PHILIPPINE JOURNAL OF OTOLARYNGOLOGY-HEAD AND NECK SURGERY VOL. 34 NO. 1 JANUARY – JUNE 2019

Intracapsular Carcinoma ex Pleomorphic Adenoma

Jose M. Carnate Jr., MD[1]
Marvin C. Masalunga, MD[2]

[1]*Department of Pathology*
College of Medicine
University of the Philippines Manila

[2]*Department of Laboratories*
Philippine General Hospital
University of the Philippines Manila

This is the case of a 37-year-old female patient presenting with a 10-year history of a gradually enlarging right infra-auricular mass. A parotidectomy was performed. The surgical pathology specimen consisted of an 18 cm diameter encapsulated nodular mass with a homogenous, cream-tan solid surface. Microscopic section showed an encapsulated neoplasm with abundant chondromyxoid stroma and tubular epithelial elements characteristic of a pleomorphic adenoma. *(Figure 1)* Randomly scattered within the tumor were foci of haphazard and complex glands. *(Figure 2)* These glands exhibited nuclear pleomorphism, luminal necrosis and mitoses compatible with an adenocarcinomatous proliferation. *(Figure 3)* Based on these features, the case was signed out as an intracapsular carcinoma ex pleomorphic adenoma.

Carcinoma ex pleomorphic adenoma is a carcinoma arising from a pre-existing pleomorphic adenoma. The antecedent benign tumor may either be a long-standing one often with a history measured in decades, or characterized by a protracted history of excisions and multiple recurrences.[1,2] The carcinoma on the other hand is either epithelial or myoepithelial in derivation. By morphologic sub-type the most commonly reported carcinoma arising in a pleomorphic adenoma is a salivary duct carcinoma or an adenocarcinoma that is not otherwise specified (NOS).[1,3] Residual pleomorphic adenoma tissue is identifiable either in its typical morphology, a chondromyxoid stroma, or a hyalinized sclerotic nodule.[1]

Aside from the type of carcinoma arising from the pleomorphic adenoma, another parameter that has to be reported is the extent of involvement by the carcinomatous component. A carcinoma that is entirely limited to within the parent tumor that is still enclosed by a complete capsule is termed an "intracapsular" or "non-invasive" carcinoma ex pleomorphic

Figure 1. Tumor composed of chondromyxoid stroma (asterisk), and tubular epithelial elements (arrow) surrounded by a complete fibrous capsule (double arrow) (Hematoxylin-eosin, 40X magnification).

Correspondence: Dr. Jose M. Carnate, Jr.
Department of Pathology
College of Medicine, University of the Philippines Manila
547 Pedro Gil St. Ermita, Manila, 1000
Philippines
Phone (632) 526-4450
Telefax (632) 400-3638
Email: jmcjpath@gmail.com

The authors declared that this represents original material that is not being considered for publication or has not been published or accepted for publication elsewhere, in full or in part, in print or electronic media; that the manuscript has been read and approved by the authors, that the requirements for authorship have been met by the authors, and that the authors believe that the manuscript represents honest work.

Disclosures: The authors signed a disclosure that there are no financial or other (including personal) relationships, intellectual passion, political or religious beliefs, and institutional affiliations that might lead to a conflict of interest.

Philipp J Otolaryngol Head Neck Surg 2019; 34 (1): 68-69

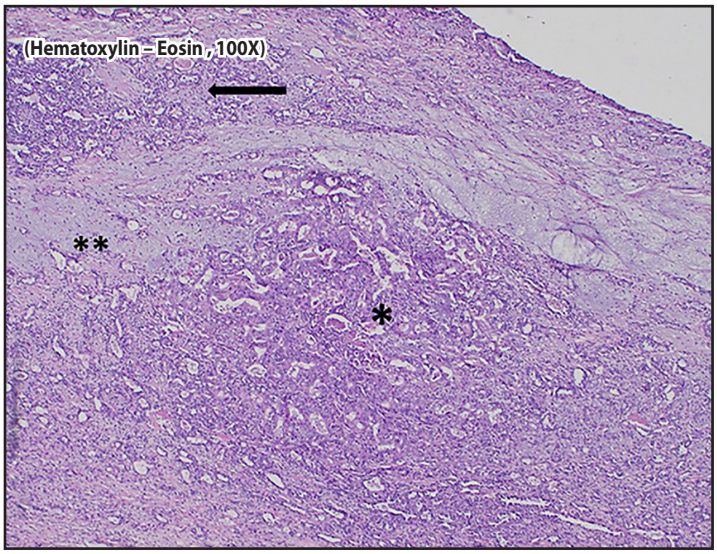

Figure 2. Haphazard and complex glands (asterisk) surrounded by chondromyxoid stroma (double asterisk) and tubular epithelial elements (arrow) (Hematoxylin-eosin, 100X magnification).

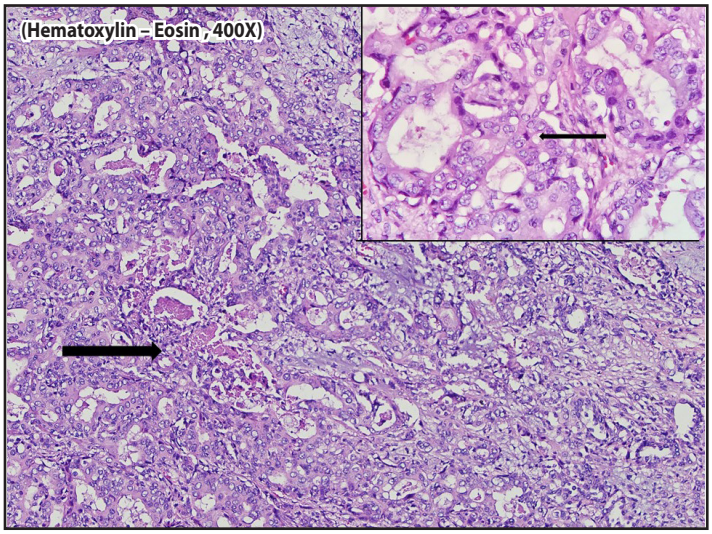

Figure 3. Haphazard and complex glands with luminal necrosis (thick arrow). Inset shows atypical nuclei and a mitosis (thin arrow) (Hematoxylin-eosin, 400X magnification).

adenoma.[1,2] Once the carcinoma breaches the capsule and infiltrates the surrounding tissue, then it is considered invasive. If the invasion is less than 4 – 6 mm beyond the capsular border, the tumor is termed "minimally invasive". Carcinomatous elements that extend beyond this threshold is termed "widely invasive".[1] This threshold is greater than the previous threshold of 1.5 mm recommended in an earlier edition of the WHO classification although the present edition does state that this threshold is preliminary and requires further validation.[1,2,4] It has to be pointed out though that quantifying invasion may not always be possible in tumors that have positive margins, those that are intrinsically unencapsulated such as minor salivary gland tumors, and those with complex multinodular growth patterns such as recurrent pleomorphic adenoma.[1] This difficulty has to be stated in the report and what conditions preclude quantifying the degree of invasion.

Non-invasive carcinoma ex pleomorphic adenoma has quite a good outcome with very low reported rates of recurrence or regional metastasis. In a review of thirty cases and a report of an additional three cases, only one case showed recurrence or metastasis.[3] This favorable outcome certainly contrasts with that of the widely invasive type where metastasis is reported to occur in up to 70% of cases.[1] Another review of ten cases showed one case developing metastasis, and recommended that non-invasive cases should thus still be followed up closely after primary treatment because regional or distant metastasis can occur.[2]

To the best of our knowledge, there are no published local data on the incidence of early malignant transformation of pleomorphic adenomas in the Filipino population. Hence, we take this opportunity to report this case. Awareness of the entity and prudent liberal sampling of these tumors may help address this gap.

REFERENCES

1. Williams MD, Ihrler S, Seethala R. Carcinoma ex pleomorphic adenoma. In: El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ. WHO Classification of Head and Neck Tumors. *IARC:* Lyon 2017 p.176.
2. Ye P, Gao Y, Mao C, Guo C, Yu G, Peng X. Carcinoma ex pleomorphic adenoma: is it a high-grade malignancy? *J. Oral Maxillofac. Surg.* 2016 Oct;74(10):2093-2104. PMID: 27131030 DOI https://doi.org/10.1016/j.joms.2016.03.037.
3. Mori T, Kunimura T, Saito K, Date H, Arima S, Matsuo K, Ochiai Y, Morohoshi T. Three cases of noninvasive carcinoma ex pleomorphic adenoma of the parotid gland and a literature survey focusing on their clinicopathologic features. The Showa University Journal of Medical Sciences. 2010 Jun;22(2):127-134. DOI https://doi.org/10.15369/sujms.22.127.
4. Gnepp DR, Brandwein-Gensler MS, El-Naggar AK, Nagao T. Carcinoma ex pleomorphic adenoma. In: Barnes L, Eveson JW, Reichart P, Sidransky D. World Health Organization Classification of Tumours. Pathology and Genetics Head and Neck Tumors. *IARC:* Lyon 2005 p. 242.