32 | 100 |

All ABCs presented with well-defined margins (Figure 8) with only one lesion presenting with periosteal reaction.

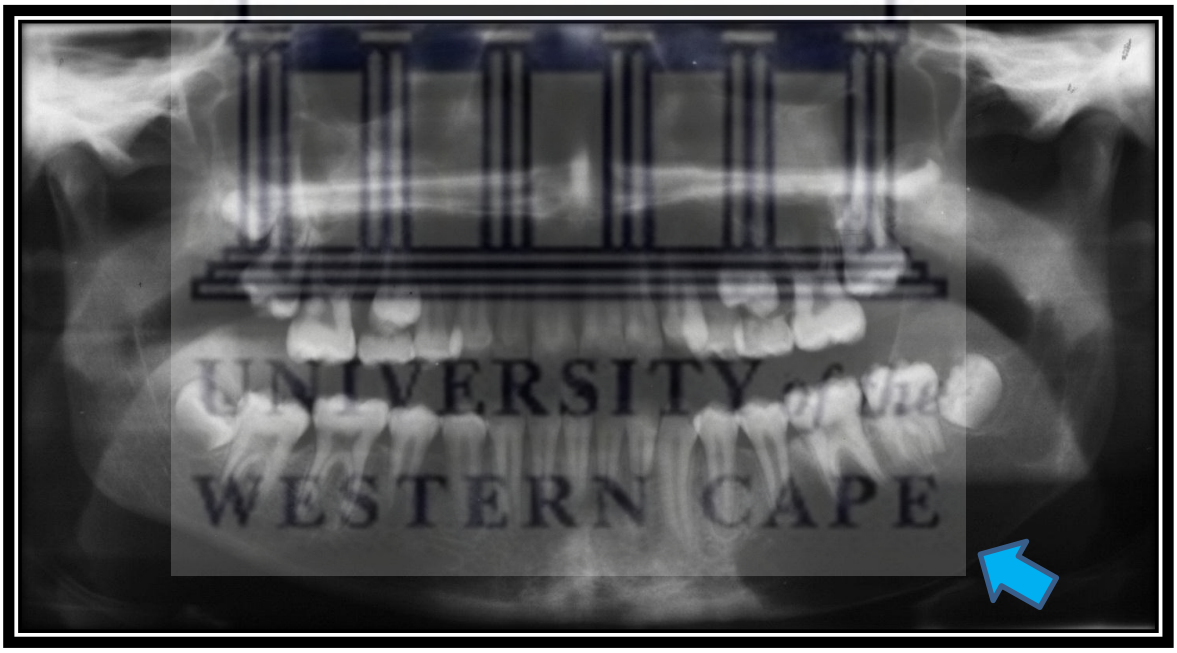



**Figure 8:** Pantomograph showing well-defined ABC in the left mandible.

### 5.4.6. Root resorption

Most SBC lesions in this study did not show signs of root resorption. There were only three cases (9.4%) which showed signs of root resorption on the dentition in close asso-

ciation with the lesion (Figure 9). SBC lesions which caused root resorption (mean size= 90.3mm) were significantly ($P< 0.0001$) larger in diameter than lesions which did not cause root resorption (mean size= 36.6mm). Only one ABC caused root resorption.

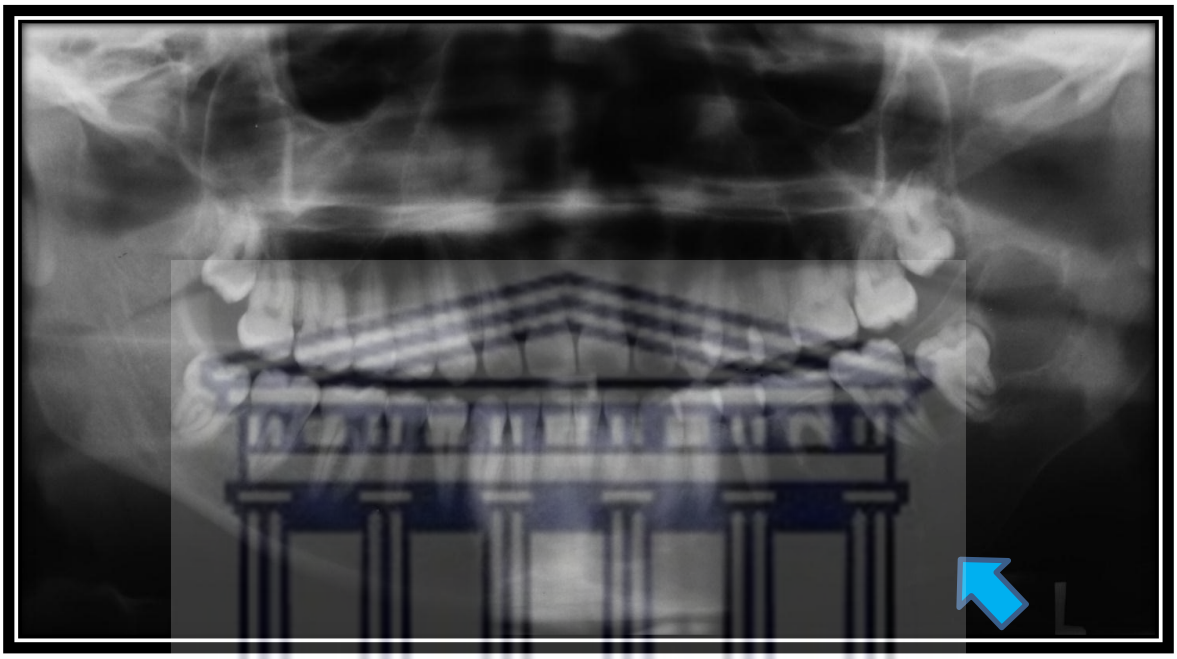


**Figure 9:** Pantomograph showing multilocular SBC with root resorption of the left lower first molar.



**5.4.7. Effects on adjacent structures**

The lamina dura of dentition in close relation with SBCs showed signs of resorption in 20 cases (62.5%) of lesions. SBCs displaced the mandibular canal in 12 cases (37.5%) and caused resorption of the cortical line around the canal in 15 cases (46.8%) (Figure 10). Twenty-four SBC lesions (77.4%) scalloped up between the roots. There were no previous signs of trauma on radiographs for all SBC lesions. None of the SBCs and ABCs were associated with any other pathological lesions detected on pantomographs or histopathology.

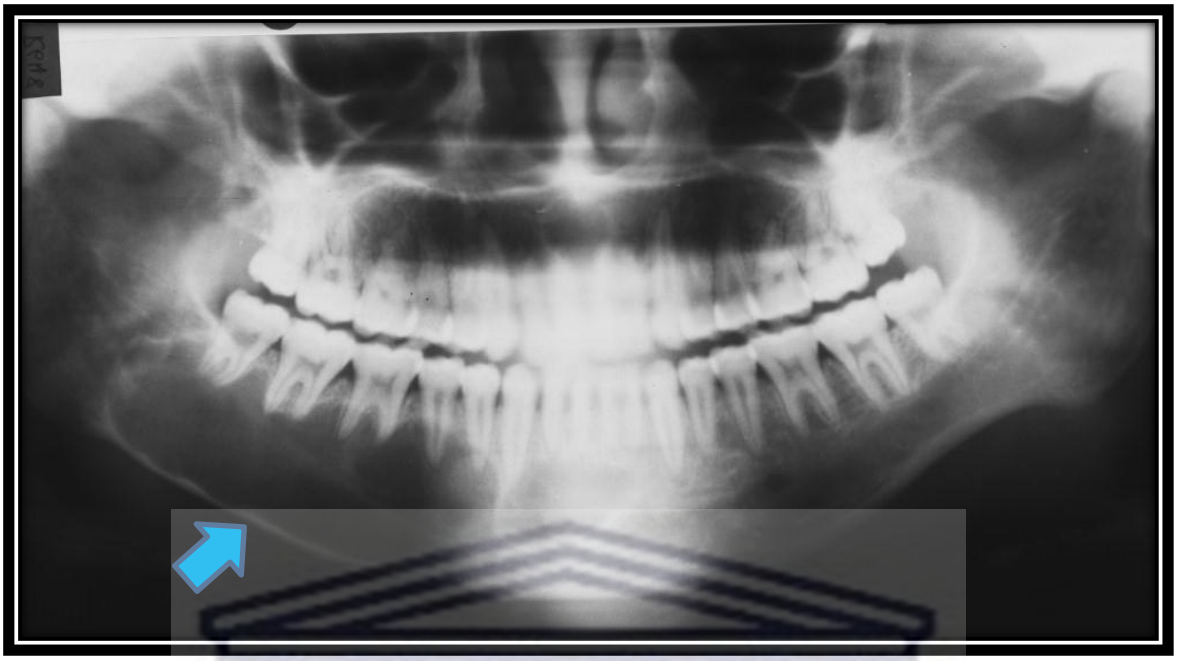


**Figure 10:** Pantomograph showing SBC with expansion of cortex and resorption of the cortical line around the inferior alveolar canal (arrow).

Expansion of the mandibular cortex was present in three ABC cases (60.0%) while thinning of the cortex was more prevalent (80.0%). The mandibular canal was displaced in 80% of cases while all ABC lesions caused resorption of the cortical line around the canal.



### 5.4.8. Size of lesions on pantomographs

SBC lesions were measured on pantomographs along their longest diameter to determine the extent of growth. The size of SBC lesions ranged from 12 mm to 125 mm (mean size= 42 mm). ABC lesions ranged from 36 mm to 89 mm in size (mean size= 60 mm) and were larger in size than SBC lesion.

On average, SBCs in the posterior regions of the jaws (mean size = 38.86 mm) were marginally larger in diameter than lesions in the anterior regions (mean size = 34.3

mm). However, this difference did not show a statistical significance (*P*= 0.54). On the other hand, multilocular SBC lesions (mean size= 70.4 mm) were significantly (*P*= 0.0044) larger in size than unilocular lesions (mean size= 36.8mm).

ABCs (mean size= 60 mm) were on average larger than SBCs on pantomographs with range from 36 mm to 89 mm.

**5.5. Initial Diagnosis**

The initial clinical and radiographic differential diagnosis of SBC lesions included ameloblastoma, traumatic bone cyst, odontogenic keratocyst and proliferative osteomyelitis. The majority of lesions (66.6%) were diagnosed as traumatic bone cyst. This is indicative of their well-known clinical and radiological feature that makes its differential diagnosis fairly accurate.

ABCs were more difficult to diagnose based on clinical and radiographic feature alone. None of the ABC lesions were initially diagnosed correctly. Common lesions as part of ABC differential diagnosis included odontogenic keratocyst, ameloblastoma, myxoma, giant cell lesions and osteitis.



**5.6. Management and recurrence**

Most SBCs were managed by surgical exploration (62.5%) and curettage (37.5%). In majority of cases (62.5%), a fluid filled cavity was found. In some cases, an empty cavity was found. None of the SBCs recurred following surgical management (Figure 11).

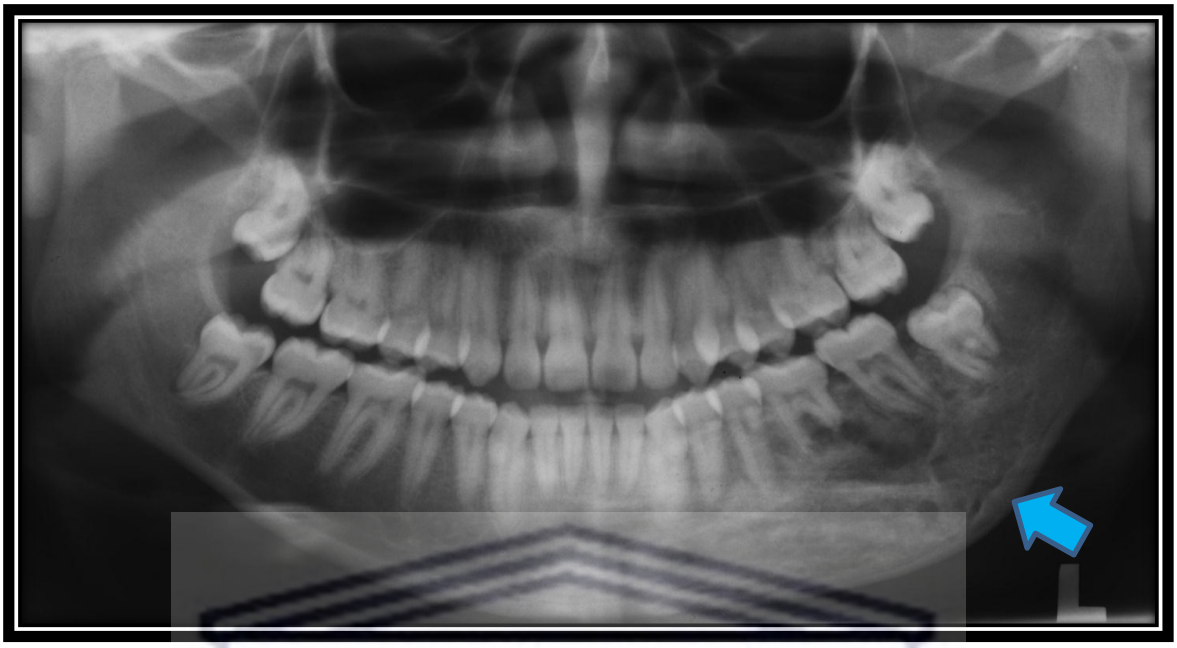


**Figure 11:** Six-month post-operative pantomograph of same patient as in Figure 9 following curettage. Note new bone formation in the left posterior mandible.

All ABCs were managed with surgical exploration of lesions with the majority of lesions showing dark venous blood. In one lesion, marginal resection of the mandible was performed without any recurrence (Figures 12 and 13). Angiography was not done on any of the lesions while none of the lesions recurred following treatment.

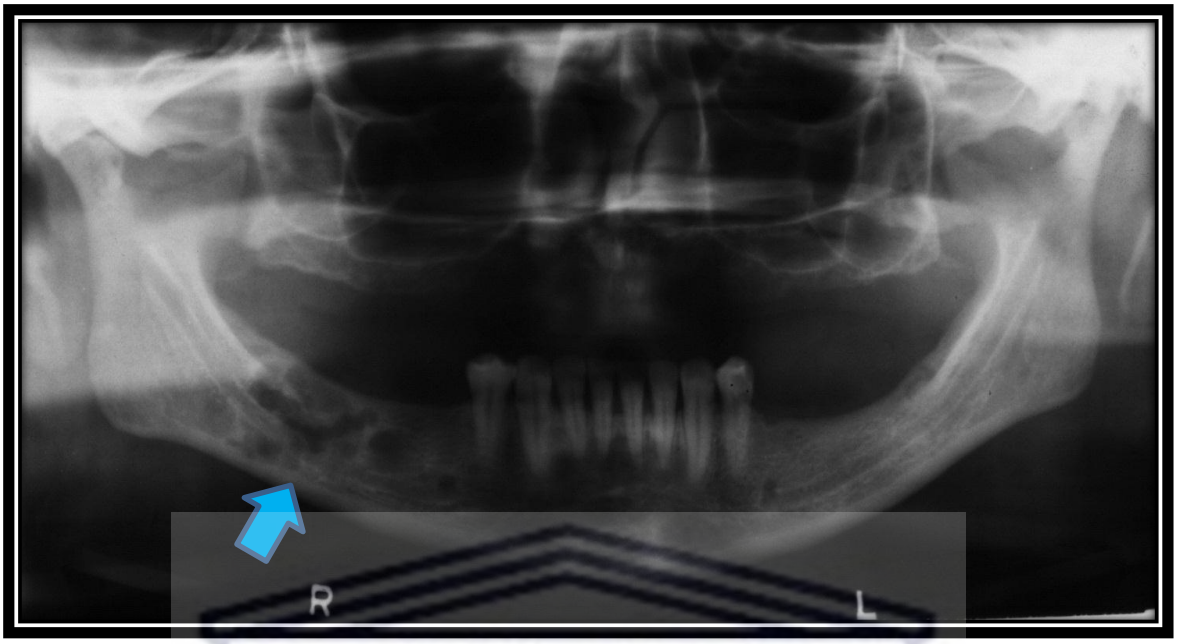



**Figure 12:** Pantomograph showing multilocular ABC in the right body of the mandible preoperatively (arrow).

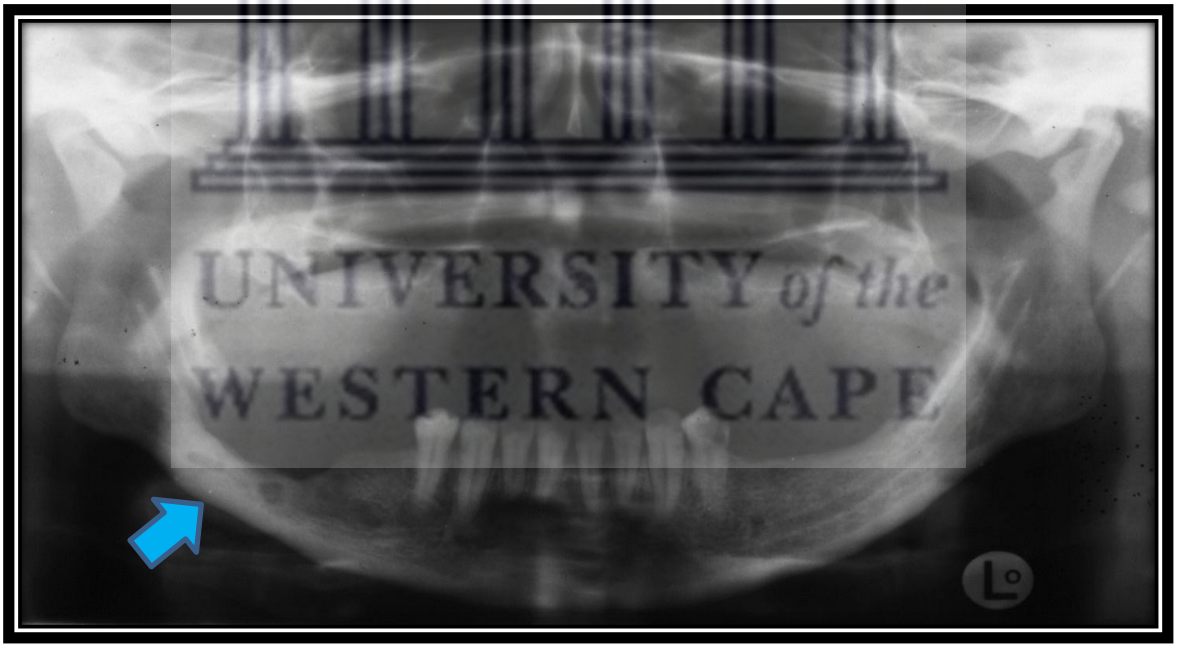


**Figure 13:** Pantomograph showing same patient as above following marginal resection of ABC in the right body of the mandible (arrow).

# Chapter 6

# DISCUSSION

Both ABC and SBC are reported to be rare lesions in the jaws with the majority of lesions are presented in the literature as case reports. There are very few reports addressing both lesions in the same study, as most previous studies report separately on the two lesions even though they have similar presentation.

## 6.1. Demographics

The incidence of SBC and ABC in the jaws is reported to be rare. ABC incidence among jaw cysts was reported as 0.4% in a South African study (Struthers and Shear 1984) and 0.15% in the United Kingdom (Jones and Franklin 2006). Similarly, the incidence of SBC ranged from 0.5-1% of all jaw cysts internationally. In South Africa, only 35 cases have been recorded by Shear and Speight (2007) over a 46-year period which equated to 1% of all jaw cysts in that sample. In this sample, the frequency of these non-epithelial cysts in the jaws was also low (0.2%).

The mean ages of patients affected by ABC and SBC were similar in this study with both lesions affecting patients in their second decade of life. Sun *et al.* (2009) reported that 72.8% of ABC occurs in the first two decades of life while more than 50% of SBC is seen in the second decade (Howe 1965; Hansen *et al.* 1974). Hence, it can be deduced that both these non-epithelial cysts affect mainly young adults.

Previous studies reported males as most affected by SBC while both genders were equally affected by ABC (Kransdorf and Sweet, 1995; Flores *et al*. 2017). However, in this sample, the majority of SBC patients were males while females were predominantly affected by ABC. These findings could be related to the epidemiological variations in different regions of the world as well as the demographics of this sample, which was composed of caucasian, mixed and black African patients while previous studies involved predominately caucasian patients.

### 6.2. Clinical features

Clinically, Shear and Speight (2007) reported that 35% of SBC cases had a swelling of the mandible which mostly involved the buccal cortex. Only rarely was the lingual cortex involved. In 67% of cases, the associated teeth were found to be vital. Hansen *et al.* (1974) reported 72% of cases were completely asymptomatic. Beasly (1976) reported a similar ratio of asymptomatic cases (77%) and found that 27% reported a history of trauma. In a review by Kaugars and Cale (1987), they noted that 25.6% of patients were symptomatic. These reported features were similar in this study with the majority of SBCs discovered incidentally and only 18.75% presented with swelling. Trauma was not reported to be a major attributing factor.

In the jaws, the most common manifestation of ABCs is a rapidly enlarging firm painless swelling. Pain is reported in fewer than half of the cases. Sun *et al.* (2009) analysed 92 cases in a literature review and found that 54.7% of cases were painless and 43.2% were painful. Bone swelling was the most frequent clinical sign. Flores *et al.* (2017) reported painless facial asymmetry in nine out of 12 cases involving ABC while paraesthesia was rarely noted. In this study, only two ABC patients (40%) were symp-

tomatic and one case presented with swelling (20%). As with SBCs, no history of trauma was noted for all ABC patients in the current study.

### 6.3. Location

In regards with the location of the presentation, SBC overwhelmingly affected the mandible with greater than 95% incidence (Kaugars and Cale 1987; Copete *et al.* 1998). The mandibular parasymphyseal and posterior regions are most widely affected (Chadwick *et al.* 2011). Flores *et al.* (2017) reported in their 42 SBC sample, that all the cases were found in the mandible; 29 cases in the posterior mandible, 10 in anterior region, and three cases in other areas. The findings in this study were very similar to what was previously reported. One case in this sample presented with bilateral SBCs which have been reported in the literature to be very rare.

ABCs on the other hand have higher incidence in the mandible (68.5%) than the maxilla (31.5%) with the mandibular ramus (31.3%) and posterior regions (24.1%) being most frequently involved (Sun *et al.* 2009). Flores *et al.* (2017) reported 83.3% of ABC lesions in the mandible and only two cases (16.7%) in the maxilla. In this study, all ABCs occurred in the mandible with the mandibular posterior regions being most commonly affected.

### 6.4. Radiological features

Radiographically, the majority of SBCs are radiolucent in appearance with a minority having internal bony trabeculae. Copete *et al.* (1998) performed radiographic analysis of 44 SBC cases and reported 61% of their sample was completely radiolucent, 32% had bony trabeculae and 7% diffusely opaque. The authors described a characteristic cone-

shaped (or dome-like shape) radiographic morphology. Sixty-six of the cones were reported to point anteriorly towards the midline, particularly in larger lesions. The dome-like shape features an upward scalloping shape between the adjacent roots of the teeth. This is considered to be a highly suggestive feature but not diagnostic of SBC (Copete *et al.* 1998). The majority of SBCs (75%) in this study appeared as completely radiolucent with ovoid and cone/V-shaped. No specific shape pattern could be identified in this sample, as many SBC lesions presented with irregular shapes. This makes diagnosis of these lesions to be more challenging on pantomographs. Advanced imaging has been describe to be more useful in diagnosing these irregular lesions.

Radiographic features of ABCs are variable depending on the type of lesion. The vascular type is generally more destructive and less defined while the solid type is less destructive and more well-defined (Motamedi *et al*, 2014). ABC lesions may appear as unilocular or multilocular radiolucencies with variably defined borders and discernable expansion of the bony cortex. In our study, four of the five cases caused ballooning of the mandible.

Flores *et al.* (2017) described the radiological features of 12 ABC lesions. Most of the lesions were described as radiolucent and one lesion was reported with a radiopaque, ground glass appearance. Some of the cases had multilocular appearance while three cases were described with poorly defined borders. Motamedi *et al*. (2014) reported their entire sample of 120 ABC lesions to be radiolucent. All ABC lesions in this study were of mixed density and presented with well-defined borders which may have been due to the stage at which the lesions presented and were diagnosed.

The size of ABC lesions in this study (mean size= 60 mm) were considerably larger on pantomographs than in the study by Sun *et al.* (2009) who reported a mean size of 48mm. Four cases displayed cortical bone reaction in their study while only one lesion displayed periosteal reaction in this sample. The larger size of lesions in our sample could be possibly attributed to the late and incidental presentation of patients to our oral health care centre. Also, some of the patients in this study were referred from distant areas in South Africa and nearby provinces (including Northern Cape, Eastern Cape and Free State). This would have, most likely, delayed their presentation for further management.

SBCs infrequently cause expansion of the cortices of the mandible due to their slow growth pattern. Flores *et al.* (2017) reported 12.8% of cases causing expansion which was similar to the findings in this study (15.6%).

**6.5. Effect on adjacent structures**

Scalloping is a prominent characteristic finding in SBC (68% reported by Copete et *al.* 1998). Scalloping appears both between teeth and away from the teeth. The presence of scalloping in this sample was slightly higher (77.4%). Chaldwick *et al.* 2011 reported that occasionally, the lamina dura can be lost in dentition in close association with SBCs (11.8%). In our sample, 20 cases (62.5%) showed resorption of lamina dura, which is much higher than reported. This may have been due to the larger size of SBC lesions in this study.

Root resorption is not a known feature of SBC and ABC in the jaws, most likely due to the slow expansion of these lesions as well as their position below the mandibular canal which is far away from the roots of dentition. Flores *et al*. (2017) reported only 4.7% of

SBCs showing signs of root resorption. In this sample there was higher incidence of root resorption at 9.4%. We were able to demonstrate that this might have been due to the larger size of SBC lesions in this study. The incidence of root resorption caused by ABC is similar to that SBC. Sun *et al.* (2009) reported 8.1% incidence of root resorption while in this study it was higher at 20%.

Mandibular canal displacement is also a known radiographic feature of SBCs. Flores *et al*. (2017) reported that two cases (4.7%) of their sample caused displacement of the canal. In this sample, 12 SBCs (37.5%) caused displacement of the canal which is higher than previously reported. This may be attributed to the larger size of lesions in this sample compared to the sample of Flores *et al*. (2017).

**6.6. Associated lesions**

El-Deeb *et al.* (1980) reported pre-existing pathology in 11 ABC lesions with an incidence rate of 21%. The most commonly encountered lesions in association with ABCs were ossifying fibroma, cementifying fibroma, fibrous dysplasia and giant cell granuloma. Sun *et al.* (2009) reported 15.2% of 92 jaw ABCs were secondary in nature. Eight cases were associated with fibrous dysplasia. In a previous South African study, Struthers (1980) and Struthers and Shear (1984B) reported that they could identify associated lesions in 33 cases. Twenty-four of them were associated with giant cell granuloma, four fibrous dysplasia, two ossifying fibroma, two cementifying fibroma, and one osteosarcoma. In our sample of five ABCs, no associated pathological lesions were identified on radiological or histopathological specimen.

### 6.7. Diagnosis

In a sample of 12 ABCs, Flores *et al.* (2017) reported that six were diagnosed through excisional biopsy and six through incisional biopsy. Three lesions were reported to be empty cavities on aspiration. Blood discharge was found in two cases, and blood-yellowish liquid was present in one case. One case was described by the authors to have a thick capsule and another was described with "soft tissue" content. All ABCs in this sample were managed with surgical exploration of lesions with the majority of lesions showing dark venous blood. Surgical exploration of these lesions in our department has shown to be an acceptable method of aiding in the diagnosis of these lesions.

Bony septa can be found and can lead to misdiagnosis because of the multilocular appearance of SBC. Oval, irregular or round borders are possible as well. Teeth are generally found to be vital. Root resorption, loss of lamina dura, cortical expansion and cortical thinning are seen in minority of cases (Wong-Romo *et al.* 2016).

SBCs should be included in the differential diagnosis list of radiolucent lesions in the jaws along with dentigerous cysts, ameloblastomas, odontogenic keratocysts, focal osteoporotic bone marrow defect, central giant cell granuloma, intraosseous vascular malformations, and others. Despite the majority of SBC lesions in this sample were not associated with impacted or unerupted teeth, ameloblastoma and odontogenic keratocysts were the most commonly cited lesions in the differential diagnosis, particularly when a multilocular lesion was present on the pantomograph.

### 6.8. Surgical management

Upon surgical exploration, SBCs are frequently found as an empty bony cavity. In some cases, there is blood, serosanguineous, or serous fluid found. Occasionally a thin membrane, granulation tissue, or blood clot can be found. Flores *et al.* (2017) reported in the 42 cases of SBCs analysed, that 25 cases were empty on aspiration biopsy, 11 cases had blood fluid content, and one case had clear liquid. During surgical exploration, fragments described as "soft tissue" were found in five cases. In this study the majority of SBC cases (62.5%) presented with a fluid filled cavity. In some cases, an empty cavity was found.

It is recommended to curette the bony margins and submit these tissues for histological examination to rule out other lesions. The surgical exploration with or without curettage usually enhances bone regeneration which is visible usually within 12-17 months. Higher recurrence is seen with lesions arising with association of cement-osseous dysplasia or when there are multiple lesions. Some authors reported higher recurrence rates among lesions exhibiting scalloped margins, loss of lamina dura, nodular bone expansion, and multiple lesions. Some authors recommend packing materials to reduce recurrence, but further studies are needed. Prognosis is generally very good (Traub *et al.* 2016).

### 6.9. Recurrence

According to one comprehensive literature review approximately 13% of reported SBC lesions recurred within two years (Sun *et al.* 2009). The recurrence rate of 13.3% was similar in both surgical techniques i.e. curettage (15.2%) and resection (11.8%). It is believed that the recurrence occurs due to incomplete removal of the lesion or of the co-

existing lesion (Sun *et al.* 2009). The non-recurrence of any lesions in this study could be explained by the absence of any associated lesions with SBCs. Another plausible explanation of the lack of recurrence of these lesions in the sample is that some lesions may have recurred but the patients may not have returned to our department due to distant outlying areas, limited transportation and the limited access to healthcare.

Chaldwick *et al.* 2011 presented two classifications of SBCs according to their recurrence rate. The first one is low in recurrence, which is characterised with intact lamina dura and smooth margins with no bony expansion. The second one is higher in recurrence and it is characterised with absent lamina dura, possible root resorption, scalloped borders, nodular expansion or the presence of radiopaque mass or multiple lesions.

# Chapter 7

# LIMITATIONS OF THIS STUDY

Since this was a retrospective record-based study, there was no standardised radiographic quality, as a number of panoramic machines were used over the years with variations of magnification and exposure parameters for each patient.

Due to the period the study covered, some information was missing in some records. In older records, it was difficult to ascertain presenting clinical features due to incomplete notes. These cases were excluded from the study.

Another limitation of this study was the absence of availability of advanced imaging modalities such as computed tomography, cone beam computed tomography and magnetic resonance imaging. The lack of accessibility of these advanced imaging modalities in South Africa's public health care sector is normally due to the high-related costs and lack of availability. In addition, many of the cases in this sample presented prior to the arrival of advanced imaging at the public hospitals in South Africa.

A major drawback of this retrospective study is the lack of follow-up for some cases. An explanation for the limited periodic follow-ups is the inability of patients to return to the Oral Health Centre as several patients reside long distances away and cannot afford transportation costs.

# Chapter 8

# CONCLUSION

ABC and SBC are rare bony pathologies in the jaws with limited case-series studies in the literature and there is lack of comparative studies on the two lesions especially in African populations. Their clinical and radiographic presentations are widely varied and their behaviour is atypical. The aetiology of these lesions is still largely unknown while their recurrence is varied in different population groups. Therefore, this study aimed to highlight the varied clinical, radiological, and intra-operative characteristics as it is vital to keep these lesions in the list of differential diagnosis especially when facing unconventional osteolytic lesions in the jaws.

The majority of the features of ABC and SBC in this study were similar to those reported in the literature. Exceptions included the larger size of these lesions in this sample and the somewhat varied shape on pantomographs when compared to other populations. Surgical exploration and/or curettage have shown to be an acceptable treatment method for the management of these pseudocysts with no recurrence rate in this sample and excellent prognosis.

# Chapter 9

# REFERENCES


1. Asaumi J, Konouchi H, Hisatomi M, Matsuzaki H, Shigehara H, Honda Y, Kishi K, 2003. MR features of aneurysmal bone cyst of the mandible and characteristics distinguishing it from other lesions. *Eur J Radiol*. 45:108-12.

2. Baqain ZH, Jayakrishnan A, Farthing PM, Hardee P. 2005. Recurrence of a solitary bone cyst of the mandible: case report. *Br J Oral Maxillofac Surg.* 43: 333-5.

3. Beasley JD. 1976. Traumatic cyst of the jaws: report of 30 cases. *J Am Dent Assoc*. 92: 145-52.

4. Bharadwaj G, Singh N, Gupta A, Sajjan AK. 2013. Giant aneurysmal bone cyst of the mandible: A case report and review of literature. *Natl J Maxillofac Surg.* 4: 107-10.

5. Biesecker JL, Marcove RC, Huvos AG, Miké V. 1970. Aneurysmal bone cysts. A clinicopathologic study of 66 cases. *Cancer*. 26: 615-25.

6. Chadwick JW, Alsufyani NA, Lam EWN. 2011. Clinical and radiographic features of solitary and cemento-osseous dysplasia-associated simple bone cysts. *Dentomaxillofac Radiol.* 40(4): 230–235.

7. Clough JR, Price CHG. 1968. Aneurysmal bone cysts. *J Bone Joint Surg Br*. 50: 110-27.

8. Copete MA, Kawamata A, Langlais RP. 1998. Solitary bone cyst of the jaws: radiographic review of 44 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 85: 221-5.

9. Dalley RW. 1999. Fibrous histiocytoma and fibrous tissue tumors of the orbit. *Radiol Clin North Am.* 37: 185-94.

10. Davies AM, Wellings RM. 1992. Imaging of bone tumors. *Curr Opin Radiol.* 4: 32-38.

11. Devi P, Thimmarasa VB, Mehrotra V, Agarwal M. 2011. Aneurysmal bone cyst of the mandible: A case report and review of literature. *J Oral Maxillofac Pathol*. 15: 105-8.

12. El-Deeb M, Sedano HO, Waite DE. 1980. Aneurysmal bone cyst of the jaws. Report of a case associated with fibrous dysplasia and review of the literature. *Int J Oral Surg.* 9: 301-311.

13. Ewing J. Neoplastic Diseases: A Treatise on Tumours. Philadelphia, Saunders; 1940. p. 126-128.

14. Fielding AF, Louden RD, Johnson AL. 1999. Simple bone cyst. *Oral Surg Oral Med Oral Pathol*. 88: 277-8.

15. Flores IL, Hamilton ME, Zanchin-Baldissera E, Uchoa-Vasconcelos AC, Chaves- Tarquinio SB, Neutzling-Gomes AP. 2017. Simple and aneurysmal bone cyst: Aspects of jaw pseudocysts based on an experience of Brazilian pathology service during 53 years. *Med Oral Patol Oral Cir Bucal*. 22: 64-69.

16. Gadre KS, Zubairy RA. 2000. Aneurysmal bone cyst of the mandibular condyle: report of a case. *J Oral Maxillofac Surg*. 58: 439-43.

17. Grecchi F, Zollino I, Candotto V, Gallo F, Rubino G, Bianco R. 2012. A case report of haemorrhagic-aneurismal bone cyst of the mandible. *Dent Res J (Isfahan)*. 9:222–4.

18. Hansen L, Sapone J, Sproat R. 1974. Traumatic bone cysts of jaws. Report of sixty-six cases. *Oral Surg.* 37: 899–910.

19. Hara H, Ohishi M, Higuchi Y. 1990. Fibrous dysplasia of the mandible associated with large solitary bone cyst. *J Oral Maxillofac Surg.* 48: 88-91.

20. Hardee PS, Whear NM, Morgan PR. 1992. Aneurysmal bone cyst of the maxilla: an association with tooth resorption. *J Craniomaxillofac Surg*. 20: 266-269.

21. Harnet J-C, Lombardi T, Klewansky P, Rieger J, Tempe M-H, Clavert J-M. 2008. Solitary bone cyst of the jaws: a review of the etiopathogenic hypotheses. *J Oral Maxillofac Surg*; 66: 2345–2348.

22. Hernandez GA, Castro A, Castro G, Amador E. 1993. Aneurysmal bone cyst versus hemangioma of the mandible. Report of a long-term follow-up of a self-limiting case. *Oral Surg Oral Med Oral Pathol*. 76: 790-6.

23. Hisatomi M, Asaumi J, Konouchi H, Shigehara H, Yanagi Y, Kishi K. 2003. MR imaging of epithelial cysts of the oral and maxillofacial region. *Eur J Radiol*. 48: 178-82.

24. Hoffmeister B, Härle F. 1985. Zysten im Kiefer- Gesichtbereich-eine katamnestische Studie an 3353 Zysten. *Dtsch. Zahnarztl. Z.* 40: 610–614 [In Swiss].

25. Horner K, Forman GH, Smith NJ. 1988. Atypical simple bone cysts of the jaws. I: Recurrent lesions. *Clin Radiol.* 39: 53–57.

26. Horner K, Forman GH. 1988. Atypical simple bone cysts of the jaws. II: A possible association with benign fibro- osseous (cemental) lesions of the jaws. *Clin Radiol*.39: 59–63.

27. Howe GL.1965. "Haemorrhagic cysts" of the mandible. *Br J Oral Surg*. 3: 55–91.

28. Hs CB, Rai BD, Nair MA, Astekar MS. 2012. Simple bone cyst of mandible mimicking periapical cyst. *Clin Pract.* 2: 59.

29. Jacobs MH. 1955. The traumatic bone cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 8: 940-9.

30. Jaffe HL, Lichtenstein L. 1942. Solitary unicameral bone cyst with emphasis on the roentgen picture, the pathologic appearance, and the pathogenesis. *Arch Surg.* 44:1004-25.

31. Jaffe HL. 1950. Aneurysmal bone cyst. *Bull NYU Hosp Jt Dis*. 11: 3-13.

32. Jones AV, Franklin CD. 2006. An analysis of oral and maxillofacial pathology found in children over a 30-year period. *Int. J. Paediatr. Dent*. 16: 19-30.

33. Kalantar MMH. 1998. Aneurysmal bone cysts of the jaws: clinicopathological features, radiographic evaluation and treatment analysis of 17 cases. *J Cranio-MaxilloFac Surg.* 26: 56-62.

34. Kaugars GE, Cale AE. 1987. Traumatic bone cyst. *Oral Surg Oral Med Oral Pathol.* 63: 318–324.

35. Khafif A, Krempl G, Medina JE. 2000. Treatment of giant cell granuloma of the maxilla with intralesional injection of steroids. *Head Neck*. 2: 822–5.

36. Killey HC, Kay LW, Seward GR. Benign Cystic Lesions of the Jaws: Their Diagnosis and Treatment. Edinburgh: Churchill Livingstone; 1977. p. 119-135.

37. Kramer IRH. 1974. Changing views on oral disease. *J R Soc Med.* 67: 271–276.

38. Kransdorf MJ, Sweet DE, 1995. Aneurysmal bone cyst: concept, controversy, clinical presentation, and imaging. *Am J Roentgenol.*164:573-80.

39. Kuttenberger JH, Farmand M, Stoss H. 1992. Recurrence of a solitary bone cyst of the mandibular condyle in a bone graft. *Oral Surg Oral Med Oral Pathol.* 74: 550–556.

40. Lee HJ, Lum C. 1999. Giant-cell tumor of the skull base. *Neuroradiology*. 41: 305–7.

41. Lenz M, Greess H, Baum U, Dobritz M, Kersting-Sommerhoff B. 2000. Oropharynx, oral cavity, floor of the mouth: CT and MRI. *Eur J Radiol.* 33: 203–15.

42. Lenz M. 2000. Imaging of head and neck tumors. *Eur J Radiol*. 33: 151–2.

43. Lenz M. Computed tomography and magnetic resonance imaging of head and neck tumors. Stuttgart: Thieme; 1993. p. 129.

44. Levy WM, Miller AS, Bonakdarpour A, Aegerter E. 1975. Aneurysmal bone cyst secondary to other osseous lesions. Report of 57 cases. *Am J Clin Pathol.* 63: 1–8.

45. Liu B, Yu SF, Li TJ. 2003. Multinucleated giant cells in various forms of giant cell containing lesions of the jaws express features of osteoclasts. *J Oral Pathol Med.* 32: 367–75.

46. Machiels F, De Maeseneer M, Chaskis C, Bourgain C, Osteaux M. 1998. Deep benign fibrous histiocytoma of the knee: CT and MR features with pathologic correlation. *Eur Radiol.* 8: 989-91.

47. Matsumura S, Murakami S, Kakimoto N, Furukawa S, Kishino M, Ishida T, *et al*. 1998. Histopathologic and radiographic findings of the simple bone cyst. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 85: 619-25.

48. Matsuzaki H, Asaumi J, Yanagi Y, Konouchi H, Honda Y, Hisatomi M, *et al*. 2003. MR imaging in the assessment of a solitary bone cyst. *Eur J Radiol Extra*. 45: 37-42.

49. Melrose RJ, Abrams AM, Mills BG. 1976. Florid osseous dysplasia. A clinical-pathologic study of thirty-four cases. *Oral Surg Oral Med Oral Pathol*. 41: 62-82.

50. Mendenhall WM, Zlotecki RA, Gibbs CP, Reith JD, Scarborough MT, Mendenhall NP. 2006. Aneurysmal bone cyst. *Am J Clin Oncol*. 29: 311–5.

51. Minami M, Kaneda T, Ozawa K, Yamamoto H, Itai Y, Ozawa M, *et al*. 1996. Cystic lesions of the maxillomandibular region: MR imaging distinction of odontogenic keratocysts and ameloblastomas from other cysts. *Am J Roentgenol.* 166: 943-9.

52. Motamedi MH, Behroozian A, Azizi T, Nazhvani AD, Motahary P, Lotfi A, 2014. Assessment of 120 maxillofacial aneurysmal bone cysts: a nationwide quest to understand this enigma. *Oral Maxillofac Surg*. 72:1523-30.

53. Motamedi MH, Stavropoulos MF. 1997. Large radiolucent lesion of the mandibular condyle. *J Oral Maxillofac Surg*. 55: 1300–4.

54. Motamedi MH, Yazdi E. 1994. Aneurysmal bone cyst of the jaws: analysis of 11 cases. *J Oral Maxillofac Surg*. 52: 471–5.

55. Neville BW, Damm DD, Allen C M, Chi AC. Oral and maxillofacial pathology. St Louis: Elsevier; 2016. p. 329-34.

56. Okuyama T, Suzuki H, Umehara I, Kuwabara Y, Suzuki S, Takagi M. 1985. Diagnosis of aneurysmal bone cyst of the mandible. A report of two cases with emphasis on scintigraphic approaches. *Clin Nucl Med*. 10: 786–90.

57. Olech E, Sieber H, Weinmann JP. 1951. Traumatic mandibular bone cysts. *Oral Surg Oral Med Oral Pathol*. 4: 1160–72.

58. Oliveira AM, Hsi BL, Weremowicz S, Rosenberg AE, Dal Cin P, Joseph N, *et al*. 2004. USP6 (Tre2) fusion oncogenes in aneurysmal bone cyst. *Cancer Res.* 15:1920-3.

59. Pankey ER, Schaberg SJ, Pierce GL, Williams TP. 1984. Clinicopathologic conference. Case 48, part II: Aneurysmal bone cyst of the mandible. *J Oral Maxillofac Surg*. 42: 118–23.

60. Perrotti V, Rubini C, Fioroni M, Piattelli A, 2004. Solid aneurysmal bone cyst of the mandible. *Int J Pediatr Otorhinolaryngol*. 68:1339-44.

61. Persson G. 1985. An atypical solitary bone cyst. *J Oral Maxillofac Surg.* 43: 905–7.

62. Revel MP, Vanel D, Sigal R, Luboinski B, Michel G, Legrand I, *et al*. 1992. Aneurysmal bone cysts of the jaws: CT and MR findings. *J Comput Assist Tomogr*. 16: 84–6.

63. Rosenberg AE, Nielsen GP, Fletcher JA. Aneurysmal bone cyst. In: WHO classification of tumors: pathology and genetics of tumors of soft tissue and bone. Lyon: IARC Press; 2005. pp. 338–339.

64. Rubin MM, Murphy FJ. 1989. Simple bone cyst of the mandibular condyle. *J Oral Maxillofac Surg*. 47: 1096–8.

65. Ruiter DJ, van Rijssel TG, van der Velde EA. 1977. Aneurysmal bone cysts. A clinicopathological study of 105 cases. *Cancer.* 39: 2231–9.

66. Sapp JP, Stark ML. 1990. Self-healing traumatic bone cysts. *Oral Surg Oral Med Oral Pathol*. 69: 597-602.

67. Schajowicz F. Tumors and Tumor-like Lesions of Bone and Joints. New York: Springer Verlag; 1981. p. 439.

68. Shear M, Speight P. Cysts of the Oral and Maxillofacial Regions. Oxford: Blackwell Munksgaard; 2007. p. 150-155.

69. Struthers PJ, Shear M. 1984. Aneurysmal bone cyst of the jaws. (I). Clinicopathological features. *Int J Oral Surg*. 13: 85 – 91.

70. Struthers PJ, Shear M. 1984. Aneurysmal bone cyst of the jaws. (II). Pathogenesis. *Int J Oral Surg*. 13: 92–100.

71. Suei Y, Taguchi A, Kurabayashi T, Kobayashi F, Nojiri M, Tanimoto K. 1998. Simple bone cyst: investigation on the presence of gas in the cavity using computed tomography. *Oral Surg Oral Med Oral Pathol*. 86: 592-4.

72. Suei Y, Tanimoto K, Wada T. 1994. Simple bone cyst. Evaluation of contents with conventional radiography and computed tomography. *Oral Surg Oral Med Oral Pathol.* 77: 296-301.

73. Sun ZJ, Sun HL, Yang RL, Zwahlen RA, Zhao YF. 2009. Review Article; Aneurysmal Bone Cysts of the Jaws. *Int J Surg Pathol.* 17: 311-22.

74. Tariq MU, Din NU, Ahmad Z, Kayani N, Ahmed R. 2014. . Cementum-like matrix in solitary bone cysts: a unique and characteristic but yet underrecognized feature of promising diagnostic utility. *Ann Diagn Pathol*. 18: 1-4.

75. Telfer MR, Jones GM, Pell GM, Eveson JW. 1990. Primary bone cyst of the mandibular condyle. *Br J Oral Maxillofac Surg.* 28: 340–3.

76. Tillman BP, Dahlin DC, Lipscomb PR, Stewart JR. 1968. Aneurysmal bone cyst: an analysis of ninety-five cases. *Mayo Clin Proc*. 43: 478-95.

77. Toller PA. 1964. Radioactive isotope and other investigations in a case of haemorrhagic cyst of the mandible. *Br J Oral Surg.* 2: 86–93.

78. Traub, F, Eberhardt, O, Fernandez, F. F, & Wirth, T. 2016. Solitary bone cyst: a comparison of treatment options with special reference to their long-term outcome. *BMC musculoskeletal disorders*, *17*, 162.

79. Tsai JC, Dalinka MK, Fallon MD, Zlatkin MB, Kressel HY. 1990. Fluid-fluid level: a nonspecific finding in tumors of bone and soft tissue. *Radiology.* 175: 779–82.

80. Ueno S, Mushimoto K, Kurozumi T, Hirase T, Takasu J. 1982. Aneurysmal bone cyst of the mandible. *J Oral Maxillofac Surg.* 40: 680–3.

81. Urs AB, Augustine J, Chawla H. 2014. Aneurismal bone Cysts of the jaws: clinicopathological study. *J Maxillofac Oral Surg*. 13: 458-63.

82. Velasco I, Cifuentes J, Lobos N, San Martín F. 2012. The unusual evolution of a simple bone cyst in the mandible: A case report. *J Clin Exp Dent*. 4: 132–5.

83. Vergel De Dios AM, Bond JR, Shives TC, McLeod RA, Unni KK. 1992. Aneurysmal bone cyst. A clinicopathologic study of 238 cases. *Cancer*. 69: 2921–31.

84. Wong-Romo G, Carrilo-Teran E, Angeles-Varela E. 2016. Solitary mandibular bone cyst. Case report and literature review. *Revista Odontológica Mexicana*. 20: e112-e119.

85. Wright JM, Vered M. 2017. Update from the 4th Edition of the World Health Organization Classification of Head and Neck Tumours: Odontogenic and Maxillofacial Bone Tumors. *Head and Neck Pathol.* 11: 68-77.

# APPENDIX 1

## Data Collection Sheet (SBC)

| | |
|---|---|
| **1. Library number** | |
| **2. Age** | |
| **3. Gender:** M=1 F=2 | |
| **4. Race:** Caucasian=1 African=2 Indian=3 Coloured=4 | |
| **5. History of trauma:** yes=1 no=2 | |
| **6. Painless swelling:** yes=1 no=2 | |
| **7. Symptomatic:** yes=1 no=2 | **Duration:** |
| **8. Location:** Ramus =1 Molar=2 Premolar =3 Ant of canine =4 Symphysis = 5 | |
| **9. Location Maxilla/mandible:** mandible=1 maxilla=2 both=3 | |
| **10. Cross midline:** yes=1 no=2 | |
| **11. Bilateral:** yes=1 no=2 | |
| **12. Size (mm)** | |
| **13. Dimension: Ant-post >Sup-inferior**: yes=1 no=2 | |
| **14. Overexposed in area of lesion:** yes=1 no=2 | |
| **15. Above inferior alveolar nerve (IAN):** yes=1 no=2 | |
| **16. Expansion of cortex:** yes=1 no=2 | |
| **17. Cortication:** Well corticated=1 delicate sclerotic rim=2 lesser cortication=3 no cortication=4 | |
| **18. Radio-density:** opaque=1 mixed=2 lucent=3 | |
| **19. Loculation:** unilocular=1 multilocular=2 scalloped=3 | |

| | |
|---|---|
| **20. Shape of lesion:** Cone or V shaped=1 Irregular=2 Ovoid=3 Round=4 | |
| **21. Scalloped up between roots:** yes=1 no=2 | |
| **22. Resorption of lamina dura**: yes=1 no=2 | |
| **23. IAN displaced:** yes=1 no=2 | |
| **24. Cortical line of IAN resorbed:** yes=1 no=2 | |
| **25. Signs of trauma: Ortho, fracture, extractions, surgery, displaced teeth, fracture line**: yes=1 no=2 | |
| **26. Root resorption**: yes=1 no=2 | |
| **27. Initial diagnosis:** | |
| **28. Management:** Exploration=1 curettage=2 other =3 none=4 | |
| **29. On surgical exploration:** Empty cavity=1 Fluid=2 Not done=3 | |
| **30. Recurrence:** yes=1 no=2 | |
| **31. Associated pathology– Cemento- or Fibro-osseous lesions: yes=1 no=2** | |

Additional notes: _____

# APPENDIX 2

## Data Collection Sheet (ABC)

| | |
|---|---|
| **1. Library number** | |
| **2. Age** | |
| **3. Gender:** M=1 F=2 | |
| **4. Race:** Caucasian=1 African=2 Indian=3 Coloured=4 | |
| **5. History of trauma:** yes=1 no=2 | |
| **6. History of previous intraosseous lesion:** yes=1 no=2 | |
| **7. Symptomatic:** yes=1 no=2 | **Duration:** |
| **8. Presented with swelling:** Fast=1 slow=2 no=3 | **Duration:** |
| **9. Location:** Mand=1 Max=2 | |
| **10. Location:** Ramus=1 Molar=2 Premolar=3 Ant of canine=4 Symphysis= 5 | |
| **11. Size (mm)** | |
| **12. Eccentrically located:** yes=1 no=2 | |
| **13. Ballooned or blowout distention:** yes=1 no=2 | |
| **14 Honeycomb or soap-bubble appearance:** yes=1 no=2 | |
| **15. Three radiological phases:** Osteolytic phase (nonspecific)=1 Growth phase (bone destruction, internal sep-tae)=2 Stabilizing phase (soap bubble) = 3 | |
| **16. Radio-density:** opaque=1 mixed=2 lucent=3 | |
| **17. Margins of lesion:** well-defined=1 ill-defined=2 | |
| **18. Periosteal reaction:** yes=1 no=2 | |
| **19. Loculation:** unilocular=1 multilocular=2 | |

| | |
|---|---|
| **20. Small radiopaque foci:** yes=1 no=2 | |
| **21. Dentition involved (teeth number)** | |
| **22. Expansion of cortex:** yes=1 no=2 | |
| **23. Thinning or destruction of cortex:** yes=1 no=2 | |
| **24. Displaced inferior alveolar canal:** yes=1 no=2 | |
| **25. Erosion of canal cortex:** yes=1 no=2 | |
| **26. Root resorption**: yes=1 no=2 | |
| **27. Root displacement:** yes=1 no=2 | |
| **28. Time from initial symptoms to presentation** | |
| **29. Initial diagnosis** | |
| **30. Management:** enucleation=1 curettage=2 resection=3 none=4 Other(cryo)=5 | |
| **31. On surgical exploration:** Dark venous blood=1 Blood soaked sponge=2 Not done=3 | |
| **32. Angiography done:** yes=1 no=2 | |
| **33. Recurrence:** yes=1 no=2 | |
| **34. Associated pathology – Intraosseous lesions: yes=1 no=2** | |



Additional notes: _____

# APPENDIX 3
# Ethical approval for study

UNIVERSITY of the WESTERN CAPE

OFFICE OF THE DIRECTOR: RESEARCH
RESEARCH AND INNOVATION DIVISION

Private Bag X17, Bellville 7535
South Africa
T: +27 21 959 4111/2948
F: +27 21 959 3170
E: research-ethics@uwc.ac.za
www.uwc.ac.za

09 May 2019

Dr M Dashti
**Faculty of Dentistry**

**Ethics Reference Number:** BM19/2/7

**Project Title:** Non-epithelial bone cysts of the jaws.

**Approval Period:** 02 May 2019 – 02 May 2020

I hereby certify that the Biomedical Science Research Ethics Committee of the University of the Western Cape approved the scientific methodology and ethics of the above mentioned research project.

Any amendments, extension or other modifications to the protocol must be submitted to the Ethics Committee for approval.

**Please remember to submit a progress report in good time for annual renewal.**

The Committee must be informed of any serious adverse event and/or termination of the study.

*Ms Patricia Josias*
*Research Ethics Committee Officer*
*University of the Western Cape*

# APPENDIX 4

## Permission to access information

Faculty of Dentistry & WHO Oral Health Collaborating Centre,
University of the Western Cape

Private Bag X1, Tygerberg 7505
South Africa
Telephone: +27 21 937 3087/6
Fax: +27 0865156459

The Dean's Office Prof Y Osman Faculty of Dentistry UWC

31 January 2019

Dear Professor Osman

**REQUEST FOR PERMISSION TO ACCESS PATIENT INFORMATION**

I am a registered Master's student in the Department of Maxillo-Facial and Oral Surgery at the University of the Western Cape. My supervisor is Prof JA Morkel and co-supervisor, Dr D Smit.

The proposed topic of my research is "**Non-Epithelial Bone Cysts of the Jaws**".

The objectives of the study are to describe the radiographic features and evaluate the possible association between the demographic profile of patients and histopathological diagnosis in patients presenting with aneurismal and solitary bone cysts.

I am hereby requesting your permission to access patient data in respect of demographic, clinical, radiological and histopathological information. To assist you in reaching a decision, the following ethical considerations will be adhered to:-

The patient's file number and identifiable patient data (names, date of birth, addresses, etc.) will not be recorded in the study.

The data that will be utilised to maintain anonymity will be the patient's record number. This number will be used for record purposed only and will only be kept for the duration

Patient records will be stored on a password-protected computer and printed information will be stored in a locked office.

Radiographs displayed in this study will not jeopardise the patient's identity, and if clinical photos are used to display the lesion, prior consent will be obtained from the patient.

Should you require any further information, please do not hesitate to contact me. Your permission to access this information will be greatly appreciated.

Regards,

_____

**Mahdi Dashti**