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Warthin Tumor Exhibiting Sebaceous Differentiation and Necrotizing Sialometaplasia

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Summary. A case of a Warthin tumor exhibiting sebaceous differentiation and necrotizing sialometaplasia is presented. This case suggests a common histogenesis for the Warthin tumor and sebaceous lymphadenoma. It supports the theory that necrotizing sialometaplasia is caused by factors which compromise or obstruct the blood supply to salivary gland tissues. The literature on sebaceous differentiation in Warthin tumor and on necrotizing sialometaplasia is reviewed.

Key words: Necrotizing sialometaplasia – Warthin tumor – Sebaceous glands – Salivary gland – Parotid gland

Introduction

Sebaceous differentiation in the normal parotid gland is a little recognized but common finding (Hamperl 1931; Meza-Chavez 1949). Sebaceous foci have been found in a variety of salivary gland tumors. These include benign mixed tumors, carcinoma ex pleomorphic adenoma, mucoepidermoid carcinomas, and Warthin tumors (Gnepp, submitted for publication). Nine Warthin tumors with focal sebaceous differentiation were found in the literature (Batsakis et al. 1972; Foote et al. 1954; Geiler 1957; Giamminola 1975; Thackray et al. 1974; Thackray et al. 1972; Zechner et al. 1973; Foote et al. 1953) however, clinical information is available in only a few of these cases.

Necrotizing sialometaplasia is a non-neoplastic, inflammatory, commonly ulcerating and self-healing lesion of human salivary glands. It occurs commonly on the hard or soft palate but may occur anywhere salivary gland tissues are found. In 1979 Donath reported six cases of necrotizing sialometaplasia involving the parotid gland. At about the same time, Lynch et al. (1979) indicated that 42 cases of necrotizing sialometaplasia had been reported in the literature. If these cases are combined with the Armed Forces Institute of Pathology's

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experience of 57 cases recently reported (Brannon et al. 1979) then at least 105 cases have been reported to date.

This is the first reported case combining Warthin tumor with sebaceous differentiation and focal necrotizing sialometaplasia.

Case Report

The patient is a 58 year old white male with a one year history of a nontender lump below his left ear. It had not changed in size during the four months prior to this admission; however, the patient noted a decrease in hearing on the right side during that time. There was a movable nontender mass located inferior to the left ear and anterior to the sternomastoid muscle. The mass measured 3 cm in diameter and appeared separate from the parotid gland. The remaining physical exam was noncontributory. Because of the clinical suspicion that the mass might represent a metastasis from an oropharyngeal carcinoma, the lesion was biopsied. The diagnosis from this biopsy was Warthin tumor. A left parotidectomy was performed. The patient is presently free of recurrent disease 14 months after his surgery.

Gross and Microscopic Findings

The parotidectomy specimen measured $8 \times 5 \times 2.5$ cm. Its cut surface revealed a focally necrotic encapsulated mass measuring $3.5 \times 2.5 \times 2.0$ cm. This mass

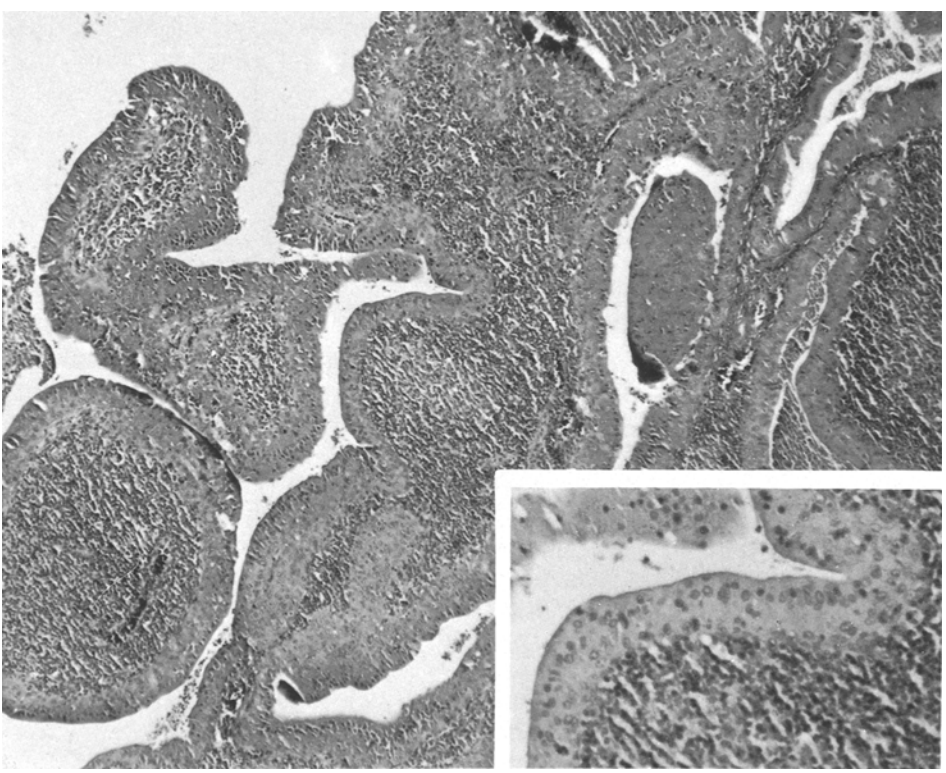


Fig. 1. Photomicrograph of the Warthin tumor demonstrating the papillary epithelial and lymphoid components of the tumor (Hematoxylin and eosin stain, original magnification $\times 125$). *Insert:* Detail of oncocytic columnar epithelium of Warthin tumor (Hematoxylin and eosin stain, original magnification $\times 350$)

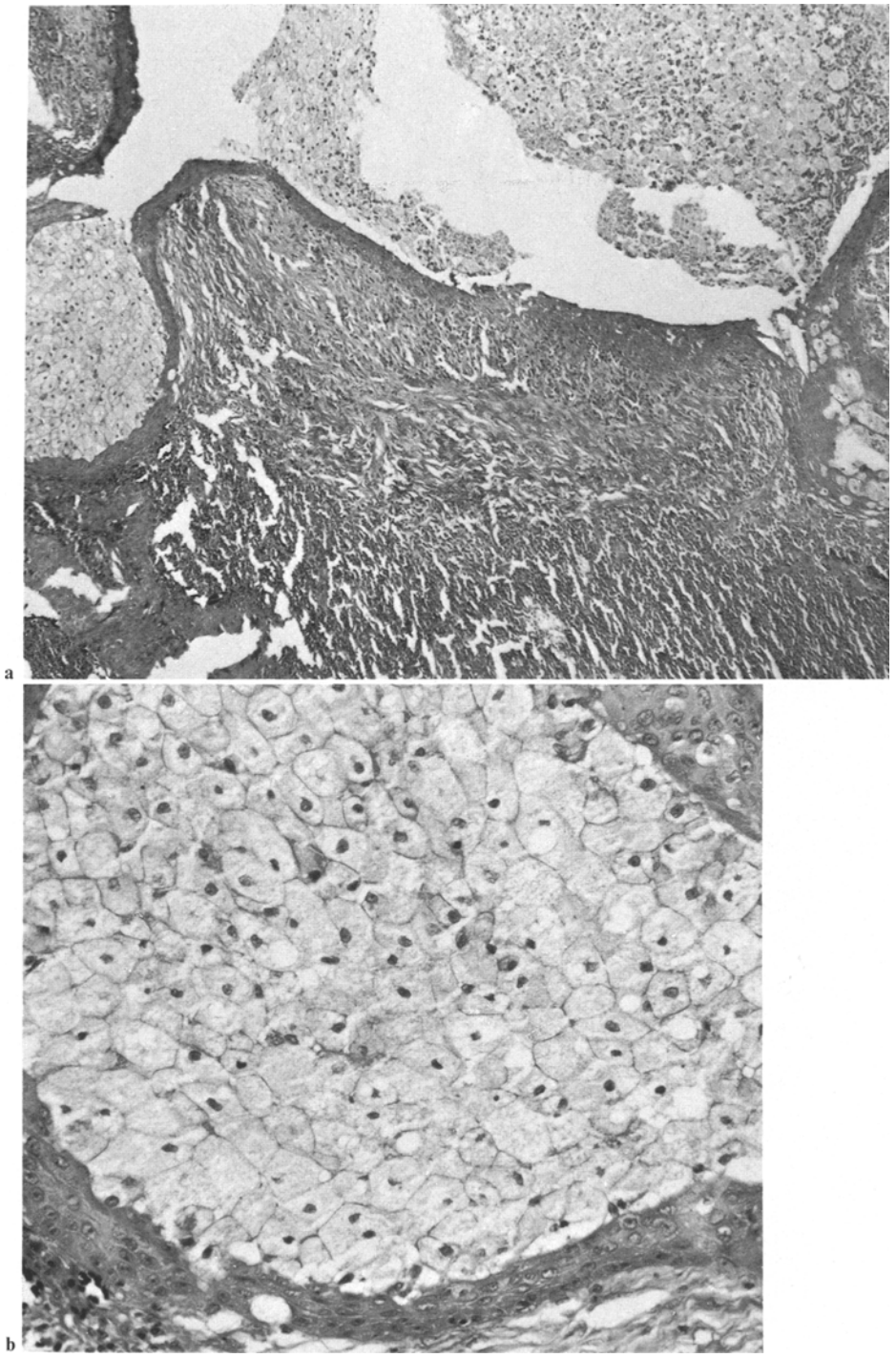
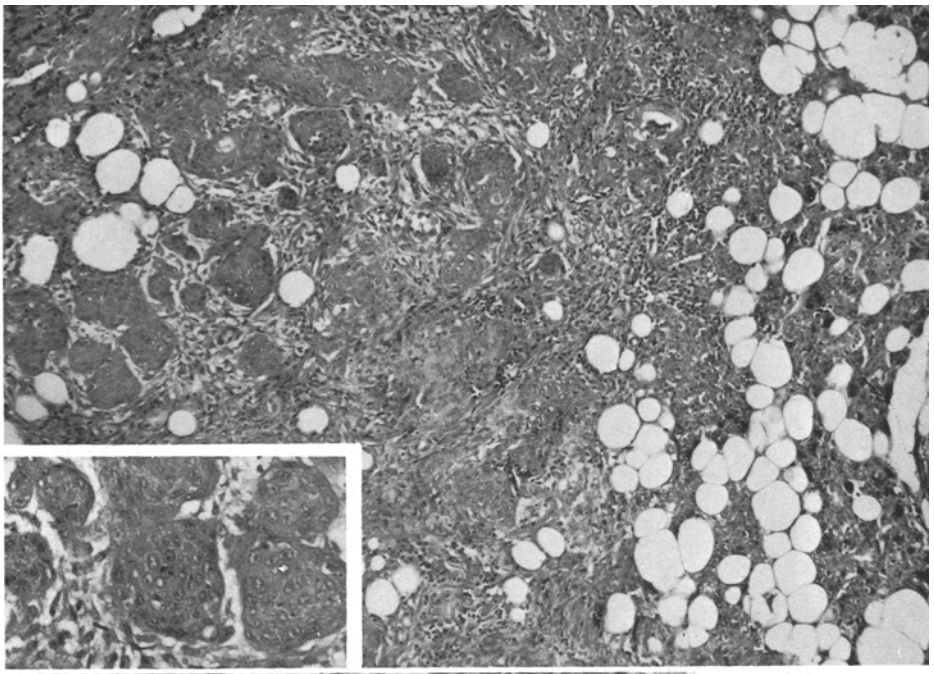
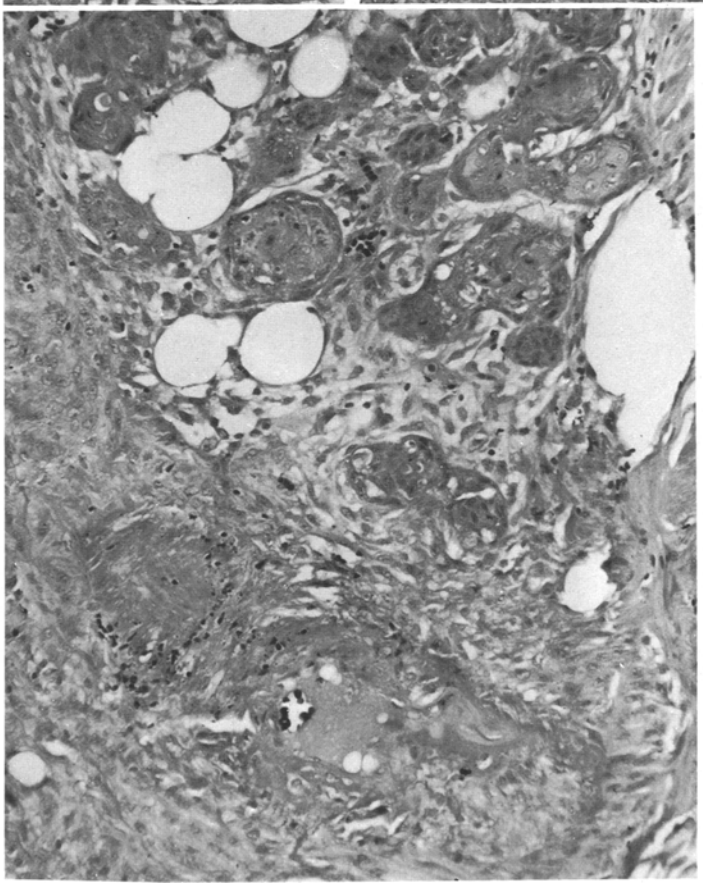


Fig. 2. a Photomicrograph of midportion of the Warthin tumor demonstrating a cystic space filled with sebum and lined by a stratified squamous epithelium. Two foci of sebaceous differentiation are evident. (Hematoxylin and eosin stain, original magnification $\times 125$). **b** Detail of sebaceous gland from 2a demonstrating the stratified squamous basilar epithelium with typical sebaceous cells in the center. (Hematoxylin and eosin stain, original magnification $\times 400$)



a



b

was soft, friable, and varied in color from pink-tan to yellow-tan to whitish-yellow. Histologic examination revealed an encapsulated tumor composed of variably sized, slightly irregular cystic cavities lined by pseudostratified columnar epithelial cells with abundant eosinophilic cytoplasm (Fig. 1). Beneath the epithelium was fibrovascular stroma containing numerous lymphocytes. In several areas immediately beneath the capsule, slitlike subcapsular sinuses filled with lymphocytes were present. In the central portion of the tumor was a dilated cystic space lined by stratified squamous epithelium. In two foci the epithelium had a different appearance (Fig. 2a). The basilar epithelium in these areas was also stratified squamous epithelium; however, toward the surface of the cyst the epithelial cells were larger and exhibited pyknotic eccentric nuclei surrounded by abundant delicate foamy cytoplasm, typical of sebaceous cells (Fig. 2b). Serial sectioning demonstrated continuity between these sebaceous glands and the larger cystic space which was filled with holocrine secretions from the sebaceous glands. The original biopsy also demonstrated similar sebaceous foci. In one area corresponding to the original biopsy site, there was focal necrosis, chronic inflammatory cells, and many blood vessels lined by plump reactive endothelium. The acini in this region had been replaced by many ductal structures which had undergone squamous metaplasia. They were composed of uniform squamous cells exhibiting a lobular arrangement (Fig. 3). In the central portion of this area, the wall of several small arteries was necrotic and infiltrated by neutrophils (Fig. 3b).

Discussion

Sebaceous differentiation in Warthin tumor has been reported ten times; however, this finding is probably more common than the literature indicates. Bernier and Bhaskar (1958) in evaluating 106 Warthin tumors concluded that "... papillary cystadenoma lymphomatosum is a salivary gland tumor arising in a lymph node ...". They based their conclusion on several factors: 1) Salivary gland tissue is commonly found in the intra-parotid and periparotid lymph nodes and may form simple cysts in clearly identifiable lymph nodes, 2) Early stages in the formation of Warthin tumors are seen in lymph nodes that have only partially replaced the nodal structure, leaving areas of clearly identifiably normal anatomy, 3) In many tumors subcapsular sinuses and/or medullary sinusoids were demonstrated.

A similar origin for sebaceous lymphadenoma has been suggested by Gnepp and Sporck (1980). They based their conclusion on finding intranodal sebaceous and salivary gland inclusions in a parotid gland and on a lymphoepithelial parotid cyst with sebaceous differentiation.

Fig. 3. a Original biopsy site with focus of necrotizing sialometaplasia exhibiting a lobular distribution of the squamous nests in a fibrous stroma infiltrated by chronic inflammatory cells. (Hematoxylin and eosin stain, original magnification ×125). *Insert:* Detail of squamous metaplasia showing uniform bland appearing nuclei (Hematoxylin and eosin stain, original magnification ×350). **b** Area of necrotizing sialometaplasia demonstrating necrotic interlobular artery infiltrated by partially fragmented neutrophils. (Hematoxylin and eosin stain, original magnification ×350)

The association of sebaceous differentiation in a Warthin tumor suggests a common histogenesis for both these tumors. This supposition is strengthened by the demonstration of microscopic foci of Warthin tumor in two otherwise typical sebaceous lymphadenomas (Rawson et al. 1950; Wasan 1971).

Necrotizing sialometaplasia is a nonneoplastic lesion of salivary glands that exhibits the histologic features of squamous metaplasia of ducts and acini, acinar and/or lobular necrosis and sialadenitis. It commonly involves the hard and/or soft palate, but may occur anywhere salivary gland tissues are found and has been noted by this author to occur in the nose, and nasopharyngeal areas. Only 19 of the 106 reported cases, including this report, involved the major salivary glands (Donath 1979; Lynch et al. 1979; Brannon et al. 1979). Necrotizing sialometaplasia may develop after a surgical procedure for a benign or malignant lesion. The time interval from initial surgery to development and diagnosis of necrotizing sialometaplasia ranged from 6 to 45 days in the study of Brannon and Corio (1979) with a mean of 17 days. They concluded that "... any factor which causes a comprise or obstruction in the blood supply to a minor or major salivary gland or to a mucoserous gland of the upper respiratory tract may result in necrotizing sialometaplasia". This case strongly supports this conclusion. The necrotizing sialometaplasia was seen only in the region of the original biopsy. The initial biopsy traumatized the area injuring the arterial supply which presumably triggered the metaplastic process.

The importance of recognizing necrotizing sialometaplasia is illustrated by several reported cases. Myers et al. (1975) reported the case of a 46 year old woman who received a diagnosis of mucoepidermoid carcinoma of the hard palate and underwent an en bloc removal of the posterior hard palate, anterior part of the soft palate and left aveolar ridge. The final diagnosis was necrotizing sialometaplasia. Arguelles et al. (1976) reported a 33 year old white female who had a wide local excision for a "well differentiated mucoepidermoid carcinoma" on the posterior third of the left hard palate. They also reported another patient, a 53 year old white male with a painful lesion of the left hard palate. An excisional biopsy was reported as a well differentiated epidermoid carcinoma. Radical surgery was averted on review of the slides.

As these three cited cases illustrate, it is important to recognize necrotizing sialometaplasia. This author has found several useful points which aid in making this diagnosis: The uniformity and bland appearing squamous cell nests arranged in lobular fashion, and the location of residual ductal lumina in one or more of these squamous nests strongly support a benign metaplastic process. A history of previous trauma or surgery is helpful; however, several cases have been reported without an obvious etiology. One must keep this diagnosis in mind in evaluating any lesion reported to arise in the areas where salivary glands are found.

Recently Seifert et al. (1980) have described four histologic subclassifications of cystadenolymphoma: 1) Typical, with an epithelial tumor component of 50%, 2) Stroma poor, with an epithelial tumor component of 70 to 80%, 3) Stroma-rich, with an epithelial tumor component of only 20 to 30%, and 4) Metaplastic with large areas of squamous cell metaplasia, stroma hyalinization and parenchymal necrosis. This latter group of tumors must be differentiated from necrotizing sialometaplasia and well differentiated epidermoid carcinoma.

The lack of cellular atypia and absence of stromal invasion would eliminate consideration of an epidermoid carcinoma. The lobular nature of necrotizing sialometaplasia involving the small ducts and the diffuse involvement of the metaplastic cystadenolymphoma, involving many of the larger ducts which would be lined by squamous epithelium should enable an observer to separate these entities on morphologic grounds.

In this case report a Warthin tumor with focal sebaceous differentiation and necrotizing sialometaplasia is reported. The association of the sebaceous foci in an otherwise typical Warthin tumor lends support for a common histogenesis of both components of the tumor.

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