International Journal of Pediatric Otorhinolaryngology 77 (2013) 1589–1592

Contents lists available at SciVerse ScienceDirect

International Journal of Pediatric Otorhinolaryngology

journal homepage: www.elsevier.com/locate/ijporl

Case report

Submucosal resection of a microcystic oropharyngeal lymphatic malformation using radiofrequency ablation

Prasad John Thottam [a,b,*], Randa Al-Barazi [a,b,*], David N. Madgy [a,b], Arlene Rozzelle [c,d]

[a] Children's Hospital of Michigan, Department of Otolaryngology, Detroit Medical Center, Detroit, MI, United States
[b] Michigan State University, Department of Surgical Services, Children's Hospital of Michigan, Detroit Medical Center, Detroit, MI, United States
[c] Children's Hospital of Michigan, Department of Plastic and Reconstructive Surgery, Detroit Medical Center, Detroit, MI, United States
[d] Wayne State University, Department of Pediatrics, Children's Hospital of Michigan, Detroit Medical Center, Detroit, MI, United States

ARTICLE INFO

Article history:
Received 8 May 2013
Accepted 27 May 2013
Available online 3 July 2013

Keywords:
Lymphatic malformation
Radio-frequency ablation
Coblation
Neck mass
Microcystic
Pediatric

ABSTRACT

Lymphatic malformations (LMs) are uncommon congenital anomalies noted to have a prevalence of 1 per 5000 births and comprise roughly 6% of all pediatric soft tissue lesions [1–3]. Recently radiofrequency ablation has been described as a surgical option for the treatment microcystic LMs in the oral cavity, more specifically the tongue. The following case describes the use of radiofrequency ablation for the submucosal removal of a large obstructing pharyngeal LM in a 4-year-old female. The mucosal sparing approach and surgical method of extirpation are discussed in detail. To the authors' knowledge this is the first description of a submucosal coblation technique being used as treatment for pharyngeal LMs.

1. Introduction

Lymphatic malformations (LMs) are uncommon congenital anomalies noted to have a prevalence of 1 per 5000 births and comprise roughly 6% of all pediatric soft tissue lesions [1–3]. They are predominately noticed before 2 years of age and occur most commonly in the head and neck region [4]. LMs do not involute with age and typically grow in proportion with the individual as they develop [4]. These anomalies can prove to be not only cosmetically deforming but also life threatening as they can acutely increase in size with infections and affect vital structures within the head and neck.

Treatment options for LM have been primarily limited to traditional surgical excision, sclerotherapy or a combination of both. Treatment predominantly has been tailored to individual LM structure and location. Recently radiofrequency ablation has been described as surgical option for the treatment microcystic LMs in the oral cavity, more specifically the tongue [5]. In the following case report, we describe the use of radiofrequency ablation, "coblation", for removal of a large submucosal pharyngeal LM impinging on the airway.

2. Case report

A 4-year-old female with a large microcystic LM involving the left side of her face, oropharynx and pharynx with extension to level T1–T2 presented to the Plastic Surgery Department with increased respiratory difficulty and witnessed apneic events despite CPAP treatments. In the past, she had undergone partial surgical excisions and sclerotherapy with sotradecol for the neck component of her LM.

Upon physical exam, there were no external signs of an enlarging cervical LM. The child was controlling her secretions but demonstrating a slight change in voice and mild retractions with respiration. A nasopharyngoscopy was performed and demonstrated an enlarging left oropharyngeal LM that was now obstructing greater than 80% of the patient's upper airway. Subsequently an MRI was performed illustrating an increase in size of the LM in both oropharyngeal and hypopharyngeal regions with significant encroachment on the airway (Fig. 1a and b). At this point the decision was made for the child to undergo a tracheostomy in order to secure her airway with anticipated intra-oral submucosal radiofrequency coblation of the lesion in the following weeks.

Three weeks after her tracheostomy, the child returned to the operating room for her procedure. Intraoperatively, the mouth was opened using a dingmann retractor and an incision was made medial to the left posterior tonsillar pillar through the mucosa only. Using blunt dissection, sub-mucosal flaps were elevated from

* Corresponding authors at: Department of Pediatric Otolaryngology, Children's Hospital of Michigan, 3901 Beaubien Street, Detroit, MI 48201, United States. Tel.: +1 330 936 6915; fax: +1 313 745 5848.
E-mail addresses: pthottam@gmail.com (P.J. Thottam), randabarazi@yahoo.com (R. Al-Barazi).

0165-5876/$ – see front matter © 2013 Elsevier Ireland Ltd. All rights reserved.
http://dx.doi.org/10.1016/j.ijporl.2013.05.037

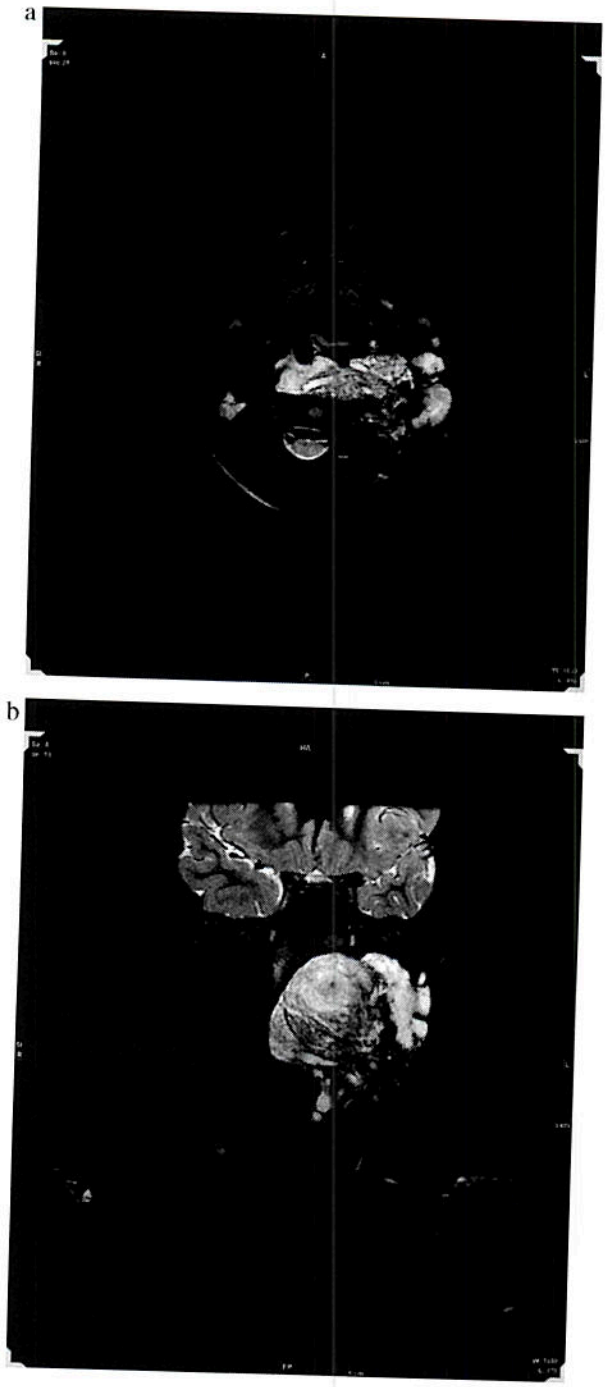

Fig. 1. (a and b) T2 MRI (a: axial and b: coronal) of large microcystic lymphatic malformation involving left parapharyngeal and retropharyngeal spaces and causing significant airway obstruction (patient is intubated).

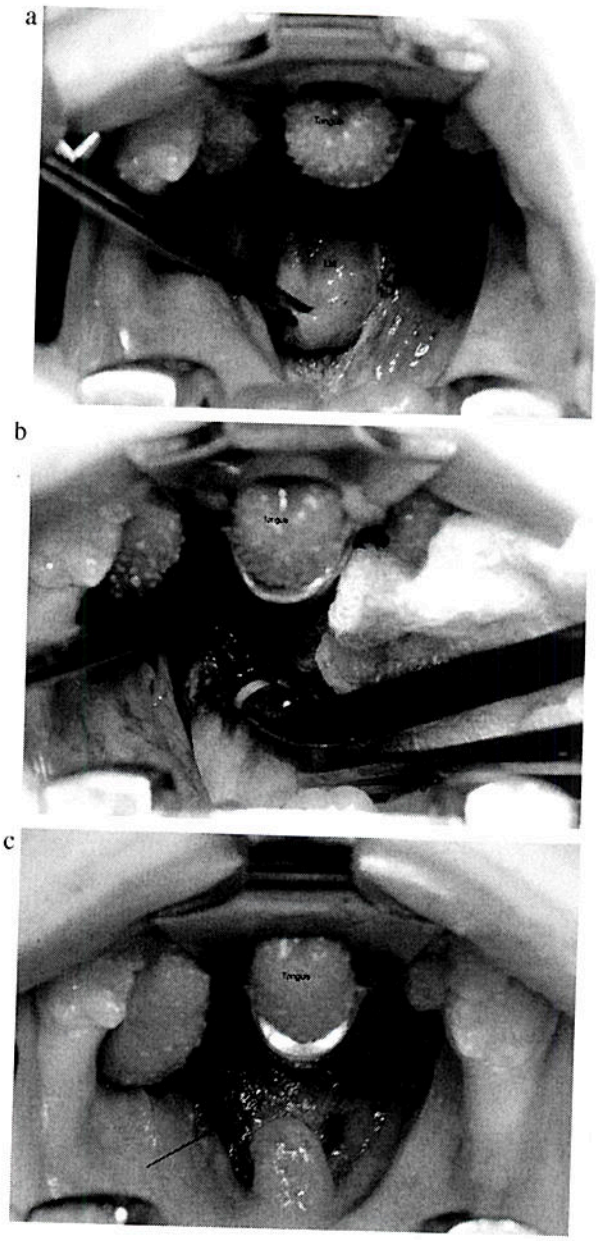

Fig. 2. (a) Surgical view of large left parapharyngeal and retropharyngeal LM being removed. Arrow is pointing to medialized oropharyngeal mucosa overlying the left parapharyngeal space. (b) Surgical view of hand-held radiofrequency ablation device being used to remove large LM from submucosal plane. (c) Surgical view of submucosal flap re-approximated and closed after removal of large LM.

the left parapharyngeal and retropharyngeal spaces in a lateral to medial fashion within the oro- and nasopharynx until adenoid tissue was identified superiorly. Inferiorly, this dissection was performed to the level of left aryepiglottic fold in the hypopharynx. This technique exposed the obstructing LM while preserving pharyngeal mucosa (Fig. 2a).

After the LM was properly exposed, a hand-held radiofrequency ablation device (PROcise EZ View Plasma Wand; Arthrocare Corp., Austin, TX) was utilized to extirpate the LM tissue using the bipolar ablation setting of 7 (Fig. 2b). Hemostasis was achieved at the same time using the coagulation setting of 3. This tissue was initially removed medially in an inferior to superior manner to give proper surgical space and visualization before addressing laterally based disease. After this was achieved, the device was directed laterally with care into the deep parapharyngeal space. Caution was taken to not disrupt the carotid sheath within this region by utilizing blunt dissection in areas of concern or pulsation. All visible LM tissue was removed from the airway. After completion the pharyngeal mucosal flaps were re-approximated and closed with 3.0 vicryl in a running-locking fashion (Fig. 2c). The patient was observed for 3 days post-surgery. During this time she tolerated a soft diet and had no respiratory events.

Three days after discharge she was readmitted for intraoral bleeding. On physical exam the patient a posterior pharyngeal bulge was noted. At that time an MRI was performed to rule out a

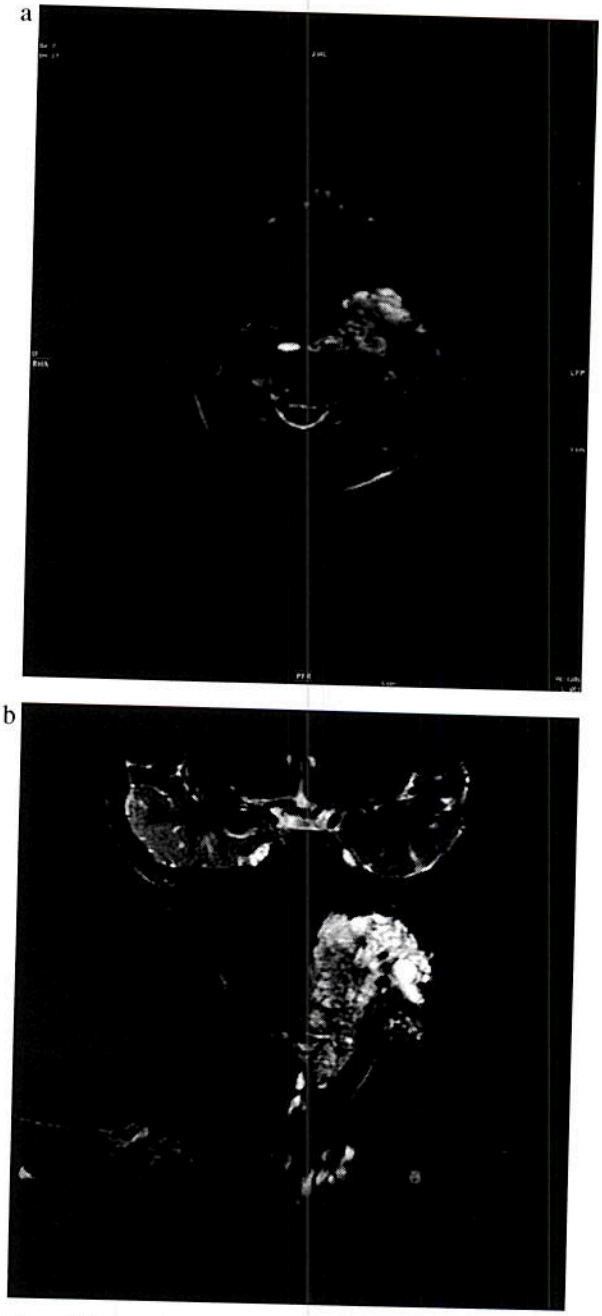

Fig. 3. (a and b) T2 MRI (a: axial and b: coronal) of significantly decreased microcystic lymphatic malformation after radiofrequency ablation. Airway is now patent.

carotid aneurysm. Fortunately, it was determined to be a LM macrocyst.

Intraoperatively, the posterior pharyngeal wound was found to be dehiscent and a blue mass was seen in the posterior pharyngeal wall obstructing greater than 50% of her upper airway. Ultrasound guided aspiration of the cyst was performed and the patient was observed for the following 2 days. No further complications associated with the surgery occurred and she was discharged home.

At her 6-month post-operative visit, the patient was eating and drinking without difficulty. She was tolerating tracheostomy capping trials without respiratory distress. A repeat MRI was performed and imaging demonstrated a significant decrease in LM size and encroachment on the airway (Fig. 3a and b). The patient was subsequently decannulated without difficulty and continues to be monitored by both Otolaryngology and Plastic Surgery.

3. Discussion

Lymphatic malformations are low flow embryological vascular anomalies theorized to develop from mesenchymal progenitor cells [4]. A subcategory of vascular malformations, LM can be further classified as macrocystic (cysts $> 2\ cm^3$), microcystic (cysts $< 2\ cm^3$), or mixed [4,6]. The characteristics of the cysts involved with these LM are of particular importance as they respond more favorably to different forms of therapy.

Sclerotherapy is frequently used for macrocystic disease and has became an attractive treatment option because it is less invasive, leaves little scarring and there is little risk of collateral damage to surrounding neurovascular structures. Unfortunately, this technique works poorly for microcystic disease, has significant post-injection swelling and possibly an increased rate of recurrence [4,7]. It is for this reason that sclerotherapy is not viewed as a favorable option for LMs directly involving the airway [4].

Traditional surgical intervention for microcystic and mixed disease has provided the advantage of more complete removal and short recovery time when compared to sclerotherapy as a form of treatment. Unfortunately, due to the complexity and invasiveness of these malformations in the cervicofacial region there is a relatively high potential for injury to surrounding structures using traditional surgical methods. Furthermore, due to the nature of these lesions, it can be surgically difficult due to altered tissue planes.

In recent literature radiofrequency ablation has been described as a new surgical option for the removal of vascular malformation such as LMs. Radiofrequency ablation produces a controlled increase in temperature within tissue resulting in denaturation and obliteration of the directed site [8]. The energy generated dissipates quickly and causes little collateral damage to adjacent structures and sensitive mucosal surfaces making it an attractive option for LM removal in the airway [8].

In this case report we describe the submucosal removal of a large microcystic LM occluding the pharyngeal airway with radiofrequency ablation. Unlike traditional cold surgical techniques, this method facilitated hemostasis and precision in the removal of a large LM while sparing the surrounding vital structures. The submucosal approach to allowed maximal preservation of mucosa, easier access to pharyngeal LM and prevented the possibility of pharyngeal granulation and scarring. To our knowledge, this is the first reported case of radiofrequency ablation for the submucosal removal of a microcystic LM involving the pharynx.

Conflicts of interest

No conflicts of interest exist for these authors. No relevant financial relationship exists between the authors and procedures or products used in this manuscript.

References

[1] J.A. Perkins, S.C. Manning, R.M. Tempero, M.J. Cunningham, J.L.Jr. Edmonds, F.A. Hoffer, et al., Lymphatic malformations: current cellular and clinical investigations, Otolaryngol Head Neck Surg 142 (2010) 789–794.
[2] Y. Bajaj, S. Hewitt, S. Ifeacho, B.E.J. Hartley, Surgical excision as primary treatment modality for extensive cervicofacial lymphatic malformations in children, Int J Pediatr Otorhinolaryngol 75 (2011) 673–677.
[3] C.M. Coffin, L.P. Denher, Vascular tumours in children and adoloscents: a clinicopathologic study of 228 tumours in 222 patients, Pathol Annu 28 (1993) 97–120.
[4] J.P. Renton, R.J.H. Smith, Current treatment paradigms in the management of lymphatic malformations, Laryngoscope 121 (2011) 56–59.

[5] N.G. Ryu, S.K. Park, H.S. Jeong, Low power radiofrequency ablation for symptomatic microcystic lymphatic malformation of the tongue, Int J Pediatr Otorhinolaryngol 72 (11) (2008) 1731–1734.
[6] C.M. Giguere, N.M. Bauman, Y. Sato, D.K. Burke, J.H. Greinwald, S. Pransky, et al., Treatment of lymphangiomas with OK-432 (picibanil) sclerotherapy: a prospective multi-institutional trial, Arch Otolaryngol Head Neck Surg 128 (2002) 1137–1144.
[7] S.J. Boardman, L.A. Cochrane, D. Roebuck, M.J. Elliott, B.E. Hartley, Multimodality treatment of pediatric lymphatic malformations of the head and neck using surgery and sclerotherapy, Arch Otolaryngol Head Neck Surg 136 (2010) 270–276.
[8] S.D. Colbert, L. Seager, F. Haider, B.T. Evans, R. Anand, P.A. Brennan, Lymphatic malformations of the head and neck – current concepts in management, Br J Oral Maxillofac Surg 51 (2012) 98–102.