



## ORIGINAL RESEARCH

# Cervical lymphangioma in adults: A report of seven cases and review of the literature

Jianwei Wang MD<sup>1,2</sup> | Yujuan Yang MD<sup>1,2</sup> | Jing Guo MD<sup>1,2</sup> | Yao Yao BS<sup>1,2</sup> |  
Luchao Dong BS<sup>1,2</sup> | Yakui Mou MD<sup>1,2</sup> | Yu Zhang MD, PhD<sup>1,2</sup>  |  
Xicheng Song MD, PhD<sup>1,2</sup> 

<sup>1</sup>Department of Otorhinolaryngology, Head and Neck Surgery, Yantai Yuhuangding Hospital, Qingdao University, Yantai, China

<sup>2</sup>Shandong Provincial Clinical Research Center for Otorhinolaryngologic Diseases

## Correspondence

Xicheng Song, Department of Otorhinolaryngology, Head and Neck Surgery, Yantai Yuhuangding Hospital, Qingdao University, No. 20, East Road, Zhifu District, Yantai 264000, China.  
Email: [drxchsong@163.com](mailto:drxchsong@163.com)

Prof. Yu Zhang, Department of Otorhinolaryngology, Head and Neck Surgery, Yantai Yuhuangding Hospital, Qingdao University, No. 20, East Road, Zhifu District, Yantai 264000, China.  
Email: [superzhang013@163.com](mailto:superzhang013@163.com)

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## Abstract

**Background:** Cervical lymphangioma usually occurs in children and is relatively rare in adults. The purpose of this study was to investigate the characteristics of cervical lymphangiomas in adults.

**Methods:** We retrospectively reviewed the medical records and analyzed clinical data obtained from seven adult patients who suffered from cervical lymphangioma from January 2008 to June 2021.

**Results:** Five (71.43%) of seven cases were asymptomatic and four (57.14%) lymphangiomas had a maximum diameter  $\geq 10$  cm. Each cervical lymphangioma was located adjacent to the carotid sheath. All the cervical lymphangiomas were completely surgically excised and were pathologically diagnosed as cystic lymphangioma. After a median 32 months follow-up period, no recurrence was observed.

**Conclusion:** Cervical lymphangiomas from adults are often large asymptomatic masses infiltrating or adjacent to surrounding vital structures which brings many difficulties to the surgery, requiring an experienced surgeon to remove the mass.

## KEYWORDS

adult, cervical lymphangioma, sclerotherapy, surgical excision, vital structure

## 1 | INTRODUCTION

Lymphangiomas are congenital, benign swellings deriving from lymphatic malformations. They most commonly occur in the neck, but may also present in the axilla, mediastinum, groin, and abdominal organs.<sup>1,2</sup> Although lymphangiomas occurred in 2% of pediatric patients may be associated with chromosomal abnormalities, the underlying mechanism of lymphatic malformation remains unclear.<sup>3</sup> A

lymphangioma usually presents with no symptoms, due to its soft characteristic. However, it can result in symptoms of compression that sometimes lead to difficulty in breathing or swallowing, by infiltrating adjacent structures.<sup>4</sup>

Approximately 90% of lymphangiomas occur in pediatric patients <2 years of age,<sup>5,6</sup> and they are rarely found in adults. Until now, fewer than 150 cases of adult lymphangioma have been reported in the literature.<sup>7,8</sup> It has been proposed that risk factors, such as cervical infection, neck trauma, and a prior neck surgery might contribute to the proliferation of lymphangiomas in adults.<sup>9,10</sup> However, the

Jianwei Wang and Yujuan Yang contributed equally to this work.

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underlying pathogenesis and clinical characteristics of lymphangiomas remain unclear. In this study, we performed a retrospective review of seven adult patients with cervical lymphangioma and described their clinical characteristics and treatment, with the goal of providing useful clinical experience to surgeons.

## 2 | METHODS

We conducted a retrospective review of patients that had been hospitalized in the Otorhinolaryngology Department of Yantai Yuhuangding Hospital from January, 2008 to June, 2021. The patients were identified by searching the electronic medical record system. The data for patients pathologically diagnosed with lymphangioma were reviewed and recorded. Tumors that were pathologically diagnosed as lymphangioma but located in areas other than the neck were excluded. Patients with cervical lymphangioma but were <18 years old were also excluded. Finally, a total of seven patients were found to be eligible for the study. These variables used for analysis were as follows: gender, age, duration of tumor onset, distension pain, pressing pain, feeling of swelling, dysphagia, dyspnea, infection, prior neck trauma, history of neck surgery, notable enlargement of the tumor before hospitalization, tumor side, tumor location, tumor size, cystic classification, tumor adjacent to the carotid sheath, tumor located medial to the sternocleidomastoid muscle, use of negative pressure drainage, duration of placement of negative pressure drainage tube, histopathological classification, follow-up duration, and tumor recurrence. Tumor size was assessed by measurements of maximum diameter that were based on imaging findings combining with intraoperative confirmations. Cystic classification was determined radiographically, with macrocystic type defined as lymphangioma comprising one or more cysts all of which were greater than 2 cubic centimeters (cc) in volume, microcystic type defined as lymphangioma comprising of multiple cysts all of which were less than 2 cc in volume, and mixed type of which macrocystic component  $\geq 50\%$  of the total disease.<sup>11</sup> The study protocol was approved by the Ethics Committee of Yantai Yuhuangding Hospital of Qingdao University (Approval no. 2020-297). This was a retrospective study and did not involve the patient's privacy or commercial interests, so the Ethics Committee approved an exemption of informed consent.

## 3 | RESULTS

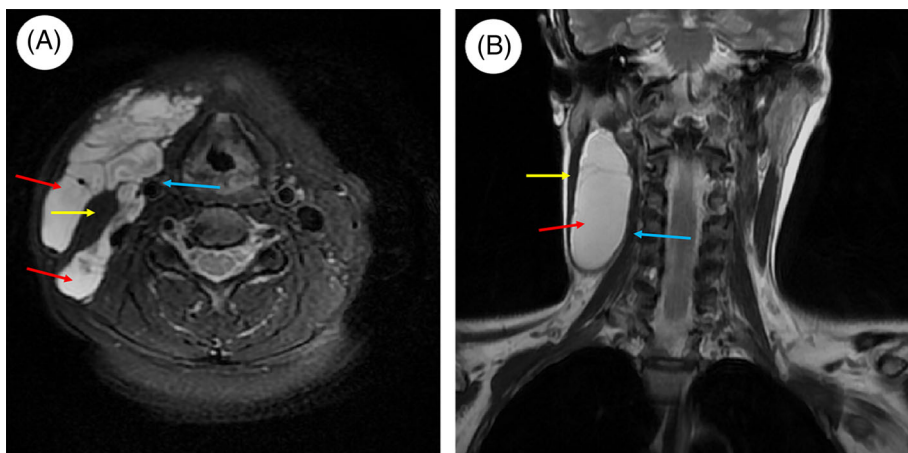
### 3.1 | Patient characteristics

A total of seven adult patients suffering from cervical lymphangioma were included in the study (Table 1). Most patients were female (five, 71.43%). The age at diagnosis ranged from 25 to 52 years old, with a median age of 43 years. All the patients had an obvious mass in the neck for 1–24 months prior to hospitalization, with a median time of 12 months. There was one patient who reported distension pain in the neck and another patient who complained of pharyngeal swelling,

TABLE 1 Patients and tumor characteristics

No.	Gender	Age (years)	Duration of onset (months)	Feeling of pain or swelling	Infection or prior neck trauma	History of neck surgery	Notable enlargement of tumor before hospitalization	Tumor side	Location	Tumor size (cm × cm)	Cystic type	Adjacent to carotid sheath	Tumors medial to the sternocleidomastoid muscle	Histopathological type	Treatment	Negative pressure drainage (days)	Follow-up duration (months)	Recurrence
1	Female	41	1	Distension pain in neck	No	No	No	Right	Parapharyngeal space	6.0 × 4.0	Macrocystic	Yes	No	Cystic	Surgery	5	55	No
2	Male	50	24	No	No	No	No	Left	Multiple regions	8.1 × 2.8	Mixed	Yes	Yes	Cystic	Surgery	4	40	No
3	Female	43	18	Swelling in pharynx	No	No	No	Right	Multiple regions	12.0 × 10.0	Macrocystic	Yes	Yes	Cystic	Surgery	2	36	No
4	Female	34	24	No	No	No	No	Right	Postclavicular	6.0 × 5.0	Macrocystic	Yes	No	Cystic	Surgery	3	32	No
5	Female	52	12	No	No	Yes	No	Right	Multiple regions	14.0 × 10.0	Mixed	Yes	Yes	Cystic	Surgery	7	18	No
6	Male	52	12	No	No	No	No	Left	Superclavicular	10.0 × 10.0	Macrocystic	Yes	Yes	Cystic	Surgery	3	10	No
7	Female	25	1	No	No	No	No	Left	Multiple regions	10.0 × 8.0	Macrocystic	Yes	Yes	Cystic	Surgery	3	9	No

**FIGURE 1** (A, B). Representative imaging scans of lymphangiomas (red arrow) infiltrating medial to the sternocleidomastoid muscle (yellow arrow) and adjacent to the carotid sheath (blue arrow).



with the remaining five patients asymptomatic. No local or systemic infection, prior neck trauma, or rapid tumor enlargement occurred among these patients. Only one patient underwent a prior neck surgery, with a cervical lipoma surgically excised 24 years ago.

### 3.2 | Tumor characteristics

All tumors were unilateral, with three located in the left side of the neck and four located in the right side. According to radiographic examinations, five patients (71.43%) were determined with macrocystic lymphangioma and another two patients (28.57%) with mixed lymphangioma. There was no microcystic lymphangioma. Furthermore, imaging examinations showed three lymphangiomas located in the supraclavicular, postclavicular regions, and parapharyngeal space, respectively. The remaining four tumors extended to multiple regions in the neck. All seven cervical lymphangiomas (100%) were located adjacent to the carotid sheath, with five (71.43%) located medial to the sternocleidomastoid muscle. We further assessed each tumor's size and found that four (57.14%) lymphangiomas had a maximum diameter  $\geq 10$  cm, and the remaining three had a maximum diameter between 6 and 8.5 cm. Those measurements were based on imaging examinations combined with intraoperative confirmations. Details of the tumor characteristics are shown in Table 1 and representative imaging scans are shown in Figure 1.

### 3.3 | Treatment and follow-up

All lymphangiomas were surgically excised with the surrounding vital structures left intact. Next, negative pressure drainage tubes were placed in each operative cavity for 2–7 days (median duration of 3 days) to prevent postoperative infection. After surgeries, one female found it a little laborious to lift her right arm, and one male experienced regional postoperative neck numbness, but both discomforts disappeared within 1 week. Postoperative pathological examinations revealed that all the tumors were cystic lymphangiomas; no simplex or cavernous lymphangiomas were present in the patients. Follow-up

data were obtained for all seven patients, at intervals of 9–55 months, with a median interval of 32 months. No patient showed evidence of tumor recurrence.

## 4 | DISCUSSION

Lymphangiomas are vascular malformations that mostly occur in the neck. Some researchers have proposed classifying lymphangioma as a congenital pathogenesis of lymphatic tissue.<sup>4,12</sup> Numerous cases of lymphangioma have been reported in the pediatric population since the disease was first described in European literature by Redenbacher in 1828.<sup>1</sup> Pediatric patients <2 years of age account for nearly 90% of cases, because routine prenatal and postnatal imaging examinations contribute to a high of diagnosis, and most symptoms are present before the age of two.<sup>13,14</sup> Most pediatric suffers are treated with sclerotherapy or surgical excision,<sup>14</sup> which might be a reason for the low incidence of lymphangioma in adults. On the other hand, some scholars attribute adult lymphatic malformations to a delayed proliferation of congenital lymphoid RESTs; however, the majority of adult cases are asymptomatic and last an average duration of  $\sim 15$  months, which adds to difficulties in verifying this hypothesis.<sup>8</sup> As lymphangiomas are rarely seen in adults and are less understood in the adult population, we reviewed the medical records of adult patients with cervical lymphangioma to reveal more characteristics of the disease that might help guide its future diagnosis and treatment. In this study, we found that airway or digestive tract constriction was not likely to occur in adult patients with cervical lymphangioma, and complete surgical excision is an effective way to treat the disease.

Most cervical lymphangiomas are unilateral and show no gender or side preference both in pediatric and adult patients.<sup>4,8,15,16</sup> The female predisposition in this study might have resulted from the small sample size. Factors including neck trauma, a prior neck surgery, and cervical inflammation have been considered as potential triggers for the development and exacerbation of lymphangiomas.<sup>4,17–19</sup> However, no worse manifestations occurred in the present patient with a cervical lipoma that had been previously excised. More research is needed to fully reveal the influence of these risk factors. An

asymptomatic mass in the neck is the main complaint of adult patients with a cervical lymphangioma, while other symptoms include difficulty in breathing or swallowing due to obstruction of the respiratory or digestive tract, as well as pain due to infection or compression of the lymphangioma.<sup>1,20,21</sup> Intriguingly, almost all cases involving airway or digestive tract obstruction occur in pediatric patients because lymphangiomas located in the tongue, epiglottis, oropharynx, hypopharynx, and parapharynx are more commonly seen in children.<sup>13,20</sup> Tumors in the oral cavity and masses in the oropharynx result in 75% and 36% of lymphangiomas involving airway obstruction, respectively.<sup>13,20,21</sup> Large lymphangiomas located in other regions of the airway or digestive tract can also cause obstruction due to compressive symptoms. However, dyspnea and dysphagia have been very rarely reported in adult patients.<sup>12,22,23</sup> One reason is that pediatric patients with lymphangiomas located inside the airway or digestive tract have often been successfully treated or the lymphangiomas involving airway obstruction might result in airway infection or even death in a few pediatric patients.<sup>24</sup> Another reason for the uncommon reports of dyspnea and dysphagia is that those symptoms would take a long time to present in adults, because they mainly arise from the development and progressive compression of cervical lymphangiomas located outside the airway or digestive tract.<sup>22,23</sup> Sometimes, even the sudden occurrence of extensive cervical lymphangiomas in adults does not result in dyspnea or dysphagia.<sup>4</sup> Thus, adult patients with cervical lymphangiomas would rarely suffer from life-threatening dyspnea or dysphagia, and mostly require clinical treatment due to the notable enlargement of cervical masses or pain due to compression or inflammation.

Histologically, lymphangiomas can be divided into three subtypes: lymphangioma simplex (capillary lymphangioma), cavernous lymphangioma, and cystic lymphangioma (cystic hygroma),<sup>25,26</sup> in which cystic lymphangioma is the most common subtype.<sup>27</sup> Treatment modalities for lymphangiomas include conservative observation, surgery, sclerotherapy, and medical management, of which surgical excision and sclerotherapy produce better responses.<sup>13</sup> Typically, lymphangiomas are treated surgically or with sclerotherapy. However, due to their proclivity to infiltrate nearby neurovascular structures and muscles, complete excision of lymphangiomas may be unachievable, and the effectiveness of sclerotherapy is commonly limited to macrocystic areas, with a high recurrence rate after treatment: approximately 22%–27% after surgery and approximately 57% after sclerotherapy.<sup>28–30</sup> In this study, all patients presented with a lymphangioma that had infiltrated the nearby carotid sheath. Meanwhile, 71.43% of the lymphangiomas located medial to the sternocleidomastoid muscle. All these situations posed a challenge for the surgeons, as they had to sufficiently expose and excise tumors to avoid missing a hidden or residual portion of the tumor that might lead to a serious recurrence problem.<sup>30,31</sup> The successful excision of tumors infiltrated deep into the interstitial space or adjacent to vital structures requires a highly skilled surgeon. In addition, the precise excision of tumors in the neck may inadvertently cause injury to the surrounding structures and result in lethal comorbidities. Thus, these patients were not only at a high risk for postoperative recurrence, but

also at a certain risk for injury to vital structures. Fortunately, all the tumors were completely excised by surgeons who had extensive experience in head and neck tumor surgery, and no recurrence occurred. Briefly, a complete surgical excision played the pivotal role in threatening this cohort of patients, and the proximity to the critically important structure and infiltrative predisposition of tumors play the primary risk factors for lymphangioma recurrence, due to the increased difficulty in complete tumor removal.

Some physicians recommend waiting for a spontaneous regression of lymphangiomas to avoid potential surgical injuries.<sup>32</sup> The hypothesis underlying spontaneous regression of lymphangiomas is that increased pressure in the lymphatic system might overcome incomplete obstructions or become distributed via alternative lymphatic routes to shrink the tumor masses.<sup>33</sup> However, spontaneous regression is rarely seen and is highly limited. Until now, spontaneous regression has been mainly observed for macrocystic lymphangiomas, and in a few mixed lymphangiomas in patients <12 years old. Only 60.66% of macrocystic lymphangiomas have shown spontaneous regression. In addition, spontaneous regression is often incomplete, and ~22% of recurrent cases require further treatment.<sup>34</sup> On the other hand, sclerotherapy has been popularized and is gradually becoming an adjuvant or alternative treatment for lymphangiomas. The most common drugs used for sclerotherapy are OK-432 (Picibanil) and bleomycin.<sup>35</sup> Previous studies have shown that the efficacy of OK-432 and bleomycin in treatment for lymphangiomas is either better than or comparable to that of surgical excision.<sup>36–39</sup> Furthermore, the complication rate of sclerotherapy performed with OK-432 and bleomycin seems to be lower than that of surgical treatment.<sup>11,40</sup> However, an effective response (shrinkage  $\geq 60\%$ ) to sclerotherapy mainly occurs in macrocystic lesions, and the disease recurrence rate is ~9%. Moreover, microcystic tumors have shown poor response to sclerotherapy.<sup>30,39</sup> Another limitation of sclerotherapy is that nearly all the relevant studies were performed in pediatric patients, and convincing evidence supporting the use of sclerotherapy for treating lymphangiomas in adult patients is still lacking. Because of the less experience of sclerotherapy in treatment for adult lymphangiomas and a 60% shrinkage being hardly accepted in the present patients, of whom the majority of the tumors had a maximum diameter  $\geq 10$  cm, the sclerotherapy failed to be the primary therapy. Thus, the efficacy of spontaneous regression and sclerotherapy heavily depend on the subtype and the specific size of lymphangioma being treated, and that dependency prevents those modalities from becoming recognized as universal and effective treatments. In addition, for patients who cannot tolerate the continuous existence of lymphangiomas, waiting for a spontaneous regression or the effects of sclerotherapy can be unacceptable. However, sclerotherapy might be useful as an adjuvant or alternative treatment when complete surgical excision is unachievable or recurrence occurs. But patients with microcystic or huge lymphangioma are probably experience poor outcomes and following recurrence. There are also some other treatment modalities for lymphangioma, such as radiofrequency ablation, but more research is needed to extend its clinical usefulness due to its limitations in treating tumors located in deep tissue spaces.

This study has some limitations. To begin with, it is a small group of adult patients, and larger-sample studies are needed. Second, all the patients were treated with surgery, and there was no comparator group to compare outcomes with a nonsurgical approach. Therefore, the true effectiveness of surgical and nonsurgical approaches in treating cervical lymphangiomas in adult patients should be further measured in future studies.

## 5 | CONCLUSION

In summary, an experienced surgeon is required to remove lymphangiomas that may infiltrate nearby tissues or adjacent to vital structures in the neck to reduce the risk of surgery and recurrence. The risk factors contributing to the development and proliferation of lymphangiomas in adult patients remain unclear. Therefore, the underlying mechanisms for cervical lymphangioma development in adult patients need to be further elucidated.

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## CONFLICT OF INTEREST

The authors declare no conflict of interest.

## ORCID

Yu Zhang  <https://orcid.org/0000-0002-1543-2629>

Xicheng Song  <https://orcid.org/0000-0002-9789-1318>

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