Radiofrequency Tissue Volume Reduction: Suggested Treatment for Lymphatic Malformation

Prakobkiat Hirunwiwatkul MD*

** Department of Otolaryngology, Faculty of Medicine, Chulalongkorn University*

Lymphatic malformation is a benign congenital lesion but is often challenging to deal with. Complete excision is its treatment of choice. It has a high rate of recurrence, since a complete excision is difficult, and it may cause cosmetic or functional defect. Radiofrequency tissue volume reduction (RTVR) is used for the treatment of sleep-disordered breathing and other areas of surgery. A case of cervical lymphatic malformation reported here is effectively treated by radiofrequency without scar or any complication.

Keywords : *Radiofrequency, Lymphatic malformation, Cystic hygroma*

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Lymphatic malformation is a congenital disease with a predilection for the head and neck and usually presents in a pediatric population[1]. Such lesions are most often seen in infancy; 80% of the cases appear within the first two years of life[2]. In the head and neck area, the most common location is the submandibular region (37%), followed by the parotid gland (31%)[3]. It typically extends into adjacent structures without respecting the fascial planes; it tends to grow slowly, and rarely it has spontaneous regression. Hemorrhage (spontaneous, trauma) and/or infection (viral, bacterial) often leads to its rapid increase in size, which possibly obstructs the initial portion of the upper aero-digestive tract or causing cosmetic deformity.

Lymphatic malformations can be characterized as microcystic, macrocystic or combined. Microcystic lesion has an old term, "Lymphangioma". It typically consists of multiple small cysts, which are commonly found in the oral cavity or oropharynx. The term "Cystic Hygroma" was once used for a macrocystic lesion. It usually consists of large, thick-walled cysts which commonly occur in the neck.

Although lymphatic malformations have a distinctive sonographic appearance, Magnetic Resonance Imaging (MRI) allows a better tissue characterization and extension[4]. T2-weighted images show hyperintense due to their high water content. Contrast administration with Gadolinium usually results in enhancement of the rims of large cysts or *septae*. This is helpful for the diagnosis and preoperative staging of cervico-thoracic lymphatic malformations in children.

Historically, treatment of cystic hygromas has ranged from a variety of medical therapies to aggressive surgical techniques. Medically, a number of approaches have been investigated. Radiation therapy has been shown to be largely ineffective and has been abandoned altogether. Recently, direct injection of sclerosing agents into the cystic lesions has received more attention.

Now there are two main options of treatment, namely: intralesional embolization with OK-432 or surgical excision. OK-432 is a lyophilized incubation mixture of group A Streptococcus pyogenes of human origin. It produces an inflammatory reaction that is followed by a gradual regression of the lesion. It may be effective treatment for macrocystic lymphatic malformations. It can be used as the primary therapy, after partial surgical excision, or in recurrent lymphatic malformations. The white blood cells induced and activated by OK-432, and the cytokines (including TNF) produced by these cells increase the endothelial permeability; and thus the accelerated lymph drainage and increased lymph flow leads to the shrinking of the cystic spaces[5-19].

Lesions involving the lip, hypopharynx and/or larynx, tongue, and the floor of the mouth had high rates of recurrence or persistent disease. Their

Correspondence to : Hirunwiwatkul P, Department of Otolaryngology, Faculty of Medicine, Chulalongkorn University, Bangkok 10330, Thailand.

treatments range from observation to extensive and multiple resections. Complete excision is the treatment of choice which has an 81% rate of cure. Partial excision is curative in only 12%, but it decreases enough of the tumor to eliminate the need for further treatment in 62%[20].

Many factors should be considered when surgical treatment is planned. They are, namely: timing of surgery, mode of dissection, blood loss and duration of procedure, staging of procedure, extension of lesion whether it involves nerves, vessels or other vital structures. To address these issues in practice, lymphatic malformations are divided into two types. Type1 is located below the level of mylohyoid muscle. In general, it is usually a macrocystic lesion consisting of large, thick-walled cysts. It may be safely resected within the first 12 months of life and sharp dissection in one setting is recommended. Type 2 is found above the level of mylohyoid muscle. It is poorly defined and frequently microcystic lesion which consists of multiple small cysts. Complete excision may not be possible and multiple staged procedures are usually needed. Surgery is recommended for type2 lymphatic malformation; if possible it should be carried out before the age of 5.

Aspiration of the cysts may be of some value in the diagnosis and management of acute respiratory problems; however, it is not for definitive therapy[21].

TCRF (temperature controlled radiofrequency) has been shown to be a safe, effective, well-tolerated and technically straightforward procedure for the treatment of sleep-disordered breathing[22-28]. In the present study, we describe a new and more conservative surgical approach to this lesion, using radiofrequency tissue ablation techniques.

Recent animal studies have been published. They show that the specific size of the lesion created in the tongue is directly proportional to the number of joules applied.[28] Lesions of standard size have been noted at an application between 800 and 1400 J. Although not previously tested in children, RFA has been used to reduce tongue volume in adults with sleep-disordered breathing. In one study, a group of 18 patients treated with RFA for significant sleep-disordered breathing obtained an overall average tongue-base volume reduction of 17%, and a 53% increase of the posterior airway space. These patients were also noted to have 77% decrease in their apnea index. An excellent safety profile was noted throughout the treatments. Although these subjects did not present with pathological tongue lesions, their significant volumetric reductions showed promises for various applications.[25]

Case Report

A 23-yr-old Thai man presented with a complaint of neck mass on the left submandibular area for 1 month (Fig. 1, 2). His neck lump was soft on palpation. Left submandibular sialography with Ultrasonography showed a large cystic mass located at the left submandibular area, size about 7x1.5 cm. The left submandibular gland was slightly enlarged without any mass. Submandibular ductal portion was normal.

Fig. 1, 2 Initial presentation of a cervical soft macrocystic lymphatic malformation on the left submandibular area

Surgical Technique

Local infiltration of 0.2 ml of 1% Xylocaine on each needle insertion sites was applied. Somnus radiofrequency probe (Somnus Medical Technologies, Sunnyvale, CA) was placed into the left submandibular mass at the point of maximal deformation and 800 to 1200 J of radiofrequency energy was applied. Additional 5 treatment sites were placed a minimum of 1.0 cm apart. Four to six sites were treated during each procedure (Fig. 3, 4).

When the active part of the radiofrequency probe was close to the marginal mandibular branch of the facial nerve, the twitching of depressor *anguli oris* was seen. The probe was removed from that point and inserted into another part of the mass in order to avoid nerve injury.

Postoperatively, the patient was on a soft diet and was advanced to a regular diet the following day. Tylenol and Tylenol with codeine were prescribed to the patient with a postoperative antibiotic (Dicloxacillin 500 mg, *tid.*) for 5 days. Further procedures will be carried out in similar fashion with 4- to 8-week intervals until the neck mass resolved.

Result

No neurological deficit was found postoperatively. The patient complained of a little pain that occurred during and after the procedure, but did not need any narcotic analgesic drug. The submandibular neck mass was slightly swollen with no tenderness and recovered from inflammation within a week. Two weeks later, the neck mass was of the same size. On the fourth week, the mass began to shrink and on the sixth week postoperatively, the authors saw the maximum effect of treatment (Fig. 5, 6).

Ultimately, the patient underwent two more treatment sessions. During the first session, he received 1000 J of radiofrequency energy per treatment area, with six areas treated each time. The patient had a moderate response with minimal discomfort. The second treatment focused on nine areas with 1000 J per each area. There was no recurrence of lymphatic malformation within the 2 year follow-up period after the third treatment.

Discussion

Radiofrequency tissue volume reduction (RFTVR) offers a relatively new and unique energy delivery technique. Radiofrequency energy produces a controlled temperature increase within the tissue that causes denaturing of local proteins, and it avoids vaporization of tissue that occurs with laser and cauterization techniques. This controlled temperature increase is unique, because it is generated not by the probe itself, as in the case with cautery or laser use, but by vibrations within individual cells themselves. This allows the skin or mucosal surfaces where the probe passes through remain undamaged during the treatment. Radiofrequency ablation also enables the volume of the treated tissue to be precisely chosen, since energy dissipates rapidly from the radiofrequency electrode in a ratio of 1/radius^4.

Despite the multiple areas of treatment, the patient demonstrated only mild to moderate edema, which peaked within the first 24 hours after th treatment and the resolution of edema was seen within

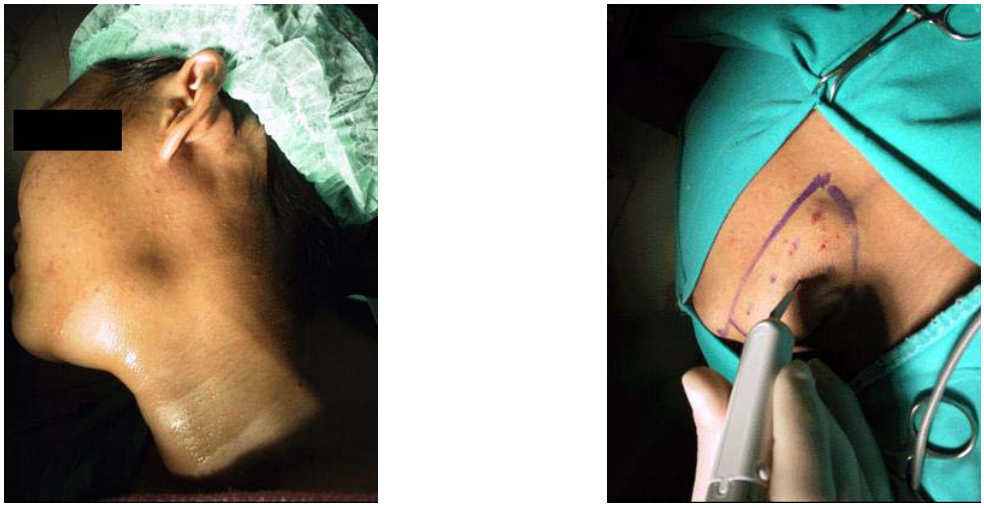

Fig. 3, 4 Surgical technique of radiofrequency tissue volume reduction, a treatment for lymphangioma

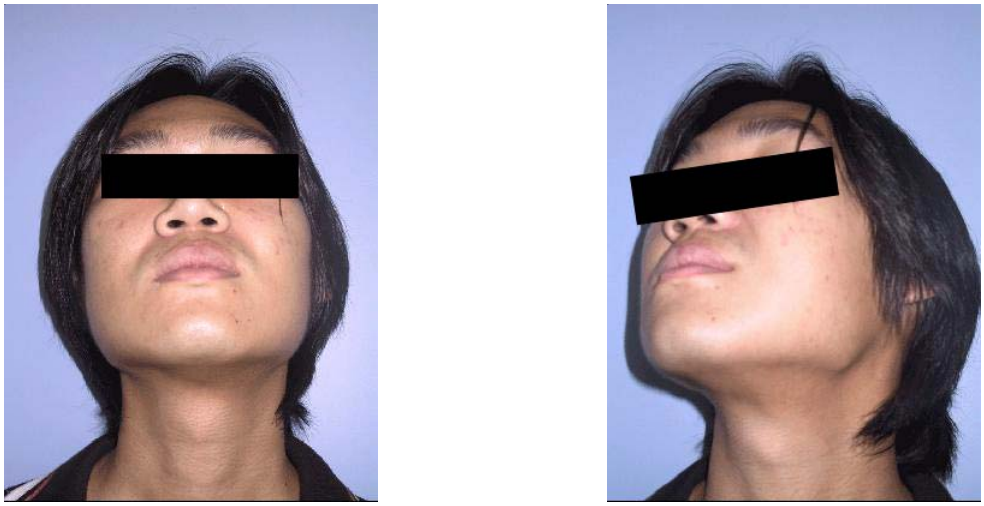

Fig. 5, 6 After two sessions of radiofrequency ablation, the left neck mass could not be seen or palpated

approximately 1 week. Over a period of 6 to 8 weeks, the lesion was progressively reduced, and it disappeared after two more treatment sessions. There was no recurrence found in the 2 year follow-up period.

Radiofrequency ablation therapy offers patients with lymphatic malformations a number of advantages. First, the technique may be used to reach precise targeted tissue with sparing of critical anatomical neurovascular components. Second, the treatment delivered is not limited by the diffusion of injected agents, as in sclerosing therapy (this appears to be a weakness in the use of OK-432). Third, the procedure entails minimal invasive techniques and avoids significant pain and morbidity involved with an incision placed on the neck. The authors think the technique offers a procedure with minimal morbidity.

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รายงานผู้ป่วย Lymphatic malformation ที่ได้รับการรักษาโดยใช้คลื่นวิทยุ

ประกอบเกียรติ หิรัญวิวัฒน์กุล

Lymphatic malformation เป็นโรคไม่ร้ายแรงที่เป็นแต่กำเนิด และมีปัญหาในการรักษา การผ่าตัดเอาออกทั้งหมดถือเป็นการรักษาที่ดีที่สุด แต่มักมีการเกิดซ้ำเนื่องจากการผ่าตัดเอาออกทั้งหมดทำได้ยาก และอาจทำให้เกิดปัญหาด้านความสวยงามหรือการทำงานของอวัยวะนั้น ๆ รายงานฉบับนี้ ได้รายงานผู้ป่วย Lymphatic malformation 1 ราย ที่ได้รับการรักษาโดยใช้คลื่นวิทยุ ซึ่งใช้ในการรักษาโรคนอนกรน พบว่าใช้รักษา Lymphatic malformation ได้ผลดี โดยไม่มีรอยแผลเป็น หรือ ภาวะแทรกซ้อนใด