Case report-Olgu sunumu

# Necrotizing sialometaplasia after lower lip wedge resection

# Alt dudak kama rezeksiyon sonrası gelişen nekrotizan siyalometaplazi

## Ahu Senem Demiröz\*, Cuyan Demirkesen, Hakan Arslan

Department of Pathology (A. S. Demiröz, MD, Prof. C. Demirkesen, MD), Department of Plastic and Reconstrictive Surgery (Assoc. Prof. H. Arslan, MD), İstanbul University Cerrahpaşa School of Medicine, TR-34098 İstanbul

## Abstract

Necrotizing sialometaplasia (NS) is a rare disease of salivary glands. It is a self limited inflammatory process associated with tissue ischemia. Both clinically and histologicaly, it can be confused with malignancy. We report a case of NS which develop after lower lip wedge resection due to squamous cell carcinoma.

Keywords: Salivary gland, squamous cell carcinoma, necrosis

## Özet

Nekrotizan siyalometaplazi türük bezlerinin nadir görülen bir hastalığıdır. iskemiye bağlı gelişen kendini sınırlayıcı inflamatuar bir süreçtir.hem klinik hem de histolojik olarak malignite ile kariştırılabilir. Bu yazıda, skuamoz hücreli karsinom nedeniyle alt dudak kama rezeksiyon yapılan bir olguda gelişen nekrotizan siyalometaplazi anlatılmıştır.

Anahtar sözcükler: Tükrük bezi, skuamoz hücreli karsinom, nekroz

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#### \*Corresponding author:

Dr. Ahu Senem Demiröz, Patoloji Anabilim Dalı, İstanbul Üniversitesi Tıp Fakültesi, TR-34098 İstanbul. E-mail: ahusenem@yahoo.com

# Introduction

Necrotizing sialometaplasia (NS) was defined by Abrams, in 1973, as a reactive necrotizing inflammatory process involving minor salivary glands of the hard palate [1, 2]. NS is a benign self limited process usually involving the oral minor salivary glands and mucoserous glands of the sinonasal tract [3]. Other sites where it has been reported include trachea, parotid gland, larynx, buccal mucosa, tongue, tonsil and retromolar trigon [2, 4].

We report a case of NS, which developed within 2 weeks after lower lip wedge resection due to squamous cell carcinoma.

# **Case report**

A 62 years old male was referred to the outpatient clinic of the plastic surgery for a lower lip lesion that had been presented for 5 months. In the physical examination ulcerated, hyperpigmented, bulky lesion with indistinct borders measuring about 1 cm in diameter was detected. Lower lip excision was performed and the diagnosis was squamous cell carcinoma developing on actinic chelitis (Figure 1).

Within 2 weeks, a recent development of ulceration on the excision site was detected. The possibility of recurrence or the presence of rest tumor was considered, therefore, re-

excision was performed. There was no residuel tumor in the re-excisional material. Scar tissue formation due to former excision was detected. Besides, in the salivary gland there were multiple foci of ductules and acinary glands composed of squamous cells without any atypical features, interpreted as squamous metaplasia (Figure 2). The aciner remnants also showed pseudoepitheliomatous hyperplasia. In some of the glands coagulation necrosis was evident. Mild inflammatory infiltrate and fibroblastic activity accompanied. These findings were interpreted as benign features as a result of degeneration and regeneration. According to these morphologic features the diagnosis NS was established.

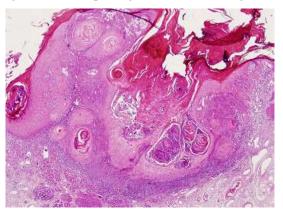


Figure 1. First lesion's epithelium is both exophitic, with focal papillomatosis and covering of hyperkeratosis and parakeratosis, and endophytic (H&EX40).

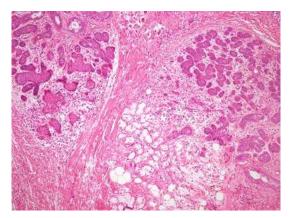


Figure 2. Multiple foci of ductules and acinary glands composed of squamous cells without any atypical features in the salivary gland (H&EX40).

# Discussion

The majority of NS cases were found on the posterior palate. And parotid was the most common gland involved. Lower lip location has also been reported with or without associated lesions [2, 5]. The histologic features of NS include coagulation necrosis of glandular acini within the lobules, extensive squamous metaplasia of ducts and sometimes mucus pooling with an associated granulation tissue and inflammatory response [2]. In contrast to our findings, cytologic atypia can be severe with single cell necrosis and hypercromatic, angulated cells. If atypia is alarming with single cell necrosis, application of immunhistochemistry panel including myoepithelial markers (smooth muscle antibody, p63, calponin), basement membrane markers (laminin, collagen type IV), E-cadherin and various cytokeratins (CK 5, CK 6, CK 7, CAM 5.2) has been suggested. Histologically, maintenance of lobular architecture is the best clue for NS [4, 6].

Most investigators believe that NS arises due to vascular changes and blockage or compromised blood supply to salivary gland lobules, resulting in ischemic necrosis or infarction. The nature and pathophysiology of the ischemia has yet to be elucidated in most cases. Possible predisposing factors are vascular injury, infection, physical and chemical trauma, smoking, alcohol abuse and recurrent vomiting [2, 4, 7, 8]. These vascular changes may follow surgical trauma to involved site as in our case. Brannon et al. [2] who reported 69 NS cases, 25 cases of NS developed after a surgical procedure. Introduction of large volume of anaesthetics in the tissue, direct injury to a blood vessel from the needle or prolonged pressure by swelling from anesthetic solution were accused for the adverse effects to local blood supply.

The mean time interval from the initial surgery to the development and diagnosis of NS was 18 days (ranged from 6-53 days) [2]. In our case the period of the formation of NS after lower lip surgery was 15 days. It is a self-healing disease usually resolving within a period of 3 to 12 weeks [2, 7]. NS does not recur, and if it is properly diagnosed preoperatively, surgical excision is not necessary [4].

In conclusion NS can be confused with mucoepidermoid and squamous cell carcinoma both histologically and clinically [5]. Its recognition is essential since misdiagnosis may result in unnecessary or inappropriate therapy [9].

# References

- 1. Abrams AM, Melrose RJ, Howel FV. Necrotising sialometaplasia: A disease simulating malignancy. Cancer 1973; 32: 130-5.
- 2. Brannon RB, Fowler LB, Hartman KS. Necrotising sialometaplasia: A clinicopathologic study of sixty nine cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1991; 72: 317-25.
- 3. Batsakis JG, Manning JT. Necrotising sialometaplasia of major salivary glands. J Laryngol Otol 1987; 101: 962-6.
- 4. Carlson DL. Necrotizing sialometaplasia: A practical approach to the diagnosis. Arch Pathol Lab Med 2009; 133: 692-8.
- Pulse CL, Lebovics RS, Zegarelli DJ. Necrotizing sialometaplasia: Report of a case after lower lip mucocele excision. J Oral Maxillofac Surg 2000; 58: 1419-21.
- 6. Rizkalla H, Toner M. Necrotizing sialometaplasia versus invasive carcinoma of the head and neck: The use of myoepithelial markers and keratin subtypes as an adjunct to diagnosis. Histopathology 2007; 51: 184-9.
- 7. Niedzielska Ii Janic T, Markowski J. Bilateral localization of necrotizing sialometaplasia: A case report. Cases J 2009; 2: 9068.
- 8. Solomon LW, merzianu M, Sullivan M, Rigual NR. Necrotizing sialometaplasia associated with blumia: Case report and literature review. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007; 103: 39-42.
- Aydin O, Yilmaz T, Ozer F, Saraç S, Sokmensuer C. Necrotising sialometaplasia of parotid gland: A possible vasculitic cause. Int J Pediatr Otorhinolaryngol 2002; 64: 171-4.