

THREE-DIMENSIONAL VOLUMETRIC ANALYSIS OF LARGE JAW CYSTIC LESIONS AFTER MARSUPIALIZATION

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ABSTRACT

The Aim: This study aimed to evaluate the effectiveness of Marsupialization on different jaw cystic lesions by volumetric analysis using three-dimensional computed tomography(CBCT).

Patients and Methods: Fifteen patients were enrolled for this study. All patients underwent surgical Marsupialization of different jaw cystic lesion. Preoperative CBCT for all patients was compared to their postoperative views after follow up period ranged from 6 and 24 months. Three dimensional volumetric analysis was performed for all cases to assess the differences in volumetric change rate, initial volume of the lesion, duration, location of the lesion, and pathological diagnosis.

Results: All cystic lesion showed great reduction in size measured on CBCT. There were no important surgical complication recorded throughout the study. The reduction rate among all patients ranged from **58.83 – 73.56 %**. The results showed that higher reduction rates were associated with a long Marsupialization period, and younger patient age.

Conclusion: Marsupialization is a valuable treatment option for different jaw cystic lesion especially with large size in all patients. The effectiveness of this treatment modality is proportional to long treatment duration, younger age group of the patients, as well as nature of the cystic lesion.

KEYWORDS: Marsupialization; cystic; 3D; volume; CBCT

INTRODUCTION

Intraosseous cysts are one of the common lesions that could be seen in the region of maxillofacial.¹ Actually, cyst is a pathological cavity that may be lined by epithelium or not, while its content may be fluid, semifluid, or empty. This differs from an abscess which contains pus.² It is challenging

to differentially diagnose cyst or cyst like lesions that may be faced daily in maxillofacial surgery, as many other lesions may have similar clinical and radiographic appearance. Cyst and cyst-like lesions differ in their origin. They could be odontogenic or non-odontogenic, with or without calcifications, benign in nature, locally invasive or with very aggressive behavior.³

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While a considerable proportion of maxillofacial cysts are asymptomatic and accidentally discovered, patients may present with persistent pain, swelling, paresthesia, teeth displacement, and pathological fractures.³ Different types of jaw cysts could be seen based on clinical and radiographic diagnosis. Their distribution varies according to their types. The most common type is radicular cysts 56%. Dentigerous cysts also have considerable prevalence 17%, while odontogenic keratocysts present about 11%.^(4,5) A Dentigerous cyst originated from dental follicle, and enlarges by fluid accumulation inside it⁶. The width of the cyst is larger than 3-4 mm^(5,7). This cyst is characterized by cyst wall attached to the neck of an unerupted tooth and enclosing its crown. Dentigerous cyst is more common in males, presented more in children and young adult, as associated with unerupted teeth, however it could be seen also during the middle age group of the patients.

Cystic appearance could be seen with some odontogenic tumors. As an examples of these tumors are central odontogenic fibroma, odontogenic myxoma, as well as odontogenic keratocyst, ameloblastoma, ameloblastic fibroma.^(3, 8) In 2005, Odontogenic keratocyst was classified by World Health Organization (WHO) as a benign tumor. It was described histologically as it has an odontogenic epithelium, mature, fibrous stroma with no odontogenic ectomesenchyme. This classification was considered at that time based on potential of aggressive behavior of keratocystic odontogenic tumors. It was advised to have a careful follow up following surgical management.⁹ In 2017, WHO reported that keratocystic tumor is not considered anymore as neoplastic lesion and reclassified it as odontogenic keratocyst which is belong to the classification of developmental odontogenic cysts.¹⁰

Another example of odontogenic tumor is Ameloblastoma. However it is considered as a benign tumor but it is invasive and has destructive growth pattern. Four main types of ameloblastoma are recognized, it may be multicystic, unicystic, extra-osseous, or desmoplastic.¹¹

Treatments of jaw cystic lesions include different

modalities that may be simple procedure or with more invasive surgical intervention. Treatment modalities include decompression, Marsupialization, enucleation, resection, or a combination of these modalities. However most commonly surgical treatment modalities of cyst of the oral cavity are enucleation and marsupialization.¹² Enucleation means complete excision of cyst lining with its content. This is permitted healing by primary intention. Marsupialization on the other hand means partial excision of cyst wall, and it is presented as Partsch's operation.¹³

As enucleation provides total removal of cyst wall and its content from interior of the bony cavity, it permits blood clot formation and healing of this cavity physiologically. It is considered a procedure of choice in small sized, readily accessible cysts, or cysts that are not involved extensively with vital teeth, soft tissues, or very close to vital structures.¹⁴ Cyst Marsupialization by simple definition, is the creation of a pouch surgically, by removing part of the overlying mucoperiosteum and accompanying cyst wall. By this maneuver a window is created which leads to interstitial tissue of bone. This is followed by suturing of the incised mucosa with the border of the remaining cyst wall that has been left.^(15,16) The reduced tension gives rise to healing and formation of bone with the surrounding bone.¹⁷

Many factors could affect the selection of proper treatment modality, such as how much the lesion extended, encroachment of the lesion on adjacent vital structures, origin, clinical behavior of the lesion, patient cooperation and systemic condition.¹⁸

Principles of treatment of dentigerous cysts are enucleation, marsupialization.^(6,19) If there is no affection on damaging successors or vital nearby structures such as inferior alveolar nerve, enucleation is a better selection.²⁰ Marsupialization is a very valuable method for management of Dentigerous cysts especially for children, as it provides preservation of the successors and natural vital structures and is considered a minimally invasive procedure,²¹ compared to other treatment methods. It has a good prognosis with no relapse.⁶

Treatment of odontogenic keratocyst (OKC) is controversial due to the aggressive behavior of the lesion and their high recurrence rate. Currently, there are two common treatments for OKC, enucleation and decompression.²² Clinician may manage OKC by enucleation with the followed by Carnoy's solution), or cryotherapy with liquid nitrogen, or by more radical intervention; surgical resection.²³ All these treatment modalities may give rise to several complications. Alternative treatment is marsupialization which creates a cystic cavity that are connected to the oral cavity,²⁴ and permits cystic fluid to come out then enucleation is the final procedure after some decompression period.

The unicystic ameloblastoma reported a lower recurrence rate if it is compared to the other types and is the only type that responds to conservative treatment.^(11, 25) Despite progress in reconstructive surgery, extensive jaw resection influences quality of life and causes numerous complications.^(25, 26) For this cause particularly, conservative treatment of unicystic ameloblastoma in young patients should be superior choice over other treatment methods.

Attempts to estimate lesion shrinkage after marsupialization have used a variety of approximate measurement methods, but many previous reports have used two-dimensional (2D) X-ray (Panorex) imaging.^(17, 27, 28, 29) Comparing CBCT to conventional computed tomography, CBCT generally results in significantly less exposure to radiation, which can be further reduced by using low-dose protocols.³⁰ Despite that CBCT give inadequate contrast of the soft tissue and standardized grayscale values still selected as first choice and gold standard in digital oral and maxillofacial surgery because it provide better accessibility as well as relatively cheaper cost.^(31, 32) In general three-dimensional (3D) computed tomography (CT) is more reliable and accurate than two-dimensional CT^(28, 33) and has been successfully used in maxillofacial surgery.^(34, 35) Three-dimensional (3D) imaging is the method of choice for preoperative cyst volume and diameter assessment; previous reports demonstrated that 3D computed tomography (CT) could precisely

measure the cyst volume, elaborate the cyst border, and evaluate the anatomical relations of the cyst.³⁶ The rationale of the present study is to assess volumetric shrinkage following marsupialization of cystic lesions through three dimensional analysis.

PATIENTS AND METHODS

The study included fifteen patients (9 males and 6 females) with ages ranged from 8 to 40 years. The selection criteria include the followings: patients with large cystic lesion as appear in 2D radiography. The size of the cystic lesion should be greater than 2 cm, patients free from any systemic diseases that could affect bone healing, and gave positive results on aspiration biopsy. The exclusion criteria include the followings: patients with medical compromises of immunodeficiency, autoimmune disease, radio or chemotherapy or trauma, as they were not the ideal candidate for ideal wound healing capacity, and patients with previous surgical intervention.

The study were limited to three types of cystic lesions, Dentegrous cyst, keratocystic lesion, and unicystic ameloblastoma. The details of the treatment plan and methodology of this study were clarified to each individual patient. Written informed consents have been collected prior to surgical intervention. It is confirmed that none of the study's procedures violated the main principles of the Declaration of Helsinki.³⁷ The study was approved by the ethics committee of faculty of Dentistry, Cairo University. Complete medical and dental history was taken for all eligible patients that fulfill inclusion criteria. Intra-oral clinical examination was carried out to check for any accompanying signs, bone expansion, color and texture of covering mucosa, conditions of related teeth. Extra-oral examination also was carried out to check for any facial asymmetry caused by the lesion, overlying skin, related lymph node, and any alterations in sensation. Preoperative radiographic examination for all patients were carried out using panoramic radiography to check for size and extensions of the cystic lesion, any relation or encroachment of the cystic lesion with vital structures, and bone support of involved teeth.

All patients had aspiration biopsy before surgical procedure to confirm cystic nature of the lesion. Preoperative CBCT were taken for all patients to examine and analyze cystic cavity in 3D fashion. For all CBCT views, every slice cut should be at least 1 mm. Measurements were done pre and every 6 months post-Marsupialization.

Marsupialization was the procedure of choice for all cases and it done in the outpatient clinic under local anesthesia using arctician 4% with adrenaline 1/100000 that was injected as nerve block for the affected region as well as mucosal infiltration around the lesion. The patient rinsed with 2% chlorohexidine mouthwash, then mucosal incision using a #15 scalpel was done in circular fashion in the planned surgical site. Some cases presented with buccal bone defect with different size, this defect is widened as needed for the success of the procedure, however other cases presented with complete intact non perforated buccal cortical plate of bone, in such cases, a hole is created through this bone to give access to the cyst wall and intra cystic cavity. This is achieved by using round bur mounted on motor driven hand-piece with copious sterile saline irrigation. The hole size was proportional to the width of the mucosal opening. After exposing the cystic lesion, a part of cyst wall tissue was incised and removed then fixed using formalin solution and sent for histological assessment (Fig 1). The edge of incised cystic lesion was fixed with oral mucosa by suturing with 3-0 silk suture. Normal sterile saline was used for irrigation the cystic cavity. The removed mucosal tissues and cyst lining was sent as incisional biopsy for histopathological examination. The cystic cavity was packed with a gauze drain. The gauze was allowed to stand for 2 days after which an alginate impression was made and an acrylic obturator was fabricated. On insertion of acrylic obturator splint, the stitches are removed. Postoperative medications for patients were prescribed which include amoxicillin 875mg with clavulanic acid 125mg* (every 12 hours), ibuprofen

* Curam 1 gm, Sandoz

400mg** (twice a day). Children patients received oral antibiotic therapy according to body weight. Instruct the patients to rinse using Chlorhexidine Hcl *** mouth wash for one minute, twice per day for 7 days. Patients that reported allergy or on other drugs, prescription was replaced putting into consideration its interaction with the other drugs. Explain to the patient patients how they can do irrigation at home with sterile saline post marsupialization surgery. Surgical wound was frequently checked and the obturator was adjusted to make room for tissue healing.



Fig. (1) Marsupialization of OKC in the region of posterior maxilla showing the hole of the cystic cavity.

The determination of cyst volume by Plan mecca Romixes software was used. The DICOM database (CT digital imaging and communications in medicine) was transferred to the software and were clipped in the axial, coronal and cross-sectional views. When superficial boundary of the intra osseous cystic lesion was demarcated, preprogrammed automated system was activated and semi-automatic segmentation was carried out. In views where superficial boundary margins was not clear, the segmentation tool was used manually. Specifying the cystic dimensions in every slice permits displaying a 3D volumetric image. This 3D image is conducted by collection of every slice in axial plane, then, 3D volume of the cystic lesion

** Brufen 400, Abbott

*** Hexitol mouth wash, ADCO

was presented with its measurements. (Fig. 2) The postoperative volume measurements was compared to preoperative volume calculate the value of reduction. (Fig. 3) Statistical analysis methods was performed with the preoperative image, 6 months postoperatively, and the last image obtained immediately at end of follow up or pre-enucleation. Statistical Analysis System (SAS) version 9.4 (SAS Institute, Cary, NC, USA) was used for statistical analyses of the results obtained. The difference is considered significant when the p value measure <0.05 .

RESULTS

Fifteen patients with large cystic lesion were included in this study. The patient's sex distribution were 9 males and 6 females. The ages of the patients were 8 to 40 years, with a mean age of 24 years. The patients included three types of cystic lesion and arranged as follows: six patients had Dentegrous

cysts, five patients had keratocystic lesion, and four patients with unicystic ameloblastoma. According to the affected site they include 7 maxillary and 8 mandibular cystic lesion. No vital complaint was recorded by any patients and surgical procedure was well tolerated by all patients. No significant complications were encountered either postoperatively or during the whole follow up time.

Regarding Dentegrous cyst group, mean reduction rate in cyst volume was 84.76% in the first 6 months. Complete resolution of the cystic lesion was achieved during for follow-up period with maximum of 24 months, and the related unerupted teeth were erupted till the bone level. (Fig. 4) In keratocystic lesion and unicystic ameloblastoma groups, the sizes of the lesion were dramatically reduced by the mean rate of 52.32% due to bone deposition and related teeth had more bone support and decreased mobility. The reduction rate and bone healing was evident in postoperative radiography

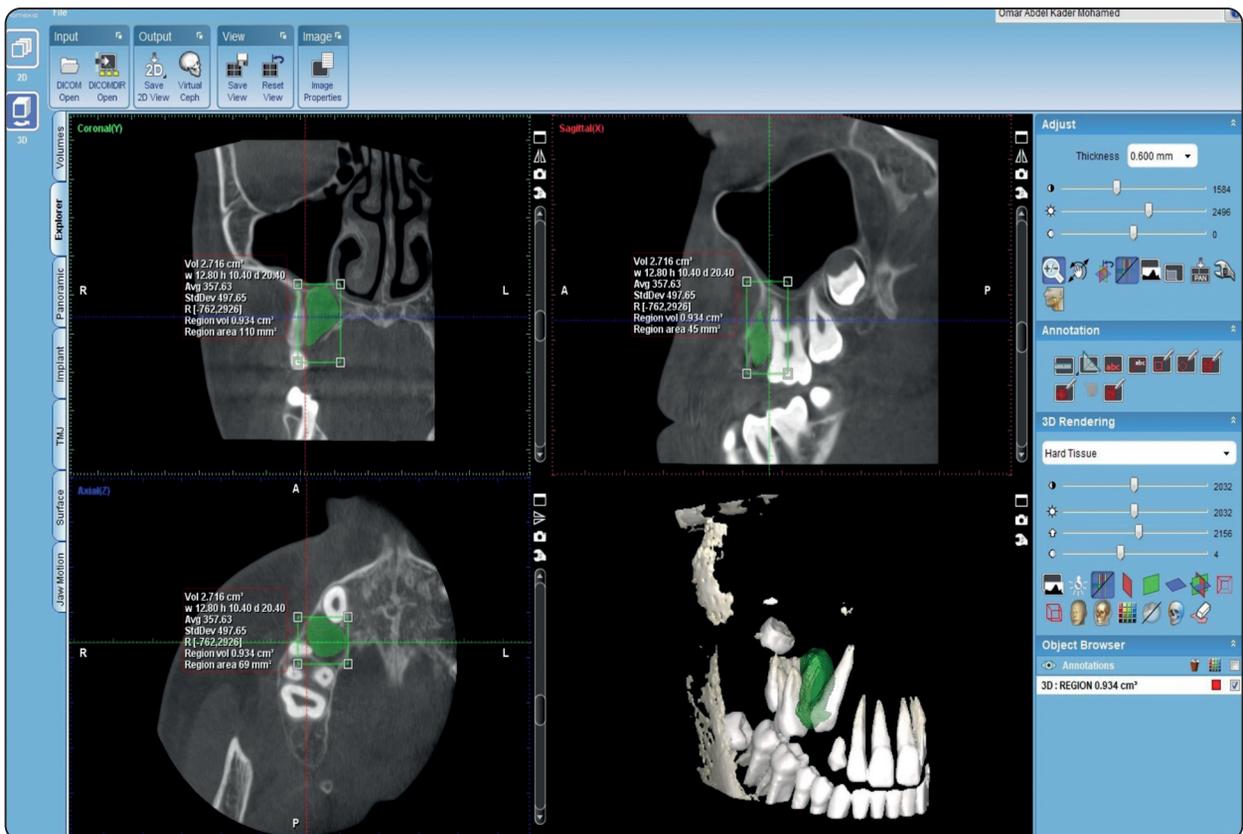


Fig. (2): 3d volumetric analysis of cystic lesion using Romexis Software

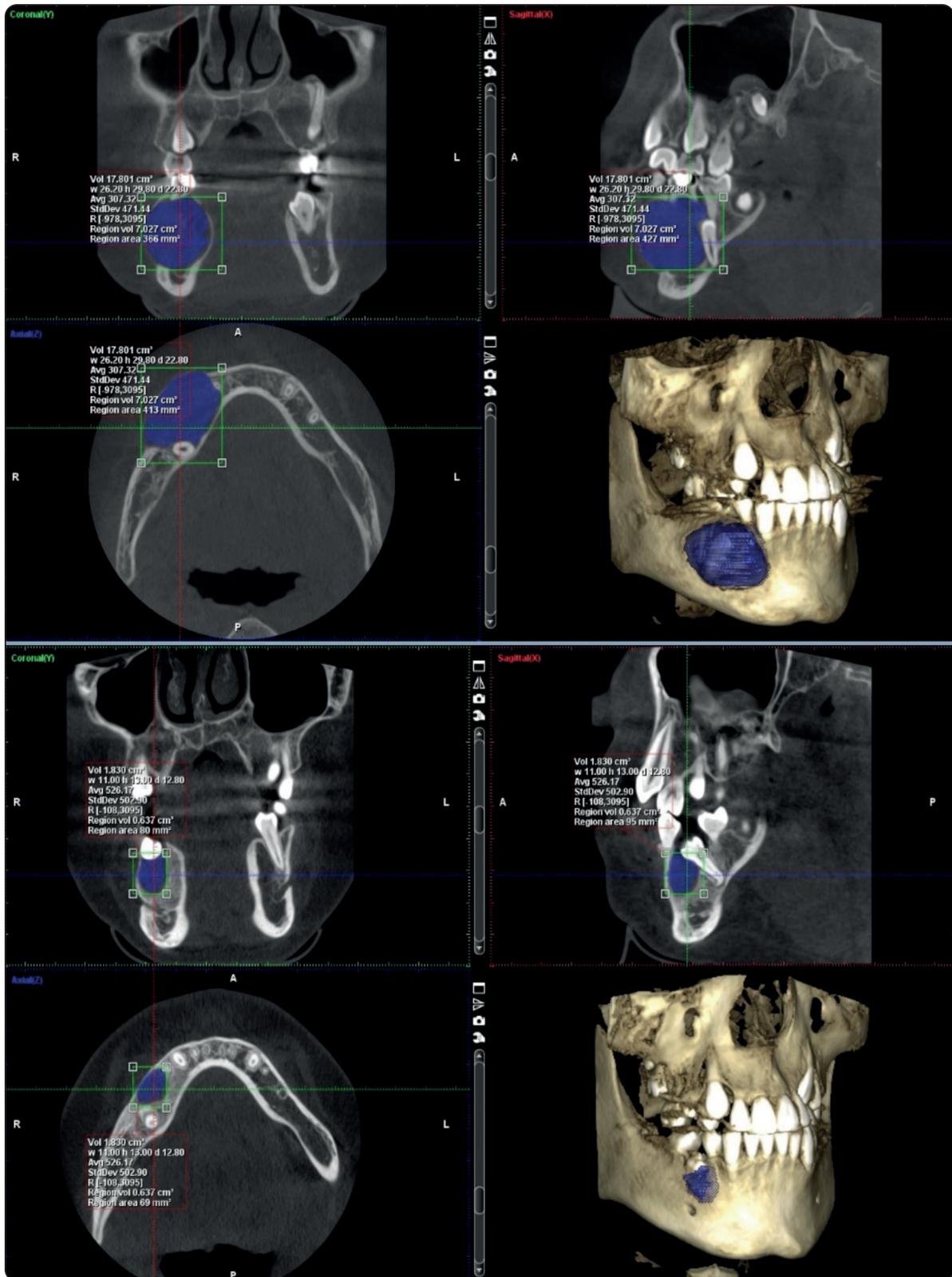


Fig. (3): Preoperative and 6 month postoperative CBCT of dentigerous cyst for 3D volumetric comparison



Fig. (4) Eruption of unerupted tooth to the bone level after 6 month post marsupialization of dentigerous cyst

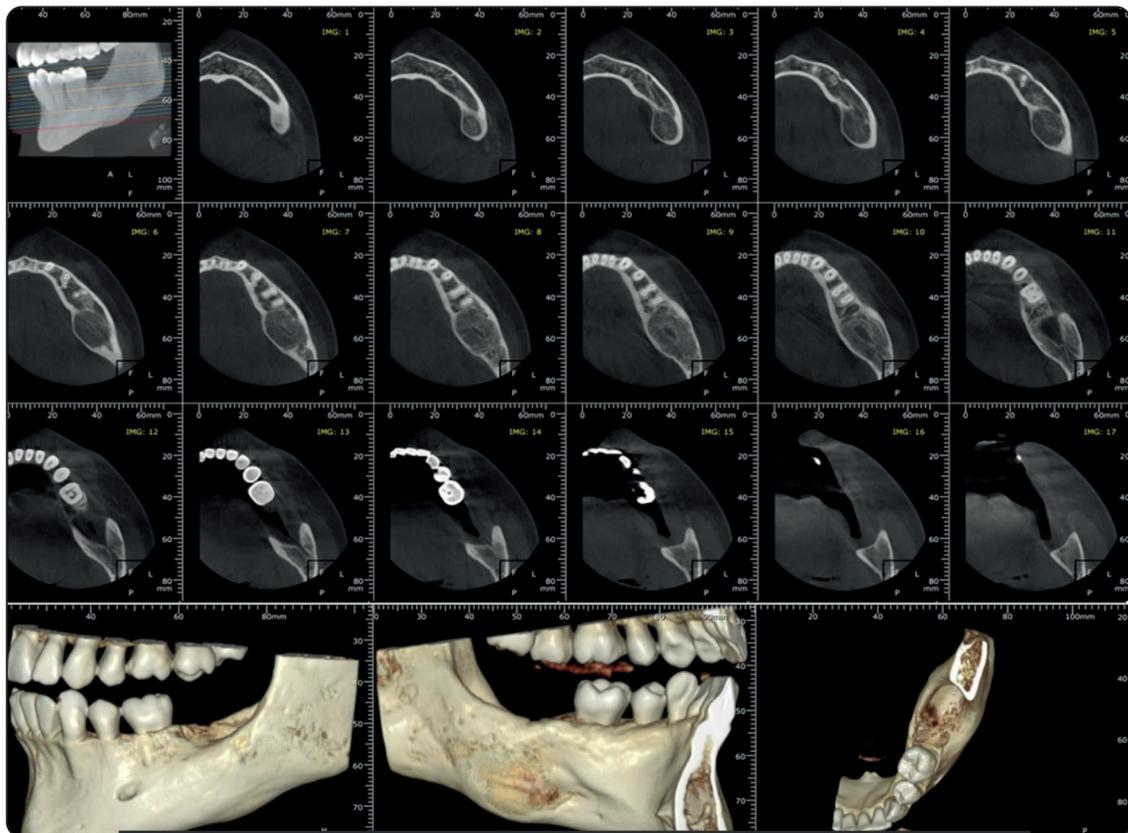


Fig. (5) The evident bone healing in unicystic ameloblastoma 1 year post operatively with pattern directed toward center of the lesion

with pattern directed toward center of the lesion. (Fig 5) (Tab 1).

In general, the patient's results reported a good reduction in the mean volume of all cystic lesions by reduction rate of 73.56 %. The average marsupialization follow up time was 15 months. The period for marsupialization varied according to nature of cystic lesions. The minimum duration was 6 months and the maximum was 24 months. Higher reduction rates were found in Dentegrous cyst group, young patient age, and long duration of marsupialization.

TABLE (1) Distribution of all cases and mean reduction in size postoperatively

Type of The lesion	No. of case	Mean of difference in size		
		6 months postop.	15 months postop.	24 months postop
DentegrousCysts	6	84.76 %	96.34%	100%
Odontogenic Keratocyst	5	58.86 %	67.45	Enucleation and curettage
Unicystic Ameloblastoma	4	45.78%	56.89	

DISCUSSION

Large cystic lesions management has been considered a debatable issue for a long time.³⁸ Management of cystic lesions include several surgical interventions mainly dependent on their size. Small lesions are managed by total enucleation, however large lesions are managed by decompression or marsupialization. A combination of these techniques may be the most suitable choice for some cases, decompression or marsupialization procedure is attempted first aiming to have reduction of the size of these lesion, subsequently it will be easier for surgical excision or removal without endangering nearby teeth or any vital structures.³⁹ Some disadvantages of the procedure reported by Aldama.⁴⁰ who preferred the full removal of the cyst

wall in order to have a good prognosis. In addition, Kruger⁴¹ stated that histological transformations could happened to developmental cyst, and it is considered important warning not leave any remnants of cyst capsule, and careful examination of the acquired surgical specimen is a must. In contrary, according to Branon⁴²this is very unlikely.

Marsupialization procedure have very important advantages such as keeping teeth involved in place, along with preserving vital structures like mandibular canal and nasal cavity and maxillary sinus from being injured or affected with more radical surgical procedure. The present study supports the findings of previous studies who assured the viability of this maneuver.

The objective of this study was to investigate the effect of marsupialization procedure in forms of the radiographic 3D volumetric reduction for different types of large cystic lesions. Despite the different origins, the multiple types of cystic lesions show a apparently the same clinical presentation; location, growth direction, expansion, or the occurrence of complications, are important factors for differentiation, however it is confirmed with biopsy which was taken as a part of marsupialization procedure. Apart from odontogenic tumor like unicystic ameloblastoma, odontogenic cysts usually not reached a very large size, it is very rare. Indeed, the odontogenic keratocyst (OKC), Dentegrous cyst could be presented with such exceptional large size. In the present study three types of cystic lesions that affect both jaws were selected based on they can reach very large size of the bony defect.

Different line of treatments are considered for dentigerous cysts, however usually marsupialization is considered a good choice and a very useful procedure especially in children and young adults for treating this type of jaw cysts, as it is considered minimally invasive surgery that preserves the natural structures as stated by Ertas and Yavuz²¹. It is also gives very good results regarding preservation and eruption of involved unerupted teeth. The longer the

period of marsupialization the better reduction in size and better healing of bone which may preclude necessity for other treatment. Our results showed accelerated reduction in the volume of Dentegrous cyst may be owing to fast regeneration capacity of younger age group of the patients.

The same assumption about marsupialization could be applied for OKC. It has been considered as a conservative management for OKC.^(43, 44) However the explanation of the effect of this procedure in reducing the size of the OKC and repair normal bone is not fully clarified.⁴⁵ it is highly complicated decision making if the aggressive surgical intervention is the better choice while cosmetic concern and functional morbidity are considered. Difference in recurrence rate between decompression followed by enucleation and aggressive interventions is not significantly.⁴⁶ Results of the present study showed agreement with Madras and Lapointe⁴⁷ who stated that enucleation when carried out after marsupialization is considered the most effective management, and cannot depend only on marsupialization as final management.

In the present study, cases of unicystic ameloblastoma had conservative treatments after marsupialization; radical resection with surgical margins was avoided because marsupialization reduced the volume of ameloblastoma in all cases. Besides, no recurrence was noticed during the follow-up period. Despite the relatively large size of the lesion marsupialization made it possible to avoid injury to vital structure or morbidity that may happened after resection. This result showed that marsupialization for unicystic ameloblastoma can prevent a major decline in a patient's quality of life after treatment.

Radiological follow-up of cyst volume after marsupialization is of paramount importance in order to measure the rate of volume reduction and the time for enucleation if needed.⁴⁸ The 2D radiography (Panorex) provide less radiation dose to the patients as well as it is cheaper in cost than

CTs. However, regarding the determination of exact volumetric changes, 2D radiography is considered with too limited value compared to 3D radiography.⁴⁹ Its shortcomings include inaccurate discrimination of cyst border and the inability to assess the cyst relation with the surrounding vital structures.²⁸ Thus, 3D imaging has become the modality of choice for preoperative assessment of cyst volume and diameter, which allows for accurate measurement of the cyst volume, demonstrates the cyst border, and evaluates the anatomical relations of the cyst. CBCT is considered superior choice compared to panoramic radiography, as it gives more information about high bone detail, accurate data about dimensions of the lesion and its association with adjacent vital structures (orbital, nasal, sinus cavities). It is also gives valuable postoperative data during follow up time.⁵⁰ In addition, Alhowalia et al. stated that CBCT is a valuable tool to have an accurate measurements of volume of cavities within bone that have been artificially created. They assessed measurements volume of simulated periapical lesions. They emphasize the importance of using appropriate software and training.⁵¹ For correct volume measurement of the lesion to be segmented, the appropriate threshold level and borders should be selected when using automated segmenting method of the software. Threshold level and borders of the lesions that not be clear may affect accuracy of the obtained measurements.⁵²

In the present study, effects of marsupialization were evaluated using 3D-CBCT and using Planmeca ProMax® 3D Max unit with 3D reconstruction software. All lesion volumes were measured at standard intervals (preoperatively, and at 6 months intervals postoperatively). This give a standard method of calculating the shrinkage rate of large cystic lesion.

In agreement with our results of this study, many studies on the 3-D volumetric analysis for assessment value of decompression surgery of odontogenic cystic lesions including ameloblastoma have reported that the reduction in volume of the

cystic lesions was related to the time elapsed leaving the lesions for decompression^(49,53,54) Specifically, Shudou et al. measured the 3-D volumetric change after marsupialization surgery for odontogenic keratocyst lesions and stated that the volume of the lesions was inversely correlated with the duration after marsupialization.⁴⁵ Jeong et al. reported that decompression was more effective when performed for more than 6 months.⁴⁹ Asutay et al. also performed 3-D volumetric analysis of the effect of decompression and reported that the percentage of volume decrease of ameloblastoma was $44.50 \pm 14.41\%$ from the preoperative volume to the volume 6 months postoperation.³⁶

Based on our results, bone defect volumetric reduction was correlated to the rate of reduction achieved. This finding implicate the conclusion that a longer marsupialization duration were combined with more successful management either by complete resolution as found with dentigerous cyst, or to make the following step of enucleation to be less complicated. This finding is the same with the younger age group of the patients and that was agreed by Song et al,⁵³ who presented an important correlations with age of the patients.

In the present study the Initial volume of the cystic lesion was not significant. This with agreement of Anavi et al. who stated that the initial size of the cyst did not affect the end results of decompression.²⁴ However this is on contrary with other study who stated that the reduction rate correlated with the initial volume.⁵⁵ Nakamura et al. make analysis to cases of cystic ameloblastoma that was managed by marsupialization. They showed that, in teenaged patients marsupialization was more effective. They also reported that with marsupialization of the younger age group there is potential for new bone formation, and that was influenced mainly by the age of patients. Besides, with every five cases in which marsupialization was extremely effective there are four cases showed unioocular appearance on radiographs.²⁵ In the present study, effect of marsupialization is affected by pathological

diagnosis, as observed with dentigerous cyst group of patients which showed accelerated reduction rate of their 3D volume. Other studies reported that OKC decompression resulted in more favorable outcomes than results of cysts of other types.^(25, 56) However Anavi et al. in their study stated that effect of marsupialization did not affected by pathological diagnosis.²⁴

In conclusion, marsupialization is a valuable treatment option contributed to volumetric reduction of different large cystic lesions of both jaws, as showed by 3D volumetric analysis. The effectiveness of this treatment modality is proportional to long treatment duration, and younger age group of the patients as well as nature of cystic lesion. In large cystic lesions, marsupialization should be favored over other methods of treatment.

CONCLUSION

Marsupialization is a valuable treatment option for different jaw cystic lesion especially with large size in all patients. The effectiveness of this treatment modality is proportional to long treatment duration, younger age group of the patients, as well as nature of the cystic lesion.

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