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Presumed congenital lymphangioma in a dog

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VetRecord Case Reports

PRESUMED CONGENITAL LYMPHANGIOMA IN A DOG: ULTRASONOGRAPHIC AND COMPUTED TOMOGRAPHY FINDINGS, PRESENTATION AND TREATMENT.

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Keywords:	canine, lymphatic tumour, Radiology, Computed tomography (CT), Ultrasonography
Topics:	Imaging, Radiology, Oncology
Abstract:	A 1-year old crossbreed dog was referred for investigation of a mass in the left hind limb with an incisional biopsy consistent with lymphangioma. Computed tomography (CT) revealed a large homogeneous, well-defined mass within the left caudal peritoneal cavity that extended through the vascular lacuna towards the left stifle joint. Ultrasound (US) revealed poorly defined multilocular cystic mass with internal septa of mixed echogenicity and poor colour Doppler flow. Lymphangioma should be considered as differential diagnosis for masses with these imaging characteristics. CT was most helpful in establishing the extent of neoplastic invasion and margins of the mass.

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<p>TITLE OF CASE <i>Do not include "a case report"</i></p>
<p>PRESUMED CONGENITAL LYMPHANGIOMA IN A DOG: ULTRASONOGRAPHIC AND COMPUTED TOMOGRAPHY FINDINGS, PRESENTATION AND TREATMENT.</p>
<p>SUMMARY <i>Up to 150 words summarising the case presentation and outcome (this will be freely available online)</i></p>
<p>A 1-year old crossbreed dog was referred for investigation of a mass in the left hind limb with an incisional biopsy consistent with lymphangioma. Computed tomography (CT) revealed a large homogeneous, well-defined mass within the left caudal peritoneal cavity that extended through the vascular lacuna towards the left stifle joint. Ultrasound (US) revealed poorly defined multilocular cystic mass with internal septa of mixed echogenicity and poor colour Doppler flow. Lymphangioma should be considered as differential diagnosis for masses with these imaging characteristics. CT was most helpful in establishing the extent of neoplastic invasion and margins of the mass.</p>
<p>BACKGROUND <i>Why you think this case is important – why did you write it up?</i></p>

CASE PRESENTATION *Presenting features, clinical and environmental history*

1 A 1-year old neutered female crossbreed dog was referred to the University of Edinburgh
2 Hospital for investigation of a large oedematous mass in the left hind limb.
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6 The patient initially presented to the referring veterinarian for investigation of left hind
7 limb lameness reported by the owner. At the first consultation the dog was not perceived to be
8 lame by the referring veterinarian but a large fluctuant swelling was noted on the medial thigh
9 and left caudal mammary region. The mass was not painful and the remainder of the physical
10 examination was unremarkable. Given no improvement with conservative treatment a surgical
11 exploration with an incisional biopsy was performed. The biopsy was taken from the
12 subcutaneous inguinal area.
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22 When the patient presented for the referral consultation the mass was similar in size,
23 involving the circumference of the leg and extending over the left caudal mammary region.
24 General physical examination, routine haematology and serum biochemistry were unremarkable.
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31 The histopathological findings for the mass lesion were consistent with lymphangioma.

INVESTIGATIONS *If relevant***DIFFERENTIAL DIAGNOSIS** *If relevant*

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34 To investigate the extent of the mass, potential metastases and to guide treatment options a
35 helical-64-slice computed tomographic (CT) study (Somatom Definition AS Siemens, Erlangen,
36 Germany) of the thorax, abdomen and hind limbs was performed under sedation. Scan settings
37 included a pitch of 1.5, tube potential of 120 kVp, reference tube current of 160 mA, slice
38 thickness of 1.5mm, and matrix 512×512. Post-contrast images were acquired within 45 seconds
39 after contrast injection and reconstructed with a low and high frequency algorithms. A bolus of
40 740mg Iodine/kg of non-ionic iodinated contrast medium (Iopamiro, Bracco, Manno,
41 Switzerland) was injected using an injector pump (Bracco, Manno, Italy) through a cephalic
42 intravenous catheter, followed by 10ml of saline solution.
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55 CT revealed a large homogeneous, relatively well-defined soft tissue mass with mean
56 attenuation of approximately 26 HU within the left caudal peritoneal cavity. The mass showed no
57 contrast enhancement. Dorsally, the mass was directly adjacent to the left psoas major muscle,
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1 ventrally to the small and large intestinal loops, and laterally to the abdominal wall. The medial
2 portion of the mass was embedding the caudal portion of the aorta from the level of cranial L5
3 vertebral body caudally (Figure 1A). The mass followed the course of the external iliac artery and
4 then femoral artery exiting the peritoneum through the vascular lacuna (Figure 1D). It extended
5 along the left thigh and was almost circumferential at the level of the stifle joint (Figure 1C). The
6 left medial iliac and popliteal lymph nodes could not be differentiated from the peritoneal and
7 extra-peritoneal portions of the mass respectively. The right medial iliac lymph node was mildly
8 enlarged measuring 8mm and the right popliteal lymph node was within normal limits. The left
9 superficial inguinal lymph node was indistinguishable from the mass and the right one was mildly
10 enlarged measuring 8mm in diameter (Figure 1B). The mass was also seen to extend
11 subcutaneously along the left ventral body wall, from the inguinal region cranially to the level of
12 L4 vertebral body. There was no evidence of displacement or compression of adjacent structures
13 associated with the mass. The remainder of the abdominal organs and all thoracic structures were
14 within normal limits.

15
16 An ultrasound (US) was performed to assess the mass further and guide treatment options.
17 B-mode and Doppler US of the abdomen and hind limb was performed (MyLab Twice Esaote,
18 Genova, Italy) with microconvex and electronic linear array probes. US- guided fine needle
19 aspirates (FNA) of the regional lymph nodes were also obtained uneventfully. US examination
20 revealed poorly defined multilocular cystic masses with internal septa of varying size and
21 thickness (Figure 2A and 2B). The cystic component of the mass was largely anechoic. The solid
22 areas of the mass were of mixed echogenicity with poor colour Doppler flow in the septa (Figure
23 2C and 2D).

24
25 Incisional biopsies of the left superficial inguinal lymph node and surrounding adipose
26 tissue were submitted by the referring practice and examined. The nodes showed ectatic sinuses
27 (Figure 3), and the surrounding adipose was disrupted by a poorly demarcated, infiltrative
28 proliferation of numerous interconnecting empty spaces and channels that were lined by single-
29 layer of normotypic endothelial cells and supported by moderately vascularised collagenous
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stroma (Figure 4). Mitoses were not evident. Infiltrative lymphangioma was diagnosed.

Different treatment options were discussed with the owners and treatment with prednisolone was started. Since then the patient has been seen monthly for follow-up appointments and continues to do well. The mass has slightly increased in size but the patient is free from clinical signs.

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TREATMENT *If relevant***OUTCOME AND FOLLOW-UP****DISCUSSION** *Include a very brief review of similar published cases*

To the authors' knowledge, this is the first report describing computed tomography and ultrasonographic appearance of canine lymphangioma in the veterinary literature. This case also highlights the important role of US and CT in investigating neoplastic lesions in dogs. Treatment options and prognosis are also discussed.

Lymphangiomas are congenital tumours arising from the lymphatic system or acquired anomalies of lymphatic drainage [1]. Congenital tumours most probably represent embryological remnants of lymphatic tissues that either arose from portions of lymph sacs that were sequestered during development or failed to connect to efferent channels [2]. Acquired or secondary lymphangiomas usually develop at sites of chronic lymphatic obstruction, due to radiotherapy, chronic infection or surgery [3]. Lymphangiomas are considered benign tumours, however they may be locally invasive [1]. They tend to have a low metastatic potential and are considered a rare pathology in a variety of animal species and humans [4]. No sex predisposition has been found and dogs and cats of all ages can be affected [4, 5]. The most common primary sites in dogs and cats are areas of the richest lymphatic drainage such as skin and lymphatic tissues although lymphangiomas have also been described in other locations such as urinary bladder, liver, lung, mesentery, kidney, tongue, spleen and mouth [4]. In humans, about 95% of lymphangiomas occur in the head, neck and axilla and the remaining 5% in lung, retroperitoneum, abdominal viscera, mesentery and mediastinum [6].

Due to the variety of radiologic presentations of lymphangioma, histological examination is necessary to confirm the diagnosis [1]. There is limited information about the US and CT appearance of lymphangiomas in the literature. In humans, ultrasonographically, lymphangiomas

1 occur as multicystic masses that may contain internal septations and are largely anechoic as was
2 observed in this case [7, 8]. Few cases of lymphangiomas are described in the veterinary literature
3 [1, 4, 9-19]. Only two case reports have been published in the veterinary literature describing
4 ultrasonographic features [9, 13]. Both described cases of cutaneous lymphangiomas in a mare
5 and a donkey where the mass was classified as a honeycomb structure without a capsule and with
6 cavities containing fluid which is similar to our case. There are a few cases in human medicine
7 where lymphangiomas were described as hyperechoic masses [6, 20]. However, increased
8 echogenicity in sonographic images is most likely related to the reverberations between structures
9 of the lymphatic system and other tissues [6, 20].

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21 Interestingly, the lymphangioma in our case appeared to be involving multiple spaces and
22 its location is atypical and has never been reported before. One case report describes
23 lymphangioma involving a pelvic limb but the ultrasonography did not reveal intraperitoneal
24 extension of this mass [10].

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31 Typically, CT shows lymphangiomas as homogeneous lesions or masses which comprise
32 of both a cystic and solid part [8]. Margins are well-defined and no contrast uptake is observed.
33 Increased attenuation of septa and wall may be seen in some cases [7]. In contrast to the human
34 literature our case did not show the typical cystic appearance. The mass was relatively
35 homogeneous and the cysts were not distinguishable. We speculate that it might be due to very
36 small size of the cysts and resolution of the images which did not allow their visualization.
37 Another theory is that the walls of the separate cysts are very thin and show attenuation which is
38 similar to their content, making them indistinguishable.

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Recent review in human literature revealed that there is no consensus on the treatment of
lymphangiomas [21]. The efficacy of different treatment options is highly variable in incidental
reports. The treatment choice depends on the size of the lesion, anatomic localisation and
potential complications. Radical surgical excision has been suggested as the treatment of choice
[22], however, the location, size and associated adhesions of the mass may preclude complete
excision and lead to a high recurrence rate [23]. Current literature also demonstrates a high level

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of effectiveness for the nonsurgical treatment of lymphangiomas with percutaneous sclerotherapy [5]. Systemic chemotherapy and interferon- α have also been attempted in human patients with extensive inoperable lesions with limited success [24, 25]. Corticosteroid therapy has been an alternative treatment option for patients facing poor surgical outcome or as an adjuvant treatment, and has been shown to be useful in reducing clinical signs [26]. Radiation therapy may also be beneficial in the management of recurring or non-resectable lesions [18].

In limited veterinary literature, there is no consensus on the treatment and only surgical excision [1, 11, 12, 17, 18, 27], radiotherapy [18] and euthanasia [4, 13, 16] have been described. Prognosis is poorly reported and ranges from death [12] to rapid recurrence [1]. In the report of successful radiotherapy treatment, the neoplasm had been excised twice beforehand. Surgical excision in our case was not possible due to the extent of the mass and its very close proximity to the aorta. Radiotherapy was initially considered but given the location and extent of the mass discovered on CT was not pursued further. Other non-surgical therapies proposed in the human literature including: diathermy, cryotherapy, fibrin glue, and percutaneous sclerosants [5] have been discussed but not pursued. Our patient is currently managed medically and at the time of writing the manuscript, progression of the disease with further clinical signs has not been observed.

Both imaging modalities were extremely valuable in the management of this case. CT played an invaluable role in assessing the extent of the disease and its relationship to other organs, whereas ultrasound provided information about the cystic morphology of the mass, which helped determine the appropriate course of treatment.

In conclusion, lymphangioma should be included among the potential causes of fluctuating mass lesions in dogs. In the authors' opinion such masses should be assessed using CT and US examinations. Ultrasonography was crucial in assessing the morphology of the mass and CT was especially helpful in delineating its exact margins. Further studies are needed to assess the incidence of lymphangioma, potential treatment options, recurrence rate and prognosis in affected dogs.

LEARNING POINTS/TAKE HOME MESSAGES *3 to 5 bullet points – this is a required field*

- Lymphangiomas are rare congenital tumours arising from the lymphatic system or acquired anomalies of lymphatic drainage.
- Computed tomography of lymphangiomas may reveal large homogeneous, well-defined mass with poor to no contrast enhancement.
- Ultrasound of lymphangiomas may reveal poorly defined multilocular cystic mass with internal septa of mixed echogenicity and poor colour Doppler flow.
- Lymphangiomas should be considered as differential diagnosis for masses with these imaging characteristics

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41 **FIGURE/VIDEO CAPTIONS** *figures should NOT be embedded in this document*

42 Fig. 1

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44 A- Post contrast image at the level of L5 showing the intraperitoneal and subcutaneous extension
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46 of the mass (between white arrows). The aortic bifurcation is shown with black arrow.
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49 B- Post contrast image at the level of coxofemoral joints showing the subcutaneous extension of
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51 the mass (between white arrows) and right superficial inguinal lymph node (asterisk).
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54 C- Post contrast image at the level of the stifle joint showing nearly circumferential character of
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56 the mass (between white arrows).
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59 D- Post contrast image in dorsal plane showing the neoplastic extension (between arrows) along
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1 the caudal vena cava (C), aorta (A) external iliac and femoral arteries exiting the peritoneum
2 through the vascular lacuna.
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6 Fig. 2

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8 US images showing the multilocular septa (A, B) and multiple cysts with anechoic fluid
9 (asterisk). Images C and D are at the level of the aortic bifurcation and show the mass extension,
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Fig. 3

Lymph node with markedly ectatic sinuses.

Fig. 4

Adipose tissue disrupted by irregular shaped lymphatic structures lined by a single-layer of
endothelial cells and supported by collagenous stroma (H+E).

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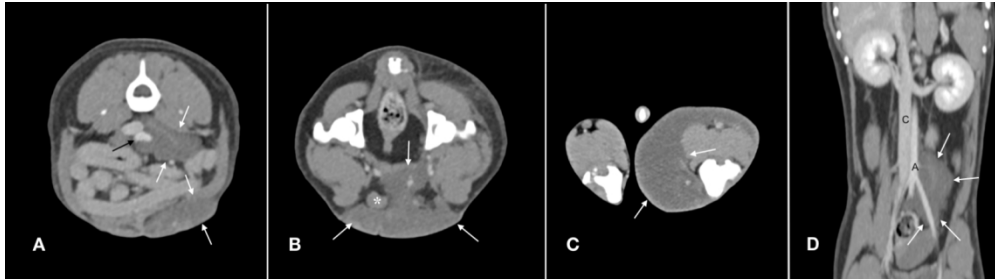
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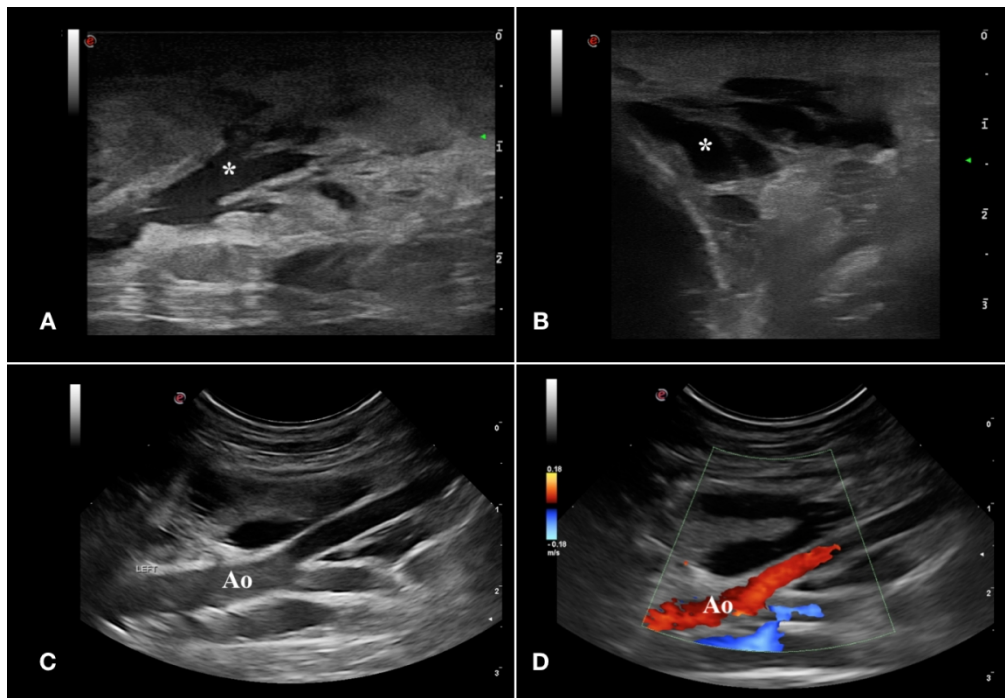
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- A- Post contrast image at the level of L5 showing the intraperitoneal and subcutaneous extension of the mass (between white arrows). The aortic bifurcation is shown with black arrow.
- B- Post contrast image at the level of coxofemoral joints showing the subcutaneous extension of the mass (between white arrows) and right superficial inguinal lymph node (asterisk).
- C- Post contrast image at the level of the stifle joint showing nearly circumferential character of the mass (between white arrows).
- D- Post contrast image in dorsal plane showing the neoplastic extension (between arrows) along the caudal vena cava (C), aorta (A) external iliac and femoral arteries exiting the peritoneum through the vascular lacuna.

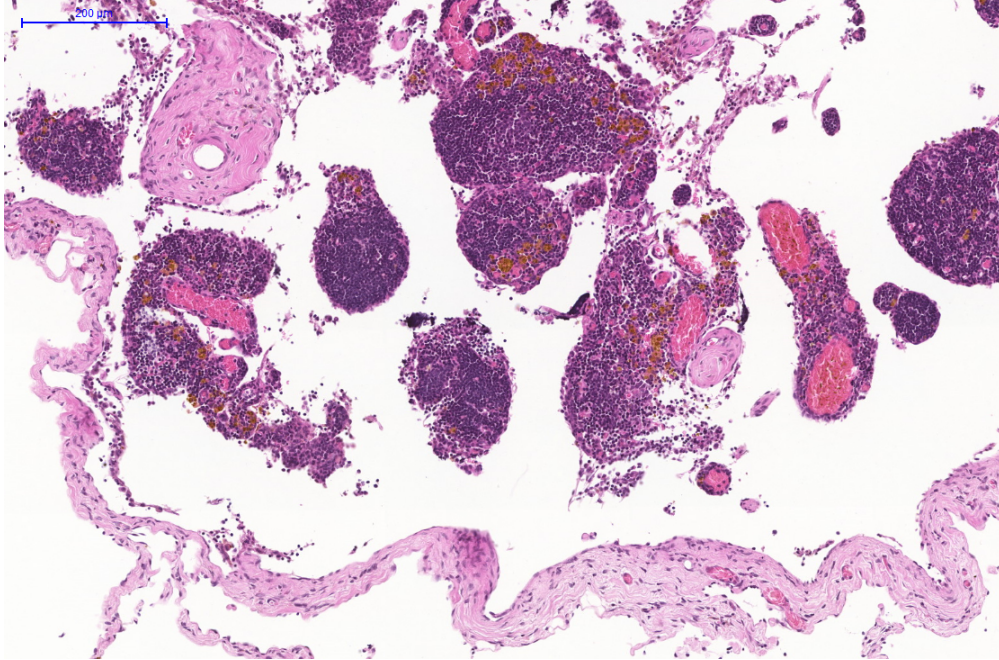
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US images showing the multilocular septa (A, B) and multiple cysts with anechoic fluid (asterisk). Images C and D are at the level of the aortic bifurcation and show the mass extension, poor margination and poor colour Doppler flow within its solid portion.

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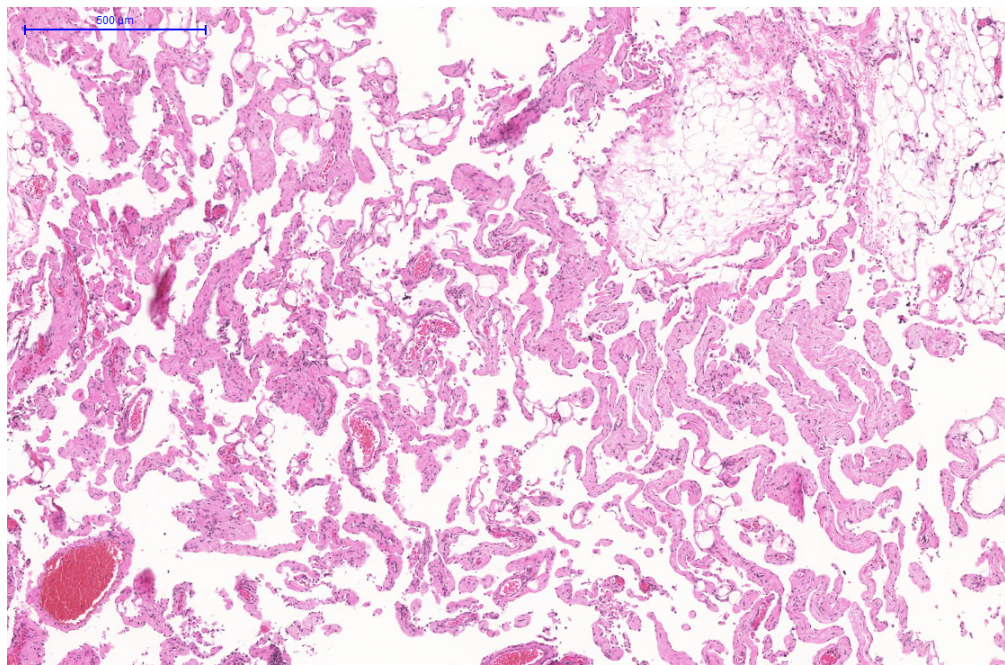


Lymph node with markedly ectatic sinuses.

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Adipose tissue disrupted by irregular shaped lymphatic structures lined by a single-layer of endothelial cells and supported by collagenous stroma (H+E).

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