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Conservative management of large mandibular dentigerous cysts with a novel approach for follow up: Two case reports

Ieva Gendviliene[1], Paul Legrand[2,3], Laura Ferreira Pinheiro Nicolielo[2], Deepti Sinha[2], Yannick Spaey[2,3], Constantinus Politis[2], Reinhilde Jacobs[2]

SUMMARY

Background. Dentigerous cysts are odontogenic cysts of the jaws accounting for approximately 24% of all epithelium-lined jaw cysts. Rarely, these cysts can become extremely large causing additional symptoms, while challenging treatment. This article represents a novel approach for radiological treatment follow up of large dentigerous cysts.

Material and methods. Dentigerous cysts were treated by marsupialization, using dedicated obturators and enucleation of the cyst afterwards. Teeth vitality in the lesion was regularly assessed by laser doppler flowmetry, follow-up of lesion regression was accomplished using semi-automatic radiographic analysis of bone healing quantification.

Results. Cystic marsupialization by means of the obturator resulted in a number of advantages compared to a conventional approach: lower risk of spontaneous fracture of the mandible, anatomical structure preservation, mandibular canal identification and preservation of tooth vitality as measured by intraoral laser doppler flowmetry. Notwithstanding that a 1-year follow-up panoramic image could not visualize a remnant bone defect nor cystic lesion recurrence, 3D-CBCT based semi-automated bone quantification could only demonstrate an increase of 46% of mineralized bone volume one year after surgery. Bone healing typically occurred starting from the periphery of the original lesion towards the inner core.

Conclusions. The presently reported cystic marsupialization was useful as a preliminary treatment for subsequent enucleation. 3-D CBCT based objective quantification of the bone volume and healing can provide new insights in lesion healing in general and more particular in the outcome of specific diagnostic and therapeutic challenges.

Keywords: cone-beam computed tomography, dentigerous cyst, surgical decompression.

INTRODUCTION

Dentigerous cysts are odontogenic cysts of the jaws accounting for approximately 24% of all epithelium-lined jaw cysts which enclose the crown and are attached to the neck of an unerupted tooth (1-3). With increasing pressure of an enlarging cyst, the unerupted tooth can be deflected from its path of eruption. They occur in the mandible's posterior area, most frequently involving the mandibular third molars, followed by the maxillary canine and the maxillary third molars (4). Dentigerous cysts are generally observed in the second and third decade of life (5).

In most cases, dentigerous cysts are asypmtomatic and usually diagnosed incidentally during routine radiological examination, but rarely, these cysts can get secondarily infected and patients usually present with symptoms such as, swelling and pain (6). Occasionally they can become extremely large and cause cortical expansion and erosion, facial asymmetry, teeth displacement, delayed tooth eruption, radicular resorption of teeth, displacement and obliteration of the maxillary antrum and nasal cavities, paraesthesia of inferior alveolar nerve, metaplastic and dysplastic changes are also known to occur (2, 7, 8).

[1]Institute of Odontology, Faculty of Medicine, Vilnius University, Lithuania
[2]Omfs Impath Research Group, Department of Imaging and Pathology, Faculty of Medicine, University of Leuven and Department of Oral and Maxillofacial Surgery, University Hospitals Leuven, Leuven, Belgium
[3]Department of Oral and Maxillofacial Surgery, Maria ziekenhuis Overpelt, Belgium

Address correspondence to Ieva Gendviliene, Institute of Odontology, Faculty of Medicine, Vilnius University, Zalgiris str. 115, LT-08217, Vilnius, Lithuania..
E-mail address: ieva.gendviliene@gmail.com

Typical radiographic features include a unilocular radiolucent lesion with well-defined sclerotic margin that is associated with the crown of an unerupted tooth (7). Cone-beam computed tomographic (CBCT) imaging is an advanced modality to diagnose large cysts as it’s sagittal, coronal and axial images eliminate the superimposition of anatomical structures (4, 9). The main advantages of CBCT imaging are its accessibility, easy handling and that it offers a real-size dataset with 3D reconstructions based on a single scan with a low radiaton dose (10). Differential diagnoses for dentigerous cysts include ameloblastoma, adenomatoid odontogenic tumor, ameloblastic fibroma, periodontal cysts and keratocyst odontogenic tumor (3, 4). The final diagnosis must be based on macroscopic and microscopic examination. Histopathologically, a dentigerous cyst reveals a nonkeratinized stratified squamous epithelium consisting of mucosebaceous, ciliated, and rarely sebaceous cells (5).

Dentigerous cysts are most commonly treated by total enucleation, marsupialization or decompression of the cyst and its removal afterwards with or without impacted tooth removal (11). The criteria for selecting treatment modality depends upon the size and site of the cyst, patient age, dentition and involvement of vital structures (12). The purpose of this article was to describe conservative management of large dentigerous cysts in the mandible’s posterior area and a novel approach for treatment follow–up.

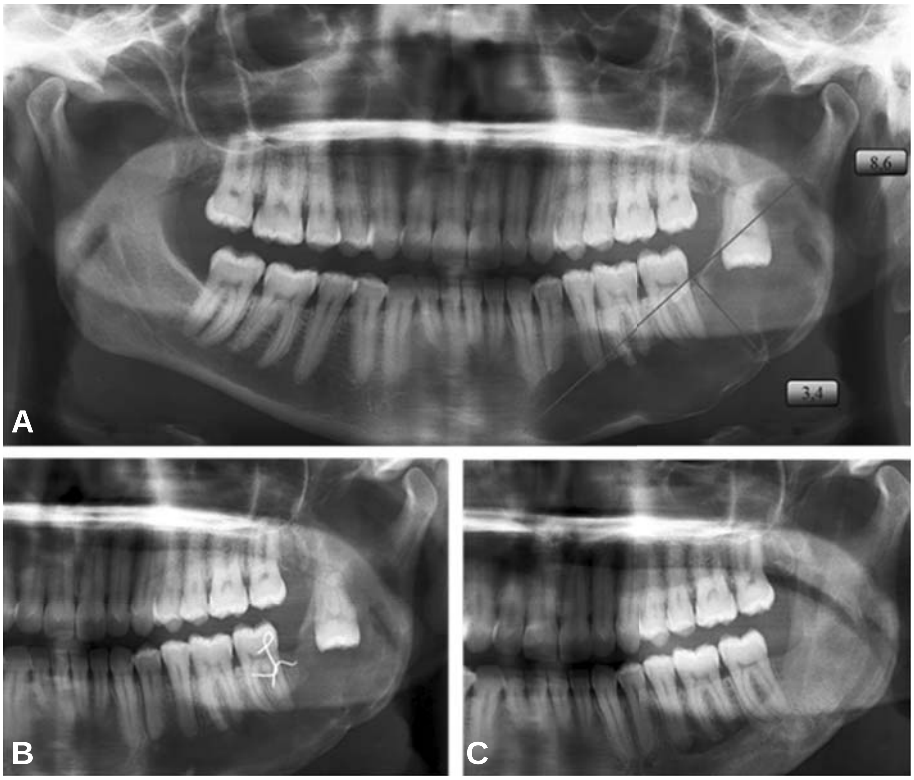

Fig. 1. Panoramic radiographic shows lesion in a 33-year-old female patient. A – preoperatively panoramic radiograph. B – panoramic radiograph after lesion decompression. C – panoramic radiograph 1 year postoperatively.

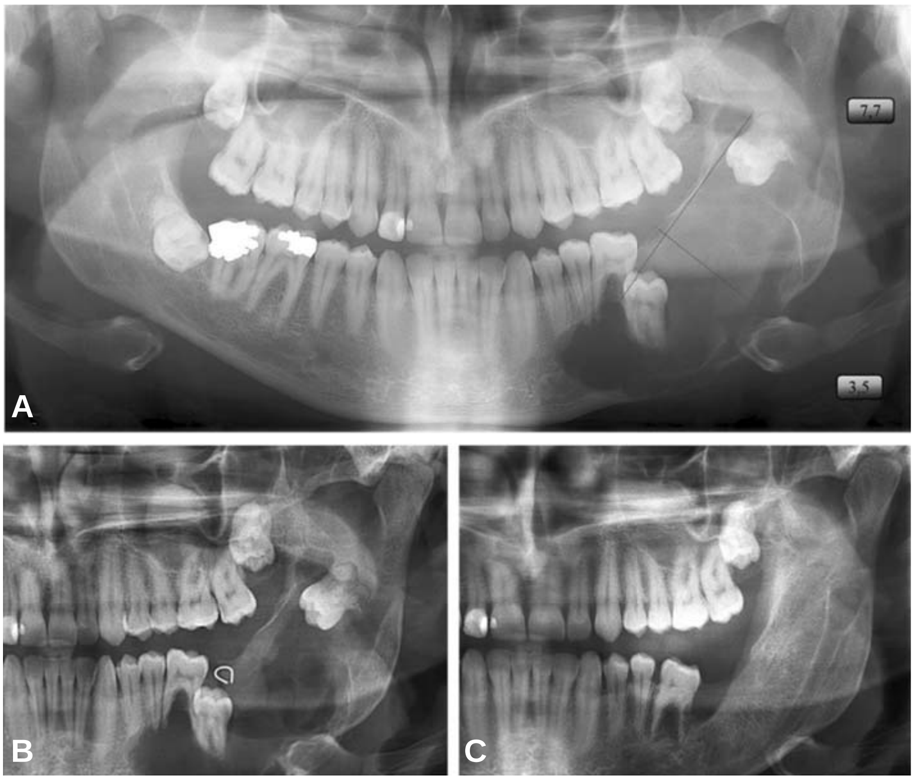

Fig. 2. Panoramic radiographic shows lesion in a 23-year-old male patient. A – preoperatively Panoramic radiograph. B – panoramic radiograph after lesion decompression. C – panoramic radiograph 4 years postoperatively.

CASE DESCRIPTION AND RESULTS

Considering the age of two patients, their occlusal status, size of the cysts, position, absence of identifiable mandibular canal, a conservative treatment modality was decided upon. The main objectives of the treatment were clinical and radiographic elimina-

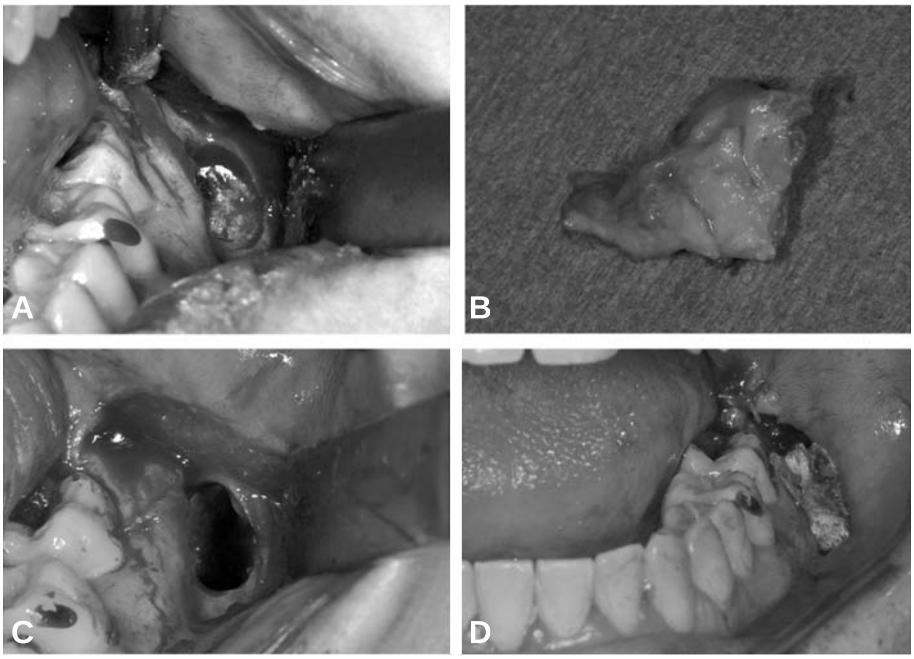

Fig. 3. Marsupialization procedure. A – creating cystic opening. B – biopsy. C – cystic opening. D – tamponation of the defect with terramycin gauze.

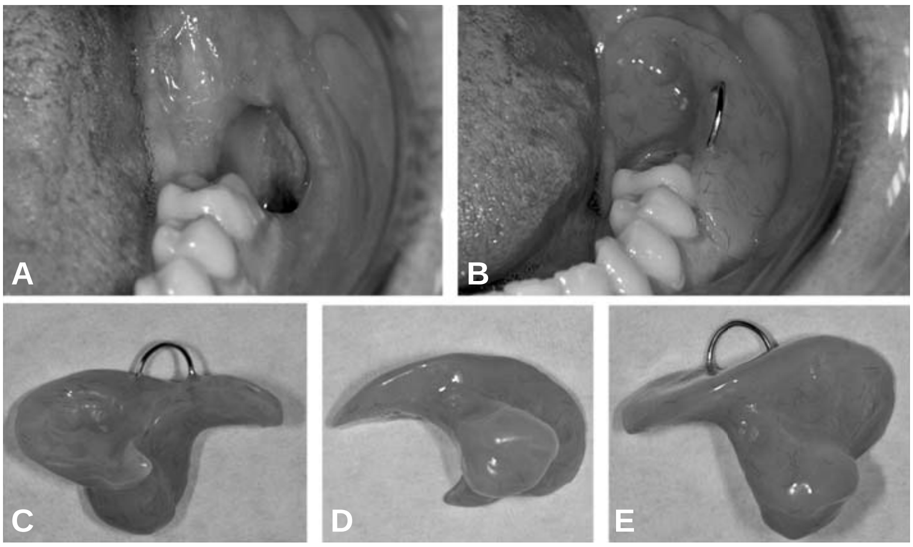

Fig. 4. Obturator making. A – cystic opening. B – obturator in the cystic opening. C, D, E – different sides of obturator.

tion of the pathologic entity with minimally invasive surgery and minimal postoperative morbidity. Case 1 – a 33-year-old female patient complaining of increased sensitivity of teeth and premature occlusal contact on the mandibular left molars (Figure 1). Case 2 - a 23-year-old male patient complaining of face asymmetry and an unerupted left mandibular second molar (tooth 37 by ISO system), with no other symptoms (Figure 2). Clinical examination revealed noticeable swelling in the posterior mandibular region. On screening panoramic imaging a large cystic lesion was detected. The patients agreed to take part in the study and signed the informed consent information sheet.

Marsupialization of the cystic lesion under local anaesthesia (Ubistesin 1:200 000, 3M ESPE Dental AG, Seefeld, Germany) was performed by creating a cystic opening of about 1 cm in diameter and removing the internal cystic contents. A concurrent incisional biopsy was obtained from the cyst lining and the cystic capsule was sutured to the oral mucosa. A Terramycine (Terramycine + polymixine B, Pfizer SA/NV, Brussel, Belgium) gauze pack was used for 5 days to tamponade the defect (Figure 3). A new one was loosely packed into the cavity and was left for for one week until the borders of the cyst were well healed. Then the gauze was removed and the wax impression for the obturator was made. Proper oral hygiene instructions were given to the patients.

The specimens were fixed with 10% formalin and embedded in paraffin. The sections were stained with hematoxylin-eosin. Microscopic examination of both cases revealed fragments of fibrous connective tissue with a chronic inflammatory cell infiltration and some islands of odontogenic epithelial rests characteristic to dentigerous cysts.

The obturator was carved in situ using warm soft pink wax plates (Modern Materials, Shur Wax X-Hard, Heraeus Kulzer, USA). Thus creating the desired shape with a metal ligature in it for easy placement into the prepared cystic opening. A duplicate copy of the wax template was made from methyl methacrylate in the dental laboratory and then the obturator was adapted to the cystic opening (Figure 4). The patients were taught how to care for the obturator and the cavity was rinsed daily with plain tap water. Rinsing was done with a disposable syringe of 10 CC and a blunt needle. The obturator was in situ all the time and never dislocated because of its good retentive form.

In both cases, decompression of the cysts lasted 9 months till anatomical structures were identified before second stage surgery. Use of the obturator resulted in a number of advantages compared to a conventional approach: lower risk of spontaneous fracture of the mandible, anatomical structure preservation, mandibular canal identification and preservation of tooth vitality. In case 1, the patient reported a slight discomfort from the obturator, more specifically during oral hygiene procedures and cleaning of the obturator. In case 2, no complaints were noted during the decompression period.

The first CBCT was taken (case 1 – 1624 DAP, case 2 – 2115 DAP) and the mandibular canal was well identifiable (Figure 5 and 6). Enucleation of the cysts was performed under intravenous sedation using a midazolam/fentanyl protocol and local anaesthesia. The lesions were examined for histopathology. Histopathological examination confirmed the clinical diagnosis of the dentigerous cysts for both patients. The findings observed by the histopathologist were fragments of fibrous connective tissue with a chronic inflammatory cell infiltration and some islands of odontogenic epithelial rests, non-keratinizing stratified squamous epithelium and reduced enamel epithelium (Figure 7).

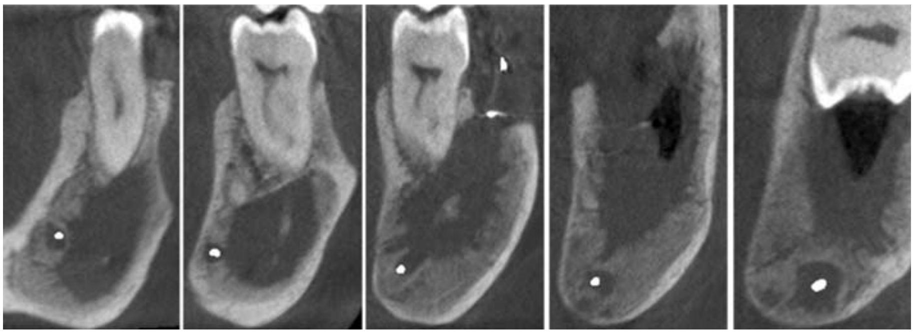

Fig. 5. CBCT image shows inferior alveolar canal follow-up in a 33-years old female patient after lesion decompression

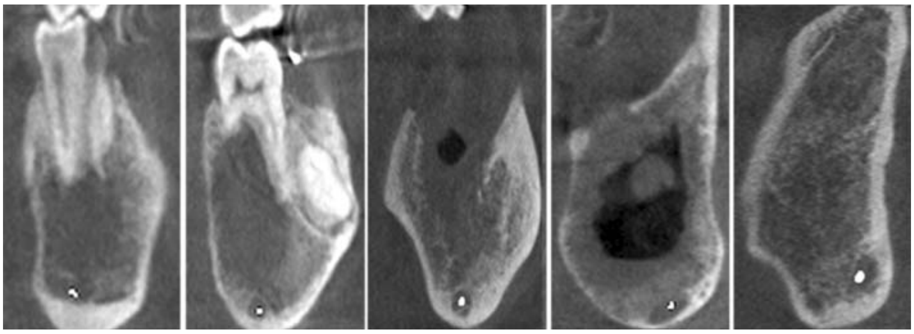

Fig. 6. CBCT image shows inferior alveolar canal follow-up in a 23-years old male patient after lesion decompression

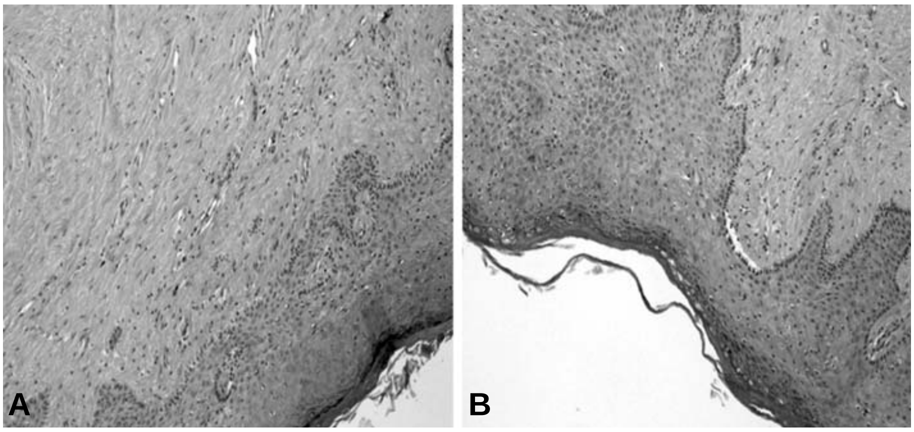

Fig. 7. Pathology report of dentigerous cysts. Original magnification 100×. Staining method: hematoxylin and eosin. A – photomicrography of a 33-years old female patient.

Sensibility, percussion and palpation tests were performed routinely during recalls. Involved teeth vitality was regularly assessed by laser doppler flowmetry with a Moor blood flow monitor (type Moor VMS - LDF 2 Laser Doppler Perfusion; wavelength, 785 (10) nm). Prior to the recording, resin splints were prepared for examined sites from premolars to molars from the polyvinyl siloxane (Aquasil, Dentsply, York, PA, USA) with the holes for the probe tips 3 mm above gingiva line in the middle of mesiodistal distance. All teeth were isolated with opaque rubber dam (ROEKO Dental Dam Latex, Coltene) before measurement. Involved teeth response were compared with the teeth response on the contralateral side of the mandible and antagonists for the female patient. However, only antagonists were measured for the male patient, as he had teeth with large restorations on the right side of the mandible and endodontics afterwards. Involved teeth response to the sensibility and palpation tests remained unchanged after decompression for both patients as compared to the results from the first visit. In case 1, teeth 36 and 37 remained more sensitive after decompression.

Cyst enucleation did not alter the response of the involved teeth to sensibility and palpation testing for both patients. In case 1, teeth 36 and 37 continued to exhibit an increased sensibility during testing, although response of both teeth became less

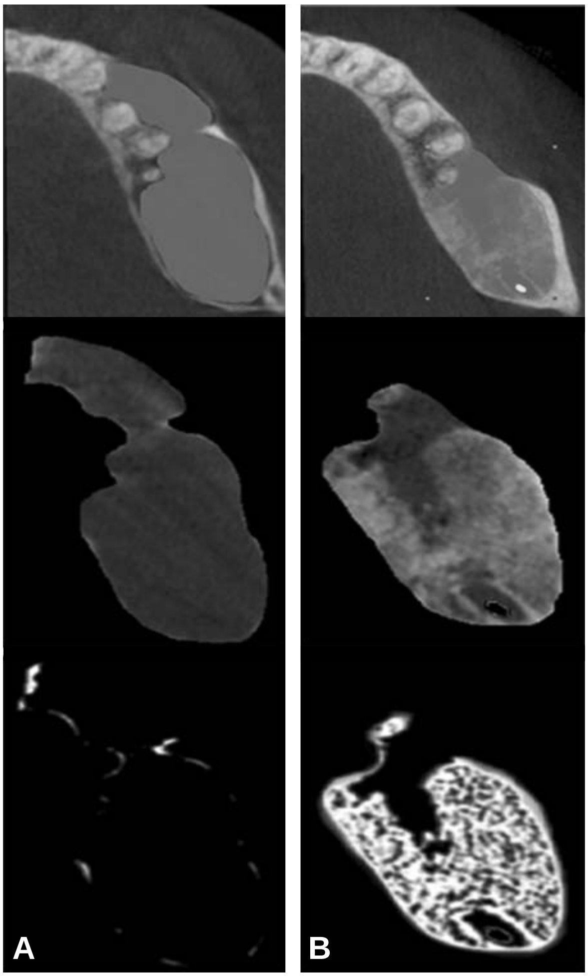

Fig. 8. Region of lesion in the axial view in a 33-year-old female patient. A – preoperative. B – 1 year postoperatively. First row: VOI selection. Second row: resulting VOI. Third row: binary image after thresholding.

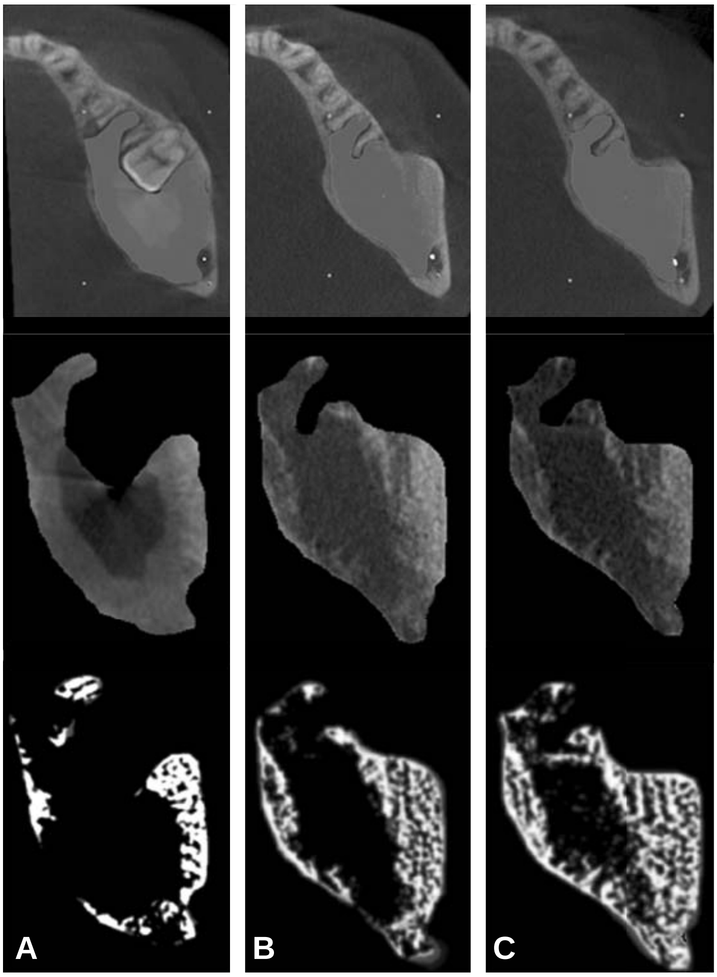

Fig. 9. Region of lesion in the axial view in a 23-year-old male patient. a. Preoperative. b. 1 year postoperatively. c. 4 years postoperatively. First row: VOI selection. Second row: resulting VOI. Third row: binary image after thresholding.

symptomatic after the second surgery. In general, teeth response to vitality testing changed minimally during the one year period of time, but an improved response was noted for the involved teeth, assuming pulp recovery (Table 1, Table 2).

Follow-up of lesion regression was accomplished using automatic radiographic analysis of formation of mineralized bone (ERB study approval ethical file number S57587). Imaging of the patients including the mandibles was performed, where a medium-field-of-view CBCT with 150 to 200 µm voxel size, was selected to allow depicting the area where the cysts were located. This was typically performed with the ProMax® Max (Planmeca, Helsinki, Finland). All data sets derived from CBCT were formatted as DICOM files.

To precisely compare data sets acquired at different time intervals, the exact same morphological structures should be compared to ensure that quantification are precisely from the same anatomical structures up to voxel accuracy. This was achieved by a fully automated and observer independent image registration based on mutual information (Maes *et al.*, 1996). The validity of the automatic registration process was verified by checking different anatomical reference points. Hereafter, for each image, a Volume of Interest (VOI) comprising only the mandibular cyst within the boundaries of the cortical bone was manually selected to evaluate the formed mineralized trabecular bone. After VOI selection, images were segmented using adaptive threshold algorithm to allow objective 3D bone quantification with CTAn software (CTAnalyser®; SkyScan, Antwerp, Belgium) (Figure 8, Figure 9). The obtained binary images within the VOI were measured following the parameters reported by Van Dessel *et al.* (2013): total volume of interest (TV), bone volume (BV), bone volume fraction (BV/TV), bone surface (BS), bone surface density (BS/TV), trabecular thickness (Tb.Th), trabecular separation (Tb.Sp), trabecular number (Tb.N), total porosity percentage (Po(tot)) and connectivity density (Conn.dn). Dimension units were given in millimeters (Tables 3 and 4) (13).

Notwithstanding that a one-year follow-up panoramic image could not visualize a remnant bone defect nor cystic lesion recurrence, 3D-CBCT based semi-automated bone quantification could only demonstrate an increase of 46% and 15% of mineralized bone volume one year after surgery in cases 1 and 2 respectively. Besides the bone volume fraction, the other parameters for bone structure quantification can be observed in Tables 3 and 4. Visually, mineral bone formation typically occurred starting from the periphery of the original lesion towards the inner core.

DISCUSSION

Dentigerous cysts should be treated surgically to avoid possible complications such as teeth displacement, resorption of teeth, bone destruction, pathologic fractures, delayed tooth eruption, displacement and obliteration of the maxillary antrum and nasal cavities and paraesthesia of inferior alveolar nerve (3). Several treatment options for dentigerous cysts have to be considered. Usually, if the cysts are not extensive, total enucleation and curettage of the cyst with removal of the impacted tooth or teeth may be the first choice (1). However, treatment of large cysts in the maxillofacial region has not been standardized.

Many authors (14-16) have recommended more conservative treatment for large cysts such as marsupialization or marsupialization with delayed cyst enucleation, especially in children or adults with dentigerous cysts in maxilla. It is believed that cyst growth occurs by a combination of osmotic and resorption pressure, coupled with release of prostaglandins and growth factors (4). Our results coincided with several studies that suggest decompression of a cyst by decreasing the intracystic pressure offers several advantages such as stimulating new bone formation around the important anatomical structures, reducing the chance of a pathologic fracture or bony discontinuity (17, 18).

The disadvantages of marsupialization are the pathologic tissue left in situ, prolonged healing time, difficult oral hygiene care in the marsupialization area, especially when dentigerous cyst is in the posterior mandible (1, 4). In order to ensure proper diagnosis and reduce recurrence risks in the cases presented, a concurrent incisional biopsy was obtained. Moreover a second procedure to enucleate the residual tissue and the impacted teeth was performed to reduce the risk of recurrence.

Total healing time for both patients was 15-18 months, but none of them had complaints about previous visits or length of treatment. However, patients have to be motivated and able to come for regular recalls for effective medical treatment. It is confirmed that effective communication has a positive impact on important outcomes including patient satisfaction, adherence to recommended treatment (19). Specifically, doctors with positive attitudes towards psychosocial issues make more statements expressing concern and empathy (20).

One issue why marsupialization technique is used more often in the upper jaw is due to the fact that it is more easily to clean the cystic cavity daily through the stent and gravity dependent drainage (4). However, stents are not fundamental in the mandible because of food impaction. Our created obturator had a design that prevented it from falling into the bone cavity or coming loose. It can easily be taken out and replaced by the patient making daily rinsing of the cystic cavity and cleaning of the obturator possible. Locating the marsupialisation window distal of the last tooth in the retromolar region makes obturator modeling more easy and

Table 1. Tests of teeth vitality in a 33-year-old female patient

Tooth Number(ISO system)	24	34	44	25	35	45	26	36	46	27	37	47
Blood flow signals (P.U.) preoperatively	7.3 (3.5)	6.2 (3.4)	7.9 (3.3)	6.4 (2.5)	6.0 (2.4)	6.1 (2.2)	5.9 (3.3)	6.3 (3.8)	7.4 (3.0)	5.9 (3.5)	6.7 (2.6)	8.5 (3.1)
Blood flow signals (P.U.) 1 year postoperatively	7.5 (3.4)	6.4 (3.8)	8.0 (3.1)	6.6 (3.4)	6.5 (2.1)	6.3 (2.9)	5.8 (3.5)	6.4 (3.1)	7.5 (2.4)	6.0 (2.2)	8.1 (3.3)	8.8 (3.3)

Table 2. Tests of teeth vitality in a 23-year-old male patient

Tooth Number (ISO system)	34	35	25	36	26	27
Blood flow signals (P.U.) preoperatively	6.9 (2.8)	6.6 (3.2)	7.5 (2.1)	6.6 (2.7)	6.8 (2.2)	5.8 (3.9)
Blood flow signals (P.U.) 1 year postoperatively	7.1 (3.2)	6.8 (2.4)	7.6 (2.1)	6.7 (2.9)	7.3 (2.1)	5.4 (2.5)
Blood flow signals (P.U.) 4 years postoperatively	7.2 (3.7)	7.4 (2.9)	7.8 (2.8)	7.8 (2.6)	6.9 (3.6)	5.9 (2.2)

limits interference with masticatory muscle activity.

While creating the obturator one should also pay attention to its shape as it should have a slimmer neck and a wider top in order to stay stable and cover the cavity opening preventing food impaction. On one hand it has to be big enough to cover the defect and keep itself in the prepared position, but on the other hand it shouldn't be too large in order to avoid the soft tissue damage and occlusion interference.

Our results coincided with a recently published study that when marsupialization is done, conservative therapy rather than root canal therapy is recommended for the teeth with root tips exposed in the cystic lesions (21). In the present cases all involved teeth had a vital pulp after decompression and enucleation of the cysts, although CBCT scanning 4 years after surgery revealed partial pulp obliteration of 36 in case 2.

Recently published study has compared reduction in volume among different cystic diseases in the posterior mandibular body or ramus by 3-dimensional CT analysis (22). Our results coincided that measurement of volume using 3-dimensional CT can improve accuracy, repeatability as the reduction of the cysts is 3-dimensional. Moreover the present clinical case reports are the first to describe an automatic quantification of bone structure for jaw cyst follow-up. Besides the cysts volume, the mineralized bone content that formed through the time was calculated, using accurate bone segmentation threshold and bone morphometric parameters.

Although CBCT is an indispensable radiographic tool for the surgical plan and follow-up of large cysts, its main drawback is that bone density cannot be calculated through extrapolation of the grey values to Hounsfield Units (HU). The relatively large amount of noise in CBCT images may lead to large errors when using these grey values in a quantitative way for differential diagnosis and follow-up of bony lesions (23).

However, bone lesion evaluation should not be based on bone density alone. A variety of pathologies may cause alterations in both the bone structure and jaw mineral content. For example, by using the micro-CT (μCT), bisphosphonate treated osteonecrotic jaws were observed to show an increased total amount of bone, higher amount of trabeculae and larger trabecular thickness in the periradicular region, while the separation between the trabeculae was reduced (24).

A recent study validated an alternative technique to assess the trabecular bone quantity and structure using CBCT without using extrapolation to HU values (13). This novel approach showed to be promising for evaluation of bone lesions, which could facilitate the quantitative and objective detection of jaw cysts and its treatment follow–up over time (25).

Table 3. 33-year-old female patient’s bone morphometric parameters in mm

	Total volume of interest	**Bone volume**	**Bone volume fraction**	**Bone surface**	**Bone surface density**	**Trabecular thickness**	**Trabecular separation**	**Trabecular number**	**Total porosity**	**Connectivity density**
Preoperatively CBCT	15687.51	336.33	2.14	3676.18	0.23	0.45	11.74	0.048	97.86	0.20
1 year postoperatively	11586.97	5617.32	48.48	28796.89	2.49	0.59	2.68	0.83	51.52	1.30

Bone surface in mm^2 per mm^3; bone surface density in mm^2 per mm^3; bone volume in mm^3; bone volume fraction in %; connectivity density in %; total porosity percentage in %, trabecular number in 1 per mm; trabecular separation in mm; trabecular thickness in mm; total volume of interest in mm^3.

Table 4. 23-year-old male patient’s bone morphometric parameters in mm

	Total volume of interest	**Bone volume**	**Bone volume fraction**	**Bone surface**	**Bone surface density**	**Trabecular thickness**	**Trabecular separation**	**Trabecular number**	**Total porosity**	**Connectivity density**
Preoperatively CBCT	31593.5	6191.43	19.60	26187.89	0.83	0.77	8.31	0.30	76.57	0.25
1 year postoperatively	20849.01	7270.90	34.87	42929.25	2.06	0.58	2.50	0.60	65.13	1.08
4 years postoperatively	20090.30	8604.56	42.83	48759.55	2.43	0.60	1.19	0.72	57.17	1.23

Bone surface in mm^2 per mm^3; bone surface density in mm^2 per mm^3; bone volume in mm^3; bone volume fraction in %; connectivity density in %; total porosity percentage in %, trabecular number in 1 per mm; trabecular separation in mm; trabecular thickness in mm; total volume of interest in mm^3.

Present results showed that parameters such as bone volume, bone surface, bone surface density, trabecular number and connectivity density increased, while total porosity and trabecular separation decreased. These results confirmed the visual observation. On the other hand, trabecular thickness showed different results between case 1 and 2. One possible explanation for this would be the lower mineral content inside the lesion in the preoperative CBCT from case 1 and consequently lower trabecular number and thickness. In case 2, the preoperative CBCT shows remaining bone that was not totally resorbed by the lesion.

CONCLUSIONS

The presently reported cyst marsupialization was useful as a preliminary treatment for subsequent enucleation. 3-D CBCT based objective quantification of the bone volume and healing can provide new insights in lesion healing in general and more particular in the outcome of specific diagnostic and therapeutic challenges.

CONFLICT OF INTEREST

The authors state no conflict of interest.

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