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Case Report
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Sebaceous Lymphadenoma of the Parotid Deep Lobe

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Abstract

Sebaceous lymphadenoma is a benign rare tumor of the salivary glands, with reported cases accounting for less than 0.2% of all salivary gland neoplasms. Its etiopathogenesis is controversial and within the postulated theories, the sebaceous differentiation in salivary glands is thought to be related to hormonal influences. We describe an unusual case of sebaceous lymphadenoma in a patient with previous breast cancer, identified as an incidental finding of a parotid deep lobe lesion on computerized tomography, and treated with surgical excision.

Keywords: Sebaceous lymphadenoma; Parotid gland; Salivary neoplasm

Introduction

Although sebaceous glands are commonly found in the salivary glands, primary sebaceous tumors of the salivary glands are very rare. The reported cases of this benign neoplasm account for less than 0.2% of all salivary gland neoplasms [1]. First described by Rawson and Horn in 1950 [2], less than 50 cases of Sebaceous Lymphadenoma (SL), have been reported in the English literature. Majority of these cases arise within the parotid gland, with rare reports involving minor salivary gland and maxilla [1,3-5]. If treated with complete surgical excision, prognosis is excellent. We describe the case of a 50-year-old woman, with an incidental finding of a deep lobe parotid tumor on Computerized Tomography (CT), diagnosed histologically as sebaceous lymphadenoma.

Case Presentation

A 50-year-old female was referred to a tertiary head and neck center with a right-sided parapharyngeal mass, identified as an incidental finding on Computerized Tomography (CT) scan for investigation of worsening telangiectasia on the background of right breast adenocarcinoma treated 11 years ago. Her medical history included breast cancer treated with neoadjuvant chemotherapy, right sided mastectomy and axillary clearance, followed by adjuvant radiotherapy 11 years prior. She was a lifelong non-smoker. Physical exam was unremarkable, with no palpable mass appreciated, along with no cervical lymphadenopathy, and an intact facial nerve. Two ultrasound guided Fine Needle Aspirate Cytology (FNAC) were performed: The first was non-diagnostic, and the second classified as atypia of undetermined significance (Milan classification III). 18-Fluorodeoxyglucose-Positron Emission Tomography (FDG PET) revealed a 25 mm lesion in the deep lobe of the right parotid gland, mildly and uniformly metabolically active (Figure 1).

Magnetic Resonance Imaging (MRI) displayed a well-circumscribed solid mass measuring 25 mm × 26 mm × 35 mm located in the deep lobe of the right parotid gland extending through the parapharyngeal space. It demonstrated intermediate signal on T2-weighted sequences, hypointense signal on T1-weighted images, restricted diffusion and post contrast homogeneous enhancement. The lesion mildly displaced the carotid sheath medially and abutted the retromandibular vein laterally, with no evidence of invasion of surrounding structures (Figure 2). Following discussion at a head and neck multidisciplinary meeting, a right transcervical deep parotid lobe excision was performed without complications. Histologic sections showed a well circumscribed lesion consisting of islands of squamous and sebaceous cells with glandular/ductal structures, oncocytic cells and a prominent associated lymphoid-dense stroma (Figure 3 and 4). The case was further discussed in the head and neck multidisciplinary meeting, and after a four-day period of routine post-operative drain tube monitoring with no complications, was discharged to the community for routine surveillance.

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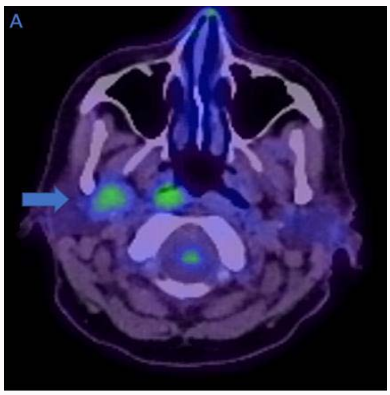

Figure 1: Axial fused18-FDG PET/CT. (A) Slightly and uniformly metabolical uptake within the right parotid deep lobe (SUVmax 2.3).

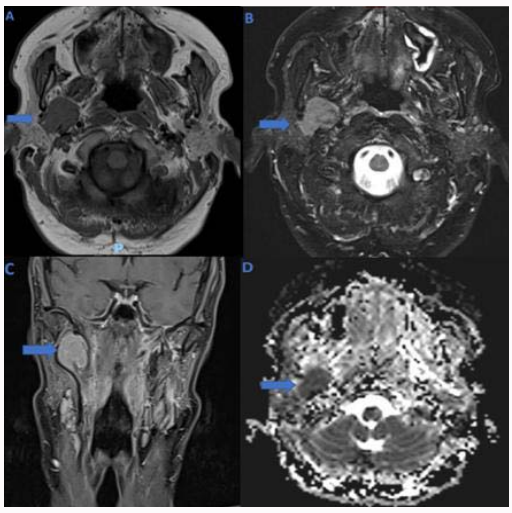

Figure 2: A) T1 axial showing hypointense signal within the well delineated parotid mass; B) T2 axial fat sat. Right parotid lesion showing intermediate T2 signal; C) T1 coronal fat sat post C+; D) ADC map. Primary parotid deep lobe mass showing restricted diffusion. Low signal on ADC map.

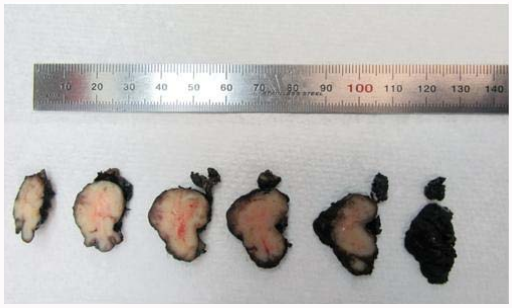

Figure 3: Right partial parotidectomy showing a circumscribed tumor with tan-white solid cut surface corresponding to the lymphoid stroma contrasting with the darker brown background of the salivary gland tissue. No lymph nodes identified.

Discussion

Sebaceous glands can be present in both major and minor salivary glands, and sebaceous differentiation has been reported within normal tissue [1]. SL is a rare benign salivary gland tumor, arising most commonly in the adult parotid gland [3]. Its etiopathogenesis is controversial. Postulated theories include origin from ectopic salivary gland inclusions within intraparotid or periparotid lymph nodes, from branchial cleft remnants, or from sebaceous metaplasia of the epithelial component of a Warthin tumor [1,4-6]. 75% of SL are diagnosed between the 6th and 8th decades, and are rare in the first 20 years of life. This is thought to be related to the fact that although sebaceous glands may be encountered in childhood, they reportedly occur more often after puberty, reaching a prevalence of almost 10% to 20% in young and middle-aged adults [7,8]. Sebaceous differentiation is thought to be related to hormonal influences, which may account for the rarity of SL in the pediatric population [1]. Clinically, SL presents as a slowly growing painless mass which may be present for many years, and is most common within the parotid gland, but has been reported in the minor salivary glands of the oral cavity, maxilla, and lip [1]. Radiological investigation with ultrasonography or cross-sectional imaging can reveal a well differentiated or encapsulated neoplasm that does not infiltrate surrounding tissues. Histopathology reveals a well demarcated biphasic lesion composed of variably sized epithelial islands set within a diffuse lymphoid background with occasional germinal centers. The epithelial nests have small dark basally located cells and larger clear luminal sebaceous cells with central nuclei and a vacuolated cytoplasm. Central cystic degeneration may be seen [3]. Although FNAC remains the cornerstone of the preoperative workup of any salivary gland lesion, the diagnosis of sebaceous lymphadenoma is challenging on cytologic material alone. This is due to the rarity of this lesion, heterogeneity of sampled components, and the cytologic overlap with other salivary gland neoplasms with sebaceous differentiation. In the literature, aspirates from histologically proven sebaceous lymphadenoma have been classified from benign, Atypia of Uncertain Significance (AUS) to Salivary Gland Neoplasm of Uncertain Malignant Potential (SUMP). The clear vacuolated appearance of the sebaceous cells and sometimes degenerative atypia on cytology may raise the false positive result of metastatic carcinoma with prominent clear cells. This highlights the problematic issue when the combination of cellular constituents are not represented on cytology and stresses the importance of clinical and radiological correlation when interpretating cytology findings [9]. The potential misclassification as metastatic carcinoma to a lymph node is of particularly relevance in our case due to the patient's history of prior breast carcinoma. A cellblock preparation, if available, is a valuable adjunct to the cytologic interpretation of these lesions as it enables ancillary testing (immunohistochemistry and/or molecular testing) to be utilized to eliminate potential differential diagnoses raised on

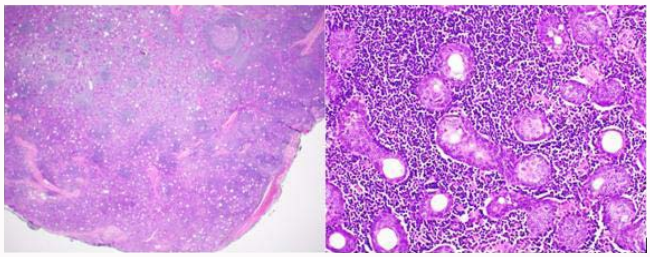

Figure 4: Histologic sections showing islands of squamous and sebaceous cells with glandular/ductal structures, oncocytic cells and a prominent associated lymphoid-dense stroma. There is no atypia in the epithelial or lymphoid population, no mucocytes/mucin, mitoses or necrosis. The constellations of features are consistent with a sebaceous lymphadenoma and argue against other types of salivary gland neoplasms which may show sebaceous differentiation.

cytology smears.

Complete surgical excision is the modality of therapy for SL and recurrence is very rare following complete removal. In addition, transformation to a sebaceous lymphadenocarcinoma has been reported but is extremely rare [10].

This would correspond to the second case described in the English literature in which an association between a parotid sebaceous lymphadenoma and breast cancer has been reported, being the first one located in the parapharyngeal space. Since this is a rare entity that has been reported in less than 40 cases, it is not unexpected that FNAs usually fail to make the diagnosis preoperatively. In this context, the clinical and radiologic features described in this report provide further information for pathologists interpreting FNA in patients with a history of breast cancer and parotid incidentalomas who initially had an inconclusive FNA. This, in turn, is essential to guide the clinician in making more evidence-based therapeutic decisions and to avoid unnecessary surgery in patients who may have significant comorbidities.

Conclusion

Sebaceous lymphadenoma is a rare salivary gland neoplasm that usually is incidentally found in the parotid gland. This is the second report describing an association between breast cancer and sebaceous lymphadenoma of the parotid gland, and the first one located in the parapharyngeal space. This may be an important clinical background for the histologic interpretation and therefore management of these patients who are usually adults with comorbidities.

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