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Polymorphous Low-Grade Adenocarcinoma of Parotid- Complicated by a Collision Tumor with Unusual Features

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Abstract: This case report concerns a patient, presented unusually with an ulcero proliferative parotid tumor; diagnosed histologically as polymorphous low-grade adenocarcinoma. (PLGA) Its rare association with a collision tumor of squamoid sebaceous carcinoma; arising from overlying adnexal elements is previously unreported. Controversies in the histological diagnosis of this entity; particularly in relation to its prognosis are emphasized and discussed, drawing comparison of similar tumors occurring elsewhere.

Keywords: PLGA of parotid- unusual clinical manifestation-collision tumor of sebaceous carcinoma- significance of papillary and solid elements.

INTRODUCTION

WHO blue book succinctly defines PLGA as a malignant tumor characterized by "cytologic uniformity, morphologic diversity and infiltrating growth pattern [1]. Many years ago, Friedmann and Lumenmann described 12 cases of this tumor under the title of lobular carcinoma because of its similarity to those arising in the breast and simultaneously a month after. Batsakis et al published cases of a similar tumor; publishing under the title of terminal duct carcinoma [2]. Following year, the current term was suggested as a clinically and morphologically distinct neoplasm, which is widely accepted [3]. Most of the papers published on this subject emphasized its predominant prevalence in minor salivary gland rather than from major salivary glands [4]. In the latter, PLGA occurred more often as a carcinomatous component of carcinoma ex pleomorphic adenoma [5].

The object of this presentation is not because of its occurrence; a rare site but because of its presentation as a collision tumor with histological features requiring further clarification.

FINDINGS

The patient under discussion, reported a slowly growing tumor in the upper part of the neck for the last three years; complicated all of a sudden prior to admission by its rapid growth with ulceration. The growth was fungating and bled on touch; an unusual occurrence in a tumor of this nature. Left parotidectomy with neck node dissection was made. The excised specimen measured 9.5x8.5x6.0cms. Multiple skin ulcerations with fungation were noticed. Cut section revealed a grayish white lobulated firm tumor with cystic areas replacing the entire parotid. Though lateral margins were free from the tumor, it was seen involving the surgical base. Microscopically the tumor showed variegated features. 40% of the tumor showed a lobular pattern; more marked at the periphery beneath the skin. (Figure-1) Indian file pattern, as well as storm of an eye appearance was not perceived. The tumor cells are relatively uniform and arranged solidly with rare acinar features. The nuclei are round and pale without pleomorphism and hyperchromasia. The lobules are separated by thick hyalinized basement membrane; as seen in basal cell adenoma; membranous type. Some of the basement membrane material was deposited in between tumor cells. Equally prominent were cells showing papillary patterns. All variants like thin papillary fronds with central cores, tubulo papillary and solid papillary elements were well represented in equal proportion (Figure 2 & 3). Mitotic activity was one per ten high power (HPF). Pseudo cribriform arrangements were rarely noticed. All nineteen lymph nodes resected show reactive hyperplasia and there were no tumor deposits. Immunochemistry using pan cytokeratin was

positive in epithelial elements. S100 and SMA were positive in solidly arranged tumor cells and vascular cores (Figure-4). Histological features of squamoid sebaceous carcinoma, a collision tumor are illustrated (Figure-5).

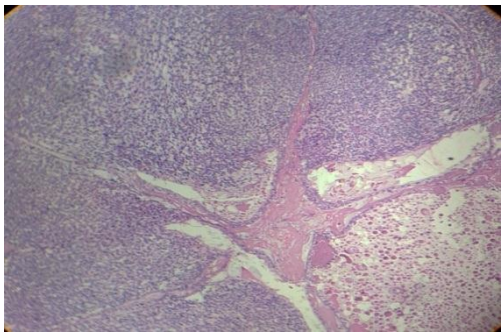

Fig-1: Solid tumor nodule with uniform cytology and cystic changes

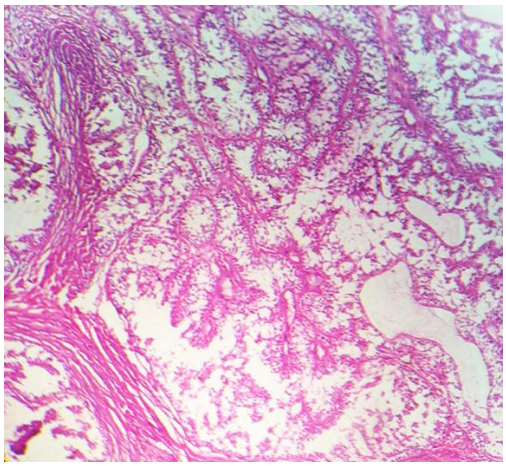

Fig-2: Papillary carcinoma with thin vascular cores

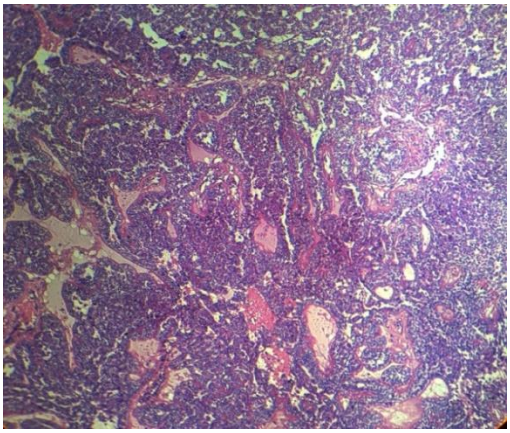

Fig-3: Tubulo papillary carcinomatous elements

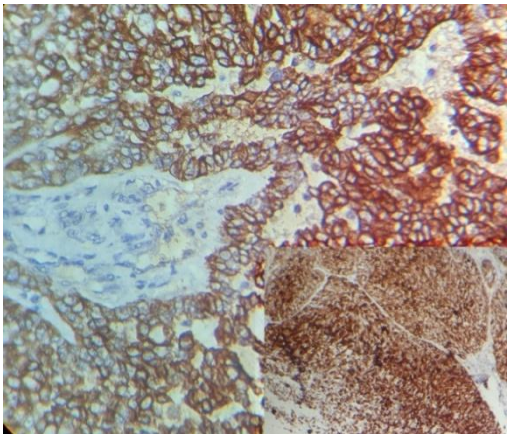

Fig-4: Cytokeratin positivity among epithelial cells; inset showing S100 positivity of solidly arranged tumor cells

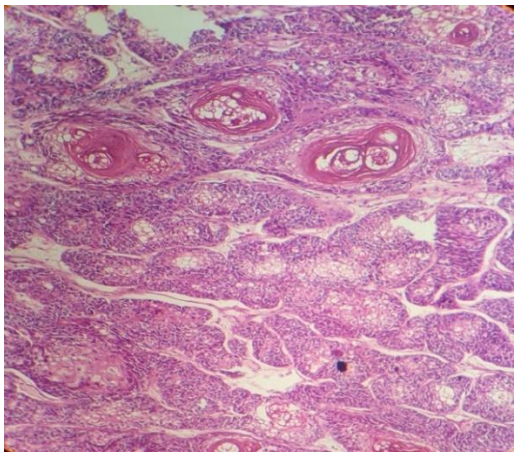

Fig-5: Squamoid sebaceous carcinoma; arising from skin

DISCUSSION

PLGA; so named because of a combination of many morphological entities, is derived from various ductal constituents present in salivary glands. The main histologic components are lobular, papillary, cribriform, small duct like structures forming concentric whorls or single file arrangements [3]. To which many more may be added like pseudo cribriform, microcystic, tubulo-papillary. Solid papillary, morphologic variants encountered in basal cell adenoma, spindle cell fascicular elements etc. Since this tumor is formed either singly or in combination with luminal and abluminal cells like basal and myoepithelial cells [6], the polymorphous appearance is a natural consequence. The bland morphology of the neoplastic cells and their tendencies for invasion are two distinct histological markers, repeatedly emphasized in many papers on this subject [7] and which are noticed in this patient also.

It is felt by the authors of the confusion surrounding the diagnosis of papillary elements; harbingers of worse prognosis, can be cleared if we consider and apply the same criteria employed in the histologic diagnosis of similar lesions in the breast. This problem is dealt with extensively in the WHO blue book on breast tumors [8]. Papillary carcinoma and its multiple morphological variants are diagnosed by assessing the proliferation of luminal cells, oriented around thin vascular cores and abluminal cells; which may be myoepithelial or basal in nature. In this connection we would like to emphasize another histological feature requiring close scrutiny is the presence of solid papillary carcinoma; a lesion usually under diagnosed in parotid but well-illustrated and elaborated in the blue book referred above. All these morphological entities including tubulopapillary pattern are noticed in the neoplasm under discussion and they bear close resemblance to those illustrated in the paper published by Evans and Luna [9]. Solid papillary carcinoma can be an in-situ lesion or one with invasive features and we consider it as an important factor in evaluating the prognosis in these cases.

Another stumbling block in judging the biologic potentialities of its solidly arranged non-papillary elements; a consideration likely to be missed if not studied immunochemically. Perez Odonez, Linkov, Huvos [10] in an analysis of 17 cases after performing immunochemical markers concluded that in PLGA luminal cell exhibited strong reaction to LMWK and vimentin while non-luminal cells showed a phenotype with basal differentiation and less commonly with myoepithelial cells. In our patient, the cells in lobular pattern showed positive reaction with SMA and

S100. Usually tumors with myoepithelial composition behave at the most as a low grade malignant tumor and we expect a similar course in our patient also. An unusual complication in this tumor, an incidental finding is the presence of a collision tumor consisting of sebaceous elements along with PLGA. Though sebaceous tumors can occur rarely as components in salivary neoplasm, in this patient uncommonly its origin is from overlying adnexal elements of skin; sharing classical morphological features of sebocytes with many epithelial pearls as in a variant of sebaceous carcinoma published as squamoid sebaceous carcinoma [11]. In spite of its innocuous morphology, it is deeply invasive.

This case is not only unique because of its combination with sebaceous carcinoma but also in its clinical manifestation and in spite of the significant presence of papillary elements. Its behavior is locally invasive without distant spread.

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