Epithelial-Myoepithelial Carcinoma of Parotid Gland : A Case Report with Immunohistochemical and Ultrastructural Studies

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Abstract

A 66-year-old man presented with a painless mass of the parotid gland. Light and electron microscopic studies verified the basic nature of the tumor as epithelial-myoepithelial carcinoma, a low-grade malignant neoplasm of the salivary gland. Pathologically, there were two types of cells; the inner eosinophilic epithelial cells lining the ducts and the outer clear cells. The former cells displayed immunoreactivity for cytokeratin and ultrastructural features of apical microvilli and desmosome. The latter cells were positive for actin, S-100 protein, vimentin and the cytoplasm contained actin microfilaments. Such pathological findings were characteristic features of this rare tumor. To our knowledge, this is the first reported case of EMC in Thailand.

Several salivary gland tumors have both epithelial and myoepithelial components such as pleomorphic adenoma (benign mixed tumor), adenoid cystic carcinoma, polymorphous low-grade adenocarcinoma, etc[1]. Therefore, mixed tumor, the most common neoplasm involving major and minor salivary glands, should be defined as a tumor that characteristically shows combined features of epithelioid and connective tissue-like growth rather than a tumor that is uniquely composed of epithelial and myoepithelial cells[1]. Epithelial-myoepithelial carcinoma (EMC), on the other hand, is extremely rare and is another example of salivary gland neoplasm that possesses both epithelial and myoepithelial elements[2]. We described herein a case of EMC of the parotid gland which appears to be the first case in this country. Immunohistochemical and EM provided the specific nature of this lesion.

CASE REPORT

A 66-year-old man developed a slow-growing painless mass of the left parotid gland of 3 years' duration. He underwent surgical excision of the mass. The lesion, however, recurred gradually 7 years later.

Physical examination revealed a large solid hard mass at the left parotid region. It was

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fixed to the skin as well as the adjacent tissue. The mass was neither tender nor pulsatile. Regional lymph nodes were not palpated. Cranial nerves were bilaterally intact.

Left total parotidectomy with facial nerve preservation was performed. The post operative course was uneventful. He also received radiotherapy 2 weeks after the surgery.

Pathologic Examination

The surgical specimen consisted of a 5 x 4.5 x 4 cm grey white mass which was firm in consistency. Fragments of tissue were fixed in 10 per cent formalin, embedded in paraffin and stained with haematoxylin and eosin (H & E), mucin, and diastase-digestible periodic-acid-Schiff (PAS). Sections of the paraffin-embedded tissue were also processed by immunohistochemical method using antibodies to actin, S-100 protein, vimentin, and cytokeratin (CAM). Moreover, a portion of the specimen was washed and refixed in buffered 2 per cent glutaradehyde solution, embedded in epoxy resin, and prepared for electron microscopic study by standard procedure.

Microscopically, the tumor consisted of many small ducts, which were lined by simple cuboidal cells with eosinophilis cytoplasm, and surrounded by several outer layers of clear tumor cells (Fig. 1A). The latter cells contained glycogen as demonstrated by diastase-digestible PAS positive cytoplasmic granules.

Immunohistochemical study of the tumor cells showed that ductal lining cells were positive for cytokeratin (Fig. 1B). The surrounding clear cells demonstrated immunoreactiveties to actin, S-100 protein, and vimentin (Fig. 1C, D, E).

Electron microscopic study disclosed the biphasic components of the tumor. The cuboidal epithelial cells displayed apical microvilli and desmosomes (Fig. 2). The clear cells were also joined to each other by desmosomes and contained 5 nm filaments in the cytoplasm (Fig. 3). According to the size of the filaments and actin positive immunohistochemistry, we consider these filaments as actin microfilaments.

The pathological diagnosis was epithelial-myoepithelial carcinoma of the left parotid gland.

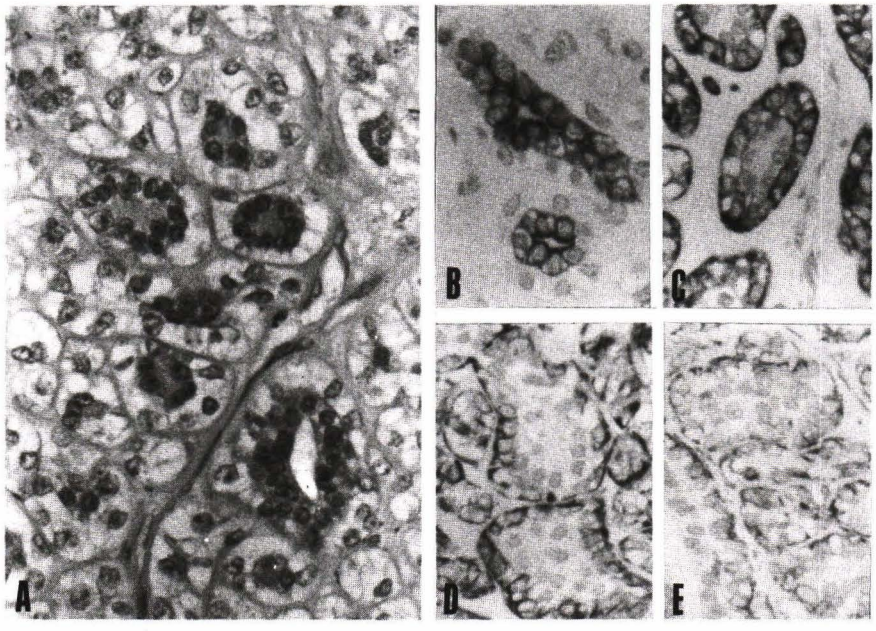

Fig. 1. Pathology of epithelial-myoepithelial carcinoma. A. Cuboidal epithelial-lined ducts surrounded by clear tumor cells. (H & E x 400) B. The ductal epithelial cells reveal positivity to cytokeratin. (H & E x 400) C-E. The clear tumor cells express actin, S-100 protein, and vimentin, respectively. (H & E x 400, each).

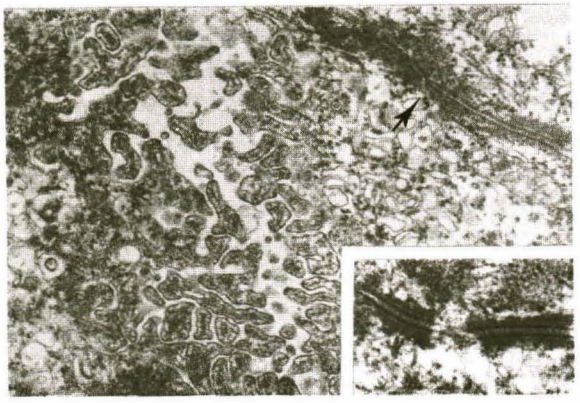

Fig. 2. Electron micrograph of epithelial cells demonstrates apical microvilli and desmosome (arrow). (x 22,500) Inset. Higher magnification shows desmosome with central line. (x 37,500).

DISCUSSION

Epithelial-myoepithelial carcinoma (EMC) is a rare type of low-grade salivary gland carcinoma accounting for 0.5-1 per cent of salivary gland neoplasms[2-5]. Its origin from intercalated ducts has been suggested[2,3]. Most tumors occur in the parotid gland. The tumor has a predominant occurrence in women particularly within the sixth and seventh decades of life with the ages ranging from 15 to 81 years and the average age of 59.4 years[2]. Clinically, this tumor is indistinguishable from other tumors of the parotid gland. It usually presents as a single painless slow-growing mass as seen in our patient. Rarely, the lesion may involve the facial nerve[2,5].

The histopathology of the EMC is unique as it is composed of small ducts lined by cuboidal-to-low columnar epithelial with eosinophilic cytoplasm and surrounded by outer layers of clear cells containing glycogen[2,4,5]. The ductal cells express immunoreaction to cytokeratin while clear tumor cells show positivities for actin, S-100 pro-

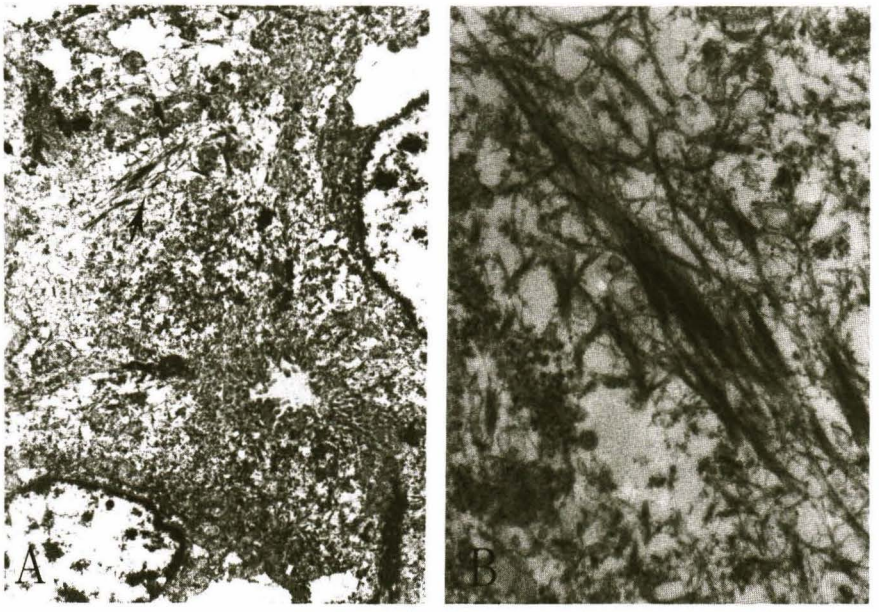

Fig. 3. A. Low-power view of electron micrograph showing adjacent myoepithelial cells containing bundles of microfilaments (arrow). B. High-power view of 5 nm actin microfilaments in a neoplastic cell (x 45,000).

tein, and vimentin indicating their epithelial and myoepithelial nature, respectively[2,3,6]. Such pathological findings aforementioned were also demonstrated in our case. Moreover, the electron-microscopic study also exhibited biphasic patterns of the tumor, supporting the immunohistochemical findings[7].

In some instances, EMC, however, must be differentiated from other neoplasms that possess clear cells such as mucoepidermoid carcinoma, acinic cell carcinoma, and clear cell variant of pleomorphic adenoma[5]. The presence of mucin-producing cells in mucoepidermoid carcinoma allows differentiation of this tumor[4,5]. Acinic cell carcinoma can be diagnosed by basophilic granularity of the cytoplasm. Moreover, clear cells in acinic cell carcinoma contains zymogen granules which are positive in PAS with and without diastase digestion[5]. Finally, lack of myxoid stroma and areas of pseudocartilage are evident against clear cell variant of pleomorphic adenoma[1,5].

Surgery is considered as the primary mode of treatment[2,3]. Total parotidectomy is recommended for parotid gland tumor. The facial nerve should be preserved if possible[2]. It should be noted that recurrence can occur after surgery as noted in our example in about one-third of cases[8]. The role of postoperative radiotherapy is controversial because of insufficient information[2].

SUMMARY

Epithelial-myoepithelial carcinoma (EMC), a rare low-grade malignant salivary gland neoplasm, was reported in a 66-year-old man. Immunohistochemical and ultrastructural studies demonstrated both epithelial and myoepithelial components of the tumor. Differential diagnoses were discussed.

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เอปิธีเลียล–มัยโอเอปิธีเลียล คาร์ซิโนมา ของต่อมพาโรติด: รายงานผู้ป่วย

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ผู้ป่วยชายไทย อายุ 66 ปี มีก้อนโตขึ้นช้า ๆ ที่ต่อมพาโรติดข้างซ้าย ผลการตรวจทางพยาธิวิทยาพบเป็น เอปิธีเลียล–มัยโอเอปิธีเลียล คาร์ซิโนมา การย้อมพิเศษ และอิมมูโนฮิสโตเคมี สนับสนุนการวินิจฉัยในผู้ป่วยรายนี้ นอกจากนี้ยังได้ทำการศึกษาโดยจุลทรรศน์อีเลคตรอนเพื่อศึกษาลักษณะจุลวิภาคของเนื้องอกชนิดนี้ด้วย เท่าที่ทราบผู้ป่วยรายนี้เป็นเอปิธีเลียล–มัยโอเอปิธีเลียล คาร์ซิโนมา ของต่อมน้ำลายรายแรกที่รายงานในประเทศไทย

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