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**Case Report**

# The successful use of bipolar radiofrequency ablation (coblation) in treatment of a lymphatic malformation affecting the upper airway

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## ABSTRACT

Lymphatic malformations of the head and neck region frequently involve the upper aero digestive tract. Patients with these lymphatic malformations may present in early infancy with sudden airway compromise. This necessitates early intervention with intubation or tracheostomy for airway stabilization. The etiology and pathogenesis of lymphatic malformations is still unclear, and a wide array of treatment modalities has been proposed. We present a case report of a neonate with a lymphatic malformation involving the upper airway, and discuss how the patient was initially stabilized with a tracheostomy, following which he underwent a series of staged procedures, including bipolar radiofrequency ablation, which eventually led to successful decannulation.

**Keywords:** Lymphatic malformation, Lymphangioma, Airway, Larynx, Coblation

## INTRODUCTION

Lymphatic malformations (LMs) are uncommon vascular malformations, with an incidence of 1 in 2,000-4,000 live births.[1] They most commonly affect the head and neck region, though they may also present as painless soft compressible masses of the back, hip, abdomen and inguinal regions. Upper aero digestive tract involvement is common in LMs that affect the head and neck. In the largest published series of LMs of the airway, 73% of head and neck LM patients had upper aero digestive tract disease, though a much smaller proportion (25%) presented with acute airway obstruction.[2] We present a case of a neonate with LM involving the larynx and oropharynx that presented with airway obstruction at birth and discuss the multi-modality treatment that led to a successful outcome in this patient. Ethics approval was obtained from the Singhealth Centralised Institutional Review Board prior to commencing the study.

## CASE REPORT

A 21-year-old Indian woman with no significant past medical history had an uneventful pregnancy until 20 weeks gestation, when a routine fetal anomaly ultrasound scan revealed a septated cystic lesion below the jaw measuring 3.4cm x 1.8cm. There was no obvious blood flow within the lesion and the amniotic fetal index (AFI) was normal. There were no other fetal anomalies detected on the ultrasound scan. The case was discussed at a birth defect clinic and was thought to be a lymphatic malformation. Amniocentesis was performed and revealed a normal 46XY karyotype. Serial ultrasounds were performed to monitor the size of the cystic mass. At 27 weeks gestation, the cystic mass had grown to 4.4cm x 1.8cm and at 34 weeks gestation, the mass measured 6.9cm x 5.2cm, with no extension into the trachea or thoracic cavity.

A planned lower section caesarian section was done at 37 weeks gestation and the baby had an initial Apgar score of 7 at 1 minute which improved to 9 at 5 minutes. His birth weight was 2640 grams. Shortly after birth, the baby required intermittent positive pressure ventilation for poor respiratory effort and this was converted to continuous positive airway pressure (CPAP) with maintenance of oxygen saturations above 95 percent. On physical examination he had intermittent stridor and there was a 5cm mass in the left submental and submandibular region (Figure 1A). The mass was soft in consistency and compressible. There were no overlying skin changes.

He was urgently referred to the otorhinolaryngology department and underwent microlaryngobronchoscopy (MLB) and intubation on day 2 of life. Findings were that of a LM affecting the tongue base and vallecula with the epiglottis largely replaced by a globular mass (Figure 2A) that obstructed the laryngeal inlet. The subglottis, trachea and carina appeared normal. An MRI of the head and neck revealed a large mixed macrocystic and microcystic involving bilateral neck spaces. In the left neck the lesion involved the posterior cervical, carotid, visceral and parapharyngeal spaces. It involved the infratemporal fossa with extension across the midline via the retropharyngeal space and nasopharynx into the right parapharyngeal space and right infratemporal fossa. There was also involvement of bilateral submandibular and sublingual regions. Caudally, the lesion extended below the left sternocleidomastoid muscle with slight extension into the left anterior-superior mediastinum, above the level of the confluence of the left internal jugular and left brachiocephalic veins (Figure 3). The larger component on the left measured 5.8 by 3.8cm and the smaller component measured 2.5 by 2.1cm on the right. The LM was thus stage V according to de Serres et al classification (bilateral infrahyoid and suprahyoid), which gave it a bad prognosis.[3]

As the laryngeal and oropharyngeal component of the mass was microcystic and deeply infiltrative, complete trans cervical resection was not possible. A decision was made for a combined trans cervical and trans oral resection of the LM. The aim of the surgery was to remove as much LM from the neck as possible while preserving vital structures. In the same sitting, in an attempt to avoid a tracheostomy, we planned to ablate the laryngeal and oropharyngeal components of the LM with carbon dioxide ($C0_2$) laser. The baby underwent excision of the neck component of the LM and $C0_2$ laser reduction of the supraglottic and tongue-base component of the LM on day 5 of life. He initially failed extubation on day 7 of life, but was successfully extubated to CPAP on day 12 of life. However, following extubation he had intermittent stridor and multiple transient desaturations. A repeat MLB on day 23 of life revealed severe edema of epiglottis and residual LM of the tongue base with complete obstruction of the laryngeal inlet. Decision was finally made to proceed with a tracheostomy to secure the airway on day 26 of life. Immediately following the tracheostomy, a second $C0_2$ laser debulking of the supraglottic lymphatic malformation was performed, and he was discharged home well with tracheostomy and regular follow up.

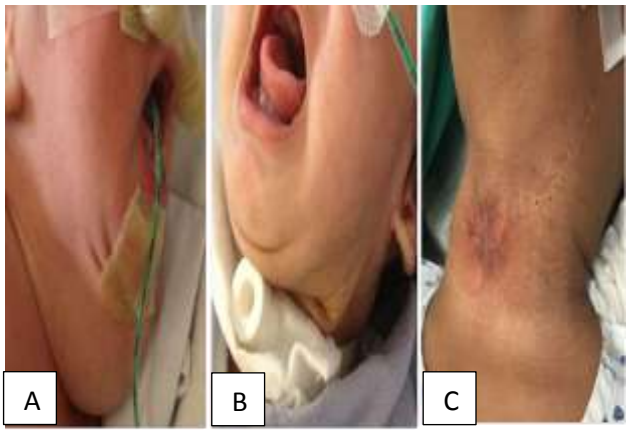


**Figure 1: Clinical photo of progress of cervical component of lm ; A) on day 2 of life; B) at 7 months old; C) at 3 years 6 months old.**

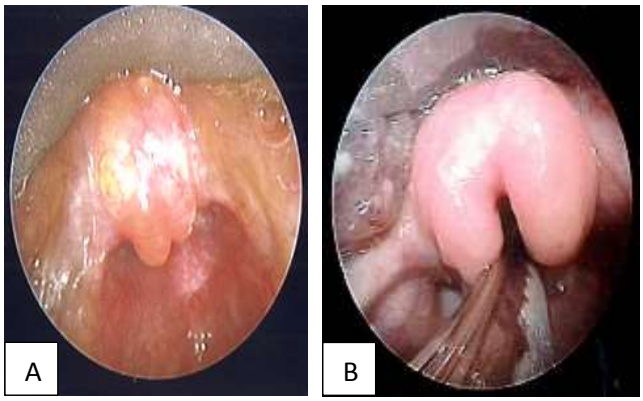


**Figure 2: MLB photo of progress of LM affecting the upper airway; A) on day 2 of life; B) at 3 years 6 months old following coblation.**

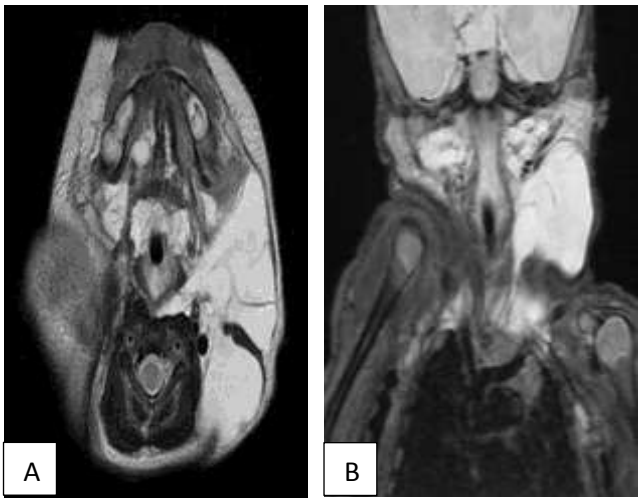


**Figure 3: MRI of LM on day 2 of life; A) axial view; B) coronal view.**

At 7 months old, he was tolerating oral feeding and weighed 8.2 kg (50th centile). He also achieved appropriate developmental milestones such as being able to roll over, reach for objects and having stranger anxiety. On examination of his neck, the cervical component of the LM was significantly smaller (Figure 1B). However, a repeat MLB showed persistent swelling of the epiglottis and tongue base. A decision was made not for further laser application as the airway component of the LM was not responding well to the treatment, and to let the child grow and see if the airway obstruction improved. MLB performed at 6 monthly intervals revealed stable but persistent airway obstruction. The child was thus unable to be decannnulated from the tracheostomy tube.

When the child was 21 months old, scattered case reports emerged on the successful use of bipolar radiofrequency ablation (coblation) for LMs involving the tongue.[4,5] Following these reports, we attempted coblation reduction of the tongue-base component of the LM, allowing the wound to heal by secondary intention. The child subsequently underwent 2 more sessions of coblation of the tongue base and epiglottic LM with significant improvement of the airway obstruction (Figure 2B). He also underwent tonsillectomy to enlarge the oropharyngeal airway. He was then successfully decannnulated at 26 months of age. There was no recurrence of the neck component of the LM at 3 years follow up (Figure 1C). The child had a good voice. He was tolerating soft diet well, and was asymptomatic except for occasional snoring.

## DISCUSSION

The etiology and pathogenesis of lymphatic malformations is still unclear, and a wide array of treatment modalities has been proposed. The primary goal of treatment of LMs is restoration or preservation of functional and aesthetic integrity, with surgery being the mainstay of treatment in many centres.[6] The operative approach should take into the account the benign nature of the disease, allowing for subtotal resection if the LM is in close proximity to neurovascular structures, which was the decision in this case.[7] The site of the lesion is a major determinant for success of surgery, and risk factors for recurrence include suprahyoid lesions, lesions affecting multiple anatomic sites, and infiltrative microcystic LM in any location.[1,8,9]

In cases where complete surgical resection is difficult due to extensive disease, sclerotherapy provides a viable treatment alternative, with the possible additional advantage that complications are reported less frequently in sclerotherapy studies than in surgical studies.[10] Examples of sclerosants used are picibanil (OK-432), doxycycline, bleomycin, ethanol and sodium tetradecyl sulfate. We did not consider sclerotherapy in our case as the primary surgeon felt the large macrocystic neck component of the LM was amenable to surgery, and following surgery, no recurrence in the neck was observed.

The management of LMs affecting the airway is challenging to most otolaryngologists. These LMs often present in early infancy with airway obstruction and potential airway emergencies. As in our case, this necessitates early treatment with intubation or tracheostomy for airway stabilization. Owing to better pre-natal imaging methods such as prenatal ultrasound and fetal MRI, the location and extent of LMs can be accurately predicted prior to delivery. In several cases, ex utero intrapartum treatment (EXIT) has been successfully achieved when congenital high airway obstruction (CHAOS) by a large lymphatic malformation was diagnosed during antenatal investigations.[11,12]

The treatment of LMs involving the airway has not been widely reported. Laryngeal (supraglottic) involvement, as was the finding in our case, is uncommon, with only 11% (15 out of 141) having laryngeal involvement in a large series of LMs of the airway.[2] Carbon dioxide laser has been effective in selected cases.[13,14] However, side effects include post-operative edema and scarring, as observed in our patient.

Recent data suggests that radiofrequency ablation is effective in reducing the size of oral cavity lymphatic malformations and improving symptoms such as pain, bleeding and infection, but no large study on its use in laryngeal LMs has been reported.[15,16] Bipolar radiofrequency ablation (coblation) has the ability to destroy tissue at much lower temperatures (40-70 degrees) than other forms of radiofrequency. This minimizes collateral damage to adjacent tissue resulting in less post-treatment swelling and scar formation. Our experience with this case suggests that bipolar radiofrequency ablation (coblation) may be superior to laser ablation in managing LMs of the tongue base and supraglottis. Larger studies, however, are required to support this hypothesis.

## CONCLUSIONS

LMs affecting the airway are challenging conditions. Children with high stage lesions that present in early infancy with airway compromise should have their airway stabilized with a tracheostomy first, followed by staged, multimodality treatment. Surgery remains a mainstay of treatment and bipolar radiofrequency ablation (coblation) is a good adjunct for treatment of LMs affecting the upper airway.

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