Long-term Outcome of Radiofrequency Ablation For Intraoral Microcystic Lymphatic Malformation

Sang W. Kim, MD; Katie Kauvanough, MD; Darren B. Orbach, MD, PhD; Ahmad I. Alomari, MD, MSc; John B. Mulliken, MD; Reza Rahbar, DMD, MD

Objective: To determine long-term outcome of radiofrequency (RF) ablation of microcystic lymphatic malformation (LM) of the oral cavity for control of recurrent infection and bleeding.

Design: Institutional review board–approved retrospective study.

Setting: Tertiary pediatric medical center.

Patients: Twenty-six patients with intraoral microcystic LM were treated with RF ablation from August 2002 through August 2010.

Intervention: Radiofrequency ablation of intraoral LM.

Main Outcome Measures: Postoperative stay, diet, pain; control of bleeding and/or infection; recurrence; and indication for retreatment.

Results: The most common complaints necessitating initial RF ablation were recurrent infection (n=10 [37%]) and bleeding (n=9 [33%]). The most common problems requiring further ablation were bleeding (n=11 [41%]) and cosmetic deformity not affecting function (n=8 [31%]). Fourteen patients (55%) were discharged home on postoperative day (POD) 3; the remaining 11 (45%) were discharged home on POD 4. Thirteen patients (52%) resumed oral diet immediately on the day of the procedure. Ten patients (38%) began eating on POD 1, and virtually every patient was on full oral intake at discharge. Fourteen patients (55%) required only acetaminophen for pain control, 11 (41%) required acetaminophen with codeine, and 1 (4%) required oxycodone. The mean follow-up time was 47 months after treatment. At the most recent clinic evaluation, 13 patients (50%) were symptom free, 8 (31%) were stable and improved without need for future treatment, and 5 (19%) required further treatment. One-half of patients in the study group underwent more than 1 RF procedure for recurrence. The number of RF ablations in this series were 1 procedure (n=13), 2 procedures (n=7), 3 procedures (n=2), 4 procedures (n=2), and 6 or 7 procedures (n=2).

Conclusions: Radiofrequency ablation is an effective treatment for localized, superficial microcystic LM in the oral cavity. Pediatric patients tolerate the treatment with rapid postoperative recovery and minimal complications. The majority of patients required a short hospital stay for observation of the airway. Virtually every patient resumed oral diet by the time of discharge. Radiofrequency ablation is the treatment of choice at Children’s Hospital Boston (CHB) for patients who present with symptomatic, superficial, and localized intraoral microcystic LM. For lesions involving deeper structures, multimodal treatments including surgical and sclerotherapy may be necessary.

Radiofrequency ablation treatment was performed by the senior otolaryngologist with the patient under general anesthesia. Owing to its mechanism of low-temperature tissue destruction, the thermal injuries to surrounding tissues are limited, and therefore patients may have a better course of recovery. Two modes of RF ablation have been described. The high-frequency mode is used for destruction of deep tissue, resulting in reduction in size from secondary fibrosis without affecting the mucosal surface. In low-frequency mode, energy is transmitted via conduction medium in small volumes for accuracy and for the removal of a thin superficial layer, with minimal thermal injury to nearby tissue.

Several articles have shown promising outcomes in treating LM in the oral cavity using the RF ablation technique, including one from our group. We report herein our long-term outcome and experience using RF ablation for microcystic LM in the oral cavity. For selective patients with lesions that are localized and superficial, we have observed excellent results with RF ablation in terms of postoperative recovery, complication rate, and clinical outcome.

### METHODS

The study group comprised of 26 children with intraoral LM who underwent RF ablation treatment at the Children’s Hospital Boston, Boston, Massachusetts, from August 2002 through August 2010. Medical records were reviewed for the location of lesions, previous treatments, and presenting signs and symptoms. Electronic medical records and inpatient medication records were accessed to document duration of hospitalization, resumption of oral intake, and pain regimen on the day of discharge; 21 patients had complete electronic medical records available. For outcome data, 4 of 26 patients were excluded owing to a follow-up duration of less than 4 months. Of the remaining 22 patients, a total of 50 RF ablation treatments were completed. Disease status was categorized based on examination by the senior otolaryngologist (R.R.) on follow-up clinic visits. On the basis of this evaluation, patients were selected for RF ablation procedure according to the extent of the lesion, specifically those that are limited to superficial and localized lesions.

Radiofrequency ablation treatment was performed by the senior otolaryngologist with the patient under general anesthesia with oral or nasal intubation. Intraoperative antibiotics and corticosteroid (dexamethasone, 0.5 mg/kg) were administered. Microcystic LM was removed in layers from superficial to deep in broad stroke with continuous saline irrigation using the coblator (Evac 70 Plasma Wand; ArthroCare Corp).

All patients were extubated in the operating room and encouraged to take liquids as tolerated. Patients were discharged home with the same pain regimen used during hospitalization and given 7 to 10 days of oral antibiotics.

### RESULTS

A total of 26 patients with localized, superficial microcystic lymphatic malformation in the oral cavity underwent RF ablation treatment at our institution (Table 1). There was an equal sex distribution in the study group. Some of these patients required more than 1 RF ablation treatment.

The most common location of the LM was the tongue (n = 19 [53%]). Thirteen patients had involvement of multiple anatomic sites; in Table 1 we report only the anatomic sites that met the selective criteria for RF ablation. These were localized, superficial microcystic LM involving mucosal surface of oral cavity based on clinical assessment. For LM lesions that were more extensive than those selected for RF ablation, patients in our series have undergone 31 surgical resections, 11 sclerotherapy procedures, and 6 laser treatments. The youngest patient in our series at the time of RF ablation was 29 months old and the oldest was 18 years old; the mean age was 10.4 years. Among the 26 patients, a total of 54 RF ablation procedures were performed during the 8-year period.

Presenting signs and symptoms that prompted RF ablation are listed in the Figure. Indications were divided into presenting problems and residual problems requiring further treatment. The most common presenting problems were recurrent bleeding and infection unresponsive to several courses of antibiotics, affecting daily life or school activities.

Thirteen patients underwent a single RF ablation treatment. Seven patients underwent 2 RF ablative treatments: 3 reported satisfactory symptom control at the follow-up visit, 3 reported intermittent bleeding, and 1 had persistent episodes of recurrent infection with swelling and bleeding. Two patients underwent 3 RF ablation treatments: 1 reported satisfactory control at the follow-up visit and the other had intermittent problems. Two patients had 4 RF ablation treatments and both reported satisfactory control. One patient underwent 6 RF ablation treatments, and another patient underwent 7 RF ablation treatments. They reported resolution of symptoms or some intermittent problems, respectively.

One patient was transferred to the intensive care unit from floor care status because of marked tongue edema on examination on postoperative day (POD) 1. The patient was treated with conservative medical management (intravenous antibiotic and corticosteroid), with resolution of swelling, and was able to be transferred to floor care within 48 hours. On POD 5, the patient again developed tongue edema, prompting a transfer to the intensive care unit for airway monitoring. Swelling resolved without affecting the airway, and the patient was discharged home on POD 10. In retrospect, this patient had a lymphatic-venous malformation that presented with intralesional bleeding postoperatively. The majority of patients were tolerating oral diet by POD 1, as given in Table 2. Postoperative oral diet regimen consisted of advancing diet from clear sips to soft con-

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Table 1. Pediatric Patients With Lymphatic Malformation of Oral Cavity

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>No. of Patients (N=26)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Total coblation procedure</td>
<td>54</td>
</tr>
<tr>
<td>Sex</td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>13</td>
</tr>
<tr>
<td>Female</td>
<td>13</td>
</tr>
<tr>
<td>Location of disease treated with coblation</td>
<td></td>
</tr>
<tr>
<td>Oral tongue</td>
<td>19</td>
</tr>
<tr>
<td>Floor of mouth</td>
<td>6</td>
</tr>
<tr>
<td>Lip</td>
<td>4</td>
</tr>
<tr>
<td>Base of tongue</td>
<td>3</td>
</tr>
<tr>
<td>Buccal</td>
<td>2</td>
</tr>
<tr>
<td>Age at coblation treatment, mean (range)</td>
<td>10.4 y (29 mo to 18 y)</td>
</tr>
</tbody>
</table>
consistency. All patients demonstrated at least 400 mL of total oral intake prior to discharge. By the time of discharge, pain was not a significant source of complaint.

Symptom-free state is defined as not having any episode of bleeding and/or infection. This is based on parents’ recount during the subsequent follow-up clinic visit with the senior otolaryngologist. All patients are followed up within 2 weeks postoperatively, then every 3 months or sooner if necessary. The clinical outcome data on Table 3 are based on assessment at the most recent clinic follow-up visit, at least 6 months after the procedure. Status of symptom control was categorized into 3 tiers: no complaints, minor symptoms, and major symptoms and/or revision procedure planned. The majority of patients were in a satisfactory symptom-free state at their most recent clinic visit (Figure). Eight patients (31%) continued to have intermittent symptoms such as bleeding and/or infection, which were effectively managed with oral antibiotic and had no significant impact on the quality of life. For the 5 of patients (19%) categorized as having major symptoms, recurrent episodes of infection, swelling, and/or bleeding were affecting quality of life, and revision RF ablation was planned.

Our preliminary report on RF ablation treatment for intraoral microcystic LM was promising.3 In this long-term follow-up series, most patients recovered from the procedure without major pain and promptly resumed oral diet, with few postoperative problems.

Several treatment modalities have been described for LM including resection, sclerotherapy, cryotherapy, electrocautery, RF ablation, corticosteroid administration, and embolization. Recurrence rates following attempted resection range from 50% to 100%.4-7 Numerous sclerosing agents have been used in the treatment of macrocystic LM including bleomycin sulfate, alcoholic solution of zein (Ethibloc; Ethicon), tetracycline, dextrose, interferon alfa-2a, fibrin sealant, and OK-432 (Picibanil; Chugai Pharmaceutical Co Ltd).8,9 OK-432 is thought to leave minimal perilesional fibrosis, making subsequent resection less hazardous.9 OK-432 is more effective for macrocystic than microcystic lesions, with 86% vs 66% reported efficacy rates, respectively.8 Alternatives for the treatment of microcystic lesions include: carbon dioxide laser, potassium titanyl phosphate laser, and RF ablation therapy in high- and low-power settings.3,8-12 Laser treatment has the advantages of less postoperative edema, less surrounding tissue trauma, and less blood loss compared with resection. Nevertheless, recurrence rate following laser treatment has been reported to range from 50% to 66%.3 A few reports have described RF ablation as an option for management of microcystic LM in the oral cavity.3,11,12 Roy et al11 described 3 children who underwent RF ablation of tongue LM without complications; all 3 showed no evidence of recurrence at the 1-year follow-up.
up visit. Grimmer et al described 11 children who had RF ablation; 62% reported in a survey that RF ablation was more effective at controlling symptoms than previous procedures (surgical excision, laser, or sclerotherapy).

Selection of the patient is critical in considering RF ablation treatment for LM. While there is no standardized medical regimen in our practice, patients in the series are typically treated with systemic antibiotics for acute infection and optimization of oral hygiene with mouth rinse and frequent dental care prior to and after the procedure. Recurrent symptoms, including acute bleeding, infection, enlargement affecting function, or cosmesis requiring frequent antibiotic therapy, were the indication for RF ablation treatment for superficial mucosal intraoral LM lesions.

In those patients who required repeated RF ablation treatment, it was notable that the indication was different than that for their initial treatment. Specifically, cosmetic deformity became the main indication for secondary treatment. Given how well patients tolerated the RF ablation during their initial treatment, patients and family developed higher expectations for overall symptom control and opted for subsequent revision RF ablations.

While the aim of this study was to describe our experience in treating superficial microcystic LM lesions involving all intraoral subunits, it is notable that the majority (53%) of the intraoral lesions in our series involved the oral tongue. Management of the microcystic LM involving the oral tongue is particularly challenging because of its propensity for recurrent infection, hemorrhage, airway compromise, feeding difficulties, and its effect on speech. Microcystic LM of the oral tongue has been classified based on the extent of lesion to plan for treatment and predict outcome.13,14 Recently Wiegand et al developed a classification system for LM of the oral tongue into 4 stages: (1) superficial microcystic LM; (2) lesion involving tongue muscle; (3) lesion involving tongue and the floor of mouth; and (4) extensive lesion involving tongue, floor of mouth, and other cervical structures. In their series, carbon dioxide laser ablation has demonstrated an excellent curative treatment for stage 1 and some stage 2 lesions. Detailed postoperative course (ie, timing of resuming oral diet, pain regimens) following carbon dioxide laser ablation was not available. For more advanced lesions, combined and/or staged approaches were necessary.13 Patients in our series who have been selected for RF ablation for their oral tongue lesions were mainly stage 1 lesions limited to the superficial mucosa based on clinical evaluation by the senior author. For patients with more extensive microcystic LM of the oral tongue, RF ablation is not the ideal primary treatment modality of choice. The characteristic feature of the RF ablation—low-temperature tissue destruction with limited injury to surrounding tissue—also serves as a limitation for lesions that are deep and involving underlying muscle.

The rate of complications was very low; however, 1 patient required transfer to the intensive care unit for respiratory observation. We recommend that every patient who undergoes RF ablation treatment of the oral cavity be observed in the hospital during the immediate postoperative period, with a low threshold for intensive monitoring if the patient develops any respiratory symptoms. Possible complications include postoperative hemorrhage leading to acute swelling and airway obstruction.

Our experience with RF ablation treatment for localized, superficial microcystic LM in pediatric patients has been very promising. Postoperatively, patients did not have significant pain issues, tolerated feeding almost immediately, and had few airway complications secondary to tissue swelling. Radiofrequency ablation is an excellent tool for management of intraoral LM for selected patients. These are ones with symptomatic, superficial microcystic lesions. For lesions involving deeper structures, multimodal treatments including surgical and sclerotherapy may be necessary.

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Correspondence: Reza Rahbar, DMD, MD, Department of Otolaryngology and Communication Enhancement, Children’s Hospital Boston, Harvard Medical School, 300 Longwood Ave, Ste LO367, Boston, MA 02115 (reza.rahbar@childrens.harvard.edu).

Author Contributions: Dr Rahbar had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. Study concept and design: Kim. Analysis and interpretation of data: Kauvanough, Orbach, Alomari, Mulliken, and Rahbar. Drafting of the manuscript: Kim. Critical revision of the manuscript for important intellectual content: Kim, Kauvanough, Orbach, Alomari, Mulliken, and Rahbar. Study supervision: Kim, Kauvanough, Orbach, Alomari, Mulliken, and Rahbar. Financial Disclosure: None reported.

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REFERENCES


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