原文題目(出處)：	Necrotizing sialometaplasia of the palate associated with angiocentric T-cell lymphoma. Annals of Diagnostic Pathology 2009;13:60-4
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內文：

Introduction

- **Necrotizing sialometaplasia (NS)** is a benign inflammatory condition that may occur in any site containing salivary gland elements,
- It most frequently involves the minor salivary glands of the hard palate.
- NS is characterized by
  - lobular necrosis with granulation tissue and inflammation.
  - pseudoepitheliomatous hyperplasia of the surface epithelium
  - squamous metaplasia of ducts and acini with preservation of the lobular architecture.
- The cause of this phenomenon is uncertain, mostly related to local trauma and injury:
  - dental injections,
  - alcohol abuse, smoking, cocaine use,
  - upper respiratory infections, prior surgery, and adjacent tumor growths .
- Most authors believe NS is the result of ischemia that leads to local infarction.
- Association of NS with neoplastic conditions is a relatively rare event

Materials and methods

- **Histologic analysis :**

  paraffin-embedded tissue and stained by routine hematoxylin-eosin method
- **Immunohistochemical studies**

  using primary antibodies to CD45 (LCA), CD43 (MTI), CD45RO (UCHL-1), CD15 (LeuM-1), CD30 (BerH2), CD4, CD8(Dako, Carpinteria, CA), CD57 (Biogenex, San Ramon, CA), Epstein-Barr latent membrane Protein (LMP-1, Dako), CD20(L26), CD3, CD56, granzyme, and TIA-1.
- In situ hybridization for Epstein-Barr virus (EBV)–encoded RNA (EBER)

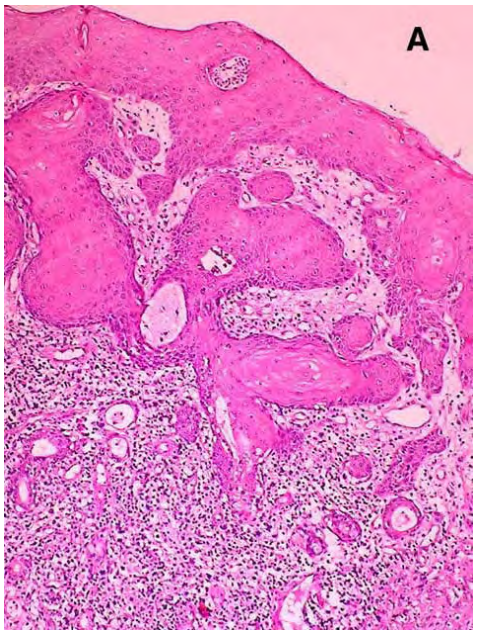
Presentation of the cases

Case1

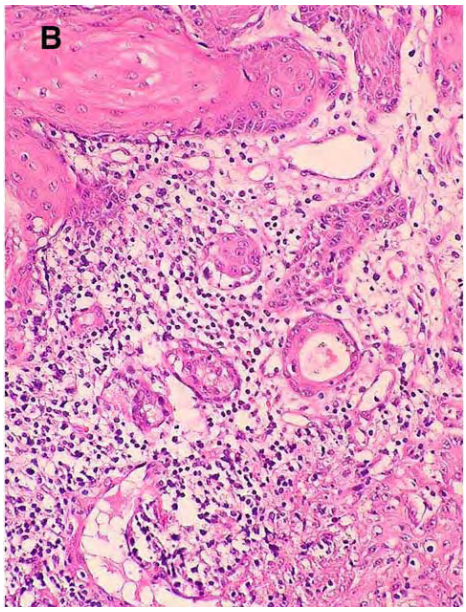
- A 24-year-old man was suffered from a rapidly growing palatal lesion discovered 2 months earlier.
  - **Physical examination** :

    a 3 × 2-cm ulcerated and bleeding lesion extending from the hard palate to the oropharynx.
- Lymphadenopathy(-)
- Needle biopsy :

the lesion was interpreted as a nonspecific inflammatory lesion



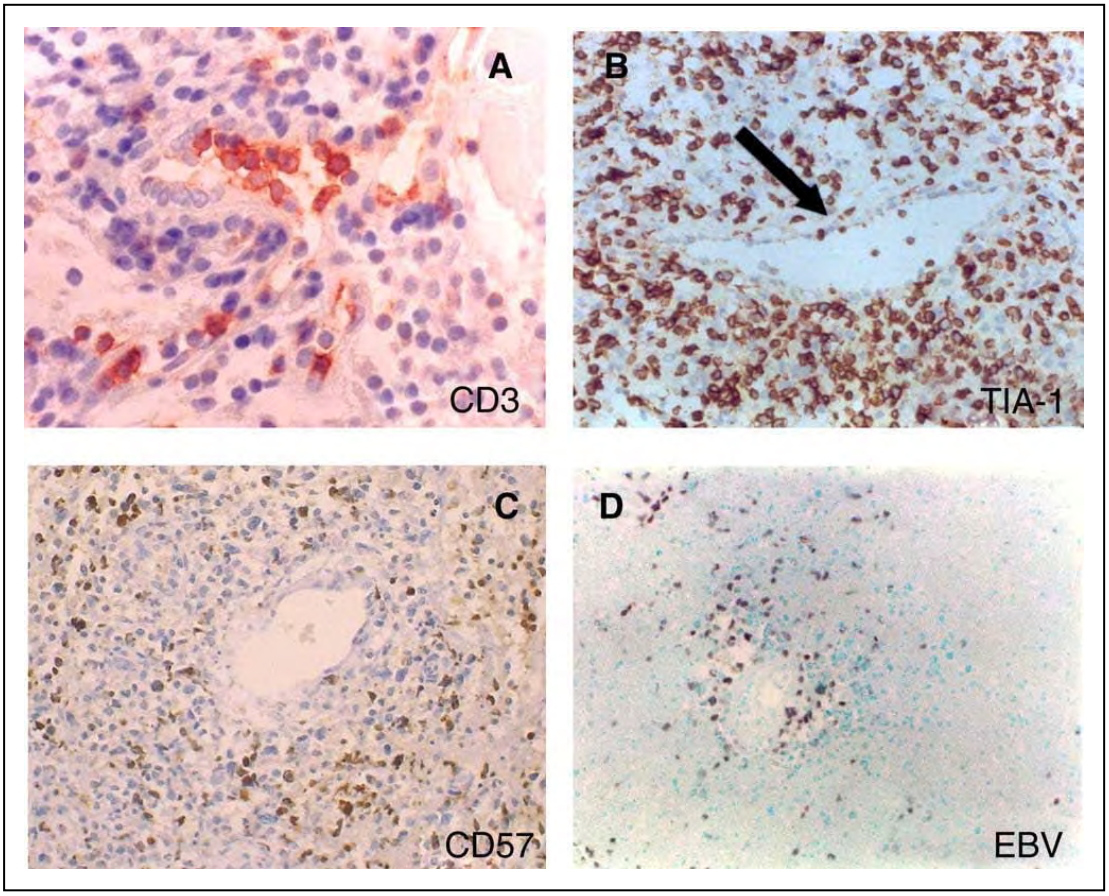
- **Diagnosis :**
  NS associated with angiocentric T-cell lymphoma
- Bone marrow biopsy :
  failed to identify any involvement by lymphoma
- He received 5 cycles of chemotherapy, but the response was poor and the lesion extended into the nasal mucosa, producing obstruction of the left side.
- The patient had intermittent fever but results of repeated cultures of the ulcer were negative for bacteria.
- He developed pancytopenia and persistent fever suggestive of virus-associated hemophagocytic syndrome.
- Finally he died of acute hepatic failure 5 months after admission.



Histologic findings

- an ulcerated lesion characterized by abundant granulation tissue associated with
  - pseudoepitheliomatous hyperplasia of the covering epithelium
  - acinar necrosis of the adjacent salivary glands
  - squamous metaplasia of ducts and acini with preservation of the lobular architecture
- A profuse lymphocytic infiltrate with a population of atypical cells surrounded the epithelial structures and extended downward to infiltrate adjacent glandular tissue
- The cell population showed a diffuse growth pattern with some patchy necrotic areas, and it was prominent around and within blood vessels.

Immunohistochemical findings

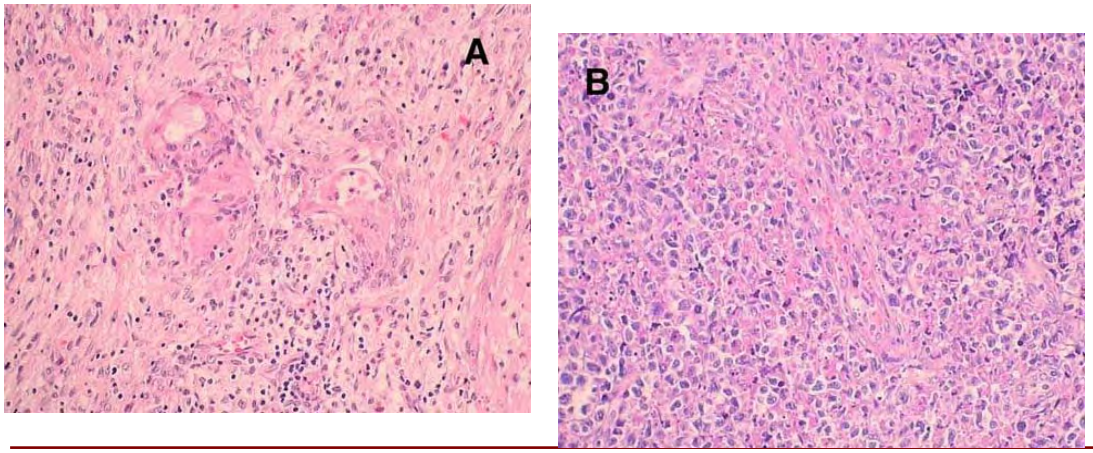


- Results for other markers including CD20, CD4, and CD8 were negative

Case 2

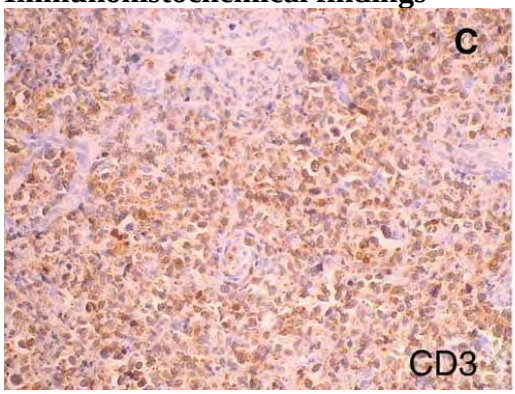
- A 39-year-old woman was suffered from rhinorrhea, otalgia, and nasal obstruction accompanied by fever and weight loss of 20 kg.
- **Computerized axial tomography scan**
  a tumor involving all paranasal sinuses with a zone of osteolysis of the medial wall of the maxillary sinus.
- **Biopsy**
  Angiocentric T-cell lymphoma
- She received radiotherapy and 4 cycles of chemotherapy,but lost to follow-up at 7 months of treatment

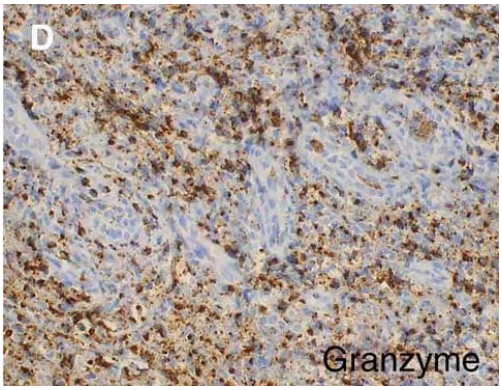
Histologic findings



- Respiratory epithelium with extensive areas of coagulative necrosis
- The accessory mucous glands featured lobular necrosis and squamous metaplasia involving ducts and acini with preservation of the lobular architecture
- A diffuse infiltrate with a population of large atypical lymphoid cells surrounded the epithelial structures and invaded the walls of some blood vessels

Immunohistochemical findings





- Results for other markers including C20, CD56, and CD34 were negative.

Discussion

- More than 75% of the reported cases of NS occurred in the salivary glands of the palate.
- It usually starts as a nonulcerated swelling, often associated with pain or paresthesia, which eventually breaks to produce a deep and well-demarcated ulcer that ranges from less than 1 to more than 5 cm in diameter and heals spontaneously after 4 to 6 weeks.
- Necrotizing sialometaplasia is believed to be a lesion that results from local ischemia, secondary to blockage of blood supply to the salivary gland tissue which is based on histology.
- Because of its clinical and microscopic features, it has been confused with squamous carcinoma and mucoepidermoid carcinoma.
- Morphologic evidence is provided by the presence of lobular infarction in the early phases of the disease.
- NS may be in association with other conditions that may produce local vascular obliteration

Nonneoplastic conditions:

atherosclerosis, local surgical trauma, allergy, thromboangiitis, obliterans (Buerger disease), dental injections, dental prosthesis, sickle cell anemia , heavy alcohol and tobacco consumption.

Neoplasm including:

Mixed tumor, monomorphic adenoma, Warthin tumor, and rhabdomyosarcoma

Angiocentric T/NK cell lymphomas (Lethal midline granuloma syndrome)

- The lesions that originate in the nasal cavity, paranasal sinuses, and hard palate constitute a diagnostically challenging group of diseases
- Most of these lesions have proved to be malignant lymphomas, with a predominance of T/NK-cell lineage in Asian and some Latin-American populations.
- It produce widespread necrosis, which is the result of vascular angioinvasion and angiodestruction by the cytotoxic cells and the surrounding inflammatory cell infiltrate.
- The 2 cases presented here could be due to local ischemia of palatine salivary

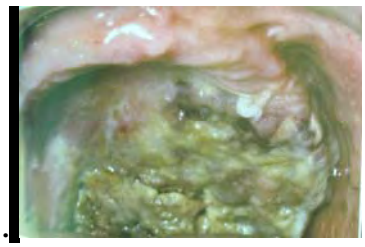
glands, possibly as a consequence of angioinvasion and angiodestruction by the neoplastic lymphocytes.

- Most authors agree that most angiocentric lymphomas are derived from Epstein-Barr virus–infected cytotoxic lymphocytes of both NK- and T cell lineages
- Case 1 was characterized as CD56−, CD57+. CD3+, TIA-1+, CD4−, CD8+, and EBER+
- case 2 as CD3+, CD56−, and granzyme+. They are not considered of NK-cell lineage but better classifiable as cytotoxic T-cell lymphomas .
- EBV is a significant and consistent finding among NK- and T-cell malignant lymphomas and, regardless of geographic origin, this infectious agent has been demonstrated in more than95% of cases .
- TIA-1 and granzyme are not limited to NK-cell lymphomas but are also seen in cytotoxic lymphomas of true T-cell lineage
- The question of how to classify EBER+, CD56−,and TIA-1+ angiocentric lymphomas of T-cell lineage occurring in similar sites with similar morphology still remains to be resolved.

Conclusions

- The author postulate that vascular occlusion by the neoplastic lymphoid cells produced ischemia and contributed to the development of the salivary gland lesion.

Questions

題號	題目
1	**Which disease is often cause destructive inflammatory condition in the palatal salivary gland and easily misdiagnoses as squamous cell carcinoma or mucoepidermoid carcinoma ?** **(A) Basal cell adenoma** **(B) Necrotizing sialometaplasia** **(C) Warthin tumor** **(D) Acinic cell adenocarcinoma**
答案(B)	出處：*Oral & Maxillofacial Pathology,second edition,SAUNDERS p.405-406*
題號	題目
2	**This 62-year-old man has a lesion over palatal side,the surface is ulcerative.**  **In histology, it shows atypical lymphoid cells infiltrating the wall and filling the lumen of a blood vessel? Which disease is the most likely?** **(A) Multiple myeloma** **(B) Angiocentric T-cell lymphoma** **(C) Burkitt's lymphoma** **(D) Plasmacytoma**
答案(B)	出處：*Oral & Maxillofacial Pathology,second edition,SAUNDERS p.524-526*