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Combined therapy of marsupialization and enucleation for maxillary dentigerous cyst management: Serial case reports

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Abstract--Dentigerous cyst is a one of the most common cysts in oral and maxillofacial field. The goal of dentigerous cyst treatment is to eliminate pathological abnormalities and preserve the tooth with minimal surgical intervention. One of the treatment is marsupialization which will reduce the pressure on the cyst which will reduce the size of the bone defect. The case study aimed to present the advantages of enucleation following marsupialization on large maxillary dentigerous cysts in terms of preservation of vital structures around the cyst cavity. This study is a serial case reports of an 18-year-old female and 15-year-old male that had their dentigerous cyst removed by using combined therapy of marsupialization and enucleation. Dentigerous cyst was treated with marsupialization followed by cyst enucleation. The combination of marsupialization and enucleation therapy can be used in the treatment of extensive maxillary dentigerous cysts because it can provide advantages in preserving the teeth and important surrounding structures and provide a good prognosis.

Keywords---Dentigerous cyst, health outcomes, marsupialization, enucleation.

Introduction

Dentigerous cyst, one of the most common cysts in oral and maxillofacial field, is a pathological cavity that is confined by a connective tissue sac bordered by stratified squamous epithelium, formed around an unerupted tooth crown, contains fluid, and occurs because its growth process originates from the dental follicle of an unerupted or growing tooth (after calcification process) (Aristizabal Arboleda et al., 2018). Dentigerous cysts are detected in children, adolescents, adults, and occasionally older people, ranging from 3-57 years old. A study in Brazil found 10 out of 17 cases of dentigerous cysts occurred in children under 15 years. They are more common in men and nearly 60% of these cysts occur in the second to third decades of life. Approximately 70% of the lesions occur in the mandible and 30% in the maxilla. Nearly 62% occurred in the molars, 12% in the premolars, 12% in the canines, and the remaining 14% occurred elsewhere in the jawbone (Adams et al., 2000).

The goal of dentigerous cyst treatment is to eliminate pathological abnormalities and preserve the tooth with minimal surgical intervention. The choice of treatment depends on the size and location of the cyst, age of the patient, tooth involved, stage of root development, position of the involved tooth in the jaw in relation to the adjacent tooth, and involvement of any vital structures (Bassetti et al., 2019). Marsupialization will reduce the pressure on the cyst which will reduce the size of the bone defect. The cyst can then be removed some time later with minimal surgical procedures, without causing a large defect (Rajae & Karima, 2021).

The aim of this case study is to inform clinicians about the advantages of enucleation following marsupialization on large maxillary dentigerous cysts, as compared to enucleation alone, in terms of preservation of vital structures around the cyst cavity

Case Description

Case 1

An 18-year-old female with the primary concern of swollen left cheek. She had surgery for her canine that was impacted five years ago. She received a ball strike to the upper left cheek a year ago. She ran into a fence in the same spot two weeks prior and the discomfort persisted for the next week. She took an analgesic; although the ache was gone, the swelling persisted. The patient had pain, especially during chewing. No systemic illness existed.

The patient's general condition and generalist status was good. Extraoral examination (Figure 1a) showed facial asymmetry on left maxillary region, palpable diffuse redness swelling with hard consistency from infraorbital to maxillary, and there was tenderness upon palpation. Intraoral examination

(Figure 1b) revealed $\pm 8 \times 3 \times 2$ cm palpable well-defined swelling on teeth 21, 63 (2°), 24 (2°), 25 (2°) regions with hard consistency and volatile area, slightly hyperemic mucosa, and there was pain upon palpation.

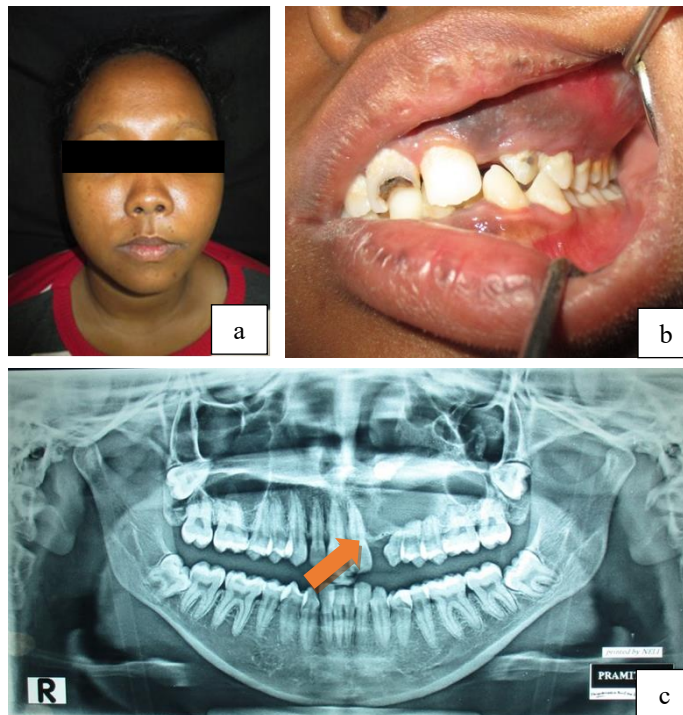


Figure 1. Preoperative (a) frontal view, (b) buccal vestibule view, (c) panoramic radiograph imaging

Radiographic examination revealed a well-defined unilocular radiolucency in the apical region of teeth 21 to 26, impacted tooth 23 that was pressed apically with its crown involved in the cystic mass, well-defined cortex of a sclerotic lesion marked with a thin radiopaque border.

A secondary infection was found on the initial assessment, so a puncture test was performed and 30 cc of clear brownish-yellow fluid was obtained (Figure 2a). The patient was then discharged and given antibiotics and analgesics. An incisional biopsy was planned when the inflammation subsided. Three months later, the patient came back with no improvements and still felt pain. A drainage incision was made on the most volatile area in teeth 24-25 regions and ± 5 cc of clear yellow fluid (cyst fluid) containing blood was obtained, followed by PZ and povidone iodine irrigation. As the lump looked smaller, a rubber drain was inserted then fixed with two 3.0 silk sutures (Figure 2b).



Figure 1. Preoperative (a) frontal view, (b) buccal vestibule view, (c) panoramic radiograph imaging

A week later, the patient underwent incisional biopsy along with marsupialization under local anesthesia. Incision was made on the mesial and distal part of the post marsupialization window. The window was widened and a cyst wall was found. An incision was then made on the wall, followed by PZ irrigation, kamicetin tampon gauze placement, and fixed with 4.0 silk sutures (Figure 2c). HPA result from the incisional biopsy showed a tissue section that partially covered with squamous epithelium and stroma consisted of fibrous connective tissue with lymphocyte, histiocyte, and plasma cells. There was no sign of malignancy and it was concluded as radicular cyst. After obturator fabrication was proceeded and kamicetin gauze was changed regularly, the obturator then placed on the patient.



Figure 3. 3 months post obturator placement: (a) Frontal view, (b) Buccal vestibule view, (c) Panoramic radiograph imaging

At the third months post obturator placement, patient experienced neither spontaneous pain, pain while inserting the obturator, nor fluid coming out from the window. Patient felt pain when the cheek is pressed but the swelling has decreased. Extraoral examination (Figure 3a) showed palpable minimal diffuse swelling on left cheek with hard consistency, no redness, and minimal tenderness. Intraoral examination (Figure 3b) revealed palpable diffuse swelling on teeth 21-22 regions with hard consistency and no redness. There was still window and minimal tenderness. Panoramic radiograph examination (Figure 3c) revealed a well-defined radiolucency on the apical of teeth 21 to 26 regions, impacted tooth 23, well-defined sclerotic and cortex border, and a thickened radiopacity around the border. This indicated an increase of bone calcification as a result of the decrease in cyst mass volume post marsupialization.

Not long after that, cyst enucleation was proceeded. Trapezoid incision was made from the distal tooth 21 until distal tooth 25. Rasparatorium was used to make the trapezoid flap and an epithelial cyst was obtained from the distal region of tooth 21 to tooth 25. Curretage was performed to separate the cyst epithelium from its cavity. The epithelium was then separated in a fragmented state and the bone was reduced using round bur along with PZ irrigation (Figure 4a). The impacted tooth 23 was lifted using bein and removed using clamps, followed by performing curretage and irrigation (Figure 4b). Spongostan was applied and the

flap was placed back, then sutured using 4.0 silk sutures. The tissue samples were taken and examined (Figure 4c).

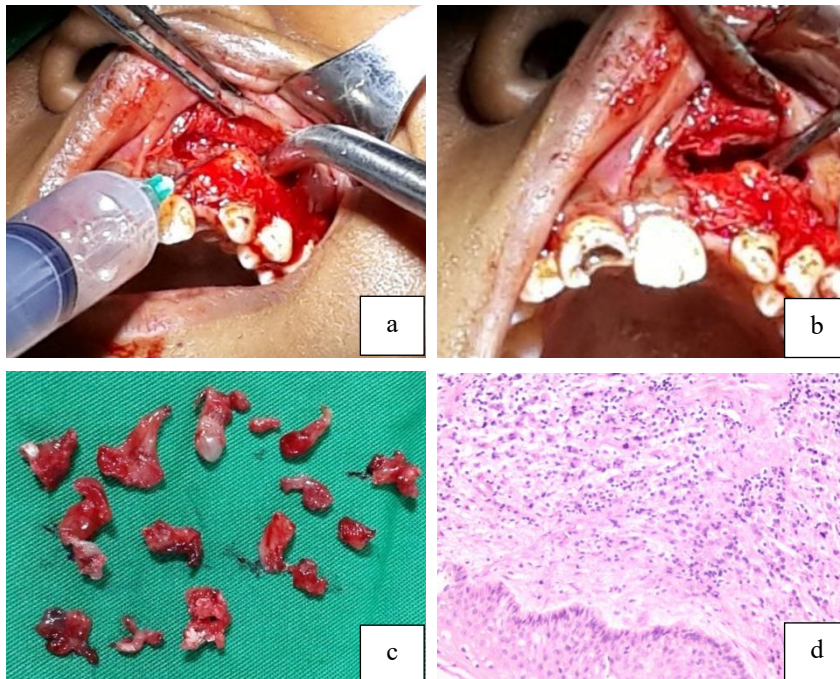


Figure 4. (a) and (b) Cyst enucleation process, (c) Cyst tissue samples, (d) HPA examination post enucleation

Post enucleation HPA examination result showed sections of tissue covered by stratified squamous epithelium with a few of lymphocytes and neutrophils, stroma of fibrous connective tissue with lymphocytes infiltration. There were no signs of malignancy and concluded as dentigerous cyst. At the 11th day post enucleation, the swelling had decreased, patient felt no pain and was able to open and close her mouth normally. Extraoral examination (Figure 5a) showed neither facial asymmetry, swelling, nor tenderness. Intraoral examination of teeth 22, 24, 25, and 26 regions (Figure 5b) revealed palpable post operation mass, good looking surgical wound, complete sutures with debris, and no redness, swelling, or tenderness. The sutures were then removed.

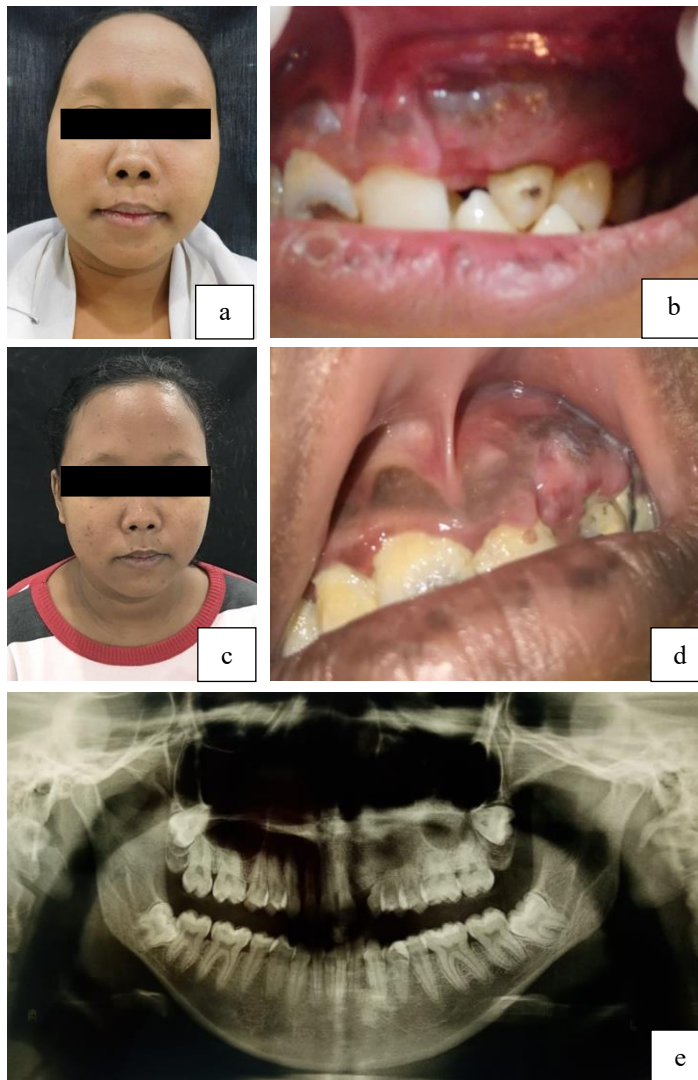


Figure 5. 11 days post enucleation: (a) Frontal view, (b) Buccal vestibule view
 6 months post enucleation: (c) Frontal view, (d) Buccal vestibule view, (e)
 Panoramic radiograph imaging

After 6-months follow up, patient had no complain at all and there was no new tumor growth. Extraoral examination (Figure 5c) showed neither facial asymmetry, hyperemia, palpable mass, nor tenderness. Intraoral examination (Figure 5d) revealed no mass on right maxillary, hyperemia, palpation, or tenderness. Panoramic radiograph examination (Figure 5e) showed radiolucent image that was getting smaller as well as a radiopacity increase on the area around the defect. This indicated that new bone formation had occurred in the area around the post-enucleation defect.

Case 2

A 15-year-old male with the primary concern of painful swollen left cheek. He started to notice a small swelling a year ago, but grew bigger slowly along with the pain. He once went to a dentist and was given antibiotic and analgesic, the pain was gone but the swelling was still there. There was neither drastic weight loss nor systemic disease.

The patient's general condition and generalist status was good. Extraoral examination (Figure 6a) showed facial asymmetry, palpable diffuse swelling in the right maxillary region with hard solid consistency, and lifted nasolabial sulcus. There was tenderness upon palpation and no redness on the skin. Intraoral examination (Figure 6b) revealed palpable diffuse mass in the right maxillary region with solid consistency, intact palpable palate, persistency of tooth 53, shallow right maxillary vestibule on teeth 12-14 regions, and neither pain upon palpation, sign of tooth 13 eruption, loose teeth, nor palatum mass.

Panoramic radiograph examination (Figure 6c) revealed a radiolucent unilocular lesion on the apical of tooth 23 extending to tooth 15 region as long as well-defined sclerotic and cortex border. There was also a radiopaque image and impacted tooth 13 that was pressed superiorly.



Figure 6. Preoperative (a) frontal view, (b) panoramic radiograph imaging, (c) buccal vestibule view, (d) Post obturator placement buccal vestibule view

The patient then underwent marsupialization along with incisional biopsy. Five days later, obturator fabrication and placement were performed (Figure 6d). HPA result from the incisional biopsy signified a cyst tissue imaging that was covered by squamous epithelium and stroma in fibrous connective tissue form with a few lymphocyte cells. There was no sign of malignancy and it was concluded as dentigerous cyst.

Panoramic imaging evaluation was carried out on the patient every 3 months. The first month image revealed no change on the size of radiolucent lesion at apical tooth 13. However, it gradually decreased at 3, 6, and 9 months post marsupialization (Figure 7a-d). A radiopaque image then began to appear on the posterior of tooth 13 as a response to the size reduction of the cyst.

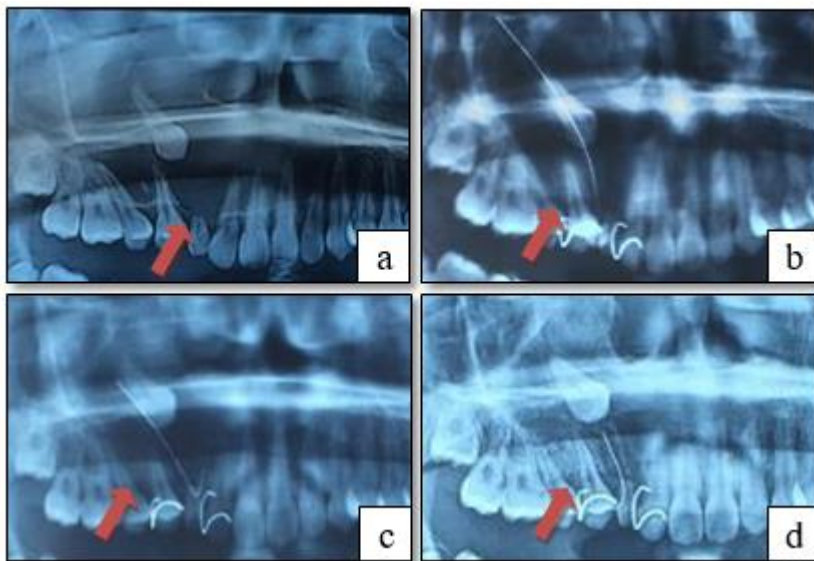


Figure 7. Panoramic imaging evaluation: (a) preoperative, (b) 3 months, (c) 6 months, (d) 9 months post marsupialization and obturator placement

The patient was examined at the 9th month post marsupialization and obturator placement. Extraoral examination (Figure 8a) showed facial asymmetry had disappeared and there was still redness swelling on the right maxillary but no tenderness upon palpation. Intraoral examination revealed post marsupialization window on tooth 53 region (Figure 8b), no palpable mass or shallow vestibule, palpable 0.5 cm window on teeth 23-15 regions with no signs of cyst, intact palpable palate, and neither redness nor pain upon palpation. The patient then agreed to proceed cyst enucleation and impacted tooth 13 odontectomy along with tooth 53 extraction under general anesthesia. Complete blood count results were within normal limits and chest X-ray examination also showed normal imaging in cor and pulmo



Figure 8. 9 months post marsupialization: (a) Frontal view, (b) Buccal vestibule view

Four weeks later, he finally underwent the procedures. A triangular flap incision was made from the distal of tooth 14, along the margin of tooth 14 to 21. The flap was then separated from the underlying bone with a raspatorium. Tooth 53 extraction was performed, followed by reducing the bone retention partially using a round bur and PZ irrigation along the window (Figure 9a). The impacted tooth 13 was lifted using a bein. The tooth was removed using clamps, then curettage and irrigation were performed (Figure 9b). Spongostan was applied and the flap was placed back, then sutured using 4.0 Vicryl sutures (Figure 9c). The tissue samples were taken and examined (Figure 9d, e). After seven days, the sutures were removed

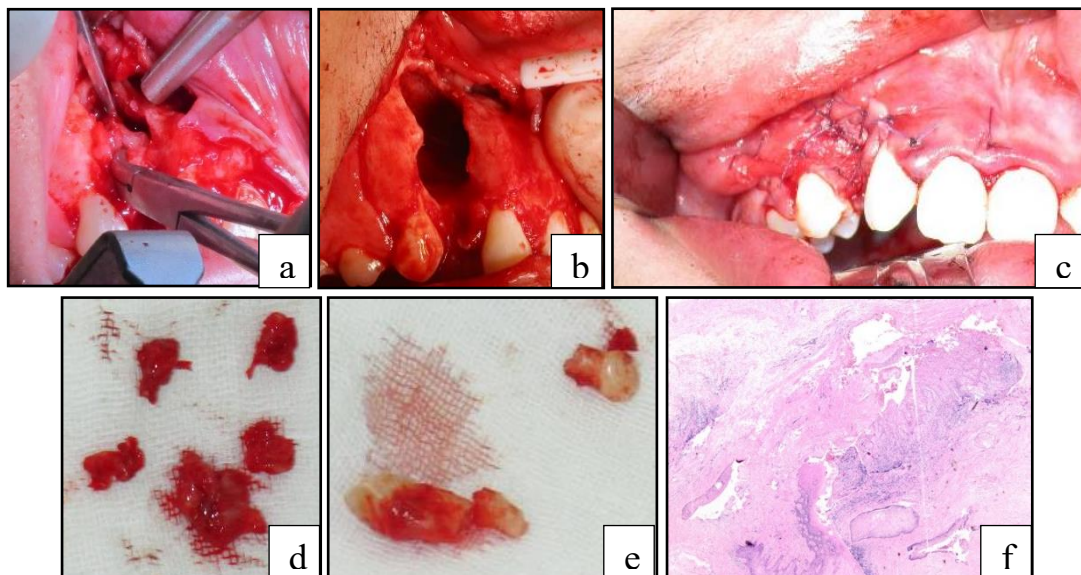


Figure 9. (a) Cyst enucleation process, (b) Post extraction, (c) Sutured flap post enucleation and extraction, (d) Cyst samples, (e) Teeth 13 and 53 fragments, (f) HPA examination post enucleation.

Post enucleation HPA examination result showed a piece of connective tissue covered by squamous epithelium and stroma with a few lymphocytes and plasma cells infiltration. There were no signs of malignancy and concluded as dentigerous cyst (Figure 9f). At the 7th months post operation examination, there were neither complaint on the surgical wound nor new tumor growth. Extraoral examination (Figure 10c) showed no facial asymmetry, hyperemia, or palpable mass. Intraoral examination (Figure 10d) revealed the window on apical tooth 14 was still not completely closed, palpable 2 mm window with 1 mm depth, and no edema, hyperemia, or mass. Panoramic radiograph examination (Figure 10b) revealed the radiolucent image was getting smaller, followed by an increase in radiopacity around the defect area. This indicated that there had been ossification around the post enucleation defect area.

After 19 months follow-up, patient had no complaints at all. Extraoral examination (Figure 10a) showed neither facial asymmetry, hyperemia, nor palpable mass. Intraoral examination (Figure 10b) revealed the window on apical tooth 14 had not close perfectly, palpable 2 mm window with 1 mm depth, and no edema, hyperemia, or palpable mass. Panoramic radiograph examination (Figure 10c) revealed there was still radiolucent image on the defect, along with a radiopacity increase in the area around the defect.

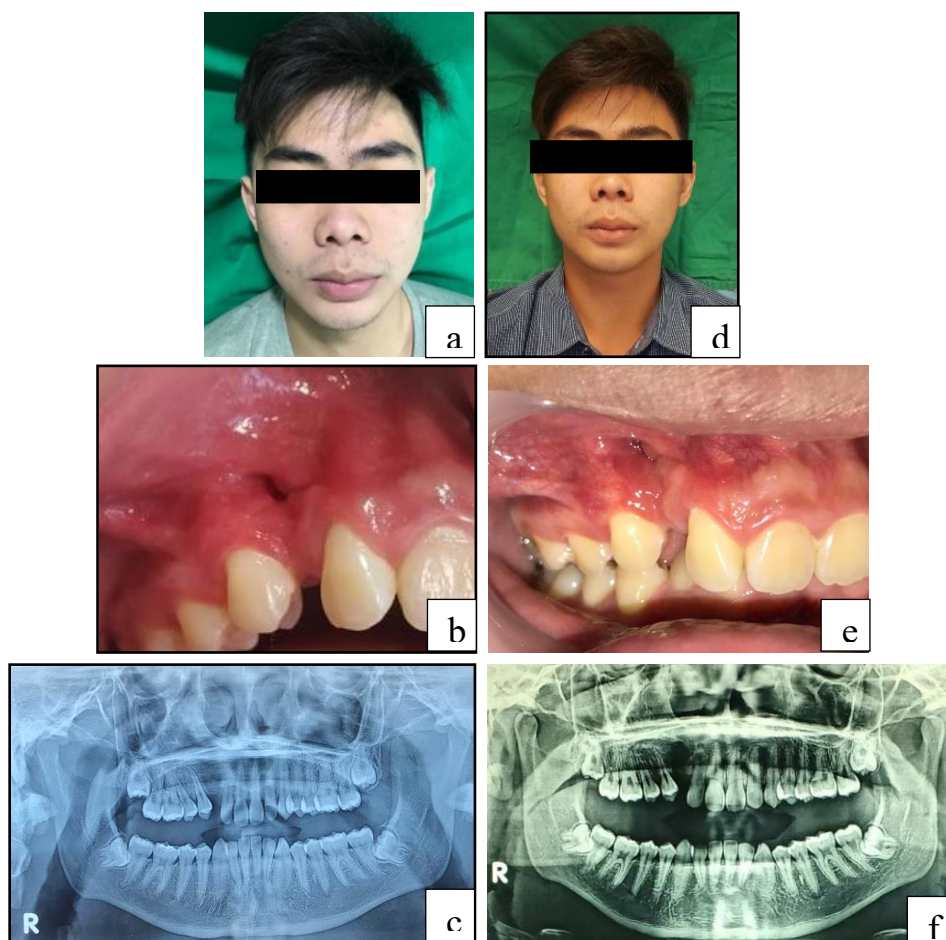


Figure 10. 7 months post enucleation: (a) Frontal view, (b) Buccal vestibule view, (c) Panoramic radiograph imaging. 19 months post enucleation: (d) Frontal view, (e) Buccal vestibule view, (f) Panoramic radiograph imaging

Discussion

This case report presented two cases of maxillary dentigerous cysts in two patients who came with complaints of swelling on the left and right upper cheeks that was getting bigger and accompanied by pain. Dentigerous cysts are usually asymptomatic unless they are very large (10-15cm), so secondary infection can cause swelling and pain. Secondary infection is common and can result in jaw expansion, so there is a possibility of pathological fracture.¹ In the first case, the patient's dentigerous cyst grew slowly at first without any symptoms, but eventually it became irritated, resulting in complaints and swelling of the maxilla (Rajae & Karima, 2021; Teixeira et al., 2011).

Cysts often develop on one tooth, but they can potentially involve several nearby teeth if the cyst is bigger. Cysts can range in size from those that grow slowly in

the pericoronal sac to those that cover part of the jawbone. Furthermore, it will cause tooth shift away from its normal position, especially in cysts that affect the teeth in the upper jaw which make it impossible to determine the origin of the cyst. In this case, the first and second patients had impacted teeth 13 and 23, respectively, which were involved in the cysts, which appeared to be displaced apically due to the pressure of the cyst mass (Aristizabal Arboleda et al., 2018; Carrera et al., 2013; Rajae & Karima, 2021).

In theory, cysts occur due to dilatation of the normal follicular space around the crown of an erupting tooth which caused by accumulation of tissue fluid or blood. Cyst proliferation occurs by a hyperosmolarity process that will cause damage to the middle part of cyst cells. Then, an osmotic gradient pressure will occur which will pump fluid outside the cyst into the cyst lumen (Aristizabal Arboleda et al., 2018; Sharma et al., 2011; Thompson, 2018). The dentigerous cyst has the potential to enlarge, causing damage to the spinal cord and expansion of the jaw. Dentigerous cysts tend to displace and resorb adjacent teeth. Teeth affected by cysts would usually move a certain distance (Spini et al., 2016). In both cases, there was no resorption of the adjacent teeth. Thus, there was no movement of the adjacent teeth and no extraction was needed, only shifting the involved 13 and or 23 impacted teeth apically.

Dentigerous cysts have unique radiographic appearance. It usually appears symmetrical, unilocular, well demarcated, and surrounds the crown of an unerupted (impacted) tooth. Except for infected cysts, the margins are not clearly defined. Cyst growth is slow and regular, giving dentigerous cysts a well-defined sclerotic margin, well-defined cortex, and characterized by a thin radiopaque line, especially if the cyst is relatively large in size or if the tooth has shifted from its position (Abd El-Fattah et al., 2021; Önay et al., 2019).

Panoramic examination in both cases had the same characteristics; unilocular radiolucency was clearly demarcated in the apical region of the surrounding teeth, impacted teeth 23 and/or 13 were pushed cranially by the mass, the crown of the impacted tooth appeared to be involved in the cystic mass, clear view of the margins of the sclerotic lesion with well-defined cortex marked by a thin radiopaque border, and the lesion appears radiolucent with well-defined borders. From the clinical and radiological examination on the two cases above, a dentigerous cyst caused by impacted teeth 23 and or 13 was established. This is also consistent with the results of postoperative histopathological examination which concluded that it was in accordance with dentigerous cysts (Aoki et al., 2018).

In the case of non-inflamed dentigerous cyst such as in the second case, the epithelial layer consists of 2 to 4 layers of nonkeratinized epithelial cells and the underlying connective tissue is flattened. The walls of the subepithelial connective tissue are not well organized and contain a large amount of glycosaminoglycan as the basic material. The subepithelial connective tissue wall is a capsule which is usually composed of a rather dense collagen network, accompanied by cells from foreign bodies. Chronic inflammatory cells are usually found. If ulceration is present, a mixture of chronic and acute inflammatory cells may be found. The connective tissue wall of the cyst is sometimes thickened and consists of fragile

connective tissue that contains a lot of scattered collagenous tissue so that the cyst is diagnosed as an odontogenic fibroma or odontogenic myxoma (Aoki et al., 2018; Danudiningrat, 2006; Spini et al., 2016; Thompson, 2018).

In the case of dentigerous cysts that experience inflammation or secondary infection, such as in the first case, the epithelial layer is hyperplastic, acanthosis occurs with the development of rete peg from squamous epithelium, the fibrous wall is denser with collagen so it is more supple, with various chronic inflammatory cell infiltrates. A keratinized epithelial surface can sometimes be seen and must be distinguished from odontogenic keratotic cysts (Aoki et al., 2018; Önay et al., 2019).

Treatment management for dentigerous cysts in small lesions could be through surgical removal. The treatment of choice for cysts is enucleation, which is done by scraping the cyst wall mucosa along with the extraction of the relevant tooth and removal of the protruding cyst wall to prevent the formation of residual cysts that result in recurrence (Teixeira et al., 2011). If the size of the lesion is large and includes extensive bone loss, marsupialization is performed by inserting a surgical drain. Marsupialization is the act of making a window in the cyst wall by taking the contents of the cyst and maintaining continuity between the cyst and the oral cavity, maxillary sinus or nasal cavity in surgery. (Aoki et al., 2018; Thompson, 2018) This procedure is recommended to remove the involvement of cysts from important structures of the oral cavity such as the teeth and inferior alveolar nerve, as well as to reduce the possibility of pathological fractures or bone damage that occurs during definitive therapy (Scariot et al., 2011).

In large cysts, decompression may be combined with enucleation. Once the cyst is significantly reduced in size, enucleation can become a simple surgical procedure. This combination of treatments is very safe compared to just doing enucleation, because it can reduce complications caused by damage to important structures found in the maxillofacial bone (Scariot et al., 2011).

In the first case, management of the dentigerous cyst was initiated with a drainage incision prior to marsupialization. This is done because in inflamed cysts, it will be difficult to obtain representative pathological tissue on HPA examination. After the inflammation subsided, marsupialization, incisional biopsy, and installation of an obturator were performed to reduce the size of the mass so that enucleation and odontectomy of the impacted tooth 23 would be easier to do. Meanwhile, in the second case, the management of the cyst was carried out by marsupialization first and accompanied by the use of an obturator. The patient was then followed up for routine control until the results of clinical and radiological evaluation showed that the dentigerous cyst was small enough for enucleation and odontectomy of the impacted tooth 23 to be performed. At the time of enucleation and odontectomy of tooth 23, after being evaluated, several teeth that were involved in the previous cyst, namely teeth 63, 24 and 25 were not extracted. This was because after marsupialization, the cysts became smaller and the teeth could be preserved. In addition, the change in cyst size after marsupialization in both cases was considered sufficient to not harm the important maxillary structures, so that immediate enucleation was considered

and followed by odontectomy of the causative tooth (Danudiningrat, 2006; Mhaske, 2009; Rajae & Karima, 2021; Sharma et al., 2011).

Marsupialization by removing intra-cystic pressure can keep the impacted tooth in the cyst cavity and help the eruption process (Teixeira et al., 2011). In the first case, marsupialization was performed so that the surrounding teeth involved in the cyst such as impacted teeth 23 and 63 24 25 which were involved in the expansion of the cyst could be preserved, hence there was no need for extraction. Similarly, in the second case, the panoramic radiograph showed expansion of the cyst starting from the tooth region 22 to 16. However, after marsupialization, the cyst was decompressed and reduced in size which only involved tooth 53 and impacted tooth 13 in the cyst cavity. Thus, at the time of cyst enucleation, teeth 53 and 13 were also extracted. This shows that the marsupialization procedure is beneficial because it can preserve the surrounding teeth involved in the expansion of the cyst so that there is no need for excessive tooth extraction during cyst enucleation (Rajae & Karima, 2021).

Spontaneous eruption of impacted teeth associated with cysts can be predicted and determined 3 months after marsupialization, although in some cases orthodontic treatment is required to assist the extrusion process (Serra e Silva et al., 2007). The 3-month period after marsupialization is a critical time to determine whether additional treatment is needed to stimulate tooth eruption if this does not occur after cyst shrinkage. After this period, it is necessary to decide between tooth extraction or orthodontics (Carrera et al., 2013). Factors that affect tooth eruption in adult patients or children under 10 years are the depth of the tooth position, inclination, incomplete root formation accompanied by an apical opening of the tooth, and the availability of space for growth (Aoki et al., 2018; Serra e Silva et al., 2007). However, in the first and second cases, teeth 13 and 23 were impacted after evaluation of the first 3 months of marsupialization in the first patient and 9 months post-marsupialization in the second patient. The position of the teeth and the inclination of these two teeth did not allow for spontaneous eruption even with the aid of orthodontic appliances. Thus, teeth 13 and 23 in both cases were considered for odontectomy during cyst enucleation.

The prognosis for dentigerous cysts is excellent and there is no chance of recurrence after complete enucleation. However, residual cysts may develop if the lesion is not completely enucleated (Rajae & Karima, 2021; Spini et al., 2016). This is consistent with the post-enucleation evaluation results in both cases. In the 6th month post-enucleation and odontectomy evaluation of tooth 23 in the first case, there were no complaint nor new cyst growth, either clinically or radiologically. The same condition also occurs in the second case. After evaluation until the 19th month after enucleation and odontectomy of tooth 13, the patient did not have any complaints nor new cyst growth, either clinically or radiologically.

The results of the evaluation of these two patients showed that the combined therapy of marsupialization followed by enucleation of an extensive maxillary dentigerous cyst can provide many advantages in preserving the teeth and surrounding vital structures, and has a better prognosis. This is in accordance with the research conducted by Scariot et al. (2011) who stated that

decompression followed by cyst enucleation is an alternative treatment for large jaw cysts, especially when the cyst is located in the maxillary sinus. This technique is very suitable for young patients because it is less harmful for important structures such as teeth and other structures around it (Scariot et al., 2011).

Conclusion

The combination of marsupialization and enucleation therapy can be used in the treatment of extensive dentigerous cysts in the maxilla because it can provide advantages in preserving the teeth and important surrounding structures and provide a good prognosis when compared to enucleation therapy alone. This case study is expected to be a preliminary study for similar follow-up studies with a larger number of samples and a longer observation time.

Acknowledgments

There are no acknowledgments to declare.

Declaration of Patient Consent

The authors certify that all appropriate patient consent forms have been obtained. In the form of informed consent, the patients have given their consent for their images and other clinical information to be reported in the journal. Patients understand that their names and initials will not be published and will remain hidden, but anonymity cannot be guaranteed.

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