

**Residual cysts of the jaws: a retrospective study of
characteristic features and management at the
University of the Western Cape Oral Health Centre**



A mini-thesis submitted in partial fulfilment of the requirements for the degree of Master of Dental Surgery in the Department of Maxillo-Facial and Oral Surgery, Faculty of Dentistry, University of the Western Cape

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**Residual cysts of the jaws: a retrospective study of
characteristic features and management at the University of
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Fadi Titinchi

KEYWORDS

Residual cyst

Inflammatory cyst

Odontogenic cyst

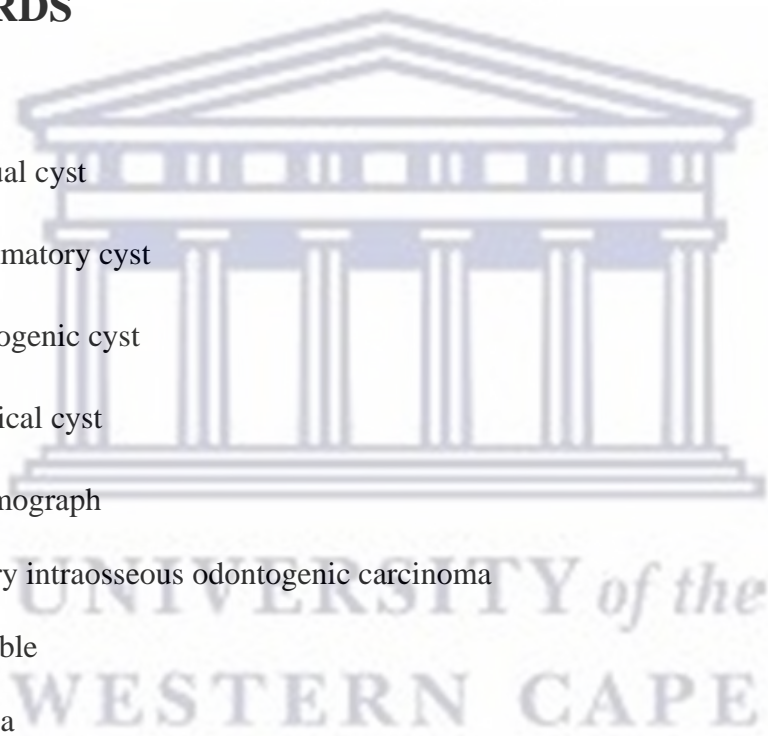
Periapical cyst

Pantomograph

Primary intraosseous odontogenic carcinoma

Mandible

Maxilla



ABSTRACT

Residual cysts of the jaws: a retrospective study of characteristic features and management at the University of the Western Cape Oral Health Centre

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Residual cysts are infrequent inflammatory cysts of the jaws which are of odontogenic origin. They are usually preceded by a radicular cyst which has developed apical or adjacent to a carious tooth. These cysts usually present with similar features as radicular cysts; however, due to absence of the causative tooth, they can mimic more aggressive cysts and tumours on radiographs. Moreover, they have the ability to destroy bone within the jaws without any symptoms.

There is paucity of studies in the literature regarding these cysts as they are often overlooked due to their lack of symptoms and innocent radiographic appearance. Hence, the aim of this study was to describe the clinical, radiological and histopathological features of residual cysts in the jaws as well as analyse their surgical management and recurrence patterns.

The records of 64 histopathologically confirmed residual cysts presenting at the University of the Western Cape Oral Health centre over a period of 30 years were

included in the study. Data recorded included demographics, clinical, radiological and histopathological features as well as surgical treatment and follow-up for any recurrences. Epi Info V7 software was used to perform statistical analysis. Statistical significance was set at $P < 0.05$.

The ages of patients ranged from 11 - 82 years (mean: 48 years). The majority of patients (46.8%) were above 50 years of age while males were slightly more affected than females. The majority of lesions occurred in the mandible (73.4%) with the posterior regions being the most commonly affected site (51.6%). Most lesions appeared as unilocular (95.3%), well-defined (93.7%) and had smooth corticated margins (85.9%). Edentulous patients were significantly more affected by residual cysts compared to dentate patients in this sample ($P < 0.01$).

Most lesions were surgically managed by enucleation (95.3%) with only one recurrence (recurrence rate: 1.6%). Marsupialization was used to manage one extensive cyst without any recurrence. Two patients (3.1%) developed primary intraosseous odontogenic carcinoma and were managed by resection and reconstruction along with adjuvant radiotherapy. Complications included a pathological fracture of the mandible post enucleation.

In conclusion, residual cysts presented with clinical and radiological features that could mimic more sinister lesions in the jaws. They should be highly suspected when elderly, edentulous patients present with cystic lesions in the jaws. All lesions should undergo biopsy and histopathological examination along with radiological correlation to arrive at an accurate diagnosis and exclude rare malignant transformation into primary intraosseous odontogenic carcinoma. Surgical management of these cysts should be

based on their presenting radiological and histopathological features. Enucleation has been shown to be highly predictable in the management of these lesions with a very low rate of recurrence.

March 2021



DECLARATION

I declare that *Residual cysts of the jaws: a retrospective study of characteristic features and management at the University of the Western Cape Oral Health Centre* 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.

Fadi Titinchi

March 2021

Signed:



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DEDICATION

This work is dedicated to my beloved parents for all their love and support throughout the years.



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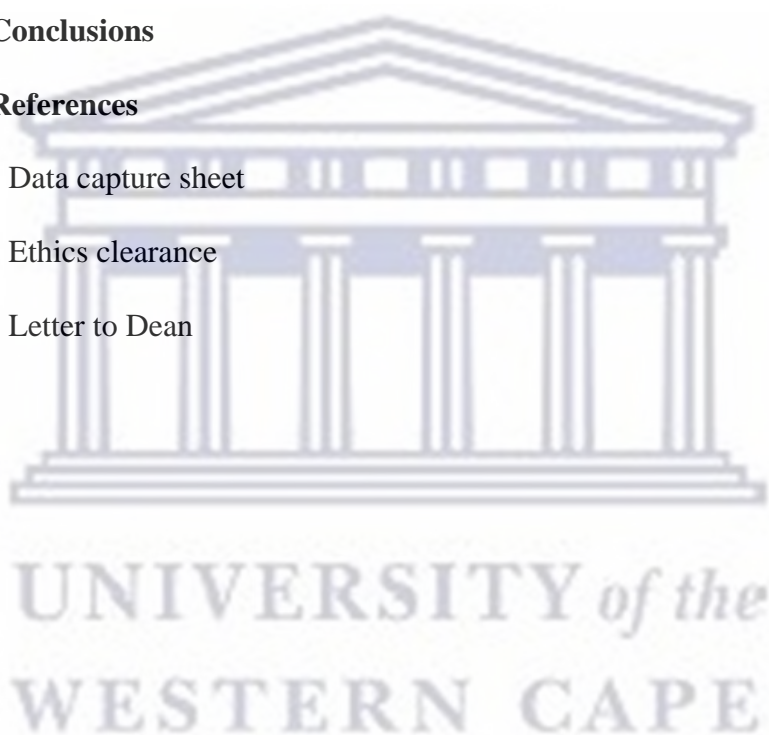


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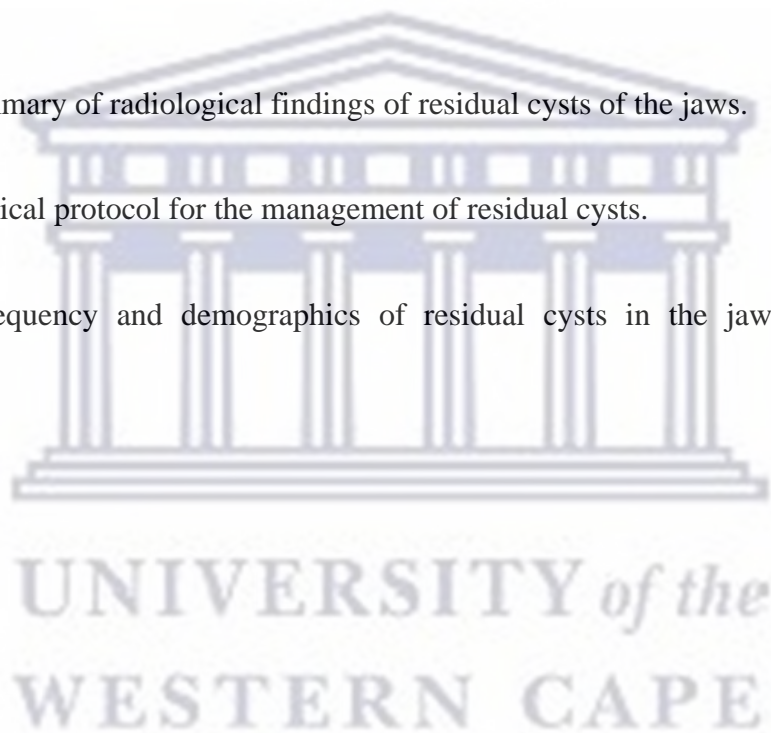
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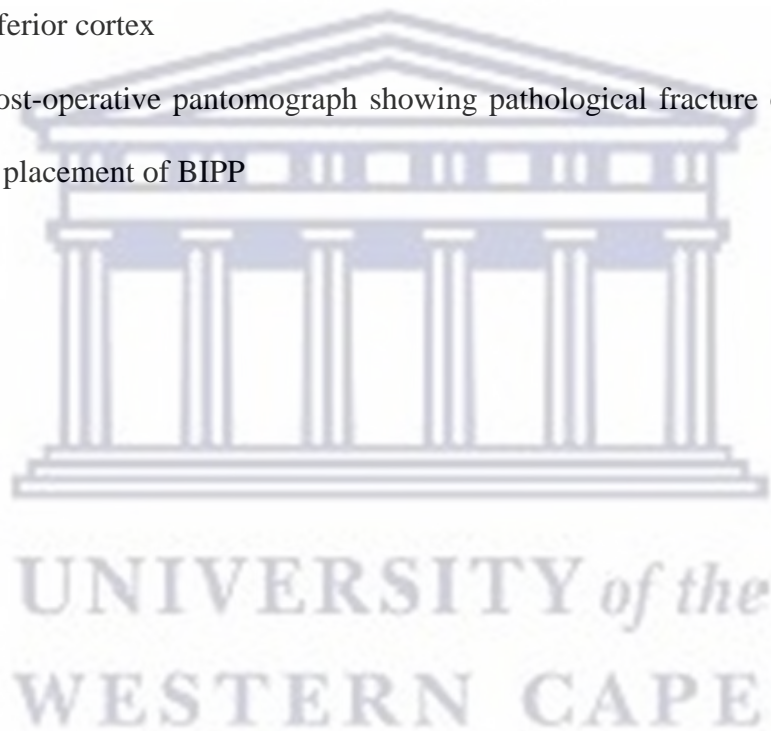
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GLOSSARY

The following terms below will be clarified for the purpose of this study:

- **BIPP:** bismuth iodine paraffin paste.
- **Cortication:** is the presence of radio-opaque rim around the borders of the lesion. It occurs as a response of the body to the tumour by formation of new bone at the periphery of the lesion resulting in radio-opaque margin.
- **Curettage:** is a surgical technique in which a curette is used to remove abnormal tissue by scooping or scraping.
- **CBCT:** cone beam computed tomography.
- **CT:** computed tomography.
- **Decompression:** is a surgical method whereby the lesion cavity is opened into the oral cavity to reduce intra-cystic pressure.
- **Enucleation:** is a surgical procedure in which the entire lesion is removed without the need for any dissection.
- **Expansion:** is the ability of the lesion to expand and grow in size within the bone.
- **Infiltration:** is the ability of a lesion to invade and spread into adjacent tissues.
- **Loculation:** is the appearance of a lesion on a radiograph in which it may consist of multiple compartments (multilocular) or a single compartment (unilocular) within the bone.
- **Margin of the lesion:** defines the border or interface between the lesion and the normal surrounding bone/tissue.

- **Marsupialization:** is a surgical technique whereby the lining of the lesion is sutured to the oral mucosa to create an opening into the lesion.
- **Resection:** is a surgical method whereby the diseased part is removed completely or partially.
- **Well-defined lesion:** is a lesion with a zone of transition of less than 1 mm from the diseased bone to the normal surrounding bone.
- **WHO:** world health organisation.



Chapter 1

INTRODUCTION

The jaws are the most frequently affected bones in the human body by cysts. This is due to the numerous epithelial rests that form a close relationship with developing dentition (Fomete *et al.*, 2016). The clinico-pathological and radiographic presentation of these cysts in the jaws may resemble each other and hence in-depth knowledge of their presenting features and behaviour is vital for their diagnosis and management (Fomete *et al.*, 2016).

Cysts of the jaws are classified according to their origin into odontogenic and non-odontogenic cysts by the World Health Organization (WHO) as shown in Table 1 (Wright and Vered, 2017). Cysts of odontogenic origin are the most prevalent type and develop from the epithelium of developing dentition. The epithelium of these cysts may arise from the dental organ, rests of Malassez, reduced dental epithelium and fragments of dental lamina. These epithelial remnants may lead to the development of a residual cyst following the removal of the involved offending dentition (Tsvetanov, 2016).

Table 1: Classification of odontogenic cysts according to 2017 WHO classification (Wright and Vered, 2017).

Inflammatory odontogenic cysts

- Radicular cyst (including Residual cyst)
- Inflammatory collateral cysts (including paradental cyst and buccal bifurcation cyst)

Developmental odontogenic cysts

- Dentigerous cyst
- Odontogenic keratocyst
- Orthokeratinised odontogenic cyst
- Gingival cyst
- Calcifying odontogenic cyst
- Glandular odontogenic cyst
- Lateral periodontal and botryoid odontogenic cyst



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Chapter 2

LITERATURE REVIEW

A cyst is a cavity lined by an epithelium-lined pouch and contains fluid, semisolid material or gas. It starts by proliferation of epithelial cells which later undergo degeneration and liquefaction, causing the development of a cyst (Adappa, 2014). The liquefied material within the cyst applies the same amount of pressure on the walls of the cavity leading to the cyst growing in a spherical shape. However, in a number of lesions the outline varies due to uneven resistance created by adjacent anatomical structures including the dentition, maxillary antrum, nasal cavity, etc. This may cause displacement of these structures and on some occasions cortical bone expansion (Adappa, 2014).

2.1. Pathogenesis

Residual cyst is usually preceded by a radicular cyst in the jaws which has formed apical or adjacent to an extracted tooth (Shear and Speight, 2007). Residual cysts usually present with the same features as those of conventional radicular cysts; however, due to removal of the cause (i.e. carious tooth/roots), the inflammatory infiltrate in these remnant cysts decreases and non-inflammatory fibrous collagen tissue is present in their walls (Shear and Speight, 2007). These cysts also have a thin epithelial lining which makes their identification by histopathological methods more challenging to differentiate from developmental cysts (Pechalova *et al.*, 2011).

The pathogenesis of radicular and residual cysts is similar (Shear and Speight, 2007). The process is initiated by the spread of bacteria from a non-vital tooth in the periapical region of the jaw. If untreated, this infection leads to the formation of a periapical granuloma which contains activated T cells that produce cytokines. These cytokines act on the epithelial remnants leading to proliferation of these remnants and differentiation into cyst formation (Shear and Speight, 2007).

The proliferating epithelium becomes oedematous through accumulation of fluid and coalesces to form microcysts lined by epithelial cells with inflammatory infiltrate. The cyst wall has a semi-permeable membrane and hence through osmosis, the cyst increases in size. Furthermore, lytic by-products from epithelial and inflammatory cells raise the osmotic pressure within the cyst leading to further expansion and formation of large intra-osseous cystic lesion within the jaws (Shear and Speight, 2007).

Some authors in the literature cast doubt about the development of these cysts as they cite the spontaneous resolution of most radicular cysts following extraction of the causative dentition (Walton, 1996). Other authors report that some residual cysts show active growth patterns in areas which have been edentulous for several years (Schaffer, 1997; Shear and Speight, 2007). The variable behaviour of these cysts highlights the importance of further detailed studies on these cysts.

2.2. Incidence

Inflammatory cysts are the most frequent type of cysts in the jaws. This group of cysts comprise of radicular and residual cysts of which residual cysts are the second most common. The incidence of residual cysts have been reported to comprise around 10% of

odontogenic cysts in the jaws and ranging from 2.2% - 13.7% depending on the region (Tsvetanov, 2016).

2.3. Demographics

Residual cysts primarily affect middle-aged patients in the third decade of life but can manifest from as early as second to the eighth decades of life (Adappa, 2014). Most authors report that the lesion has slightly higher predilection for males than females (Khosravi *et al.* 2013; Shear and Speight, 2007).

2.4. Clinical features

The majority of residual cysts in the jaws are asymptomatic and are detected incidentally on routine radiography (Shear and Speight, 2007). However, if the cyst becomes secondarily infected, patients may report pain and swelling and become aware of the lesion. As the cyst gradually increases in size, they can cause tooth displacement and mobility (Tsvetanov, 2016).

Residual cysts in edentulous areas can cause expansion of the alveolar ridge preventing prosthetic restoration of the affect area caused by the resultant bony expansion. These cysts can also cause significant bone resorption and displacement of vital areas such as inferior alveolar canal, maxillary antrum and nasal cavity. Mandibular pathological fractures may rarely occur (Tsvetanov, 2016).

2.5. Radiological features

Residual cysts commonly present as unilocular, well-defined, radiolucent lesions on radiographs. They are usually round or oval in shape with a thin sclerotic border

(Nortje, 2020; Shear and Speight, 2007). These cysts can lead to displacement or resorption of adjacent anatomical structures and dentition. Tooth bearing areas of the jaws are most frequently involved with the maxilla (60%) more commonly affected than the mandible (40%). The maxillary anterior region is reported to be the most frequent site of the cyst in the jaws (Shear and Speight, 2007; Tsvetanov, 2016).

Residual cysts can lead to expansion of cortical plates and invade the maxillary sinus or displace the inferior alveolar canal. However, an infection within the cyst can make the cortex less apparent on radiographs. The inner part of the lesion is usually radiolucent however dystrophic calcifications can be seen in well-established cysts (Jamdade *et al.*, 2012).

Symptomatic cysts are reported to be slightly larger in size than asymptomatic lesions, but the latter also demonstrated a reduction in size with increasing age of the cyst. This is explained by the fact that these cysts are usually asymptomatic and will progressively reduce in size with epithelial atrophy (High and Hirschmann, 1986).

2.6. Histopathological features

Histological examination demonstrates that the cyst is usually lined by squamous epithelium which can range from 1 to 50 cell layers in thickness. In majority of lesions, the epithelial lining is about 20 cell layers in thickness (Jamedade *et al.*, 2012). The epithelium arises from the cell remnants of Malassez, however in certain lesions it can originate from the respiratory epithelium of the antrum usually as a result of the lesion communicating with the maxillary antral lining (Jamedade *et al.*, 2012).

Epithelial lining of residual cysts can show linear or hair-pin shaped calcifications called rushton bodies. These calcifications are rarely seen. The origin of these hyaline bodies was previously unclear but it is currently understood that they signify a secretory by-product of odontogenic epithelium. The cyst wall usually displays cholesterol clefts along with multi-nucleated giant cells, erythrocytes, and foci of hemosiderin pigmentation (Tsvetanov, 2016).

A chronic inflammatory infiltrate is usually noted within the cyst when the causative tooth is extracted and can continue due to lysosome or other chemical deposits from necrotic or dying cells and may sometimes initiate an acute inflammatory response. If the radicular cyst is accidentally left undetected following tooth removal, certain level of inflammation may continue and thus residual cyst, even though to a lesser extent than radicular cysts, have the ability to continue expanding (Muglali *et al.*, 2008).

High and Hirschmann (1986) reported on the histological and radiographic changes of residual cysts as they aged. They were able to demonstrate a slow rise in the mineralized material within the cyst lumen as the cyst aged. These mineralized deposits become histochemically prominent in residual cysts that are present for more than eight years and radiographically six years later. Cholesterol crystals were also noted in the residual cystic contents.

2.7. Differential Diagnosis

Residual cysts can mimic a number of benign cysts and tumours in the jaws. The list of differential diagnosis should include unicystic ameloblastoma, odontogenic keratocyst (OKC), traumatic bone cyst, glandular odontogenic cyst (GOC) and lateral periodontal cyst (Tsvetanov, 2016). When residual cyst exhibits calcifications, one should

differentiate it from other mixed lesions on radiographs such as odontoma, root rests, mixed stage of cemento-osseous dysplasia, adenomatoid odontogenic tumour (AOT) and calcifying epithelial odontogenic tumour (Pindborg tumour) (Sridevi *et al.*, 2014). Accurate diagnosis can be achieved by correlating the clinical, radiographic and histological findings (Boffano and Gallesio, 2010).

2.8. Management

Since residual cysts present with similar clinical and radiographic features as more aggressive cysts and tumours, incisional biopsy or enucleation and/or curettage should be performed on all lesions to confirm the diagnosis by histopathological examination (Tsvetanov, 2016).

Residual cysts can be managed by various surgical methods to reduce intra-lesional pressure within the cyst (Allon *et al.*, 2015). Enucleation is the most common method whereby the entire cyst lining is removed usually at the time of biopsy. Larger cysts and those in unfavourable anatomical locations are managed by marsupialization. This method involves suturing the cyst lining to the oral mucosa over a period of time to minimize intraluminal pressure leading to reduction in cyst size. This is followed by enucleation of the residual cyst lining (Tsvetanov, 2016).

Decompression is another treatment option that lowers the pressure within the cystic cavity. This technique involves making a smaller window in the cyst wall, which is kept patent by suturing a device to it. Although both marsupialization and decompression are based on the principle of releasing intramural pressure to shrink the cyst size; at the same time they allow gradual bone deposition around the lesion (Enislidis *et al.*, 2004).

2.9. Prognosis and recurrence

Residual cysts have low recurrence rate following enucleation and hence the prognosis following surgical management is good. If during surgical removal the cystic lining and wall is severely fragmented, leaving epithelial rests in the cavity, or if a periapical granuloma is only partially enucleated, a residual cyst can develop in the area after a long period of time (Jamdade *et al.*, 2012). Therefore, periodic follow-up is essential to detect any recurrences and malignant transformation (Sridevi *et al.*, 2014). No studies have reported on the recurrence rate of residual cysts in the literature.

Residual cysts tend to resolve slowly over-time as the squamous epithelium lining of the cyst is thin and act as a semi-permeable membrane and continues to permeate cellular material into the cyst's lumen. Both these methods could cause a delay in resolving of the cyst (Sridevi *et al.*, 2014).

2.10. Development of odontogenic tumours from residual cyst

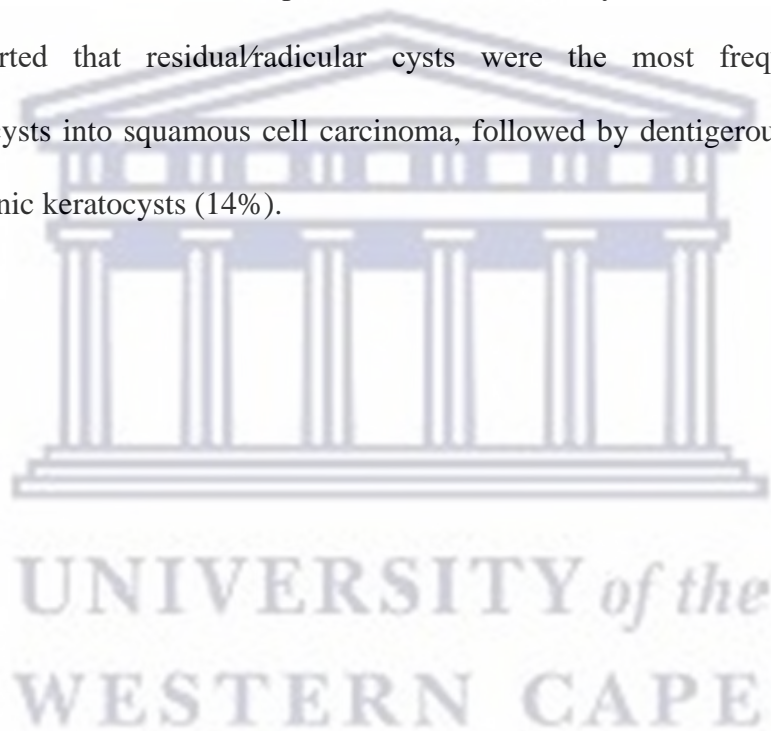
Some authors have reported that ameloblastoma and adenomatoid odontogenic tumour can originate from the cystic lining of residual cysts (Mahajan *et al.*, 2014; Lee, 1970; Giansanti and Somerson, 1970). Unicystic ameloblastoma has been reported to emerge from the lining of residual cyst, radicular cyst and dentigerous cyst. Lucas (1954) was the first to report that neoplastic change can arise in the epithelial lining of certain odontogenic cysts as ameloblastoma and residual cysts are of similar embryonic origin.

2.11. Potential malignant transformation

Primary intraosseous odontogenic carcinoma is described as a squamous cell carcinoma originating within the jaws without primary communication with the oral cavity and

arises primarily from rests of odontogenic epithelium (Bodner *et al.*, 2011). It is a sporadic malignant neoplasm and presents almost entirely in the jaws. The estimated incidence of primary intraosseous odontogenic carcinoma has been reported to be 1 - 2% of all oral cancer. It has predilection for older males and involves mostly the mandible (Bodner *et al.*, 2011).

Of all odontogenic cysts, radicular/residual cysts have the highest potential for malignant transformation of their epithelium. In a review by Bodner *et al.* (2011), the authors reported that residual/radicular cysts were the most frequently (60%) transformed cysts into squamous cell carcinoma, followed by dentigerous cysts (16%) and odontogenic keratocysts (14%).



Chapter 3

AIM AND OBJECTIVES

3.1. Aim

The aim of this study was to describe the clinical, radiological and histopathological features of residual cysts of the jaws presenting at the University of the Western Cape Oral Health centre as well as to analyse their surgical management and recurrence patterns.

3.2. Objectives

- 1) To describe the clinical and demographic features of residual cysts of the jaws.
- 2) To describe the radiographic and histopathological features of residual cysts of the jaws.
- 3) To determine if any of residual cysts underwent malignant transformation.
- 4) Analyse treatment methods and recurrence rates.
- 5) To determine the potential of residual cysts to transform into primary intraosseous odontogenic carcinoma.

3.3. Research questions

- 1) What are the clinical, radiographic and histopathological features of residual cysts of the jaws? Are there any distinct radiographic signs that differentiate them from other odontogenic cysts and tumours?
- 2) What is the potential of these cysts to develop primary intraosseous odontogenic carcinoma?
- 3) What is the most appropriate surgical method to manage these cysts?
- 4) What is the recurrence rate of these cysts following surgical management?

3.4. Rationale for the study

There is paucity in the literature regarding the clinical and radiographic presentation of residual cysts of the jaws (Nortje, 2020; Shear and Speight, 2007). This is especially important as they may mimic more aggressive cysts and tumours of the jaws. They have also been reported to rarely transform into primary intraosseous odontogenic carcinoma. Even less is known regarding their surgical management when they become extensive in size (Shear and Speight 2007). Hence the rationale for this study was to describe the presenting features of a large case-series of these cysts as well as their management and recurrence patterns.

Chapter 4

MATERIALS AND METHODS

4.1. Study design

This was a retrospective descriptive study of residual cysts of the jaws. It was designed to study the clinical, radiographic and histopathological features of this cystic lesion as well as its surgical management and recurrence patterns during a period of thirty years from 1989 to 2019.

4.2. Study sample

The study sample was selected manually by compiling all clinical and radiographic information from the records of the Departments of Maxillo-Facial and Oral Surgery and Diagnostics & Radiology at the University of the Western Cape Oral Health centre. All diagnoses were confirmed histopathologically prior to the inclusion in this study.

4.3. Selection criteria

Inclusion criteria:

- Histopathologically confirmed diagnosis of residual cyst of the jaws.
- Patient records containing details regarding demographic data and treatment methods.

- Each record included at least one pantomograph.

Exclusion criteria:

- Records with incomplete patient data or insufficient history.
- The histopathological diagnosis was inconclusive.
- The diagnostic quality of the radiographs was poor or inadequate.

4.4. Data collection

A standardized data collection sheet were used to collect information from each file (Appendix 1). All data collected was entered onto a 2010 Microsoft Excel spreadsheet. The data recorded included the patient's age (in years), gender and ethnic background. The presenting signs and symptoms of the patient as well as the history of the lesion were collected. The detailed location of the lesion within the jaws was noted. Histopathological features were recorded including any cases that developed into primary intraosseous odontogenic carcinoma. Surgical management and follow-up was also noted.

4.5. Radiographic examination

The radiographs included in this study were scrutinised by two pre-calibrated examiners. Each image included in the study was examined separately by each examiner followed by correlation of the findings. If there was a disagreement between the two examiners, then an independent examiner (Maxillofacial Radiologist) scrutinised the case and a decision was reached by unanimous agreement.

All radiographs included in this study were examined on a standard viewing box in a dark room. All digital radiographs were examined on a standard 19 inch computer screen. No magnification was used to examine radiographs in this study.

The location of the lesion was classified into varying anatomical regions in the jaws. The anterior region of the mandible extended from the left canine (33) to right canine (43) and in edentulous patients from the left mental foramen to right mental foramen. The posterior region of the mandible extended from canine (33/43) to the angle of the mandible, for both sides. The anterior region of the maxilla extended from the left canine (23) to right canine (13) while the posterior region extended from canine (13/23) to the maxillary tuberosity on both sides.

The size of lesions on radiographs was measured in millimetres using the widest diameter from one end of the lesion to the other. The overall shape of the lesion was noted along with the radio-density and locularity. Radio-density was classified as either radio-lucent, radio-opaque or mixed (radio-lucency with radio-opacity). Lesions were categorised as either unilocular (only one compartment) or multilocular (multiple adjacent compartments within the cavity). The margins were classified as either well-defined or ill-defined. The effect of the lesion on adjacent structures (adjacent dentition, mandibular canal and/or maxillary antrum) was recorded.

The dentition affected by the lesion was recorded to demonstrate the extent of the lesion and whether it crossed 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 was analysed using t-test and chi-square test to draw correlations between various parameters such as lesion size, gender, locularity, radio-density etc. Microsoft Excel spreadsheet version 2010 was used to process the data digitally and compute means, percentages, etc. Epi Info V7 software was used to perform statistical analysis. Statistical significance was set at $P < 0.05$.

4.7. Ethical considerations

This study was a retrospective record-based analysis of patients presenting with residual cysts of the jaws at the University of the Western Cape Oral Health centre. The research protocol for this study was presented to the Faculty of Dentistry of the University of the Western Cape research committee and ethics clearance was obtained from Biomedical Research Ethics Committee (BMREC) of the University of the Western Cape (approval number: BM20/5/13) prior to commencement of the study (Appendix 2). Permission was also obtained from the Dean of the Faculty of Dentistry, University of the Western Cape prior to accessing patient files (Appendix 3).

Patient details recorded as part of this study did not include any identifiable information such as their name, address or date of birth. The file number was recorded as part of data collection sheet for reference purposes only. All electronic data was saved on a password protected Excel spreadsheet and computer. All physical data collection sheets were kept in a secure office and were destroyed after completion of the study. No patient details were displayed on radiographs included in the study. Clinical photographs included in the study obscured the patients' identity.

Chapter 5

RESULTS

A total of 75 residual cysts were diagnosed over a period of 30 years at the Department of Maxillo-Facial and Oral Surgery, University of the Western Cape Oral Health centre. Only 64 cysts in 62 patients met the inclusion criteria and were included in the study. Overall, the frequency of residual cysts amongst all odontogenic cysts was relatively low (3.2%).

5.1. Demographic data

The ages of patients ranged from as young as 11 years to 82 years (mean: 48 years). The majority of patients (46.8%) were above 50 years of age. Males (54.8%) were slightly more affected than females (45.2%). Table 2 summarises the distribution of patients amongst the different age groups.

Table 2: Distribution of ages and gender of patients diagnosed with residual cysts of the jaws.

Age group	Males	Females	Total	Percentage
0-10	0	0	0	0
10-20	2	2	4	6.25
21-30	2	3	5	7.81
31-40	6	7	13	20.31
41-50	8	5	13	20.31
51-60	9	5	14	21.87
61-70	5	3	8	12.50
71-80	2	0	2	3.12
81-90	0	5	5	7.81
Total	34	30	64	100

5.2. Clinical presentation

Symptoms were present in 57.8% of patients which included swelling (64%) (Figure 1), pain (25%) and pus discharge (6.3%). Only one patient presented with paraesthesia.

Symptoms were present for a mean period of 6.8 months prior to presentation. Most symptomatic lesions occurred in the posterior mandibular regions (77.8%).

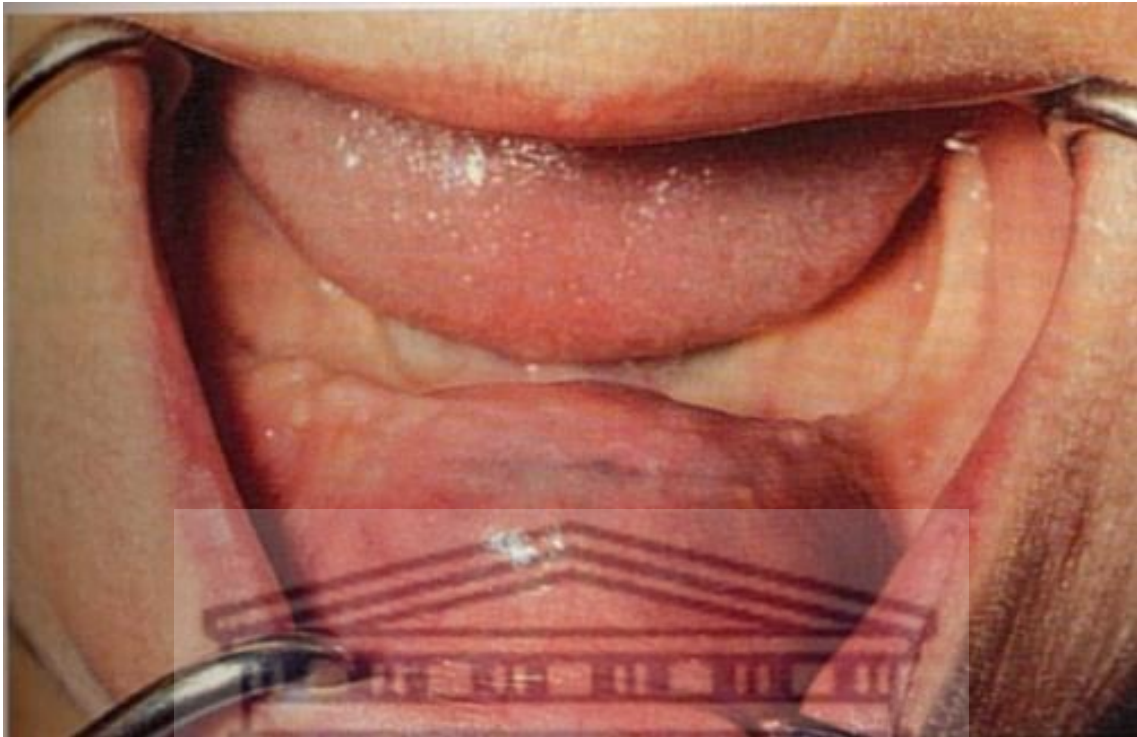


Figure 1: Intraoral clinical photograph showing extensive swelling in the anterior mandible caused by residual cyst.

5.3. Radiographic features

The majority of lesions occurred in the mandible (73.4%) with the posterior regions being the most commonly affected site (51.6%). Most lesions appeared as unilocular (95.3%), well-defined (93.7%) and had smooth corticated margins (85.9%). Bony expansion was not a common feature while only one case caused root resorption.

The size of cysts on pantomographs ranged from 6 mm to 125 mm in greatest diameter (mean size: 39.4 mm). The lesion demonstrated the ability to cause displacement of adjacent anatomical structures including mandibular canal (45.7% of all lesions located in mandibular posterior region) and maxillary antrum (66.6% of all lesions located in

maxillary posterior region). One lesion demonstrated calcification within the cyst (Figure 2). Summary of radiological findings is presented in Table 3.

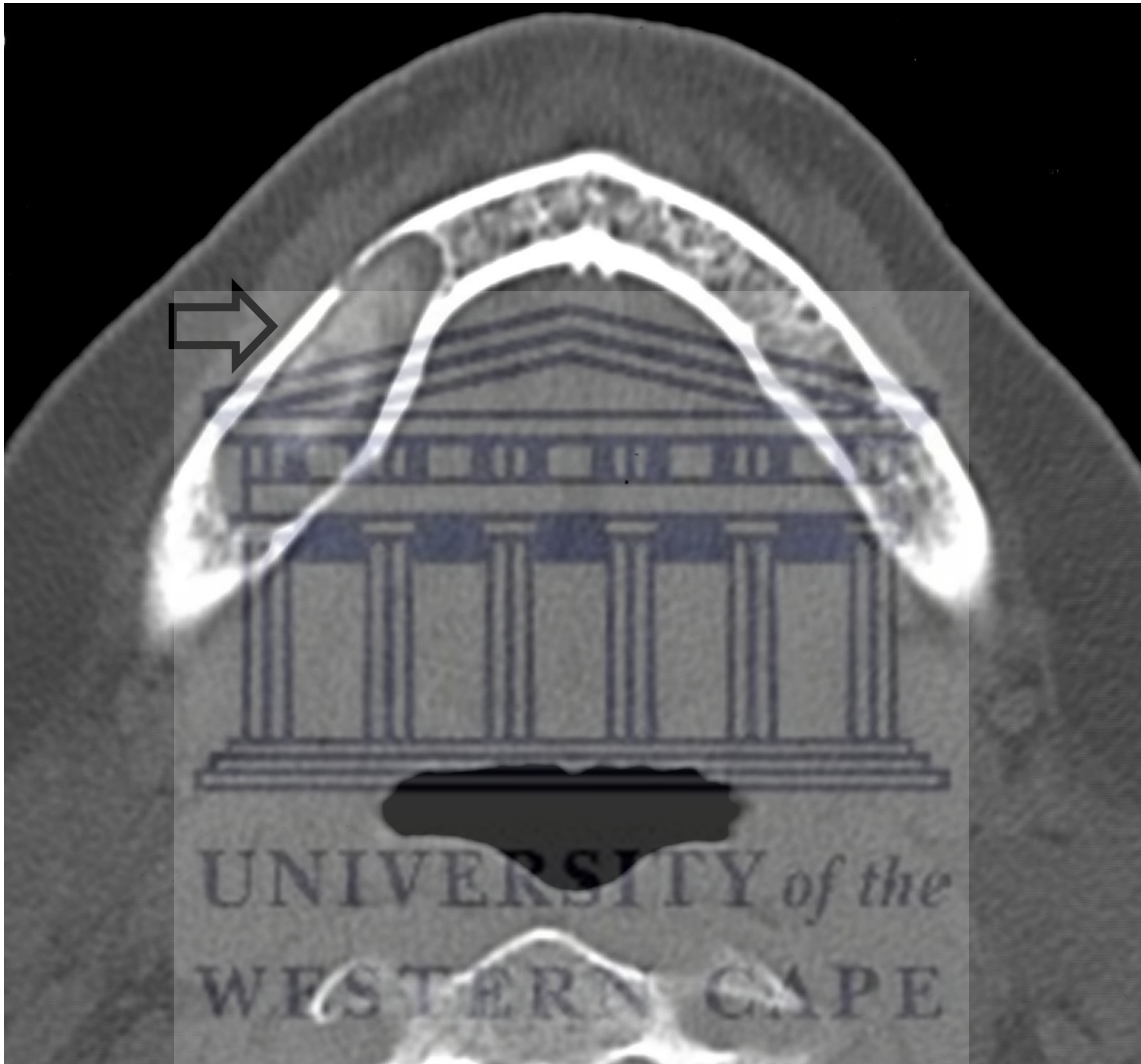


Figure 2: Axial CT showing calcified material within the lumen of a residual cyst in right mandible.

Table 3: Summary of radiological findings of residual cysts of the jaws.

	Anterior Mandible	Posterior Mandible	Anterior Maxilla	Posterior Maxilla	Total
Locularity					
Unilocular	13 (20.3%)	32 (50%)	10 (15.6%)	6 (9.3%)	61 (95.3%)
Multilocular	1 (1.6%)	1 (1.6%)	0	1 (1.6%)	3 (4.7%)
Demarcation					
Well-defined	14 (21.8%)	31 (48.4%)	9 (14.1%)	6 (9.3%)	60 (93.7%)
Ill-defined	0	2 (3.2%)	1 (1.6%)	1 (1.6%)	4 (6.3%)
Shape					
Irregular	2 (3.2%)	15 (23.4%)	2 (3.2%)	4 (6.3%)	23 (35.9%)
Round	3 (4.7%)	9 (14.1%)	5 (7.8%)	1 (1.6%)	18 (28.1%)
Oval	9 (14.1%)	9 (14.1%)	3 (4.7%)	2 (3.2%)	23 (35.9%)
Margins					
Smooth	12 (18.7%)	29 (45.3%)	9 (14.1%)	5 (7.8%)	55 (85.9%)
irregular	2 (3.2%)	4 (6.3%)	1 (1.6%)	2 (3.2%)	9 (14.1%)
Bony expansion					
	5 (7.8%)	12 (18.7%)	2 (3.2%)	2 (3.2%)	21 (32.8%)
Sclerotic margin					
	13 (20.3%)	24 (37.5%)	6 (9.3%)	4 (6.3%)	47 (73.4%)
Root resorption					
	0	1 (1.6%)	0	0	1 (1.6%)

Interestingly, most patients in this sample were edentulous (60.9%). Seventeen lesions (26.5%) were incidentally found on routine panoramic radiographs (Figure 3). One-third of incidentally found lesions occurred in edentulous patients. These incidental lesions were significantly smaller in size than symptomatic lesions ($P < 0.05$).

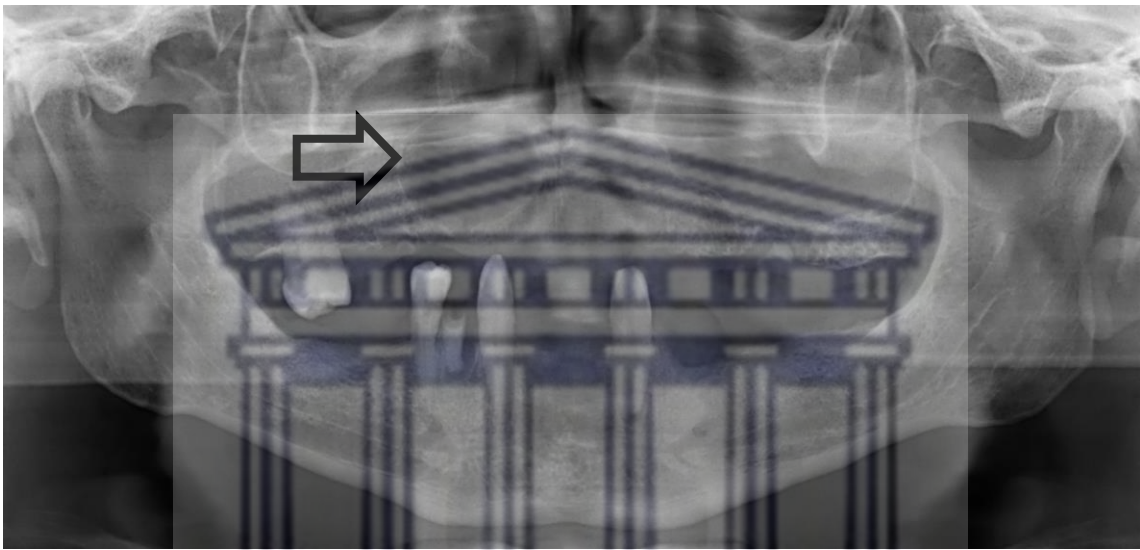


Figure 3: Pantomograph showing incidentally found residual cyst in the right maxilla.

5.4. Initial Diagnosis

Due to the high frequency of these cysts in elderly patients with radiological features that resemble other cystic lesions of the jaws, odontogenic keratocyst and ameloblastoma were high on the list of differential diagnosis in this sample (Figures 4 and 5). Only thirty (46.8%) of lesions were accurately diagnosed by the consulting clinician based on clinical and radiological features alone. This should raise further suspicion when such lesions are encountered in elderly, edentulous patients.

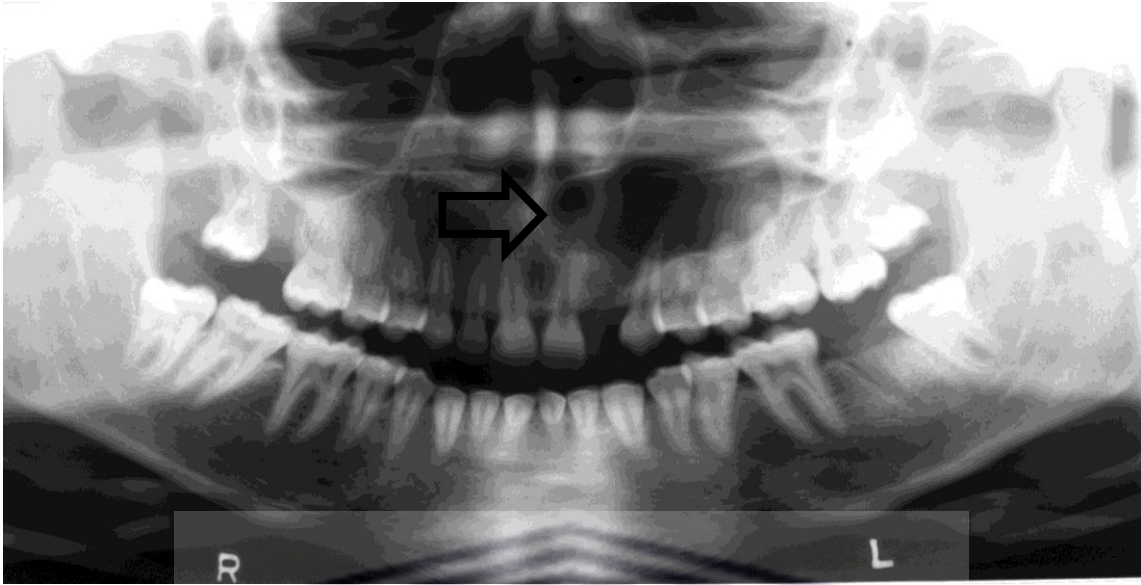


Figure 4: Pantomograph showing radiolucent lesion in left maxilla initially thought to be an OKC.

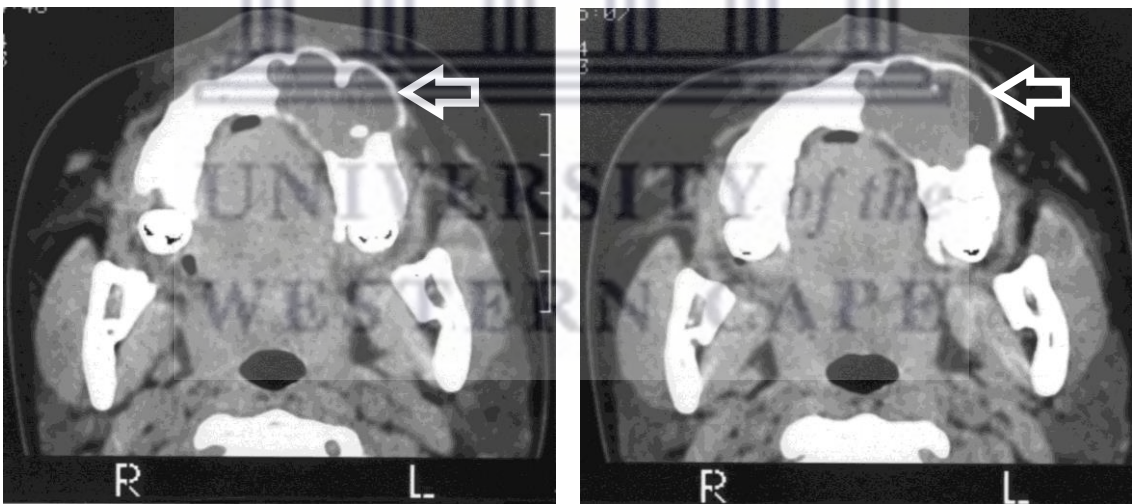


Figure 5: Axial CT images of same patient in Figure 4 showing scalloping of margins of the lesion mimicking an OKC.

5.5. Histopathological features

All lesions showed a cyst lined by proliferative stratified squamous epithelium with some lesions showing foci of mucous metaplasia. Dense chronic inflammation was noted within the fibrous wall of the cyst (Figure 6). Cholesterol cleft was not a prominent feature (39%) and only five lesions showed calcifications (7.8%).

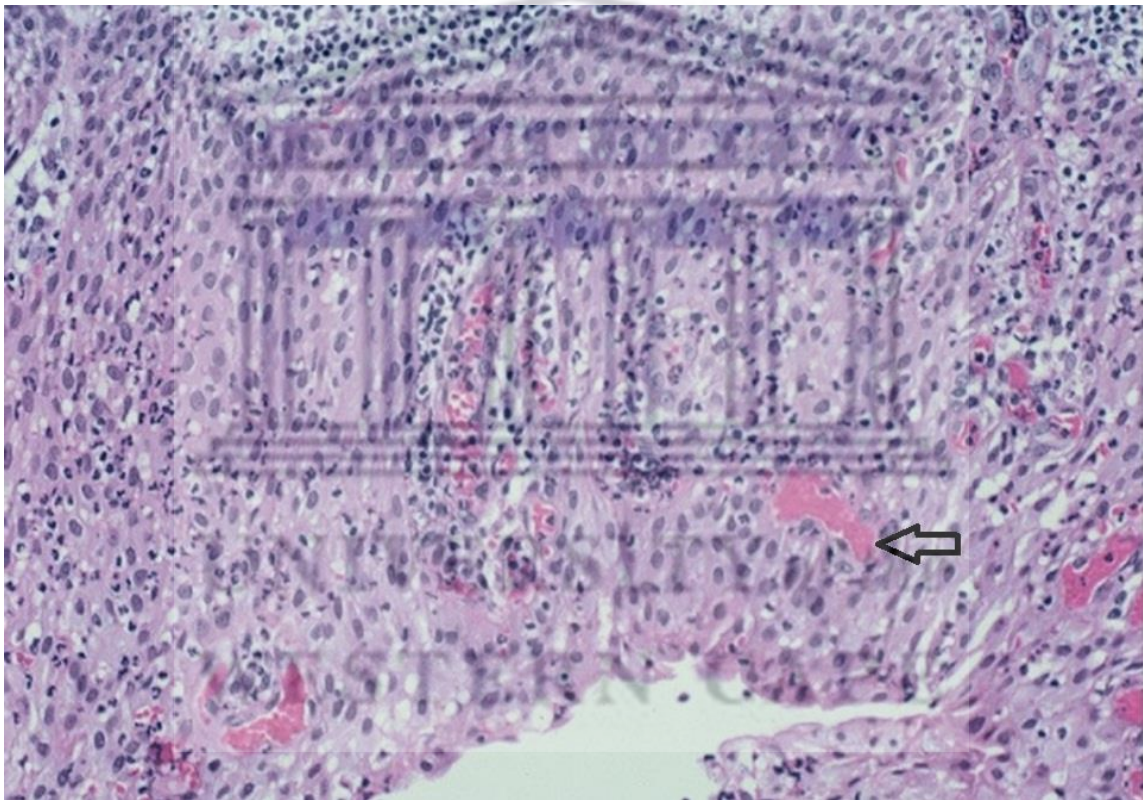


Figure 6: H & E stained histological section showing hyaline bodies (arrow) in the epithelial lining with dense inflammatory infiltrate.

Two residual cysts showed signs of primary intraosseous odontogenic carcinoma within their lining. Both lesions occurred in the mandible and caused extensive destruction with cervical lymph node involvement at the time of presentation (Figure 7). No residual cyst in this sample gave rise to benign odontogenic tumours such as ameloblastoma or adenomatoid odontogenic tumour.

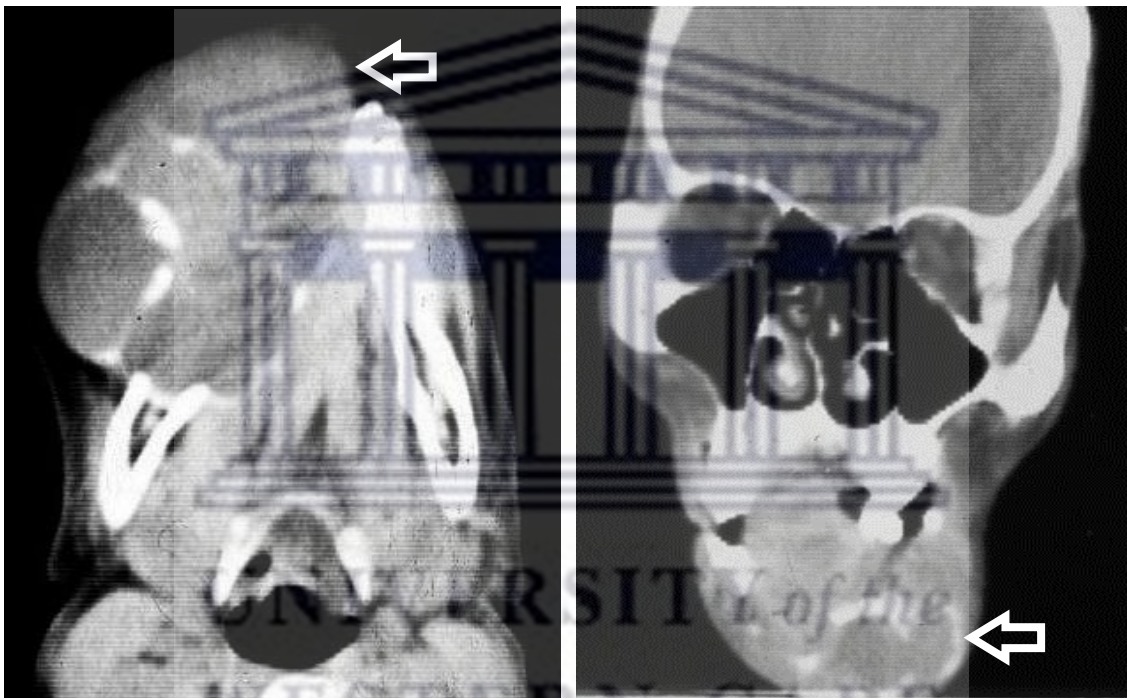


Figure 7: Axial and coronal CT showing destructive, ill-defined primary intraosseous odontogenic carcinoma arising from residual cyst.

5.6. Management

Most lesions were surgically managed with enucleation of the cystic lining (Figures 8 and 9) while only one case initially underwent marsupialization due to its extensive size followed by enucleation. Large lesions that underwent enucleation were packed with

bismuth iodine paraffin paste (BIPP) impregnated gauze for an initial period of two weeks followed by incremental removal of BIPP weekly over two to three visits depending on the size of the defect (Figures 10 and 11). This aided in eliminating the dead space and avoided the need for bone grafting. Both patients with malignancy in the cyst lining underwent resection with one patient demising eight months later.

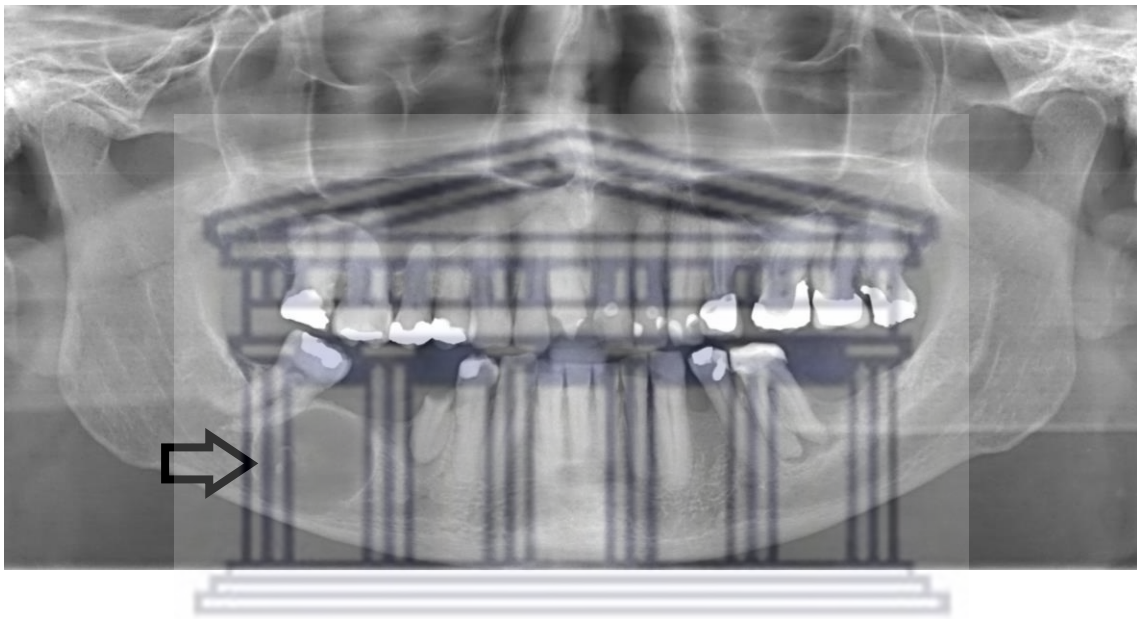


Figure 8: Pantomograph showing residual cyst in right posterior mandible that underwent enucleation.

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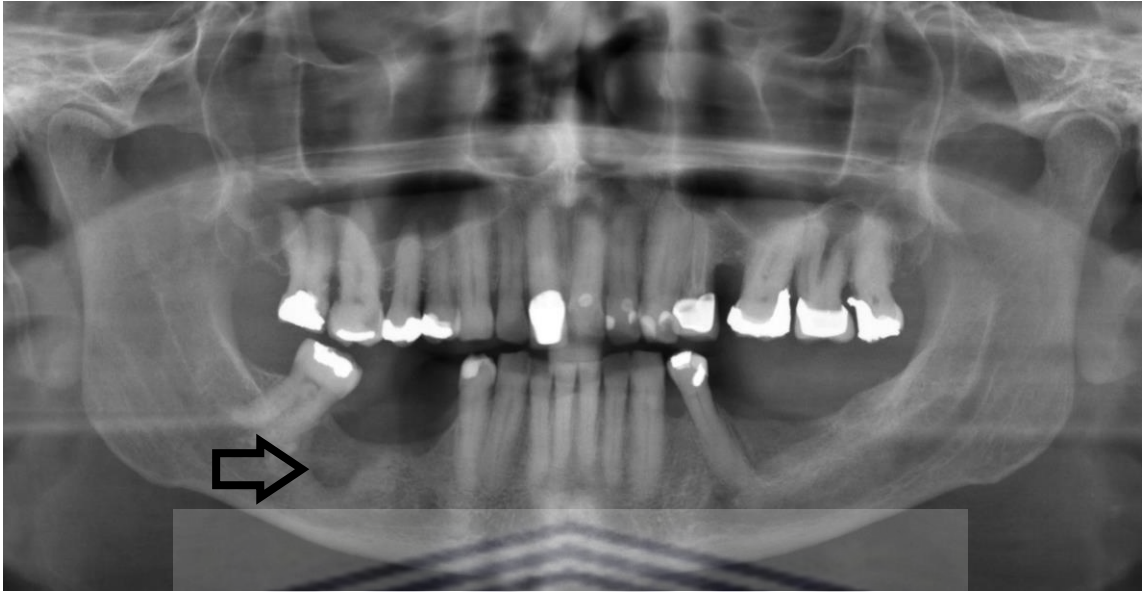


Figure 9: Pantomograph of same patient in Figure 8 showing bone regeneration 14 months post enucleation.

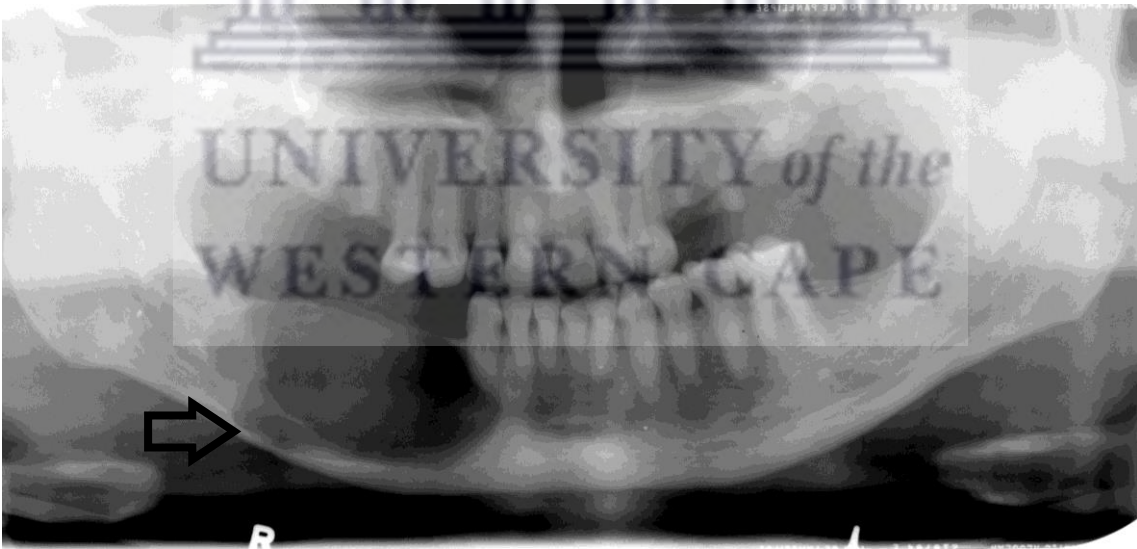


Figure 10: Pantomograph showing a large residual cyst in the right mandible.

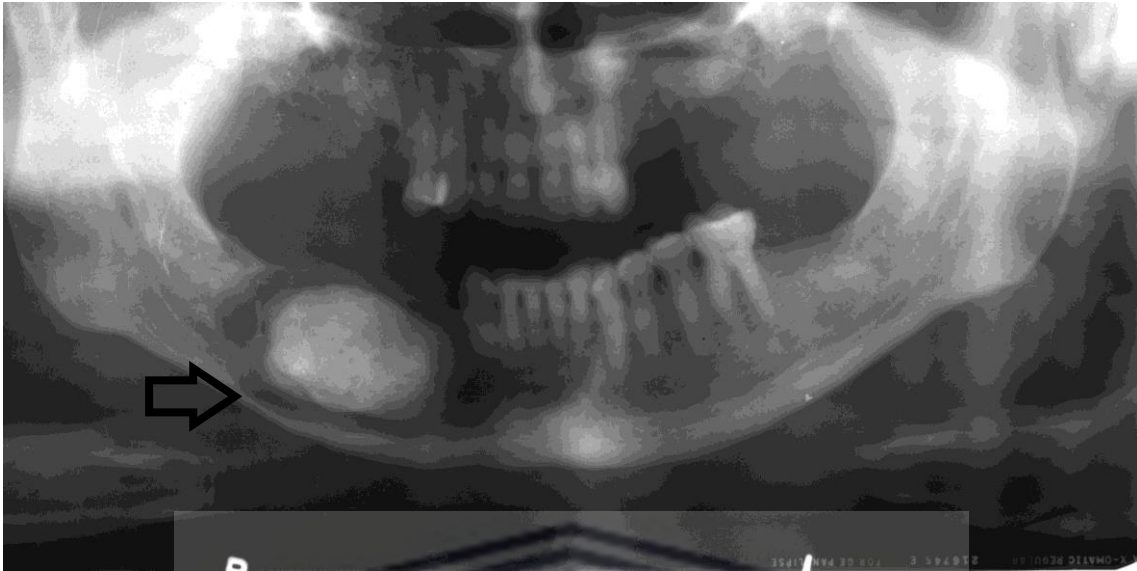


Figure 11: Pantomograph of same patient following enucleation and placement of BIPP to eliminate the dead space.

A surgical management protocol is formulated based on the findings of this study (Table 4). Enucleation of the entire cyst lining was found to be appropriate for most residual cysts as the majority of lesions were less than 40 mm in diameter. One extensive lesion measuring 125 mm in diameter underwent marsupialization to reduce its size initially. Resection with reconstruction was utilized for residual cysts that developed primary intraosseous odontogenic carcinoma.

Table 4: Surgical protocol for the management of residual cysts.

Surgical method	Clinico-pathological features
	- Size of lesion less than 4 cm on pantomograph
Enucleation/curettage	- Easily accessible anatomical location - Unilocular lesions
Marsupialization/decompression followed by enucleation	- Size of lesion greater than 4 cm on pantomograph - Difficult to access regions (e.g. posterior maxilla) - Unilocular or multilocular lesions - Thinning of mandibular cortex with risk for pathological fracture
Resection and reconstruction	- Presence of malignant transformation (primary intraosseous squamous cell carcinoma) - Extension/invasion of adjacent soft tissues

5.7. Recurrence

Although residual cysts are not known to recur, one patient who underwent enucleation of cystic lesion in maxilla returned with recurrent lesion four months post-operatively (recurrence rate: 1.6%). This was most likely due to inadequate enucleation of the lesion which extended into the maxillary antrum (Figures 12, 13 and 14). Other adverse

complications include one patient who suffered a pathological fracture of the mandible post enucleation of the lesion (Figures 15 and 16).



Figure 12: Pre-operative pantomograph reconstruction of cone beam computed tomography (CBCT) of large residual cyst in the left maxilla extending into maxillary sinus.

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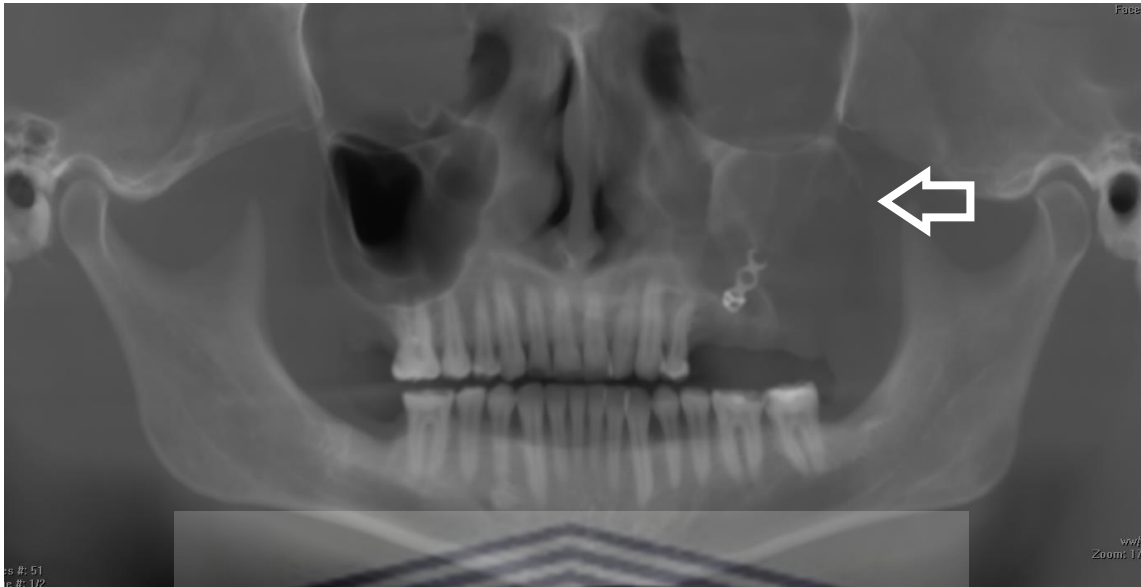


Figure 13: Four month post-operative pantomograph CBCT of same patient in Figure 12 showing recurrence of residual cyst in left maxilla post enucleation.

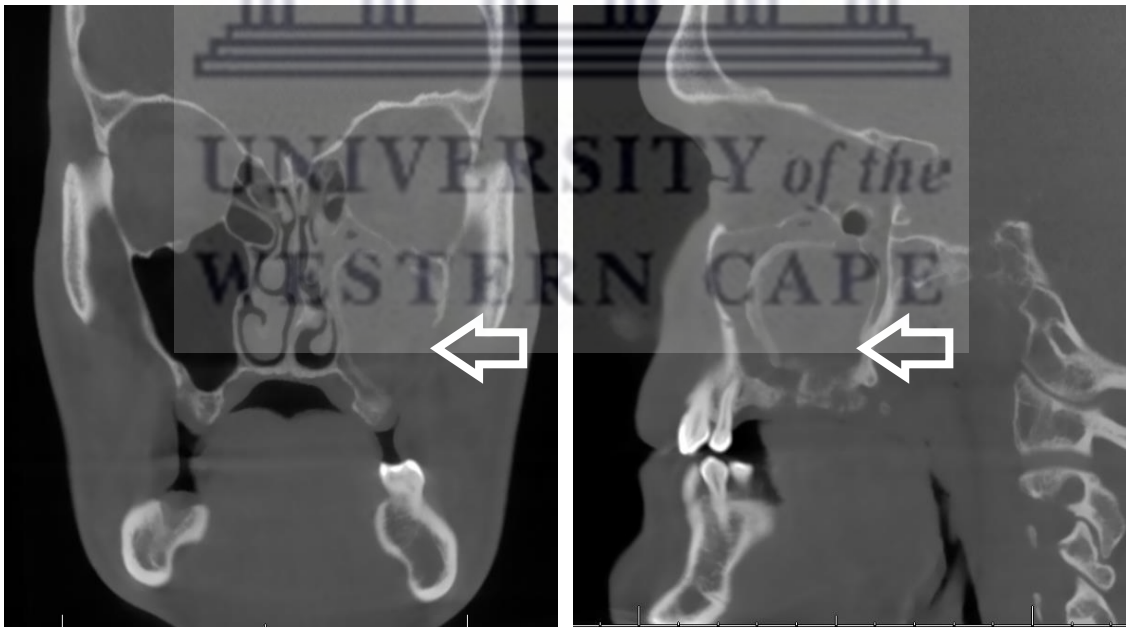


Figure 14: Post-operative coronal and sagittal CBCT showing recurrent residual cyst in left maxilla of the same patient in Figure 13.

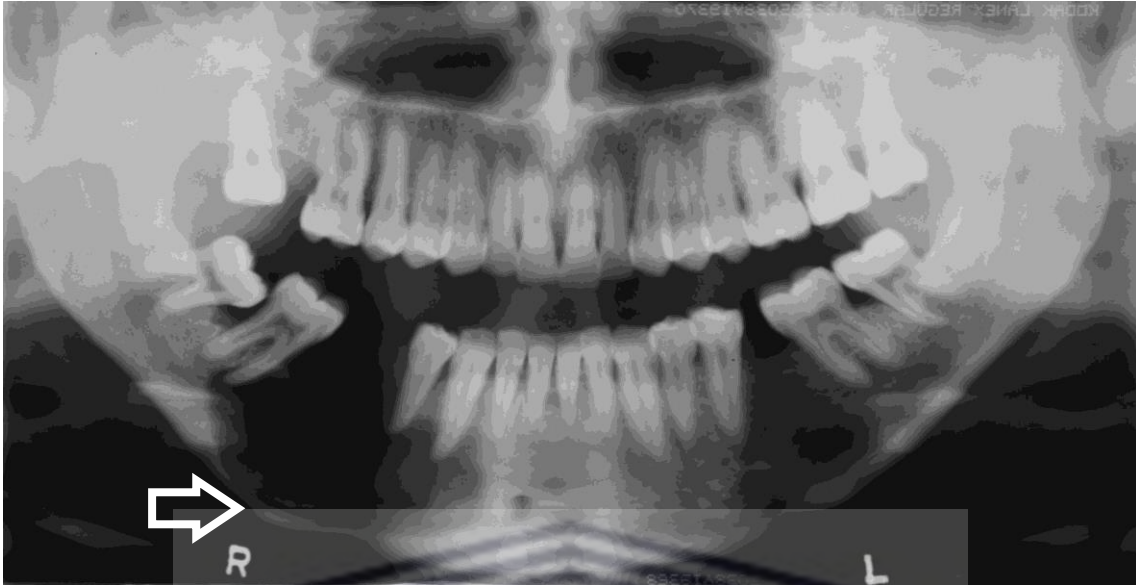


Figure 15: Pantomograph showing residual cyst in the right mandible with thinning of inferior cortex.



Figure 16: Post-operative pantomograph showing pathological fracture of the mandible and placement of BIPP.

Chapter 6

DISCUSSION

Residual cysts are often overlooked and assumed to be unambiguous in its presentation. For this reason, there are only a handful of detailed studies in the literature. This is one of the largest detailed reports in the literature on residual cysts. It highlights its variable clinico-pathological presentation and its ability to continually grow undetected mimicking more aggressive cysts and tumours.

6.1. Demographic data

Inflammatory odontogenic cysts are the most common type of cysts in the jaws. They are comprised of radicular and residual cysts as well as inflammatory collateral cysts (including paradental cyst and buccal bifurcation cyst). The frequency of residual cysts have been reported to range from 2.2% - 13.7% of odontogenic cysts in the jaws as shown in Table 5 (Tsvetanov, 2016; Pechalova *et al.*, 2011). In this sample, the frequency was relatively low due to high rates of early extractions of carious teeth which prevented progress into inflammatory cysts.

Table 5: Frequency and demographics of residual cysts in the jaws in the literature.

Author	Region	Frequency	Ranking amongst odontogenic cyst	Mean age range (decade)	M:F ratio	Maxilla: Mandible ratio
Present study	South Africa	3.2%	4th	4th-5th	1.2:1	1:2.8
Núñez-Urrutia <i>et al.</i> (2010)	Spain	4.3%	4th	3rd-4th	NA*	1:1
Kambalimath <i>et al.</i> (2014)	India	6.0%	5th	6th	3.5:1	NA*
Ledesma-Montes <i>et al.</i> (2000)	Mexico	4.9%	4th	3rd	0.88:1	NA*
Ochsenius <i>et al.</i> (2007)	Chile	11.0%	4th	5th-6th	1.15:1	2:1
Acikgoz <i>et al.</i> (2012)	Turkey	13.7%	3rd	NA*	NA*	1:0.85
Khosravi <i>et al.</i> (2013)	Iran	12.9%	3rd	3rd	1.77:1	1:1.06
Avelar <i>et al.</i> (2009)	Brazil	5.9%	3rd	2nd	NA*	1:1.16
Mosqueda-Taylor <i>et al.</i> (2002)	Mexico	2.2%	4th	NA*	NA*	NA*
Hu and You (1989)	Korea	NA*	NA*	3rd-4th	1.23:1	2.3:1

NA*: not available.

The majority of patients presenting with residual cysts are middle-aged but the lesion can manifest from as early as second to the eighth decades of life (Adappa, 2014). In this study, patients were older than previously reported most likely due to limited access to advanced dental imaging in community clinics which delays the diagnosis of these lesions. Most authors report that the lesion has slightly higher predilection for males as was shown in this sample (Shear and Speight, 2007; Khosravi *et al.*, 2013). Kambalimath *et al.* (2014) reported in their sample that males were 3.5 times more commonly affected than females.

6.2. Clinical features

A large number of residual cysts are symptom-less and found incidentally on routine dental radiography (Shear and Speight, 2007). In lesions that become secondarily infected, some patients may present with pain and inflammation. More than half of patients in this sample were symptomatic with swelling being the most common symptom. These symptoms are non-specific as other odontogenic lesions can have similar presentation but they should direct the clinician to investigate further.

High and Hirschmann (1986) reported in their sample that symptomatic cysts were more commonly found in the anterior maxilla, with a frequency of 37%. This is due to the high prevalence of palatal invaginations and trauma which can lead to pulpal necrosis in the dentition in this region. This study was in disagreement with this finding as most symptomatic lesions occurred in the posterior mandible. This further complicates distinguishing this cyst from other odontogenic cysts and tumours as they also have a high tendency to occur in this region.

The majority of patients in this sample were edentulous, most of them presented with ill-fitting dentures due the bony expansion caused by these cysts. Interestingly, Bodner *et al.* (2011) reported that the majority of cystic lesions in edentulous jaws were in fact residual cysts. Hence, based on the evidence presented in this study and other studies, residual cysts should be high on the list of differential diagnosis of cystic lesions in edentulous jaws.

6.3. Radiological features

Most studies that reported on the radiological features of residual cysts concur that these lesions present as unilocular, well-defined, radiolucencies. They are usually round or oval in shape with a thin sclerotic border (Shear and Speight, 2007). This fairly predictable radiological presentation was also noted in this study. These cysts can also cause expansion of the involved bone and displacement of anatomical structures including mandibular canal, maxillary sinus and nasal cavity as demonstrated in this study (Tsvetanov, 2016).

In most studies from Europe, the maxilla is reported to be more commonly affected than the mandible (Pechalova *et al.*, 2011; Tsvetanov, 2016). In this sample, the mandible was overwhelmingly more involved than the maxilla. This finding is explained by the mechanism of tooth loss where by in Europe most causes of tooth loss are accidental injury to the maxillary incisors whereas in developing countries, dental caries is the main instigating factor for development of these cysts, which usually occurs in the posterior regions of the jaws.

6.4. Histopathological features

Histological examination demonstrates that the cyst is usually lined by squamous epithelium. In newly formed cysts, the epithelial lining can exhibit signs of proliferation and arcing with marked inflammatory infiltrate but as the cyst gradually enlarges, the lining becomes dormant and consistent with varying amount of differentiation to mimic stratified squamous epithelium (Tsvetanov, 2016). The histopathological features of these cysts in this study are consistent with these finding. Occasionally, disseminated mucous cells or zones of ciliated pseudo-stratified columnar epithelium can be seen (Tsvetanov, 2016).

Calcifications within the lining of residual cysts are seen occasionally as shown in this sample. These calcifications usually present in long-established lesions and those with chronic inflammation. The calcified masses may be barely noticeable, fine grains of radio-opacity, or large irregular particles. Dystrophic calcifications occur due to the deposition of calcium salts in areas of chronic inflammation or in necrotic tissues (Sridevi *et al.*, 2014; High and Hirschmann, 1986). These calcifications further complicate the diagnosis of these cysts as a number of other odontogenic lesions may have similar presentation such as calcifying odontogenic cyst, dentinogenic ghost cell tumour and calcifying epithelial odontogenic tumour (Sridevi *et al.*, 2014).

Histopathological examination remains the main method of diagnosis of these lesions along with radiological correlation. However, heavily inflamed odontogenic cysts pose major diagnostic challenges as the inflammatory changes disguises the true features of the cyst on histological specimen (Boffano and Gallesio, 2010). In these cases, clinical

and radiological correlation along with histochemical markers becomes vital to arrive at an accurate diagnosis (Boffano and Gallesio, 2010).

6.5. Surgical Management

Residual cysts are usually surgically managed by enucleation, marsupialization or decompression depending on numerous anatomical and patient specific factors (Allon *et al.*, 2015). Smaller lesions can be enucleated entirely at the time of biopsy and is the ideal treatment choice. Enucleation was shown to be an acceptable method in this sample with low morbidity and recurrence rate. However, there are some contraindications to enucleation which include large lesions, difficult accessibility, proximity to adjacent vital structures, and patient's age. Hence, high-risk patients, younger individuals and the elderly should be managed with minimally invasive procedures to reduce morbidity (Torul *et al.*, 2018).

Larger residual cysts particularly those extending into adjacent anatomical structures should be managed with marsupialization over a period of time to reduce the size of the cyst followed by enucleation of the remaining cyst lining (Tsvetanov, 2016). Marsupialization is an accepted treatment method that entails suturing of the cystic lining with the oral mucosa with the aim of reducing the intra-cystic pressure and promoting new bone formation around the defect (Anavi *et al.*, 2011). This method was used for one extensive lesion in this sample that extended into the ramus of the mandible. A major disadvantage of this technique is that it requires multiple procedures under general anaesthesia.

Another method to reduce intra-cystic pressure is decompression. This method entails creating a window within the cyst wall that remains open to the oral cavity by placing a

device such as a drain or cannula. This method is also followed by enucleation of the shrunken cyst lining and allows for new bone deposition over time. Decompression is regarded as the more appropriate treatment option in patients that warrant a more conservative approach (Enislidis *et al.*, 2004). This method was not utilized in this population as most cysts were amenable to enucleation and obliteration of dead space with BIPP impregnated gauze packs.

6.6. Recurrence

Residual cysts are not known to recur frequently however no data is available in the literature regarding their recurrence rate. This is the first study to report on the recurrence rate of these cysts in a large series of patients and hence no comparison could be made. The main reasons for recurrence of these cysts are usually due to inadequate surgical access and fragmentation of the cystic lining leading to re-proliferation of the epithelium (Jamdade *et al.*, 2012). Therefore, periodic follow-up is essential to detect any recurrences and malignant transformation (Sridevi *et al.*, 2014).

Following surgical intervention, the defect gradually fills with bone however in certain cases, where the mandibular cortex is involved in an edentulous patient, then pathological fracture can occur as was demonstrated in one of the patients in this sample (Tsvetanov, 2016). Packing BIPP impregnated gauze within the cavity and gradually removing it over several weeks has been shown to be effective method in preventing post-operative infections and aiding bone formation at the same time. No other study has reported on this technique in the management of residual cysts.

6.7. Primary intraosseous odontogenic carcinoma

Radicular/residual cysts have the highest potential of all odontogenic cysts to transform in primary intraosseous odontogenic carcinoma (Bodner *et al.*, 2011). They have a predilection for older males and predominantly involve the mandible (Wenguang *et al.*, 2016). Although both cases in this sample occurred in the mandible, one patient was a middle aged female which did not fit the profile of patients affected by this devastating lesion. Bodner *et al.* (2011) reported that of all primary intraosseous odontogenic carcinoma reported in the literature, 60% developed from residual/radicular cysts followed by dentigerous cysts (16%) and odontogenic keratocysts (14%). This further highlights the need to perform a biopsy of all cystic lesions of the jaws to differentiate these inconspicuous appearing lesions.

The pathogenesis of malignant transformation of odontogenic cyst epithelium remains unknown. Some authors proposed that the prolonged chronic inflammation may be a precursor factor for malignant change in the cyst epithelium (Bodner *et al.*, 2011). This finding is supported by the presence of chronic infiltrate of lymphocytes and plasma cells in the cystic lining of malignant cyst epithelium (Bodner *et al.*, 2011).

When primary intraosseous carcinoma is detected in the cyst lining, then the management approach should be selected based on the extent of the lesion (Bodner *et al.*, 2011). If the carcinoma is within the cyst and has not invaded the adjacent bone, additional surgical management must be avoided and the patient should be monitored regularly. If the margins of the lesion are positive or there is carcinoma invading the surrounding bone as in both cases in this sample, additional management is indicated.

This entails combination of resection, radiation therapy, and chemotherapy with neck dissection if indicated (Bodner *et al.*, 2011).



Chapter 7

LIMITATIONS OF THIS STUDY

A number of challenges were faced during the course of the study. These are inherent to many retrospective analytical studies. These are listed below:

- A few records had to be excluded from the study due to incomplete information. In some records, it was challenging to obtain the patient's presenting clinical information, especially for earlier cases.
- The quality of radiographs utilized in this study could not be standardised due to the retrospective nature of this work. A number of different panoramic systems were used over the years with a variation in radiation output with a move towards digital systems in recent years.
- Only few patients underwent advanced imaging modalities (namely CT, CBCT and MR imaging) in this study sample. There are only a handful of case reports in the literature on the advanced imaging of this cyst and therefore inclusion of such imaging would have been valuable in this study. Advanced imaging was only used selectively by the treating clinician and reserved only for diagnostically and surgically challenging cases. This is to avoid high costs and unnecessary radiation exposure to the patient.

- A large number of patients were lost on follow-up. This has been noted in other studies conducted by the author. Frequently, many patients report that they are unable to attend their follow-up appointments as they do not have funds for transportation while many others reside far away from University of the Western Cape Oral Health centre.



Chapter 8

CONCLUSIONS

Residual cysts presented with clinical and radiological features that could mimic other destructive lesions in the jaws. These cysts occurred more commonly in older and edentulous patients and should be high on the list of differential diagnosis. They should not be taken lightly based on these features alone as they have the potential to cause significant bone resorption over time. All lesions should undergo biopsy and histopathological examination along with radiological correlation to arrive at an accurate diagnosis and to exclude rare malignant transformation into primary intraosseous odontogenic carcinoma. Surgical management of these cysts with enucleation is highly predictable with a very low rate of recurrence. Marsupialization/decompression should be reserved for extensive lesions in difficult to access anatomical regions.

Chapter 9

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Appendix 1

Data Collection Sheet

Record number				
Age (years)				
Gender	Male=1	Female=2		
Ethnicity	Caucasian=1	African=2	Mixed race=3	Indian =4
Symptomatic	yes=1	no=2		
Period (months)				
Pain	yes=1	no=2		
Swelling	yes=1	no=2		
Paraesthesia	yes=1	no=2		
Pus discharge	yes=1	no=2		
Edentulous jaw	yes=1	no=2		
Period since removal of causative tooth (months)				
Incidental finding	yes=1	no=2		
Jaw	Mandible=1	Maxilla=2		
Location	Anterior =1	Posterior=2	both=3	
Shape	Irregular=1	Round=2	Oval=3	
Radio-density	opaque=1	mixed=2	lucent=3	
Demarcation of lesion	well-defined=1	ill-defined=2		

Margins	Smooth=1	scalloped= 2	irregular=3	
Sclerotic margin	yes=1	no=2		
Size (longest diameter in mm)				
Loculation	unilocular=1	multilocular=2		
Displaced IAN canal	yes=1	no=2		
Displaced Maxillary antrum	yes=1	no=2		
Displaced nasal cavity	yes=1	no=2		
Expansion of cortex	yes=1	no=2		
Root resorption	yes=1	no=2		
Displacement of teeth	yes=1	no=2		
Initial diagnosis				
Histology				
Malignant transformation	yes=1	no=2		
Pathological fracture	yes=1	no=2		
Management	enucleation=1	marsupial ization=2	resection =3	none= 4
Intra-op findings				
Recurrence	yes=1	no=2	Not available=3	
Follow-up period (months)				

Additional notes:

Appendix 2



UNIVERSITY of the
WESTERN CAPE



23 June 2020

Dr F Titinchi
Faculty of Dentistry

Ethics Reference Number: BM20/5/13

Project Title: Residual cysts of the jaws: a retrospective study of characteristic features and management at the University of the Western Cape Oral Health Centre.

Approval Period: 23 June 2020 - 23 June 2023

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 annually by 30 November for the duration of the project.

Permission to conduct the study must be submitted to BMREC for record-keeping.

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

Director: Research Development
University of the Western Cape
Private Bag X 17
Bellville 7535
Republic of South Africa
Tel: +27 21 959 4111
Email: research-ethics@uwc.ac.za

NHREC Registration Number: BMREC-130416-050

FROM HOPE TO ACTION THROUGH KNOWLEDGE.

Appendix 3

**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

Faculty of Dentistry

UWC

04 April 2020

Dear Professor Myburgh,

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 (Student No: 2768296). My supervisor is Prof JA Morkel.

The proposed topic of my research is "Residual cysts of the jaws: a retrospective study of characteristic features and management at the University of the Western Cape Oral Health Centre".

The objectives of the study are to describe the clinical, radiographic and histopathological features of patients presenting with residual cysts of the jaws at the University of the Western Cape Oral Health Centre as well as their management and recurrence patterns.

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 purposes only and will only be kept for the duration of the study.

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.

Sincerely,

Fadi Titinchi