血管淋巴樣增生併嗜伊紅血球增生症—病例報告

Angiolymphoid Hyperplasia with Eosinophilia

中國醫藥大學附設醫院　口腔顎面外科

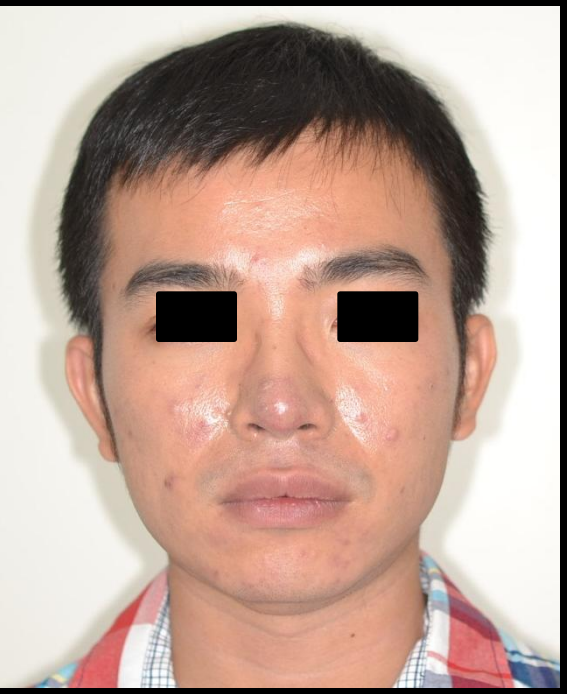
報告者—簡杏宜

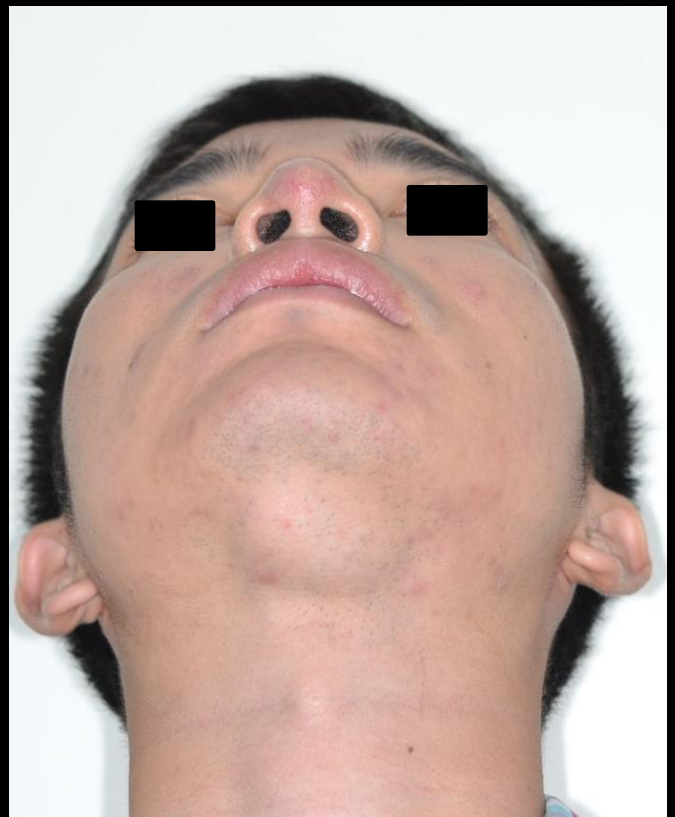
指導者—張加明醫師　薛水上醫師

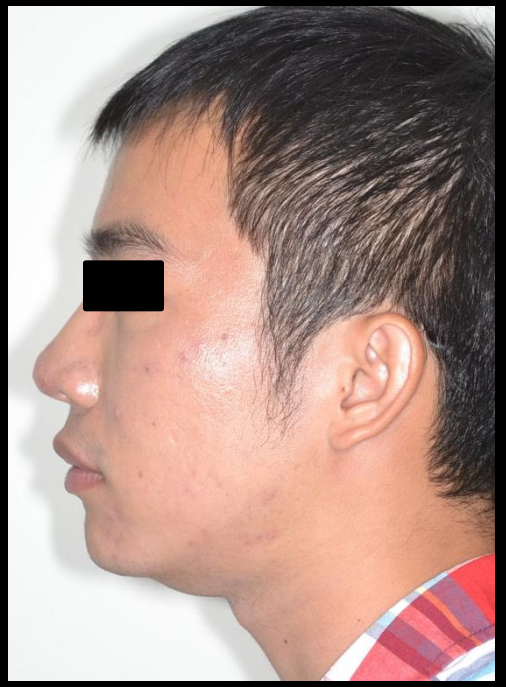
Basic Data

- **Name：黃XX**
- **Gender：male**
- **Age：28 y/o**
- **就診日期: 2011.07.08**
- **Chief complaint**
  - –Painless mass over submental area for two months.

Physical Examination

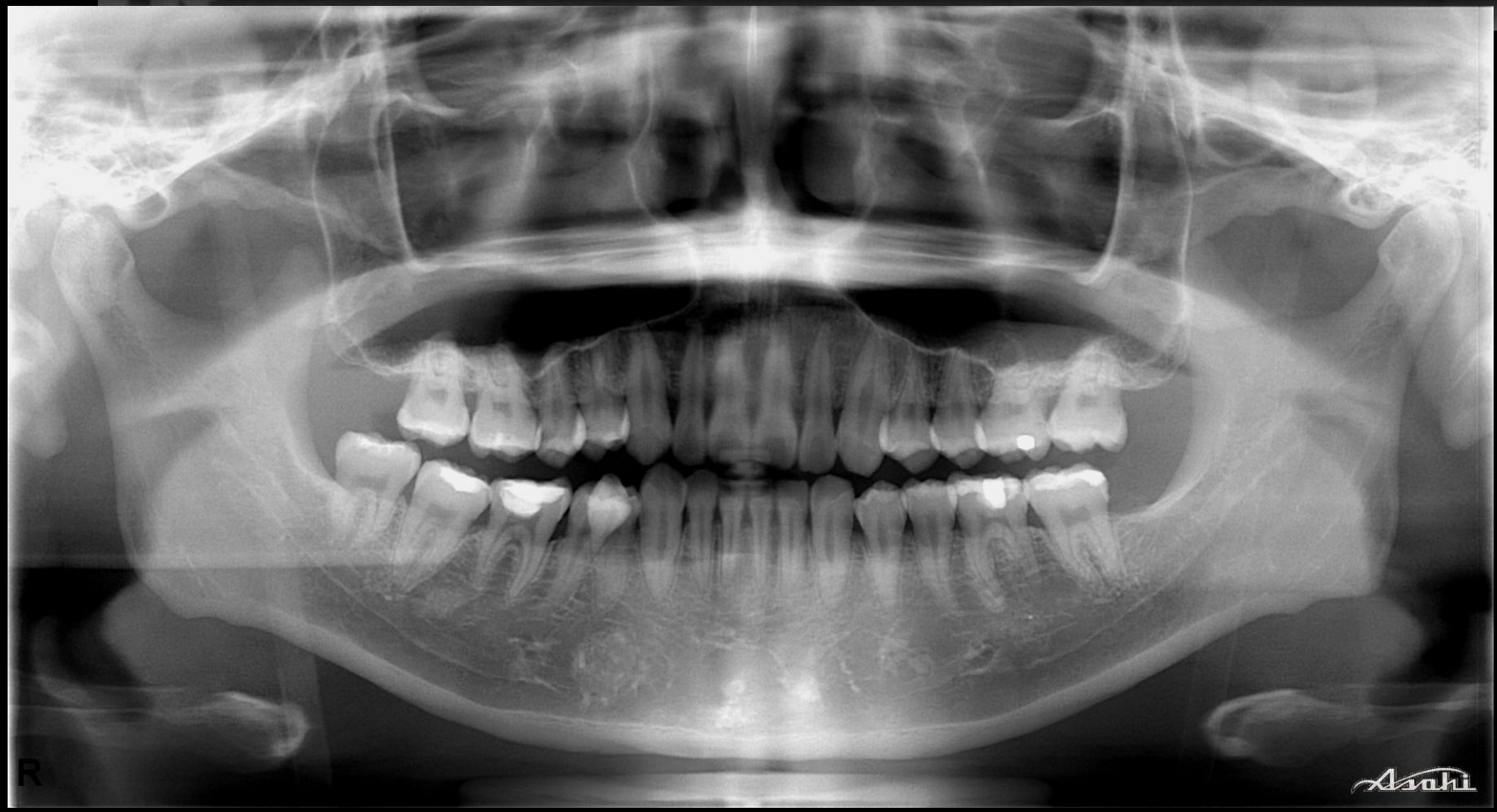






2011/07/08

Radiographic Findings



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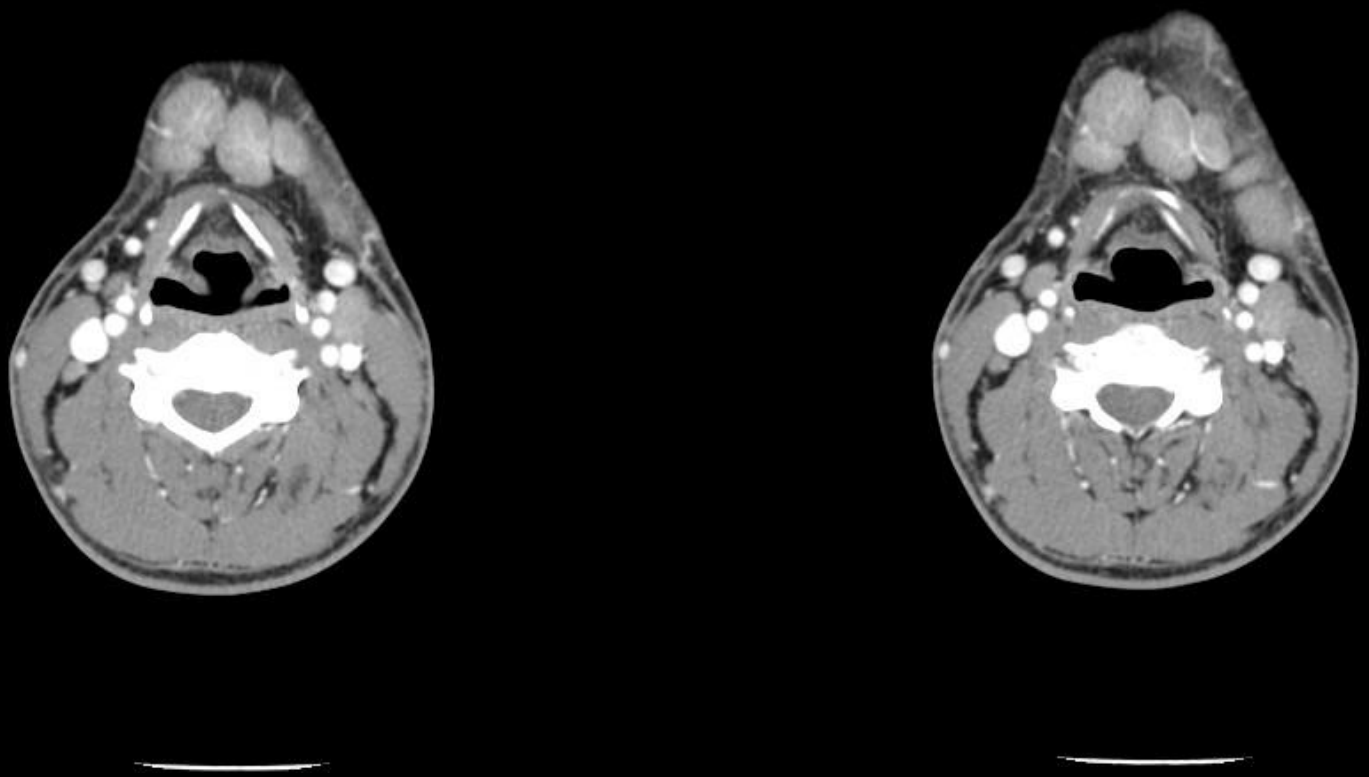
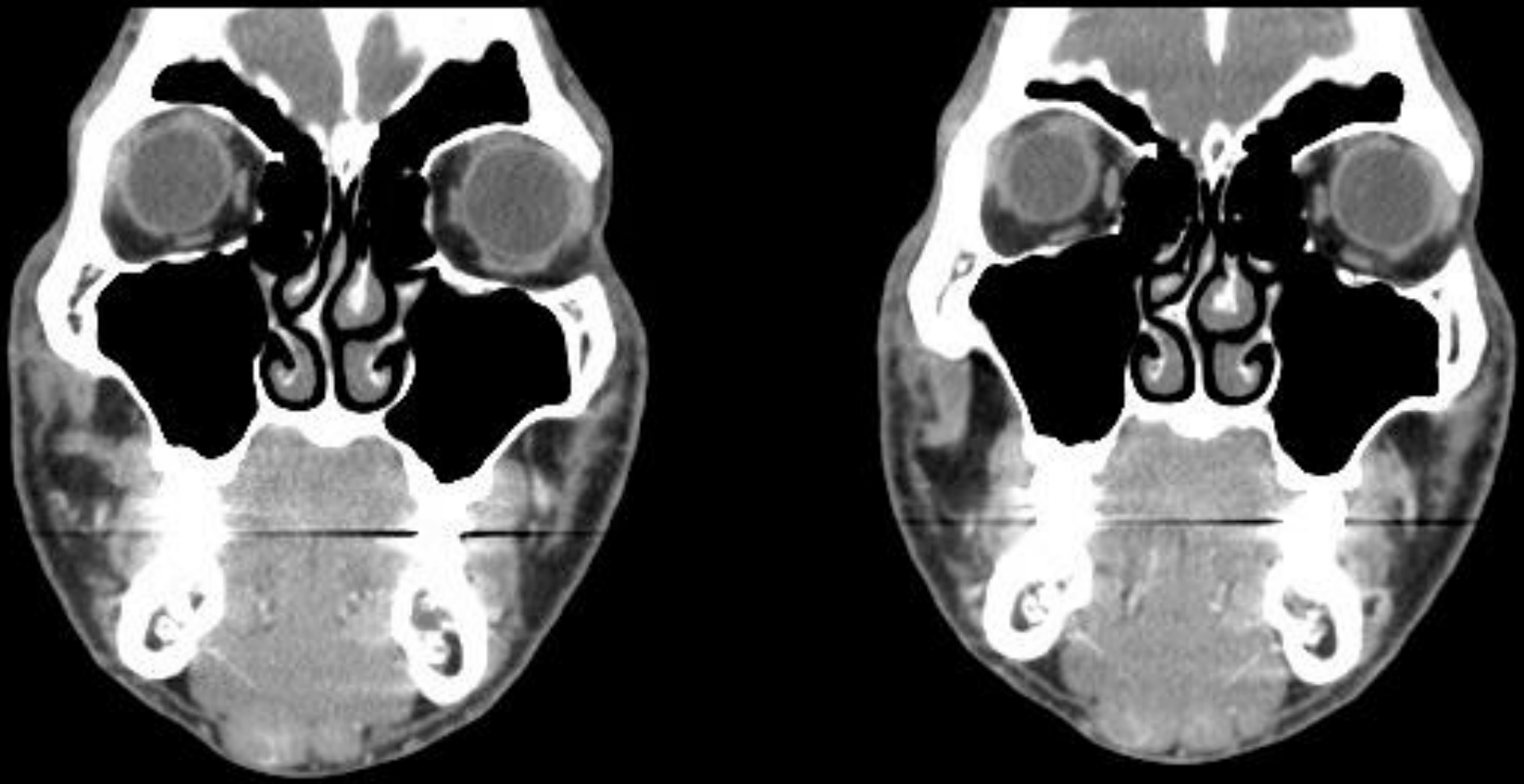
Radiographic Findings



2011/07/29

Radiographic Findings



2011/07/29

Blood Data

- ANA (-)
- IgG : 1540 mg/dL (751 – 1560)
- IgE : 78.13 IU/mL (<165)
- Eosinophil : 1.7 % (0 -7)
- **C3 : 87.2 mg/dL (79 - 152)**
- **RA : 33.5 Iu/mL (< 20 negative)**
- **Creatinine: 0.89 (0.5 ~ 1.3)**

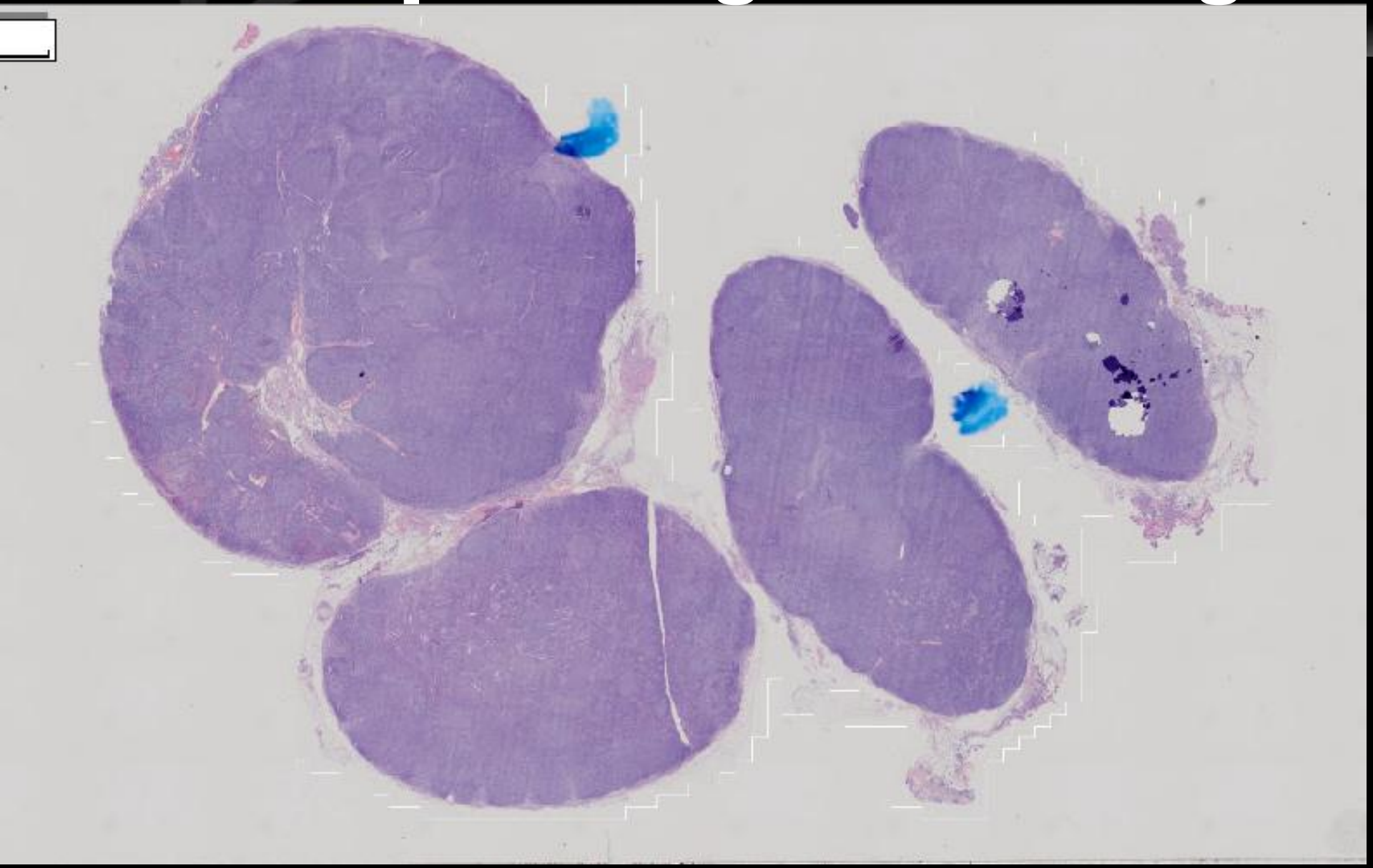
- **Clinical impression:**
  - **Angiolymphoid hyperplasia with eosinophilia**
  - **Kimura's disease**
  - **Lymphoma**

- **Treatment :**
  - **Excisional biopsy of the submental mass at the submental area under GA**

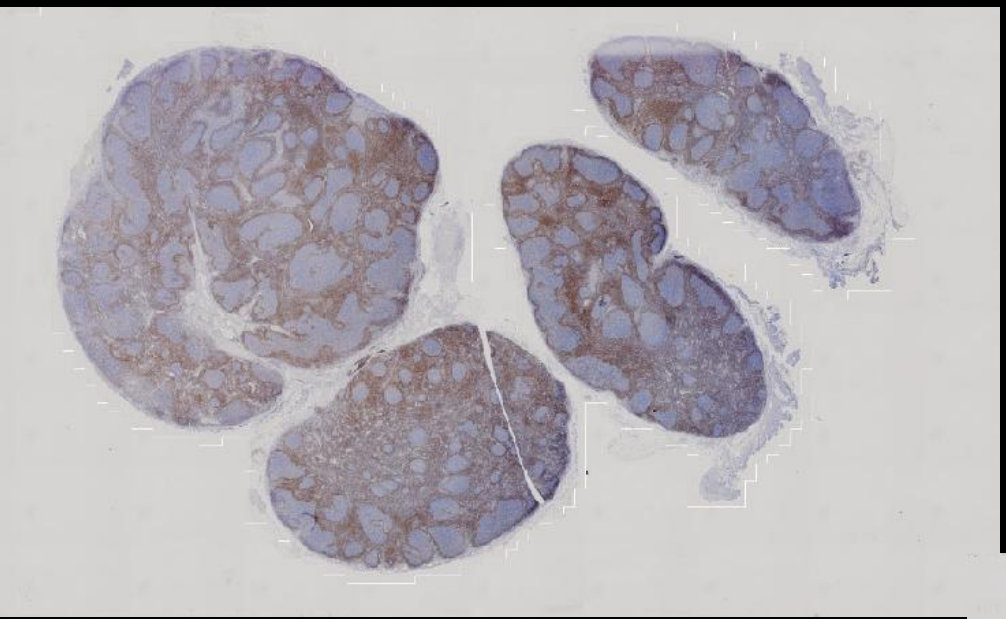
Operation

- Submental incision
- Excision of the submental mass

Histopathological Findings

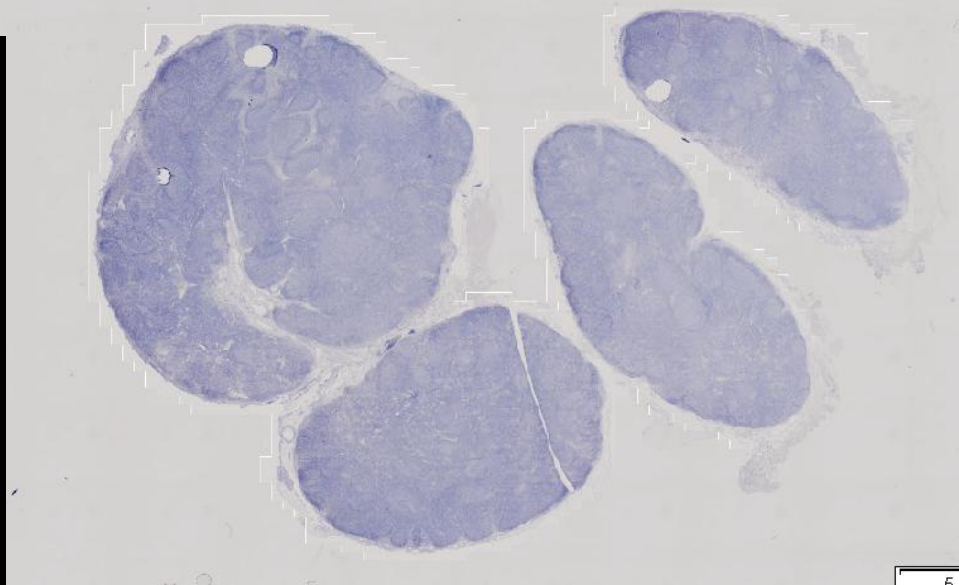


Histopathological Findings

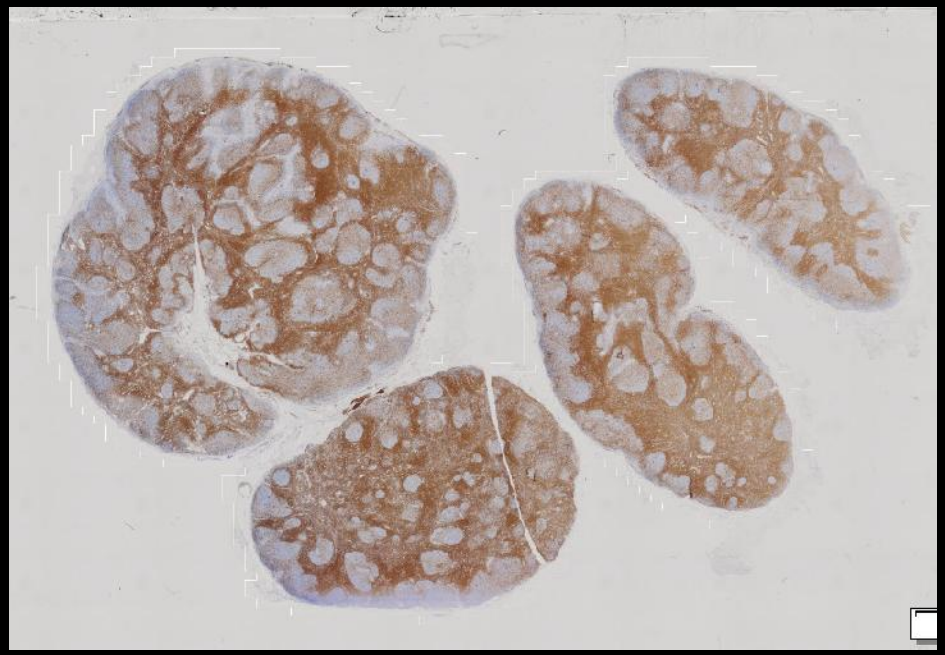


Bcl 2(-)

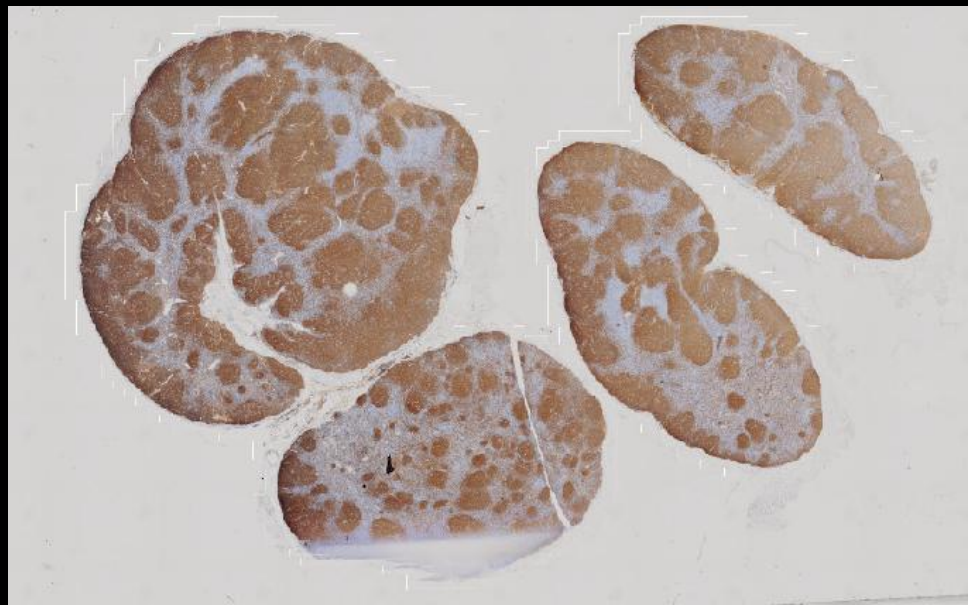
Cyclin D(-)



Histopathological Findings

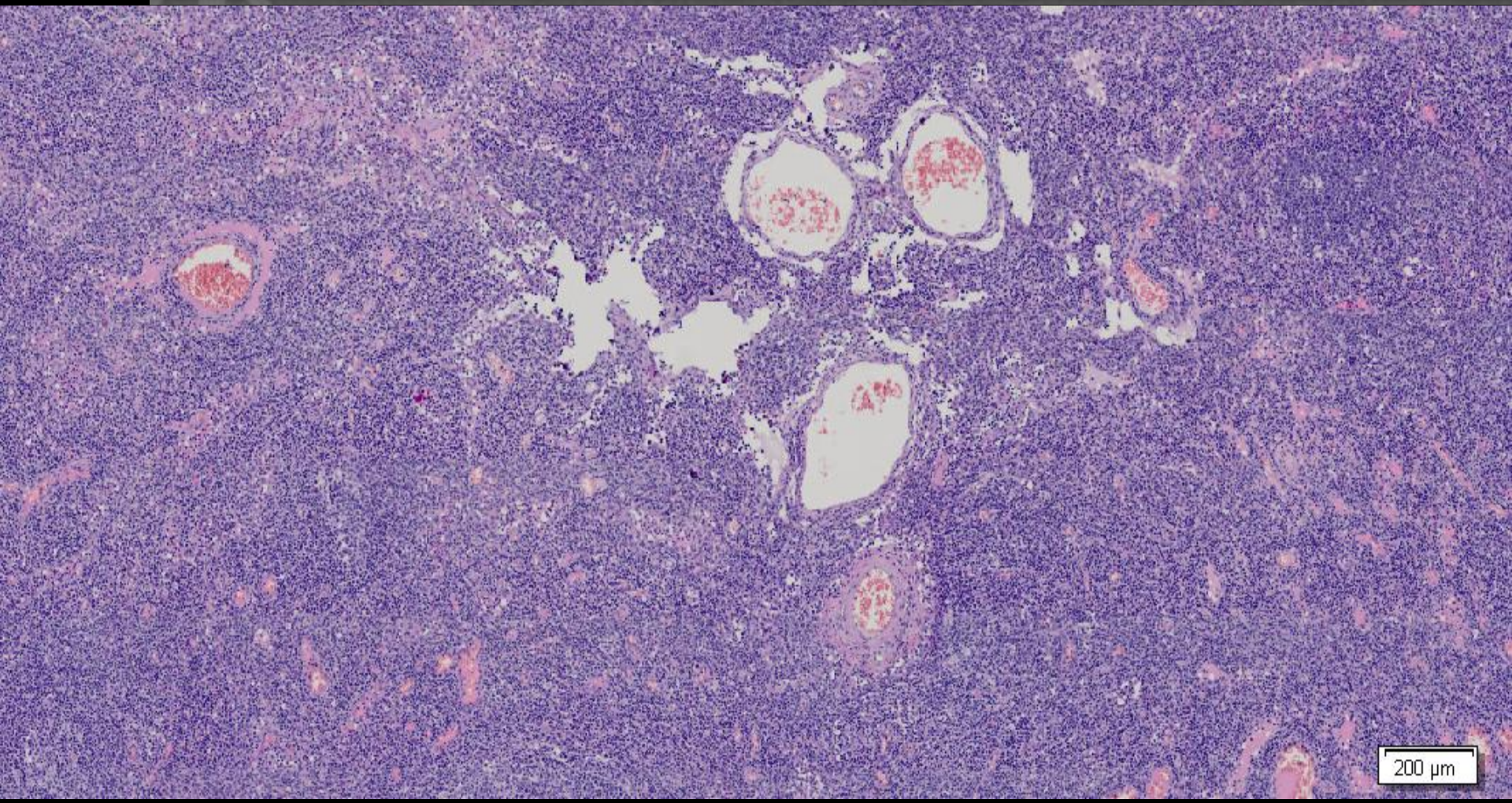


CD 3, T cell

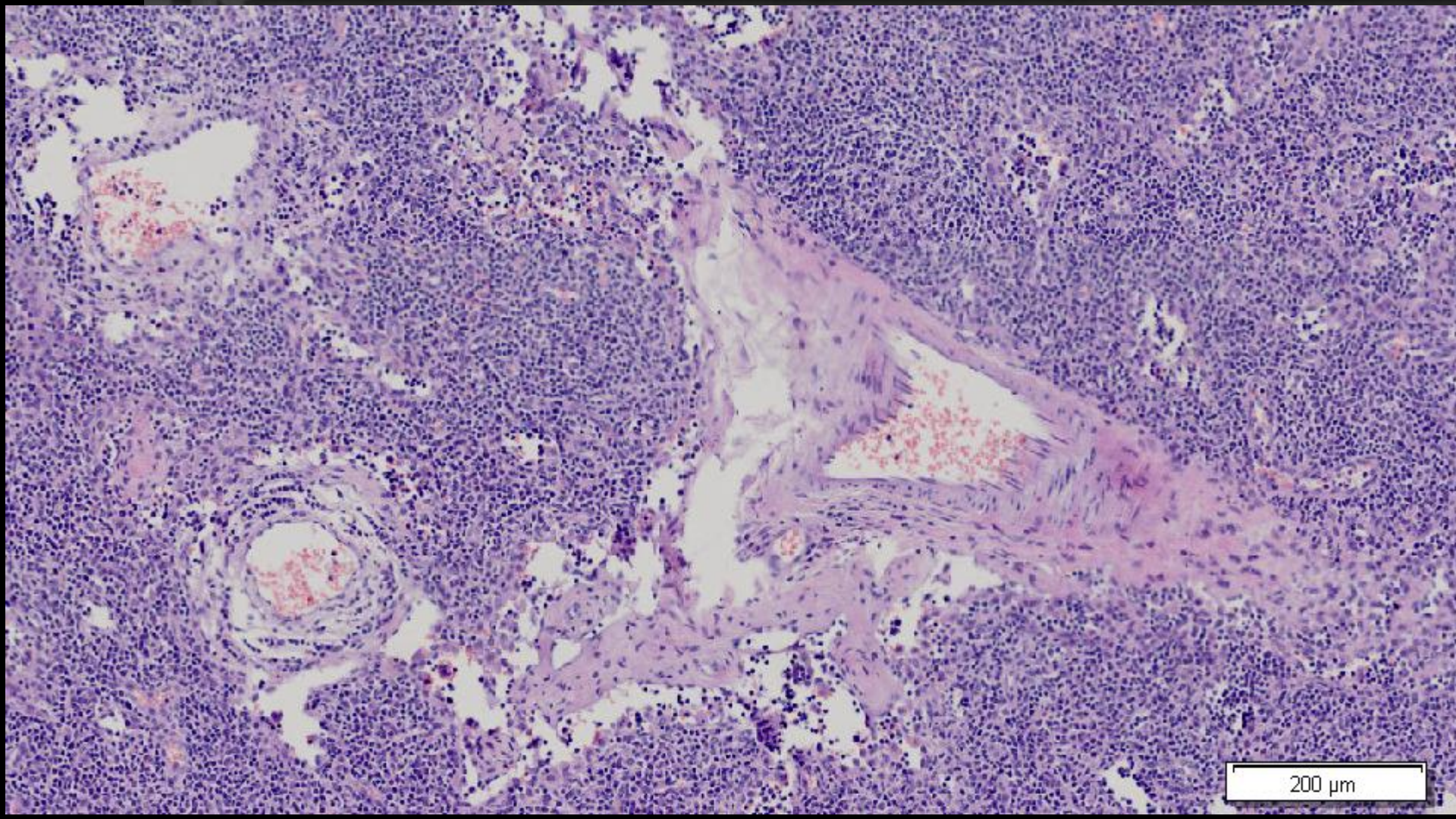


CD20 , B cell

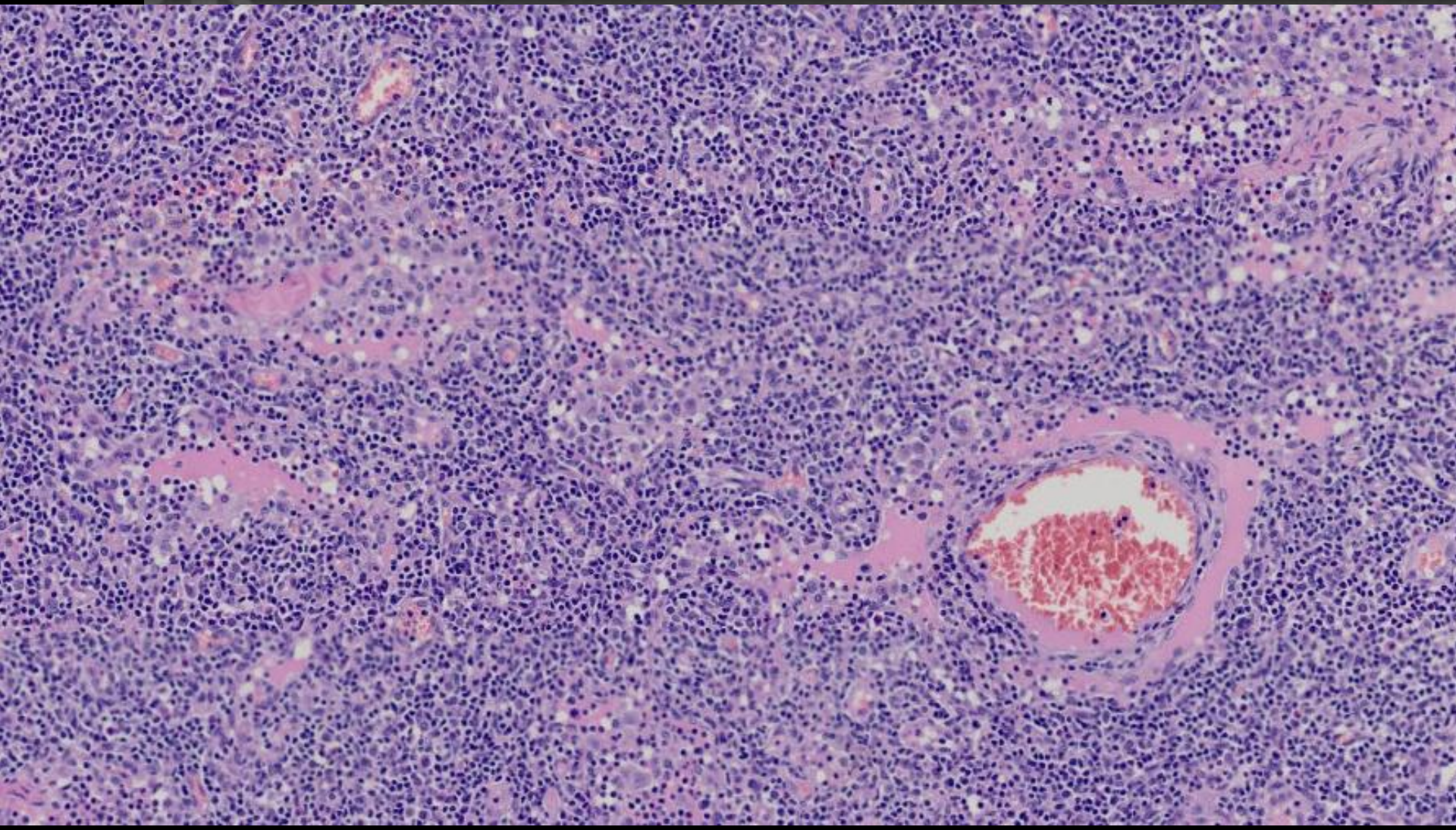
Histopathological Findings



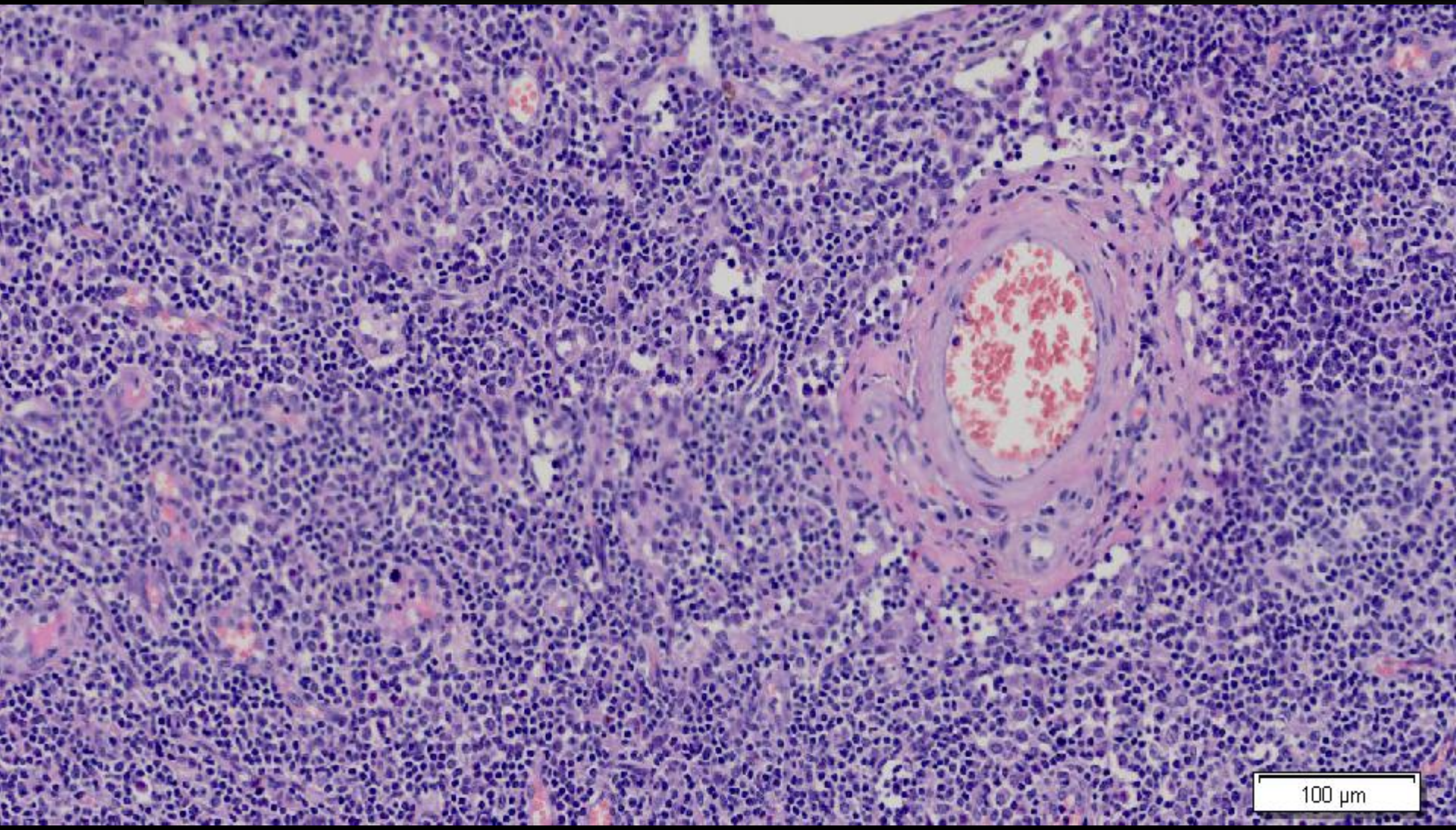
Histopathological Findings



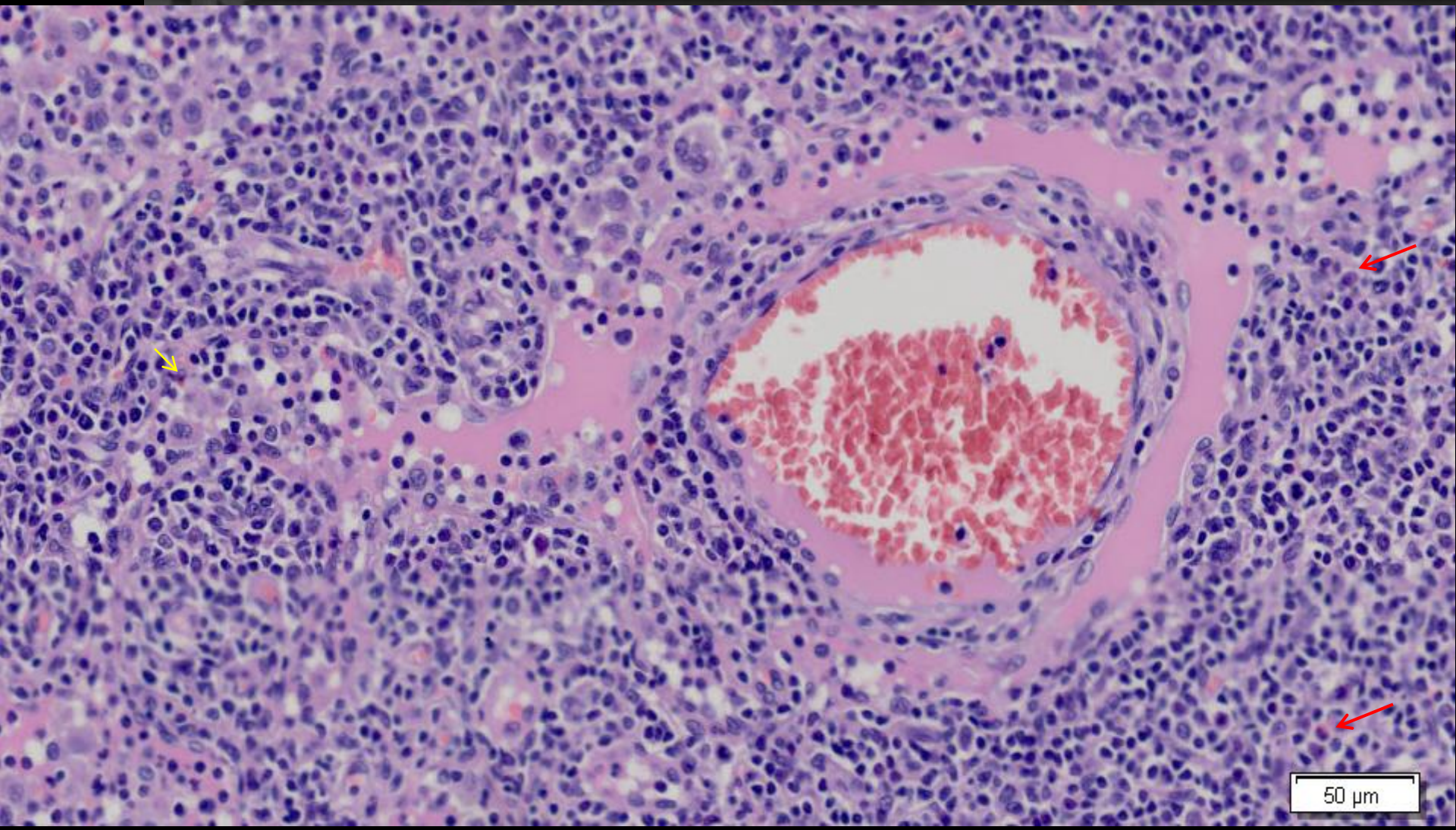
Histopathological Findings



Histopathological Findings



Histopathological Findings



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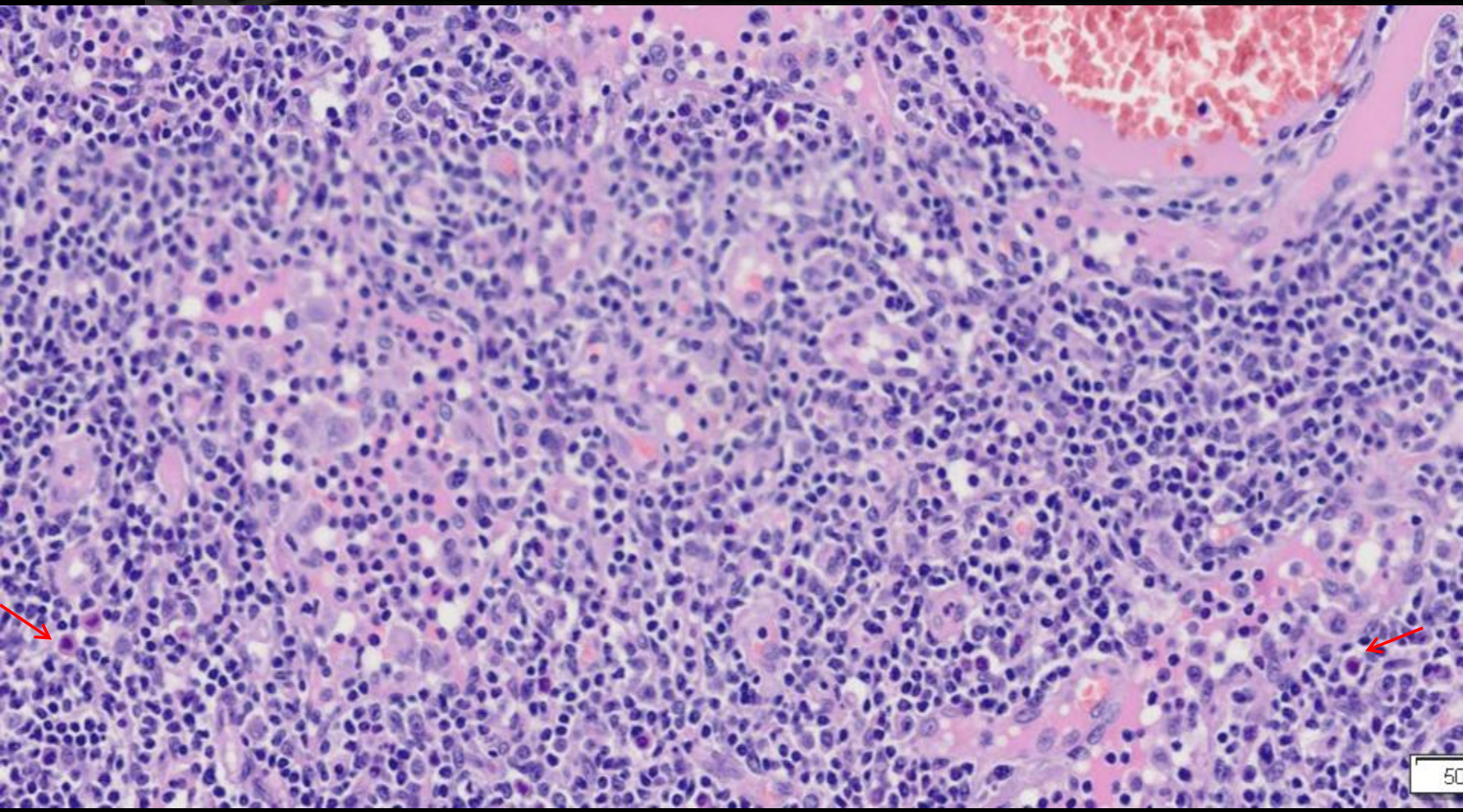
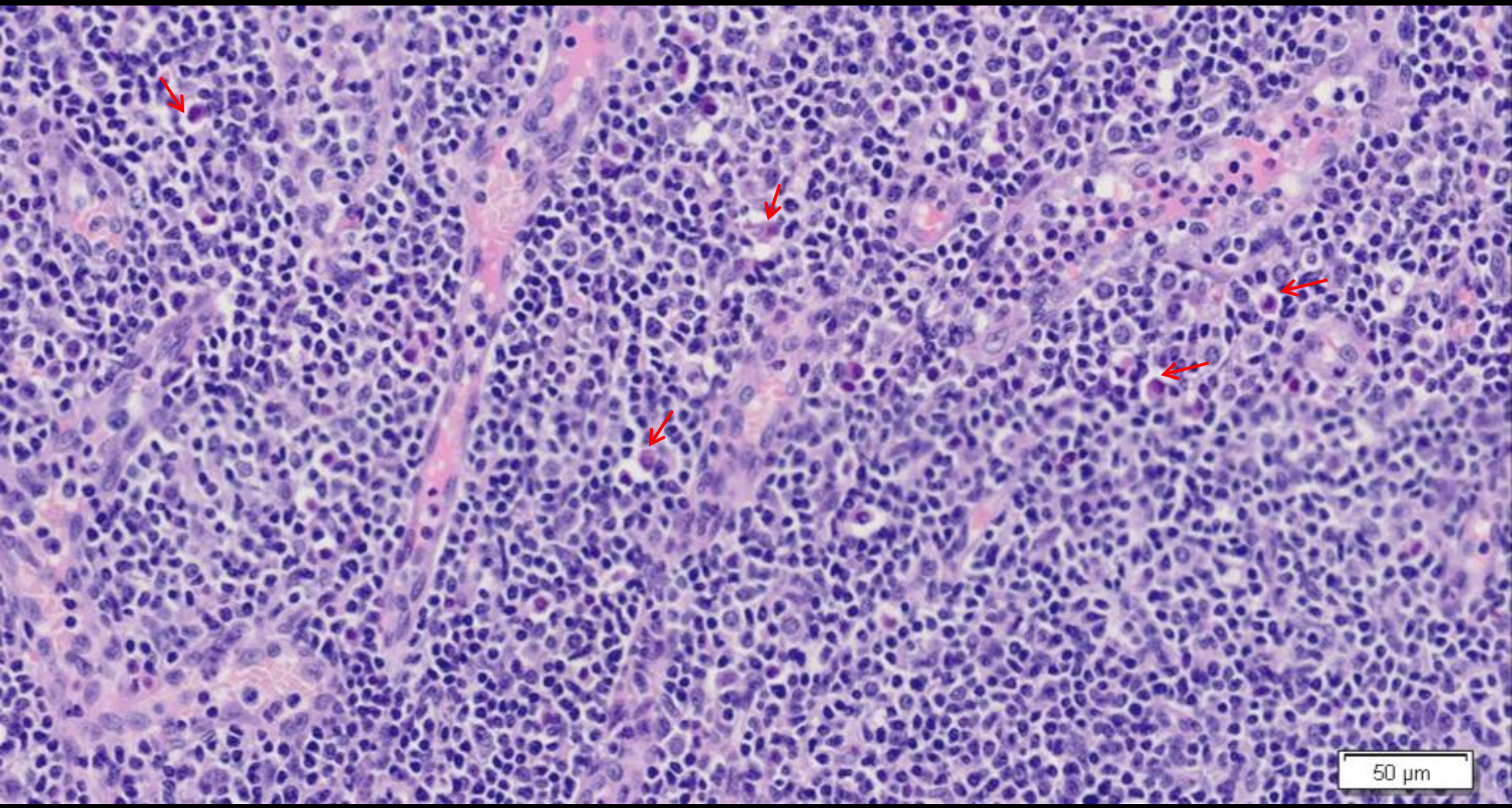
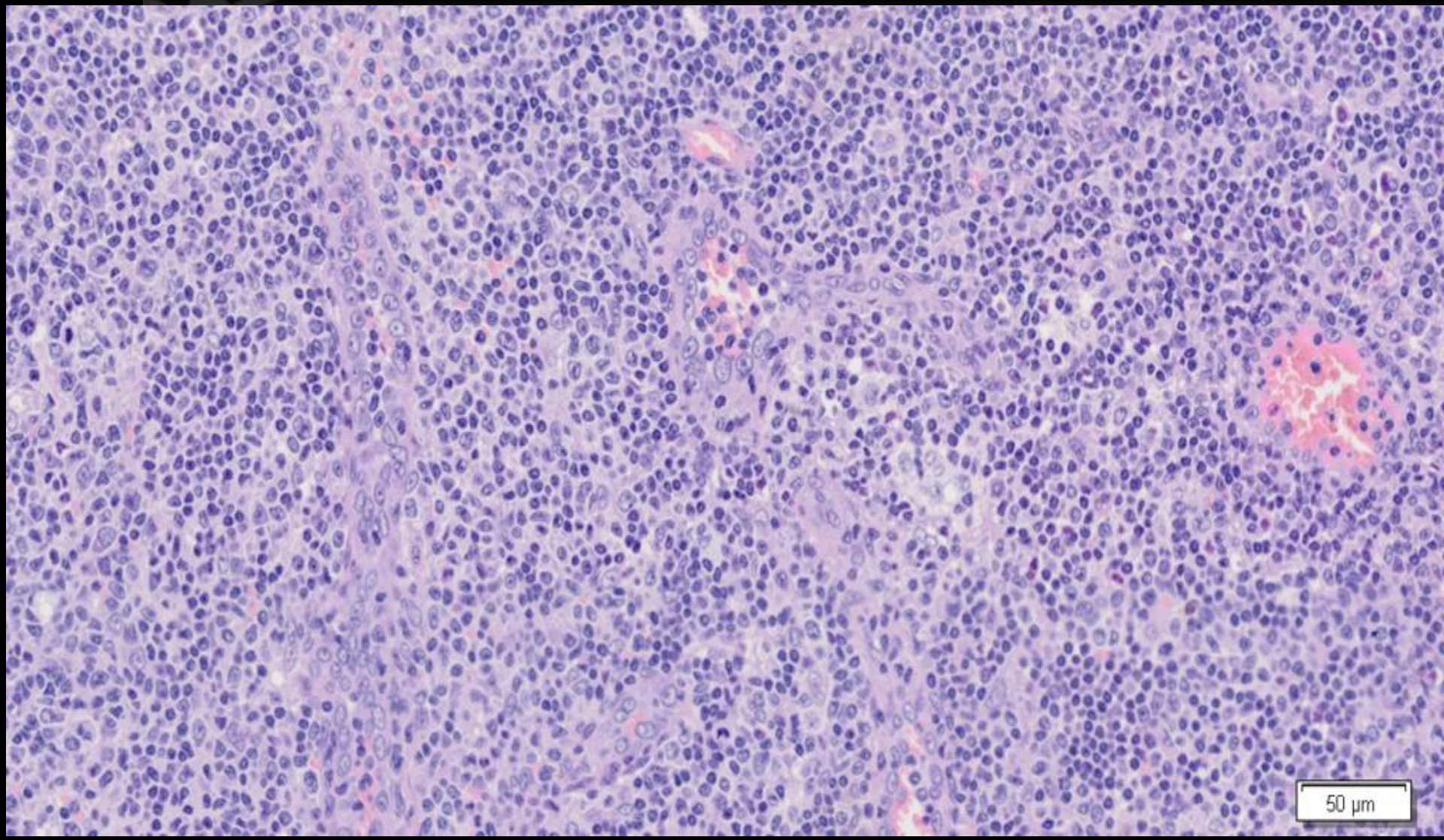


Endothelial cells are plump with abundant eosinophilic cytoplasm. The endothelial cells are surrounded by a mixed of lymphoid cells and eosinophils

Histopathological Findings



Histopathological Findings



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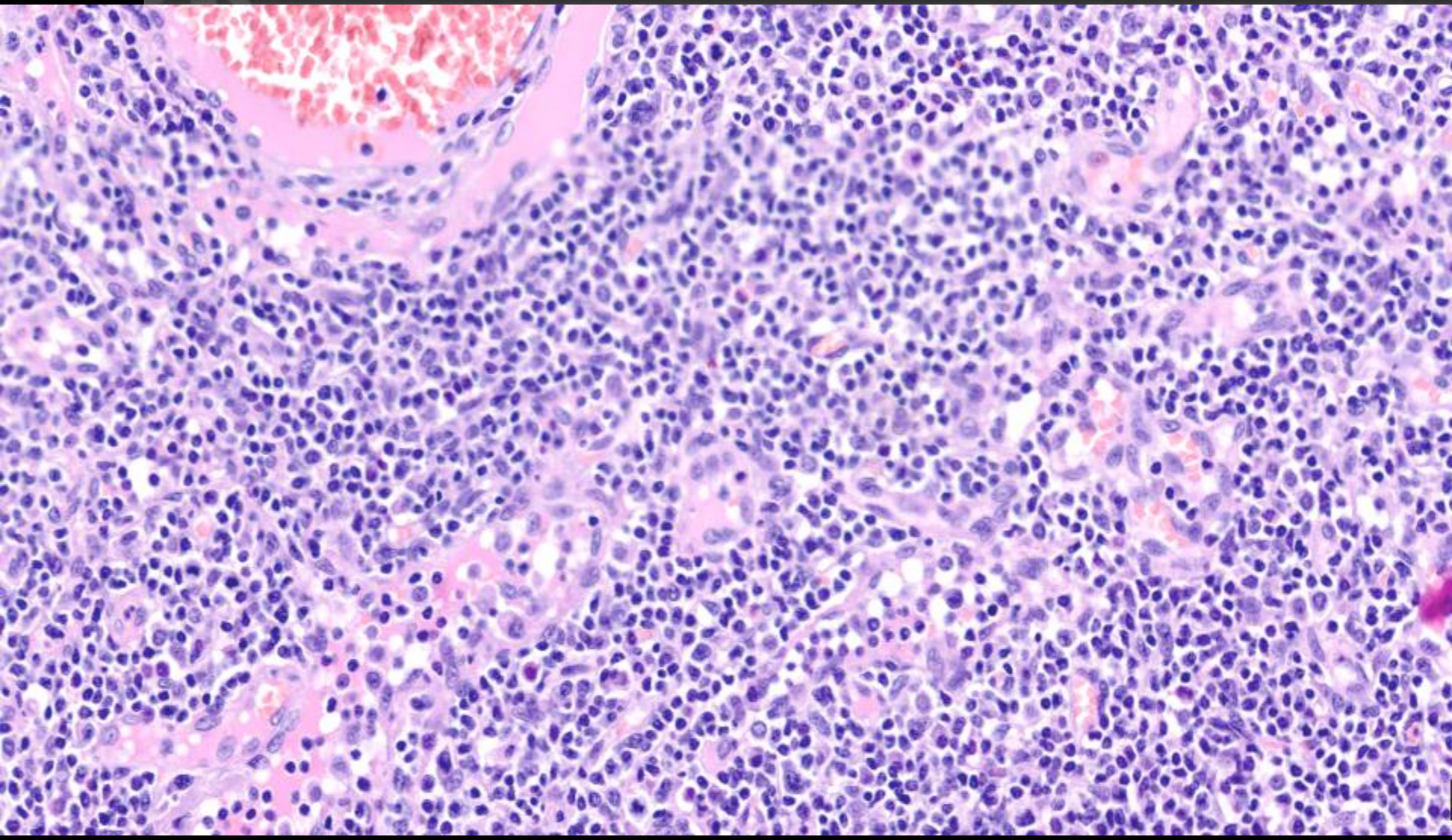
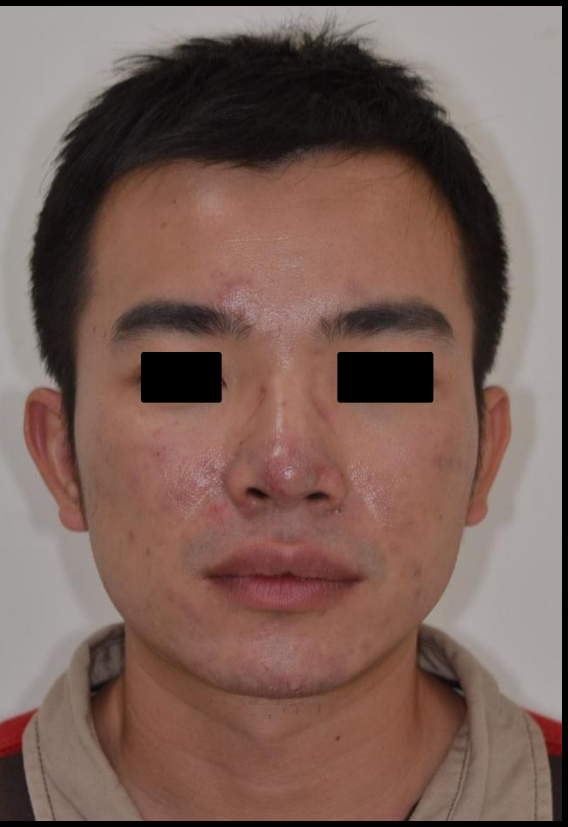
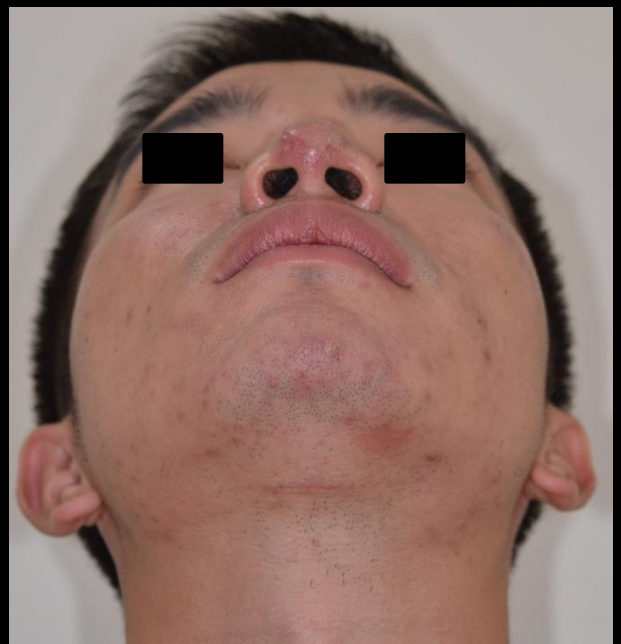


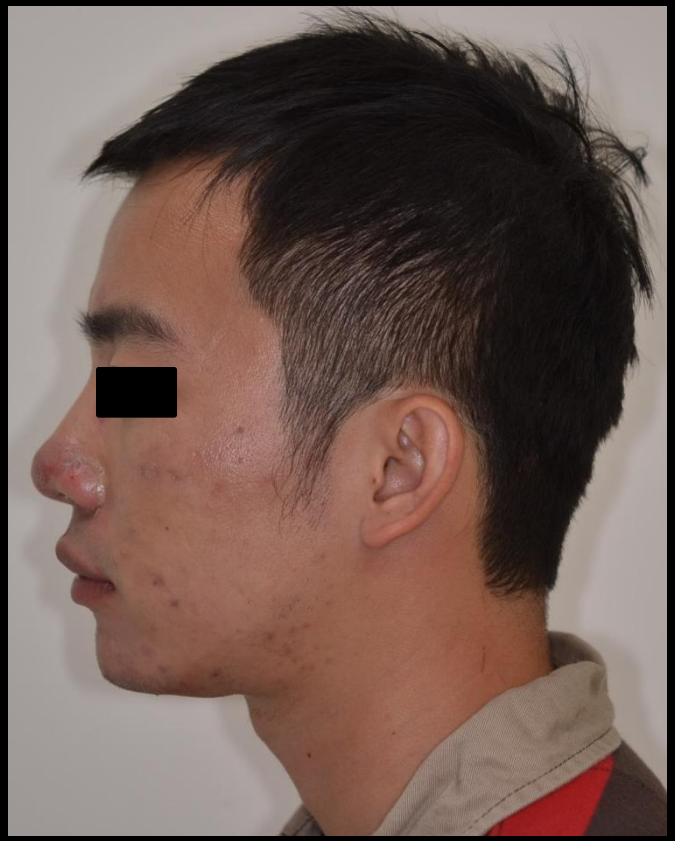
Diagnosis

- Angiolymphoid hyperplasia with eosinophilia (ALHE)

Post-op 3 months F/U







2011/12/16

DISCUSSION 1

Introduction of the Disease

Terminology

- Subcutaneous angiolymphoid hyperplasia with eosinophilia

  Wells and Whimster in 1969

- Epitheloid hemangioma or histiocytic hemangioma

Differential Diagnosis

- Kimura's Disease
  - Chronic, allergic inflammation
- Lymphoma
  - Bcl2(-), cyclin D (-) → reactive lymphoid hyperplasia, not a lymphoma
  - CD3, CD20 → T cells and B cells distribution

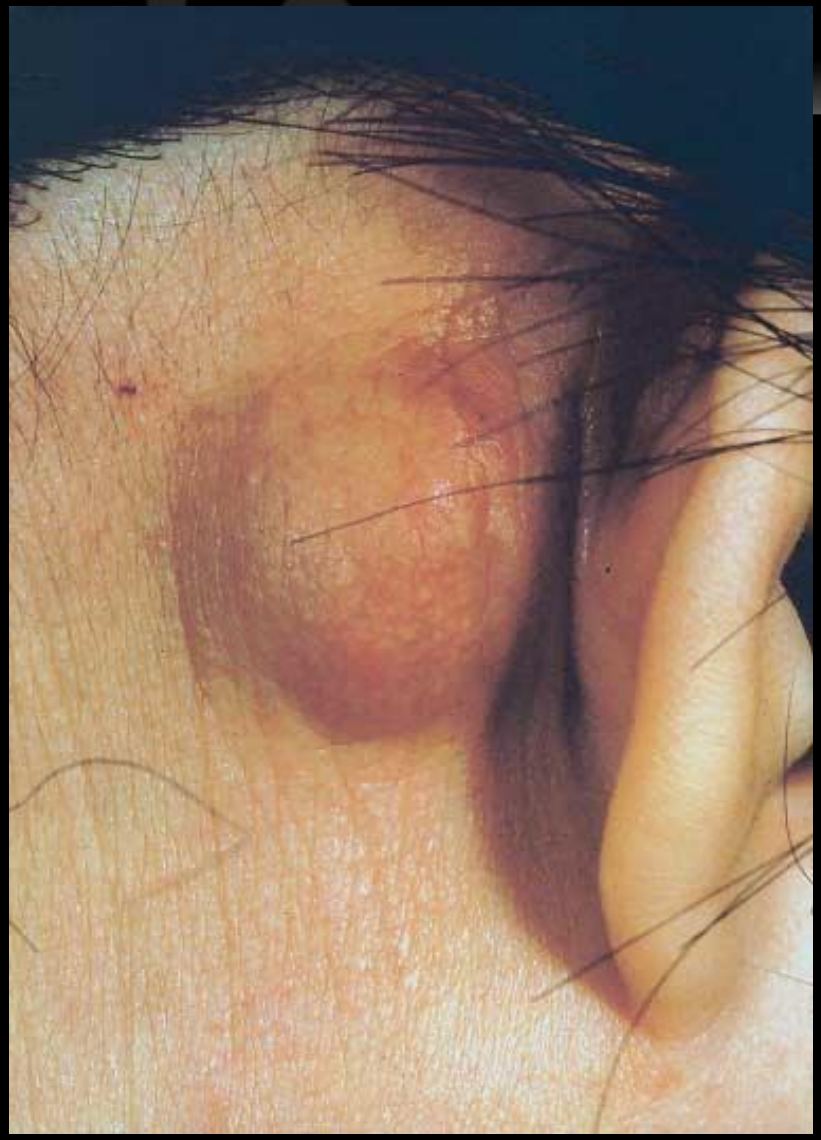
Differential Diagnosis

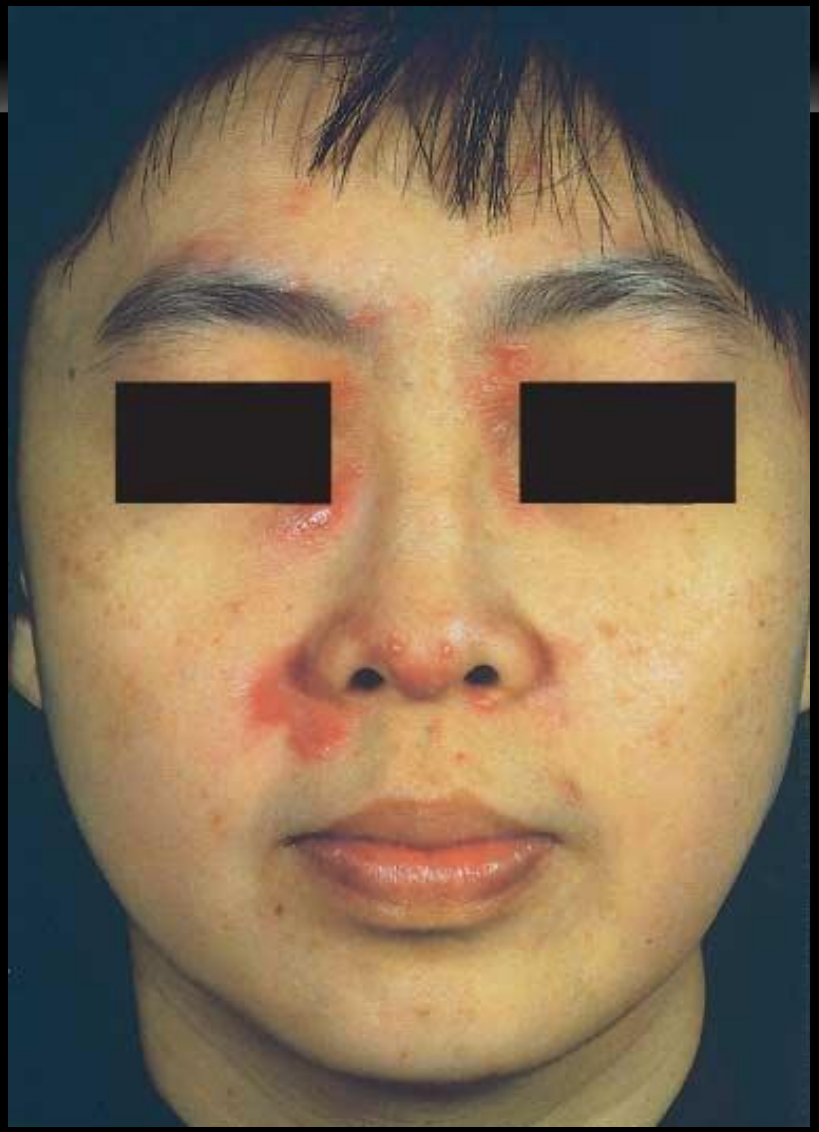
Kimura’s Disease & ALHE

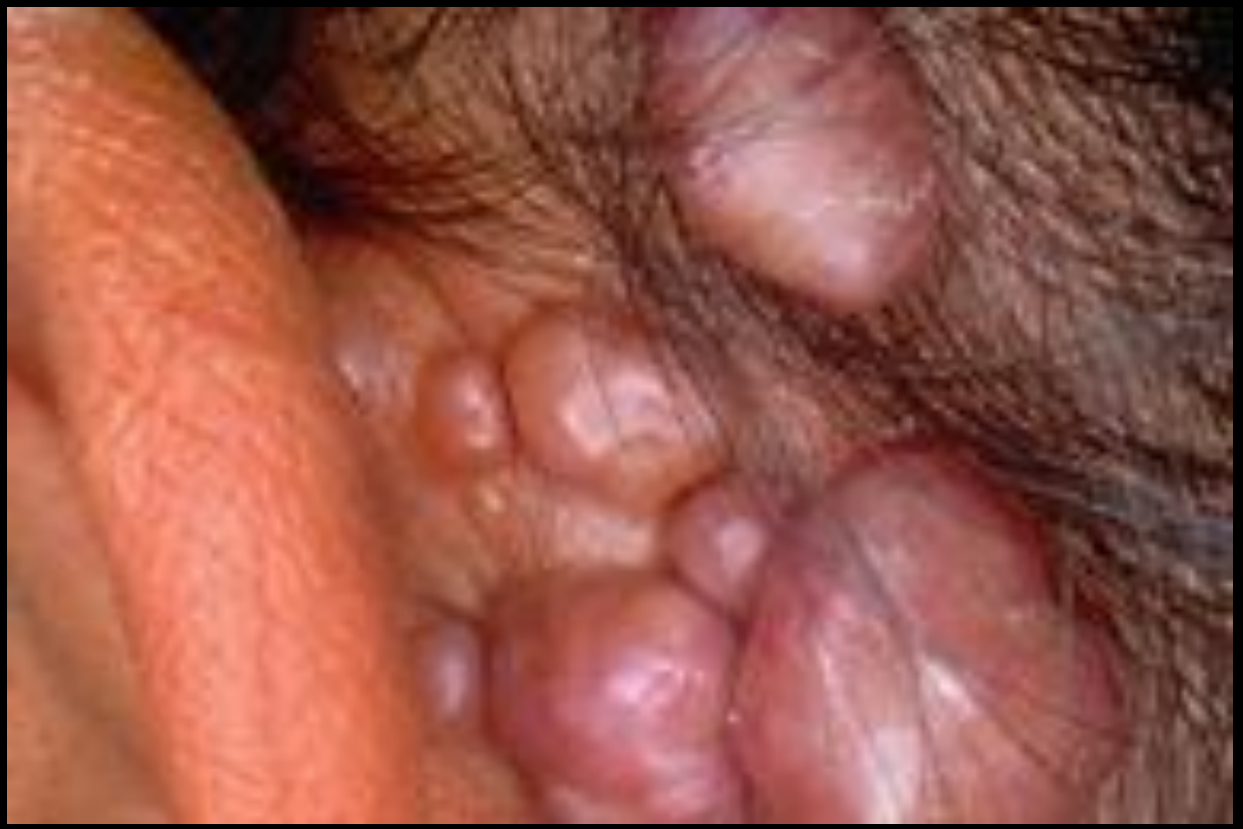
DISCUSSION 2

Clinical Characteristics

	Kimura's disease	ALHE
Sex	Male predominance	Female predominance
Age	2nd – 3rd decades	3rd – 5th decades
Race	More common in Orientals	Occurs in all races
Presentation	Discrete subcutaneous mass	Small dermal papules or nodules
Size	> 2 cm	< 2 cm
Number	Single or multiple	Usually multiple
Overlying skin	Usually normal	Usually erythematous skin





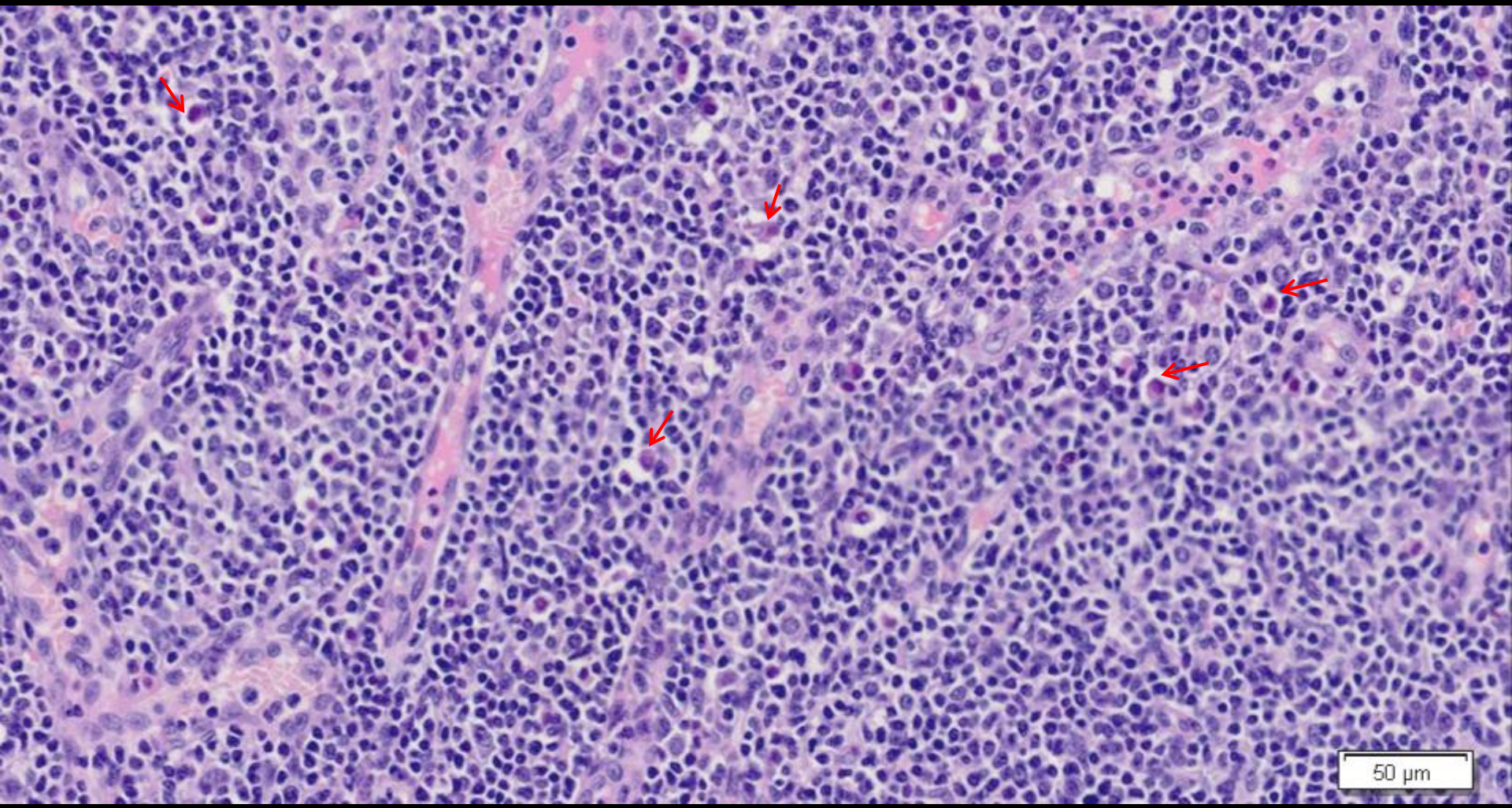


	Kimura's disease	ALHE
Location	Deeper (head &neck)	More superficial (head & neck)
Duration	2 months to 10 years	3 weeks to 12 years
Pruritus	No	May be severe
Lymphadenopathy	Common	Uncommon
Blood eosinophilia	Usually > 10% of total WBC	Usually < 10% of total WBC
Serum IgE	Usually elevated	Usually normal
Glomerulo-nephritis	Occasional	Rare

DISCUSSION 3

Histopathological Characteristics

	Kimura's disease	ALHE
Depth	Subcutaneous, muscle	Dermis, subcutaneous
Vessels	Some degree of vascular proliferation	Florid vascular proliferation
Endothelium	**Flat or low cuboidal**	Cuboidal to dome-shape (epitheloid or histiocytoid)
Inflammation	Abundant lymphocytes and plasma cells	Sparse to heavy infiltrate of lymphocytes and plasma cells
Lymphoid follicles	Always found	May be present
Eosinophils	Abundant	Sparse to abundant
Eosinophilic abscess	Present	Rare
Sclerosis	Significant at all stages	Not a prominent feature


50 µm

DISCUSSION 4

Treatments

Kimura's Disease	ALHE
Steroid therapy	Medical treatment Intralesional corticosteroids Radiotherapy Topical tacrolimus, isotretinoin, and interferon alfa-2b.
Radiotherapy	
Cryotherapy	
Surgical excision(the recurrence rate : 25%)	Surgical treatment Excisions, including the arterial and venous segments at the base of the lesion. Carbon dioxide laser Cryosurgery
Cytotoxic agents : cyclosporin and pentoxifyline	

Conclusion

- Even though clinical characteristics of the patient mimic Kimura's disease, histopathological features confirms its diagnosis.
- A prominent vascular component lined by plump epitheloid endothelial cells is a characteristic of ALHE.
- Nevertheless, both Kimura's disease and ALHE are benign conditions which tend to recur despite any treatment, no malignant change has been reported.


Thank you for your attention

Reference

- Seregard S. Angiolymphoid hyperplasia should not be confused with Kimura's disease. *Acta Ophthalmol Scand 2001; 79:91-3*
- Bei'er Lin. Angiolymphoid hyperplasia with eosinophilia of the eyelid with spontaneous regression. *Opth Plastic & Recons. Surgery 2007;24:308-310*
- Daniel Esmalili. Simultaneous Presentation of Kimura Disease and Angiolymphoid Hyperplasia with Eosinophilia. *Opth Plastic & Recons. Surgery 2007;24:310-311*
- R.K Hejmadi. Angiolymphoid hyperplasia with eosinophilia (epitheloid haemangioma) occuring within multiple deep lymph nodes and presenting with weight loss and raised CA-125 levels. *Virchows Arch 2006;448: 366-368*
- Massimo Fusconi. Angiolymphoid hyperplasia with eosinophilia. *Otolaryngology-Head and Neck Surgery 2006;135: 816-817*