Marsupialization for Treatment of Jaw Cysts: Indications and Limitations
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Abstract:
Background: Removal of jaw cysts that reach large sizes, result in facial deformity, injury to the adjacent neurovascular bundles or jaw fracture. Marsupialization means creating a window into the cyst for decompression and is an adjuvant treatment. Limitations of this technique are not explained completely.

Materials and Methods: Patients suffering from a variety of jaw cysts that were managed by marsupialization with or without subsequent enucleation were included in this study between 2007 and 2014. At the 3rd and 6th month, follow-up radiographs were ordered.

Results: There were 16 patients (20 cysts) that were included in this study. There were two cases (2/16) that decision was made to change the treatment planning. Half the cysts were histologically proven odontogenic keratocyst. Two-thirds of the patients, who needs to enucleation subsequent to the successful marsupialization, were managed under general anesthesia.

Conclusion: Limitations of this technique are: Inability to examine the whole cyst microscopically, questionable eruption of the permanent tooth involved with the cyst, dependency on the patient cooperation, and difficulty of irrigation fluid circulation in multilocular jaw cysts, and finally, if the created window has large dimensions then soft tissue closure after final cyst enucleation is the problem.

Key Words: Enucleation, jaw cyst, marsupialization

Introduction
Frequency of the cysts in mandible and maxilla is much more than the other skeleton.¹ The reason is the presence of odontogenic epithelium in jaw bones that can proliferate to the variety of the odontogenic cysts.²³ The presence of the teeth also is the other important factor, through the spread of infection from pulpal or periodontal origin to the jaws.² The presence of inflammation within the jaw bones is a trigger for beginning of the cysts in the alveolar process by itself.

Sometimes, jaw cysts reach large sizes and their removal results in facial deformity, injury to the adjacent neurovascular bundles, and permanent loss of nerve function.²⁶ Probability of mandibular fracture during removal of large cysts or in the early period after surgery is a real danger if remaining cortical bone is very thin.²⁸ We call these features: “enucleation with complication.”

Loss of permanent teeth when a large cyst occurs in the mixed dentition and involves the teeth buds is the rule. Marsupialization minimizes any disruption to future dental development. Finally, elderly patients with severely compromised health conditions and the presence of large cyst within the jaw fall in this category. All above-mentioned conditions are indications for marsupialization.

In most articles, benefits of this technique are noticed, and drawbacks are in the shadow. In this article, experience of the authors with the technique of marsupialization for the jaw cysts is presented. Limitations of this technique are highlighted.

Materials and Methods
Patients suffering from jaw cysts that were managed by marsupialization with or without subsequent enucleation were included in this study between 2007 and 2014. An irrigation port or two windows were placed into the cyst for twice daily irrigation. After doing this surgery, the patients were recalled every month. At the 3rd and 6th month after operation, follow-up radiographs were ordered, and the further decision was made about how to continue. If the considerable reduction in the size of the lesion was obtained, then subsequent probramed enucleation was considered, and the marsupialization was considered as successful.

Increase in the radiographic size of the cyst or static dimension, as well as signs of soft tissue invasion, was considered as “not responding” and subsequent enucleation was considered without proposed benefits of marsupialization.

Results
Table 1 shows the results. There were 16 patients that were included in this study. Mean age of the patients was 32.2 ± 26. 20% of the cysts were located in the maxilla.
31% of the patients were in mixed dentition. Sex distribution was equal. Two patients had more than one cyst (2 and 4 cysts), one of them with Gorlin-Goltz syndrome. 20 jaw cysts were managed by marsupialization while 15/20 needed to subsequent enucleation, two-thirds under general anesthesia. Marsupialization was done under local anesthesia in all cases.

From five cysts that were in the preadolescence, one was radicular cyst correlates with the remnant roots of the mandibular first molar. The other four cysts were involved with the tooth germ that half of them respond ideally to the marsupialization with the spontaneous eruption of the tooth buds into the mouth without the need for orthodontic treatment. The other two cases responded to this procedure by dramatic decrease in the size of the cyst but needed to further enucleation with the loss of permanent tooth involved with the cyst.

There were two cases (2/16) that decision was made to change the treatment planning. In a case, toward “enucleation with complication,” other case toward resection.

Half the cysts were histologically proven odontogenic keratocyst/tumor (OKC). Dentigerous cyst with (15%) and a radicular cyst with (10%) were in the second and third place. Glandular odontogenic cyst, calcifying epithelial odontogenic cyst, and mural ameloblastoma, each with one case, were in the minority. In two cases, the exact pathologic diagnosis was not possible because any specimen had been send to the laboratory. One patient developed recurrence of the cyst (OKC) in the same site 3 years after the surgery.

The most common method for marsupialization was acrylic devices and infusion set that was prepared by the surgeon, each with eight cases. Acrylic resin extension, penetrating into the cystic cavity and attached to the upper denture or space maintainer was the other tool. Oral airways, and cut hypodermic syringe each with one case, were the other used devices, for marsupialization (Figure 1).

### Discussion
Considering the marsupialization as a conservative surgical approach in the management of large jaw cysts has many beneficial effects.8 Marsupialization in jaw cysts means creating a window into the cyst for decompression. This window in the maxillary cysts can open into the nose, maxillary sinus, or oral cavity.9,10 For mandibular cysts, the intraoral window is the sole choice.

Removal of the cyst in the mandible and maxilla, when the cyst encroaches into the neurovascular bundles. Inferior alveolar
nerve in the mandible and infraorbital nerve in the maxilla will result in the lost sensibility of the lips after surgery.\textsuperscript{12} This condition aggravates if the cyst needs to adjunctive treatments such as curettage and peripheral ostectomy as well as the application of Carnoy’s solution or cryotherapy that is recommended in the cysts with high recurrence rates such as OKC and glandular odontogenic cyst.\textsuperscript{13-15} Successful treatment by marsupialization will reduce this possibility by depositing bone above the neurovascular bundle after cyst decompression (Figure 2).

Another advantage of marsupialization is the prevention of pathologic fracture in weak mandible.\textsuperscript{16}

General anesthesia in geriatric patients with severely compromised medical conditions has high risks. Marsupialization followed by secondary enucleation of the cyst under local anesthesia can reduce these risks although the probability of medical emergencies will increase during this procedure that the surgeon should be ready to confront it.\textsuperscript{17}

Two-thirds of the patients, who needs to enucleation subsequent to successful marsupialization, were managed under general anesthesia. This is because of the young age of the patients in some cases and the fact that despite the reduction in the size of the cyst in comparison with the beginning of the treatment, yet the size of the lesion is so large that needs to general anesthesia for complete cystectomy.

There was a case of huge mural ameloblastoma that had endangered the airway. After decompression, this condition improved (Figure 3).

Facial bulging is the other reason for using this technique. After regression of the facial bony expansion, the cyst can be removed without excessive loss of the bony contours of the maxilla.

Marsupialization had been recommended in the treatment of the high recurrent cysts (mainly OKC).\textsuperscript{18} The proposed theory is metaplasia of the epithelium in these cases (Figure 4).\textsuperscript{19,20} Study of Pogrel and Jordan on the OKCs treated by decompression showed a reduction in anti-apoptotic gene BCL-2 Marker that is found in high concentration in the epithelial lining of KCOTs while the loss of Bcl-2 from marsupialized cysts was observed.\textsuperscript{21} Another reason for the reduction in the size of OKC is by inhibiting interleukin-1alpha expression and epithelial cell proliferation.\textsuperscript{22} Mitotic activity of the epithelial cells also decreases after marsupialization.

Final hypothesis emphasis on the fact that decreases intracystic negative pressure following marsupialization may enhance bone formation around jaw cysts.\textsuperscript{23}
The fact that majority of the cysts in this series was OKC come from nature of the cyst that enlarges, without expansion. These cysts are often discovered when they reach large sizes. Fibrosis in the cyst capsula and thickened cyst wall are the other advantages of marsupialization that leads to enblock enucleation of the cyst.

Marsupialization in treatment of jaw cysts has some limitations that are listed below:

- Histopathologic examination of the whole cyst epithelium is not possible. Some cysts may undergo ameloblastomatous or carcinomatous (mucoepidermoid or squamous cell carcinoma) changes. That need to more radical surgeries. Obtaining and sending the biopsy specimen for histopathologic examination is necessary.
- Marsupialization of a cyst lesion to allow tooth eruption, necessarily will not lead to the resolution of the cyst and tooth eruption into the oral cavity. If the involved permanent tooth entrapped in the cyst lumen or ectopically displaced by the cyst, then expecting the tooth to erupt after marsupialization is not realistic. Patient age, as well as impacted premolar angulation to the adjacent teeth axes, the cusp depth, space/tooth size, eruption period and cyst size, as measured on initial images are predictors in the success of eruption.
- Patient/parents cooperation has paramount importance in this procedure because of prolonged treatment of marsupialization. The most important factor in this technique is the patient understanding of the treatment planning and spending time for effective irrigation. This technique is not recommended in mentally retarded patients with multiple jaw cysts because this group has not high collaboration in daily self-irrigation even with the help.
- Multifocal nature of OKC should be considered. A separate window for each cyst is mandatory.
- Large radiographically multilocular mandibular cyst extending to the ramus may not respond well to marsupialization.

Posterior part of such cysts is far from access by irrigation solution and creating a window, posterior to the mandibular anterior border of the ramus is not possible.
- Decompression followed by enucleation with primary closure should be considered when the surgeon decides to choose the best device for marsupialization. Creating a large window is not recommended because of inability to primary close the window after programed enucleation of the cyst. Dehiscence of the wound if the mucosa is sutured under tension or the need for soft tissue flaps (if the window is very large), are limitations of these devices (Figure 5). Creating marsupialization window in the palate is not recommended because primary closure after cyst enucleation is impossible.
- Treatment of mural ameloblastoma and calcifying epithelial odontogenic cysts by marsupialization followed by enucleation is not accepted universally and is very dependent on the case.
- Marsupialization is done under local anesthesia, so very young children are not good candidates for this procedure.
- Breakage of the acrylic device and detachment of infusion set (inward/outward) is possible that needs to immediate replacement. Adjustment and reduction of acrylic extension that has penetrated into the cystic lumen is mandatory in programed intervals.

Conclusion
Marsupialization is a good adjuvant technique for the management of large cystic lesions involving the jaws. Attention to the limitations of this technique is mandatory. Inability to examine the whole cyst microscopically, the questionable eruption of the permanent tooth involved with the cyst, dependency on the patient cooperation, difficulty of irrigation fluid circulation in multilocular jaw cysts and finally
problem of soft tissue closure after final cyst enucleation (if the created window has large dimensions), all are in the list.

References