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Computed Tomography Diagnosis of Cervical Lymphatic Malformations: A Retrospective Study

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Abstract

Objectives: To investigate the imaging findings and diagnostic points of cervical lymphatic malformations on CT plain and contrast-enhanced CT.

Methods: We retrospectively analyzed the clinical and imaging data of 19 patients diagnosed with cervical lymphatic lymphatic malformations in our hospital from May 2018 to October 2023, 8 patients underwent CT plain scanning and 11 patients underwent contrast-enhanced CT.

Results: All 19 patients with cervical lymphadenopathy showed hypodensity in the lesion area, with masses of different sizes, visible unicompartmental and multicompartmental cystic masses, visible segregation within the lesion, and enhancement of the cyst wall and segregation, with no enhancement of the central fluid component.

Conclusions: Computed tomography is able to clearly show the imaging features of cervical lymphatic malformation, which is valuable in diagnosing and guiding the treatment of this disease.

Keywords: Cervical region; lymphatic malformation; X-ray computed; tomography; diagnosis

Abbreviations: CT-Computed tomography; GE-General Electric; kV-Kilovolt; mA-Milliampere; MPR-Multi-plannar reconstruction; %-Percentage

Introduction

Lymphatic malformations, which used to be known as lymphangiomas, are malformations due to abnormalities in the development of lymphatic vessels or genetic abnormalities in specific cells [1], and can be categorized as microcystic, macrocystic, and mixed based on the size of their cysts. Microcystic consists of cysts smaller than 2 cm, macrocystic consists of cysts larger than 2 cm, and the mixed type has cysts of varying sizes. Cystic lymphatic malformations occur more frequently than the other types of lymphatic malformations, and can consist of a single or multiple large cystic lesions [2, 3].To the best of our knowledge, there have been mostly case reports of cervical lymphatic malformations in the literature, and the present single-center study collected cervical lymphatic malformations in a total of 19 patients with more complete clinical, imaging and histopathological data, including age, sex, laterality, medical history and Computed tomography(CT) findings. A case review of all patients was performed to summarize their CT features with the aim of improving the understanding of this disease.

Material and Methods

The retrospective study was approved by the ethics committee of "Beijing Shijitan Hospital" . The informed consent requirement was waived.

Study Population

Patients diagnosed with cervical lymphatic malformation by clinical, imaging and pathology from May 2018 to October 2023 in our hospital were retrospectively analyzed. Inclusion criteria: (1) patients with surgically and pathologically confirmed cervical lymphatic malformation; (2) patients with complete clinical and CT examination data; exclusion criteria: (1) patients with incomplete or poor quality imaging data, such as poor quality CT images; (2) patients with other serious diseases, such as malignant tumors, infections, etc., which may affect the imaging manifestations of lymphatic malformations and interfere with diagnostic results; (3) patients with incomplete clinical and CT examination data. A total of 19 patients were finally enrolled, including 7 males and 12 females, with ages ranging from 11 to 68 years old and an average of 37.7 years old. Clinical symptoms included: 13 patients with neck mass, 2 patients with axillary mass, 5 patients with dyspnea, 2 patients with pleural effusion, 2 patients with pericardial effusion , and 2 patients with abdominal mass, as shown in Table 1.

Table 1: Clinical baseline characteristics in Cervical lymphatic malformation patients (n = 19)

Case	Gender	Age	Medical history[a]
1	Female	18	Cystic mass in neck and abdominal cavity for more than 2 months
2	Female	51	Lower abdominal wall mass for 25 years, pleural effusion on the right side for 16 years
3	Male	59	Left neck mass for more than 30 years
4	Female	68	Neck mass for 5 years
5	Female	42	Swelling of the right upper extremity 1.5 years after resection of right cervical lymphangioma
6	Male	22	Chest tightness, pericardial effusion for more than 1 year
7	Female	52	Chest tightness and shortness of breath for more than 10 months
8	Female	15	Right neck mass for 6 months
9	Male	30	Left neck mass for more than 2 years and gradually increasing in size

10	Female	38	Right neck mass 13 years after surgery, right neck mass 1 week
11	Male	13	Lymphangioma 13 years, with wheezing for 5 years, aggravated for 2 months
12	Female	50	Chest tightness and breath holding for 3 years
13	Male	11	Massive pericardial effusion for 2 months
14	Female	52	Neck mass for more than 6 years, enlarged for more than 3 months
15	Male	22	Multiple swellings throughout the body for more than 10 years
16	Female	31	Chylothorax for 14 years, bilateral neck and axillary masses for 13 years, wheezing for 1 year
17	Female	39	Cystic mass in right neck for 8 months
18	Male	49	Left cervical lymphangioma 23 years after surgery, left cervical mass found for 4 months
19	Female	54	Right neck mass for over 17 years

[a]The same patient may have two or more symptoms

CT Image Acquisition

256-slice Brilliance iCT (Philips Medical Systems), Revolution CT (GE Medical Systems) and SOMATOM Force CT (Siemens Medical Systems) were used, with the following scanning parameters: the patient was lying on their back, with head slightly tilted back so that the neck was parallel to the bed surface, and the two external ear holes were equidistant from the bed surface, and a lateral localized image of the neck was taken, and a CT image was scanned in the region of the localized image selected to be extended from the skull to the root of the neck. The CT image was scanned from the skull to the root of the neck, extending to the upper mediastinum if necessary, with a tube voltage of 80~120kV, a tube current of 250~300mA, a layer thickness of 5mm, and a layer spacing of 5mm, and a non-ionic contrast agent (iodohexol 300mgI/ml) was injected using an UlrichINJECT CTmotion™ (Germany) contrast agent high-pressure injector at an injection rate of 2.0 ~3.0ml/s, the total amount of saline injection was 15ml, and the total amount of contrast agent injection was 60-100ml, the injection site of all patients was the anterior elbow vein, the arterial phase was 30-35s, and the venous phase was 70-80s,with a 1.0 pitch and a matrix of 512×512, the original images were transmitted to the post-processing workstation for multiplanar reconstruction to reconstruct the soft tissue window and the bone window, and the reconstructed layer was 2 mm.

Radiological Evaluation

Two radiologists with diagnostic experience read the films in a double-blind fashion, observing and judging the images, observing the borders of the lesion (clear/unclear), shape (regular/irregular), density of the envelope contents (low/high/moderate), intervals (present/absent), and enhancement (present/absent). Disagreements in the evaluations were resolved by discussion and consensus.

Statistical Analysis

The statistical analyses were performed using IBM SPSS Statistics v26.0(Statistical Package for the Social Sciences,Inc,Chicago,IL,United States).The mean±SD was done as quantitative variables and percentage(%) as qualitative variables.

Results

Radiological Findings

All patients showed hypointensity at the lesion site(Figure1~6), 6 cases of left lymphangioma, 9 cases of right lymphangioma, 4 cases of bilateral lymphangioma, 8 patients underwent CT plain scanning, 11 patients underwent CT plain scanning + enhancement, 4 patients had well-defined lesion boundaries, 2 patients saw segregation within the lesion, 2 patients saw mild enhancement in the cystic wall, and enhancement of the peritoneum and segregation was seen in 1 patient. The image features are listed in Table 2.

Table 2: CT features in Cervical lymphatic malformation patients (n = 19)

Case	Side	Characteristics of lesion	Maximum diameter(mm)	Enhancedment	CT value(HU)
1	Left	Multiple low-density lesions of variable size	17*16*19	—	16
2	Right	Multiple striated nodular hypodense foci	34*19*27	+	33
3	Left	Large cystic hypodense foci with irregular morphology, well-defined borders, and internal segregation	56*67*98	—	19
4	Right	Irregular hypodense shadow with well-defined borders and internal segregation	86*114*134	—	18
5	Right	Cystic low density mass	24*31*32	—	20
6	Bilateral	Cystic low density mass	13*29*34	—	11
7	Right	Cystic low density mass	29*40*28	—	16
8	Right	Oval low density shadow	26*33*85	—	30
9	Left	Multiple mass of cystic hypodensity	160*107*113	—	17
10	Right	Cystic low density mass	15*20*40	—	23
11	Right	Cystic low density mass	25*31*17	—	15
12	Bilateral	Multiple rounded hypodense Shadows	23*14*26	—	19
13	Left	Multiple cystic hypodense shadows	25*30*35	—	17
14	Left	Diffuse low density shadow	65*57*65	+	20
15	Bilateral	Multiple cystic hypodense shadows of variable size	32*46*51	—	21
16	Bilateral	Multiple rounded hypodense shadows	84*119*111	+	18
17	Right	Cystic low density shadow	27*41*41	—	24
18	Left	Shadows	43*51*49	—	38

19	Right	Multiple cystic hypodense shadows	72*85*64	—	22

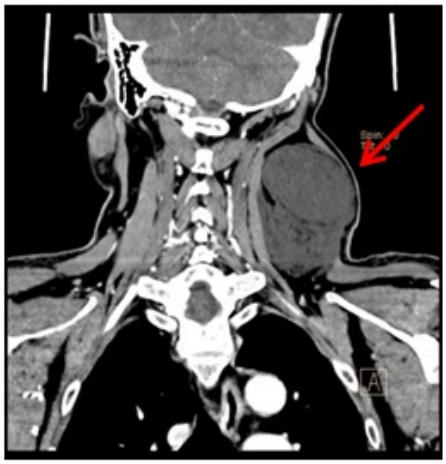


Figure 1: Male, 59 years old.Left cervical mass (arrow), irregular pattern, cystic hypodense shadow with well defined borders, with strips of hypodense segregation, the margin of the lesion is flat on the second cervical vertebral body, and the lower margin is located on the left supraclavicular bone, with compression and displacement of adjacent blood vessels and muscles, with well-defined demarcation.

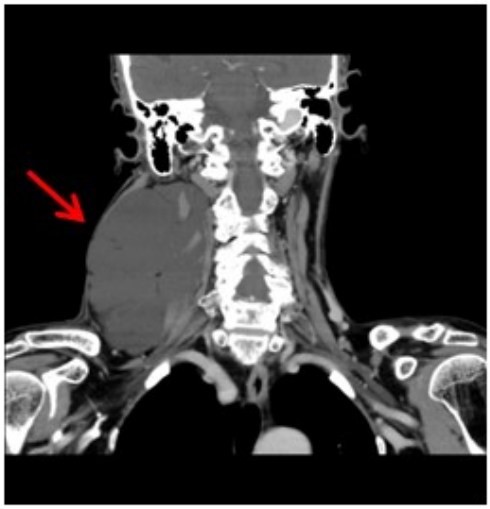
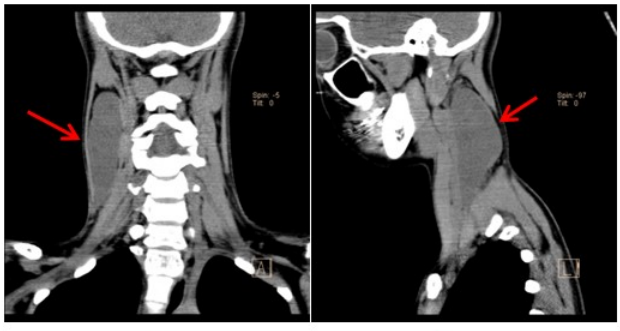


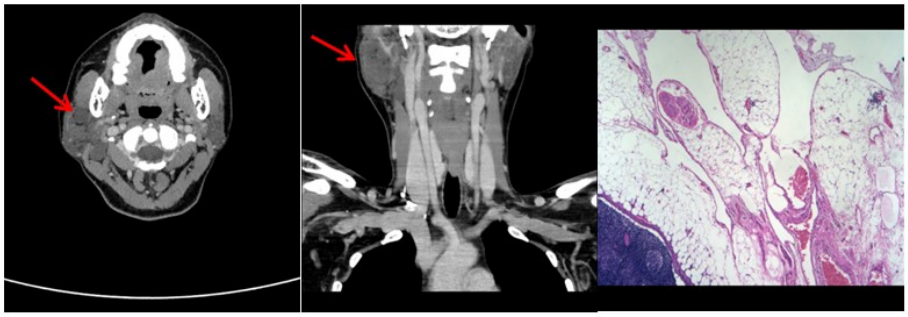
Figure 2: Female, 68 years old. Right neck mass(arrow), irregularly shaped, cystic hypodense shadow with well defined borders.



3A 3B

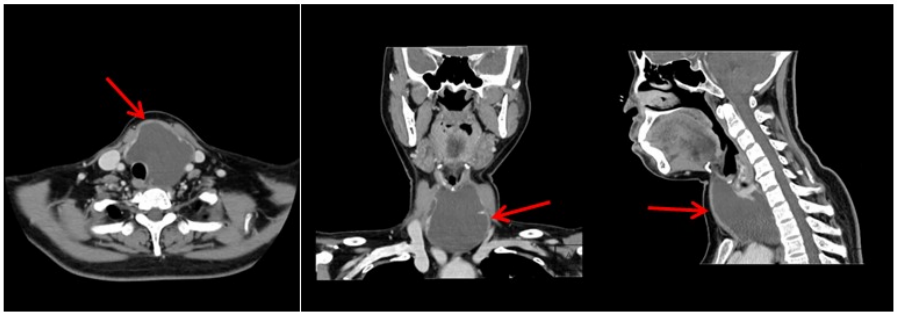
Figure 3A~B: Female, 15 years old.(A)Coronary position.(B)sagittal position.Right neck mass(arrow), hypodense shadow, wel-

l-defined, homogeneous density, adjacent soft tissue compressed and pushed.



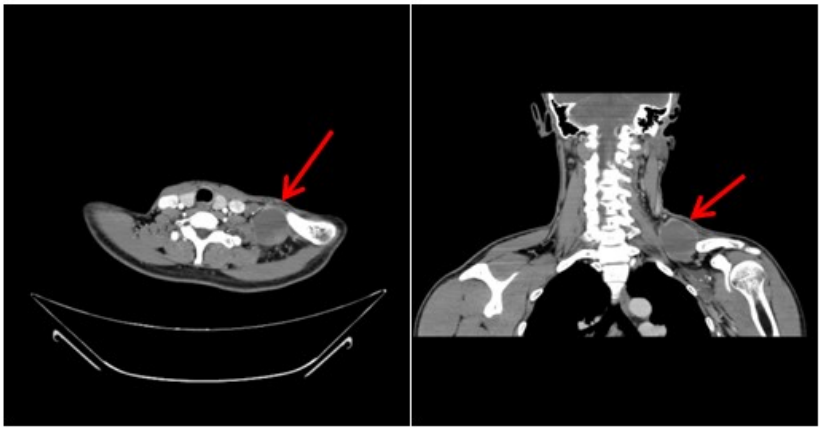
4A 4B 4C

Figure 4A~C: Female, 38 years old.(A)Axial position.(B)Coronary position. (C)Histopathological section.Right neck mass (arrow), irregular pattern, low density foci, multiple small lymph nodes seen in the neck.High magnification photomicrography supported the diagnosis of Cervical lymphatic malformation.



5A 5B 5C

Figure 5A~C: Female,52 years old.(A)Axial position.(B)Coronary position.(C)sagittal position.Low-density shadows are seen in the neck(arrow), and segments are seen in the mass; enhancement scan shows enhancement of the envelope and segments, and no enhancement of the low-density shadows is seen.



6A 6B

Figure 6A~B: Male, 49 years old. (A)Axial position.(B)Coronary position.Left neck mass(arrow), cystic hypodense shadow, CT after direct lymphangiography, , iodized oil residue was seen in the left venous angle area, left neck, left subclavian to axillary.

Histopathological Features

Histopathological examination of the neck lesions in 13 patients, microscopic examination showed a large number of vasodila-

tation in the fibrous tissue and transverse muscle tissue, the lumen was of different sizes and irregular morphology, the thickness of the wall of the tube was different, the lymphocytes were visible in the cystic wall, and the wall of the tube was covered with smooth muscle, and immunohistochemistry of CD34 (+), CD31 (+), D2-40 (+), and Ki-67 (+) was seen in 6 of 13 patients, and in combination with the clinic, The lesions were consistent with lymphatic malformation.

Discussion

Lymphatic malformations can occur at any age or site and usually occur when the lymphatic system fails to connect to the normal jugular veins [4], mainly due to abnormal development of the primitive lymphatic vessels or abnormal proliferation of lymphatic vessel endothelial cells [5], most people are diagnosed at birth, most often in the cervical-facial area, followed by clavicular and axillary regions, the most frequent occurrence of cervical-facial area may be due to the fact that lymphatic system is predominantly in this region dominates the region, which is rich in lymphatic vessels [6, 7], although it is the cervical lymphatic malformations are benign and usually asymptomatic clinically, the most striking sign of the lesion is manifested by a soft, painless and compressible mass, which varies widely in size, very large masses can cause dysphagia and dyspnea, as well as pain due to infection or compression, however, even very small lymphatic malformations have the potential to become clinically important disease because acute infections and hormonal changes can cause significant growth of lymphatic malformations and, as a result, severe aesthetic and functional impairments may occur, including respiratory or gastrointestinal impairments, speech disorders, malocclusion, and loss of vision [3], and they sometimes lead to complications that can be severe and life-threatening [2, 8, 9].

Cervical lymphatic malformations are most commonly seen in children under 2 years of age and are rare in adults [8, 10], almost all cases involving airway or gastrointestinal obstruction occur in pediatric patients, dyspnea and dysphagia in adult patients are rarely reported, in our study a total of 5 patients patients presented with dyspnea symptoms of chest tightness and wheezing, 1 was a male child 13 years of age and 4 were adults. There is no gender preference in lymphatic malformations [1, 7, 11, 12], in most of the reviews, the incidence is equal in male and female ratio [2, 3, 13], in our study, there were 7 male and 12 female patients, this female tendency may be due to the small sample size, which was also reported in the study by Wang J [12] et al. Cervical lymphangiovenous malformations may present as unicompartmental or multicompartmental cysts [4, 14], some scholars [6] have also suggested that lymphangiomas are mostly unicompartmental with a few of unicompartmental type, in our study, 9 patients had multicompartmental cysts, which is in agreement with the literature reports. Literature reports cervical lymphangiomas both in children and adults can be bilateral or unilateral [2, 12, 13], in our study, 6 cases of left sided lymphangiomas, 9 cases of right sided lymphangiomas, and 4 cases of bilateral lymphangiomas, which is in agreement with previous literature reports.

A combination of clinical, radiological and pathological approaches can help in the early diagnosis of this type of lesion.Radiology is playing an increasingly important role in the diagnosis of lymphangiovenous malformations.Various diagnostic methods have been mentioned in the literature.Methods for the evaluation of cervical lymphangiovenous tumors include ultrasound, CT and MRI.CT is the preferred method as it aids in the diagnosis and precise surgical localization [10].For the most accurate surgical evaluation, a CT scanning [6, 7] can be performed to determine the anatomy of the lymphovascular malformation as well as important structures in close proximity to the surrounding area [1, 2], in addition, CT is more advantageous in detecting calcifications [13], especially contrast-enhanced computed tomography, which also detects the condition of the vasculature [9], and plays a crucial role in characterizing the cystic component, determining the extent of the lesion, and guiding the therapeutic decision [16]. CT manifestations of cervical lymphatic malformations include hypodense attenuation of the lesion, masses of variable diameter and size, well defined cystic structures with thin walls and irregular morphology [1, 5, 9, 11, 15, 16], absence of calcification, and visible septations of the lesion [2, 17], and in enhanced CT scans, there is usually a thin, homogeneous wall

around the core that attenuates like water, and in case of infection, a thicker wall and enhanced Enhancement was seen in the cyst wall and septum but not in the central fluid component [1, 15], in our study, all 19 patients showed cystic watery hypodense foci, with variable lesion sizes, the largest mass being 160*107*113 mm and the smallest 17*16*19 mm, 4 patients had well-defined lesion boundaries, 2 patients had septum seen within the lesion, 2 patients had cyst wall enhancement, and 1 Enhancement of the peritoneum and septum was seen in one patient, and no enhancement of the central fluid component was seen, which was consistent with previous reports. The CT values of the lesions varied according to the nature of the contents of the mass [6], and the mean CT value of the lesions in 19 patients was 20.9 HU, suggesting hemorrhage and celiac fluid.

The definitive diagnosis of cervical lymphatic malformations is histopathology, which shows multiple dilated lymphatic vessels [4, 7, 15, 16] with weakened endothelial cells separated by a small amount of mesenchyme, and a stroma that contains moderate to dense aggregates of lymphatic fluid with varying numbers of lymphocytes [9, 15], and in pathologic practice, an immuno Positive histochemistry can distinguish lymphatic malformations from venous malformations [9, 18], in our study, histopathological examination of neck lesions in 13 patients showed microscopic examination of a large number of dilated vasculature in fibrous and transverse muscle tissues, with lumens of varying sizes and irregular morphology, walls of varying thicknesses, lymphocyte aggregates visible in the capsule wall, and smooth muscle on the wall of the vasculature. There were 6 patients with immunohistochemistry CD34 (+), CD31 (+), D2-40 (+), which was consistent with lymphovascular malformation.

Conclusion

The limitations of this study include the limited number of patients enrolled, the limited pathology sampling, and the lack of patient follow-up. Future studies should employ a randomized controlled design with a large sample size.

In conclusion, our case series describes the CT manifestations of cervical lymphadenopathy with certain imaging features, including lesions that are hypointense, variable in size, well-defined borders, single or multiple cystic foci, and enhancement of the cystic wall and septum may be seen on enhancement scans, with no enhancement within. CT examination is highly accurate in the diagnosis of cervical lymphatic malformations and aids in the diagnosis when combined with history, clinical and other investigations, thus greatly facilitating early diagnosis, the development of individualized treatment plans, and the improvement of the patient's prognosis.

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Declarations

Guarantor

The scientific guarantor of this publication is Rengui Wang

Conflict of Interest

The authors have no conficts of interest to declare

Statistics and Biometry

No complex statistical methods were necessary for this paper

Informed Consent

Written informed consent was waived by the Institutional Review Board

Ethical Approval

Institutional Review Board approval was obtained

Study Subjects or Cohorts Overlap

No study subjects or cohort overlap has been reported.

Methodology

- Retrospective
- Cross-sectional study/diagnostic or prognostic study/observational /
- Performed at one institution

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