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CASE REPORT

A case of necrotizing sialometaplasia in the upper lip

Üst dudakta nekrotizan sialometaplazi: Olgu sunumu

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Necrotizing sialometaplasia is a benign, reactive, and self-limiting inflammatory disorder with clinical and histologic features resembling carcinoma. A fifty-eight-year-old woman presented with a firm submucosal mass on the right side of the upper lip, measuring 0.5 cm. Histopathologic examination of the incisional biopsy specimen was reported as adenoid cystic carcinoma, resulting in removal of the mass with a large excision and reconstruction of the primary site. However, final histopathologic diagnosis of the excised mass was necrotizing sialometaplasia. No recurrences occurred during a three-year follow-up. This report draws attention to the difficulty in distinguishing between necrotizing sialometaplasia and adenoid cystic carcinoma.

Key Words: Lip neoplasms/diagnosis/surgery; salivary gland diseases/pathology; sialometaplasia, necrotizing/diagnosis/pathology/surgery.

Nekrotizan sialometaplazi benign, reaktif ve kendi kendini sınırlayan enflamatuvar bir hastalıktır. Bu hastalığın önemi, klinik ve histolojik özellikleri açısından karsinoma benzemesidir. Elli sekiz yaşında kadın hasta sağ üst dudakta, 0.5 cm boyutunda, mukoza altında sert bir kitle ile başvurdu. Biyopsi sonucunun adenoid kistik karsinom olarak bildirilmesi üzerine kitle geniş bir çevre doku ile çıkarıldı ve saha aynı seansta rekonstrükte edildi. Çıkartılan kitleye histopatolojik inceleme sonucunda nekrotizan sialometaplazi tanısı kondu. Olgunun üç yıllık izleminde nüks saptanmadı. Bu yazıda nekrotizan sialometaplazinin adenoid kistik karsinom ile karıştırılabileceği vurgulandı.

Anahtar Sözcükler: Dudak neoplazmları/tanı/cerrahi; tükürük bezi hastalıkları/patoloji; nekrotizan sialometaplazi/tanı/patoloji/cerrahi.

Necrotizing sialometaplasia (NS) may be histologically misdiagnosed as squamous cell or mucoepidermoid carcinoma. It is a reactive, inflammatory, and usually self-limited benign disorder essentially affecting the minor salivary glands and manifests clinical and histologic features that simulate malignancy.[1] It is most commonly found in the hard palate, but has been reported in all areas where salivary gland tissue exists.[2] The aim of this report is to present a case of NS of the upper lip which was misdiagnosed as carcinoma and also to draw attention to some misleading features of this disease.

CASE REPORT

A fifty-eight-year-old woman presented with a mass in the upper lip, which appeared a year ago. She had no other complaints and no history of smoking or consumption of ethyl alcohol, nor did she

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recall any traumatic event. Examination revealed a firm submucosal mass on the right side of the upper lip, measuring 0.5 cm. The mass assumed the size of a lentil grain two months ago, after which it progressively enlarged.

An incisional biopsy was performed and histopathologic examination of the specimen resulted in an initial diagnosis of an adenoid cystic carcinoma. The mass was removed with a large excision and the primary site was reconstructed. Histopathologic study of the excised mass was consistent with a diagnosis of NS. No recurrences were detected during a follow-up period of three years.

Microscopically, the specimen was covered with intact squamous epithelium, beneath which there were minor salivary glands. In one area, normal structure was somewhat distorted, but maintained its lobular architecture. The glands underwent metaplastic changes around the major duct (Fig. 1). Both multiple and serial sections showed no areas of adenoid cystic carcinoma. What lead to an erroneous conclusion for adenoid cystic carcinoma following the initial biopsy was the observa-

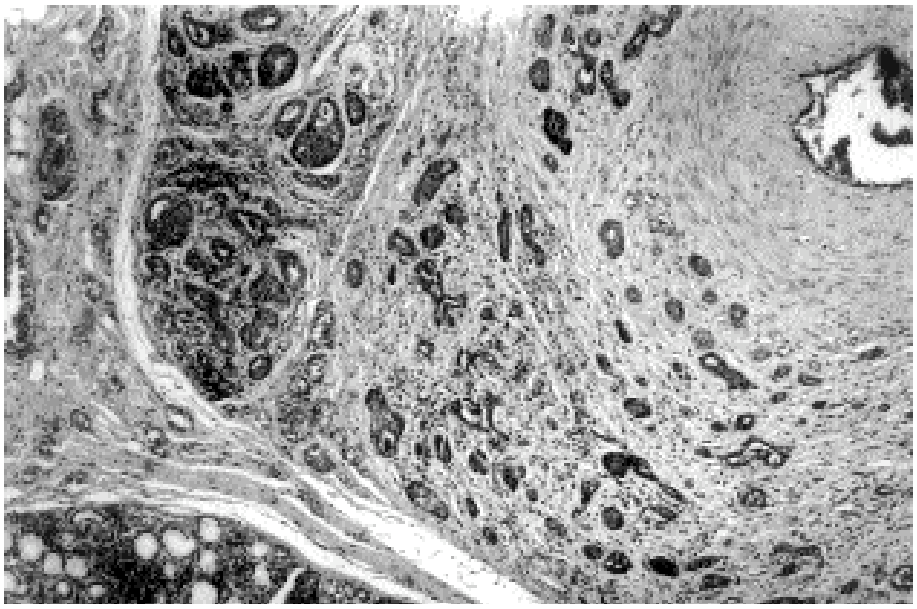

Fig. 1 - *Normal minor salivary gland at the lower left corner and a major excretory duct at the upper right corner. The glands are of a squamoid appearance (H-E x 50).*

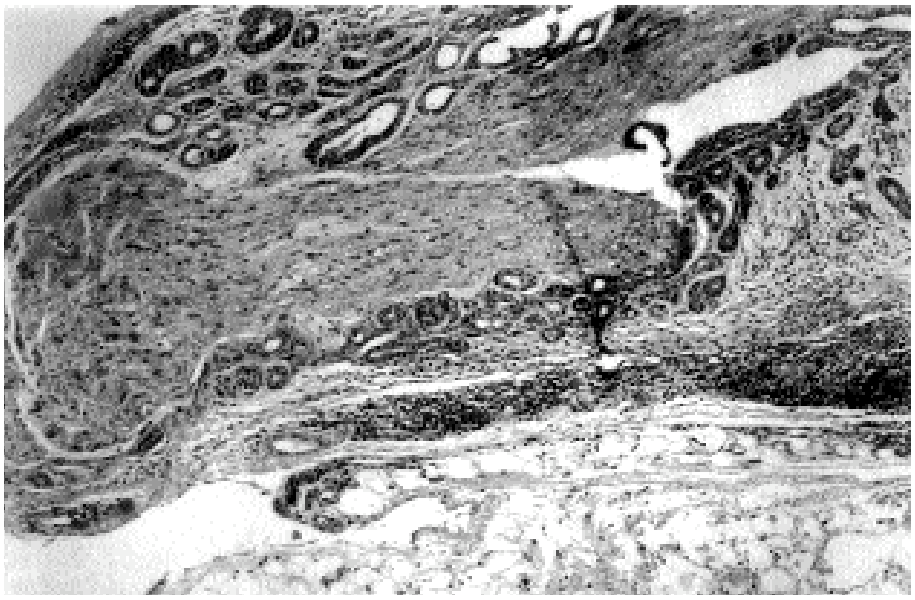

Fig. 2 - *The initial biopsy specimen showing glandular structures around a peripheral nerve (H-E x 50).*

tion of glandular structures around a peripheral nerve (Fig. 2).

DISCUSSION

Necrotizing sialometaplasia is a benign, self-limiting inflammatory disorder with clinical and histologic features resembling carcinoma. It mainly affects the minor salivary glands, though it may also be found wherever salivary tissue is present. It most often occurs in the oral cavity, the junction of the hard and soft palate being the most frequently involved site. The lesion presents as a deeply-seated and sharply demarcated ulcer, but a non-ulcerated swelling or an incidental histologic finding may also be the only manifestation.[1-3] It is thought to result from ischemia or a tissue injury that is caused by impaired blood supply due to anesthesia, dental trauma, or surgery.[1,3]

Unlike squamous cell carcinoma or mucoepidermoid carcinoma, histologic features of NS include lobular necrosis and metaplastic epithelial cells lining the small salivary gland ducts, with preservation of a lobular appearance of the involved gland.[1,3] In either the minor or major salivary glands, the main histologic feature is infarcted salivary lobules with subsequent repair and metaplasia.[1,4]

The treatment of NS varies from wide resection to local excisional biopsy.[4,5] Since spontaneous healing may occur within some weeks, no treatment may be required other than follow-up.[3] In our case, the lesion persisted for months and showed progressive enlargement. Moreover, the initial diagnosis of the biopsy specimen was reported as adenoid cystic carcinoma. Literature reports emphasize the confusing histologic features of NS in the differential diagnosis with squamous cell carcinoma, mucoepidermoid carcinoma, or adenoid cystic carcinoma.[2,4,6]

This case report illustrates that NS may be found as a submucosal mass in the upper lip and highlights a potential risk that it may be misdiagnosed as a malignancy and, thus, be handled accordingly.

REFERENCES

1. Sneige N, Batsakis JG. Necrotizing sialometaplasia. Ann Otol Rhinol Laryngol 1992;101:282-4.
2. Granick MS, Solomon MP, Benedetto AV, Hannegan MW, Sohn M. Necrotizing sialometaplasia masquerading as residual cancer of the lip. Ann Plast Surg 1988; 21:152-4.
3. van der Wal JE, van der Waal I. Necrotizing sialometaplasia: report of 12 new cases. Br J Oral Maxillofac Surg 1990;28:326-8.
4. Russell JD, Glover GW, Friedmann I. Necrotizing sialometaplasia. J Laryngol Otol 1992;106:569-71.
5. Myers EN, Bankaci M, Barnes EL Jr. Necrotizing sialometaplasia. Report of a case. Arch Otolaryngol 1975;101:628-9.
6. Imbery TA, Edwards PA. Necrotizing sialometaplasia: literature review and case reports. J Am Dent Assoc 1996;127:1087-92.